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Experiences of peer navigators implementing a bilingual multilevel intervention to address sexually transmitted infection and HIV disparities and social determinants of health

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ABSTRACT (ENGLISH)

Introduction

Sexually transmitted infections (STIs) and human immunodeficiency virus (HIV) disproportionately affect young gay, bisexual and other men who have sex with men (GBMSM) and transgender women of colour. We explored the experiences of community-based peer navigators (‘Community Navigators’) who participated in *Impact Triad*, a bilingual multilevel intervention developed by our community-based participatory research partnership to reduce STIs and HIV and address social determinants of health (e.g., employment, education, social support and discrimination) among young GBMSM and transgender women of colour.

Methods

Individual in-depth interviews were conducted with 15 Community Navigators who participated in *Impact Triad*. Themes were identified through constant comparison.

Results

Community Navigators' mean age was 31.4 years. Seven were self-identified as African American/Black, 5 as Latine, 2 as multiracial/multiethnic, 1 as Asian American, 10 as cisgender men, 4 as transgender women and 1 as gender nonbinary. Thirteen themes emerged in three domains: (1) key aspects of the Community Navigator role (e.g., desire to serve as a community resource, the importance of being part of the communities in which one was working, the value of having an official role, being connected to other Community Navigators to problem-solving and sustaining intervention aspects long-term); (2) experiences implementing *Impact Triad* (e.g., engaging community members, meeting prioritized needs, building trust, using social media, increasing awareness and knowledge and challenges related to COVID-19) and (3) lessons learned for future interventions (e.g., facilitating access to broader resources, building additional skills and increasing interactions among Community Navigators).

Conclusion

Interviews identified important learnings about serving as Community Navigators and implementing *Impact Triad* that can guide future efforts to address STI/HIV disparities and social determinants of health through community-based peer navigation.

Patient or Public Contribution

Throughout this intervention trial, our partnership worked collaboratively with a study-specific community advisory board (CAB) comprised primarily of young GBMSM and transgender women of colour. Members of this CAB participated in all aspects of the trial including trial design, intervention development, recruitment and retention strategies, data collection and analysis, interpretation of findings and dissemination.

FULL TEXT

INTRODUCTIONSexually transmitted infection (STI) and human immunodeficiency virus (HIV) disparities

There is an urgent need to address disparities related to STIs and HIV in the United States. STI and HIV rates are highest among young persons,[1,2] and gay, bisexual and other men who have sex with men (GBMSM) and transgender women, and particularly those who are persons of colour, also carry a disproportionate burden of STIs and HIV. For example, GBMSM comprise approximately 4% of the US adult male population but 43% of all syphilis cases in the country and 86% of all HIV diagnoses among men,[2–4] and if current rates persist, one in two African American/Black and one in four Latine GBMSM may be diagnosed with HIV during his lifetime.[5] (The term 'Latine' uses a gender-neutral 'e', which replaces the gendered endings 'a' and 'o' as in 'Latina' and 'Latino', and is similar to 'Latinx'; this term is increasingly used within Latine LGBTQ communities). It is estimated that 14% of transgender women in the country are living with HIV, with prevalence rates as high as 44% among African American/Black and 26% among Latine transgender women.[6] Furthermore, the US South has high STI rates compared to other regions of the country[1,7] and has been referred to as the 'new' and 'latest' US HIV epicentre.[8]

Social determinants of health

The issues contributing to STI and HIV disparities experienced by young GBMSM and transgender women of colour are complex. Social determinants that influence the health of these communities include individual, sociocultural, environmental, system and policy factors. For example, GBMSM and transgender women, particularly those who are persons of colour, are more likely than heterosexual and cisgender counterparts to experience limited access to employment and education and face high rates of discrimination in a range of settings including health care, workplaces and schools.[9–12] In addition, employment status, education level and related factors such as poverty, income and health insurance coverage have been associated with STI and HIV incidence and outcomes.[13,14] There has been a call to broaden the focus of STI and HIV prevention efforts to address these 'upstream' factors that profoundly impact sexual health, in addition to individual-level factors such as increasing correct and consistent condom use and STI and HIV screening.[15,16]

Community-based participatory research (CBPR) and community-based peer navigation

CBPR, which engages community members and community organizations as partners in all phases of the research process,[17] has been identified as an important approach to understanding and reducing disproportionate STI and HIV burdens.[18,19] Community-based peer navigation leverages natural helping within existing social networks and has been used in STI and HIV prevention efforts with promising results.[20–24] There is a need for further development and evaluation of community-based peer navigation interventions to address STIs and HIV as well as contributing social determinants using CBPR.

Centers for Disease Control and Prevention (CDC) Community Approaches to Reducing Sexually Transmitted Diseases (CARS) initiative and the *Impact Triad* intervention

CARS is a unique initiative of the US CDC that promotes the use of community engagement to increase STI and HIV prevention, screening and treatment and address related social determinants of health within communities disproportionately affected by STIs and HIV. The initiative focuses on identifying and implementing innovative community-driven strategies that leverage community assets.[25] As part of the CARS initiative, our CBPR partnership developed and tested *Impact Triad*, a bilingual (English and Spanish), multilevel intervention designed to reduce STIs and HIV and improve social determinants of health among young GBMSM and transgender women of colour in a high-incidence community within the US South.

Purpose

A better understanding of the implementation of community-based peer navigation interventions is essential to strengthening future STI and HIV prevention efforts among communities facing health disparities. As a component of the process evaluation of *Impact Triad*, we qualitatively explored the experiences of community-based peer navigators within the intervention, using individual in-depth interviews.

METHODSDevelopment of *Impact Triad*

The *Impact Triad* intervention trial was conducted by the North Carolina Community Research Partnership. This partnership is a long-standing CBPR partnership comprised of community members, community organization and clinic representatives and academic researchers.[18] Throughout this trial, our partnership worked collaboratively with a 15-member community advisory board (CAB) comprised primarily of young GBMSM and transgender women of colour.

We first conducted a community-driven needs assessment to identify and better understand needs and priorities related to STI and HIV prevention, screening and treatment and social determinants of health among young GBMSM and transgender women of colour locally, as well as existing community assets. Through this process, the CAB and CBPR partnership prioritized four social determinants of health as particularly salient for young GBMSM and transgender women of colour and their risk for STIs and HIV: employment, education, social support and discrimination. Based on needs assessment findings, the CAB and CBPR partnership developed intervention strategies to reduce STIs and HIV and improve these social determinants of health and integrated these strategies into the *Impact Triad* intervention, the details of which are described elsewhere.[26]

Briefly, *Impact Triad* includes three primary multilevel strategies: community-based peer navigation, use of social media and antidiscrimination trainings for community organization staff. The intervention involved training young GBMSM and transgender women of colour as community-based peer navigators, known as ‘Community Navigators’ in English and ‘Navegantes Comunitarios’ in Spanish, to carry out helping activities with members of their social networks in the community within the context of their daily lives. In addition to one-on-one and group-level in-person helping to share information and resources related to STI and HIV prevention, screening and treatment and prioritized social determinants of health, Community Navigators created and updated intervention-related social media accounts (e.g., Facebook and Instagram) and used their own social media accounts for messaging with social network members. Community Navigators were also involved in the development of brief online video testimonials designed to raise consciousness among community organizations about the challenges and barriers young GBMSM and transgender women of colour face when accessing services related to STIs and HIV and social determinants of health and ways to facilitate access (https://www.youtube.com/channel/UCd7gOGhBerT0w1CTq5BwMcQ).

Community Navigator recruitment and training

Fifteen young GBMSM and transgender women of colour identified as informal leaders among their communities were recruited and trained to serve as *Impact Triad* Community Navigators. To be eligible to serve as a Community Navigator, a participant was 18 years of age or older; self-identified as a person of colour; self-identified as a man or as a transgender woman; reported sex with men and provided informed consent.

Community Navigators completed four 4-h training sessions, in two cohorts of seven to eight Community Navigators each. They were trained to serve as (1) health advisors to provide information to social network members to meet needs and priorities related to STI and HIV prevention (e.g., condom and pre-exposure prophylaxis [PrEP] access and use), screening and treatment, as well as social determinants of health (e.g., employment and education), offering guidance on where and how to access available services; (2) opinion leaders to bolster healthy and reframe unhealthy norms and expectations related to STIs and HIV or social determinants of health (e.g., social support) and (3) community advocates to bring the voices of young GBMSM and transgender women of colour to local community organizations by sharing feedback for improvement based on the perspectives of social network members.

Community Navigators were trained to use an adapted version of the ‘ask-advise-assist’ model,[27] represented by the acronym ‘IMPACT’ in English or ‘IMPACTO’ in Spanish with each letter representing a step in the natural helping process. A low-literacy wallet-sized reminder card was developed to serve as a ‘cheat sheet’ for Community Navigators outlining these steps to support others.[26] Upon graduation, Community Navigators received a framed certificate of training completion, an identification badge, a t-shirt and bag with the *Impact Triad* intervention logo and intervention materials (e.g., condoms, penis models and informational brochures) to carry out their work within their social networks and communities of young GBMSM and transgender women of colour. Community Navigators worked informally and formally with members of their social networks for 12 months and met monthly as a group with one another and with members of the CBPR partnership in convenient locations within trusted community organizations to plan, coordinate and evaluate their activities.

Community Navigators were provided a $50 stipend for each training session and each month of the 12-month intervention implementation. They also were provided $50 per month to compensate for transportation costs.

Qualitative interviews

After the conclusion of intervention implementation, individual in-depth interviews were conducted with all 15 *Impact Triad* Community Navigators. Standardized interview guides were developed in English and Spanish with careful consideration of wording, sequence and content to explore experiences with the intervention and current issues affecting the health of young GBMSM and transgender women of colour. Abbreviated sample items from the guide are outlined in Table 1.

Table 1 In-depth interview guide with domains and abbreviated sample items

Impact Triad project
Tell me about how you became involved with the *Impact Triad* project.
When you became a Community Navigator, was your role how you thought it would be? What was different than you expected?
What did you like about being a Community Navigator?
What was hard about being a Community Navigator?
Training
How well prepared were you to serve as a Community Navigator? What other training would have helped you?
What suggestions for other topics should we have included?
How new for you was the information you received?
Recruitment
Tell me how you recruited the young gay and bisexual men and transgender women of colour who are part of your social network to participate in *Impact Triad*.
Community Navigator experience
What are some challenges that you faced as you began working with other young gay and bisexual men and transgender women of colour in your community?
What were some of the things that made it easier to talk to other young gay and bisexual men and transgender women of colour about sexual health? To talk about social determinants of health, such as employment, education, social support and discrimination? What were some of the things that made it difficult?
What were the most popular topics that young gay and bisexual men and transgender women wanted to discuss? What topics did they not seem comfortable talking about?

How did you use social media to work with young gay and bisexual men and transgender women of colour?
What do you think worked well with social media?
What challenges did you have using social media? Were you able to work them out?
Think back to before *Impact Triad* started. Are the topics that you and your friends talk about now any different? How have the ways your friends provide support to one another changed?
How were the monthly meetings with the *Impact Triad* coordinators and the other Community Navigators?
PrEP
How do the people who were part of your group feel about PrEP?
How interested are they in PrEP?
What barriers do they face to using PrEP?
Alcohol and drug use
How common is alcohol use among the people who were part of your group? Among other young gay and bisexual men and transgender women of colour?
How about drug use? What types of drugs do they use?
How common is it to use alcohol when having sex?
How common is it to use drugs when having sex? What drugs are used during sex?
What does alcohol and drug use have to do with risky sexual behaviours?
Social determinants of health
In *Impact Triad*, we focused on employment, education, social support and discrimination as social determinants of health. What else affects the health and well-being of young gay and bisexual men and transgender women of colour?
How big of an issue is housing for young gay and bisexual men and transgender women of colour? How big of an issue is access to food? How do these issues affect their health?
Individual perceptions and empowerment

What do you think healthy sexuality means to young gay and bisexual men and transgender women of colour in our local community? Have your ideas of what healthy sexuality looks like changed since participating in *Impact Triad?*
What do you think would be good ideas or ways to prevent STIs and HIV among young gay and bisexual men and transgender women of colour living in your community? What do you think would be good ideas or ways to improve social determinants of health?
Do you think you will continue your work as a Community Navigator? Why?

Because of the COVID-19 pandemic, interviews were conducted via telephone. Two trained bilingual members of our CBPR partnership conducted the interviews in English (*n* = 12) and Spanish (*n* = 3). Interviews averaged 45 min in length. All Community Navigators provided written consent and were compensated $50 for participating in an interview. Human subject protection and oversight were provided by the Wake Forest University School of Medicine Institutional Review Board.

Data analysis and interpretation

All interviews were digitally recorded and transcribed. Constant comparison, an approach to grounded theory, was used to analyse interview data. Constant comparison combines qualitative coding with simultaneous comparison; initial observations are continually refined throughout data collection and analysis.[28] Three analysts read and reread interview transcripts, compared and contrasted content categories based on each analyst's interpretation of the data and identified emerging themes. After preliminary themes were developed, analysts came together with other CBPR partnership members via WebEx (a videoconferencing platform) in multiple meetings to discuss and reconcile final themes using an iterative process. Themes were also presented to the CAB in a meeting via WebEx for refinement and validation; CAB members were invited to respond to the themes and offered insights regarding themes that resonated with their own experiences and themes that they considered priorities for informing future efforts to improve the health of young GBMSM and transgender women of colour. At each stage in the analysis and interpretation process, a consensus approach was used, resolving discrepancies through discussion.

RESULTSCommunity Navigator characteristics

Key characteristics of the *Impact Triad* Community Navigators at the time of enrolment in the intervention trial are presented in Table 2. The average Community Navigator age was 31.4 years. Seven Community Navigators (47%) identified as African American/Black, 5 as Latine (33%), 2 as multiracial/multiethnic (13%) and 1 as Asian American (7%); 10 identified as cisgender men (67%), 4 as transgender women (27%) and 1 as gender nonbinary (7%); 10 identified as gay (67%), 4 as heterosexual (27%) and 1 as bisexual (7%); 4 had a high school diploma or lower (27%) and 11 had at least some college (73%); 5 were in school full-time (33%) and 12 were employed (80%).

Table 2 Select characteristics of *Impact Triad* Community Navigators (*n* = 15)

Characteristic	Mean (SD) or *n* (%)
Age (years)	31.4 (8.4)
Racial/ethnic identity	
African American/Black	7 (47)
Hispanic/Latine	5 (33)

Multiracial/multiethnic	2 (13)
Asian American	1 (7)
Gender	
Cisgender man	10 (67)
Transgender woman	4 (27)
Gender nonbinary	1 (7)
Sexual orientation	
Gay	10 (67)
Heterosexual	4 (27)
Bisexual	1 (7)
Education level	
Less than a high school diploma or equivalent	1 (7)
High school diploma or equivalent	3 (20)
Some college	5 (33)
2-year degree	1 (7)
4-year degree	4 (27)
Master's degree, professional degree or more	1 (7)
Student status	
Not in school	10 (67)
In school full-time	5 (33)
Employment status	
Employed	12 (80)
Disabled and not working	2 (13)

Unemployed	1 (7)

Qualitative findings

Thirteen themes emerged from the interviews and were organized into three domains: (1) key aspects of the Community Navigator role, (2) experiences implementing the *Impact Triad* intervention and (3) lessons learned for future interventions.

Key aspects of the Community Navigator role

Community Navigators identified several important aspects of fulfilling their role within *Impact Triad*. These themes, along with selected quotations, are presented in Table 3.

Table 3 Key aspects of the Community Navigator role (qualitative themes with select quotations from in-depth interviews)

Desire to serve as a community leader
Community Navigators valued serving as leaders and as resources to address social determinants of health within their communities of identity.
I liked getting to know other Latinos, and that I learned more about STDs and helped the community to find resources that helped them to live better. (Participant [P] 15)
I am educated and went to college, but I never felt like I achieved anything. After being a Community Navigator I feel like I have something to give to the community and it's great. (P11)
Importance of being part of the communities in which one was working
Being members of the communities in which they worked was fundamental to trust building and successful intervention implementation by Community Navigators.
We keep it one hundred, real, and hit the street, because that is what people respect and is how we earn trust, because we are demonstrating how we are part of this world. (P12)
Yo creo que mi credibilidad—ellos creen en mí—y que soy parte de la comunidad, eso fue lo que me ayudó. [I believe that my credibility—they put their faith in me—and that I am part of the community, that is what helped me out.] (P2)
Value of having an official role
Having official titles and roles contributed to Community Navigators' self-assurance and validated their work in communities.
Podíamos hablar de temas de sexualidad sin que fuera tabú. Y era más fácil teniendo el respaldo de una institución y un programa para resolver algunas de las preguntas que tenían algunos de mis amigos y les daba pena hacer. [We were able to discuss topics about sexuality without it being taboo. It was also easier having the backing of an institution and program to assist in answering the questions that some of my friends had but were shy to ask.] (P1)

I was extremely well prepared and was ready to become a Community Navigator for all the good, the bad, ugly, and different. We all prepared for everything no matter what happens. (P11)
Connections built with other Community Navigators
Being connected to the other Community Navigators across gender identities, sexual orientations and racial/ethnic identities increased a sense of belonging, resource sharing, creativity and problem solving.
Well for me, I am a helper. I have always been an advocate for the transgender community but didn't have the opportunity to be part of a group before, so having that opportunity was even better than what I thought it would be. It was very special. (P6)
I think [the monthly Community Navigator meetings] were probably the most impactful for me, honestly. Connecting with other Navigators and being able to talk about things that we had experienced. (P7)
Sustaining intervention aspects long-term
Community Navigators sustained aspects of the intervention after implementation officially ended.
I will continue work as a Community Navigator because it isn't a job but is something that needs to be done, even if I am not paid for it. It just needs to be done. (P10)
I think I will always be a Community Navigator, formally or informally, because I know something that I can always use and I will always be involved doing something to help others. (P7)

Desire to serve as a community leader

Community Navigators expressed that they valued serving as leaders and as resources to address social determinants of health within their communities. Among the Community Navigators, there was a common sense of intrinsic motivation to engage, be involved with and help their communities of identity—LGBTQ communities and communities of colour. Community Navigators reported that they were drawn to the *Impact Triad* intervention because of the opportunity to gain knowledge to share within their social networks and because they recognized the importance of both promoting sexual health and addressing social determinants that can make it difficult for community members to take care of their health. Community Navigators also appreciated that they made a genuine impact, which furthered their desire to continue being leaders within their social networks and communities. They felt pride in their roles as Community Navigators, saw themselves as fulfilling a need for social support within their communities, enjoyed building leadership skills that could potentially create opportunities for future careers (e.g., public speaking skills) and demonstrated a desire to continue utilizing the knowledge and training they received postintervention.

Importance of being part of the communities in which one was working

Community Navigators emphasized that being members of the communities in which they worked was fundamental to trust building and successful intervention implementation. Community Navigators had shared experiences and identities and felt a sense of belonging with others in their communities and were known and seen as leaders in their communities before becoming involved with *Impact Triad*. These connections led to increased initial levels of rapport with community members within Community Navigators' social networks, facilitated the process of recruiting social network members to participate in the intervention and were helpful for further building trust to have conversations about a wide array of health-related topics.

Value of having an official role

Community Navigators reported that having official titles and roles that were linked to well-known and well-respected institutions (i.e., Wake Forest University School of Medicine and Triad Health Project, an established AIDS service organization in Greensboro, North Carolina), and having received formal training through these institutions, provided a sense of reassurance that helped to facilitate relationship building with members of their communities. As a consequence, Navigators felt more confident in their knowledge about sexual health and social determinants and perceived that they were more trusted to talk with others within the community about sensitive health topics. In addition, Community Navigators highlighted the importance of having a badge indicating their role in the intervention and emphasized that receiving a stipend for their time furthered their sense of legitimacy and validated the work they performed to assist their communities.

Connections built with other Community Navigators

Community Navigators shared that being connected to the other Community Navigators across gender identities, sexual orientations and racial/ethnic identities increased a sense of belonging, resource sharing, creativity and problem solving. Community Navigators expressed that monthly meetings with the other Community Navigators and intervention coordinators were a time and place for growth because they brought together diverse experiences and perspectives and provided an opportunity to exchange ideas with one another, as well as discuss challenges and ways to overcome them. Navigators noted that it was valuable to know that other Community Navigators, including those with different gender identities, sexual orientations and/or racial/ethnic identities than their own, had found themselves in similar situations and to learn from one another. Thus, these conversations fostered an environment where Community Navigators felt welcome to express themselves and their ideas about promoting health within their communities through engagement and education. While each Community Navigator carried out helping activities independently with their social network members, the Community Navigators also worked as a team by providing social support to one another. These connections took place both in meetings and more informally in one-on-one interactions outside of meetings (including via social media) and continued after implementation ended.

Sustaining intervention aspects long-term

Community Navigators reported hoping to continue their roles to improve sexual health and social determinants within their communities after intervention implementation officially ended. Many expressed a conviction to share the knowledge they had gained through the intervention, stressing that their social network members and other community members continued to come to them for information. Community Navigators reported continuing to engage in helping activities such as condom distribution and disseminating information through social media because the need within their communities persisted and because it felt natural to continue to draw on the training and resources provided by *Impact Triad*.

Experiences implementing the *Impact Triad* intervention

Another set of themes related to Community Navigators' experiences implementing *Impact Triad*. These themes and selected quotations are presented in Table 4.

Table 4 Community Navigator experiences implementing the *Impact Triad* intervention (qualitative themes with select quotations from in-depth interviews)

Engaging community members
Community Navigators identified effective and innovative strategies to engage community members.
Con las actividades, al momento de juntarnos también teníamos comida, refrescos, botana. A veces aprovechábamos que había algún evento deportivo importante o programas como los Oscar. [With activities, when we gathered we would bring food, drinks, snacks. Sometimes we would take advantage of big sporting events or programs like the Oscars to get together.] (P1)

I posted fliers and updated social media about when there was going to be a testing event at Triad Health Project, and just let them know what events were happening and what we were doing. (P8)
Les di la prioridad a mis amigos a los que yo sabía que podrían estar tal vez en más riesgo, tuvieran menos información o que en conversaciones privadas me habían comentado que nunca se habían hecho la prueba del VIH. O algunos amigos que podrían no estar utilizando condones o otros métodos de prevención como PrEP. [I gave priority to my friends that I thought would be most at risk, or had less information, or in private conversations had mentioned to me that they had never been tested for HIV. Or other friends who might not be utilizing condoms or other prevention methods such as PrEP.] (P1)
Meeting prioritized needs
Community Navigators met social network members' prioritized needs by educating and supporting them to access and use local resources.
I helped my social network members to find some resources. For instance, most of the guys that I invited didn't know how to get condoms, how to get PrEP, or how to make an appointment to get STD testing. So that was very helpful, because every time they were asking me for more information and more condoms and things like that. (P15)
Some of my friends didn't even know where to get condoms or medications or that there are educational resources available. It's good to have associations with organizations like Triad Health Project or FaithAction [immigrant-serving community organization in Greensboro, NC]. It made them interested or willing to talk more about it. (P15)
Getting the information out there is important because I find that a lot of people in our communities don't even know that PrEP exists. (P6)
We learned about continuing one's education, employment opportunities, and stuff like that. I think it's better if [social network members] hear from someone they know about the resources available for them. (P9)
Building trust
Communicating about sexual health and addressing stigma and misinformation were difficult but overcome by building and nurturing trust.
It's important to support each other, help people with whatever they need, have open conversations, and build trust, because when you build trust you can change and improve people's lives. (P15)
Yes, my friends definitely feel more comfortable now talking to me about sex, how to put a condom on correctly, and things like that. It's not awkward anymore. They feel completely comfortable talking about STDs and PrEP and some of them asked me to go with them to get tested and asked me questions about resources, and I have been able to give out materials with the information they need. (P6)
When [social network members] start to trust you, it's easier to talk about anything, so trust made it easier. (P7)
Using social media

Use of social media complemented personal interaction to further facilitate dissemination of information and support.
I was making posts about testing events and testing sites, informing [my social network members] about different STIs and also about jobs, job fairs, and places that may be hiring. Also about housing. I used Facebook, Grindr, and Jack'd. (P4)
I think social media helped us so a lot more people could be reached. I don't go to many places so being on social media allows me to get out in the world when I am not going to do it in person. (P9)
I used social media to share what I think is important and to show information and resources that I come across. I like to say from my own experiences and other people's experiences too, what people go through, so there is someone seeing it. (P13)
I posted on my Snapchat stories, usually about where to go and get tested and about getting tested regularly. I posted on Facebook on my timeline and created stories. (P14)
Increasing awareness and knowledge
Intervention successes included increased awareness of prevalent health disparities and increased knowledge about sexual health and about resources to address disparities.
I told them all about the 'sexual pandemic' and then they became curious. I taught the group how to wear a condom correctly, and about how to be safe or get PrEP if they need it. (P14)
I educated [my social network] about some of the bad statistics that often face the LGBTQ communities such as HIV rates, unemployment, and things like that that affect us. (P6)
Challenges related to COVID-19
The COVID-19 pandemic hindered implementation and social media became the main strategy to reach social network members.
It was way more difficult to continue with the communication. I guess that's the blessing of social media but, yes, [the pandemic] changed everything. (P12)
It made it very different because I feel certain things worked better in person, like teaching people how to put a condom on. [The pandemic] made it a little difficult. (P6)
[The pandemic] definitely impacted how many people I was able to reach, because I spread the message in the halls at school just talking to people and it affected my ability to do that. (P14)

La pandemia afectó a mi red social mucho porque nos dejamos de reunir, y la intimidad y el espíritu del grupo de estar reunidos en un solo lugar, la convivencia y la confianza que se crea en el grupo, se pierde y se vuelve más individual. [The pandemic affected my social network a lot since we stopped getting together, and the intimacy and group spirit from gathering in the same place, the atmosphere of trust that was created within the group, this was all lost and everything became more individualized.] (P1)

Engaging community members

Community Navigators shared that they had identified effective and innovative strategies to engage community members. Community Navigators utilized various strategies to reach the individuals within their social networks. At the beginning of intervention implementation, Community Navigators focused their recruitment efforts on social network members who they perceived as being more vulnerable or at risk and as needing social support and emphasized to these social network members the benefits of their participation. Through this process, Community Navigators were able to reach a broad range of individuals, including engaging nongay identifying MSM by building trust to overcome concerns about having their sexual behaviours exposed. In addition, Community Navigators used creative ways to share information about health and local resources through social media posts and by making in-person events and gatherings as welcoming and comfortable environments as possible for social network members.

Meeting prioritized needs

Community Navigators reported meeting social network members' prioritized needs by educating and supporting them to access and use local resources. Community Navigators noted that their role provided them with insight that allowed them to pinpoint needs and priorities within the community. Community Navigators were able to effectively link social network members with the resources they needed, which helped social network members stay engaged in the intervention. Through communication and interaction with social network members, Community Navigators identified and filled knowledge gaps and guided social network members to resources that would meet their needs related to social determinants (e.g., employment and education) and sexual health (e.g., how to access condoms and PrEP and how to make an appointment for STI or HIV screening).

Building trust

Community Navigators emphasized that communicating about sexual health and addressing stigma and misinformation were difficult but overcome by building and nurturing trust. Many of the Community Navigators reported initial challenges in communication with social network members related to sensitive topics due to stigma and discomfort. Though Community Navigators had their social network members' trust more generally, they had to work on building further trust specifically around sexual health. To surpass this barrier, Community Navigators fostered an open and safe environment and worked to normalize talking about sexual health. To make social network members feel more comfortable, Community Navigators eased into conversations about sexual health, first starting with a topic that was less sensitive or emphasizing common experiences that the Community Navigators and social network members shared, including feelings of stigma. Social network members opened up to the Community Navigators and to one another over time, and conversations about sexual health happened more naturally over the course of intervention implementation as Community Navigators continued to promote a sense of community within their social networks.

Using social media

Community Navigators described using social media to complement personal interaction to further facilitate dissemination of information and support. Community Navigators reported that personal interactions were an important part of the intervention because they provided a more personable approach, particularly during the early stages of implementation and when recruiting individuals within their social networks to participate. Social media was also an essential tool utilized by Community Navigators throughout the course of the intervention in addition to these personal interactions, and after the COVID-19 pandemic began many Community Navigators adapted their interactions to rely solely on social media. Community Navigators posted on their social media accounts about resources or community events, and these posts benefited not only their immediate social network but also extended their reach to their social network members' larger networks and individuals whom they would not have interacted with otherwise. Expanding reach in these ways had a dual impact on Community Navigators' work by strengthening both their confidence as Community Navigators and their rapport within the community. In addition, Community Navigators noted other advantages of social media, including that it was a direct and instant form of communication between Community Navigators and social network members and, in some cases, made conversations about sensitive topics easier and more comfortable than in face-to-face communication.

Increasing awareness and knowledge

Community Navigators considered important intervention successes to include increased awareness of prevalent health disparities and increased knowledge about sexual health and about resources to address disparities. Community Navigators expressed that, among their social networks, there was a general lack of awareness of how young GBMSM and transgender women of colour, and particularly those in the US South, were disproportionately affected by STIs and HIV and by social determinants such as unemployment. When educating social network members about these disparities many Community Navigators reported witnessing a ‘light bulb’ moment where social network members internalized information about sexual health and local employment and education resources.

Community Navigators also successfully promoted broader definitions of sexual health, noting that within their communities sexual health had typically been equated with condom use or abstinence only. Through participating in *Impact Triad*, Community Navigators and members of their social networks came to understand sexual health as also involving screening for STIs and HIV, access to treatment and other prevention strategies such as PrEP.

Challenges related to COVID-19

Community Navigators reported that the COVID-19 pandemic hindered implementation and impacted Community Navigators in various ways. Because of the need for physical distancing, social media became the primary way the Community Navigators communicated with their social network members. Through social media Community Navigators maintained contact with their social networks, but Community Navigators expressed that members of their social networks felt isolated from one another. The connectedness Community Navigators fostered within their social network groups was reduced by the pandemic. In-person interactions were postponed, and the closeness and ‘spirit’ or morale of the groups were negatively affected. Furthermore, Community Navigators faced additional challenges teaching social network members about certain sexual health topics such as how to correctly use condoms when they could not carry out these activities in person. However, Community Navigators were resilient and adapted quickly to utilizing social media to continue to maintain relationships with participants and engage and teach them.

Lessons learned for future community navigation interventions

Finally, Community Navigators highlighted learnings to inform future similar interventions. Related themes and selected quotations are presented in Table 5.

Table 5 Lessons learned for future community navigation interventions (qualitative themes with select quotations from in-depth interviews)

Facilitating access to broader resources
The *Impact Triad* intervention should be expanded to facilitate access to a wider range of resources.
I think financial aspects, access to education, resources, transportation, and education are all important. (P5)
I think we should work more on housing, because I have learned about other programs that are available for housing. (P4)

Without access to food, it will affect your mental health and your physical health. It's a real problem, and then what you are willing to do to get food or what you are willing to do to get housing can affect your future. The problem is disproportionately bigger [among GBMSM and transgender women of color] than other populations. (P13)
Building additional skills
Community Navigator training should include activities to develop a more in-depth and diverse skillset, including more practice communicating about sensitive topics and promoting healthy coping strategies.
It would be good to learn more about how to talk or how to deal with people who use drugs and also how to talk to people who are newly diagnosed with HIV. (P11)
It would help to have training about having difficult conversations and how to be honest with people without making them feel that you are judging them. (P7)
I think we need to do more with trauma and mental health, honestly. A good way to help sexual health is to talk about trauma because we live in a society that bombards us with traumatic things all the time. (P13)
Increasing interactions among Community Navigators
Future iterations of *Impact Triad* should foster more interaction among Community Navigators.
Sería bueno poder reunirnos con navegantes por años de otros proyectos que nos ayuden a buscar nuevas oportunidades y conocimiento para los navegantes. [It would be good to get together with Navigators that have worked for years on other projects who could help us find new opportunities and provide advice for new Navigators.] (P2)
As Community Navigators, we had our meetings every month, and we talked about any challenges, shared our thoughts, and provided some feedback. It was really friendly, to be honest. But I would like to have more interactions with the whole group instead of just once a month. (P15)

Facilitating access to broader resources

Community Navigators reported that the *Impact Triad* intervention should be expanded to facilitate access to a wider range of resources. Throughout intervention implementation, Community Navigators heard from their social network members that it would be beneficial to receive information about additional resources related to social determinants of health, such as transportation, food insecurity, financial literacy, housing and substance use, which also contribute to sexual risk. Other barriers such as access to health care more generally, including insurance coverage and mental health care, were also suggested for inclusion as outcomes in future iterations of *Impact Triad*. Community Navigators noted that young GBMSM and transgender women of colour in different geographical areas experienced different needs, highlighting the importance of expanding the resources shared through the *Impact Triad* intervention. Community Navigators also suggested potentially adapting the intervention to reach broader communities, such as non-LGBTQ communities, or to meet the unique needs of specific subgroups such as bisexual men.

Building additional skills

Community Navigators indicated that future trainings should include activities to develop a more in-depth and diverse skillset, including more practice communicating about sensitive topics and promoting healthy coping strategies. Community Navigators shared that their training prepared them well for interacting within their social networks but that they experienced challenges in a few areas. Community Navigators felt they would benefit from more skill-building, particularly through role-playing, that emphasized effective ways to talk about sensitive and complex topics, such as how to engage in dialogue around a recent positive HIV diagnosis. Many Community Navigators noted that they found it difficult to have conversations about mental health, healthy coping strategies and how to overcome fear among social network members to access services when needed. They noted that more training would be useful to build their skills and increase their self-efficacy to have these difficult conversations.

Increasing interactions among Community Navigators

Community Navigators expressed that future iterations of *Impact Triad* should foster more interaction among Community Navigators. Community Navigators reported that they formed a support network with each other during the monthly meetings with one another, during which they shared ideas, challenges and successes. Many expressed a desire for a greater frequency of these meetings to discuss barriers and receive feedback. To further increase interaction and learning opportunities among Community Navigators, Community Navigators suggested that in future iterations of the intervention, former Community Navigators could be invited to mentor newer Community Navigators and share their experiences, thus expanding the network of trained Community Navigators and building on real-life experiences of serving as Community Navigators.

DISCUSSION

Analysis of in-depth interviews with the *Impact Triad* Community Navigators both highlighted the ways that intersecting factors magnify health risks among GBMSM and transgender women of colour and illustrated the strengths of the intervention in raising awareness and disseminating information pertaining to sexual health and social determinants of health. One of the greatest contributing factors to the success of the *Impact Triad* intervention was the Community Navigators' ability to forge and nurture connections and relationships with members of their social networks and then build on this foundation to discuss sensitive topics with confidence and increase awareness, knowledge and use of local resources among these social network members. This finding aligns with existing literature that points to the potential of community-based peer navigators to reach marginalized communities affected by health disparities and to connect community members to services because community-based peer navigators understand community needs and strengths on a personal level and reflect the ways that members of their communities communicate and interact.[29–31] The desire expressed by Community Navigators to sustain their work to continue to improve the overall health of their communities further illustrates the potential impact of the intervention.

Another factor playing a role in the success of *Impact Triad* was the Community Navigators' flexibility and ability to pivot and adapt to the unprecedented circumstances brought by the onset of the COVID-19 pandemic. Initially, Community Navigators' interactions with members of their social networks included in-person communication, which suddenly became no longer possible during the early phases of the pandemic. Adapting to the pandemic led to increased integration and utilization of social media platforms to cultivate and strengthen relationships. Shifting to an entirely virtual implementation of *Impact Triad* modified previous perceptions of how to connect with individuals and was consistent with documented changes in the ways in which networks provide social support in the context of COVID-19 with shifts away from physical interaction and toward the use of technology.[32] In some ways, these changes increased accessibility, allowing Community Navigators to overcome logistical barriers to engagement based on locations, schedules and work demands; these learnings could also greatly benefit research efforts in rural and other more isolated communities. However, restrictions due to COVID-19 did affect camaraderie among Community Navigators and members of their social networks. Community Navigators noted that there was a personal aspect of face-to-face contact that social media could not replicate. Nonetheless, our findings demonstrate that the versatility of social media is a major asset in conducting intervention research. Other intervention studies in recent years have reported making similar adaptations due to COVID-19,[33–35] and future studies should continue to explore ways to achieve the connection typically built and reinforced through in-person interaction in a virtual setting.

Interviews also identified implications for future iterations of the *Impact Triad* intervention to seek to further address and improve the quality of life among young GBMSM and transgender women of colour. A common thread across

Community Navigators' experiences involved the intricate ways that interconnected social determinants impact health. To have a more profound impact, future efforts can expand the domains of resources and education provided by Community Navigators by incorporating into Community Navigator training information on how to address needs such as housing, food insecurity, transportation and others, in addition to employment, education, social support and discrimination. Furthermore, leveraging the experiences of existing Community Navigators to provide mentorship and support to future Community Navigators can lead to an increasingly cohesive group dynamic and greater community capacity. For example, future Community Navigator trainings and monthly meetings could include components in which Community Navigators from previous cohorts are present to share their experiences, offer ‘tips’, and answer questions from new Community Navigators.

Limitations

It is possible that social desirability bias could have influenced Community Navigators' responses to interview items. Given that interview data were collected and analysed by members of our CBPR partnership that had developed and implemented the *Impact Triad* intervention, Community Navigators may have felt inclined to describe *Impact Triad* and their experiences with the intervention in ways that were positive and less comfortable sharing constructive criticism or difficulties they had faced as part of *Impact Triad*. We tried to mitigate this possibility and encourage open and honest responses by ensuring confidentiality and emphasizing that there were no right or wrong answers and that Community Navigators were experts in their own experiences. Furthermore, interviews were conducted by CBPR partnership members who were not directly responsible for training and supporting the Community Navigators.

CONCLUSIONS

These interviews identified important learnings about serving as Community Navigators and implementing *Impact Triad* and provided information to guide future efforts to address STI and HIV disparities and critical social determinants of health through community-based peer navigation. Further research is warranted given that community-based peer navigation remains an understudied yet promising approach to health promotion and disease prevention among communities and populations experiencing health disparities.

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CONFLICT OF INTEREST

The authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request.

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Corrigendum

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FULL TEXT

In the article by Fløtten et al.,[1] the affiliation of Kjersti J. Ø. Fløtten to the Institute for Clinical Medicine, Faculty of Medicine, University of Oslo, Oslo, Norway, should have been listed in addition to her affiliations to the Department of Integrated Care and Health Promotion, Akershus University Hospital, Lørenskog, Norway, and the International Network of Health Promotion Hospitals and Health Services (HPH), Taskforce on Children &Adolescents (HPHCA), Trento, Italy.

We apologize for the error.

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New Home, New You: A retrospective mixed-methods evaluation of a health-related behavioural intervention programme supporting social housing tenants

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ABSTRACT (ENGLISH)

Background

Social housing tenants are at greater risk of engaging in unhealthy behaviours than the general population. Housing association employees are in an ideal position to contribute positively to their tenants' health. ‘New Home, New You’ (NHNY) is a joint venture between a social housing association, a city council and a community healthcare provider in the South West of England. It was designed with the aim of improving the health and well-being of social housing tenants.

Objectives

The aim of this retrospective evaluation was to establish whether social housing tenants were benefiting from this health-related behavioural intervention in terms of their mental well-being and health-related behaviours.

Methods

This was a mixed-methods evaluation. The outcome evaluation was a secondary analysis of quantitative data collected during the NHNY project. The process of delivering and receiving the intervention was evaluated using qualitative, semi-structured interviews with housing officers and tenants who had participated in the programme. The development of the intervention was evaluated through a focus group and additional semistructured interviews with key stakeholders. Quantitative data were analysed using the Statistical Package for the Social Sciences. Qualitative interviews were analysed using thematic analysis.

Results

Six key stakeholders and a total of seven housing officers from several teams and seven tenants were interviewed. Of the 1016 tenants who were invited to participate in NHNY, 226 enroled in the programme. For participating tenants, the scope for health-related behaviour change was greatest in relation to eating and smoking. Small positive statistically significant changes in mental health were found between the 6- and 12-month mean score and between the baseline and the 12-month score.

Conclusions

The findings indicate that NHNY may have been beneficial for some participating tenants. Housing officers can have a significant role in promoting health messages and embedding behaviour change among their tenants. Although the programme was implemented as a service improvement rather than a controlled trial and focused on a particular intervention and geographical area, other housing associations may find this evaluation useful for considering whether to adopt some of the principles applied in NHNY in other settings.

Patient or Public Contribution

A social housing tenant representative provided input on the methodology and methods used to evaluate NHNY, as well as the information sheet.

FULL TEXT

INTRODUCTION

Health outcomes are strongly related to the conditions in which people are born, grow, live, work and age.[1] Evidence shows that people from low-income groups and those residing in deprived areas are more likely to experience poor health outcomes and have a lower life expectancy.[2,3] For example, in a large US study which analysed data on income and mortality from 1999 through 2014, the gap in life expectancy between the richest 1% and poorest 1% of individuals was 14.6 years for males and 10.1 years for females.[4] A social gradient in health can also be seen for health-related behaviours, with people of lower socioeconomic status (SES) being more likely to eat unhealthily, be sedentary and smoke, compared with those of higher SES.[2] Furthermore, although there has been a general increase in the prevalence of poor mental health, those of lower SES are disproportionately affected.[5]

Social housing tenants are more likely to have lower SES and to report financial hardship, therefore being made more likely due to the social determinants of health to engage in unhealthy behaviours than the general population.[6] It has been demonstrated that they have higher levels of chronic conditions and health risk factors, including smoking and sedentary lifestyles, compared with those living in other forms of housing.[7] In addition, they are 1.5 times more likely to experience poor mental health.[8] Both physical and mental health are important to overall well-being, while the effect of well-being on health is substantial and comparable to other risk factors such as an unhealthy diet.[9]

Given the potential benefits that health-related behaviour changes can bring to individual health and public health, it is no surprise that health research devotes considerable time to identifying effective behavioural interventions.[2] Considering the morbidity and mortality associated with chronic health conditions, the importance of addressing modifiable health-related behaviours in high-risk populations has been highlighted.[6] Promoting behaviour change among people at the lower spectrum of income is considered a major means of achieving a reduction in health inequalities.[2]

Although limited by its observational nature, previous research has identified an association between housing, the built environment and aspects of mental health and well-being. Housing associations can contribute substantially to the health of their tenants.[10] There are various ways in which housing can impact well-being, for example, through providing a property in good material condition, or through proving a trusted, central organization, a tenant can approach if they need help, advice or support. Assured tenancy with a social landlord can also give people new hope and the opportunity to focus on improving their health by changing their health-related behaviours. People in the housing provider workforce, especially those supporting people living in social housing, have opportunities to speak with new and current tenants about their health and well-being and support them to make behavioural changes.[11] Although, there is some evidence to suggest that gaining a tenancy in affordable and appropriate accommodation has a positive impact on health and well-being, this association is complex.[12] This may reflect both the limitations of current evidence and the complexity of the relationships between housing and well-being for vulnerable people with complex needs.[12] To date, there is little academic literature to evidence the impact of public health intervention in social housing settings[12] and a paucity of research specific to residents of social housing.[6] An evidence review on housing associations and housing interventions[10] concluded that there is a need for ‘evidence of the health and wellbeing impacts of housing associations' community-centred work to be produced and published’ to develop the evidence base in this field.

While there are many health-related behaviours that influence a person's health and well-being, including sleep and social contact, the ‘One You’ campaign[13] focuses on the four behaviours of exercise, diet, drinking (alcohol) and smoking, for which there is robust evidence that they influence health outcomes and people's well-being. Evidence demonstrates that people who are involved in decisions about their health are more satisfied with the services they receive and feel that the decisions made were the most appropriate for their circumstances.[14] Thus, involving individuals in decisions about their health can ensure that people make informed decisions about their behaviours. Plymouth Community Homes (PCH) is the largest social housing landlord in Plymouth city and provide homes to over 35,000 people in the area. ‘New Home, New You’ (NHNY) is a joint venture between PCH, Plymouth City Council and Livewell Southwest. The programme has been designed with the aim of improving the health and well-being of PCH customers. Applicants for PCH properties will have been waiting for a suitable home for any number of years or months. Often those waiting will have been living in properties that are unsuitable for them, such as being overcrowded, not adapted to their disabilities or even with no home at all. Being given a home for life that meets people's needs is a good opportunity for them to decide to make other positive changes to their lives. Recognizing that housing association employees have a unique role in engaging with people at this moment in their lives,[11] there was an opportunity to assist with the city-wide health and well-being agenda by ‘Making Every Contact Count (MECC)’[15,16] and training PCH staff to provide them with the tools to help people make those changes. Thus, the NHNY project was established. This approach aligns with the Ottawa Charter's principles of Health Promotion, namely strengthening community actions and developing personal skills.[17] The project supports new tenants and transferring tenants to consider and achieve self-set health and well-being goals, through interaction with suitably trained housing officers.

Aim

The aim of this evaluation was to establish whether social housing tenants were benefiting from a health-related behavioural intervention in terms of their mental well-being and health-related behaviours (moving, smoking, alcohol consumption and eating) and whether the intervention was delivered and received as intended. As this was a pilot intervention, the evaluation also investigated the wider strategic learning about developing the intervention.

Staff training

All housing officers and some managerial staff at PCH received training as part of the NHNY project. A 3-h training session was developed and delivered by Livewell Southwest, 6 months before the launch of the NHNY project. The content was specific to Plymouth city and the NHNY project and aimed to develop skills in motivational interviewing, health considerations and signposting to suitable support services and opportunities in the community and to national resources. All housing officers (including any new staff) received a refresher training session from Livewell Southwest, 18 months after the first training (12 months after the launch of NHNY). Internal staff training about the process and monitoring of NHNY was provided to all staff before the start of the NHNY project, and ongoing support with the administration was given to staff throughout.

Programme delivery and monitoring

Participation in the programme was entirely voluntary and offered at the time of sign-up to a standard tenancy (as distinct from housing with support, provided for older people with additional needs). If tenants agreed to take part in the NHNY project, they were offered a one-to-one conversation with their housing officer, who had received the health and well-being-related training.

Before this meeting, their current health and well-being were assessed using two questionnaires. The two data collection tools used were the Short Warwick-Edinburgh Mental Wellbeing Scale (SWEMWBS)[18–21] and the ‘How Are You’ (HAY) quiz.[22] The housing officer then explored with the new resident whether there was anything they would like to improve in relation to smoking, diet, physical activity, alcohol consumption and/or mental well-being and if so, provided the tenant with support to achieve it using goal setting and motivational interviewing. Thereafter, the housing officer suggested ways to improve health and well-being. They also signposted people to appropriate organizations for help and advice relating to their goals.

Tenants also received a welcome pack upon moving into their new homes. In addition to the useful household items included in PCH's usual welcome pack (pack of tea, coffee, tea cloth and other items), NHNY participants also received well-being-related items such as toothbrushes and toothpaste, vouchers for replacing smoking with e-cigarettes (where relevant), along with information about health and well-being services. In addition to this, participating tenants received a fortnightly, free, home-delivered bag of fresh vegetables for 3 months. PCH also provided free cookery sessions for those who wish to learn how to cook healthy meals using the vegetables provided as part of the project. The housing officer then met with the tenant at 1 month and after 6 months and then visited or telephoned them at 12 months for a catch-up and to complete the two questionnaires again.

METHODOLOGYTheoretical framework

The programme theory for NHNY draws upon the ‘behaviour system’ of ‘capability, opportunity and motivation’[23] and involves:

- (1) Education (provision of information to improve capability and motivation)
- (2) Persuasion (motivational interviewing to increase motivation)
- (3) Incentivisation (enhanced ‘Welcome Pack’ on moving in and fortnightly vegetable bag delivery to improve motivation)
- (4) Training (cooking lessons to improve capability)
- (5) Enablement (access to resources to improve capability, motivation and opportunity).

Design

The University of Plymouth was asked to evaluate the impact of the intervention retrospectively. This was a mixed methods study design, which included a quantitative and qualitative methodology.

The three aspects of the evaluation were:

Outcome evaluation

This was a retrospective evaluation, which analysed data collected during the NHNY project. All analysed data had already been collected and were provided by PCH, following the signing of an agreement policy between the University of Plymouth and PCH. The University was not involved in the choice of the NHNY data collection tools. Consent to participate and share data anonymously in any written report was gathered before the time of data collection by the housing officer. It was agreed that all data would be held securely and kept in line with the PCH data storage and protection policies and that they would be used to examine the impact of the project.

Participants in this data set signed up to NHNY between the start of the programme (23rd October 2017) and 31st December 2018. The sample consists of all NHNY participants who agreed to share their data.

The survey outcomes for health-related behaviours were gained through the completion of the HAY quiz.[22] The HAY quiz was chosen by NHNY stakeholders because it functions as an interactive tool to promote improvement in health-related behaviours for those completing it, as results are accompanied by specific advice. It was also publicly available and easily accessible. The HAY quiz enables the assessment of an individual's health-related behaviours with regard to eating, alcohol, smoking and exercising.

The SWEMWBS was chosen to assess the mental well-being of participants throughout the programme, and it is a validated tool for measuring the change in intervention studies.[18–21] The short version includes seven more rigorously tested statements each of which describes a positive state of well-being and is rated by participants on a 5-point Likert scale.

Process evaluation

The process of delivering and receiving the intervention was evaluated using qualitative, semi-structured interviews with housing officers and tenants who had participated in the programme. Interviews also touched upon reported behaviour changes, as part of housing officers' and tenants' experiences of the programme.

Housing officers were invited after a presentation by the evaluation team on a staff away day. Tenants were invited by their housing officer after completion of the programme, as the evaluation was conducted retrospectively and researchers did not have direct access to tenants' contact details. This was followed up by a telephone conversation with the interviewer (Z.A.) to answer tenants' questions and arrange the interview. Participants provided their informed consent in writing before participating.

The semistructured interviews were conducted face-to-face (Z.A.). Housing officers were interviewed in private meeting rooms on PCH premises during their working days. Tenants were interviewed in their own homes or at a suitable location on university premises, at their preference. Tenants were provided with a £30 voucher in recognition of their time spent participating in the evaluation. Topic guides were developed to guide the interviews with tenants and housing officers. All interviews were digitally recorded and transcribed by a staff member at PCH. Interviews took place between October 2019 and February 2020, immediately pre-COVID-19 pandemic.

Formative evaluation

The development and implementation of the intervention were evaluated through a focus group and additional semi-structured interviews with key stakeholders who had been involved from the outset. Potential participants were invited by email and were provided with a participant information sheet. Participants provided their informed consent in writing before participating. The focus group (Z.A. and M.P.) and telephone interviews (Z.A.) followed a qualitative approach, allowing participants to respond to general questions, based on a topic guide, in their own words. The focus group and interviews were digitally recorded using a digital audio recorder and transcribed by a staff member at PCH following the signature of a confidentiality agreement.

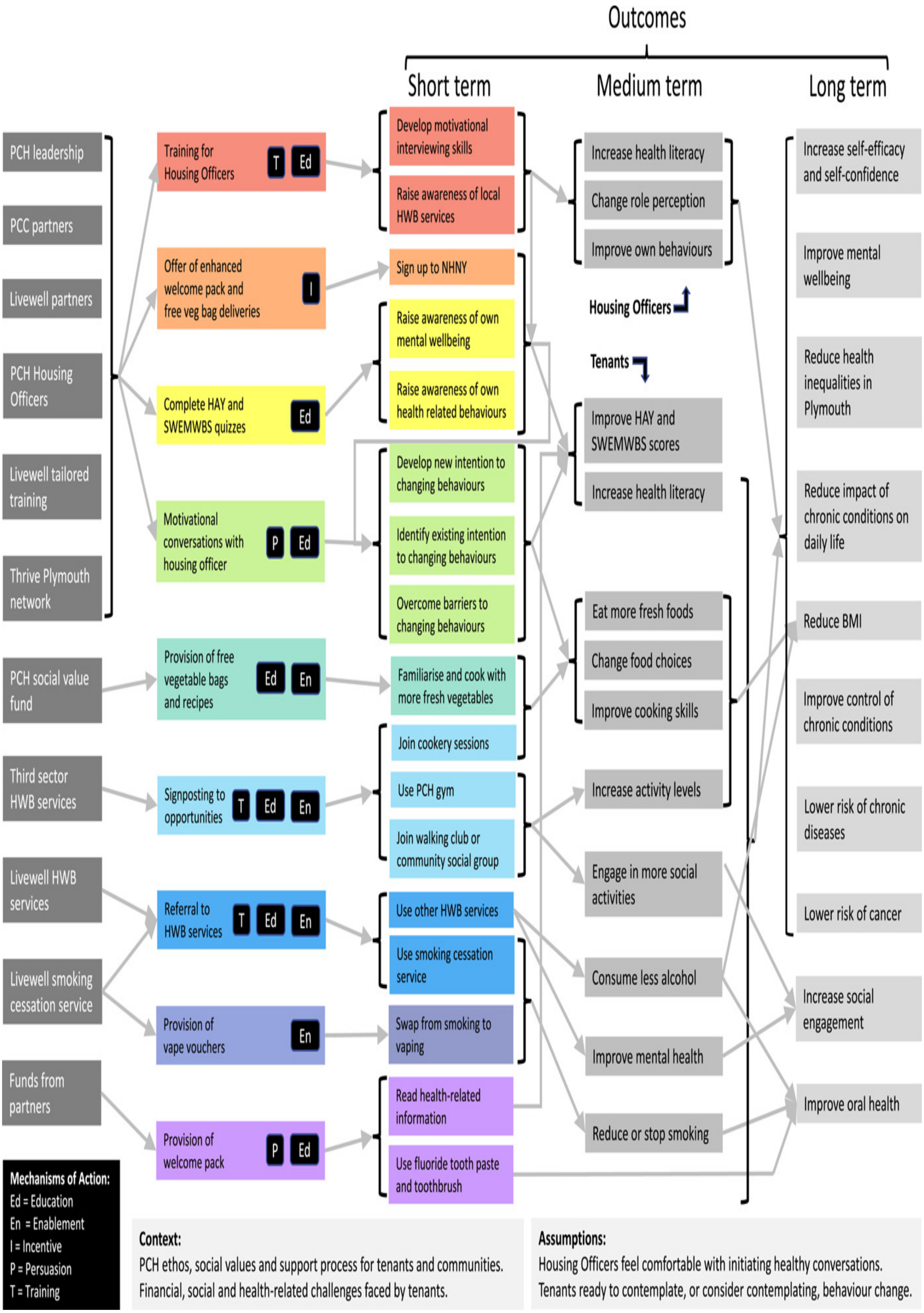
Data analysis

The Statistical Package for the Social Sciences (SPSS, version 24) was used for the analyses of quantitative data (M.P.). Continuous and categorical variables are presented as means (standard deviation [SD]) and frequencies (%), respectively. Listwise deletion was applied for missing information. The normality of data was tested before statistical tests were conducted. A *p*-value of less than .05 was considered to suggest statistical significance.

Interview transcripts were uploaded onto NVivo 12 software. Data were analysed using thematic analysis as described by Braun and Clarke.[24] Verbatim transcripts were coded line-by-line and key themes and subthemes were developed (Z.A.). Coding decisions and themes were interrogated (Z.A., M.P., J.S.) and adjusted part way through and at the end of the analysis, thus ensuring rigour in analysis. All transcripts were checked for accuracy against the original recordings and corrected where necessary by Z.A.

FINDINGS

A logic model showing all intended inputs, outputs and outcomes is shown below (Figure 1). This was initially developed during the evaluation planning process and was updated after taking into account information gathered from people who participated in the evaluation.



Context:

PCH ethos, social values and support process for tenants and communities.

Financial, social and health-related challenges faced by tenants.

Assumptions:

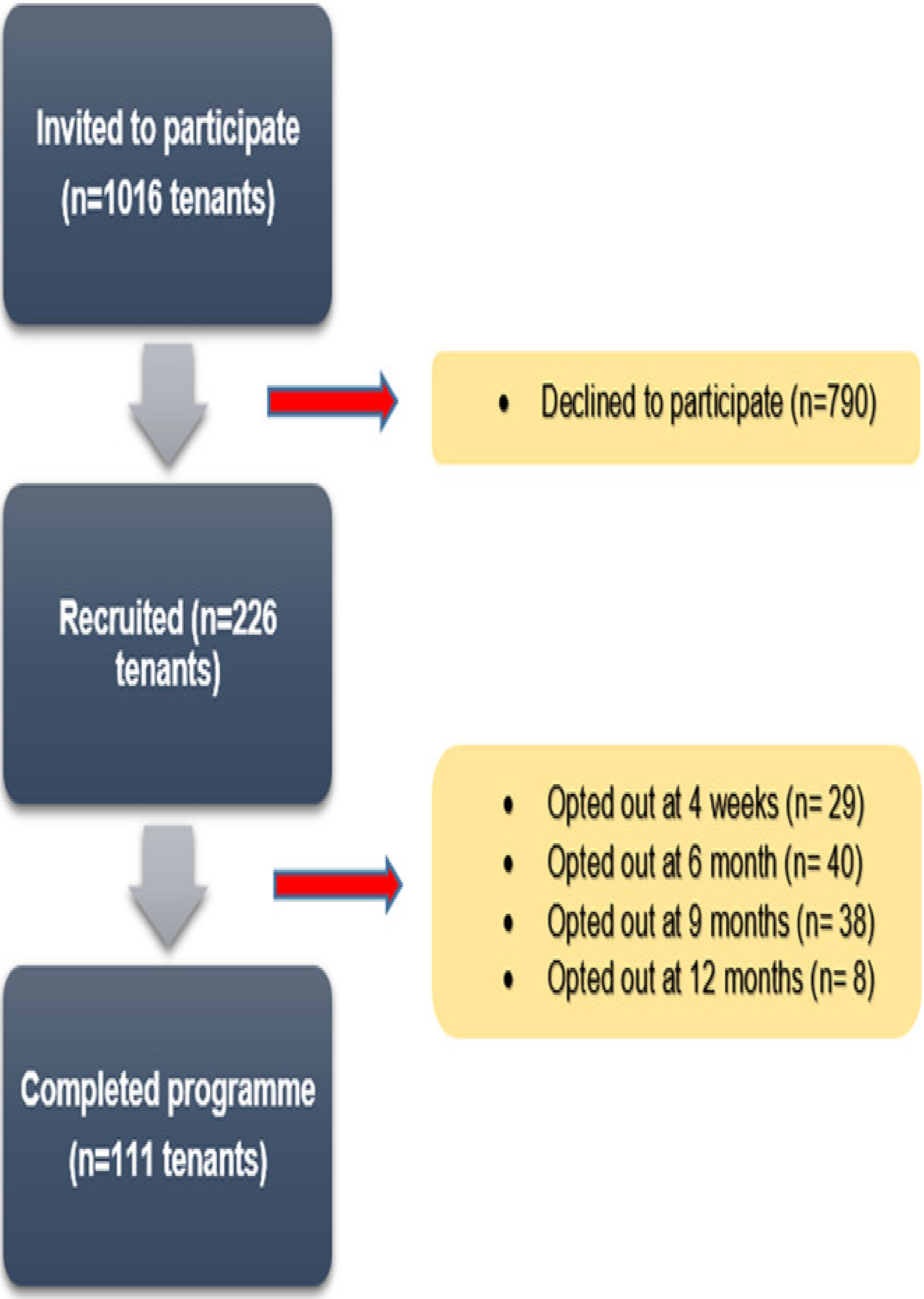
Housing Officers feel comfortable with initiating healthy conversations.

Tenants ready to contemplate, or consider contemplating, behaviour change.

Enlarge this image.

Outcome evaluation

Figure 2 below presents the flow of participants during NHNY.



Enlarge this image.

Of the 1016 individuals who were invited to participate in NHNY (these would have been named as main tenants), 226 signed up for the intervention, yielding a response rate of 22.2%. Of these, 111 completed the intervention. Therefore, the programme was limited in its reach, and the retention of participants was low.

Demographic characteristics

The baseline results below (Table 1) and those at Months 6 and 12 refer to the 111 participants who completed the NHNY programme. The column in grey shows the characteristics of those who dropped out (*n* = 115).

Table 1 Baseline characteristics of NHNY participants

[IMAGE OMITTED. SEE PDF.]

Abbreviations: NHNY, New Home, New You; PCH, Plymouth Community Homes; SD, standard deviation.

The average age of the participants who completed the NHNY programme was 36.6 years, which was similar to the mean age for all eligible tenants (38.9 years) and younger than the overall PCH tenant population (52.1 years). There were 35 (31.5%) males and 76 females (68.5%), which is similar to the split of gender among those who were invited (35.7% male: 63.2% female) and the overall PCH tenant population (39.3% male; 60.5% female and 0.2% unknown). The majority of the participants completing NHNY identified as White British (*n* = 103, 92.8%), reflecting the low ethnic diversity in the city (93%).[25] The percentage of White British among the overall PCH tenant population was 78.4%. The age and ethnicity of those recruited to the intervention may differ from the overall tenant population due to the inclusion of tenants of housing with support within the overall figures.

Comparing people who completed the programme with those who dropped out, there were no significant differences in proportions in terms of gender (χ^2 (1) = 0.145; $p = .703$), age (χ^2 (5) = 3.106; $p = .684$), ethnic group ($p = .609$—*exact test*) or annual household income (χ^2 (7) = 5.615; $p = .585$).

HAY quiz

Most frequently, participants who completed the programme scored category 2 at baseline, which is considered ‘low’. A Wilcoxon, two-tailed test comparing the baseline median score to the 6-month median score showed that there was a positive change which was statistically significant ($Z = -2.915$, $p = .004$). The same test indicated significantly statistically significant positive changes between the 6-month median score to the 12-month one ($Z = -3.663$; $p <.001$) and between the baseline and the 12-month score ($Z = -5.563$; $p <.001$).

SWEMWBS

Baseline SWEMWBS results were recorded for 103 of the NHNY participants who completed the programme, with an average (mean) transformed score of 23.28 (SD 4.23), which was not significantly different from the England average (mean 23.5; SD 3.90).[18]

A paired *t*-test (two-tailed) showed that there was an increase of .001 in the score at 6 months compared to the baseline, which was not statistically significant ($t_{95} = 0.002$; $p = .999$). The same test indicated statistically significant positive changes between the 6-month mean score (23.56) and the 12-month score (24.74) (mean difference: −1.18; $t_{98} = -2.451$; $p = .016$) and between the baseline (23.29) and the 12-month score (24.51) (mean difference: −1.22; $t_{99} = -3.425$; $p = .001$).

Process evaluation

A total of seven housing officers from several teams were interviewed. Seven tenants also participated in interviews, including four women and three men. Their ages ranged from early 30s to mid-60s and all identified as being of White British ethnicity. Only one tenant interviewed was raising a young child.

The following key themes were identified in the participants' interviews:

- (1)
  Challenges faced by tenants
- (2)
  Tenants' perceptions of the value of their tenancy
- (3)
  Housing officers' perceptions of their roles
- (4)
  Staff training
- (5)
  Invitation to participate
- (6)
  Implementation
- (7)
  Effects on tenants
- (8)
  Staff perceptions of programme delivery

This section summarizes information about these themes and includes selected quotes from a range of housing officers and tenants who were interviewed, for illustration.

Challenges faced by tenants

Housing officers and tenants reported that PCH tenants may be experiencing financial constraints, health issues, vulnerability or deprivation when they begin a tenancy. Housing officers mentioned financial constraints, health inequality, vulnerability and deprivation. Tenants often reported health issues.

We were pleased we had got in. We thought, we have got a roof over our heads, we are alright, we can manage. (Tenant)

I think the biggest impact has been Universal Credit really …people can get themselves into a lot of trouble with the rent really quickly, due to that. (Housing Officer)

Tenants' perceptions of the value of their tenancy when facing challenges

Tenants often described how beneficial their new PCH tenancy was for their physical and mental health and social circumstances, independent of their participation in NHNY.

…I just thought when I moved in here it would be like moving into anywhere else I moved into …but it's not like that …they look after you. It makes you feel safe…. (Tenant)

Tenants linked these positive impacts to the way they were treated as tenants, the assurance of a long-term tenancy and the suitability of their PCH home to their needs. PCH tenancies had released tenants from difficult circumstances in their previous accommodation, such as overcrowding or being confined to unsuitable accommodation due to impairments.

Older tenants, with experience renting privately, appreciated the security of feeling settled in a ‘forever home’

(Tenant).

…in private [rented accommodation] …you can't make it your own home. Where we can make this our own home. (Tenant)

Housing officers' perceptions of their roles

Recognizing the challenges and inequalities faced by their tenants, most housing officers described having a role to play in helping tenants to manage or overcome these issues, which were frequently linked to suitability for a tenancy or maintaining the tenancy. The social work of a housing officer had actively drawn several people to take up the role and was perceived to be valued by PCH. Some also talked about the communication skills they used to tackle sensitive issues with tenants in the course of their work.

…I work with social services, the schools, the police, we do a lot of joint working …There is a lot of social work, I will find, within the [housing officer] role…. (Housing Officer)

Some took a holistic approach to support tenants, whilst recognizing there were limits to the time that could be committed and that some tenants will not engage. Others felt that health and well-being was a different type of work, which they were not qualified to do and which would be intrusive upon all tenants.

…there's always been about level of thinking about someone's health, but this has moved beyond just that, this has moved to like, what can we do to really try and improve someone's general wellness and how they get on with their lives…. (Housing Officer)

Staff training

Some housing officers felt their colleagues talked more about mental health and about wanting to change their health-related behaviours since receiving NHNY training and some reported making changes. Others were already aware of their less healthy behaviours but had not been motivated by the training to make changes.

…it is an eye opener, to be perfectly honest it is, but have I actually done anything about it? …maybe, maybe somewhere in the back of my mind it's stuck with me. (Housing Officer)

Some housing officers had become aware of local health and well-being services to which they could signpost tenants, because of the training. Some felt the focus on motivational interviewing skills would help colleagues who were uncomfortable broaching personal issues with tenants; though some of the housing officers interviewed felt they personally had this ability already.

Invitation to participate

Housing officers reported that the sign-up process had changed because it was time-consuming and housing officers were uncomfortable with asking people questions about health-related behaviours. The process moved online, with tenants completing the health-related behaviour questions after the first meeting.

…it's morphed so that we send out this email asking people if they are interested and that has the links on it. (Housing Officer)

Housing officers suggested tenants may decline because they are not interested, or they have a lot to do linked to moving home. Housing officers indicated that completing online quizzes may be a barrier affecting capability, especially for older tenants and people who do not have digital devices.

Some housing officers felt there were no trends, demographically, in who joined the programme. One housing officer suggested that tenants' intentions to change their health-related behaviours may affect their motivation to participate.

It depends on someone's time of life as well, doesn't it, whether they are wanting to make a change or not, you know. (Housing Officer)

Tenants whose lives were being impacted by chronic health conditions described the programme as an opportunity to obtain support to improve their current health situation.

Housing officers reported that, for some tenants, access to vegetable bags helped to mitigate food poverty. Most tenants interviewed signed up for the free vegetable bag deliveries, with some signing up for this reason alone.

They just said you get a free pack of veg every couple of weeks, so I thought well, why not? Don't look a gift horse in the mouth. (Tenant)

Implementation

Some tenants and housing officers viewed the programme as providing people with support to make changes, without receiving criticism. Housing officers found that over several visits they could build up trust and create opportunities to go into more depth with health-related conversations.

However, housing officers reported that completing the quizzes and conversations was time-consuming alongside their usual procedures.

…when we do a pre-tenancy assessment it takes an hour at least, just to go through that, and then to add the New Home New You onto that as well, it was quite a lot…. (Housing Officer).

Staff were aware that the programme tailed off and tenants lost interest when the free vegetable bag deliveries stopped. Visits were sometimes replaced by phone calls, and it could be difficult to contact people for follow-up. They also felt that worry and poor mental health were barriers to participation.

…there are people that are very low mood and how you can support them is another thing again really. (Housing Officer)

Staff suggestions included offering the programme to other tenant groups, increasing staff interaction and tangible support and improving digital access. Tenants who completed the programme were generally happy with it, suggesting only that the vegetable bags could include more variety and extend for a longer time.

Effects on tenants

Most tenants reported greater awareness of the health impacts of their behaviours. Some had changed their perceptions regarding their health and their capability to influence it. Several reported that they had lost weight and some had noticed existing health conditions and pain had improved. Some felt their relatives had also benefited.

…because I'm cooking healthier, he's eating healthier and he's quite happy, he's lost a bit of weight as well and he's quite pleased about it…. (Tenant)

All tenants interviewed appreciated the vegetable bags, which had led to sustained healthier food purchases and meal preparation for those who felt their diet could be improved. A few participants felt the programme had no impact other than being a source of free vegetables.

Few tenants actively discussed mental health, though several described feeling more settled since moving home.

Tenants reported they had engaged in walking, exercise classes or using a gym, which they had maintained afterwards. Some reported a social or mental health benefit to getting outdoors however some were constrained by physical health or finances.

Housing officers reported that participants who smoked were often interested in stopping and they felt well prepared to assist with vouchers or referrals to smoking cessation services. Tenants who were interviewed and who smoked did not use these services. Tenants and housing officers reported that drinking a lot of alcohol was rare.

Staff perceptions of programme delivery

The housing officers who were most enthusiastic about the programme saw it as an opportunity to establish rapport with tenants, to understand their world and preempt problems. However, some housing officers felt hypocritical about promoting health-related behaviours that they did not follow themselves.

Housing officers often reported that there was a fine balance between offering to support tenants and intruding upon their personal lives.

…it also felt a little bit intrusive, because some of the questions are really personal, particularly the mental health sort of side of things… (Housing Officer)

Housing officers frequently reported feeling uncomfortable about the language used in the HAY quiz, to the extent that quizzes had become an online task. In addition, they found that response options did not always reflect tenants' behaviour changes.

Most housing officers valued feedback on the programme's impact on tenants, as this made them feel their efforts to engage tenants were worthwhile.

Formative evaluation

Six key stakeholders participated in describing the prerequisites which enabled them to develop and implement the NHNY programme and the challenges encountered in the early stages.

Connections, leadership and social values

The leaders described having a vision for improving health and reducing inequalities to increase people's life chances. They had the power and influence to initiate interventions, which enabled leaders to act upon organizational values of being ‘more than just bricks and mortar’ (Stakeholder). This was seen as pivotal to developing and investing in the programme.

…it was always felt that when someone moves into a new home …it was the perfect opportunity for them to make a change… (Stakeholder)

Challenges of defining and resourcing the programme

It took time to agree on the scope of the programme and the roles of different organizations, due to differing priorities around the programme's purpose, emphasis and development process. This frustrated some stakeholders.

…there was a process at the start where we had lots of meetings …about what we were going to do, was it ethical, would it do any harm and that seemed for us to drag on quite some months… (Stakeholder)

The cost of vegetable bag deliveries, welcome packs, housing officer's time and the time spent devising and delivering training, were absorbed by stakeholder organizations.

The success of utilizing the moment of change

NHNY was considered to enable staff to support tenants at a key moment of change, making the best use of existing staff contacts with new tenants. It was felt to have the potential to increase housing officers' capability and motivation to review their own health-related behaviours.

Staff perceptions and motivation

Initially, stakeholders found that staff perceptions of their role and motivation to deliver NHNY varied considerably. By seeking staff feedback and acting upon suggestions, stakeholders adapted NHNY to make it more acceptable and feasible for housing officers to deliver the programme.

Dedicating staff time

Allowing time for staff training, engaging tenants and delivering NHNY appeared to be critical to programme delivery but also at a premium, particularly for initial staff training.

…a lot of praise has to go to the [external training] team …who did put together a bespoke training programme for the housing officers. (Stakeholder)

Monitoring and recording participant data

Stakeholders had worked through numerous challenges around what to measure, how to record data and managing the administrative burden of the programme. Stakeholders decided to provide incentives (free vegetable bags) to overcome possible barriers to participation linked to the formal consent process.

DISCUSSION

Our findings indicate that although limited in its reach, NHNY may have been beneficial for some participating tenants, possibly creating an opportunity and enhancing capability for health-related behaviour change, particularly for participants who were already internally motivated to change. The current evaluation has identified a number of factors influencing participant engagement and acceptability as well as housing officer engagement. Housing associations may adopt some of the principles applied in NHNY in other settings to promote their tenants' well-being.

Our evaluation has shown statistically significant improvements in some health outcomes of some participating tenants. However, only 22.2% of tenants who were invited took the opportunity to participate in the NHNY programme. Whilst staff did not report any clear trends in their interview responses, some of the more engaged tenants who were interviewed reported that significant pre-existing physical health issues had contributed to their interest in participating in the programme. Of those who accepted the invitation, approximately 49% completed the programme at 12 months. This is in line with other research which demonstrated low retention rates with vulnerable adults.[26] The timing and complexity of the NHNY sign-up process appeared to be a barrier to participation for tenants. Proactive methods to contact participants (e.g., text messaging) could help improve follow-up rates.[26] More broadly, ‘a continuously dynamic process of monitoring intervention progress and tailoring strategies to particular circumstances’, has been recommended for maximizing retention.[26,27]

The mental well-being scores for NHNY participants compare well with the England average,[18] despite the recent adversity experienced by some tenants reported by tenants and housing officers. Overall, there appears to be potential to improve equity of access to support with mental health and health-related behaviour change by adapting the programme design to enable people who are experiencing poorer mental health or who are not ready to contemplate behaviour change to engage with support from housing officers. It, therefore, appears prudent as per the principles of MECC,[15,16] to embed conversations about health in general housing officer activity so that tenants have opportunities to engage without having to formally contemplate and consent. Given the complex interplay of factors affecting mental well-being, consideration should be given to the ways in which housing associations can connect with external agencies such as mental health support, the police and social services to provide such support to their tenants.[5]

Given that participants frequently commented on the value they placed on the vegetable bags, this was clearly a significant component of the NHNY intervention as a whole. Interview data suggested this may have been beneficial to participants on several levels, leading to sustained healthier food purchases and meal preparation for those who felt their diets could be improved. Considering that lack of access to affordable and nutritious foods are common barriers to healthy eating among tenants in public housing,[28,29] social housing providers should consider whether free vegetable deliveries could form part of an organization's existing support process for new tenants experiencing food poverty, without obligation to sign up to a programme.

The evaluation showed that there was a statistically significant improvement in the overall HAY quiz score over the course of the NHNY programme, for those who completed the programme. When viewing individual changes in HAY scores, an overall improvement in individual participants' scores was seen in 43.9% of participants. The finding that effects were not uniform across the participants is consistent with existing literature. The scope for health-related behaviour change was greater in relation to eating and smoking. Similarly, in a mixed-methods study involving public housing residents, smoking among residents was found to be extremely high, while only 22% and 29% of 88 participants reported consuming more than one serving of fruit or one serving of vegetables, respectively, per day.[6] Considering that human self-regulation draws on limited resources, interventions may be more effective when they focus on one behaviour change at a time,[2] ideally the one chosen by the tenants themselves.

There was also a statistically significant improvement in reported mental well-being among participants by the end of the NHNY intervention. The Warwick Medical School guidelines indicate that this change of 1.22 points is borderline clinically meaningful. Although this positive small change could well be attributed to NHNY, we cannot exclude the possibility that other factors contributed to this. Housing may facilitate positive changes in the mental health of permanent supported housing tenants.[30] However, in many cases, there is limited, or lack of evidence of the effect of housing interventions on well-being and effects are not necessarily uniform across groups of vulnerable people.[12] Some of the participants interviewed after the NHNY programme described these positive feelings in relation to the sense of security and suitability that their overall PCH tenancy provided. Owing to the design of the intervention, it is not possible to ascertain the extent to which the NHNY programme contributed to the positive outcome changes, independent/in addition to this sense of security. A project adopting a randomized controlled design would provide the strongest evidence that improvement in outcomes was attributed to the intervention, although it is recognized this design is difficult to achieve for community-based health promotion interventions.

Owing to being landlords, housing associations could reach people that public health interventions may not otherwise reach.[10] As was the case with NHNY, housing officers can make important contributions to the well-being of their tenants and may be involved in the delivery of services.[10] PCH staff varied in their willingness to engage in conversations about health with tenants. This appeared to relate to their varying perceptions about the boundaries of the housing officer role and feeling a sense of intrusion if asking about tenants' behaviours before establishing rapport and trust, which is in line with perceived challenges among other housing associations' staff.[11] A cultural shift, whereby staff understand the benefits of such programmes, is therefore particularly important in similar contexts.[11] For housing officers to reach their full ‘public health potential’, effective training (e.g., understanding of public health, and behaviour change techniques) must be provided.[11]

The administration of the programme, which some housing officers perceived to be onerous, and the language used in the HAY quiz, which made them feel uncomfortable, appeared to de-motivate housing officers, even when they were committed to the programme. Housing officers welcomed the simplification of the administrative process and separation of housing officers from the quiz completion process, which may reduce barriers to staff engagement. This highlights the need to involve housing officers in programme development, implementation and monitoring. Such an approach can also enhance a feeling of ownership and also help staff understand a project.[11] The organizational commitment to NHNY appeared to assist some housing officers to put a greater focus on supporting tenants' and communities' well-being and opportunities, beyond tenancy maintenance. In fact, organizational readiness is considered an important element for the implementation of MECC,[11] while strong leadership is imperative in bringing about a cultural change.

Limitations

The generalizability of results to highly urban areas with diverse ethnic groups may be limited. The generalizability of the study is further attenuated by the low response rate and subsequently small sample size. Owing to the design of the study, it is not possible to ascertain through the evaluation the extent to which the NHNY programme contributed to these changes. Although having a control group would have allowed us to control for confounding factors, this was not possible due to the retrospective nature of the evaluation and the design of the intervention. In addition, tenants' responses to the HAY quiz and SWEMWBS, are at risk of bias, as common with other surveys, participants may have been reluctant to be open with housing officers (or with themselves) about their health-related behaviours. The HAY quiz is not a validated tool for measuring behaviour change. Therefore, it is difficult to infer what change is clinically meaningful. Furthermore, as also reported by housing officers, the HAY quiz scoring system did not capture all changes that tenants had made to their food consumption or smoking activity, such as switching to vaping. Lastly, participating tenants were recruited to take part in interviews by their housing officers after completing the programme. It is likely that housing officers would have been more successful in recruiting tenants with whom they had built up a good rapport during the programme. It is also possible that tenants who took part in the interviews were those who were motivated to change their behaviour and may have provided relatively positive feedback. Therefore, the tenants who were interviewed may not be representative of all participating tenants, and the transferability of the findings to those who dropped out or who were not successful in changing their behaviour may be limited. Housing officers who were interviewed may also have had a more positive perception of the programme than those who did not respond to the invitation, and this may have been reflected in their feedback.

CONCLUSIONS

The findings indicate that NHNY may have been beneficial for some participating tenants. During the pilot phase, many tenants did not take up the opportunity or had limited engagement in the programme. The NHNY pilot has generated key learning points and presents an important opportunity to show how the intervention could be improved. The programme has already been adapted to overcome some barriers to participation and to extend tenants' opportunities to engage in health-related conversations with housing officers and to access other support services. In particular, to improve uptake, administrative processes have been streamlined. The programme has become more goal-focused, with tenants defining their chosen goals before receiving free vegetable bags, making it easier for staff to provide appropriate signposting. In addition, measures have been put in place to monitor and evaluate programme outcomes and the effectiveness of the adaptations.

Housing officers can undoubtedly have a significant role in promoting health messages and embedding behaviour changes among their tenants. Although this study focused on a particular intervention and geographical area, other housing associations may find this evaluation useful for considering whether to adopt some of the principles applied in NHNY in other settings. Improved interventions that target health-related behaviour change among social housing tenants are warranted.

AUTHOR CONTRIBUTIONS

Martha Paisi, Zoe Allen, and Jill Shawe made substantial contributions to the evaluation conception and design, revised it critically for important intellectual content, gave final approval of the version to be published, and agreed to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. Martha Paisi and Zoe Allen drafted the manuscript and contributed to acquisition and analysis of data.

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CONFLICT OF INTEREST

The authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request.

ETHICS STATEMENT

A confidentiality agreement was put in place between the University of Plymouth and PCH, before data sharing. Both the formative and process evaluation and outcome evaluation were approved by the Faculty of Health and Human Sciences Research Ethics Committee of the University of Plymouth (ref: 18/19-1147 and ref: 13/14–240, respectively).

DETAILS

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Stories for Change: The impact of Public Narrative on the co-production process

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ABSTRACT (ENGLISH)

Introduction

Involving service users in health service design and delivery is considered important to improve the quality of healthcare because it ensures that the delivery of healthcare is adapted to the needs of the users. Co-production is a process used to involve service users, but multiple papers have highlighted the need for the mechanisms and values guiding co-production to be more clearly stated. The aim of this paper was to evaluate the mechanisms and values that guided the co-production approach of the Stories for Change project, which used Public Narrative as part of the co-design process to create change in National Health Service maternity services.

Methods

This study was conducted using a rapid feedback evaluation approach. Semistructured interviews ($n = 16$) were the main source of data, six of which were maternity service users, with observations (5 h) and documentary analysis also carried out in parallel. RREAL sheets were used for data analysis to organize data based on key topics of interest.

Results

This study identified three broad mechanisms and values underpinning the co-production approach: creating an open and safe space to share ideas, learning how to tell stories using Public Narrative and having service providers who play a key role in strengthening the health system listen to stories compelling them to action. This study identified the main areas for improvement of the Stories for Change project related to recruitment, the inclusion of participants, the co-design process, the Skills Session and the Learning Event.

Conclusion

Our study provided a deeper understanding of the co-production approach that addresses the need to uncover the mechanism and values underlying co-production and co-design approaches. This study expands on the literature pertaining to the influence of storytelling in creating meaningful change in health care. We propose a co-design methodology that uses Public Narrative as a model for service user engagement to help inform future healthcare development processes.

Patient or Public Contribution

The experiences and perceptions of maternity service users and health professionals informed this evaluation. The project organizers were involved in the manuscript preparation stage by providing feedback, and service users wrote a commentary on the project from the lived experience perspective.

FULL TEXT

INTRODUCTION

In recent years, there have been multiple calls for action to improve maternity services in the National Health Service (NHS) in England. Various formal independent reviews have been carried out in different regions of England with families of babies who had faced negative experiences in NHS care. These highlighted the need to improve the safety of maternity care in the NHS and put forward recommendations and actions needed to reduce the deaths of newborns and improve quality care. These reports included the Ockenden Report for Shrewsbury and Telford,[1] the Thematic Review of incidents relating to Maternity Care at the Nottingham University Hospitals NHS Trust,[2] and ‘Reading the Signals’ for East Kent.[3] The main concerns raised in these reports included the disparities in outcomes for women of Black, Asian and Mixed ethnic groups and those who live in more deprived areas,[4] which led to a call for urgent action in a letter published in the *BMJ*.[5]

Alongside this, there has been increasing importance placed on and interest in the involvement and engagement of service users in health and social care.[6,7] Involving service users is considered an important approach to improving the quality of health care because it ensures that the delivery of health care is adapted to the needs of the users, incorporating patient choice in the design of services and fostering a shared decision-making model that embodies democratic principles and accountability.[8,9]

Langley et al.[10] identified co-production as a method by which to integrate social factors into policy and practice. The diversity in evidence that can be incorporated into co-production processes, and the relevance to patients, help connect evidence to policy and practice.[6,10] Reviews of the literature on co-production have pointed to the lack of clear definitions related to what constitutes co-production, with diverse and overlapping ideas over how this process is carried out.[6,10] This includes the interpretation of ‘co-production’ as an umbrella term for the ‘co’- terminologies, such as ‘co-design’, compared to its interpretation as a distinct term.[6] The term ‘co-design’ has also been highlighted to have unclear definitions,[7] illustrating the unclear processes of service user involvement in health and social care. Rather than the necessity for clearly delineated definitions, these papers highlight the need for increased clarity over the values, principles and mechanisms that underpin the practices of co-production and co-design.[6,7,10]

Our paper sets out to build on the literature as we respond to this need through our evaluation of a co-production project called Stories for Change, which sought to deliver a learning initiative based on co-production, using co-design methodology to involve service users in the redesign of maternity health services. Through our evaluation, the values and mechanisms that underpinned the co-production approach are made explicit, and, consequently transparent and replicable.

Stories for Change background

In response to the inequity experienced in maternity care in the NHS, NHS England and NHS Improvement (NHSEI) developed a perinatal equity strategy.[4] In October 2021, as part of this strategy, the South East region proposed a project with the objective of delivering a learning initiative on improvements in NHS maternity services based on co-production with maternity service users, creating the Stories for Change project.[11] The evaluation team adopted the term ‘maternity service users’ as this was the term chosen by NHSEI. The project included a co-design group, and project organizers, who were made up of a service user representative, a subject matter expert on the Public Narrative approach, and a project administrator. The requirement for partaking in the co-design group was having given birth in the NHS in the South East region within the last 2 years (2020–2022). All experiences were welcomed, mothers did not have to have faced explicit wrongdoing, but needed to have the desire to suggest improvements based on their experiences.

The Stories for Change project aimed to co-produce solutions for the improvement of maternity services in the NHS. Based on the previous literature highlighting the complexity and variety in definitions, we have decided to make explicit the relevant definitions that guided this project's co-production process. In this context, co-production refers to the involvement of patients as partners in the improvement of the quality of health care, by being included in its design and production.[8] Co-design refers to a specific activity within co-production, which entails their involvement in the process of designing a service, relating both to its functionality and to how it is experienced by service users.[8]

This project used the Double Diamond approach as a co-design process. This approach embodies four steps to co-design: discovering the service users' problems, defining the challenges in different ways, developing different answers to the problems and finally, delivering the solutions.[12] This process incorporated the use of Public Narrative to develop the leadership resources of the co-design group and to identify the service users' problems and develop solutions. This is a leadership practice put forward by Marshall Ganz that aims to use public stories to call others to action.[13–15] The approach uses a method of storytelling based on values that draw on our emotional resources to galvanize others into creating change,[13,15] and the project aimed to use this approach to empower the leadership of the co-design group to create change. The Public Narrative framework links a story of self to a story of us, which connects the speaker's personal values to the values of the audience, and leads to a story of now, which grounds these values into a call for action.[13,14] Another aspect of the co-design process was the agreed norms and aims that were discussed and developed between the co-design group and project organizers at every session, and included: active listening, camera on when possible, being present, respecting their diversity and everyone having a voice. The learning initiative centred the co-design, content and delivery around developing the participants' skills, confidence and capability to use the Public Narrative approach to effectively lever change.

The project started in January 2022 and consisted of three co-design group sessions, where the co-design group members and project organizers would meet with the aim of developing the co-design group members' stories. A Public Narrative skills session was held, where the co-design group members developed their Public Narrative skills, and other maternity service users were invited to learn the methodology. The session created the opportunity for additional service users to join the project. The learning initiative culminated in the Learning Event in April 2022, where the co-design group members presented their stories to NHS staff, to use their own lived experiences and voices to convey what could be improved in NHS maternity services. The co-design group members decided who would be invited to this event, and some of the NHS staff invited were drawn from the same services as the co-design group members, to directly express improvements they felt could be made.

METHODSStudy design

This study was designed as a rapid feedback evaluation with interviews as the main source of data.[16] Using a rapid feedback evaluation allowed us to continually collect data and provide feedback within a limited timeframe.[17] We undertook iterative processes of data collection and analysis, carrying out the two stages in parallel to share emerging findings and to inform subsequent data collection.[18]

The rapid evaluation was guided by the following questions:

- 1. What was the programme theory guiding the Stories for Change project? What were the expected outcomes?
- 2. What were the factors acting as barriers and facilitators to the implementation of co-design and co-production?
- 3. What was the perceived impact of the Learning Event on creating change?
- 4. How was the Public Narrative approach perceived by the different groups involved?
- 5. What recommendations did participants have for future similar projects?

Sampling and recruitment

Purposive sampling was used to recruit a sample of 16 participants (see Table 1 for the sampling strategy). We aimed to interview the different stakeholders involved in the co-production process, this included the co-design group members, the project organizers and NHS staff. We interviewed six co-design group members, who were maternity service users learning the Public Narrative approach to present their stories to the NHS staff. Six were interviewed before the Learning Event, and five participated in a second interview after the Learning Event. We interviewed the three project organizers, whose role was to teach the Public Narrative approach, run the co-design group sessions and guide the delivery of the Learning Event, with two acting as co-facilitators and one as the project administrator. We also interviewed seven NHS staff who attended the Learning Event, to whom the stories were delivered and with whom the co-design group members hoped to co-produce solutions. The NHS staff recruited were all stakeholders in maternity services, which included managers, consultants, those involved in their Local Maternity and Neonatal System (LMNS), the project's co-sponsors and Maternity Voices Partnership (MVP) co-chairs. An MVP is an NHS working group made of stakeholders in maternity services, including service users, providers and commissioners that work together to improve maternity care.

Table 1 Sampling strategy for interviews.

	First interview	Second interview	Total interviews
Co-design group members	6	5	11
Project organizers	3	0	3
NHS staff	0	7	7

	9	12	21

The project organizers worked with the evaluation team to recruit co-design group members and NHS staff, by promoting the evaluation and inviting them to take part. Those who showed interest in participating were then contacted by one of the researchers (S. M.) via email. Participant information sheets and consent forms were shared, and interviews were arranged.

Data collection

In-depth, semistructured interviews were conducted with the project organizers, co-design group members and NHS staff over Zoom and Microsoft Teams from 14 February 2022 to 23 May 2022. The interviews were conducted by two researchers on the evaluation team working in parallel, using an interview topic guide, which was created based on the research questions guiding the study. The interviews were audio recorded, and interview data were entered into RREAL sheets, which helped to organize and summarize data in real-time, based on key topics of interest.[19] Organizing data in this way allowed the researchers to maintain consistency throughout data collection and identify the key findings of the study in a short amount of time.[16]

The interviews were conducted in two stages to capture the process of co-designing and co-producing the programme from beginning to end. The first stage was carried out with nine participants, who were co-design group members and project organizers. The interviews captured their role in the project, their expectations, their experience of co-design and co-production and any barriers and facilitators to the co-design and co-production process. The second stage of interviews was carried out with 12 participants, which included co-design group members interviewed in the first stage to capture their experience of the process over time, and NHS staff who attended the Learning Event. These captured their views of the Learning Event and what change they hoped the project would lead to. A total of 21 interviews were carried out. The interviews in both stages captured participants' views of the Public Narrative approach and recommendations for future similar projects.

Documentary analysis and observations were carried out to ensure triangulation using different data collection methods. This allowed us to include information that was not brought up in the interviews, address any knowledge gaps and analyse the intended objectives of the programme compared to what is happening in practice.[16]

Data analysis

We used an inductive-deductive approach to data analysis, with the research questions guiding the analysis, whilst the evaluation team was sensitive to new themes arising from the interviews. Following rapid qualitative data analysis approaches, RREAL sheets were used to identify recurrent topics across study participants and enabled emerging findings to be shared in real-time during the evaluation.[20,21]

The RREAL sheet is a working document that enables the synthesis of data as data collection is ongoing.[21] It allowed for the identification of gaps during data collection, and collaborative interpretation as regular team meetings were held to discuss findings.[21] The RREAL sheets also helped identify when we reached data saturation, as it made clear when no new information was arising. The RREAL sheets were subsequently used to guide in-depth analysis, after which the evaluation team discussed which quotes to use to illustrate the key findings. The manuscript was then shared with the project organizers, who reviewed the results and provided feedback.

Through using different RREAL sheets, research teams can make comparisons between different factors influencing the results, and thus for this evaluation, we used different RREAL sheets for each population and study stage (see Supporting Information: Appendix A for an example).[21]

RESULTS

The key findings related to the mechanisms and values underpinning the co-production approach and recommendations for future projects and to create change. The mechanisms and values included: the collaborative and inclusive design of the project, power sharing, facilitation of an open and safe space, the use of stories and being called to action. Co-design group members were also invited to share their lived experience perspective of taking part in this co-production initiative.

Collaborative and inclusive design of the project

The co-production approach was considered a process that was both collaborative and inclusive. Participants felt that collaboration was central to the co-production process. Co-design group members reflected on how the language used about collaboration was present in every part of the project.

The language that is used is all about collaborating, cooperating with one another, being part of this journey with us. (Co-design group member)

Another component of the collaborative design of the project that was beneficial to the co-design process was the diversity of skill sets amongst the project organizers. Participants believed that their mixed skillset created a balanced and dynamic group.

[Project organiser] is chairing the meeting, she's coming at it from a different way where she had to get the objectives covered and outcomes […] [project organiser] is there in the background providing additional support to all of us, but [project organiser] is on the other side providing emotional support […] quite a good dynamic of people. (Co-design group member)

The project was thought to be inclusive due to the recruitment process. Participants agreed that the project had recruited a diverse group of mothers, mentioning factors including ethnicity, background and job roles.

Remuneration enabled participation for some of those involved. Some mothers stated that it influenced them to get involved at the start, and participants expressed that it showed that their expertise and time were being appreciated. Participants had the option to get their childcare costs covered, and some decided to take that opportunity as they felt that it allowed them to be more present in the meetings. Remuneration also helped participants prioritize the project when they were busy.

I think it helps as well with commitment of time because everybody is so busy it helps to prioritise it. (Co-design group member)

Project organizers acknowledged that people who the healthcare service had not worked with before would not have been aware of the project as recruitment was done through MVPs and social media accounts, which reached people with whom they were already connected. At the time of recruiting, they were also not aware that providing remuneration would interact with state benefits, which potentially led to the exclusion of participants from more deprived backgrounds.

We really want to make sure that we remunerate people for their time, which is great in practice but in reality […] the majority of the time if someone is in receipt of any state benefit they are unable to claim this money. (Project organizer)

Some co-design group members voiced that systemic factors could have acted as potential barriers to certain populations participating. These included mothers who did not speak English well, who did not have adequate digital access to participate, and who did not have childcare options.

I wonder if actually some of the people who suffer the most from less effective care, less compassionate care, are people who don't speak very good English, people who don't have access to being able to join online groups, people who don't have the childcare support to be able to come join in those online groups. (Co-design group member)

Power sharing

Co-design group members expressed that power-sharing was the norm. This meant that project organizers were genuinely collaborating with co-design group members and not only using them as representation. This made co-design group members feel that their expertise was being valued. They compared this project to others, where they felt that including the voice of service users was purely tokenistic.

This [group] is important because the power balance is right, whereas other groups and feedback requests and things like that you often feel like actually it's not real, you're going to say your thing but then the people running it are going to do their thing anyway, whereas this feels more like everyone's voice is important and they actually want to hear what needs to change rather than they've already decided. (Co-design group member)

There were differing ideas in relation to the structure of the design. The project organizers had intentionally structured the project without an agenda to work with the co-design group members to shape the content. At the beginning of the project, whilst some saw the nonhierarchical nature of the co-design process as a facilitator to creating a collaborative space, others saw it as impractical. Participants acknowledged that this was due to shifting the power back to the service users, but some thought that it led to a lack of defined roles, and, therefore, practical steps, to guide the process. However, as the project progressed and participants reached the stage of planning for the Learning Event, all the co-design group members agreed that the session added the practical component necessary to ground their stories and ideas.

The practicalities of the co-design process are still a bit vague […] If everyone is brought to the same level, as in if the facilitators are really putting the power back to the group, that's a really nice idea but sometimes you need someone to guide it […] there needs to be some sort of concrete steps. (Co-design group member)

Facilitation of an open and safe space

One of the most important mechanisms to the co-production process was the open and safe space. This was a key theme that arose from interviews with the co-design group members, which led them to feel empowered. The trusting relationship that was built in co-design group sessions meant that they openly shared their ideas and stories. It enabled them to honestly address the changes that need to be made in the maternity system.

It's very open and free flowing, which makes me feel that there is a lot of trust there, we've been trusted to share what we think is best. (Co-design group member)

NHS staff reflected on how the Learning Event provided a space for service users and service providers to communicate with each other directly, which prompted open conversations.

I think it worked very well because I think that in the subgroups, at least the group I was in, I felt that it really helped other professionals to open up and to connect with those people who had told the stories. (NHS staff)

The project organizers were considered important influences on the creation of this space. The co-design group members were supported by the project organizers, who made the mothers feel that they could express sensitive issues such as racial bias and consent. Co-design group members considered that the project organizers helped to facilitate conversation in a productive, yet not decisive way.

You can even express anything that is sensitive, like racial bias, like lack of consent […] but you forever feel supported while discussing it which is great. (Co-design group member)

They were very good at prompting discussion without necessarily leading it. (Co-design group member)

The use of stories

The use of stories was a key component of the Stories for Change project, and central to the co-production approach of this project. The project organizers designed the project with the hope that through the service users constructing their stories using the Public Narrative approach, they would feel empowered and would influence the NHS staff to make a change.

To use the stories for change framework to build the skills, confidence and capability of current and recent service users to advocate on their own behalf for changes that matter for them. (Project organizer)

They also felt heard due to the common factor bringing them together in this project, which was that they all wanted improvements in the NHS maternity services. Telling their stories was deemed a silver lining of their birthing experiences, as speaking about it and producing change is something constructive that came from a bad experience.

I was thinking ‘my situation is quite different to everybody else’ […] but having ten faces on the screen and gradually get to realise that we are all in the same boat, it adds quite a lot of power. Everyone is gradually becoming quite empowered and that is going to have more impact going forward, for the people who want to listen and also for us. (Co-design group member)

A key theme that emerged was the impact of emotion in the stories. Project organizers highlighted the importance of the use of emotion in stories to create the motion needed to lead to change and action. NHS staff found that there was an emotional resonance that other methods of sharing information do not have.

There's an emotional resonance there, that immediately you can connect with those people that you can't do by other means, you know a poster wouldn't have the same impact. (NHS staff)

Called to action

The NHS staff felt that the use of stories was a method that was conducive to learning and to creating change when staff were overworked.

The ‘stories’ bit in the title made it right […] to actually have an event that was focused in that with other key stakeholders there was really important. (NHS staff)

For NHS staff who attended the Learning Event, listening to the stories made them feel called to action. They perceived the stories to be attention-grabbing, as this approach meant stories were told in a way that kept listeners alert. They acknowledged that listening to what could be improved in the services from maternity service users themselves was essential to giving the power back to the service users and making the appropriate changes.

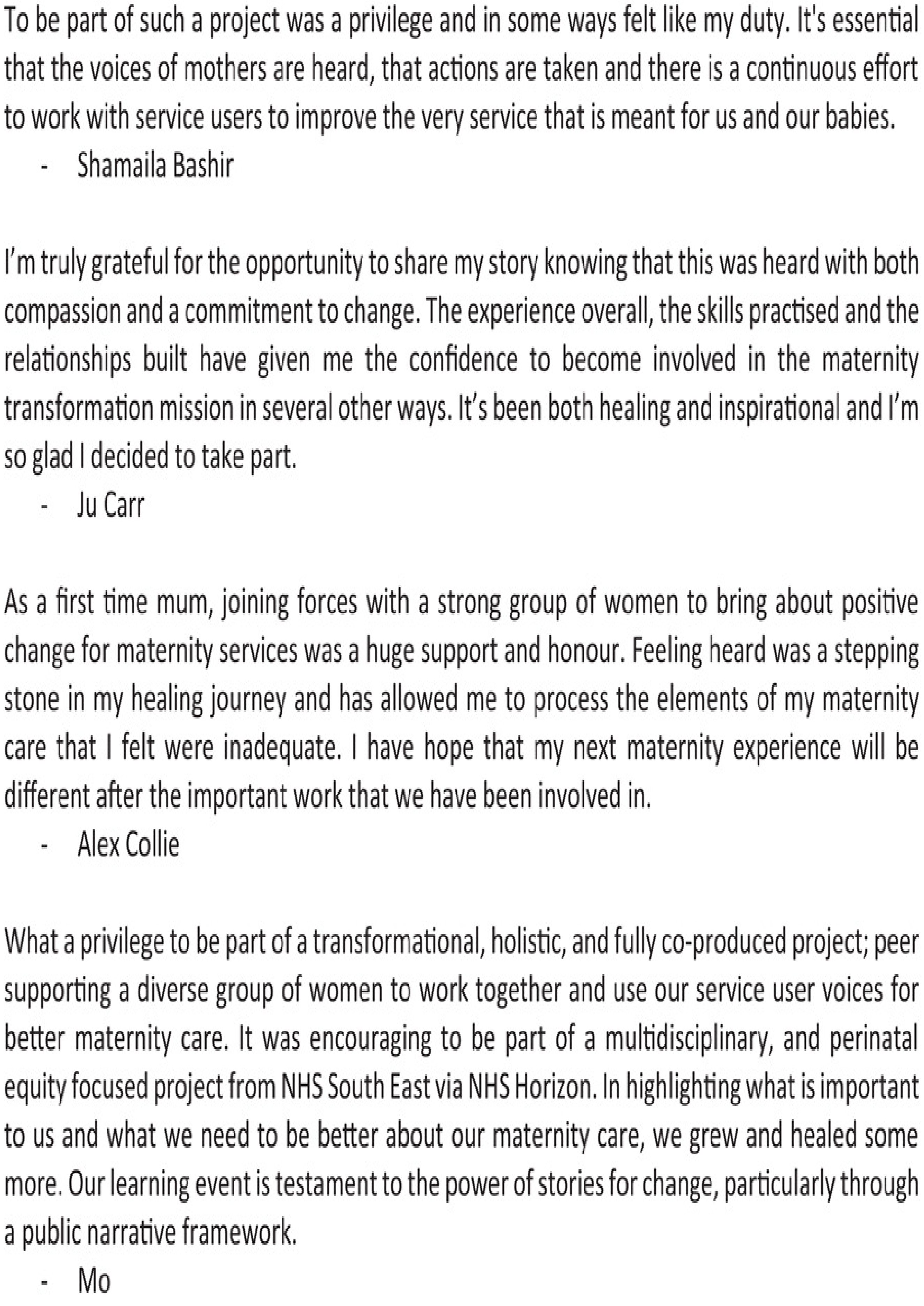
That really important point of power sharing and giving that power back to the mothers and not thinking that you're the one that's sitting there with all the power. (NHS staff)

It made me feel that I am now in a position, a hot seat basically, to make a change with them. (NHS staff)

NHS staff and co-design group members reflected on changes that started taking place as a result of the Learning Event. For example, NHS stakeholders stated that they would include information from the event in their revalidation reports, MVP co-chairs recognized the importance of getting to know different cultures in their region, and maternity service users were involved in the development of training programmes in maternity services. The event also spurred a drive to use stories as a form of data, and participants stated that the Stories for Change project would lead to more similar events. On a personal level, NHS staff expressed how it would change the way that they interacted with their patients, as it opened their eyes to the importance of taking a more compassionate approach.

Lived experience perspective

Service users who took part in the Stories for Change project were also invited to contribute by sharing their lived experience perspective on the project. Figure 1 presents a commentary on the service users' experiences of taking part in this co-production project.



To be part of such a project was a privilege and in some ways felt like my duty. It's essential that the voices of mothers are heard, that actions are taken and there is a continuous effort to work with service users to improve the very service that is meant for us and our babies.

- Shamaila Bashir

I'm truly grateful for the opportunity to share my story knowing that this was heard with both compassion and a commitment to change. The experience overall, the skills practised and the relationships built have given me the confidence to become involved in the maternity transformation mission in several other ways. It's been both healing and inspirational and I'm so glad I decided to take part.

- Ju Carr

As a first time mum, joining forces with a strong group of women to bring about positive change for maternity services was a huge support and honour. Feeling heard was a stepping stone in my healing journey and has allowed me to process the elements of my maternity care that I felt were inadequate. I have hope that my next maternity experience will be different after the important work that we have been involved in.

- Alex Collie

What a privilege to be part of a transformational, holistic, and fully co-produced project; peer supporting a diverse group of women to work together and use our service user voices for better maternity care. It was encouraging to be part of a multidisciplinary, and perinatal equity focused project from NHS South East via NHS Horizon. In highlighting what is important to us and what we need to be better about our maternity care, we grew and healed some more. Our learning event is testament to the power of stories for change, particularly through a public narrative framework.

- Mo

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Recommendations for future projects and to create change made by participants

During interviews, participants put forward recommendations for future similar projects targeted at specific stakeholders to ensure change in healthcare processes (Table 2). These include improving the recruitment and inclusion methods in the design process, factoring in diversity and types of remuneration offered to participants and improving the co-design process by sharing their stories with each other earlier to facilitate improvement.

Table 2 Recommendations for future similar projects and to create change (made by participants).

Theme	Respondent group	Recommendations
Recruitment process	Project organizer	Long-term networking is needed to connect with a variety of communities and established groups in these communities. Acknowledge the importance of building new relationships. Outreach workers (separate to community midwives) could follow up with people who register their pregnancy to give them the opportunity to be included in the project from the start. Ask what type of remuneration participants would want as state benefits may interact with remuneration and participants may not want to just receive money.
	Co-design member	Local community outreach to include participants that would not hear about the event through social media or MVPs.
Inclusion in the co-design group	Project organizer and co-design member	Could include partners in future projects. Include more people from deprived backgrounds through improving recruitment and considering how to remunerate participants.
	Co-design member	Could include someone that represents midwives as they would bring a different, more frontline perspective.
	NHS staff	Include mothers of the global majority who have English as their second language.
Co-design process	Project organizer	Include daily themes (such as daily tips) in the WhatsApp group to create more connection.

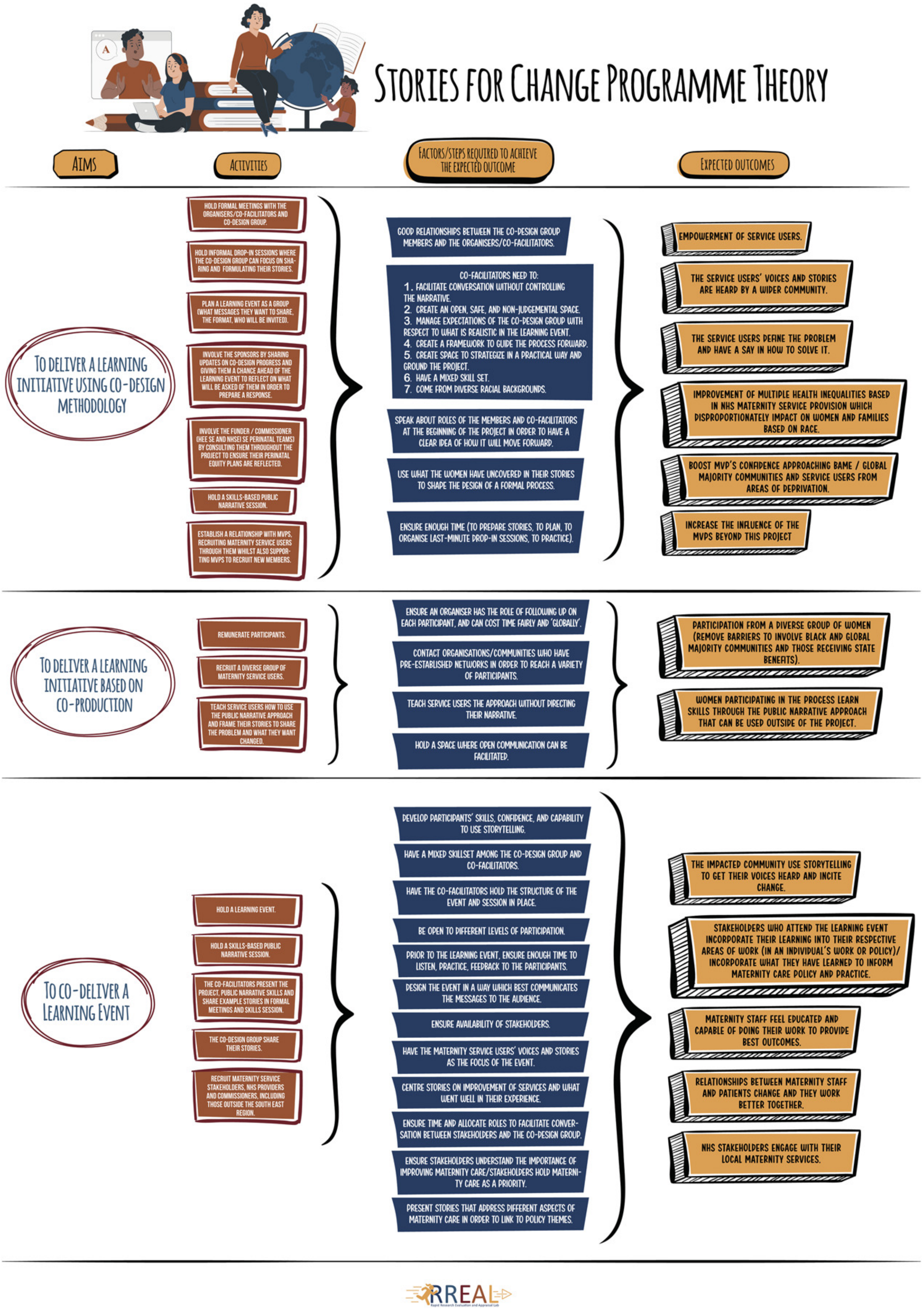
	Co-design member	At the start of the process, speak about which aspects of the project will be co-designed and which will be co-facilitated to mark the roles more clearly. Spend a whole day together to accelerate the process, either in person or online. Share their stories with each other earlier to better understand each other's experiences, what improvements they want to call for, and how to present them. Increase the number of sessions that they hold from the beginning to have more allocated time to create and share their stories and plan the event. Create an agenda before the meeting so they know in advance what would be discussed, and this would help them make a plan around what to do with their children during the event. Create a shared document to compile the notes that the co-design members make reflecting on the process of the project.
Skills Session	Project organizer and co-design member	Provide more time to set up and practice the Public Narrative approach.
	Co-design member	Provide more time in breakout rooms. Make the Skills Session more of a workshop with role play and practice as the co-design members had already learnt the skills. Invite participants earlier so that more people could attend.

Learning Event	Co-design member	Encourage service user involvement through more social media use and a wider range of platforms (including Instagram). Hold more rehearsals. Implement a structure where everyone is prompted to talk in breakout rooms to encourage everyone to be present and to hear everyone's views. Encourage attendance from representatives from each co-design member's local trust. Spend more time in the breakout rooms focussing on the pledges.
	NHS staff	Invite more consultants. Invite health visitors. Share more stories. Hold in person Learning Events.
In order to promote change: Accountability to commit to creating change	Co-design member	Create a timeline and action plan to track the changes. Use social media to follow up on the decision-makers' promises and keep the movement going. Hold a meeting every 3–6 months to get an update from the decision-makers. Involve service users at the local level to ensure that they are listened to, that they can monitor the change, and keep the importance of the human experience on the service providers' minds. Hold a ‘you said, we did’ board for the professionals to show commitment to their actions.
	NHS staff	Hold an annual event to review progress. Constantly review progress through a health equity lens.

In order to promote change: Reaching a wider audience	NHS staff	Bring the project to every region and NHS hospital, or to individual provider trusts. Use eye-catching advertisement to publicize the project. NHS employers should allow their staff time to attend and hear the stories. Learning Events should be held at board meetings as a mandatory event to reach people who do not want to listen to these stories.

Abbreviation: MVP, Maternity Voices Partnership. **Programme theory**

Based on interviews, observations and documentary analysis, we developed a programme theory for the project, which can be found in Figure 2. This programme theory serves as a useful framework on which to base future projects that wish to use the Stories for Change co-production model.



STORIES FOR CHANGE PROGRAMME THEORY

AIMS | ACTIVITIES | FACTORS/STEPS REQUIRED TO ACHIEVE THE EXPECTED OUTCOME | EXPECTED OUTCOMES

TO DELIVER A LEARNING INITIATIVE USING CO-DESIGN METHODOLOGY

Activities:
- HOLD FORMAL MEETINGS WITH THE ORGANISERS/CO-FACILITATORS AND CO-DESIGN GROUP.
- HOLD INFORMAL DROP-IN SESSIONS WHERE THE CO-DESIGN GROUP CAN FOCUS ON SHARING AND FORMULATING THEIR STORIES.
- PLAN A LEARNING EVENT AS A GROUP (WHAT MESSAGES THEY WANT TO SHARE, THE FORMAT, WHO WILL BE INVITED).
- INVOLVE THE SPONSORS BY SHARING UPDATES ON CO-DESIGN PROGRESS AND GIVING THEM A CHANCE AHEAD OF THE LEARNING EVENT TO REFLECT ON WHAT WILL BE ASKED OF THEM IN ORDER TO PREPARE A RESPONSE.
- INVOLVE THE FUNDER / COMMISSIONER (HEE SE AND NHSEI SE PERINATAL TEAMS) BY CONSULTING THEM THROUGHOUT THE PROJECT TO ENSURE THEIR PERINATAL EQUITY PLANS ARE REFLECTED.
- HOLD A SKILLS-BASED PUBLIC NARRATIVE SESSION.
- ESTABLISH A RELATIONSHIP WITH MVPS, RECRUITING MATERNITY SERVICE USERS THROUGH THEM WHILST ALSO SUPPORTING MVPS TO RECRUIT NEW MEMBERS.

Factors/steps:
- GOOD RELATIONSHIPS BETWEEN THE CO-DESIGN GROUP MEMBERS AND THE ORGANISERS/CO-FACILITATORS.
- CO-FACILITATORS NEED TO:
  1. FACILITATE CONVERSATION WITHOUT CONTROLLING THE NARRATIVE.
  2. CREATE AN OPEN, SAFE, AND NON-JUDGEMENTAL SPACE.
  3. MANAGE EXPECTATIONS OF THE CO-DESIGN GROUP WITH RESPECT TO WHAT IS REALISTIC IN THE LEARNING EVENT.
  4. CREATE A FRAMEWORK TO GUIDE THE PROCESS FORWARD.
  5. CREATE SPACE TO STRATEGIZE IN A PRACTICAL WAY AND GROUND THE PROJECT.
  6. HAVE A MIXED SKILL SET.
  7. COME FROM DIVERSE RACIAL BACKGROUNDS.
- SPEAK ABOUT ROLES OF THE MEMBERS AND CO-FACILITATORS AT THE BEGINNING OF THE PROJECT IN ORDER TO HAVE A CLEAR IDEA OF HOW IT WILL MOVE FORWARD.
- USE WHAT THE WOMEN HAVE UNCOVERED IN THEIR STORIES TO SHAPE THE DESIGN OF A FORMAL PROCESS.
- ENSURE ENOUGH TIME (TO PREPARE STORIES, TO PLAN, TO ORGANISE LAST-MINUTE DROP-IN SESSIONS, TO PRACTICE).

Expected outcomes:
- EMPOWERMENT OF SERVICE USERS.
- THE SERVICE USERS' VOICES AND STORIES ARE HEARD BY A WIDER COMMUNITY.
- THE SERVICE USERS DEFINE THE PROBLEM AND HAVE A SAY IN HOW TO SOLVE IT.
- IMPROVEMENT OF MULTIPLE HEALTH INEQUALITIES BASED IN NHS MATERNITY SERVICE PROVISION WHICH DISPROPORTIONATELY IMPACT ON WOMEN AND FAMILIES BASED ON RACE.
- BOOST MVP'S CONFIDENCE APPROACHING BAME / GLOBAL MAJORITY COMMUNITIES AND SERVICE USERS FROM AREAS OF DEPRIVATION.
- INCREASE THE INFLUENCE OF THE MVPS BEYOND THIS PROJECT

TO DELIVER A LEARNING INITIATIVE BASED ON CO-PRODUCTION

Activities:
- REMUNERATE PARTICIPANTS.
- RECRUIT A DIVERSE GROUP OF MATERNITY SERVICE USERS.
- TEACH SERVICE USERS HOW TO USE THE PUBLIC NARRATIVE APPROACH AND FRAME THEIR STORIES TO SHARE THE PROBLEM AND WHAT THEY WANT CHANGED.

Factors/steps:
- ENSURE AN ORGANISER HAS THE ROLE OF FOLLOWING UP ON EACH PARTICIPANT, AND CAN COST TIME FAIRLY AND 'GLOBALLY'.
- CONTACT ORGANISATIONS/COMMUNITIES WHO HAVE PRE-ESTABLISHED NETWORKS IN ORDER TO REACH A VARIETY OF PARTICIPANTS.
- TEACH SERVICE USERS THE APPROACH WITHOUT DIRECTING THEIR NARRATIVE.
- HOLD A SPACE WHERE OPEN COMMUNICATION CAN BE FACILITATED.

Expected outcomes:
- PARTICIPATION FROM A DIVERSE GROUP OF WOMEN (REMOVE BARRIERS TO INVOLVE BLACK AND GLOBAL MAJORITY COMMUNITIES AND THOSE RECEIVING STATE BENEFITS).
- WOMEN PARTICIPATING IN THE PROCESS LEARN SKILLS THROUGH THE PUBLIC NARRATIVE APPROACH THAT CAN BE USED OUTSIDE OF THE PROJECT.

TO CO-DELIVER A LEARNING EVENT

Activities:
- HOLD A LEARNING EVENT.
- HOLD A SKILLS-BASED PUBLIC NARRATIVE SESSION.
- THE CO-FACILITATORS PRESENT THE PROJECT, PUBLIC NARRATIVE SKILLS AND SHARE EXAMPLE STORIES IN FORMAL MEETINGS AND SKILLS SESSION.
- THE CO-DESIGN GROUP SHARE THEIR STORIES.
- RECRUIT MATERNITY SERVICE STAKEHOLDERS, NHS PROVIDERS AND COMMISSIONERS, INCLUDING THOSE OUTSIDE THE SOUTH EAST REGION.

Factors/steps:
- DEVELOP PARTICIPANTS' SKILLS, CONFIDENCE, AND CAPABILITY TO USE STORYTELLING.
- HAVE A MIXED SKILLSET AMONG THE CO-DESIGN GROUP AND CO-FACILITATORS.
- HAVE THE CO-FACILITATORS HOLD THE STRUCTURE OF THE EVENT AND SESSION IN PLACE.
- BE OPEN TO DIFFERENT LEVELS OF PARTICIPATION.
- PRIOR TO THE LEARNING EVENT, ENSURE ENOUGH TIME TO LISTEN, PRACTICE, FEEDBACK TO THE PARTICIPANTS.
- DESIGN THE EVENT IN A WAY WHICH BEST COMMUNICATES THE MESSAGES TO THE AUDIENCE.
- ENSURE AVAILABILITY OF STAKEHOLDERS.
- HAVE THE MATERNITY SERVICE USERS' VOICES AND STORIES AS THE FOCUS OF THE EVENT.
- CENTRE STORIES ON IMPROVEMENT OF SERVICES AND WHAT WENT WELL IN THEIR EXPERIENCE.
- ENSURE TIME AND ALLOCATE ROLES TO FACILITATE CONVERSATION BETWEEN STAKEHOLDERS AND THE CO-DESIGN GROUP.
- ENSURE STAKEHOLDERS UNDERSTAND THE IMPORTANCE OF IMPROVING MATERNITY CARE/STAKEHOLDERS HOLD MATERNITY CARE AS A PRIORITY.
- PRESENT STORIES THAT ADDRESS DIFFERENT ASPECTS OF MATERNITY CARE IN ORDER TO LINK TO POLICY THEMES.

Expected outcomes:
- THE IMPACTED COMMUNITY USE STORYTELLING TO GET THEIR VOICES HEARD AND INCITE CHANGE.
- STAKEHOLDERS WHO ATTEND THE LEARNING EVENT INCORPORATE THEIR LEARNING INTO THEIR RESPECTIVE AREAS OF WORK (IN AN INDIVIDUAL'S WORK OR POLICY)/ INCORPORATE WHAT THEY HAVE LEARNED TO INFORM MATERNITY CARE POLICY AND PRACTICE.
- MATERNITY STAFF FEEL EDUCATED AND CAPABLE OF DOING THEIR WORK TO PROVIDE BEST OUTCOMES.
- RELATIONSHIPS BETWEEN MATERNITY STAFF AND PATIENTS CHANGE AND THEY WORK BETTER TOGETHER.
- NHS STAKEHOLDERS ENGAGE WITH THEIR LOCAL MATERNITY SERVICES.

RREAL
Rapid Research Evaluation and Appraisal Lab

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DISCUSSION

Our study provided a deeper understanding of the mechanisms and values underlying a co-production approach that aimed to involve service users in the redesign of maternity services. This study identified three broad factors in facilitating co-production: creating an open and safe space to share ideas, learning how to tell stories using the Public Narrative approach and having service providers who play a key role in strengthening the health system listen to stories compelling them to action. These different factors provide insight into the influence of storytelling in creating meaningful change in healthcare, which could help inform future healthcare development processes.

The Public Narrative approach is a leadership practice that aims to express values through storytelling, and ultimately call people to action.[13] This evaluation has shown that this method was conducive to contributing to co-design in health care. Co-design aims to value experiential knowledge,[22] which is what the Public Narrative approach enables. The Double Diamond approach incorporates four steps, three of which are the discovery, defining and developing of the problems and solutions.[22] Through storytelling, the co-design group members were able to centre their values and experiences in the process of uncovering the problems and solutions in NHS maternity services. The broader context within which the Public Narrative was used fostered the creation of the service users as leaders, empowering them to use their stories and expertise to call for change. The findings highlighted that they were able to create an environment in which to openly uncover their stories and propose solutions, highlighting how the relationship between the approach and the values guiding the sessions enabled the use of the Double Diamond model to act as the co-design process in this project.

Historically, the power balance between the professional and service user has been measured based on the product as opposed to the process, centring the importance around who the final decision-maker is rather than the process of interaction between the different actors that leads to the production of solutions, as was the case with the Arnstein's iconic ladder of citizen participation.[23,24] The project's co-production approach entailed co-design methodology, which was shown to enable interaction between different actors, as maternity service users were incorporated into the process of redesigning services.[8] Our study found that this co-production approach is dependent on the nonhierarchical nature of the design process, which promoted power sharing. The Stories for Change project is an exemplar of the mixing of different forms of knowledge that represent an authentic co-production experience.[25]

Our study found that having the space to share ideas allows for the recognition of everyone's expertise. Project organizers intentionally designed the project without content in order for the co-design group members to have control over designing solutions. The facilitators and co-design group members created a safe environment in which NHS staff could learn and have direct conversations with service users and consequently were open to changing their points of view and ways of working.

Findings in relation to other studies

Our results highlighted the importance of examining the process of producing solutions, as participants emphasized the importance of a collaborative and inclusive project and an open and safe space in empowering the service users and calling the NHS staff to create change. This aligns with previous studies identifying that successful co-production was the consequence of having a ‘space to talk’ and a ‘space to change’, referring to the spaces that recognize everyone's expertise and allow for individuals to change their views.[25] In our study, these spaces allowed for diverse forms of knowledge to interact on equitable grounds, which is considered important in dismantling the hierarchy of knowledge between professionals and service users in co-production initiatives.[24–26] The Public Narrative approach gives unique tools by which to challenge the knowledge hierarchy that maintains health professionals on a superior level, as it gives service users a technique by which to share their expertise of lived experience and connect with their audience.

Another study that supports our study was conducted by Abma et al.[27] who promote a user involvement model called the Dialogue Model. Their study highlighted the importance of having a ‘safe environment’ as a foundation on which to develop the patients' voices, which consequently acknowledges power differentials and creates the space necessary for dialogue with professionals.[27] Our study showed that in the context of maternity services, a safe environment is necessary to create a trusting relationship between stakeholders, as this fosters the honest sharing of stories and open dialogue between NHS staff and co-design group members.

Strengths and limitations

To our knowledge, the present study is the first to use Public Narrative as a tool for service user engagement in health care. This is a strength as it makes explicit the mechanisms and values used to guide co-design, which allows for future replication of this approach. Second, given the time limitations associated with developing healthcare policies, our rapid methodology allows for scientifically rigorous methods of ensuring that patients' voices are systematically identified and incorporated into healthcare policies in a timely manner.

The findings of this study should be considered in relation to its limitations. The data collection started when the project commenced, which meant that the first interviewees approached had little time to reflect. This was overcome by interviewing some interviewees in the second round, but not all took part in a second interview. Furthermore, due to the rapid nature of the evaluation, there was a short data collection period following the Learning Event, which meant that the impact of the project could not be properly evaluated. This also meant that we were unable to evaluate the whole Double Diamond model, due to the last step being the delivery of the solutions. Although the idea for the solutions has been delivered, we could not test and evaluate the impact of these solutions in maternity services. There may be biased perspectives on the positive impacts of the project, as the NHS staff interviewed maybe those who are more likely to attend such events, and who are more invested in creating change in maternity services than other NHS stakeholders.

CONCLUSION AND IMPLICATIONS FOR POLICY AND PRACTICE

Our study has shown a method by which co-production can be done meaningfully with service users. We found that a co-production approach that uses co-design methodology with Public Narrative as a user involvement strategy empowers maternity service users and calls NHS staff to act and create change. Key to the co-design process is the creation of an open and safe space that facilitates the dismantling of the hierarchy of knowledge between service user and professional, necessary to ensure that power is redistributed and that service users are genuinely part of designing solutions.

The results of our study carry with them some implications. First, this study suggests that creating impact through a co-production approach can play an instrumental role in ensuring that patients' voices are accounted for in healthcare policy development processes. Second, the specific recommendations for incorporating stories in healthcare processes can also be applied to other co-design projects that are unsure how to explicitly undergo co-design. We recommend that future studies implement and evaluate the approach in other healthcare contexts.

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CONFLICT OF INTEREST STATEMENT

The authors declare no conflicts of interest.

DATA AVAILABILITY STATEMENT

Audio recordings of the interviews analysed in this study cannot be shared publicly to protect the privacy and anonymity of participants. If you wish to obtain access to this data, please contact the UCL ethics committee at ethics@ucl.ac.uk and/or the corresponding author.

ETHICS STATEMENT

Ethics approval was obtained from the UCL Research Ethics Committee (REC) (6862/008).

DETAILS

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The dynamic nature of patient engagement within a Canadian patient-oriented kidney health research network: Perspectives of researchers and patient partners

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ABSTRACT (ENGLISH)

Introduction

Canadians Seeking Solutions and Innovations to Overcome Chronic Kidney Disease (Can-SOLVE CKD) is a pan-Canadian health research network that engages patients as partners across 18 unique projects and core infrastructure. In this qualitative study, we explored how research teams integrated patient partners into network research activities to inform our patient engagement approach.

Methods

To capture a breadth of perspectives, this qualitative descriptive study purposively sampled researchers and patient partners across 18 network research teams. We conducted 4 focus groups (2 patients and 2 researchers; $n = 26$) and 28 individual telephone interviews ($n = 12$ patient partners; $n = 16$ researchers). Transcripts were coded in duplicate, and themes were developed through an inductive, thematic analysis approach.

Results

We included 24 patient partners and 24 researchers from 17 of the 18 projects and all core committees within the network. Overarching concepts relate participants' initial impressions and uncertainty about patient engagement to an evolving appreciation of its value, impact and sustainability. We identified four themes with subthemes that characterized the dynamic nature of patient engagement and how participants integrated patients across network initiatives: (1) Reinforcing a shared purpose (*learning together, collective commitment, evolving attitudes*); (2) Fostering a culture of responsive and innovative research (*accessible supports, strengthened process and product*); (3) Aligning priorities, goals and needs (*amenability to patient involvement, mutually productive relationships, harmonizing expectations*); (4) Building a path to sustainability (*value creation, capacity building, sustaining knowledge use*).

Conclusions

Our findings demonstrate the dynamic and adaptive processes related to patient engagement within a national, patient-oriented kidney health research network. Optimization of support structures and capacity are key factors to promote sustainability of engagement processes within and beyond the network.

Patient or Public Contribution

This project was conceived in collaboration with a Can-SOLVE CKD patient partner (N. F.), with lived experience of kidney failure. He also co-designed the study′s protocol, led focus groups and researcher interviews, and contributed to data analysis. L. G. has lived experience as a caregiver for a person with CKD and facilitated patient partner focus groups. The patient partners, both of whom are listed authors, provided important insights that shaped our interpretation and presentation of study findings.

FULL TEXT

INTRODUCTION

Chronic kidney disease (CKD), defined as persistent structural or functional abnormalities of the kidney, is a significant public health concern that affects approximately 10% of the global population.[1,2] Care for persons with CKD is complex in that disease manifestations are varied and treatment options encompass both strategies to slow progression and manage complications.[3] In addition to its high associated morbidity, mortality and healthcare costs,[4] CKD management is complicated by a lack of high-quality evidence guiding clinical care.[5–7] Moreover, areas of highest kidney health research activity do not necessarily align with the priorities of health system users, including patients, whose involvement as partners in research has been increasingly sought to enhance research relevance and application in practice.[8]

The experience of living with a condition such as CKD means that patients, and those who care for them, hold valuable insight into how service delivery could be optimized to meet their needs.[9] Alongside international strategies that support purposeful and meaningful involvement of patients and the public in health research, such as the Patient-Centered Outcomes Research Institute (PCORI) in the United States[10] and National Institute for Health Research (NIHR) Centre for Engagement and Dissemination (formerly INVOLVE) in the United Kingdom,[11] Canada's Strategy for Patient-Oriented Research (SPOR) was launched to provide support for patient-oriented research; that is, health research that engages patients as partners, focuses on patient-identified priorities and enhances healthcare delivery and outcomes.[12,13] The Canadians Seeking Solutions and Innovations to Overcome Chronic Kidney Disease (Can-SOLVE CKD) Network is one of five pan-Canadian chronic disease patient-oriented research networks supported through SPOR. The network's mandate is to overcome challenges to uptake of research evidence into practice through engagement with knowledge users (i.e., patients, caregivers, clinicians, policymakers) across the research cycle, from inception to implementation of findings.[14] The active engagement of patients as partners in Can-SOLVE CKD reflects a shift from the traditional view of patients as research subjects toward a collaborative model of co-production of relevant, useable health innovation.

Several frameworks for patient and public involvement in research offer guidance on engagement approaches and practical strategies to promote meaningful collaboration, typically related to individual or smaller-scale projects.[13,15–17] However, the broad scope of initiatives across Can-SOLVE CKD meant that a single, uniform approach to patient engagement was neither feasible nor desirable. As diverse network teams operationalized and adapted engagement resources in response to their unique needs, it was unclear how patient partners could or should engage with researchers in their mutual pursuit of network objectives. Attempts to understand the perspectives of patients and researchers in this context are critical for establishing meaningful engagement processes and productive research partnerships.[18] With an appreciation for the dynamic nature of patient engagement within Can-SOLVE CKD, the purpose of this qualitative study was to explore how the network integrated patient partners into a wide range of research activities and initiatives to inform engagement structures and supports.

METHODSStudy Design and setting

The Can-SOLVE CKD network encompasses 18 unique, multidisciplinary research projects spanning kidney health populations (i.e., nondialysis CKD, kidney failure, transplantation, paediatrics) and research pillars (i.e., 1 basic science, 4 translational and 13 clinical projects). Infrastructure to support patient engagement, knowledge translation and capacity building was provided through several committees centred around patients (e.g., Patient Governance Circle,[19] Indigenous Peoples' Engagement &Research Council [IPERC][20]). Over 50 patient partners and 130 researchers from across Canada have contributed to network research and governance.

The network referred to existing patient engagement resources initially to establish patient co-leadership, team member roles and expected contributions across projects; more formal guidance documents and reporting processes were developed after the first year. In this qualitative descriptive study,[21,22] we outline how these processes took shape. We adhered to the consolidated criteria for reporting qualitative research[23] (COREQ) and to the ethical conditions outlined by the health research ethics boards from the University of Calgary (REB18-0131) and University of Montreal (#17-138-CERES-D).

Participant recruitment

We employed a purposive, maximum variation sampling approach to identify and invite eligible individuals from Can-SOLVE CKD to participate in this study. This sampling approach enabled selection of information-rich individuals to characterize unique experiences and central themes.[24] Any adult (≥18 years) patient partner or researcher within the network was eligible, although we aimed to sample across stakeholder roles and types of engagement activities to reflect a breadth of perspectives. This included deliberate sampling of patient partners and researchers with various roles on research project teams (i.e., project co-lead, research advisor and/or study conduct [for patient partners]; principal investigator, co-investigator or research support staff [for researchers]) and/or core network committees (i.e., co-chair or member of the Training and Mentorship Committee, Patient Governance Circle, IPERC and/or Knowledge Translation Committee). The network considers a ‘patient’ as someone who has personal experience living with kidney disease of any type or stage, informal caregivers, such as family or friends, and living kidney donors. Eligible researchers included principal investigators, research staff and/or research trainees on project teams. Many researchers were also healthcare professionals with experience in care delivery to persons with CKD.

Data collection

Data were collected through focus groups and semistructured interviews. The use of both focus groups and interviews in a study is a form of methods triangulation, whereby the data arising from group interaction and discussion can complement and enhance that obtained through in-depth, reflective interviews.[25] Focus groups were conducted during an annual network meeting held 2 years into the network's 5-year funding term and lasted 90 min. Each focus group was comprised of either patient partners or researchers to allow for in-depth discussion, manage potential power differentials and enable expression of differing viewpoints in a safe setting. An observer took field notes to supplement the recordings and track important nonverbal communication or actions by participants. Individual interviews were conducted by telephone or videoconferencing to expand on issues raised during focus groups and other experiences related to research engagement. Interviews were conducted by one investigator and lasted 40–60 min. N. F. conducted patient partner interviews and M. J. E. conducted researcher interviews to minimize unfavourable dynamics, except in instances of perceived conflict or pre-existing relationships. The question guides were developed by investigators based on familiarity with the engagement literature to explore perceived roles, experiences and challenges of engaging across projects/disciplines in kidney health research (see Supporting Information). Interviews and focus groups were audio-recorded, transcribed verbatim and uploaded to NVivo 12 (QSR International Pty.) to facilitate data organization and retrieval. Demographic and network-related information were collected to characterize our sample and inform our purposive sampling approach.

Data analysis

Data collection and analysis took place concurrently so that emerging concepts could be explored in subsequent interviews. Two investigators (N. F., M. J. E.) inductively analysed de-identified transcripts using thematic analysis.[26,27] Through a systematic process, investigators first independently reviewed transcripts, noting initial impressions and developed a preliminary coding framework. This was refined through coding 10 transcripts (5 patient partners; 5 researchers), discussing code definitions and scope and then systematically applying codes to remaining transcripts. Coded extracts and relationships between concepts were then examined to develop themes based on meaningful clusters. Emerging themes were verified in relation to the data set for rigour and discussed among the broader research team to ensure consistency, credibility and relevance of findings. The comprehensiveness of our data set allowed us to develop subthemes that encompass important yet distinct concepts comprising each main theme. We presented preliminary findings at a subsequent Can-SOLVE CKD Network annual general meeting that most study participants attended, and incorporated audience feedback and suggestions into our final thematic reporting. We maintained an audit trail of all analytic steps.

RESULTS

Forty-eight individuals from across Canada participated in this study. We conducted 2 patient partner and 2 researcher focus groups (each with 4–10 individuals) and 28 interviews (12 patient partners; 16 researchers). Six patient partners participated in both an interview and focus group. Participants were represented on 17 of the 18 network projects and all core network committees. Of 24 participating patient partners, 14 contributed to 3 or more research project teams and 9 sat on more than 1 core network committee. Patient partner responsibilities and expected commitments varied across projects and committees, including attending regular meetings, providing input on study processes and materials, co-developing tools, engaging with communities and disseminating findings. Of the 24 participating researchers, 17 contributed to only one research project team and 4 sat on at least 1 committee. Demographic characteristics for patient partner and researcher participants are provided in Tables 1 and 2, respectively.

Table 1 Characteristics of participating patient partners ($n = 24$).

Characteristic	Number
Sex	
Female	16

Male	7
Prefer not to answer	1
Age (year)	
<40	5
40–64	15
≥65	4
Location of residence	
Western Canada	15
Ontario	8
Quebec	0
Eastern Canada	1
Territories	0
Duration in Can-SOLVE CKD Network (year)	
≤1	4
2–3	14
>3	6
Number of projects and/or committees	
1	5
2–3	11
>3	8

Table 2 Characteristics of participating researchers (*n* = 24).

Characteristic	Number
Sex	

Female	15
Male	9
Age (year)	
<40	4
40–64	20
≥65	0
Location of residence	
Western Canada	15
Ontario	8
Quebec	0
Eastern Canada	0
Territories	0
Prefer not to answer	1
Duration in Can-SOLVE CKD Network (year)	
≤1	1
2–3	6
>3	17
Number of projects and/or committees	
1	16
2–3	7
>3	0

We characterized four overarching themes according to participants' accounts of the dynamic nature of patient engagement within the network (Supporting Information): (1) Reinforcing a shared purpose; (2) Fostering a culture of responsive and innovative research; (3) Aligning priorities, goals and needs and (4) Building a path to sustainability. Participants' initial apprehensions about patient engagement evolved into appreciation of its value and potential impact on kidney health innovation. The concept of a research ‘paradigm shift’ was articulated by one researcher participant (R12) and reflected across most patient and researcher interviews—participants recognized the increasing support for and embeddedness of patient engagement in routine health research practices and were enthusiastic about being on the forefront of this new era. They emphasized how formalized but flexible engagement support was necessary to align network and patient partner research priorities and to develop practical solutions to real-world problems facing healthcare delivery. Both patients and researchers pointed to a need to anticipate sustainability of network infrastructure, engagement resources and research products. Identified themes and subthemes are described in the following section, with additional exemplar quotes provided in Table 3.

Table 3 Additional supporting quotes from patient partners and researchers presented by theme and subtheme.

Patient partner quotes	Researcher quotes
Theme 1: Reinforcing a shared purpose	
Subtheme: Learning together	
‘My job has been trying to help everyone figure out what patient engagement looks like, what activities we should be doing, trying to ensure that there is some sort of communication flow’. (P3) ‘It's been a teaching and educational exercise over the last three years’. (P3) ‘Those are all pieces of input that I put in there, and [Researcher-X] and the team have all really bent over backwards for me, my lack of knowledge and me being really green to this stuff. And I've tried my best, to be honest’. (P7) ‘Patients didn't know what they were getting into, researchers also had no idea what they were getting into, and I think overall there are some things that completely surprised them. And in general, they [researchers] are very impressed by patient engagement and the patients that have been involved’. (P11)	‘I'm just learning and doing as I go, hoping I'm always keeping the patient in the best interest and including them’. (R2) ‘We didn't have any guidelines; we just learned from experience and leveraged our experience working with First Nation community members and adapting some of those philosophies when working with patient partners’. (R3) ‘I figured out just asking them [patient partners]: “Does this work? Doesn't this work? What do you think about this?” It's the best way to do it, it becomes a lot easier’. (R4) ‘In terms of trying to fit a formal structure around it, yes [first time engaging patients]. But we do a bunch of work with the patients particularly who have been participants in our trials in an informal structure’. (R9) ‘I was expecting a little better structure. I think a lot of this has been on the job learning’. (R14)
Subtheme: Collective commitment	

‘When I think of a patient lead or patient-oriented research, I see the patient at the very centre and their role being clearly defined from the very beginning’. (FG-P1) ‘I think there's something really opening up, too, in terms of determining direction and choice of research based on what patients are saying. And I think the beginning of patient research from that perspective is truly exciting’. (FG-P2) ‘The research team really had a patient-oriented focus and [that was top of mind] …it's more like we're driving the bus or trying to drive the bus’. (P3)	‘Ultimately the end goal is for us to improve kidney health for people living with chronic kidney disease, and so, yes, I think at every stage of the process we value their [patient partner] feedback and we try to incorporate it pretty much all the time’. (R3) ‘I think you have to be open minded, and you have to acknowledge that these people bring a unique perspective and that we're all here to learn something from each other. So, open your mind and learn something from them because they've got a lot to teach’. (R8) ‘When you think about it, how ridiculous it is to have a service industry, which in some ways we are, that completely ignores the input from their customers. [Person-X] spoke about that at one of the initial Can-SOLVE meetings, and it was like a penny dropped that this was clearly wrong and that we do need to engage patient partners’. (R13) ‘I think they have to continue to keep us honest, right? What we promised and what we wrote, we have to carry through it’. (R14)
Subtheme: Evolving attitudes	
‘I was surprised that doctors were coming over and tapping me on the shoulder and saying: “I have someone for you.” Doctors have seen the value we bring and have invited us to participate’. (P4) ‘The biggest [surprise], I guess, is that my voice was being recognized, [being] acknowledged’. (P11) ‘I think the researchers need to not be afraid of asking us’. (P12)	‘I actually never really thought I would have patients on my research team, but for me, I think it's new and I learned from them’. (R2) ‘What I've learned is more perspective. I think working with these kids, the biggest advantage has been giving me perspective, as a clinician, where they're coming from. I had no idea five years ago what life was really like for these kids’. (R7) ‘I think there has been a shift for sure, and that [researchers] are starting to shift more. I hear them talking a lot more about including patients …Asking people what they want before we tell them what they want’. (R10)
Theme 2: Fostering a culture of responsive and innovative research	
Subtheme: Accessible supports	

<table>
<tr><td>‘I usually have a sit-down meeting once every two weeks, just about business in general and keeping her [other patient partner] up to date on everything’. (P6)
‘Knowing when to pull back or allow space for people to be healthy and to be well, because we don't want to make the people that are involved in the network any more sick or tired than they already are’. (P9)
‘I thought getting to know the other patients was really helpful in terms of sort of understanding what was going on especially the patients that had sort of been there before Can-SOLVE was officially Can-SOLVE’. (P12)</td><td>‘We find that corresponding by phone individually is very helpful for [patient partners] as well …First a conference call with all the patients and then individual follow-up with each of them’. (R1)
‘It's taken some work to get us where we are now, but that's not because we don't have the traits, it's just that there had to be some circling and getting to know each other’. (R4)
‘Patients are no different than researchers—they have different priorities, different challenges, different personalities, and they have the challenges posed by their health as well’. (R12)</td></tr>
<tr><td colspan="2">Subtheme: Strengthened process and product</td></tr>
<tr><td>‘The researchers were saying, “Well, we can't focus on that because it's phantom.” And we were saying, “It may be phantom to you, but it's not phantom to us. Just because you can't now tell us what's causing it doesn't mean it's not real.” And they started changing their tune’. (P8)
‘[We are trying] to get people the best background information as possible that we can. Same thing with Wabishki Bizhiko Skaanj, we started right from scratch with that one. Like, there was nothing, they just told us, “We need a cultural sensitivity training,” and the slate was clean’. (P10)
‘If we're there in the initial conversations we can help shape [the projects], and something very simple [can have] a profound positive impact on how to present it to patients’. (P12)</td><td>‘There's a place for [foundational research], but there's also a big place for applied research that's going to make a difference. And if you want to make a real difference, the first thing you have to do is ask the people whose lives will be impacted, what kind of a difference they want made’. (R10)
‘In the end we're still going to need health research, whether we can find people to buy in or not. And where I come back to you is it is a paradigm shift. It is required. It's going to make the research better, so we have to keep working that way’. (R12)
‘[Patient partner] gave us some very helpful feedback about the reluctance for patients to engage in studies that look promising and yet there's an untreated comparative group. We had to deal with that …by amending the protocol so that if the studies were positive, then patients would be invited for an open-label extension of that study’. (R13)</td></tr>
<tr><td colspan="2"><i>Theme 3: Aligning priorities, goals, and needs</i></td></tr>
<tr><td colspan="2">Subtheme: Amenability to patient involvement</td></tr>
</table>

‘They [researchers] don't expect enough of us, in general. They try to give us like really simple small things. We've encouraged [Researcher-X], as part of our project, to like give us more and we'll tell [them] when it's too much’. (FG-P1) ‘You can see in the teams when the leader is very passionate, the rest of the team follows. But in the ones that are okay, that's kind of good, the patient engagement piece for those projects …kind of like slips through the cracks a bit’. (FG-P2) ‘In the first two years [we] soft pedaled to give the projects a little bit of time to get up and going and figure out what works and what doesn't. [Now] we are becoming far clearer in regard to the expectation as it relates to patient engagement’. (P3) ‘Our coordinator, has been right there dealing with me, dealing with my lack of technology, and she [is the] the go-to’. (P7)	‘We're able to kind of take it [patient engagement] to the next level. But I think as a research team who's maybe new to patient engagement, might not have a lot of experience, they might not be as open as we are to being adaptable to patient partner's needs’. (R3) ‘The only other thing that is a bit difficult is that the pace of [research], for a physician and a researcher, is different than the pace of what patient partners [are expecting]’. (R4) ‘I think unfortunately Can-SOLVE is very adult focused and integrating youth into the adult world doesn't really work. In order to bring these kids and their parents into an upper level of Can-SOLVE, I think it needs to be something that they can relate to’. (R7) ‘I would say that the type of research we're doing in this project lends itself really well with the patient partnerships at all phases. But I can totally appreciate that some other types of research would not, and that'll be okay too’. (R11)
Subtheme: Mutually productive relationships	

‘[In] the ideal world patients are co-authors on papers. I have already been a co-author on several papers [and] that is where we should be going. That's the way research should be thinking of our input. I mean, it's only to their benefit that the patients are [involved]’. (FG-P1) ‘I [gave] my feedback on whether I think it's beneficial on behalf of the patients I work with. I think of a patient lead or patient oriented research, I see the patient almost at the very center …[When] I go back to my area I want to be able to say to the patients: “This is what your role will be, this is what you'll give back, and this is what you'll get”’. (FG-P1) ‘My expectations, whatever knowledge I could gain, that I could share with people with kidney disease, and to make an improvement in outcomes for people with kidney failure’. (P7) ‘Hopefully [there will be] less patient engagement [in the future] because there would be less kidney disease’. (P9)	‘I felt like they [patient partners] needed to be heard, for sure …at the time to be really mindful of listening to them, but also making sure that we were staying on track’. (R2) ‘We've been inclusive but judicious with what they're [patient partners] involved with. I think that where they've been involved is very helpful, and where they're not involved it probably wouldn't have change anything in that regard’. (R5) ‘I think it's important for patient partners to realize that their view represents their view, not the global patient view. There are different patients, just like different doctors have different opinions about how a condition should be managed. Different patients also have very different perspectives about what they want to know and how they want to be treated …Early on in Can-SOLVE, sometimes there was a sense that if you didn't agree with them you were somehow ignoring the patient voice’. (R8) ‘I think that their input into what had been identified as the path of the study was helpful and did change the direction of things. I hope they feel the same way. I'm not sure that the patients have had the same satisfaction from their involvement in the study. It's awkward making that leap from what patients want for their own care and experience to what's the best science project here’. (R16)
Subtheme: Harmonizing expectations	

‘We talk a lot about the learning process, not just for us as patient partners being involved …but it's the growing pains that researchers have to go through as well in terms of what to expect from us’. (FG-P1) ‘I think [my] responsibilities are so different on every aspect of Can-SOLVE that I'm involved in. I've learned that that's okay, different things work for different provinces and different groups. I think that sometimes we try and find a “one-size-fits-all” …but what we're finding is that it doesn't work across everything’. (FG-P2)	‘I think the ideal patient partner is someone who comes to the table and recognizes the importance of their position on the research project and really leverages that position to speak the truth of what they exactly feel …People might not be open to providing [us] with their true feedback. But as a patient partner that's exactly what we're looking for. We want them to tell us exactly how it is’. (R3) ‘We're going to have discussions around these things, about how much time they want to do, [what] they want to give to it, what the work involves, and how they want to approach it’. (R10) ‘What we try to do is be very clear as to what the ask is, what we anticipate the timing is, what the touch points are, and how much commitment it would involve, and we try to be very clear upfront’. (R14)
Theme 4: Building a path to sustainability	
Subtheme: Value creation	
‘[Researcher] brought up a really interesting point and it just hit me like a ton of bricks, I thought: “Wow, we're not even really taking good care of our donor.” And all our emphasis has been on the recipient and his care, but not the donor’. (FG-P2) ‘[It is a] very large task to ask patients to take on the Transplant Ambassador Program in their hospital. Now we saying: “Can you recruit other people and train other people and have this program running really well?” I have a sense of ownership and pride in this program]’. (FG-P2) ‘Being on this committee means I am reading more [health information] …and now I am not afraid to ask questions’. (P11)	‘I can take any problem in healthcare, and I can apply infinite resources to that problem, and it will never be perfect. So, what we have to do is take a step back and say, how much value are we generating from this particular intervention? My sense is we're applying a lot of resources to patient engagement and no matter how many resources we apply, it likely won't be enough. I think what you're going to see is the pendulum swinging back the other way a little bit’. (R5) ‘The purpose of this is still science, right? It is the best science working with our patient partners, but also we have to have a results phase, and these entities have to be productivity and research driven. And that's also what I hear from our patient partners is, “Are we sticking to our timelines? Are we getting results?”’ (R14)
Subtheme: Capacity building	

'It is a very large task to ask patients to take on the Transplant Ambassador Program in their hospital. Not only are we saying, "This is your project, your baby, manage it," but also "Can you recruit other people and train other people and have this program running really well?"' (FG-P2) 'I'm just helping to move us along. Everybody needs to be articulate and know about Wabishki Bizhiko Skaanj, and we have some tools to help you …Everybody on the project should really be up to speed on what's happening and move forward that way'. (P10)	'I would consider it as part of my postdoc training because it is the way of research now. That is something essential that I need to develop, and if I want to have more impactful research, I really do need to engage patients'. (R6) 'We're pretty good at project management type stuff and we might be good at getting the people together into the room. But then, step back a little bit and move other people forward. Stop trying to be the one who says we have to develop this thing in this way'. (R10)
Subtheme: Sustaining knowledge use	
'In a perfect world we'll have all 18 projects completed and papers published and the knowledge translation from the projects will very quickly get out there in the world. That work is really a lot of blood, sweat and tears, and I'd hate to see it just put in a box. I hope it's the basis for kidney research going forward'. (P5) 'I focused my interest on the knowledge translation piece because it was something that really interested me. I didn't really know a lot about it, but the idea of translating information into a way that lay people could understand really interested me. I have had a lot of opportunities to do speaking engagements'. (P12)	'[We] got a lot of feedback from both researchers as well as the patients on the implementation process. It's hard for them to maybe talk about the technical side of things, but their voices are heard in the sense of the technicality of things …That's the whole point of this, to make it so that when you go to practice it becomes part of your routine care for the patient'. (R2) 'Very few studies now come with these blockbuster results, and I think it's important then to be able to put it in language that is clear, that the information is written so that people can understand it, and then see how they can integrate this into their care'. (R14)

Note: Participant identifiers are indicated in parentheses following each quote. Those beginning with P denote patient participants, those beginning with R denote researcher participants, and FG refers to focus groups.

Reinforcing a shared purpose

Despite having little experience in patient engagement before their joining the network, nearly all patient and researcher participants described approaching their partnerships with an open mind and eagerness to work together to advance the research. The network's clear mandate, strong leadership and shared commitment to patient engagement formed the necessary foundation upon which research partnerships could flourish.

Learning together

Participants reported varying levels of experience and comfort with patient-oriented research before the network's inception. Although several patient partners and researchers had contributed to early network planning and its funding proposal, they described being underprepared for collaborating subsequently within network project teams. Some researchers drew on prior experiences working with patient advisory groups outside of Can-SOLVE CKD, and others expressed initial apprehension and dismay over a lack of guidance on patient engagement specific to their projects. Most patient partners described having little to no prior research experience other than as a research participant, and participants outlined a co-learning exercise as they developed processes together and formalized collaborative engagement structures. As one patient said,

> We were falling into the trap of wanting to do too much because we were enthusiastic, but we had to find that balance of what we could conceivably do with our time as volunteers and what was better done by people who are staff. I think in the first year, we always joked about building the plane as we're flying it. (P12)

Collective Commitment

Despite initial apprehension about how to collaborate in research, researchers identified a strong rationale for patient engagement from their projects' outset. They indicated that research engagement could enable realistic, honest and pragmatic discourse about end-user needs and outlined a commitment to ensure their work resonated with patient partners. Patient partners described a variety of motives for engaging in the network (e.g., ‘build skills’, ‘leave a legacy’, ‘obligation’ to community) but indicated it was critical first to understand why their involvement was needed and how they could best contribute. They explained how mutual appreciation of each other's roles paved the way for confident, prepared and productive research and a patient leadership model where, as one patient explained, ‘we're driving the bus’ (P3).

Evolving attitudes

Several patient partners observed that researchers seemed uncertain initially about how to involve them in the research and hesitant about approaching them with research-related questions or requests. Although many patients described being uncertain themselves, they expressed an openness to being approached about collaborative problem solving. They indicated these partnerships were critical to move research forward and mitigate concerns about patient engagement pitfalls, such as tokenism. As relationships developed and confidence grew, patient partners described an increasing appreciation of having their voices recognized, and researchers described gaining a grounded perspective of the implications of their work to those living with kidney disease. This translated into growing receptivity to patient engagement among researchers and perceived value in better understanding patients' lived experiences. As one nonclinician researcher described,

I don't see any of these individuals [in a clinical setting], so I can't really understand what they're going through. I think this really humanizes the work …It's really great to get that patient perspective and it's also extremely motivating when you hear these individual stories. (R6)

Fostering a culture of responsive and innovative research

The Can-SOLVE CKD Network's culture of openness and responsiveness to the needs of diverse projects and members was highlighted across nearly all interviews and focus groups. Patient partners described increasing comfort and confidence with collaborating in research as a result of the formal (e.g., tools, guidance documents, workshops) and informal (e.g., peer mentorship) support structures that evolved over time. Participants appreciated the network's emphasis on patient engagement as a way of making research more meaningful and achieving greater impact from the resulting health innovations.

Accessible supports

Participants acknowledged the network's flexibility and responsiveness to patient partners' needs, which varied across individuals and over time. This included recognizing patient partners as people first, with responsibilities and priorities outside of their network commitments. Researchers described adapting research processes to accommodate patient partners (e.g., technical assistance, conducive scheduling) and remaining flexible when unforeseen obstacles arose, such as patient partner illness. Patient participants related the network's responsiveness to its nonhierarchical structure and emphasis on equity and inclusion among its membership.

Many researchers and patient partners turned to internal network resources to inform their engagement strategies, which matured over the network's term to offer practical, accessible guidance in areas including knowledge translation, engagement competencies and cultural safety. Several researchers had also accessed resources outside of the network (e.g., provincial SPOR resources), but few described connecting with other network teams for support. In contrast, nearly all patient partners articulated the importance of peer mentorship and supported one another informally in the unique experience of partnering in kidney health research. One researcher commended a patient partner for serving as a key support for others on their research team,

She [patient] was enabling all the partnerships, and not only enabling them but ensuring everything was being done appropriately, serving as a resource to patients when they had questions. (R11)

Strengthened process and product

Researchers anticipated that improved research processes resulting from patient engagement would strengthen the resulting health innovation. They provided several examples of how patient partner involvement enhanced patient-facing materials, interview question wording and outcome selection. A few researchers discussed how ethical issues about trial design raised by patient partners, such as randomization and untreated comparator groups, influenced how they framed research methodology to participants. Researchers anticipated enhanced applicability of health innovations developed in partnership with patients, and patients observed how even seemingly small changes to research approaches can influence outcomes. Patient partners explained how a movement toward greater patient leadership prompted establishment of several core network resources and community engagement strategies (e.g., Indigenous health research learning pathway).[28] One patient who contributed to both research and core committee activities focused on Indigenous health explained,

I think that's one way that patient partners can help change the way research is done—the way we're engaging with communities, especially Indigenous communities. For me, it's an important thing. (P9)

Aligning priorities, goals and needs

With the diversity of researchers, patient partners and initiatives across the network, participants described how alignment between individual and research-related goals, priorities and needs was critical for the network's success. Although several patients initially questioned how to best apply their experience and expertise to the various initiatives, expectations surrounding patient engagement became clearer as the research progressed and comfort and familiarity with one another grew. Researchers described being attuned to patients' obligations and priorities and striving to ensure their collaboration remained mutually rewarding, respectful and enjoyable.

Amenability to patient involvement

Initially, researchers and patient partners wondered whether certain project types might be more amenable to patient engagement than others. For example, researchers wondered if projects that were more slowly paced, biomedical or with inexperienced teams may struggle with effective patient involvement. However, patient partners cited research teams' commitment, and responsiveness as key elements supporting engagement regardless of project type. As one basic scientist explained, ‘There is no need for a science background that is particularly advanced’ for patients to contribute to biomedical research (R13); technical concepts could be relayed in a straightforward way, and patients could contribute to other important, nontechnical aspects of research design. Patient partners described relying on communication and trusting relationships to overcome hurdles to engaging in less clinically focused projects. Some researchers and patient partners identified challenges to integrating paediatric patient partners into larger network circles, which they related to a lack of supports to meet the distinct engagement needs of this population.

Mutually productive relationships

Researchers discussed difficulties balancing patient engagement with their research objectives and milestones. Participants relayed infrequent instances where researchers and patient partners had competing agendas or ideas regarding their project's direction. Awareness of each other's needs grew through increased familiarity among the team, habituation to research processes and mechanisms for revisiting priorities. Researchers described being mindful of patient partners' other commitments and trying to limit their requests for involvement to areas of highest yield for both parties. Patient partners reinforced that for them to contribute most effectively, all parties needed to be aligned in their vision and goals. Patient partners suggested that acknowledgement of their contributions to the research, such as through co-authorship on manuscripts, would foster trust and a desire to collaborate on future research initiatives.

Harmonizing expectations

Patient partners and researchers across most projects described having open conversations about project goals initially, whereas more systematic processes to establish expectations came later with a better understanding of team members' needs and project direction. Patient partners acknowledged the diversity in network projects and membership and rejected a singular approach to engagement. As one patient said during a focus group,

Different things work for different provinces and different groups. I think that sometimes we try and find a one-size-fits-all [approach] …but what we're finding is that it doesn't work across everything. (FG-P2)

Some researchers described tempering initial high expectations for prompt, observable changes to their research and growing to appreciate other benefits of research partnerships, including community connection, honest feedback and compassion rooted in lived experience.

Building a path to sustainability

Researcher and patient participants described how sustainability of the network's research, infrastructure and patient engagement had been a priority for the network from the outset. They conveyed a sense of pride and ownership of the network's achievements, and indicated it was a shared responsibility of network membership to ensure the collaborative relationships and best engagement practices continued beyond the network's funding term. Key drivers of sustainability identified across researcher and patient partner interviews included demonstrating value of the research arising from patient engagement, building capacity for research engagement through skills development and hands-on learning, and supporting the uptake, or implementation, of research generated by the network in real-world settings.

Value creation

Researchers explained how systemic changes to research engaging patients must demonstrate not just impact, but value for them to be sustainable long-term. Few questioned whether the potential impact of patient-oriented research was enough to justify continued high costs, resources and human capital. However, others re-iterated the tangible and/or anticipated benefits to health innovation, healthcare delivery and patient outcomes to justify its longevity. Several patient partners noted personal impacts of their involvement in Can-SOLVE CKD, such as enhanced disease-related knowledge, research skills and confidence during research and clinical encounters. Participants explained how continued awareness in public and research circles was needed to sustain patient engagement beyond the network's funding term and dedicated patient-oriented research funding opportunities. One researcher expressed the following concerns,

It [patient-oriented research] could be just another [funding] program that comes to an end …If they're able to get a bit more momentum behind the public to speak to this—by public, I mean members of the public but also voters, because it's the minister of health that ultimately will decide on these issues—then I think that will help with the sustainability of it. (R12)

Capacity building

Patients and researchers explained how early identified gaps in support provided opportunities to develop internal resources that could enhance and sustain patient-oriented research capacity. Patient partners described how resources that were under development at the time of interviews (e.g., communities of practice, Indigenous health research learning pathways)[29] were expected to promote research skills development and sustain culturally safe research practices. Building on the strong bonds formed among patient partners within the network, they discussed formalizing peer-led training of patient partners in the how-to of patient leadership. One researcher observed colleagues becoming increasingly comfortable with patient engagement as ‘the way of [doing] research now’ (R6), and others described how principles of collaborative research were being integrated with their institutions' graduate and postgraduate research training programmes. For established researchers and trainees, most patient engagement training took place through hands-on, experiential learning and were supplemented by network tools and resources.

Sustaining knowledge use

Patient partners and researchers indicated that moving the knowledge generated through network activities into the applied setting was critical to sustain use of health innovations over time. They discussed the importance of considering real-world implementation and evaluation at the outset of their projects, and aligning their patient engagement approaches with this goal of relevant, usable research. Patient partners discussed being naturally attracted to areas where they could advance research application and observe changes in kidney care and patient outcomes on a larger scale. Both researchers and patients outlined how patient partners had contributed to traditional, academic strategies for sharing research knowledge (e.g., co-authorship on published reports) as well as community engagement activities to share tools and personal narratives related to network initiatives. Both researchers and patient partners described a commitment to seeing their initiatives through to completion, and suggested that patient engagement was critical for promoting use of research in clinical practice or communities affected by kidney disease. One researcher remarked,

The other big thing [is] implementation and dissemination. We develop this tool, but how do we ensure uptake? Patient input is the key component. (R1)

DISCUSSION

In this study, we provided a real-world example of the dynamic nature of engaging patients as research partners in a national kidney health research network. In exploring perspectives across a wide variety of network activities, we identified several themes capturing the adaptive processes and growing confidence of the network to integrate patients as partners. Mutual appreciation of the purpose of patient-oriented research formed the foundation upon which patient partners and researchers aligned their priorities and moved projects forward. In response to identified gaps in initial engagement guidance offered by the network, members co-developed internal resources and processes that formalized a concerted, inclusive approach to partnership across initiatives. This promoted greater transparency and accountability and helped align the expectations of both researchers and patient partners. The network's emphasis on impactful, applicable research from the outset encouraged participants to think ahead to how network infrastructure, health innovations and patient engagement could be sustained long-term.

Several frameworks exist in the academic literature to guide patient engagement in research,[13,15,16,30,31] although their application in the context of a diverse network like Can-SOLVE CKD has not been clearly defined. Recognizing that high-level frameworks leave room for customization and flexibility; our findings support the approach taken by network members of adapting existing guidance and developing novel strategies to support engagement where gaps were identified. These findings align with those of Greenhalgh et al.,[15] who assert that a single, off-the-shelf framework may be less useful than a menu of evidence-based resources. This opportunity to customize engagement approaches based on network members' needs led to the creation of several tools and products to support research processes targeted to patient and researcher audiences alike.[28,29,32,33] The Can-SOLVE CKD Network is not unique in its application of guiding frameworks for patient engagement and commitment to key principles of inclusivity, support and mutual trust.[34–37] Whereas others have reported similar challenges to partner with patients in research, such as avoiding tokenism and balancing individual needs with research objectives,[17] our study is among the first to describe how research practices, supports and perspectives adapted to new and unforeseen issues arising over the course of a large-scale network partnership. In one qualitative study of researchers' and patients' experiences in a patient-oriented research programme centred on case management, participants described strengthened relationships over time, the importance of strong structural supports and balancing diverse interests.[38] Our study extends and enhances these findings through its emphasis on the dynamic nature of network partnerships, responsive leadership and implications for sustainability across a broad scope of research projects and network initiatives.

The network environment was a key success factor, specifically its emphasis on relationship-building across initiatives. Initially, much time was devoted to establishing interpersonal connections, devising ways to collaborate effectively and overcoming steep learning curves for both patients and researchers. This emphasis on relationship building reflects a growth mindset,[39] whereby researchers and patient partners worked in concert to first determine their specific projects' needs and then built upon their experiential knowledge toward relevant, usable research products. This growth occurred to a large extent organically alongside increasing confidence, competency and trust among network members. Consistent with our findings, other reports have emphasized cohesive relationships and positive team interactions as a way of facilitating patient engagement and ensuring meaningful engagement experiences.[16,40–42] In response to the variability in Can-SOLVE CKD project needs and patient partners' commitment to improving engagement practices, the network developed internal resources with membership input over time that created more structure and expectations around patient engagement (see Toolkit for Project Leads, available at https://cansolveckd.ca/resources/). These supports matured over time, enabled teams to tackle challenges as they arose and fostered a culture of responsiveness and innovation as the network's capacity grew.

Recognizing the significant financial and human costs of engaging patients in a national research network, participants described how impactful interventions and measurable outputs would be critical for sustaining the network and patient engagement more broadly. As participants reflected on their journey, pockets of innovation were highlighted, including patient leadership in research projects (e.g., Transplant Ambassador Programme),[43] knowledge mobilization and strategies for equity, diversity and inclusion. One example that featured prominently in interviews was the Wabishki Bizhiko Skaanj learning pathway,[28] which was developed to address identified gaps in Indigenous health research competence in the network. The opportunity to leverage strong connections between patient partners across the network could further enable sustainable research engagement and capacity, whereby patient partners with varied research and kidney disease lived experience could assist, support and train one another using principles of peer mentorship.

Our findings raise important questions about the next steps for the network and generated research knowledge upon completion of the funding term. As most network projects focused on developing and testing strategies to address identified gaps in kidney care delivery,[14] several participants questioned how the resulting research would make its way into practice to influence healthcare delivery and patient outcomes. With the increasing adoption of collaborative models of health research, opportunities to engage patients in the dissemination and application of research are emerging.[33,44] As patients will ultimately use or benefit from the output of health research activities, their engagement in research should not necessarily cease once a study has concluded or a health intervention has been generated—continued engagement in identifying barriers and facilitators to implementation, tailoring interventions and evaluating outcomes are critical for promoting uptake and sustainability in different contexts.[44] Moreover, building capacity in the know-how of patient engagement among trainee and early-career researchers can foster a ‘business as usual’ attitude toward research partnerships that translate to real-world applications.[45,p.4] These issues require further study, practice and adaptation as appropriate to promote sustainability of network initiatives.

Many of the challenges to patient engagement described by Bird et al.,[40] such as power imbalances, were overcome by the supportive research environment. In ensuring patient embeddedness across all network activities and access to capacity-building resources, patient partners' and researchers' initial scepticism gave way to new skills, enhanced confidence and tangible research outputs over time.[46–48] Strong relationships coupled with shared purpose largely averted tokenism and its consequences, such as poor engagement experiences and lack of demonstrable impact.[49] However, engagement success relies on many factors, some of which are outside the control of research teams.[50,51] The varying research skills and engagement experience of participants meant that additional resources were needed to address early challenges to research collaboration. Also, the vast scope of research disciplines and project foci within the network necessitated tailored approaches to supporting each team's unique needs, which were not always possible given resource and capacity constraints. As a result, some communities may have been unintentionally less well supported, such as paediatric patient and caregiver partners for whom the network's ‘adult-focused’ engagement structures may not be conducive. Several barriers to diversity within patient-oriented research have been identified, such as lower literacy, communication challenges and socioeconomic disadvantage, which can lead to the underrepresentation of certain groups.[52] Recognition of the diversity of individuals engaging as research partners is a critical first step toward tailoring engagement infrastructure and processes to meet their needs and those of the research. Going forward, the network has prioritized an equitable and inclusive approach to its research activities and patient partnerships that includes guidance on culturally safe health research practices, considerations for sex-/gender-based and paediatric research engagement, resources to build capacity and confidence in research engagement, and mechanisms to communicate concerns and address challenges as they arise.

The main strength of our study lies in its capture of diverse perspectives across a national patient-oriented research network, although we acknowledge some limitations. We did not attempt repeated data collection from participants over time, which may have enabled the refinement of concepts related to change or evolution. However, our staggered recruitment approach over the later network stages captured nuances in how patient engagement practices adapted and resulted in tangible products. Perspectives were limited to those who agreed to participate, which for focus groups meant individuals present at the in-person annual network meeting. Invitations for phone or virtual interviews were extended to all network members and aimed to reduce barriers to participation for all interested individuals. Given the close relationships among network members that our study highlighted, it is possible that participants tempered their responses or presented overly optimistic assessments of their engagement experiences. To address this, a research team member with whom participants were unfamiliar led interviews/focus groups and gave assurances of confidentiality. Lastly, this study was undertaken exclusively with Can-SOLVE CKD Network members, although we anticipate that our broad characterization of findings will be transferrable to both large-scale, patient-oriented research contexts and individual teams seeking guidance on patient engagement.

CONCLUSION

Across a diverse, national patient-oriented kidney health research network, approaches and attitudes toward patient engagement evolved over time as relationships solidified, confidences grew and priorities became better aligned. Optimizing support structures was a driver of effective engagement and took place both through the adaptation of existing resources and the development of new ones to address identified gaps. The sustainability of the network's innovations and patient partnerships was emphasized across participants and highlights the need for future work to examine the impact and value of network products and explore avenues for expanding patient-engaged kidney health research capacity.

AUTHOR CONTRIBUTIONS

Meghan J. Elliott and Nicolas Fernandez conceived of this study, participated in its co-design, conducted interviews/focus groups, data analysis and drafted the manuscript. Tamara L. McCarron participated in data analysis, results interpretation and manuscript preparation. Kara Schick-Makaroff contributed to the study design and provided methods support, including qualitative data analysis and interpretation. Leah Getchell assisted with participant recruitment, data collection during focus groups and interpretation of findings. Braden Manns contributed to the study design and provided operational support and mentorship to the research team. All authors provided a critical review of the manuscript and approved the final version.

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CONFLICT OF INTEREST STATEMENT

The authors declare no conflicts of interest.

DATA AVAILABILITY STATEMENT

The data sets used and/or analysed during the current study have not been made publicly available due to the potential identifiability of participants from transcript data. Data may be available upon reasonable request to the corresponding author.

ETHICS STATEMENT

This research project was approved by the Conjoint Health Research Ethics Board (CHREB) at the University of Calgary (REB18-0131) and the Comité d'éthique de la recherche en santé (CÉRES) at the University of Montreal (#17-138-CERES-D). All participants provided informed consent for participation in this study.

DETAILS

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Experiences of human papillomavirus self-sampling by women >60 years old: A qualitative study

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ABSTRACT (ENGLISH)

Background
Human papillomavirus (HPV) self-sampling has shown to be acceptable and feasible across cultures and effective in reaching women who do not participate in regular cervical cancer screening. However, most of these studies have included younger women. There is a lack of knowledge of how older women reason about HPV self-sampling.

Objective
The aim of this study was to describe how women (>60 years old) experience the offering of self-sampling of HPV, compared to having a sample collected by a healthcare professional.

Design and Participants
The study had a qualitative explorative design. Four focus group discussions were conducted with women 60–69 years old (*n*=22). Data were analysed using principles of interpretive description.

Results
Five themes were identified: self-sampling—convenient and without pain, lack of knowledge, worries related to HPV self-sampling, need for information and taking a societal perspective.

Conclusion
Women aged >60 years found that HPV self-sampling was convenient and easy to perform. Further, they stressed the importance of being able to remain in the screening programme in advanced age and that self-sampling could be

a possible solution. This study also revealed a lack of knowledge among women in this age group regarding HPV infection, how the disease is transmitted and its relation to cervical cancer.

Public Contribution

Women who had performed HPV self-sampling participated in the focus group discussion.

FULL TEXT

BACKGROUND

Screening as secondary prevention to reduce the risk of cervical cancer has been established for decades, shown to be effective and organized as a population-based programme.[1] In the last 10 years, knowledge of the human papillomavirus (HPV) as a necessary prerequisite to cervical cancer has changed the prevention methods from primary cytology testing to analysis of HPV.[2–4] HPV vaccination as primary prevention is now offered in many countries, mainly to young girls before sexual debut as the main target group.[5] As a complement to the cytological analysis, an HPV test (testing for high-risk types 16 and 18 that cause 70% of all cervical cancer) is provided in several European countries, as well as in the United States,[6,7] recommended by the WHO.[8] HPV testing has even been shown more effective than screening by cytology.[9,10]

Participation in prevention programmes is a key to reducing the incidence of and mortality from cervical cancer. Different barriers to attendance, as well as factors that encourage women to attend the screening, are well-described in the literature, for example, by our previous studies.[11–14] Nonattenders of the cervical cancer screening programme have been described as having an increased risk for cervical cancer.[15,16] Self-sampling of HPV has been implemented and has shown to be effective in reaching women not attending the regular screening,[17,18] as well as being experienced as acceptable and feasible across cultures.[18,19]

Several studies have been conducted evaluating HPV self-sampling,[20] but most of the research has included younger women <60 years of age. To exclude older women from studies is problematic: HPV screening may be especially important for this age group, as cytology as a primary screening method has low sensitivity for women >60 years. Additionally, older women are often excluded from regular invitations to screening programmes, and cervical cancer found often is identified late, leading to a more invasive form of cervical cancer.[21–23]

This study is part of a project evaluating HPV self-sampling in women aged 60–69 years compared to having a sample collected by a healthcare professional, with respect to conformity (sensitivity, specificity). In Sweden, HPV testing has been recommended by the National Board of Health and Welfare since 2016 as the primary screening method for women 30 years and older, taken by midwives in the screening programme. Additionally, the upper age limit for screening was increased from 59 to at least 64 years of age, where the woman now is called for her last sampling at 70 years of age.[24] Larger studies on the prevalence of HPV and how screening works are lacking in the older age group.[25] At the same time, one-third of all cervical cancer cases are diagnosed in the age group over 60 years.[21,26] A large number of studies have been conducted to evaluate self-sampling with varying results, but studies in an older age group are scarce.[27–30]

Previous studies have emphasized factors such as information, communication and trust as essential in a well-functioning screening programme with high coverage.[18,31] However, knowledge is lacking on how older women >60 years reason about cervical cancer and its prevention. Additionally, there is a lack of studies that include women's own perspectives. Studies evaluating the self-sampling of HPV are often cross-sectional or trials[32–38]; very few qualitative studies exist, but these have either used a theory-based interview guide that also guided the analysis,[39,40] or focused on specific groups of women with different ethnicity.[41–44] Therefore, the framework for this study is based on naturalistic enquiry[45] that fosters an inductive approach, meaning that instead of relying on predetermined conceptualizations, aims to generate new knowledge and understanding by interacting with the participants in their own lived experience. The aim of this study was to describe how women >60 years of age experience the offer of self-sampling of HPV, compared to having a sample collected by a healthcare professional.

MATERIALS AND METHODSDesign

This qualitative explorative study using focus group discussions (FGDs) was part of a larger study on the evaluation of vaginal self-sampling of HPV (https://researchweb.org/is/fourol/project/228071 Reg. no 228071) in all women aged 60–69 years in Örebro County in Sweden (*N* = 2258) who were invited to a ‘catch-up’ screening programme, arranged and funded by the health care during 2018–2020. This catch-up programme is part of the population-based cervical screening programme in the county and aims to be a bridge between the earlier screening programme that women exit at 59 years and the ‘catch-up’ to 70 years. Exclusion criteria were being deregistered from the population-based screening programme due to prior hysterectomy or own request or having a history of invasive carcinoma. Women were given information about the main study along with the invitation to the ‘catch-up’ screening programme.

Participants

All women who had given informed consent to the main study on vaginal self-sampling of HPV were informed about this focus group study. A strategic sample of women included in the main study, based on variations in HPV test results, were asked to take part. FGDs were held in two different strata: women with the same result in the self-sample and in the midwife-obtained sample and women with different results in the two tests. The women were contacted by a researcher who explained the study and invited them to participate. If the women agreed to participate, they were sent further written information on the study. A total of 22 women agreed to participate in the four FGDs (two groups in each stratum). The FGDs included five to six participants each.

Data collection

Data were collected in FGDs, a form of group interview that capitalizes on communication between research participants to generate data. Further, it is known that potentially delicate issues are more suitable for group discussions than individual interviews.[46–48] All FGDs took place at the university and were held in Swedish, moderated by the first author (K. B.) and co-moderated by the last author (M. H.-N.), both with experience and training in performing FGDs. All FGDs began with the moderator welcoming the participants and providing information on how the discussion would be conducted. The FGDs began with a discussion on the question ‘What is your experience with HPV testing?’ and followed an open interview guide. The FGDs lasted between 35 and 45 min, and all interviews were audio-recorded. Data collection and the first steps of the analytical process were conducted in parallel, as women's reasoning in prior FGDs was discussed and confirmed in the next one.

Data analysis

The audio recordings of the FGDs were transcribed verbatim by a professional secretary. Data analysis was inspired by interpretive description.[49,50] First, transcripts were read by two of the researchers separately using constant comparative techniques and involved asking the questions ‘What is going on here?’ and ‘What is this about?’ while reading sections of the text. The researchers identified patterns, generating initial codes inductively. Moderators' notes of reflections during the data collection were taken into consideration in the analysis. The preliminary coding structure was revised through a comparison of the coded transcript excerpts. This was followed by searching for themes, reviewing themes and defining and naming themes.[51] Second, the emerging themes were interpreted through a ‘clinical lens’, based on professionals' backgrounds as nurses and prior research conducted on cervical cancer screening. Preliminary results were continuously discussed during the analytical process in the larger research group, which included researchers with different professional backgrounds (GP/pathologist, biomedical scientist, registered nurses) and midwives working with cervical cancer screening. For examples of the analysis process, see Table 1. Quotations were identified from the FGDs to illustrate the findings and increase trustworthiness.[45]

Table 1 Examples of the analysis process

Meaning unit	Code	Theme
W: It's like less painful or unpleasant to.	Less pain and discomfort	Self-sampling—convenient and without pain

And especially for those who felt uneasy in the gynaecology chair	Avoid the gynaecology chair	
W: I could take the test when I had time, and when I felt like doing it. I didn't have a time to fit. So that was great.	Be able to control time and place	

RESULTS

The findings are presented in the five themes formed in the analysis, all related to women's experiences of HPV self-sampling.

Self-sampling—Convenient and without pain

Overall, the women described the self-sampling as easy to perform and found the instructions were clear. Even though most of them were retired from work, they expressed that it was convenient to take the sample at home on an occasion that was suitable for them. Not having to schedule a time and not having to travel were described as advantages of the self-sampling. When taking the self-sample at home, they could do it slowly and did not have to hurry. They expressed that they had experiences of feeling rushed and of being on a conveyer belt when visiting the midwife for Pap smear tests and that there was no time for conversations or asking questions.

W*1: When you go to your midwife to take the sample they don't have a lot of time. It is much like a conveyer belt. (FGD 3, *n* = 6)

*W = woman

The women shared experiences of how having a sample taken by the midwife often could be painful. However, they described that the self-sampling did not hurt, as the sample was collected from the vagina and not from the cervix. They also were relieved that they could take the self-sample without entering the gynaecological examination chair, which could be more difficult in advanced age and which several of the women described as a vulnerable situation.

W1: It was so easy, so convenient. It was no trouble at all to take the test. Yes, simple you got all the equipment you needed, and the form was already filled out. It was just for us to put the stuff in the envelope and put it in the mailbox.

W2: It was very good, there were good instructions. It wasn't difficult.

W3: No.

W4: At first, I thought like, ‘Oops, how is this going to work?’ But, soon after, I thought that I should be testing. There was nothing strange at all.

W5: No, and you didn't have to ‘climb’ the gyno chair. Because that can feel unpleasant. I have always attended. I've come when I've been summoned. It isn't that fun, but there are things that are worse. (FGD 1, *n* = 5)

Lack of knowledge

It was obvious from the FGDs that the women lacked knowledge about the central aspects of cervical cancer and its prevention. Several women did not know what HPV was or that the HPV test and the Pap smear were not the same tests.

W1: Then I have to ask what it is [HPV], because I have not heard of it?

Interviewer: No, you have not heard of it at all?

Several W: No. (FGD 2, *n* = 5)

Almost all of the women expressed that they had attended regularly at the population-based cervical cancer screening over their lifetime but never reflected on why they took the Pap smear and the intervals. Several women expressed that they did not know that they would not be offered an invitation to the cervical screening programme, anymore.

W1: Because I was not called, so then I thought, I have to call and ask. ‘No’, she said. ‘You have turned 60 year, you know, so you cannot come anymore’. I thought ‘What?’ (FGD 1, *n* = 5)

It emerged in the FGDs that it was not clear to most of the women that the screening programme had changed over time.

W1: What is it really you look for when you take the test at the midwife's …I did not have knowledge, I think. I go there, and they take that test, and then I trust it. But then there is [information] about this virus …if it exists and then how it has affected? (FGD 2, *n* = 5)

The women described thoughts about how they could take the test by themselves, as they lacked knowledge of the technical procedure of the tests. They reflected on whether the test taken by themselves really was correct, as the test (Pap smear) they had done by the midwife feels and sometimes hurts compared to the self-test.

W1: I thought …do you have to scratch the cervix or is it [the HPV virus] …everywhere in the vagina I wondered. How do I know that I find the right spot?

W2: I felt the same.

W3: Yes, me too.

W2: Or wondered…

W3: Yes, because when you are at the midwife, you can feel how she almost…

W2: Yes.

W1: Yes, exactly …maybe it is all over the space. I don't know.

W2: No, but that was exactly the same as I thought. Because they say, 'Oh, it might hurt a bit'. (FGD 1, *n* = 5)

Reflections about whether the tests were the same or not were also especially apparent among those women who received a positive HPV result when the test was done by themselves and a normal result when done by the midwife. They expressed frustrations about the different test results and about not being given any explanation as to why the tests differed.

W1: The test showed that I have the virus.

W2: The same for me.

W1: And I got scared, so I had to call you at the hospital …and got an explanation from—not the person I talked to at first, but from someone else. I got the explanation that the self-test sample is not performed the same way as the midwife test. The midwife test is from the cervix but the self-test is just from the area near the cervix. But I got scared, and I think about this almost every day. (FGD 3, *n* = 6)

Some women knew that the HPV test was about the prevention of cervical cancer but expressed little knowledge of the disease itself, such as its causes and possible prevention. Even those women who had experienced cytological abnormality and treatment for that expressed uncertainty about the causes and progression of the disease. They were surprised that they could have gotten the infection many years ago and not have had any symptoms. Some women thought that a positive HPV test was equal to having cancer. Other women described how a positive HPV test led to a fear of developing cervical cancer.

W1: Yes, the information, you know, one goes and wonders, 'Will I get cervix cancer now?' (FGD 4, *n* = 6)

For several women, it was unknown that HPV is a sexually transmitted infection. Few women described a connection between HPV and sexual partners and cervical cancer, and some mentioned the HPV vaccination for young girls as a connection. However, most of them were not aware before entering the study that HPV could appear in women in their age group. They discussed HPV being a 'new' infection and something that occurred only in teenagers and young people.

W1: We who struggle with the same old man, it is not as important…. (FGD 4, *n* = 6)

W3: In our age it isn't of that great importance [HPV vaccination], if we already caught it [HPV], it doesn't matter where we got it from.

Several women: No, no.

W3: But it is, of course, important information for young persons. (FGD 4, *n* = 6)

Worries related to HPV self-sampling

The women expressed several different kinds of worries related to the HPV self-sampling. There were women who described being worried that they had performed the self-sample in an incorrect way. They were concerned about whether they had rubbed the swab deep enough to collect any virus. There were also worries about whether the self-sample was as reliable as the sample taken by the midwife. And there were also some women who expressed a worry about other diseases that could be detected by a midwife beside the HPV test that was taken. It was seen as beneficial to have extra control to have it confirmed that one was healthy.

W1: But are there other …diseases or symptoms or …that a midwife can see when you are sitting there that you may also be able to treat? (FGD 1, *n* = 5)

Women in the FGDs who had a positive HPV test also expressed other worries—worries about from whom they had got the HPV virus and worries about being infected with HPV without knowing. Not knowing for how long they had been infected with HPV generated reflective thoughts about how they had lived their lives when they were younger. There were also women who became suspicious towards their partner, wondering whether their partner had cheated on them or where else they could have got the HPV virus.

W1: …then I asked my husband, 'Have you been with anyone else?' Because HPV can't be for 30 years, can it?

W2: Well…

W3 &4: Yes.

W1: Well, the feeling at first, 'Have you been with anyone else?' No, well, it does matter. Like that, but otherwise this [the self-test] is great. (FGD 3, *n* = 6)

W1: Yes, but I know, because I have been cohabiting for 10 years, and it makes you wonder, when did I get it [HPV virus] …A certain fear …My first thought was, 'Did I get it from you or did I get it before I met you?' Well, it is not strange to ponder about this.

W2, 3: No, no.

W1: Because you don't ask a man about such things.

W2: No.

W3: No. (FGD 4, *n* = 6)

A positive HPV test also caused worries about having cancer and whether they were going to die. One woman described how she identified different signs in her body that could be related to being ill:

W1: Yes, I was worried, and I must say that I googled, too, and then I experienced and thought that I started to smell strange around me …so then I called actually to the clinic and asked…. (FGD 4, *n* = 6)

They also expressed concerns about whether the information from the doctor and midwife was reliable. There were also descriptions of how seeking information on the Internet could increase their worries. There was an overload of information, and they described information that was in some ways contradictory. It was expressed that it could be difficult to know which websites, besides Sweden's 24-h helpline 1177, provided reliable information.

W1: …to get an answer [a positive HPV test] and get an understanding of what this answer means, because that's what you may not know …And then maybe you get worried and then you go to the Internet and read, 'Yes, this is how it can be', but talk to a person then about 'I feel this worry. Is it justified or not?' and get it explained. It is probably the most important thing. (FGD 2, *n* = 5)

Need for information

The women expressed that information was crucial, especially if the HPV self-sampling was to be implemented for all women. Not having contact with a midwife made the information even more important.

W1: HPV self-sampling, if it is to become a routine then, then I think they need to think about the information, because now we all have different experiences. Then you meet with no one. (FGD 4, *n* = 6)

In the FGDs, two different needs for information were raised, the need for information regarding how to perform the test, but also a need for information when receiving a positive test result.

W1: Yes, information is so important, because you go and wonder like, 'Well, will I get cervical cancer now?' (FGD 4, *n* = 6)

Although, it was appreciated that the information included images on how to perform a test, the women still perceived that they lacked information regarding how to perform the test. They also lacked information that the self-sample kit was safe and that it was not possible to do it wrong. Also, some women expressed that they would have been helped by information on how to perform the test if one had fragile tissue or vaginal prolapse. These women described that they at first could not perform the self-sampling and that they had contacted the midwife at their health centre and been advised to use Vaseline or Estriol to facilitate the sampling.

Further, the women expressed that an instructional video on the Internet could be a complement to the written instructions, even though there were illustrations in the instruction booklet. An instructional video could be useful for all women, but especially for women who could not read instructions in Swedish.

It was emphasized that the information regarding how to perform the self-sampling, when to expect a result and what happens if the result is positive is of extra importance as there is no contact with health care and no opportunity to ask questions.

W1: That you get a phone call, not paper.

Several W: No, no.

W1: It always raises questions, and then you cannot get an answer on a piece of paper.

W2: No, but exactly, I called. You feel that you want to know, why is there a difference, yes.

W4: Yes, yes, I had to come up with such a real examination, but then after that I did not get any information and I missed it. So I was at the clinic here, and they did a real check …It was okay, but you would still have wanted a message afterwards that ‘this is what it looks like or this is what it looks like; it does not come out’. And I still have not received it. It is tedious. (FGD 3, *n* = 6)

Taking a societal perspective

The FGDs revealed that the women felt that they were contributing to society by taking part in this self-sampling study. All the women had attended the standard screening programme over the years, but they expressed that self-sampling could be an alternative for women who do not attend the standard screening programme. For example, immigrants or women with negative experiences of gynaecological examinations or abuse might find the self-sampling acceptable. Further, as well as self-sampling being seen as a way to increase the participation rate, it was also seen as an option for ongoing testing of women over 60 years of age, as it is not resource intensive.

W1: …to present the arguments for [continuing HPV testing] instead of, as I said before, ‘Well, now, aren't we even allowed to do this [HPV testing], anymore? There are no nurses, and you are not allowed go to the primary healthcare centre, and now shall you do the HPV self-test as if you were a gynaecologist?’ I can really hear what it would be like if you did not get the information about the positive aspects of self-testing.

W2: Yes.

W3: Uh-huh.

W1: And also, I think that we all feel that if we can contribute to reducing the costs, it feels sensible.

W4: Definitely! (FGD 1, *n* = 5)

Self-sampling was described as a way of freeing up time and resources for the midwives. On the other hand, one woman expressed a concern that self-sampling could increase societal costs, as more women would participate in the screening programme, and as the HPV self-sample seems to be more sensitive than the test taken by the midwife, which would lead to increasing numbers of follow-up examinations.

W1: It's about the information about why and in what way you contribute …Because I mean, just like you are in, if many more go, you can prevent, if you should have cancer or get cancer, that it spreads before you get help, and in the long run, everyone benefits from it. And that healthcare can focus on those who really need help. (FGD 1, *n* = 5)

Most women were aware of the HPV vaccination of young girls, and they thought it was important also to vaccinate young boys. Further, they discussed whether not only women but also men should be tested for HPV. In one of the FGDs, the heteronormative aspect of HPV testing and vaccination was reflected on.

DISCUSSION

The present study examined how women aged 60–69 years experienced HPV self-sampling. To our knowledge, this is the first qualitative study on experiences of HPV self-sampling on women in this age group, compared to having a sample collected by a healthcare professional.

Overall, the women were positive towards self-sampling and found it convenient and easy compared with going to the midwife. This is in line with results from earlier studies on younger women.[39] Even though most of the women in our study were retired from work, they still appreciated being able to perform the test whenever they wanted.

The women were familiar with the HPV vaccination among young girls, but they had not reflected on the fact that older women could also be infected with HPV. Further, not all of the women were aware that HPV is a sexually transmitted infection and that it could cause cervical cancer. A knowledge deficit is also reported in younger women in earlier research, where the women also described a lack of knowledge and a need for more education on HPV and cervical cancer.[52,53] This result is surprising, in that despite those women >60 years having regularly attended screening programmes during the majority of their lives, they lacked knowledge about screening methods and the causes and prevention of cervical cancer. Thus, it highlights that most women participate in screening without knowing why, and there remains a need to find different ways to communicate information so that women could give more informed consent based on evidence. Several of these women had suggestions of how to improve information, for example, through instructional videos, especially important for women who do not speak Swedish. They discussed how it is difficult to navigate and judge different information published on the Internet and how to get women to only read established and evidence-based websites. In the FGDs, many thoughts were raised about cervical cancer and how it could be detected in time. There may be a need to target just this age group of women with, for example, information about risks for cancer and its prevention, but also more practical information such as how to perform the self-test with fragile tissue or vaginal prolapse. Prior studies have investigated different interventions, mainly focussed on providing information to younger age groups of women[54,55]; however, it is a methodological challenge to evaluate the effect of such interventions. In this study, no women expressed that they had difficulty understanding how to perform the self-sampling test. However, several described uncertainties about having done it correctly. Williams et al.[53] showed that women have low confidence in their ability to take the self-sampling test correctly. In recent studies comparing the self-sampling of HPV to having a sample collected by a clinician, the results showed a concordance between the two testing methods.[17,20,56–58] This stresses the need to provide the women with information that the HPV self-sampling test is equal to having a sample collected by a clinician, and is reliable.

It was obvious in the FGDs that these women expressed a trust that other abnormalities or diseases could be detected during cervical cancer screening but concluded that these could be missed in a self-sampling HPV test. It might be in some way a 'false' trust that it is possible to detect diseases during this short examination/test. In any case, it might be understood in the light of it being more common in this age group to have established contact with a midwife over one's lifetime or to have had regular gynaecological examinations and/or opportunistic screening by a private clinic. It might be different in younger age groups, as much of the behaviour is related to socialization over time in health prevention programmes. This trust in the midwives could also explain why many of these women expressed being disappointed not to be offered regular invitations to screenings, anymore. In the context of being older and having an increased risk for diseases such as cancer, they pointed out that they need to be checked and that the self-sampling tests could be an alternative. This must, however, be considered along with the increasing risk of 'false' positive HPV tests that would need to be followed up and an increasing need for correct information about what a positive HPV test means.

In the light of the Covid-19 pandemic, it is even more urgent to evaluate and implement HPV self-sampling, as the conventional HPV screening programme is set on pause to minimize the risk for transmission of infection.

LIMITATIONS

A potential limitation of this study is that all women in the FGDs had performed the self-sample as well as having had the sample performed by the midwife. Even though the main study invited all women registered in the 'catch-up' screening programme, there is a risk of a selective sample, not targeting women who were difficult to reach or who had low levels of participation in population-based screening. We made an effort to arrange a focus group with women who had not performed the self-sample to capture descriptions of reasons not to perform the self-sample. These women either declined to participate in a focus group or did not show up for the focus group. This might have led to a more positive description of the self-sample, even though the included women did describe the disadvantages as well as the advantages of self-sampling. Future research focusing on women who are hesitant to use HPV self-sampling is needed to get more complete knowledge.

CONCLUSION

Women >60 years of age found that HPV self-sampling was convenient and easy to perform. Further, they stressed the importance of being able to remain in the screening programme in advanced age and that self-sampling could be a possible solution. This study also revealed a lack of knowledge among women in this age group regarding HPV infection, how the disease is transmitted and its relation to cervical cancer. Information targeting just this age group of women is needed, along with practical instructions on how to perform the test with fragile tissue or vaginal prolapse.

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CONFLICT OF INTEREST

The authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

The data are not publicly available due to privacy or ethical restrictions.

ETHICS STATEMENT

Ethical approval was obtained from the Regional Ethical Review Board of Uppsala, Sweden (reg. no: 2017/365).

DETAILS

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Involving patients and carers in patient safety in primary care: A qualitative study of a co-designed patient safety guide

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ABSTRACT (ENGLISH)

Background

Involving patients is a key premise of national and international policies on patient safety, which requires understanding how patients or carers want to be involved and developing resources to support this. This paper examines patients' and carers' views of being involved in patient safety in primary care and their views of potentially using a co-designed patient safety guide for primary care (PSG-PC) to foster both involvement and their safety.

Methods

A qualitative study using semistructured face-to-face interviews with 18 patients and/or carers in primary care. Interviews were transcribed and analysis was conducted using an inductive thematic approach.

Results

Overall participants expressed enthusiasm for the PSG-PC as a tool to support patients and carers to be involved in patient safety in primary care. However, for some participants being involved in patient safety was seen as taking on the role of General Practitioner and had the potential to add an additional workload for patients. Participants' willingness or ability to be involved in patient safety was influenced by a range of factors including an invisible, often underacknowledged role of everyday safety for patients' interactions with primary care; the levels of involvement that patients wanted in their care and safety and the work of embedding the PSG-PC for patients into their routine interactions with primary care. Participants identified components of the PSG-PC that would be useful to them, in particular, if they had a responsibility for caring for a family member if they had more complex care or long-term conditions.

Conclusion

Involving patients and carers in patient safety needs a tailored and personalized approach that enables patients and carers to use resources like the PSG-PC routinely and helps challenge assumptions about their willingness and ability to be involved in patient safety. Doing so would raise awareness of opportunities to be involved in safety in line with personal preference.

Patient or Public Contribution

Patient and public involvement were central to the research study. This included working in partnership to develop the PSG-PC with patients and carers and throughout our study including in the design of the study, recruiting participants, interpretation of findings.

FULL TEXT

BACKGROUND

Patient safety is a key global health priority as health systems have increasing levels of demand for services alongside financial constraints, fragmentation, the changing role of technology in care and information transfer and an increase in long-term condition management.[1–5] Patient safety has been broadly defined as ‘the avoidance, prevention and amelioration of adverse outcomes or injuries stemming from the processes of healthcare’.[6] For example, in primary care diagnostic and medication incidents, were most likely to result in harm or severe harm.[7] There has been a focus on preventing the most common causes of harm such as prescribing, diagnosis and treatment in primary care.[3,7–10] Patient–provider communication issues also contribute to patient safety incidents either directly or indirectly.[1,11] Yet who should be involved and responsible for patient safety remains unclear.[12] Increasingly the role of patients and carers has been advocated as an additional component of patient safety to prevent serious incidents by preventing harm before it occurs to make care safer.[13–17]

Involving patients in their health care and research is an essential element for improving both the quality and safety of care and addressing inequalities within health and social care.[14,18] Involving patients in health care is one source of creating a resilient system that allows flexibility and adaptation within a complex healthcare system.[19–24] This flexibility may be what is needed to improve patient safety, and health outcomes and meet the needs of patient-centred care.[20,25,26] Patient safety issues can be identified throughout patient experiences with health care, from access or diagnosis to medication management, treatment and self-management with primary care managing repeated uncertainties along episodes of care.[7,27–29] Involving patients more explicitly in their care needs to be considered within an understanding of the dynamic, nuanced nature of involvement as well as patient–clinician dynamics situated within the wider contextual factors which may influence involvement.[6,15,30,31] This approach builds on the growing debate within the wider patient safety literature. More broadly within the patient safety literature, there has been a shift from a ‘Safety-I’ perspective (which can be defined as a context in which as few things go wrong as possible) to a ‘Safety-II’ approach (which focuses on how things go well in everyday work by understanding the uncertainties and trade-offs).[32] It has been argued that these two perspectives on safety reflect two distinct but complimentary views of patient safety.[33] There is a need to learn from everyday work in the development of initiatives for patient safety that build on a deeper understanding of safety.[33]

Assumptions are often made within the policy and broader literature that patient involvement is an equally desired role by all patients without recognizing the role of the system, contextual or individual (e.g., health literacy) factors that might influence an individual's capacity or desire to be involved.[30] How patients identify their eligibility for health care is influenced by multiple factors, from the individual themselves, and their social contexts through to macro-level influences on services from resource allocation to service design, this complex interplay of factors have been termed candidacy.[34] This is important when considering that patients and their carers have a unique perspective as they move within and across system transitions and have the potential to reduce variability experienced from health care which may have an impact on outcomes.[20,35] The majority of research on patient safety in primary care has been descriptive, with few studies focusing on interventions aimed at involving patients in their safety.[1,27,36–39] Patients' capacity or willingness to raise safety concerns may be influenced by previous experiences of health care, especially where previous experiences have left them feeling vulnerable.[8,12,36,40–42] For example, patients living with multiple long-term conditions or with more complex care needs experience duplication and fragmentation of care and experience.[36,43]

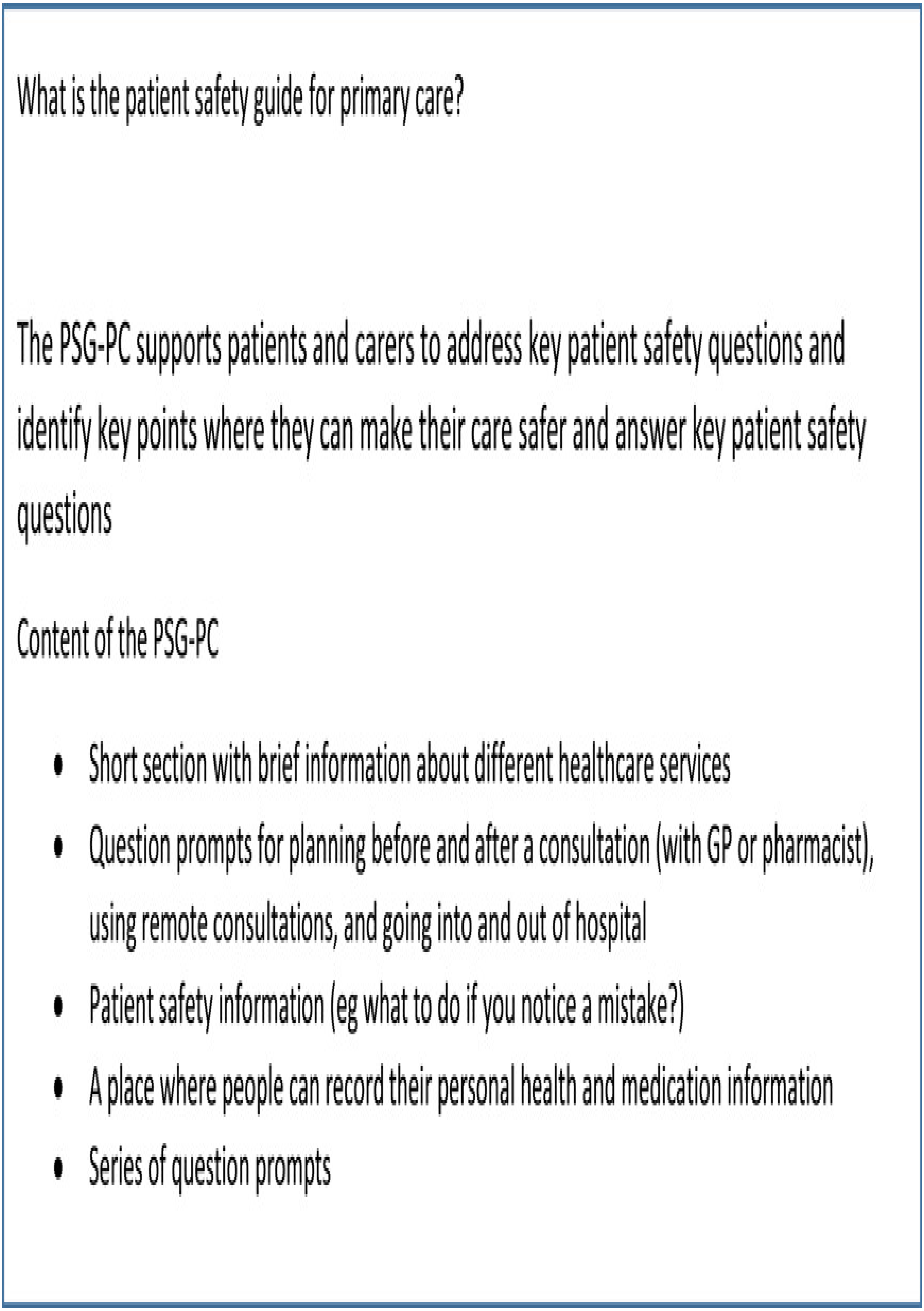
One approach to involving patients in patient safety is to create an ongoing dialogue which builds trust, clarifies expectations and ensures understanding between patients and healthcare professionals.[10,39,44] Yet this approach is based on an assumption that everyone will want and be able to be actively involved in their care and patient safety. There has been limited research which has examined what this would look like in practice and whether patients have the willingness or capacity to take on this additional work.[38,45,46] This expectation of the role of patients in patient safety needs to be examined to ensure that initiatives to involve patients do not create or reinforce inequity or compound patient safety risks.[47]

There have been tools and handbooks developed for patients to support their involvement in their care and safety in secondary care (including safety tips, treatment plans and fall prevention) but an equivalent version does not exist in primary care.[15,48–53] This study is part of a wider project to develop and test the patient safety guide for primary care (PSG-PC).[15,36] The PSG-PC supports patients and carers to address key patient safety questions and identify key points where they can make their primary care interactions safer and be active partners in their care. The PSG-PC has been developed using an experience-based co-design approach in partnership with patients, carers, members of the public and healthcare professionals (including GPs and pharmacists) and the development has been reported in detail elsewhere.[15,36,47,54,55] The PSG-PC was co-designed to support effective communication between patients and clinicians, which was considered a key component of involving patients and carers in patient safety in primary care.[15] The aim of this study was to explore patients' and carers' views of being involved in patient safety in primary care and their views of potentially using the PSG-PC to support involvement.

METHODDesign

An in-depth qualitative study design was adopted using semistructured interviews which examined patients' and carers' views of being involved in patient safety in primary care and their views of potentially using a PSG-PC (see

Figure 1) to support involvement. Semistructured interviews were chosen to allow participants to discuss without limitation the areas they recognized as most important as well as to include topics that the research team, including members of the patient and public involvement group for the study, identified as relevant.[56] the PSG-PS has been developed using a participatory approach based on the experience-based co-design approach,[15] for this part of the wider project we aimed to explore the views of people who had not been involved in the development of the prototype to enhance the potential acceptability and feasibility of the intervention.[57] This study is part of a wider project to develop and test the PSG-PC,[15,36] and used a prototype of the PSG-PC to explore patient and carers' views of being involved in patient safety and using a tool such as the PSG-PC to support this.



What is the patient safety guide for primary care?

The PSG-PC supports patients and carers to address key patient safety questions and identify key points where they can make their care safer and answer key patient safety questions

Content of the PSG-PC

- Short section with brief information about different healthcare services
- Question prompts for planning before and after a consultation (with GP or pharmacist), using remote consultations, and going into and out of hospital
- Patient safety information (eg what to do if you notice a mistake?)
- A place where people can record their personal health and medication information
- Series of question prompts

Enlarge this image.

Recruitment

Patients and carers were recruited using a snowball sampling approach through advertisements and contacts including with voluntary groups and community centres across Greater Manchester and social media (Twitter).[58] Eighteen patients and/or carers were interviewed. Fourteen participants were female, and 18 were White. We recruited participants from a range of characteristics of different socioeconomic status, geographical areas, gender, experiences of using services and different numbers and types of conditions. Participants ranged from having no long-term conditions to a maximum of seven long-term conditions (see Table 1). As this study was examining a prototype that was only available in English, potential participants were excluded if they could not speak or read English. They were invited to contact the research team by email or phone to express interest, and potential participants were sent a participant information sheet. Recruitment was completed when no new themes that were relevant to the aims of the project emerged from the data.[59]

Table 1 Participant demographics

Participant ID	Age	Gender	Employment status	Health condition
PS01	72	Female	Retired	Chronic obstructive pulmonary disease (COPD) or asthma
PS02	69	Female	Retired	Diabetes; arthritis
PS03	65	Male	Kitchen porter	Thyroid problem
PS04	71	Female	Retired	Chronic obstructive pulmonary disease (COPD) or asthma; eczema
PS05	45	Male	Town and country planning	Chronic fatigue syndrome; anxiety or depression
PS06	53	Female	Carer	Arthritis; anxiety or depression; lupus
PS07	58	Male	Retired	Angina or heart attack; high blood pressure; kidney problems; COPD or asthma; stomach problems; arthritis; anxiety or depression
PS08	58	Female	Administrator	None reported
PS09	25	Female	Postgraduate student and project manager intern	Polycystic ovary syndrome
PS10	58	Female	Medically retired civil servant	Thyroid problem

PS11	69	Female	Retired	High blood pressure; arthritis
PS12	71	Male	Retired	High blood pressure; anxiety or depression; high cholesterol; heart murmur
PS13	71	Female	Retired	Stomach problems; arthritis; anxiety or depression
PS14	52	Female	Teaching assistant	None reported
PS15	64	Female	Retired	High cholesterol
PS16	40	Female	Project work	None reported
PS17	34	Female	Volunteer at primary school	Anxiety or depression; schizoaffective disorder
PS18	34	Female	Student	COPD or asthma; stomach pains; anxiety or depression

Data collection

Interviews were conducted by either R. L. M. or S. G. face-to-face between January and March 2017. Written consent was obtained from all participants. The topic guide was developed by researchers in collaboration with the patient and public involvement contributors. Interviews were conducted in participants' homes. Interviews ranged from 22 to 59 min (average of 39 min). Interview participants were offered a £20 voucher to compensate them for their time. Reimbursing participants for participation is becoming more commonplace which may influence peoples' willingness to participate in research, however, it has been argued that is unethical to not reimburse participants to in part address power imbalances between paid researchers and unpaid participants.[60,61] To reduce the potential of the payment influencing the participants' comments, it was explained to participants that the research was interested in their opinions and that there were no right or wrong answers and it was explained that the vouchers were reimbursement for their time and were free not to answer questions and could end the interview at any time.

The interviews were digitally recorded and professionally transcribed. Interviews explored openly patients' and carers' views of being involved in patient safety in primary care and their views of potentially using the PSG-PC to support involvement, and what they liked and did not like about the PSG-PC. Interviews took an open approach, using vignettes to explore the limitations of the guide and how people could use it in practice which enabled participants to examine in depth what would or would not be acceptable for them in line with a person-based approach to intervention development.[57] This included exploring their experiences of using primary care services, health and medication management, giving feedback, their view on the PSG-PC and what information is important, or not, to support people to be involved in patient safety. The topic guide was modified iteratively throughout data collection and analysis.

Data analysis

Data analysis was ongoing throughout the study and followed an inductive thematic approach.[62,63] All authors met regularly to discuss emerging themes. All authors analysed transcripts and commented on the interpretation of the data and agreed on the key themes and concepts. Key themes were identified through discussion. Key themes and quotes were circulated to all the authors for comments and discussion. A final set of themes and subthemes were agreed upon by all authors. Nvivo 11 qualitative analysis software was used to support analysis. The sample size was determined by thematic saturation, which was identified when there was a point where no new themes or codes were developed from the analysis.[64]

Patient and public involvement

A patient and public involvement group was established to support the wider study to co-develop the PSG-PC along with other stakeholders and information about that has been detailed elsewhere.[15] Our patient and public involvement group were involved in the qualitative study to help design and format the questions and support recruitment. R. L. M. also presented the initial themes and led a discussion about what they meant in terms of developing the PSG-PC and interpreting types of involvement in patient safety that participants expressed which fed into the subsequent refinement of the PSG-PC.[15,36]

RESULTS

Eighteen patients and/or carers were interviewed. Fourteen participants were female, and 18 were White. Eight participants were employed, eight were retired and two were students. Participants' ages ranged from 34 to 72 years old (mean age of 56 years) (see Table 1). The main themes related to the invisible role of everyday safety for patients with interactions with primary care; the levels of involvement of patients in patient safety in primary care and the role of the PSG-PC and the work of embedding the PSG-PC for patients into routine interactions with primary care.

The invisible role of everyday safety for patients with interactions with primary care

Participants described initially their contact with primary care and their role in patient safety in an implicit way. Participants' accounts detailed everyday experiences where they identified issues that could be considered potential safety issues. The PSG-PC could help prevent this as it includes a prompt to consider medication allergies to make explicit their role in identifying medications that they have had previous allergic reactions to as patients moved across healthcare settings. For example, one participant was prescribed multiple times a selective serotonin reuptake inhibitor which they were allergic to and was recorded in their patient records and they described a breakdown in trust as they did not feel listened to or in partnership in their own care. The importance of systems and tools at home to manage complex medications to prevent safety issues was often unrecognized but when they broke down it highlighted the importance of them generally preventing medication-related patient safety issues. One participant described the importance of systems to manage their medications to avoid accidental overdoses of medication to manage schizoaffective disorder:

I wouldn't survive without my pillbox …I do my pills every week. I think if I didn't have that I'd probably be in a bit of a state. One time I accidentally took an overdose of the sodium valproate …I rang the support team and they said, well, you need to get to hospital straight away. (PS17)

Most participants described supporting family members (usually elderly relatives) or friends as informal carers and described it as often being unrecognized. They detailed the complexity of supporting relatives and coordinating care whilst supporting their family members. This often involved remembering the person they were caring for (often complex) medical history, multiple appointments as well as the everyday logistics of getting them to appointments. Participants described adopting this role with little support from health services and the additional burden to them. The following participant described the challenges of caring for their mother and getting her to attend hospital appointments. Her mother had multiple health conditions, needed a wheelchair and had memory problems and by the time they arrived at the consultation, she had often forgotten key things that needed to be discussed.

I think it [PSG-PC] would [useful]. Because [mother] was very reluctant to go to a consultant at the hospital. Mainly because of the disruption of going …So, if you've got what you want to ask written down, you can concentrate on making her comfortable in our case, and you could always refer to this when you got there. You're not worrying about oh will I remember everything. (PS02)

Levels of involvement of patients in patient safety in primary care and the role of the PSG-PC

Participants' accounts detailed a continuum of levels of involvement in patient safety in primary care that they would feel willing or able to adopt which would influence whether they would want to use the PSG-PC. For some participants, this meant that they would not use the PSG-PC, other participants described selectively choosing sections that were relevant to them and not using other sections. This continuum was not necessarily static but depended on a combination of contributing factors (such as their health status, their perception of the role of the patient or implicit boundaries of responsibility for different elements of care). For some participants, they viewed being involved in patient safety as part of being a ‘patient’ and their patient identity and working in partnership with their GP.

I think that is good for people to have ownership over their medical problems anyway and work, sort of, almost in partnership with the GP to figure out, you know, how to move forwards. (PS09)

Conversely, some participants described their involvement with their healthcare professional in more transactional terms of trying to get the most out of a short consultation which threatens the development of trust and reinforces power dynamics that prioritize the professionals' agenda with limited to develop relationships. For example, the following participant described the challenges of a 10-min appointment and felt pressure to not overrun whilst also trying to raise issues that they wanted to discuss:

I think [consultation] could be a bit longer …it's about ten minutes. So, you just have to keep to the point. It's not social …It's more to the point and that's it. (PS02)

A few participants did not feel they had an explicit role in patient safety as they felt that this was within the healthcare professionals' roles to maintain the patient records as accurate and up to date. For these participants, initially, their accounts depicted a clear boundary of responsibility in healthcare settings with them not necessarily identifying their role as part of an implied ‘safety net’ but the PSG-PC challenged their assumptions about their role and they could identify the potential to ensure information held about them is accurate.

And the doctors, with all the computers and what have you should know what I'm on anyway. And they should be able to refer back to that [patient record] …not always true …I think it [PSG-PC] would be a very good idea. (PS02)

One participant recognized that the questions and prompts in the PSG-PC may have helped them to plan as they became ill and may have helped to avoid delays in diagnosis or management which they experienced.

This [to feedback] never occurred to me when I first became poorly and I think I could have prevented a lot of what went wrong …You know, my management of my illness a lot better. (PS17)

Another component of involving patients in patient safety can include being willing to feedback (either positive or negative) to healthcare professionals. Some participants were comfortable giving and described it as just part of being an informed patient.

Yeah, it's good for them to know, good for the patient to know, you know, feedback what's happening, what's going to happen? You know, say how you feel, and stuff like that, you know. (PS03)

Those participants who had provided feedback described their uncertainty about how to provide feedback in general practice and often felt unsupported. For example, the following participant did not know how to make a complaint about their GP and felt disempowered when told to speak to the GP directly:

I've made a formal complaint in the past about one of the GPs, and I did a letter to the practice manager because that's the only thing I knew …I didn't know anything about the complaints procedure, all I knew is to write to the practice manager rather than the GP themselves, but then they asked me in to speak to the actual GP themselves, so I didn't take it any further. (PS16)

The work of embedding the PSG-PC for patients into routine interactions with primary care

Participants described a range of ways that they could identify the PSG-PC that may be relevant to themselves or others. For example, most participants identified that the PSG-PC could be used as an aide memoire to support them during a consultation to remember key points or questions they wanted to raise. Other participants reflected that the PSG-PC layout was logical linking with the sequencing of a consultation and could help reduce the ‘door knob’ effect which is particularly important for participants managing long-term or complex conditions as they carried information across healthcare providers. The following participant described how they would get anxious before an appointment and then forget the main points they wanted to discuss with their GP:

Yeah, you could have that as …a prompt sort of before you go in and say, right …Whereas at the minute I think you're so worked up at going anyway and, like you say, you do forget, you know, when you come out of it and you think, oh, maybe I should have asked that, but then that time's gone …so, it's making the most of that time. (PS01)

Some participants thought the PSG-PC would be particularly useful as a prompt if trying to introduce sensitive or potentially embarrassing symptoms.

[PSG-PC] just gives you that route as to train of thought …you know, phrasing wise …How can I announce it, do I just give it to the GP blunt …a prompt, you know memory wise, you know, how to introduce it …sometimes it's difficult to think of ways to, sort of, like, introduce the, you know, whatever you've got. (PS03)

However, in contrast for a few participants having a prompt could potentially be an additional burden or distraction from a consultation and additional task loading could overburden them. One participant described their experience of consultations where they felt that adding in the guide would distract them as they already found it challenging to interpret the medical terms.

And sometimes when I'm in a session with the doctor, I'm so absorbed by trying to get out what the issue is, and trying to talk to them, even though I have notes of, okay, have I talked about this, and this, I can't comprehend them, it's like it becomes all jargon when I'm inside the appointment. (PS18)

Whilst most participants recognized that the PSG-PC could be useful it was typically for particular, more relevant sections and they would selectively use components depending on what they needed at that time.

Just from what I've seen maybe there were things there that weren't important for me but I know that they are important for other people who perhaps have less understanding or through age are not comfortable. (PS13)

Participants were shown a paper version of the guide and participants discussed a range of opinions for using an online or mobile application-based version with the majority wanting both options as a way of making it accessible and inclusive.

Both [paper and digital formats] for me …I think I prefer paper, but obviously, I know the way the world is going most things are going electronically anyway …I think that some people feel that they are being, not left out, sidelined because they haven't got the accessibility to a computer, whereas everyone can get a pen or pencil. (PS08)

The majority of participants expressed that they would use the PSG-PC, however, one participant felt that using the PSG-PC would involve them taking on the role of the GP.

I actually think this booklet, it's like the patient has to do the GP's job for him. (PS16)

DISCUSSION

The study analysis highlighted a more nuanced understanding of how patients and carers want or the extent to which they feel able to be involved in patient safety in primary care. In particular, it highlighted the invisible work and often implicit work that patients and carers are already doing to foster fostering patient safety (e.g., avoiding serious adverse events by identifying medications for which participants had a known allergy). The PSG-PC could support patients and carers to draw out this tacit patient safety knowledge and make it explicit and therefore has the potential to support patients to be active in patient safety and their care. However, for some participants, the PSG-PC could add an additional burden when they are already overwhelmed with their health or the responsibility of caring for a family member. Other studies have highlighted patients who may wish to raise safety concerns may be vulnerable to or perceive a power imbalance that may influence their willingness or ability to raise patient safety issues if they would like to.[12,40] The systems in which healthcare provision occurs can reinforce existing inequities and influence patients' ability to raise problems or their perception of entitlement to care which creates a context in which involvement in patient safety may be limited.[65] Despite this, the majority of participants in this study considered the PSG-PC that could support their involvement in patient safety, and they would selectively use the parts of the guide which were most relevant to their particular context. This suggests that the PSG-PC might be considered a complex intervention with the safety issues that it aims to address are dynamic and emergent. This is an important component of conceptualizing how these types of interventions aimed at involving patients in patient safety can be responsive to support the adoption or nonadoption of an intervention in the future.[66] Embedding the

PSG-PC more broadly into care and consultation pathways was identified as an important part of implementing the PSG-PC by patients but was deemed limited unless healthcare professionals were also engaged with its use. Medication management is one area where patients and carers often more explicitly identify patient safety issues.[67] Participant accounts often depicted taking this role for granted and not linking it to patient safety which may influence their willingness to adopt an explicit role in patient safety as this could be seen as additional work rather than recognizing their existing contribution to enhancing safety. Furthermore, whilst patients may have a more clearly identifiable role in medication management for patient safety, the role of communication is also an important contributing factor.[15,40] This is a multifactorial component with the timeliness and clarity of information a key dimension of fostering an environment in which patient involvement in patient safety may occur.[12,15,40] Participants' accounts depicted assumptions about the information that the general practitioner had available during the consultation and the accuracy of the patient record. It was often taken for granted by participants that this information would be up-to-date, accurate and used to inform consultations and for participants that had this belief, it limited the information participants felt was necessary to include during a consultation. However, patient records may be poorly maintained, have missing information and have inconsistencies between entries.[68] This is particularly important as the information that patients or carers disclose during a consultation may be based on a potentially erroneous assumption that may have consequences for patient safety (e.g., a medication with known allergies) and places the burden of responsibility on patients to monitor medication prescriptions. It is important to understand these uncertainties and trade-offs conceptually from a Safety-II perspective.[33] The PSG-PC was identified as being able to support this as it may foster better communication and transparency so both patients and healthcare professionals have a more shared understanding of what has happened and what may need to be done in the future.

Another area of patient involvement in patient safety which has received the most attention is the role of patients providing feedback to healthcare providers and particularly in hospital settings.[30,69] For example, in the UK national patient safety syllabus a key component is learning from events and lived experiences but this is based on an assumption that people know how and where to give feedback.[14,15,70] Participants' accounts within this study challenged this assumption as participants' accounts depicted a range of preferences and experiences for giving feedback in primary care with most not knowing how to give feedback or having negative experiences if they had given feedback.

Previous research has examined how patients conceptualize responsibility for health care and the boundaries between patient and professional roles that involving patients in patient safety may occupy.[12] This study extends this to identify how the role that a patient or carer may identify as being willing or able to adopt to foster patient safety may influence their use of the PSG-PC. This is of particular importance as much of the national and international policy and academic literature highlights the importance of involving patients in patient safety but there is considerably less focus on how to do it.[1–12,14] Accounts depicted a continuum of levels of involvement in patient safety as opposed to a categorical approach (i.e., they are involved or not) which was flexible and dynamic depending on a range of contributory factors. Similarly, Heavey et al.[12] found with patients recently discharged from the hospital, that their narratives of responsibility for safety in the hospital were based on either personal expertise and/or a duty of self-care or alternatively an expectation of safety was the focus of professionals duty and expertise. Accounts in this study identified a varying role of expectation for involvement with accounts ranging from more individualized, personal responsibility, which resonated patient safety conceptually with self-care, through to it being the GPs role. This influenced the extent to which participants felt able to shift the boundaries and expectations of their role which in turn influenced their willingness or ability to share responsibility for patient safety and the PSG-PC.[71,72]

Strengths and limitations

This study draws on patient and carers' experiences and prospective accounts using the PSG-PC to support them to be more active in patient safety which extends previous work which identifies that patients should be involved but crucially not *how* to involve them using a co-design approach.[20] This is a strength of this study as it supports an understanding of how different participants identify if they feel able and willing to be involved in patient safety in primary care. One limitation of this approach is that, while we obtained participants' initial thoughts participants had not used the PSG-PC in practice. Future work needs to examine how patients and carers use the PSG-PC and whether it is acceptable.[28] In this study, due to resource constraints, only patients and carers were interviewed and it is a limitation that we did not interview other stakeholders. Interviews were conducted by members of the research team and whilst participants were asked open questions and purposefully questioned about what they did not like about the guide, in line with intervention development studies.[57] Participants were recruited through a snowball sampling approach and whilst the team recruited people from a range of backgrounds there is a limitation that only the views of people who volunteer are reported and the views of people who may not have the time or capacity were not included.[73] Participants' views were based on a paper version of the PSG-PC and future research should explore the development of mobile phone applications. One main limitation of the study is that the PSG-PC was only available in English as this was a prototype and the first stage in developing and testing the PSG-PC, further work is needed to culturally adapt the PSG-PC or develop versions for marginalized groups that may have specific needs (e.g., text-to-speech option for people with vision impairment). The changing context of health care since the start of the severe acute respiratory syndrome coronavirus 2 (SARS-CoV-2) COVID-19 pandemic has acutely brought to the fore the need for a nuanced approach to patient involvement in patient safety that is responsive to changing models of care (e.g., video chat).

CONCLUSION

By examining patients' and carers' views of being involved in patient safety this study shows the need for a nuanced and personalized approach. The PSG-PC has the potential to support patients' and carers' involvement. This study identified challenges to assumptions about patients' and carers' willingness and ability to be involved in patient safety (e.g., that all patients want to be involved in patient safety) and the potential adoption of the PSG-PC in routine practice.

AUTHOR CONTRIBUTIONS

Rebecca L. Morris led the design of project, data collection, analysis, interpretation and drafted the manuscript. Stephen Campbell conceived the project and Sally Giles collected data. All authors developed the analysis and interpretation and critically contributed to the prototype development and manuscript.

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CONFLICT OF INTEREST

The authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

Research data are not shared.

ETHICS STATEMENT

Ethical approval for this study was granted by Yorkshire &The Humber-Sheffield Research Ethics Committee (REC reference: 16/YH/0496).

DETAILS

Subject:	Anxiety; Health care policy; Patients; Medical personnel; Safety; Caregivers; Thyroid gland; Co-design; Primary care; Chronic obstructive pulmonary disease; International policy; Citizen participation; Public involvement; Patient safety; Chronic illnesses; Arthritis; Asthma; Health care; Recruitment; Interviews; Blood pressure; Patient satisfaction; Hypertension; Individualized; Family physicians; Chronic fatigue syndrome; Willingness; Qualitative research; Hyperlipidemia; Embedding
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Observations on strategies used by people with dementia to manage being assessed using validated measures: A pilot qualitative video analysis

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ABSTRACT (ENGLISH)

Background

Analysis of video data was conducted of validated assessments with people with dementia as part of a feasibility control study comparing a lifelong learning service with other dementia services.

Objective

The aim was to provide a new perspective on what occurs during the assessment process when using validated measures in research and explore which strategies people with dementia use to manage their participation.

Design

Video recordings were made of pre- and postintervention assessments of people with dementia. An initial pilot analysis of 10 videos of the pre-assessments was conducted.

Setting

Lifelong learning services and other dementia services situated in six municipalities in Northern Denmark took part in this study, with 55 people with dementia participating.

Results

The themes identified were: ‘State of mind’ and ‘Mental resources’, showing how these aspects influenced the participants' reactions and the strategies they used.

Discussion

The results are discussed in relation to how individual personality traits influence the assessment process and the way a person with dementia will manage the situation.

Conclusion

The assessment situation is complex and can be influenced by the strategies adopted by individuals with dementia as they try to manage the assessment process.

Patient or Public Contribution

People with dementia supported the decision-making for the choice of validated measure used within this study.

FULL TEXT

INTRODUCTION

Growing evidence shows that people with dementia can report their views and experiences in research.[1,2] However, an area that has been little researched is how people with dementia react while being assessed using validated measures or what strategies they use in this situation. Validated measures in health research enable the assessment of the quality of care, the effectiveness of interventions and supporting decision-making in clinical care and intervention settings.[3] Such measures also enable an understanding of the cause and effect of health conditions and interventions. They have an important role in testing hypotheses to support decision-making in health and social care.[3] In dementia care and research, the use of validated measures also helps to provide a perspective on the way an individual's dementia is progressing, and therefore to understand how best to support them. Such tools are also an important part of the diagnosis. In the United Kingdom, NICE[4] recommends the use of assessments of cognition, functional ability and mental state when diagnosing dementia. More is known about the experience of receiving a diagnosis of dementia than the impact of participating in validated measures. The communication of a diagnosis of dementia requires sensitivity, indicating that the process can be stressful and overwhelming.[5] In Xanthopoulou and McCabe's[6] study, participants with dementia reported they found the assessment outcome and subsequent diagnosis difficult to hear, and they were scared and upset to receive the diagnosis.

Literature on using validated measures tends to report the outcomes and rationale for their validity and use. Literature reviews on using measures with people with dementia, and more widely in using patient outcome measures, have cautioned not to burden participants or cause harm.[7–9] However, there is little discussion about what this means in practice. Ward et al.'s[10] review on evaluating cognitive stimulation highlighted that insufficient information is given about how the assessment of people with dementia is conducted. They also noted that little is reported on how these tests are experienced and what impact there is on the subjective interpretation of the tests by people with dementia, something that has been criticized in cognitive stimulation effect research.[11–13] However, it is important to identify measures that are acceptable for both the research community and for those diagnosed with dementia, including reducing any impact in terms of distress, confusion, anxiety or burden in participating.[8] Heggestad et al.[14] argue that the assessment process can be humiliating, and people with dementia may experience a loss of dignity in taking a test. This can have a negative impact on how they see themselves and can be a reminder of the progression of their dementia. Therefore, research to explore how people with dementia experience an assessment process will provide insight to support them through this process be it for diagnostic or research purposes.

This paper provides findings from observed assessments used in a research setting. The authors provide insights into this little-researched area, and ways to support the person with dementia, researchers and clinicians undertaking assessments. This is the second paper to present findings from this research, with the first available through Thoft et al.[15] The first article highlighted the strategies of the researcher in undertaking assessments using validated measures. The aim of this paper is to provide a new perspective on the assessment process used in research and explore how people with dementia react and identify strategies they use when being assessed with validated measures.

MATERIALS AND METHODS

This paper presents findings from a video analysis of conducting validated measures with people with dementia. This was part of a wider feasibility and pilot study that was conducted on lifelong learning services in Denmark. Lifelong learning is an education-led programme that provides lessons to support cognitive function, decision making and activities of daily living. It is based on the premise that people living with dementia can learn, develop and grow.[16] The project assessed an intervention group (Lifelong Learning intervention) and a control group (treatment as usual, e.g., services at day-care centres). The study was conducted in six municipalities in Northern Denmark. Participants were tested at the outset of the study and after 5–6 months. Participants were assessed using five validated measures: Mini-Mental State Examination (MMSE)[17]; Quality of Life in Alzheimer's Disease Scale (QoL-AD)[18,19]; General Self-Efficacy Scale[20]; Rosenberg Self-Esteem Scale[21]; Hawthorn Friendship Scale.[22] A detailed method, background to the wider study and facilitator strategies are presented in Thoft et al.[15] and Sørensen et al.[23] This paper provides an overview of the methods in relation to the video analysis.

Public and patient engagement

People with dementia and staff from the lifelong learning intervention took part in a workshop to identify the most appropriate measures to use for the wider study. Their input was gained through discussions about what they felt was important to research about the intervention and informed the final choice of validated measures used. These workshops will be the focus of future analyses.

Video analysis

Fifty-five participants were recruited into the main study (*n* = 30 intervention group; *n* = 25 control group). All participants undertook pre- and postassessments, which were recorded using one video recorder. This was positioned to capture the participants' facial features and reactions, while also capturing the table, paperwork and side/back view of the assessor. Videos were chosen because they captured both verbal and nonverbal reactions and enabled multiple reviews of actions and behaviours which may not be identified in person.[24]

The decision to conduct an initial pilot analysis was based on the pragmatics of undertaking the analysis and testing the outcomes of this approach. Video analysis is time-consuming, requiring multiple viewings, by several researchers. To ensure that this would elicit valuable and viable data, the team first conducted this as a pilot stage, with plans to extend this analysis. This paper, therefore, presents the findings from this initial stage. A stratified sample of 10 pre-assessment videos was analysed. This stratification included: equal distribution across the intervention and control group (*n* = 5 per group); each locality in which the service was delivered; level of dementia (high and low MMSE-score) and diversity of gender. The pre-assessment videos were chosen to avoid recall or familiarity with the measures as this could be a risk if including the postassessment videos. The demographic profile of the participants from the analysed videos is reported in Table 1.

Table 1 Demographic of participants in analysed videos.

Video no.	Gender	Type of dementia	Age	Intervention/control	Video length
7	Male	AD	74	Control	57 min 37 s
31	Male	AD	77	Control	25 min 54 s
34	Male	OTHER	73	Intervention	58 min 30 s

49	Female	OTHER	65	Intervention	31 min 31 s
55	Male	Not specified	89	Control	37 min 30 s
60	Female	AD	62	Intervention	37 min 5 s
71	Male	OTHER	54	Control	25 min 49 s
75	Male	OTHER	74	Intervention	22 min 33 s
82	Female	OTHER	68	Control	33 min 52 s
86	Female	AD	76	Intervention	25 min 45 s

The videos were analysed using an adapted version of Ridder's[25] video analysis approach. The identified videos were watched in full by four members of the research team to develop an analysis framework, which was tested and adapted using one video, and focused on identifying participants' reactions. The resulting framework was used to code all videos. A video graph was developed for all the videos in Excel. This notes by timeframe each reaction to the assessment situation, including physical movement, facial expressions and verbal comments, alongside researcher reflections on the action. Viewing the videos in full and reviewing this graph allowed the research team to identify clips for a deeper microanalysis that explored key moments and interactions during the assessments. These were coded alongside the verbal interaction to provide a detailed account of what occurred. Thirteen clips were chosen from the 10 videos for further microanalysis (see Table 2). The final analysis stage was to draw themes from across each microanalysis and the video graph (see Thoft et al.[15] for further details of the method, video and participant demographics).

Table 2 Extract from the microanalysis.

Video no. 49; (22.07–25.12): Meaningful event: Facilitation and interpretation of questions	Assessment of event ‘I feel/think…’ or ‘The student seems to…’	Reflection of event *How can you see this response, emotions, engagement, interactions*	Supporting text *Transcription of clip*
Description of the event, who was involved and what occurred	*Interpretation of what occurred, taking into consideration the student's perspective*	*Researcher's reflective comments on the action*	*Extracts from the transcription*

R and P are sitting across each other around a round table. P is facing the camera. There is a question paper on the table and R has a pencil which she is using to show the questions. R asks the Self-efficacy question. P is leaning forward and has one hand on the table holding a pencil and the other is held up against her mouth. She is looking at the paper.	It feels as though the atmosphere is relaxed and they both seem at ease.		Self-Efficacy—When I meet a problem, I am able to identify several solutions.
P leans back and adjusts her clothing and then leans forward again in the same position. There is a pause as P thinks.	It feels as if R is giving P time to think as P seems to be concentrating on how to answer the question.	This is an example of facilitation as it recognizes that a person with dementia needs time to understand and process through their answer.	
P moves her hand from her mouth to the table and talks about a previous answer and that her answer now is going against the previous answer. She points towards the paper as she talks and is looking at the paper. There is a slight joking tone in her voice and a slight smile.	It seems that P is questioning herself and her previous answer. It does not feel as though P is upset by this as she smiles and has a jovial tone to her voice. It may be that the paper is being used by P to recall her previous answer as she points to it and is focused on the paper.	There is recognition and recall by P of her previous answers. This moment shows that P is reflecting on the answers and remembers her answers. The paper acts as visual clue to support P's attention, focus and memory.	P—Yeah, but well, it contradicts the other thing, right? But well, it can be that I should… (pointing at the paper).
Evaluation *Identifying the themes emerging from the clip*	Facilitation—Some good facilitation is shown through listening and giving time for the participant to answer and giving options for the responses. There is a dilemma in the role of the facilitator in how much to support or not during the test and how much time the participant needs to answer….		

As Table 2 demonstrates, the video analysis provides a description of the action alongside the reflections and observations of this action by the researchers. Supporting evidence is also provided through transcripts of the dialogue in the videos. The results are presented as observations that were made by the researchers with supporting statements provided in the form of descriptions of the action or participant's quotations.

The research team consisted of two senior researchers with previous experience in leading dementia-related research, and research with the lifelong learning service in Denmark (called the Aalborg Dementia School, at The Knowledge Centre for Dementia, Aalborg Municipality). One had expertise using video analysis methods. Two other researchers completed the team, having a background in nursing and expertise in qualitative research. All members of the team undertook the analysis in Denmark working in pairs to analyse each video. The researcher with video analysis expertise provided training, with review sessions at intervals during the analysis for the team to discuss the approach and how to correctly log and review the data.

Ethics

Participants were recruited through their service. Each service attended a meeting with the lead researcher to inform them about the aims and process of the research. Participants were informed about the project through a participant information sheet and were able to discuss this with a member of the research team. This emphasized that their participation was voluntary and was not related to their continued use of their respective services. Participants completed a consent form before participating in both pre- and postdata collection phases. Where required, consent was discussed and gained with support from a family or staff member, although no proxy consent was used. All participants were self-consenting. Danish legislation requires research studies to be based on informed consent and not on ethical approval from a national or public agency.[26] The video recordings were not allowed to be shown outside of the research team due to the requirements of confidentiality and anonymity as stated by the Danish ethical requirements. All names used in the article are pseudonyms. In keeping with good research practice, the Regional Committee on Health Research Ethics was also consulted. It was judged that no further application was needed in relation to LBK nr 1083 of 15/09/2017 definition of a Health Science Research Project and the Committee law §14, stk. 1, jf. §2, nr 1-3. These reference Danish ethical laws and recommendations of the Danish Ministry of Higher Education and Science that ensure participant safety and rights under the Danish Code of Conduct for Research Integrity.[27]

RESULTS

The 13 clips varied in length from 17 s to over 3 min. This reflected the nature of the interactions, which were often short responses to questions asked during the assessment. Two core themes were identified about the way people with dementia react and the strategies they used while being assessed using validated measures. These were: ‘State of mind’ and ‘Mental resources’.

State of mind

State of mind was observed as both positive and negative, with a positive outlook supporting the person with dementia to find the assessment process less stressful.

A positive state

An individual's state of mind could impact how they experienced and responded to being assessed. State of mind was identified through emotional state/mood, emotional responses and body language.

Participants commented on their emotional state, for example, Anni said that she is normally a ‘cheerful person’. This was also apparent in the way she presented during the assessment, especially when recalling memories of her family. She smiled and laughed as she shared her thoughts. Even when she responded incorrectly, Anni smiled while responding. For example, Anni was asked to provide the address where the assessment was taking place (MMSE), she did not know, but smiled and laughed as she recalled it was near a 10-pin bowling alley where her husband was currently playing.

Arne also commented on his mood. He asked the researcher for feedback on whether he was responding correctly during the self-efficacy measure. The researcher commented that there was no right or wrong answer only what Arne was feeling. Arne commented that he was in a ‘good mood’ and together they reflected that if he had been in a bad mood, it could have impacted his responses:

Arne: Yes, yes, but now I'm in a good mood today. (Smiling and laughing)

Researcher: You are right, because if you are in a bad mood, I think it would look different—don't you think?

Arne: Yes, I think. (video 75)

This exchange suggests that when in a good mood, a person may respond more positively than when in a bad mood, thus having a potential effect on the test situation. The easy relationship observed between Arne and the researcher may also have had an impact on his mood, helping to ease the test situation.

A negative state

The mood exhibited by participants was not always positive with some showing signs of disappointment or frustration, characterizing a more negative state of mind. For example, Lone showed disappointment when she could not recall her surname. Her body language and expression changed. She leaned forward, her smile disappeared into a sigh and she looked to the side while saying: ‘Suddenly I couldn't remember it…’ (video 31). Even though Lone succeeded in answering the question, given time to think, her tone and body language expressed, what the research team considered disappointment. It may also have been a moment of recognition of the challenges caused by her dementia.

Participants also expressed frustration. This was mostly observed in relation to the participants' loss of ability to answer questions. This was usually directed towards themselves and their dementia. For example, Hans was telling the researcher about his former language skills:

Earlier, I had five languages (showing five fingers). I was good as hell at languages and now I can, I can just speak a little Danish …And Swedish (talks in Swedish) I can't speak that anymore—I can't understand the damn prose. And that sucks when you are on a visit there. (video 34)

During this dialogue Hans was initially relaxed in his body language, resting one arm on the table and leaning his head in the other hand, while speaking in a soft tone of voice with a slight smile on his face. This changed as he talked about his declining skills. He became increasingly restless, leaning backwards and quickly forward while pointing with his finger, brows furrowed, raising his voice and firmly placing both hands on the table. This was observed as frustration towards his failing abilities and recall of the skills he used to have.

On some occasions, participants showed contradictory verbal and nonverbal expressions. This was observed in Bo who was asked to repeat the three words in the MMSE. Bo was smiling and laughing without being seemingly happy. Bo had a tense, forced, almost unnatural smile, and although he was laughing, his body language showed nervousness or discomfort, as he was tapping his finger and moving his legs, looking away and leaning back while answering: ‘That is worse! (laughing)’ (video 7). This was observed as a reaction to not being able to answer the question.

Other verbal and nonverbal signs were observed. Examples of this include looking down at the table seeming disappointed, changing tone of voice and body language showing anger and frustration, for example, making strong hand gestures and smiling ruefully to express discomfort when being confronted with difficulties due to dementia.

Mental resources

Participants were observed to use the mental resources of reflection, humour and bodily movement. All the participants at times were engaged and concentrating, showing different skills to help complete the assessments.

Reflective skills

Reflective skills were observed in many participants. When Grethe was asked a question about her marital relationship (QoL-AD), she replied that the responding category ‘excellent’ did not fit her usual wording; ‘It's probably excellent. No, good. I have difficulties using the word excellent—good means more to me than excellent’ (video 82). Grethe was able to reflect upon personal preferences towards the meaning of the categories showing her language and interpretation skills. The researchers experienced that several participants found it unnatural to use the category excellent.

Some of the participants also talked through their reflective process. Bente recognized that an answer she gave in the self-efficacy questionnaire about ‘When I am confronted with a problem, I can usually find several solutions‘ contradicted her earlier answers where she said could not manage difficult situations or unexpected events:

Yeah, but well, it contradicts the other things, right but it can be that I have to change …I think about different solutions that is what I am thinking about? (fidgeting with her shirt, looking down at the paper). (video 49)

She explained that she thinks about different solutions, but assesses her abilities as ‘moderately true’ and that she can come up with solutions to her problems. Other participants reflected by comparing their abilities before their dementia diagnosis and their present abilities, and by comparing their skills to those of others. The participants would use words such as ‘before’ and ‘now’, showing that their answers were considered in light of their diagnosis. This was particularly noticed during the QoL-AD, as Anni commented: ‘Well, normally I would say it is good enough, I think so. I don't think it's bad, my memory’ (video 86). Even though the participants were confronted with their decline, they were observed to identify several solutions on how to handle a problem when asked in the self-efficacy test and were aware of managing dementia in their everyday lives by seeking help from others, as Bente stated: ‘I can get help’ (video 49).

Supporting concentration

The participants took the tests seriously, and these were completed without breaks (although these were offered), and by asking questions. Their concentration was particularly noticeable by their use of physical contact with items, such as pencils or test paper. Here the items seemed to work as a physical prompt or sensory stimulus. For example, when Knud responded to the self-efficacy question ‘I am able to do things as good as most people’ (video 71), he was observed to follow the questions with a pencil and took time to think through his answer. The test paper for all the measures, apart from the MMSE, was placed on the table for the participant to see. Some used this, reading the questions, and pointing or touching the paper as they responded. The visual cues provided by the paper and pencil were observed to support their ability to answer.

Participants were also observed to use pauses, and look to the side before answering a question, seemingly to give their response consideration and make sure they gave an accurate account of their experience. However, looking off to the side also led to a loss of focus as the participants could lose track of the question asked.

Shared connection

Participants often looked at the researcher for confirmation or support when answering the questions. This sense of shared connection was also evident through their use of humour, which was observed with some participants making a joke about the question or their answer. This seemed to act as a coping strategy to mask their insecurity or difficulties in undertaking the assessment. Bente was joking about her handwriting, commenting: ‘my writing is not good’ (video 49) while apologizing to the camera, leaning back and laughing. Anni used laughter when she was not able to recall what day it was during the MMSE test: ‘Thursday? Wednesday …The days have been changed over here. Now I can't remember if its Wednesday or Thursday! (laughing)’ (video 86). This seeking confirmation and the shared humour seemed to establish a form of shared connection between the participants and the researchers.

Nonverbal communication

Nonverbal communication in the form of facial expressions, gestures and bodily movement was observed across all the videos. Gestures were observed as a strategy to support individuals when faced with symptoms of their dementia, for example, challenges with language. During the MMSE Hans used gestures to explain which region he lived in. He drew a map of Denmark in the air, pointing towards the Northern part of Denmark. Hans was not able to verbalize his answer so used nonverbal communication instead. Also, during the MMSE in response to which floor they were on, Arne looked out the window, gesturing to show the building was built on terraced land. By doing this, he showed awareness of the building's challenging geographical layout, even though he was not able to verbally provide the correct floor level.

Participants were also observed to use movement, fidgeting and self-touch, for example, hugging themselves, keeping hands clasped or folded, resting them on the table, leaning backwards and forward in the chair and tapping fingers against the table. It was noted that these movements were most often used at times of potential stress.

DISCUSSION

This paper sheds light on a little researched area, to understand what takes place during a formal validated assessment process in research with people with dementia. The rationale for exploring this interaction was twofold, to provide an understanding of the assessment process and people with dementia's reactions to this, and to identify ways of providing support for the individuals at a time that could be stressful.

One of the key findings related to the way personality and mood can influence a person's response, as one participant stated, being and calling oneself a cheerful person can be a way of showing one's personality and may affect the reactions towards the assessment. This individual did not seem to react negatively regardless of whether her answers were correct or not. It may be that this participant lacked insight into the progression of their dementia and how this affected her memory. Stress, hope or personality have been reported[12] as having the potential to impact assessment scores, while people with dementia and caregivers have identified that individual traits can influence their choices during research.[8] How these factors can affect a score requires greater investigation, especially when these assessments are used to determine care pathways and the impact of interventions.

Another key strategy was the use of touch and movement to support people with dementia, whether this was through fidgeting, hugging themselves or touching the table and/or the answer sheet. This worked to ground the individual in the moment and act as a comfort and memory aid. People with dementia have been observed[28] to use touch to connect in the moment and that this can support the sharing of memories, while the touch of paperwork or holding a pencil can support attention and concentration in a research context.[29] Such connections may indicate increased physical and cognitive arousal, and fidgeting has been associated with increased motor and sensory activity in the brain.[30] While there is limited research to explain the function of fidgeting, there appear to be links to increased neural activity and arousal[30] that may be a physiological support mechanism for people with dementia under test-like situations. The participants in this study were observed to fidget by tapping the table, moving their legs and making varied hand gestures, using this nonverbal communication as a way to express their emotions, both positive and negative and to support their concentration.

Stress can support our decision-making and social interactions, however, too much can negatively impact our behaviour and our cognitive function.[31] One way to manage stress is through tactile stimulation.[31,32] Self-touch has also been associated as a coping mechanism for managing stress, such as hugging oneself or touching a face or hands.[31] Skovdahl et al.[32] describe touch as a way of supporting communication, particularly nonverbally. Therefore, the provision of a pencil or paper as a tactile object for people with dementia to use, and an understanding of body language may be a way of supporting people with dementia in undertaking an assessment and helping them to answer to the best of their abilities.

Humour was observed to work as a coping strategy when responding to the validated measures and seemingly acted to smooth over worries or tensions and to mitigate where an individual was unsure of what response to give. The use of humour to manage stressful situations, as observed in this study, has also been studied in health-professional and patient interactions.[33–35] Laughter can also result from a release of tension as a ‘basic biological form’ (p. 4),[36] which helps to reduce stress and help the individual to relax. Mallett and A'Herne[37] identified that patients, in clinical settings, used humour to deflect conflict, particularly if associated with criticism. This use of humour may be expected as people with dementia use humour as a form of tension release when under stress.[36] However, the use of humour by people with dementia is also considered a natural part of their communication,[38] and that humour is a strategy which is used as an expression of their ‘personhood and autonomy’ (p. 341). Humour has also been shown to make it easier for mistakes to be made, to laugh about these mistakes and to relieve stress when being with other people.[29] While much of this research has been carried out in clinical settings, the effect of humour is similar to that which was observed in this present study and eased tense situations, supported decisions and showed individual personalities. The use of humour was a coping mechanism that could be adopted to provide a more comfortable setting and ease relationships to aid the assessment.

What is starting to be evidenced is that many factors can impact how people with dementia respond to validated measures. These factors can aid their responses but also may be detrimental. Differences in personality, mood, ways of interpreting questions or response options or responding nonverbally can all influence the final assessment score. As an example of this, in Scandinavian countries there is a cultural law—the Law of Jante—that is drawn from Sandemose[39] and in Anglo-Saxon societies as the ‘tall poppy syndrome’.[40] This sets out certain personality and cultural ways of being, for example, not thinking too highly of oneself, or boastful of one's successes.[39] In an ethnographic study[41] of Jante, it was reported that Danes were often worried about standing out. They downplayed successes and conformed to societal norms, fearing retribution for being too boastful. The use of Likert scales that ask a participant to respond positively about one's abilities, as in this present study, therefore may be affected by this Law of Jante and how a participant responds. This law was noted by the researchers to be particularly relevant to the older generation and may have resulted in ‘good’ rather than ‘excellent’ responses, as one participant exemplified. It is therefore a question about how researchers take account of this within the way they score and report their findings. Further research is needed to understand how much these factors need to be considered and how they are managed. At present, there is little evidence that these are considered, and a possible starting point would be for researchers to monitor such factors and include this within their write-ups so that a fuller picture develops.

Another factor worth consideration is the involvement of people with dementia in determining the core domains that led to the validated measures used in this study. This was viewed as an important aspect of the study as it ensured that the measures were reflective of the needs and experiences of those who used the service. This is not often considered when deciding on validated measures for people with dementia.[42] Evidence from patient outcome measures research finds that such inclusive practice can lead to greater health and practice benefits, and more reliable evidence associated with the experiences of those being assessed.[43] The production of guidance to ensure a robust and open process is followed would be a valuable resource. An example from the findings of this study also highlights the need for people with dementia to be involved in the use of and design of validated measures.

The authors acknowledge that while some findings from this study may be expected, the way that validated measures are experienced by people living with dementia is not often considered in the literature. Therefore, it is not known if or how researchers or clinicians take account of mood, personality, and so forth, when conducting assessments. The authors believe that this is an aspect that could be more openly discussed as it can impact the outcomes for evidence of the impact of an intervention, but more importantly, on the care a person with dementia receives. Only with more open conversations and research can we find a way to mitigate these variables or develop more guidance on when an assessment should or should not be used. For example, the research team are taking lessons learned from this pilot forward for a new larger-scale evaluation of the lifelong learning service across Denmark, Norway and the United Kingom where it is now being run, and this has influenced the training provided to assessors on how to undertake the assessments.

Limitations

The key limitation is the number of videos analysed in this pilot analysis. The ability to generalize the findings is limited, however, this study has provided novel information on a situation that is not often researched. The identification of factors that could impact how people with dementia react and respond to validated measures warrants further investigation. People with dementia were not part of evaluating the assessment process to share what or how they had experienced the situation. This may be an area for future research so that findings are not based on observation alone but also on personal experience. A further limitation was the potential for the researchers' responses and behaviour to impact participants' responses, potentially influencing how they responded. Further research or training on how to mitigate this would be a valuable consideration for the future.

CONCLUSIONS

What has emerged is the complexities of assessing people with dementia. People with dementia are using different strategies to manage their emotional responses to being assessed. These responses may hinder or help their answers and as such this opens a potential area for further research as responses to validated measurers may not provide an absolute answer. They rather need to be considered in relation to how the individual responds physically and verbally during the assessment and their cultural background. What this study provides is insight into the assessment process, highlighting that there may be more to consider when interpreting findings from validated measures and that there are approaches that can support the person with dementia to manage what can be a potentially stressful situation.

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CONFLICT OF INTEREST STATEMENT

The authors declare no conflicts of interest.

DATA AVAILABILITY STATEMENT

Anonymized data are available on request due to privacy/ethical restrictions.

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'Being a mother is not child's play': The capabilities of mothers in a low-resource setting in South Africa

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ABSTRACT (ENGLISH)

Background
The importance of a child's first 1000 days has now been widely accepted by the medical fraternity. Yet, we do not know much about caring practices in low-resource settings.

Aim
This study aimed to investigate the caring capabilities of mothers in a low-resource setting.

Method
In this study, in-depth interviews were conducted with 18 mothers with children aged 30 months or younger to better understand the arrangements, means and ends that inform developmental health in a low-resource setting in South Africa. The study was conducted in a low-income area, the former black township of Mangaung in Bloemfontein. The mothers were recruited via pamphlets, and two interviews followed. Because of Covid-19, interviews took place via mobile phones, in Sesotho, the local language in the area. Trained fieldworkers conducted, translated and transcribed the interviews. We used thematic analysis and the capabilities approach as the theoretical framework to analyse the responses from the mothers.

Findings
We used the following organizing themes: pregnancy and ante-natal care, nutrition, cognitive and physical development, the home environment and access to health care. Although short-term reactions to pregnancy were often negative, the longer-term responses showed that the respondents have agency. Most of them could change their nutrition habits, breastfeed and receive adequate nutrition support from the public health system. Most experienced joy when their children reached milestones (cognitive and others), although they became anxious if milestones were not reached. They emphasized children's play and had dreams for their children's futures. Technology was often mentioned as playing a role in their children's development. A large proportion of the respondents had disrupted homes (because of absent or abusive fathers), but some had stable homes. Most of them showed substantial capability to overcome adverse home environments. The public health system helped them deal with their health problems and their children's health problems, although it also created anxiety in many cases. Our data show how they develop their capabilities and overcome obstacles organically in the face of resource limitations. Despite pregnancies being unexpected and unplanned and fathers being absent, the respondents accepted the pregnancy, adjusted their diets and social behaviour, showed agency by attending primary healthcare facilities and ensured that their children received the required vaccinations. Their extended families played an important role in providing care. Despite the sacrifices, the respondents expressed joy and helped their children function by eating, playing, socializing, learning and using their senses.

Conclusion
Our sample of mothers have the agency to adapt to the demands of parenthood and childcare and overcome adversity. Our data support the notion that mothers are held disproportionately and unfairly responsible for achieving the first 1000 days ideals. Despite considerable curtailment of their functionings and capabilities, they nevertheless showed agency to ensure their health and their children's health. A holistic approach should consider these findings in designing policy interventions for children's developmental health.

Patient and Public Contribution
We used paid fieldworkers to interact with the research participants.

FULL TEXT

INTRODUCTION

The concept of a child's first 1000 days (from conception to the second birthday) has become prominent in public health. The argument is that if one makes the right ‘investments’ during this period, the child will receive long-term benefits. The research motivating this argument mainly uses quantitative approaches, with large data sets. The idea of investing in the first 1000 days originates from research showing that the circumstances of the first 3 years of life have a far-reaching influence on the brain's final anatomical and functional architecture and consequently a person's psycho-socio-behavioural functioning.[1] Events of this early period influence competency and behaviour in adulthood.[2]

Effective interventions during this period may improve children's developmental outcomes and reduce inequality.[3]

However, the biomedical emphasis on the first 1000 days has unfairly pathologized maternal responsibility. By emphasizing biological processes, policies and programmes, this approach tends to neglect the sociopolitical and bio-political aspects that help or hinder caregivers and families in managing childcare and family life in a way that is meaningful to them. Criticism has been directed at policy interventions that visualize the mother as having no history, that focus on the 1000 days as a target and that aim to enhance the child's prospects while ignoring sociopolitical concerns.[4]

Good nutrition, a stable emotional attachment and the avoidance of toxic stress are primary factors in brain development. Three levels of broader factors are important in a child's development: individual and family (micro), neighbourhood, town and community (meso) and social and political arrangements (macro).[5] In research on the first 1000 days, a biomedical approach dominates. As a result, this research neglects the issue of caregivers and their contexts, especially in low-resource settings. In this paper, we argue that qualitative data add valuable perspectives to biomedical data, particularly concerning the demands of parenthood and the responsibilities that the first 1000 days entail. We draw on the capabilities literature to argue for a more complex approach to the mother's role and child development during this period.

Maternal, neonatal and infant deaths are high in low-resource settings.[6] Most research focuses on understanding and preventing the problem from a biomedical perspective. Papers and books commonly refer to ‘preventing’ these deaths. Appropriate care is also important for neurocognitive functioning and child development. For example, a systematic review found that play and reading improve neurocognitive functioning in children aged 0–3 years.[7] However, providing care should go beyond a biomedical approach. Mitchell notes that: ‘Meeting mothers' needs in the care of their babies is important to mothers, family members and professionals, and deserves greater attention’.[8] A systematic review pointed to the cost-effectiveness of participatory action and learning to improve maternal and neonatal survival.[9] Evidence points to the importance of postnatal education programmes[10] and a dialectical relationship between families and hospital procedures.[8] Maternal depression is also associated with poor child development outcomes and research has pointed to the importance of lay health workers providing support for women suffering from depression.[11] Programmes focusing on maternal depression led to better child development outcomes. In a recent study, Garcia et al.[12] found that, although there was no statistical evidence that fathers' involvement improves child development outcomes, it showed some promise in a low-resource setting. Research also points to the importance of attending antenatal care visits and breastfeeding for child development.[13]

The biomedical approach focuses largely on the role of care in preventing infant and child mortality. But care can also be seen in broader terms. The African proverb that says ‘it takes a village to raise a child’ embodies this broader approach.[14] If it takes a village to raise a child, the mother has collective knowledge and support, material and immaterial, that she can harness for childcare. It is our view, this form of capital is a critical part of the immediate eco-system of the child and essential in the healthy development of the child.

The biomedical focus on mothers' needs is important but lacks an understanding of what mothers do and value and its focus could place undue pressure on mothers to comply with mechanical aspects of caring. Our paper thus uses the capabilities approach to understand what mothers do and value in caring for their children.

The capabilities approach derives from work by Amartya Sen in the 1980s and Martha Nussbaum in the 1990s.[15,16] Sen[17] described this approach as ‘an intellectual discipline that gives a central role to the evaluation of a person's achievements and freedoms in terms of his or her actual ability to do the different things a person has reason to value doing or being’. The term ‘Capabilities’ refers to the abilities that allow people to pursue meaningful ends in their lives.[17,18] They enable people to convert their endowments into meaningful achievements (‘functionings’) and pursue their goals. It thus values meaning and dignity above conventional metrics like income.

The capabilities approach has two central concepts: capabilities and functionings. Capabilities are the freedom and opportunities that people have. Functionings are the doings and beings that people use to achieve their capabilities. For example, travel is a functioning, but freedom to travel is a capability.[18] People's different abilities to convert resources into functionings are called conversion factors. The resources could be personal (intelligence, reading skills), social (public policies) or environmental (climate, location). The difference between beings and doings is important. ‘Being’ could refer to being a caregiver, while ‘doing’ refers to providing care. In understanding capabilities and functionings, the difference between ends and means is important. For Robeyns, ‘we should always be clear, when valuing something, whether we value it as an end in itself, or as a means to a valuable end’.[18] Other concepts used in the capabilities approach are agency and structural constraints. Agency is a person's ability to bring about changes, while structural constraints are laws, norms and institutions that inhibit people's capabilities. Research within the capability approach has challenged the biomedical or biostatistical understanding of health,[19] and particularly as applied to children[20] and child growth.[21]

In this paper, we argue that qualitative data add valuable perspectives to the biomedical data, particularly concerning the demands of parenthood and the responsibilities that the first 1000 days entail. We draw on the capabilities literature to argue for a more complex approach to the mother's role and child development during this period.

METHODSStudy area and respondents

Our participants were from the Freedom Square area of the former black township of Mangaung in Bloemfontein, South Africa. Since 1994, Mangaung residents have been able to access property without racial restrictions. The scrapping of the restrictions meant that upper-middle-class people moved out of this former black township. According to the 2011 census, the average monthly household income in Mangaung was R5100 (725 USD in 2011). We recruited respondents by distributing recruitment pamphlets in Freedom Square in 2020, which initially developed as an informal settlement and was upgraded between 1992 and 1994. Household incomes in Freedom Square are even lower than in the wider former township area. Marais and Ntema[22] recorded an average annual household income here of R22,000 (230 USD in 2008). However, despite the low-resource setting of our study, mobile technologies and television were commonly available.

All our respondents were mothers of children between 18 and 30 months of age. We did not record their marital status, but their responses give the impression that many did not have a husband or steady partner. Six respondents said that the child's father was making no financial contribution. This meant that they were solely responsible for the child's daily needs and socio-emotional care. All the respondents depended on the public health system, except for one with private medical aid. However, some respondents occasionally opted to use private medical practitioners. All our participants had access to mobile phones.

Data collection

We trained two fieldworkers who spoke the dominant local language, Sesotho, to conduct the interviews. The fieldworkers conducted the interviews during July and August 2020. Verbal informed consent was obtained from all participants, with the consent documents sent to the participants through Whatsapp (a commonly used messaging service in South Africa). Because of the Covid-19 lockdown, interviews took place via mobile phones. We set out to do two interviews with each respondent. Respondents received a voucher of R100 per interview for their time. We completed 19 baseline and 18 follow-up interviews and only used the 18 interviews for which we had both (see Table 1). The interviews were recorded and transcribed and translated by the fieldworkers.

Table 1 Profile of the participants

	Still at school?	Gender of child	Number of children	Planned pregnancy	Considered an abortion?	Breastfeeding?	Stopped using alcohol	Giving birth by C-section	High blood pressure problems	Ref to social grant	Father making a financial contribution
1	No	Male	2	No	No mention	No mention	No mention	C-section	Yes	No mention	Yes

2	No	Male	3	No	No mention	Yes	No mention	C-section	Yes	No mention	Yes
3	No	Make	2	No	No mention	Yes	No mention	C-section	Yes	Yes	No mention
4	No	Male	1	No	Yes	Yes	No mention	No mention	No mention	No mention	No mention
5	No	Female	3	Yes	No mention	Tried, had problems	No mention	No mention	Yes	No mention	Yes
6	No	Female	3	No	No mention	Yes	Yes	No mention	No	Yes	No
7	Yes	Female	2	No	Yes	No mention	Yes	No mention	No mention	Yes	No
8	No	Not available	Not available	No	No mention	First 20 days, then got sick	No mention	C-section	Yes	Yes	No
9	No	Not available	Not available	No	No mention	Yes	No mention	C-section	No mention	No mention	Yes
10	No	Not available	Not available	No	No mention	Yes	No mention	No mention	No mention	No mention	No
11	No	Male	2	Yes	No mention	Yes, but stopped because the child was not gaining wait	Yes	C-section	No mention	No mention	Yes
12	No	Male	Not available	No mention	No mention	Yes	Yes	No mention	No mention	No mention	Yes
13	No	Male	Not available	No	No mention	No mention	No mention	No mention	No mention	No mention	No

14	No	Not available	Not available	No mention	No mention	Yes	No mention	No mention	No mention	Yes	No-mention
15	Yes	Not available	Not available	No	No mention	Yes	No mention	No mention	No mention	No mention	Yes
16	No	Not available	1	No	No mention	Yes	No mention	No mention	No mention	No mention	Yes
17	No	Male	1	No	No mention	Yes	No mention	No mention	No mention	No mention	Yes
18	No	Male	4	No mention	No mention	Yes	No mention	C-section	Yes	No mention	No mention

The interview framework

We asked one question during the first interview: ‘Tell us the story of raising this child’. We probed for the mothers' wishes for their child, the extent to which these wishes were realized and the boons and difficulties of raising a child. The interviews were conducted to elicit the mother's story of her pregnancy, her child's birth and the first 2 years of her child's life, and fieldworkers were trained to probe selectively. During the second interview (about 2 weeks later), the interviewers clarified any unclear information from the first interview and asked six additional questions drawn from our reading of the first round of transcribed interviews:

- (1) How often did the child get sick, and what did you do when the child was sick?
- (2) What was the most challenging aspect of raising this child?
- (3) What was your main achievement in raising this child?
- (4) What did you do to help the child to develop physically?
- (5) What did you do to promote the child's brain development?
- (6) Is someone else looking after the child regularly? Why, who and why do you trust this person?

Data analysis

We used thematic analysis, starting with themes from the literature of the first 1000 days and amending the themes on the basis of the mothers' stories. The first two authors conducted coding independently and then discussed it to settle on a common set of themes. A preliminary coding framework was developed from the literature. The transcripts were dissected using the preliminary framework. Abstract themes were first identified, followed by elegant themes. The themes were then arranged into a network to distinguish basic and organizing themes.[23]

FINDINGS

We discuss the experience of mothers within the five essentials of the first 1000 days as our organizing themes: pregnancy and ante-natal care, nutrition, cognitive and physical development, home environment and access to health care.

Pregnancy and ante-natal care

Ante-natal care and maternal health are key policy concerns in South Africa, where the maternal death rate remains high.[24] We identified two subthemes: short-term reaction to falling pregnant and longer-term responses. Of our 18 respondents, 13 spoke about aspects of falling pregnant, and 10 of these had had unplanned pregnancies. They said things like ‘I was angry’, ‘I was not happy’, ‘I was shocked’, ‘I did not want the child’, ‘It wasted my time’, ‘It took time to pull myself together’, ‘I was surprised’ and ‘I was afraid’. A few respondents had not immediately realized that they were pregnant. One of them had visited the clinic for hypertension and only then discovered that she was 6 months pregnant. Two respondents had considered abortion but decided against it on religious grounds. Two said that pregnancy had affected their education. However, three respondents expressed their excitement at falling pregnant.

Despite about half of the respondents falling pregnant unintentionally, most of them had the capability to accept the pregnancy eventually, seek medical care and bond with the child. Respondents used words like ‘excitement’ and ‘a joyful moment’. One of them described how she got used to being pregnant, felt more at ease, started getting excited about the baby and was very happy when the baby was born.

Nutrition

We identified three subthemes regarding nutrition: behaviour change during pregnancy, breastfeeding and the role of the public health system in monitoring children's diets. Many of our respondents said that falling pregnant forced them to change their behaviour, for example, by stopping drinking alcohol and starting to eat healthily ‘for the sake of the child’. Concerning alcohol, one respondent said:

I could stop drinking alcohol so I could raise my child properly, with respect. Because when you're drunk, you get out of control, you know? You don't even know how to speak correctly, and even the child won't know you because you are drunk. So that is something I wanted to change.

This respondent did not relate the use of alcohol to foetal alcohol syndrome. Instead, her motivation was to become a responsible parent.

Breastfeeding was common because staff at the public clinic emphasized the importance of breastfeeding. Respondents were able to explain the nutritional value of breastfeeding because they had visited a public clinic. However, going back to work was a problem for continued breastfeeding, as one explained:

When he was still with me before I went back to work, he was breastfeeding and did not want a bottle. He never got used to it—he refused it. And when the child drinks milk from a cup, it is different from when he is breastfeed. So, he lost weight a little bit.

The clinic staff weighed the children and recorded weight gains or losses. Respondents demonstrated agency in trying hard to comply with advice from the clinic despite practical realities and the lack of accommodation for breastfeeding mothers in the workplace. Knowing that the nurse would weigh the baby made some respondents anxious. They felt that the nurses scolded them when there was little increase in the child's weight. Finally, several respondents talked about the difficulty of affording food and having to go hungry but ensuring that the child had food.

Cognitive and physical development

We identified the following subthemes: reactions to reaching or not reaching milestones, an understanding of the importance of play, the role of the public health system, dreams for their children and the role of technology. Most respondents had an idea of children's expected development milestones, such as sitting up at about 6 months, walking at about 1 year, recognizing their names and saying words. They enjoyed seeing their child reach these milestones, expressing delight if the child was crawling and standing very young and excitement at seeing them grow and talk. These expressions of joy stood in contrast to their descriptions of surprise and fear when they found out that they were pregnant. However, external organizations can overemphasize these milestones, causing anxiety. Some of our respondents had worried that not achieving the goals according to the guidelines meant that something was wrong.

Most respondents talked about their children's play (identified as a capability by Nussbaum).[15] Some of them related play to social development and learning, as the following quotes seem to show:

I love playing with kids, spend some of my time playing with them, and you know that a child grows very well when playing with other kids.

Sometimes I leave him to play free but making sure there is no danger. When he wants to touch things, I let him be so that he can feel different things.

However, none of the respondents mentioned that play contributes to cognitive development. None of them said that they had been told this by the clinics or doctors. Safety was a primary consideration. One respondent mentioned locking house doors to keep the child safe.

Opinions were divided about how much the clinic staff had helped them understand and foster physical and cognitive development. Some felt that the staff had been extremely helpful. One respondent said:

The clinic showed us how to speak to the child normally and said we need not use baby language. He will understand. I think that helped. Because now when I talk to him he responds, even when he responds with his eyes. The simple thing is making a conversation with him because language development is important. When you bath him, talk to him. Tell him: ‘I'm bathing you, clothing you or putting on your shoes’—such simple things.

This quote emphasizes the importance of conversations with the child and confirms that this message originated from the primary health system. This understanding that normal daily activities contribute to physical and cognitive development and connect the child to people in the household is important in a low-resource setting.

Some respondents believed that crèches and schools were crucial for physical and cognitive development. One said that her child was not learning anything when he was at home and that at the crèche, he would ‘be able to learn something new every day’. Another explained her decision as follows:

I think the importance of taking a child to crèche is so that he can start writing at a young age. So he can know the right way to hold a pen—not necessarily writing, but how to hold a pen. Also, knowing how to play with building blocks so that he may know how to build things with his hands.

Despite the value that mothers saw in early childhood development (ECD) centres, some expressed concerns about the quality of care. However, many respondents were worried about the cost of sending the child to a good crèche or school.

Many respondents had dreams for their children. The following quote summarizes the feeling of most of them:

My dreams about my children are to see them achieving more things than I have achieved myself. I want my children to study further. I want them to be respectful to people because one can be educated and all, but not have a respectful nature, and they are just nothing without respect.

Respondents also mentioned the advantage, and possible disadvantage, of technology. We heard comments such as ‘he is fighting for the phone’, ‘he knows everyone's phone in the house’, ‘he loves watching cartoons’ and he ‘repeats the songs from the TV’. Although overexposure could be negative, these endowments do have the advantage of connecting the children to the outside world.

However, beyond the discussions about achieving good physical and cognitive development, perhaps the best summing up was from the respondent who described her achievement as just ‘seeing him being okay, seeing him around and him being here with me’.

Home environment

We identified three subthemes: disrupters, stabilizing factors and the mother's agency to overcome adverse home environments. The two main disrupters were neglect by the children's fathers and financial distress. At least half of the respondents had difficult relationships with partners who did not acknowledge the child, did not contribute financially to raising the child or abused alcohol. One respondent said: ‘My wish was that the love that I give the baby, I wished that the father would give it as well’.

Financial concerns included paying someone to look after the child and buying formula milk, diapers and creams to avoid rashes. The distress caused by money worries was evident in one respondent's words:

When my children need something, I can't give them that thing. I can't give it to them even if I want to. Things like that make me wish that I didn't have children. I think that's the most painful thing.

Several respondents mentioned financial sacrifices like walking rather than using a taxi, and not buying formula milk and baby food but putting the child on homemade food early. However, despite this lack of support from fathers and financial difficulties, our respondents showed agency. We heard many comments like this about the fathers: ‘I don't even care about him’, ‘I have pride’, ‘I'll raise the baby for the father’, ‘I am going to carry my problems’ and ‘I was trying to do everything myself’.

The respondents mentioned several stabilizing factors: a good relationship with their partner, help from the extended family, a trustworthy caregiver and the South African child support grant.

A positive relationship with the child's father was seen as especially important during pregnancy. One respondent said that she ‘had someone who was supporting me throughout my pregnancy, so I ended up enjoying being pregnant’, and it helped that ‘at least his father listens to me when I talk to him’. Another said that when she came back home with the baby, ‘the baby's father was the one who looked after us, just the two of us’.

Respondents mentioned their mothers and grandmothers in particular as important sources of care and financial support for the children. One respondent said:

I receive support from both sides, especially from his grandmother. She loves him. She is very protective of the home. When I tell her that the child is sick, she comes with everything.

Respondents said that it was not easy to find the right person to care for the child when they were at work. But most were satisfied, saying that they trusted this person, that the child was clean, the person is taking good care of the child, the child is growing well and even that they received good advice from the caregiver.

A few respondents mentioned the stabilizing role of the child support grant that the South African government provides. They thought that the amount was too small (26 USD at the time of the study), but were nevertheless most thankful for the contribution. Some used it to buy formula milk, diapers or food or to pay for babysitting.

Most respondents mentioned that attachment developed between them and their children. They said things like ‘we are very close’, and described children following them, sharing telephone discussions and not wanting to leave them. One of the mothers stressed the importance of mother–child bonding, saying that if ‘it is not okay’, the child will be mentally harmed.

In a low-resource setting, exercising agency often involves making sacrifices. They spoke about not getting enough sleep, being tired and not having enough time. They spoke of household chores, headaches and nerve problems, and referred to the child as a 'load to carry'. They spoke of crying and feeling overwhelmed. On the social side, they spoke of a diminished social life, fewer friends (and dating), routine changes, reduced alcohol intake, missing out on breakfast or an afternoon nap, limited freedom and being controlled by the child. One mother said, while laughing: 'Being a mother is not child's play [Sesotho, *Ho ba mme ha se papadi*—literally, "To be a mother is not a game"]'. She explained as follows: 'Because, everything you do, you have to think about the child'. Another referred to 'baby stress'.

Access to health care

In our analysis of the interview transcripts, we identified three main subthemes of health care: health problems during pregnancy, child health problems and the role of the health system, public or private, in managing the problems.

Maternal death at birth is abnormally high in South Africa, at 100 deaths per 100,000 live births.[25] Respondents reported several health concerns. Six mentioned gestational hypertension. Seven had given birth by Caesarean section. Other problems during pregnancy that they mentioned were excessive vaginal discharge, heartburn, sleeplessness, excessive sleeping, pressure on a hip joint, dizziness, feeling weak, back pain, headaches, abdominal pain, vomiting and feeling ill. One mentioned a benign tumour.

There were 18 references to stress during pregnancy. Various reasons cited for the stress were the pregnancy being unplanned, not accepting the situation, work-related difficulties such as travelling, worry about the financial situation (how to provide for the baby after birth) and lack of support from the child's father.

Some mentioned feeling depressed following delivery. One respondent said: 'After having the baby, I had convinced myself that there was nothing else I could do. I had given up on life'.

Access to adequate health care and skilled providers is key to maternal and child health. Respondents reported mixed experiences in attending ante-natal clinics. One respondent expressed her diligence as follows:

Once the doctor confirmed that I was pregnant, I started attending ante-natal classes. I went to the clinic as early as possible so that if something was wrong with the baby—they can see it earlier, you see? I didn't miss my appointments. I made sure to take the pills the way they told me to. I even changed my diet, a little.

The respondents were generally thankful for receiving appropriate services from the public health service and most said that once they received confirmation of pregnancy, they attended according to their programme. Two said that they appreciated the text messages reminding them about their appointments.

The respondents were generally aware of their children's needs. They mentioned respiratory problems, including pneumonia, and urinary and teething problems. Some reported only minimal child health problems. All the respondents emphasized that they wanted their children to grow up healthy. One mother said that to achieve this, 'I will have to look after him properly'.

The respondents described many worries. One said, 'When he is still a baby and just cries the whole day, you worry'. Another said, 'He would get sick, probably, two to three times in a month'. A child's illness became a stress factor for the mother. One respondent said that this stressed her because 'I would feel like I should have been the one who was sick, not him'. Respondents noted that the frequency of illnesses decreases as the child grows older: 'After he turns a year old, then those problems become less'. Three respondents reported serious health problems that required the child to be hospitalized, saying that 'it was scary' and 'I was in pain'.

Despite these concerns about the child, we heard many remarks indicative of the respondents' resourcefulness, in other words, agency, in managing difficult circumstances. One respondent said: 'If the child doesn't want to eat, you shouldn't give up. You have to be patient with your child'. Another said that since the child was a baby, she had ‘tried, by all means, to ensure that he grows up healthy’.

Respondents mostly went to a primary healthcare clinic when the child was sick or, for those who could afford it, a private medical practitioner. Respondents attended the postnatal clinic for the child's EPI vaccines and for advice, as one explained:

He had to have vaccinations according to the book ‘Road to Health’, from three days to six weeks—injections and those drops. The second thing was tracking his growth, weight, height and seeing if he had reached milestones and was not delayed or being held back. They would ask if he could respond to my gestures, and smile and laugh. If you don't attend those sessions, you won't know when something is wrong with your child.

However, despite the clinics, doctors and Google, misconceptions remained about what causes health problems. One mother blamed herself, and reported some astonishing misinformation that she had received from a clinic:

When this child was sick, I would worry a lot because his flu was too much. He had a problem with his chest and lost his breath. We always had to spend money to get to the doctor. I think he had flu because I liked cold stuff. I used to eat a lot of cold stuff, and it affects the baby, so when the baby is born, they are born having flu.

Affording private medical care was a problem for many mothers. Considering some of the resource-related problems explained earlier, this is understandable. Several respondents complained about the cost of health care and medicines.

One respondent spoke about getting the necessary support ‘at the right time’, and said that going to the clinic made her aware of her responsibility. However, their views on service quality at the postnatal clinics were mixed.

Some respondents related instances of inadequate service from the public health facilities. One said that after not receiving adequate care from the public health system, one of the private hospitals helped her. Another said: ‘When I take my child to a doctor, they check him from head to toe. They make sure that they don't miss anything’. We heard complaints about poor treatment, such as: ‘They didn't say much. They said I should bring the baby for his injections. If he doesn't get his injection, he will fall ill’. One respondent complained about the clinic staff not believing her when she said that her child could not urinate. Some respondents were scared of the nurses: ‘But if you dare miss your clinic date—yoh! They spit fire’.

DISCUSSION AND CONCLUSION

In this discussion, we analyse our findings within the capabilities approach and against the literature on the first 1000 days and caring (see Table 2).

Table 2 Summary of the main findings

The health outcomes recommended in the first 1000 days literature require certain functionings by the mothers. Examples of these include going to a clinic or doctor during pregnancy. Most of our respondents were late in confirming their pregnancy and half of them had unplanned pregnancies. However, once they and their children were part of the primary healthcare system, they remained in the system. Most respondents spoke about adjusting their diets and social behaviour after falling pregnant. There was a concerted effort to stay healthy during pregnancy. Despite pregnancies being unplanned, all the respondents eventually accepted being pregnant and enjoyed it (a capability identified by Nussbaum).[15] This behaviour confirms the literature that shows the importance of attending antenatal healthcare facilities[13] and confirms Nussbaum's[26] capabilities related to emotions. It is possible to link postnatal care and mothers and such a link would benefit the children.

We noted important functionings and capabilities related to the relationship between the mother and the child. Doings include attending primary healthcare facilities with the child and ensuring that the child was vaccinated. Most respondents realized the importance of providing good nutrition, allowing their children to play and facilitating learning by creating educational opportunities. The mother's functioning supports child development and confirms the literature on the importance of attending antenatal clinics[13] and the role of play in the cognitive development of the child.[7] Some mentioned the value of talking to the child and making eye contact—both important for neural development. Despite absent fathers, respondents said that they were able to care for their children on their own and with the help of extended families, confirming the important roles of the extended family noted in the literature. The literature shows that although the father's involvement is important, most mothers will be able to raise children adequately. Our evidence shows that mothers are doing this and that they find support from the grandmother and extended families and the village.[12] Most of the responses suggest that the child was well nourished and the mother had bonded with the child. Despite making sacrifices, most respondents said that they enjoyed having the child. All of this gave them the capability to promote their child's growth and cognitive development. It enabled them to dream about the child's future. In the respondents' descriptions of the children's behaviour, we noted several functionings: eating, playing, socializing, learning and using their senses. Most of these are simple requirements for the cognitive and child development indicators identified by research. These functionings lead to growth, health and cognitive development capabilities. Despite several structural constraints, we found many examples of the respondents exercising agency.

We think that there are three specific values attached to the use of a qualitative approach. First, the respondents' stories brought to light their ability to change their behaviour. Biomedical literature often emphasizes interventions to change behaviour and ignores the agency that people have. For example, the first 1000 days literature stresses the importance of ante-natal care, maternal health (in a primarily biomedical sense) and a stable home environment. This can become a mechanical way of thinking about raising children. Our approach allowed us to understand how mothers can deal positively with difficult circumstances during the first 1000 days.

Second, linked to the above, the first 1000 days literature's expectations could give mothers a sense of failure, a common problem identified in the literature.[4] Although our respondents did receive some very good advice from the public health system, some of them feared going to the clinic to be reprimanded by the staff. The responses show how focusing on some metrics could deter mothers from going to the clinic. Although the literature points to the important role of antenatal clinics,[13] our evidence shows that the nature of the support from these clinics is crucial in keeping women in the system.

Third, the 1000 days literature tends to overemphasize specific health issues and does not always take a holistic approach. For example, respondents referred to their children achieving the capability of play. Yet, none of them related play to cognitive development. From their responses, we could not detect that the public health facilities emphasized play. One mother reported being advised to talk to her child regularly. We would like to see more of these less mechanistic approaches supported by the health system.

Although we do not intend to generalize from our small sample, we acknowledge that our recruitment method may have caused some bias. The voucher that we provided may mean that we attracted a disproportionate number of respondents who were very poor. Our sample may also be disproportionately made up of women who were managing motherhood well, in other words displayed agency, as those who were struggling might not have had the energy to participate in the study, or been eager to reveal their failure.

In conclusion, we would argue that our respondents' evidence supports the notion that mothers are held disproportionately and unfairly responsible for achieving the first 1000 days ideals. Many issues that make it difficult for mothers to enhance their children's developmental health are not internal or personal. Rather, they are the result of negative social arrangements such as the absence of supportive fathers, work environments that do not support mothers returning to work and inequalities (racial, gender and otherwise) in the quality of services available to mothers. The constraints within a low-resource setting place unfair pressure on mothers to provide for their children's daily needs and facilitate socio-emotional stability, physical growth and cognitive development. They have to manage with limited means and are constrained in their ability to convert these means into the desired end: their children's developmental health. The 18 mothers in our sample experienced a disproportionate curtailment of their functionings and capabilities. A holistic approach should take this into account in designing policy interventions for children's developmental health.

AUTHOR CONTRIBUTIONS

Michael Pienaar: Conceptualization; formal analysis; writing – original draft. **Lochner Marais**: Conceptualization; formal analysis; writing – original draft. **Mosaathebe Serekoane**: Conceptualization; data curation; methodology; writing –review and editing. **Kobus Marais**: Conceptualization; writing –review and editing. **Jan Cloete**
: Conceptualization; data curation; methodology; writing –review and editing. **Molefi Lenka**: Conceptualization; data curation. **Carla Sharp**: Conceptualization; writing –review and editing.

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CONFLICT OF INTEREST

The authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request.

ETHICS STATEMENT

The research has ethics approval from the Faculty of Health Science Ethics Committee at the University of the Free State. Participants provided consent.

DETAILS

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Exploring patients' perspectives of gestational diabetes mellitus screening and counselling in Ontario: A grounded theory study

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ABSTRACT (ENGLISH)

Introduction

Gestational diabetes mellitus (GDM) is associated with adverse perinatal outcomes. Approaches to screening for GDM continue to evolve, introducing potential variability of care. This study explored the impact of these variations on GDM counselling and screening from the perspectives of pregnant individuals.

Methods

Following a Corbin and Strauss approach to qualitative, grounded theory we recruited 28 individuals from three cities in Ontario, Canada who had a singleton pregnancy under the care of either a midwife, family physician or obstetrician. Convenience and purposive sampling techniques were used. Semi-structured telephone interviews were conducted and transcribed verbatim between March and December 2020. Transcripts were analysed inductively resulting in codes, categories and themes.

Results

Three themes were derived from the data about GDM screening and counselling: ‘informing oneself’, ‘deciding’ and ‘screening’. All participants, regardless of geographical region, or antenatal care provider, moved through these three steps during the GDM counselling and screening process. Differences in counselling approaches between pregnancy care providers were noted throughout the ‘informing’ and ‘deciding’ stages of care. Factors influencing these differences included communication, healthcare autonomy and patient motivation to engage with health services. No differences were noted within care provider groups across the three geographic regions. Participant experiences of GDM screening were influenced by logistical challenges and personal preferences towards testing.

Conclusion

Informing oneself about GDM may be a crucial step for facilitating decision-making and screening uptake, with an emphasis on information provision to facilitate patient autonomy and motivation.

Patient or Public Contribution

Participants of our study included patients and service users. Participants were actively involved in the study design due to the qualitative, patient-centred nature of the research methods employed. Analysis of results was structured according to the emergent themes of the data which were grounded in patient perspectives and experiences.

FULL TEXT

BACKGROUND

Gestational diabetes mellitus (GDM) is one of the most frequent metabolic disturbances of pregnancy, affecting up to 20% of pregnant individuals in Canada.[1] Factors contributing to the rising rates of GDM in Ontario include variations in screening approaches and diagnostic thresholds, and increased incidence of delayed childbearing, obesity, and excess gestational weight gain.[2–5]

Two approaches to GDM screening are endorsed by Diabetes Canada (DC) and the Society of Obstetricians and Gynecologists of Canada (SOGC): the ‘preferred’ two-step, method of a nonfasting, 50 g oral glucose challenge test (50 g OGCT) followed by a fasting 75 g oral glucose tolerance test (75 g OGTT) upon abnormal results, as well as an ‘alternate’ method of the one-step, fasting 75 g OGTT.[6–8] A randomized controlled trial, comparing the incidence of GDM between screening approaches, revealed a nearly doubled incidence rate of GDM in the group who underwent a one-step 75 g OGTT, when compared to the group who was administered a 50 g OGCT followed by a 100 g OGTT (16.5% vs. 8.5%, respectively).[9]

However there is a lack of consensus between regulatory bodies on the optimal GDM screening approach. For example, DC and the SOGC recommendations differ from that of the International Association of the Diabetes and Pregnancy Study Groups (IADPSG), which supports the use of the one-step 75 g OGTT with lower diagnostic cut-off values.[7,8,10,11] Comparatively, the American College of Obstetricians and Gynecologists (ACOG) recommends the use of an OGTT with a higher glucose load (100 g) and a longer assessment period (3 h).[11,12] A recent randomized controlled trial examined the rate of GDM diagnosis across groups with different diagnostic threshold criteria, with the lower glycemic criteria group reporting over double the rate of GDM diagnoses compared to the higher glycemic criteria group (15.3% vs. 6.1%, respectively).[13]

There has also been debate over universal versus risk-based GDM screening. The SOGC and DC shifted from selective screening in the late 1990s to recommending universal screening.[6] Differences and uncertainty of diagnostic thresholds between approaches, the most appropriate glucose load, the number of abnormal values required to determine a GDM diagnosis, the importance of early trimester and postpartum screening, as well as whether to practice risk-based or universal screening, has led to discrepancies in the true prevalence of GDM in Canada.[6]

From the patient's perspective, GDM care includes a range of experiences such as counselling, screening, diagnosis, management and postpartum follow-up. Patients who received a GDM diagnosis have reported feelings of self-blame, failure, confusion, and fear, signifying uncertainty and guilt.[14–16] Lack of time and continuity of care have been identified as barriers to communication between patients and healthcare providers (HCPs); however, discussion of the GDM condition, associated risks and potential outcomes have been shown to promote greater acceptance of the diagnosis for the patient.[14]

Despite evidence indicating patient motivation to protect the health of their baby, many challenges impeding positive behaviour change and treatment compliance have been reported.[17] These have included a lack of access to GDM services, financial barriers, lack of communication with HCPs and poor adherence to lifestyle changes such as diet and exercise.[17] Given the multifaceted nature of GDM for patients, the aim of this study was to explore the impact of variations in GDM counselling and screening from the perspective of patients.

METHODS

We conducted a qualitative, grounded theory study with pregnant participants to explore their experiences of gestational diabetes screening and counselling practices. We sought to recruit participants from various geographic locations in the province, and who received care from different antenatal care providers. Patients who had had a singleton birth within the 5 months before data collection, who also received antenatal care from a midwife (MW), family physician (FP) or obstetrician (OB) within Hamilton, Ottawa or Sudbury, Ontario, Canada, were eligible to participate. Semi-structured interviews took place between March 2020 and December 2020 and were conducted over the telephone for approximately 30–45 min. Convenience and purposive sampling were used for recruitment, using social media and posters within the community. These sampling methods were chosen based on the limited geographic regions targeted and the ability to identify participants with lived experiences with the research topic. Additionally, given the lack of strict selection criteria and the qualitative methodology employed, these sampling techniques were most appropriate.

The geographic regions identified for inclusion were selected to increase subpopulation variability and to utilize existing contacts to assist in recruitment. A minimum of three patients who received care from each health profession and from each geographic region was identified as the desired sample size (totalling 27 participants) based on experience with similar studies by the research team, but with the intention of continuing recruitment until we reached saturation.[18,19] An interview guide was developed by our research team and utilized during the interview process, using a mix of open- and closed-ended questions to elicit the participant's perspectives on their experiences with GDM counselling and screening (see Supporting Information: S1).

In keeping with grounded theory, as described by Corbin and Strauss, data analysis began at the same time as data collection, to make use of the iterative process of constant comparison.[20,21] Interviews were transcribed verbatim and entered into Nvivo 11 software.[22] Data analysis began with open coding. Initial open coding of three transcripts was completed by three independent researchers to ensure consistency and agreement in the coding process.[23] Next, codes were grouped to form axial codes which provided a framework from which the open codes could be synthesized into hierarchically structured categories.[24–27] Lastly, during the selective coding process, further grouping was completed to form themes that, when brought together, generated a theory grounded in the data.[27]

Interim analyses were shared at team debrief meetings, and the Principal Investigator reviewed the coding at each stage of analysis.[23] The research team was comprised of students and experts from a variety of disciplines, including midwifery, maternal-foetal medicine, obstetrics and health research methodology. Investigator triangulation was used to review, validate and come to an agreement on disputed codes between researchers. These approaches were employed to minimize bias, strengthen credibility and add breadth to the emerging phenomena.[28]

RESULTS

A total of 28 participants were included. Demographic characteristics were obtained and are presented below (Table 1).

Table 1 Participant characteristics by the antenatal care provider

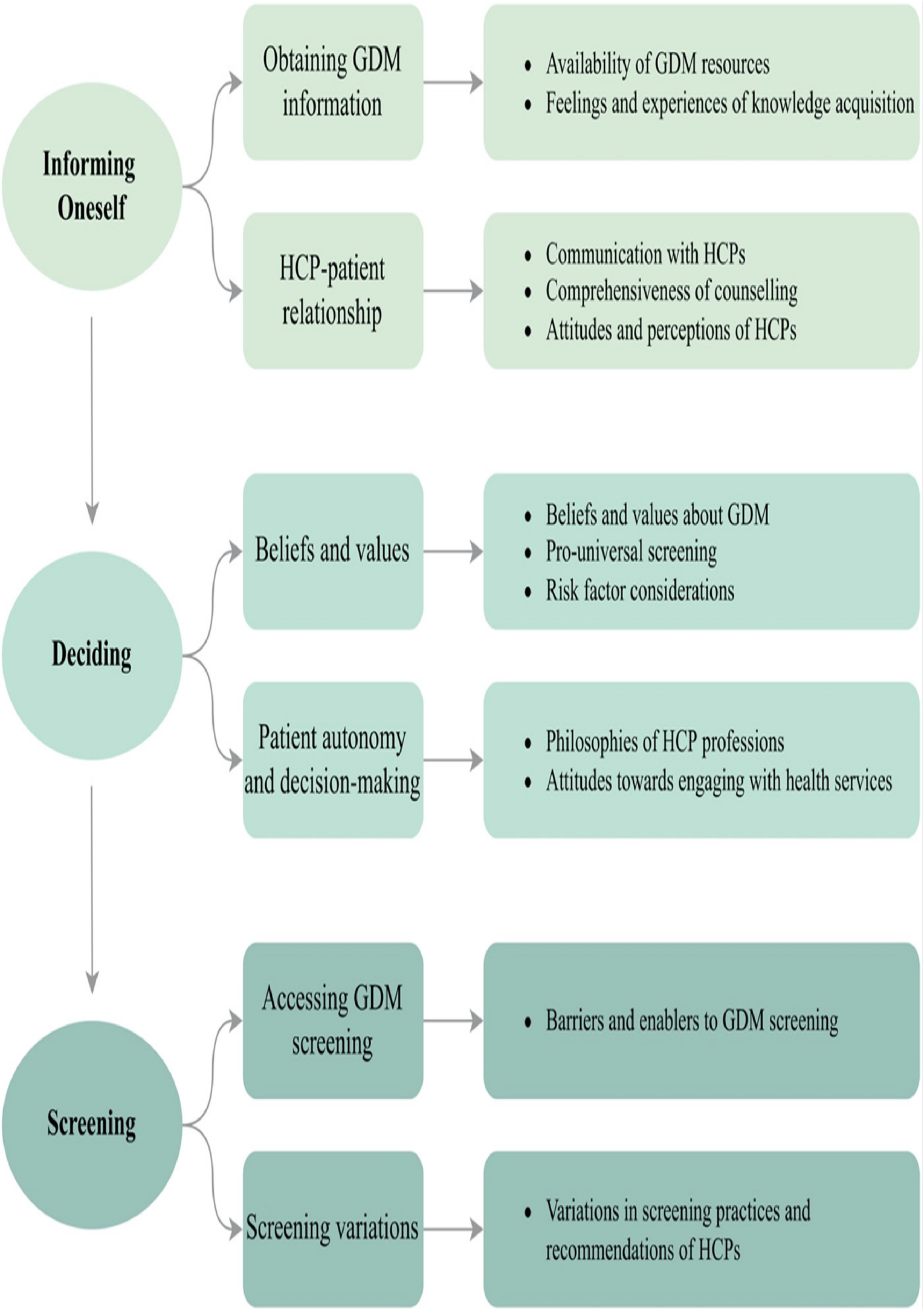
Characteristics	Antenatal care provider			
	Midwife (*n* = 12)	Obstetrician (*n* = 10)	Family physician (*n* = 6)	Total (*n* = 23a)
Geographic region				
Hamilton	5	3	2	10
Ottawa	4	4	3	11
Sudbury	3	3	1	7
Maternal age (years)				
15–24	0	0	1	1
25–34	6	6	1	13

35–44	3	4	2	9
Ethnic or cultural origin(s)				
East Asian	0	2	0	2
White	9	8	4	21
Hispanic	0	1	0	1
Other	0	0	0	0
Highest level of education				
High school	0	2	1	3
Bachelor's degree	4	5	2	11
Master's degree	4	1	1	6
Postgraduate certificate	0	1	0	1
Doctorate	1	0	0	1
College diploma	0	1	0	1
Past live births, including most recent (#)				
1	6	8	3	17
2	3	1	1	5
3	0	1	0	1
BMI (kg/m^2)				
18–24	5	4	4	13
25–30	4	0	0	4
30–35	0	2	0	2
35–40	0	3	0	3
>40	0	1	0	1

Neonatal birth weight (lbs)				
<6	1	1	1	3
6–7	3	1	1	5
7–8	2	5	1	8
8–9	2	2	1	5
>9	1	1	0	2
Gestational age at delivery (weeks + # days)				
<35	1	0	0	1
35–37 + 6	1	2	1	4
38–39 + 6	2	7	0	9
≥40	4	1	3	8
Diagnosis of GDM in most recent pregnancy (Yes/No)				
Yes	2	1	0	3
No	7	9	4	20

[a] Demographic characteristics data from five participants were not obtained.

Our findings are summarized in the themes arising from the data: ‘informing oneself’, ‘deciding’ and ‘screening’ (Figure 1). We found that all participants, regardless of geographical region, or antenatal care provider, moved through these three steps during the GDM counselling and screening process.



Enlarge this image.

Informing oneself

The theme of 'informing oneself' reflected the first stage of the patient's care experience. This theme was underpinned by the factors that influenced the understanding of GDM, particularly pertaining to screening, prevention and management.

Accessing GDM resources was influenced by the availability of sources outside of direct antenatal care to improve participants' understanding and satisfaction with their GDM care experiences. External sources included websites, pamphlets, information sessions led by hospital staff, prenatal classes, independent research, conversations with family or friends and Facebook pregnancy groups. One person articulated:

I like it to be in a paper format, like a pamphlet, or some type of brochure. When I started going just to my OB/GYN, they would say, ‘here's some additional resources that you can read more about it’. That would have been really helpful with the gestational diabetes as well. (P2, FP)

Positive experiences with GDM information provision were described as factors that strengthened participants' knowledge acquisition, reduced pregnancy anxiety, and enabled participants to develop positive lifestyle habits through diet management and exercise. Negative experiences associated with a perceived lack of GDM information provision included feelings of frustration due to limited knowledge acquisition, as well as uncertainty in dietary and lifestyle modifications before and between testing:

I remember I started to change my diet, but then I wondered, is this going to impact the [GDM] test? If I get negative, would I go back to my old diet, being that I wasn't sure whether I should make the changes before, or would I have to wait until after the test? (P3, MW)

Participants' relationship with their antenatal care provider was an important consideration in obtaining GDM information. Some participants felt that the counselling they received was thorough, whereas others felt it was minimal. In general, more comprehensive counselling among patients in midwifery care was noted.

A range of discussion topics covered during counselling was reported and included: adverse outcomes, screening options such as the OGTT or the OGCT, risk factors associated with GDM and logistical considerations of the screening procedure such as the timing of fasting if required, and the type of sugar beverage administered.

Most participants, regardless of the care provider, reported feeling that they received sufficient information and were able to ask questions as needed. Many participants expressed appreciation for a provider who was accessible, supportive, demonstrated a calm demeanour and listened attentively. However, many reported that they would have liked to receive more counselling on specific topics, including prevention and management of GDM, signs and symptoms to be aware of, customized diet recommendations, and how to best prepare for the testing procedure:

I don't know if there's preventative measures that you can take to just prevent developing it. That would be helpful in terms of your diet or exercise. I don't feel like I got that information. (P3, MW)

Lack of communication emerged as a barrier to accessing information, including restrictive timelines and protocols for appointments, lack of follow-up regarding the participant's GDM test results and inability to discuss health concerns due to restrictions in the scope of practice, or lack of educational training to provide patients with requested information.

Deciding

The theme of ‘deciding’ explored factors in the decision-making process pertaining to GDM screening, including participant beliefs, values and healthcare autonomy. Prior beliefs about GDM and personal values regarding knowledge acquisition were explored; many participants viewed knowledge as a tool to prevent potential medical complications in pregnancy.

Almost all participants expressed pro-universal GDM screening value statements. Beliefs that informed this included the following: (a) GDM is largely an asymptomatic condition, (b) GDM can affect anyone, regardless of risk profile, (c) screening is a minimally invasive and low-risk procedure (the benefits outweigh the harms) and (d) screening improves one's awareness and motivation for the health of themselves and their developing baby:

From what I've seen, it's pretty random. I've seen people who are quite thin and healthy, people who aren't so healthy. Because of the effects it can have on the baby, I think it's important to be screened, because you don't necessarily know until it might be a bit late, and be causing significant effects. (P2, OB)

The degree to which participants were involved in the decision-making process was particularly influential within this stage. Decisions included whether or not to be screened for GDM; choice of screening parameters such as the OGTT or OGCT; and the gestational time period to complete screening.

Being provided with an agency to make decisions was articulated as being very important for participants. For those who received care from a midwife, most reported that GDM screening was presented as optional and that they were able to make an informed decision based on the information provided. This was summarized best by one client, who stated:

With the midwife, she definitely made everything an option because she just wanted me to have more of an informed choice. So she told me 'these are the tests that we highly recommend', but really it was always up to me whether or not I wanted to do a test. (P9, MW)

For those who were not presented with GDM screening as 'optional' they described being instructed to be screened for GDM as it was simply 'the thing to do'. One participant described this clearly when they stated, 'I don't think it was presented to me as an option. It was presented to me as everyone gets screened, so I should get screened' (P5, OB).

Furthermore, when asked about perceptions of the differences in counselling practices between antenatal care providers, many expressed the general belief that midwives have more time to provide comprehensive counselling, offering more opportunities for the client to make an informed decision:

OBs and FPs have such little time to sit and discuss things. Based on my experience of how it's gone in the past, I feel like there wouldn't be as much discussion and more just 'you need to do this screen, here is the [requisition], go and do it'. (P10, MW)

Attitudes and motivation for engaging with health services were factors that influenced the level of importance that participants placed on healthcare autonomy. Some participants placed a significant emphasis on personal autonomy in their healthcare decisions. The participants that expressed the importance of making autonomous decisions were largely clients of midwives:

I think it's important for us that we are provided with information and that we are able to make the decision. We are supposed to live in a society where we are not forced to do things that we are not comfortable with. I think by having a midwife and them always making sure that they are informing my decisions, it's an awesome thing and obviously very empowering knowing that you are able to make these decisions on your own. (P12, MW)

Receptivity to screening was influenced by the risk factors that participants presented with and the extent to which they expressed concerns about its impact on the health of themselves and their babies. Many participants for whom screening was presented as optional ultimately expressed their receptivity to being tested given their risk factors:

[Screening] was available to me, and my OB thought it was a good idea based on the fact that I am older, I am overweight …and obviously with my family history of type 2. (P7, OB)

Screening

The last theme was related to experiences of completing the GDM screening test. Factors influencing the participant's access to testing were key determinants in their satisfaction with the screening process. Logistical barriers included challenges in obtaining childcare, inconvenient location of the laboratory, difficulty in scheduling time off work and transportation challenges. Individual reactions and experiences that presented barriers included distaste or aversion for the sugar beverage administered, difficulty coordinating fasting times before the test, emesis or nausea and discomfort with in-person assessment during the COVID-19 pandemic.

Factors that enabled screening were the absence of financial cost, ease of coordinating fasting times and taking time off work, the ability to attend the laboratory in a convenient location and on the weekend, a supportive partner that could transport the participant to and from their appointment, available childcare and minimal physical discomforts such as nausea.

Variations in screening practices included gestational timing, the type of testing approach recommended, and the locations where screening was offered. A few participants received earlier screening in their pregnancy due to the presence of risk factors: 'My baby was trending quite large for my third pregnancy, so I did get screened earlier …I think given my BMI and whatnot …they just wanted to check and make sure that I didn't have it' (P1, OB).

While most participants were screened within the recommended window of 24 to 28 weeks of gestation, a few individuals reported screening between 20 and 24 weeks gestation. No participants reported screening past 28 weeks. Of the 28 participants interviewed, 23 received the nonfasting 50 g OGCT; less than half required the follow-up 75 g OGTT. The remaining five received the one step, fasting 75 g OGTT. Participants from rural areas experienced more limitations in lab capacities and screening times compared to cities.

DISCUSSION

This study explored the experiences of GDM counselling and screening from the perspectives of patients who received antenatal care from either a midwife, family physician or obstetrician in Hamilton, Sudbury or Ottawa, Ontario. The goal of this paper was to provide a qualitative analysis to explore the impact of variations in screening guidelines and changing patient population trends related to GDM counselling and screening practices.

Our findings highlight the progression of an individual's experience engaging with GDM health services through three stages: ‘informing oneself’, ‘deciding’ and ‘screening’. The findings within the stage of ‘informing oneself’ aligned with literature that supports the importance of comprehensive and personalized care provision according to the lived experiences and preferences of the individual.[14,29,30] For example, much of the literature that explored patient perceptions GDM diagnosis highlighted feelings of self-blame, failure, confusion and anxiety.[14,29,30] These negative feelings were largely attributable to a lack of communication with their care provider, self-perceptions of risk factors and a lack of information regarding adverse outcomes.[14,29,30] Our participants also expressed how lack of communication, support or information provision impacted their experience.

Our findings highlight how autonomy and empowerment were tools for facilitating screening uptake and changes in health-seeking behaviours.[31,32] For many participants, the autonomy that they had in decision-making reflected their confidence in, or motivation for complying with, their care provider's recommendations. For example, many midwifery clients expressed strong motivation for being an active participant in the decision-making process. These participants were more likely to value informed choice approaches, and were generally more expressive about their healthcare desires than those who received care from a physician. Instead, those receiving physician-led care expressed enthusiasm to comply with their providers recommendations if it meant protecting the health of their baby. We also found that factors such as reactions to the screening test and logistical considerations in accessing laboratory services, at the individual, organizational and health systems levels influenced participants' experiences. Barriers to obtaining screening reported in the literature were consistent with those expressed by participants in our study, including time restraints, inconvenient locations and transportation challenges.[17,33]

In alignment with the literature, our findings indicated that there is a need for GDM care to be provided in a manner that is comprehensible, personalized and accessible, to best accommodate the lifestyle choices of diverse patient populations.[30,34] In particular, our study highlighted the importance of knowledge sharing as a facilitator in the decision-making process.[14,29,30,35] Knowledge sharing is a reciprocal process that promotes patient empowerment, and encourages humility of the provider to foster a relationship built on mutual respect and rapport.[35]

Given the evolution of screening guidelines in recent years, care providers have had to regularly integrate these changes into their clinical practice. We had hypothesized that patients would be aware of and possibly confused by the variations in screening guidelines over time and this would be reflected in patient data. However, the findings showed the minimal impact of screening guidelines inconsistencies on the experiences of participants. Instead, logistical challenges, accessibility of the screening and personal preferences arose as primary influences on the participants' experiences.

One of the aims of this study was to explore the differences, if any, in the GDM counselling and screening practices of antenatal care providers across professions and across geographic regions within Ontario. Our findings revealed that while there were considerable differences in the participants' counselling experiences across care provider groups, there were very few differences across geographical regions. For example, those who received care from a midwife offered similar sentiments regardless of their geography, highlighting consistencies in professional philosophies across geographic regions. However, we acknowledge the restrictions of our sample to three geographic regions, which may not reflect the spectrum of individual and professional philosophies across the country.

The interpretation of access was also important to consider within the context of our study. Access can be conceptualized by reciprocal interaction between health structures and the ‘consumer’.[36] It encompasses both accessibilities of providers, organizations, institutions and health systems to provide services, as well as the abilities of the consumer to receive such services, such as the ability to perceive, seek, reach, pay for and engage with health services.[36,37] As highlighted in our findings, information provision was a key factor in subsequent decision-making and GDM screening uptake. However, to adequately interpret our findings, we must consider the multitude of agents that impact an individual's access to healthcare.[36,37]

Lastly, the authors acknowledged that patient self-selection of care providers was a key consideration in the findings. Those who selected to have midwifery-led care may be inherently different that those who selected physician-led care, particularly with respect to desired autonomy during decision-making.[38]

Our study was unique in that it was one of the first to explore the experiences of patients seeking services pertaining to GDM counselling and screening, and compare across antenatal healthcare services, in a Canadian context. Furthermore, this study uniquely highlighted the direct impact that policy-level guidelines have on patients and providers. Given the lack of qualitative evidence on this topic, the findings from this study provide valuable insight into what factors patients are most impacted by when seeking GDM counselling and screening.

Strengths of our study included the multidisciplinary nature of our team and our recruitment approaches to maximize participant variation to reflect the diversity that exists within the greater Ontario population, and to explore the range of social, cultural, economic and environmental factors that contribute to the experiences of health-seeking patients. This enabled a range of perspectives that formed the basis through which comparisons of clinical practices across health sectors could be made.[39]

A limitation of our study was potential selection bias, given that those who volunteered to participate may be more willing to do so based on their personal beliefs about the topic. Furthermore, demographic characteristics were not obtained from five participants due to data collection documentation errors, presenting another source of bias. Additionally, our sample size reflected a lack of ethnic diversity, consisting of majority Caucasian identifying participants, with minimal to no participants from other ethnic groups. This may have further contributed to selection bias, with other populations not being well represented in our data. The COVID-19 pandemic may have also presented possible selection bias which may have impacted recruitment for this study given the uncertainty in restrictions and research protocols. Also, during this time emergency alternatives to GDM screening protocols were published and may have introduced another variation in care.[40] Lastly, language was also a limitation as interviews were only conducted in English.[41]

CONCLUSION

Our findings indicate that patients engage in GDM counselling and screening with a motivated mindset to protect the health of their babies. During the process of informing/deciding/screening, the informing stage and knowledge acquisition were crucial steps for facilitating decision-making and screening uptake. However, there were differences in the perceptions of the comprehensiveness of GDM counselling between antenatal care providers. The desire for patients to be active participants in decision-making is a reflection of their selection of care providers. Useful next steps to improve the patient experience include training for health professionals, and the creation of patient information resources that are adapted to the needs, preferences and lifestyles of patients, as well as a greater emphasis on information provision to facilitate patient autonomy.

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CONFLICT OF INTEREST

The authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

The data sets generated and/or analysed during the current study are not publicly available due to the lack of consent from the study participants to share the data publicly but are available from the corresponding author at a reasonable request.

ETHICS STATEMENT

Ethical approval was obtained from the Hamilton Integrated Research Ethics Board (HiREB Project: 7916). All participants provided consent before participation in the study.

DETAILS

Subject:	Pregnancy; Patient-centered care; Perinatal; Patients; Social networks; Data analysis; Pregnancy complications; Autonomy; Information sharing; Decision making; Gestational diabetes; Interviews; Grounded theory; Counseling; Health services; Qualitative analysis; Research methodology; Gynecology; Motivation; Diabetes mellitus; Research methods; Midwives; Clinical trials; Medical screening; Glucose; Family physicians; Data collection; Uptake; Patient participation; Medical personnel; Antenatal care; Midwifery; Prenatal care
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Ethical, legal, organizational and social issues related to the use of scalp cooling for the prevention of chemotherapy-induced alopecia: A systematic review

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ABSTRACT (ENGLISH)

Introduction

Scalp cooling (SC) aims to prevent chemotherapy-induced alopecia. The goal of this systematic review is to tackle ethical, legal, organizational and social issues related to SC.

Methods

A critical appraisal of the literature was carried out using a systematic review design. MEDLINE, Embase and Web of Science databases were searched up until 2 June 2021. Studies addressing these aspects in English or Spanish were considered. Representatives of both patient associations and professional scientific societies related to the topic participated in the design of the protocol and the review of the findings.

Results

A total of 17 studies were included. Articles were critically appraised using the MMAT and SANRA. Findings were organized into four categories: (1) ethical aspects focused on equal access, gender equity and doctor–patient communication supported by Patient Decision Aids (PtDAs); (2) patient perspective and acceptability; (3) professional perspective and acceptability; (4) organizational aspects focused on accessibility and feasibility.

Conclusion

Cancer patients' expectations when using SC need to be adjusted to reduce the potential distress associated with hair loss. PtDAs could help patients clarify their values and preferences regarding SC. Equal access to technology should be guaranteed.

Patient or Public Contribution

In this systematic review, the representatives of the patient associations (Ms. María Luz Amador Muñoz of the Spanish Association Against Cancer [AECC] and Ms. Catiana Martinez Cánovas of the Spanish Breast Cancer Federation [FECMA]) participated in the review of the study protocol, as well as in the results, discussion and conclusions, making their contributions. In the type of design of these studies (systematic reviews), it is not usual to

have the direct participation of patients, but in this one, we have done so, as it is a systematic review that is part of a report of the Spanish Network of Health Technology Assessment Agencies (ETS).

FULL TEXT

INTRODUCTION

Alopecia is one of the most common and visible adverse effects of chemotherapy, which affects approximately 65% of all patients undergoing chemotherapy.[1,2]

Although chemotherapy-induced alopecia (CIA) is not life-threatening and in most cases is reversible, it can have a significant impact on a patient's quality of life, especially in psychological and social terms.[3,4] While it has long been considered an acceptable side effect in the treatment of patients, the increasing number of cancer survivors and a better understanding of the associated psychological processes have led to a greater consideration of CIA as a relevant problem.[5] The negative psychosocial effects associated with CIA are strongly related to the diversity of sociocultural values and symbolic assignments attributed to hair.[6–8] As an essential element in personal identity, hair loss causes high levels of stress and anxiety, and makes it difficult to perform daily activities, especially those in social contexts.[9] The clear visibility of alopecia for patients and the people around them acts to identify the cancer and usually, when alopecia appears it becomes the moment of public recognition of the disease.[9,10] As a result, people with CIA may begin to perceive certain changes in the attitudes towards them of the people they relate to, ranging from sympathy to rejection.[11,12]

Scalp cooling (SC) has been used since the late 1970s as a system to prevent CIA.[13] Reduction in scalp temperature induces vasoconstriction, which limits the arrival of chemotherapeutic agents to the scalp and also produces a reduction in metabolism in the cells present in hair follicles at the time of highest chemotherapeutic concentration in the blood plasma. This reduces their vulnerability to the antimitotic and antimetabolic effects of these drugs.[14] The scalp must attain a subcutaneous temperature (between 1 and 2 mm) below 22°C,[15] which is equivalent to an epicutaneous temperature of 19°C,[14] although greater preventive effects could be achieved with temperatures close to 15°C.[16] A correct adjustment of the SC system to the patient's head is essential to attain these temperatures consistently and homogeneously.[17,18] The SC activation should commence 5 and 30 min before the infusion of cytostatic drugs and continue until completion.[18] After completion of the infusion, the SC must remain for a more or less prolonged time depending on the pharmacokinetics of the chemotherapeutic agents used, with postinfusion cooling times varying from 15 min to 4 h.[18]

In 2021, the Spanish Network of Agencies for Health Technology Assessment (HTA) and Services of the National Health System (RedETS) drew up an HTA report[19] commissioned by the Spanish Ministry of Health on the effectiveness, safety and cost-effectiveness of SC devices for the prevention of CIA. This Systematic Review (SR) was conducted as part of this HTA report. This report commissioned by the Ministry of Health of our country has the main objective of informing the political decision to include the technology in the common portfolio of health services of the National Health System, but it is also useful in decision-making for clinicians and patients.[20] Particularly, the ethical, legal, organizational and social issues are relevant when considering the equity in access, feasibility and acceptability of the technology, which are key elements to take into account for a successful implementation of the technology.

The research questions were: what are the ethical and legal implications of the use of SC? What are the attitudes, perceptions and experiences of patients and healthcare professionals regarding SC systems to prevent CIA? Are there any organizational aspects that may affect the accessibility or feasibility of the SC?

METHODSProtocol and registration

The SR of the literature followed the methodologic guidelines drawn up by the Cochrane Collaboration[21] with reporting in accordance with the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) statement. The prespecified protocol for this review was registered in PROSPERO (registration number CRD42021268228). The PRISMA checklist is available in Supporting Information: File 1.

Design and participation of stakeholders

In this SR, the representatives of patient associations such as the Spanish Association Against Cancer (AECC) and the Spanish Breast Cancer Federation (FECMA) participated in the review of the study protocol, as well as in the results, discussion and conclusions, having the opportunity to make their contributions. In addition, representatives of professional scientific societies related to the topic such as the Spanish Society of Medical Oncology (SEOM) were invited to participate. Although direct patient participation is not common in systematic reviews, we have integrated it into this study because we believe that the involvement of patients and other stakeholders in the research process is crucia.[22]

Search strategy

The electronic databases MEDLINE (using the Ovid platform), EMBASE (Elsevier interface) and WOS were searched from database inception to June 2, 2021. The search strategy included both controlled vocabulary and text-word terms related to ‘chemotherapy’, ‘alopecia’ and ‘hair loss’. The search was restricted to studies published in English or Spanish. The full search strategy used for each database is available in Supporting Information: File 2.

Selection criteria

Studies were eligible for inclusion if they fulfilled the criteria shown in Table 1.

Table 1 Study selection criteria

Design	Experimental, quasi-experimental, observational, qualitative or mixed-methods studies, systematic and narrative reviews and theoretical articles
Population	Adults (>18 years) with cancer eligible to receive, who are receiving or have received intravenous chemotherapy treatment and/or healthcare professionals using SC technology
Intervention	Application of the SC system
Outcome	Ethical aspects (patient values, morals, culture and autonomy, risk benefit ratio, human rights, dignity), legal aspects (laws, regulations, data protection, human rights, property and responsibility, market regulation), organizational aspects (process or work flow, planning or implementation, informational and training needs, acceptability) and social aspects (impact, perspective of patients and caregivers, experiences and preferences with cancer and the use of SC, important outcome measures for patients, self-management, information and support needs, acceptability)
Language	English or Spanish
Date of publication	No restrictions

Study selection

Two reviewers independently screened and in duplicate the title and abstract of retrieved references to identify potentially eligible studies. The full text of these references was then screened again in duplicate to confirm eligibility. Doubts and discrepancies between reviewers were resolved by discussion and, when no consensus was reached, a third reviewer was consulted.

Data extraction and quality assessment

Data were extracted from studies included by one reviewer and checked by a second reviewer using a piloted form in Excel format devised by the authors that included the following items: general information (authors, publication year, country and funding), study design, population, measures used, main findings, study limitations and conflict of interest.

Two reviewers independently and in duplicate assessed possible methodologic limitations of the studies included. We planned to assess quantitative, qualitative and mixed methods studies using the Mixed Methods Assessment Tool (MMAT).[23] The quality of systematic reviews and narrative reviews were assessed with the aid of the AMSTAR-2[24] and SANRA tools,[25] respectively. Descriptive studies, qualitative studies and mixed methods studies were then assessed using MMAT.[23] This tool permits to appraise the methodological quality of five categories of studies: qualitative research, randomized controlled trials, non-randomized studies, quantitative descriptive studies and mixed methods studies. For each relevant study, we used the corresponding criteria to appraise the study's quality, conduct the appraisal process and determined an overall quality score for each study. In the case of SANRA, used for the assessment of narrative articles, although the authors of the tool do not establish cut-offs for different grades of quality, we considered a score of ≤5 as low, 6–8 as moderate and 9–12 as high quality.

Data synthesis

A narrative synthesis of the main themes found was performed, taking into account criteria of relevance (applicability to the context specified in the review question), coherence (how clear and well supported are the data from the primary studies and from the synthesized outcomes provided by reviews) and adequacy (the degree of richness and amount of data supporting a review conclusion).[23,26]

RESULTS

From a total of 107 records initially identified, after eliminating duplicates, through the database search, 44 potentially relevant publications were selected after the title and abstract screening, of which 17 studies were finally eligible for inclusion according to the pre-established selection criteria (Figure 1). Table 2 shows the main characteristics and overall quality of the studies included. Detailed quality assessments of the studies are available in Supporting Information: File 3. The studies were published between 1996 and 2020, all in English. Five are descriptive studies,[28–32] four narrative reviews,[33–36] three qualitative studies,[37–39] two mixed methods studies,[40,41] two theoretical articles about implementation experience[42,43] and one observational study.[44]

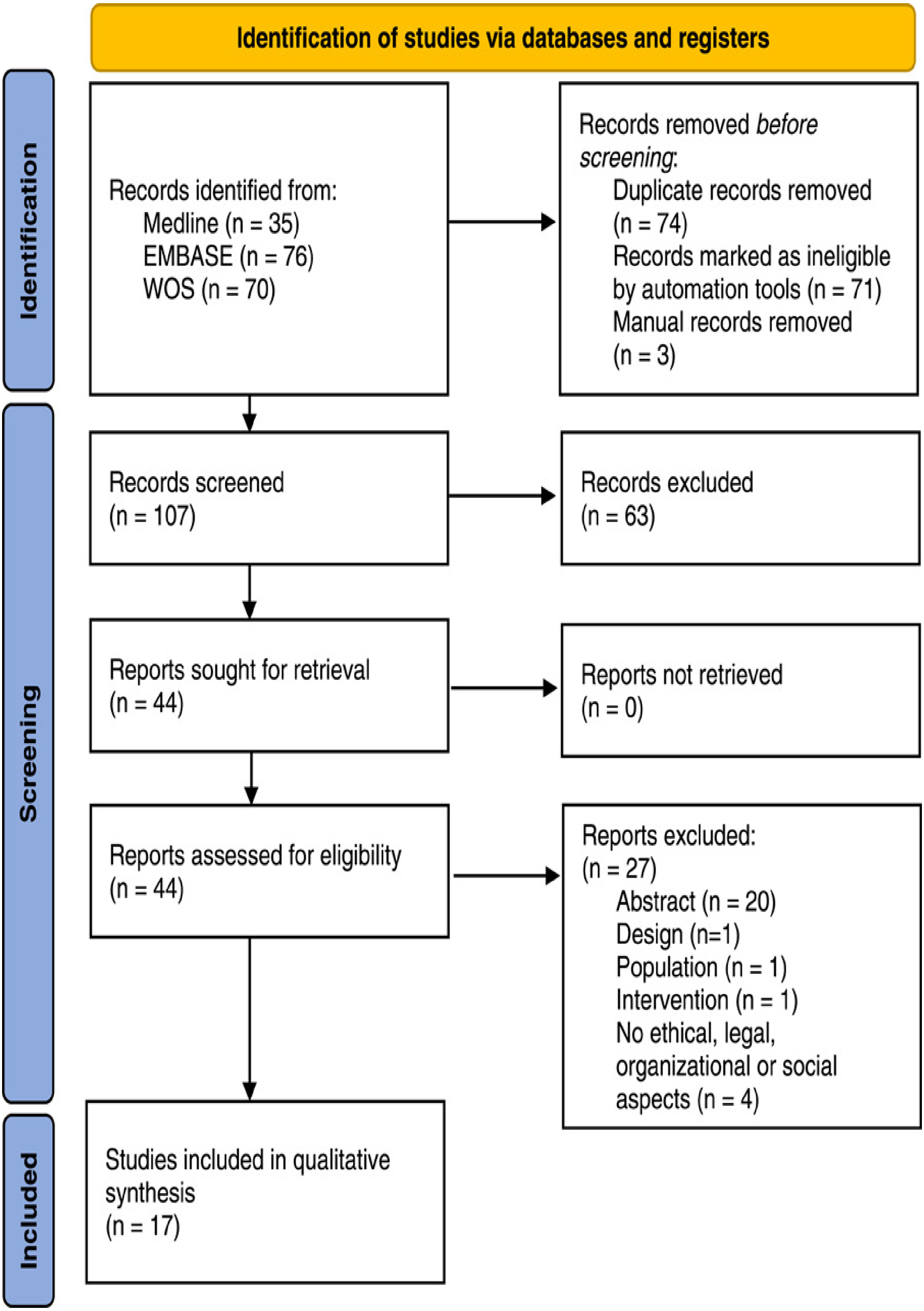

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Table 2 Main characteristics of studies included

References	Objective	Methods	Population (*N*)	Quality
Bitto et al.[28]	To assess the effects of the SC system on quality of life in patients with BC	Descriptive study using a questionnaire	BC patients (70)	Higha
Breed et al.[33]	Raise awareness of the impact of CIA to demonstrate the potential for prevention of CIA by SC	Narrative review	Patients undergoing chemotherapy	9 (High)b
Dougherty[37]	Know the opinion of patients about hair loss and SC	Qualitative study through semistructured interviews	Cancer patients undergoing intravenous chemotherapy	Lowa
Fischer-Cartlidge et al.[42]	Describe the experience of a large multisite organization that implemented an SC programme	Theoretical article: Implementation experience	Patients undergoing chemotherapy	NE
Heery et al.[43]	Describe the implementation of a SC programme in a community breast health centre	Theoretical article: Implementation experience	Patients with BC undergoing chemotherapy, users of DIGNICAP® (67) and PENGUIN™ (25). Patients with other types of cancer users of both systems (14)	NE
Lemieux et al.[29]	To know the opinion of health professionals on the use of SC in the reduction of alopecia in patients with BC and participation in an RCT	Descriptive study using a questionnaire	Patients with BC undergoing chemotherapy	Lowa
Massey[44]	To assess the efficacy of SC technology in patients receiving treatment for BC and to assess their views about its comfort and acceptability	Observational study	Patients with BC undergoing chemotherapy (94)	Lowa
Mols et al.[30]	To explore the attitudes of the nursing staff on ECC technology and the perception and opinion of alopecia	Descriptive study using a questionnaire	Patients with BC undergoing SC (98) and without the technology (168)	Lowa
Peerbooms et al.[31]	To know the familiarity, opinions and attitudes of patients and oncology professionals about SC technology	Descriptive study using a questionnaire	Nursing professionals (49) Oncology professionals (100) Former cancer patients (177)	Lowa

Peterson et al.[34]	To present a pragmatic workflow for collaborative efforts of healthcare professionals to provide SC in patients undergoing treatment regimens known to affect hair follicles	Narrative review	Patients undergoing chemotherapy	8 (moderate)b
Randall and Ream[40]	To explore the attitudes of the nursing staff on technology and the perception and opinion of alopecia	Mixed methods study (questionnaires and interviews)	Nursing staff Questionnaire (13) and interviews (3)	Lowa
Roe[35]	To explore the issue of CIA from the patient's perspective and SC as a preventive measure, along with a review of the evidence on the risk associated with the development of scalp metastases after SC	Narrative review and discussion article	Patients undergoing chemotherapy	9 (high)b
Shaw et al.[38]	To explore patients' perceptions and experience on SC	Qualitative study through focus groups and interviews	Patients with BC undergoing chemotherapy, both users of cooling systems and not (17)	Higha
Shaw et al.[39]	To qualitatively explore healthcare professionals' perceptions, barriers and facilitators to the implementation of SC in Australian cancer centres	Qualitative study using telephone interviews	Health professionals working in oncology (21) in five cancer centres, in some of which SC therapy was performed and in others it was not	Higha
Van Den Hurk et al.[32]	To explore the severity and burden of hair loss in cancer patients treated with chemotherapy, level of satisfaction with wigs, hair growth and body image	Descriptive study using a questionnaire	Patients with BC undergoing SC (98) and without the technology (168)	Lowa
Van Den Hurk et al.[41]	Provide up-to-date online information on CIA and SC to help patients cope with CIA and their choice of SC	Mixed methods study	Focus groups: patients with BC with CIA (9) and patients with BC who received SC (6). Semistructured interviews: SC-eligible BC patients (11). Questionnaires: nursing staff (10)	Lowa

Young and Arif[36]	To explore clinicians' barriers to the application of SC as well as to present data on recent studies	Narrative review	Medical and nursing professionals	6 (moderate)b

Abbreviations: BC, breast cancer; CIA, chemotherapy-induced alopecia; NE, not evaluated; RCT, randomized controlled trial; SC, scalp cooling.

a

Assessed with MMAT (Mixed Methods Assessment Tool).

b

Assessed with SANRA (scale for the quality assessment of narrative review article).

The studies included in the review were synthesized according to their relevance, coherence and adequacy, and different standard tools were used to assess the quality of the studies, depending on their design (Table 2). All the quality assessments of the studies are available in Supporting Information: File 3. We found that the quality of four of the five descriptive studies[29–32] was considered low, and only the study by Bitto et al.[28] had high quality. Aspects evaluated with this tool were the sampling strategy, the representativeness, if the measurements are appropriate, the risk of nonresponse bias and if the statistical analysis is appropriate. Although only three qualitative studies, the quality of two of them was considered high[38,39] and one[37] low. The aspects assessed for qualitative studies were the adequacy of the qualitative approach, the method of data collection if the findings are adequately derived from the data, the appropriate interpretation of results and the coherence. Two mixed-methods studies were also found,[40,41] but the quality was rated as low, the same as the observational study included.[44] The aspects assessed were the adequacy of the rationale, the integration of the different components of the study, whether the outputs were adequately interpreted, the divergences and inconsistencies between quantitative and qualitative results, and if the quality criteria of each method were followed. The SANRA tool[25] was used to evaluate the quality of narrative reviews, and thus the four narrative reviews included were of a moderate-high quality.[33–36] With this tool aspects such as justification, explicit aims, description of the literature search, referencing, scientific reasoning and appropriate presentation of data were assessed.

A thematic analysis of the review results was performed and the main findings were categorized according to four relevant dimensions: (1) ethical aspects focused on equal access, gender equity and doctor–patient communication supported by Patient Decision Aids (PtDAs); (2) patient perspective and acceptability; (3) professional perspective and acceptability; (4) organizational aspects focused on accessibility and feasibility. No results were found on legal aspects. A summary of the findings is shown in Table 3.

Table 3 Main findings

Finding	Studies that contribute to the findings
Ethical aspects focused on equal access, gender equity and doctor–patient communication supported by Patient Decision Aids (PtDAs)	
Health professionals should learn when to offer SC according to the characteristics of each patient. Patient education should include expected results, normal hair loss that may occur and proper care and precautions for hair. Professionals should consider possible causes of distress for their patients.	Dougherty,[37] Roe,[35] Peterson et al.[34]

Patient experiences influence oncology healthcare professionals' attitudes towards SC technology.	Shaw et al.[39]
Online decision-making tools, scientific information and practical advice on CIA and SC have been developed, as well as a value clarification exercise. https://www.scalpcooling.org/ (last access 11 August 2021).	Van Den Hurk et al.[41]
Patient perspective and acceptability	
Patients who undergo SC may have a mismatch between their expectations and their experience, as well as feel greater distress if their hair begins to fall out despite SC therapy, compared to those who do not undergo SC, since that for the latter, alopecia is an expected result of chemotherapy.	Breed et al.,[33] Shaw et al.[38]
Factors favoring acceptability are faster hair growth, the attitude of the nursing staff towards the SC.	Shaw et al.,[38] Shaw et al.[39]
Barriers to acceptability are the fact that the SC technology does not guarantee that the hair will be kept, having to spend more time in the hospital, the potential risk of skin metastases to the scalp, thinking that it may be too cold, inability to of tolerating the cold cap for 5 h, prioritizing hair colouring to mask grey, finding using colour powder for touch-ups too time-consuming or finding that washing hair only once a week was intolerable.	Peerbooms et al.,[31] Van Den Hurk et al.,[41] Heery et al.[43]
Professional perspective and acceptability	
Health professionals should discuss SC with both men and women in a way that enables men to discuss their concerns. They should not assume that men do not have concerns about hair loss, as they may also want to keep their hair during chemotherapy.	Randall and Ream,[40] Breed et al.,[33] Roe[35]
Nursing staff may be charged with informing their patients about the SC to provide them with sufficient knowledge about the risks and benefits, to make an informed decision.	Young and Arif[36]
Although oncology professionals consider the implementation of SC technology in their chemotherapy unit acceptable, they do not consider it feasible for patients to remain in the ward 90 min after the chemotherapy session to end the SC therapy.	Lemieux et al.[29]

Approximately 50% of oncology professionals consider the implementation of SC technology in their chemotherapy unit to be acceptable.	Lemieux et al.[29]
Some facilitators for the acceptability of the professionals include considering it as a service for their patients, the fact that patients actively request it, the participation of personnel in the decision-making for implementation and the commitment between the medical and nursing staff. Attitudes towards the need for intervention on alopecia and towards SC influenced professionals to defend the technology within their centres and offer the treatment to their patients.	Peerbooms et al.,[31] Shaw et al.[39]
Some barriers are the lack of evidence on efficacy and safety, little evidence about the risk of metastases to the scalp skin, logistic difficulties and lack of organizational support.	Peerbooms et al.,[31] Shaw et al.,[39] Fischer-Cartlidge et al.[42]
Professionals perceive that SC supposes an increased workload of the nursing staff. The development of protocols and records for the evaluation of results in daily practice in hospitals that use SC technology is recommended. Planning to manage changes to your workflow is an important precursor to implementation.	Breed et al.,[33] Shaw et al.[39]
Organizational aspects focused on accessibility and feasibility	
Equal access of patients to SC is a concern for the group of professionals since the number of patients exceeds the availability of machines, which can lead to unequal access to care in a universal health system.	Shaw et al.[39]
The subsequent cooling time of the SC is added to the treatment time, so the chemotherapy session space is not available to another/patient. It is necessary to ensure that patients, regardless of whether or not they use SC technology, do not exceed waiting times for treatment.	Roe,[35] Shaw et al.[39]
To address accessibility issues, it has been proposed: (a) to match the postcooling period with the administration of cytostatic agents that do not produce alopecia, when those are prescribed, (b) provide an additional room to which the patient can move with SC technology during the postcooling period and (c) assess using SC technology to enable two patients to be treated at once in open rooms.	Fischer-Cartlidge et al.[42]

For successful implementation, the support of the organization is necessary. Organizational support includes both increased funding for nursing time and provision of additional space to accommodate increased treatment time, as well as SC therapy-free spaces to reduce expectations of access for patients whom cannot receive this. Regarding the implementation of SC systems, it is necessary to consider the need for an interprofessional team, working with facility teams, training, taking into account medical resources and legal considerations, integrating technology into documentation, records and orders and good planning. The oncology nursing team could conduct patient education and reinforce adherence, which may positively affect their outcomes with SC technology.	Fischer-Cartlidge et al.,[42] Heery et al.[43]

Abbreviations: CIA, chemotherapy-induced alopecia; SC, scalp cooling.

Ethical aspects focused on equal access, gender equity and doctor–patient communication supported by PtDAs

The main concern expressed by healthcare professionals was the patients' access to SC, as for many there can be a lack of equal access.[39] On the one hand, this problem is mainly related to the limited availability of devices that affects accessibility and patient expectations.[39] On the other hand, in the healthcare context, although a large percentage of professionals (85%) believe that both women and men need support to discuss their concerns about CIA,[40] there seems to be a tendency for nurses to recommend SC more to women than to men.[40] Thus, the availability of SC and adequate information provided to the patient to make a decision are two essential attributes to contributing to equal access and gender equity.[33,35]

Communication between professionals and patients about how to cope with alopecia and available therapies to reduce it, such as SC, encourages evidence-based informed and shared decision-making. According to the nursing professionals' point of view, patients are not sufficiently informed about the associated risks of SC. Moreover, most nurses consider that patients are discouraged due to the required long duration of its use.[40] Communicating SC characteristics on correct hair preservation, restoration, care and maintenance precautions (expected outcomes and appropriate, patient-adjusted expectations) can reduce concerns and distress about CIA.[34] However, according to individual characteristics, it is necessary to know the most appropriate timing of treatment to enhance a positive experience with SC.[35,37] A study highlighted that the nursing staff is one optimal profile to inform about SC to chemotherapy-treated patients', as medical staff tend to offer SC to a lesser degree.[36] The findings of a qualitative study conducted with patients treated with SC to reduce CIA indicate that information about the efficacy of SC provided by oncology staff was verbal and within the framework of professional experience.[38] Patients reported that the information received on the SC process in terms of tolerability and hair care preservation, during treatment, was insufficient. However, patients with CIA who did not undergo SC reported that they did not receive information from their healthcare professional about SC as part of the treatment choice process. These patients were informed about SC through their peers, with no possibility of access to SC.

Finally, to facilitate effective communication about CIA and SC, and encourage informed decision-making among practitioners and patients, some authors have promoted the development of a PtDA in web format.[41] The PtDA is included in a website (https://www.scalpcooling.org/) which contains scientific information on alopecia and SC. Patients can complete a value clarification exercise here about their hair and how they perceive their personal likelihood of hair loss, as well as the safety of SC, ultimately receiving an overview with their own reasons for whether or not to choose the SC procedure, which can be printed out to discuss with healthcare professionals. When patients use PtDAs to make decisions, they gain more valid and accurate knowledge about the risks and benefits of available alternatives and are more likely to make decisions more consistent with their values.[41]

Patients' perspective and acceptability

The acceptability of SC among patients was high when applied in cases where this therapy has shown good results.

In the pilot study by Dougherty,[37] 50% of participants deemed that SC was worthwhile, and 50% also reported that they would use SC again. In another study, 82.22% of participants who obtained positive results with SC would recommend its use compared to 11.11% of those who obtained negative results.[28]

A factor affecting the acceptability of SC is the need for information before, during and after SC treatment.[41] Other factors influencing the acceptability of SC are interpersonal speed differences in hair growth (to initiate or continue therapy with SC). Nursing staff attitudes towards SC were also considered a key factor for initiation and continuity with SC.[39]

In contrast, reported barriers to accepting SC were the nonguarantee that SC maintains hair; increased hospital stay; possible risk of skin metastases on the scalp; coldness of the technique; perception of pain when using SC; additional time requirement, preferring hair colouring to mask grey hair, finding using colour powder for touch-ups too time-consuming or finding washing hair only once a week intolerable.[31,38,41,43]

From the patients' perspective, the majority of patients with breast cancer report that SC is poorly known before cancer diagnosis (73%).[31] Regarding the information received, 63% of cancer patients are satisfied with the information provided by hospital staff about SC, while 56% of patients dissatisfied with the information reported not having received information about SC.[31] Regarding comfort, the study of Massey[44] revealed that 85% of participants considered SC comfortable.

An important aspect to consider is the patients' feelings after SC treatment are: improved self-esteem (52%), improvement in quality of life (50%), disappointment (27%), hope (21%), insecurity (10%) and grief (6%).[31] From a psychological point of view, uncertainty regarding hair preservation in patients using SC may cause further distress, and severe alopecia despite SC may lead to additional disappointment.[33] In this sense, the qualitative study performed by Shaw et al.[38] on women with breast cancer found that despite the explanation received about the possibility of hair loss even using SC, some women did not match prior expectations with SC experience and experienced considerable anguish when losing their hair. Another source of stress for women who used the SC was hair care, due to the lack of information about the products to use, the frequency with which to wash their hair or because they did not know what information they should give to their hairdressers. Other women reported less hair loss than expected and were satisfied with SC's contribution to maintaining a better quality of life during chemotherapy. Nevertheless, women who did not use SC experienced hair loss but, as this was an expected consequence of chemotherapy, they reported less shock and were more likely to monitor and plan for hair loss by shaving their heads once hair loss began. In this study, the level of pain and discomfort associated with SC was considered insufficient to discontinue treatment, although some women reported that discomfort was too great to repeat a future treatment.

While positive results with SC predict better body image, compared to people who have negative outcomes or to those who do not use SC,[30,32] women using SC have a greater fear of radical mastectomy, scalp metastasis and alopecia.[32]

Professional perspective and acceptability

The acceptability of SC for healthcare professionals seems to be moderate. Although for most (85%) oncology healthcare professionals, SC is known to prevent CIA; only 50% consider its implementation in the chemotherapy unit acceptable.[29] On the one hand, the main facilitators for professionals' acceptability of SC are: seeing SC as a patient service[31]; responding to patient requests for access[39]; patient satisfaction with the experience[39] and professionals' personal attitudes towards the need for alopecia intervention and personal attitudes towards SC.[39] On the other hand, limited evidence on the efficacy and safety of SC mainly on the risk of scalp skin metastases, are the most important reasons for medical staff not supporting its use.[31,39,42] In addition, logistic difficulties in the hospital are the main reasons precluding nursing staff from supporting SC.[31] The nonavailability of SC in facilities on a routine basis, as well as the additional time and professional effort required by nursing staff, and limited access to SC were identified as the main barriers by healthcare professionals (not only nurses).[39] The lack of knowledge about which patient profiles benefit most from SC,[39] as well as difficulty identifying billing costs and processes without a similar intervention[42] have also been pointed out as barriers. Regarding the lack of knowledge, Peerbooms et al.[31] reported that 60% of nursing staff sought information about SC in scientific resources and exchanged knowledge with other professionals; 16% claimed the need for frequent SC training.[31]

Finally, Lemieux et al.[29] reported that 88% of oncology healthcare staff believed that clinical trials on SC were necessary, and consequently, 85% would recommend their patients start a clinical trial on SC.

Organizational aspects focused on accessibility and feasibility

Feasibility seems to be one of the main problems from healthcare professionals' perspectives. Thus, 72% of healthcare professionals do not consider it feasible to extend patients' stay in the treatment room to receive SC after the chemotherapy session has finished.[29] In terms of workload, limitations in trained nursing staff were considered an impediment to the routine use of SC.[39] Lack of continuous healthcare professional training, as well as lack of knowledge about SC, generates worse results in patients, which fosters the view that it is not worth the extra time and extra nursing work.[39]

Regarding the implementation of SC, several studies have reported barriers and facilitators.[39] Reported barriers include system-level change in patient flow and working practices; lack of organizational support to increase funding for training and nursing staff time, and provision of additional space to see to increased time during chemotherapy treatment.[39] From the professionals' point of view, these authors found that physician and nurse involvement in decision-making was a relevant facilitator to implement SC. The study by Heery et al.[43] considered that a key aspect of SC implementation is to ensure the availability of SC-trained individuals to accompany patients eligible to participate in SC programmes. During the implementation of SC, the best-trained people were female healthcare providers, patients or former caregivers. Ensuring patient adherence is also an essential component of SC implementation.

Some initiatives to improve the implementation of SC have been reported. According to Fischer-Cartlidge et al.,[42] some improvement initiatives are (1) matching the postcooling period with the administration of cytostatic agents that do not produce alopecia when those are prescribed; (2) enabling an additional room where the patient can move to with the device during the postcooling period and (3) assessing the possibility of using machines that enable two patients to be treated at the same time in open rooms. The need for real-world data monitoring and evaluation is also relevant.[33] Recording success rates with SC by involving patients in recording hair loss, together with resource use and costing data will add valuable information at health policy, managerial, clinical and patient decision levels. In this way, the outcomes assessment in daily clinical practice in some hospitals in The Netherlands proved to be very effective for chemotherapy programmes.[33]

DISCUSSION

The main goal of this SR was to assess the ethical, legal, organizational and social issues involved in SC use by oncologic patients. Overall, the quality of the studies included was low-moderate. Findings were organized according to relevance into the following topics: (1) ethical aspects focused on equal access, gender equity and doctor–patient communication supported by PtDAs; (2) patient perspective and acceptability; (3) professional perspective and acceptability; (4) organizational aspects focused on accessibility and feasibility.

First, the review has pointed out the necessity to avoid the possibility of gender inequity when implementing the SC system. Thus, to avoid gender inequity, SC therapy should be offered equally to men and women, which avoids the assumption that men care less than women about maintaining their hair. In this regard, having clear protocols on which population to offer this therapy to and how to offer it can help minimize the influence of professionals' attitudes towards both CIA and SC.[31,39] However, the introduction of SC technology should not affect the ability to treat other patients, so waiting times for chemotherapy treatment should not be affected in any case.[35,39] This poses an organizational planning challenge, especially in properly managing the postcooling time that needs to be added to the chemotherapy regimen time. This is one of the key aspects that must be correctly planned before implementing the technology. Other ethical issues related to SC, such as whether SC should have some restrictions regarding age considerations, were not included in this review because no evidence was found addressing this problematic topic.

From the patients' perspective, the present review has shown how important it is to have enough detailed information before and during the use of SC, not only regarding the technology, but also hair care in general. When

SC is available, adequate information and communication dynamics need to be implemented with all patients so that they can decide according to their values and preferences. To do this, decision support tools could be of great value, [41] since these tools aid patients to clarify their own values about CIA and the impact of this on their own lives. The experience and feelings of each patient may be different, and for some of them, the prior expectations about SC might not be met by the results, which can lead to feelings of considerable anguish if they lose their hair. Due to the aforementioned, the nurses' and doctors' communication with patients and information provided about SC therapy is essential to reduce an imbalance between expectations and patient outcomes; as well as to identify potential sources of distress regarding hair preservation.[33,37,38] Beyond providing information on the efficacy and safety of SC, information on hair care before, during and after the sessions is an important aspect for patients in chemotherapy treatment.[41] Patients can complete a value clarification exercise about their hair and how they perceive their personal likelihood of hair loss, as well as the safety of SC, ultimately receiving an overview with their own reasons about whether or not to choose the SC procedure, which can be printed out for consultation with healthcare professionals. When patients use PtDAs to make decisions, they gain more valid and accurate knowledge about the risks and benefits of available alternatives and are more likely to make decisions more consistent with their values.[41]

From an organizational point of view, patient access to SC should be ensured in circumstances where the demand exceeds the availability of devices to minimize access inequalities.[39] For this reason, the number of devices that each centre will require, the increase in nursing staff required, as well as the adequacy of additional space, must be carefully analysed and managed.[42] The possibility of using machines that enable two patients to be treated at the same time in open rooms, as well as optimizing the use of SC machines is another factor that may help reduce the nursing workload.[42]

Regarding the implementation of SC, the literature points out some aspects worth highlighting. A crucial element for the successful implementation of this technology is planning ahead to manage changes in the workflow of the healthcare system. Healthcare managers should design an implementation plan with the help of the centre's interprofessional team, which could increase the acceptability of SC technology by professionals once it is implemented.[39] Good planning should also include the recording systems to be introduced, which will enable the technology's impact to be assessed.[33] Increased workload of the nursing staff can be reduced by strategies such as involving patients in the hair loss registry, preferably online.[33]

Moreover, Nangia[45] stressed in a commentary that, due to the fact that the use of these devices requires new processes to be implemented by infusion centres, some providers and administrators may think it is too much effort to offer this service.

This SR provides results that are of interest to all the stakeholders involved in SC. Firstly, for patients who are interested in using SC, this SR can provide a bigger picture of the available evidence about patients' perspectives on this technology, which can be helpful to better inform their personal decisions. Second, doctors and nurses who may be interested in using SC in their centres could find these findings of great value. Third, the results of the present SR can also have an impact on policymaking when the healthcare systems are considering whether to introduce SC. In the case of SC, as the report[19] shows, at the present time, it is not possible to recommend the use of SC for the prevention of CIA in women with breast cancer from the evidence available on effectiveness and cost-effectiveness. This is mainly due to the low certainty associated with the long-term safety of the technology. In these cases of uncertainty, it is even more relevant for policymakers to consider other factors involved in the technology, such as those considered in this SR. Considering all these factors can help to improve decision-making from the policy-making point of view. And finally, it also might be interesting for the industry to better understand the main concerns that healthcare professionals find regarding the feasibility of the technology.

Strengths and limitations of the review

This study has some strengths and offers valuable information for patients, healthcare professionals, managers and policymakers. Despite the recent publication of an SR on the efficacy and safety of SC,[13] no specific publications have reported on patients' and professionals' perceptions and attitudes in terms of ethical, legal, organizational and social aspects. The SR by Rugo et al.[13] reported on the effectiveness of SC and overlooked the attitudes and perceptions of patients and healthcare professionals, as well as the ethical, legal, organizational and social issues involved in the widespread use of SC. The present review is the first to address patients' and professionals' perspectives on SC therapy, as well as ethical, legal and organizational aspects. Another strength of the study is the ability to integrate all the actors involved in an evaluation process in a single search, from the organizational and institutional level to the patient's point of view, including the professionals' point of view.

However, this SR has certain limitations. First, the possibility that some studies have not been included because they are not drawn up in English or Spanish or because they are not indexed in the databases consulted. Another limitation is the type of study design; studies with a more robust methodology, which takes into account all patient aspects (from ethical to acceptability) are necessary.

This SR has tried to encompass all factors involved in the assessment of technology, from organizational to more person-centred levels. Future SRs could focus on an analytical and critical review of this model, offering sustainable improvements in each step proposed. One study included in this review reported that, when patients use PtDA, in addition to helping them clarify their values and preferences, they had more realistic expectations about the possible benefits and side effects of their treatment options.[41] Evidence has shown that when healthcare professionals receive training focused on a person-centred care (PCC) model in the hospital setting, it improves their knowledge of the model and enables them to educate their patients in its use, with the help of PtDAs.[46] Therefore, future research could focus on the evaluation of training programmes on the PCC model for healthcare professionals in this area, and on the development and assessment of PtDAs to make better-informed decisions together with their practitioner.

CONCLUSIONS

This SR provides important learning points relevant to patient care, decision making and organizational policy. When ethical, legal, organizational and social issues related to the use of SC for the prevention of CIA were examined, the importance of equal access, which includes the need to offer SC to everybody, without assuming gender differences, to address concerns about hair loss and impaired perception of body self-image was revealed. From a communication and educational perspective, the PtDa was found to be relevant to assist patients in clarifying their values and preferences, as well as the need for good communication with the healthcare staff team to adjust patients' prior expectations to reduce the potential distress associated with hair loss during SC use. This is important because the SR revealed that even when patients have received information, their expectations can differ and when patients use a PtDa they have more accurate expectations of the possible benefits and drawbacks of their options and are more likely to make decisions that are consistent with their values. In summary, we believe that this SR can benefit patient decisions, communication among healthcare staff and broader organizational considerations about SC. It can also help policymakers in deciding whether to include SC in their services, integrating other aspects complementary to efficacy and cost-effectiveness. The integration of all relevant ethical, legal, organizational and social aspects can facilitate a more accurate and better-informed decision-making process.

AUTHOR CONTRIBUTIONS

Janet Delgado Rodríguez and Vanesa Ramos-García: Investigation; conceptualization; supervision and data analysis; writing –original draft; review and editing. **Diego Infante-Ventura and María del Mar Trujillo-Martín**: Methodology; writing –original draft, review and editing. **José Carlos Suarez-Herrera and Antonio Rueda-Domínguez**: Writing –review and editing. **Pedro Serrano-Aguilar**: Writing –original draft, review and editing.

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CONFLICT OF INTEREST

J. D. R., V. R. G., D. I.-V., P. S. A. and M. M. T. work for the same research entity that received the funding. The remaining authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

Data are available on request from the authors.

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The elephant in the room: Exploring the influence and participation of patients in infection-related care across surgical pathways in South Africa and India

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ABSTRACT (ENGLISH)

Objective

The irrational use of antibiotics is a leading contributor to antibiotic resistance. Antibiotic stewardship (AS) interventions predominantly focus on prescribers. This study investigated the influence and participation of inpatients in infection-related care, including antibiotic decision-making, within and across two tertiary hospitals in South Africa (Cape Town) and India (Kerala).

Methods

Through ethnographic enquiry of clinical practice in surgical pathways, including direct nonparticipant observation of clinical practices, healthcare worker (HCW), patient and carer interactions in surgical ward rounds and face-to-face interviews with participants (HCWs and patients), we sought to capture the implicit and explicit influence that patients and carers have in infection-related care. Field notes and interview transcripts were thematically coded, aided by NVivo 12® Pro software.

Results

Whilst observational data revealed the nuanced roles that patients/carers play in antibiotic decision-making, HCWs did not recognize these roles. Patients and carers, though invested in patient care, are not routinely involved, nor are they aware of the opportunities for engagement in infection-related decision-making. Patients associated clinical improvement with antibiotic use and did not consider hospitalization to be associated with infection acquisition or transmission, highlighting a lack of understanding of the threat of infection and antibiotic resistance. Patients' economic and cultural positionalities may influence their infection-related behaviours. In the study site in India, cultural norms mean that carers play widespread but unrecognized roles in inpatient care, participating in infection prevention activities.

Conclusion

For patients to have a valuable role in AS and make informed decisions regarding their infection-related care, a mutual understanding of their role in this process among HCWs and patients is crucial. The observed differences between the two study sites indicate the critical need for understanding and addressing the contextual drivers that impact effective patient-centred healthcare delivery.

Patient or Public Contribution

Ethnographic observations and interviews conducted in this study involved patients as participants. Patients were recruited for interviews after obtaining signed informed consent forms. Patients' identities were completely anonymized when presenting the study findings.

FULL TEXT

INTRODUCTION

Antibiotic use in human populations remains a key driver of antibiotic resistance (ABR) worldwide.[1,2] Unregulated access to antibiotics for patients and the public is an additional risk for the emergence and spread of ABR.[3–6] However, the lack of access to effective and affordable antibiotics continues to cause more harm than the risk of ABR.[7] Optimizing antibiotic use requires finding a balance between the need for regulation and control of excessive antibiotic use versus ensuring adequate access to those who need them. To do this effectively, we need to engage with the public and patients to bring about a culture of civil society responsibility for antibiotic use.[8]

Whilst ABR remains a global challenge, its impact is more significant in low- and middle-income countries (LMICs).[9] In the context of surgery, addressing behaviours and practices related to infection management and antibiotic use across the surgical pathway (before, during and after surgery) is key to tackling important drivers of ABR and to decreasing the burden of surgical infections globally.[10–12] Currently, antibiotic stewardship (AS) interventions focus predominantly on prescribers, rarely considering the patient's role in antibiotic decision-making and consumption.[13] This is at a time when patient-centred care, including infection-related care, is becoming increasingly relevant.[14] Very few studies exist on patient involvement in AS, particularly in LMIC settings.[15] There is also a knowledge gap in healthcare professional views on patient involvement in infection-related care. This is an important gap to explore as patient involvement is in part dependent on the willingness of healthcare staff to participate in the process.[16,17] Managing inpatient care, in ways that recognize the patient's role as a participant, remains challenging particularly in surgical pathways.[15,18] The surgical patient's involvement in pre- and postoperative care is crucial to optimized recovery, and yet remains difficult to achieve.

Arnstein[19] provides a foundation for research into patient involvement, describing eight rungs of citizen participation from least to most inclusive: Manipulation, Therapy, Informing, Consultation, Placation, Partnership, Delegated power, Citizen control. Different models for types of patient involvement have been described, one of which is ‘co-production’, or patient participation, defined as ‘user co-delivery of professionally designed services’.[20] At the point of care delivery, this type of involvement describes an approach where health professionals work together with patients (and/or carers) as partners to achieve optimal care. A partnership approach may not, however, be appropriate for all patients under all circumstances, and it has been argued that ‘participation should be defined by whatever level the patient is most comfortable with’.[21]

To become effective participants in their own infection-related care, however, patients need to understand the basics and complexities of infection transmission and resistance and the role of AS to address ABR. Efforts to raise patient awareness have included written information on websites and leaflets and posters targeting healthcare users and the public, to mention a few. However, patients and the public in general may be unable to grasp the immediate threat of ABR, partly due to a lack of effective communication strategies and confusion related to the advice and information provided on the subject.[22–24] Additionally, the social determinants of health influence clinical interactions and outcomes,[25,26] posing limitations to patients' ability to effectively participate in their own health care.[27] In relation to infection-related care, these factors and how they may influence health-seeking and health-providing behaviours remain understudied.[27]

This study aims to explore patient involvement in infection-related care and the social and cultural factors that influence this from the perspectives of health professionals and patients and their carers in two tertiary hospitals in South Africa and India.

Study context

South Africa is an upper middle-income country and India is an LMIC. Over 76% of the global antibiotic consumption between 2000 and 2010 was attributable to the BRICS countries, with India leading the group as the largest consumer of antibiotics in human health.[28] Availability and accessibility to health systems are a challenge in LMICs. In South Africa, the majority (up to 70%) of patients access healthcare through the public sector.[29] In India, whilst public sector healthcare services are available free of cost to patients, due to limited staff and supplies at government facilities, many seek care from the private sector (reported up to 65%), paying out-of-pocket.[30] Annually, 3646 surgeries per 100,000 population are performed in India in comparison to the global estimate of 5000 surgeries per 100,000 patients.[31] South Africa's surgical capacity has been noted to be below international requirements, with a greater concentration of available resources in urban areas.[32,33] Kerala, the state where the study site in India is located, is an atypical state with high literacy rates and better healthcare access and infrastructure compared to the rest of the country.[34] The participating study sites were selected because despite operating in health systems with limited resources, they perform significant number of surgical interventions and have established strategies to rationalize the use of antibiotics.

Adult gastrointestinal (GI) and cardiovascular and thoracic surgery (CVTS) specialties at academic tertiary referral hospitals in South Africa (site A) and India (site B) were included in this study. Surgical specialties were selected to represent high infection and/or mortality risks.[10,35] Site A is a 950-bed government-funded public hospital in Cape Town, while site B is a not-for-profit 1350-bed private tertiary centre in Kerala. The GI team at site B features an experienced team of GI, hepatobiliary and pancreatic surgeons. The liver transplantation team within the department is credited with 850 liver transplants and is the largest Liver Transplant Programme in Kerala. The CVTS specialty at site B is one of the busiest in the country, with over 2000 cardiac surgeries being performed annually.[36] Both sites provide subsidized care for some patients based on income status, run established AS programmes and play key roles in AS initiatives in their respective contexts.

Conceptual framework

We investigated the explicit and implicit influence and participation of patients and carers in infection-related decision-making across the participating multidisciplinary teams. Whilst we used the definition of culture coined by Spradley: ‘the acquired knowledge people use to interpret, experience, and generate behaviours’,[37] its application to the clinical context is built upon our existing research spanning different countries in the last 10 years.[11,12,38–42] The existing research describes the role of hierarchies and the need for clinical autonomy in infection-related decision-making in inpatient settings, wherein senior doctor autonomy overrules policies.[11,38,40] Recognizing the gap in knowledge, we have expanded on this research to consider the role of patients and carers in infection-related decision-making.[15,42]

METHODSStudy design

Data were collected between June 2018 and November 2019 by trained researchers through nonparticipant ward round observations and semi-structured face-to-face interviews with patients, patient carers and healthcare workers (HCWs). Documentary analysis of inpatient records, and the policy and guidelines on antibiotic prescribing provided contextual knowledge.

Nonparticipant observations

Data were gathered from general ward and intensive care unit (ICU) rounds. Four trained researchers and their trainer took notes of their observations, specifically on the following: place, the people involved, actions of participants, related activities carried out, tasks and results that participants tried to accomplish, emotions felt or expressed, the major events that occurred, the discussions that took place, who led the discussions, who acted upon identified plans. In Site B, additional data were gathered from the outpatient clinics, operation theatres and during departmental meetings. A previously used and tested data collection guide[39] facilitated data consistency.

Face-to-face interviews

Study participants were recruited using convenience sampling and participation was voluntary, at a place and time convenient for the participant. A semistructured interview guide was used for the interviews, differentiated for patients and HCWs. In addition to this, questions that came up during observations were put forth for discussion. Interviews were conducted by the four trained researchers (two trained researchers at each site), with or without the study lead who had provided training.

Study participants

All HCWs involved in patient care in the surgical specialties of interest were eligible to participate. This included HCWs with different roles, experiences and expertise in the surgical teams and those from nonsurgical teams who had input into the care of surgical patients (e.g., the AS team). Patients admitted to any of the surgical specialties of interest were eligible for inclusion in the study. For the interviews, patients who were prescribed therapeutic antibiotic(s) while under the care of the surgical team(s) were invited to participate. Participants were selected using the purposive sampling technique.

Data analysis

Before analysis, a coding framework created by the four trained researchers and the study lead was validated through group discussions. Data from each setting were thematically analysed by researchers. Field notes and interview transcripts were analysed using the grounded theory approach—a method extensively used by the research team and published[11]—aided by NVivo 12® Pro software. Analyses of data were iterative and recursive, using constant comparison. Following analysis, the researchers discussed emerging themes for revision as required. Redundant themes were removed and other themes were collapsed or expanded as necessary. The analysis process for each study site was undertaken separately to avoid analytical bias between sites.

The different data collection methods of ward round observations, face-to-face interviews and HCW and patient/care interactions provided cross-validation and triangulation of findings. To mitigate professional biases, our research team included two pharmacists, one anthropologist, one nurse, three infectious diseases specialists and five physicians from a range of surgical specialties (GI, general, cardiothoracic and emergency). The diversity of backgrounds enabled us to consider our role-related biases and examine different perspectives throughout the analysis.

RESULTS

In site A, data were collected from 72 h of observations, including 960 episodes (659 in GI and 301 in CVTS) of bedside discussions with patients and/or their carers. Interviews were conducted with 61 HCWs and 7 patients. In site B, data were collected from 138 h of observations, including 883 episodes (399 in GI and 484 in CVTS) of bedside discussions with patients and/or their carers. Interviews were conducted with 44 HCWs and 6 patients with/without their carers.[12] Whilst attempts were made to recruit more patients during the data collection phase of the study, we found recruitment to be challenging, as many patients, unfamiliar with participation in research, were not comfortable with providing consent to be interviewed. The observations of communication between the HCWs, patients and their carers during ward rounds provided rich insight into the boundaries of patient and carer roles and participation in infection-related care. Analysis of data from across the study sites identified the following themes: lack of understanding of the threat of infection and ABR and the patients' positionality—both culturally and economically—and how this may influence the extent of their involvement in decision-making.

A lack of understanding of the threat of infection and ABRAwareness of infections

While some of the patients knew that antibiotics are for treating infections, general awareness seemed to be lacking regarding the specific infection being treated (X1; Table 1). Some patients associated clinical improvement with antibiotic prescription and use, while others discussed the need for antibiotics to treat a virus (X2, X3, X4; Table 1). Prescribers also feel the demand for antibiotics from some patients in site B (X4; Table 1). Patients report a feeling of stigmatization with having an infection, with some demonstrating a lack of understanding of the processes involved in their care (X5; Table 1).

Table 1 Excerpts from study data (interviews and field notes).

Theme	Excerpt ID	Excerpt from data

A lack of understanding of the threat of infection and ABR	X1	‘There was no reason for the presence of germs in my blood. I was told that it was due to blood transfusion. Three persons came to give blood from my son's workplace. Then younger son's friends also came. They are not bad boys. We cannot say the germs are from their blood; or cannot say the germs are in my body …I cannot blame anybody. Any way I was given antibiotic and I had it’. Patient, CVTS, India
	X2	‘Most of the people actually they want some medicine like antibiotic …when they come for review, they will ask why I was not given an antibiotic, my wound is open and all, so the problem is there, they expect some antibiotics from us. They think that antibiotic is secure for them’. GI Surgeon, India
	X3	‘I am getting antibiotics. They are going to give me a certain type of antibiotic …the bottle is now not here …but it is a special antibiotic for that virus’. Patient, GI, South Africa
	X4	‘…there is always a feeling that, you know, if the patient becomes unwell and comes back to the ICU, many of them would ask you that is it because you have not given them an antibiotic’. GI Surgeon, India
	X5	‘In the beginning, I felt so bad; I thought it was my fault (referring to contact precaution notices). Now I see them on other doors and so, I don't feel bad anymore’. Patient, CVTS, South Africa
	X6	Senior Registrar (SR) 1 informed the patient that she can be discharged tomorrow but patient was very reluctant. SR1 suggested that he will discontinue all IV medications tomorrow and the day after the patient can be discharged to the guest house. He suggested the patient can stay there for two days and see if there is any recurrence of pain. If there is no issue, then they can go home. Patient appeared to be happy with that option. SR1 pointed out that if she stays in hospital for longer there is a chance of getting infection. Field notes, GI surgery, India

	X7	‘…that was after the operation. they gave me a tablet, yes, one or two …yes, for the pee, maybe, yes, maybe that was the antibiotic …I couldn't keep my pee in. They gave tablets because every time I have a pee0. What is that? What do you call that? Isn't that infection?’ Patient, GI Surgery, South Africa
	X8	‘If somebody is educated and aware, these are the people who would not want to use antibiotics. Even for the children, they would say that, you know, antibiotics are not good’. GI Surgeon, India
	X9	‘Well, it is not that we are going just to educate regarding the antibiotic. Obviously, it comes up in conversations, and then we take it from there. It is not like we go and counsel every patient about the antibiotic use, and in our state, I think it is not that bad, you know, compared to other states of you know India because literacy level is high, and you know even people understand that you do not need the antibiotic for everything’. GI Surgeon, India
	X10	The update mentioned that the patient had a temperature of 38.8°C the day before. Consultant 1 asks the patient if he has burning upon urinating to which the patient responded no. Consultant 2 looks at the wound and asks if the patient is on antibiotics. Consultant 1 confirms that the patient is on antibiotics and provided the name and route of administration. Consultant 2 tells the patient that he (the patient) has temperature spikes, probably due to an infection. Consultant 2 tells the patient that he is already on antibiotics but that they need to sort out the cause of the spike in temperature. Field Notes, GI Surgery, South Africa
	X11	‘After the suture removal, the surgical resident informed the carers that wound is still infected and antibiotics are being administered. He also informed that there is a suspicion of urinary tract infections and advised to call and enquire about urine culture report to change the antibiotic if needed’. Field notes, GI surgery, India

	X12	‘We do visit them and talk to them once they are shifted to the ward. Sometimes they would want to know how is it after the transplant, what is the dietary changes needs to be made. There is a dietician to direct them, but still we go and talk to them. We educate them about the infection control like they have to hand wash regularly, the bystander should wear the mask’. MSW, GI Surgery, India
Patients' positionality, both cultural and economic, may influence the extent of their involvement in decision-making	X13	The consultant turns to the patient and asks if he knows when the next stage of his procedure will take place. The patient says that he does not know, adding lightly that he did not know there is another stage. The consultant tells the patient that he must ask questions about anything that is not clear to him (the patient)—to any one on his management team—and proceeds to explain the stages of the patient's management, what has been done so far and what may still need to be done. Field Notes, ICU, South Africa
	X14	‘The patient doesn't compel us to start the medication because they are not aware of those things. They do say that they are feeling feverish. I saw the wound sores while bathing. They don't know that the medicine needs to be started for that. They tell the doctor also that the wound was wet, there was discharge and things like that, but they don't suggest an antibiotic to give them…’. GI Staff nurse, India
	X15	‘Often, we will talk to a patient and if the patient says, “I don't want to know, just take the responsibility,” and it happens more often than not …when you get to that, it just becomes habit forming …it's the only way I have known …the profile changes completely when you are dealing with private patients because they are generally much better educated and they have much higher demands’. CVTS Surgeon, South Africa

	X16	‘Not as a rule, you would see patients saying these doctors didn't wash their hands when they examined me …I think we haven't really got to the point where the patients are litigation conscious, we don't have that culture yet. It is getting there, and it depends on the patient, some patients are more informed than others. It will change I am sure particularly in the private sector. But it is a good change, probably what we are trying to get out there. Everybody takes responsibility for infection control’. AS Staff, South Africa
	X17	‘We can't get through to the staff it feels like you are talking to a brick wall …so, maybe we should just start empowering our patients. We have designed a few patient pamphlets ourselves, and in the little pamphlet we ask, “did you ask the health care worker, did the health care worker wash his or her hands before they touched you? Were you able to remind them?” …but our patients come from poor socioeconomic backgrounds, most of our patients so they are just grateful for the care they are going to receive and we know, it is human nature, if I am going to say to you, please wash your hands before you, I am going to get your back up and so you know what I am just going to do the bare necessities here and I am going to walk away and I am not going to communicate with a patient so that is the nature, patients don't want to be victimized’. IPC Staff, South Africa
	X18	‘My husband and sons took all the decision about my care while I was in hospital. It is all their choice. They won't ask anything’. Patient, CVTS, India
	X19	Carer of a patient came in [patient not present]. The surgeon greeted him. Carer explained the patient's history to the surgeon and showed the reports of the tests done in an outside hospital. After reviewing the reports, the surgeon explained to the carer that the patient cannot be operated on due to advanced age and clinical condition. He suggested that it would be better to continue palliative care for the patient. He apologized to the carer for not being able to help them. Field notes, GI surgery outpatient clinic, India

	X20	‘I had a doubt since I am a nurse, I know normally after any surgery, they start with antibiotics, right? But I didn't see them giving any injection at all. When I asked the nurse, the nurse told that now without antibiotic it will heal, there is no need for antibiotics. On the fourth day, my daughter who is also a doctor, will be calling asking for the culture result. I too would ask but they would tell it has not come yet. I know, in our hospital if there is no outcome in 24 hours, they will give preliminary as no growth, but here even after four days, the results are not out yet. Even after the culture result came that night, they did not inform any doctor and they did not start any medications too. They did not change the antibiotic based on the growth’. Patient carer, CVTS, India
	X21	The consultant then mentions that the patient's family is coming from a coastal town approximately four hours away and that they need to talk to the family. Field Notes, CVTS, South Africa
	X22	‘We refer them all to the doctors. They are here all the time …we just say, “Doctor the patient or the family is asking questions, can you explain to them?” Then he explains to them’. Nurse, CVTS ICU, South Africa
	X23	‘If I am able to convince the patient, the patient will go back happily. If I am not able to convince the patient, well the patient will go from me to another surgeon, and then to another surgeon, ultimately to a surgeon who will actually prescribe an antibiotic and then he would be happy there, so he will go off doctor shopping’. GI Surgical Resident, India
	X24	‘I was transferred here when my funds ran out. If I had known of the wonderful care I would have received here, I would have said from the beginning, “Take me to this hospital” …There in private, it was all about the money, but here it is all about the care’. Patient, GI Surgery, South Africa

	X25	‘….so even then in some patients, who have complete financial restraints, we will have to maybe reduce the doses or take into consideration other drugs…. the other thing that happens is they will want to go to another hospital also, so probably somewhere in medical college where the medicine is free, we would recommend that’. Pharmacist, GI surgery, India
	X26	‘I think it is more a bit of, a case of, they're in a hurry, you know; like the lady that was helping me to eat—you know, and I've noticed—this morning when there was time, she had on an apron but when she came through now she didn't wear an apron, but, it is all right’. Patient, GI surgery, South Africa
	X27	‘Patients’ role is also there because some patients themselves ask the doctor, suppose they come to outpatients and they would have read up on something and they will be asking whether they should be on antibiotics. In such cases, the patient has to be reassured that they do not require an antibiotic’. GI Surgical Resident, India
	X28	‘Sometimes it is just the patient's pressure, that the patient might not feel that I am a good enough doctor in case I have not satisfied the patient's prescription as well and satisfying the patient is also a very important part of our practice. Whether we satisfy them by prescribing what they want or by convincing them that they actually do not need it. Either way the patient has to be satisfied, right, and ultimately I think the patient is satisfied. The patient wants results. So with an antibiotic or without an antibiotic, if the patient actually can get well, if that convinces the patient beyond any doubt that yes, he did not need an antibiotic, or did he need an antibiotic at that point’. GI Surgical resident, India

	X29	‘Some of the patients do tend to ask for antibiotics because they have been used to these five days of antibiotic [to be] necessary. We do tell them that we follow whatever is the standardized protocol all over the world, i.e., give prophylactic antibiotic and repeat every four hours during the surgery. Postop, unless there are signs of infection or anything, we do not give any antibiotic, in that case usually they understand. I hope they do not go back and buy it on their own’. GI Surgeon, India
	X30	‘I find it much simpler to prescribe three days of ofloxacin to somebody rather than face a litigation based on completely unscientific allegation by somebody from outside’. GI Surgeon, India
	X31	‘Patient expectations, I think defensive medicine, a genuine fear of harming the patient by withholding therapy, and using inadequate diagnostic tests. It's really hard, I'm not saying it's easy …most general practitioners in South Africa are in private practice and it's a business; so, if you don't give your patient what they want, they'll go somewhere else and your livelihood is at risk’. AS Physician, South Africa
	X32	‘It is a real problem, and in India, there is an even bigger problem out in the community, so as you know, patients who go to see a clinician, a general practitioner, usually get antibiotics. So even if it is a viral fever, they might end up getting an antibiotic …you will be surprised that some of the antibiotics that you would think three times before using even in tertiary care centers, is used very frequently [in a small hospital]’. GI Surgeon, India
	X33	‘The cost of antibiotic especially when they undergo treatment in a hospital like this [is an issue]. Many patients would not be able to afford high end antibiotics. Sometimes, we will [switch therapy] to lesser antibiotics because we have no other option’. GI Surgeon, India

	X34	‘If we had started IV antibiotic from the beginning, probably she would have improved quicker. We had initially begun treatment with intravenous antibiotic but since she could not afford it, it was changed to oral. Even though the microbiologist had suggested [stronger] antibiotics, we could not prescribe those as she wasn't willing to buy them. We prescribed a [stronger] antibiotic towards the end when we could arrange it for free from the hospital pharmacy. However, this could only be given for two days’. GI Surgeon, India
	X35	‘In India, there is no restriction on over-the-counter medicines, you will find the pharmacist dispensing [without prescription]. People are consuming antibiotics much more than what it used to be in the past’. GI Surgeon, India
	X36	‘Absolutely, [previous exposures to multiple antibiotic] limits our choice of antibiotics …misuse of antibiotics or over usage of antibiotics and not giving proper courses of antibiotics, well, that has caused a resistance and now we are now called the country of superbugs. It is just because anybody here, right from a registered medical practitioner to a doctor, can prescribe antibiotic; even the patient can actually go and even the pharmacists can prescribe an antibiotic and that is what has led to us you know to a problem that we have you know multidrug resistance bacteria which are not sensitive to any antibiotic’. GI Surgical Resident, India

Abbreviations: CVTS, cardiovascular and thoracic surgery; GI, gastrointestinal; ICU, intensive care unit; IPC, infection prevention and control; MSW, medical social worker.

We observed a general lack of awareness of healthcare-associated infection (HCAI) risk among patients at site B. Patients and their carers preferred to remain in hospital as they felt they were safer there, where healthcare attention was closer than at home, despite reassurances from the surgical team that they were fit for discharge (X6; Table 1). The terms used by patients to describe infection in site A highlight how the perspectives of infection and illness differ between patients (X3, X7; Table 1). Patients referred to antibiotics as treatment for ‘viruses’ or ‘germs’ (X1, X3; Table 1).

HCWs considered the patient's socioeconomic status and/or level of education to be a factor in their understanding of antibiotic use. Surgeons at site B considered patients with higher educational qualifications to have a better understanding of antibiotic use and misuse (X8, X9; Table 1).

HCW communication with the patient

The content and level of communication and engagement varied across the specialties. At site A, communication patterns of surgeons with patients were individualistic and depended on the communication style of individual HCWs, whereas at site B, communication patterns were department- or specialty-specific. In site A, patient engagement depended on the approach of the individual senior surgeon leading the ward round and varied from a simple greeting to details inviting the patient to respond. In site B, there was consistency within specialties in the approach to patient engagement. In site B, the senior surgeons in the GI specialty took time during the rounds to speak with patients and/or their carers regarding the patient's progress and the next steps for treatment and hence the ward rounds took more time. The ward rounds led by CVTS senior surgeons were found to be much quicker, with less time spent interacting with patients and/or their carers. Across both departments, there were separate ward rounds—different from the early morning rounds—led by the surgical residents, wherein more detailed discussions were conducted with patients and/or their carers regarding any issues related to their care (care already received, care being received and care yet to be received). Across both sites, the most senior member present on the ward round was the one who engaged with the patient. This communication included discussions on their health status and plan of care (X10; Table 1). Whilst the name of the antibiotic was not mentioned in discussions with patients and/or carers, the indication for the prescribed antibiotics was generally discussed (X11; Table 1).

At site B, medical social workers (MSWs) who act as patient liaison provided psychosocial support to the patients and their families. During routine interactions with patients and carers, they also educated them on infection prevention and control (IPC) practices (X12; Table 1); however, education on ABR was not included.

Patients'positionality, both cultural and economic, may influence the extent of their involvement in decision-making

Limited patient roles

Some HCWs encouraged patients to get involved by actively engaging them in care discussions (X13; Table 1). The decision to treat and the choice of therapy for a given infection are considered to reside with the treating doctor, with patients generally not aware of opportunities that may exist for their involvement in decision-making (X13, X14, X15; Table 1). Even if the context allowed patients to contribute or be involved in care, they may not feel empowered to do so (X16, X17; Table 1).

A greater reliance on the voice and role of the carer (often the nominated head of family or adult child of the patient) in decision-making was observed in site B (X18, X19; Table 1), where hospital policy provides for a carer to stay with the patient. Patients' close family members played key roles in decision-making around patient care, from the need to seek medical help to which doctor to visit and the decision to have or not have a procedure, so much so that at times, patients were excluded from this decision-making process (X18, X19, X20; Table 1). Family members also felt to have the authority to question decisions taken by the healthcare team, stemming from the need to protect their family members (X18, X20; Table 1).

Carer involvement was more limited at site A. Healthcare teams presented updates on patient progress to each other, to the patient and to family members who may be visiting (X21; Table 1). However, no form of engagement by the patient carer or family in antibiotic discussions or in antibiotic decision-making was observed. Nurses sometimes provided updates (infection care updates that may include information on antibiotic use) at the family's request; however, they voiced their preference to have the treating doctors do so (X22; Table 1).

Patient's choice of healthcare provider

In the Indian healthcare systems where patients can select healthcare providers, they may have greater access to antibiotics by choosing doctors more likely to prescribe them (X23; Table 1). The patient's choice of healthcare provider is also influenced by their financial status, where patients who exhaust their ability to self-fund care are transferred from private to public hospitals (X24; X25; Table 1).

Patients at site A expressed gratitude for the quality of care received, given their initial perceptions of care in a public healthcare environment (X24; Table 1). This awareness of the prevailing work pressures experienced by their healthcare teams as well as their gratitude for care sometimes impeded their ability to voice observed shortcomings in the care that they were receiving (X26; Table 1).

Patient and/or carer demand for and access to antibiotics

Whilst prescribers may want to practice restraint in antibiotic prescribing, this is countered by a demand for antibiotics from patients and their families. This may be due to patients' own research on illness before seeking medical care (X27; Table 1). Prescribers in site B reported to have made efforts to educate patients and carers who demanded antibiotics (X28, X29; Table 1).

Fear of patient complaints and litigation can drive the decision to prescribe antibiotics, even though the prescriber may understand this to be irrational (X30; Table 1). The general perception, however, was that irrational prescribing is more prevalent in primary and secondary health centres than in tertiary care. Factors considered to influence inappropriate antibiotic prescribing included lack of good diagnostics and competition in community and private sectors for patients (X31; X32; Table 1).

Out-of-pocket expenses incurred to patients in healthcare systems that do not have universal health coverage act as an additional factor in suboptimal antibiotic prescribing. In site B, the high cost of antibiotics adds to the financial burden already placed on patients by surgery. As such, some patients may not be prescribed the most appropriate antibiotic. In some cases, where the most appropriate antibiotic is initiated, the course may not be finished due to cost constraints (X33; X34; Table 1), with implications for ABR. This was generally not the case at site A, where cost was not a factor, most likely because it was a public hospital where the financial cost of care incurred to patients was minimal.

The unregulated access to and consumption of antibiotics before presentation at the hospital were recognized by participants in site B as factors in the development of ABR (X35; X36; Table 1).

DISCUSSION

This study explored the inpatient's influence and participation in infection-related care in surgical specialties in India and South Africa. The cultural differences are manifested in how care is provided and the extent to which patients and carers are involved in decision-making. The patients' positionality, both economic and cultural, may influence their engagement with and expectations of care, including infection-related care and antibiotic use. Missed opportunities for effective patient engagement in AS and IPC remain in patient pathways. This is an important gap, given that infection is one of the unintended consequences of surgery,[43] especially in LMICs, where the burden of surgical infections is higher.[44] In health care, effective communication is key to positive patient outcomes.[41] This communication occurs at different levels with HCWs, patients, carers and the public. The communication needs of these different groups may vary. Patients bring their own beliefs, which need to be understood before they can be changed or influenced. While patient engagement could be affected by various clinical- and administration-related pressures, some senior surgeons, regardless of these, consistently engaged the patient in discussions related to their care, including infection care decisions.[41]

Our data show that patients do not necessarily associate hospitals with infection. We need to go beyond the assumption that patient, carer and public education alone will address the identified gaps in IPC and AS and that it will foster optimized practices.[42] The identified gaps can be addressed, perhaps by using those around the patient with additional specific roles in relation to patient education and engagement. In site B, MSWs, identified to be closely engaged in patient interactions, can provide pre- and postsurgical infection care advice and training to facilitate better patient participation in infection care.

Opportunities exist in targeting the involvement of HCWs in patient engagement, and participation in infection care and AS. HCWs themselves may also benefit from context-appropriate communication skills to effectively engage patients. Depending on the context, patient carers also need to be engaged in interventions. Opportunities for patient and HCW engagement can provide learning, for both healthcare providers and recipients, on the effective means and outcomes of such engagement.

Table 2 presents recommendations for optimizing inpatients' and their carers' involvement in antibiotic decision-making. Implementation of patient-centred interventions has demonstrated improvements in short-term knowledge of infections and antibiotic therapy among participants.[45] Patients have been known to influence antibiotic prescribing,[42,46] and co-design of AS- and ABR- focused interventions with patients may help influence attitudes and behaviours in relation to antibiotic consumption. Such efforts will also support evolving patient roles from passive recipients to active participants in care.

Table 2 Recommendations for optimizing patient involvement in antibiotic decision-making.

Recommendation	Description
Efforts to check patient needs and understanding of information provided	Patients bring lay knowledge and expertise to their own care, which needs to be understood and acknowledged. Patient education needs to go with checks for patient comprehension, to assist with healthcare worker understanding where further information needs to be provided.
Identifying champions for patient-centred antimicrobial stewardship communication	Doctors may spend limited time with patients on the ward and nurses may be limited by a low nurse to patient ratio. It would be beneficial to identify a champion for communicating with patients regarding their care. Medical Social Workers in India, who have a key role to play in patient communication, are an example. Opportunity may exist to utilize community health workers (CHWs) or home-based carers for such roles in South Africa. CHWs are already involved in medical specialties, especially in primary healthcare centres, where they assist with care communication and support of the patient in HIV and/or tuberculosis care.[55]
Improving education to patient and carers on healthcare-associated infections, antibiotic use and antibiotic resistance	Evidence-based educational materials should be prepared for patient and carer education. This education could be delivered through one-to-one counselling, leaflets or pamphlets or other suitable means. The materials should be concise, contextually appropriate and in a language devoid of medical jargon that the patient/carer can easily understand.
Identifying role for pharmacists and nurses in providing one-to-one education for patients	A one-to-one education/counselling session may be beneficial for patient education. This session should take place ideally at a time that is convenient for the patient and should also be flexible as it may need to be repeated over time as needed. There may be opportunity for expanding the role of pharmacists and nurses in patient education. Limitations in resources (funding and time constraints) pose challenges to this and will need to be addressed.

Individualistic or not, the decision-making in one's health is heavily influenced by the society and its perspectives on health.[47] In different cultures, positionality of individuals within the family and wider society can determine health-seeking behaviours and the individual's level of involvement in relation to his or her own care needs.[48,49] The role of close family members and/or carers in patient care differed across the two sites. In site B, carer engagement is supported, first, by hospital policy that mandates their presence and involvement in patient care. Second, there was a paternalistic practice of protecting the patient from what was considered sensitive information in relation to their own health, thereby excluding them from decisions about their own care, especially for patients with serious illnesses, while the carer took charge. Carers also play key roles in inpatient and post-discharge infection-related care.[42] This may be a wider reflection of the cultural norms in India, where the wider family support network is more depended upon in health care.

A key point to consider is that patients will have their own knowledge and lay beliefs, which may influence their behaviours and expectations of care received,[42,50] including expectations for being prescribed antibiotics. The prescribers' accounts and experiences confirm this, particularly their fear of patients choosing a different prescriber who might be more willing to comply to the patient's demand for antibiotic prescriptions. This creates competition among doctors and hospitals, especially in the case of private hospitals, and can act as a perverse incentive to prescribe antibiotics, as described in a previous study among formal and informal health providers in West Bengal, India.[51] In pluralistic health systems, where patients choose from a range of formal and informal healthcare providers, and the opportunity for self-medication is higher,[52] policy and regulation alone will not address the overuse of antibiotics. Such self-medication practices may have implications for ABR, for instance, where self-funding patients may be unable to afford full antibiotic courses. There needs to be sustainable engagement within civil society and the public as well as amongst HCWs on the threat of ABR, underpinned by an understanding of the context and specific patient/public behaviours that drive ABR, to bring about effective change in collective behaviours.[8]

In health care, current opportunities for meaningful and effective communication between patients and healthcare providers remain limited, particularly around IPC and AS.[15] Active engagement with and participation of patients in their care decision-making is critical, particularly in countries where lack of adequate universal healthcare means that patients use out-of-pocket expenses to fund healthcare needs. Inappropriate antibiotic use may also stem from their over-the-counter access, especially where antibiotic procurement cost is significantly less than that associated with a doctor's consultation or hospital admission. Literature on antibiotic self-medication practices in an urban population in Kerala showed that 3.31% of 775 adult participants reported antibiotic self-medication, of which 36% were procured using doctors' previous prescriptions, with convenience as the major reason for self-medication.[53] Other states in India with lower literacy rates report higher levels of over-the-counter antibiotic sales.[53] A study from the UK investigated the educational level of older patients and their access to healthcare, highlighting that patients with more education navigate the health system and access services more effectively.[54] HCWs in Site B also associated higher education levels with better understanding of antibiotic-related decision-making and use and vice versa.

Our study has limitations. It was conducted in a public hospital in South Africa and a private hospital in India. Both study sites are atypical hospitals at the forefront of implementing AS interventions and are not necessarily reflective of practices across other hospitals in each country. In addition, while the study focused on patient and carer involvement in infection care, HCW interactions were critical to providing insight and contextualization. This work presents important information on the extent of the surgical patient's involvement in antibiotic decision-making and identifies opportunities for a more patient-centred AS engagement. Longitudinal data collection also facilitated multiple observations, reducing the likelihood of the Hawthorne effect. In addition, the application of a data collection guide, multiple data sources and researcher reflexivity helped to minimize subjectivity in the data collection process and to validate the findings.

CONCLUSION

This study has provided new insight into surgical inpatients' involvement in infection-related care, including AS, across two diverse settings. To have a valuable role in AS and make informed decisions related to their care, a better understanding and channelling of the knowledge and experiences that patients and carers bring to their own care needs is crucial. The universal patient-centred approach to care, modelled through an individualistic lens, may not be responsive to the cultural determinants of health and ABR in settings like India, where a community of individuals connected to the patient has a voice in patient care with greater access to and demand for antibiotics. More effort is required to fully integrate and channel patient and carer experiences and outlooks in initiatives to address ABR, especially as it relates to the demand for and access to antibiotics.

AUTHOR CONTRIBUTIONS

This is a multicentre study involving multiple surgical departments at each site. All the authors represent either the research team or the surgical team. All the authors have critically reviewed the manuscript and have approved the final version to be published.

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CONFLICT OF INTEREST STATEMENT

The authors declare no conflicts of interest.

DATA AVAILABILITY STATEMENT

Data on which this publication is based are available via a secure server. Access to the data can be provided upon reasonable request. The data that support the findings of this study are available on request from the corresponding author. The data are not publicly available due to privacy or ethical restrictions.

ETHICS STATEMENT

Ethical approval was obtained from both the study sites (Ref: 499/2018 and IEC-AIMS-2018-INF.CONT-005A). Prior to study commencement, the surgical teams were sensitized to the study and their approval was sought and obtained. Buy-in from the surgical leads eased access to the teams. For observations, researchers obtained group consent from the specialty leads and verbal consent from participants. Posters were put up in wards to make participants aware of this study taking place in the ward. Written informed consent was obtained from participants before each interview. All participant-identifiable data were anonymized before data analysis.

DETAILS

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Nothing about us without us: A co-production strategy for communities, researchers and stakeholders to identify ways of improving health and reducing inequalities

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ABSTRACT (ENGLISH)

Introduction

Co-production with communities is increasingly seen as best practice that can improve the quality, relevance and effectiveness of research and service delivery. Despite this promising position, there remains uncertainty around definitions of co-production and how to operationalize it. The current paper describes the development of a co-production strategy to guide the work of the ActEarly multistakeholder preventative research programme to improve children's health in Bradford and Tower Hamlets, UK.

Methods

The strategy used Appreciative Inquiry (AI), an approach following a five-step iterative process: to *define* (Step 1) scope and guide progress; to *discover* (Step 2) key issues through seven focus groups ($N=36$) and eight in-depth interviews with key stakeholders representing community groups, and the voluntary and statutory sectors; to *dream* (Step 3) best practice through two workshops with AI participants to review findings; to *design* (Step 4) a co-production strategy building on AI findings and to *deliver* (Step 5) the practical guidance in the strategy.

Results

Nine principles for how to do co-production well were identified: power should be shared; embrace a wide range of perspectives and skills; respect and value the lived experience; benefits should be for all involved parties; go to communities and do not expect them to come to you; work flexibly; avoid jargon and ensure availability of the right information; relationships should be built for the long-term; co-production activities should be adequately resourced. These principles were based on three underlying values of equality, reciprocity and agency.

Conclusion

The empirical insights of the paper highlight the crucial importance of adequate resources and infrastructure to deliver effective co-production. This documentation of one approach to operationalizing co-production serves to avert any misappropriations of the term ‘co-production’ by listening to service users, stakeholders and other relevant groups, to develop trust and long-term relationships, and build on the learning that already exists amongst such groups.

Patient or Public Contribution

The work was overseen by a steering group (*N* = 17) of individuals, both professional and members of the public with experience in undertaking co-production, and/or with some knowledge of the context of the two ActEarly field sites, who provided regular oversight and feedback on the AI process.

FULL TEXT

INTRODUCTION

This paper reports on the process of co-producing a co-production strategy for a large UK consortium of researchers, stakeholders and communities called ‘ActEarly’, which aims to improve the health opportunities of families living in deprived areas. The importance of working in partnership with communities to address issues which impact their health and happiness is well recognized.[1] Reviews have found that health interventions which engaged communities in their development or delivery have a positive impact on health outcomes.[2] However, effective partnership working with communities can be challenging, and there can be adverse impacts if opinions are sought, but little change is demonstrated. Co-production is a ‘complex social phenomenon’, and the relationships between processes and outcomes can be ambiguous; outcomes may include ‘soft’ variables that are ‘hard’ to measure in practice, such as improved trust, shared responsibility, levels of influence and ownership over projects.[3] Evidence about ‘what works’ or ‘how’ to do co-production is limited. Smith et al's.[4] scoping review of co-production practice and future research priorities in UK-funded applied health research urges researchers to be clearer in reporting ways in which they are operationalizing co-production by providing a set of values and operating principles through which co-production could be implemented.

Context of ActEarly

The ActEarly UK Preventative Research Programme aims to promote health and well-being in early life in two multicultural areas of the United Kingdom with high rates of child poverty, Bradford in West Yorkshire, and the Borough of Tower Hamlets in East London.[5] Living in an area with high levels of child poverty often coincides with exposure to other economic, physical, cultural, learning, social and service environmental risk factors, which can predispose children and their families to poorer mental and physical health outcomes. Co-production is at the heart of ActEarly, launched in 2019, which uses a ‘City Collaboratory’ model to unite communities, researchers and stakeholders, including local governments, the NHS and the third sector to identify, co-produce and implement system-wide early life upstream prevention interventions.[5]

The tricky problem of defining co-production

What unites nearly all researchers and practitioners involved in co-production is a recognition of the difficulty in defining it. It is noted as a slippery,[6] woolly[7] and muddled[8] concept, the benefits of which may be diminished if the definition is unclear or misapplied. Co-production and co-design are conceptualized in a wide range of ways, and the elasticity of the term has been referred to as both its strength and its limitation.[3] Co-research has been used as an umbrella term to encompass a family of approaches, such as ‘participatory’, ‘emancipatory’ and ‘inclusive’ research, which reflect a turn towards involving communities in the process of knowledge production.[9] Co-research aims to put principles of empowerment into practice, by offering participants greater control over the research process while providing opportunities to learn and reflect upon their experience.[10] Co-production builds on this, focusing on the delivery of more responsive, personalized public services in an equal and reciprocal relationship between professionals, people using services, their families and their neighbours.[11–15] Where activities are co-produced in this way, there is great potential for both services and neighbourhoods to become far more effective agents of change. These principles are increasingly applied to the production of knowledge, and co-production is now a mainstream term in health research.[16] Similarly, the UK National Institute for Health and Care Research (NIHR) see co-production as an approach in which 'researchers, practitioners and members of the public work together, sharing power and responsibility from the start to the end of the project, including the generation of knowledge'.[17,p.1]

Key considerations for effective co-production include the recognition that it is context-dependent and that it requires building trust and creating opportunities for genuine power sharing and respect amongst all partners. The approach to co-production used as a starting point by the ActEarly consortium builds on others' definitions and sees it as a collaborative process involving researchers, practitioners, decision-makers and the public working together, sharing power and responsibility.[5]

Rather than seeking universal definitions of terms such as co-production, Masterson et al.[18] recommend that future applied research should focus on articulating the underlying principles and values that need to be translated and explored in practice. For this reason, this paper sets out the principles and values underpinning co-production to inform ActEarly's work undertaken with its communities.

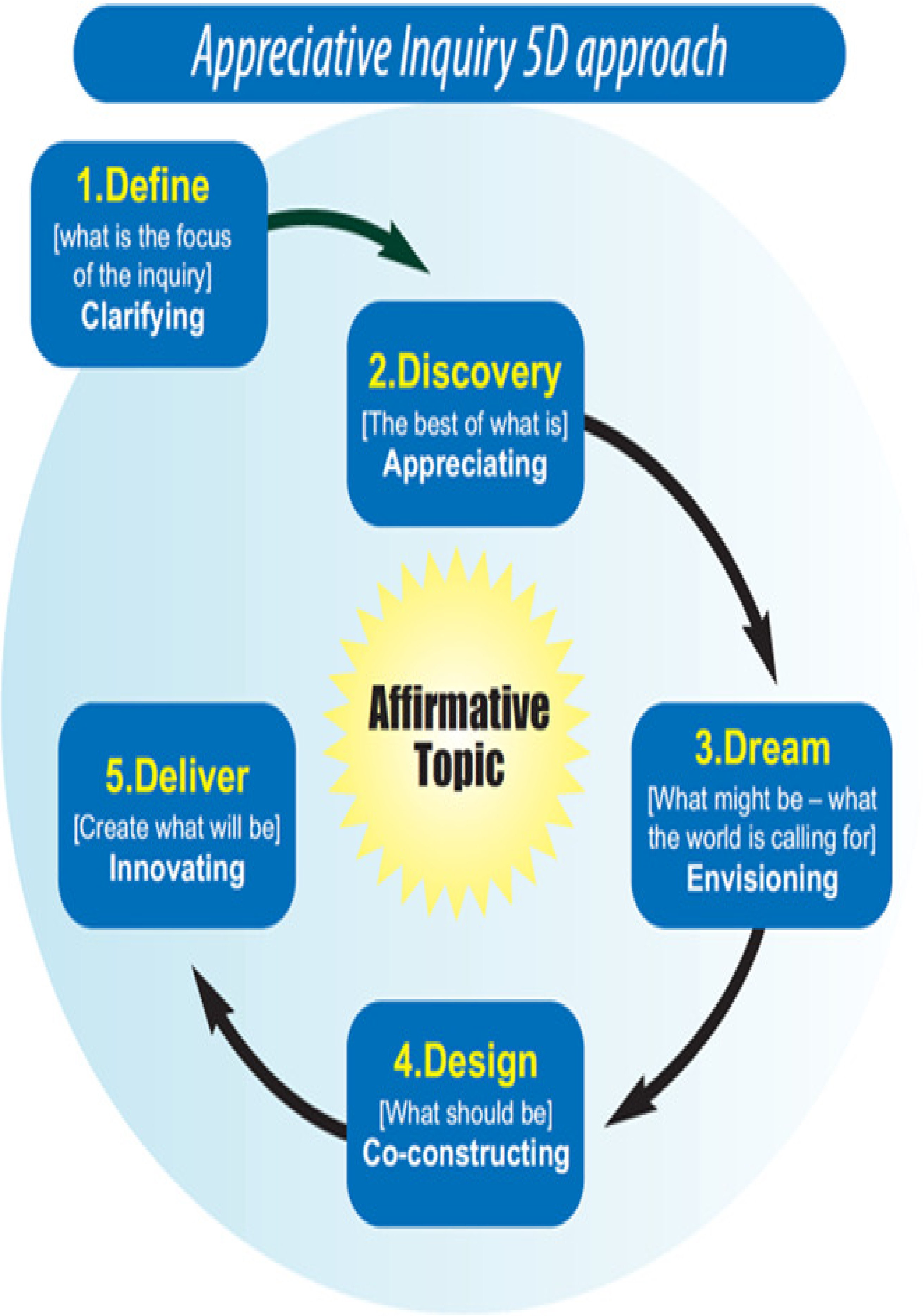
Aims and objectives: Co-producing a co-production strategy

The aim of this study was to develop a co-production strategy to inform the work undertaken with these communities within and beyond the ActEarly programme. Working with communities and stakeholders in Bradford and Tower Hamlets, our objectives were to use an asset-based Appreciative Inquiry (AI) approach to co-produce a set of guiding principles and core values which would form the basis of the co-production strategy.

An assets-based approach was used, which focussed on the positives of what has worked in these areas and by concentrating on existing assets. This was achieved by going to local organizations and community groups in both ActEarly locations and allowing them to guide what should be included in the strategy; the aim being to produce an end product—the strategy report itself—that carries the values embedded within it.

METHODS

The ActEarly Co-production strategy was designed through the application of AI. A fundamental principle of AI is its focus on assets and strengths within communities rather than focusing on deficits and problems.[19] The basic assumption is that in every human system, something works right and contains certain elements that make it vital, effective and successful.[20] AI helps people to focus on what is working well, the positive core and identifying strengths by engaging them in inquiries and stories that highlight and then leverage those strengths.[19] The deliberately affirmative assumptions of AI about people, organizations and relationships are in stark contrast to more traditional forms of research that seek to analyse or diagnose problems. The AI took the 5D approach (so-called because it is based on five elements that start with D), which is depicted in Figure 1 below and with each domain described below.



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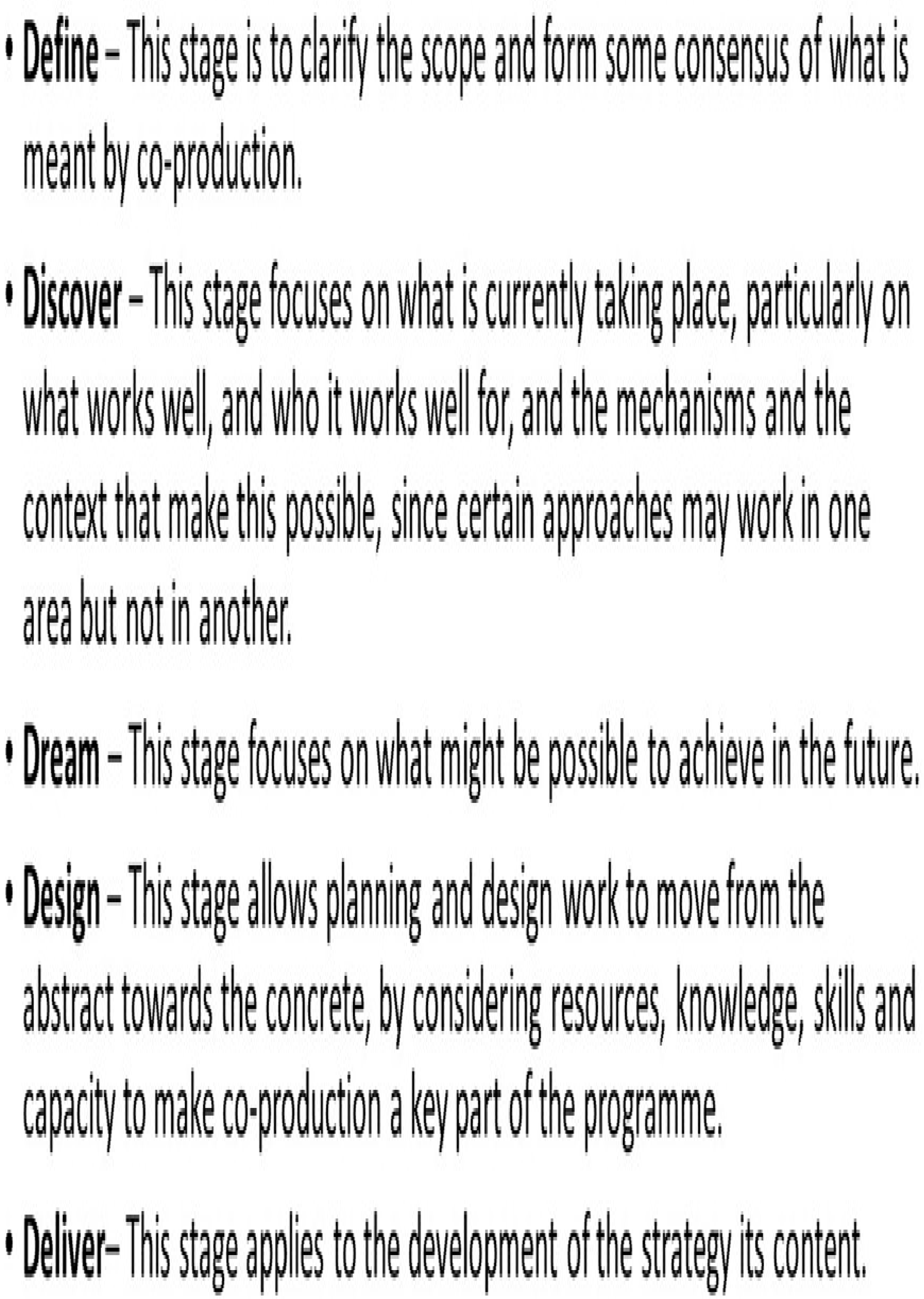
Step 1: ***Define***

The research process was overseen by a steering group, made up of 17 individuals (professionals and members of the public) with experience in undertaking co-production, and/or with some knowledge of the context of the two ActEarly field sites. The steering group provided oversight and feedback on the AI process, and, where necessary, suggested adjustments to the methods proposed. They helped to identify participants adequately reflecting the diversity of the populations across both ActEarly sites.

Step 2: ***Discover***

Seven focus groups ($N = 36$) and eight in-depth interviews with key stakeholders were undertaken from March to November 2021 across the two ActEarly field sites. The majority of data collection was online, with the exception of two focus groups, and one interview, which was held in person. Participants included stakeholders representing community groups, residents' associations and people employed in community engagement work across the voluntary sector and the statutory sector. A long list of potential participants in each location was identified, most of whom were already known to the research team and the wider work of ActEarly, such as Public and Patient Involvement groups associated with the Bradford Institute for Health Research. We also included groups and networks that may have had limited opportunity to connect with ActEarly. The groups, and individual stakeholders, were approached by the research team via telephone or email and a request was made to organize a session at an appropriate time for the research team to deliver an online workshop or interview. A snowball sampling approach was used, and at the end of each interview or focus group, participants were asked if there were other key stakeholders that should be included in the research. This approach was continued until a saturation point was achieved with the comments and themes being raised in the interviews and focus groups. The topic guides for these discussions were designed around the 5Ds model in Figures 1 and 2. The transcripts and field notes from the focus groups were systematically reviewed by two members of the research team and grouped according to the emergent themes, ideas and concepts. Thematic analysis (TA) was used to identify the preliminary results from this stage of the AI process.



- **Define –** This stage is to clarify the scope and form some consensus of what is meant by co-production.
- **Discover –** This stage focuses on what is currently taking place, particularly on what works well, and who it works well for, and the mechanisms and the context that make this possible, since certain approaches may work in one area but not in another.
- **Dream –** This stage focuses on what might be possible to achieve in the future.
- **Design –** This stage allows planning and design work to move from the abstract towards the concrete, by considering resources, knowledge, skills and capacity to make co-production a key part of the programme.
- **Deliver–** This stage applies to the development of the strategy its content.

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Step 3: ***Dream***

The preliminary results of the AI process were then presented at two workshops in November with the patient and public contributors and researchers. Thirty-five people attended the 2 hour in-person workshop in Bradford, and 17 people attended the 2 hour online workshop in London. These workshops provided an opportunity to present back to participants the preliminary findings from the AI process and to dream about best practices. The team opened up a discussion with participants about the importance of co-production to ActEarly, who was engaged during the AI process and preliminary findings. Participants were then asked to reflect on what the team should avoid, what they should eliminate and what they should enhance, or do more of. Following breakout group discussions on these topics, participants were then asked to collaboratively develop one idea that the team could take forward. Discussions at these two workshops informed the next steps of strategy development. Field notes were taken by facilitators attending the workshops, which were supplemented by content recorded in Google Jamboards for the online workshop.

Step 4: *Design and analyse*

The co-production strategy was designed using data collected from focus groups, interviews and workshops. Conversations were recorded and the transcripts and field notes were systematically reviewed by the research team and grouped according to the emergent themes, ideas and concepts. Two members of the research team analysed the data using TA, a qualitative data analysis technique used 'for identifying, analysing and interpreting patterned meanings or themes in qualitative data'.[21,p.79] Through systematic and transparent coding of the key themes and methodical and honest reporting of the findings, the researchers sought to try to minimize bias. The findings were checked against some of the wider contextual issues and preliminary themes coming out of the focus groups in some of the more in-depth interviews with practitioners. These components were then re-evaluated, regrouped as necessary and gradually refined and linked to other conceptual categories. To ensure rigour, the key themes and components coming out of the analysis were initially analysed by the two researchers separately and then brought together to compare and refine. The researchers attempted to maintain a high level of thoughtful and deliberate planning throughout the AI process, and a diligent and ongoing application of researcher reflexivity, as well as honest communication between the researchers, and the participants in the AI process through the Step 3 workshops. The preliminary findings were also discussed with the steering group (N = 17) made up of both professionals and members of the public with experience in undertaking co-production, and/or with some knowledge of the context of the two ActEarly field sites. The steering group provided regular oversight and feedback on the AI process and helped to reduce and draw out any researcher influence on the results.

Step 5: *Deliver*

A strategy and practical guidance were developed and disseminated to the ActEarly consortium and wider stakeholders.

RESULTS

The findings from the AI process identified nine main themes that characterize effective co-production (referred to hereafter as principles of co-production) and three core values. The following sections set out the key themes from the analysis of the data from the focus groups and interviews, the dream and design workshops and other aspects of the AI research. Direct quotations are presented as being from a focus group (FG) or interview (INT), and then whether the stakeholder is in Tower Hamlets (TH) or in Bradford (B).

Principles of co-production

The AI process generated nine guiding principles on how to deliver successful co-production which are depicted in Figure 3, and then further described in more detail below.

Nine guiding principles for co-production

Principle 1:	Power should be shared amongst all partners
Principle 2:	Embrace a wide range of perspectives and skills
Principle 3:	Respect and value 'lived experience' and how different forms of knowledge can be expressed
Principle 4:	Ensure that there are benefits to all parties involved in co-production activities
Principle 5:	Go to communities. Do not expect people to come to you
Principle 6:	Work flexibly
Principle 7 :	Avoid jargon and ensure communities have access to the right information at the right time
Principle 8:	Relationships with communities should be built for the long term
Principle 9:	Make sure co-production initiatives are adequately resourced

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Principle 1: Power should be shared amongst all partners

Successful co-production requires a recognition of the imbalanced starting positions faced by many of the stakeholders in co-production. There needs to be an intentional effort from those who hold power to show that they are willing to share it and a focus on building towards that sense of equality.

It needs a lot of groundwork to prepare organisations to share their power—people who currently hold the power need to be committed to co-production process. (FG3, TH)

With young people we are always aware that …we have more power because we're older but we have to deliberately make a choice to …really listen to what they're saying because it's so easy to, because we're all older, lived longer, to dismiss some of the things that they say. (FG10, B)

Principle 2: Embrace a wide range of different perspectives and skills to ensure these are represented in the project

Nearly all participants in the AI process discussed the importance of ensuring that communities feel properly heard and listened to, which is linked to the theme of building relationships and developing trust (Principle 8).

Get up in to the communities and real groups to better understand the truth, and properly listen to people and what they're saying about what the issues are. (FG8, B)

Everything that's gone well so far has been because we've listened to the mothers, we've understood the mothers, and we've taken forth their ideas, therefore they've felt as if that they have been involved, they've been listened to, they are instrumental in making change. (INT16, B)

Many participants mentioned the importance of partnership working, whether that entails larger, more established and better-resourced organizations working with smaller ones, or just different stakeholders and partners working together to affect change. The success of partnership working seems to come from the credibility and trust that established relationships can bring, as well as the sharing of resources, and also the focus and balance that such partnerships can bring.

Better collaboration between participants and stakeholders—without it, the work loses focus and balance. From a participant's point of view, they don't care about difficulties communicating between. (FG3, TH)

Working with local community organizations as gatekeepers can help to ensure access to a diverse range of community views, but some participants mentioned the need to be more ambitious and move beyond working solely with a small group of gatekeepers to involving a wider audience. A particular suggestion involved working with designated community champions or community partners to deliver successful co-production:

It really helps to have a lot of champions in the community—they have unbounded energy, resilience and capacity. Recognising those people and bringing them together has worked well. …In some communities, projects need to be endorsed by the right community members to really work. (INT15, TH)

The key is to have localised grassroots partners and to make sure the focus and needs identified are based in the community and tap into local expertise, not a parachuting-in viewpoint. Having a grassroots bottom-up perspective is key in anything of this sort. (INT14, B)

Principle 3: Respect and value of lived experience and how different forms of knowledge can be expressed and transmitted

Participants drew attention to the importance of respecting and valuing the 'lived experience' held by different people, and how different forms of knowledge can be expressed and shared.

People can connect if they see different people involved and committed to the work. Different sectors are represented, and you feel that your voice is being heard and your experiences are valued. (FG3, B)

The ways in which knowledge is valued reflects how it is produced, meaning that in many cases, members of the public have been limited to the role of participants in research studies, where data are collected about them, without their influence in the design, data collection or eventual output. However, approaches such as citizen science and co-production aim to change this. An active and well-engaged Patient and Public Involvement group in Bradford explained how they support researchers by helping them consider practical and ethical issues from a community perspective. Whilst such inclusion should be celebrated, very few examples came forth to demonstrate the more advanced forms of inclusion whereby community members produce knowledge and conduct research with professionals as a partnership. Several contributors made a case for better inclusion of different forms of knowledge and lived experience to feed into research systems, and how damaging it can be if this is not the case.

I think the worst thing from the cohort …that we work with is to hear them and not listen to them and not follow through with something they're doing, because that's how you cause that disengagement again …because they're going to think that no-one takes their ideas or experiences into account. (INT12, B)

The benefits of drawing on a plurality of sources and types of knowledge include improved validity and better engagement from target communities. This is possible because local knowledge is the mundane, yet expert understanding of, and practical reasoning about, local conditions derived from lived experience. When the lived experience is pressed into service, using the right approaches, it has the potential to augment research and implementation efforts.

Principle 4: Ensure that there are benefits to all parties involved in co-production activities

Ensuring that communities receive equal benefits was mentioned often in discussions with stakeholders throughout the AI process. Too often, those leading a co-production project make things work for their own purposes (e.g. to complete a Public Patient Involvement exercise or to complete the development of a funding bid), and those being engaged do not receive anything in return. This can erode trust, meaning that participating groups are used as a resource in the materialistic sense and not as an asset to help agencies achieve improvements. One way to ensure participating groups benefit in their contribution to a project is to provide visible feedback, with clear pathways to feed into policy. This can be achieved through, a clear and consistent feedback loop that articulates what people expressed during the project, the project team responses and actions, in addition to a commitment for partners to take forward the outputs of projects.

If there is no feedback, people will question the point and not want to be engaged in the future. There has to be a feedback loop explaining to people, for example, why change didn't happen. (INT6, TH)

Representation of different communities is needed since this will give confidence to communities that they are being taken seriously …If they don't see things change then they can't be bothered to contribute. Change breeds confidence. (FG2, B)

Principle 5: Go to communities. Do not expect communities to come to you

Participants also mentioned the importance of going to where people and communities are, rather than expecting them to come to researchers. Equally, talking to people in their own environments informally was seen as a good way to connect. Some participants framed successful co-production as being about keeping motivated and connected to the direction those participating in the project want to go in and being mindful of giving support when needed and stepping back where space is needed for growth. Such requirements necessarily require a flexible disposition and successful co-production is an ongoing dialogue with communities and not a one-off event.

Practice through research …it's about coming along to things already set up to not duplicate or re-invent the wheel. It's about coming out to groups—it's better to meet in their environments—so services meeting communities halfway—currently it's a very top-down approach with little co-production with communities …We need to change the way we work, be more practical and do things differently. (FG11, B)

Principle 6: Work flexibly

Working flexibly, including adapting activities to be able to work both face-to-face and online as appropriate, and talking to people in their own environments in informal ways was seen as crucial to successful co-production. For this to work well, it is necessary to pay attention to the varying timescales of different sectors and organizations, as well as the importance of involving communities from the beginning rather than including some co-productive element to a project as an add-on further down the line. Alignment of competing timescales can be challenging due to different priorities and foci and can be further intensified by political cycles, which can force timescales in different directions. Working flexibly also includes learning through trial and error, and being open to adapting and changing approaches and methods used in a project:

Sometimes people can make things too complicated as they're sticking to a formular rather than adapting to context …with co-production you have to change as you go along, and you have to be open to change your methodology. (INT6, TH)

Principle 7: Avoid jargon and ensure communities have access to the right information at the right time

All those involved in a project need to have access to information in the appropriate cultural context for them, such as the appropriate language, and to be clear about what is being aimed for in a project. Using the appropriate terminology and language is crucial to the feedback process, to ensure participants understand how a project has evolved and if or how changes have been adopted. Reflecting on the language used in co-production—particularly where multiple professions and stakeholder groups are involved—cannot be underestimated, since each profession or group will have their own terminologies and ways of framing or talking about particular issues, based on their own experiences and other contextual factors.

We need to use appropriate language for different stakeholders and service users to explain what we're doing. (INT15, TH)

Professionals use language that can exclude people—like acronyms. People feel out of place if they ask things to be explained to them. The language of co-production involves an understanding that participation for residents or communities is harder as people don't feel confident. It's important to avoid using terminology or acronyms that might make this worse. (FG2, B)

Avoiding jargon and using more collaborative terms, such as aims, values and ethos, can help to resolve potential conflict due to the lack of shared vision. However, there needs to be as much clarity as possible from the outset of a project around what is being aimed for, to avoid a ‘drift’ in direction, and to keep things moving towards the desired goals of all involved. Participants warned of overpromising on outcomes and a lack of clarity about what is on the table from the outset.

Principle 8: Relationships with communities should be built for the long term and not for the short term

Participants noted the importance of the level of trust communities have in both the process and the people delivering activities. This was seen as a deal-breaker as people have seen many ideas come and go and noticed who makes a difference and which services are not actively making a difference. The act of doing things together builds on this level of trust and creates an opportunity for services or research projects to return in the future to capitalize on this trust:

Community events and group activities work well because they build relationships, they continue to build relationships—so there's an element of relationship building within the activity that you're doing. (FG10, B)

Building individual relationships even when convening groups, or largely doing group work is really important. It's about knowing where people are coming from, how best to support them. The human approach and connection with people is very important. (FG3, TH)

Participants mentioned that communities feel disengaged and frustrated when research teams ‘parachute’ in to work with them on short-term projects, and then leave without actioning community priorities, but funders often work on short-term funding cycles, and this mismatch in timeframes produces its own challenges. Participants highlighted how co-production works best when it is focused on tangible action and when people can see how their contributions have been followed up.

Co-production works best around tangible action—people enjoy contributing to things they are seeing develop in front of their eyes. Some things have different timescales. (FG3, TH)

Our strength has always come from the fact that if they've said something as random as it may sound, we follow through with it even if it costs money, but that's how we've gained the trust of them because we follow through. (INT16, B)

Principle 9: Make sure co-production initiatives are adequately resourced

The topic of adequate time and resources to do co-production successfully was frequently discussed. Building strong links with the voluntary and community sector is important, but this takes time and funding for all parties involved. Participants raised issues with funding specifically for co-production, suggesting that it is challenging to appropriately allocate time and resources between co-production and other mechanisms for engagement and priorities. They also mentioned the issue of capacity, and that it takes time for everyone to be in the same place, which is exacerbated by the way research funding works—with short timelines, insufficient budgets, and there is always an added extra that gets cut back.

To make a meaningful difference …[co-production] has to be built into commissioning and grant making, so that people are resourced to be able to work in that way. (INT12, B)

You can often fall into trap of chasing funding based on perceived need, rather than working with community to identify what's really needed. (INT14, B)

A competitive environment for the voluntary sector in terms of seeking funding can lead to a resistance to sharing data and collaborating which poses a challenge to notions of partnership working and reciprocity raised above. There is also a very real danger of not recognizing the labour that goes into doing co-production which means participants are not properly rewarded for their time and intensive work.

Dream workshops

The discussions during each workshop were captured by a graphic illustrator, and the outputs are shown in Figures 4 and 5. below. The outputs from these events were used to assist the research team in formulating the actions to be taken as a result of the AI process.

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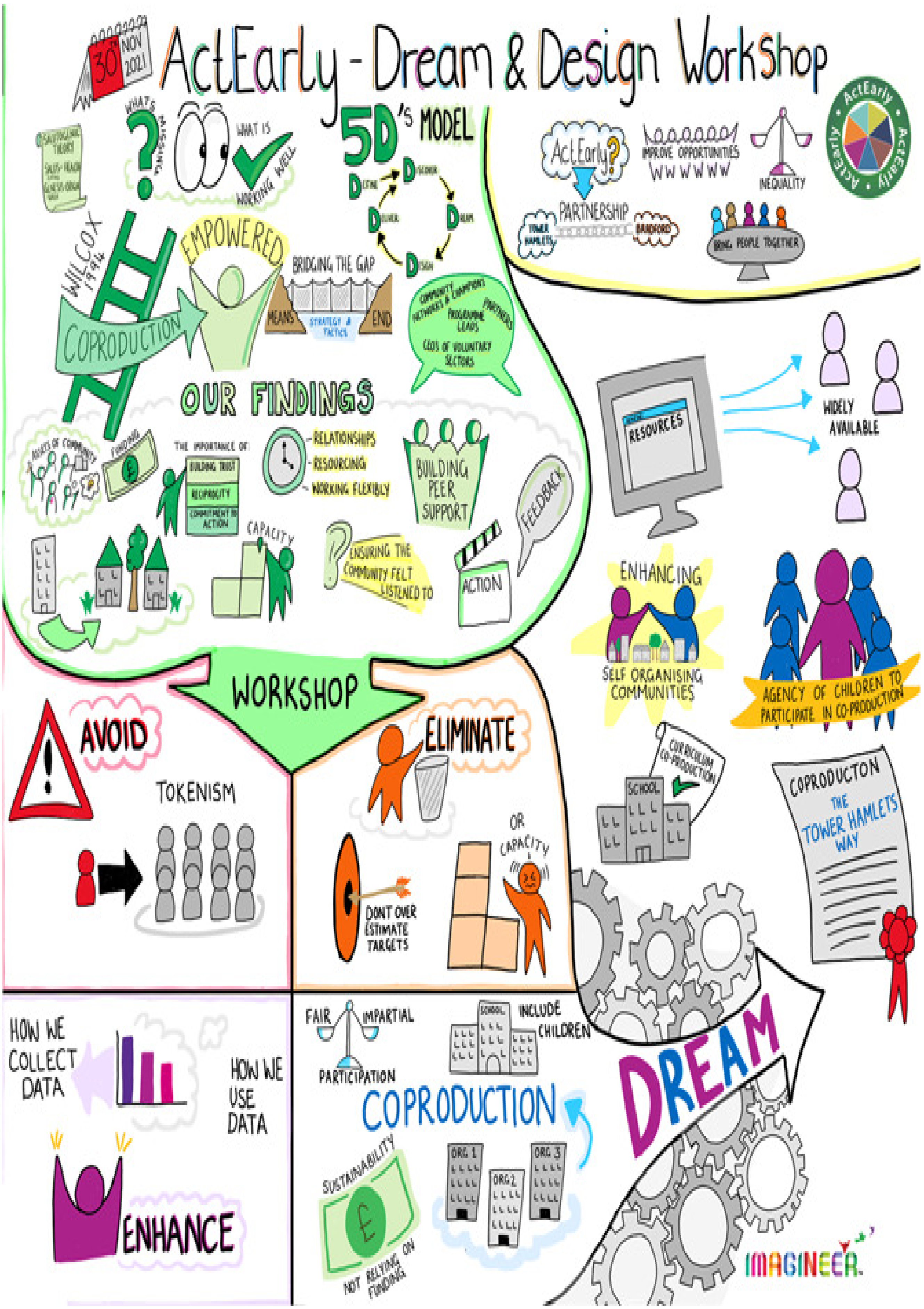

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In the Bradford workshop, participants recognized the politics and changing power dynamics at play between different actors, stakeholders and organizations. They highlighted the importance of building long-term commitments and building on existing knowledge and expertise that already exists in communities, whilst asking for political decision-making to be representative of the communities being served. They discussed the possibility of developing a community-led co-production hub as a shared learning environment, a place for all ages and communities to come together, and a space for agencies to be ‘guests’ in a community space. Participants also discussed the importance of investing in diverse community champions to coordinate a bottom-up approach to responsibly using resources effectively and to procure for social value, whilst also recognizing the limitations of what is possible.

In the Tower Hamlets workshop, discussions focussed on ensuring and developing co-production capacity within organizations that is sustainable and for the long term; identifying opportunities and repurposing existing structures and mechanisms to promote co-production and training professionals to see co-production as an integral part of day-to-day activities. The discussion also explored the potential to include children as co-producers, and the development of a recognized approach to co-production in Tower Hamlets, or an awards scheme and celebration of successful co-production activities.

Core values underpinning co-production

Through reflection and dialogue with all stakeholders in various forms throughout the AI process, three core values emerged that underpin the principles stated above that make co-production distinct from other forms of inclusive approaches. These values demonstrate what co-production is comprised of.

Equality

All those participating in co-production must feel they are equal contributors to the process of design and delivery, and this requires inclusion from the very start of a project, all the way through to evaluation. Equality does not mean treating everyone the same but necessarily focuses on treating participants differently by respecting and accommodating their difference.

Agency

Respecting the goals and values of community members may well be (and probably will be) different to the organizations that have commissioned a project. Finding ways to respect this and negotiating and accommodating this difference is a crucial underlying value of successful co-production.

Reciprocity

This refers to the mutually beneficial exchange of knowledge and resources in a context of partnership, where all parties have something to gain. This will be different for each party, but this must be expected and respected.

Co-producing the co-production strategy

Together the nine principles and three core values form the underpinnings of the co-production strategy to guide the ActEarly consortium. Together with the steering group overseeing the process, a set of specific actions or practical suggestions was developed to take forward the principles and values of the co-production strategy (see Supporting Information: File 1).

DISCUSSION

This paper reports on the process of co-producing a co-production strategy for a consortium of researchers, practitioners, stakeholders and communities who aim to implement system-based approaches to improving children's health in two locations in England. The co-production strategy also includes a list of practical suggestions for researchers and practitioners wishing to implement the values and principles of the co-production strategy, recognizing that some principles will be easier to enact than others. The principles developed through the AI process are consistent with previous work to capture the foundational principles of co-production. For example, Harrison et al's.[22] narrative reviews identify, quantify and summarize the conceptual foundational principles of patient stakeholder engagement in research and best practice activities and found that the most commonly reported foundational principles were ‘respect’ and ‘equitable power between all team members’. For co-production to flourish, attention needs to be turned to appropriate, long-term resourcing of community assets (e.g., community centres, schools and faith settings) that can act as ‘gate-keepers’ to communities, and also for the researchers and services who need to work with communities.[23] Smith et al.[4] draw attention to the fact that researchers operationalize co-production in various ways, often without the necessary financial and organizational support required and the right conditions for success. Projects ‘parachuting’ in to work on specific topics and then leaving after that project ended, or those not appearing to fulfil community needs, were seen as very damaging to trust.[24] It would seem prudent to develop appropriate community infrastructure at a ‘place’ level, providing hubs that can connect communities with researchers and stakeholders and provide opportunities for longer-term dialogue. Central to this longer-term dialogue is allowing communities to be the driving force in the identification of their own priorities, and that funders should tailor their commissioning cycles to suit. Experience of priority setting in Bradford,[25] under the umbrella of the ActEarly programme has found that communities identify a range of issues important to children's health and happiness and that with commitment and joint working, these can be translated into shared research and service agendas. This is also supported by previous work to capture the core principles in co-design and research, which have found consistent results.[22,26] Following the development of our co-production strategy, the ActEarly research team is developing specific actions to progress opportunities for dialogue with communities. These include a commitment to organize feedback sessions in Bradford and in Tower Hamlets, through regular open space meetings in each location, as an opportunity to check in, to share information about what projects are taking place, and also to share knowledge about what has worked, and areas where things have gone less well. Such meetings could also serve as an opportunity to feed into an evaluation of co-production—to review whether the process worked in the way it was intended, and if not, why not. These sessions would be dedicated spaces for reflecting on what was produced, what worked and what did not—a factor that is particularly important to ensure participants do not feel that it is their fault if a process or activity has not worked out. This is consistent with Witteman et al.'s[26] offering pragmatic, actionable lessons for developing effective research partnerships between different stakeholders such as patients, caregivers, clinicians and researchers.

Another outcome of the AI process is the need to better understand when co-production has worked well, and what are the criteria for its success. Furthermore, appropriate attention and consideration need to be given to monitoring, data capture and evaluation, and to reflect on how routine data is gathered, how well it is gathered and how any change affected is different as a result of having used co-production.[3,13] Participants warned against too many metrics since communities work at their own pace, and there is a need to reduce pressure on monitoring reports with too many targets and outcomes set by external partners. Concomitant with this, there is a need to focus on long-term and sustainable outcomes that build on trusted relationships and continue long after a project or piece of research has finished.

Strengths and limitations

A strength of the approach used to co-produce a co-production strategy was the use of an asset-based AI approach to include a wide range of stakeholders and communities in its development.[2] This process, in and of itself, strengthened relationships between stakeholders and has built on existing community knowledge and assets, enabling the research team to base the strategy on enhancing existing approaches, rather than re-inventing the wheel. AI practices change the character of interpersonal interactions, including changing perspectives, focusing upon and learning from past successes, and forming relationships and a common vision.[19,27] Furthermore, the use of the AI process meant that the co-production strategy developed is grounded in research conducted at a local level and represents an articulation of how the stakeholders and local groups in the two ActEarly sites understand themselves and their work.

The key audience for this work is a multidisciplinary group of researchers, theme leads, policy makers and other interested parties that make up the ActEarly consortium. Perhaps the most challenging aspect of the strategy was to try to reconcile the differing perspectives of the consortium stakeholders with what the community groups in each location articulated to the research team. Further to this, in some instances, there were some differences of opinion that could not be reconciled. This, however, underlines the importance of bringing together diverse groups and stakeholders so that they might work together to develop projects and services and approaches that are acceptable, and feasible. The strategy was developed in two places with similar, high levels of deprivation and ill health. The parallels of opinions across the two sites were striking, however, it may be that other ‘places’ have different opinions and priorities. An outcome of this strategy development work is to encourage all those undertaking co-productive activities to engage with their communities at the start to set the principles and underlying values of the process, perhaps using the strategy developed here as a starting point. For example, Fleming and Rhodes[28] discuss the challenges of embedding lived experience in research, and the NIHR[17] guidance on co-production highlights the assumption implied in co-production that those with lived experience are often best placed to design and deliver research. The challenge lies in actually acknowledging this lived experience and embedding it in the research process.

CONCLUSION

Effective co-production necessitates a level of commitment, proper resources, openness to work flexibly and to listen and reflect on shared dialogues and priorities. It requires appropriate resourcing and infrastructure, and a long-term vision. Whilst co-production presents many opportunities, it is crucial to recognize that there is not a 'one size fits all' approach, and there is a critical need to accept the diversity in approaches to co-production, and to better tailor these approaches to context, different stakeholder groups and the different stages of the research and implementation process. This requires extensive reflection on the use of chosen approaches in practice and a more systematic reporting process on the learning that ensues. This includes where processes did not work out, failures and areas for improvement. Many might find putting the principles and values outlined here into practice daunting if they do not consider themselves to be 'skilled' at co-production. The aim of this paper is to invite others to read the ActEarly co-production strategy and find both explanations and practical guidance on how to put co-production into practice. By providing a set of principles, along with practical recommendations, others can find ways to reflect on their current practice and explore new ways of working and find ways of improving co-production activities they are involved with. The paper constitutes an invitation to others to build on the methods and findings presented here, with the overall aim of allowing for the better operationalization of co-production principles and to guard against the tokenistic use of, and potential hollowing out of, the term 'co-production'.

ACKNOWLEDGEMENTS

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CONFLICT OF INTEREST

The authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

The data collected as part of this research that support the main findings of this study are not available for reuse. Data are available on request due to privacy/ethical restrictions.

ETHICS STATEMENT

The project was reviewed and received ethical approval from the University College London Research Ethics Committee ID 19659/001.

DETAILS

Subject:	Health disparities; Principles; Long term; Prevention programs; Community organizations; Workshops; Values; Childrens health; Reciprocity; Listening; Researchers; Best practice; Interviews; Infrastructure; Stakeholders; Uncertainty; Learning; Child poverty; Equality; Power; Focus groups; Medical research; Consortia; Appreciative inquiry
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Location:	United Kingdom--UK
Identifier / keyword:	Appreciative Inquiry; co-production; ethnicity; health inequalities; public health; strategy development
Publication title:	Health Expectations; Oxford
Volume:	26
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The use of arts-based methodologies and methods with young people with complex psychosocial needs: A systematic narrative review

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ProQuest document link

ABSTRACT (ENGLISH)

Background

Arts-based methodologies and methods (ABM) can elicit rich and meaningful data with seldom-heard groups and empower participants in research. Young people with complex psychosocial needs could be better engaged in research using arts-based approaches to overcome communication and literacy issues as well as distrust of those with power, including researchers. A critical review of the use and impact of ABM among this population is timely.

The purpose of this review is to synthesize and examine the experience and use of ABM with young people with complex psychosocial needs.

Methods

A systematic narrative literature review was conducted with a search of the literature from 2009 to 2021. All abstracts were reviewed independently by two authors and full papers were screened for eligibility against inclusion and exclusion criteria. Data synthesis focused on a descriptive numerical summary and a thematic analysis focused on key patterns across papers relating to the review objectives.

Results and Discussion

A total of 25 papers were included. The most common issues of focus were mental health ($n=10$) and homelessness ($n=11$) and methods using Photovoice ($n=12$) and Body Mapping ($n=5$). Individual interview data ($n=20$) were the most commonly analysed, followed by created works ($n=19$). Less than half the studies involved young people in the interpretation of the data collected. Knowledge translation was not described in almost half the studies, with public exhibits ($n=7$) and forums with service providers ($n=4$) being the most common activities. Key themes across the studies were valued over traditional methods in eliciting data, ABM as an approach to engage these young people in research and the impact of the use of ABM on participants and on key stakeholders through knowledge translation.

Conclusions

The growing field of ABM presents opportunities to enhance research with young people with complex psychosocial needs by promoting meaningful exploration of experiences, engaging participants in research and strengthening knowledge translation. The involvement of young people in the interpretation of data and ensuring that knowledge translation occurs are key areas for future attention.

Patient or Public Contribution

The findings of this review will inform future research to improve the engagement of young people with complex psychosocial needs in research and promote power sharing between researchers and research participants.

FULL TEXT

BACKGROUND

Arts-based research methodologies and methods (ABM) are receiving increasing attention in health and social sciences because of their potential to elicit deeper, richer, more meaningful data from participants' perspective.[1] Drawing on narrative, visual, audio and experiential forms of artmaking, arts-based research can elucidate otherwise hidden knowledge as participants give meaning to their experiences in forms beyond the spoken or written word.[2–5] ABM can involve the integration of artmaking in the data collection process, often in conjunction with other more traditional data collection methods such as interviews, focus groups or ethnographic research. ABM can also be used in the research process to create, disseminate or translate research, often through exhibitions, installation or performance.[1] For the purposes of this review, ABM are understood as those in which participants engage in a creative practice of self-expression at any point in the research process. Practices of creative self-expression might include the use of art forms such as poetry, drawing, mapping, collage, photographs/photovoice, participatory video, digital storytelling (DST), drama and image theatre or theatre.[6,7] We define methodologies as the assumptions and principles in a particular research approach and methods as the research action, that is, the techniques for gathering data or evidence.[8] We understand methods as the tools, techniques or procedures used to gather the evidence.[8] In past reviews, authors have noted that methodology and method have been used interchangeably in the arts-based research contexts,[1] hence why we have included both terms.

ABM have been used in a range of settings and with different participant groups.[3,9–11] With a focus on the dissemination of knowledge in arts-based health research, Fraser and al Sayah examined what methods are considered as arts-based research and how these are used in health where studies involved children and adults.[11] Photographs and drawings were found to be the main methods used and studies aimed to produce new knowledge or translate knowledge to practice in the varying contexts of HIV, heart disease and life-threatening conditions.[11], p.127 Leavy[12] asserts that ABM can complement or even improve upon traditional qualitative research methods, as it brings meaning-making to the forefront of the process of data collection and creation, enriching understanding and empowering participants through the participatory nature of art production. ABM are frequently grounded in participatory methodologies that aim to empower participants as partners in research.[11] Empowerment can mean participant control over art-making decisions, data selection or data analysis and other areas of the research process including guiding research to explore areas and themes relevant to their experience.[11] ABM can therefore promote more meaningful dialogue than more traditional qualitative methods and support discussions around complex or sensitive issues.[11,12] ABM have also been shown to produce data that are viewed as relatable, informative and impactful by both expert and wider audiences, priming them for use in knowledge translation through exhibitions, installation or performance.[1,11,12] The emotive capacity of art can promote audiences into thinking differently, deeply and empathetically about others' experiences.[12]

While ABM have been used to good effect for conducting research with some marginalized population groups, there has been minimal exploration of the benefits and barriers to these methodologies and methods with young people with mental health and related issues.[1] D'Amico et al.[7] discuss the use of arts-based, visual and digital methods and ‘their potential to enhance the quality of data collected and to engage and empower child and youth participants’ (p. 529) facing global adversity. De Vecchi et al.[13] note in their scoping review the ‘lack of uptake’ (p. 191) of digital storytelling in mental health research despite ‘implications for the development of recovery-oriented mental health services’ (p. 191). Challenges to the use of ABM are time and resource constraints.[14] The provision of tools, time and support for the creation of art projects can be expensive and may not scale to larger sample sizes.[15] Ethical issues, such as ownership, anonymity and risks of retraumatization, are other challenges to the use of art in research.[11,16]

A critical review of the use and impact of ABM among young people with complex psychosocial needs is timely. We understand complex psychosocial needs as multiple levels of need, for example, a diagnosed mental health issue, but also including issues such as unstable housing, involvement with the social care and justice systems and co-occurring drug and alcohol issues.[15,17–25] This definition and the scope of included studies in the current review are addressed in further detail in the methods section. There are many challenges in research, including the use of arts-based methodologies with young people with complex needs, particularly those with poor literacy and high levels of distrust of those with power, including researchers.[14,26] The rapidly changing field of social media and technology use among young people also underscores the need to assess the use of other technologies beyond the traditional photovoice, photo elicitation and other visual storytelling devices used in past research with young people.[14] Ostensibly, these methodologies and methods are appropriate for this population because they sit within an emancipatory research paradigm, emphasizing empowerment and knowledge creation within the process of the research rather than merely the end-product.[27,28]

Arts-based methodologies are thought to extend traditional research knowledge boundaries to value and include participants voices and to seek to provide participants with greater power in the research process.[16] This is because artmaking in the research process creates space for participants to safely control the expression of their perspectives. This has been powerfully demonstrated for participants who are often marginalized or silenced within society.[29,30] Often, participants' expressions lead to a ‘deeper understanding’ (p. 683) of the way people experience their health and well-being by not only engaging participants in the research process but also by motivating policymakers and healthcare professionals into action to improve services.[16] Published studies highlight several beneficial outcomes from using arts-based, visual and digital methods in research such as their ability to provide counter-narratives and reduce stigma. De Jager et al.[31] found that studies using Body Mapping methods empowered participants by enabling them to produce a first-person nonverbal narrative. The method was found to engage young participants and balance power dynamics between the competing world views of participant and researcher. In their systematic review of digital storytelling in research, de Jager et al.[3] note therapeutic benefits for participants and potential to reach policymakers and create positive social change.

This systematic review is unique, as previous systematic reviews have tended to focus on the use of a specific arts-based, visual and digital research methodologies, while we are instead focussing on a particular population subgroup: young people with complex psychosocial needs and the use of any arts-based methodology or method to engage them in research. More specifically, the objectives of the review were to identify and synthesize the evidence about

- (1)
  The types of arts-based research methodologies and methods used in research to measure experience; access to and/or experience of health and related social service and impact or outcomes of services among young people with complex psychosocial needs.
- (2)
  How arts-based methods and/or creative outputs were used at different research stages.
- (3)
  Reported improvements in data elicitation about access, experience and outcomes of health and related social services compared to more 'traditional' qualitative approaches.
- (4)
  The involvement of young people in meaning-making.

Synthesis of the evidence about the methodologies and methods used, their impact on data collected, policy and practice and young people themselves will inform thinking around new innovations, use and benefits of arts-based visual and digital research methodologies and methods to improve policy and health programmes for young people with complex psychosocial needs.

METHODS

The systematic review was conducted in accordance with PRISMA guidelines[32] and has been registered with PROSPERO (Examining the use of arts-based, visual and digital methods in research with vulnerable youth, registration number CRD42020161675).

Search strategy and information sources

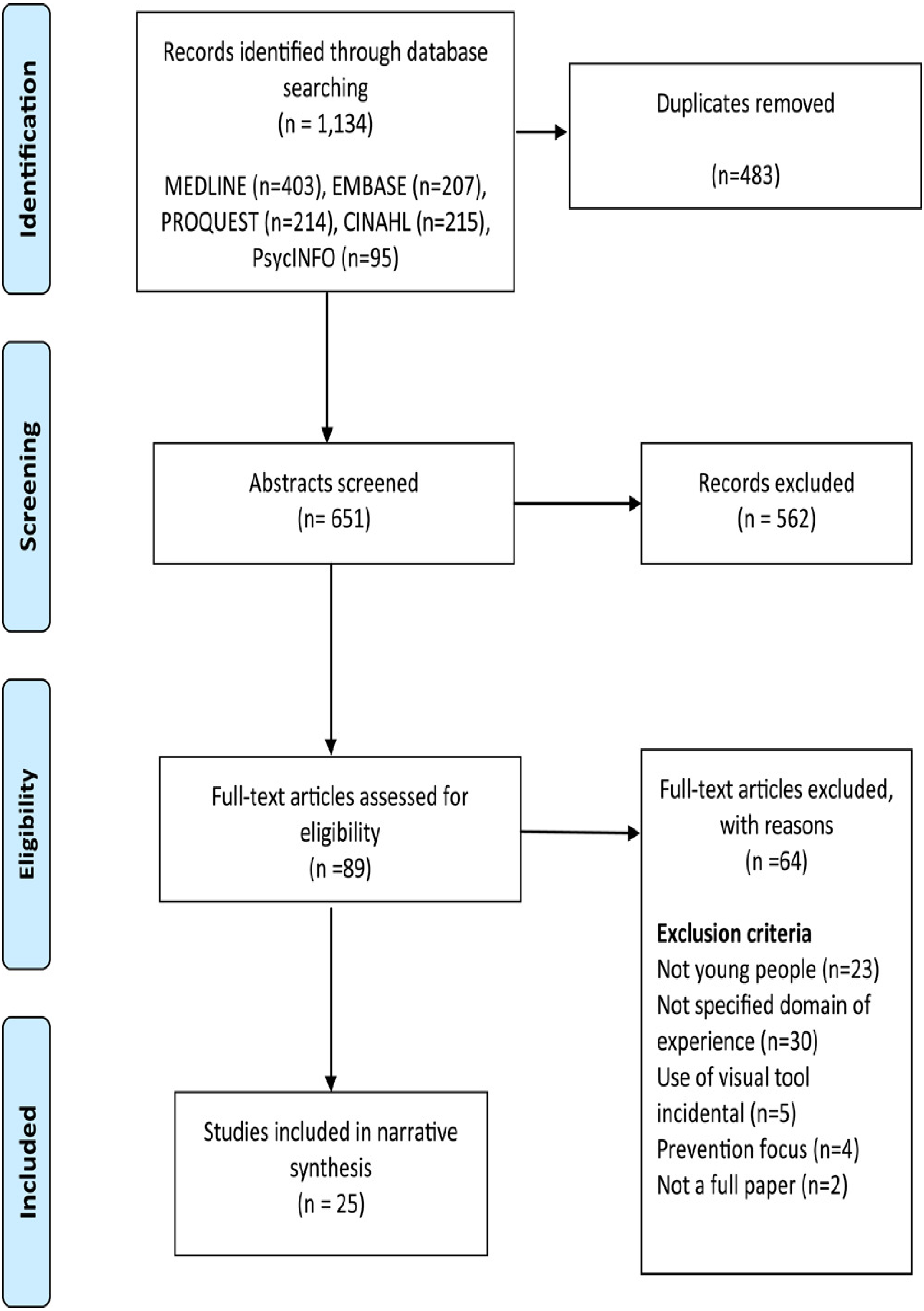
Four electronic databases were systematically searched: Medline, ProQuest, Embase and CINAHL. A comprehensive set of search strategies was implemented to identify all relevant studies (see Supporting Information: 1). Pilot searches were run to test the terms using key articles expected to meet our inclusion criteria and minor changes were made to search terms to ensure that key papers were included.

Inclusion criteria

Studies included were those using ABM, either on their own or in combination with or other qualitative or quantitative research methods, with young people aged between 10 and 24 years with complex psychosocial needs (e.g., have directly experienced mental health difficulties that would meet criteria outlined in the Diagnostic and Statistical Manual of Mental Disorders or have experiences of homelessness in the community). Young people who are homeless are a deliberate focus of this review, given that the incidence of mental health disorders in this group is significantly higher than among young people in the general community and they are less likely to have a diagnosis.[23,24] Those studies that were focused on young people with a diagnosed learning and developmental disorder or intellectual disability were not included after discussion by the full research team. It was agreed that they constituted a group of participants for whom arts-based approaches would be valuable, but that ABM would require substantial tailoring specifically to their needs, which warranted a separate and more focused systematic review. Detailed inclusion and exclusion criteria are provided in Supporting Information: 2.

Review process

Articles identified were exported from each individual database using export tools in EndNotex9 bibliographic software and duplicates were removed.[33] At least two independent reviewers from the research team were involved in the processes of screening for eligibility, extraction and data synthesis. At full-text review, the reasons for exclusion were recorded. The results of the search in the Preferred Reporting Items for Systematic Reviews and Meta-analyses (PRISMA) flow diagram[32] are provided in Figure 1.



Enlarge this image.

Data extraction

A data extraction tool was developed and piloted by the research team. The data extracted included details about the population, setting, domain of experience, study methodology and methods, data collected and analysed (including young people's involvement) and knowledge translation strategies. The data extraction tool with an example is provided in Supporting Information: 3.

Data synthesis

The synthesis first focused on a descriptive numerical summary of countries, the papers that focused on health issues, methodology, arts-based method(s) used, data collected and analysed, approach to analysis and knowledge translation strategies. Thematic analysis was then used to identify patterns across articles regarding how ABM are used in research with young people with complex psychosocial needs, focusing on the value of ABM, the involvement and impact on study participants and challenges.[34] To achieve this analysis, two reviewers:

- (1)
  Familiarized themselves with the studies and findings in each of the papers referring to the extraction table and full papers as required;
- (2)
  Generated an initial coding scheme to organize data thematically discussed with the full research team;
- (3)
  Amalgamated/organized codes to generate potential theme and
- (4)
  Reviewed themes in terms of their salience across the reviewed articles and in relation to the review objectives.

Assessment of methodological quality

This systematic review was concentrated on the research process and approach, methods used, data obtained, impact on participants and knowledge translation rather than overall study quality as the focus of systematic reviews is to assess evidence of interventions on health outcomes. However, data were extracted in relation to evidence about the value of ABM in improving data quality, impacts of the research on service delivery or policy and the impacts on study participants, for example, whether ABM enhance the quality of data in terms of richness, recruitment, retention and engagement of participants, which are key challenges in research on sensitive issues and with ‘vulnerable’ participants.[35]

RESULTSDescriptive results

ABM were used with young people with complex psychosocial needs in studies across several diverse countries and continents. Most studies were carried out in the United States (*n* = 6) and Canada (*n* = 5), followed by Australia (*n* = 4), Central/South America (*n* = 4), Africa (*n* = 2) and one each in South Korea, the United Kingdom, France and Sweden.

Domains

Issues or conditions most often studied were mental health (*n* = 10), homelessness (*n* = 11), general complex support needs (*n* = 2) and addiction (*n* = 2).

Methodological frameworks

Participatory methodologies (*n* = 10) were the most common, followed by interpretative phenomenological and hermeneutic (*n* = 6), unspecified qualitative methodology (*n* = 7), grounded theory (*n* = 1) and narrative approaches (*n* = 1).

Arts-based methods

The definitions of arts-based methods and exemplar papers from this review are provided in Table 1. Photovoice (*n* = 12) was the most commonly used method, followed by Body Mapping (*n* = 5). These findings were consistent with a systematic review conducted by Fraser and al Sayah,[11] who found that visual methods, such as photos and Body Mapping, were the most used ABM.[2]

Table 1 Definitions of arts-based methods and use in included studies

Arts-based method	Definition	*n* (%)	Paper examples
Photovoice (including Photo-blogging/participatory photography)	Photovoice is an established method developed initially by health promotion researchers Wang and Burris.[64] The method involves participants taking photos guided by a research question, which are then used to help them reflect upon and explore the reasons, emotions and experiences that have guided their chosen images.	15 (58 %)	The study of Bender et al.,[54] 'Asking for Change' Photovoice intervention with young people who were homeless aimed to (1) build relationships and connection, (2) teach social, emotional, leadership and photography skills and (3) empower young people to be social change agents. Guided by a professional photographer, young people were each provided with an electronic tablet to take photos about their lives to share and discuss with the group. A mixed-methods evaluation showed that the approach was feasible and highly acceptable, created new opportunities to connect and was associated with improvements in communication skills, social connectedness, resiliency and well-being.
Photo or visual-elicitation	Photo and visual elicitation methods involve using a photograph or other visual stimulus as a support during a research interview.[65]	3 (11 %)	The study of Lecomte et al.[37] aimed to understand the role of food in family interactions amongst teenagers with Bulimia Nervosa. As part of their method, participating teenagers were asked to take a photograph of the table after a family meal before clearing to discuss with the researcher during an interview. The selected photograph was displayed on a computer screen during the interviews and was the basis for the questions asked. The same picture was also used for an interview with their parents.

Body Mapping	Body Mapping involves creating life-sized artworks of the human body to visually depict an individual's perception of their body, identity and experiences. Most commonly, the method involves drawing an outline in pairs of a person's body in a position they wish to be represented on a large piece of drawing paper and then using a range of art materials to fill in the Body Map in response to questions posed in a workshop.[31]	5 (19 %)	The study of Macken et al.[2] aimed to explore the use of Body Mapping as a research method with young people in residential treatment for drug and alcohol issues and to examine how Body Mapping can engage young people in exploring their strengths and sources of support during treatment. They found that Body Mapping produced richer data and accessed new narratives not usually captured in more traditional interviews, in a subsequent interview where young people explained their Body Map.
Digital storytelling	Digital storytelling (DST) involves using multimedia consisting of images/segments of video with background music and voiceover narrative to explore experiences.[66]	1 (4 %)	The study of Boydell et al.[56] involved young people diagnosed with psychosis. Participants attended a DST workshop session to produce individual digital stories describing how they manage psychosis in everyday life. Overall, young people found the story creation process both challenging and rewarding. For instance, although creating the story was emotionally difficult and ‘scary’ for participants, they reported feeling better and a sense of relief when it was completed. Participants found it difficult to make decisions about what part of their story to tell.
Filmmaking	Filmmaking was involved in only one included study, which involved co-production of a film to disseminate research findings.	1 (4 %)	The study of Dunn et al.[52] details their efforts to find ways to provide information about depression and help-seeking beyond traditional academic audiences, specifically to other young people experiencing depression. This involved the creation of a film through a collaborative workshop where young people shared their experiences, decided on the tone, tenor and message of the film, identified their primary audience and produced the bulk of the audio and visual material.

Song writing	Song writing was a method used in only one included study, which involved co-writing a song to analyse and summarize data generated from qualitative methods.	1 (4 %)	The paper of Fairchild and McFerran[55] described a music workshop that involved collaborative song writing with children experiencing homelessness and family violence. This included engaging participants in creative and child-centred forms of data generation, analysis and presentation of the findings. The researchers focused on the use of group song writing to co-create knowledge and understanding and to ensure that the final product represented the young people in ways that they could recognize and resonate with.

Data collected and analysed

Table 2 shows the types of data used in analysis in these studies. Individual interview data (*n* = 20) were the most common data type, followed by created works (*n* = 19), field notes (*n* = 8) and group interview data (*n* = 7). Many studies used more than one type of data for analysis and often used created works as prompts and devices to explore issues during interviews.

Table 2 Use of arts-based methods and/or creative outputs at different research stages (*n* = 25)

Research stage	*n* (%)
Data collection:	
All studies involved young people's participation in the creation of art or image	25 (100)
Data used in analysis:	
Artwork and associated descriptors[36,38–41,43–46,48–54,56,57,59,60]	19 (76)
Individual interviews[36–40,42–44,46–50,53–59]	20 (80)
Group interviews or discussions[36,38,45,49,51,52,57]	7 (28)
Researcher field notes[40,45,46,49,50,55,57,58]	8 (32)
Quantitative self-report measures[44,55]	2 (8)
Other[40,44,55,60]	4 (16)
Knowledge translation:	

Not described[37,39,40,42,45,48–50,56–59]	12 (48)
Forums with service providers[38,41,44,46]	4 (16)
Exhibition of artworks[36,44,51–53,55,60]	7 (28)
Other[38,43,60]	3 (12)

Approach to analysis

Thematic analysis (*n* = 9) was the most common approach to analysis, followed by unspecified qualitative analysis (*n* = 8), content analysis (*n* = 4) and visual narrative analysis (*n* = 2). In our review, almost a third of the studies did not report any particular theoretical orientation in their analysis. In their systematic review of ABM, Fraser and al Sayah identified a need for a more explicit discussion of theoretical underpinnings in this area of research.[11]

Knowledge translation

Knowledge translation, when described, occurred through public exhibits (*n* = 7) and forums with service providers (*n* = 4). In nearly half the studies, knowledge translation activities were not described. See Table 2.

Thematic analysis resultsEnriching data through arts-based research methods

Almost all studies emphasized that the use of ABM elicited richer data than solely relying on traditional qualitative methods. Several studies reported the potential of arts-based research methods to elicit information that is not readily expressed in words or text.[36–40] Several authors argued that ABM allowed for participants' viewpoints to be represented with greater authenticity and depth.[2,37,40–44] Young people were also reported to be able to better disclose and share complex and difficult experiences and emotions.[2,36,44–47]

Engaging young people with complex psychosocial needs

ABM were found to be highly engaging for young people who are traditionally hard to reach and retain in research.[36,41,45,48–50] Several authors noted its utility in initiating and facilitating dialogues with young people, particularly about difficult and confronting issues.[2,37,42,46,51,52] Studies also described the use of created art as prompts to initiate an interview, generate deeper exploration or at the end of interviews as a means of keeping young people engaged.[37,51] This finding was consistent with Bagnoli's review of ABM, in which they found ABM to be useful as an ‘ice breaker’.[53] ABM were also reported to be useful in holding young peoples' attention, particularly through research elements requiring concerted participation.[2,43] The research process was reported to be enjoyable and valuable by participants who found the use of art to be more familiar and comfortable than the spoken word.[2,42,45,49,52,54]

Sharing power

Participation is considered central to ABM, as collaboration with researchers empowers participants and allows a richer representation of experience. We were therefore interested in how often ABM allowed for substantive participatory involvement in analysis. In the studies reviewed, there were several forms of participation in analysis: identification of themes or topics (*n* = 8), co-analysis by guided group discussion (*n* = 7), selected pictures for analysis (*n* = 4) and analysis through art production (writing lyrics, etc.) (*n* = 2). In our review, 12 studies (44%) involved some form of participation in analysis. Several authors noted that ABM promoted a more democratic process in research and valued young people as partners and experts[2,38,42,47,49,51,52,54] and promoted a reciprocal empathetic relationship between researchers and young people.[2,38,43] Due to the creative nature of ABM, the skills and assets of young people were seen to be more valued.[42,52] Young people were also reported by the authors to be more able to influence the direction of research through participation than in research using more traditional methods.[36,48,49]

Impact on participants and therapeutic effect

In several studies, authors noted that ABM improved young peoples' self-efficacy or self-worth.[36,54–56] The collaborative nature of ABM was reported to facilitate the development of participants' communication and social skills.[45,48,52,54,57] In some studies, participants were able to share common experiences with peers and developed new support networks.[43,45,46,54] Young people were reported to have gained new creative skills and took pride in the creation of art they found to be meaningful.[45,48,52,56] ABM also served as an emotional outlet for young people and promoted self-reflection, offering them an opportunity to discover unexpected strengths, resiliency, optimism and process their past experiences.[2,36,38,41,45,47–51,54,56–59] In one study, ABM was noted as having a calming effect.[2] Bender et al.,[54] who used a validated self-report measure, found that ABM improved young peoples' social connectedness, resiliency and personal well-being.

Challenges in the use of arts-based research methods

The most often cited challenge to implementing ABM was that it was resource- and time-intensive.[41,52,54,56] Ethical issues, such as ownership over art, consent and the presentation of photographs including minors, were also reported.[45,47,48,50,54,59] Issues pertaining to group interactions, participant attention and retention arose due to the creative and relatively unstructured nature of the research, as well as the complex psychosocial needs of the participants in a few studies.[51,55] Some researchers expressed a concern that ABM may expose young people to retraumatization by triggering past experiences[50,52] and highlight the need for support to be provided during and following ABM projects.

DISCUSSION

Several key themes emerged from our analysis and synthesis of the use of ABM across a diverse range of studies with young people with complex psychosocial needs. ABM's value over traditional methods, the engagement of young people in research and the impact on participants and on others through knowledge translation were key areas of focus in this systematic review.

In almost all studies, ABM were found to elicit rich and insightful data from young people with complex psychosocial needs. This is an important finding, as research with these young people can be challenging, including in terms of data quality.[60] A major barrier to productive dialogue with young people in research has been found to result from their discomfort and hesitancy to express themselves through words.[14,26] ABM bypasses this challenge, effectively capturing nonverbal and preconscious information.[2,36–38,40,61] For example, Willis et al.[39] found that ABM were effective in 'externalizing somatic and emotional experiences' (p. 2). Importantly, ABM also enabled young people to identify strengths and positive traits,[43] which are often overshadowed by a more deficit discourse in more conventional approaches.[1,2,10] A focus on resilience and optimism in research can also translate to interactions outside of the research process as found by Macken et al.[2] in their study, in which young people took their body maps to counselling sessions to explore their strengths and sources of support. While most studies in this review reported positive psycho-social outcomes from participation in the research, some studies noted a concern regarding retraumatization.[50,52] Attention to ensuring appropriate psychological support is important in future studies.

Qualitative research methods are argued to be more effective if they can engage participants and therefore collect data that better reflect the experiences and meaning-making of the participants.[62] In this review, ABM was found to be highly engaging for young people and promoted interaction with researchers and in the research process.[2,36,37,41–43,48–51] By holding young peoples' interest and curiosity, the use of art allowed for a 'progression from factual, descriptive questions to more personal ones' (p. 3).[36] The use of art appeared to foster an openness to self-expression and communication that allowed for more involved, sustained and enjoyable participation in the data collection process and shifted the unequal power dynamics often inherent in research.[2,14,42,49,52,54]

ABM are often grounded in a participatory methodology [14] as we found was used in 10 of the 25 included studies. Leavy[12] asserts that in ABM, participants are often viewed and valued as equal collaborators, shifting the traditional researcher-researched hierarchy. As there is no correct way of understanding art, ABM democratizes meaning and decentralizes power between researchers and young people.[12] From this position, it is argued that compassionate and productive reciprocal relationships can be formed that lessen the barriers of age and authority.[38,52] As a result of equalizing power differentials, young people can more readily engage with research and share their experience.[3,50] Several studies in this review noted the importance of participation in ABM to promote meaningful engagement and therefore more authentic data.[38,49,50] Importantly, empowering young people with a voice and a platform can actively address the stigma and inequity and underlying power differentials that they may experience both in research and outside of research.[38,45,48,52] In nonparticipatory methodologies, participants can be detached from the analysis and meaning-making about the data they produced. In contrast, when young people were involved in the identification and development of themes, as was the case in almost half of the studies, researchers reported greater confidence that the findings produced reflected participants' lived experience.[14,63]

ABM can have a profound effect on participants. Young people reported being empowered through the acquisition of creative skills and the production of art.[48] An opportunity to create something meaningful can enable young people with complex psychosocial needs to build their sense of self, identity and self-esteem and change their and others' perceptions of what they can accomplish.[45,48,52,56] ABM can also have direct therapeutic benefits.[3] In our review, ABM appeared to draw out young peoples' strengths, resilience and optimism.[36,38,41,45,47–49,54,56–58,63] However, Forge et al.[51] expressed concern that participant retention was impacted by significant barriers in the lives of the young people, with only two studies reporting making participation in the research flexible to account for these barriers.[42,52]

While the use of art in research has been found to increase the accessibility of findings to the general public, key questions remain about how ABM can facilitate knowledge translation and influence policy and practice.[1,12] In our review, 12 studies (48%) did not describe knowledge translation activities. The lack of attention to knowledge translation in almost half of the reviewed studies was consistent with the findings of de Jager et al.[3] and highlight an area that requires attention in future studies to maximize the impact of the use of ABM. While ABM can have important positive impacts in the research setting, it is vital that meaningful and tangible changes are focussed on service provision and policy.[12] A common theme throughout many of the reviewed studies was young peoples' strong desire to share their stories and advocate for issues through their art, underscoring the importance of knowledge translation activities.

FUTURE RESEARCH

Future research should explore any differences in the feasibility and acceptability of different methods and their benefits for young people with distinct issues, such as those experiencing mental health issues compared to those who are homeless and also for those with different gender identities and diverse ways of thinking, learning, processing and behaving, often referred to as neurodiverse individuals. The issue of retraumatization or the triggering of past difficult experiences was raised in some studies. Future research using ABM should identify and evaluate approaches to keeping participants safe and the types of supports that are provided to mitigate any harms.

CONCLUSION

ABM is a growing field, demonstrated by having 22 of the 25 papers identified and included that were published in the last 5 years (since 2017). The use of ABM with young people with complex psychosocial needs was reported to elicit rich and authentic data, while engaging participants and empowering them as partners in research. ABM was reported as adding substantial utility to more traditional qualitative research approaches including promoting meaningful exploration of participant experience. ABM was found to have benefits for young people, who gained creative skills, social networks and an opportunity for self-reflection. Knowledge translation is a key area for future attention, given the potential of the data generated through ABM to impact on policy and practice.

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CONFLICT OF INTEREST

The authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

The author has provided the required data availability statement, and if applicable, included functional and accurate links to said data therein.

DETAILS

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‘It's like a never-ending diabetes youth camp’: Co-designing a digital social network for young people with type 1 diabetes

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ABSTRACT (ENGLISH)

Introduction

Living with a chronic condition such as type 1 diabetes (T1D) affects everyday life and support from others experiencing a similar situation can be helpful. A way to receive such support is to use an online network where people can connect and share experiences. Research has described the benefits of using such tools for connecting patients. The aim of this study was to describe the co-design of a social network for young people with T1D and to describe their experiences when using this network.

Methods

A co-design approach was used, following three steps adapted from Sanders and Stappers (2008). In all, 36 adolescents with T1D participated. Data in the form of recordings and notes from telephone interviews, workshops and focus groups were collected and then analysed using content analysis. Numerical data from the digital platform were also used.

Findings

For the interpersonal values, supporting, learning and relating to emerge, the framework of the network must be appealing and user-friendly. The limits of time and place are eliminated, and there is a possibility for many more to join in.

Conclusion

Co-design ensures that what stakeholders think is important forms the basis for the design. The interpersonal values that are promoted are ones that only the exchange of lived knowledge and experience can generate. It is complementary to the support that healthcare professionals can offer; thus, this kind of social network is important for improved, coproduced care.

Patient or Public Contribution

The participants in the present study were persons living with T1D. They were active co-creators from the start to the end. An adult person with experience of living with T1D was involved as an advisor in the research team when drafting the manuscript.

FULL TEXT

INTRODUCTION

Type 1 diabetes (T1D) is a noncurable chronic condition that requires treatment and ongoing self-management ‘24/7’. Adolescents with diabetes and their caregivers spend less than 1% of their time a year visiting a diabetes healthcare provider.[1] Knowledge about the disease and the need for support are important factors in adolescents' lives; having a chronic condition may lead to the feeling of being different.[2] Between the ages of 11 and 15, most young people perform much of the daily diabetes management, and in mid-teens (ages 15–17) take responsibility almost entirely on their own.[3] Living with T1D has a huge impact on adolescents' daily life,[4] and they troubleshoot and make decisions in day-to-day activities on their own; therefore, they seek out and understand the importance of getting support from others in a similar situation.[5–7] Even though healthcare professionals are aware of this need, it may be difficult to organize groups within the healthcare system and even if they do, few young people join in.[8] Consequently, using other forms more focused on young persons, such as online communities, could represent one solution.

Online social networks

An online social community or network involves people sharing experiences and supporting each other in online activities.[9,10] Recent research describes the benefits of using different tools for connecting patients with chronic conditions to their peers.[11] For example, young women with T1D found comfort in receiving social support on forums on the Internet from others in the same situation; it helped them maintain a balanced view of their lives and to manage life transitions.[12] Adolescents and young adults with T1D who use social media in their everyday lives achieved better control compared to patients who did not use social media.[13] In the United States, 23%–39% of young people seek peers online.[14] Moreover, online social networks could be useful tools for patients and/or their caregivers to learn about blood glucose devices and receive technological assistance from other members. Through closed groups, members may help others in the network by spreading awareness about the condition itself, and providing emotional support and/or technical assistance when building on members' shared experiences.[15]

Existing research exploring diabetes online communities shows that people with T1D seek out diabetes online networks because it is challenging to identify a peer in real life. The shared experience has been mentioned as the most frequent topic discussed in several studies, and the sense of normality and validation of lived experiences are also central.[10] The enthusiasm about using networks goes beyond information support and adds the value of emotional solidarity, shared feelings and experiences.[16] The immediate response and orientations from other members give opportunity to acknowledge the community as a safe space. This is not possible in the contacts with healthcare providers.[17] A recent scoping review of diabetes online communities reported promising results showing several benefits and relatively few negative outcomes. This points to the importance of participatory frameworks with the inclusion of users in the design and in the parameter-setting stages, to better capture community elements and potentially increase social validity and usability of future networks.[10] This indicates that the users should be involved in the design process when developing a social network. The aim of this study was to describe the co-design process of a social network for young persons with T1D and to describe the experiences of using this network.

METHODSParticipants

A co-design approach was used involving joint exploration and articulation of needs and solutions.[18,19] A sample of convenience[20] was created, in which three outpatient children's departments, serving about 700 patients, in the southeast of Sweden were involved. The inclusion criteria were young persons between 13 and 17 years of age diagnosed with T1D at least 1 year before the initiative (Table 1). Twelve persons from each department were included (*n* = 36). Diabetes nurses at the participating departments provided contact information to parents whose adolescent children fulfilled the criteria. Members were informed that participation was voluntary, and that they could withdraw at any time without explanation, following the Declaration of Helsinki. Approval was given by the Swedish Ethical Review Authority (D.nr 2018/449-32). Co-designers were, in addition to the young persons, two researchers with experience of working with diabetes care, a software developer and two community managers from a health tech company.

Table 1 Description of participants' age and gender in the various steps during the study

Step	Activity	Invited	Participated	Age and background information	Gender
I	Telephone interviews	*N* = 36	36	13–17 years (median 15)	18 girls
				Range of years since diagnose 1–14 years (median 5)	18 boys

	Workshops	N = 36	21a	13–17 years	16 girls
					5 boys
III	Summarizing focus group interview or Telephone interviews	N = 36	6	13–17 years	5 girls
			3		1 boy
					3 girls

a

Reasons for not participating in the workshops were not having the time/opportunity on the day of the workshop or not interested in getting involved.

Procedure

The process of co-design followed three steps, adapted from Sanders and Stappers[19] (Figure 1). In the first step, telephone interviews were conducted to map out the general use of social media. These formed the basis for the semistructured workshops, where content and structure were enclosed for the online social network. The second step consisted of the iterative process of developing and testing the social network, and in the third step, a focus group was conducted to describe the experiences of using the network.

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Step I

Young persons with T1D were interviewed separately by phone (Table 1). The interviews were semistructured according to an interview guide (Table 2). Interviews were recorded and transcribed verbatim. The purpose was to understand these persons' use of social media and their ideas for developing a network for young persons with T1D. Then, they were invited to one of three semistructured workshops led by a software manager and two community managers.[21] Of these, 21 persons (hereinafter referred to as members) participated in the workshops. The members had a meal together, while the software manager coordinated the discussion about habits of using social media in general: when, how, where and why they used different types of social media. The following discussion was then focused on what the intended social network would be like, including the content, technical features and user-friendly functions. The discussion also focused on how the members wanted to use such a network and what type of content was important. Members wrote post-it notes, which were grouped based on subject areas and clustered into different subjects, and were later used to create different areas within the social network. Two researchers observed the conversation and took notes.

Table 2 Interview guide used in the telephone interview in Step I

Question area	Questions
Background questions	Age, duration of diabetes diagnosis
	What kind of social media do you use today and how do you use them?
Thoughts about a social network for youths with T1D	If there was a social network for youths with T1D: what would be in it for you?
	What functions would you prefer?
	What would make you use such a network?
	Do you believe in the idea about such a network?
	Why?
	Why not?
	What are your thoughts about sharing your experiences of diabetes with others?
	What do you think about adults' presence in such a network?
Question about participation in the co-designing process	Would you like to be involved in designing such a network together with other young people with diabetes and a software manager?

Step II

A framework for social networks was used and was adjusted based on the members' views and requirements described in Step I. An invitation was sent to all 36 members to test the network over 12 weeks. During these weeks, an iterative process of developing content and structure and adjusting for errors was performed simultaneously. Members could create their own posts, write their opinions and views about the content and interact with each other.

The Community Manager also posted questions, news and polls to engage members. All new posts led to a notification by email and a push notification in the app in members' smartphones. Members could comment on other persons' posts and they could ‘clap’, which had the same function as ‘like’ on other social media. There was also an ‘eye’ symbol with a number indicating how many members had read the post. Before the members got access to the community, each member was paired with another member as a ‘buddy’. Each buddy-couple received tasks to discuss; the community manager initiated these tasks. This buddy function was a way to meet the need for individual support described in previous research.[22] Data were collected from the community platform following the number of posts, comments and likes for members and the community manager.

Step III

After the test period, all members were invited to a focus group interview led by the community managers. This was recorded and transcribed verbatim (Table 1). Two researchers observed and took notes during the focus group. The members were asked about their experiences, views and ideas for improvement of the network. The purpose of this focus group was to describe the members' experiences of participating in the development of the network and using the network.

Data analysis

The telephone interviews in Step I were read several times to make sense of the data. Then, the text was coded by the first author and entered into a spreadsheet. The codes were grouped into subcategories by the first and last author. A thematic analysis was performed using an inductive approach strongly linked to the raw data.[23] Numerical data (Step II) from the community platform were compiled. The focus group interview (Step III) was analysed using inductive content analysis.[24] First, meanings and sentences were assigned a code. In the open coding phase, codes were grouped into categories and sorted under higher-order headings. Similar subcategories formed higher-order categories. To achieve trustworthiness, the other authors in the research group read and discussed the analysis.

RESULTS

In the telephone interviews (Step I), members described incentives for becoming a member of a social network, such as the opportunity to connect with others and share experiences together. Another incentive could be to provide and receive information and support. Furthermore, important prerequisites were described, especially the presence of an adult, that the network has many members and that all members have their own experience of T1D. The workshop's discussions focused on functions, opinions and the members' habits around using apps, the internet and smartphones. This formed the basis for designing the social network. Members stated the importance of simplicity, such as easy access and a clear overview. Quotes illustrating the participants' opinions are presented in Table 3.

Table 3 Quotes from telephone interviews and workshops illustrating the incentives and prerequisites for using a social network focused on young people with T1D (Step I)

Incentives and prerequisites	Quotes from interviews (Step I)
Connecting	I think it's a good idea, because then you can write with each other about the disease if you don't want to share it with someone else who might not understand it
T1D only	It feels good to know that it is only for those with diabetes. It might feel better then.
Support	I think it would be really, really nice, because since there aren't very many people with diabetes that you know, it would be great to have everyone gathered in one place because you sometimes feel quite alone

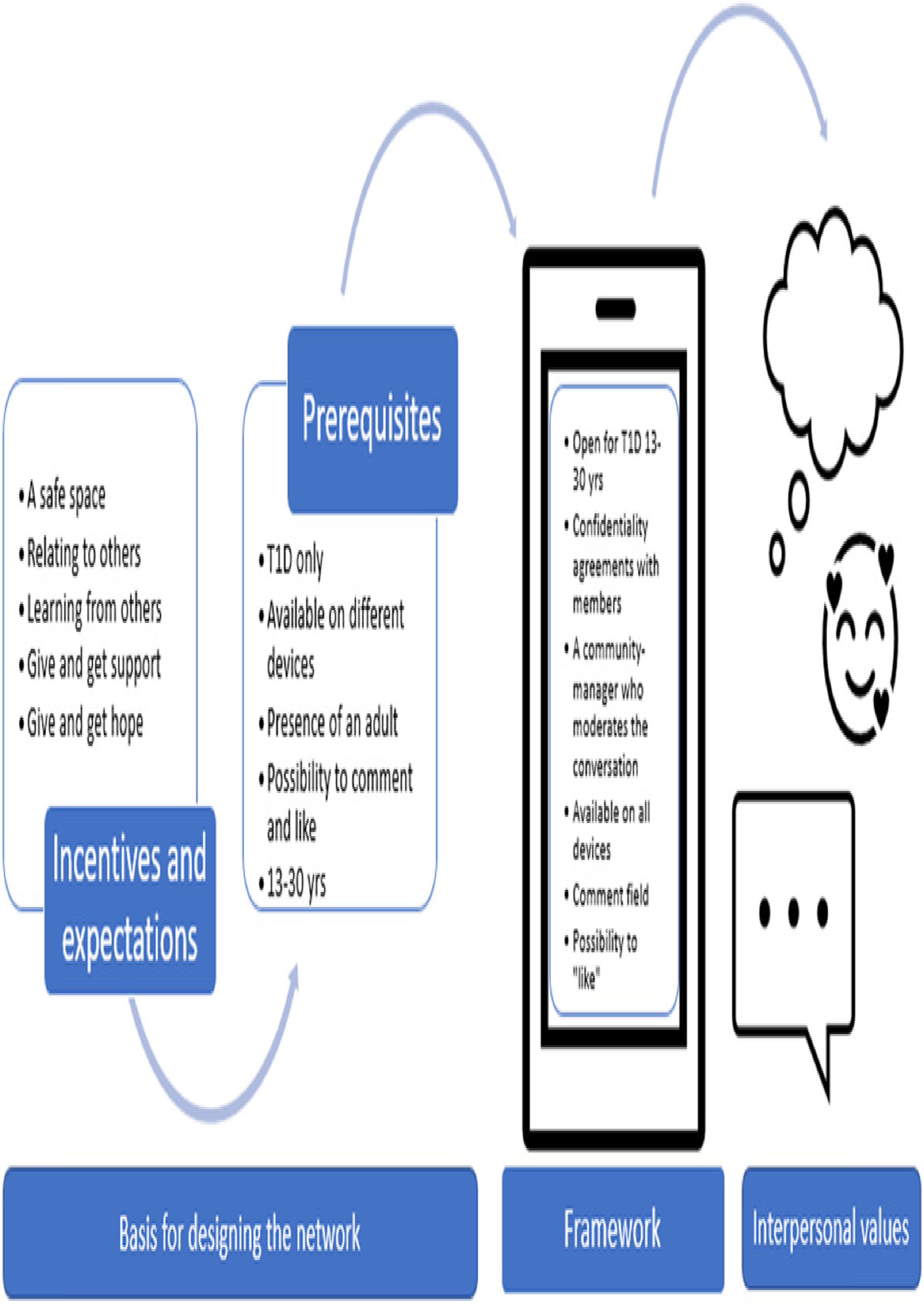
Share experiences	Maybe more that you post things there about tips and advice that you yourself think work
Presence of an adult	It might be more special if it's only for young people, but it's good if there are adults who can keep an eye in case something bad is shown

After clustering the subjects, five areas of different subjects emerged (news about diabetes, free questions, missions, polls and challenges—the last three created by the community manager). Then, members formulated rules for the social network. These included using a positive tone in the posts and avoiding negative reactions that could be interpreted as offensive. All members agreed that the community manager should be responsible for coordinating content, outsourcing assignments and polls and questions during the testing period.

All 36 members were invited to join the test period (Step II) and 33 logged in to the network. The members were encouraged to report any technical problems, which contributed towards improving the functionality simultaneously. Most posts and interactions were initiated by the community manager (87%). The posts concerning diabetes-specific questions and information about diabetes received most feedback from the members. There was almost no contact between the buddie couples. Some of the members tried to get in touch but received no response from their buddy.

To describe experiences of using the platform and of being a member of the network (Step III), all members were invited to a focus-group discussion. The content analysis of this discussion revealed two categories: framework and possible interpersonal values (Figure 2 and Table 4). Quotes to illustrate the categories are presented in Table 4.

The *framework* included ‘functionality’, exemplified by aspects such as accessibility and that the interface should be attractive irrespective of the devices used (smartphone, tablet or computer). Polls, posts and how notifications should be presented were discussed and also what kind of functionality was desirable. The members did not find any use for the buddy function, but it was argued that this kind of functionality may better suit members recently diagnosed with T1D.



Enlarge this image.

Table 4 Quotes from the focus group that illustrate categories from the content analysis (Step III)

Category	Subcategory	Quotes from focus group (Step III)
Framework	Functionality	Great with that ‘eye’ showing how many people had read and reacted on the post
	Access and rules	I don't think we want to be there if there are a lot of parents just reading too
Interpersonal values	Relating	I have seen that there are more young people who feel a little like me. It's a bit what I take with me and it's probably what's nice that you have that app, that you have people who may be the same age as yourself as who you can easily reach
	Supporting	Yes, if I had had this [buddy function] when I got diabetes in the first place, maybe I had appreciated a buddy to ask—but you have that support anyway on the network
	Co-learning	This thing about starting to drink [alcohol] …then it would have been good to have someone who was a bit older who had a bit more experience…

The second part about the framework was described as ‘access and rules’. The rules were about how to engage in the network and which members should be allowed. For example, members preferred that young adults were invited into the network. Issues about transition into adult care and how to prepare for moving away from parents could be important topics to raise in an extended network. The need for more activity from members was expressed since ongoing activity is important for a social network to serve its purpose. Another important issue was about not allowing parents to be part of the network, because their presence may deter members from engaging.

The second category included the interpersonal values that can be developed while on an online social network (see Table 4 and Figure 2). One of the interpersonal values described is about relating; that members share the same experiences and can understand each other in a deeper way. They described several examples in everyday life that those with their own experiences of living with diabetes could understand. It could be the feeling of having a bad day (such as a stubborn high blood sugar) and how others describe that feeling and relate to it. Another value was about supporting, that members could bolster each other. This was illustrated by the solidarity that they could offer each other when they feel a sense of hopelessness. This way of giving and receiving hope was expressed in terms of not being alone in the struggle with the condition in everyday life. Members pointed out that there is a lower risk of a judgemental attitude between those who share the same experience, which then added to the feeling of support. Finally, ‘co-learning’ was mentioned, describing how members could learn different things from each other's lived experiences. This was more implicit, exemplified by the network being a platform for sharing concrete examples of how to do things, for example, what to consider when drinking alcohol and advice concerning food and physical activity. Members could learn from these concrete examples and apply them to their own situation.

Being a member on a social network has potentials for young persons with T1D. However, to accomplish those potentials described as interpersonal values in the present study, the network need an appropriate framework. When all these things come together, it could be considered as a safe space to meet others with the same experiences. It is not restricted to any physical place or an organized group of selected members. The limits of time and place are eliminated and there is a possibility for many more to join in. This was described by the members as ‘participating in a never-ending diabetes youth camp’. Due to the nature of a virtual network, facilitated by a community manager, the safe space for meetup becomes a reality. A camp is limited to time and place and is therefore not available to everyone.

DISCUSSION

To our knowledge, this study is the first to use a co-designed approach with young persons with T1D to create a digital network. Young people have a lot to contribute to the design as they can describe their incentives for using digital social media and how they use it.

The result shows that members want the presence of adults on the site as a prerequisite for the safe space that they value. This is in accordance with the idea of 'facilitated networks' as a possible configuration of value-creating services.[25] Teenagers can be empowered by sharing lived experiences with young adults with T1D,[26] which was confirmed in our study. Members place a high level of trust in their peers and follow their advice about lifestyle changes,[11] which indicates the importance of the presence of healthcare professionals in the network, to minimize the risk of incorrect advice being shared, which could otherwise be a risk.[13] Members expressed the need for functionality, access and rules. Others have described that online networks need mentors, guides and moderators to be present. Such a presence can provide the necessary structure and direction to shift a negative or unproductive social networking process into one that could positively influence social support.[27] In the current social network, the risk of bad behaviour and misinformation has been minimized by the presence of a community manager. Continuous interactions within the network are needed, which are dependent on members' engagement and the willingness to share personal experiences.[27] A critical minimum number of members has been confirmed by White et al.,[28] who report that a rapid increase in membership and level of participation in the network indicates motivation and increases the possibilities for exchange of information. The low level of activity from and between members in this study may indicate that the test is too small or that the group was too homogeneous. Even if there is a small percentage of members who are active on a forum, it is worth keeping in mind that persons who choose not to post content or comment on others' postings may still benefit from observing or being part of the community.[29]

We found that membership in a social online network offers a platform to both seek out and provide tailored social support around diabetes management. Our results about the interpersonal values; supporting and co-learning as incentives for being a member in the network is confirmed by previous studies.[16,29] Members highly value being part of a digital social network since it increases knowledge, improves self-care and reciprocates emotional support.[10,29,30] Knowledge about T1D can form the basis for successful self-management.[27] Belonging to a social network where members can receive emotional support and mutual reciprocity and being part of other members' lived experiences are important driving forces for using diabetes online social networks.[10]

To conclude, the findings in this study are consistent with previous literature about online networks for persons with T1D and provide a more in-depth understanding of the nature of online social networks. The results can be applied to a wider perspective of online networks to foster peer-to-peer support for other chronic conditions as well. The use of co-design adds the value of directing content and structure to meet the needs of potential new members in future networks. It also ensures that structure and content are designed based on what users consider important and not based on assumptions made by others.

Methodological considerations

A strength of this study was that we could work iteratively, due to the close collaboration between members, technology developers, community managers and researchers, although the several steps of data collection in the co-design process were time-consuming. Further, a potential limitation is that people who agree to participate may not be representative of the population. Members were recruited by the diabetes nurse, and we cannot control if the diabetes nurse may have asked specific persons to join the study due to special characteristics. This may have affected the sampling procedure. The members that were included in the present study had several years of experience of living with T1D, which may have influenced the design of the network. The needs can vary depending on how long a person has lived with diabetes. In addition, we started with a rather small group of members, which resulted in a low degree of activity in the network. On the other hand, this was part of the learning process and further cycles with a larger group of users are called for. The network was based on an existing technology framework, which may have affected the members' creativity in the design of features and appearance.

CONCLUSIONS

We identified the potential advantages of joining a network for adolescents with T1D. Relating, supporting and learning together is something that the exchange of lived knowledge and experience can generate. This cannot be provided by healthcare professionals. By using co-design, it was possible to straightaway build on what the young persons described as important. The participation of a facilitating healthcare professional was considered necessary by the users, to make the network a safe space to share and learn from. For future research, we recommend exploring the content in the network that could provide information about what is important for a wider group of young persons living with diabetes, and to also use this as a channel for patient feedback to diabetes teams to enable them to make improvements towards better, coproduced care.

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CONFLICT OF INTERESTS

The authors declare that there are no conflict of interests.

DATA AVAILABILITY STATEMENT

The focus group and interview data (transcripts) that support the study conclusions are unavailable for public access because informed consent to share the complete transcripts outside of the research team was not obtained from the study participants.

DETAILS

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Co-creating system-wide improvement for people with traumatic brain injury across one integrated care system in the United Kingdom to initiate a

transformation journey through co-production

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ABSTRACT (ENGLISH)

Background and Objective

There is a need for better integration of services across communities and sectors for people living with traumatic brain injury (TBI) to meet their complex needs. Building on insights gained from earlier pilot work, here we report the outcomes of a participatory workshop that sought to better understand the challenges, barriers and opportunities that currently exist within the care pathway for survivors of TBI.

Methods

A diverse range of stakeholders from the acute and rehabilitation care pathway and the health and social care system were invited to participate in a 3-h workshop. The participants worked in four mixed subgroups using practice development methodology, which promotes person-centred, inclusive and participatory action.

Results

Thematic analysis identified shared purposes and values that were used to produce a detailed implementation and impact framework for application at both the level of the care interface and the overarching integrated care system. A variety of enablers were identified that related to collective values and behaviours, case management, team leadership and integrated team working, workforce capability, evidence-based practice and resourcing. The clinical, economic, cultural and social outcomes associated with these enablers were also identified, and included patient safety, independence and well-being, reduced waiting times, re-admission rates, staff retention and professional development.

Conclusion

The co-produced recommendations made within the implementation and impact framework described here provide a means by which the culture and delivery of health and social care services can be better tailored to meet the needs of people living with TBI. We believe that the recommendations will help shape the formation of new services as well as the development of existing ones.

Patient or Public Contribution

Patient and public involvement have been established over a 10-year history of relationship building through a joint

forum and events involving three charities representing people with TBI, carers, family members, clinicians, service users, researchers and commissioners, culminating in a politically supported event that identified concerns about the needs of people following TBI. These relationships formed the foundation for the interactive workshop, the focus of this publication.

FULL TEXT

INTRODUCTION

Severe traumatic brain injury (TBI) has a profound impact on people, their carers and families, especially evident when transitioning from hospital to home and the community.[1] Care is experienced as fragmented, and there is an urgent need for better integration across health and social care and the voluntary sector to enable people with TBI to be at the heart of care.[2]

TBI is a major challenge both globally[3] and for the UK healthcare system.[4] As a major cause of long-term disability, TBI can affect all areas of daily life, reducing the quality of life significantly for both the person and their carers.[1–3]

The transition from hospital to home and the community across patient pathways tests the principles of integrated health and social care systems to its limits,[2] and so getting it right for this group of citizens would provide key lessons for all.

Whilst this challenge affects all societies internationally,[3] this paper shares early steps towards transforming services across one integrated care system (ICS), Kent and Medway, in England based on a 10-year history of building stakeholder relationships. Founded on insights about how the care pathway is experienced[2] and using the key principles associated with co-production[5] and practice development (PD),[6] this service development initiative describes and outlines a specific process that has been used to begin to develop more integrated person-centred, safe and effective care and services across the health and social care system. Working in partnership with people who have experienced TBI, their families, carers predominantly through key charities, and other stakeholders, the global approach of ‘what matters to you’[7] underpins the purpose and direction of the transformation required.

The local context

An epidemiological report[8] published by Kent and Medway Observatory identified that the incidence of TBI has increased for two successive years (2017–2019),[8] with the total number of in-patient hospital admissions for TBI increasing from 3645 (2016/17) to 4295 (2018/19). There is no available comparative hospital admission data for this period nationally. The latest regional data for the South East of England for 2019–2020 shows that the rate of finished admission episodes for head injuries has increased by 12% since 2005–2006.[9]

These rates of TBI in Kent and Medway are consistent with European rates. Other key findings from the Observatory report can be found in Box 1.

1.**BoxFindings from the Kent and Medway Observatory report 8**

TBI represents approximately 1% of all admissions (elective and emergency) to hospitals in Kent and Medway.

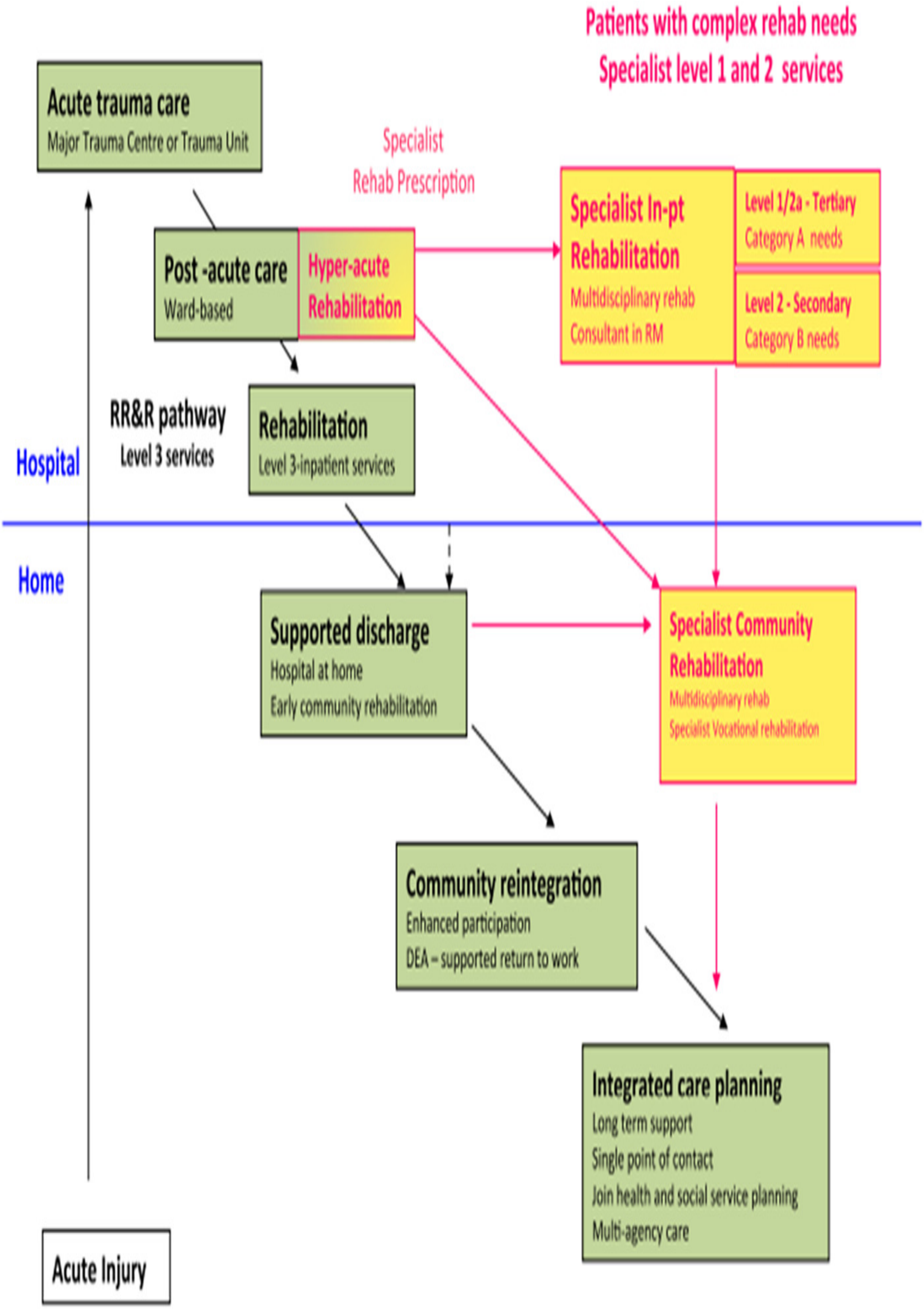
A higher level of deprivation appears as a risk factor for TBI in Kent and Medway, both in terms of incidence (by initial admission) and for subsequent readmission.

Male sex and older age both appear as risk factors for TBI in Kent and Medway.

West Kent appears to have a significantly higher rate of TBI than the rest of Kent and Medway.

White British people count for the majority of patients with TBI, but the incidence is highest in ethnic minorities in Kent and Medway.

East Kent has an established in-patient Neuro-rehabilitation Service, which provides specialist multi-disciplinary care to people diagnosed with TBI. Following treatment, individuals are discharged and referred to community services for postdischarge support. However, the postdischarge community pathway (Figure 1) identified in the rehabilitation care standards[10] lacks definition, and there are limited resources and capacity to meet the ongoing rehabilitation needs of TBI survivors after discharge.



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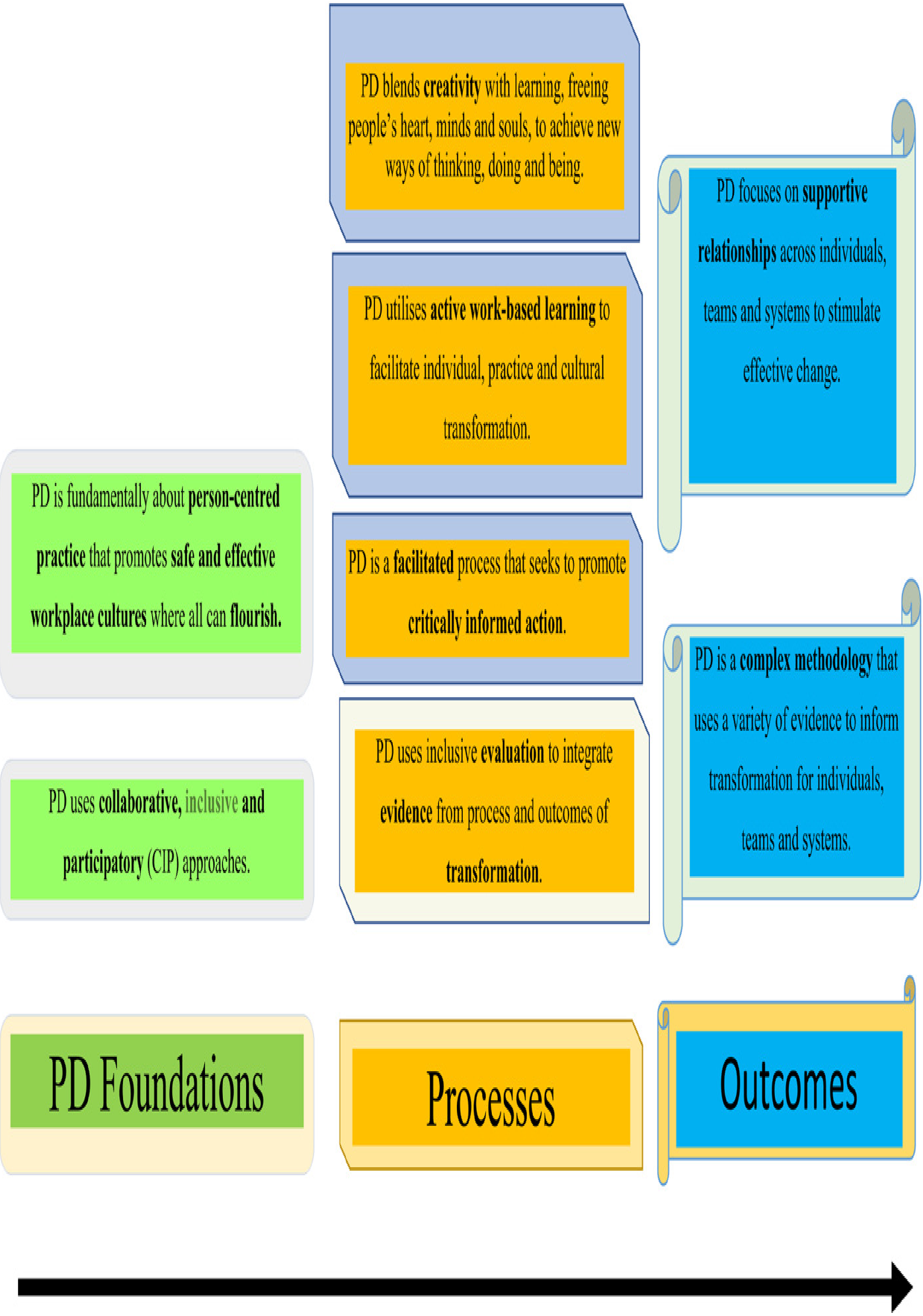
The newly created ICS for Kent and Medway in England has inherited 10 years of development work under the auspices of the Kent Brain Injury Forum (KBIF), a vibrant community of people living with TBI, and their families and carers (experts by experience). These groups have been supported by three charities, all of which have regularly come together to exchange best practices at annual brain injury conferences organized and chaired by the Service Director of the Neuro-rehabilitation Unit. Key themes emerging from these meetings catalysed an initial research study[2] to gain insight into the experiences of people diagnosed with severe TBI, who were being discharged home from the local in-patient service. Insights were also gained from their carers during the first-month post-discharge. This study concluded that patients and carers struggled to say what meaningful support had been given for transition into community living. It reported that, following discharge, there were new unanticipated needs that remained unresolved. This study also confirmed that patients and carers require further support in the longer term after the first month postdischarge in the transitional period.

In light of these postdischarge difficulties and the increasing incidence of TBI in Kent and Medway, the Neuro-rehabilitation Service Director in East Kent decided to convene a consultation with service providers and users at a stakeholder event in the House of Commons in October 2018.[11] The overarching aims of the event were to (1) raise awareness of the needs of TBI survivors and of the importance of specialist rehabilitation and care services, (2) engage diverse stakeholders across all sectors and (3) expand the East Kent service to include unmet TBI needs in Medway and West Kent (this predated both the COVID-19 pandemic and the new ICS structures introduced in July 2022). This event comprised 31 attendees, including 6 experts by experience (1 individual living with TBI and 5 family members who were carers). One of the family members was a mother of a TBI patient and an active member of the KBIF charity at that time. Also in attendance were two member of parliament (MP)s: the Chairman of the All-Party Parliamentary Group on Brain Injury Care service development, and a local MP.

After this launch event, an interactive follow-up co-production workshop was organized to explore ways of creating a more effective care pathway that was based on a better understanding of what matters to people who have experienced a TBI, their families and carers. Here we describe the insights and recommendations drawn from that workshop.

Methodology

The interactive workshop adopted a ‘practice development’ methodology, which is detailed in Figure 2.


PD is fundamentally about **person-centred practice** that promotes **safe and effective workplace cultures** where all can **flourish.**

PD uses **collaborative,** inclusive **and participatory** (CIP) approaches.

PD blends **creativity** with learning, freeing people's heart, minds and souls, to achieve new ways of thinking, doing and being.

PD utilises **active work-based learning** to facilitate individual, practice and cultural transformation.

PD is a **facilitated** process that seeks to promote **critically informed action.**

PD uses inclusive **evaluation** to integrate **evidence** from process and outcomes of **transformation.**

PD focuses on **supportive relationships** across individuals, teams and systems to stimulate effective change.

PD is a **complex methodology** that uses a variety of evidence to inform transformation for individuals, teams and systems.

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PD is a useful tool for formulating new innovations and services and is conducted 'with' rather than 'on' people. In this instance, PD was used to co-create the purpose, direction, activities and outcomes to inform how TBI services across this ICS should be formulated. PD was selected to guide the initiative because of (1) Its underpinning values—the provision of seamless, person-centred, safe and effective care across communities with continuity, enabling everyone (providers and recipients of care/services) to thrive and flourish; and (2) Ways of working that are collaborative, inclusive and participative to co-create outputs with stakeholders and service users enabling active engagement and ownership drawing on the expertise and experiences of all.

PD has much in common with the concept of 'co-production'. While there is no single agreed consensual definition of co-production in the literature, Langley et al.[12] suggest that 'co-production' can be seen as a way of working with people as 'knowledge users'[12] (p. 112) while Kothari et al.[13] define co-production as 'a model of collaborative research that explicitly responds to knowledge user needs in order to produce research findings that are useful, useable and used' (p. 1). In 2018, the NIHR published guidance on how to co-produce a research project[14] which identified key principles and features, whilst acknowledging that there is no 'one size fits all approach' (p. 5). The five key principles identified relate to power sharing, inclusivity of perspectives, relationship building, respecting and valuing all participant contributions and gaining benefits from working together. These principles were all integral to the way that the workshop was conducted on the day and served to enhance the PD methodology used.

METHOD

A wide range of diverse stakeholders was invited to take part in a 3-h workshop. These were all connected to the East Kent Neuro-rehabilitation Service, and also included representation from key partner agencies and charities across the acute and rehabilitation care pathway and system. Three key charities (KBIF, Kent MS Therapy Centre and Headway) attended along with a carer representative, expert clinicians, managers and a neuro-rehabilitation commissioner, all of whom had been unable to participate in previous meetings. The workshop was co-facilitated by the Neuro-rehabilitation Service Director and a Professor in Practice Development and was coordinated and supported by a core stakeholder group. Twenty-five individuals participated overall (Table 1).

Table 1 Stakeholder representation from across Kent and Medway in the workshop.

Total number of participants	Clinicians	Family carers	Secondary care managers	Charity representatives	Commissioners	Researchers	Legal representative solicitor
25	10	1	7	3	2	1	1

The event was intentionally organized on a central hospital site to enable those hospital clinicians to participate who had been unable to leave the hospital to attend the initial launch event in London. The PD processes used in the workshop are summarized in Table 2.

Table 2 Practice development methods, workshop processes and outputs.

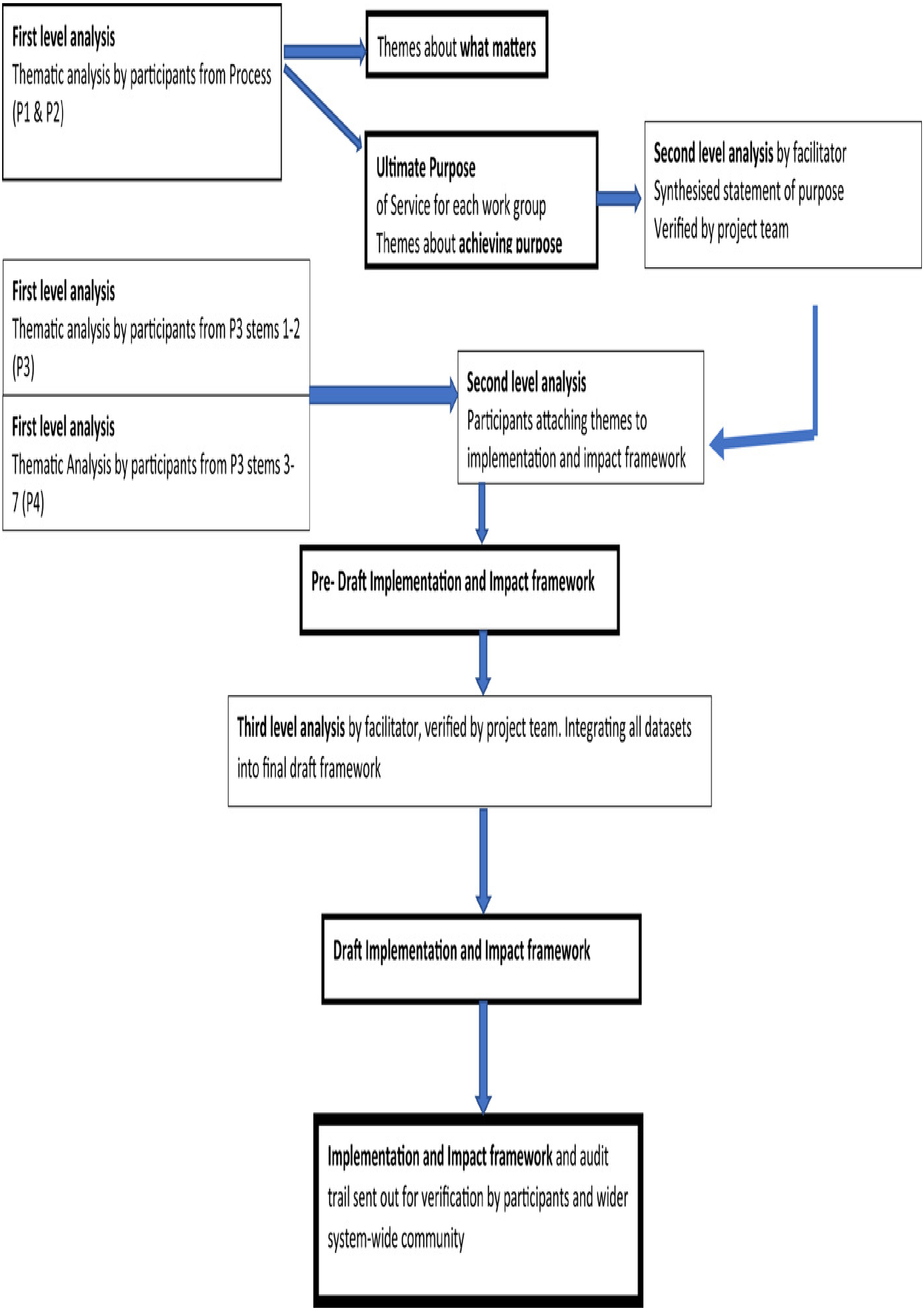
Process 1 commenced before the workshop to enable participants to take an informed decision to participate, and to know both what to expect and the preparation required. What mattered to participants in relation to services for people with TBI would be an important starting point guided by participants' experiences, values and expertise. Participants needed to know that their voice mattered, and this would be enabled through agreed ground rules to enable openness, honesty, creativity and learning for shared mutual understanding. Outputs from the workshop would be synthesized from group activity collaboratively and would not be linked to any one individual or organization. Whilst the workshop was under the umbrella of collaborative service improvement rather than research, the need for a safe psychological environment was essential to enable everyone's voice to be heard. Anonymity for participants was protected in relation to the data arising, which was group-level data, not individual data.

Process 2 aimed to distil what mattered to participants about their experiences of receiving or providing TBI services and would help identify learning from past successes, challenges, obstacles and opportunities paving the way for a formal values clarification exercise. Participants were self-selected to a chosen group focusing on either successes, challenges, obstacles or opportunities and distilled 3-4 common themes from their individual experiences.

Process 3 involved participants working together in premixed stakeholder groups to complete a values clarification exercise to identify the shared purpose and ways of working, enablers, key system, and service features, and expected outcomes. Small group work with mixed stakeholder participants was intended to endorse a safe space for different perspectives, with everyone's voice expected to be heard. Everyone's contribution was verbally shared and captured on ‘post-it’ notes.

Collaborative analysis (Process 4) achieved by clustering the post-it notes from the previous activities was shared across the four workshop groups. Participants were asked to identify common themes, use the language of participants to describe themes, and clarify meaning where this was not clear rather than making their own interpretations.

The themes generated collectively were then allocated to an implementation and impact framework by participants (Process 5). This framework comprised three parts—the enablers required to support system transformation and improved services for people with TBI; the activities or attributes that would characterize what the system and the pathway would be doing when caring for people in Kent and Medway with TBI, and lastly, the anticipated impact that would guide the evaluation of the system's effectiveness. Figure 3 describes the analysis process to explain how the results were generated.



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Process 6 involved a creative exercise to help participants to think ‘outside the box’ and to envisage a new future for the service, which could be developed in future sessions.

The final workforce process (P7) focused on creative evaluation, an important part of PD to capture both collaborative learning and how the workshop was experienced in relation to the collaborative, inclusive and participative space intended.

RESULTS

The interactive processes between individuals and groups within this workshop, which can be described as co-production (Langley et al.[12]), enabled participants to identify and share core priorities and enabled the development of an implementation and impact framework to guide integrated services for people with TBI at two levels:

- (1) at the micro level—which refers to the frontline teams interfacing directly with people living with TBI across the care pathway;
- (2) at the macro level, which identifies what the ICS needs to do to support an improved care pathway.

The key values and priorities that were identified included:

- (1) Restoring the quality of life after brain injury with stories that tell this;
- (2) Involving families to ensure support is provided for all;
- (3) Collective objective-setting;
- (4) Integrated services across all parts of the pathway;
- (5) Funding to achieve values and meet demand.

The shared purpose that emerged from the four mixed working groups to guide the transformation of services (see Box 2) was:

To enable people to optimise their rehabilitation potential and return to a meaningful life, maximising their quality, participation and independence

2:**BoxSynthesizing the ultimate purpose for care for people with traumatic brain injury from the four workshops group purposes**

Synthesized shared purpose	Groups 1–4 agreed ultimate purposes ×4
Ultimate purpose is: *To enable people to optimize their rehab potential and return to a meaningful life, maximizing their quality, participation and independence*	To enable people to achieve their optimal rehab potential and long-term quality of life

	To maximize access and opportunity to enable the patient to achieve optimum function and participation and support families and wider services in this journey
	Reducing dependence and maximizing quality of life
	To return to a meaningful life

Two interrelated implementation and impact frameworks were developed from themes generated at the workshop for the purpose of identifying the enablers, activities and indicators for evaluating impact with a specific focus on (1) individuals with TBI and their direct care teams, (2) the broader ICS that supports the TBI pathway.

The implementation and impact framework summarized in Table 3 focuses on what participants feel is needed by people with TBI, their carers and families at the micro-system level and across the pathway. The framework identifies the key activities that the service needs to focus on for those directly experiencing it, namely: engaging people actively about what matters in partnership with those providing it; delivering ongoing holistic care and support and flexible patient pathways that include active follow-up. The enablers include collective values and behaviours that support people with the activities identified from the perspective of both those providing and experiencing care; case management, team leadership with the integrated team working; workforce capabilities and evidence-based practice across the pathway; time and support for staff to be responsive. The emergent impact is expected to improve patient participation and safety, as well as the wider quality of life and independence.

Table 3 An implementation and impact framework for people following traumatic brain injury at the microsystems level and the teams providing direct care.

The system-level enablers and activities required to support the pathway and the micro-system level are presented in Table 4. Key enablers include systems leadership to drive integration, workforce development, a learning culture that supports education and research and stakeholder commissioning for a joined-up pathway.

Table 4 A system-wide implementation and impact framework for supporting people and workforce teams caring for people following traumatic brain injury across place.

Enablers	Activities/attributes	Outcomes/impact

Systems leadership that drives integration •(1) Effective leadership that integrates team experts with ‘joined-up’ working across the whole pathway and patient journey, for example, specialist services input to include neuropsychiatry and neurobehavioral/neuropsychological expertise •(2) Multidisciplinary interagency and interprofessional working, sharing and supporting across all health and social care sectors *Workforce development and resources* •(1) Workforce development through a multiprofessional career and capability framework •(2) Resources: people with skills, time, equipment, facilities, for example, day hospital; •(3) Funding to pursue innovation and technology *A learning culture that supports education and research* •(1) A forum for learning that is resourced •(2) Education opportunities for all: individuals, partners and agencies in care •(3) A diverse research programme, comprising multiple groups and integrated into the pathway to improve both short and long-term	Creating engagement opportunities for people with emphasis on care in the community Enabling active participation in education and learning by staff, patients and the wider community Championing and celebrating people, continuous improvement and innovations, for example, improving options for appropriate supportive living in the community Using population health data for planning Broader engagement with society	*Outcome measures of performance, quality and clinical outcomes* •(1) Impact indicators for person, families and team (see Table 3) *System process indicators* •(1) Reduced length of stay •(2) Reduced admissions •(3) Re-admission rates •(4) Reduced waiting list to access services/clinics •(5) Reduced referrals to mental health, criminal justice, alcohol and drug abuse •(6) Early intervention •(7) Increased synergy across health and social care •(8) Removal of systems barriers *Learning culture* •(1) Lifelong learning for people and staff *Population* •(1) BAME population integration and pathway monitoring *Society impact* •(1) Involvement of Brain Injury volunteer organizations •(2)

patient and family outcome. These groups will also be able to make an educational contribution *Stakeholder commissioning for a joined-up pathway with*: •(1) Equity of access to services •(2) Integrated communication system •(3) Infrastructure for population health management (data) *Funding* •(1) Opportunities for establishing long-term funding across sectors and expansion of the service county-wide		Normalizing, campaigning, legislation

An example of how themes were generated to populate Tables 3 and 4 is provided in Box 3, where the enabler of ‘effective systems leadership’ identified by one workgroup is illustrated from the five individual ‘post-it’ notes that comprised the theme.

3:**BoxAn example of first-level data analysis from group written post-it notes to generate a theme title for one enabler at the systems level**

Theme identified by workshop group working with enablers	Individual post-it notes written by individuals comprising the theme
Effective leadership that integrates team experts and joined up working across whole pathway and patient journey	Team, good leadership and followship without silos Integrated team experts across the whole pathway Integrated team efforts across the whole pathway Mutual Synergy and joined up working Support across system barriers—health and social services

Key activities that the ICS needs to embrace include: creating social engagement opportunities for professional stakeholders; promoting active participation in ‘learning for all’ events; championing and celebrating continuous improvement and innovation and using population data for planning. The outcomes and impact are those most relevant to people with TBI and their families, however, staff outcomes, such as well-being and job satisfaction, were also recognized as influential on the quality of care experienced.

Evaluation of the workshops

Evaluation is vital not just to ascertain whether outcomes were achieved and how they inform continuous learning and improvement, but also to establish whether collaboration, inclusion and participation produced a genuine co-productive experience.[6] In keeping with the principles of PD to use inclusive evaluation approaches that endorse co-production that focus on what matters to people, PD also blends creativity with learning to free peoples' hearts, minds and souls to achieve new ways of thinking, doing and being (see Figure 2). Participants on their tables were encouraged to capture words that best described their experience of the workshop. Participants were then given guidance about how to structure a Zen poem, termed a Haiku, with its first line of five syllables, the second line of seven syllables and the third line of five syllables and were invited to work with their chosen words to develop a collaborative poem that expressed their combined experience. The resulting expressions are shared with the other groups, and are frequently experienced by participants as powerful, uplifting and energizing and genuinely convey their lived experience. In other contexts, creativity can also include the use of nature, mandalas, artwork, images and metaphors to express an individual or group journey and learning. The evaluation (Box 4) demonstrated a positive, collaborative and empowering experience for participants that felt friendly, respectful, stimulating and thought-provoking, and reflected achievement of the top rungs of the co-production and co-design ladder—that is, ‘doing within an equal and reciprocal partnership’.[15]

4:**BoxEvaluation poems (Haikus) to convey the experience of each work group**

‘We feel empowered Stimulated, united Curious—next steps?’	‘Creative, friendly Mutually respectful Inspirational’
‘Collaborative Thought-provoking &well-led Positivity’	‘Expansive interest Friendly collaboration Welcoming, Mixing’

DISCUSSION

The shared direction and purpose co-created from the workshop were to find better ways to ‘optimise rehabilitation potential and return to a meaningful life, maximizing their quality, participation and independence’.

Developing a shared purpose is the starting point when embarking on a journey of collaborative transformation, major change and sustainable improvement,[16] but its achievement requires shared meaning at all levels of the system and genuine engagement with people and their families about what matters to them.[7]

Our workshop highlighted several core principles of care pathway improvement:

- (1)
  Focus on powerful emotions for good[17];

- (2)
  Inform conversations by case managers and professionals about personal goals, needs, care planning and continuity of care, identified as lacking in previous research[1,2];

- (3)
  Measure what matters and what is valued[18] rather than what is mandated[19] and

- (4)
 Inform continuous learning and improvement.[18,20]

Facilitating and supporting emotional work with people is a prerequisite for compassionate person-centred care, which in turn requires team members to have emotional intelligence, and access to the emotional support necessary for sustaining staff wellbeing.[21] A body of powerful stories can convey the real impact to inform learning, improvement and research supporting transformation by using evaluation approaches that focus on what works or does not work, and for whom in what contexts.[22]

There is a need for improved continuity of care across pathways with flexible follow-up and access to support contacts, and better communication.[1,2] Case managers would be pivotal to enable continuity and improvement of care by ensuring that the patient and carer are at the centre of service provision.[2] Support with meeting the complex care needs of people with TBI is required not just for carers and families, but also by general practice, and community care partners involved in care management across the system.[2]

The UK Brain Injury charity, Headway, defines a case manager as being ‘responsible for overseeing and managing the overall care of the person with a brain injury. They will prepare an individually tailored care plan or treatment programme for and with each client, which is designed to meet the person's specific health, social and emotional needs’.[23]

Whilst Headway recognizes that case managers come from different professional backgrounds, they assert that in the United Kingdom, they are only usually available through private referrals and interim compensation payments. By contrast, in Australia, a community-based case management system exists[24] providing consistent direct, holistic, client-centred services, with decision-making directed equally by staff and clients. That said, the healthcare system in Australia is funded differently and is based on healthcare insurance paid by Australian taxpayers. The challenge across the United Kingdom would be to consider how an ICS could potentially accommodate and support the introduction of such a role.

Mindful of Berwick's[18] plea in the context of the third era of healthcare, ‘to measure only what matters, and mainly for learning’ (p. 1239), demonstrating positive impact and progress towards achieving person- and family-centred goals, therefore, require indicators and measures that reflect the needs of people experiencing TBI.

Whilst broad performance indicators linked to experiences of care are routinely collected for services in England, such as the Friends and Family Test and patient satisfaction surveys, little effort is put into using this data to support service improvement.[19] The professional skills training required to help teams and systems take forward any insights gained from these data are also often missing, as are the governance systems that integrate learning with improvement and research at the meso and macro system levels.[22]

There is a large range of standardized outcome measure indicator scales which are currently used by medical and health care professionals within the clinical practice to establish individual baseline performance and measure change over time, for example, the Glasgow Coma Scale[25] and Modified Ashworth Scale.[26] These measures are scientific instruments with associated psychometric properties (validity, reliability and responsiveness), which are used to support and guide treatment interventions and also to enable individuals with TBI and their families to better understand associated change and progress, which in turn can serve to support and encourage motivation and engagement in the recovery process. Given that the majority of outcome measures have been developed and designed by clinicians in partnership with academics, the current study underlines the need to co-produce additional outcome measures to ensure that the needs of people with TBI (and their carers) are properly assessed.

Together with the framework articulated in Tables 3 and 4, these core principles highlight action priorities at the care interface that:

- (1)
  Use and embed emotional touchpoints[17] with other measures that support learning, enable improvement and celebration, as well as identify the strategies that work, using co-production approaches;
- (2)
  Consider how people living with TBI and their families can be potentially supported to contribute to improving the care pathway for people post-TBI in the community;
- (3)
  Consider how people living with TBI and their families can help co-design relevant outcome measures for use across the care pathway.

Embedding what matters at the microsystem level through using tools derived from experience-based design, such as emotional touchpoints, provides deep emotional insights when used in everyday practice.[17] Emotional support has been identified as a key unmet need for both people with TBI and their carers[2] and this tool can be tailored to people following TBI, their carers and families to explore both positive and negative emotions associated with unmet needs across pathways as well as successes.[17] Emotional touch points focus on emotional challenges related to different points across the pathway experienced by people as they cope with a disability, enabling both catharsis and a focus on the emotional support required.[1,2] Positive emotions can also be shared with those at earlier stages in their rehabilitation journey to inspire hope and share strategies that work.

Beyond the care interface, the workshop also concluded that the underpinning health and social care system needs to support frontline teams through the professional development and sustenance of high-quality multidisciplinary professional teams with capabilities wrapped around individual citizens rather than the professions. This is quite a challenge for the NHS, which is founded and based on individual professions which are regulated by independent regulatory organizations. Legislative barriers exist that specifically define the scope of practice within which a medical and healthcare professional can legitimately and professionally work, so attempting to change roles or practice would require allied legal change. The concept of ‘trans-disciplinarity’, often used to describe professionals working across traditional boundaries, is poorly defined in research, although Van Bewer[27] asserts that it involves ‘sharing of knowledge, skills and decision-making with a focus on real-world problems’ (p. 346). Given the importance of the patient and family within healthcare, we suggest that the transdisciplinary healthcare team should include individual patients and related nonprofessional stakeholders.

This area of action should endeavour to:

- (1)
  Appoint systems leaders with the prerequisite skill set to ensure a joined-up approach across ICS and TBI pathways;
- (2)
  Develop a multiprofessional capabilities and career framework (including case management) around each individual's TBI journey;
- (3)
  Create further opportunities for genuine engagement approaches with people and communities through co-production;

- (4)
  Enable the co-evaluation of person-centred, safe and effective resources and services by creating a learning culture across the system at every level, inclusive of all.

System levers and enablers required to support pathways for people with TBI include systems leadership[28] and workforce development,[29] a place-based learning culture[30] and stakeholder commissioning with indicators of system effectiveness that enable resources (people and infrastructure) to be used for optimal effect across communities. Workforce transformation is essential for supporting integration across the system, and three enablers have been recognized, namely (1) Systems leaders with the skills to break down barriers and organizational silos; (2) Facilitation expertise to draw on the workplace as the main resource for learning, developing and improving and (3) Recognition of the need to wrap capabilities around the person, citizens and communities.[28,30]

Problems with vertical integration across pathways have been identified as detrimental in the first months at home, whilst case management across the pathway for people and their carers, together with quality team leadership, can positively address this for people experiencing TBI.

Workforce enablers at the systems level include the need to grow interprofessional capabilities tailored to the person with TBI and their carers, recognizing the unique expertise that emerges from collective leadership and working.[31] This kind of approach would place the ability to collaborate and share best practices at the heart of staff recruitment and development. Developing the right staff capabilities for a quality integrated service where learning is at the heart of the system is a priority.

Developing people-centred learning cultures at every level of the system is another essential enabler[30] and, as intimated, relies on forming successful partnerships with people experiencing TBI to ensure effective ongoing service evaluation and innovation.[30] These partnerships will help move from a deficit model focused on learning from safety incidents towards one that values everyone's contributions in relation to what matters. This involves growing carers as facilitators of change[31] and increasing the availability of relevant placements and career progression opportunities to develop the practice of health and social care professionals.[30] These learning cultures require skilled facilitators who can foster an approach to learning that recognizes the financial uncertainty and ambiguity within which the cultures must nevertheless flourish.[6,29,30,32,33] We suggest that ICSs will know if their services are right for people experiencing TBI if the co-produced individual outcomes are met and system-level indicators show reduced lengths of stay, admissions and re-admission rates, referrals to mental health, criminal justice and substance abuse support services. Getting this right across the system would not only have a positive impact on people with TBI, their carers and families but also on communities and society more generally.

Beyond these recommendations, we suggest that it would also be useful to gain knowledge of what kinds of physical and mental health issues are experienced by people living with TBI in the much longer term. Insight into the nature of health needs experienced chronically, as opposed to soon after discharge, could further inform the framework proposed here (see Brett and colleagues[34–36]).

Strengths and limitations

We acknowledge that the implementation of the current framework and sustenance of allied values will depend on multiple factors. The NHS is a complex system within which numerous teams, services and individuals are working under constantly changing conditions, organizational climates and political influences. Together these affect the extent to which a front-line team can work in partnership with people post-TBI. The COVID-19 pandemic has changed work environments and practises and has especially affected the NHS work environment. We are also conscious that individuals who have survived TBI are a vulnerable group who may well have been impacted more negatively during the pandemic and lockdown periods. It is fortuitous that this workshop event took place prepandemic, which enabled a large group of stakeholders to take part without any undesirable health risks. We do note, however, that the workshop would perhaps have been enhanced by even greater citizen involvement. To reiterate a key point from above, we also understand that one learning outcome from the pandemic has been that there is a need to work in much greater partnership with people across the system and build collaborative working relationships. The introduction of ICSs across England provides a template through which to achieve this.

CONCLUSION

The recommendations made here formulated through a process of co-production, provide an initial framework by which the experiences of people living with TBI can be improved via system-wide transformation. Co-produced improvements are needed postdischarge at both the micro-level across pathways (to improve the experience and contribution of people with TBI at the care interface) and systems-level (to improve staff culture, recruitment and progression and increase multidisciplinary expertise that is joined-up). More broadly, the co-produced framework model that has emerged could offer a template for other neurological rehabilitation services that are likewise in need of reform.

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CONFLICT OF INTEREST STATEMENT

The authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

The anonymized group-level data collected is not publicly available as this was undertaken as a collaborative service improvement initiative where participation would be protected safely by anonymity and privacy.

DETAILS

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Provision of and trust in COVID-19 vaccines information: Perspectives of people who have had COVID-19

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ProQuest document link

ABSTRACT (ENGLISH)

Aim

The aim of this study was to understand the provision and need, quality of and trust in COVID-19 vaccines information from the perspectives of people who have had COVID-19 infection.

Method

People who have had a COVID-19 infection were approached via their general practice and invited to participate in the Nivel Corona Cohort. They completed questionnaires at baseline (Q1), and at three months (Q2). Outcome measures were based on health information-seeking behaviour, as used in the Comprehensive Model of Information Seeking. Antecedents (i.e., gender, age, education level, health literacy) were used from Q1, and one's beliefs and experiences (i.e., trust in the information and healthcare system, how applicable the information is), information carrier factors (i.e., information quality perceptions and via which sources), health-information seeking actions (i.e., decision to vaccinate and information sufficiency) and vaccination status from Q2. Data were analysed using descriptive analyses, analysis of variance tests (F-tests) and χ^2 tests with the statistical software STATA.

Results

Of the respondents (N=314), 96% were vaccinated at least once, mostly after having had the virus. Most retrieved information about COVID-19 vaccines on the website of the National Institute for Public Health and the Environment (79%), broader via the internet (56%), or from family and friends (35%). Almost all had trust in the information (89%) and healthcare system (94%). Most found the information applicable to their situation (67%). Moreover, most perceived the information as correct (71%) and did not perceive the information to be misleading (85%), while fewer people found the information reliable (59%) and clear (58%). Overall, the majority indicated that the information met their expectations to make a well-informed decision to vaccinate (89%).

Conclusion

Different characteristics of people who had COVID-19 and sought information were identified, which is important to offer tailored information. People who had COVID-19 in this study, mainly middle-aged, vaccinated and highly educated, were generally positive about the vaccines information, but overall the reliability and clarity could be improved. This is important for a high vaccination uptake, booster programs and coming pandemics.

Patient or Public Contribution

The questionnaire was reviewed by patients who had COVID-19, one of whom is a health services researcher.

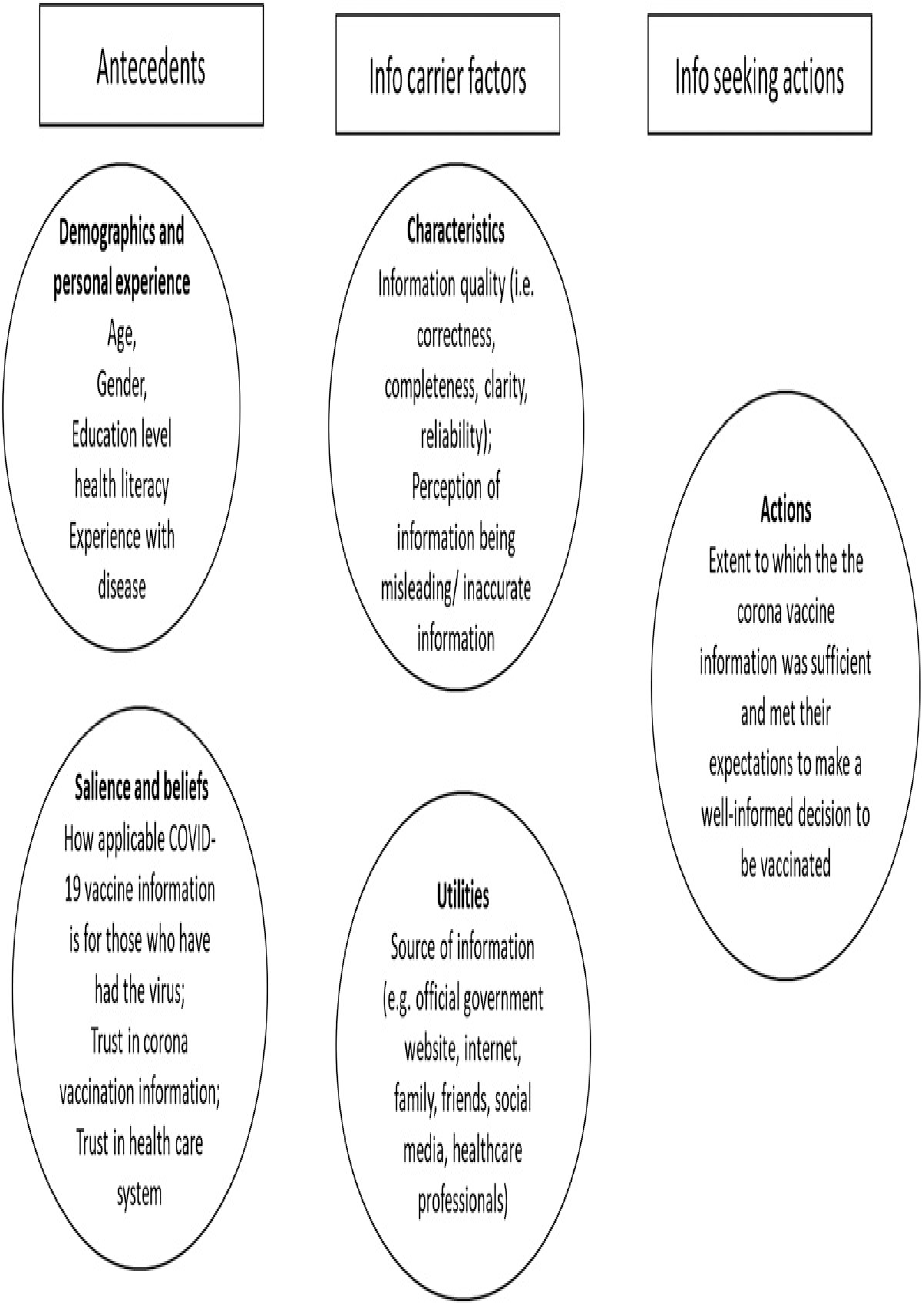
FULL TEXT

INTRODUCTION

Reliable and clear information is important for a high vaccination uptake,[1] including booster programs and pandemics. To understand the provision of vaccines information and trust therein, the coronavirus disease 2019 (COVID-19) is used as a case study. An effective way to tackle the COVID-19 pandemic is through mass vaccination globally, though acceptance of the vaccines is often a major challenge.[2] The COVID-19 pandemic is a unique situation, where vaccines were developed more rapidly than usual. In the beginning, this may have had a negative impact on the trust that people had in the developed vaccines and could, therefore, negatively influence the choice to vaccinate.[3,4] Over the course of time, people have chosen to vaccinate, though, now with the emerging strains of COVID-19 and the need for boosters,[5] provision of information and trust therein are key aspects in being well-informed to decide to vaccinate.

Specifically, the provision of COVID-19 vaccines information and trust therein amongst people who have COVID-19 is of interest. At the start of the COVID-19 pandemic, it is likely that there is a high-risk perception experienced by people as it is a new disease.[6,7] Preventative measures, such as vaccination programs, are steered by risk perception, as trends also show that uptake of the preventive measures was higher at the beginning of the pandemic than later on. As time passes, this risk perception may decrease as there is more information and experiences with the virus. People who have had COVID-19 may have a lower risk perception as they have had the virus and may be less inclined to vaccinate, or instead, they may be more keen to vaccinate as they have experienced the virus severely and want to prevent being infected again. Hence, it is of interest to investigate this group's information-seeking behaviour to tailor future vaccination campaigns, or the necessary COVID-19 boosters due to the emergence of new strains.

There are various constructs that help capture how people seek information, and to consolidate these various aspects, an adapted version of the Comprehensive Model of Information Seeking (CMIS) is used (Figure 1).[8,9]



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Factors that motivate (antecedents), one's background characteristics (demographics), how applicable or relevant the information need is (salience) and one's prior feelings regarding the need to seek information (beliefs) influence one to seek information.[8] In the case of COVID-19, trust in the information and healthcare system in relation to the intention to vaccinate[10-12] is essential. Trust in vaccines information is key because the general public is often unable to fully assess the correctness of public health recommendations.[13]

Moreover, potentially more than ever before, during a global pandemic, people wish to be well-informed and ask for more health-related information.[14] Important factors are, for example, users' perceptions of the information (characteristics). Previous studies show that people use different information sources, both offline and online, to access health-related information.[15–18] Healthcare providers are currently still the most trusted source of information, though, seeking online information via the internet has gained popularity and has become an important information source to the public.[19,20] Seeking online information allows for direct answers, readily available and (mostly) accessible information in large quantities. Though this information may not be easily comprehensible to the wide public, the trustworthiness and objectivity of the information should also be examined carefully.[21,22] Due to the newness of the virus and uncertainties about the effectiveness of the vaccines, it is interesting to see where people who have had COVID-19 seek or receive their COVID-19 vaccine information and their perceptions of the quality of this information.

Lastly, information-seeking actions such as whether one decides to vaccinate based on the sought or received information is important. Provision of information that meets the expectations of the public is key for those to decide to vaccinate, specifically trust that COVID-19 vaccines are safe and effective.[2,3,23] However, the most common reasons for hesitation or refusal to be vaccinated with the COVID-19 vaccines were fear of side effects, safety and effectiveness.[24-27] It is therefore important to understand whether the provision of the current information on the vaccines is tailored in such a way that people who have had COVID-19 decide to vaccinate.

Taking the different factors from the CMIS model into account, we investigated the characteristics of people who have had COVID-19, that sought or received COVID-19 vaccines information, where they sought the information, what they thought of the information, and which health information-seeking behaviours and actions they took. The aim of this study was to understand the provision and need, quality of and trust in COVID-19 vaccines information from the perspectives of people who have had COVID-19 infection. This is a unique target group as their views on the vaccines may be different than those who have not had the virus.

METHODSStudy design

Nivel (Netherlands Institute for Health Services Research) has set up a cohort of people who have had COVID-19 to provide insight into the course, severity and short- and long-term consequences of COVID-19. This panel design provides for a wide range and large sample of people reflecting the heterogenous patient population in daily general practitioner (GP) care practice. In this study, we conducted a secondary analysis of the cohort data.

Setting

This study took place in the period when the Netherlands experienced the third wave, and the alpha, followed by the delta variant, were the most prevalent.[28] The participants in this study had COVID-19 in the first half of 2021. At that point in time, about two million Dutch inhabitants had been diagnosed and registered with COVID-19. Also, 32,438 people had died in the Netherlands with COVID-19, either registered or probable cause of death.[28] Further, during this time, the start of the country-wide vaccination rollout took place.

Nivel Corona Cohort recruitment process

In Table 1, the different recruitment phases of GPs and patients for the Nivel Corona Cohort are described.

Table 1 Nivel Corona Cohort recruitment process

Recruitment phase	Activity

•1. Eligible general practitioners to participate in study	In May 2020, GPs participating in Nivel Primary Care Database (Nivel-PCD) were enquired to assess feasibility of the study. In total, 90 practices had shown interest in the study after a call from the research team. In February 2021, a selection of 25 GPs were invited via e-mail to participate in this study. GPs were selected based on completeness of morbidity data in 2019, having delivered weekly data in 2020, using R83.03 to code COVID-19 cases and having sufficient COVID-19 cases.
•2. Recruiting general practitioners to participate in study	Of the25 GPs invited to participate in the Nivel Corona Cohort, 18 practices participated. The selected GPs showed interest in participation, had sufficient quality and completeness of routinely registered electronic health record (EHR) data and had a sufficient number of COVID-19 registrations.
•3. Selecting patients from general practitioners to participate in study	People in the Nivel Corona Cohort were recruited from GPs that participate in the Nivel-PCD and could only participate after invitation. Nivel-PCD collects EHR data from around 500 GPs spread throughout the Netherlands. Nivel-PCD receives data on a weekly basis from approximately 350 practices, with more than one million listed people, allowing to identify prevalent and incident COVID-19 cases. Data in Nivel-PCD is pseudonymized at the GP, Nivel does not receive directly identifying data such as name or address.[29]
•4. Diagnosis of COVID-19 for an individual patient	In the Netherlands, GPs use the International Classification of Primary Care to code the complaints and diseases their patients present to them. People with COVID-19 were detected based on ICPC-1 code R83.03 (COVID-19). This ICPC-code was introduced by the Dutch College of General Practitioners to register COVID-19 as of November 2020. The diagnosis of COVID-19 for an individual patient could be in their EHR when the patient consulted their GP directly, or when the patient contacted the Municipal Health services (GGD), who provided the national testing facilities. The GGD sent information on positive tests to GPs via automated coupling, using the R83.03 code, under the requirement that patients gave consent.

•5. Generation of study pseudonym for each patient	A study pseudonym was generated for each patient, that allows for data linkage between Nivel-PCD and the patient's filled in questionnaires. In Nivel-PCD it is possible, but only via a trusted third party, to link the pseudonyms with a patient identification number that is known only in the practices' domain.[29] This allowed us to initially flag eligible people for the Nivel Corona Cohort and to let GPs subsequently conduct a check on whether they were indeed eligible for participation.
•6. Inclusion/exclusion of patients	People were excluded from this study if the GP indicated that the patient was not eligible to participate (i.e., due to the burden of filling in the questionnaire for this person or cognitive or personal problems hindering participation, or due to not being proficient in the Dutch language).
•7. Invitation of patient through trusted third party	The GP provided names and addresses of suitable people to the trusted third party who invited people on behalf of their practice. The invitation contained the patient's study pseudonym.

Abbreviation: COVID-19, coronavirus disease 2019.

Participants

If a patient decided to participate, they were asked to register online with the study pseudonym. During the registration process, the patient was asked for informed consent for study participation and data linkage. Once registered, the patient received invitations per e-mail to questionnaires at enrolment and after three months.

Person inclusion for the Nivel Corona Cohort started in April 2021, and people were recruited both retrospectively and prospectively. At the start of the study, the GP invited all people with a known COVID-19 infection in the past six weeks, and then every two weeks after the start of the study, newly diagnosed people were invited to participate.

Data collection

For this particular study, we used data from the two first online questionnaires sent to people in the Nivel Corona Cohort (Q1, the start of participation and Q2, at three months). Since we used data from both questionnaires (Q1 and Q2), we only included the people in this study who filled out both the first and second questionnaires. A reminder was sent after one week. People who did not respond or refused to participate in the first questionnaire were marked in the database and were not approached for further participation. We used data from people who filled in their second questionnaire (Q2) before the 28th of January 2022. All data were stored on Nivel's protected server. The questionnaire data was pseudonymized.

See Supporting Information: Appendix 1, for an overview of the questionnaire topics, outcome measures and types of responses possible, based on the CMIS. See Table 2 for the constructs of the adapted CMIS to fit the context of COVID-19 vaccine information-seeking behaviour.

Table 2 Study constructs in the context of the CMIS

CMIS construct	Description of measures
Antecedents	Background characteristics (Q1 questionnaire), first moment a participant filled in the questionnaire, were used.

	•(1) gender •(2) age •(3) level of education (low, middle, high)
	The categorization of the education level is in accordance with Statistics Netherlands (Low: primary education, prevocational secondary education (VMBO), Middle: senior general secondary education (HAVO), pre-university education (VWO), senior secondary vocational education (MBO); High: higher vocational education (HBO) and university education (WO).[30] In Q2, second filled in questionnaire, questions on health literacy (based on the Chew's Set of Brief Screening questions [SBSQ])[31–33] were posed.
	Personal relevance factors (salience and beliefs) posed:
	•(1) the degree in which people have trust in the sought/received information as well as trust in healthcare system. •(2) how applicable or relevant the vaccines information was for them.
Information carrier factors	In the Q2 questionnaire, we collected specific information on the COVID-19 vaccines information. All questions pertaining to COVID-19 vaccines information included the information people actively sought themselves as well as the information they received from healthcare providers or other source. These two forms of receiving information (actively searching and passively receiving) are not split up in the questions asked.
	These questions included:
	•(1) perceived quality of information, •(2) whether the information was misleading or inaccurate, •(3) type of information sources used to search for information on the COVID-19 vaccines.
	See Supporting Information: Appendix 1, for the types of responses.

Information seeking action	Based on whether the people who have had COVID-19 sought or received information (and which information), we asked whether the information met their expectations to make a choice to vaccinate (yes/no), and if no, an open question was posed about what they would have desired regarding the information provision.
Overall process*	The original CMIS shows the information-seeking process as a linear process. The arrows, from left-to-right, in this model suggesting information seeking as a process that follows the factors from left-to-right. However, one can start and stop searching at different points based on beliefs and perceptions that may change over time. Hence, in the adapted model in this study (Figure 1), there are no arrows, but instead the factors presented with each important CMIS theme (antecedents, information carriers and actions).

Abbreviations: CMIS, Comprehensive Model of Information Seeking; COVID-19, coronavirus disease 2019.

Data analysis

We used descriptive analyses (frequencies [*N*, %], means [SD]) to describe the population and outcomes. For the personal factors, we looked at differences in age, gender, level of education, health literacy, vaccination status, (in)sufficient trust in the COVID-19 vaccination information, and trust in the healthcare system in relation to the outcomes. The outcomes included were: whether (and where) the person sought/received information, perceptions of information quality, whether the information was misleading or inaccurate, and whether the information was sufficient/met the person's expectations to make a well-informed decision to vaccinate.

The differences were analysed using analysis of variance tests (F-tests) and χ^2 tests. We chose group comparisons as this is an explorative study looking at different facets of information-seeking behaviour and how this differs amongst different types of COVID-19 people (e.g., younger vs. older people, higher educated vs. lower educated). Tukey post hoc tests revealed the difference in means in the groups of respondents with different background characteristics. A significance level of $p < .05$ was used. We used the statistical software STATA version 16 for the analysis of the questionnaires.

RESULTS

We invited 1851 people for the Nivel Corona Cohort. In total, 442 people filled in the Q1 questionnaire, at the start of their participation in this study. Of these respondents, 314 also filled in the Q2 questionnaire (net response rate 70%).

COVID-19 exposure and vaccination

All people have had COVID-19, of which many (58%) had the virus one–three months before filling in the Q1 questionnaire. The majority (89%) of the respondents received the COVID-19 vaccines after they had COVID-19. Over the period of three months (Q1 and Q2), there was an increase in the number of respondents vaccinated with at least one vaccine (68%–96%). An overview of sample characteristics is reported in Table 3.

Table 3 Background characteristics of sample population

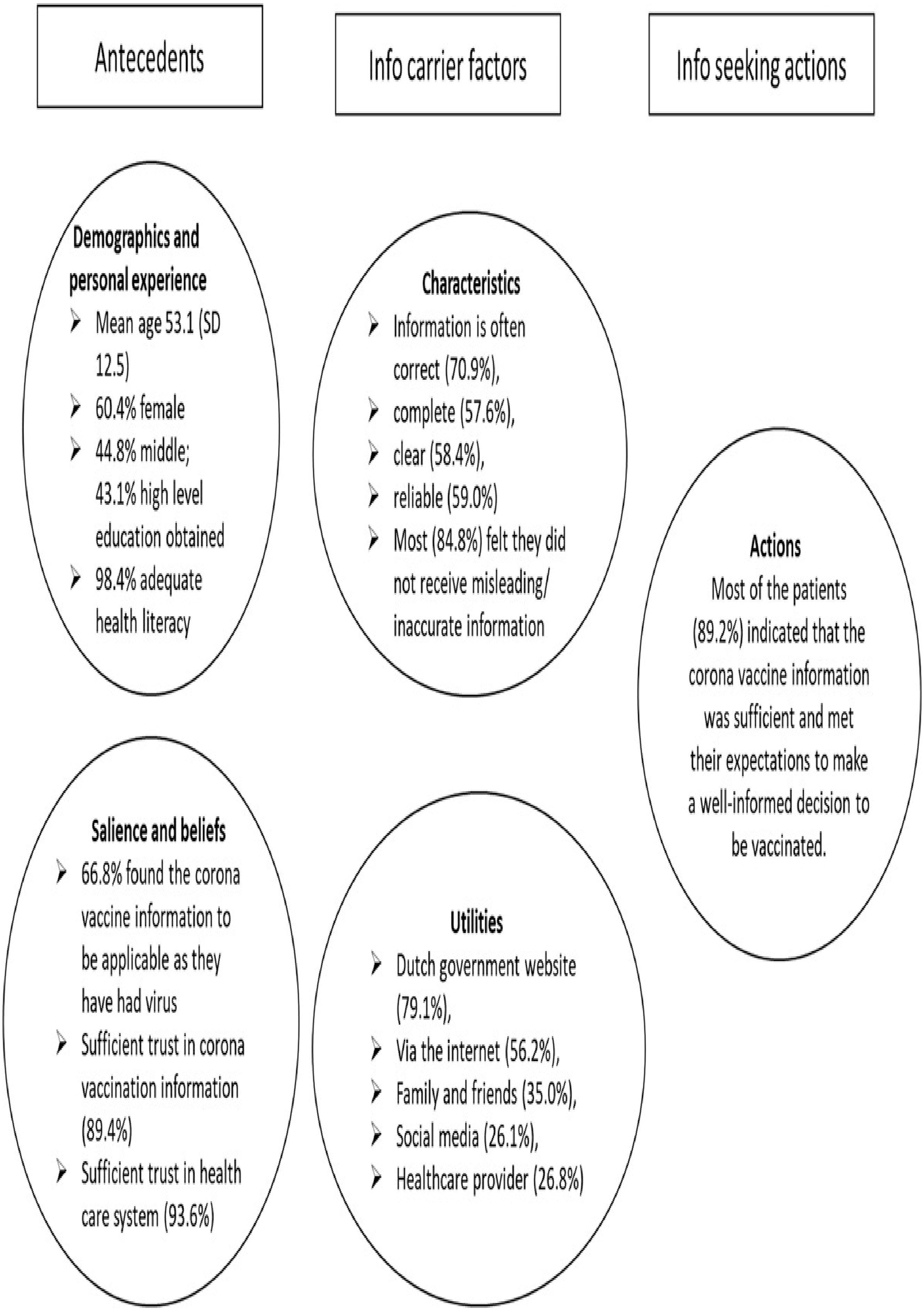
Characteristics	*N* (%)
Age (*N* = 311) (years)	
<40	33 (11)
40–64	234 (75)

≥65	44 (14)
Gender (*N* = 314)	
Male	118 (38)
Female	196 (62)
Country of birth (*N* = 307)	
The Netherlands	292 (96)
Other	15 (4)
Education (*N* = 290)	
Low	35 (12)
Middle	130 (45)
High	125 (43)
Health literacy score (*N* = 314)	
Adequate health literacy	309 (98)
Inadequate health literacy	5 (2)
Vaccination status (*N* = 311)	
Vaccinated with at least one vaccine	298 (96)
Not (yet) vaccinated	13 (4)
Trust in COVID-19 vaccines information (*N* = 274)	
Sufficient	245 (89)
Insufficient	29 (11)
Trust in healthcare system (*N* = 297)	
Sufficient	278 (94)
Insufficient	19 (6)

Abbreviation: COVID-19, coronavirus disease 2019.

Adapted CMIS for COVID-vaccines related information

The factors that influence information-seeking behaviour of people with COVID-19 selected in this study are presented according to the CMIS, as shown in Figure 2.



Demographics and personal experience

- Mean age 53.1 (SD 12.5)
- 60.4% female
- 44.8% middle; 43.1% high level education obtained
- 98.4% adequate health literacy

Characteristics

- Information is often correct (70.9%),
- complete (57.6%),
- clear (58.4%),
- reliable (59.0%)
- Most (84.8%) felt they did not receive misleading/ inaccurate information

Actions

Most of the patients (89.2%) indicated that the corona vaccine information was sufficient and met their expectations to make a well-informed decision to be vaccinated.

Salience and beliefs

- 66.8% found the corona vaccine information to be applicable as they have had virus
- Sufficient trust in corona vaccination information (89.4%)
- Sufficient trust in health care system (93.6%)

Utilities

- Dutch government website (79.1%),
- Via the internet (56.2%),
- Family and friends (35.0%),
- Social media (26.1%),
- Healthcare provider (26.8%)

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Antecedents

Most people were female (62%), obtained a middle (45%) or high level (43%) of education, and were middle-aged (mean: 53.1, SD: 12.5). The majority had a self-perceived adequate health literacy (98%), implying that the role of health literacy cannot further be analysed as the number of respondents with an inadequate health literacy score was too small (Figure 2, *antecedents*).

Personal relevance factors (salience and beliefs)

About two-thirds of the people (67%) found the information provided or sought on the COVID-19 vaccines to be applicable to them as they have had the COVID-19 virus, specifically people who have more trust in the information (*p* <.0001) and those with more trust in the healthcare system (*p* <.0001).

Moreover, the respondents who indicated that the information was applicable to them mainly indicated in an open question that the information they sought was complete, clear and accurate. They also indicated that the information was applicable to their situation as they had only needed one vaccination as they are people who have had COVID-19. Those who did not think the information was applicable to them indicated in an open question that the information was too general and could be applicable to all people (not specific to those who have had the virus, that is, when to vaccinate after the recovery period), that the side effects, long-term effects and effect of natural immunity against the virus were not clearly indicated as much information had not been based on scientific evidence, and/or that some information is contradictory (i.e., one or two vaccines necessary for people who have had COVID-19).

Lastly, the majority of the respondents (89%) had a sufficient amount of trust in the information on the COVID-19 vaccines and trust in the healthcare system (94% of respondents).

Information carrier factorsPerceptions on information quality

Respondents were asked to assess the quality of the COVID-19 vaccines information (Table 4). The majority, 71%, of the respondents often found the information to be correct. Fewer respondents often found the information reliable (59%) or clear (58%). More than half (58%) indicated that the information was often complete, and two-thirds of the respondents, 67%, sometimes found the information to be contradictory. Also, respondents were inquired how applicable they perceived the information to be for their situation. Less than half, 41%, indicated this to be the case.

Table 4 People who had COVID-19 perceptions on the information they received/sought about the COVID-19 vaccines

	Always	Often	Sometimes	Never
The information is correct (*N* = 295), *N* (%)	23 (8)	209 (71)	63 (21)	-
The information is complete (*N* = 297), *N* (%)	18 (6)	171 (58)	101 (34)	7 (2)
The information is clear (*N* = 298), *N* (%)	27 (9)	174 (58)	93 (31)	4 (1)
The information is reliable (*N* = 300), *N* (%)	37 (9)	177 (59)	94 (31)	2 (1)
The information is up to date (*N* = 299), *N* (%)	26 (9)	177 (59)	91 (30)	5 (2)
The information applies to me (*N* = 298), *N* (%)	13 (4)	122 (41)	155 (52)	8 (3)
The amount of information is just right (*N* = 298), *N* (%)	11 (4)	152 (51)	125 (42)	10 (3)
Information from different sources contradicts each (*N* = 293), *N* (%)	13 (4)	62 (21)	196 (67)	22 (8)

Abbreviation: COVID-19, coronavirus disease 2019.

Perceptions on information quality in relation to background

People with different background characteristics had varying perceptions of information quality. Primarily, higher educated people, those who had sufficient trust in the COVID-19 information and in the healthcare system, and who were younger (primarily <40 years old) were more positive about the information quality, see Supporting Information: Appendix 2.

Misinformation regarding the COVID-19 vaccines

Most (85%) of the respondents (*N* = 277) felt that they did not receive misleading or inaccurate information regarding the COVID-19 vaccines. The respondents (*N* = 42) who did find the information on the COVID-19 vaccines to be misleading or inaccurate, mentioned various reasons in an open question. People who found the information misleading or inaccurate, specifically indicated the information about the side effects (*N* = 3), (long-term) consequences, safety in use and effectiveness of the vaccines (*N* = 9), but also regarding the related deaths (*N* = 2). Also, people found the quality of the information poor (i.e., not reliable, misleading, inconsistent, contradictory, incomplete and/or not based on enough scientific evidence) (*N* = 10). Some found the information based too much on political views (*N* = 3), not suited for those who have already had the virus and the effects of natural immunity (*N* = 3) and based on conspiracies and those who are against vaccinating (*N* = 3).

Information source about the COVID-19 vaccine

The most used information source was the central Dutch site on COVID-19 vaccines information, the National Institute for Public Health and the Environment (RIVM) website (Table 5). The majority of the respondents (79%) sought or received information about the COVID-19 vaccines on this website. Additionally, respondents sought or received information on the internet via different search engines (56%), via family and friends (35%), social media (26%) and healthcare providers (27%). About one-quarter of the respondents (22%) used the Dutch website with primary care information from the GP (thuisarts.nl). Lastly, a minority of the respondents searched for information on different websites (13%).

Table 5 Information sources where respondents found information on the COVID-19 vaccine

Information source	*N* (%)
On the central Dutch site on COVID-19 vaccination information	242 (79)
On the internet via a search engine, such as Google, Bing or Yahoo	172 (56)
With family/friends/acquaintances	107 (35)
On social media	80 (26)
With a doctor or other healthcare provider	82 (27)
Other	29 (10)
On the website of Thuisarts.nl	66 (22)
On another website	39 (13)
At a hospital	27 (19)
In a patient organization	10 (3)

With a health insurer	2 (1)
I did not seek nor receive information	20 (7)

Abbreviation: COVID-19, coronavirus disease 2019.

Primarily younger, higher educated, females were more likely to seek information on the RIVM website. Generally, older people and lower educated sought information on social media. For other comparisons of where people sought or received information and their background characteristics, see Supporting Information: Appendix 3.

Those who did not seek nor receive information (7%) gave various reasons. Some indicated that they did not see the added value ($N = 2$) nor the need ($N = 5$). Others indicated that there was already enough presented in the media ($N = 1$), or specifically in the newspaper ($N = 1$). Some respondents already had enough trust in the information or in others/faith in science or had enough information ($N = 2$). Some expressed being tired of the situation ($N = 3$). Others mentioned they were already informed or their situation was clear ($N = 5$).

Information seeking actionInformation provision and decision to vaccinate

Most of the people (89%), on the whole, agreed with the question about how sufficient the information was and whether this met their expectations, and whether to make a well-informed decision to vaccinate. People with sufficient trust ($p < .0001$) in the information on the vaccines and in the healthcare system ($p < .0001$) were more likely to believe that the information provided on the COVID-19 vaccines was sufficient to make a well-informed decision to vaccinate. At the same time, about one-third (32%) indicated that the information was sometimes-to-never reliable (41%) nor clear (32%).

In case the information was not sufficient ($N = 30$), respondents gave the following points that could have been better with regard to the provision of information: more honesty/transparency, such as information based on facts and research, and the need for the potential consequences and disadvantages of the vaccines to be more openly discussed. Also, there is a desire for the pros and cons to be made more explicit. Moreover, respondents indicated that quite a lot is still unknown, and some respondents would want less emphasis on only the positive elements, and instead more transparency. Lastly, some expressed the need for more clarity and clear communication, for example, why the AstraZeneca vaccine was mainly limited to the 60–64-year-old group and what the added value is in comparison to natural immunity.

DISCUSSION

The sample population who sought or received information about the COVID-19 vaccines on a major governmental-related website for this, via the internet, or via family and friends. Most people found the information correct and did not perceive the information to be misleading, while fewer people found the information reliable and clear.

Revisiting the CMIS model, the antecedents are important factors to start the information-seeking process. These are often motivators to start the search process, which can reduce uncertainty, confirm/disprove certain beliefs or ideas, or satisfy curiosity.[34] While, most people were vaccinated with at least one vaccine at the moment of filling the second (Q2) questionnaire, we cannot conclude on the motivations to vaccinate. It is also important to note here that it is not necessarily everyone's decision to only take a vaccine after infection, but that this also has to do with the fact that vaccinations only started then. Nevertheless, we can comment on characteristic types of people who used which information sources and what their perceptions were on the (quality of the) information during their information-seeking process.

Certain types of people were more inclined to seek health information, such as women, those higher educated and younger people.[35–37] This also is in line with our sample population, primarily younger, higher educated, females and those who had sufficient trust in the healthcare system were more likely to seek information on the RIVM website, who sought information on the Dutch government website, a trustworthy and reliable website. The representativeness of the cohort was checked (unpublished manuscript, can be requested by the corresponding author). Comparisons were made between the Nivel Corona Cohort and the groups of selected and invited patients that did not participate. The Nivel Corona Cohort particularly included specific types of people, generally older and more often female.

Trust, an important antecedent, is also a motivator to seek information. A large group of the respondents had trust in the COVID-19 vaccines information and healthcare system, while about 1 in 10 found the information misleading and inaccurate. Based on the Integrative Model of Organizational, trustworthy information can be defined with traits such as technical knowledge, widely open and accessible, factual, reliable and consistent.[38] Lack of trust is also often associated with perceptions that the information is misleading or inaccurate. Also, this was shown in this study, in the partly tautological relationships (e.g., people who had less trust in the information were more likely to agree that information was misleading or inaccurate) as the three central dimensions of trust are benevolence, integrity and ability, which are related to accuracy.[38]

Moreover, during the information search process, the antecedent factors can still cause an individual to stop their search. Reasons for this may be that they become overwhelmed by the information. This is why it is important to enquire about the perceptions of information quality. Overall, the respondents found the information to be applicable and complete, whilst, there were also respondents (more than half) who found the information to be contradictory on an occasional basis, and less reliable or clear. Evaluating one's perceptions of the quality of the information is a challenge, as these are based on subjective perspectives. However, these factors do act as a starting point to further enquire about what changes the information-seekers' (or lack thereof) needs and desires in terms of patient-tailored information. For example, despite the small proportion of the respondents who found the information misleading, or not applicable to them, we can follow up on their suggestions that the information could have been more transparent and more clear. Specifically, information can be more clear on whether people who have had the virus need one or two vaccine(s), what the long-term consequences are of the vaccine and the effects (i.e., herd immunity and build-up of antibodies) of the vaccines. Evidence shows that transparent communication may harm vaccine acceptance here and there, however, transparency increases trust in health authorities.[29] On the contrary, vague and sometimes reassuring communication does not increase vaccine acceptance either. Ambiguity in communicating information may lead to lower trust and higher endorsement of the spread of misinformation.[27] In return, to vaccinate large amounts of people, it is crucial that people trust in the fact that COVID-19 vaccines are safe and effective,[2,3,23] as well as the effectiveness of boosters in the future.[5]

Lastly, information-seeking action, the majority of the study sample population indicated that the information about the COVID-19 vaccines was sufficient. Also, the information met their expectations to make a well-informed decision about whether or not to be vaccinated. For those who indicated no, we also enquired the reasons why, which in return helps to tailor the needs of people who both actively search for information and the group who prefers not to. Reasons why people do not seek information or avoid information are because information can conflict with their prior knowledge, beliefs and attitudes, or potentially causes heightened emotion such as anxiety or stress about the information.[39]

Situations where uncertainty is present can cause increased anxiety and risk perception[40,41] amongst individuals seeking healthcare/treatment. As a result, this can decrease well-informed and optimal healthcare decisions as well as avoidance behaviours.[40,41] In the case of COVID-19, this is a unique situation, because there was more uncertainty regarding how fast the vaccines were developed, and there was an increased level of infection-related uncertainty, in the context of a global pandemic.

Due to rapidly emerging vaccines and without, at the time of development, sufficient evidence as to their effectiveness and health impact,[40] this can cause uncertainty for people to vaccinate. Our study adds the perspective of people who have already had COVID-19 and their perceptions of the COVID-19 vaccines information, and whether this influences their decision to vaccinate. Their perceptions may be different to those who did not have an infection yet and choose to vaccinate as they had already had COVID-19, including views on risk-perception of the virus/need to vaccinate.

An important way to decrease the spread of the virus is by mass vaccination uptake.[2] Though, to realize this, people should be well-informed and feel confident to make a choice to vaccinate. Solely providing information about COVID-19 vaccines is not sufficient. More importantly, the information should be tailored to the needs of the people seeking vaccine information. One of those elements was whether people who already had had a COVID infection and therefore might have other questions or information needs regarding vaccination than those who did not experience an infection. To be able to tailor information, it is important to be aware of the perceptions of and the trust in the information of different groups, and our study contributes to insights into the perceptions of those who already experienced an infection. The people with COVID-19 that found the information misleading/or inaccurate indicated that this was generally due to safety and efficacy reasons related to the vaccines. This is in line with research conducted in the United Kingdom (Freeman et al.[42]) whereby people who were strongly hesitant towards the vaccine were less likely to see the collective benefit[42,43] of vaccination, and instead had more concerns about the safety and fast development of the vaccines. A way to manage uncertainty in healthcare is by communicative practices, whereby the information moderates the effect of the uncertainty.[44] Reliable and accurate information, as well as information about the safety and effectiveness of the vaccines, can decrease the uncertainty about whether to vaccinate, also in those who already had an infection.

Over time, the need for effective communication strategies for uncertain healthcare-related situations has increased, as well as the nature of uncertain situations has become more complex. Therefore, there is a need to know how to tailor information to accommodate this uncertainty.[45] The lessons learned from this study give room to further tailor information about vaccines in future pandemics or vaccination campaigns. The communication strategies (i.e., using the perceptions about the quality and how/where people seek information about vaccines sought/received in those who might already feel immunized because they experienced an infection) are important to more accurately target patients about vaccines in future vaccination campaigns, booster programs or pandemics. Based on our results, to make sure to tailor the information to their needs, respondents indicated that more honesty and transparency in the information is needed, that information is backed up by facts and research, and there is more overall clarity in the communication and information (e.g., whether people need one or two vaccine(s) if they have had the virus).

Strengths and limitations

One of the strengths of this study included the varied sample of people, reflecting the general population. We recruited people from 18 practices, and people varied in the severity of COVID-19. We also included those with mild complaints. Another strength was using participants drawn from practices, providing a well-defined population, in which COVID-19 has really been diagnosed. If not drawn from a practice, then the sample population would have consisted of self-reported diagnoses.

An important limitation is the small number of people who had not had a vaccine in the sample. In our study, we have a relatively large number of people who were vaccinated, hence we cannot draw conclusions about unvaccinated people. The moment of the questionnaire could have played a role. In hindsight, including the information on COVID-19 vaccines questions in Q1 and Q2, both before and after being vaccinated, would have been more appropriate. Now we could only report on the group of people who have primarily already been vaccinated. According to the Dutch governmental website on COVID-19 vaccination, 89% of people aged 18 and older have now had at least one COVID-19 vaccine.[46] In our sample, the mean age was 53 years old, and people over 55 or 65 remain the most receptive group to vaccination, which may explain the high vaccination rate.[26,47]

Another limitation is the small group of people with inadequate health literacy (N = 5), suggesting we could not study this sample. It is important to note that not all low-educated people have inadequate health literacy, as shown in our sample, only one person indicated inadequate self-reported health literacy and a low obtained education level. The small group of people with inadequate health literacy may be related to the ease or difficulty that people with a lower health literacy may experience when completing questionnaires. In our study, we also had a large group of people with, on average, a high education level. Amongst various potential reasons why, one particular reason why this may not have such a large role is that there is not a big difference in vaccination coverage between low, middle and highly educated people in the Netherlands. According to Statistics Netherlands, about three-fourths (77%) of the highly educated people 25 years and older indicated that they intend to vaccinate, while slightly fewer low educated (68%) and people who obtained a middle-level education (69%).[48]

Additionally, a large part of the questions is based on single-item and self-reported measures. We asked people who have had COVID-19 what they think of the COVID-19 vaccines information. The information we collected (whether something is reliable, clear or trustworthy) is someone's opinion, however, it could be that these people read misleading information and did not realize it. Our approach was to see whether people have the feeling that the information is adequate and correct, and not whether this was actually the case. For example, we did not visit the information sources to test the accuracy or the trustworthiness of the information.

Lastly, participants were not explicitly asked about the information content they sought/received. They were only asked which information source they used to seek/receive COVID-19 vaccines information and their perceptions of the information.

Clinical implications

Tailoring COVID-19 vaccines information to specific people's characteristics, and increasing clarity and transparency are important for accommodating the information needs of different types of people. Primarily younger, higher educated, females and those who had sufficient trust in the healthcare system were more likely to seek information on the Dutch government website, a reliable and trustworthy source. More attention should go out to set up ways to make the COVID-19 vaccines information provision more inclusive, for example, males, lower-educated people, and those that have less trust in the government/healthcare system. The reasons why people had (or lacked) trust in the information about vaccines and the healthcare system, as well as motives to vaccinate, could also be further investigated. Also, while we collected data on migration background, it proved that the vast majority of participants were of Dutch descent. Migration background might be an interesting aspect in relation to vaccine uptake decisions and trust in the vaccine for a future study. Potentially selecting GPs in communities with a higher population of migrants may result in a more heterogenous and diversified study population. These lessons learned can increase effective communicative strategies in future pandemics, vaccination campaigns or booster programs.

CONCLUSION

Different characteristics of people who had COVID-19 and sought information were identified, which is important to offer tailored information. Among this vaccinated, and generally higher educated, middle-aged, female population, people who had COVID-19 were generally positive about the vaccines information, but overall the reliability and clarity could be improved. Reliable and clear information is important for a high vaccination uptake; for other vaccine programs, including booster programs and coming pandemics. More research is necessary to draw conclusions on the perceptions of the COVID-19 vaccine information in the group of unvaccinated people.

AUTHOR CONTRIBUTIONS

Laura Schackmann, Karin Hek and Liset van Dijk contributed to the conceptualization and design of this project. Data analysis was led by Laura Schackmann under the supervision of Karin Hek. Data interpretation and a critical review of the results were discussed with all the authors. Laura Schackmann wrote the first draft of the paper under the supervision of Liset van Dijk. All authors contributed to reviewing and editing subsequent drafts and reviewed the final manuscript.

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CONFLICTS OF INTEREST

L. v. D., M. V. and L. S. received funding from TEVA Pharmaceuticals for a study not related to this study. L. v. D. and M. V. also received funding from AstraZeneca for a study not related to this study. The remaining authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request.

ETHICS STATEMENT

The Medical Ethics Committee (METc) of the Vrije University Academic Medical Centre (VUMC) approved the protocol (METC protocol number 2020.0709). The METc VUMC concluded that this study is not a clinical research with human subjects as meant in the Medical Research Involving Human Subjects Act (WMO). The study was also approved according to the governance code of Nivel-PCD under number: NZR-003120.081. All participants gave informed consent before starting the questionnaire. Participation in the study included giving permission to link data from the questionnaire to the Nivel-PCD data of this person. The anonymity of all respondents is guaranteed.

DETAILS

Subject:	Public health; Information retrieval; Health care; Websites; Viral diseases; Immunization; Health services; Infections; Participation; COVID-19; General practice; Health behavior; Vaccines; Trust; Reliability; COVID-19 vaccines; Tests; Health information; Coronaviruses; Decision making; Pandemics; Health literacy; Information systems; Questionnaires; Disease prevention; Statistical analysis; Variance analysis; Chi-square test; Patients; Electronic health records; Viruses; Perceptions; Information seeking behavior; Primary care; Uptake; Middle aged people; Information sources; Friendship
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Supporting the parent-to-child transfer of self-management responsibility for chronic kidney disease: A qualitative study

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ABSTRACT (ENGLISH)

Introduction

As children with long-term conditions (LTCs) mature, they are usually expected to assume responsibility from their parents for self-management of their condition. Little is known about what supports families with this handover of responsibility, including the role of healthcare professionals (HCPs). This study aimed to explore what supports young people with chronic kidney disease (CKD) to assume self-management responsibility and parents to relinquish control.

Methods

A qualitative study, using a grounded theory approach was conducted. Individual and dyadic interviews and focus groups were carried out with 16 young people aged 13–17 years old with CKD, 13 parents, and 20 HCPs. Participants were recruited from two UK children's renal units.

Findings

Building and maintaining trust, fostering positivity, learning from mistakes, forming partnerships and individualized support, facilitated the transfer of self-management responsibility. However, HCPs' focus on developing partnerships with young people meant some parents felt excluded, highlighting uncertainty around whether support should be child- or family-centred. Although tailored support was identified as critical, aspects of local service provision appeared to impact on HCPs' capacity to implement individualized approaches.

Conclusion

This study has identified what supports the handover of responsibility, and, importantly, HCPs' current, and potential role in helping young people to assume responsibility for managing their LTC. Further research is needed to explore how HCPs' involvement balances child- and family-centred care, and how HCPs can adopt personalized, strengths-based approaches to help ensure the support that families receive is tailored to their individual needs.

Patient or Public Contribution

Patient and public involvement was integrated throughout the study, with young adults with CKD and parents who had a child with CKD actively involved in the study's design and delivery.

FULL TEXT

INTRODUCTION

Self-management has become an increasingly important aspect of health care across all age groups, due to the growing prevalence of long-term conditions (LTCs).[1] Although definitions of self-management vary, Lorig and Holman[2] suggest it involves medical, role and emotional management to enable the individual ‘to manage the symptoms, treatment, physical and psychosocial consequences and lifestyle changes inherent in living with a chronic condition’ (p. 178). As children are usually dependent on, or share condition management with their parents, alternative terms such as ‘supported self-management' and ‘responsibility sharing’ have been used in childhood LTCs,[3,4] As children mature, they are expected to assume responsibility from their parents for self-management of their LTC.[5] However, this expectation has been challenged, and studies suggest that for some families, shared parent-child management is preferable to the young person managing their LTC independently.[6,7]

Healthcare policy and research focuses on adolescence and the transition between child and adult services as the main developmental phase to acquiring self-management skills.[8,9] Consequently, healthcare professionals (HCPs) tend to view the assumption of self-management responsibility as a process that starts when the young person is around 13 years old and ends with the transfer to adult services.[10] Studies suggest, however, that families can start this process at an earlier developmental stage,[6,11,12] and some guidelines recommend that HCP support to develop self-management skills should start in early childhood.[4] This uncertainty around the optimal time for children to assume responsibility is compounded by studies highlighting adolescents' difficulties engaging in self-management, resulting in adverse consequences for their health.[13] Additionally, the conflation between children's age and competency and the tendency of HCPs to view children as a homogenous group,[14,15] underlines the need for individualized support with the transfer of responsibility.

An integrative review that explored the parent-to-child transfer of self-management responsibility found that this transfer was a complex, individualized process.[16] The review identified how children and parents adopted various strategies to facilitate the transfer of responsibility, but there was limited evidence about the approaches used by HCPs and ambivalence around what was helpful. Where research explored what supported children to assume responsibility, this was primarily from the perspectives of children and parents; the views of HCPs were noticeably absent. Due to this gap in the literature, the review suggested further research was needed with all key stakeholders, including children, parents and HCPs, to gain a better understanding of the transfer process and what supports families with the handover of responsibility.

Research around the transfer of self-management responsibility has mostly focused on diabetes and asthma, two of the most prevalent childhood LTCs.[16,17] As LTCs differ in severity and self-management demands can vary, a condition-specific approach can be useful when studying the parent-to-child transfer of responsibility.[18] Therefore, this study focused on chronic kidney disease (CKD), a complex LTC related to irreversible kidney damage, with a wide range of causes and complications.[19] Children with CKD can be classified by stages 1–5, based on the rate at which the kidneys filter waste products; stage 5 indicates end-stage kidney disease, which means renal replacement therapies, such as dialysis or kidney transplantation, are needed.[20] Although CKD shares some self-management tasks with other LTCs, condition-specific demands include renal diets, fluid restrictions or targets and dialysis, either carried out in a hospital or home setting. In the United Kingdom, 13 specialist renal centres manage the care of children with CKD stages 3–5.[21] As the majority of CKD management tasks are undertaken outside of the renal centre (e.g., in the child's home or school), and because CKD is a lifelong condition, child and family assumption of management responsibility is critical.

Studies suggest children, especially during adolescence, experience difficulties engaging in CKD self-management.[22] Adolescents have higher levels of kidney transplant loss compared to younger children and adults[23] and less than 20% of adolescents on dialysis were perceived by HCPs to have assumed self-management responsibility at transfer to adult care.[24] While the literature suggests the parent-to-child transfer of self-management responsibility is an important aspect of children's development, there is limited research on this transfer process involving children with CKD, and, crucially, how the process can be supported. Therefore, this study aimed to address this gap by exploring what supports young people with CKD to assume self-management responsibility and parents to relinquish control.

METHOD

The study used a constructivist grounded theory methodology.[25] Grounded theory is useful in exploratory research, as it aims to construct a theory that offers in-depth understanding and explains the phenomenon being studied.[26]

Sampling and recruitment

Participants were recruited from two UK children's kidney units. Purposive sampling was initially used as the aim was to achieve maximum variation in relation to (1) young people aged 13–18 years old with CKD stages 3–5 and their parents/carers, and (2) HCPs from a range of disciplines in the renal multidisciplinary team. As the study progressed, theoretical sampling was used to sample young people with CKD stages 3–4, to generate data to elaborate and refine the emerging categories. One clinician from each of the kidney units identified potential participants and gained consent for R. N. to provide them with study information. A total of 49 participants took part in the study comprising 16 young people (Table 1), 13 parent/carers (11 mothers, 1 step-father, 1 carer) and 20 HCPs (5 renal paediatricians, 4 nurses, 4 social workers, 3 clinical psychologists, 3 play workers, 1 dietitian).

Table 1 Characteristics of young people (*n* = 16)

Young people's characteristics	Girls (*n* = 9)	Boys (*n* = 7)	Total
Age			
13	1	2	3

14	1	3	4
15	2	1	3
16	4	1	5
17	1	0	1
Ethnicity			
White	4	3	7
South Asian	3	2	5
Black	2	1	3
Other	0	1	1
CKD stage/treatment			
Pre-emptive transplant	0	3	3
Dialysis	4	3	7
In-centre haemodialysis	1	3	4
Home dialysis	3	0	3
Transplant	5	1	6

Abbreviation: CKD, chronic kidney disease.

Data collection

Semi-structured interviews and focus groups were conducted to generate data. Young people and parents were offered the opportunity to be interviewed together or separately, and HCPs participated in either individual interviews or focus groups (Table 2).

Table 2 Data collection methods

Method	Type of participant/number	Length (range, in minutes)
Individual interview (*n* = 21)	Young people = 7 Parents = 4 HCPs = 10	24–78

Paired interview (*n* = 9)	Young people/parent dyads = 9	46–93
Focus group (*n* = 2)	13 HCPs	
	Focus group A = 9 ×HCPs (renal paediatricians = 3; clinical psychologists = 2; social workers = 2; nurse = 1; play worker = 1). 3 of these HCPs also took part in an individual interview	46
	Focus group B = 4 ×HCPs (social workers = 2; clinical psychologist = 1; play worker = 1)	54

Abbreviation: HCP, healthcare professional.

R. N. conducted all data collection, although the larger focus group (A) was co-facilitated by V. S. Interviews and focus groups took place in person in the family home or hospital setting, or by telephone and were guided by a topic guide. As part of theoretical sampling, topic guides were revised as the study progressed (Supporting Information: 1). Interviews and focus groups were digitally recorded and transcribed verbatim. To address some of the methodological and ethical issues of conducting research with children, task-based methods were used to generate data.[27] For example, in later interviews, participants were asked to consider the suggestions generated during earlier interviews around what supported the transfer of responsibility. Each individual suggestion was written on a piece of card, which was handed to participants, with the request that they consider each suggestion in turn.

Data analysis

Data collection and analysis were conducted concurrently, using an iterative, inductive process. Initial codes were developed by line-by-line coding, with the aim of identifying actions and processes in the data. Focused coding, in conjunction with constant comparison, involved evaluating the initial codes to identify analytical, and theoretical categories.[25] A supplementary approach was used to analyse how interaction contributed to data generation in the paired interviews and focus groups.[28] NVivo11 was used to code and manage data. To ensure trustworthiness and credibility, reflexivity and regular discussion between authors were incorporated into the analytic process.

Patient and public involvement (PPI)

PPI was integrated throughout the study, with two young adults with CKD and two parents of young people with CKD involved in the study's design and delivery. Table 3 summarizes the PPI at different stages of the study.

Table 3 PPI contributions

Stage of study	Advice sought	Methods
Initial research idea/before study started	Relevance of research idea; study methods; plain English summary for funding application	Online meeting
		Email
Applying for ethical approval	Participant information leaflets	Email
Data collection	Topic guides and task-based methods used during interviews	Face-to-face meeting
		Email

Data analysis	Discussion of study findings	Online meeting
Dissemination	Plain English summary of study findings for participants	Email

Abbreviation: PPI, Patient and public involvement.

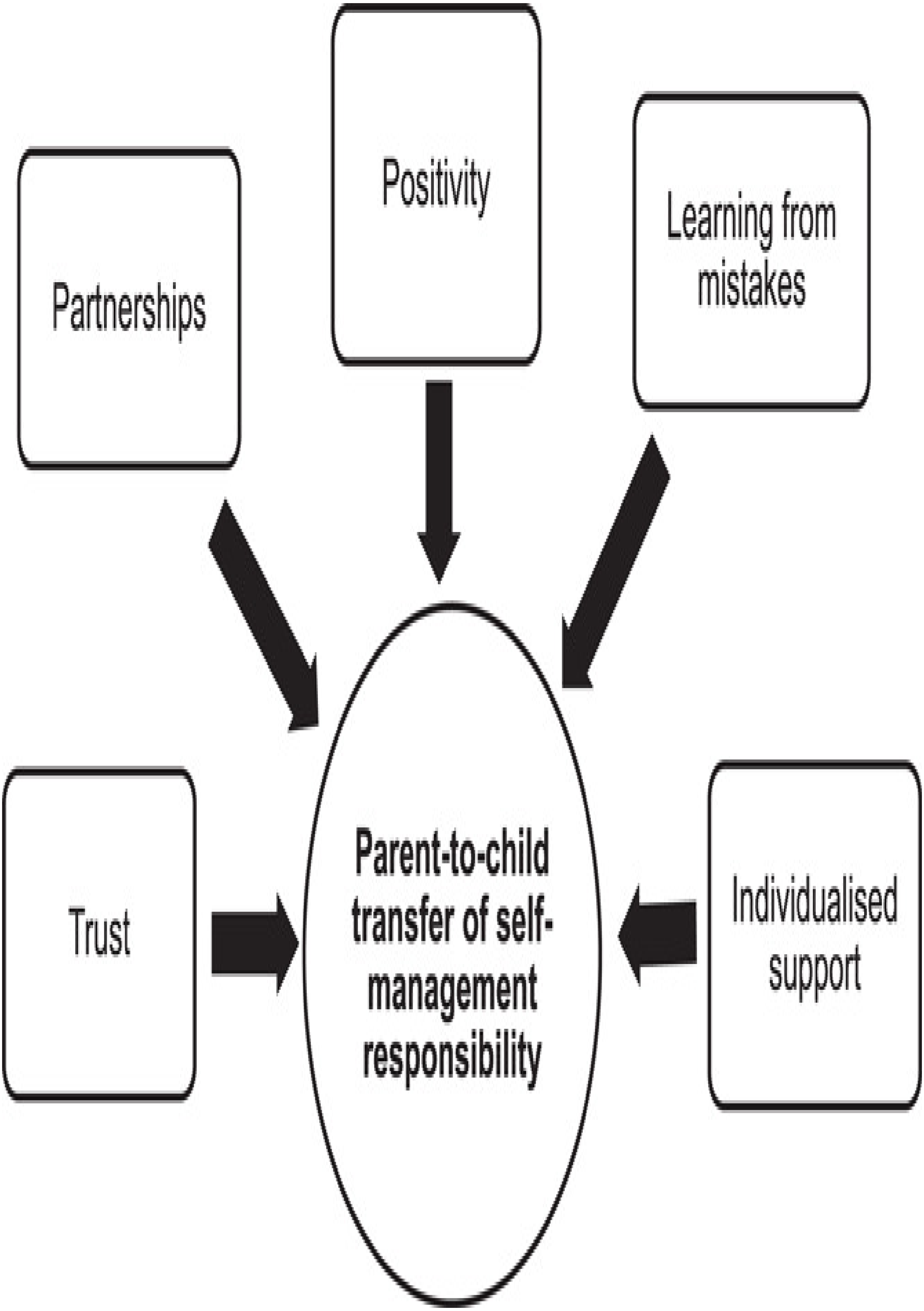
The impact of PPI on the study was manifold. For example, during discussions, none of the PPI contributors used the term ‘self-management’, instead describing young people ‘being in control’ and ‘taking charge’ of their health care; this had a significant impact on the language used with participants throughout the study, especially during data collection. PPI contributors' advice to change some of the language and design of the participant information leaflets made the leaflets more accessible and, through provision of improved information, potentially supported participants to make an informed decision about whether to participate.[29] Topic guides were revised based on feedback to: ask additional questions to explore other aspects of self-management PPI contributors thought relevant; alter existing questions so they were easier to understand; and adjust the order of the questions. Consultation with PPI contributors about study findings suggested the emergent categories and theory resonated with their own experiences of the transfer of CKD self-management responsibility.

Ethical issues

Participants were provided with age/developmentally appropriate information, and all provided informed assent/consent. Participants were assured of confidentiality and anonymity. In the data extracts presented, participants are identified by the type of participant (young person, parent, HCP) and the participants' numerical study identifier (1–20). The young person's age and gender are included in the data extracts to provide contextual information.

RESULTS

A grounded theory, *shifting responsibilities*, was constructed from the data, consisting of a core category (*shifting responsibilities*) and two connected subcategories (*developing independence* and *making changes*). Further details about the grounded theory, core category and subcategories have been reported previously.[12] This paper focuses on a specific aspect of the second subcategory, making changes, to explain how young people's, parents' and HCPs' adjustments to their behaviour and communication supported the parent-to-child transfer of self-management responsibility. This included behaviour and communication that: built and maintained trust; formed partnerships; fostered positivity; supported learning from mistakes, and was responsive to young people's and parents' individual preferences and needs (Figure 1). A gradual transfer, developing a routine, and connecting with others with CKD were also perceived to support the transfer process and have been described elsewhere.[12]



Enlarge this image.

Building and maintaining trust

Young people, parents and HCPs perceived trust was needed for the transfer of self-management responsibility.

This included trusting relationships between young people and their parents and between the young person–parent dyad and HCPs. Additionally, some young people suggested trusting themselves was an aspect of assuming responsibility. When young people were able to demonstrate they could consistently engage in self-management, parents started to trust that their child could be relied on to perform self-management:

It became a habit, I got good at taking them [medication], there was that trust. Then I stopped taking them. I think that trust is there again, but when I stopped taking them, I was obviously not being responsible. (YP1, 14-year-old girl)

Parents adjusted their behaviour and communication as trust was built with their child; for example, they reduced how often they reminded their child to take their medication or monitored their renal diet. Trust was essential for parents to feel able to relinquish control. However, as the quotation above highlights, maintaining trust was difficult, especially if the young person disengaged, even temporarily, from self-management. When trust was lost, the transfer process was disrupted as parents tended to reassume responsibility and an increased level of control. Some HCPs' perceived they had a role in supporting parents to start trusting their child was able to self-manage. This included identifying opportunities where the young person would be able to demonstrate to their parents that they could be trusted, such as connecting themselves safely to their dialysis machine, or following their renal diet when outside the family home:

With the diet, one thing happens at home, and another thing happens at school or when they're out with their friends. One way that I tackle it, is for them to take more responsibility for what happens when they're not at home first. If they can show their parents that they're managing well when they're out on their own, and the parents can trust them to make the right decisions, then that shows them that they are capable of managing …it's trying to build up the trust between the child and their parents. (HCP2)

Trusting relationships between the young person–parent dyad and HCPs were perceived as supporting the transfer of responsibility:

If they trust in you, I think that's very helpful. I've looked after most of these people for the last 14 years, I'm a familiar face. We've got a relationship, we've built up trust over time, that really helps. To analyse the problems, the young person has got to be open first. (HCP8)

This quotation suggests that trust needed to be two-way, that HCPs needed to be able to trust families, as well as young people and parents trust HCPs. There was a sense that as young people assumed self-management responsibility, they needed to be 'open' with HCPs, which was more likely if there was a trusting relationship. Some young people described how being able to trust themselves, or having confidence in their ability to manage their condition, was part of assuming responsibility. Their accounts suggested that this impacted how much their parents were able to trust them and relinquish control:

I can't trust myself with food, because I like a lot of food that I'm not supposed to eat. Sometimes I won't be able to contain myself from not eating it. My mum, she cares too much about me to stop reminding me about the things I eat, so she won't hand me that responsibility that easily. (YP15, 15-year-old boy)

Approaches used to support young people to trust themselves and develop self-confidence, included HCPs and parents acknowledging when young people were managing their condition. This will be discussed in more detail in Section 3.3, fostering positivity.

Forming partnerships

Partnerships between the young person, their parents and HCPs were perceived to support the transfer of self-management. Young people and parents described how 'teamwork', which included undertaking self-management activities together, supported young people to become increasingly involved in managing their CKD. HCPs adopted a range of approaches to encourage partnership including directing communication primarily at young people rather than parents; exploring young people's concerns and their motivation to assume responsibility; joint goal-setting; findings solutions together; acting as an advocate for the young person and helping young people to negotiate with their parents around the transfer of responsibility. Young people appeared to value being treated as an equal; they described how interactions with HCPs that encouraged partnership, supported their assumption of responsibility:

It's a two-way thing. They [HCPs] want your take on it, because they don't want to be saying things and then me leave and be, ‘Forget that. I'm not doing that’. They ask our opinions, how it would work. They are very supportive in that way. It's your opinions and their opinions, but they mostly want your take on it, so you can help them understand. I like the independence, they're treating me like an adult rather than a kid. (YP1, 14-year-old girl)

Although most HCPs encouraged young people to attend appointments on their own, there were conflicting views among HCPs around how much parents should be included and whether they were a barrier or facilitator to young people taking responsibility for condition management. The few young people who had attended appointments on their own appreciated having the opportunity to focus on issues important to them and talk more openly with HCPs, compared to when their parents were present. Parents, however, appeared more ambivalent about HCPs' decisions to include or exclude them from consultations; while they seemed to accept that HCPs forming a partnership with their child was a necessary stage in their child assuming responsibility, they also struggled with relinquishing control. Some HCPs emphasized the need to partner with the young person–parent dyad and perceived parents' involvement was critical to supporting young people to assume responsibility:

It does need to be in tandem because they are closely entwined. The danger of doing it in isolation is that the young person comes home and goes, ‘Mum I've talked to this nurse, I want to take my own meds’, and the parent goes, ‘No bloody way!’ Unless you're doing it together, I mean it could work, but it's going to be more successful if you're doing it as a combined approach. (HCP17)

Fostering positivity

Young people, parents and HCPs described how the transfer of responsibility was often a difficult process, in particular when young people struggled to integrate self-management into their daily life. Therefore, behaviour and interactions that fostered positivity, such as acknowledging when the young person had been able to manage their condition, and focusing on what was going well, were perceived to support the transfer of self-management. Parents, in particular, emphasized the importance of keeping positive, even when their child was struggling with self-management:

Sometimes she'll [child] say, ‘I'm doing well with my tablets, aren't I?’ I'll be like, ‘Oh, yes’. I try to be positive about it, but I can't say if she's had any tablets yesterday. I try to look at the positive stuff, she could be a lot worse than what she is, behaviour wise, but it is a concern to me. (Parent7, 16-year-old girl)

HCPs' accounts suggested they were aware of the need to acknowledge a young person's strengths. However, there was a sense this rarely happened, as appointments tended to focus on problems, including the young person's difficulties with assuming responsibility:

Sometimes patients do nine tasks out of ten really well, but the focus in clinic will be on the one they're not doing, which is disheartening on the young person, because they probably really tried, and it's the one thing that they've not managed to stay on top of. Conversations tend to be so negative, that it puts them right off trying again. Somebody needs to say, ‘Well done for doing your medicine, turning up today, engaging in your healthcare, but we need to work a little bit on…’. (HCP1)

As this extract suggests, HCPs making changes to their interactions with young people to acknowledge what they had achieved and provide positive feedback was perceived to support young people's motivation to continue engaging in self-management.

Supporting learning from mistakes

When young people had difficulties with assuming self-management responsibility, learning from mistakes was perceived to be helpful. Some young people acknowledged the impact on their health when they stopped engaging in self-management, and this prompted them to resume responsibility:

I definitely learnt from my mistake. I keep my water bottle near now. I make sure I'm keeping on top of things. I have all my medications properly, and check and double-check that I've got all my medications. (YP14, 16-year-old girl)

Although parents were aware of the potential risks of their child making mistakes with self-management, they accepted making mistakes was ‘normal’ and could provide opportunities for their child to learn:

I'd tell parents with teenage children, when they make mistakes, let them see. Let them understand that sometimes they will make mistakes. Don't teach them there's no mistake, no, then you make them so rigid, let them be free with you. Tell them it's a mistake and this is the repercussion, so they know. (Parent3, 15-year-old girl)

As this extract suggests, acknowledging that young people would make self-management mistakes could potentially encourage young people to be 'free' or honest with their parents when they were struggling with self-management. HCPs accounts also indicated how learning from mistakes could facilitate the transfer of responsibility. Some HCPs described discussing with families how mistakes could provide opportunities for young people to develop an understanding of the consequences of their self-management decisions:

Being a teenager is about making mistakes, it's learning from your mistakes. But we don't want them to make mistakes that cause them harm …I talk to the family, I say making mistakes is the learning process, let them make mistakes safely, not letting them make any mistakes is not safe. (HCP8)

However, as this quotation highlights, the emphasis was on making mistakes 'safely' due to the awareness that some self-management mistakes could have a significant impact on the young person's health.

Individualizing support

Young people, parents' and HCPs' accounts suggested that the transfer of responsibility was completely individualized to each family. Contextual issues, such as the: young person's chronological and developmental age; family relationships and physical and social environment, interacted with and influenced the transfer process. A young person's progression through the CKD stages and the condition-specific self-management requirements, such as starting dialysis or receiving a kidney transplant, were also perceived to impact the young person's assumption of responsibility and parents' willingness to relinquish control. During a dyadic interview, a 16-year-old girl and her mother discussed how responsibility shifted after she had received a transplanted kidney:

Young person: Before my transplant I was responsible for taking my tablets of an evening, and you would just know. You wouldn't even—,

Parent: She only took two tablets. She took them at night and at that point I never used to check in. Now and again I used to say, 'Have you taken your tablets?' when I said goodnight, but it's not like it is now. I think it's the importance of the tablets, because tacrolimus [immunosuppressive medication], if you forget it, it's massive …I was a lot more slapdash then. (YP8, Parent 8, 16-year-old girl)

HCPs accounts indicated they were aware that the transfer of responsibility was experienced differently by each family. The importance of individualizing support to each family's needs was discussed in focus groups, as HCPs generated ideas around what facilitated the transfer of self-management:

HCP8: For some people, meeting other patients would be hell, for some it would be great …there isn't one size that fits all.

HCP1: It's tailoring it. Like you say, some people wouldn't engage, some don't like digital technology, but they'd like the face-to-face. It's finding what fits.

Although HCPs believed support needed to be individualized, national and local transition guidance around young people moving from paediatric to adult services, underpinned service provision. Consequently, HCP involvement in the transfer of responsibility tended to start when young people were around 13 years old and finished when they transferred to adult services. Some HCPs accounts revealed their frustration that the young person's chronological age, rather than their ability to self-manage, determined when they moved to adult services:

HCP8: We are driven by age …that drives when we do transition rather than the patient.

HCP11: It depends as well where you work. We have a [NHS] Trust that mandates that we move patients over at the age of 16 …but there are other Trusts where between 16 and 19, young people are offered a choice, 'Do you want to go to paediatric services, or move up to adult services?' So how we practise as clinicians is dictated by the management who decide how they want to do things within this Trust. (A National Health Service (NHS) Trust is an organizational unit in England and Wales that provides health services, and generally serves either a geographical area or a specialized function).

These extracts highlight potential tensions between HCPs' belief in the need for individualized support and what they were able to provide in practice.

DISCUSSION

Previous studies have explored the parent-to-child transfer of self-management responsibility, but little is known about what support young people and parents' need as responsibilities shift.[16] This study contributes to knowledge by identifying what facilitates this transfer process, and, importantly, HCPs' current and potential role in helping both young people to assume responsibility, and parents relinquish control. Findings suggest there were similar views among young people, parents and HCPs about what supported the transfer of responsibility. However, some tensions appeared to be evident, in particular around the formation of partnerships between HCPs and young people that excluded parents, and the provision of individualized support. By highlighting what facilitates the transfer of responsibility, study findings both support and extend the existing literature, and have implications for practice.

Behaviour and communication that built and maintained trust were perceived to help the transfer process. This finding supports existing research that found parents needed to trust their child to relinquish control.[30,31] However, by exploring HCPs' perspectives, this study extends the current understanding of the HCP role, suggesting HCPs could contribute to the development of trusting parent-child relationships. Previous studies recognized that situations, when the child was away from the family home (e.g., to attend school, or socialize with friends), could be anxiety-provoking for parents, and therefore, recommendations were made that HCPs should provide reassurance to parents about their child's self-management ability.[32] In contrast, this study's findings suggest that by actively identifying situations when the young person has the opportunity to demonstrate to their parents they can be trusted to engage in self-management, HCPs can help build and maintain trust. The importance of trusting relationships between young person–parent dyads and HCPs has been highlighted in previous research. Sullivan-Bolyai et al.[33] found parents lost trust in HCPs when HCPs believed the deterioration in young people's health was a consequence of parents' transferring responsibility to the child before they were ready. The inclusion of HCPs in this study, however, extends knowledge in this area by suggesting trust is two-way, as young people–parent dyads need to trust HCPs, and HCPs need to trust families.

Some young people in this study believed they needed to be able to trust themselves to assume self-management responsibility. This suggestion that young people with CKD benefit from developing confidence and belief in their own ability aligns with the concept of self-efficacy.[34] Although the literature proposes that enhancing self-efficacy can facilitate young people assuming responsibility,[18,35,36] there is limited empirical research to support this. Colver et al.[9] suggest HCPs should encourage self-efficacy and recommend further research 'to identify the most effective and efficient ways to promote young people's knowledge and confidence in the management of their LTC' (p. 77). By identifying approaches that can support young people's belief in their self-management ability, such as fostering positivity and connecting with others with CKD,[12] this study's findings have implications for practice.

Young people, parents' and HCPs' perceived partnerships supported the transfer of responsibility. Previous research has highlighted the importance of collaborative child–parent relationships, as young people are more likely to learn self-management from their parents, rather than HCPs.[22,37] Participants' accounts suggest that HCPs formed partnerships primarily with young people, rather than the young person–parent dyad, as they perceived this encouraged young people to assume responsibility. As reported previously, HCPs tended to view the transfer of responsibility as part of the transition between child and adult services.[12] Consequently, UK transition guidance shaped HCP involvement, including the importance of young people attending clinic appointments without their parents.[8,38] Consistent with previous research, young people in this study valued meeting with HCPs on their own, as they felt more able to talk openly without their parents present.[22] While some parents were positive about their children attending appointments without them, others struggled with being excluded and wanted to be kept informed.[39] The conflicting views among HCPs about whether parents are a facilitator or barrier to the transfer process and parents' ambivalence about their inclusion or exclusion from consultations,[40] extend the debate around whether HCP involvement should be child- or family-centred.[41] Although it has been recommended that triadic collaboration is fostered between young people, parents and HCPs during the transfer of responsibility,[9,42] only a few HCPs in this study seemed to view parents as supporting the assumption of responsibility. Therefore, few aimed to form a partnership with the young person–parent dyad. The uncertainty around how HCPs balance child- and family-centred care during the parent-to-child transfer of responsibility indicates further research is needed.

Parents and HCPs perceived the transfer of responsibility was supported by fostering positivity. Only a few previous studies exploring diabetes self-management identified positive reinforcement and offering rewards as helping young people to assume responsibility.[33,43] However, as neither of these studies included HCP participants, further research exploring how HCPs can adopt a ‘strengths-based approach’, as recommended by UK transition guidance, is needed.[38] Consistent with previous studies, young people learnt from making mistakes with self-management.[44,45] Parents and HCPs were aware, however, that some mistakes could have a significant impact on the young person's health and, as a result, there was ambivalence about learning through trial and error.[46] Although the existing literature recommends HCPs increase opportunities for experiential learning so young people can learn from the mistakes they make,[33] there is limited evidence to suggest that HCPs have utilized this strategy. Potentially due to including HCP participants, this current study extends knowledge in this area, finding that HCPs discussed with parents how making mistakes ‘safely’ was part of their child assuming responsibility.

In this study, HCPs described the importance of tailored support to meet the individual needs of young people and parents. Previous literature has discussed the need for HCPs to consider children as individuals and avoid having a uniform policy around when, and how the transfer of responsibility occurs.[15,47] However, as a consequence of UK transition guidance underpinning HCPs support to young people assuming responsibility, HCP involvement tended to be service-led, rather than based on family needs.[12] This highlights a potential tension between HCPs' beliefs that support needs to be individualized and what occurs in practice. Although guidance recommends HCPs adopt individualized or personalized approaches,[1,4] there is limited evidence around how HCPs use these approaches in practice to support the transfer of responsibility. Further research to explore how HCPs construct and implement individualized support to facilitate the parent-to-child transfer of responsibility is needed.

Strengths and limitations

Having PPI to advise on the design and conduct of this study was a major strength and impacted on the quality and relevance of its findings. An equal focus on HCPs' perspectives, alongside those of young people and parents, assisted with gaining an in-depth and holistic understanding of what supports young people to assume self-management responsibility. Although there was diversity in the sample, especially in relation to young people's age, ethnicity and CKD stage/treatment and HCPs' discipline, selection bias may have occurred due to reliance on clinicians in the kidney units for recruitment. Diversity could have been increased further through the recruitment of a greater number of fathers. Dyadic and focus groups can generate unique ethical and practical challenges, as power relations and family/group dynamics can potentially inhibit some participants from speaking.[48,49] However, adopting techniques such as task-based methods, the researcher aimed to encourage young people and ‘quieter’ group members to contribute to discussions.

CONCLUSION

This study has explored what supports the parent-to-child transfer of self-management responsibility for CKD. Study findings have contributed to knowledge, and, importantly, have identified HCPs' current and potential role in facilitating young people to assume responsibility and parents to relinquish control. These new insights have implications for practice, highlighting how families would benefit from individualized support that helps to: build and maintain trust, form partnerships that include parents, foster positivity and support learning from mistakes. Conflicting views around whether parents are a barrier or a facilitator to young people assuming responsibility indicate further research is needed to understand how HCPs can balance child- and family-centred care when supporting the transfer process. Finally, further research to explore how HCPs can adopt personalized and strengths-based approaches in practice would help ensure the support that families receive is tailored to their individual needs.

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CONFLICT OF INTEREST

The authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

Research data are not shared.

ETHICS STATEMENT

Approval was obtained from the UK Health Research Authority (226365), a National Health Service (NHS) Research Ethics Committee (18/YH/0210) and the NHS Trust Research and Development Departments.

DETAILS

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Exploring COVID-19 experiences for persons with multiple sclerosis and carers: An Australian qualitative study

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ABSTRACT (ENGLISH)

Objective

The COVID-19 pandemic continues to impact communities around the world. In this study, we explored the COVID-19 experiences of persons with multiple sclerosis (MS) and carers.

Methods

Using a qualitative approach, interviews were undertaken with 27 participants residing in Australia (10 persons with MS, 10 carers and 7 MS service providers). Demographic and background data were also collected. Interviews were analysed using an inductive iterative thematic analysis.

Results

Across all groups, participants consistently recognized pandemic challenges and impacts for persons with MS and carers, especially due to disruption to routines and services. Emotional and mental health impacts were also highlighted, as anxiety, fear of contracting COVID-19 and stress, including relationship stress between persons with MS and carers and family members. Some persons with MS also mentioned physical health impacts, while for carers, the challenge of disruptions included increased demands and reduced resources. In addition to acknowledging challenges, persons with MS and carers also gave examples of resilience. This included coping and adapting by finding new routines and creating space through rest and breaks and through appreciating positives including the benefits of access to telehealth.

Conclusion

Additional support is required for persons with MS and carers in navigating the impacts of COVID-19 as the pandemic progresses. In addition to addressing challenges and disruptions, such support should also acknowledge and support the resilience of people with MS and carers and enhance resilience through supporting strategies for coping and adaptation.

Patient and Public Contribution

Service user stakeholders were consulted at the beginning and end of the study. They provided feedback on interview questions and participant engagement, as well as service user perspectives on the themes identified in the current study. Participants were provided with summaries of key themes identified and invited to provide comments.

FULL TEXT

INTRODUCTION

During the COVID-19 pandemic, many communities have been dominated by public health directives to limit the spread of disease and reduce negative health impacts. In Australia, this included restrictions, such as lockdowns or limiting social mobility, as well as other directives such as mask-wearing.[1] The potential impact of COVID-19 has been particularly salient for individuals who have existing chronic neurological conditions, such as multiple sclerosis (MS).[2] In Australia, researchers estimated in 2017 that there were at least 25,000 persons with MS (103.7/100,000), increasing from previous estimates and varying across states, with the highest prevalence in Tasmania and the lowest in Queensland.[3] Throughout the pandemic, public health directives meant that usual MS healthcare services were unavailable or substantially altered,[4] with reduced outpatient and in-home care services[5] and increased reliance on telehealth.[4] These service changes were received with varying acceptance across the MS community.[6,7] Research on the potential impacts for persons with MS during the pandemic have been mixed. Some report reductions in positive health behaviours, such as decreased physical activity, poorer sleep and dietary behaviours.[8,9]

as well as increased levels of anxiety,[10–12] depression[12–14] and greater fatigue[14] compared to persons without MS. Research also identifies COVID-specific concerns such as fear of contracting the virus, perception of higher risk in persons with MS and increased loneliness associated with confinement and minimized social contact.[14] Social concerns for persons with MS also extend to less social support, financial issues and employment concerns.[7,13]

In contrast, a recent rapid systematic review argues that while anxiety, stress and depression may be higher in persons with MS during the pandemic compared to others, they may not be substantially different to prepandemic levels.[15] Additional research shows minimal MS symptom change,[7] as well as reporting positive impacts of COVID-19, including increased family time and improved capacity to manage fatigue.[7,16] Some research also recognizes individual and social contexts that support coping and resilience during the pandemic.[17] Persons with MS may employ supportive strategies such as active coping and acceptance[17] and maintain health behaviours such as physical exercise that reduce the impact of disability and depression in this context.[8,18] Together, these behaviours may result in greater resilience, predicting less depression and anxiety.[10]

Family and care of persons with MS may also be impacted during a pandemic. Restrictions may dramatically change daily interpersonal environments and disruptions to support services are often addressed by the immediate family. Given the importance of the relational network of persons with MS,[19] an understanding of this social context is imperative, yet research is limited. Research looking at informal carers of people with various chronic health conditions in the context of the COVID-19 pandemic reported an increase in caregiving intensity and burden[20] as well as greater symptoms of depression, anxiety, fatigue and sleep problems compared to noncarers.[21] For MS carers specifically, research recognizes increased responsibility for care in response to limited home services, coupled with the lack of support for these family members to care for their own health and well-being.[7] Australian and European caregivers of persons with MS express increased anxiety about worsening MS symptoms in their loved ones and fears of COVID-19.[7,16] To date, there appear to be no other studies that focus on pandemic experiences and impacts for carers or family members of persons with MS.

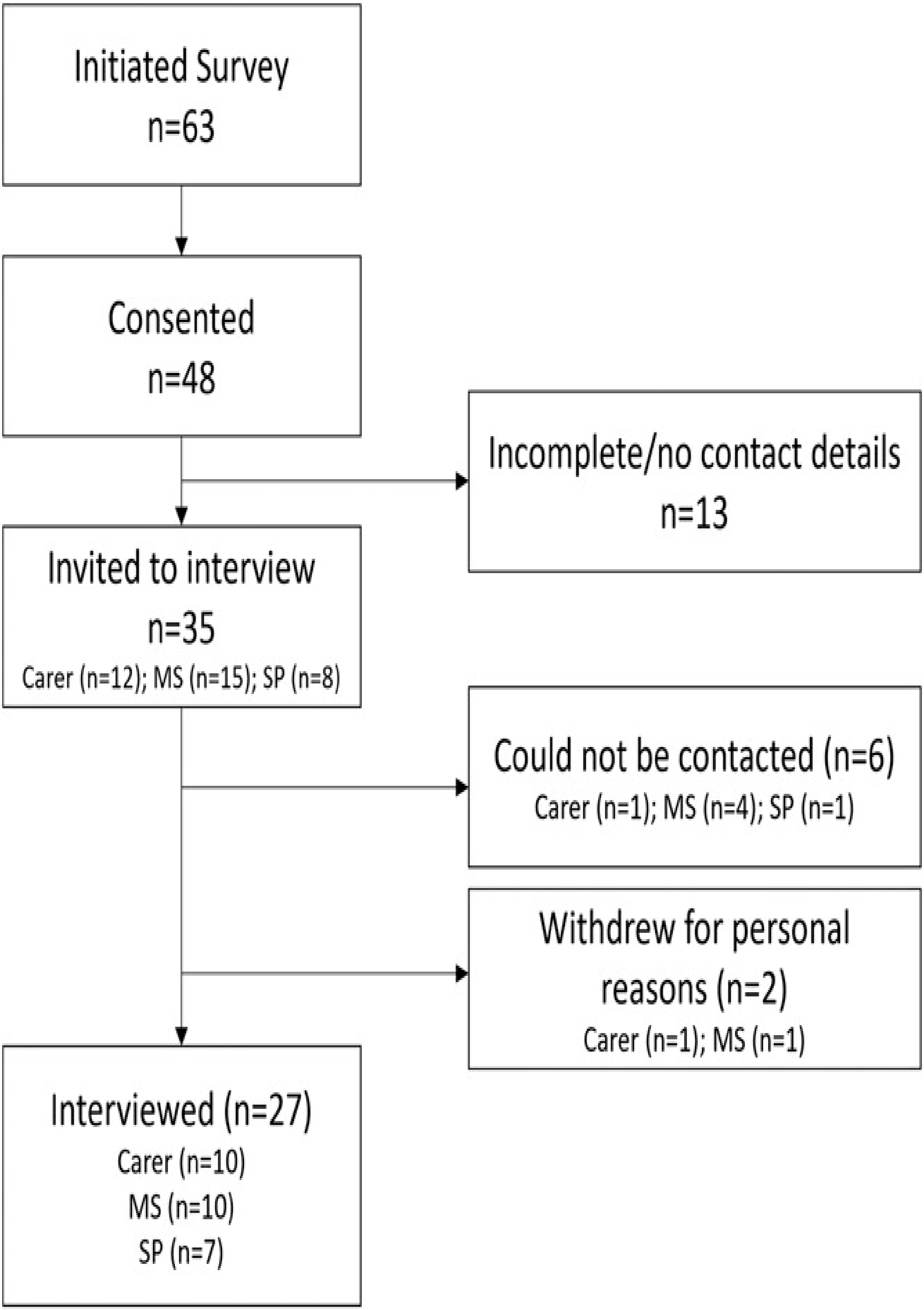
There are concerns more generally about declines in carer psychological well-being in the absence of sufficient support for family carers during the pandemic,[22] with international calls for carer support and caregiver-centred interventions.[22,23] Yet there is little research specifically on the impact of COVID-19 on MS carer and family experiences to guide the development of such interventions tailored for the MS community. Using a qualitative approach to deepen understanding of the issues, this study builds on previous research by integrating multiple perspectives: persons with MS, carers and family members and MS service providers. MS service providers can provide a key perspective on potential impacts such as service access and engagement, especially in the context of responding to community crises such as COVID-19.[7] To date, there appear to be no other studies that integrate the service provider perspective in relation to carer experiences during COVID-19. The primary aim of the current study was to explore how the pandemic influenced the experiences of persons with MS and MS carers and loved ones, including changes to the care experience in both personal and service contexts.

METHODSStudy design

This study was part of a broader qualitative study investigating caregiving experiences in MS. For the current study, the data set focused on a subsection of the data corpus in which participants commented specifically on their experience during the pandemic and how the pandemic had impacted care experiences. A university ethics review board granted approval for the study, and consumer representatives provided input into the study design and interview process. All participants received relevant study information and provided consent before participating in interviews. Participation in the interviews was voluntary, without remuneration. Interviews were conducted between September 2020 and August 2021. During this time, the different states and territories of Australia were experiencing varying levels of pandemic impacts, from low levels of transmission, restriction and minimal social disruption (e.g., Western Australia) to higher levels of transmission and government-directed societal lockdown (e.g., Victoria).

Participant recruitment

To sample from a broad range of perspectives, we recruited participants across Australia from three different groups: persons with MS, carers/loved ones and services (e.g., health professionals, support service providers). Potential participants with MS and carer participants were recruited through electronic advertising (e.g., newsletters, social media) via MS organizations, as well as a database of participants in previous studies who had consented to be contacted about future studies. Potential participants from services were also recruited via MS Australia and the Australian state-based MS organization (i.e., via social media announcements and emails). All interested individuals were directed to an online survey site (Qualtrics), which provided information about the study and collected basic information (demographics, background information) as well as consent to being contacted for the interview. A flow chart of participation through to interview can be seen in Figure 1.



Enlarge this image.

Persons with MS could be included if they were above 18 years of age, had an established definite diagnosis of MS and received regular support or assistance with activities from carers (e.g., transportation, shopping and meal preparation). Carers could be included if they were above 18 years of age, had a close connection to a person with an established diagnosis of MS and had regular and frequent contact to provide care and support for the person with MS. Service providers could be included if they were involved in health-related services/management of persons with MS (including health professionals, service managers or MS community support and advocacy). As individuals who have ongoing contact with persons with MS and their carers, this group provides an additional perspective on experiences such as changes to engagement in services during the pandemic. To capture the broadest range of experiences, individuals were not required to be linked to each other.

Participants

Thirty-five individuals provided contact information for the interview (15 persons with MS; 12 Carers; 8 service providers), but six could not be contacted. One person with MS and one carer withdrew for personal reasons before the interview could be conducted. Twenty-seven participants completed interviews (10 persons with MS; 10 carers; 7 service providers). To our knowledge, the participants were independent of each other. This sample represented all the individuals who expressed an interest and could be contacted before the study closing.

Participants identified being in age groups from 25 to 75 years of age. Participants with MS and carer participants predominantly identified their partner/spouse as the main caregiving relationship, except for one person with MS who identified their friend as the main carer and two carers identifying their adult child with MS as the person they support. Participants with MS were represented across age groups 25–75 years of age, while carers were mostly older (55–75 years of age) and service providers were mostly younger (25–44 years of age). Participants across all three groups were predominantly female (85% of all participants).

Participants were recruited from across Australia and from most states and territories. Most service providers were healthcare practitioners (four physiotherapists, one counsellor, two coordinators of well-being/disability support for persons with MS and carers). All service providers reported seeing clients with a variety of MS types and disability levels. See Table 1 for participant characteristics across groups.

Table 1 Participant information

	MS	Carer/family	Service provider
Participants	10	10	7
Gender			
Female	9	8	6
Male	1	2	1
Age			
25–34	2	1	3
35–44	3	1	3
45–54	2	1	0
55–64	2	5	1

65–74	1	2	0
State/territorya			
Australian Capital Territory	2	0	0
New South Wales	2	4	0
Queensland	1	1	1
South Australia	1	0	0
Victoria	1	2	2
Western Australia	3	3	4
MS type of person with MSb			
Relapse remitting	8	5	-
Primary progressive	1	1	-
Secondary progressive	1	4	-
Years since the diagnosis of person with MSb			
0–4	2	2	-
5–9	3	3	-
10–14	3	0	-
15–19	1	3	-
20–24	1	0	-
25–30	0	1	-
30 or more	0	1	-
Disability (PDDS) of person with MSb			
Mild	3	2	-
Moderate	6	2	-

Severe	1	6	-
Carer-burden (CTiMMS means)c			
Activities of daily living	-	1.83	-
Instrumental care	-	3.20	-
Psycho-emotional care	-	2.48	-
Social-practical care	-	2.53	-

Abbreviations: MS, multiple sclerosis; PDDS, Patient Determined Disease Steps.

a

No participants were from Tasmania or Northern Territory.

b

MS-related information from carers refers to the person with MS that they provide care for.

c

CTiMMS range: 0 (No help)–4 (lots of help).

Procedure

Interviews were conducted by two members of the research team (H. C. and P. M.-L.), who have experience in interviewing and qualitative research. None of the participants had a pre-established relationship with the interviewers. Question development was informed by previous research, reviewed following initial pilot interviews and feedback from stakeholder representatives (a person with MS and a carer). As part of the broader semistructured interview, participants were asked questions relating to caregiving and carer involvement in health care for persons with MS. Interviews were conducted by phone and lasted between 30 and 50 min (average of 42 min). For the current study, participants were invited to comment on the impact of COVID-19 personally and any changes to the care experience that they had observed. The invitation was an open-ended discussion and general, allowing for variability in impact given the diversity of geographical experiences with the pandemic (e.g., exposure to the virus, health directives, lockdown duration, restrictions). Clarifying questions were based on participants' responses. All interviews were audio-recorded and transcribed for analysis, removing any identifying information. Following each interview, the researcher created a summary of key ideas identified in the interview and sent it to the participant with an invitation to review and provide feedback or to provide further comments. Any returned comments were incorporated into transcripts for analysis.

Demographic and clinical descriptors

During initial recruitment via our online recruitment survey, we gathered data on gender, age and state or territory from all participants. From persons with MS, we gathered data on MS type at diagnosis, years since diagnosis, disability level and carer context (relationship type, living arrangements, etc). The level of disability was gathered using the Patient Determined Disease Steps (PDDS), which correlates highly (*r* = .78) with the clinician-rated Expanded Disability Status Scale.[24] We categorized these into no or mild disability (score of 0–2), moderate disability (score of 3–5) and severe disability (score of 6–8). For carers, we asked about the number of years they had been a carer for their associated persons with MS, what type of MS the person has and the level of carer support. Levels of carer support were established using the Caregiving Tasks in MS Scale (CTiMSS).[25] The CTiMSS comprises 24 items measuring the amount of help provided to care recipients on scales from no help (0) to lots of help (4). These are grouped into four domains assessing basic activities of daily living (e.g., toileting, feeding), instrumental activities of daily living care (e.g., preparing meals, transportation), psycho-emotional care (e.g., managing fatigue, mood swings) and social-practical care (e.g., providing companionship, assisting with physical exercises). Mean scores were calculated for each subscale, with higher ratings indicating more carers' help in given tasks. Service providers were asked to describe their work role and their experience working with persons with MS.

Data analysis

The current study analysed the data set from interviews specifically pertaining to COVID-19 experiences. This was predominantly generated in response to the specific question on COVID-19 at the end of the interview. To maximize the data set, spontaneous comments relating to COVID-19 experiences provided in other parts of the interview were also included. Analysis of transcripts was undertaken using NVivo software (Release 1, 2021; QSR International Pty. Ltd.). Given the exploratory nature of the research, coding and development of themes followed an inductive iterative process used in thematic analysis[26] with steps to enhance trustworthiness and rigour.[27] Analysis was positioned as experiential and inductive and incorporated both semantic and latent coding.[28] The stages of analysis included initial analysis and development of preliminary codes undertaken by two members of the research team (H. C. and P. M.-L.); review of open coding and development of a broad framework; review of emerging themes and subthemes with reference to the raw data; producing the report. To support triangulation across these stages, code and theme development was discussed between coders, with further review and input through regular meetings with members of the research team. Findings have been reported using COREQ guidelines for qualitative research.[29]

RESULTS

Several key themes were identified in the data including acknowledging *challenges and impacts*, *coping and adapting* to the challenges of COVID-19 and *appreciating positives* despite the challenges that emerged because of the pandemic. Table 2 provides a summary of key themes, subthemes and examples of codes.

Table 2 Overall themes, subthemes and example codes across perspectives: Participants with MS, carers/family, MS service providers

Themes	Subthemes	Example codes
Challenges and impacts	Disruptions	Personal disruptions; social disruptions; service disruptions
	Physical and mental health	Fear of infections; anxiety and stress; mental health challenges; physical health impacts
	Reduced resources	Demands and pressures; increased carer burden; lack of support; energy and fatigue
	Relational and social	Isolation; living pressures; relational stress; employment impact
Coping and adapting	Creating space	Getting out; respite and taking breaks; time apart
	Routines and activities	Maintaining and replacing activities; adapting to telehealth; maintaining physical health; self-care; social activities
Appreciating positives	Service continuity	Maintaining access to services
	Technology supports	Telehealth; online connections

	Strengthening relationships	Time together; becoming closer

Abbreviation: MS, multiple sclerosis.

Consistency of themes across groups

Each major theme was reflected in responses from participants from all three groups and was present in responses across more than half of those interviewed. Interestingly, service providers' comments predominantly focused on challenges and impacts, especially subthemes relating to pandemic-related disruptions and impacts on emotional and mental health. Ideas relating to the main themes of coping and adapting and appreciating positives were more often raised by persons with MS and carers and family members rather than service providers. Group variations in themes and coding are expanded in the sections below.

Challenges and impacts

The strongest theme described pandemic-related challenges and impacts. This included disruptions to services and personal routines due to health directives such as lockdowns and restrictions. Some participants also described behavioural changes due to personal fears and concerns about COVID-19.

Disruptions

Participants across all groups typically highlighted observed disruptions as a result of COVID-19, especially service disruption for persons with MS. Some participants highlighted the impact on physical health and social engagement. We also heard that while telehealth was welcomed, it was not always easy or sufficient.

But I think what one of the major things that has a negative effect on (husband with MS) is, say, neuro physio and things like that, that's all been kind of cancelled, which has happened before in the past, which does have quite an impact on his body, quite a bit. (Carer, Female)

The disruptions in personal routines and social events for persons with MS as well as carers and family members were also noted. This included personal health routines, travel, recreational activities and family events. In some cases, access to these activities appeared to be due to limits imposed through health directives, and in other cases, it was connected to concerns about COVID-19 itself.

Well before Covid I was actually doing an exercise program with MS but because of possibly picking up a bug in a gym I basically stopped going back. So I suppose that that was the biggest impact. When it started I haven't been bothered to go back. (MS, Male, severe disability)

I think the main one is probably the socialisation aspect. There's a lot of people too scared to go out. There's a lot of care support workers who don't do social things with clients now because of all the fears of the bugs being transmitted, the virus being transmitted around. So if anything, we've lost a lot of socialisation. (Service Provider, Female)

As suggested in the excerpts, we also heard from participants about the personal and social impact of these challenges on their physical, mental and relational health.

Physical and mental health

Participants across all groups commented on the emotional response to the pandemic and its challenges, as well as concerns about the physical and mental health of persons with MS and carers/family members. These connected both directly to the pandemic (e.g., fears of infection), as described previously, because of responses to the pandemic, such as disruption to services or reduced access to activities. The interviews included reference to a broad range of emotions such as fear, anxiety, stress, frustration and depression, including personal experiences in themselves or observed in persons with MS and carers and family members.

So, it would be nice to have the fear and concern not as kind of like an everyday thought and concern. (Carer, Female)

We moved treatment dates around because we didn't want to be too immuno-compromised or coming into the hospital during Covid outbreaks, and so that is stressful, like I think I'm more stressed and anxious than I would have been otherwise and so that obviously then has an impact because it just means everything is harder to navigate, blood tests are harder to navigate, so it does make me grumpier I think. (MS, Female, moderate disability)

Participants described health concerns about contracting COVID-19, describing their perceptions of higher risk and how this may influence their choices in accessing services and engaging in activities.

Reduced resources

Participants across all groups acknowledged that disruption to MS-related services was often accompanied by increased demands and lack of support, especially for immediate family and carers who assumed the support that would ordinarily be provided by services. We noted observations of the emotional and physical impact of navigating the pandemic, placing strain on personal resources.

It's a lot harder obviously being at home and with the treatment I wasn't very mobile either so it was so much heavier on him (participant's carer)—he had to do everything really for at least 3 months. (MS, Female, moderate disability)

A feeling of, you know that carer's burnout are really coming to fruition where the supports weren't able to come into the home and they had to do everything themselves and the quality of their health declining as well and their ability to look after themselves, declining. So, you know, onset of depression, symptoms of anxiety going, ‘How am I going to do this on my own?’ was a big one, especially if the individual with MS also had mental health issues. (Service Provider, Female)

Importantly, in the context of COVID-19, carer participants rarely described this as an additional burden, instead, there was more recognition that this was a necessary change, whilst acknowledging it was a difficult challenge.

Social and relational impacts

In some interviews, participants reported impacts on their own relationships as pandemic-related constraints, such as lockdowns, increased living pressures and relationship stress. For other participants, the experience of restrictions was linked to isolation and loneliness, particularly for persons with MS.

So, my kids were off still doing their days because if they were working or whatever, but for me it was total isolation. So, it was very depressing. And your self-worth goes into your boots, I was lonely. (MS, Female, moderate disability)

And probably I'm seeing a family, you know, people are working from home, the carers are working from home because people can't go out as much possibly for all families, but especially ones with disability, and MS that it puts more strain on the relationship from just living in each other's pockets. (Service Provider, Female)

Participants in our interviews described a broad range of experiences, challenges and difficulties in response to the pandemic, some related to pandemic-related restrictions and the flow on effects of these and others related to concerns about contracting COVID-19 directly. While these concerns and impacts understandably dominated the descriptions of the COVID-19 experience, they were not solely defined by complaint or helplessness, rather we also heard in participant interview responses intended to promote coping and adaptation to very difficult circumstances.

Coping and adapting

Across interviews with persons with MS and carers, we heard examples of attempting to respond to the pandemic and related restrictions in helpful ways, such as by creating space and through routines and activities.

Creating space

In the idea of creating space through the pandemic, participants referred to the impact of lockdowns and trying to manage confinement, highlighting the importance of space, either in physically different locations or in finding space for self. We heard descriptions from participants about the importance of ‘getting out’, and for carers, the need for setting boundaries, finding respite, taking breaks, spending time apart and having space and time for themselves.

It's a little concerning, like all these issues. High risk for COVID. And, um, yeah, it's putting a bit more stress on us to be at home, like locked down 24/7 together. Um, so we've kind of been taking time apart, you know, for her to watch her TV shows and for me to play video games like we kind of separate into our own thing. (Carer, Female)

So consequently, she's sort of locked herself in since then and we haven't been out. And I get out by going shopping and I mean, shopping for food and that sort of thing. I don't buy big lots, so I go down three or four times a week. (Carer, Male)

Routines and activities

The above excerpts also highlight for some participants the importance of maintaining meaningful activities and finding alternative ways of meeting physical health and social needs. We heard in some interviews of persons with MS and carers the importance of maintaining routines and activities to meet these needs.

I would go, drive to the coffee shop, take my walking pole and get a takeaway coffee and go for a walk for half an hour, so I developed a routine in my day and that routine was what basically saved my mental health. Otherwise, I wouldn't have seen anyone. So that coffee man became so important. (MS, Female, moderate disability)

Overall, we heard from some participants examples of efforts to overcome initial challenges, disruptions or personal concerns, to navigate the pandemic in helpful and adaptive ways. Similarly, in some interviews, we also noted participants' efforts to recognize and appreciate positive experiences and unexpected benefits.

Appreciating positive experiences and benefits

In interviews with MS and carers and family members, and occasionally service providers, there was some recognition of positive experiences. This included, for example, experiences that offset challenges, such as service continuity rather than service disruption. A few participants also observed strengthening of relationships in the pandemic context. For others, benefits emerged in response to the pandemic, such as the use of technology for telehealth or to maintain social connection, as well as to help manage time and effort associated with face-to-face events.

Our access to caregiving has been great, sometimes they're all maxed up and everything, but they have been able to come. But auxiliary services sometimes didn't come like the house cleaning and things but that wasn't really an issue. (Carer, Female)

The fact that he can make an appointment with our GP and instead of having to use up all his energy available that day on just getting to the GP to have a meeting for just a script and that's it. The fact that he could have a five-minute conversation and it's done is just brilliant. A lot of the museums and the zoo's and things like that were online, and he was able to do that with the kids which was fantastic. (Carer, Female)

In interviews, we noted that there was an acknowledgement of positive experiences, and we also heard positive evaluations in appreciation and gratitude, through descriptive words such as *great, lucky, bonus, thankfully and brilliant*. Examples of coping and resilience were minimally present in service providers' observation, who tended to focus predominantly on the experiences of challenges and negative impacts on persons with MS and carers.

DISCUSSION

The current study brought together perspectives of persons with MS, carers and family members and service providers, to explore pandemic-related experiences of persons with MS and carers and family members. With limited research on MS carers during the COVID-19 pandemic,[7,16] this study used qualitative methods to gain a richer understanding of the MS carer experience during this time. We heard salient themes surrounding the challenges of disruptions, as well as impacts on health, relationships and resources. Yet we also heard examples of resilience, with persons with MS and carers finding ways to cope and adapt and appreciating positive experiences and benefits gained amidst the pandemic. Interestingly reports of positive adaptation tended to be absent from accounts of service providers, which may reflect a potential blind spot in their knowledge of the pandemic experience for persons with MS and their carers.

Many key themes identified here are reflected in previous studies, including disruptions due to COVID and the significant shift away from home services towards telehealth.[4,5,30] As with our research, MS participants in previous studies welcomed telehealth as a method of increasing access to services,[6] although concerns remain regarding ease of use for some people recognizing that it is not always an adequate replacement.[7] As the world adapts to the current pandemic and how it may unfold, further research is needed that gains a more nuanced understanding of telehealth use for the MS community, to ensure equitable development in response to diversity in ability, economic resources and metropolitan proximity.

In describing salient impacts of the pandemic, in the current study, the emotional, physical and mental health impacts for persons with MS partly echo previous research identifying experiences such as anxiety and stress[12–14] and COVID-related fear and concerns.[10–12] Unlike some studies,[14] there was not a strong theme of fatigue for persons with MS. This may be partly due to participant self-selection, where individuals with significantly low mood and fatigue are perhaps less likely to engage in interviews. However, there were not consistently strong reports of low mood, fatigue or sleep problems from carer and service provider participants about persons with MS. Rather, anxiety and stress were a more common theme for MS participants, but further research is needed to monitor changes as communities progress through different phases of the pandemic.

This study adds to the existing literature by providing greater insight into COVID-19 pandemic experiences for MS carers and families. Their experiences of additional demands due to service disruptions were highlighted by persons with MS, service providers and carers themselves. This is consistent with the experiences of carers with other conditions during COVID-19[20] as well as MS carers in previous studies.[7] Similar to impacts on persons with MS, carers and family members acknowledged stress and anxiety, as reported in previous research,[16] although unlike previous research,[7,22] carers and family members in the current study did not specifically report feeling less supported as a result of the pandemic. Nevertheless, reports of increased demands with reduced resources for MS carers add weight to the need for carer-centred interventions[22,23] that support resourcing and resilience. There is also a benefit in exploring the role of broader MS relational networks[19] and their experience in these times.

Previous research has identified factors that may shape different experiences of the pandemic, such as perceived COVID-19 risk and personal resilience,[10] coping and acceptance[17] and recognizing benefits as well as challenges of pandemic-related restrictions.[7,16] The current study provides some insight into possible facilitating strategies, such as creating physical or emotional space, developing routines and activities or through appreciation and gratitude. Some of these strategies have historically demonstrated potential benefits in other contexts.[31,32] This suggests a need to bolster MS community research and services that explore coping, adaptation, resilience and positive resourcing at multiple levels, including personally, relationally and systemically.

There are important limitations. First, interviewees predominantly indicated they were female, which may influence reported experiences and responses, including in coping and adaptation. Future studies should consider gender differences, as well as the impact of culture on care expectations and perceptions. In addition, while experiences were sampled across the country, adding to the diversity and richness of different experiences, it also impacts the consistency of findings. COVID-19 impacts such as service delivery, social isolation and disconnection from usual activities were reported more by participants experiencing significant lockdowns and restrictions, yet these experiences were subject to geographical and temporal variability as the pandemic progressed and health directives changed across the course of the study. Similarly, there were differences between groups in experiential variables (e.g., age, length of diagnosis, location), which may have introduced additional variability. This, along with the small sample size, limited any kind of subgroup analyses. As the pandemic evolves, future research may need to consider a more refined process for evaluating the impact of pandemic-related variables on persons with MS and their carers and family members, especially as we prepare for potential future pandemics.

CONCLUSION

Overall, the current study is relatively unique in providing a focused qualitative exploration of COVID-19 impacts and experiences of MS carers and family members, in addition to persons with MS. The findings have several implications. First, the study reiterates the value of MS relational networks, recognizing them as a valuable resource that needs to be resourced and considered in policy development, especially when preparing for future crises and chronic community stressors. Second, ensuring such preparedness requires strong investment into carer-centred interventions that bolster carer resilience and enhance their well-being. Finally, this study recognizes the importance of coping and adaptive responding that can build resilience, which service providers may miss if focusing extensively on challenges. To this end, service providers could more actively adopt a strength-based approach, recognizing and supporting these strategies when engaging with persons with MS and their carers. Investing in these changes will benefit both carers and persons with MS as community stressors unfold.

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CONFLICT OF INTEREST

The authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

The data set from the current study is not openly accessible as participant consent was given with the understanding that access to the data was only made available to the research team. Requests regarding the data set can be sent to helen.correia@murdoch.edu.au.

ETHICS STATEMENT

The study was approved by the Murdoch University Human Research Ethics Committee (Approval 2019/057). Participants provided informed consent before participation and provided verbal consent at the beginning of each interview.

DETAILS

Subject:	Pandemics; Mental health services; Mental health; Anxiety; Relatives; Coping strategies; Medical personnel; Social networks; Disruption; Health status; Coping; Caregivers; COVID-19; Multiple sclerosis; Resilience; Interviews; Fatigue; Participation; Medical research; Stress; Public health; Routines; Mental depression; Age groups; Qualitative research; Coronaviruses; Telemedicine; Disease transmission
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Developing a research community within an online healthcare feedback platform

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ProQuest document link

ABSTRACT (ENGLISH)

Introduction

Care Opinion is an online feedback platform supporting patients to author stories about their care. It is not known whether authors would be willing to be involved in improving care through research. The aims of this study were to explore the views and preferences of Care Opinion authors about joining an online research community and to pilot new research community functionality.

Methods

Five hundred and nine Care Opinion authors were invited to take part in an online survey in June 2019. Survey items included questions about participants' willingness to take part in research and their preferences for supporting processes. Data were analysed descriptively. Authors were invited to consent to join a research community and were asked to participate in three pilot studies.

Results

One hundred and sixty-three people consented to take part in the survey (32%). Participants indicated they would like to know the time commitment to the project (146, 90%), details about the organization carrying out the research (124, 76%) and safeguarding information (124, 76%). Over half indicated that they did not know how to get involved in healthcare research (87, 53%). Subsequently, 667 authors were invited to join the research community, 183 (27%) accepted, and three studies were matched to their expressed preferences for project attributes or organization type.

Conclusion

Many people who leave online feedback about their experiences of healthcare are also willing to join a research community via that platform. They have strong preferences for supporting University and NHS research. Eligibility and acceptance rates to join pilot research studies varied. Further work is needed to grow the research community, increase its diversity, and create relevant and varied opportunities to support research.

Patient or Public Contribution

Four members of the Safety In Numbers patient and public involvement and engagement (PPIE) group advised about survey development.

FULL TEXT

INTRODUCTION

There has been a marked increase internationally in healthcare organizations collecting and responding to feedback from patients about their experiences of health care using different methods.[1-5] Traditionally, people who have received treatment have had limited routes to provide feedback about the care they have received. For example, face-to-face feedback during episodes of care, surveys, letters and cards to care providers, the Patient Advice and Liaison Service and healthcare organization complaints interfaces have been the main routes through which feedback can be provided. New technologies, such as online platforms and social media, now provide formal and informal opportunities for patients to offer open feedback and recount often in-depth stories. These platforms can provide an understanding of patients' experiences during and after having received care to healthcare teams and to other patients.[6-8] One such online platform is Care Opinion, which supports patients to author stories about care they have experienced and offers the opportunity for staff to respond to those stories, which they do with varying levels of engagement.[9]

The growing popularity of online platforms such as Care Opinion indicates that people are willing to offer feedback about their care in this way. Patients have described being motivated to do so for a range of reasons, including improving the care they and others receive, improving healthcare services, publicly thanking and praising staff or a service, generally to empower patients or to inform other patients.[9,10] A recent study by Mazanderani et al.[8] found that those who leave feedback do so as an act of caring about other patients, and caring about the healthcare system and those working in it. Whilst it is established that some people are willing to give feedback to improve services, it is not known whether people who offer this type of feedback are willing to be further involved in improving health care. Given that improving healthcare services is a major motivator for offering feedback, it is possible that those who do so may be willing, if asked, to be further involved in healthcare improvement as participants or advisors to research.

Further, considering the widely reported difficulties in recruiting people into healthcare research studies,[11,12] exploring new and different routes to recruitment, especially those that may offer the ability to select participants based on their health conditions and their demographics, may offer a solution to many healthcare research projects. Routes to recruitment that do not involve placing an additional burden on stretched healthcare services may be of particular benefit because they would not require the input of clinical teams, for example, in identifying participants, or of healthcare administration staff in the distribution of study recruitment materials. Indeed, during the first two waves of the COVID-19 pandemic, most non-Covid related research was discontinued within the NHS, and it was not possible to access patients via healthcare organizations to take part in the research. It is now, more than ever, important that other viable recruitment options are identified.

Involving and engaging users of health care in the design and management of research can also be challenging for researchers, who may default to using established patient and public involvement groups convened by healthcare organizations.[13] There is an ever-deeper emphasis on the involvement of patients in healthcare services research to increase its quality and impact. The potential for patients and the public to be involved spans the different stages of research,[14] from developing priorities and setting the research programme,[15] during the development of research protocols, assessing the appropriateness of research instruments,[16] and of course as participants. Further opportunities are available during the data analysis and write-up stage to ensure that emerging themes are interpreted from lay perspectives.[17] There is potential for those who are willing to provide feedback about their experiences online to contribute to research, either as participants, advisers or co-researchers, to improve care. The aims of this study, therefore, were to:

- (1)
  Examine the views and preferences of Care Opinion authors (people who have shared their stories on the Care Opinion platform) about joining an online research community;

- (2)
  Assess whether those who join a research community then agree to take part in research studies, if eligible;

- (3)
  Pilot new research community functionality within the Care Opinion website.

MATERIALS AND METHODS

We undertook a quantitative study using an online survey method and used the results to inform the development and pilot testing of research community functionality within the Care Opinion platform.

Survey development

The survey was developed by a team of three researchers with backgrounds in applied health and social research (B.F. and B.K.) and psychology (J.K.O'H.), and a Care Opinion team member with a background in clinical medicine, public health and health services research (J.M.). Four members of the Safety In Numbers patient and public involvement and engagement (PPIE) group advised about survey items. Survey items were developed based on a review of the literature about frameworks supporting patient and public involvement in research,[18] and included items about the participants, for example, their age, ethnicity, gender, and employment status. Survey items also included questions about participants' perception of the importance of healthcare research, their willingness to take part in research, the research-related activities they would be willing to get involved in, the types of research organizations they would be willing to engage with, whether they would require payment to be involved in research, and their preferences for processes supporting research involvement. Answer options comprised Likert scales, yes/no options and open questions. Two sets of questions, one measuring prosocialness[19] and another exploring quality in service-user involvement,[20] were adapted, although the results of those items are not reported here. Four people piloted the survey and gave feedback on the length of time the survey took to complete and the wording of the survey items. The survey was programmed so that respondents were not forced to answer every question.

Participants and recruitment

Participants were adults who had submitted a story to Care Opinion during May and June 2019 and had consented to further contact from the Care Opinion team. To reduce the risk of causing distress, we excluded authors who described end-of-life care or care resulting in serious negative effects on physical or mental health. A recruitment email was sent by Care Opinion inviting people to take part in the study with a link to the questionnaire hosted on the SurveyMonkey site. A link to an information statement about the study and a consent form was embedded at the beginning of the survey. In total, the invitation email was delivered to 509 care opinion authors in July 2019. A reminder email was sent 2 weeks later, and the survey remained open for 1 month.

Data analysis

Descriptive statistics, including counts of responses to questions and associated percentages, were computed using the software programme SPSS version 27. Missing data were not deducted from the total possible responses. Open questions were explored using content analysis. Responses were grouped into categories and counts, and percentages of categories were calculated. In this article, we report responses to a subset of survey items.

Piloting the research platform within Care Opinion

Following the survey analysis, we used the results to inform the design of a ‘proof of concept’ research community on the Care Opinion platform. We added a feature to the platform enabling authors to join the community and express their preferences for communication. A further feature enabled Care Opinion staff to send project invitation emails to research community members, allowing invitees to accept or decline the invitation as they chose.

To create an initial community population, we defined a sample of adults who had posted a story on Care Opinion between February and April 2020, and we emailed those authors with an invitation to join the pilot community. We sent an email a reminder after 1 week. Subsequently, we invited community members to participate in three different types of research study over the following 9 months. The first study sought unselected participants to respond to a survey about medical uncertainty in the emergency department. The second sought interviewees for a study of experiences of patients and family members involved in a serious incident in an NHS hospital or mental health service in England. For both of these studies, we sent automated email invitations via Care Opinion to community members whose consent preferences matched the study (e.g., research organization, type (survey, interview), location (online, phone, home visit). For the second study, eligibility was set out in the invitation. The third study required interviewees who were over 75 and had experienced a recent hospital stay, or they were a family member or carer of such a person. Given the very narrow requirements of this study, and after an initial automated email invitation to research community members detailing eligibility produced disappointing results, we undertook a wider search of stories posted in the past year (i.e., beyond our research community cohort) which was then manually filtered to identify potentially suitable authors for an email invitation.

RESULTS

A total of 163 people (32%) consented to take part in the survey. Of these, 110 were female (67.4%) and 52 were male (31.9%). This reflects the population of Care Opinion authors, which has a higher proportion of women compared to men (64% vs 36%). Most respondents were white (151; 92.6%), and over a third were retired (57, 34.9%). Respondents were of different ages, with 55–64 being the largest age group. A quarter of respondents were under 45 years old. The majority of respondents were over 55 years old (91, 55%). This compares well to the population of Care Opinion authors, where the median age is 55. The demographics of survey respondents are shown in Table 1. Most participants had used the Care Opinion site to leave a story about their own care (115; 70.6%), 37 had left a story about a relative's care (22.7%), and the remainder had posted a story about the care of a partner (8, 4.9%) or friend (1, 0.6%).

Table 1 Demographics of survey respondents (base *n* = 163)

	Number (%)
Gender	
Female	110 (67.4)
Male	52 (31.9)
Missing	1
Age range	
18–24	4 (2.5)
25–34	18 (11.0)
35–44	20 (12.3)
45–54	30 (18.4)
55–64	50 (30.7)
65+	40 (24.5)
Missing	1
Ethnicity	
White	151 (92.6)
Asian or British Asian	5 (3.1)
Mixed race	2 (1.2)

Black or Black British	1 (0.6)
Other	3 (1.8)
Missing	1
Employment status	
Retired	57 (34.9)
Full-time employed	52 (31.9)
Part-time employed	24 (14.7)
Not currently employed	10 (6.1)
Homemaker	3 (1.8)
Education	2 (1.2)
Other (e.g., carer, volunteer, self-employed and disabled)	15 (9.2)

Views about healthcare research

The vast majority of the 161 people who answered the question about whether healthcare research was important, reported that it was very (149, 91.4%) and fairly (12, 7.4%) important. Table 2 shows responses to three further questions about taking part in healthcare research. The majority either agreed or strongly agreed that taking part in healthcare research improves health care (156, 95.7%), and agreed or strongly agreed that patient and public involvement in healthcare research is important (160, 98.2%). Over half of respondents either agreed or strongly agreed that they did not know how to get involved in healthcare research (87, 53.4%).

Table 2 Responses to questions about taking part in healthcare research (base *n* = 163)

	Strongly agree	Agree	Neither agree nor disagree	Disagree	Strongly disagree	Missing
Taking part in healthcare research improves healthcare	111 (68.1%)	45 (27.6%)	5 (3.1%)	2 (1.2%)	0	0
Patient and public involvement in healthcare research is important	130 (79.8%)	30 (18.4%)	1 (0.6%)	0	0	2

I don't know how to get involved in healthcare research	22 (13.5%)	65 (39.9%)	39 (23.9%)	28 (17%)	7 (4.3%)	2

Taking part in a research project

Answers to questions about how authors would respond to an invitation to take part in a research project are in Table 3. Most respondents reported that they would probably or definitely not immediately opt out (132, 80.9%). Most respondents would probably or definitely agree to help University research (122, 87.1%) and even more would probably or definitely agree to help NHS research (151, 92.6%), whilst less than half would probably or definitely agree to help research by private companies (71, 43.5%). The majority would be happy to consider invitations about any kind of research (103, 63.2%), but NHS research received the most support and private companies the least.

Table 3 How Care Opinion authors would respond to an invitation to take part in a research project (base *n* = 163)

	Definitely not	Probably not	Not sure	Probably	Definitely	Missing
I would opt out immediately	68 (41.7%)	64 (39.3%)	22 (13.5%)	4 (2.5%)	1 (0.6%)	4
I would only want to receive invitations about research which related to the story I posted	35 (21.5%)	42 (25.8%)	24 (14.7%)	49 (30.1%)	12 (7.4%)	1
I would agree to help university research	2 (1.2%)	2 (1.2%)	16 (9.8%)	88 (54%)	54 (33.1%)	1
I would agree to help NHS research	4 (2.5%)	1 (0.6%)	6 (3.7%)	77 (47.2%)	74 (45.4%)	1
I would agree to help research by private companies	10 (6.1%)	16 (9.8%)	65 (39.9%)	55 (33.7%)	16 (9.8%)	1
I would be happy to consider invitations about any kind of research	4 (2.5%)	17 (10.4%)	39 (23.9%)	68 (41.7%)	35 (21.5%)	0
I would only help research if I could do it online	21 (12.9%)	29 (17.8%)	59 (36.2%)	36 (22.1%)	17 (10.4%)	1

Information required about the research

The information participants would like before taking part in the research is in Table 4. The most frequently selected type of information they would like was the time commitment to the project (90% of respondents selected this option), followed by details of the organization carrying out the research, safeguarding information, travel commitments and timelines.

Table 4 Information respondents would like about the research before taking part (base *n* = 163)

Information respondents would like before deciding whether to be involved	Number (%)
Your time commitment to the project	146 (89.6)
Details about the organization carrying out the research	124 (76.1)
Safeguarding information for you if you took part	124 (76.1)
Travel commitments/locations	119 (73)
Timelines for the project	111 (68.1)
Key people involved in research	74 (45.4)
Details about payments for your contribution to the project	51 (31.3)

Note: Participants could select multiple options to this question.

Respondents' views about using the Care Opinion platform for research

We asked a series of questions about preferences for taking part in research via the Care Opinion platform to guide the design of the research platform. Respondents' answers are in Table 5. The majority of respondents agreed or strongly agreed that people should be able to opt-in or out of being asked to help with research (156, 95.7%), that people should be able to limit how often they are asked to help with research (130, 79.7%), that people should be able to see what kind of organization is asking for their help (158, 96.8%), and that people should be able to read information about the research before they decide whether to help (153, 93.9%).

Table 5 Respondents' views and preferences for taking part in healthcare research (base *n* = 163)

	Strongly agree	Agree	Neither agree nor disagree	Disagree	Strongly disagree	Missing
People who share a story on Care Opinion may have experiences which would help research studies	112 (68.7%)	44 (27%)	6 (3.7%)	0	0	1
People who share a story on Care Opinion would not want to help research	5 (3.1%)	1 (0.6%)	38 (23.3%)	76 (46.6%)	41 (25.2%)	2

I would like to see people who share stories on Care Opinion also helping research	80 (49.1%)	64 (39.3%)	17 (10.4%)	0	0	2
I would like to be able to see if a particular story author has also helped a research study	41 (25.2%	57 (35%)	48 (29.4%)	14 (8.6%)	1 (0.6%)	2
If I helped a research study, I would like people to be able to see that on Care Opinion	45 (27.6%)	51 (31.3%)	47 (28.8%	16 (9.8%)	2 (1.2%	2
I don't think Care Opinion should be inviting people to help research	4 (2.5%)	3 (1.8%)	17 (10.4%)	73 (44.8%)	64 (39.3%)	2
I am more likely than other people to help research because I am an author on Care Opinion	18 (11%)	53 (32.5%)	58 (35.6%)	22 (13.5%)	10 (6.1%)	2
People should be able to opt in or out of being asked to help	94 (57.7%)	62 (38%)	3 (1.8%)	2 (1.2%)	1 (1.2%)	1
People should be able to limit how often they are asked to help	60 (36.8%)	70 (42.9%)	19 (11.7%)	9 (535%)	3 (1.8%)	2
People should be able to see what kind of organization is asking for their help	110 (67.4%)	48 (29.4%)	3 (1.8%)	0	0	2
People should be able to read information about the research before they decide to help or not	110 (67.5%)	43 (26.4%)	6 (3.7%)	3 (1.8%)	0	1

Types of research people would take part in

More than 80% of respondents answered the open question about the research they would support, and some offered multiple types of research in their answers. Responses are in Table 6. Many respondents (59, 36.2%) gave answers indicating that they would support research relevant to their own condition or story, or a specific health condition such as arthritis, mental health conditions, types of cancer and chronic pain or ageing. Respondents also indicated they would support research relating to services, such as rehabilitation, improving services in care settings such as hospitals, and management of the NHS and social care (26, 16%). Others indicated they would like to help with research that supported the patient experience, patient satisfaction, perceptions of treatment and what matters to patients and families (13, 8%). A small number would support research focussing on a particular group, such as children, older people and ethnic minorities (5), whilst four people indicated they would support a particular research type, such as qualitative, surveys or research to improve methods. The remainder of responses (3) related to health promotion, and stem cell research.

Table 6 Types of research participants would and would not help with and concerns about being asked to take part (base *n* = 163)

Research participants would help with	Number (%)	Research participants would not help with	Number (%)	Concerns about being asked to take part in research	Number (%)
Research relevant to own condition or story	59 (36.2)	None	84 (51.5)	None	70 (42.9)
None/unsure/any	38 (23.3)	Profit-making/private sector	8 (4.9)	Resources	18 (11.0)
Research related to services	26 (16.0)	Clinical, invasive or drug research	7 (4.3)	Privacy	15 (9.2)
Research supporting patient experience	13 (8.0)	Research that will not change anything	3 (1.8)	Opting in and out	6 (3.7)
Research focussing on specific group	5 (3.1)	Animal research	3 (1.8)	Legitimacy	5 (3.1)
Research using a specific method	3 (1.8)	Learning disability research	2 (1.2)	Ethics	4 (2.5)
Other	3 (1.8)	Children/childhood research	2 (1.2)	Lack of impact	2 (1.2)
		Time-consuming research	2 (1.2)	Distract from Care Opinion's main objectives	1 (0.6)
		Addiction research	1 (0.6)		

The majority responded to the open question about the research they would support (69%), and the answers covered more than one type of research. Most indicated there was no research they would not help with (84, 51.5%). Eight people responded that they did not want to support profit-making or private-sector research (4.9%), and seven people did not want to support clinical, invasive or drug research (4.3%). Other responses included research that would not bring about change or that they could not contribute to. Three people did not want to support research involving animals, two people did not want to support learning disability research and two people did not want to support research with children or about childhood. Two people indicated that they would not help with research that took too much time. Finally, one person did not want to help with addiction research.

Nearly three-quarters (71.2%) of participants answered the open question about whether they had concerns about being asked to take part in research, and their answers covered multiple concerns. Many had no worries (42.9%). Eighteen people had concerns relating to resources, for example, travel, the time it would take, or being bombarded with requests (11%). Fifteen people had worries about privacy, for example, being identified or judged, and data security (9.2%). Seven people had concerns about being able to opt-in or out (3.7%). Other concerns related to the legitimacy of organizations carrying out the research, ethical issues, such as other people's motivations for taking part in research and the emotional burden on participants, that the research would lack impact or that research would impact Care Opinion's main objectives.

RESULTS OF THE PILOT TEST OF THE RESEARCH COMMUNITY

Of the 667 authors invited to join the research community, 183 (27.4%) accepted, and 5 (0.7%) declined. On joining, and based on survey responses, authors were able to set their preferences for participation (research organization type, acceptable communication channels). We invited members of this community to participate in three research projects. For the first two research studies, we sent automated email invitations via Care Opinion to community members whose expressed preferences matched the project attributes. For the first research project, a survey of views on medical uncertainty, we invited 128 research community members whose preferences matched the project. Of these, 67 authors (52.3%) accepted the invitation, and 49 (38.2%) went on to complete the survey. For the second, a more specific project, we invited 139 members willing to receive invitations from a university, of whom 11 authors (7.9%) were eligible and accepted the invitation whilst 30 (21.5%) declined. This project required participants to have experience a ‘serious incident’ in their care.

For the third project, seeking people over 75 with a recent hospital episode, or their carers, 23 research community members were invited, of whom 2 (8.7%) accepted an invitation. To extend the sample, we undertook a wider search of Care Opinion stories (not restricted to research community authors) posted in the past year, which included the words (‘elderly’ or ‘old*’) and (‘ward’ or ‘discharge’). This resulted in 441 candidate stories, of which 52 were tagged as relevant by the researcher. The authors of these stories were invited to participate by email, resulting in seven further authors (13.5%) agreeing to interview.

DISCUSSION

This project examined the views and preferences of Care Opinion authors about joining an online research community and pilot-tested a research community within the Care Opinion platform. It found that survey respondents perceived healthcare research to be important, that patient and public involvement in research is important, that the majority would be willing to help with any type of research and thought that people who shared a story on Care Opinion had experiences that would help research studies. People reported that they should be able to see the organization asking for their involvement and that they should be able to read information about research before deciding. Respondents were more positive about helping NHS and university research than other types of research, and respondents expressed clear preferences for being able to opt-in and out and limiting the number of invitations received. They wanted to know the expected time commitment to a project in advance and about safeguarding information. Based on the positive responses of Care Opinion authors about being involved in research, the results of the survey were used to develop a pilot research community platform. Over a quarter of those invited joined the community and over a third of those invited to take part in the first study did so. Finding participants for studies with more specific inclusion criteria was more challenging.

Motivation and barriers to taking part in research

This research found that whilst some survey respondents would support research relevant to their own condition, others were happy to support any research, and some who went on to join the research community did indeed respond to a request to respond to a general survey during the pilot. Previous research has found that people's motivation to take part in research is primarily altruistic.[21] However, Bradley et al.[22] found that patients joining research teams were motivated by their own individual needs and wanted to get involved in research that was relevant to their own health condition. Taking part in research was viewed by their participants as contributing towards the public good and the trustworthiness of researchers was judged based on their organization and profession. Further, Dixon-Woods and Tarrant (2009)[23] discussed how people may be reluctant to join research if they are sceptical of the intentions of the researchers, especially when there is a cost to them in the form of, for example, providing personal data or undergoing tests. Respondents in this study indicated that they were more positive about supporting research conducted by the NHS or a university than research by private organizations, which emphasizes the importance of trust to Care Opinion authors. The involvement of patient organizations has been found to have a positive impact on participant recruitment,[24] so clearly, the perceived legitimacy of research is influenced by the organizations conducting and supporting it.[25] Indeed, a recent review indicated that being part of a trusted research team is crucial to successful patient involvement in research.[26] It is possible, then, that the overall legitimacy of such a research community may be damaged by allowing access to organizations outside the UK NHS and higher education. Further consultation around this area will be needed before wider access is considered.

Participants in the survey were overwhelmingly positive about being involved in research, yet when authors were invited to join a community, only 27% did so. The dissonance between expressed intentions to get involved in research and people's actions when invited to do so align with behavioural models, such as the theory of planned behaviour,[27] which proposes an ‘intention-behaviour gap’ between the intention, formed through attitudes, norms and perceived behavioural control, and the desired action. Achieving a conversion rate of fewer than one-third of invited authors indicates that barriers to joining for those who are willing need to be fully understood and addressed. Furthermore, as few authors matched our third pilot project—aimed at people over 75 with a recent hospital episode—the community would need to expand to yield sufficient participant numbers for projects with detailed and specific eligibility criteria. More consideration is also needed about facilitating certain groups, such as older populations, to be involved. Iteratively developing recruitment methods may support increased participation,[28] and involving members of the research community in developing these methods will be important. A systematic review has demonstrated patient and public involvement to improve participant recruitment to studies, particularly if the people involved have experience relevant to the subject of the research, such as the experience of a particular health condition.[29]

Nevertheless, less than 1% of invited authors declined to join the research community, indicating that there was limited direct opposition to doing so, rather than that there might be a lack of internal or external motivation to join on receipt of the invitation. Similarly, how authors are matched to research opportunities requires further consideration. The completion rate for the first pilot study, a general survey, was 38%, however, for the second study, for which the eligibility criteria were specific and related to experiencing a serious incident in care, only 8% accepted the invitation to take part once those criteria had been communicated, and many more (22%) declined. Further information is needed to understand the reason for those declines. It is, of course, possible that more challenging topic areas, particularly for people who have had negative experiences of care or, indeed, have been harmed by their care, may lead to reluctance to take part. In addition, it was not possible to know the health status of people invited to join the community or to take part in pilot studies, so fluctuating health may also have been a barrier.

More than three-quarters of survey respondents indicated that information about safeguarding would be required before they agreed to take part in a study, and some also had worries about privacy, for example, being identified or judged. Interestingly, there were a large number of definitive declines to the second pilot study about serious incidents, where potential participants may have had fears about the consequences of being identified. Other studies have found that a range of fears, including confidentiality, act as barriers to taking part in research, albeit in clinical trials.[30] Some survey respondents in this study also had practical concerns about being involved in research or would want information about practicalities involved in taking part, such as time commitments and travel arrangements. Such practicalities can be a barrier to participation, even for those who are motivated to take part.[31]

Reducing the burden of taking part should therefore be a priority for researchers.

Extending the role of online feedback platforms

This work has demonstrated that care feedback platforms are a potential route to engage people in research and can be used to augment other methods of data collection and analysis. Internationally, online feedback routes are increasingly available,[32] although only to those who have the resources to be able to access them. Dudhwala et al.[33] drew a distinction between feedback routes that are approved by healthcare organizations and used by them to solicit feedback, and those that are unofficial and not actively used to ask for or review patient feedback. Care Opinion primarily falls into the first of these categories and it is possible that its more established role as a broker of patient feedback heightens trust. As the number of people leaving online feedback grows, the opportunities to involve contributors are also increasing. Improving care is a strong motivation for sharing online feedback, so it is not surprising that our study found that people who posted online feedback also thought that healthcare research is important and that it can improve care. Other research has shown that the motivation for leaving online feedback is to care for or empower other patients and to care for the NHS.[7,8,10] Care Opinion authors in this study seemed similarly motivated. However, over half of survey respondents did not know how to get involved in healthcare research, indicating that platforms such as Care Opinion could play a valuable role in linking those willing to give their opinions and recount their experiences of healthcare with organizations needing access to participants to deliver their research. There are obvious caveats for researchers to consider, including the need to appraise potential bias introduced by recruiting solely from those who are already willing and have the confidence to author stories online about their care. Further, trust in research conducted via an online feedback platform needs to be explored, especially amongst healthcare staff who can perceive online feedback platforms poorly, for example, as overly negative and not useful in improving care.[34]

Recommendations for future research

This research was conducted within the National Institute for Health and Care Research Yorkshire and Humber Patient Safety Translational Research Centre, and more feasibility work needs to be conducted before broadening the reach of the platform to other researchers. To build on this research, further work should take place to increase the number of authors who accept an invitation to join the community and to increase motivation to take part in research by providing interesting and varied research involvement and participation opportunities. Care needs to be taken to increase the diversity of the research community beyond those who responded to the survey, who were majority white and over 55. Finally, to fully develop the research community, more work needs to be done, based on the results of the survey reported here, to develop functions to allow Care Opinion authors to set additional and more specific preferences about the type of research they wish to be involved in, which will support better-targeted research invitations.

Limitations

Those who took part in the survey were mostly female, white, retired or employed, and 55 years or older, which is not representative of the population as a whole, although the majority of healthcare users are older and female, and participants were more representative of the population of Care Opinion authors. Demographics of non-responders to the survey are not available, which is also a limitation. In addition, whilst the research community was piloted within Care Opinion, a formal feasibility study was not conducted so the experiences of those who took part in the pilot testing of the research functionality and subsequent research projects were not explored. The three pilot research invitations were limited in that they invited Care Opinion authors to be participants in research, rather than to be involved in the co-production of research or other involvement activities. This work was conducted with people who had experienced care in the UK, which is predominantly provided by the National Health Service (NHS). Levels of satisfaction with the NHS at the time of data collection were relatively high (60% very or quite satisfied),[35] which might have influenced people's willingness to be involved in NHS-based research. Satisfaction with the NHS has since fallen.

CONCLUSION

Many people who leave online feedback about their experiences of health care on the Care Opinion platform are also willing to join a research community via that platform and take part in the research. They have strong preferences for supporting NHS and university research. Acceptance rates to join pilot research studies varied, and further work is needed to grow the research community, increase its diversity, create relevant and varied opportunities and reduce barriers to taking part.

AUTHOR CONTRIBUTIONS

Beth Fylan drafted the manuscript, analysed the data and contributed to research and online community design. James Munro drafted the manuscript, contributed to the research design, led online community development and facilitated the research pilot projects. Jane K. O'Hara led the research and contributed to online community development. Binish Khatoon drafted the research protocol, drafted the survey and commented on the manuscript. Rebecca Lawton advised on online community development, oversaw the pilot research projects and commented on the manuscript.

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CONFLICT OF INTEREST

James Munro is Chief Executive Officer of Care Opinion CIC.

DATA AVAILABILITY STATEMENT

The survey data reported in this article are not publicly available to meet the terms of ethical approval for the study and to protect participant privacy.

ETHICS STATEMENT

Ethical approval was granted by the Biomedical, Natural, Physical and Health Sciences Research Ethics Panel at the University of Bradford on 10/04/2019—reference E721.

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Development and early-stage evaluation of a patient portal to enhance familial communication about hereditary cancer susceptibility testing: A patient-driven approach

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ABSTRACT (ENGLISH)

Introduction

Genetic testing for hereditary cancer syndromes (HCSs) can improve health outcomes through cancer risk mitigation strategies. Effective communication between tested individuals and their family members is key to reducing the hereditary cancer burden. Our objective was to develop a patient portal to improve familial communication for patients undergoing HCS genetic testing, followed by an early-phase evaluation.

Methods

The portal was developed following the completion of 25 semistructured interviews with individuals having undergone HCS susceptibility testing at BC Cancer. Following initial development, we recruited patients and healthcare providers to provide critical feedback informing portal refinement. Quantitative feedback was summarized using descriptive statistics, and qualitative feedback was synthesized by two reviewers who engaged in iterative discussion within the research team to prioritize recommendations for integration.

Results

The patient portal includes four key components consisting of (a) targeted educational information about hereditary cancer and HBOC syndrome associated risks and testing process overview, (b) a general frequently asked questions 'FAQ' page informed by the qualitative interviews, patient partner feedback, and consultation with the HCP, (c) guidance to support familial communication including a video developed with a patient partner describing their lived experience navigating the communication process and (d) a series of lay summaries of genetic test findings to support information transfer among family members. Thirteen healthcare providers and seven patients participated in user testing. Domains within which participant recommendations were provided included presentation, educational content and process clarification.

Conclusions

This investigation demonstrates the value of continual integration of patient and provider preferences through the development of tools endeavouring to assist with complex genomics-informed decision-making. Our work aims to broaden the population-wide impact of HCS testing programs by improving communication processes between probands and their potentially affected family members.

Patient or Public Contribution

This work involved a patient partner who was actively engaged in all aspects of the research investigation including protocol development, review and editing of all study documentation (including that of the previously published qualitative investigation), interpretation of results, as well as reviewing and editing the manuscript. Patient partners and healthcare professionals were recruited as research participants to provide critical feedback on the patient portal.

FULL TEXT

INTRODUCTION

Early detection of hereditary cancer syndromes (HCSs) has the potential to reduce cancer burden through genomics-informed prevention and treatment.[1–5] HCSs, such as Hereditary Breast and Ovarian Cancer (HBOC) syndrome, Lynch syndrome, and Li-Fraumeni syndrome explain a substantial proportion of cancer diagnoses.[3,6,7] The detection of pathogenic germline variants within families commonly involves the use of multigene sequencing.[1,7] BC Cancer, the sole publicly reimbursed cancer service provider for a Canadian province (British Columbia [BC]), offers genetic testing through the Hereditary Cancer Program (HCP) for individuals with a suspected HCS (throughout BC and Yukon territory).[7]

To generate patient benefit from HCS screening programs, communication between probands (the first tested individual within a family[5]) and their biological relatives is critical.[6,8] Family members who are made aware of the presence of an HCS may choose to initiate cascade (presymptomatic) genetic testing and if indicated, undertake recommended cancer risk reduction strategies (e.g., enhanced screening frequency or prophylactic surgical interventions).

The value of interfamilial communication is not only pertinent to positive genetic test findings. In the event of negative results, biological relatives may either be reassured about their cancer risk,[9] invited to have genetic testing themselves if they remain eligible despite their family member's results, or receive increased cancer screening recommendations based on the family history of cancer alone. For these reasons, interfamilial communication about test results and familial risk is critical to ensure accurate awareness of cancer predisposition.

Currently, in Canada, interfamilial communication is a necessary prerequisite to cascade testing and reducing concomitant disease burden. Given jurisdiction-specific privacy laws preventing testing facilities from contacting potentially affected family members directly,[9–13] it is the responsibility of tested individuals to initiate communication about results and eligibility for cascade testing with their family members. Although some jurisdictions have implemented direct contact approaches where communication about testing eligibility is led by testing facilities, such policies raise ethical and legal considerations. Some question whether a direct approach has the potential to cause undue distress among potentially affected family members or disrupt interfamilial relationships.[14]

Within jurisdictions where cascade testing is facilitated through interfamilial communication of a positive finding, it is well established that probands face a variety of challenges throughout the communication process with family members.[15] Strained or distant relationships, limited genetic literacy, and lack of understanding about cascade testing have been identified as barriers to effective communication.[15–17] Conversely, female gender, closer kinship, and the perception that family members will respond favourably to the information are known facilitators of familial communication and results sharing.[18,19] Due in part to known barriers to communication, cascade testing within affected families remains suboptimal.[5,6,17,20,21] The development of strategies that respond to communication challenges necessary for ensuring the success of hereditary cancer testing programs aiming to reduce familial cancer burden.

Patient-facing decision support tools and educational resources present an opportunity to enhance familial communication for HCS testing. Decision support tools are increasingly being developed to address informational complexity and uncertainty and to assist patients with decisions related to genetic testing, preferences for the return of results and recommendations following the return of results.[22] For example, resources have been developed and evaluated to guide genomics-informed reproductive decision-making,[23,24] to clarify preferences for the return of incidental or secondary findings,[25–28] as well as to assist individuals deciding whether to undergo genetic testing.[29–31] With an established need to enhance cascade testing for HCSs, interventions have been developed and evaluated specifically for *BRCA1* and *BRCA2* testing and other familial cancer syndromes.[13] Such interventions include but are not limited to multistep skills-building sessions with genetic counsellors, informational booklets, webinars, and motivational interviewing.[18,32–34]

Building off existing evidence, we sought to engage patients and healthcare providers at BC Cancer to develop a patient portal to identify and mitigate barriers to familial communication. A primary feature of this investigation was to develop a resource for independent use by patients undergoing HCS testing at BC Cancer as an adjunct to genetic counselling appointments, for feasible implementation into standard care.

We developed and conducted user testing of a patient portal to support individuals undergoing HCS susceptibility testing at BC Cancer's HCP. For the purposes of early-stage development and refinement within this clinical context, the patient portal was developed for individuals with suspected HBOC, receiving *BRCA1* and *BRCA2* genetic testing, who account for the largest proportion of patients referred to the HCP (approximately 80%). Given the high throughput of *BRCA1* and *BRCA2* testing within the HCP, this clinical context was determined to be appropriate within which an initial tool could be developed, evaluated, and expanded upon for feasible clinical implementation at BC Cancer.

MATERIALS AND METHODSBackground

The portal was developed through a multiphase, patient-oriented approach. We first conducted 25 semistructured qualitative interviews with individuals having undergone HCS susceptibility testing at BC Cancer. Detailed methods and results of the qualitative investigation are published elsewhere.[15] Following interviews, two qualitative analysts summarized (1) key barriers to communication, (2) recommended strategies to support patients communicating genetic testing information to their families and (3) reported preferences for the design, content and structure of a patient portal.

Patient portal development

Informed by the qualitative investigation, the patient portal was developed to respond to three prioritized patient portal elements, described in Table 1. Briefly, participants hoped for (1) increased support throughout the entire testing and return of results trajectory, (2) a resource for accurate information about testing, results and HCS management and (3) and multipronged, informed guidance about communicating a diagnosis of an HCS to family members. Among the latter, suggested strategies to enhance communication with family included guidance about initiating conversations with family members, as well as recommendations for managing attempts at communication perceived as unfruitful or challenging.

Table 1 Key portal features informed by qualitative investigation

Participant prioritized portal element	Patient portal integration	Portal page
Support throughout the pretesting, testing, return of results and communication with family trajectory	Introductory educational information to explain why testing has been recommended as well as potential benefits and outcomes Flow diagram to explain to patients the process of genetic testing Content tailored to each stage of the testing process (e.g., before testing, testing process, results, patient and familial recommendations following the return of results and communicating with family about findings)	Home
A single educational resource enhancing access to accurate information about testing, results and HCS management	Educational material (*BRCA1* and *BRCA2* genes, common terminology, population level risk estimates for individuals with and without HCS) Benefits of testing (e.g., risk reduction strategies for affected families) Links to external educational resources	Home For Patients Talking to Your Family
Guidance about communicating a diagnosis of a hereditary cancer syndrome to family members	Brief informational video presenting a patient experience communicating with family about their hereditary cancer syndrome diagnosis Written guidance about how to initiate conversations with family members Key benefits to communicating with family members about test results Email template text to facilitate written communication family members Lay summary of results for patients and their family members	Talking to Your Family Results summary pages ‘Discussing genetic testing with your family’ email template

Abbreviation: HCS, hereditary cancer syndrome.

The research team partnered with a professional web developer, located in Vancouver, British Columbia to build the online portal platform.[35] Content was co-developed by the multidisciplinary research team of medical oncologists, genetic counsellors, medical geneticists, health services researchers and a patient partner. The patient partner (C. H.) contributed to each aspect of the research project, including but not limited to study protocol development, critical review of study documentation, interpretation of results, portal content development, and revision and outcomes dissemination.

User testing instrument and development

User testing feedback was collected using an online, REDCap survey.[36] The survey included six questions adapted from a published instrument that applied an ordinal scale to elicit critical feedback about portal features.[25,37] The survey sought feedback about clarity, length and amount of information presented, and the presentation of risks and benefits. Participants were also asked to respond to a series of open-ended questions about key portal components (Supporting Information). Finally, developed in collaboration with genetic counsellors, HCP co-directors and our patient partner, participants were presented with three examples of a lay summary of test results and asked to provide feedback for improvement.

User testing participant recruitment

Our sampling frame for user testing included patients and healthcare providers recruited from BC Cancer between February and March 2022. Eligible patient participants were members of BC Cancer's Patient and Family Experience Program, a program that invites individuals experienced with cancer to participate in research investigations in a patient partner role. Potential participants were recruited via an online newsletter. Eligible healthcare providers included medical oncologists, genetic counsellors, geneticists and those involved in clinical and leadership roles at BC Cancer's HCP. Using a convenience sampling approach, the research team generated a list of eligible healthcare providers to approach for participation, via email. Potential participants who expressed interest in providing portal feedback first completed an online consent form through REDCap. Following the provision of electronic consent, participants were then given a link to the patient portal as well as the online feedback survey, also administered using REDCap. Patient partners were provided with an honorarium of $75.00 CAD. Healthcare provider participants were not provided with an honorarium.

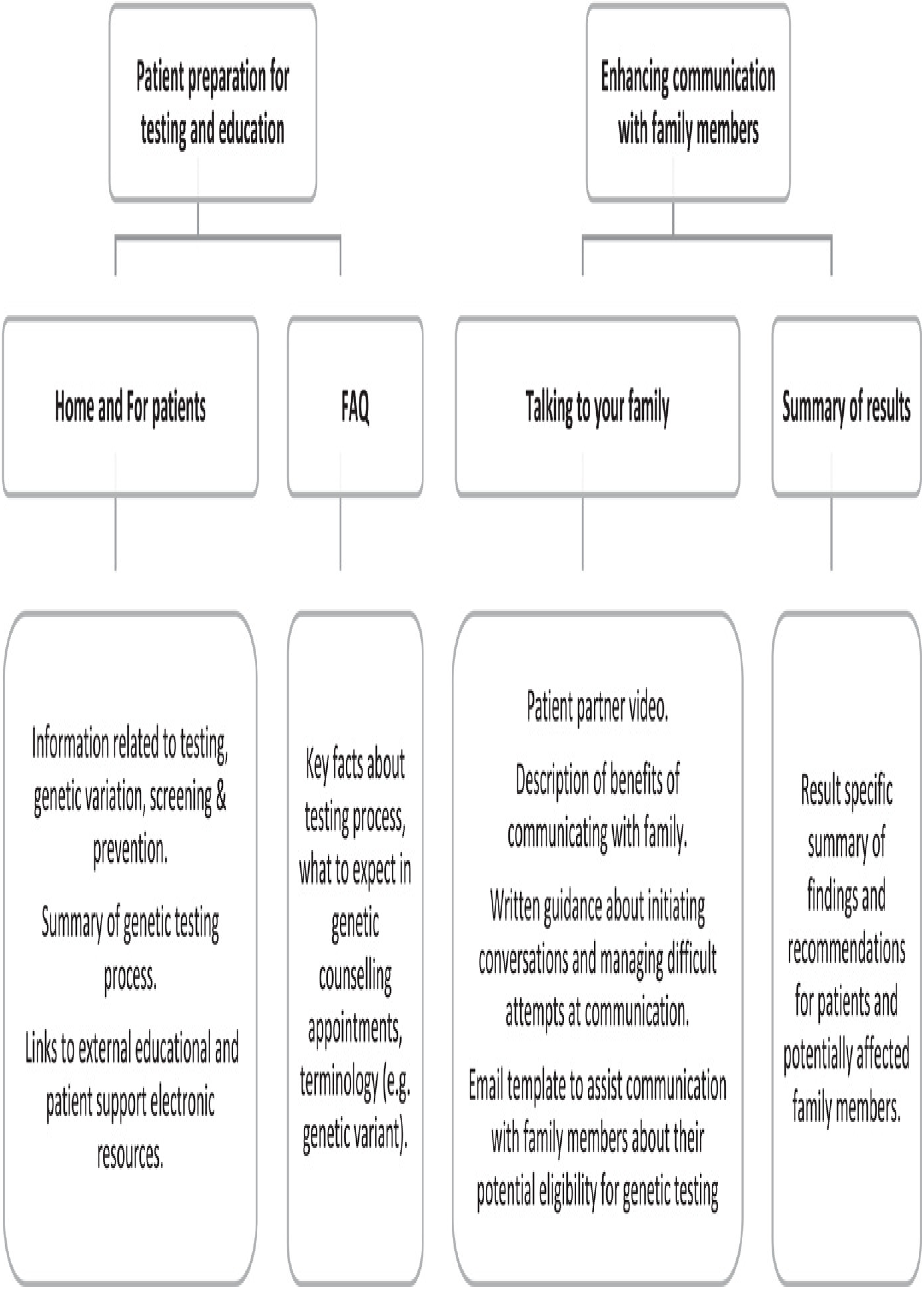
Data analysis and summary of feedback

Quantitative feedback was summarized using descriptive statistics. Qualitative feedback was reviewed by two co-authors (S. P. and R. L.). The primary analyst (S. P.) synthesized all qualitative feedback, summarized key findings and presented results to the larger research team for discussion. Through an iterative process of review and discussion, the research team prioritized suggestions for integration into the patient portal.

RESULTSPatient values integration into the patient portal

Following the completion of 25 qualitative interviews and a summary of results, investigators prioritized three key elements to inform the structure and content of the patient portal, as detailed in Table 1.

Based on participant feedback that spanned educational challenges and communication barriers throughout the process of testing, the portal was designed to be provided to individuals considering HSC testing, before their pretest genetic counselling appointment. The patient portal was designed as a single website, with each major component briefly described in Figure 1.



Enlarge this image.

To address participants' voiced unmet needs and preferences, the portal included six separate web pages spanning four components. The ‘Home’ page was designed to prepare patients for the testing process and their first pretesting genetic counselling appointment, to present key facts related to HCSs and provide links to each additional page. A 'For Patients' page provides information about *BRCA1* and *BRCA2* genes, prevention and screening strategies available to those with HBOC, as well as links to additional informational resources identified by HCP healthcare providers.

The 'Talking to your Family' page presents information about how and why to communicate with family, through a series of questions and answers. Questions addressed in the 'Talking to Your Family' page include but are not limited to, 'Why should I talk to my family if I have received a positive (or negative) result?', 'How can I prepare to talk to my family?' and 'What if I do not feel I can speak with my family members about testing?'

Within this portal page, we also included a template email text for individuals who would prefer to broach conversations using written communication. The email template text, developed in close consultation with our patient partner and HCP healthcare providers, is intended to inform family members that the individual has undergone testing with the HCP, and a written means by which to encourage family members to contact the HCP about testing, directly. 'Talking to Your Family' content also addresses why and how individuals with negative or uninformative findings should consider communicating their results to their family. For example, in describing why it is important to speak with family members about a negative carrier testing result, we highlight the importance of testing for potentially at-risk siblings who may harbour an HCS even if the proband does not.

As an additional component to the 'Talking to your Family' page, investigators developed a brief informational video describing one patient's lived experience communicating with their family about their HCS diagnosis. The video, approximately 4 minutes in length, describes the patient partner's experience relaying a diagnosis of an HCS to various members of her family. She describes initial and continued attempts with family members, and how she handled different reactions from relatives. Finally, she provides suggestions and recommendations, based on her own experiences, as to how to broach conversations about genetic risk using multiple approaches.

A general frequently asked questions ('FAQ') page provides a series of questions including additional information about testing eligibility, the testing process and where to go for more information. For example, questions addressed on the FAQ page include but are not limited to, 'What is a genetic variant?', 'Can anyone have genetic testing for hereditary cancer?' and 'Whom can I contact if I have any questions?'

As an adjunct to the patient portal to further enhance proband education, we developed lay summaries of *BRCA1* and *BRCA2* test results. To ensure applicability to all individuals undergoing HCS testing, lay summaries were developed for both *BRCA1* and *BRCA2* carrier and index findings, including positive, negative, uninformative, and variants of unknown significance. Summaries were developed to be provided to tested individuals following discussions with their genetic counsellors about test results, to enhance understanding and facilitate conversations with family members. Summaries were embedded into the patient portal through a password-protected healthcare provider page, wherein providers could email patients directly.

User testing results

A total of eight patient partners responded to the recruitment newsletter and provided informed consent via REDCap, seven of whom returned completed surveys. Thirty-five healthcare provider email invitations were sent, 20 responded to the invitational email and indicated an interest in participating, with 14 providing informed consent and 13 healthcare providers providing survey feedback.

Characteristics of user testing survey participants

Among the healthcare provider sample, 5 were oncologists, and 8 were genetic counsellors, with 12/13 self-reporting female gender. Healthcare providers reported discussing HCS a median of six times per week. All patient participants self-reported female gender, with a median age of 58 (range: 38–70). The majority of patients reported a previous diagnosis of cancer (*n* = 6), previous genetic testing for an HCS (*n* = 4), White ethnicity (*n* = 5) and university or college education (*n* = 6). Participant characteristics are described in Table 2.

Table 2 Characteristics of patient and clinician participants

Healthcare providers	*N* = 13
Speciality	
Oncologist	5
Genetic counsellor	8
Female	12
How many times per week do you discuss hereditary cancer genetic testing? (median, range)	6, 0–16
Patient partners	*N* = 7
Female	7
Age (median, range)	58, 38–70
Previous cancer diagnosis	6
Type of cancer	
Breast	3
Lymphoma	1
Melanoma	1
Multiple other	1
Previous genetic testing	4
Hereditary cancer syndrome diagnosis, yes	1
Missing	1
Ethnicity	
White	5
Other	
Jewish	1
Norwegian	1

Education	
High school	1
University degree	4
College (nonuniversity)	2

Summary of quantitative feedback

Among quantitative survey questions, healthcare providers demonstrated broader variation in their responses regarding the presentation of information, risks and benefits, as well as amount and clarity of information, as compared to patient participants (see Table 3). Most participants reported that the way general information was presented was either good or excellent (*n* = 16/20). Similarly, most participants reported that the risk and benefits presentation, as well as the description of questions and answers, was good or excellent (*n* = 14/20 and *n* = 15/20, respectively). Fourteen (14/20) of the participants thought that the amount of information presented was just right, and all but 2 individuals (*n* = 18/20) reported that everything or most things were presented clearly. All patient participants stated that they would ‘probably’ or ‘definitely’ recommend the patient portal, with 8/13 healthcare providers stating the same.

Table 3 Summary of quantitative patient and healthcare provider feedback (app content)

	Patient (*n* = 7)	Clinician (*n* = 13)
The way general information was presented		
Excellent	5	3
Good	2	6
Fair	0	3
Poor	0	1
The way risks and benefits were presented		
Excellent	3	2
Good	3	6
Fair	1	5
Poor	0	0
The way questions and answers were presented		
Excellent	2	3

Good	4	6
Fair	1	4
Poor	0	0
Amount of information		
Just right	6	8
Too little	1	1
Too much	0	3
Missing	0	1
Clarity of information		
Everything clear/balanced	2	3
Most things clear	4	8
Some unclear		2
Recommend the e-health app to others		
Definitely recommend	4	3
Probably recommend	3	5
Probably not recommend	0	4
Definitely not recommend	0	0
Missing	0	1
Preferred term to describe a genetic variant		
Gene change	0	3
Genetic variant	4	2
Genetic mutation	3	3
Genetic alteration	0	2

Gene problem	0	1
Pathogenic (if using term variant)	0	1
Abnormal result	0	1

Summary of qualitative feedback

All patients and 12 healthcare providers completed written qualitative comments. In general, feedback was detailed and varied. Following the analysis of the open-ended qualitative survey questions, we identified three key categories that reflect common and comprehensively articulated suggestions, namely (a) presentation, (b) educational content and (c) process clarification, as described in detail in Table 4. Key illustrative quotes are presented in Supporting Information. Due to the fact that participant feedback related to the patient portal content and results summaries were overlapping in terms of suggested recommendations, we present an aggregated summary of qualitative feedback.

Table 4 Summary of patient partner and healthcare provider qualitative feedback

Domain	Key feedback
Presentation: Ensuring information is clear and easy to navigate	Use lay and neutral language throughout Present balanced risk estimates and apply multiple ways of communicating risk Present all information in a succinct and digestible format Clarify location of all educational material within the patient portal
Educational content: Information to support personal understanding and communication with family	Include additional FAQ questions to clarify testing eligibility, follow-up process, recommendations for patients and family members following the return of results and implications of a hereditary cancer syndrome diagnosis (e.g., genetic nondiscrimination legislation) Define all technical and scientific language
Process clarification: Enhanced transparency around proband and cascade testing processes	Clearly describe HCS testing eligibility for probands and family members (according to result type and genetic family member relation) Clarify process for retesting in the future Highlight downstream benefits related to cascade testing (for relevant genetic relatives)

Abbreviation: HCS, hereditary cancer syndrome.

Presentation

Participants offered critical feedback about the presentation of information to improve the navigation and organization of the portal. Suggestions included the use of lay and succinct language throughout in an effort to reduce informational complexity, as well as ensure users are aware of the location of additional, more detailed educational material. Complexity and amount of scientific information were highlighted throughout qualitative responses as both necessary to ensure adequate understanding and as a potential challenge in terms of informational overload. As such, participants suggested the presentation of simplified information and phrasing, with clearly indicated links to additional, more detailed information.

Both patient and healthcare provider participants also suggested the need for neutrality in phrasing, and the careful selection of language to describe risk. For example, when presenting risk estimates (e.g., the lifetime risk of breast cancer for individuals harbouring a *BRCA1* variant), one healthcare provider participant suggested ensuring a neutral presentation of risks by presenting probabilities both positively and negatively (e.g., the probability of being diagnosed with cancer, as well as the probability of not being diagnosed with cancer over the individual's lifetime), to ensure a balanced understanding of risk. Similarly, one participant suggested softening causal language and acknowledging uncertainty related to the causal nature of pathogenic variations and the effectiveness of risk reduction strategies. In summary, participants prioritized the clear and succinct provision of accurate information, reduced informational complexity and improved ease of portal navigation.

Educational content

As stated, participants consistently acknowledged the complexity of information involved in HCS susceptibility testing, alongside a need to equip patients with information to make decisions about their health and their families. In addition to recommending concise language, participants also suggested the addition of information pertaining to key aspects of the testing and follow-up process. For example, suggestions included clarifying that the evidence base around variant pathogenicity is evolving and therefore classifications have the potential to change over time, more detailed information related to available cancer screening strategies, the distinction between germline and somatic variants, further detail regarding testing eligibility, as well as detailed information about existing genetic nondiscrimination legislation.

Related to both reducing informational complexity and providing succinct educational information, participants highlighted the need to clearly define and standardize scientific terminology. There was no singular preference among participants about the appropriate terminology to describe a gene variant (Table 4). Healthcare providers varied in their preference, while most patient partners selected ‘genetic variant’ followed by ‘genetic mutation’ as their preferred term. Within the qualitative feedback survey, participants further suggested consistency of terminology through the selection of a single term to describe a genetic variant, throughout the portal.

Process clarification

In addition to detailed feedback regarding the user interface and educational information, participants highlighted the need to clarify current genetic testing and follow-up processes within the patient portal. Patients and healthcare providers suggested the addition of detailed information about testing eligibility for both probands and family members, the return of results process, as well as the role genetic counsellors play in providing continued educational support following the return of sequencing results. For example, one patient participant suggested clearly presenting scenarios within which an individual would be eligible for HCP testing, such as—as the participant suggested—the presence of a strong family history of cancer alongside a current cancer diagnosis. Others wished to clarify whether it was the responsibility of the patient or the testing facility to initiate contact to determine eligibility for retesting in the future. Incorporating and clearly detailing information pertaining to the testing, diagnosis, cascade testing, and risk reduction strategy initiation process was critical, as reiterated through the qualitative feedback.

DISCUSSION

We present the development and preliminary evaluation of a patient portal to enhance communication between individuals undergoing HBOC susceptibility genetic testing and their genetic family members. Unique to this investigation is a patient and healthcare provider-guided approach to directly address experienced communication barriers. Our work placed patient perspectives at the centre of each stage of the research process, from identifying communication challenges and recommendations for enhanced support,[15] to portal content, design and initial evaluation. This work builds off existing investigations addressing inherent complexities associated with genomics-informed decision-making.[22–24,29,30]

User testing of patient-facing decision support tools is an established and valued approach to ensure usability, comprehension and the capture of relevant educational information.[25,38] Through user testing, we identified a need for enhanced clarity around testing eligibility and process to enable effective communication with their relatives, while presenting educational information using approachable and concise language. Educational and process elements were raised as key to enhancing patient understanding and enabling discussions with family.

The broad and varied nature of participant recommendations suggests a need to develop flexible patient-facing resources that are responsive to patient preferences. For example, our work demonstrates the value of generating and presenting information in a manner that acknowledges preferences for varying levels of informational detail, as well as a spectrum of approaches to navigating familial communication. Our evaluation strategy was designed to seek critical comments regarding the structure and content of the patient portal, integrate preferences, mitigate experienced challenges, and implement recommendations. Patient and healthcare provider co-development is critical to ensure comprehensive integration of the priorities and preferences of those for whom the patient portal is intended.[25] Findings presented through this process can be used to develop and broaden the scope of existing tools beyond *BRCA1* and *BRCA2* testing.

Previous attempts to increase interfamilial communication have demonstrated variable impacts in their ability to encourage individuals to initiate conversations and increase the uptake of cascade testing.[18,20] Published evaluations demonstrate challenges in generating patient and familial health benefits through the development of educational resources. A subsequent phase to the current investigation should consider comparative effectiveness to determine the extent to which our patient portal promotes conversations between probands and their family members, cascade testing and uptake of secondary prevention strategies among affected families. Here, we present the initial development phase, upon which additional evaluation can be investigated.

Our work can further be used to support ongoing efforts to increase cascade testing among affected families, including facilitated and direct contact of relatives by testing facilities.[5] Frey et al. recently conducted a single-arm feasibility study to evaluate the use of a facilitated approach, wherein testing facilities assisted probands by providing education around hereditary cancer risk, identifying potentially affected family members and communicating with relatives about cascade testing. The intervention also included genetic counselling for at-risk relatives and distributing at-home testing kits for those willing to undergo cascade testing. Investigators found that the facilitated approach suggested an increased uptake of cascade testing, although no comparative analyses were conducted due to the absence of a standard care control arm. Although future evaluations are required to determine whether and under which conditions facilitated and direct contact approaches are appropriate, emerging evidence demonstrates that patients may be supportive of policy shifts allowing for greater assistance from testing facilities in communicating with affected family members.[39] As the role of facilitated and direct contact continues to be investigated cross-jurisdictionally,[12] educational and communication supports such as the patient portal presented here can be incorporated as an adjunct to further enhance education about HCSs, the role of cascade testing and to support communication efforts.

Limitations

This work should be interpreted in light of limitations. Firstly, the portal was developed and evaluated in English. For this reason, the applicability and relevance to non-English speakers are limited and may fail to capture broader, pertinent educational and communication challenges. Addressing such disparities by translating the patient portal into additional languages relevant to jurisdiction-specific testing populations (e.g., Chinese, French and Punjabi) and seeking critical feedback from individuals representing non-English speaking groups will further broaden the potential impact of the tool. Of note, the patient portal does not capture information beyond the scope of *BRCA1* and *BRCA2*—associated HBOC, such as Li-Fraumeni syndrome, Lynch syndrome or more moderate-risk HBOC genes. The next steps in this research should seek to incorporate a breadth of HCS and elicit user feedback from affected individuals.

Second, critical feedback received during user testing may not be generalizable to the broader patient population for whom the portal was developed. The majority of participants who provided survey feedback were female, with most patient participants reporting postsecondary education and White ethnicity. Owing to study timelines, we were unable to continue recruitment efforts beyond March 2022, limiting our ability to enhance sociodemographic diversity. As a result, the spectrum of relevant information and guidance presented may not be adequately addressed in the current iteration of the portal.[40]

Our limitations speak predominantly to the ability of the patient portal to serve as a valued and effective resource, relevant to a diverse patient population. Broadening the reach of patient resources seeking to increase awareness about genetic testing and promote familial testing for potentially affected individuals will further support the mitigation of known healthcare inequities. Existing evidence suggests that multiple patient characteristics are associated with awareness of genetic testing and the uptake of publicly reimbursed healthcare services. Such factors include but are not limited to gender, language, ethnicity, income and education.[4,41,42] Further efforts to ascertain critical feedback and ensure relevance to individuals representing a diversity of perspectives are warranted.

CONCLUSIONS

We present the development and preliminary evaluation of a patient portal to facilitate familial communication about HCS susceptibility testing. This portal was designed to enhance genetic literacy and equip tested individuals with guidance and resources for communicating hereditary cancer risk with their families. Engaging patients and healthcare providers at each stage of portal development as both participants and research team members has ensured a responsive approach to addressing voiced unmet needs and communication barriers within the patient portal. As a critical first step to enhancing the impacts of population base screening programmes, this work presents a patient-guided evaluation to guide efforts to improve familial communication for patients undergoing HCS testing.

AUTHOR CONTRIBUTIONS

Samantha Pollard: Conceptualization; data curation; formal analysis; investigation; methodology; project administration; writing –original draft; writing –review &editing. **Deirdre Weymann**: Conceptualization; investigation; methodology; writing –review &editing. **Rosalie Loewen**: Data curation; formal analysis; project administration; writing –review &editing. **Jennifer Nuk**: Investigation; methodology; writing –review &editing. **Sophie Sun**: Funding acquisition; investigation; methodology; writing –review &editing. **Kasmintan A. Schrader**: Funding acquisition; investigation; methodology; writing –review &editing. **Chiquita Hessels**: Conceptualization; investigation; validation; writing –review &editing. **Dean A. Regier**: Conceptualization; data curation; formal analysis; funding acquisition; investigation; methodology; resources; software; supervision; validation; writing –review &editing. Samantha Pollard had full access to all the data in the study and takes responsibility for the integrity of the data and the accuracy of the data analysis. All of the authors gave final approval of this version to be published and agreed to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

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CONFLICTS OF INTEREST

S. P. and D. W. co-direct IMPRINT Research Consulting Ltd. and have received payments from Roche and AstraZeneca, unrelated to the current investigation. The remaining authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

Link to the patient portal will be made available upon reasonable request to the corresponding author.

ETHICS STATEMENT

The study was approved by the University of British Columbia and the BC Cancer Behavioural Ethics Board (H18-00644). Informed consent was obtained from all subjects involved in the study.

DETAILS

Subject:	Cancer; Research; Feedback; Susceptibility; Verbal communication; Relatives; Health care; Health status; Information transfer; Genetic screening; Web portals; Genomics; Teams; Decision making; Video communication; Interviews; Developmental stages; Mitigation; Genetic testing; Patients; Education; Editing; Health professional-Patient communication; Medical personnel; Communication; Medical screening; Risk communication; Risk reduction; Treatment preferences; Health risks
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‘No one's ever said anything about sleep’: A qualitative investigation into mothers' experiences of sleep in children with epilepsy

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ProQuest document link

ABSTRACT (ENGLISH)

Introduction

Sleep problems in children with epilepsy (CWE) are common. However, little is known about parental experiences and feelings about managing sleep in their CWE. To provide the most appropriate services' provision, it is essential that the lived experience of parents of this patient group and the issues and problems that they face in managing their child's sleep is understood.

Method

In 2018, nine mothers of CWE (aged 5–15 years) were interviewed about their perceptions and experiences around their child's sleep, sleep problems and their management, the impact of sleep difficulties on the child and their family and available support.

Results

Four themes were identified that represented the nature of the child's sleep problems, including settling and night-waking issues, parasomnias and child anxiety around sleep. Seven themes represented mothers' experiences of managing their child's sleep and any associated problems, including the longstanding challenging nature of child sleep issues, management strategies adopted, challenges related to managing sleep over time, the link between sleep and seizures, the negative impact of poor sleep on daytime functioning, role of antiseizure medication and maternal concerns about child sleep. One theme represented the perceived lack of information, help and support available.

Conclusions

Findings suggest there are unmet needs in supporting parents to deal with sleep, sleep problems and their management in CWE.

Patient or Public Contribution

This individual study was conducted under the umbrella of the CASTLE research programme (see https://castlestudy.org.uk/). Parents who have lived experience of parenting a child with epilepsy were co-applicants for the programme and were involved in the original conception, aims, design and funding application for the research programme (including the project reported in this paper) and advised on project design. Mothers of CWE who have lived experience of managing sleep and sleep problems in their child were participants who shared their experiences through the interviews, which formed the data of the current study.

FULL TEXT

INTRODUCTION

Epilepsy is a common chronic neurological condition, characterized by recurrent seizures; the prevalence in children and adolescents under 18 years of age is estimated to be 0.32%–0.55%.[1,2] Many children with epilepsy (CWE) experience co-existing deficits in behavioural, cognitive, attention, academic and psychosocial domains,[3–5] as well as reduced quality of life in comparison to children without epilepsy.[6,7]

Appropriate quantity and quality of sleep are crucial to healthy well-being. However, sleep and epilepsy have a complex and bidirectional relationship[8] and have been described as ‘unfortunate bedfellows’.[9] In CWE, commonly reported sleep difficulties are problems around initiation (settling and falling asleep), maintenance (experiencing night or early morning wakings), duration, daytime sleepiness as well as sleep anxiety.[10–12]

Research into sleep deprivation of parents of children with complex needs has found that one of the biggest challenges was the need to be available or vigilant during the night and that the impact of this sleep deprivation is ‘relentless’ and ‘draining’.[13] Over two-thirds (68.6%) of parents in a previous study of parents of CWE reported having concerns about their child having night-time seizures.[14] These worries and concerns are likely to contribute to parental sleep disturbance, which is more common in parents of CWE than in parents of children without epilepsy.[15] In a sample of parents of CWE (with intractable epilepsy), 75% reported that their sleep was affected, and of these, 48% reported the impact on their sleep was stressful.[16]

Traditional treatment goals for CWE tend to focus on seizure management, even though the importance of the need to develop child epilepsy care beyond ‘seizure control with minimal adverse effects’ has been a longstanding recommendation.[17] Yet sleep or problems around sleep are not usually an aspect of standard care that is regularly addressed, even though sleep is a key consideration reported by parents, CWE and healthcare professionals.[18,19]

Behavioural and educational approaches have shown signs of being effective treatments for many of the sleep difficulties experienced by these children and their families.[20] Research suggests that there may be special considerations for managing sleep in CWE, which could helpfully be addressed to likely increase families' engagement with and feasibility of intervention.[21] A greater understanding of these considerations could helpfully direct future intervention development[22] and ensure broader service provision is focused on the areas of most need. However, little research has explored the lived experience of sleep and sleep problems in CWE on parents.

The current paper aimed to address the research questions: 1) explore the types of parentally reported sleep problems faced by CWE and their families, 2) identify parents' experiences and feelings around managing their child's sleep and any associated problems and 3) identify parents' perception of available help and support when parenting a CWE around sleep.

MATERIALS AND METHOD

This descriptive qualitative study was conducted as part of the CASTLE (Changing Agendas on Sleep, Treatment and Learning in Epilepsy) programme of studies. Parents were asked about their child's sleep, any sleep problems, the experience of parenting a CWE around sleep and their perception of available support as part of a wider interview to inform the development of an online sleep intervention.[21,22]

Participants and recruitment

Participants were nine co-habiting mothers of CWE (six boys and three girls who ranged in age from 5 to 15 years, with median = 10 years and mean = 10.3 years, SD = 2.9). See Table 1 for descriptive details about the children of the interviewees, including their epilepsy.

Table 1 Descriptive details about the children with epilepsy of interviewees

	Age	Gender	Type of epilepsy	Duration since diagnosis	Seizure timing
P1	10	M	Benign rolandic	<1 year ago	Transitioning between sleep and wake
P2	15	M	Benign rolandic (a-typical)	>3 years ago	Daytime and during sleep
P3	10	F	Benign rolandic	<1 year ago	Transitioning between sleep and wake
P4	11	M	Benign rolandic	Between 1 and 3 years ago	Transitioning between sleep and wake
P5	5	F	Benign rolandic	Between 1 and 3 years ago	Daytime. 1 year without seizures (due to medication)
P6	9	M	Focal	>3 years ago	During sleep
P7	13	F	Unspecified	>3 years ago	During sleep and transitioning between sleep and wake
P8	7	M	Focal	>3 years ago	Transitioning between sleep and wake
P9	13	M	Generalized	>3 years ago	Daytime and transitioning between sleep and wake

Children suffered from a wide range of sleep problems, which allowed mothers to share their experiences of a broad range of sleep-related issues, see Table 2 for a summary of the children's maternal-reported sleep problems.

Table 2 Type of current and past parent-reported sleep problems[a]

	Settling	Night waking	Early morning waking	Sleep-related anxiety	Poor sleep quality	Daytime sleepiness	Morning waking difficulty	Co-sleeping	Room sharing	Sleep terrors	Sleep walking	Other
P1 (child aged 10 years)	C	C		C	C			C		P	P	
P2 (child aged 15 years)	C	C	C	C		C			P	C	C	
P3 (child aged 10 years)		C		C			P	C		C		
P4 (child aged 11 years)	C	C		C					P			
P5 (child aged 5 years)	C	C		C		C		C	C			
P6 (child aged 9 years)	C	C	C			P		P				
P7 (child aged 13 years)	C						C					
P8 (child aged 7 years)		C	C					C				
P9 (child aged 13 years)	C	C			C	C						C (Possible restless leg syndrome)

a

C—Current sleep problems refer to problems which are currently present, but it should be noted that the duration of these problems is generally longstanding (since infancy or beyond), with most problems also being also present in the past. P—Some parents also reported past sleep problems, which relate to issues that have now been resolved but which were significant problems at some point.

Participants were recruited by responding to online advertisements, which were placed on the websites of epilepsy organizations and charities (e.g., Epilepsy Action), the CASTLE study and researchers' university websites. Participants were recruited between March and July 2018, with interviews taking place between March and July 2018. The criteria for participation were being the parent of a child with epilepsy, based in the United Kingdom and having sufficient English skills to partake in the interview and also willing to review a draft online sleep intervention which was part of the larger CASTLE programme of studies reported in a separate paper.[22]

Measures and data analysisInterviews

A semi-structured interview schedule was developed by the researchers that asked about key topics related to the child's sleep and relevant to the development of an online sleep intervention for a wider study (Supporting Information: 1 for full interview schedule; Cook et al.[21] and Wiggs et al.[22] for development of the sleep intervention).

Analysis

Data were thematically analysed according to the six standardized stages outlined by Braun and Clarke.[23] Data familiarization was achieved through the reading and rereading of transcripts before the data were fully coded by a researcher not involved in the interview process (GC). This study employed an inductive analytic strategy driven by the data and participants' own words. Coding was reviewed and discussed amongst the research team, to address any discrepancies and reach agreement. A number of codes were combined following discussions, for example, the codes ‘frequent overnight awakenings’ and ‘night waking issues’ contained similar content and were combined. Other codes that did not relate to mothers' perceptions or experiences around their child's sleep were set aside. During the ‘search for themes’ stage, one of the researchers (GC) reviewed the codes and clustered them into potential themes and subthemes. Next, coded extracts of raw data were revisited, and the themes/subthemes reviewed across the whole data set to ensure that they comprised an accurate reflection of the transcripts. During the ‘defining and naming themes’ stage, the research team reviewed the proposed analysis and refined the specific details of themes, as well as agreed names and descriptions. This involved ongoing discussion and iterative amendments until agreement was reached. The final set of themes and subthemes were agreed upon between all authors.

Procedure

Once parents had expressed an interest (through responding to online adverts), they were provided with an information sheet and given an opportunity to ask any questions. Eligible participants who wished to participate were required to complete a consent form. Interviews were then completed by two researchers (PG and LW) at a time and in a format convenient for participants (two face-to-face, six by telephone and one via video call). Interviews were audio recorded, transcribed verbatim and thematically analysed.

RESULTS

Interviews lasted, on average, 61 min. Themes are presented in line with the study's research questions; four represent the nature of maternally reported sleep problems; seven (and associated subthemes) represent mothers' experiences and feelings around managing their child's sleep and any associated problems and one relates to mothers' perceptions of the available help and support to them around sleep. Themes (and subtheme(s) where appropriate) are presented and described below, with detailed illustrative quotations appearing in Table 3.

Table 3 Summary of themes and subthemes supported by detailed quotations

Research question 1: Parentally reported sleep problems faced by CWE and their families

Theme: Settling issues	‘She had trouble falling asleep, so falling asleep sometimes lasted more than an hour to make her fall asleep’ (P5). ‘He used to just, like, when we put him to bed it would be, like, 2 hours or so afterwards before he'd finally managed to drop off to sleep. Yeah, he really used to struggle to get to sleep and anything like that…’ (P4). ‘If he can't sleep he's up down, annoying his brother, in and out of his sisters room, up and down the stairs …wandering around the house making a noise’ (P6). ‘…it got to the point, even at that age, where you dreaded bedtime with her. You were, like, oh god it's bedtime. It just came over you like a black cloud, your heart sunk, we both looked at each other, like, it's bedtime’ (P7).
Theme: Night-waking issues	‘He would go to bed fine and go to sleep fine but it's just in the middle of the night when he wakes up …it would be that bit that would cause the problem for us’ (P1). ‘So whether they were seizures waking him up or whether he was just not falling to sleep properly, not going into deep sleep and he was just dozing’ (P2). ‘I'm not sure if they were seizures waking up, because obviously he has quite a lot of seizures. So whether they were seizures waking him up or whether he was just not falling to sleep properly, not going into deep sleep and he was just dozing’ (P2). ‘It's just the amount of times he wakes up and gets up in the night and unless you sit and watch him all night I don't know how you would prevent him getting up all night’ (P6). ‘He'd go to sleep at sort of 8.30/9 and then wake up at between 7 and 8 so he was, but he would always in the night he would wake up and come into us and we'd have to go and put him back’ (P1).

Theme: Parasomnias	‘Well he's 11 this November probably from the age of 7 he's suffered with night terrors intermittently. So we had a few episodes of that and then he did a bit of sleepwalking and he's never since sort of being the age of 7 years old he's never really fully settled at night time’ (P1). ‘But she very much still every night still shouting ‘no, no, no, no' and shouting out … During her sleep yeah, she has a recurrent dream …when she's having the dream and she was shouting “mum, mum” and I'm like “I'm here”’ (P3). ‘Sometimes it was very difficult to tell if he was just wandering around because he didn't want to go to sleep because he was anxious or he'd had a seizure or he was literally sleep walking …he was asleep, so he did sleep walk quite a lot’ (P2).
Theme: Child anxiety around sleep	‘He's always been quite a nervous sleeper you know he's always had to have a light on and things’ (P1). ‘He was having a lot of difficulty sleeping because he was worried that if he went to sleep, because we had quite a few instances where he went to bed as normal and when he woke up he was in hospital. Because sometime in that night he'd had a tonic clonic and we had to call an ambulance because his tonic clonics tend to be about 10 minutes long and his oxygen levels drop, so we have to call an ambulance. So he did go through a stage of not wanting to go to sleep and I think I wrote in there that he had a massive fear that he was going to die in his sleep. So I think that's, it's not just the seizures it's the emotional side of it as well’ (P2).
Research question 2: Parents' experiences and feelings around managing their child's sleep and any associated problems	
Theme: Longstanding challenging nature of child sleep issues	‘I said to him yesterday how's your sleep and he said to be honest I don't think I've slept through the night for the last 6 months’ (P2). ‘He used to just be saying I can't sleep mum I can't sleep all the time and you, it's just awful when they want to sleep but they can't get to sleep’ (P4). ‘It's so draining for her and us really, you know. Because when she's awake she's like Bagpuss the whole house is awake. So she's got a little sister so then she's awake because [child's] awake …It affects everyone’ (P7).

Theme: Management strategies for child sleep	‘He's never slept and we've gone through lots of different things, of early to bed, late to bed, exercise before bed, everything’ (P2).
Subtheme: Co-sleeping	‘Practical wise it was easier for him to be in with us because he is there all night. Rather than sort of lying there uncomfortably with my arm up in the air trying to hold his hand and then creeping out of the room half an hour later’ (P2). ‘Ever since he had his first seizure he co-sleeps with his dad because we don't feel comfortable to let him sleep on his own …we'd just be up all night checking on him …it's us more than him’ (P1). ‘But the main thing was we all slept together …I think I was probably 50/50 us wanting to sleep with him as well as him wanting to sleep with us’ (P2). ‘Well he [child] slept in my bed and my husband slept on the floor in our room bless him, on and off for about 3 years’ (P2). ‘…we have to deal what we're living with at the minute and just get on with it and as long as we all get sleep …however we feel, we just feel a lot calmer and more settled if one of us is with him’ (P1).

Subtheme: Sleep environment adaptations	‘It's petrifying isn't it if you wake up and it's pitch black and you're not sure, so he did have various night lights throughout the years, until he was a bit older and now he's got a sort of electric one he can flick on when he wants to by the side of the bed’ (P2). ‘We had one of those safety pillows and I think that's a massive help and it's a massive reassurance for [child] and for parents that you've got one of these safety pillows. That if they do have a tonic clonic face down …they can breathe through the pillow, yes I think that's a massive thing’ (P2). ‘The early nights and everything that we're doing, we've got we've bought black out curtains, we've bought well 2 sets of black out blinds and curtains, we have a fan going, we have all these kind of little interventions that we've put in place just anecdotal’ (P3). ‘Now whether or not that [blackout blinds] cancels out the noise of the rest of the just the general household as well. Because then her brother will be going to bed an hour later. You know there's just those things that I'm very conscious of might wake her up again’ (P3).

Subtheme: Use of monitoring devices	‘He had a couple of tonic clonics in the middle of the night, which we did pick up on because I had this video monitor and you know I had like an alarm and everything …I have that on my phone pointed at a video monitor. So that if he moves on the video monitor even if I don't hear it, it'll pick up the movement as well and sound an alarm, so we did beef up on the seizures’ (P4). ‘Once I got all the monitors setup and everything, I was kind of happy that I wasn't going to miss any [seizures], if he did have any’ (P4). ‘We have monitors as well and I think which have given her some security. I think they help her to go to sleep knowing that she has a monitor …She feels less anxious because she's got a bed monitor and she's got a room like a video monitor on’ (P3). ‘Obviously it's movement [that is monitored by the device] and he's a bit of a rough sleeper so that goes off quite often …I just check on the video monitor and if I can see he's just turning over or doing a normal movement then I know he's fine. If I can't see him properly on the monitor then I'll just pop in and make sure he's alright’ (P4). ‘My problem with the seizure detection monitor is that I've not seen it work, it's not proved itself to me because we've only had it 2 weeks and she hasn't had that kind of seizure’ (P3).
Theme: Challenges related to managing sleep over time	‘It's very hard if she's resistant to want to go bed …because of your seizures. You don't want to bring it into that just before you go to bed. But for an older child they've got to take ownership of it a little bit’ (P4). ‘We have had relaxation DVDs in the past, before she was diagnosed with epilepsy …They were kind of like a story that went into relaxation …We did use those for a good while …but then she outgrew them’ (P3).

Theme: Link between sleep and seizures	'…a vicious circle because the seizures were caused by lack of sleep and …they're getting stressed because they're not getting enough' (P4). 'I know for a fact that if we put him back in his own bed he would be waking up in the night and then obviously we're told that maybe one of the causes of the seizure is lack of sleep and tiredness so we don't want him to, we're scared if he goes back in his own bed and he's waking up all the time is it going to bring on a seizure' (P1). 'I was kind of neurotic with trying to make sure that he was getting enough sleep. Because I knew that that was a major trigger for his seizures, because it was when he was tired, he always had more' (P4).
Theme: Negative impact of poor sleep on daytime functioning	'Sleep obviously has a massive effect on the way he functions during the day' (P2). 'If he's been up then he really does suffer the next day at school, whether that's a knock on effect that he's tired so he has more seizures and its just a constant knock on effect' (P2). 'He doesn't do well without sleep and I think that's the other big thing, he does need a lot of sleep and again I don't know whether that's because he wakes numerous times or whether he's a teenager' (P2).
Theme: Antiseizure medication	'I mean she still wakes up but since we, I've noticed improvements since we started medicating, that's also the truth' (P5). 'We finally found a medicine that stopped the tonic clonics and him just having the partial focals that then I think that that was the turning point that it seemed to be much easier to be much easier to cope with that we knew he wasn't in his bedroom having tonic clonics on his own' (P2) 'Because we go to clinic and they say she needs to sleep because you know the better the sleep the less seizures but then they go but she won't sleep because of the condition and the medicine. Side effect is it won't, she won't sleep' (P7).
Theme: Maternal concerns about child sleep	'I think it's just that now because at the point of diagnosis they said this is you know the one thing you can do. It feels more pressured' (P3).

Subtheme: Maternal anxiety around night-time seizures	‘The fear of it [a seizure] happening and us not hearing him or not being there is just unbearable to think about so for us as a family it's just working [co-sleeping] at the minute that he's with us’ (P1). ‘It's drilled into you, you need to call an ambulance if the seizure goes on after 5 minutes. If I'm not in her room, if I'm not half awake and alert to when she starts the seizure, how do I know it's gone over 5 and I think that's the main worry for everybody really. So probably for me, the only thing that would make me more comfortable is if I am confident that something would alert me’ (P3). ‘The only thing that would make me more comfortable is if I am confident that something would alert me [to child seizure]’ (P3).
Research question 3: Parents' perception of available help and support when parenting a CWE around sleep	
Theme: Lack of information, help and support available	‘I think even in all the pamphlets and leaflets and whatever we've been given over the years, nobody's ever mentioned sleep’ (P2). ‘Sleep is the one thing that we can do, help but then not really much assistance comes along with that at that point of diagnosis’ (P3). ‘His consultant gives you all sorts of stupid advice and you're like really, “don't let him drink coffee” and I'm like he's 9 I don't let him drink coffee anyway’ (P6). ‘…we are given nothing in hospital, you know or when we've been to A&E, there's nothing that you are told, as I say all we were told at that point is you must get good sleep and so it would be great if somebody then said and here have a look at this …and I would have come straight home and gone straight onto it’ (P3).

Abbreviation: CWE, children with epilepsy. **Research question 1: Parentally reported sleep problems faced by CWE and their familiesSettling issues**

Mothers commonly reported settling issues, *‘She had trouble falling asleep’* (P5). In many cases, it was the duration in settling their child to sleep which was problematic, such as, *‘2 hours or so afterwards before he'd finally managed to drop off to sleep’* (P4).

In some families, the CWE settling issues negatively influenced the wider family, *‘annoying his brother, in and out of his sister's room…’* (P6). For some, the extent of settling issues influenced their feelings and experiences around putting their child to bed, *‘…you dreaded bedtime with her…’* (P7). Settling issues were problematic for mothers, families and CWE but were one of the most commonly reported sleep problems.

Night-waking issues

Mothers also commonly reported issues around night wakings, *‘…it's just in the middle of the night when he wakes up…’* (P1). For some mothers, there was ongoing uncertainty around the cause, and some mothers felt helpless about how they could address their child's night waking, *'…I don't know how you would prevent him getting up all night'* (P6).

These night wakings would also disrupt parents' own sleep, *'…in the night he would wake up and come into us and we'd have to go and put him back'* (P1). There was an ongoing challenge for parents in dealing with frequent night wakings, perhaps particularly in CWE when the underlying cause of the waking is unknown.

Parasomnias

Nightmares, sleep waking and sleep terrors were common parasomnias experienced by some CWE, *'…he's suffered with night terrors intermittently …and then he did a bit of sleepwalking…'* (P1). For some mothers, their child's parasomnias were shocking and required parental intervention, *'when she's having the dream and she was shouting "mum, mum" and I'm like "I'm here"'* (P3). Some mothers specifically highlighted the problems of distinguishing and understanding these behaviours in their CWE. For example, *'…it was very difficult to tell if he was just wandering around because he didn't want to go to sleep because he was anxious or he'd had a seizure or he was literally sleep walking…'* (P2).

Parasomnias could be longstanding and challenging for parents and children to manage.

Child anxiety around sleep

A difficulty faced by many mothers was their child's sleep-related worries, concerns or anxieties, *'He's always been quite a nervous sleeper'* (P1). Some mothers highlighted that the child's epilepsy and the possibility of sleep-related seizures was a source of anxiety for their child, *'…he had a massive fear that he was going to die in his sleep'* (P2). For some mothers, the challenge was managing a calm and relaxing bedtime when child anxiety was most commonly expressed or shared at this time, *'That's usually when it comes out [child's anxieties or worries] …just when we're doing the bedtime'* (P3).

A key factor in dealing with sleep in CWE was managing the child's worries that could surface around sleep or the anxieties the child had about sleep due to epilepsy.

Research question 2: Parents' experiences and feelings around managing their child's sleep and any associated problemsLongstanding challenging nature of child sleep issues

For many mothers, the sleep problems experienced by their child were longstanding, *'So when he was diagnosed when he was 7 [now 11] and I think right from the beginning he had problems with sleep'* (P2). Some mothers acknowledged the challenges they felt about supporting their child's sleep problems, *'It's just awful when they want to sleep but they can't get to sleep'* (P4).

In some cases, it was not just the mothers, but also the children, that acknowledged their disturbed sleep *'He said to be honest I don't think I've slept through the night for the last 6 months'* (P2). For many mothers, the sleep problems experienced by their CWE negatively impacted the whole family, *'It's so draining for her and us …It affects everyone'* (P7). Overall, the evidence in this theme suggested that sleep problems were common and longstanding, and their influence was negative for the CWE, their parents and the wider family unit.

Management strategies for child sleep

Many mothers reported adopting certain approaches or practices to facilitate what they perceived to be safe sleep for their children. These were broad and varied in nature and encapsulated within three subthemes.

Co-sleeping

Some mothers found advantages of co-sleeping (bed and/or room sharing), *'Practical wise it was easier for him to be in with us because he is there all night'* (P2). For some parents, a key motivation for co-sleeping was so that they could monitor their child and their safety overnight, which allayed some parental concerns, *'…we don't feel comfortable to let him sleep on his own'* (P1).

In other cases, co-sleeping was beneficial as having parents nearby helped to alleviate some of their child's fears or worries, '*I think I was probably 50/50 us wanting to sleep with him as well as him wanting to sleep with us'* (P2). However, adopting co-sleeping practices had a knock-on impact on the sleep arrangements of the rest of the family, *'[child] slept in my bed and my husband slept on the floor'* (P2).

For some, co-sleeping was reactively adopted to facilitate improved familial sleep, *'…we have to deal with what* *we're living with at the minute and just get on with it …we just feel a lot calmer and more settled if one of us is with him*' (P1). Co-sleeping was a commonly reported strategy desired and adopted by parents and their CWE to provide support and reassurance around child safety.

Sleep environment adaptations

Another key strategy was making changes or amendments to their CWE's sleep environment. There were a number of motivations for doing so, for some, it was to make it more comfortable and/or comforting for their child, *'…he did have various night lights throughout the years'* (P2).

Some parents made use of specific products to provide reassurance to themselves and their child, *'We had one of those safety pillows and I think that's a massive help and it's a massive reassurance for [child] and for parents'* (P2). In some cases, specific changes were implemented at prominent transitions, for example, when transitioning the CWE to solo sleeping to provide additional assurances.

It was common for mothers to make multiple amendments in an attempt to help manage their child's sleep, even if the evidence base for these specific adaptations was not clear. For example, one family reported, *'We have all these kind of little interventions that we've put in place, just anecdotal'* (P3).

There was a range of different amendments that mothers made to their child's sleep environment, with the motivations predominantly revolving around offering reassurance and alleviating some safety concerns.

Use of monitoring devices

A number of mothers reported using various different types of monitoring devices with their CWE. These allowed parents to monitor their child and/or be alerted to any seizures and intervene if needed. For example, *'We did pick up on [child's seizures] because I had this video monitor and you know I had like an alarm and everything'* (P4).

The use of monitoring devices was beneficial predominantly due to the reassurance they were perceived to provide; for the mother, *'Once I got all the monitors setup and everything, I was kind of happy that I wasn't going to miss any [seizures], if he did have any' (P4) and for the child, 'We have monitors as well and I think which have given her some security'* (P3).

There was also a range of issues and concerns about the use of these devices. These were around them going off unnecessarily, *'…loads of false alarms'* (P4) and the alerts could have a disruptive effect on parental sleep because *'[alarm] goes off quite often'* (P4). In addition, the focus monitors appropriated could be a challenge for mothers who did make use of such devices, *'Another parent I've heard had a monitor in their room just the video monitor and they said they sat and watched it'* (P3).

Some mothers had reservations about the use of these methods as they did not feel these methods were acceptable for their child, *'…kind of feels like an invasion of privacy'* (P6). Another concern for some was the reliability and efficacy of monitoring devices, *'It's not proved itself to me'* (P3).

Monitoring devices were relatively widely used and appeared to offer highly sought-after reassurance to both parent and child that seizures would not be missed. However, these systems had limitations for some families.

Challenges related to managing sleep over time

The majority of mothers reported that many of the challenges in managing their child's sleep changed over time.

As children became older, they were offered more independence, *'I can't honestly say whether he was a lot better at sleeping or he was more independent at sorting himself out'* (P2). Mothers also acknowledged that as their child aged and developed, the child needed to take additional responsibility for their epilepsy, *'…for an older child they've got to take ownership of it a little bit'* (P4).

The type, success and suitability of parental management techniques also evolved over time, *'We are very conscious that as [child] gets a little bit older she might not want me [to co-sleep with her]'* (P3). Mothers of CWE clearly highlighted that managing their child's sleep and any sleep problems were challenging, and the type and nature of these challenges tended to change over time.

Link between sleep and seizures

A number of mothers felt their child's sleep and seizures were linked, *'I know from experience with [child] that if he doesn't get enough sleep then he will have more seizures'* (P2). In some cases, mothers believed that addressing their child's sleep problems would have a positive impact on their seizure management, *'If I could get sleep sorted I think it would massively change her seizures. It's all to do with her sleep'* (P7).

The knowledge of the relationship between sleep and seizures resulted in maternal concerns about ensuring their child obtained enough sleep, *'I was kind of neurotic with trying to make sure that he was getting enough sleep'* (P4). Mothers were clearly aware of and, in many cases, had experienced first-hand the relationship between sleep and seizures.

Negative impact of poor sleep on daytime functioning

A number of mothers clearly identified that poor sleep had a negative impact on their child's day-to-day functioning, *'He can be quite sleepy during the day, it depends on what he's doing and obviously if he's seized the night before he's more likely to be tired'* (P6).

A definite link was made by mothers between their child's sleep and potential negative implications for their child's academic endeavours, *'If he's been up then he really does suffer the next day at school'* (P2). There was a challenge for some mothers in differentiating between poor sleep due to the child's epilepsy or developmental stage, *'…I don't know whether that's because he wakes numerous times or whether he's a teenager'* (P2). From their own experiences, many mothers linked poor sleep with having negative implications for children's daytime functioning and sleepiness levels, although the cause of poor sleep was not always clear.

Antiseizure medication

A number of mothers reported antiseizure medication having an impact on their child's sleep. For some, antiseizure medications had a positive impact on their child's sleep, *'I've noticed improvements since we started medicating'* (P5). A benefit of anti-seizure medication was that it provided additional reassurance for the child, *'…not kind of worrying about going to sleep like he used to'* (P4).

Another benefit was the additional reassurance this provided that night-time seizures were not being experienced by the child and so could be missed by parents, 'We finally found a medicine that stopped the tonic clinics *…it seemed to be much easier to cope with that when we knew he wasn't in his bedroom having tonic clonics on his own'* (P2). However, for some families, the child's antiseizure medications posed challenges due to their negative impact on child's sleep, *'…then they go but she won't sleep because of the condition and the medicine'* (P7).

The belief and/or experience that medication-assisted in reducing and/or assisting in managing seizures gave some CWE and their parent's reassurance around seizures which in turn had a beneficial impact on their child's sleep. However, this positive experience was not shared by all families.

Maternal concerns about child sleep

A number of mothers reported having worries and concerns about sleep for their CWE. For some mothers, even attempting to establish and maintain healthy sleep in their childhood itself was anxiety-inducing. The fact that sleep is the one aspect highlighted to parents as modifiable added to maternal anxiety about sleep.

Maternal anxiety around night-time seizures

Regardless of the nature of the child's seizures (i.e., frequency and severity), some mothers held concerns about the possibility of night-time seizures. Particularly noted was the role of the parent in monitoring the child's safety and the impact this had on many families sleeping arrangements, *'The fear of it [seizure] happening and us not hearing him or not being there is just unbearable to think about'* (P1).

The specific timing of some children's seizures and the challenges of parents monitoring night-time seizures generated maternal anxieties, *'If he's getting up in the middle of the night and I'm fast asleep it's much easier to miss if he had a seizure'* (P4). These concerns appeared to be compounded by an awareness of the critical role of parental or medical intervention for child safety.

The timing and safety of children who were likely to experience seizures around sleep or overnight contributed to specific maternal worries and anxieties about sleep.

Research question 3: Parents' perception of available help and support when parenting a CWE around sleepLack of information, help and support available

Most mothers had experienced a lack of relevant help and support about sleep issues, *'We've not had any advice* *beyond that. Apart from "get good sleep, that's the one thing you can do"'* (P3). This was particularly noted as a salient issue around the time of a new diagnosis.

In some cases, sleep advice that had been offered by healthcare professionals had not been well received or deemed as helpful by mothers. Some even suggested that this lack of information needed to be addressed and suggested the need for standardized advice in the form of a resource that parents could be directed to by healthcare professionals. For example, one mother reported, *'…it would be great if somebody then said and here have a look at this …I would have come straight home and gone straight onto it'* (P3). It was clear that help and support for child sleep problems were lacking in this clinical group. There is a key need for additional resources or interventions that address sleep in CWE to support parents throughout their epilepsy journey.

DISCUSSION

Mothers reported four themes which represented the nature of the child's sleep problems. These included settling and night-waking issues, parasomnias and child anxiety around sleep. Seven themes represented mothers' experiences of managing their child's sleep and any associated problems. These encompassed the longstanding challenging nature of child sleep issues, management strategies adopted, challenges related to managing sleep over time, the link between sleep and seizures, the negative impact of poor sleep on daytime functioning, the role of antiseizure medication and maternal concerns about child sleep. One theme represented the perceived lack of information, help and support available.

The current study contributes to existing empirical evidence that suggests sleeplessness problems and parasomnias are common in CWE[12,14,24] and that CWE are susceptible to experiencing sleep-related anxiety around sleep, particularly if they frequently experience seizures or have had traumatic seizure experiences associated with sleep.[25] There are clear added challenges for parents of CWE when managing their children's sleep and any sleep problems. These concerns commonly resulted in additional maternal monitoring of the child during the night, which is likely to negatively impact both maternal anxiety and sleep, the latter of which is noted to be commonly disrupted in parents of CWE.[14,15,26,27] It appeared that the use of some management strategies, such as room sharing or co-sleeping, were often reactively implemented in an attempt to help the CWE and their family obtain the best quantity and quality of sleep that they could, in the circumstances. However, practices such as co-sleeping can have a detrimental effect on the parent/caregivers' and children's sleep.[14]

In many cases, it appeared that parents employed multiple, sometimes diverse, strategies. A number of mothers reported using different products or devices primarily to help provide some reassurance about their child's safety and security, principally not missing night-time seizures. There is a wide range of monitoring devices available, but the evidence for their efficacy is scant for children.[28] Mothers' experiences or concerns around false alarms are also reiterated in the literature in the context of challenges around sensitivity and specificity around these devices.[29] There is currently no individual seizure-monitoring device, which is effective for detecting all types of seizures in adults or children.[30] In addition, a topic of key concern to parents is Sudden Unexpected Death in Epilepsy (SUDEP),[31] yet no devices have been demonstrated to prevent SUDEP.[32] The lack of good quality evidence for the use of seizure monitoring devices may make it challenging for parents to select the most appropriate device and to have confidence in its usage.

Mothers were aware of the links between sleep and seizures, yet felt that they lacked guidance about how to address or improve their child's sleep, including from their healthcare teams. This appeared to heighten maternal anxieties and feelings of 'helplessness'. This finding emphasizes the need to ensure adequate help and support is available to help support healthy sleep in CWE, as identified in previous work.[19,21] Current results suggest this is crucial around the time of diagnosis and concur with previous research that there is a need for ongoing information and support, beyond diagnosis.[33] The availability of ongoing, sleep-related support is an important unmet need for this clinical group.

Some research groups (such as CASTLE) have recognized this gap in sleep-related help and support of CWE and their families and have begun to explore sleep interventions for this clinical group, the efficacy of which is currently being investigated through a clinical trial (see https://castlestudy.org.uk/). For this group of children, the impact of antiseizure medications on sleep is also a priority, given that antiseizure medication was noted by mothers in the current study to have mixed effects on sleep, as has been reported by others.[34]

The current findings need to be considered in the context of some limitations. The sample size (*n* = 9) was smaller than intended and reflected recruitment challenges. However, it included mothers of a range of diverse participants (child age, epilepsy type and severity and sleep problems), and this heterogeneous group is a distinct strength as this allowed us to obtain a range of parental experiences and perceptions. The original intention had been to recruit both mothers and fathers. However, our final sample solely consisted of mothers. It is not clear if fathers share the same concerns and approaches to managing their child's sleep. In addition, the involvement and availability of the father in co-habiting households may directly or indirectly influence maternal feelings and behaviours and understanding these dynamics within families needs further elucidation in future work. Importantly, our sample was also self-selecting and so is unlikely to be representative of all parents of CWE. The current study focused on the parental experience of managing their child's sleep, but it is also important to identify how CWE feel about their sleep and its management. Future work could explore parental and child lived experiences in a larger and more diverse sample. Such information can inform the development of support for families. For example, additional content of the interviews considered in the current study addressed what parents wanted from an online sleep intervention. These have been reported elsewhere,[21] and some of the key issues highlighted were considered in the development of an online intervention specifically designed for this clinical population.[22] Addressing sleep in CWE, and its management may have a twofold benefit of improving both the CWE and their parent's sleep quality.

Given all of the authors are experienced sleep and/or epilepsy researchers, while every attempt was made to put aside what we know about sleep and its management in CWE, it is possible that our existing knowledge and understanding (of both the wider literature and conducting qualitative studies with this patient group) influenced the data collection (e.g., following up with questioning on specific sleep issues or problems) and in the analysis of the data (e.g., identifying codes informed by our existing knowledge). However, every attempt was made to foreground parents' words and experiences in the data collection and analysis.

CONCLUSION

To provide the most appropriate services and support for sleep issues in this patient group, it is essential that the lived experience of parents and the issues and problems that they face in managing their child's sleep are understood and acknowledged. The current paper has highlighted the key areas of child sleep behaviour and associated issues which mothers with lived experience have faced and, in many cases, desire help and support for. Current findings suggest that there are unmet needs in supporting parents to deal with sleep, sleep problems and their management in CWE. Given the link between sleep and seizures, sleep should be an intervention target for health services; if appropriate provision is provided for CWE and their parents, this may result in improved outcomes for multiple child and parental outcomes.

AUTHOR CONTRIBUTIONS

All authors contributed to the conception and design, acquisition of data, analysis and interpretation of data; and drafting the article or revising it critically for important intellectual content.

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CONFLICT OF INTEREST

The authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

The data sets presented in this article are not readily available because the data are qualitative interviews and cannot be shared. Requests to access the data sets should be directed to amber.collingwood@kcl.ac.uk.

ETHICS STATEMENT

Ethical approval was obtained through Oxford Brookes University Research Ethics Committee (study reference 171108). All participants provided written informed consent.

DETAILS

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When attitudes and beliefs get in the way of shared decision-making: A mediation analysis of participation preference

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ABSTRACT (ENGLISH)

Introduction

Certain sociodemographic characteristics (e.g., older age) have previously been identified as barriers to patients' participation preference in shared decision-making (SDM). We aim to demonstrate that this relationship is mediated by the perceived power imbalance that manifests itself in patients' negative attitudes and beliefs about their role in decision-making.

Methods

We recruited a large sample (N=434) of outpatients with a range of urological diagnoses (42.2% urooncological). Before the medical consultation at a university hospital, patients completed the Patients' Attitudes and Beliefs Scale and the Autonomy Preference Index. We evaluated attitudes as a mediator between sociodemographic factors and participation preference in a path model.

Results

We replicated associations between relevant sociodemographic factors and participation preference. Importantly, attitudes and beliefs about one's own role as a patient mediated this relationship. The mediation path model explained a substantial proportion of the variance in participation preference (27.8%). Participation preferences and attitudes did not differ for oncological and nononcological patients.

Conclusion

Patients' attitudes and beliefs about their role determine whether they are willing to participate in medical decision-making. Thus, inviting patients to participate in SDM should encompass an assessment of their attitudes and beliefs. Importantly, negative attitudes may be accessible to change. Unlike stable sociodemographic characteristics, such values are promising targets for interventions to foster more active participation in SDM.

Patient or Public Contribution

This study was part of a larger project on implementing SDM in urological practice. Several stakeholders were involved in the design, planning and conduction of this study, for example, three authors are practising urologists, and three are psychologists with experience in patient care. In addition, the survey was piloted with patients, and their feedback was integrated into the questionnaire. The data presented in this study is based on patients' responses. Results may help to empower our patients.

FULL TEXT

INTRODUCTIONModels of patient–provider relationship

In recent years there has been a fundamental societal shift in how we think about patients' role in treatment and decision-making.[1] Moving away from the outdated paternalistic decision-making model, in which the patient typically has a passive role with little autonomy and responsibility, the gold standard is shared decision-making (SDM).

SDM is an interactive process in which patients and healthcare providers share information about available treatment options, risks and benefits and personal preferences for specific treatment options. SDM entails decision-support counselling (deliberation of options) and joined implementation of the decision.[2,3] Thus, SDM is neither paternalistic (i.e., the physician decides alone) nor does it leave the patient alone.[2,4,5] A recent integrative model of SDM[4] defined the nine most essential elements necessary for SDM: define/explain the problem, present options, discuss pros/cons, patient values/preferences, discuss patient ability/self-efficacy, provider recommendation, check/clarify understanding, make or explicitly defer the decision, arrange follow-up.[4,6] SDM has been recommended as an ethical imperative and is considered a hallmark of patient-centred care.[7–9] The ideal SDM process[6] fosters a patient's right to self-determination and places value on his or her voice. Thus, enabling the delivery of truly individualized care,[10] which is at the heart of patient-centred care.

Although there appears to be a widespread consensus among stakeholders (e.g., patients, policymakers and providers) on the relevance of SDM and despite the increasing international efforts to promote SDM, implementation has proven to be difficult and slow.[8,11] As part of a larger project to promote SDM in urological practice, we, thus, set out to investigate the obstacles that prevent patients from participating in SDM.

Relevance of participation preference

SDM has quantifiable benefits; the involvement of patients in treatment decisions is, for example, associated with better treatment adherence,[12–14] more accurate knowledge about their condition and recommended treatments[15] and less decisional conflict.[16] Ultimately, this leads to higher satisfaction among patients[16,17] and physicians[7] alike. Some studies even report a higher quality of life.[18,19] Beyond these sizable effects, SDM is also considered the ethical imperative for treatment decisions as it fosters patients' right to self-determination.[9,20] Consequently, most modern healthcare systems have implemented legislation to facilitate SDM.[8]

Despite these benefits, not all patients seek participation in decision-making to the same extent.[15,21–23] Although a general trend towards a stronger preference to participate has been observed over the last decades,[24] about a quarter of patients (Md = 27% in a review of 31 studies[23]) express a low desire for autonomy. Research has recently started to identify reasons for this.

Existing research reported lower participation preference in patients with certain sociodemographic characteristics. In particular, older patients, less educated individuals and men[18,25–27] express lower interest in active participation. However, a comprehensive review indicates that the exact nature and mechanisms involved in such sociodemographic variables' on participation preference remain to be specified in cancer patients.[28] Thus, one goal of this study was to investigate the association of sociodemographic variables on the participation preference of urooncological patients.

Power imbalance as a potential barrier

It has recently been suggested that a seemingly low desire for patient autonomy may be due to a perceived lack of personal ability to participate rather than a genuine lack of actual desire.[29] In particular, the power imbalance (i.e., the perceived permission to participate) in the physician–patient relationship has been hypothesized to be a critical determinant of patients' ability to participate in medical decision-making. Patients often underestimate the value of their contributions to the consultation, that is, their ability to understand medical information and the importance of their personal preferences,[30,31] which contributes to an increased feeling of a power imbalance. Consequently, these individuals tend to delegate decisions to medical experts. The asymmetry in the physician–patient relationship is deepened if patients feel dependent on their physician's benevolence.[32]

For example, patients may fear being labelled a difficult patient and the negative impact that such a label could have on their care.[33–35] Thus, they refrain from asking too many questions or asserting their interests, such as actively participating in decision-making. All too often, the normative belief prevails that a ‘good’ patient is characterized by conformity and passivity, which may overshadow patients' desire for autonomy.[36] Several studies report that even when patients witness a potentially harmful and preventable event during their care (e.g., a lack of hygiene measures), a majority of (cancer) patients opt not to speak up or assert their interest.[37–39] This phenomenon has previously been labelled as white-coat silence,[40] and it is closely related to patients' attitudes regarding their role in decision-making and beliefs about what constitutes a ‘good’ patient. This effect is particularly pronounced when the stakes are high, as is the case for life-threatening illnesses due to the increased dependence on the physician. In extreme cases, patients may even describe themselves as being hostages to the situation.[41]

Considering patients' attitudes and beliefs about their role thus appears essential in optimized SDM. However, these patient variables are not routinely assessed even when SDM is carefully implemented. Therefore, it is crucial to investigate the potential barriers that attitudes and beliefs impose and may curtail patients' potential to participate in medical consultation actively. Thus, we seek to analyse the relationship between patients' attitudes and beliefs about their role and their participation preference.

Attitudes' and beliefs' influence on behaviour

The relevance of attitudes and beliefs for behaviour preference (i.e., participation preference) is well-founded in psychological theories such as the theory of planned behaviour.[42] From this perspective, a particular behaviour (e.g., participation in decision-making) is preceded by an intention (or behavioural preference).[43] In the SDM context, this implies that the act of participating requires a certain participation preference. The theory of planned behaviour postulates three important determinants for such a specific behavioural preference: attitudes towards the behaviour, control beliefs and normative beliefs. We, therefore, examine whether the relationship between patient characteristics and participation preference is mediated by attitudes towards the behaviour (participating in decision-making), control beliefs (presence of factors that facilitate or impede participation, e.g., lack of knowledge), as well as normative beliefs (what defines a ‘good’ patient).

Importantly, prior work has identified the potential of targeting attitudes and beliefs regarding SDM in healthcare professionals to foster change in behaviour intentions and behaviour itself.[44] However, there are no investigations on the role of attitudes and beliefs in patients.

Current study: SDM in urooncology

SDM has been used and evaluated in a diverse range of medical subdisciplines. A recent review by the American Urological Association (AUA) concluded that it is woefully underutilized[45] in the urological domain. However, SDM is increasingly requested in treatment guidelines and has proven to positively impact urological patients' knowledge, decisional conflict and quality of life.[45]

Thus, in the context of a larger project to foster SDM in general urological practice, we set out to study barriers that impede urological patients' desire and ability to participate. Integration of SDM and engagement of patients is especially relevant in urology because patients in urology and urooncology often face very difficult and highly preference-sensitive decisions,[46,47] in which patient participation can have favourable effects. For instance, early-stage prostate cancer treatment demands patients and physicians to collaboratively decide among many treatment choices in equipoise, such as active surveillance, surgery and various forms of radiation treatments.[47,48] Regarding the participation preference of patients in urology, there have been a few studies on prostate cancer. In this patient group, there is a relatively high preference for participation compared to other tumour entities[22,23]; however, around 10%–15% of patients report a low participation preference.[15,23,49]

While previous research has typically addressed the influence of stable sociodemographic variables[18,25–28] on participation preference, we also aimed to gather information on potential mediators. We hypothesize that the previously reported association between certain patient characteristics and a low preference for participation[18,26,50] is mediated by a perceived power imbalance, which we expect to be reflected in patients' attitudes towards active participation, control beliefs and normative beliefs about the patient role.

With this research, we aim to identify potential targets for interventions that may help to empower patients who are reluctant to engage in SDM. Unlike stable sociodemographic characteristics, attitudes and beliefs are more accessible to change[43] and, therefore, may be ideal targets for structured interventions to help foster patient engagement and increase patient participation in the SDM process. For example, rather than assuming that older cohorts will most likely not be as open to active participation in decision-making, knowing more about the mechanisms in this process may encourage clinicians to pay close attention to their attitudes and beliefs.

METHODSParticipants

We recruited a sample of *N* = 468 consecutive patients who had a scheduled appointment or presented unscheduled with urgent symptoms at the outpatient clinic of the Department of Urology and Urosurgery at University Medical Center Mannheim, Germany. We specified the following inclusion criteria: at least 18 years old, adequate proficiency in German and the ability to give informed consent. We excluded 3 patients with obviously invalid responses (i.e., only marked the middle response category on all questionnaires) and 15 patients (3.2%) with more than 50% missing data. In addition, 16 patients (3.4%) dropped out after giving informed consent.

Excluded patients (*n* = 31, 6.6%) were compared to those in the final sample (*N* = 434) by Pearson χ^2-tests. When necessary (a cell with an expected cell count below one), we calculated Fisher's exact test.[51] There were no differences between excluded patients and the final sample regarding the type of appointment (appointment or unscheduled), diagnosis and occupational status (all *p*'s >.117). However, more excluded patients lived alone ($\chi^2(1) = 20.56$, $p \leq .001$, $\varphi = -0.21$), were female ($\chi^2(1) = 6.761$, $p = .017$, $\varphi = 0.121$) and had a lower level of education (no university degree) ($\chi^2(1) = 6.421$, $p = .011$, $\varphi = -0.117$). All effect sizes are considered small.[52]

The final sample of *N* = 434 was between 19 and 89 years old (*M* = 62.5, SD = 13.5), primarily male (87.3%) and with a scheduled appointment (88.7%). Patients had a variety of urological diagnoses (42.2% urooncological).

Data collection and measures

Patients were approached before their consultation with a urologist by a nurse or a research assistant; they were informed about the protocol. After they signed informed consent, patients provided sociodemographic characteristics and filled in a set of self-report questionnaires, including the Autonomy Preference Index (API)[53,54] to assess participation preference and the Patients' Attitudes and Beliefs Scale (PABS).[55] Additional questionnaire data, for example, on patients' anxiety and depression, are reported elsewhere.[56,57] Medical diagnoses were retrieved from patients' electronic health records.

Demographic and medical variables

We collected standard sociodemographic information about patients' age, gender, level of education, marital status and living arrangement. We dichotomized the following variables: education (0 = without and 1 = with a university degree), living arrangement (0 = living alone or 1 = with others) and occupation (0 = currently unemployed/retired or 1 = currently either employed/in training). In addition, medical diagnoses were categorized as (0) nononcological versus (1) oncological. For consistency, we applied dichotomization cut-offs established in previous studies.[56,57]

Participation preference and intention to participate

The primary outcome of our study is patients' behaviour preference for participating in medical decision-making. Patients' preferences for involvement were measured with the German version of the API.[53,54] The measure consists of two subscales: decision-making (API-dm) and information-seeking (API-is). The German API-dm subscale has four items, which assess generic participation preference, and the API-is contains seven items, which assess information seeking. On both subscales, a linear transformation (0–100) is applied.[58] Higher scores indicate a stronger desire for autonomy. We report descriptive statistics for the API-is subscale. However, because there were high ceiling effects and little variance, the subscale was not used in further analyses (compare, e.g., Benbassat and colleagues[27,59,60]). The primary outcome of our study, patients' participation preference, was thus assessed with the API-dm subscale. The API-dm is often used on its own to assess participation preference, and it has been validated in various languages (e.g., German) and settings.[24,61] Internal consistency for the scale was good $\alpha = .85$.[58]

In addition, we generated an item to ask patients about their intention to participate: ‘Regarding your upcoming consultation: Do you intend to participate in decision-making?’. The item was rated on a 6-point Likert Scale ranging from (‘Absolutely not’ to ‘Absolutely yes’).

Attitudes and beliefs

We assessed patients' attitudes and beliefs about their role in shared treatment decisions using the PABS.[55] The PABS was translated to German by the first author and independently back-translated by a bilingual speaker. There were no discrepancies between the two versions. The German version can be obtained in Supporting Information: Appendix A. The PABS consists of 12 items on 2 subscales: 7 items for positive attitudes (e.g., ‘I have the right to make my own medical decisions, after all, it's my life’) and 5 for negative attitudes (e.g., ‘It would offend my doctor if I were to make my own decision(s)’). Patients rate their agreement to the items on a 5-point Likert scale ranging from ‘do not agree at all’ to ‘fully agree’. The original English items were generated by multiple patient focus groups, strengthening the content validity of the scale.[55] Internal consistency for the negative and positive subscale was acceptable with $\alpha = .71$ and .73, respectively.[55] Total scores were calculated by summing up all items on the respective subscales (positive attitudes and beliefs and negative attitudes and beliefs) and linearly transforming them to range from 0 to 100.

Statistical analyses

A drop-out analysis was conducted to assess systematic differences between completers and noncompleters (see Section 2.1). We compared patient characteristics for patients with urooncological versus nonurooncological diagnoses with χ^2-tests. Furthermore, patient subgroups were compared (independent sample *t*-test) regarding participation preference, attitudes and beliefs. Univariate descriptive statistics are reported for participation preference, attitudes and beliefs and intention to participate, along with the Pearson correlation coefficients for the bivariate associations between these variables. The association between nominal scaled sample characteristics (e.g., gender) and the dependent variable participation preference were analysed with the correlation ratio η.

The expected mediation (i.e., sociodemographic variables—attitudes and beliefs—participation preference) was assessed with a path model (based on structural equation modelling). We checked the multivariate normality assumption between all included endogenous variables with the MVN R-package.[62] The path analysis was calculated with the lavaan R-package[63] using R.[64] The complete list of the R-packages is listed in Supporting Information: Appendix B. All other analyses were conducted in IBM SPSS, Version 27.0. Significance for all tests was set at $\alpha = .05$. Where applicable, we report effect sizes; interpretation is based on Cohen's taxonomy.[52]

RESULTSSample characteristics

The final sample (*N* = 434) was between 19 and 89 years old (*M* = 62.5, SD = 13.5), mostly male (87.3%), and German nationals (97.5%). The majority of patients had scheduled appointments (88.7%). Patients had a range of urological diagnoses (42.2% urooncological: 27.2% prostate cancer, 8.1% bladder cancer, 6.3% other urooncological tumour entities, 0.7% missing information). Further sociodemographic characteristics are reported in Table 1.

Table 1 Sample characteristics and comparison of oncological with nononcological patients

Characteristic	Full sample (*N* = 434)		Oncological patients (*n* = 183)		Nononcological patients (*n* = 251)	
	n	%	*n*	%	*n*	%
Gendera						
Male	378	87.1	169	92.3	209	83.6
Female	55	12.7	14	7.7	41	16.4
Nationality						
German	423	97.5	181	98.9	242	96.4
Other	11	2.5	2	1.1	9	3.6
Living arrangement						
Alone	93	21.4	35	19.1	58	23.1
With others	341	78.6	148	80.9	193	76.9
Higher education						
No	277	63.8	115	62.8	162	64.5
Yes	157	36.2	68	37.2	89	35.5
Occupation status						
Unemployed	11	2.5	3	1.7	8	3.2
Apprenticeship/training	24	5.5	7	3.9	17	6.8

Employed	173	39.9	56	31.1	117	46.8
Retired	222	51.2	114	63.3	108	43.2

Note: Diverging numbers of patients from the total sample size are due to missing values.

a

The option ‘divers’ was available but chosen by no patient.

The subgroup of oncological patients (*n* = 183) contained significantly more males ($\chi^2(1) = 7.295$, *p* = .007, φ = −0.13), more were retired ($\chi^2(3) = 17.17$, *p* = .001, φ = 0.2), and they were on average older (*M* = 66.7, SD = 11.2) than the nononcological patients (*M* = 59.4, SD = 14.3), *t*(429.497), *p* ≤.001, with a medium effect size of Hedge's g_s = −0.559.[52] See Table 1 for further comparison of both subgroups. However, the two diagnostic groups did not differ in their attitudes and beliefs, intention to participate or the primary outcome participation preference (all *p*'s ≥.104).

Patients' participation preference and associated variables

Overall, most patients wanted to participate to some degree in decision-making (*M* = 44.8, SD = 26.5). Descriptively patients reported higher scores for positive attitudes (*M* = 64.9, SD = 14.4) than for negative attitudes (*M* = 55.8, SD = 12.8). Univariate descriptive statistics of metric variables are presented in Table 2. Additionally, we included bivariate correlations in Table 2. Patients' self-reported preference for information seeking (*M* = 95.9, SD = 8, Range = [0–100]) and intention to participate in the upcoming consultation (*M* = 5.4, SD = 0.9, Range = [1–6]) had ceiling effects, and because of the variance restriction in these two variables, they are precluded from further analyses.

Table 2 Descriptive statistics and bivariate correlations of participation preference with predictor variables

Variable	1	2	3	4	5	6	*n*	*M*	SD
•1. Participation preference (API-dm)	-						427	44.8	26.5
•2. Information preference (API-is)	.036	-					433	95.9	8.0
•3. Age	−.166**	.114*	-				434	62.5	13.5
•4. Intention to participate	.258**	.152**	.152**	-			422	5.4	0.9
•5. Positive attitudes (PABS-P)	.383**	.166**	−.016	.305**	-		426	64.9	14.4
•6. Negative attitudes (PABS-N)	−.317**	−.044	.149**	−.14**	.02	-	423	55.8	12.8

Note: Differences in the number of patients (*n*) are due to missing data.

Abbreviations: API, Autonomy Preference Index; API-dm, decision-making subscale of the API; API-is, information-seeking subscale of the API; PABS-N, negative subscale of the Patients' Attitudes and Beliefs Scale; PABS-P, positive subscale of the Patients' Attitudes and Beliefs Scale.

*

p ≤.05;

**

$p \leq .01$.

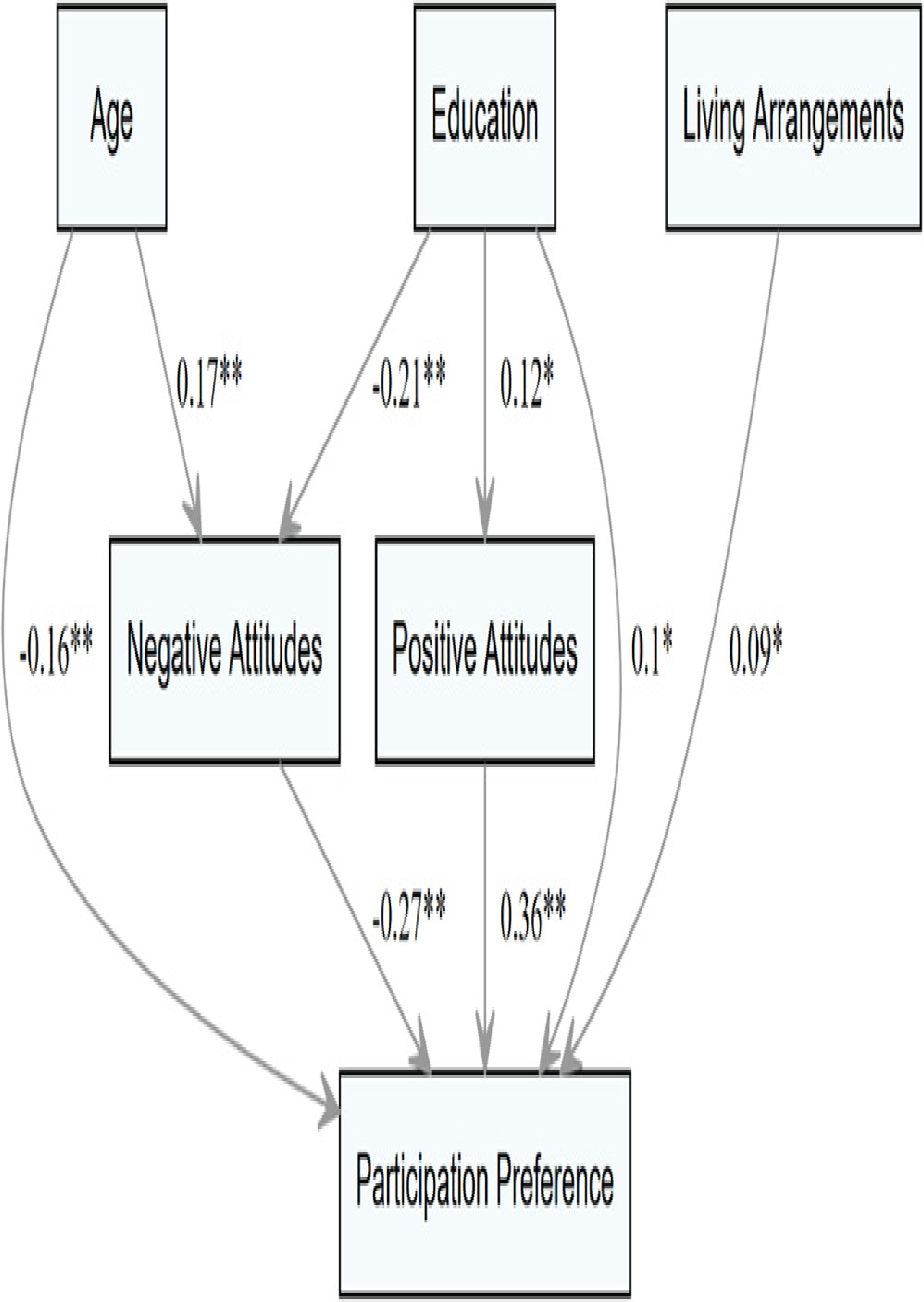
The primary outcome, patients' participation preference (API-dm), correlated highest with the two attitudes subscales (positive attitudes: $r = .383$, $p \leq .01$; negative attitudes $r = -.317$, $p \leq .01$), with medium effect sizes.[52] Age correlated negatively with participation preference, with older patients reporting a lower participation preference ($r = -.166$, $p \leq .01$), corresponding to a small effect size. Furthermore, patients' participation preference was significantly associated with education ($\eta = 0.191$, $p \leq .01$), occupation ($\eta = 0.2$, $p \leq .01$) and living arrangement ($\eta = 0.107$, $p \leq .05$), each with a small effect size.

The associations of patients' gender and type of diagnosis with participation preference were nonsignificant ($p \geq .14$) and, thus, excluded from the multivariate mediation analysis. Furthermore, we excluded occupation from the mediation analysis due to substantial multicollinearity with age ($\eta = 0.7$, $p \leq .001$).

Based on the results of the bivariate analyses described above, the following variables with a significant association with participation preference were included in the mediation model: age, education and living arrangement as exogenous predictors; positive and negative attitudes as endogenous mediators of the effect and participation preference as the endogenous outcome variable.

Mediating effects of patients' attitudes and beliefs

The assumptions of multivariate normality for all endogenous variables included in the path analysis (negative attitudes, positive attitudes and participation preference) were examined with the Mardia Test of Skewness ($b1p = 11.2$, $p = .34$) and Mardia Test of Kurtosis ($b2p = 1.4$, $p = .16$)[65] as well as with the Henze–Zirkler test (HZ = 1.001, $p = .1$). We included patient characteristics which were significantly bivariate associated with participation preference as exogenous predictors in the path analysis (i.e., age, education and living arrangements). Positive and negative attitudes and beliefs were entered into the model as mediators. Since all paths are theoretically plausible, no constraints were set. Thus, the model is fully saturated (no fit characteristics can be reported). The path model is depicted in Figure 1. The model explained 27.8% of the overall variance in our primary outcome participation preference.



Enlarge this image.

We provide an overview of all effects in the path analysis in Table 3. All three exogenous predictor variables (patients' age, education and living arrangements) significantly affected participation preference (Table 3). Higher age (*ß* = −0.05, $p \leq .01$), a lower level of education (no university degree) (*ß* = −0.2, $p \leq .001$) and living arrangement (living alone) (*ß* = 0.12, $p \leq .01$) were associated with lower participation preference.

Table 3 Effects in the mediation analysis between sociodemographic variables and participation preference

Abbreviations: API, participation preferences as measured with the Autonomy Preference Index; Edu, level of education; Liv, living arrangements of the patients; Neg, negative attitudes; Pos, positive attitudes.

Negative attitudes partly mediated the effects of age and education on participation preference. Higher age and lower education levels were both associated with more negative attitudes (Figure 1), which significantly predicted lower participation preference (*β* = −.27, $p \leq .001$). More positive attitudes further mediated the effect of education on participation preference. Last, living arrangements directly affected participation preference but had no significant relationship with positive or negative attitudes.

CONCLUSIONDiscussion

Shared decision making (SDM) is generally recommended in medical consultations, but there are barriers to its successful implementation. Some of these barriers (i.e., attitudes and beliefs) are clearly more accessible to change than others (i.e., age). We examined their combined effects in patients who faced preference-sensitive decisions in urological consultations. Interestingly, we found that patients' attitudes about their role in the interaction and their personal beliefs about acceptable behaviours in the decision-making process were strong predictors of patients' participation preference in decision-making.

With reference to earlier studies, we replicated the association of participation preference and sociodemographic characteristics,[26] for example, age, education, occupation and living arrangements in urological patients. Specifically, we found that older patients and those who are less educated and live alone have a lower generic participation preference. More importantly, as a noteworthy extension of the existing literature, our data show that patients' attitudes and beliefs mediate their influence on participation preference. This novel finding aligns with recent theoretical considerations suggesting that less educated and older patients may not feel empowered to participate even if they wanted to.[30,41]

Overall, the predictors in our mediation path model explained around 28% of the variance in patients' participation preferences, which corresponds to the strong explanatory power of our model.[52] In previous research, multiple regressions were mainly used to analyse factors influencing participation preferences and explained between 3% and 21% of the variance.[66–68] While our mediation path model explained a substantial amount of variance and, more importantly, uncovered the mediating effects of attitudes and beliefs, it still leaves room for improvement. Future studies could include other relevant influencing factors, for example, a measure of perceived behavioural control.

Our findings underscore the importance of assessing patients' attitudes and beliefs regarding their role in decision-making. Instead of assuming that older or less educated patients would not want to participate, we found that these patients are encumbered by negative attitudes and beliefs about their role. To enable and adequately support patient participation, clinicians should challenge patients' negative beliefs (e.g., ‘It would offend my doctor if I were to make my own decision(s)’).

Furthermore, our findings emphasize the general notion that decision-making is not always rational. Instead, negative emotions (e.g., fear of being labelled as a bad patient[29,33]) can induce avoidance, even if this behaviour incurs costs (see our experimental work; e.g., Pittig et al.[69]). In patients with negative attitudes and paternalistic beliefs, this may hamper patients' expressing their personal needs.

Patients are thought to be especially prone to experiencing power imbalance in the relationship with their physicians when the stakes are high, for example, when they face a life-threatening disease.[41] However, we did not find a difference between the oncological and the nononcological subsamples. A possible explanation is that the nononcological patients in our sample presented with complex and long-lasting medical conditions and consequently faced substantial burdens as well (see Köther et al.[57]). In line with this, it is possible that patients' attitudes, beliefs and participation preferences are not directly associated with illness severity but rather with the perceived burden and impairment associated with it.

In addition, there were significant differences between the oncological and the nononcological subsamples, for example, the urooncological sample contained more male patients, they were on average older and more were retired. Thus, the high homogeneity in this sample and lack of variance could have limited the possibility of finding differences in attitudes and beliefs. This might limit the generalizability to the bigger concept of the influence of illness severity (i.e., life-threatening or not). However, it does not speak against the generalizability to urological patients, as the described differences in the distribution of gender, age and occupation are typical in urology versus urooncology.

Regarding the information preference of the patients, we found considerable ceiling effects in the API-is, similar to previous reports.[27,59,60,70] Unfortunately, the lack of variance obtained by the measurement prevented us from including it in further analysis. However, it is important to recognize that urological and urooncological patients generally have a genuinely high preference for being informed, even when reporting a lower preference for participation. The finding of a universally high preference for information is consistent with studies on other patient groups, for example, patients at the general practitioner[70] or patients with end-stage renal disease[71] or a study with over 5000 older adults who all have a very high desire for information.[72]

Thus, to treat uro(onco)logical patients according to their preferences, they should readily receive information on diagnostics, treatment options and side effects regardless of their preference for participation in the decision-making process.

Limitations

One limitation of our study is the utilization of the attitudes and beliefs scale as a proxy for a power imbalance. Even though attitudes and beliefs, as measured by the PABS,[55] were the strongest predictors of participation preference and capture essential aspects of the power imbalance in the patient–physician relationship, such as attitudes towards the behaviour, control and normative beliefs, the questionnaire does miss out on some relevant facets. While we recognize the importance of having a short questionnaire, we argue that the five items on the negative subscale (PABS-N) are missing some important concepts.

For example, the PABS-N lacks items that assess perceived dependency on the physician, the avoidance of speaking up or how the patients value having a good relationship with the physician. An additional direct measure of perceived behaviour control, for example, self-efficacy in the context of medical decision-making, would be an interesting extension in future studies. Moreover, there is a limitation in the way the PABS was translated. While we did assess the quality of the translation with an independent bilingual speaker, as well as a short pilot with urooncological patients, the validity of the translation could have been improved by further conducting in-depth cognitive interviews. To address these limitations, we are currently developing a questionnaire that is explicitly designed to capture the construct of power asymmetry in the relationship in broader terms.

Furthermore, generalization from the chosen urological sample to other patient groups maybe not be warranted. Our sample was primarily male and of higher age. While this is expected in a urological sample, it is unclear if our results generalize well to other medical conditions, for example, if the found mediation is only prevalent in primarily male patients. While a patient's gender and primary diagnosis did not have a meaningful effect on participation preference or attitudes and beliefs, it remains an open question if attitudes and beliefs have a mediating effect on patients' participation preference in other cancer types or patient groups.

Finally, structural equation analysis is generally not suitable for identifying causal relationships. However, with respect to the direction of the mediation, it is plausible that age may influence attitudes, whereas the opposite relationship is impossible.

Conclusion and future directions

With increasing international efforts to better implement SDM,[8,73] it is important to identify barriers that limit patients' ability to participate actively. We found low participation preference in older male patients with less education to be mediated by patients' attitudes and beliefs about their role in decision-making. However, such patient characteristics cannot be (easily) modified. In accordance with psychological theories, it may be much more promising to empower patients by actively targeting their attitudes and beliefs (see Ajzen and colleagues[43,44]). This may open up new avenues for interventions that prepare patients for the decision-making process. In SDM, the fear of burdening the physician by asking too many questions[74] may be amenable to change. Such interventions should aim to redefine perceptions of what constitutes a ‘good patient’. Successful implementation of SDM may require a shift in the power asymmetry.[75] One way to achieve this may be to explicitly challenge patients' attitudes and beliefs about the inferiority of their role in the decision-making process.

An obvious first step may be to reassure them that participation will not result in a negative consequence but instead could be a valuable resource for the decision-making process. Interestingly, small interventions may have a profound impact, as even the most simple instructions have previously shown promise in changing responses to others.[76] For example, SDM implementation research has previously tried to engage patients, that is, by giving them question prompt lists (e.g., the cancer consultation preparation package[77] or the AskShareKnow campaign[78]). Such question prompt lists showed some promising first results, as they increased patient activity and led patients to ask more questions.

Yet, it is often difficult to employ such interventions in clinical practice most importantly due to time restraints. Outsourcing these interventions, for example, to online decision aids, may ameliorate this. In urooncology, existing decision aids generally have not addressed patients' attitudes and beliefs,[79] and thus, they may miss an opportunity to pave the way for SDM. Also, participating in online self-help groups was shown to reduce negative attitudes in (cancer) patients and should be encouraged by clinicians.[80] Implementation of SDM in urology and urooncology is especially important,[45] given that patients, in the course of their treatment, face many preference-sensitive decisions.[47,48,81] Research on SDM has documented the positive impact patient participation can have in urology.[82–84] At its core, SDM requires a fundamental shift in power,[85] which can only be achieved by the active empowerment of patients in urology. Future studies should examine if targeting attitudes and beliefs, for example, in decision aids, holds its promise and helps empower patients in the process of SDM.

It is important to consider that patients' negative attitudes and beliefs and the accompanying fear of being perceived as a ‘bad patient’ may arise from experience. A patient may become quickly disheartened if he/she is willing to participate but experiences a lack of time or responsiveness from the clinicians. In line with the title of a recent SDM intervention study, ‘Changing patients but not physicians is not enough’,[77] it could be a valuable measure for SDM adaption to also prepare physicians to embrace their patients' willingness to participate in decision-making.

Finally, our study highlights the importance of patients' attitudes and beliefs regarding the perceived power asymmetry before the consultation for their active participation. Building on that, an interesting path for future studies would be the investigation of the experienced power asymmetry in the medical encounter and the influence on the perceived participation after the encounter.

Practice implications

Our data emphasize the importance of addressing patients' attitudes and beliefs about their patient role in SDM. Targeting attitudes and beliefs can potentially be a valuable intervention to overcome traditional barriers to active participation, such as higher age or lower education. The role of patients' attitudes and beliefs about active participation should be considered in the assessment and targets of future decision aids to empower patients. It may ultimately help to overcome power asymmetry and foster patient-centred healthcare.

AUTHOR CONTRIBUTIONS

Björn Büdenbender: Methodology; data curation; formal analysis; writing –original draft; writing –review &editing; visualization. **Anja K. Köther**: Data curation; writing –review &editing. **Britta Grüne**: Investigation; data curation. **Maurice S. Michel**: Funding acquisition; resources. **Maximilian C. Kriegmair**: Funding acquisition; conceptualization; resources; supervision. **Georg W. Alpers**: Conceptualization; supervision; writing –review &editing; funding acquisition; resources. All authors approved the final version of the report.

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CONFLICT OF INTEREST

The authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

The data and scripts that support the findings of this study will be deposited on MADATA (University of Mannheim, https://madata.bib.uni-mannheim.de/id/eprint/406) Research Data Repository (doi:10.7801/406) and made available by the authors, without undue reservation, to any qualified researcher.

ETHICS STATEMENT

This study was approved by the research ethics Committee II of the medical faculty Mannheim of the University of Heidelberg (MA-2019-635N). The authors confirm that all personal identifiers have been removed or disguised so the patients described are not identifiable and cannot be identified through the details of the story.

DETAILS

Subject:	Urology; Patients; Patient-centered care; Cancer therapies; Prostate cancer; Decision making; Quality of life; Decision analysis; Medical decision making; Power; Participation; Imbalance; Attitudes; Sociodemographics; Preferences; Negative attitudes; Patient participation; Outpatients; Group decision making; Public participation; Autonomy; Beliefs
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Engaging participants with research findings: A rights-informed approach

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ProQuest document link

ABSTRACT (ENGLISH)

Background

Sharing research findings with participants is recognized as an ethical imperative for the research community. However, most discourse on this topic in mainstream public health takes a paternalistic approach, with researchers retaining the power to choose if, when, and how research findings are shared.

Methods

Fieldwork took place from August 2018 to January 2019 and again from August 2019 to December 2019 among two communities in the south Indian state of Kerala. We integrated participant engagement with study findings into the research protocol, using various collaborative strategies identified during the design stage, forming partnerships with participants and determining appropriate forms of dissemination for different participant groups during fieldwork.

Results

Findings from previous research projects undertaken with these communities by other researchers had not been shared with them. This was interpreted by the communities as researchers not being interested in making a difference to their situation. In the current study, building reciprocal relationships that minimized power disparities, and providing outputs in tailored formats that promoted active engagement were key factors that enabled participants to engage with results. This engagement added value by enabling us to co-develop study recommendations. This process also enabled the community to have ownership of the results and use them to advocate for health system change to improve access to health care.

Conclusion

Research should be transformative for participating communities. Participants have a right to know the results of the research they participate in since their knowledge provides the research data which can in turn promote community change. Operationalising this requires researchers to build partnerships with participants and their communities from the outset. The role of participants must be reimagined, and adequate resources should be built into the research process. This is both socially responsible and ethical, but also improves the impact and legitimacy of research for the participants and the communities that they represent.

Patient or Public Contribution

Participants of our research contributed to the design of various aspects of the engagement processes including the venue, the formats used for engagement, interpretation of the findings and recommendations from our research.

FULL TEXT

BACKGROUND

Health research has the innate capacity to affect the lives of both the participants and the larger communities within which the research is embedded since the knowledge generated can enable societies to better organize themselves to advance their health goals.[1] Given this significance, historically a lot of attention has been paid to ensuring scientific rigour and the integrity of processes such as research design, conduct and reporting.[2] Viewed through this classical lens, the key stakeholders in the research process are usually the ‘expert’ researchers[3] who plan, implement and lead all aspects of an investigation, along with institutions such as funding bodies and universities which have oversight of the research process. Research participants have a time-limited role that ends once they provide the data that was sought from them.[4,5] However, the last two decades have seen a change in this approach and in the understanding of who the ‘expert’ is.[6] Several theoretical frameworks, declarations and organizations have been established to change this status quo and find meaningful ways to involve participants in all stages of the research process.[4,7–11] Key frameworks to have emerged include the Strategy for Patient-Oriented Research-Patient Engagement (SPOR),[7] the INVOLVE framework[9] and the Patient Centre Outcomes Engagement Rubric.[11] Whilst each framework is unique, they all share a high regard for the value of the lived experiences of research participants, an emphasis on engaging participants as equal partners across all stages of research (not just using tokenistic gestures) and a belief in the importance of sharing research findings with participants through a genuine and engaging process.

Most health research participants would like to know the results of the studies to which they have contributed.[4,12] It has been proposed that sharing study findings should be the norm, since they may have significance not just for the participant, but also for their families and communities.[13] Evidence suggests sharing results can also increase participants' sense of ownership of research outcomes, improve trust between researchers and participants and encourage participation in future research.[14] Studies have also shown that participants consider receiving research results as a right.[14–16] Indeed, it has been argued that there is a moral and ethical imperative to share study findings with participants.[17,18] The Declaration of Helsinki for example states that ‘all medical research subjects should be given the option of being informed about the general outcome and results of the study’.[3] It is also recognized that sharing study findings provides better access to participants and acceptability for the research[9,11] and dissemination of results is often included as a requirement in institutional or funding protocols.[19] Researchers as well as funding agencies and other bodies that govern research generally support sharing results with study participants.[20,21] It is increasingly recognized that sharing results with participants should go beyond merely providing a plain language summary of key findings; it begins with engaging participants meaningfully across different stages of a research project using multiple strategies[22,23] and concludes with a shared process of discussion and interpretation of the study findings, to establish their purpose in the real word setting.

Morello-Frosch et al.[24] discuss three possible approaches to understanding the process of sharing research results with participants:

- 1.
  A clinical ethics framework where the focus is on reporting back individual results, especially in situations where clinical action is required to protect the participant. Here, decisions about sharing results rest with scientists and medical experts and the ownership of the data rest with the researcher(s).[25]

- 2.
  A community-based participatory framework where there is a strong focus on communicating both aggregate and individual results to participants using protocols that are jointly developed by both the research team and the participants. Decisions about sharing results are made jointly by the researchers and the participating community.[17] The ownership of data primarily rests with the participants from whom the data has been collected.[26]

- 3.
  Citizen-science ‘data-judo’ where the emphasis on reporting back both aggregate and individual results to participants is guided primarily by the policy goals that the research set out to achieve. The lead here is taken by communities and advocacy groups who have marshalled their own scientific resources to conduct a study with a view to influencing policy change.[27]

Despite this, the reality is that in many cases health research participants still do not receive study results[4,28] nor are they engaged in the feedback of findings. This is often the case with clinical trials but is also true for community-based participatory health research.[4] It has been argued that sharing results is not a simple task. The extent to which participants are engaged is driven by factors including the nature of the research data (the intended user, data sensitivity, ownership, target audience)[29]; the infrastructure available to facilitate data sharing[30–32]; organizational and research context (culture of the organization and discipline-specific practices)[23,33]; the individual characteristics and motivation of researcher(s) (including academic position, prior experience in sharing data as well as expected returns)[23] and existing practices that govern research data sharing (open access publications, institutional and research repositories, sharing upon request, etc.).[34] Open access, strongly promoted by several funding agencies and institutions,[35–38] while making research outputs widely available, primarily seeks to fulfil academic requirements, including establishing researchers as experts in their field. The formats of journal articles also limit the availability and useability of research findings to academic and policy audiences.

Whilst not formally rewarded by existing academic processes, the fact that this topic finds consistent mention in the literature reiterates how important it is for the research community. After all, research is a form of problem-solving, and the value of research lies in finding answers that will ultimately benefit society—either by increasing knowledge, changing attitudes or providing practical solutions. The true benefit of research is therefore only realized once that research is shared with broader society. It is this that motivated us to develop the dissemination processes discussed in this paper. We engaged research participants with our study findings during a community-based research project that explored access to health care for socially excluded communities in the south Indian state of Kerala. Key findings from this research have already been published,[39,40] but were also shared with the communities that participated. We involved our participants at every stage of the study in a range of processes focused on ensuring they could actively engage with project findings once initial data collection and analysis were complete. In this paper, we reflect on our experiences of engaging different participant groups, examine why a rights-based perspective is a more effective approach to address this issue, and discuss strategies that can be adopted by researchers to improve the dissemination and application of their findings at the participant level.

METHODSStudy setting and participants

This study reports on the sharing of results of an ethnographic study carried out in the south Indian state of Kerala between August 2018 to January 2019 and August 2019 until December 2019.[39,40] The study sought to understand how social exclusion impacts access to health care for two marginalized communities: Indigenous communities and older widows living alone. Participants from Indigenous communities were recruited by theoretical sampling, whilst older widows living alone were identified using both theoretical and snowballing sampling strategies. The third group of participants was also recruited from the healthcare professionals (community healthcare workers, nurses and doctors) and key informants (Key informants are those who possess specialist knowledge on account of their experience, expertise and sometimes their position within a community or an organization.) connected to the two communities. These participants were recruited by using stratified purposive sampling.

Methodology

Participants were engaged with results during several phases of the ethnographic study: (1) Building participant rapport and trust; (2) Data collection via interviews, focus group discussions, participant observations and concurrent analysis; (3) Member checking and sharing emerging and final results with participants (the focus of this paper).

Dissemination planning

The decision to ensure that research findings were shared in a meaningful way with participants was made during the research design stage, with an initial dissemination plan integrated into the study protocol. Our belief that participants have the right to know the study findings were also influenced by our respect for the agency of participants. This stance evolved from our previous individual experiences as health researchers, combined with a collective appreciation for the importance of a tailored approach to meaningfully engaging participants with findings. This encouraged us to start discussions early with participants to identify which approach to sharing findings would be most effective for them. It also ensured that we set aside adequate time and resources to engage participants with findings at the end of the study.

Forming partnerships with participants

Ethnographic research places a great emphasis on extended contact with participants and building reciprocal relationships, as this helps avoid power disparities and gives better access to participants' lived realities. In this study, we were particularly conscious of working with socially excluded communities and the healthcare workers who provide services to these communities. Building partnerships of trust and mutual respect was therefore essential for a successful research project. This partnership-building occurred in two stages during the research: Once initial ideas for the research were formalised, the first author, who lived with the communities during the course of the research, held discussions with members of the population groups we expected to work with, to gauge the relevance of the topic to their lives and obtain feedback on our plans for data collection. For the Indigenous communities, meetings were held with a community-based organisation, three Indigenous village chiefs and four healthcare workers providing services to the communities. Regarding the older widows, discussions took place with healthcare workers including two doctors and three community health workers providing services to the widows. Meetings with individual widows at this stage were not feasible due to the dispersed nature of older widows in the geographical area of research.

During the initial months of fieldwork, we took time to understand the specific contexts of different participant groups, respecting local protocols and customs during interactions, and discussing any concerns expressed about research processes and participation:

- (1)
  Among the Indigenous communities, traditional protocols governing the interaction of outsiders with the community, such as meeting the village chief and seeking his permission, were followed.

- (2)
  With older widows, initial interactions were facilitated by a community health worker who was a woman from the area and known to the participants. In addition to explaining key aspects of the research, the initial visit was used as an opportunity to meet the local area representative, be introduced to the neighbours and discuss suitable times and venues when data collection could happen. This was deemed necessary as communities were wary about outsiders visiting older widows living alone.

- (3)
  Healthcare workers were met at the health facilities where they worked. The initial visit provided a forum to break the ice, engage in discussions about the relevance of the research and answer any questions.

Such extended and multi-layered contact enabled the first author to become familiar with the participants and move from being the ‘outsider expert’ to an ‘accepted’ member of the group. This also enabled the first author to better understand existing power differentials such as those between the health system and community participants, as well as between the first author as an expert outsider and study participants. This supported the first author to build relationships that were reciprocal and respectful.

Tailoring dissemination

Having different groups of participants required us to evolve appropriate mechanisms that promoted active engagement and sharing of research findings with each group. This involved negotiating the appropriate location and arrangement for disseminating study findings.

The findings were also shared with a community-based advocacy organization in Attapadi, Kerala. This was to ensure that dissemination of study findings extended beyond immediate participants to relevant community-based groups working in local Indigenous health and wellbeing. We were unable to identify a suitable organization to disseminate findings related to older widows living in the community, as most community-based groups working with older widows catered primarily for those in residential care.

FINDINGSLegacy of nondissemination

During the initial phase of fieldwork, the impact of previous research findings not being shared was a key barrier to building rapport. Several research groups had carried out data collection with the Indigenous communities, but hardly anyone returned to share findings. This lack of engagement by researchers post-data collection, made community members feel researchers were far more interested in ‘getting their work done’ rather than helping them address their situation. This feeling was echoed by other participant groups, including older widows. Some even wondered if researchers accurately reported the information that they had shared in their outputs. They felt that this was unfair as they had spent time and shared information about their lives with researchers. Such experiences also meant that communities were reticent about participating in further research as they considered such exercises futile.

People like you come here for research, but once they finish their work, there is no sign of them. They don't tell us what they have found out and neither do we know what has happened to the report that they wrote. Once they have collected whatever they want no one ever comes back. (Indigenous Community FGD, TI10)

Health system participants (especially doctors) pointed out the benefits of disseminating the results locally and the limitations of not doing so. Many felt that research could only initiate change in the health system where it was carried out if researchers shared their findings in a locally meaningful way.

I don't think so far anyone has come and spoken about their findings here. Usually what happens is that people will do some surveys or carry out interviews. But once that is over, they do not come back. We won't even know where they are or what happened to them after that. (Healthcare provider IDI, MO_4)

Inaccessible research outputs

Most research findings from previous studies conducted in these communities were available as official reports and/or journal articles. However, accessing and making sense of their contents required resources such as internet access and specific skills including proficiency in written English and an understanding of health research terminology. These were significant barriers for community participants (Indigenous people and widows), who were unable to read reports or documents from previous research. Healthcare professionals were also unaware of where to find the outputs of previous research and how to apply the findings to their practice. What evolved in our consultations was that a verbal presentation followed by a discussion of the results either in an individual or a group setting would be far more effective for all participant groups. Merely receiving a copy of a research report was not going to help them engage with the findings or apply them to their lives.

I don't know what they are writing about us. I hope they are writing what we say (laughs). But what can we do about it? We can't read what they write, and they also don't tell us what they finally write in those reports. (Indigenous community FGD, TK12)

Familiar forums foster engagement

Among the Indigenous communities, the village assembly presided over by the village chief was chosen as the appropriate forum to share findings. Aggregate findings were presented one by one followed by a discussion about the key issues. Using this traditional forum and discussion style provided the Indigenous communities with a familiar context which enabled greater engagement and empowered participants to comment on the findings and correct our interpretation.

With the widows, individual meetings were held to talk about the findings on a one-to-one basis in their homes. This suited them better as these participants lived by themselves. Gathering them together would have disrupted their daily routine and put them in the company of others they did not know well. This would have diminished their ability to engage with the findings, ask questions and comment on them freely.

Among healthcare providers, one-to-one discussions were favoured. As with community participants, aggregate findings were shared followed by a discussion and comments on the findings. This allowed participants to discuss findings in some detail, clarify our interpretation and provide further input. Some participants preferred to learn about the findings at the health facility, while others asked for the discussion to be held away from their place of work.

Contextualizing findings increase understanding

Our discussions with the different groups of participants also established that different presentation formats would be more effective for each group. Before presenting study findings, discussions were held with a small number of participants to explore the best format to present the results. These discussions helped us to understand the importance of using imagery, language and contextual information relevant to each population group instead of a generic approach. For example, while discussing the gradual decline of Indigenous healing traditions and the increasing acceptance of western medicine, parallels were drawn to how the official language of the state (Malayalam) had taken over tribal dialects, resulting in tribal terms being replaced with Malayalam words. Thus, the Indigenous word for mother ‘avva’ was replaced by the Malayalam word ‘amma’. Using this language resonated with the community and led to discussions about how the neglect of healing traditions should be seen within the overall context of Indigenous ways of life (including language) not being valued. Among the widows, the key findings were presented within the context of the altered family structure that the state of Kerala had witnessed in the last two decades where the social understanding of what constituted a ‘family unit’ and responsibilities for caring for older family members had changed considerably. This resonated with the lived context of our research participants and set the tone for engaging discussions as they were able to relate our findings to their lives. Likewise, healthcare providers were able to engage better with the findings when they were presented within the context of their experiences of providing health care.

Two-way communication adds value

Two-way communication that occurred as a result of the various processes adopted during the dissemination of findings added value to the research in two specific ways. First, it confirmed that the results captured the lived reality of the participants. Second, this process facilitated the co-construction of findings and enabled us to jointly evolve recommendations from the study. One finding, for example, was about the importance of decentralizing the delivery of healthcare services. During our discussions, some of the widows pointed out that there was a specific aspect of decentralization that would help them the most. Participants explained that while the neighbourhood clinics had made a difference in their ability to access healthcare services, they still had to travel great distances for diagnostics. This feedback enabled us to evolve a specific recommendation about decentralization that was relevant to the community.

Handling multiple perspectives

Some of the study findings showed that the planning and delivery of healthcare services made access difficult for communities. In the case of Indigenous communities, the centralization of health care was a decision taken to ensure that most services were provided under the supervision of specialist doctors. However, this had the impact of excluding many of the villagers who lived far away from this hospital. With the widows, the issue of physical infrastructure that was not friendly for older adults was highlighted as a deterrent to visiting public healthcare facilities even when care was required. We had anticipated that some of the health system participants, particularly those in charge of the local health system might be unhappy with findings that appeared critical of their service provision. We addressed this by ensuring that the language used while describing results did not suggest that any specific individual or institution was responsible for any negative findings. Thus, while discussing the issue of centralization, we encouraged participants—particularly those from the health system—who disagreed with the findings, to present their side of the story. Second, we were careful to communicate to all participants that our findings were meant to identify systemic issues rather than imply criticism of one individual or institution.

The group discussion format employed to engage Indigenous participants enabled greater engagement and discussion among all those present. However, it also posed some unique challenges. For example, in one of the early village meetings, a community member objected to findings that he felt portrayed the local health system in a poor light. We prepared for this by clearly communicating at the beginning of each meeting that it was acceptable to disagree with the results that were being shared and that everyone present had a right to discuss the results and disagree or comment on them.

Using results to advocate for change

An important, though unplanned, the outcome of the process used to share findings with participants was how they were able to own the results and use them to advocate for change. The findings from our study were used as part of the Indigenous manifesto that was released before the legislative assembly elections in the state of Kerala in 2021. This manifesto was shared with the major political parties in the state of Kerala thereby promoting visibility and debate on the study findings. Over the last 2 years, the results were also used to lobby and advocate for decentralization and primary health care in Attapadi and the appointment of 41 Indigenous young women as front-line community healthcare workers. This was made possible because the research findings were:

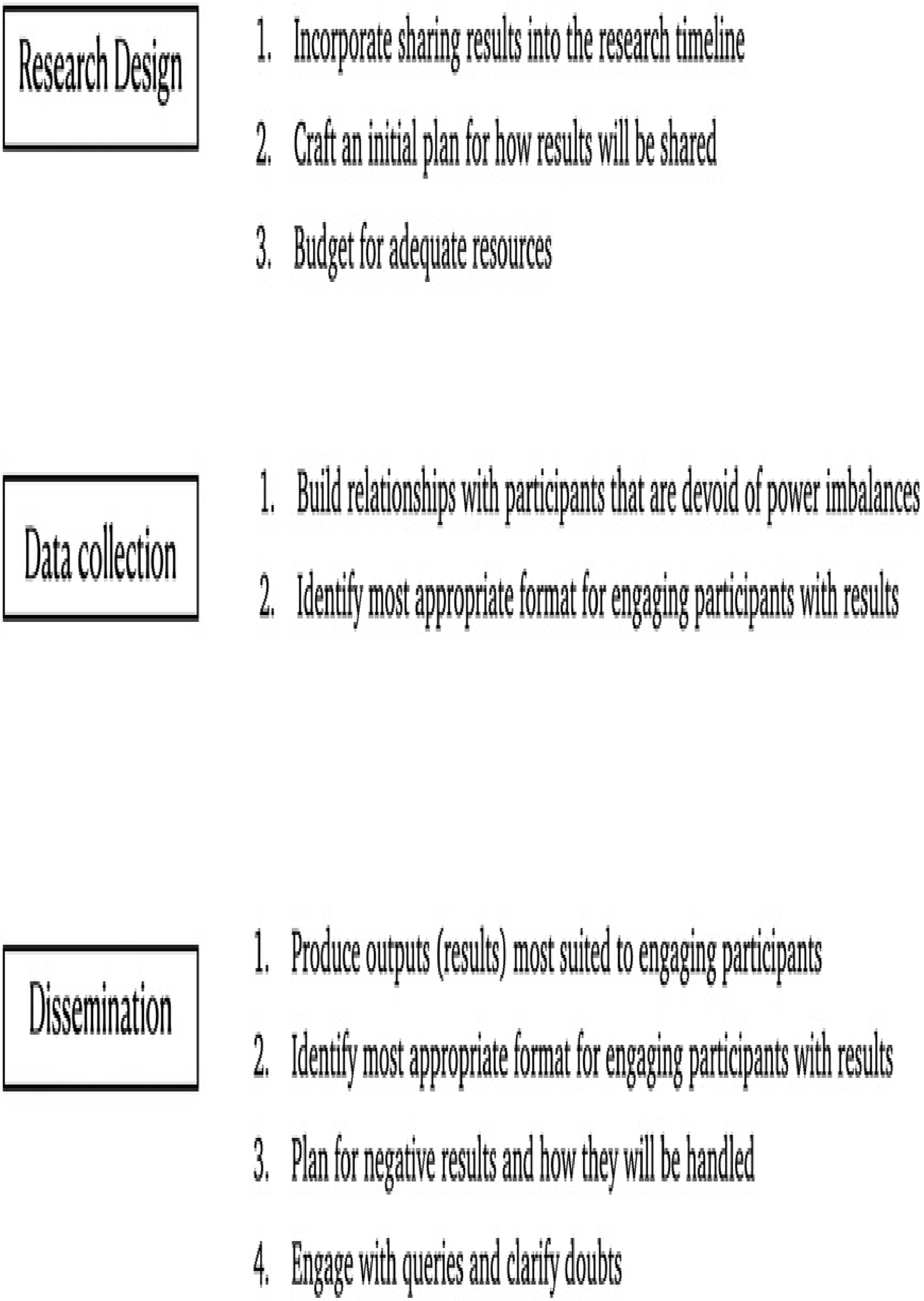
- 1. discussed and shared in a manner that made them accessible to participants and the wider community to which they belonged, promoting ownership of study results;
- 2. shared with a community-based organization working among the Indigenous communities which was involved in health advocacy. This organization was able to use the research findings to drive policy change.

This also demonstrated the importance of sharing results and engaging key stakeholders such as community-based organizations as allies to drive change and impact.

DISCUSSION

The last three decades have seen a growing interest in engaging research participants and ensuring that the findings of health research are freely available to those beyond the traditional consumers of research outputs. Most discussions about sharing research findings state that participants would like to know about the findings.[12,15,41] However, it is also acknowledged that in practice research findings are not shared as often as they should be.[15,22,41] Indeed, whilst health researchers agree on the importance of making research publicly accessible, it is often admitted that it is ‘easy to become untethered from these foundational principles’.[42] The failure to share research findings with participants has been shown to impact researcher credibility and an individual's future participation in research.[4,43,44] We found this to be true in our study. Most participants had taken part in research previously, and not knowing what was written about them made them feel that research and researchers were more concerned about their own careers than improving community living conditions.

Publishing in open access journals is highly recommended as a way of making research results widely available. However, journal publications are unsuited to the effective dissemination of results to research participants and in fact, heighten the power difference between local communities and researchers.[45,46] A lot of research published about the Indigenous communities who took part in this study was available online in the form of reports, journal articles and newspaper reports. However, for participants to access these, understand the key issues and relate this to their lives, requires a specific set of skills including fluency in English, access to the internet, as well as knowledge of technical terminology common in such outputs. Not having these skills is a key barrier to participants' access to research findings. A more effective mechanism for dissemination to participants is to arrive at formats of sharing via negotiated consensus. These formats should take account of participant profiles including language spoken at home, knowledge-sharing traditions (oral vs. written), local context and culture. Doing this enabled us to arrive at three different formats of sharing study findings which helped participants to engage with findings effectively. For this to occur, the value of sharing research findings with participants must be incorporated into the research design, and steps should be taken throughout the research process to engage with participants and identify the most effective formats for them to engage with the findings (Figure 1). While the actual sharing of results occurred towards the end of the study, engaging with participants throughout the research process was critical in ensuring that we moved beyond the tokenistic sharing of results to active participant engagement. The various engagement activities from forming partnerships to tailoring dissemination and promoting two-way communication ensured that our participants had control over some of the decisions that were taken regarding the sharing of results such as the format, the location and the forums that were used. These processes ensured that participants moved from nonparticipation or tokenism to involvement at the level of citizen power as described by Arnstein while discussing citizen participation.[47] Achieving this from our experience requires that the sharing of results needs to be planned early and is in keeping with previous recommendations.[2]



Research Design

1. Incorporate sharing results into the research timeline
2. Craft an initial plan for how results will be shared
3. Budget for adequate resources

Data collection

1. Build relationships with participants that are devoid of power imbalances
2. Identify most appropriate format for engaging participants with results

Dissemination

1. Produce outputs (results) most suited to engaging participants
2. Identify most appropriate format for engaging participants with results
3. Plan for negative results and how they will be handled
4. Engage with queries and clarify doubts

Enlarge this image.

While the literature provides several reasons for research findings not being shared with participants, one issue that gets little attention is the fact that current reward structures for researchers rarely incentivize the sharing of research findings beyond publications in high-impact journals.[22,28] This is unlikely to change until the role of the academic is redefined to enable researchers to move beyond theory development and testing to embrace research as a strategy for social change. There is a movement within academia to change this approach with various frameworks (e.g., Participatory Action Research [PAR], transformative research approaches and feminist methodologies) espousing the value of working with study participants as equal partners throughout the research process, including sharing study findings. However, a lack of validation at a systemic level makes this a challenge, one which can only be overcome with a change in how we (mainstream public health academics) conceptualize and teach about the role of health research, the values that we instil in research students, and the criteria by which academic success is judged. Cross-disciplinary insights particularly from fields focused on social causes such as sociology and social work could also be of benefit.

One question that researchers might have about sharing findings is, what will participants and their communities do with the findings that have been shared with them? We found that one of the important outcomes of the process we followed, was the co-construction of study findings which gave the community ownership. This in turn enabled the community to use them to drive change in the local health system and raise some of the issues identified in larger debates about access to health care with government officials. The involvement of a community-based advocacy group, which also participated in the research, acted as a catalyst in this process. This clearly shows that communities have agency and when research findings are shared with them in the right format and committed community organizations are included in the dissemination process, they can use that information to take positive action and drive locally relevant change. This process of co-construction by the participants and their communities further helped to address the power differentials that are a norm between researchers and study participants, by giving them the opportunity to decide how they are defined[48] and providing appropriate input to the study findings. This agreed interpretation of study data ensured that the findings were not only transparent, accessible and relevant to participants' lives, but more importantly, it strengthened the validity of the findings, ensuring they were an accurate reflection of participant values, beliefs and experiences.

Communicating effectively with participants about study findings requires researchers to work with participants to identify the most suitable way to do so.[49] In our experience, this is key if participants are to fully engage with results. Attending the village assembly for the Indigenous community participants, and one-to-one meetings at a location chosen by the older widows and health system participants, enabled greater engagement, as opposed to a one size fits all approach. Avoiding technical jargon and using local idioms, images and phrases where feasible is also essential. This may well be an area where researchers need training in forms of communication that are effective when communicating with lay audiences.

Rights-based approach to engaging participants with results: A way forward

Research participants make an important contribution to the success of any research by sharing their information. Without this contribution, research is not possible. This we believe grants participants the right to know the outcomes of the research they were involved in. Most discussions on engaging participants and sharing research results promote this either as an ethical stance that the researcher should take or as a way to add value to the research.[4,7–11,24,25] A rights-informed approach goes beyond these by perspectives not only including in its ambit ethical and accountability arguments but also acknowledging that participants, by virtue of their contributions to the success of the research, have a right to know the outcomes. Second, it frames the sharing of results as an obligation to participants, rather than an optional activity that can be left to the discretion of researchers or institutions. This also addresses the power differentials that other approaches confer on researchers and institutions when it comes to sharing results.

As previously noted, transformative research frameworks such as PAR already promote sharing research findings as an effective strategy for addressing the power imbalance between participants and researchers.[26,50] We believe, however, that sharing findings with participants should not be restricted to frameworks such as PAR but rather is relevant to all health research. It is the right of every research participant, not only those who participate in community-based research.

When research participants do not have access to the results, it further adds to the distance between researchers and participants. Consequently, research remains something that is distant and disconnected from people's daily lives and their improvement.[51] Engaging participants with results is an effective way to address this and makes the benefits to participants and their communities far more tangible. It also helps to ensure that research participants welcome and value research as a means to solving issues pertinent to their lives thereby encouraging the participation of individuals and communities.

Recognizing, respecting and valuing the rights, agency and ability of research participants to engage in the dissemination process should not be an optional activity. It is essential for bringing together the key research processes of investigation, identification and implementation of ideas. It bridges the gap between the discovery and delivery of new knowledge. A rights-informed approach to engaging participants with study findings is not only socially responsible and an ethical imperative, but it also has implications for the influence and impact of research where it most counts—study participants and the population they represent.

AUTHORS CONTRIBUTIONS

Mathew Sunil George carried out the data collection and drafted the manuscript with contributions from Rakhal Gaitonde, Rachel Davey, Itismita Mohanty and Penney Upton. Mathew Sunil George, Rakhal Gaitonde and Penney Upton were involved in the analysis of the qualitative data. All authors were involved in the design of the larger study from which the data for this manuscript emerged. The authors read and approved the final manuscript.

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CONFLICT OF INTEREST

The authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

The data sets generated and/or analysed during the current study are not publicly available as they are qualitative in nature but are available from the corresponding author upon reasonable request.

ETHICS STATEMENT

The Human Research Ethics Committees of the University of Canberra (20180074) and the Indian Institute of Public Health Delhi (IIPHD_IEC_03_2018 provided ethical approval for this study. Regulatory permissions were obtained from the Kerala Department of Health (GO(Rt)No2677/2018/H&FWD) and the local administration in Attapadi.

Permissions were also obtained from the local health officials before fieldwork began. All participants gave informed consent before data collection.

DETAILS

Subject:	Health care access; Fieldwork; Ethnography; Public health; Community; Funding; Legitimacy; Researchers; Information sharing; Health care; Community involvement; Change agents; Partnerships; Open access publishing; Ethics; Dissemination; Research design; Medical research; Power; Community participation; Ownership; Data collection; Research projects
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A framework for implementing Patient and Public Involvement in mental health research: The PATHWAY research programme benchmarked against NIHR standards

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ProQuest document link

ABSTRACT (ENGLISH)

Background

Patient and Public Involvement (PPI) in research has become a key component recommended by research commissioners, grant award bodies and specified in government policies. Despite the increased call for PPI, few studies have demonstrated how to implement PPI within large-scale research studies.

Objective

The aim of the current study was to provide a case example of the implementation of a patient advisory group in a large-scale mental health research programme (PATHWAY) and to benchmark this against UK standards.

Method

A PPI group was incorporated throughout the PATHWAY research programme, from grant development to dissemination. The group attended regular meetings and supported participant recruitment, evaluated patient-facing documents, supported the piloting of the research intervention and co-developed the dissemination and impact strategy. The implementation of PPI throughout the project was benchmarked against the UK standards for PPI.

Results

The inclusion of PPI in the PATHWAY project provided tangible changes to the research project (i.e., improving study documents, co-developing dissemination materials) but also proved to be a beneficial experience to PPI members through the development of new skills and the opportunity to provide a patient voice in research. We show how PPI was involved across seven study phases and provide examples of implementation of the six UK standards. The study did not include PPI in data analysis but met all the UK standards for PPI. Challenges regarding practical components (i.e., meeting frequency, language use), increasing diversity and PPI members' knowledge of research were highlighted as areas for further improvement.

Conclusions

We provide a case example of how PPI can be implemented throughout a research lifecycle and we note the barriers faced and make suggestions for PPI in future implementation and research.

Patient and Public Contribution

PPI members were involved throughout the lifecycle of the research programme. The PPI lead was a co-author on the manuscript and contributed to report writing.

FULL TEXT

INTRODUCTION

Patient and Public Involvement (PPI) in research has become prominent in health and social care services research.[1] PPI activity as defined by the National Institute for Health Research NIHR[2] is an ‘active partnership between the public and researchers in the research process’ and is demarcated as doing research ‘with’ and ‘by’ the public rather than ‘to’, ‘about’ and ‘for’ the public.

Involving patients and the public in research is strongly recommended by current policy and practice in the United Kingdom,[3] and funding bodies such as the NIHR and The Research Excellence Framework Higher Education Funding Council emphasize the inclusion of PPI in research programmes.[4–6] In their 2015 report ‘Going the Extra Mile’, the NIHR outlined that by ‘2025 we expect all people using health and social care, and increasing numbers of the public, to be aware of and choosing to contribute to research…’,[6,p.10] supporting the public's call for further involvement.

The inclusion of PPI in research has demonstrable benefits such as ensuring that the research conducted is relevant, is culturally and logistically appropriate, enhances recruitment and retention and improves the quality of outputs.[7–10] Not only does PPI improve research but it also benefits PPI members. Brett et al.[7] conducted a systematic review of the impact of PPI on service users, research and communities across 65 studies. Service users reported personal benefits including feeling empowered, valued and making meaningful contributions to the research community. In addition, they reported improved knowledge of research and felt that they were able to further develop skills such as interviewing and public speaking.

While it is evident that PPI has a number of potential benefits to researchers and PPI members, concerns have been raised that it can be tokenistic, managerialist, time consuming and requires significant resourcing.[11,12] In addition, evidence of the value and impact of PPI remains limited, with inconsistency in what effective PPI in research is, what this might look like[11] and how to assess impact.[11] Several systematic reviews have found a poor understanding of how members of the public can be involved and the true impact that PPI can have on research.[7,13–16] Such limitations were attributed to the lack of guidance and consistency on how to report and evaluate the use of PPI, which has resulted in the development of tools and guidelines to increase transparency in reporting.[17,18] More recently, the NIHR[19] published six UK standards to ensure PPI is effectively implemented, which are (1) inclusive opportunities; (2) working together; (3) support and learning; (4) communications; (5) impact and (6) governance. The standards provide a benchmark for PPI along with indicators that improvement can be monitored against.

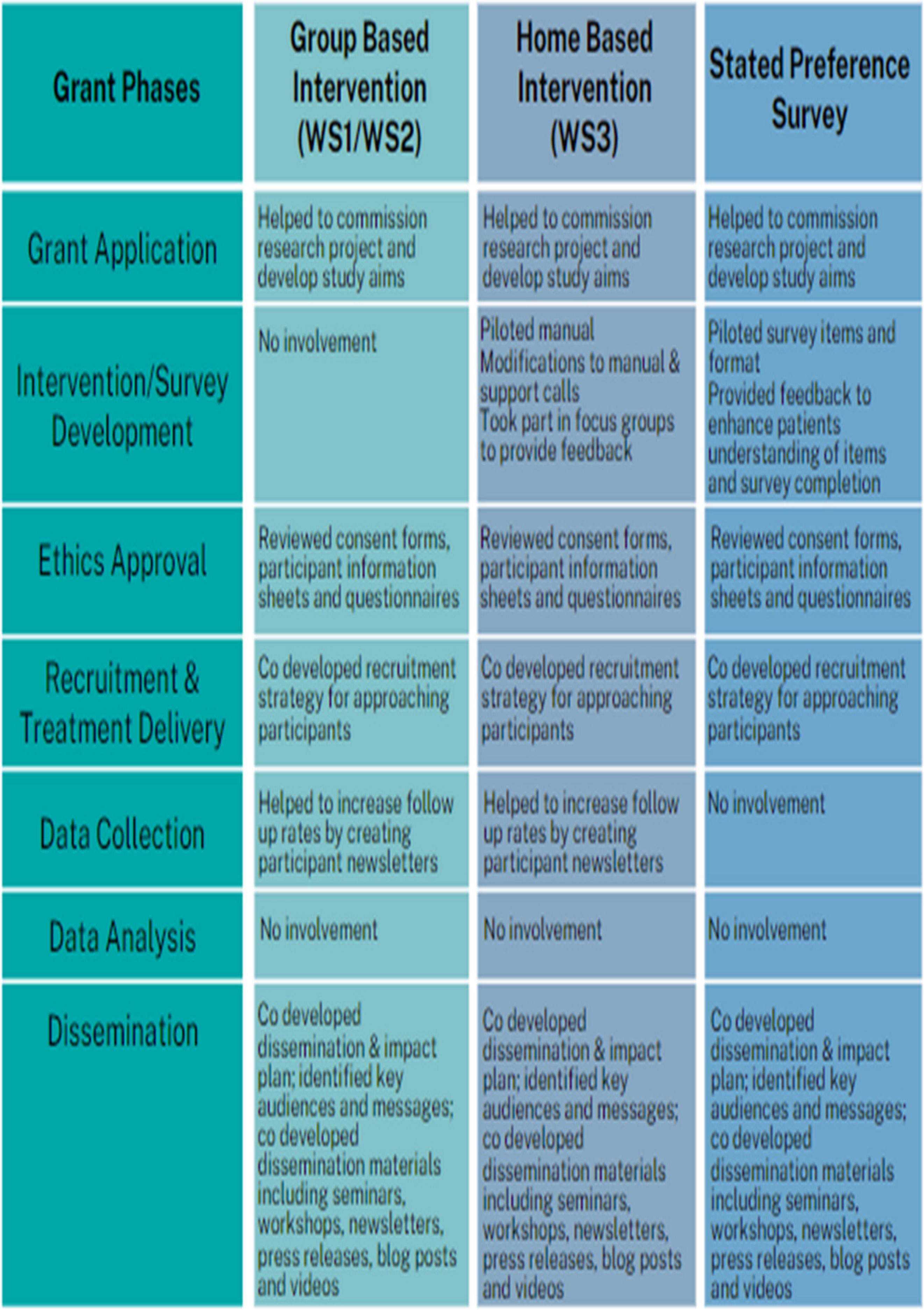
While increased guidance on the reporting of PPI in research has been mandated,[5,18] few studies have provided a case example of how PPI can be effectively implemented throughout the lifecycle of a project. As such, the aims of the present paper are to (1) report on how PPI was utilized in a large-scale multidisciplinary research programme (PATHWAY) and provide a framework for how PPI can be used and (2) benchmark the PATHWAY trial against the UK standards for PPI.

OVERVIEW OF THE PATHWAY PROGRAMME

The PATHWAY Programme is a 7-year project funded under the NIHR Programme Grants for Applied Research (RP-PG-1211-2001). The programme included two feasibility trials and two randomized controlled trials with integrated qualitative and health economics analyses (NCT02420431; NCT03129282; NCT 03999359). The overall aim of the programme was to reduce symptoms of anxiety and/or depression in cardiac rehabilitation (CR) patients by integrating metacognitive therapy (MCT[20]) in CR. PATHWAY evaluated whether MCT delivered in addition to usual CR is more effective than attending usual CR sessions alone in reducing symptoms of anxiety and/or depression. MCT was evaluated in two formats across two different trials, group-based delivery at CR services (Group-MCT) and home-based self-help with additional telephone support (Home-MCT) (for details of the programme, see Wells et al.[21–24]).

FRAMEWORK FOR PPI: THE PATHWAY TRIAL

PPI was integrated throughout all stages of the research programme. Figure 1 presents an overview of the framework for the implementation of PPI. The level of involvement and contribution made by PPI advisors changed throughout the research study and to members' personal preferences. PPI was implemented with the support of a PPI lead who worked in collaboration with both the research team and PPI group. The PPI lead developed and oversaw all aspects of involvement and acted as a facilitator and liaison between both groups, assisted in removing barriers to involvement, acted on concerns raised by the PPI group and evaluated the experiences of the group.



Grant Phases	Group Based Intervention (WS1/WS2)	Home Based Intervention (WS3)	Stated Preference Survey
Grant Application	Helped to commission research project and develop study aims	Helped to commission research project and develop study aims	Helped to commission research project and develop study aims
Intervention/Survey Development	No involvement	Piloted manual Modifications to manual & support calls Took part in focus groups to provide feedback	Piloted survey items and format Provided feedback to enhance patients understanding of items and survey completion
Ethics Approval	Reviewed consent forms, participant information sheets and questionnaires	Reviewed consent forms, participant information sheets and questionnaires	Reviewed consent forms, participant information sheets and questionnaires
Recruitment & Treatment Delivery	Co developed recruitment strategy for approaching participants	Co developed recruitment strategy for approaching participants	Co developed recruitment strategy for approaching participants
Data Collection	Helped to increase follow up rates by creating participant newsletters	Helped to increase follow up rates by creating participant newsletters	No involvement
Data Analysis	No involvement	No involvement	No involvement
Dissemination	Co developed dissemination & impact plan; identified key audiences and messages; co developed dissemination materials including seminars, workshops, newsletters, press releases, blog posts and videos	Co developed dissemination & impact plan; identified key audiences and messages; co developed dissemination materials including seminars, workshops, newsletters, press releases, blog posts and videos	Co developed dissemination & impact plan; identified key audiences and messages; co developed dissemination materials including seminars, workshops, newsletters, press releases, blog posts and videos

Enlarge this image.

Members of the public were recruited via patient networks, with the following eligibility criteria: experience of one or more of the following was required: (1) heart disease; (2) psychological distress and/or (3) being a carer of someone with one or more of these conditions. A group of 10 advisors was formed following an expression of interest from 16 people. Of the initial 10 advisors, 3 members left. The most common reason for withdrawing was due to illhealth. Demographic characteristics of the 10 PPI advisors and the PPI Lead are included in Table 1. Written consent was sought and obtained from all PPI contributors for their quotes, questionnaire responses and demographic information to be used in this paper.

Table 1 Descriptive characteristics of the PPI advisory group

PPI Member ID	Gender	Age at beginning of the study?	Ethnicity	Marital status	Qualification	Are you a CR service user?	Do you have experience in research?	Do you have experience as a PPI member in other research studies?
01	M	Not reported	Not reported	Married	Not reported	Y	N	N
02	F	70	Black African	Divorced	Degree	N (mental health nurse)	Y	Y
03	F	53	Black	Divorced	Degree	Y	Y	Y
04	M	67	White background	Married	Vocational qualification	Y	Y	N
05	M	69	White British	Married	Vocational qualification	Y	N	N
06	M	61	White British	Married	Diploma	Y	N	N
07	F	65	White British	Married	Degree	Y	Y	N
08	F	Not reported	Not reported	Not reported	Not reported	Y (Carer)	Y	Y
09	M	63	White British	Married	Degree	Y	N	N

10[a]	M	Not reported	Not reported	Not reported	Not reported	Not reported	Not reported	Not reported

Abbreviations: F, female; M, male; N, no; PPI, Patient and Public Involvement; Y, yes.

[a] PPI member left the group before data collection.

Stage 1: Grant development and research governance

Consultations with service users at two cardiac service user group meetings (Service User Research Endeavour Group at Liverpool Heart and Chest Hospital NHS Foundation Trust and the Ticker Club at University Hospital of South Manchester NHS Foundation Trust) were conducted. Two meetings were held, which focused on (1) the research background and preliminary ideas and (2) discussions related to the research proposal and PPI plans. Further consultations, funded by a Research Design Service (RDS) NW bursary, were conducted with Ticker Club members regarding the acceptability and feasibility of the research. An advisor from the RDS NW and a service user were included as co-applicants on the grant application and reviewed the lay summary and PPI sections of the grant application.

The PPI Lead and the PPI Chair were members of the PATHWAY executive committee, while a PPI member independent of the research team and PPI advisory group was a member of the trial steering committee. As part of the executive and trial steering committee, PPI representatives provided input on study design, conduct, consultation about emerging findings and dissemination and impact planning. Attendance at executive committees allowed the PPI lead and chair to contextualize research findings and review them with the advisory group.

Stage 2: Research deliveryEthical approval

All patient-facing material was reviewed and approved by the PPI group before its submission to ethics. This included service users ensuring that the information provided in consent forms and participant information sheets was sufficient and written in a lay format. The advisory group also reviewed the order of study documentation in the information packs to ensure that it was engaging and inviting for prospective patients.

Intervention development

Service users were involved in the development of the home-based MCT manual, focusing specifically on ensuring that the format and readability of the manual were appropriate for patients.

Home-based MCT

PPI members contributed to the layout of the home-based manual. Four PPI members piloted the home-based manual. In doing so, members completed each module in the manual along with the telephone support calls, which for the purposes of the pilot were delivered by the research team. This allowed researchers to evaluate the timing and content of the telephone support calls. Following the pilot, PPI members provided feedback on the manual and support calls via feedback meetings. PPI members commented that (1) more emphasis should be placed on the ease of use and flexibility in using the modules to enhance compliance; (2) a question section should be added to give the patient the opportunity to clarify/discuss any issues with the therapist in the subsequent telephone call; (3) an estimation of the amount of time to complete each module should be included; (4) the language at times was too technical, which resulted in minor modifications to the text and (5) overall the modules were easy to follow. All of the suggested changes were implemented in the manual ahead of the feasibility trial.

While piloting the home-based manual benefitted the research team, the advisory group also noted that piloting the self-help manual supported them in developing further knowledge and understanding of MCT, anxiety and depression. Some advisors felt that piloting the manual changed their beliefs about their own thinking and their personal ways of coping with and understanding anxiety and/or depression symptoms. One PPI member reported that ‘I think it's great that psychological, the emotional side is now being sort of trying to be managed because it's not been there. …I didn’t know until we started getting the paperwork what it was [anxiety and depression] …and now I know why I didn’t want to go out of the house for six months. I never ever suffered with depression before, but I now know why' (PPI member 3).

Recruitment and treatment delivery

The PPI group also aided in co-developing a recruitment strategy for approaching participants across all workstreams. This ensured that the language used when approaching and inviting potential participants to take part was appropriate and engaging. In addition, for the health economics workstream, the group provided feedback on potential sources of recruitment.

Data collection and analysis

To increase the rate of return of follow-up questionnaires, service users were asked for suggestions on how to enhance follow-up participation and decrease the drop-out rate. Suggestions included the creation of a participant newsletter, which highlighted the importance of questionnaire completion and its benefit to patients and NHS services while providing trial updates and maintaining patient engagement. The newsletter was co-produced between PPI members and the research team and provided a significant benefit in improving the rate of return for follow-up questionnaires.

Service users were not involved in the data analysis, although qualitative and quantitative data were discussed with the PPI group, which aided in ensuring that key messages were presented in a format that was accessible to the public; for example, service users commented on the importance of including the treatment recovery rate, as they felt this was an important outcome for patients.

Stated preference research

A stated preference survey using a discrete choice experiment was designed to estimate preferences for psychological care within the CR pathway.[25] PPI members advised on the design of the discrete choice experiment, including feedback on potential key attributes and levels to be used in choice set questions, refining these to create a feasible design and feedback on participant demographics that may affect preferences for care. The PPI group also completed and commented on a draft survey to check that materials were easy to follow and to estimate the time that would be required by participants. Finally, the group commented on the likely impact of the COVID-19 pandemic on stated preference responses (including the response rate and elicited preferences). The PPI work was published as a case study focusing on the use of PPI in stated preference research in health.[26] The PPI lead (L. B.) was an author of the case study and contributed to report writing.

Qualitative process evaluation

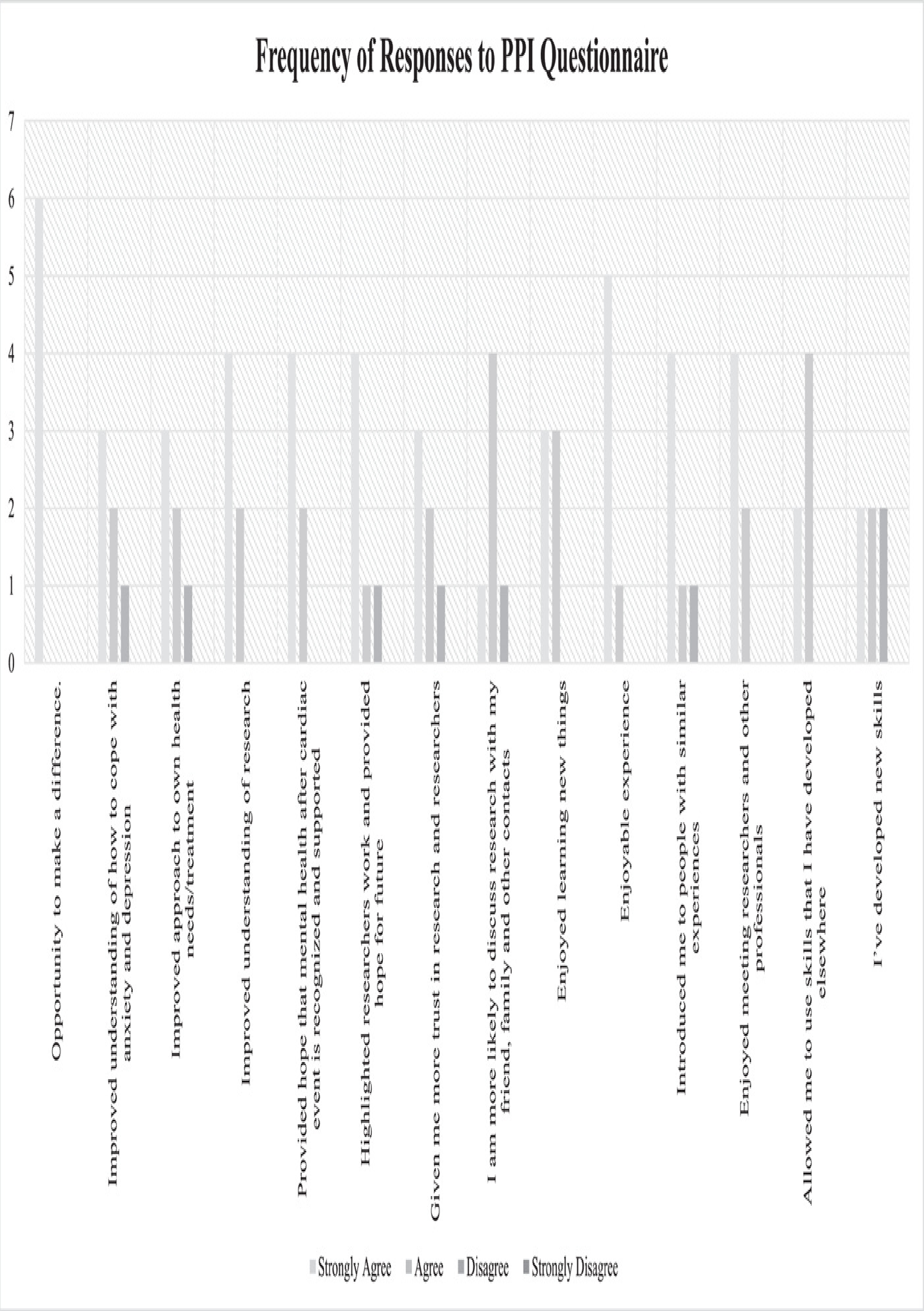
A qualitative process evaluation was embedded into the PATHWAY trial. The PPI group was consulted about the design of the qualitative process evaluation and the materials used to collect qualitative data. Initially, the group provided feedback on the longitudinal nature of the qualitative component of the research and confirmed that they felt this would not be burdensome for participants. The group was also consulted about the qualitative interview topic guides, with the aim of ensuring that all areas of relevance were covered during interviews, the correct language was used and topics that could be understood to be potentially sensitive were handled in an appropriate way.

Stage 3: Dissemination

Group activities were held with the PPI group to aid the development of the dissemination plan with a focus on (1) identifying key messages, target audiences, format and publication/venues and (2) styles of dissemination. Discussions were conducted to determine results of relevance to the public, explanations of future steps and language use in dissemination work. The group aided in the development of resources with members being involved in creating videos, blogs and presentations of their experiences taking part in PPI, which helped to provide a patient voice. They also played a key role in the development and delivery of a live event disseminating the study results for patients, clinicians and members of the public: helping to inform the format of the event and taking part in the event by discussing why they chose to be involved in the project and the role they played in the research.

PPI group feedback

Evaluations of the PPI approach were conducted throughout the project through feedback meetings, telephone calls and a summative questionnaire completion (see Supporting Information: Appendix 1) by the PPI lead. The summative questionnaire was developed by the study team to assess the impact that taking part in a PPI group has had on them. These evaluations encouraged members to raise concerns, highlight the benefits of participating in research and determine how the group met NIHR standards. The summative questionnaire was completed in the final year of the study. Results were analysed and anonymized by the PPI lead before being shared with the PPI and research teams. Six members completed the questionnaire. Overall, PPI members felt that taking part provided them with the opportunity to make a difference, improved their understanding of research, made them more likely to discuss research with friends and family and improved their understanding of coping with anxiety and depression and how they personally manage it (Figure 2). Only one PPI member felt that taking part in the PPI group did not improve their understanding of anxiety and depression, their own mental health coping or their perspectives on researchers. This was due to their occupation and previous research experience. All PPI members noted that taking part did not have any negative impacts on them and four out of six would take part in further PPI. The two members who said they would not take part in further PPI noted that this was due to a deterioration in their physical health.



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BENCHMARKING PPI

To assess the implementation of PPI in PATHWAY, we compared our use against the NIHR UK standards (see

Table 2). In addition to benchmarking against UK standards, we note the challenges faced and ways in which the standards could have been further implemented. Feedback meetings were held with the PPI group to assess how they felt the team met the NIHR UK standards. See Table 3 for a summary of key recommendations for future PPI groups.

Table 2 Implementation of NIHR UK standards

Indicators	Research phase	PATHWAY implementation
1. Inclusive opportunities	GA	Recruited a range of public contributors (patients and carers) from varying sources (i.e., cardiovascular charities, public involvement groups)
	GA	Removed barriers by asking about any support needs from the first meeting and ensured this was supported throughout
	All	Providing financial support for their time and travel
	All	PPI members were given the choice for how they would like to be involved based on members' interest and/or skills
2. Working together	All	Collaborative approach from small tasks (i.e., meeting dates) to co-productive of materials and dissemination activities
	All	Decisions made collaboratively with researchers
	GA	Created terms of reference with the PPI group, which was reviewed quarterly
	All	Ongoing review of roles and expectations
	All	Individual members' ideas and contributions recognized, felt valued and listened to
	All	Meeting minutes circulated following each meeting
	All	Open dialogue and communication
3. Support and learning	All	Clear point of contact for information and support, always accessible to answer questions
	All	Supported individual's needs (i.e., visual disabilities)
	All	Designated resources to support public involvement

	All	Informing the group about PPI activities external to the project, especially as the project ends
	D	Learning by doing (co-presenting with researchers)
4. Communications	All	Flexibility in providing feedback to members following meetings (via email or phone)
	All	Feedback provided at meetings on how the research team has implemented feedback or justification for why feedback could not be implemented was provided at each meeting.
	D	Co-developed a dissemination and impact plan
5. Impact	D	Co-developed a dissemination and impact plan
6. Governance	All	Service users involved throughout all phases of the study
	All	Service user representation on the executive committee and steering committees
	All	Money and resources allocated for public involvement
	All	Terms of reference reviewed quarterly

Abbreviations: All, all phases; D, Dissemination; GA, grant application; PPI, Patient and Public Involvement.

Table 3 Key recommendations for future PPI groups

UK standard	Recommendations
1. Inclusive opportunities	Ensure PPI is adequately costed for, tools such as the INVOLVE calculator can aid with financing
	Ensure a range of PPI members are recruited using various ways of advertising (online, local groups) and allow flexibility in ways of returning an expression of interest forms
	Provide PPI members with flexibility and choice in how they would like to provide feedback (i.e., in meetings, phone, via email) and when (i.e., more/less contribution at different phases)
2. Working together	Create a term of reference document at the outset that includes aspects such as frequency of meetings, times, amount of notice required before a meeting is to be scheduled, to aid in setting expectations

	Ensure a collaborative approach and co-production of materials and dissemination and impact planning
3. Support and learning	Ensure groups have a clear point of contact (i.e., designated researcher or PPI lead)
	Ensure individual's needs are met (i.e., visual disabilities)
	Ask PPI members if they would be interested in learning about other PPI opportunities to support their engagement in the PPI group
	Allow members to co-present with researchers to aid in developing new skills
4. Communications	Ensure feedback is provided to outline why suggestions made by the PPI group have or have not been implemented
5. Impact	Involve PPI members in developing the research dissemination and impact plan (identifying who to disseminate to, selecting key messages for varying audiences)
6. Governance	Involve PPI throughout all governance levels

Abbreviation: PPI, Patient and Public Involvement.

Standard 1: Inclusive opportunities

Feedback from the PPI group highlighted that they felt inclusive opportunities were met early on with the adaptations made to support the needs of individual members and provide choice and flexibility around the type of work the group would be involved in. All group members were reimbursed for their time following UK standards along with travel expenses, to avoid financial barriers to involvement. Considerations raised for future involvement highlighted that members may have disabilities that make taking part in group settings difficult and therefore one-to-one options could have been provided for involvement. While the diversity of the PPI group is representative of those patients recruited to the PATHWAY trial, recruitment to the PPI group could have utilized a broader recruitment strategy. Participants were predominantly recruited via local patient networks or online advertisement; however, future research could aim to recruit directly from the patient services they aim to conduct research with.

Standard 2: Working together

Feedback meetings, telephone calls and questionnaires were used throughout the project, to evaluate the PPI approach and allow challenges to be raised and addressed to improve working together and was led by the PPI lead. This was met positively by the PPI group as it presented an opportunity for them to talk anonymously away from the research team. The phone calls in particular were considered to offer an opportunity to raise concerns that would not normally be raised in a larger group. Feedback from evaluations resulted in changes to our PPI approach. For example, the room layout was changed from a classroom style to a round table, more time was allocated to discussions rather than presentations and more clarity was given about which areas of the research could not be changed and why. In addition, at the start of meetings, we implemented a ‘you suggested, we did’, where we reviewed changes that we were able to implement based on discussions from the previous meeting. When changes were unable to be implemented due to research processes, these were clearly explained to the PPI group. Small actions such as using doodle polls and introducing all new members of the team were also felt to have played a role in creating an atmosphere of equality and clarity between the PPI and research teams. By the end of the study, our approach to working together was felt to strength, as one member said, ‘It has been a fulfilling experience. Too often patients' views are not considered I felt that we were listened to at every stage of the research and our suggestions were treated with respect' (PPI Member 7).

In the future, the group commented that further assistance was needed for individuals reintegrating into the group when they had been away for personal or health reasons. This could have been facilitated by a one-to-one meeting with the PPI lead and PPI member before the meeting to update on any changes to the group and progress made during missed meetings.

Standard 3: Support and learning

Advisory group members felt well supported; measures were put in place to accommodate those with additional needs or disabilities from the start of the project and were reviewed regularly. In addition, there was a range of learning opportunities offered to the group throughout the project, such as working with the team on presentations. PPI members also communicated that taking part in PPI had increased their trust in research and researchers and made them more likely to discuss research with friends and family. PPI members commented on the increased understanding they gained, 'I have gained a better understanding of all the difficult decisions that researchers have to make' (PPI Member 5). There were however a number of ways in which learning opportunities could have been further supported. While the advisory group ultimately felt that sufficient information had been given, at times confusion around some of the processes, protocols and unfamiliarity with the psychological intervention (MCT) had resulted in feelings of being under-utilized. For example, although the psychological intervention used within the trial (MCT) was explained to the group, this was not done until later in the research programme. One member commented that,

when we first started most, possibly all of us didn't understand the nature of the actual treatment. It was quite a long way down the line before we saw the materials … and understood more about how the treatment would work. I think some training earlier on in the process would have put us in a position where we were better informed to contribute. (PPI Member 6)

The group also felt that, while the research team provided sufficient information on the research process, this was a high volume of information, and providing them with a handbook or online training materials outlining the research processes would have been useful: allowing them to look up aspects of the research and refresh on processes in their own time. Providing such information at the start of the research process could have also aided in setting expectations with the PPI team around what they could and could not change. For example, PPI members wondered if it was possible to change the wording on validated questionnaires, which was outside the scope of what they were able to do. However, the research team did provide an explanation as to why this was not possible, the implications for the project and how questionnaires are developed.

Throughout the project, we had PPI representation at both the steering and executive team meetings and the principal investigator met with the PPI lead at these meetings and also at the PPI groups. To support PPI members and to ensure they felt comfortable attending these meetings, the PPI lead would accompany the PPI member to the meetings. PPI members were also provided with a copy of the previous meeting minutes, the meeting slides and the meeting agenda before the meeting. Following the meeting, the PPI lead and the research fellow were available for the PPI member to contact and ask questions if they felt uncomfortable to asking these during the team meeting.

Standard 4: Communication

The PPI group felt that the varying types of communication (group-based, one-to-one, emails, phone calls) and amount of communication outside of meetings were sufficient. However, the initial frequency of meetings, format and approach of meetings required improvements. While recaps of previous meetings were originally provided at the start of each meeting to reduce the impact of long gaps between meetings, it was viewed by members as wasting time and detracting from project business, as such email communication between meetings was increased to reduce the length of the recap at subsequent meetings. The group felt that they were able to speak without opinions being dismissed and that this also allowed them to say when they did not understand. They felt the research team was responsive and adjusted their language (i.e., used plain language) when speaking with PPI members. However, one member highlighted that expressing a different opinion to the rest of the group can be intimidating and can result in the feeling that they have to agree with the consensus. As such, members were specifically encouraged to express alternative views, and varying formats were offered to provide feedback (i.e., one-to-one, emails, phone calls), which is paramount in ensuring open and honest feedback from members.

Standard 5: Impact

The advisory group felt that their input was taken on board and the research team was very open to feedback from them. Being involved in the co-development of the home-based MCT intervention was felt by the group to be their most significant impact as they could see the adaptions following their suggestions. Advisors felt their input on this also had the most impact on patients as it ensured the accessibility of the home-based intervention.

Standard 6: Governance

The structure of governance included a PPI lead and PPI chairman as members of the executive committee and an independent PPI member as a member of the steering committee. This helped to ensure that PPI representation was considered at all project governance levels. Reviews of the PPI approach were conducted throughout the project and adjustments were made where needed. Future considerations were suggestions surrounding funding for PPI activities, as there were some additional opportunities that the group was unable to undertake due to funding not being allocated for them. Therefore, future planning for public involvement should include the allocation of funding for additional PPI opportunities such as attendance at conferences. Further suggestions include clarification of what payments can cover (i.e., taxis to attend meetings) and ensuring clear procedures around paperwork and timing of payments to ensure clear expectations on how long a payment may take.

DISCUSSION

PPI can play an important role by providing a patient's perspective that can be utilized throughout all phases of a research study. The current paper illustrated how PPI can be implemented as well as how national PPI standards might be met, and implementation improved upon further.

Implementation

Throughout the research programme, PPI resulted in tangible changes in patient-facing documents, the layout of the home-based intervention, the development of newsletters and participation in dissemination events. In doing so, PPI members aided in ensuring the appropriateness of the language being used, the format of interventions and aided in ensuring research findings were presented in an accessible and user-friendly manner. The implementation of PPI in our PATHWAY programme was consistent with previous studies, which have advocated for PPI throughout all phases of research.[12,19,27] Beyond playing an integral role in the research programme, PPI also provided benefits to PPI members, an often-overlooked benefit to implementing PPI in research. In line with previous research,[12,27,28] PPI members noted that the study allowed them to develop new transferrable skills, allowed them to have a voice, connect with others who had similar experiences and connect the study results to the impact and benefit for patients.

Benchmarking and future learning

To assess if PPI met national guidelines for quality and consistency, we benchmarked our strategy against the UK standards for public involvement.[19] While some authors have argued that the UK standards could lead to inflexible PPI practices,[29] the standards allowed us to reflect on the way PPI was utilized and highlighted ways in which it might have been improved.[30] Conducted a systematic review of PPI frameworks, which found that there were 65 different PPI frameworks that could be grouped into five categories: power-focused; priority-setting; study-focused; report-focused and partnership-focused. While this highlights that there are several frameworks available with specific objectives to help guide PPI. But specificity may compromise transferability, and[30] note little use beyond the groups that originally developed them. Guidelines that are broader and provide principles to work towards such as INVOLVE may provide more flexibility and stimulate use across objectives.

One of the main sources of difficulty that we observed between researchers and advisory group members was the advisory groups' lack of knowledge of the research processes, which is commonly noted as a barrier.[12,27,31] In the future, we recommend that researchers should hold a training event at the study's outset, which focuses on research processes/timelines, confidentiality and what can happen at the end of a research project. This should assist in setting realistic expectations for what the research can achieve within a limited time frame and what is within the scope of the research project. For example, in research projects evaluating an intervention that is found to be effective, it would be useful to provide an outline of the duration and the steps that are needed to be undertaken before new treatments become accessible to the public. We found that the PPI group held unrealistic expectations concerning the implementation of research findings. Additionally, studies may also look to provide a research handbook for PPI groups that members can use to refer back to throughout the project and in their own time.[32]

In addition, there were practical challenges that were encountered in implementing PPI including frequency of meetings, format and delivery of meetings and language use. At times, there were long gaps between meetings, which created inconsistency and feelings of underutilization. In the future, setting clear expectations at the study's outset can aid in establishing accountability between researchers and the PPI group. According to Ocloo et al.'s[28] systematic review of the barriers and enablers of PPI across health and social care research, clear definitions of roles, goals of participation and specification of the time commitment required were important factors in improving PPI.

While a common challenge of PPI surrounds issues of power dynamics,[12,27–31] the PATHWAY programme did not (to our knowledge) encounter observable difficulties in this area; however, we are mindful that such dynamics might exert unseen influences. Our interpretation of the lack of observable problems in this domain is that PPI was included at programme governance levels (steering committee, executive team committees) and a designated PPI lead acted as a bridge between the PPI group and researchers. We also incorporated PPI throughout the project and solicited ongoing reflections on PPI utilization that was facilitated by the PPI lead. Such strategies may have helped to reduce the ‘us vs. them’ culture often noted as a barrier in implementing PPI. However, further assessments of the mechanisms that may mitigate such dynamics are required.

Limitations

While the PATHWAY programme incorporated PPI throughout the lifecycle of the research project, further assessment of the impact and effectiveness of PPI in PATHWAY is required. Assessment of PPI was conducted using a questionnaire developed by the study team, but this measure has not been validated and items focused largely on assessing the impact of taking part in a PPI group. It would have been useful to include assessments of the barriers and enablers to PPI engagement during sessions and to examine the personal frameworks used by PPI members in assessing and advising on the research process. Similarly, an evaluation was not conducted on the impact that PPI had on the researchers themselves.[12] evaluated researchers' experiences and perceptions of PPI in 36 health researchers and noted the complexity of researchers' experience in involving patients and the public in research. While researchers noted a range of practical challenges (i.e., lack of time, insufficient funding), they also noted that PPI was accompanied by a range of emotions from cynicism to enthusiasm, noting that the impact that PPI has on research is both beneficial and burdensome.

CONCLUSION

The current paper provided an account of how PPI was implemented throughout the lifecycle of PATHWAY. PPI was a component of our clinical trials, stated preference survey and qualitative research. We benchmarked PPI against UK standards, and although we appear to be compliant, we found specific areas for improvement. We provide an overview and examples of our practice in the PATHWAY programme and make specific suggestions that could be useful in guiding future studies.

AUTHOR CONTRIBUTIONS

Lora Capobianco and Adrian Wells contributed to conceptualization, investigation, writing – original and subsequent drafts and editing and project administration. Cintia Faija, Bethany Cooper, Lindsey Brown, Rebecca McPhillips and Gemma Shields contributed to the writing –review and editing. Adrian Wells contributed to supervision and funding acquisition.

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CONFLICT OF INTEREST

A. W. is the director of the MCT Institute and developer of MCT. A. W. has written books on cognitive behaviour therapy and MCT. The remaining authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

Data may be made available on request to the corresponding author.

DETAILS

Subject:	Mental health; Standards; Divorce; Life cycle analysis; Advisors; Report writing; Grants; Advisory groups; Citizen participation; Patients; Data analysis; Public involvement; Public policy; Recruitment; Job requirements; Documents; Respiratory distress syndrome; Dissemination; Medical research; Implementation; Meetings; Research projects
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Co-design development of a decision guide on eating and drinking for people with severe dementia during acute hospital admissions

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ABSTRACT (ENGLISH)

Introduction

Using co-design processes, we aimed to develop an evidence-based decision guide for family carers and hospital professionals to support decision-making about eating and drinking for hospital patients with severe dementia.

Methods

Following a systematic review, we interviewed people with mild dementia, family carers and hospital professionals in England. We then held co-design workshops with family carers and hospital professionals. In parallel with the workshops, we used a matrix to synthesize data from all studies and to develop a decision guide prototype. The prototype was iteratively refined through further co-design workshops and discussions among researchers and Patient and Public Involvement (PPI) representatives. We conducted user testing for final feedback and to finalize the decision guide.

Results

Most participants acknowledged the limited benefits of tube feeding and would not use or want it for someone with severe dementia. However, they found decision-making processes and communication about nutrition and hydration were emotionally demanding and poorly supported in acute hospitals. The co-design groups developed the aims of the decision guide to support conversations and shared decision-making processes in acute hospitals, and help people reach evidence-based decisions. It was designed to clarify decision-making stages, provide information and elicit the values/preferences of everyone involved. It encouraged person-centred care, best-interests decision-making and multidisciplinary team working. From user testing, family carers and hospital professionals thought the decision guide could help initiate conversations and inform decisions. The final decision guide was disseminated and is being used in clinical practice in England.

Conclusion

We used rigorous and transparent processes to co-design the decision guide with everyone involved. The decision guide may facilitate conversations about nutrition and hydration and help people reach shared decisions that meet the needs and preferences of people with severe dementia. Future evaluation is required to test its real-world impacts.

Patient or Public Contribution

People with mild dementia, family carers and hospital professionals contributed to the design of the decision guide through the interviews and co-design workshops. PPI members helped design study procedures and materials and prepare this manuscript.

FULL TEXT

INTRODUCTION

In the severe stages, people living with dementia often develop eating and drinking problems, for example, swallowing difficulties, holding food in the mouth, changes in appetite and other changes in eating habits.[1,2] These eating and drinking problems may worsen during acute hospital admissions where people have to adapt to strict and busy hospital routines,[3,4] and their personal care, including help with eating and drinking, can be suboptimal.[5] Although there is no evidence regarding the benefits of tube feeding for people with severe dementia[6] and it is rare in the United Kingdom, they may still receive futile interventions, resulting from the influences of cultural beliefs, legal restrictions and family requests.[7–9]

With the declining capacity of the person with severe dementia to make decisions, decisions in hospitals about their nutrition and hydration are usually made by healthcare professionals with input from family carers, under the Mental Capacity Act (England and Wales).[10–12] People with mild dementia may also have difficulty discussing their wishes and decisions[13,14] and want to leave future decisions about eating and drinking to family carers and professionals.[15] However, it is still unclear how such decision-making can be best supported, and both family carers and professionals often find making these decisions challenging.[8,16,17]

Decision aids or guides (henceforth decision guide(s)) are tools developed to help people participate in decision-making about healthcare options, clarify and communicate their personal values and promote deliberation between everyone involved about options.[18] Studies report people who had used a decision guide had more opportunities to discuss care decisions with their clinician, had more accurate risk perceptions and participated more in decision-making.[19]

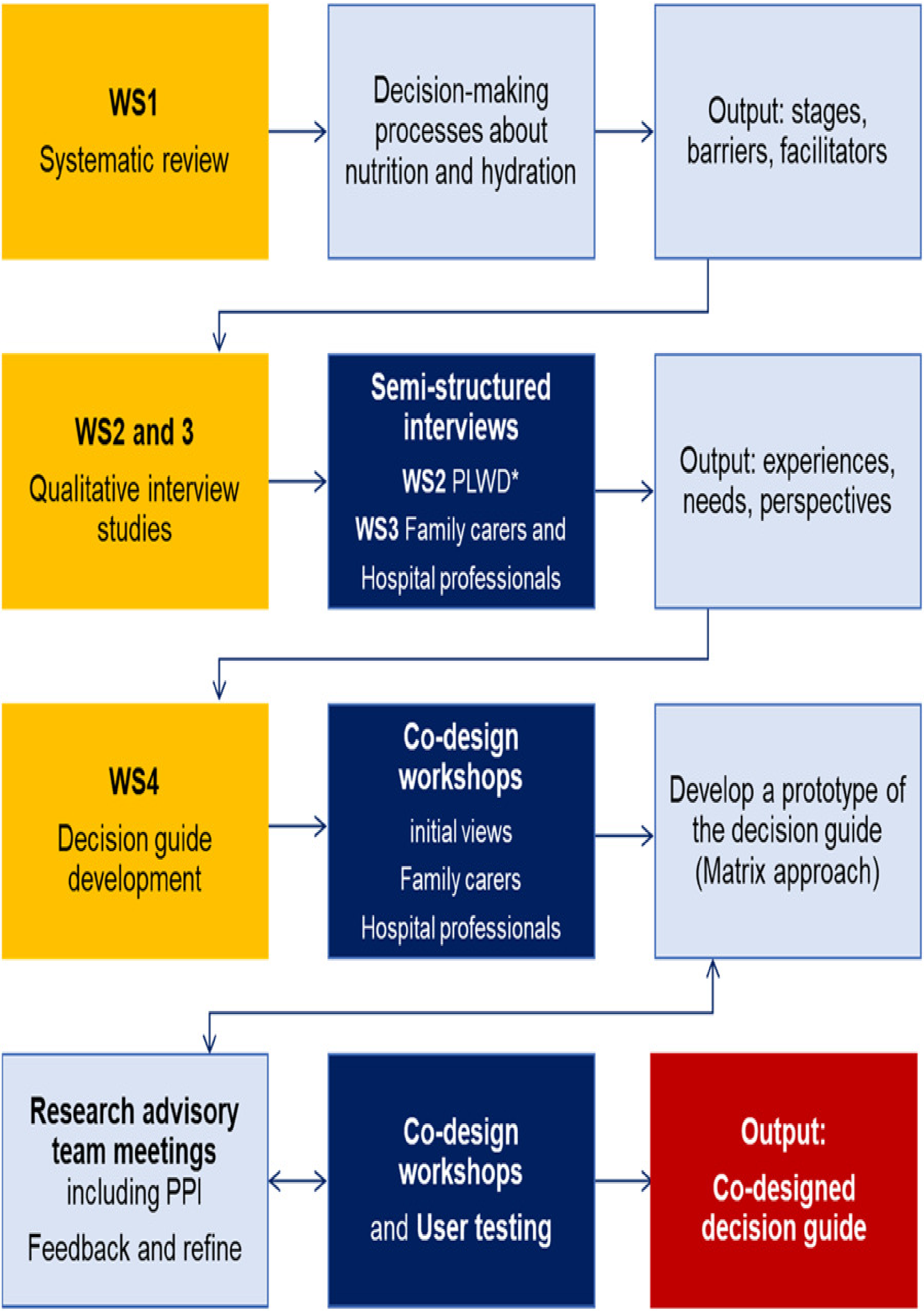
From our previous studies, decision guides regarding care for people with severe dementia, including at the end-of-life, seemed to be helpful[20–25]; however, the earlier guides have been developed for either practitioners or family carers to make decisions for people with severe dementia and not specific to eating and drinking issues in acute hospitals, which are particularly challenging. This co-design study, therefore, aimed to develop a decision guide covering nutrition and hydration for people with severe dementia in acute hospitals. The decision guide was expected to be used together by family carers and hospital professionals in making difficult decisions. The specific research objectives were to:

- 1. Using a matrix approach, synthesize data from a systematic review and co-design workshops and interviews with people with mild dementia, family carers and hospital professionals;
- 2. Understand problems and possible solutions around eating and drinking decisions for people with severe dementia in acute hospitals;
- 3. Identify the focus and aims of the decision guide;
- 4. Map the problems and possible solutions identified from the synthesis into the decision guide;
- 5. Identify suitable format, content and dissemination of the decision guide.

METHODS

In developing the decision guide, we used a co-design approach, in which researchers work in partnership with end users to gain deeper insights into the world of research participants and develop research outputs that are based on their experiences, perspectives and needs.[26,27] We based our co-design processes on the 2005 Double Diamond design model proposed by the British Design Council which involves ‘discovering’ problems and needs of the users, ‘defining’ the challenges and areas to focus upon, ‘developing’ potential solutions and ‘delivering’ solutions that work.[28] The co-design research involves a range of methods, including interviews, surveys, focus groups or workshops,[29,30] and can be used to produce a variety of outputs, such as theoretical frameworks, commissioning statements, documents and interventions.[29] It has been used to co-design many healthcare interventions, including decision tools in dementia and palliative care.[24,31]

As shown in Figure 1, the co-design processes in this study comprised four sequential workstreams; Workstream 1 (WS1): a systematic review of decision-making processes about nutrition and hydration for people with dementia[8]; WS2: semistructured interviews with people with mild dementia about their perspectives regarding future possible eating and drinking problems[15]; WS3: semistructured interviews with family carers and hospital professionals about care for nutrition and hydration for people with severe dementia during acute hospital admissions[32] and WS4: a co-design workshop to develop a decision guide about nutrition and hydration for a patient with severe dementia in an acute hospital. WS1–3 have been reported in separate publications, and this article focuses on the last WS4.



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Population and participants

In WS4, we screened potential participants against the eligibility criteria in Table 1.

Table 1 Eligibility criteria for participants in this study

Family carers
Inclusion criteria
Family member or friend who is/was a key decision-maker for a person with severe dementia (current or bereaved/former)
Participants must be able to provide informed consent
Participants must be able to read and speak English
Participants must be over the age of 18 years
Exclusion criteria
Family carers bereaved in the past 3 months
Hospital professionals
Inclusion criteria
Hospital professionals in a caring role, either health or social care, for people with severe dementia
Experienced in providing dementia care and contributing to decision-making related to nutrition and hydration in acute hospital setting
Participants must be able to provide informed consent
Participants must be able to read and speak English

Due to limited time, financial support and accessibility caused by COVID-19, we did not include people with dementia as participants in the online co-design workshops (WS4). We appreciated that online workshops with people with dementia would require far more complex and sensitive processes.[33] For example, ongoing mental capacity assessments and constant training and technical support, which need adjustments to the individual's changing capabilities and needs as dementia progresses. However, we included the experiences and needs of people with mild dementia identified from WS2 interviews throughout the co-design processes and development of our decision guide.

Participant recruitment and consent process

Due to COVID-19, we recruited family carers from Join Dementia Research (JDR) and online social media. JDR is an online self-registration service that enables volunteers with memory problems or dementia, family carers of those with memory problems or dementia and healthy volunteers to register their interest in taking part in the research.

We recruited hospital professionals (henceforth professionals for brevity) using online social media. Potential professional participants were also invited via contacts of the research team and snowballing methods.[34] As a result, few professional participants had a prior work-related relationship with the research team, but we monitored its influence on research processes and reassured the participants about voluntariness and confidentiality. To obtain rich information, we purposively sampled professionals from the range of roles that would be part of a multidisciplinary team providing care for people with severe dementia.

We also invited family carers and professionals who had participated in the WS3 interviews who had expressed their interest in subsequent studies. Participant information sheets and consent forms were sent to potential participants who were given sufficient time to consider the study. We obtained written consent from participants before the workshops.

Research advisory team meetings

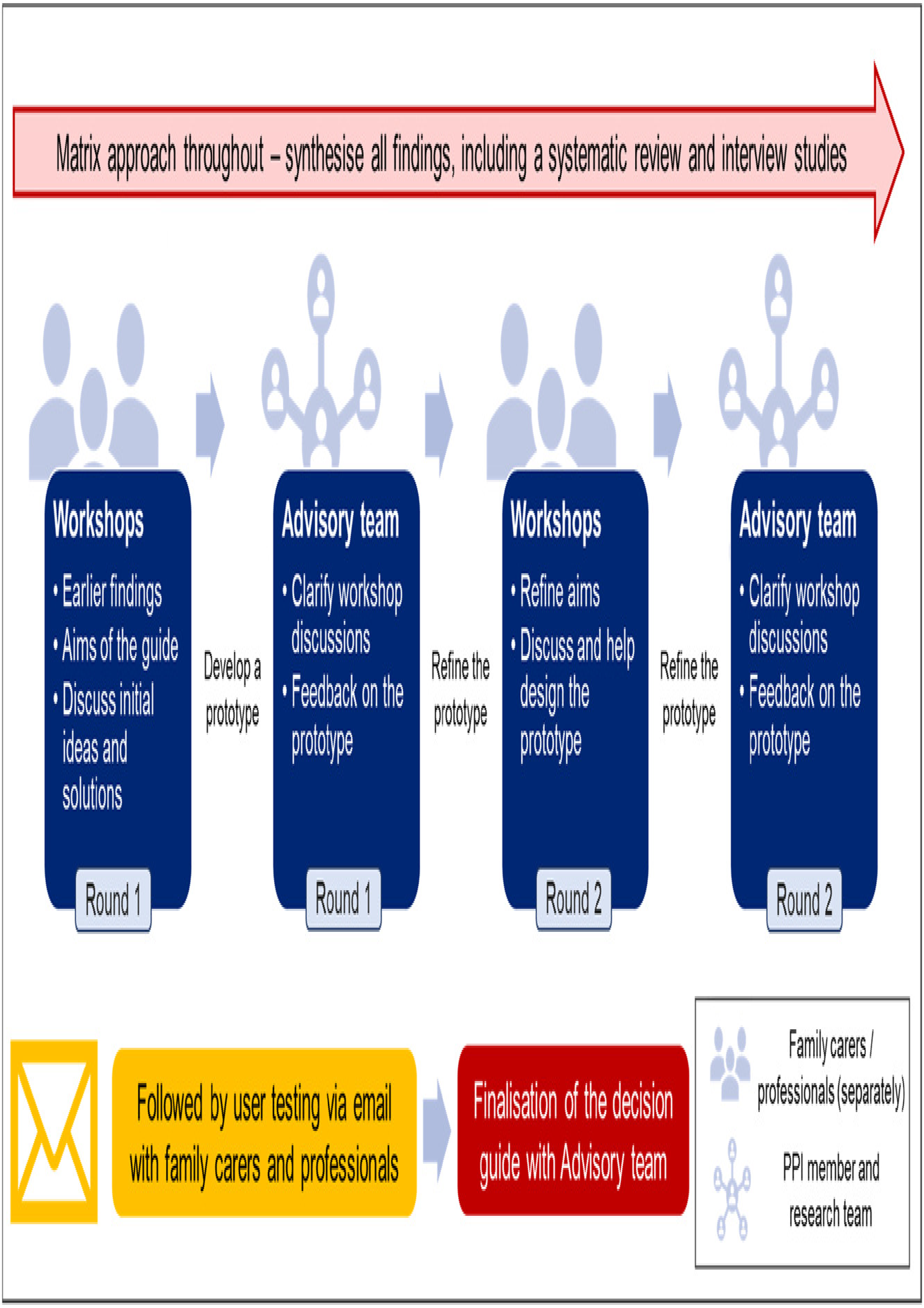
We held three research advisory team meetings to discuss the interpretation of discussions from the co-design workshops and feedback on the prototype. They consisted of research team members and a Patient and Public Involvement (PPI) member, who was a family carer. The research team comprised old age and consultation liaison psychiatrists, a psychologist, a social care researcher, a speech and language therapist, a gerontologist and a linguist. We shared our general views that palliative care for older people with dementia should focus on the quality of life and avoid futile interventions. This may impact our interpretation of data generated in this study and the development of our decision guide. However, we reflected on our assumptions and involved a PPI member in providing feedback on the interpretations and development throughout the research processes.

Data collection methodsContributions from the earlier workstreams to the design of this study

The systematic review (WS1) informed the structure of case scenarios used in the discussions in co-design workshops. Interview findings with people with mild dementia, family carers and professionals (WS2–3) were fed into the co-design process, in terms of identifying problems and needs, providing feedback and solutions and generating ideas.[26,30,35] We also asked interview participants for suggestions about the design of our decision guide. In summary, from WS2, people with mild dementia want to postpone discussions about future eating and drinking problems, partly due to fears of being a burden to family and of being treated like a child. They wish to maintain a good quality of life rather than be kept alive at later stages by artificial nutrition and hydration.[15] In WS3, family carers often experienced frustration with delays and repeated conversations about eating and drinking with different professionals, while professionals felt unprepared for decision-making and found it challenging to work across the multidisciplinary team.[32] In particular, as people with dementia were not included in WS4, throughout the co-design workshops we advocated for them by raising their views and needs identified from WS2 and including the findings in the final decision guide (Supporting Information: File S2).

Co-design workshops with family carers and professionals

From May to September 2021, we held two co-design workshops with family carers, two co-design workshops with professionals and three research advisory team meetings. All workshops and meetings were online using Zoom software. Several versions of the prototype were discussed and iteratively refined through the co-design workshops and research advisory team meetings (Supporting Information: File S1). We conducted user testing to get final feedback and finalize the decision guide (Supporting Information: File S2 for the final decision guide). A flowchart of co-design workshop activities in this study is shown in Figure 2.



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Although we acknowledged that a mixed group approach can unleash ideas that people might otherwise hold back and allows them to hear about other people's experiences,[36] we conducted co-design workshops with a homogenous group to minimize power imbalance among participants with different backgrounds,[37] especially between family carers and professionals. However, we summarized and brought the ideas generated from one group to another to stimulate further discussions.

Timeframes and number of participants of co-design workshops and research advisory team meetings are displayed in Table 2. The specific steps and activities of the co-design process are described below.

Table 2 Timeframes and number of participants in workshops and research advisory team meetings

Online workshops or meetings	Month/year	No. of participants
Family carer workshop (round 1)	05/2021	8
Professional workshop (round 1)	06/2021	6
Advisory team meeting (round 1)	06/2021	7
Family carer workshop (round 2)	07/2021	6
Professional workshop (round 2)	08/2021	8
Advisory team meeting (round 2)	08/2021	7
User testing (individual, email)	08–09/2021	26
Advisory team meeting (round 3)	09/2021	7

Preworkshop preparation

We posted printed materials to participants in advance and asked them to use the materials on the day of the workshops (Supporting Information: File S3). They could record any ideas on these papers. This enhanced physical engagement and helped those with technology difficulties or who were less confident in speaking during the workshops. Participants were also able to understand the nature and expectations of our workshop activities and prepare some ideas to share, which is important in public engagement in research.[38]

Co-design workshop procedures

All co-design workshops lasted approximately 1.5 h and were facilitated by the first and second authors. We started by introducing workshop agreements mainly covering confidentiality and generating mutual respect. We emphasized some basic principles of a co-design process and encouraged participants to express their ideas, listen to other people and ask questions whenever they wished (Supporting Information: File S3). We also navigated other methods of communication in the online software, for example, the use of the ‘Raise Hand’ and chat box functions to create a ‘safe’ and ‘brave’ space for participants to share their experiences and ideas and express disagreement.[36]

We used an online interactive board (Google Jamboard) to help facilitate and visualize discussion during our online workshops. However, we acknowledge that some people are not familiar with these technologies, and their use may hinder full engagement or make them feel uncomfortable. We, therefore, helped them type their views on the board and asked them to discuss them freely during the workshops. If participants wanted to express any ideas in writing, they could do so in the chat box function in Zoom.

Focus of discussions: First round of co-design workshops

During the first workshops, we introduced the study background, described the overall study aims, presented findings from our WS2–3 interviews to participants and then started workshop activities. For both family carer and professional workshops, in the first rounds, we initially identified the difficulties of having conversations and decision-making about eating and drinking for people with severe dementia during their hospital stay. We later focused on and creatively thought about possible solutions to the problems and initially discussed the design of our decision guide.

K. A., E. L. S. and N. D. then mapped the discussions in workshops and findings from our earlier workstreams into matrices (see below) as well as matters discussed in the first whole advisory team meeting to develop a prototype of the guide.

Focus of discussions: Second round of co-design workshops

In the second round, discussions focused on the decision guide development. We recapped previous discussions and presented the prototypes (Supporting Information: File S1) to co-design participants. A printed copy of the prototype was also sent to the participants in advance. This was to help participants comment on the physical version and visibility of the prototype, for example, font size, icons and colours. The co-design groups then reflected on the prototype and generated ideas to improve it. They also discussed and refined the aims of the decision guide. After that, K. A., E. L. S. and N. D. developed a further iteration of the prototype based on the discussions and feedback from the co-design groups with additional comments from the second advisory team meeting.

Postworkshop involvement

After each workshop, we created an electronic copy of the online discussion board and shared this with participants (Supporting Information: File S3). We encouraged participants to send us any additional thoughts on the workshop discussion, edits on the prototype and suggestions for the workshop process. Using a prepaid envelope, they could send us back the printed workshop materials with their notes and edits. We did not audio-record workshops, but facilitators made detailed notes. We then compiled a report for research advisory team meetings. There was no formal analysis of the notes, but we scrutinized, summarized and aggregated the data into the matrices (Supporting Information: File S4).

User testing

From August to September 2021, we conducted user testing by sending a near-finalized prototype (Supporting Information: File S1) together with a feedback form (Supporting Information: File S5) to 40 people. The feedback form was adapted from a user manual for assessing the acceptability of a decision guide for osteoporosis treatments, as proposed by O'Connor and Cranney[39] and is freely available to use. Those invited to this user testing were participants in our interview and workshop studies and people who had expressed interest but could not join the workshops.

The feedback form for user testing asked participants to rate the content of each section and comment on the amount of information, and the length of the nearly finalized version of the prototype. We also sought its perceived usability, that is whether the participants perceived the prototype would help start conversations and make decisions. The form also provided spaces for free-text comments.

We offered participants the option of giving only qualitative feedback if preferred, either via emails, online meetings or telephone calls. Descriptive analysis was used to present the quantitative data, for example, frequency, proportion and percentage. There was no formal analysis of free-text comments, but we included them in the matrices.

Data synthesis using a matrix approach

To develop the decision guide, we used a matrix to collate all findings from every workstream. The matrix was initially structured using a decision-making model developed in the WS1—systematic review—to create the headings for rows. It helped prioritize problems and identify possible solutions to conversations and decision-making about eating and drinking problems in acute hospital settings. It also informed the content, format and mode of delivery of the guide.

Data from every workstream were iteratively populated throughout the development process in WS4, and we subsequently modified key components of the matrix to enhance its clarity and thoroughness. Table 3 shows the key components of the matrix and indicates if data from the individual workstream was available for each component. A copy of the populated matrix is available in Supporting Information: File S4. Throughout the processes, we used the minimum criteria of the International Patient Decision Aid Standards (IPDASi) version 4.0 framework to inform the necessary components of our decision guide[40] (Supporting Information: File S6).

Table 3 Matrix table for synthesizing and developing a decision guide

Key components	WS1 systematic review	WS2 PLWD interviews	WS3 family interviews	W3 professional interviews	WS4 workshops and research advisory team meetings
Common admitting conditions	NA	NA	x	x	x
Common eating/drinking problems at home or in care home	x	x	x	x	x
Common eating/drinking problems in hospitals	x	NA	x	x	x
Identify the problems and decisions	x	x	x	x	x
Initiate discussion or conversation	x	x	x	x	x
Exchange information: understanding disease and interventions	x	x	x	x	x
Exchange information: explaining disease and interventions	x	x	x	x	x
Acknowledge emotions of all involved	x	x	x	x	x
Clarify values of eating/drinking problems and interventions	x	x	x	x	x

Clarify values of approaching the *decisions*	x	x	x	x	x
Consideration of feasibility	x	x	x	x	x
Communicate preferred choices	x	x	x	x	x
Make a final decision and communicate the decision	x	x	x	x	x
Provide eating/drinking interventions	x	NA	x	x	x
Monitor and evaluate the support	x	x	x	x	x
Renegotiate the decision	x	x	x	x	x
Postdischarge education and support	NA	NA	x	x	x
Facilitators	x	NA	x	x	x
Barriers	x	NA	x	x	x
Decision guide: format	NA	x	x	x	See the last three rows below
Decision guide: techniques	NA	NA	NA	NA	See the last three rows below
Signpost additional information	NA	NA	NA	NA	x
Other components	x	NA	NA	NA	x
Decision guide: WS4 workshops and research advisory team meetings					
Aims and expected outcomes	x				
Preferred format	x				
Possible techniques to be used	x				

Abbreviations: NA, not applicable; PLWD, people living with dementia; WS, workstream. **FINDINGSParticipant characteristics**

Most family carers and professional workshop participants continued their participation in our WS3 interview study. Many participants joined both co-design workshops for each group. The co-design workshop participant characteristics are shown in Table 4.

Table 4 Co-design workshop participant characteristics

Participant characteristics	1st Family workshop (*n* = 8)	2nd Family workshop (*n* = 6)	1st Professional workshop (*n* = 6)	2nd Professional workshop (*n* = 8)
Participants from the interview study (WS3)	5	5	4	6
Age (years)				
Mean	46.5	46.5	42.2	44.8
Range	39–63	29–63	29–54	29–57
Sex (female: male)	5:3	4:2	6:0	7:1
Marital status				
Married or in a civil partnership	5	3	4	8
Single never married/in a civil partnership	2	3	1	0
Co-habiting with partner	1	0	0	0
Divorced	0	0	1	0
Ethnicity				
English/Welsh/Scottish/Northern Irish/British	5	3	4	6
Asian/Asian British	1	0	2	2
Black Caribbean/Black British	0	1	0	0
Any other European White	2	2	0	0
Family carer participants				
Current caring situation				
Current carer	1	2	-	-

Bereaved or former carer	7	4	-	-
Relationship to the person with dementia				
Child	7	6	-	-
Grandchild	1	0	-	-
Hospital professional participants				
Current occupation				
Physician	-	-	1	2
Nurse	-	-	3	3
Speech and language therapist	-	-	2	2
Clinical psychologist	-	-	0	1
Years of working with people with dementia (year)				
Less than 1	-	-	0	0
1–5	-	-	2	2
5–10	-	-	1	0
More than 10	-	-	3	6

In user testing, 26 participants provided feedback and suggestions (response rate to invitation 65%). Of 26 participants, 22 participants used the feedback form. Four participants preferred to give qualitative feedback; two used emails, one chose an online individual meeting and another a telephone call. We did not collect demographic data, so the data are not presented here.

Key findings from workshops and research advisory team meetings

From the co-design workshops and data synthesis using matrices, it seemed most participants were concerned about conversations or communications between family carers and professionals rather than decision-making per se. For example, most would prefer risk feeding, in which people with dementia will be helped to eat as long as it does not cause them distress and to not use tube feeding. However, this can involve long and difficult conversations before reaching this decision. Through the co-design process, the co-design groups and research team developed the aims of the decision guide as being to support conversations and communication between family carers and professionals to reach informed shared decision-making about eating and drinking problems in patients with severe dementia.

Co-design workshops with family carers and professionalsCommon problems in conversation and decision-making for eating and drinking and possible solutions

A voting exercise was used to identify and prioritize the problems and set some direction for group discussions about possible solutions. Separate lists of potential problems in the conversation and decision-making for carer and professional workshops were predetermined and informed by the findings of WS1–3. We presented this list to participants and asked them to choose the problems that they perceived as most important. Using Zoom's voting function, they could choose multiple options from the list, and their choices were anonymous; however, we also explicitly asked them to raise any other issues they wished to discuss (Supporting Information: File S3).

From Table 5, family carers identified ‘finding the right staff to talk to’ (7 of 8 first workshop participants) as the most concerning problem, followed by difficulty ‘adapting to hospital rules and routines’ (5/8) and ‘addressing personal and cultural beliefs’ (5/8).

Table 5 Family carers and hospital professionals in the first round's identification of the most important points about conversations and decision-making for eating and drinking[a]

Family carers	*N* = 8	Hospital professionals	*N* = 6
Finding the right staff	7	Know overall dementia prognosis	4
Hospital rules and routine	5	Explain treatment options	4
Personal and cultural beliefs	5	Hospital rules and routine	3
Available treatment options	3	Time pressure	3
Understanding about eating/drinking problems	3	Initiate conversation	3
Time pressure	2	Disagreement between people	3
Initiate conversation	2	Personal and cultural beliefs	2
Emotional difficulties	2	Explain eating/drinking problems	2
Disagreement between people	0	Confusing/underrecognized roles	2
		Emotional difficulties	1

a

Participants could choose more than one option (multiple choices).

In workshop discussions, family carers repeatedly mentioned difficulties in finding a constant point of contact and felt frustrated by having to say the same things to different professionals. They suggested having a live document or signs over beds that people could always access and update information about eating and drinking in the hospital.

Family carers emphasized that empathy and mutual respect were important to help conversations run successfully. Therefore, professionals should offer sufficient time and be sensitive to their emotions and readiness for the discussion. This would then enhance the shared decision-making processes.

From Table 5, professionals selected both 'knowing the overall progression and prognosis of dementia' and 'explaining available treatment options' as the most important issues during conversations and decision-making (4 of 6 first workshop participants).

Professionals wanted to talk to the family member or carer who knew the person with dementia best to understand the overall progression of dementia and hear about existing eating and drinking problems and needs. This was in line with a person-centred care approach. They also suggested using food and bowel charts as objective evidence to identify eating and drinking problems in hospitals and initiate conversations. These charts could help families to see eating and drinking changes and to build a more realistic understanding of the situation.

Professionals thought it was important that family members should talk to a member of the professional team who is familiar with and confident enough to discuss common eating and drinking problems of people with severe dementia. Otherwise, family members risk creating unrealistic expectations that may influence ensuing conversations. Many professionals wanted to inform families about some specific interventions provided in their hospitals, for example, finger foods, cultural menus and protected mealtimes. This was to allow families to come to hospitals and help communicate with the person and offer their preferred food and drinks. Professionals would also like to have electronic systems on which they could update the information they received from family members and share it with the wider health and social care providers.

All participants emphasized the cultural and personal meaning of eating and drinking, which should form part of the conversation. Language and communication difficulties could prevent people from explaining the situation or expressing their wishes. Professionals wanted to emphasize to family members that dementia was a terminal illness and to explore and validate their understanding and expectation about eating and drinking, especially at the end of life, while family carers wanted to know what the signs of end-of-life in dementia were, especially in terms of eating and drinking difficulties.

Content, format and mode of delivery of the decision guide

Throughout WS4, several versions of the prototype were presented and discussed in the co-design workshops and research advisory team meetings (Supporting Information: File S1). Most participants agreed it could be a single version for both family carers and professionals to enhance a culture of trust and ensure that everyone would start the conversations from the same perspective. Therefore, both family carers and professionals stressed the decision guide should be sensitive to, but not underestimate, levels of understanding and knowledge of end-users. For example, information should not make family carers feel patronized.

Although some participants suggested that the decision guide could be a booklet, most family carers and professionals agreed to keep it shorter (no more than two pages of A4) because it could be difficult for family carers or professionals to read long documents. The decision guide could be a conversation starter and signpost for people to further information resources elsewhere.

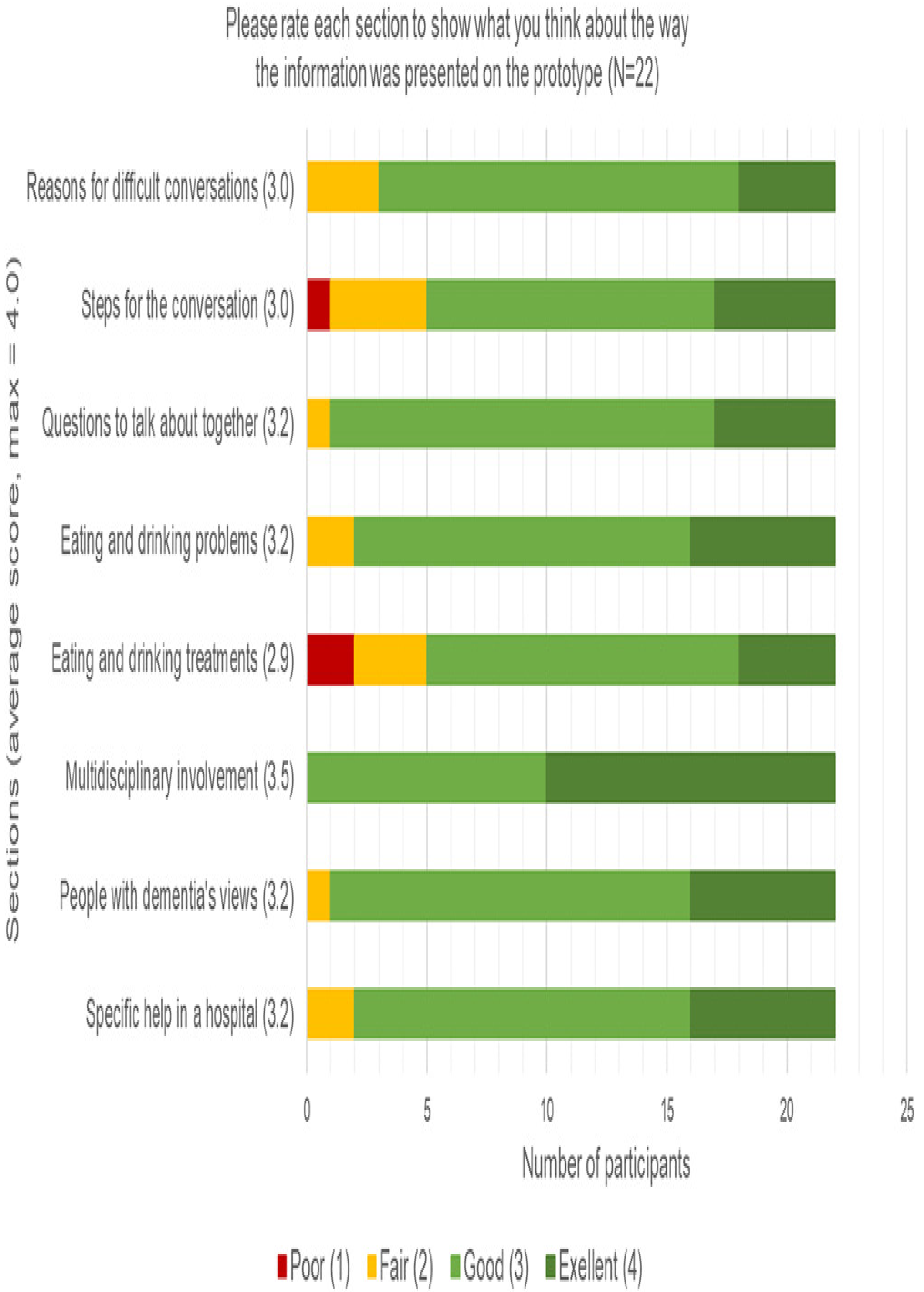
Research advisory team meetings

Between the two rounds of workshops, the research advisory team helped interpret and expand the discussion from the workshops, and feedback on the prototype. For example, the team helped review the aims of the decision guide and suggested it could help professionals focus on engaging families in conversations, and vice versa, rather than mechanically going through all eating and drinking options. The decision guide might also benefit senior professionals for use in educational or training programmes for junior colleagues.

Food and bowel charts were thought useful subject areas to start the conversation and agree upon a care plan, but, at the end-of-life, professionals said they would not frequently record food, drink or whether the person has passed urine or had opened their bowels, so people could then stop using the charts. The team noted that signs over patients' beds were often left behind when patients moved to another bed or ward, so it would be more practical if the family kept the guide or information with them, ready to discuss with professionals.

User testing with family carers and professionalsContent of each section of the prototype

User testing was conducted on the nearly finalized version of the prototype (Supporting Information: File S1). As shown in Figure 3, participants could rate each section of the prototype according to the Likert scale of ‘poor, fair, good, and excellent’ (Supporting Information: File S5). To understand the overall rating, we converted these responses to the score of ‘1, 2, 3, and 4’, respectively, and then calculated the average scores of each section from all participants, as reflecting the Likert scale.



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From the user testing, participants rated ‘good–excellent’ (average score 3.1–4.0) for most sections of the prototype.

The introduction regarding a multidisciplinary approach was the most well-received section. The eating and drinking treatment section was least favoured by participants, and one of the three sections was rated as ‘fair–good’ (average score 2.1–3.0), along with the ‘reasons for difficult conversations’ and the ‘steps of conversations’ section. This indicated these sections warranted further attention and reiterations.

Overall presentation and perceived usability of the prototype

From Table 6, most participants thought the amount of information was ‘just right’ (68.2%) presented in ‘just right’ length of the prototype (86.4%). Every participant thought that the prototype could help people start a conversation about the eating and drinking problems of people with severe dementia in hospitals. Participants had mixed views about whether the information presented in the prototype was slanted particularly towards any eating and drinking treatments.

Table 6 Feedback on overall presentation and perceived usability of the prototype from the user testing

Feedback topics on the prototype	Family (*N* = 9)	Professional (*N* = 13)	Total (*N* = 22), *N* (%)
Participant type (total *N* = 26)a			
Participating in the interviews only	6	4	10 (38.5)
Participating in the workshops only	1	2	3 (11.5)
Both in interviews and workshops	3	6	9 (34.6)
People who expressed interest	0	4	4 (15.4)
Length of presentation			
Too long	0	3	3 (13.6)
Just right	9	10	19 (86.4)
Amount of information			
Too much information	2	5	7 (31.8)
Just right	7	8	15 (68.2)
Presentation slanted towards			
Use of tube feeding	0	1	1 (4.6)
Use of eating and drinking with accepted risks	0	7	7 (31.8)
Balanced	9	5	14 (63.6)

Do you think we included enough information to encourage a family member and hospital team get the conversation started?			
Yes	9	13	22 (100)
Would you have found this prototype useful when making your decision about eating and drinking for people with severe dementia in a hospital? (total *N* = 20)b			
Yes	7	10	17 (85.0)
No	0	3	3 (15.0)

a

Including four participants who provided only qualitative feedback.

b

Missing data from two former family carers who might have perceived this question was no longer relevant to them and so skipped the question.

Three professional participants (from 20 participants) thought information on the prototype might not be helpful in making their decisions. In free-text comments, a professional participant explained that the information might not be useful for themself but could be for other professionals:

I personally wouldn't find this form useful for me …This is all information I am well aware of. BUT I do think the information is useful for doctors, however as a prompt to think of topics/issues they should be thinking about but often are not on a busy acute ward. I also think it does cover all of the key points for them.

Another two participants considered it was ‘too generic [and] each patient was different’ and that as ‘a general overview of the problem without necessarily helping [people] to reach a conclusion [and] as a prompt to initiate conversation it was helpful’. This was in line with the aims of the decision guide as defined by the co-design groups to create a conversation facilitator for families and professionals rather than a self-study document to reach a decision or conclusion by themselves. Similarly, professional workshop participants emphasized that the guide should not be prescriptive, and families and professionals should have a discussion to reach a personalized care plan.

Qualitative feedback from free text comments, emails and one-on-one meeting

The participants' main concerns and suggestions concerned clarification of the purpose and of the end-users of the guide at the beginning. Some suggested that every part was applicable and accessible for both family carers and professionals, in terms of comprehensibility and relevance. Although many participants thought the information was acceptable in its length and volume, they suggested that we condense the text and check the flow to enhance readability.

Some provided free-text comments about the clarity and sensitivity of the language used. For example, some professional participants thought that the explanation about the disadvantages of tube feeding might be ‘too daunting’ to families, while family carers did not make any comments about this point. It was considered important to emphasize the multidisciplinary nature of involvement in eating and drinking for people with severe dementia in acute hospitals. It was thought worth explaining the roles of each professional in decision-making processes as well.

Most participants mentioned they liked the guide because it contained sufficient, useful and concise information within a compact format. Overall, the sections had a clear layout using headings, blocks and colours; hence, they were easy to follow and could help get a conversation started.

Finalization of the decision guide

Based on the feedback and suggestions, we amended the prototype guide and cross-checked with the matrix table to ensure consistency with findings from all workstreams. For example, when co-design workshops suggested emphasizing the person-centred and holistic approach, we then checked the sections (rows) of the matrix table across all workstreams (columns) that might be relevant to this, for example, the sections of ‘Acknowledge emotions of all involved’, ‘Clarify values of eating/drinking problems and interventions’ and ‘Clarify values of approaching the decisions’ (see Table 3 and Supporting Information: File S4).

In our last research advisory team meeting, the team helped interpret the findings from user testing and provided suggestions for the decision guide. Following this meeting, we continued refining and checking the guide met current evidence, guidelines and data synthesized from all workstreams. This was also checked against the IPDASi v4.0 criteria[40] (Supporting Information: File S6). We worked with a professional designer to help redesign and convert the prototype into a decision guide. Following a final check with the research advisory team and workshop participants, we agreed that the decision guide could be finalized (Supporting Information: File S2).

Dissemination of the decision guide

From 1 December 2021, the final version of the decision guide (Supporting Information: File S2) was uploaded to the UCL Division of Psychiatry website and released via Twitter. We also emailed a copy of the decision guide with the link to the UCL Division of Psychiatry webpage to all participants, colleagues and relevant organizations. It was well-received as people shared or recommended the guide to others. We heard that the decision guide has been circulated via email within local professional groups and uploaded to some organizations' electronic resources. One Admiral Nurse (dementia specialist) in an NHS trust offered to help with implementation.

Feedback on workshops and co-design process

Throughout the co-design process, we sought feedback and suggestions about workshops and the co-design process from participants, including from the feedback form in user testing (Supporting Information: File S5). Many participants sent us emails after workshop sessions saying that they found the workshops engaging and thought-provoking. In one of our first workshops, one participant suggested a queueing system for people to speak up because some participants might be not confident enough to do so. In the following workshops, we encouraged participants to use the ‘Raise Hand’ and ‘chat box’ functions, and this helped us to manage the queue and other people to build up discussions from their comments in the chat box more effectively.

Twelve workshop participants responded to the user testing process. In the feedback form, workshop participants mentioned the workshops were ‘well organised’, ‘easy to join and communicate’ and ‘naturally’ interactive, and it was nice to meet and exchange views with other people. It also made them feel that ‘[their] views mattered and counted which was great’ (former carer).

Some workshop participants suggested that the workshops could be ‘longer to cover all the points that were intended’ (current carer). Some were interested in having a workshop to bring persons with dementia, family carers and health professionals together to share ideas. Some thought it would be better to keep the same participants in all workshops because change could affect group dynamics and prevent discussion.

DISCUSSION

We applied systematic and transparent methods to develop a decision guide to be used by families and professionals covering nutrition and hydration for people with severe dementia in acute hospitals. We used various sources of evidence including a systematic review and interview and workshop studies, the data from which were synthesized. All processes interweaved principles of PPI in research and shared decision-making in health and social care which have been encouraged in dementia and palliative care.[41,42] The study set a context within that families and professionals (the end-users) can be heard and co-design the guide with researchers, while the guide itself supported the shared decision-making approach to dementia care decisions among those involved.

Practical points about a decision guide regarding the care for people with severe dementia in acute hospitalsAims and users of the decision guide

Consistent with our decision guide, the most common components of decision-making guides appear to be tools to improve communication between persons with dementia, family and healthcare professionals.[43] Our decision guide was co-designed to be used in clinical discussions as a conversation facilitator for both families and professionals in a single document. Decision guides used in a discussion with other people involved in the decision-making were also considered more beneficial than self-guided tools.[44]

Decisions about eating and drinking interventions, either about risk feeding or artificial nutrition and hydration, involve personal values and cultural beliefs.[8] We expect the guide would help, for example, people initiate conversations, families understand the working processes of the hospital team, and professionals recognize emotions and the personal meanings for the person with dementia and their family associated with eating and drinking. The decision guide provided sufficient and honest information to encourage further discussion. This is consistent with previous co-designed research of a discussion tool about choices of care for people with dementia[35] and decision guides in general.[19]

Content presentation of the decision guide for dual users

In the co-design process, family carers wanted honest and full information about all nutrition and hydration treatments in the guide. However, some professionals were not keen to include tube feeding because it is not recommended for people with severe dementia,[6,42] and so, if it was included, it could be seen as an alternative 'option'. However, the core principles of a decision guide are to help a decision-maker to make an 'informed' and 'value-sensitive' decision.[19] With the agreement of workshop participants and the research advisory team, we have included information about tube feeding in the guide and presented it in an honest way, based on current evidence.[6,45]

Commenting on the presentation, family carers perceived the information as balanced and not distressing, but professional participants had very mixed opinions on this (see Table 6). It is worth noting that our decision guide was not a 'patient decision guide' that would be used by persons living with dementia, but by families and professionals to make decisions on behalf of the persons. Therefore, it was not entirely about the preferences of families and professionals, but rather it aimed to prompt everyone to consider important points and place the person with dementia at the centre of the discussion, in line with a person-centred care approach.[46]

Key lessons from the co-design process

Consistent with the literature,[36] the co-design process through workshop activities helped participants in this study feel heard by other participants who shared the same experiences, ideas and interests, and so they did not feel alone. The co-design process helped them recognize other aspects of eating and drinking problems that they had never thought of, but could then prompt creative solutions or learning from other people's practice which they might consider using in the future.

Table 7 shows some lessons identified from this study that could be applied in future co-design projects. Some of these lessons are discussed in more detail below.

Table 7 Lessons identified from this study for further co-design research

Outline expectations about roles and workshop activities in advance
Start workshops with informal introductions (ice-breaking)
Set workshop rules to create safe space and open discussion
Explain and agree workshop objectives and agendas
Consider any power hierarchy between different stakeholders
Be alert to possible exclusion from actual co-design participation (tokenism)
Carefully plan and manage any lack of agreement among co-design groups and between researchers
Observe and respond to person's emotions and needs
Observe and respond to group dynamic and relationships
Offer flexibility about time and location (more convenient for online workshops)
Consider varying ability and skills of each participant to take part in workshop activities
Consider suitability of workshop activities for intended participants and platform
Provide technological support, especially for online workshops
Offer variety of participating methods to maximize participation
Minimize any mismatch between the design of activities and the expected outcomes
Address any difficulty translating general conceptual ideas into practical solutions
Keep ongoing relationships and involvement with participants after the workshops
Provide sufficient time for each co-design workshop
Consider time and financial constraints of entire research project

Facilitate group dynamics and relationships

From the user testing feedback, a consistent group of participants across the co-design process enabled us to sustain group dynamics and relationships and led to richer discussions. In a previous co-design study, mutual understanding and trust within a co-design group enabled a collaborative, compassionate and open mindset to collectively build on members' ideas and create solutions.[36] However, making a commitment to participate in every session may be difficult for some participants, as can be seen, as some participants could not join the second workshop due to their unavailability. It was suggested participants are offered flexibility about their ability to participate.[47] We explicitly informed participants that they were free to join all or any of the workshops, and we tried to accommodate the most convenient dates for most participants.

Online platforms

All workshops in this study were online, and logistically this made the study an efficient use of time. It minimized the scheduling problems which can be a major problem of many face-to-face workshops.[48,49] However, the online platform hindered our use of certain types of co-design exercises that could be useful, for example, patient journey or mind mapping, and paper prototyping and sketching. Some online interactive platforms can support these exercises, but these could have been too complex for some research participants. Training participating end-users in research skills has been found helpful,[27] which may include skills for these technologies. However, this was not feasible in this study due to time and financial constraints.

Acknowledge and manage the lack of agreement

There was sometimes no agreement of ideas and preferences about the decision guide between researchers, family carers and professionals. This difference of opinion is common in co-design studies and can compromise the study process and outputs and/or create feelings of tokenism and frustration and disappointment at the missed co-design opportunity.[27] However, it is the essence of co-design research to facilitate participants in creatively thinking about ideas and agreeing, to some extent, with group conclusions, moving forward with specific solutions to the problems.

Design of workshop activities: General concepts versus practical solutions

Workshop exercises can sometimes make it hard for participants to reach specific discussions and solutions within the workshop's pace and flow. For example, we found it easier for participants to instantly come up with the general aims of the guide and solutions to eating and drinking problems. It was more challenging when we specifically asked them how to frame the information in the guide, and this made one of our workshops rather quiet. It is important to carefully design and simplify exercises to ensure that they would not deter contributions to the discussions. This should be accompanied by sufficient time to respond using a variety of participatory methods, which was found to be effective in this study, for example, chat box and printed materials.

Strengths and limitations

This study used a systematic and transparent approach to co-design a decision guide. It was based on our series of studies about the perspectives, experiences and needs of everyone involved, including people with mild dementia, family carers and professionals. Each study was done in sequence and informed subsequent studies. We involved PPI members and experts in palliative care and dementia care to feedback on study materials and interpret findings. We used the matrix table to synthesize all data and develop the guide, which allows for transparent reporting and was found beneficial in another co-design study.[24]

All family workshop participants were recruited from JDR, and most of them continued their contribution to our interview study. These could be ‘super users’ who are constantly involved in research projects and might not represent the typical population of people with dementia and their carers.[47] They could be a selected group of people who are keen on helping ‘design’ and participating in co-design research.[29] However, many family carers in our study were quite new to research involvement and had relatively mixed experiences and backgrounds. We triangulated family discussions with feedback from professional workshops and research advisory team meetings, including a PPI member, to help advocate for persons with dementia and other families.

Implications for policy, clinical practice and future research

Families and professionals can freely use the guide to prepare for or follow during such conversations and decision-making (Supporting Information: File S2). However, we acknowledged possible barriers to implementing our decision guide in clinical practice, including indifference on the part of healthcare professionals and organizational inertia.[50] Regarding participants' feedback, the guide could be used in clinical teaching to junior healthcare professionals or clinicians who are not fully confident in this area.

With adequate time and funding resources, further studies may strive to develop a process to involve people with dementia in co-design workshops and to combine groups of participants and equalize their power, thereby fostering mutual and holistic understanding. A decision guide, for example, in a pictorial format may support people with dementia to indicate their preferences and further help family carers and professionals make decisions.

Future larger evaluation research may explore our decision guide's acceptability in real-world settings. Standard tools may be used to measure, for example, decisional conflict, patient-clinician communication, participation in decision-making, decisional regrets and satisfaction with choices, as in previous studies.[19,44] A future development study may include an electronic format of a decision guide that would have more interactive and separate sections or layers of information and automatically direct users to information relevant to them.[35]

CONCLUSION

Through a rigorous co-design process, we developed a decision guide to support conversations and decision-making regarding nutrition and hydration for people with severe dementia in acute hospitals. From user testing, the decision guide was perceived to be useful in initiating conversations and making decisions. Co-design workshop participants reported very positive experiences of being involved in our co-design process. The final version of the decision guide was widely disseminated and is being used in clinical practice; however, it would be needed to be tested in future evaluation research.

AUTHOR CONTRIBUTIONS

Kanthee Anantapong conceived and designed the study, recruited participants, acquired data from co-design workshops, analysed and interpreted data, developed a prototype of the decision guide and wrote the manuscript. Andrea Bruun acquired data from co-design workshops and contributed to the development of workshop materials and the interpretation of the data. Anne Walford, Christina H. Smith and Jill Manthorpe contributed to the development of workshop materials and the interpretation of the data. Elizabeth L. Sampson and Nathan Davies designed the study, supervised data collection and data analysis and contributed to the interpretation of the data. All authors reiterated the prototype of the decision guide, finalized the decision guide and revised and approved the final version of the manuscript to be published.

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CONFLICT OF INTEREST

The authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

The data that supports the findings of this study are available in the supplementary material of this article.

ETHICS STATEMENT

Ethical approval was granted by the Health Research Authority (Camden &Kings Cross Research Ethics Committee, REC reference: 20/LO/0049).

DETAILS

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Understanding support systems for Parkinson's disease management in community settings: A cross-national qualitative study

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ProQuest document link

ABSTRACT (ENGLISH)

Background

Health and social care systems face difficulties in managing multimorbidity, disease burden and complex needs in long-term conditions such as Parkinson's disease.

Objective

This study aimed to develop a European understanding of how health and social care professionals can collaborate with stakeholders from different organizations and sectors to enhance the management of Parkinson's disease in a community setting by identifying the existing gaps in this process and how people with Parkinson's disease and their family carers could benefit from these partnerships.

Methods

A mixed-methods sequential study was conducted in Denmark, Norway, Spain and the United Kingdom. The findings from the qualitative phase are presented. Individual semistructured interviews were analysed using Braun's and Clarke's thematic analysis. A meta-ethnography approach was used to analyse and synthesize cross-national findings.

Results

A total of 41 healthcare professionals and 39 stakeholders from different disciplines and sectors were interviewed in the four countries. The participants acknowledged a lack of awareness of available resources and poor communication between the different support systems in the management of Parkinson's disease. To promote multiagency collaborations, the participants highlighted the need to organize services along the Parkinson's disease journey, patient involvement and strategic involvement of carers in organizing resources and Parkinson's disease care pathways. According to the participants, the benefits from multiagency partnerships could lead to an enhanced continuity of care and specialized knowledge, mobilization of resources in the community, personalized support and improved access to services.

Conclusions

Policymakers are called upon to create formal structures that facilitate multisectoral collaborations to promote an integrated system of care for the management of Parkinson's disease in the community. To address this challenge, we propose five strategies showing how organizations can work together to optimize the use of resources and enhance the management of Parkinson's disease throughout the illness trajectory.

Patient or Public Contribution

Patient and Public Involvement groups made up of stakeholders, healthcare professionals, patients with Parkinson's disease and family carers participated in the design of the study, the development of the interview guides and the validation of the findings.

FULL TEXT

INTRODUCTION

The overall number of people diagnosed with Parkinson's Disease (PD) has been growing progressively globally. In

2019, approximately 8.5 million individuals had received a PD diagnosis.[1] This estimation is expected to increase to 12 million people in 2050,[2] indicating that compared to other neurological conditions, PD has the fastest-growing rate in most countries.

Previous evidence has shown the direct and indirect costs associated with the management of PD, which affects both patients and family carers in relation to hospital admissions, medication, nonmotor symptoms and productivity loss.[3] The consequences of PD on an individual level may result in the need for continuous support to manage multiple aspects of everyday life, including mobility, work, medication, safety, social life and emotional stability.[3] In addition, cognitive deterioration in the person with PD (PwPD) may involve a financial burden for the PwPD, the family carer and the health and social care system.[4–6] Thus, support is needed from a long-term perspective and often increases with the progression of the illness.

Life with PD usually takes place in the community, where PwPD and their family carers have to learn to cope with the PD and its consequences.[7] Current guidelines contain information regarding medication, symptom management, patient and professional relationships and communication and assessments.[8] However, with a clear focus on an acute episodic model of care, healthcare services are under pressure and may neglect nonbiomedical consequences of PD (biographical disruptions, negative emotions, strained relationships, nonmotor symptoms and a restriction of meaningful activities), which constitute the most essential burden for patients and families and are the leading causes of hospital re-admissions and a poor quality of life.[7,9,10]

Furthermore, the existing National Healthcare Systems' personalized self-care plans and tools fail to capture how people live with and adjust to PD from the PwPD's and the family carers' perspectives. These demands on health and social care systems globally and the limited resources lead to gaps in the care pathways related to manage multimorbidity, disease burden and complex needs and to reach disadvantaged populations, which are understood in this study to be those having immigrant status and/or an ethnic minority background, being older, being socially vulnerable, living with disabilities due to long-term conditions and being a caregiver.[11]

Self-management programmes for long-term conditions are evolving and are now increasingly seen as a collective initiative involving personal networks and other community resources, which go beyond those traditionally known as formal services.[12] Consequently, this work builds on new understandings of how stronger collaborations between the levels of care and additional support can enhance existing self-management approaches for PD on a community level [13,14] while also reaching disadvantaged areas through more integrated action plans.[11,12] Previous research has shown that community resources such as voluntary organizations can improve health outcomes through broader forms of support that include the provision of information, physical or social activities, and are better able to reach disadvantaged populations compared with the health and social care services.[15,16]

Furthermore, European recommendations[8,17] are taking a strategic leap when it comes to placing patients and their families at the centre of decision-making processes and also regarding the importance of involving various agents in the management of long-term conditions, including PD. Nevertheless, despite these initiatives, the relationships between agencies are still not clearly established or understood. The lack of awareness of what support is available in the community can lead to an overlap in activities, limited use of community resources and action-planning gaps.[18–20] Understanding how systems of support for PD management in the community work is essential to enhance the reach of services. Moreover, it is paramount to identify the successful initiatives used by different countries and to learn from established good practices.

In response to the previously mentioned knowledge gaps, the overall aim of this paper is to develop a European understanding of how health and social care professionals can collaborate with stakeholders from different organizations and sectors to enhance the management of PD in the community, and to identify the existing gaps in the collaboration and the potential benefits for PwPD and their family carers.

In particular, the following research questions will be answered:

Q1. How could professionals and stakeholders from different levels of care and sectors work together to improve PD management in the community?

Q2. What are the gaps in the collaboration?

Q3. What could the potential benefits of partnerships for PD management be?

METHODSStudy design and setting

This article presents the qualitative phase of a sequential mixed-methods study conducted in Denmark, Norway, Spain and the United Kingdom. This study is part of the OPTIM-PARK project, which aims to enhance the process of living with PD by designing multisectoral care pathways to optimize the use of community resources across European countries. In this paper, we report findings from the qualitative phase, which is part of the development stage of The UK Medical Research Council framework for developing and evaluating complex interventions.[21] A strength of this study is the Patient and Public Involvement (PPI) from all countries in different phases to maximize the relevance, applicability and transferability of the findings. The study was reported using the Consolidated criteria for reporting qualitative research (COREQ) (see Supporting Information: File 1).

Participants

A purposeful sampling of health and social care professionals and stakeholders was chosen in each participating country. A total of 40 participants were selected to ensure a broad representation of profiles in each group:

- (1)
  Health professionals from different disciplines that provide support directly or indirectly to PwPD and family carers. The exclusion criteria were an unwillingness to participate in the project or they were not involved in the direct care or support of PwPD.

- (2)
  Stakeholders from different sectors that directly or indirectly impact in the management of PD and the development of care pathways for PD or other long-term conditions. The exclusion criteria were an unwillingness to participate in the project or a lack of involvement in their role in the strategic planning of community PD care.

Participants were recruited through the strategies shown in Figure 1. Two healthcare professionals and five stakeholders decided not to participate in the interview due to lack of time.

National Health Services	Organisations & Community groups	Other secondary strategies
- Parkinson specialiast consultations (secondary care services). - PD clinics. - Primary care mail out targeting people with PD. - Databases of clinicians.	- Parkinson's disease national and local support groups. - Other local organisations likely to have people with PD as members. - Local community groups when relevant (healthy living).	- Information and invitation through local pharmacies. - Local radio, newspapers, websistes of PD organisations. - Other Parkinson's trust or foundations. - Public municipal home care offices.

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Data collection

Semistructured individual interviews were conducted between April and October 2020 and supported by an interview guide (Table 1), which was developed by all partners (Table 2) and refined by the PPI groups in Spain and the United Kingdom. Interviews initially took place face to face (*n* = 16), although due to the Covid-19 pandemic, the majority had to be carried out by telephone (*n* = 30) and video conference (*n* = 34). The interviewers (all women) in all countries (Table 2) had extensive experience in conducting in-depth qualitative interviews.

Table 1 Interview schedule for semistructured interviews with stakeholders and health and social care professionals

Topic fields	Questions
Available resources/services/organizations:	What community resources are you aware of for people with Parkinson's and family carers?
	What systems of support are you aware of for people with Parkinson's and family carers?
	What kind/type of support do they provide?
	How did you hear about these resources?
	How can they contribute to more positive living with Parkinson's?
Collaboration between professionals, organizations and levels of care to improve PD management in the community.	Who is responsible in your organization for liaising with other organizations, professionals, policymakers?
	Have you ever, or do you currently, signpost or refer people with Parkinson's or family carers to any of these resources?
	Is there an official pathway within your service to signpost or refer?
	What role do you think different organizations have in the management and daily living of Parkinson's and caring for people with Parkinson's?
	Where do you think the responsibility lies in the provision of care and day-to-day living with Parkinson's and caring for people with Parkinson's?
Strategies to maintain these collaborations	What kind of relationship do you have with the different Parkinson's organizations?

	What kinds of relationships do you have with people living with patients and family carers?
	How have you established relationships with these organizations or resources?
	Have any of these relationships changed over time?
The benefits of these collaborations for PD management	What kind of intervention would you find most useful in your current practice/role?
	Do you feel represented in care pathways for Parkinson's?
	How would you define an ideal care pathway in which you have an active role?

Table 2 Research partners and Patient and Public Involvement (PPI) representatives involved in this study

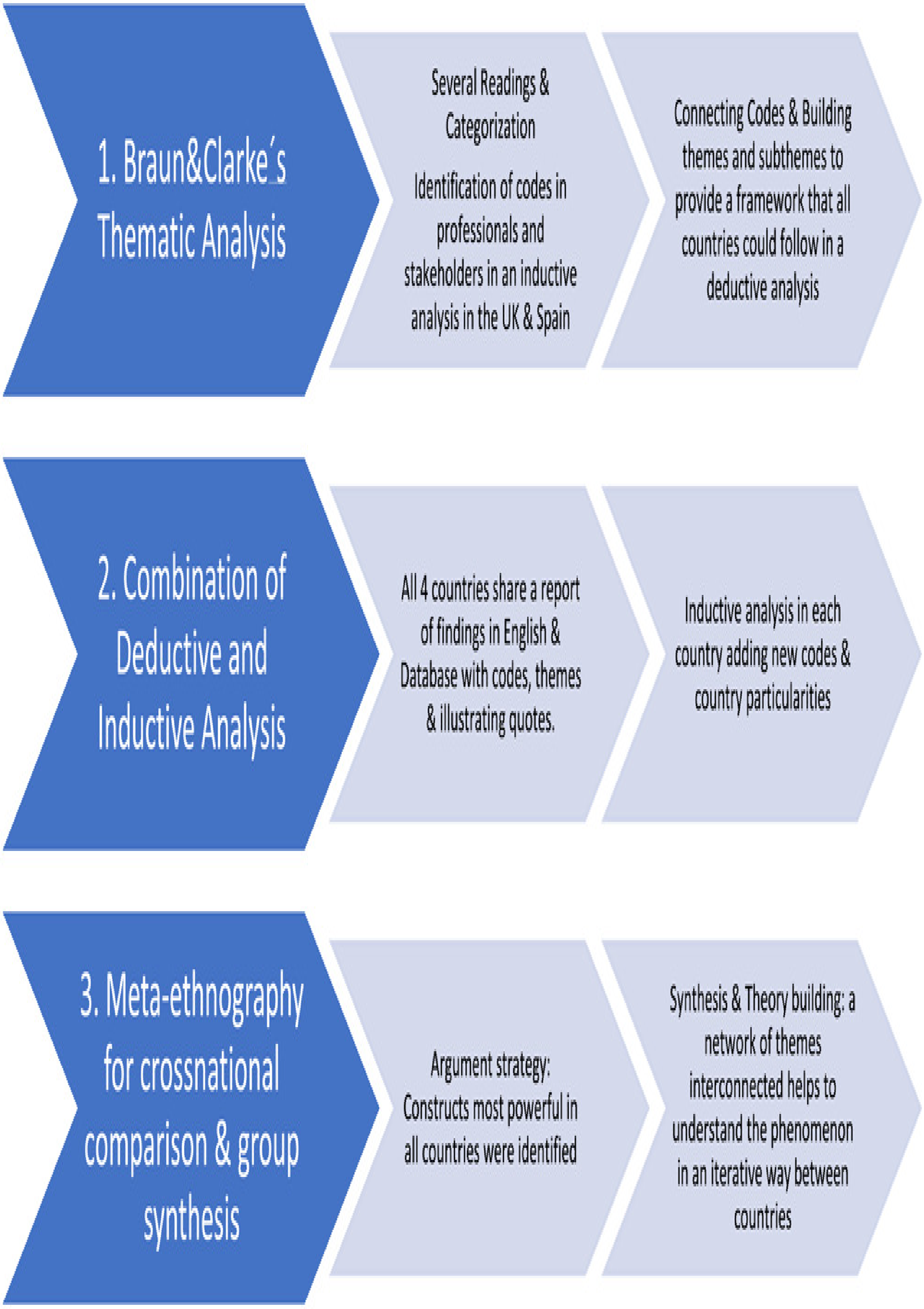
	UK	Spain	Norway	Denmark
Research partners	3 members: 1 Psychology, 1 Nursing, 1 Geriatric Medicine	3 members: Nursing	3 members: 2 Occupational therapy, 1 Nursing	2 members: Nursing
PPI	8 members: 1PwPD; 3FCs; 1 user organization; 2 professionals (1 from specialist care, 1 from community care), 1 stakeholder	10 members: 2PwPD; 2FCs; 1user organization; 4 professionals (1 from specialist care, 3 from community care); 1 stakeholder	6 members: 2 PwPD; 1FC; 1 user organization; 2 professionals (1 from specialist care, 1 from community care)	8 members: 1PwPD; 1 FC; 1 user organization; 5 professionals (1 from specialist care, 1 from community care)

Abbreviations: FC, family carer; PwPD, people/person with Parkinson's Disease.

All the participants were also asked to complete a sociodemographic form. The recorded interviews lasted between 32 and 118 min, with an average of 60 min.

Data analysis

All the interviews were transcribed and analysed following Braun's and Clarke's[22] thematic analysis combining deductive and inductive approaches (see Figure 2).[23–26] The analysis started with an inductive approach with several readings and the categorization of the full transcripts of the professional interviews from Spain and stakeholders interviews from the United Kingdom to provide a framework of analysis connected to the research questions that the other participating countries could follow. All the countries completed their national analyses following the framework provided using a deductive approach and also created additional codes/themes whenever relevant using an inductive approach. An excel database for each analysed group of participants including codes, themes, quotes and a description of the themes was created and shared among all countries.



Enlarge this image.

All the interviews were analysed in the original language of each country, and country-specific reports were written in English explaining the process followed, and included the findings with quotes for each particular group of participants. A total of 81 themes and 186 subthemes emerged from the analysis across all countries.

Once the country-specific reports and findings were received, a cross-national comparison was initiated, which involved multiple readings and discussions across teams towards an analytic synthesis. A meta-ethnography approach (lines of argument synthesis) was applied,[27] which helped to interpret and explain the findings across groups and countries, not in an attempt to create generalizations, but to ensure translation from one qualitative case study to another. Using the lines of argument strategy,[27] the most powerful constructs representing the entire data set from all countries were identified. This led to an agreed conceptual framework that incorporated a network of interconnected themes that are presented in the results and enhanced understanding of the phenomenon under study. This process led to comparative cross-national synthetic constructs elaborated in the discussion.

Ethical considerations

Following required ethical approval, the participants received a study invitation and were informed of the plans to maintain the participants' confidentiality and anonymity. They all signed an informed consent form. The participants were then allocated a study number and all the names were removed from the analysis and the written national reports.

RESULTS

In total, 41 healthcare professionals and 39 stakeholders were interviewed across the four countries (Table 3). Most participants were women comprising professionals (85.4%) and stakeholders (79.5%), with an average age of 48.5 and 51.1 years, respectively. In relation to the profile of professionals, many nurses (31.7%), physicians (26.8%) and therapists (34.1%) participated. The role played by the stakeholders in their organization was mainly managerial (48.7%) and direct work with other groups, people with PD or carers (30.1%). Moreover, at least 30% of the participants from each country met the requirement of working actively and directly with vulnerable groups.

Table 3 Description of samples in all countries and cross-national

	Professionals				Stakeholders				Total PROF	Total STH
	UK (*n* = 10)	Spain (*n* = 11)	Norway (*n* = 10)	Denmark (*n* = 10)	UK (*n* = 11)	Spain (*n* = 10)	Norway (*n* = 9)	Denmark (*n* = 9)	41	39
Female	8	10	10	7	10	8	5	8	35	31
Male	2	1	0	3	1	2	4	1	6	8
Age	48 (35–63)	45.5 (31–62)	48.2 (28–67)	52.6 (37–60)	50.6 (38–74)	54 (42–67)	51.7 (38–77)	49.6 (39–59)	48.5 (28–67)	51.5 (38–77)

Years in this position	21.6 (11–41)	22.5 (6–42)	12.9 (5–20)	22.2 (5–35)	9 (0.17–24)	12.9 (2–43)	8.6 (1–16)	7.3 (1.5–20)	19.9 (5–42)	9.5 (0.17–43)
Years working with people with PD level	8.5 (1–29)	18.2 (1–39)	7.5 (1–20)	11.1 (3–20)					14.9 (3–42)	
National					6	4	7	2		19
Regional						5	2	1		8
Local					5	1		4		10
Others								2		2
Professionals' profiles										
Nursea	2	4	4	3					13	
Physicianb	3	4	1	3					11	
Speech and language therapist	2		1	1					4	
Physiotherapist	2	1	1	2					6	
Occupational therapist		1	2	1					4	
Clinical Psychologist	1								1	
Social worker		1							1	
Training instructor			1						1	
Role in the organization										
Research					2	3				5
Direct workc					5	1	2	4		12
Managerial					3	5	6	5		19
Policy						1				1

Other: Strategic lead—service design team					1		1			2

Abbreviation: PD, Parkinson's Disease. a

Nurse: There was variability in the profile of nurses in all countries: PD specialist nurse; Specialized in Neurology; Primary Care Nurse; Nurse (Deprived population).

b

Physician: There was variability in their profile in all countries: geriatrician; consultant; Physician—Neurologist; GP (Family doctor).

c

Direct Work: usually refers to direct work with: groups; people with PD; carers.

A total of two themes and five categories emerged from the cross-national analysis and these are presented below (for additional quotes, see Supporting Information: File 2).

Towards more connected systems of support

This theme describes the existing gaps and challenges in the health and social care systems, and the fragmented communication and support in PD management perceived by both professionals and stakeholders. It also covers the benefits that multiagency partnerships could potentially bring to the care and support systems in the four countries, according to the participants.

Staff capacity and training

The stakeholders and professionals identified difficulties in the current systems of support such as an increased workload and overstretched services. The interviewees in the four participating countries discussed the increase in demand and caseload, the reduction in commissioned services and a reduced consultation time, which have all impacted the way the care and support is delivered. As a result, PwPD may call upon alternative support systems such as the family, voluntary organizations and other services to cover the care that the health system cannot provide. Most stakeholders and some professionals in some countries considered that the voluntary organizations were in a better position than the healthcare professionals and had more time to cover informational, social and emotional needs. Both the professionals and stakeholders perceived that the involvement of alternative support systems in PD management was largely dependent on each country's formal system and available funding, the changing political landscape and the individual's commitment to sustain the available support in a specific region, and also that there was a lack of a formal organizational structure and co-ordination between sectors and organizations.

It's only volunteers that work with these things, so it depends on what resources are available locally. In some places, there is a person or someone very passionate about something that becomes something big there because someone has a lot of energy to do it, and in other places it can be different. (NO-SH-003)

Some of the benefits identified by all countries from potential multiagency collaborations were the complementary roles in care and support provision. The professionals (Denmark [DK], Spain [SP], United Kingdom [UK]) and stakeholders (SP, UK, Norway [NO]) highlighted the specific advantages of collaborating with community organizations and the voluntary sector, such as organizing social activities, for example, walking groups, theatre and dance, which could provide peer support, a feeling of belonging and being part of a community, something that the clinical community cannot provide. In any case, identifying other hubs of support in the community was seen as a great opportunity to promote the PwPD's independence from the overstretched health system.

I quite often will suggest just the [name of organisation] website […] actually Parkinson's cafes give people the opportunity to come together once a month, to have a chat, to get some support. (UK-SH-008)

In addition, the professionals from all countries commented on the staff's lack of PD specialized clinical skills, from primary care, community services and health centres, which could potentially lead to clinical misjudgements. Many participants indicated the need for education to improve care and support. The participants from Norway and Denmark shared current training opportunities, for example, the Parkinson Net model in Norway, and in Denmark, passionate professionals often educate other professionals about PD symptoms and care.

Health centres have very few patients with PD, so I have actually been out teaching at several of the centres, just to give them the most basic knowledge about PD. (DK-HCP-002)

Moreover, in an attempt to foster a multiagency or more connected model of care, it was suggested that all parties could share training resources and best practices to complement each other and ensure continuous professional development. As such, all the agencies could benefit from the existing resources and expertise and avoid duplication, while addressing existing training gaps. The stakeholders and professionals from all of the countries agreed that linking up multidisciplinary and multisectoral teams might facilitate potential continuity of care, better management and knowledge mobilization, which is currently missing. Partnerships were also considered as a path to accessing specialized care that was not formally established in PD care pathways.

You don't see a social worker going with the doctor or nurse for a home visit. When, well, yes, it is important for each one to make their report, okay, but also to see the relationships a bit, right? […] the representatives of the institutions have to negotiate and reach agreements. (SP-SH-002)

Awareness of, and communication between, support systems

Health and social care professionals in all four countries acknowledged that they were not always aware of local resources and support that were available as these were constantly changing and very diverse. This made it difficult and frustrating for professionals to navigate the existing resources and to check if the services were still available in their region. An additional issue raised by the interviewees in all countries was that some people could be missing out on the support available because they choose not to be part of the local Parkinson's association.

Our association is of great importance to those who choose to and do sign up; those who choose to participate. Because some choose not to, they can be difficult to get in touch with. And they are probably the ones who need it most, right? (DK-HCP-010)

The participants in all of the countries identified that working in silos contributed significantly to the fragmentation of support and communication in PD management. In many cases, due to the bureaucracy and lack of communication between professionals and the different sectors, it has been difficult for clinicians to maintain an overview of the PwPD's history, for example, the admissions, discharges and follow-ups were not communicated between clinicians.

The community services are not necessarily told if, for example a PwPD falls, and he gets physiotherapy in a private clinic. Then he might tell the physiotherapist, but that information never goes any further. (DK-SH-004)

In discussing the potential benefits of working together, the participants in Norway, the United Kingdom and Spain shared examples of past, existing or ideal collaborations, such as when PD nurses had worked closely with consultants and local PD groups, the collaborations between health and social care services and family. According to the participants, reducing the burden of PwPD, maximizing clinical time and thus improving care might be some of the potential benefits from shared information record systems. In addition, sharing communication channels could be cost-effective and time-saving. All the participants agreed that effective communication between the levels of care and sectors could create a more connected system, with decision-making processes involving treatment management being shared between patient, carers and different professionals.

It would be a great advance, to create a truly multidisciplinary team […] we would have a fluid communication that could avoid making the patient dizzy, that the problems are not solved, that ends up in the hospital or in the ER hours and hours, using a resource that is not necessary at that time. (SP-HCP-001)

Managing the complexity and support needs of a neurodegenerative disease

This theme captures the complexity of care and support for PwPD and their families to address their increasing vulnerability and social, mental and health needs at various stages of PD. It also illustrates the potential benefits of multiagency, and across organization partnerships throughout the PD journey, such as enhanced support to PwPD and their families through their active engagement.

Timely, meaningful and broader support

Issues concerning the inconsistent support and the lack of long-term sustainability of the management of PD were discussed by the interviewees in all countries. These inconsistencies could be due to both the geographical location and the complex needs of the PD journey. For example, Norwegian health professionals highlighted that in some municipalities, PwPD did not receive personalized support due to the remote geography of the country.

Some [professionals] have a pure Parkinson nurse position and can be reached all week, from Monday to Friday, some can be reached once a week, while others can only be contacted for a few hours per week. And sadly, this differs greatly from place to place. There is no standardized plan for this. (NO-SH-004)

The participants in Denmark and Spain noted the lack of support towards people living with advanced stages of the condition, for example, cognitive decline, and end-of-life care.

I would like that there were more resources available for PwPD in the later stages. They are often forgotten. We have offers for all other stages, but in the later stages …arghh … I think something is missing. (DK-SH-005)

This postcode ‘lottery’ and lack of standardized provision of support were perceived as potentially creating health inequalities. Professionals from different disciplines and sectors in all countries acknowledged the imperative input to support PwPD in a long-term perspective, from diagnosis to the advanced stages and criticized the poor management of mental health issues.

We definitely had some patients who desperately needed psychological help […] who were really struggling with coming to terms with the diagnosis, that you know, suffered significant anxiety and depression. (UK-HCP-007)

Patient involvement and engagement

Management of the complexity and support needs along the PD journey could not be achieved without patient involvement and their continuous engagement. The lack of patient involvement in the design of services was mentioned by all countries, except the participants from Norway, where PPI in both public and voluntary organizations is well established, and where the PPI representatives are viewed as essential partners. In other countries, PwPD may not be involved in co-production of care, or treatment plans, and several of the professionals had not considered this.

Many of the participants also experienced a lack of interest in and a low attendance to some of the available support services by PwPD. A barrier to engage in certain resources according to the Norwegian and the UK stakeholders was that the support available was not always flexible and responsive to the individual's particular needs. The professionals from all countries noted that language and location were potential barriers to the attendance to some resources. Professionals also discussed the need for different types of support to appeal to PwPD in different illness stages, from diagnosis to bereavement, and preferences, for example, social groups with many elderly people may not be appealing to younger PwPD, or those who have not accepted their diagnosis may consider it stigmatizing or a forecast of future deterioration.

you can send out a letter to say the department is changing […] feedback from our patient group was what the hell is this? there was a big lesson learning there, in terms of any literature that we are going to send out to patients, we probably need to get patients to read it before we send it out! (UK-SH-011)

Building partnerships between disciplines, sectors, PwPD and FC could lead to personalized PD care and ensure continuous engagement with the decision-making processes. This approach could enable PwPD to be partners and gain sense of control over their PD (self) management.

He is an active patient, that is, you as a health professional will accompany him, you will help him to cope well with his illness, but the one who has to manage his illness is him. (SP-HCP-001)

Support to and involvement from carers

Although family carers were generally considered to be a relevant support in managing the complexity of PD, professionals in the United Kingdom and stakeholders in Spain acknowledged that carers were not always involved in designing and implementing care plans with the PwPD, and that carer engagement should start early. All countries acknowledged the need for support to the informal carers, who provide the care and may experience severe stress.

We use the carers; we don't take over the tasks that they have. If carers become exhausted and there is a need for assistance, then that's what we're working towards rather than us starting to relieve carers so that they won't get worn out. (NO-HCP-09)

A potential outcome from multiagency partnerships could be proactively offering more support for carers, rather than solely reactively. Carers often lacked the initial knowledge and skills to deal with PD but could be quite resourceful and were proactively seeking help to access information and community/formal resources. Moreover, carers could be signposted to professional services and community resources to prevent burden and stress and offer opportunities for respite time if required.

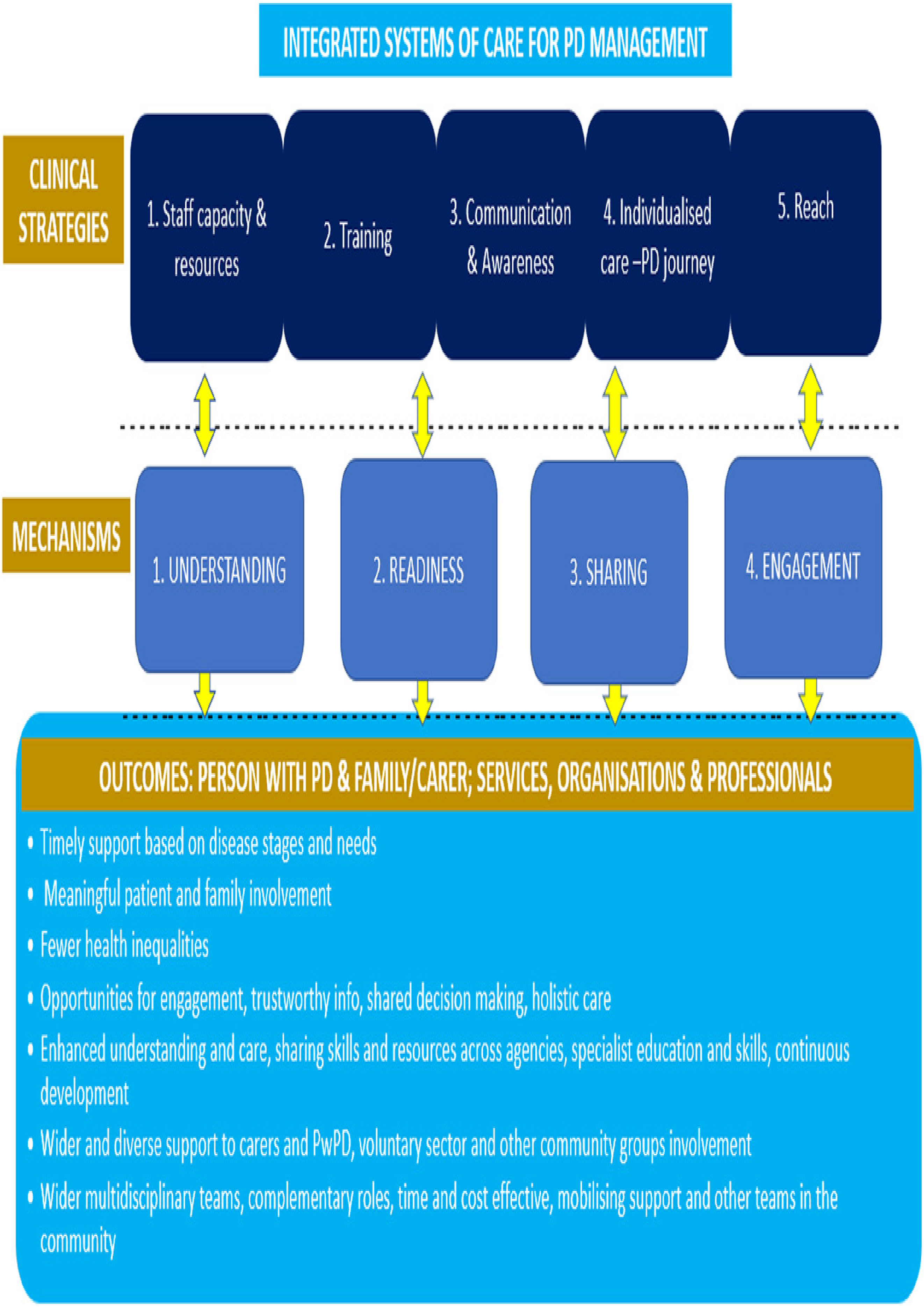
the family and carers are really active at that diagnosis point and that wasn't really featuring in our service offer …we hadn't realised that family and carers were actually the people doing all the information seeking at that moment. (UK-SH-010)

DISCUSSION

This qualitative study has shown a European understanding of how health and social care professionals and other stakeholders from different agencies and organizations can work together to enhance PD management in the community, what the existing gaps in this process are and how people with PD and family carers could benefit from these partnerships.

The main gaps in PD care identified in our study by the participants were overstretched services, lack of awareness of available resources and support, a limited trained workforce, disjointed services and fragmented communication, inconsistent and limited support, in particular, in mental health issues and advanced stages, and poor patient and carer involvement. Identifying these barriers to multiagency partnerships in PD management is an essential step in planning strategies to address them in European health systems. Recent studies[6,28–30] have also identified three of these barriers, the lack of interdisciplinary management and ongoing support, especially regarding psychological needs, and advanced stages, and the fragmentation of health and social care in other countries.

To address these gaps, our main findings from health and social care professionals and stakeholders are integrated in Figure 3, which proposes five strategies and four underpinning mechanisms that could make it easier for different organizations to work together to improve PD management in a community setting.


INTEGRATED SYSTEMS OF CARE FOR PD MANAGEMENT

CLINICAL STRATEGIES

1. Staff capacity & resources
2. Training
3. Communication & Awareness
4. Individualised care –PD journey
5. Reach

MECHANISMS

1. UNDERSTANDING
2. READINESS
3. SHARING
4. ENGAGEMENT

OUTCOMES: PERSON WITH PD & FAMILY/CARER; SERVICES, ORGANISATIONS & PROFESSIONALS

- Timely support based on disease stages and needs
- Meaningful patient and family involvement
- Fewer health inequalities
- Opportunities for engagement, trustworthy info, shared decision making, holistic care
- Enhanced understanding and care, sharing skills and resources across agencies, specialist education and skills, continuous development
- Wider and diverse support to carers and PwPD, voluntary sector and other community groups involvement
- Wider multidisciplinary teams, complementary roles, time and cost effective, mobilising support and other teams in the community

Enlarge this image.

The first strategy is to have the right *staff capacity and resources* to implement integrated systems of care for PD management. This includes the staff having sufficient *training* to obtain *specialist PD skills*, which is the second strategy. To achieve these multiagency collaborations, a macro-level formal structure to formalize partnerships and care pathways for the PD management in the community might be proposed by policymakers. This could result in shifting priorities towards individualized care and a common vision and agreed agendas. At a meso level, co-ordination is proposed that will enhance the connections between agencies, levels of care, professionals, the voluntary sector, community organizations, PwPD and family carers and help to navigate and mobilize resources to overcome the staff shortages. These connections could be achieved by an awareness of the roles and resources, specialized training, shared communication systems, complementing expertise and sharing best practice, of the creation of community hubs and identifying PD champions/navigators.

The relevance of creating formal partnerships involving all agencies, that is, the voluntary sector, the community, PWPD and family carers, in PD management has not yet been explored. However, according to the WHO and some comprehensive community-based programmes,[17,20,31–33] a multisectoral approach has previously been shown to provide benefits in addressing health problems and reducing health inequalities through sharing objectives, pooling resources and optimizing them by avoiding duplication of activities. Furthermore, a multisectoral approach could facilitate two changes: an increase in the number of healthcare professionals who specialize in PD, and community care as the major context for PD management. According to previous studies,[28,34,35] these changes are essential to improve care for PwPD, especially the most vulnerable, the elderly, to reduce unexpected hospital admissions, carer burden, costs, pressure on the medical system and to enhance the patient's experience and their quality of life. This important change in PD management, from the care delivered mainly in hospitals towards care in the community and in the patients' home, is needed in many countries to achieve a patient-centred perspective and to address health and nonhealth needs.[28,35] It is in the community context that PwPD face multiple motor and nonmotor symptoms including cognitive decline[6] and where PwPD and their family carers face the adjustment process to their new personal, familiar, social and professional roles.[5,36] Hence, it is important that all health and social care professionals involved in PD management acquire specialist training in PD and an in-depth knowledge of the role of the different disciplines involved.[28] The training delivered to multidisciplinary teams in the Dutch ParkinsonNet to increase specialization in PD is an example that has shown improvements in patient outcomes and care costs.[37]

The third strategy is effective *communication between and across* services, organizations, PwPD and their families and *awareness* of what support is available (see Figure 3). The strategy identified in our study is in line with previous international studies that have demonstrated that working with community organizations (beyond the healthcare system) is associated with better health outcomes in people with long-term conditions.[13,31] However, there is a gap in these studies as this has not been studied in PD. We propose that individual assessments of social support, from individual social networks and neighbourhoods, and participation in community organizations and the voluntary sector could also bring benefits for PwPD in terms of self-management and health outcomes. Moreover, improving communication between health and social care professionals, regarding the levels of care, community organizations, the voluntary sector and PwPD and family carers, should be a priority for policymakers to foster multisectoral collaboration and integrated systems of care for PD management.[29,31]

The fourth strategy is *individualized care* along the *PD journey* that promotes timely, meaningful and wider support. The management of PD through this model is paramount to address care fragmentation, poor interdisciplinary care and promote timely access to services and therapies.[28,38] To promote individualized care throughout the PD journey, it is essential to identify in healthcare a single point of access or a care coordinator, which is an urgent need according to PwPD[28,29] and long-term guidelines.[8] The care coordinator, or single hub, could play a leading role in the assessment process of each person, liaise and work with all health and social care services, the voluntary sector and community organizations and ensure that all referrals to any service or organization start working well for the person.[8,29]

The final strategy is to *reach to PwPD and their families* to ensure meaningful involvement and continuous engagement. We propose, from a micro-level perspective, that PwPD and their families can become valuable partners that can influence these partnerships and advocate personalized support by their continuous engagement, involvement in clinical decision-making and the management of their condition and preferred support.

In addition, it is proposed that the PwPD, and their family carers if appropriate, are involved in their needs assessment, as it has been highlighted in other long-term conditions.[8] We also propose the need to include the family carers in these assessments to identify any caring, physical and mental health needs.[8] Fostering self-management for PwPD is also paramount for a person-centred approach but also requires ensuring educational and support opportunities.[29,39]

The adoption of this model may result in positive outcomes that are relevant to services, organizations, healthcare professionals, PwPD and their family carers, as described above and shown in Figure 3. Future research should explore the implementation of a multisectoral approach for PD management in a particular context. Future development of tools that help healthcare professionals and stakeholders connect, share resources and optimize communication could also constitute a breakthrough to a more personalized, integrated and cost-effective PD care.

Limitations

Although we have found important commonalities across country findings, we also acknowledge the existence of cultural differences and the variety of health and social care systems, as well as the use of both inductive and deductive thematic analyses, which could lead to a loss of national findings. However, the wide experience of researchers who undertook all interviews, the involvement of at least two researchers in each country in all analyses, the application of the meta-ethnography approach (lines of argument synthesis) and the validation from the PPI groups have minimized this.

To our knowledge, this is the first exploratory study across four European countries engaging with a variety of participants to understand how different agencies can collaborate to enhance PD management in a community setting. This study has provided new insights and understanding that could facilitate changes across other countries with established healthcare systems and encourage a more connected system of care in PD and other long-term conditions.

CONCLUSIONS

Policymakers are called upon to create formal structures that facilitate multisectoral collaborations between healthcare, social care, community organizations, the voluntary sector and other agents to promote an integrated system of care for PD management in community settings. To address this challenge, five strategies of how different organizations can work together to enhance the management of the different needs throughout the PD journey and the optimization of the resources of the health and social care are proposed.

AUTHOR CONTRIBUTIONS

Dia Soilemezi: Formal analysis; investigation; methodology; validation; visualization; writing – original draft; writing – review &editing. **Ana Palmar-Santos**: Formal analysis; investigation; methodology; validation; visualization; writing – original draft; writing –review &editing. **M. Victoria Navarta-Sánchez**: Conceptualization; data curation; formal analysis; funding acquisition; investigation; methodology; validation; visualization; resources; writing – original draft, writing –review &editing. **Helen C. Roberts**: Conceptualization; formal analysis; funding acquisition; investigation; methodology; validation; visualization; writing – review &editing. **Azucena Pedraz-Marcos**: Formal analysis; Investigation; Methodology; Validation; Visualization; Writing –original draft; Writing –review &editing. **Anita Haahr**:

Conceptualization; data curation; formal analysis; funding acquisition; investigation; methodology; validation; visualization; resources; writing –review &editing. **Dorthe Sørensen**: Formal analysis; investigation; methodology; validation; visualization; writing – review &editing. **Line K. Bragstad**: Data curation; formal analysis; investigation; methodology; resources; validation; visualization; writing – review &editing. **Ellen G. Hjelle**: Formal analysis; investigation; methodology; validation; visualization; writing –review &editing. **Silje Bjørnsen Haavaag**: Formal analysis; investigation; methodology; validation; visualization; writing – review &editing. **Mari Carmen Portillo**: Conceptualization; data curation; formal analysis; funding acquisition; investigation; methodology; project administration; resources; validation; visualization; supervision; writing –original draft; writing –review &editing. All authors have contributed to the manuscript substantially and have agreed to the final submitted version.

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CONFLICT OF INTEREST

The authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available on request from the corresponding author. The data are not publicly available due to privacy or ethical restrictions.

ETHICS STATEMENT

This study obtained the ethical approval from the required ethics committees: University of Southampton—IRAS number: 265184; Research Ethics Committee in Hospital Universitario La Princesa number: 3995, CEIm 02/20; Norwegian Centre for Research Data reference number: 986940. Participants gave informed consent before taking part in this study.

DETAILS

Subject:	Ethnography; Support systems; Collaboration; Community; Organizations; Health care; Medical personnel; Disease management; Policy making; Caregivers; Neurodegenerative diseases; Quality of care; Mobilization; Strategic planning; Parkinson's disease; Interviews; Professional relationships; Citizen participation; Patients; Qualitative analysis; Public involvement; Stakeholders; Chronic illnesses; Social care; Movement disorders; Partnerships; Individualized; Health professional-Patient communication; Patient participation; Professionals; Nursing; Qualitative research
Business indexing term:	Subject: Strategic planning Professional relationships Professionals Stakeholders

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‘To me, it's ones and zeros, but in reality that one is death’: A qualitative study exploring researchers' experience of involving and engaging seldom-heard communities in big data research

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ABSTRACT (ENGLISH)

Background

Big data research requires public support. It has been argued that this can be achieved by public involvement and engagement to ensure that public views are at the centre of research projects. Researchers should aim to include diverse communities, including seldom-heard voices, to ensure that a range of voices are heard and that research is meaningful to them.

Objective

We explored how researchers involve and engage seldom-heard communities around big data research.

Methods

This is a qualitative study. Researchers who had experience of involving or engaging seldom-heard communities in big data research were recruited. They were based in England (n=5), Scotland (n=4), Belgium (n=2) and Canada (n=1). Twelve semistructured interviews were conducted on Zoom. All interviews were audio-recorded and transcribed, and we used reflexive thematic analysis to analyse participants' experiences.

Results

The analysis highlighted the complexity of involving and engaging seldom-heard communities around big data research. Four themes were developed to represent participants' experiences: (1) abstraction and complexity of big data, (2) one size does not fit all, (3) working in partnership and (4) empowering the public contribution.

Conclusion

The study offers researchers a better understanding of how to involve and engage seldom-heard communities in a meaningful way around big data research. There is no one right approach, with involvement and engagement activities required to be project-specific and dependent on the public contributors, researchers' needs, resources and time available.

Patient and Public Involvement

Two public contributors are authors of the paper and they were involved in the study design, analysis and writing.

FULL TEXT

INTRODUCTION

Patient and public involvement and engagement (PPIE) has become embedded in health research and within the NHS,[1] and is used in healthcare services[2] to put the public perspective at the centre of the discussion[3] and improve professionalism among medical practitioners.[4] It helps to align priorities shared by researchers and the public[5] and it helps researchers understand the lived experience of patients and the public.[6] There is also an ethical argument that those who pay (taxpayers) should have a say on how their services and research are shaped.[7] We follow the National Institute for Health and Care Research (NIHR) definition of public involvement and engagement.[8] Public involvement in research means that work is ‘being carried out “with” or “by” members of the public rather than “to,” “about” or “for” them’. We use the term ‘public contributor’ to describe this role. Conversely, public engagement stands for activities ‘where information and knowledge about research is provided and disseminated’.

Big data

There are multiple definitions of big data in the literature.[9] In this paper, we define big data research as reusing routinely collected medical data for research purposes. This can happen by linking large medical data sets from various sources. When initially collecting medical data, the public (or the researcher) might not be aware that their data may be later reused for research. Many big data research studies use opt-out consent, where patients need to inform someone, usually their medical provider, that they do not want their medical data to be reused for research. Public support is needed for these projects to be able to take place,[10] and a systematic review has shown that the public generally supports the reuse of their medical data.[11] However, they can be concerned that their data might be misused, for example, sold to private companies.[12] PPIE can assist in alleviating these concerns.[13] Hill et al.[14] found that talking about and explaining the research process around big data improved their study participants' support in reusing their medical data. Public contributors can also contribute to the decision process on who can access medical data for research purposes, thus ensuring that a social licence exists.[15] Social licence is more than meeting legal requirements and requires public trust that researchers will conduct their work ethically.[13] Poor governance can lead to a deterioration of the social licence.[16]

Seldom-heard communities

In addition to the ‘usual’ public, it is important to capture the voices of groups in our communities who are less frequently heard. Successful PPIE requires the inclusion of seldom-heard communities,[5,17,18] and researchers should aim to include them,[19] but how to do it in a meaningful way remains challenging.[20–22] Such communities are often easy to ignore, but not including them can make research findings ungeneralizable to all parts of society and miss the nuances of experiences specific to those groups[23] and will not provide solutions for all communities.[24] PPIE should be inclusive of and accessible to everyone.[5] Not including seldom-heard voices can reflect the power structures at play and perpetuate health inequalities. This is important as these communities might experience poorer social and health outcomes. For example, the Covid-19 pandemic disproportionately affected people from ethnic minorities.[25]

The terminology and definitions in this area are contested. Some of the terms used include hard-to-reach,[23,26] seldom-heard,[27,28] seldom-listened,[29] peripheral voices,[30] marginalized[31,32] and underserved.[33] The key characteristic of these definitions is that these communities are less included in research than other groups in mainstream society.

Within the UK legal context, the Equality Act 2010 uses the term 'protected characteristics'. These are age, disability, gender reassignment, marriage or civil partnership, pregnancy and maternity, race, religion or belief, sex and sexual orientation. The Act provides antidiscrimination laws and embeds requirements for diversity and inclusion for public bodies but is not always directly applied to research. However, it can be influential in how researchers approach diversity in their work.[18] We will use 'seldom-heard' as this shifts the responsibility for inclusion to researchers rather than blaming the public, as implied by the 'hard-to-reach' wording. Their inclusion (or a lack of it) is not a fault of these communities.[33] When presenting the results, we kept the original terms used by participants when quoting them. However, we recognize that use of any terms might not necessarily represent how these communities would like to be described.

Research aim

Despite understanding the importance of PPIE, there is limited knowledge of how this can be effectively facilitated in big data research.[34] A previously published system logic model identifying key elements of PPIE in big data research recognized the inclusion of seldom-heard communities as a key component,[34] and therefore, there is a need to understand how to ensure all voices are included.

This paper explores researchers' experiences of involving and engaging seldom-heard communities in big data research.

METHODSTheoretical position

This study adopts social constructionism as its theoretical lens when understanding and analysing data.[35] We believe that multiple realities and perspectives exist among researchers. These are subjective and socially constructed and thus depend on participants' cultural, political and historical backgrounds. Researchers (and thus their work) are shaped by their relationships with public contributors. From the social constructionism perspective, the dynamics of social interactions are essential to understand how new knowledge is achieved.[36] Thus, in our analysis, we focused on the processes around PPIE rather than its structures.

Social constructionism can be used to justify a more collaborative form of inquiry.[35] This can be achieved by conducting research together with the public contributors. Collaborative work can be seen among our participants who involve the public in their work but also in our project, as we involved two public contributors as co-researchers.

Participants and data collection

Alongside big data researchers, we included facilitators of PPIE activities in big data projects. Facilitators (some of whom might be qualified researchers) are in charge of the overall organization of the PPIE progress; they co-ordinate, organize and facilitate activities and act as intermediaries between researchers and public contributors.[37] They often are recruited at research institutions to support specific big data research projects. Throughout the paper, for clarity, we will refer to both groups as researchers. All participants had to have an experience of involving or engaging seldom-heard communities or aiming to reach them.

We recruited through Twitter, bulletins and established networks within big data research such as Health Data Research UK. Interested participants contacted the author for further details and to register their interests. Interviews were conducted on Zoom between March and June 2022. Interviews were later transcribed and anonymized, with all participants assigned pseudonyms.

A semistructured interview guide was developed to elicit participant experiences of PPIE with seldom-heard communities. We also included an opportunity for them to speak about communities that they planned to reach or tried to engage but were unsuccessful. After the first interview, co-authors met to reflect on the topic guide. One follow-up question on what participants perceived as a seldom-heard community was added to the topic guide.

Only limited demographics were collected from participants to protect their anonymity. Twelve participants took part in the study. We reached data saturation when no new themes appeared in our analysis.[38] Participants were based in England (n = 5), Scotland (n = 4), Belgium (n = 2) and Canada (n = 1). The majority were women (n = 11) and there was one man. Their experience of research and PPIE ranged from two and a half years to 20 years, with an average of 9 years. We also asked them to describe themselves as researchers (n = 6) or facilitators (n = 9), although they could have chosen both options. Six participants were from an ethnic minority background.

Data analysis

We conducted a reflective inductive thematic analysis.[39–41] This method allowed us to identify patterns across all interviews systematically. Thus, we unpacked the realities experienced by researchers. We used both semantic and latent coding. Semantic coding shows more explicit patterns within the data and stays as close as possible to what participants said. This allowed us to share specific practical examples of PPIE strategies. Latent coding provided more implicit and interpretive reflection on the data. Initially, one interview was coded jointly by three authors (P. T., S. A., N. T.) in Word. Then, the author (P. T.), an experienced qualitative researcher, inductively coded the remaining interviews, supported by NVivo 12. We met as a team on multiple occasions to discuss the data analysis and develop and refine further themes.

Public contributors can be meaningfully involved in qualitative analysis[42] and trained to conduct reflexive thematic analysis.[43] Two authors (S. A. and N. T.) who are public contributors received training in reflexive thematic analysis (focusing on being reflective, coding process and refining themes). They were involved in the designing the study, initial coding and developing and refining the themes. They are both experienced public contributors and S. A. also acts as the Data Ambassador for Care and Health Informatics theme within the Applied Research Collaboration North West Coast. This role involved raising awareness and knowledge about big data research. Two authors (S. A. and N. T.) also took part in an exercise reflecting on how their backgrounds influenced what they perceived in the data and what they brought to the analysis.

The research-active authors also reflected on their academic backgrounds (P. T., K. F., S. E. R. and L. F.). Research team consisted of qualitative researchers with experience of involving and engaging the public, and those who conducted research in big data. These different perspectives allowed us to bring distinct views to the data analysis and furthered our understanding of the experiences of our participants.

FINDINGS

We present four themes that explore how researchers involved and engaged seldom-heard communities in big data research: (1) abstraction and complexity of big data, (2) one size does not fit all, (3) working in partnership and (4) empowering the public contribution. All themes appeared throughout all interviews, which provides an indication that these experiences were commonly shared among participants (even if participants were based in different countries). We have provided additional quotes in Supporting Information: Appendix 1 that offer further examples of how the participants involved and engaged seldom-heard communities around big data research.

Abstraction and complexity of big data

Big data can be an abstract and difficult topic to explain to the public. Participants said that conversations about big data include technical, specialist's vocabulary, jargon, references to legislation and regulations. Researchers found it challenging to discuss the complexity of this kind of research with public contributors in lay terms:

Big data is a really complex environment to navigate both in terms of the research, but also in terms of like the regulatory aspects and legislative aspects. (Sophia)

Sometimes, the difficulty in explaining big data research impacted on participants' experience of involving the public. Public contributors can have a role in advising (or deciding) if researchers may access routinely collected health data for research purposes. Here, the public contribute to the governance groups of these initiatives. Researchers who worked with these groups found it hard to explain to the public the purpose of big data research. They struggled to contextualize the concept of big data to the public if it did not directly refer to the public contributors' health condition or a topic that might interest them. The following extracts illustrate that challenge as the participant refers to bringing public contributors to support big data infrastructure:

Project (…) was just looking at the infrastructures of big data. It was really challenging to actually put that into a context that was relevant to members of the public; they kind of said ‘well we don't even know if you want us to be involved, we don't really see how we can be because this is all to do with linking up datasets with each other and it's all very technical, and it's not really anything to do with our living experience as patients or as members of the public’. So that was that was quite a hard project actually to think about. (Sienna)

It is not only public contributors who can be confused by big data jargon. Some participants who were not data researchers said that their familiarity with the topic was more akin to the public contributors rather than data researchers they worked with on the project. They might feel uncomfortable asking questions or requesting clarification. The public contributors often were more confident in asking these kinds of questions. This was seen as a very positive element of PPIE by a participant:

I'm sometimes really pleased when [public contributors] ask questions. Because I'm like oh, good, I don't know if I could have asked that, but so I'm really pleased that you did. I probably should have known that, but I don't, so I'm glad you asked it. (Robyn)

Participants felt that promoting the benefits of big data research, being transparent in how data are used and building trust with the public would ensure that some negative media stories around big data research could be counteracted. They believed that overall, the general public would be supportive of data sharing to improve healthcare. They recognized the need for effective communication between researchers and the public. In individual projects, they suggested training and supporting the public contributors around big data research but described it as a slow and time-consuming process.

One of the things that we really do is kind of work with our staff to make sure that they are able to explain it in kind of like plain English. If we were to have a session about something like trusted research environments, which can be kind of like a technical. Then we would work with staff to actually plan the presentations (…) to make sure that the language is right, we also hold drop-in sessions once a month so that members of the public that we work with can come in and say ‘I have a question’. (…) And so we bring in some of our more technical staff because I have no technical knowledge myself. (Harriet)

Participants spoke about how communication must continue outside the research projects and involve the broader community. The public contributors involved in big data research are essential to helping further engagement with their communities. As they become more familiar with big data research, their knowledge can be utilized to engage with the general public and raise awareness of big data research. They can help explain what big data research is about, its benefits and how it works. Here, a participant speaks about explaining in lay terms a technical term related to data:

When it comes to data and infrastructure and things, it can be very complex. There's lots of big words like pseudonymisation [laughing] and things like this, so we worked with the public members to create this animation, which gives a snapshot of what the project's like and it's an accessible snapshot. (Robyn)

This theme shows that talking about big data can be complex and challenging. However, there was an agreement that PPIE around big data research takes the researcher away from numbers and allows them to bring a human face to the data. The following excerpt explains this:

I love doing this type of analysis of, you know, hitting the buttons and seeing the graphs come up and seeing results. It's really exciting, but you miss that contact with people. And having that PPI group, there was a really good way for me to touch base and think about what the numbers meant. And think about the stories behind some of the data. And connect it to people's lived experience and I think that's really important. (…) To me, it's ones and zeros, but in reality that one is death. So it's really important to have that in front of your mind, and I think that brings it home when you've got a group of people in front of you who are really interested in what you're doing and to whom it could potentially make a difference. (Zoe)

One size does not fit all

This theme elicits the need for researchers to be flexible and often innovative when involving public contributors in big data research. Participants did not have one prescription on how to successfully work with the public contributors.

How PPIE looked in the participants' work differed based on the project needs, public interest or experiences. Public contributors can be involved in different roles within projects around big data research. These included contributing to the review of the data access process and as co-investigators or members of advisory groups for specific projects. The following quote shows how public contributors can assist with decisions over whether and how researchers can access routinely collected medical data for research purposes.

That's a group of around eight members of the public who we meet with on a quarterly basis to get their views on our kind of engagement plans (…) and also to get them to become more part of our project approval process is something they've been really keen to do, so we're looking at our kind of review process. Researchers who want access to routinely collected health and social care data puts their applications in and it goes through a rigorous, multistage approval process and one of those that we're looking to do is to have the public voice within that so their vote, their part of it would be an assessment of the public value of the projects that come in. (Alex)

Participants said that public contributors can have a much more active role and co-share responsibilities with researchers:

We have two co-leads. One of them is myself and but the other one is a member of the public, so that from the very beginning, I am working very closely with [the public contributor] so that we can kind of shape this programme together, making sure that the public views are fed in right from the very beginning and as part of that we've also got a leadership (…) and so in this leadership team, it's half public contributor, patient-public contributors and the other half would be kind of like professionals such as myself. (Harriet)

How to work with each community might depend on their needs. Many participants spoke about the need to understand the specific community that they were planning to work with. Here, a participant suggests a pre-engagement engagement to understand what PPIE should look like:

It's just really interesting about doing that pre-work to set up the scope and the scale of the engagement work and then to set up the environment that would be the safe as possible, so it's almost like a pre-engagement engagement where you're really setting up the safe environment to allow for good public engagement to happen for diverse members. (Victoria)

Who represents seldom-heard communities differed among participants. Participants often spoke about aiming to be reflective of the community. However, they recognized that it was not always possible (or feasible) to reach everyone who might potentially contribute. They admitted that because of their recruitment methods, limited resources or time, the public contributors who were generally involved often represented a limited range of demographics. Each community is different and might require different PPIE strategies. They argued that the recruitment should be specifically tailored to the group they wanted to reach. The communities that were most often involved in PPIE were generally white and elderly. The seldom-heard communities they wanted to involve included ethnic minorities, people experiencing homelessness, traveller communities or different age groups (especially younger people). However, they also wanted to reach people with particular health conditions or improve male representation. The following quote illustrates how participants perceived their role in encouraging diversity:

We do try to reach out to seldom-heard groups. We are currently undertaking an audit of our group to see how, where we're lacking, 'cause I suppose within the patient and public involvement there tends to be a certain type of person who volunteers and has got the time. So tend to be retired, tend to be white more often than not, and so we are keen to widen our demographic (…) we're not just interested in ethnicity (…) it tends to be quite a lot of women as well that volunteer, so you know, increasing, men, also increasing our younger population. (August)

Working in partnership

PPIE is not conducted in a silo. The participants worked with others (organizations, charities, public services and public contributors) with the aim of being inclusive and to reach more diverse communities, especially around big data research. This theme explores these different actors' roles in successful PPIE.

These partnerships have the potential to fill the gaps in researchers' understanding of local communities. Some participants recognized that researchers themselves could be a hard-to-reach group. Meetings can be held during working hours or be otherwise inaccessible to public contributors. Others recognized that the diversity of their teams is important and might reflect how well they involve and engage communities.

I think while we don't have as much diversity as we could in our staff, it's harder for us to communicate or share those messages or understand the groups that we're trying to reach. (Arabella)

Charities and organizations already provide existing links with the community and offer that bridge for researchers to reach the seldom-heard groups. They can assist with recruitment and engagement strategies. However, there is a risk that a researcher will not necessarily improve the diversity of their group but rather take over the demographic composition of the group they engaged with, as this participant explains:

So it was mainly about because I was kind of piggybacking on a charity, on several charities groups. It was down to who they had picked up and they were already actually meeting via Zoom this charity, so I kind of inherited their diversity or degree of diversity. (Zoe)

However, as much as these partnerships can be helpful, establishing them is not easy. It can be time-consuming to build that trust with the charity, and participants recognized that this needs to be an ongoing relationship that should benefit both parties.

Some participants also said that that relationship could be confusing to the potential public contributors if there is more than one research team working on that project (and thus trying to involve them). The following extract shows how one of the participants struggled to get some patient groups involved because they already had been working with other researchers:

I contacted several [patient groups] in [the city] to see if they would be interested in doing some PPI workshops with them or telling them a bit more about the research we're doing. (…) They didn't necessarily know that they it was the right thing for them at the time, but also they'd had so many researchers getting in contact with them that it's they said it's just really difficult for us to choose who we work with and if they've already got a relationship with somebody else. Then they may choose to work with them obviously instead. (Sienna)

Researchers can act as facilitators of PPIE or bring in trained experts (who might not necessarily be familiar with big data research). The facilitators' role is to act as this connecting bridge during work, an intermediary between researchers and the public contributors.

What we are trying to do is bring these people on board and explain to us what it is, and we try to turn it into more lay language and sometimes with [public contributors], engage them to have a conversation so that they can actually challenge the experts rather than us doing it. So we are more of an inbetweener in that sense. (Kimberly)

PPIE is also about involving individual public contributors. Participants often spoke about how interested and passionate public contributors can become about their involvement. These partnerships require working together and respecting each other. Some participants spoke highly of public contributors they worked with:

And one thing that I think that is often forgotten is about [public] members is that they are just, they're not just patients or they're not just a member of the public. These are very talented, very skilled people. You know they've got their own life skills. You know they've got their own careers. They've got all of the skills and knowledge from that, and I think it's great that they want to volunteer with us and help share some of that. (Robyn)

Only when truly working in partnership with public contributors can it lead to their empowerment. This is the focus of the next theme.

Empowering the public contribution

Participants felt that for involvement to be successful, there must be a power balance between researchers and the public contributors. Empowerment gives public contributors the ability to contribute to the involvement process fully. This can be achieved through ongoing support and ensuring that they become more familiar with big data research or projects that they are involved in. As the following qoute illustrates, this is a continuing process.

Giving a sort of chance for people to ask questions, which was the nice thing about that project is that it wasn't a one-off, people could go away, look up something for themselves and then they could come back and be like what's this and they'd post a link and then we'd come back and answer those questions. So it was quite a nice kind of two-way in that sense. (Drew)

Most participants felt that public contributors need to be supported at each stage of the involvement process but also recognized that this can be time-consuming and requires additional work. Some suggested an open-door policy where public contributors could reach researchers anytime and thus also feel like a part of the team. WhatsApp groups for public contributors can be a safe place to discuss the project further. Public contributors should receive training or induction both around the project and PPIE (especially if they are involved in a research project for the first time). One of the techniques that supported the public in understanding the jargon around big data research was a ‘live dictionary’, which could be updated as people asked questions throughout the lifetime of the research project.

But one of the things that we've created is an ongoing glossary. And if there's any words or phrases that the [public] members don't understand, it's a case of pop it into that glossary, and someone will answer it for them. (Robyn)

However, participants recognized that not all training can be equally helpful and that some institutional resources were more bureaucratic and could potentially discourage people from being involved. This is illustrated by the following quote talking about the focus on training offered by the academic institution to new public contributors involved in the research:

[The training] is quite formal and it's about like the whole university obviously it's not about big data, it's not really keyed towards seldom like heard groups or different types of groups, so I think there's other types of training that could still be useful for people, even if it's just, you know, stories of being involved that are from people who are more like them. So I think it could be a little bit of a little bit tailored, and some of it's very dry if I'm honest. (Zoe)

After receiving all this training and support, some participants felt that there is a danger that the public contributors start offering more of an expert view rather than a lay person perspective. There is a fine balance between understanding the project enough to be able to provide a nuanced contribution and where public contributors become what one can describe as ‘usual suspects’ of people who keep getting involved and thus become more like professionals. One participant spoke of a successful approach to dealing with this challenge:

it is a really fine line between building their knowledge to get involved and becoming an expert in that and kind of losing that public perspective (…) to kind of help with that; we do also have members of the public in a role for only specific amount of time. So, for example, now [advisory board]. They're only there three years, and then we kind of refresh the board, so with that, we're constantly bringing in that kind of like newer public perspective as well. (Harriet)

Empowerment must be felt in practice and involvement needs to be genuine. Public contributors must feel that they make a difference. In the ‘one size does not fit all’ theme, a researcher spoke about the public contributors' panel assessing if researchers can access medical data for research purposes. The participant described how the public contributors perceived this and how it could be expanded for more empowerment:

‘Do you agree with our decisions over whether these were approved or not?’ And in the main, they aligned with what the decisions had been, but on a couple of occasions, they were like ‘we don't see the public value in doing this. It's not well explained’, so is either it wasn't when explained or the public value wasn't there, and so that going more of a point of challenge for them and made it quite clear that they wanted to be part of the genuine process of review. (Alex)

Participants pointed out that only when there is a real sense of empowerment can public contributors' involvement impact positively on the research projects. There are multiple ways by which public contributors can shape projects. Participants named the following contributions: ensuring that the research questions address the public interest, co-analysing study results, advising if researchers' ideas and thoughts are on the right track (e.g., appropriate wording used or right engagement strategy put in place) and public contributors doing sense-checking and contributing to potential engagement strategies with the broader public. The following quote shows the variety of involvement and its impact:

Extremely impactful, (…), it's actually led to changes in the direction of our work, but in cases where that hasn't necessarily happened, that they've been more supportive of what we're kind of thinking and it has changed the way that some are kind of like thinking about the topic of public trust and public confidence, for example, and we only ever used to think like the wording that we would use as an organisation was we need to earn public trust. We need to build public trust but then through the [advisory board] through exploring that a bit more, we've kind of changed our way of thinking, so it's more about demonstrating trustworthiness in the use of data and building public confidence. (Harriet)

This theme has shown that public involvement should not be an afterthought and needs to be a genuine (but often time- and resource-consuming) process that can have a significant impact on researchers' work. This can be especially seen in the following extract:

It is difficult to do really well, and it takes a lot of time and a lot of resources, and I think people underestimate that. I also think there's a culture towards PPI as a tick box. (Penelope)

DISCUSSION

Our findings have shown that talking about big data ‘with’ (rather than ‘to’) public contributors can be challenging, but that PPIE can be meaningful for both researchers and public contributors. The findings elicited how researchers and their research can benefit from involving and engaging seldom-heard communities. Table 1 summarizes the key recommendations. This adds to the previous literature on meaningfully including a diverse range of communities[44] and is relevant to other areas of health and social care research. PPIE requires time and resources,[45] and not all communities are often equally involved.[46] However, our participants have shown that inclusion around big data research (because of the complexity of the topic) takes additional time and resources to succeed (even in contrast to other health research). This can be seen in extra activities such as a ‘pre-engagement engagement’, which was suggested as a baseline for successful working with the community. Our findings challenge the perspectives of some researchers who believe that public contributors rarely care about or can understand big data research and thus are not able to be involved in decisions around whether medical data can be reused for research.[47] Involving and engaging seldom-heard communities in big data might be more challenging than in other forms of health research but it is important as big data research offers an opportunity to reduce health disparities.[48] Without seldom-heard voice input, this might not happen.

Table 1 Key recommendations on involving and engaging seldom-heard communities around big data research.

•1.
Provide information in lay language and, where not possible, explain in simple English. Ensure that these explanations are available at any point to the public contributor (e.g., through an online dictionary).

•2.
Rotate public contributors on a ‘big data panel’ every 3 years to bring in new ideas and lay perspectives.

•3.
Reach out to new communities for at least 50% of the new attendees, potentially using charitable/partner organizations to help.

•4.
Identify relevant seldom-heard communities for each project.

•5.
Consider strategies to add additional diversity on multiple characteristics (e.g., LGBTQ+ and ethnic minority, or disability).

•6.
Adequate and ongoing training/support for PAs should be provided to empower them so that they can truly contribute.

The findings confirm that defining a group as a seldom-heard group is context-specific.[33,49] The participants named numerous types of seldom-heard communities involved and engaged within the context of their work. Researchers should reflect on who would be the most seldom-heard group within the context of their study and recognize that this might include more than one community. The concept of superdiversity[50,51] could provide researchers with further guidance on moving away from looking at a single characteristic (e.g., ethnicity) of the community and focusing instead on diversity within diversity. This would ensure that the needs of communities within communities are considered.

Researchers need to take time to plan PPIE well as they design their projects. NIHR guidance[33,52] recognizes this and recommends working with communities on a long-term basis. Our findings have shown the importance of building and maintaining relationships with organizations, especially charities. This confirms previous research that shows that links to the third sector are crucial in building trust.[53–55] They often act as gatekeepers but also have the potential to act as a partner. There is, however, a risk that researchers would not reach many communities as they might be limited to the partner organization's level of diversity.

There is a growing trend to establish a pool of volunteers interested in participating in PPI activities.[56] This approach might appeal to those who have time, resources and feel comfortable with working with institutions. However, this risks public contributors becoming ‘usual suspects’ of people who are involved regularly and thus not providing new contributions. There is the danger that they will become more expert than researchers themselves, thus no longer providing lay experiences and views in the project. There remains a contentious issue: how to strike a balance between public contributors being capable of contributing fully but also retaining a lay perspective.[57] One of our participants suggested the need to change public contributors on advisory boards every 3 years. This offers a solution to deal with the challenge of ‘usual suspects’ and brings a fresh public perspective but adds more work on the part of the researchers to recruit, provide training and support new public contributors on the project. The other option is to sense-check any work with the broader public.

Researchers should also ensure that any involvement is not tokenistic and enables power-sharing between researchers and the public contributors.[21] There is no one ‘right’ way to do it, and the approach depends on the project's needs (or resources) and the public contributors' interests. However, their interests should not be confused with their understanding of the topic, and researchers should provide training to improve public contributors' knowledge, thus facilitating their ability to contribute. This genuine empowerment was seen as crucial among our participants when discussing big data research with public contributors. Although not mentioned by our participants, some public contributors, for example, coming from Indigenous communities, might also require researchers to respect their values to feel truly empowered.[58]

STUDY LIMITATIONS

The study participants came from diverse communities, for example, various ethnic minority backgrounds. However, we did not record if they are a part of other seldom-heard communities, for example, LGBTQ+ or people living with disabilities. We only explored the perspectives of the researchers, and there is a possibility that the public contributors (including those coming from seldom-heard communities) would have a different view on their PPIE activities around big data research. As big data is a fast developing and diverse research area, new ways of involving and engaging will emerge, so future research should further explore how researchers involve and engage public contributors and how concepts of super diversity could be utilized.

CONCLUSION

Our study explored how researchers involve and engage public contributors (especially seldom-heard communities) in a meaningful way in big data research. The findings highlight that there is no one right approach to doing PPIE and that PPIE strategies are project-specific and depend on the public contributors, researchers' needs, resources and time available. We encourage others to reflect on their involvement strategies and hope that these results will support researchers who want to involve more seldom-heard communities in complex research topics such as big data.

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CONFLICT OF INTEREST STATEMENT

The authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

Anonymized data are available upon reasonable request.

ETHICS STATEMENT

We received ethical approval to conduct this study from the ethics committee at University of Liverpool under the number 10063.

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Accompanying patients in clinical oncology teams: Reported activities and perceived effects

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ABSTRACT (ENGLISH)

Introduction

Since 2018, four establishments in Quebec, Canada, have decided to implement the PAROLE-Onco programme, which introduced accompanying patients (APs) in healthcare teams to improve the experience of cancer patients. APs are patient advisors who have had a cancer treatment experience and who conduct consultations to complement the service offered by providing emotional, informational and educational support to patients undergoing treatments (e.g., radiotherapy, chemotherapy, surgery), mostly for breast cancer. We aimed to explore the evolution of APs' perspectives regarding their activities within the clinical oncology teams as well as the perceived effects of their intervention with patients, the clinical team and themselves.

Methods

A qualitative study based on semistructured interviews and focus groups was conducted with APs at the beginning of their intervention (T1) and 2 years afterwards (T2). The themes discussed were APs' activities and the perceived effects of their interventions on themselves, on the patients and on the clinical team.

Results

In total, 20 APs were interviewed. In T2, APs' activities shifted from listening and sharing experiences to empowering patients by helping them become partners in their care and felt generally more integrated into the clinical team. APs help patients feel understood and supported, alleviate stress and become partners in the care they receive. They also alleviate the clinical team's workload by offering a complementary service through emotional support, which, according to them, helps patients feel calmer and more prepared for their appointments with healthcare professionals. They communicate additional information about their patients' health journey, which makes the appointment more efficient for healthcare professionals. When APs accompany patients, they feel as if they can make a difference in patients' lives. Their activities are perceived by some as an opportunity to give back but also as a way of giving meaning to their own experience, in turn serving as a learning experience.

Conclusion

By mobilizing their experiential knowledge, APs provide emotional, informational, cognitive and navigational support, which allows patients to be more empowered in their care and which complements professionals' scientific knowledge, thereby helping to refine their sensitivity to the patients' experiences.

Patient or Public Contribution

Two patient–researchers have contributed to the study design, the conduct of the study, the data analysis and interpretation, as well as in the preparation and writing of this manuscript.

FULL TEXT

INTRODUCTION

It is estimated that in 2022, 60,000 Quebecers were diagnosed with cancer, which represents 158 new cases per day.[1] This number has been on the rise for several years and is expected to continue rising in the coming years due to testing delays and backlogs following the pandemic.[2] Cancer is the country's leading cause of death, and Quebec is one of the provinces in Canada with the highest incidence and prevalence of cancer.[3,4] In this context, cancer prevention and treatment are a public health priority. In response, Quebec has a cancer directorate within the Ministry of Health and Social Services that has adopted multiple measures to reduce the incidence and prevalence of cancer, but also to improve the quality, safety and experience of care and services. The care and service partnership[5] constitutes one way of achieving these goals by recognizing patients' experiential knowledge, status as full members of the care team and capacity for self-determination to make decisions about themselves based on their needs and values.[6]

Moreover, the assessment of cancer patients' experience highlighted that emotional support was the most lacking aspect among the six areas of patient experience assessed in health and social service organizations in Quebec and across Canada.[7] This need is all the more significant in the context of a pandemic where patients expect and hope to receive emotional support and benevolent accompaniment. In oncology, peer support has usually been provided by ‘patient navigators’ comprised of nurses, social workers, educators, as well as former patients.[8] By helping patients access healthcare, patient navigators have facilitated and hence accelerated diagnosis and treatment journeys. Patients have benefited from these programmes as it was reported they participated in improving their health by, for instance, increasing adherence to treatment, bringing comfort and guiding them through the healthcare system.[9,10] This could be considered patient-centred care, where patients' needs and preferences are integrated into the delivery of care, moving away from medical paternalism.[11]

However, the care and service partnership goes beyond patient-centered care and can also be exercised at the clinical level by introducing accompanying patients (APs) into the clinical teams to meet patients' need for emotional support.[5] APs are patient advisors who have acquired specific experiential knowledge related to living with cancer, using services and interacting with healthcare professionals.

They are, therefore, in an optimal position to provide a distinct and unique touch to new patients' support by helping them, for instance, navigate, understand and eventually accept their health situation. APs can also accompany patients to facilitate their transition from acute care to front-line teams and community cancer teams.[12] They can improve patients' quality of life by promoting healthy lifestyle habits and reducing symptoms of anxiety and depression[13] and have positive impacts on healthcare professionals (e.g., work satisfaction, empathy), managers, and decision-makers (e.g., to better take into account the patients' experience) and the APs themselves (e.g., finalize their recovery).[14]

The PAROLE-Onco programme aimed to integrate APs into the clinical teams of four different healthcare establishments in Quebec, Canada.[15] Selected APs were trained and coached to intervene with patients,[15] while giving them space to innovate in their own ways to accompany patients based on their experiential knowledge. Since 2019, healthcare professionals have introduced, during medical appointments with patients, APs accompanying services as an additional resource, and patients were free to accept or refuse such a resource. Research coordinators or clinical staff members monitored all procedures and collected essential clinical data on patients who had consented to participate in an anonymous and confidential manner to match them with an AP with a similar profile. Patients then made appointments with their AP according to their needs.

To date, the perspective of APs directly involved at a clinical level has been poorly documented. We aim to assess the evolution of APs' perspectives regarding their activities over time when APs and the perceived effects of their intervention on themselves, on the patients and on the clinical team.

METHODS

Data were collected on two separate occasions, at the beginning of the PAROLE-Onco programme, where APs started APs (T1), and 2 years later (T2).

Settings

Table 1 presents the four establishments that were included in this study: the Centre hospitalier de l'Université de Montréal (E1), the Centre Hospitalier Universitaire de Québec-Université Laval (E2), the Centre intégré universitaire de santé et de services sociaux (CIUSSS) de l'Est-de-l'Île-de-Montréal (E3) and the CIUSSS de la Mauricie-et-du-Centre-du-Québec (E4). Each establishment recruited its own APs (29 in total), and one site (E3) set up monthly meetings including a doctor and a psychologist to better accompany APs. Some APs did not have the opportunity to accompany patients since they were involved in the preparation phase before the intervention began. Therefore, they were not included in the data collection. The programmes in which APs were implemented include two in breast cancer (E1 and E4), one in breast oncogenetics (E2) and one in breast and gynaecologic cancers (E3).

Table 1 Establishments' characteristics

Establishment	E1	E2	E3	E4
Programme	Breast cancer	Breast oncogenetics	Breast and gynaecologic cancer	Breast cancer
Number of APs in total in T1	5	4	5	3
Number of APs in total in T2	9	2	14	1

Abbreviation: AP, accompanying patient.

Data collection

Data were collected via semistructured interviews and focus group discussions. All APs from the four establishments were invited to participate in T1 and T2. Participants were contacted by telephone or email to participate and to sign electronically the consent form approved by the Research Ethics Committee. No compensation was offered. All participants consented to partake in the research and be recorded. Due to the context of the COVID-19 pandemic, the interviews were conducted either by telephone or videoconference, and the focus group discussions were carried out by videoconference. The questions in T1 (Supporting Information) aimed to identify, among other information, the roles of APs and the effects of their interventions and were co-created and pilot-tested with two patient–researchers (patients included in the research team; M.-A. C. and M. D.). T1 data collection events were realized 4 months after APs were first introduced in the four establishments. Two years later (T2), the data collection aimed to assess the change in the APs' perspective regarding their roles and the effects of their interventions by presenting the T1 results. APs discussed how elements have changed since the new APs joined the team or whether new elements have emerged. Therefore, no interview guide was used in T2. Transcripts of the interviews and focus group discussions were prepared. All data collection events were carried out in French and were subsequently translated into English.

Participants

In total, for the two rounds of data collection (T1/T2), we were able to interview 20 different APs (T1: n = 10, T2: n = 10). A summary of data collection in T1 and T2 is presented in Table 2, and Table 3 presents a description of the participants.

Table 2 Summary of data collection events

Data collection events	T1	*N* (participations)	T2	*N* (participations)
Focus groups	2	7	5	19
Interviews	8	8	0	0
Total of participations		15		19
Total of participants		10*		16**

Abbreviation: AP, accompanying patient.

*

Out of a total of 15 participations, 5 APs participated in 2 events. Therefore, there are 10 different APs in T1.

**

Out of a total of 19 participations, 3 APs participated in 2 events. Therefore, there are 16 different APs in T2, of which 6 also participated in T1. Therefore, there were 10 new APs that were interviewed since T1.

Table 3 Description of participants

Characteristics		T1	T2
		N (participants)	*N* (participants)
	E1	4	7
	E2	1	2
	E3	2	6
	E4	3	1
	25–34 years old	0	1
	35–44 years old	0	2
	45–54 years old	2	2
	55–64 years old	5	7
	65–74 years old	3	4
	Born in the province of Quebec	9	14
	Born outside of Canada	1	2

	Partial studies (high school, college or university)	1	3
	Has a university degree	9	13
	Works part-time	1	1
	Works full-time	0	3
	On leave (illness or maternity)	0	3
	Volunteering	2	3
	Retired	7	6
	Breast cancer	7	9
	Oncogenetic trajectory	2	3
	Breast cancer and oncogenetic trajectory	0	1
	Gynaecologic cancers	0	1
	Metastatic cancer	1	2

Abbreviation: AP, accompanying patient.

In T1, of the 10 APs that were involved in the four establishments and that have accompanied patients, all of them agreed to participate. One focus group with E4 was held in June 2019 (*n* = 3 participants). Another focus group was held in September 2019 E1, E2 and E3 (*n* = 4). The two focus groups were led by the principal researcher (M.-P. P.) and lasted 58 and 178 min, respectively. Moreover, eight individual interviews were held between April and May 2020 and were conducted by research coordinators (K. B. and M. I.-N.). They lasted between 30 and 63 min. Out of the 10 APs, 5 participated in two data collection events (individual interviews and focus groups).

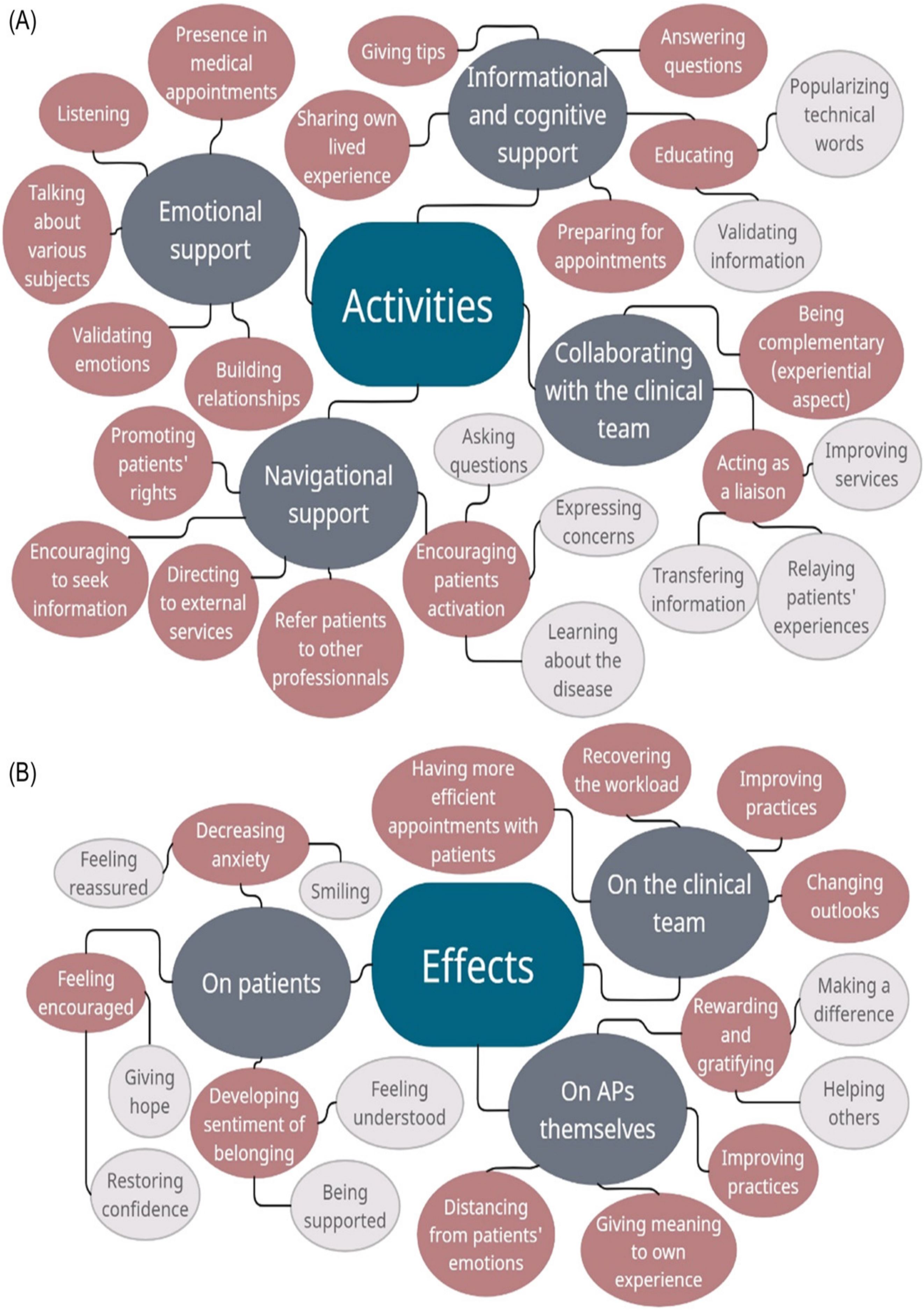
In T2, of the 20 APs that were APs, 16 agreed to participate (4 did not reply to our invitation). Of the 16 participants, 6 have participated in T1. The other four APs that participated in T1 were not reinvited in T2 because they were no longer involved in the PAROLE-Onco programme due to personal issues. Therefore, there were 10 new APs that were interviewed in T2. An initial focus group with E1 and E3 (*n* = 3 participants) was held in September 2021 and lasted 35 min. At that time, the APs had been APs for 12–22 months. Four other focus groups for each establishment (*n* = 16 participants in total) were held between March and May 2022 and lasted between 80 and 115 min. The range of months of involvement was between 6 and 32 during this period. The events were led by the principal researcher or a research assistant (J. P.). Of the 16 APs, 3 participated in two data collection events.

Data analysis

To analyse data, we followed the six-step guideline of Braun and Clarke.[16] First, all interviews were transcribed to familiarize ourselves with the data. Second, several meetings between the authors, including two patient researchers, took place to construct the codebook that contained four main categories: (1) APs activities regarding patients and clinical teams, (2) PAs perceived effects of their activities on the patients, (3) on the clinical team and (4) on themselves. Then, we used a thematic analysis approach to better ‘understand a set of experiences, thoughts, or behaviors’ pertaining to these categories.[17] We used an inductive approach to theme identification—or patterned responses that occurred in the data set.[17] Coding was done using the QDA Miner Software (version 6.0.2.). Steps 4 and 5 consisted of grouping some themes together to define APs activities. The final step is the writing of this manuscript.

RESULTS

The qualitative analysis enabled us to group APs activities into four categories: emotional support, navigational support, informational and cognitive support and collaborating with the clinical teams (Figure 1A). Elements of responses pertaining to the effects of APs activities can be found in Figure 1B.



Enlarge this image.

ActivitiesEmotional support

In T1, one of the main roles reported by APs was to listen. Since APs have undergone similar experiences, they can better understand what patients are going through, and therefore lead conversations patients could not have with their loved ones: ‘The fact that we have experienced the situation, we are able to be more empathetic towards patients […]. The patients tell me things that they could not say to a partner because they do not want to disturb them’ (E4-19). In T2, APs reported moving from an unconditional listening role to a more ‘active’ role towards patients. APs now put the emphasis on discussing and validating the patients' emotions to help them understand and accept their journey with cancer while also reducing their anxiety and reassuring them: ‘what I have experienced with several women is validation, validating them in what they feel, in the choices they can make’ (E2-01).

Overall, APs try to not only talk about the disease, treatment and care trajectories, but also about the difficulties experienced at home, in interpersonal relationships and in daily activities, thus participating with the patients in building relationships based on trust and openness. Some mentioned in T2 that they have accompanied the patients' loved ones to bring comfort to the whole family. Some also said they have accompanied patients to their medical appointments, especially patients who may have barriers that limit their ability to interact with their physician.

Navigational support

Various resources inside and outside the hospital are offered to the patients, and one of the roles of APs, as mentioned both in T1 in T2, is to act as patient navigators. Not only are they familiar with the range of hospital services offered, but they also ‘know the entire chain of operations for having gone through it’ (E2-01). APs noticed that ‘often women are not told about this. […] They don't know they have access to this, and they always think that you have to pay too’ (E1-02). APs therefore ‘encourage them to get the right information’ (E2-02) and make sure to direct patients to the external services made available to them to complete the support sessions they offer, if needed. They also suggest referring patients to other professionals, be it a psychologist, a nutritionist or a social worker, if they feel that the patients' degree of distress lies beyond their area of expertise and experiences undergone to effectively meet the latter's various needs.

In T2, APs realized that the patients are often not informed of their rights. This touches on building patients' ability to advocate for their own rights. They mention their new role in ensuring they know their rights and become comfortable using them. Encouraging them to ask questions and to assume responsibility for fulfilling their desire to understand and learn about their disease are some of the aspects discussed during the meetings with their patients. Therefore, APs help patients make their own decisions by encouraging them to think through the situation, ask questions and express their concerns and uncertainties: ‘here are patients who are afraid to ask questions because they don't want to be perceived as annoying patients. You always have to reassure them. We say “no, it's your right.” We have to encourage them’ (E1-05).

Informational and cognitive support

The accompanying sessions with the patients allow APs, in T1, to share their own lived experiences with discernment without making it an example to be followed. Their role is not to teach, but rather to use their own experience as a way to answer patients' questions. Having experienced the system at hand, APs could serve as resource persons for individuals who are unaware of how to navigate a new healthcare structure: ‘We're able to guide them and encourage them. We are not there to pity them and take care of them. We're really there to support them and say, “Look, I've been there. Here are the steps”’ (E4-19).

In T2, APs explained that they give tips they have learned throughout their own journey with patients instead of giving advice which, according to them, they are not trained to do, nor do they have the expertise to give opinions that include clinical details: ‘I don't like giving advice because I feel it's not part of my mandate… it's really sharing [experiences]’ (E2-02). Another activity mentioned in T2 is that of helping patients to understand and validate the information received by the clinical team, and thus help them prepare for their appointment with healthcare professionals. Indeed, as opposed to T1, they can help educate patients by popularizing some technical information transmitted by the healthcare professionals and talking to patients using the same language as them: ‘we have the same words because we have often experienced the same emotions, so we will share the same words that the professional will not share’ (E1-01). Some APs specify that for medical information, patients instinctively know to direct their questions to healthcare providers. With APs, they prefer to ask questions about the establishment and the care pathway: ‘They will ask more questions about their facility: Did you stay in the hospital long? Was it hard? Did you have any pain? That kind of questions’ (E2-02). By answering patients' questions based on their experiential knowledge, they ‘help patients become partners in their care [by having] a kind of educational role’ (E1-07).

Collaborating with the clinical team

In T1, some APs felt they were not integrated into the clinical team, but that ultimately it could bring a value-added resource to healthcare professionals: ‘it would improve the contact they have with their patients’ (E4-22). In T2, they specify that they have a complementary role with the clinical team with respect to the emotional and experiential aspects of the disease versus the therapeutic aspect provided by the clinical team. Some mentioned that ‘the professionals, they can't know if they haven't lived it …It's just a fact’ (E1-01). Therefore, APs form a different relationship with patients than healthcare professionals can, and they complete the range of services offered to the establishment. APs felt that they are (or should be) ‘a link in a chain of all the different professionals, that [they] are part of the group’ (E1-04), although some feel they have not yet fulfilled that role.

Moreover, APs mentioned in T2 that their role also consists of acting as a liaison between the patients and the clinical team. For example, with patients' consent, they can transfer information to the clinical team. It is done by updating them regularly about their patients' health journey and their patients' personal situation through the provision of medical information about treatment and the disease they may not know: ‘we also serve to update the doctor on important facts that can have an impact on the patients' health’ (E3-07). They can also relay how the patients experience their care and how the healthcare professionals can improve it. Even if information transmission is not homogeneous across establishments, some APs have developed good communication with team members: ‘the pivot nurse was a good ally. I would call her, leave a message, and she would call me back the same day’ (E3-07).

Perceived effects of their intervention on patients

Both in T1 and T2, APs have the perception that patients are less stressed at the end of a meeting after they were listened to and were able to be reassured: ‘I am always told that “It makes me feel good to talk to you”’ (E1-10). There is no need for a full session to have that effect, ‘even 5 minutes with a patient in a corridor, in the elevator, the person is happy, she has really lowered her anxiety level’ (E1-03). After talking with a patient, APs could sense that they were leaving them with a smile: ‘we ended …I'm not telling you with bursts of laughter but with a smile. I'm sure the patient on the other end of the phone line smiled’ (E4-22). In T2, APs added the fact that their accompanying sessions help restore their patients' confidence and hope, and develop the patients' feeling of belonging as they feel understood and supported in their life experiences: ‘I find it positive for patients to be with other people who have had cancer from which they have recovered, that there is long-term healing that exists. I find it encourages them to continue’ (E3-15).

Nonetheless, while in T2, the rapid and positive effects of their support on patients are pointed out, some APs mentioned that a few minutes are not sufficient to delve deep into the patients' concerns and questions, and thus have a positive long-term impact on them. Sometimes, several meetings are necessary before a certain progression in the patients' journey is seen.

Perceived effects of their intervention on the clinical team

Both in T1 and in T2, APs shared that they could facilitate the task of healthcare professionals by preparing patients to meet and feel comfortable with the information they receive from their physicians. They think that it could be easier for healthcare practitioners to have patients that are calm during a medical appointment: ‘If [the] patient is in a good mood, understands and feels safe because she has been spoken to, it is much easier to care for that patient. She will be a lot more open to treatment. I'm sure of that’ (E4-22).

In addition, since health teams can be understaffed and overwhelmed, APs can ‘recover a little from the overload of work’ (E1-02). In T2, they put more emphasis on the complementary aspect of their role to the health professional's therapeutic and curative function through their emotional support and their backing in the process of the patients' adaptation and acceptance of the disease: ‘we're a bit of a buffer between the two; we come to sooth a lot of things that the work staff doesn't always have time to sort out or that the patient doesn't dare to say' (E3-02). Also, the information shared between APs and patients could make the appointments more efficient for the clinical team by ensuring that the tasks are separated. This way, patients can be directed to other resources that offer services that the healthcare team may not be able to provide: 'The health professionals, to advise massage therapy …They didn't have cancer, so the process of reconciliation with the body, they don't know it that much' (E4-22).

By relaying how the patients experience their care and how the healthcare professionals can improve it, APs mentioned feeling heard, and receiving openness and appreciation from the clinical team. For example, when patients made suggestions to improve how patients are received at the hospital, APs met with the staff and received positive feedback, and 'they said it changed their whole outlook. As a result, what I understood was that it was to be an integral part of their training' (E3-16). In turn, this link that is created with the clinical team encourages the staff members to ask APs more questions, consult with them and ask for their opinion.

Perceived effects of their intervention on themselves

In T1, they mentioned that being an AP is rewarding, and it satisfies their need to help others: 'I'm retired, but still feel the need to do things for other people. So that satisfies my needs well. And that's something rewarding' (E2-01). They feel like they are making a difference, and this benefits them both as their discussions also serve as a learning experience: 'It's a plus in both directions. When I talk to someone, it makes me feel just as good to see that I have lightened their mood, as I have helped them. She helps me' (E1-04). In T2, the APs discussed how the different patients they encounter represent a learning and experiential opportunity for them to improve their caregiving abilities and skills. Also, having the opportunity to share allows APs to give meaning to their own experience, and helping someone gives them a sense of purpose. However, some APs can find it emotionally difficult to listen to patients' distress: 'For sure sometimes it can be hard for us. […] We may have lived with cancer, yes, but we haven't experienced all the distress that people can experience' (E4-22). But overall, APs in T2 are more capable of distancing themselves from their patients' life stories to prevent their emotions from taking over their role as unbiased listeners. They felt as if they have developed ways to help them maintain control over their emotions and lighten the heaviness of listening sessions, whether through the community of practice meetings they organize between APs, which help them to share ideas about the more difficult encounters they might have, or by adopting the neutral attitude discussed above.

In T2, however, not all APs continue to consider their work as gratifying. Some perceive their role only as an opportunity to give back, which does not necessarily bring them anything personally: 'The word gratifying is not what resonates with me anymore' (E1-07).

DISCUSSION

The objective of this study was to assess the evolution of APs' perspectives regarding their activities when APs, as well as the perceived effects of their intervention on the patients, on the clinical team and on themselves.

Different activities played

Like many studies on peer support interventions for cancer,[9,10,18–21] our study shows that the primary activity of APs is to listen to patients and validate their emotions to facilitate their acceptance process of the disease and increase their ability to fight cancer in a positive way. This is done by sharing their own lived experiential knowledge and tips they acquired throughout their own journey with illness. They also share information not only about their experiences with the disease and treatments but also about community resources, a role that is also reflected in the work of Fisher et al.[22] and Jacobson et al.[23] They allow patients to visualize the care pathway and thus gain a better understanding of the different steps they will have to go through. In T2, APs' activities shifted from listening and sharing experiences to empowering patients by helping them become partners in their care. It is possible that the 'listening role' is a less threatening first step to finding a place within the care team, but time and experience APs have given APs the ability to try and take on a more active role in the clinical team. Other functions, like advocacy support, are potentially more contentious, and it is not surprising that it appears in T2 rather than in T1. Thus, these APs also have a patient navigator role as presented in the literature,[8,9] and they are all former patients of the establishment and have all been led through the same trajectory. Another capacity emphasized by APs was their ability to help patients better prepare for their medical appointments and better understand their illness, treatments and the consequences of decisions made. Often patients are reluctant to ask professionals to clarify information provided to them or ask questions, or take their place in the decision-making process. By playing this role, APs can provide a safe space in which to ask questions.[24] This educational activity is also found in the literature[25–28] but places less emphasis on APs playing a counsellor role. In our context, they help patients to explore coping resources in a nonconfrontational way using reflective listening rather than persuasion. Finally, they can talk about professionals and introduce them to patients in reference to their own experience of the patient-professional relationship. Such a role is rarely reported in the literature outside of mental health.[29] Therefore, APs provide meta literacy support,[30] characterized by support on behavioural (patient behaviour), social-emotional and cognitive levels, and not only at the educational level.

Particularities to be a member of the clinical team

While there are many studies on the contribution of peer support programmes in cancer care,[9,10,18–21] there are few reports that address peer mentoring in which APs are integrated into the clinical team, except in the area of mental health.[31] Our results show that, in T2, some APs felt more integrated into the clinical team and were able to communicate and collaborate with healthcare professionals, although not all establishments have succeeded in fully integrating APs. Introducing APs as full members of the clinical team translates into APs' having access to the relevant medical information on the patients with their consent to better understand the context of their accompaniment. It also means being able to interact with healthcare professionals when they identify situations that require the contribution of professionals and the possibility of leaving a note in the patient's medical file, with the patient's consent, summarizing the main points of the exchanges that may be relevant for the team.

Being former patients of the establishment and thus being highly familiar with the professionals, APs become the ‘transmission agent’ between the professionals and the patients. On the patients' side, they encourage the development of a bond of trust with the professionals. They also embody hope in the team's ability to care for them, as the APs are there to tell them. For healthcare professionals, the feedback on the patients' health journey and personal life allows them to better understand the patients' reality and thus better respond to their needs to help them have a better experience. Also, APs emphasized the distinction of roles within the clinical team, as they did not consider that discussing treatment and clinical details was their responsibility. They were comfortable giving advice based on their own experience and did not seek to provide professional counselling. APs develop complicity with the patient based on a shared experience. This bond can bring to light important clinical situations that would otherwise not have been reported to the clinical team. By becoming a member of the team, they can suggest that other professionals, such as psychologists, would be able to meet patients' different needs. Again, such a role is not very present in the literature available on peer support programmes except in mental health.

AP's perception of the effects of their interventions

Through this research, we were able to show that the APs had perceived a certain number of effects of their accompaniment to the patients. The first effect that stands out is the decrease in anxiety, whether it be at the time of the examination (genetic, biological, radiological, etc.), the announcement of the diagnosis, the choice of treatments and the end of the treatments. Having a safe place to discuss their fears and anxieties and being supported by people who have successfully dealt with them and are still alive allows them to lower their anxiety levels. By being less anxious, patients are then better able to retain the information given to them, be more able to prepare for their appointment and dare to ask questions. Such a change in patients' behaviour allows them to be more involved in their care, to regain power over their health,[17] and to develop a partnership with their healthcare professionals.[32,33] APs foster a bond of trust between the clinical team and the patients by sharing their own relational experiences with the team. This lived experience allows patients to identify with and feel more comfortable communicating with their professionals.[34] As discussed by Fisher et al.,[22] one of the key features of peer support revolves around encouraging self-empowerment, as supporters focus on a person-centred approach. In T2, APs also emphasized restoring patients' confidence through their accompanying sessions. The authors considered supporters' role in helping patients cope with negative emotions and insecurities, just as APs mentioned discussing with patients their fears and worries.

For professionals, as evidenced by the role of APs within the team, they make them more aware of the patients' perspective and experience and may therefore realize that they may have to change their behaviour, in particular by improving their communication abilities. This contributes to improving the quality of care, as highlighted by Gates and Akabas[35] and to humanizing the care process.

For APs themselves, Brodar et al.[19] mentioned that peer supporters could become emotionally charged following their encounters with patients as they can be reminded of their own experience with cancer. It was therefore suggested that there should be more support from clinical staff as well as from other peer supporters to create a sense of community which could comfort APs during difficult times and help them give meaning to their own experiences. However, in our study, such a need did not emerge. This can perhaps be explained by APs meeting regularly in a community of practice where they can share their accompaniments and find support from the other peers present. To APs, APs are seen more as a learning opportunity, which helps give meaning to their own journey with their illness while also giving them a sense of accomplishment. Such a result has been mentioned by Solomon[34] ; being a peer provider offered the latter personal growth as it increased their confidence in their capabilities to support and their ability to cope with the illness as well as their self-esteem.

Limitations

The concept of APs as an integral member of a clinical team is quite recent. Our study is exploratory and requires further study over time and quantitative studies to test different models. We also recognize that APs have different perceptions of their integration, and thus the results may not be an exact representation for all APs, nor do all APs practice every activity mentioned above. Through their own experience and with time, they have developed their own way of APs. Therefore, it would be important to further explore the different accompanying profiles of APs in the future. Similarly, the contexts in the four establishments are different and, accordingly, our results cannot be generalized. Moreover, here we have presented APs' perspective of their roles and their effects on themselves, the patients and the clinical team, but it is also important to assess the challenges and facilitators of their integration into the clinical team. Those results are presented in another manuscript in preparation. Future work could assess how the roles of APs and their effects on their loved ones would change if they were paid as opposed to working as volunteers, as is currently the case. In addition, it would be important to assess the patients' as well as the clinical teams' perspectives on APs. Data collection for the two populations is currently underway. Also, of the 29 APs that were included in the clinical teams at the four establishments, 20 participated in the study because some had changed positions or were unable to respond to our request. However, in our data collection process, both in T1 and T2, we felt that we had reached data saturation.

CONCLUSION

This article assesses the evolution of APs' perception of their role and the effects they can have on people affected by breast cancer, mostly, on healthcare professionals and on themselves. It highlights that APs provide emotional, informational, cognitive and navigational support that allows patients to be more empowered in their care. As they gain experience, APs progressively endorse a broader set of roles within the teams. APs also help patients become partners in their care. They are able to mobilize their experiential knowledge to complement professionals' scientific and experiential knowledge. By integrating them into teams, they can also help professionals more effectively take into account patients' lived experiences in the way they respond to their needs. In this way, they contribute to improving patients' experience of care, but also the professionals' sensitivity to patients' experiences. However, to be able to respond to patients' needs and fit into teams, organizational factors may be more or less favourable. In a second article, we, therefore, propose to focus on the issues identified by APs and examine how healthcare establishments can further facilitate integrating APs into their team.

AUTHOR CONTRIBUTIONS

Marie-Pascale Pomey, Monica Iliescu-Nelea, Cécile Vialaron, Karine Bouchard, Louise Normandin, Marie-Andrée Côté, Mado Desforges, Israël Fortin, Isabelle Ganache, Catherine Régis, Zeev Rosberger, Danielle Charpentier, Lynda Bélanger, Michel Dorva, Djahanchah P. Ghadiri, Mélanie Lavoie-Tremblay, Antoine Boivin, Jean-François

Pelletier, Nicolas Fernandez, Alain M. Danino and Michèle de Guise have conceived and designed the project. Marie-Pascale Pomey, Jesseca Paquette, Monica Iliescu-Nelea, Cécile Vialaron, Rim Mourad, Karine Bouchard, Louise Normandin, Marie-Andrée Côté, Monica Iliescu-Nelea and Pénélope Pomey-Carpentier have participated in data collection and analysis. All authors have made substantial contributions to this study and have participated in the writing of this paper.

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CONFLICT OF INTEREST

The authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

Data sharing is not applicable to this article as no data sets were generated or analysed during the current study.

ETHICS STATEMENT

This study received ethical approval from the Research Ethics Committee (17.260) of the Research Centre of the University of Montreal Hospital Centre (CRCHUM).

DETAILS

Subject:	Intervention; Oncology; Radiation therapy; Health care; Medical personnel; Listening; Parole; Breast cancer; Radiotherapy; Teams; Interviews; Chemotherapy; Emotions; Patients; Data analysis; Alleviation; Learning; Social support; Patient satisfaction; Focus groups; Pandemics; Surgery; Medical research; Health professional-Patient communication; Emotional support; Data collection; Cognitive ability
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Self-management interventions for children and young people with sickle cell disease: A systematic review

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ProQuest document link

ABSTRACT (ENGLISH)

Background

Increasing numbers of interventions are being developed to support self-management for children and young people (CYP) with sickle cell disease (SCD), but no systematic review has systematically synthesized this evidence regarding their characteristics, effectiveness, acceptability and feasibility for all published intervention types.

Methods

The Joanna Briggs Institute guidelines for mixed-method reviews were followed. A systematic search of eight databases and key journals was conducted from their inception to November 2021. Primary research of self-management interventions targeting CYP with SCD aged 8–24 years and reporting any health/social outcome and acceptability data were included. Design-specific standardized critical appraisal instruments were used. Two independent reviewers screened and appraised the articles. A third reviewer resolved disagreements.

Results

Of 1654 articles identified, 38 studies were included. Methodological quality was moderate. Most studies evaluated SCD education, psycho-behavioural, psychosocial and skills training and/or social support interventions. They appear to demonstrate short-term improvements in knowledge, social functioning and medical adherence outcomes. Interventions that were multifaceted in content, combined technological platforms and in-person group-based formats and involved peers, family and care providers were more acceptable and effective. The long-term impact of interventions was limited, including CYP's involvement in the intervention development and implementation.

Conclusions

There is inconclusive evidence for any self-management programme. Nonetheless, support from family, peers and care providers appears to be important for self-management interventions' effectiveness and acceptability. Future research needs to prioritize CYP involvement in both intervention design and delivery, their wider social context and include CYP with SCD from non-Black backgrounds.

Patient and Public Contribution

Three young people with SCD recruited acted as the review advisors. They were formally trained in the review process and involved in every aspect of the review: the design, conduct and interpretation of the findings. CYP involvement in the interventions' development and implementation was analysed as part of the review. This systematic review was conducted as part of a wider research project titled: *Understanding fatigue experiences of CYP with SCD to guide the co-development of a fatigue self-management intervention*. Two of the young advisors involved in the review were also involved in the development of the project funding application.

FULL TEXT

INTRODUCTION

Sickle cell disease (SCD) is globally the most common paediatric genetic blood disorder. Approximately 300,000 babies are born with the condition annually, with a projected 30% increase in incidence by 2050.[1] Most births occur in Africa, but annually an estimated 2000 and 300 babies are born with SCD in the United States and the United Kingdom, respectively.[2] Alterations in the shape of red blood cells characterize SCD, leading to blood vessel occlusion and inflammation, infarction, organ damage, pain and profound chronic anaemia.[3] Consequently, children born with SCD experience comorbidities, have high healthcare needs and reduced life expectancy.[4,5]

With medical advances, mainly in high-income countries, children born with SCD transition into adulthood. However, SCD is associated with significant medical and psychosocial challenges that often worsen when children and young people (CYP) reach an age to become responsible for managing their condition. As CYP with SCD mature, they have increased decision-making autonomy and self-care independence. Self-management is the purposeful performance of specific learned tasks, activities and behaviours to manage the medical, psychosocial and life impact of a chronic illness.[6-8] SCD self-management strategies include symptom monitoring, following treatment plans, and ensuring health maintenance practices. Self-management is crucial as CYP with SCD begin caring for themselves and their illness.

SCD-related management strategies can negatively impact CYP's perceived quality of life, disrupt school and work attendance and participation and influence their social interactions and relationships.[9-15] This may reduce commitment to treatment plans and health maintenance practices.[16] Effective self-management support can help mitigate treatment challenges, engage CYP, improve health and social outcomes and reduce illness burden and health costs.[16,17] Consequently, recent efforts have focused on developing and implementing interventions to support and improve self-management.

Previous reviews have focused on specific psychological,[18] medication adherence,[19] transition programmes[20] and e-health interventions,[21] and assessed their effects on particular outcomes. No systematic review has been identified that focused on all types of self-management interventions for CYP with SCD and included the full range of health and social outcomes that have been evaluated. This review aimed to: (1) summarize the range and characteristics of self-management interventions for CYP with SCD; and (2) critically evaluate the effectiveness, acceptability and feasibility of different self-management interventions, identifying factors influencing their acceptability and implementation.

METHODS

The Joanna Briggs Institute (JBI) guidelines for conducting mixed-method reviews[22] informed the conduct of this review. The review protocol was registered (PROSPERO: CRD42021286422). The review is reported according to the expanded Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) reporting checklist.[23]

Search strategy

First, eight databases: Ovid MEDLINE, EMBASE, PsycINFO and EBM Reviews, ASSIA, CINAHL, Web of Science and Engineering Village were searched from their inception to November 2021. A comprehensive search strategy was developed and modified appropriately across the databases to reflect the various MeSH terms. The following main concepts: ‘child’, ‘young people’, ‘sickle cell disease’, ‘self-management’, ‘self-care’ and ‘intervention’ guided the development of the search strategy, with support from a medical reference librarian. See Supporting Information: File 1 for full search strategies applied across the databases. Second, searches of online abstract archives of key paediatric and haematology journals (*Blood*, *Journal of Pediatric Psychology*, *Journal of Pediatric Hematology Oncology, Pediatric Blood Cancer*, *Journal of Adolescent Health*), trial registries (*ISRCTN*, *ClinicalTrials.gov*), review registries (*PROSPERO*, *Cochrane Database of Systematic Reviews, JBI Systematic review Register*) internet-based resources (*Google*, *Google Scholar*) and OpenGrey from their inception to November 2021 supplemented database searches. Third, reference lists of included studies and systematic review[18-21] also provided sources for studies not identified in the database searches. The 3-step search strategy was implemented by one reviewer (B. A. P.). Authors of recent and ongoing studies were contacted for available publications and/or unpublished data. The review included evidence that met the eligibility criteria detailed in Table 1. The search was unrestricted by country or setting, language or publication year.

Table 1 Eligibility criteria

PICOS framework	Inclusion criteria	Exclusion criteria
Population	CYP aged 8–25 years with SCD. The upper age limit is consistent with definitions of young people by the UN, WHO and UK NHS. The lower age limit is informed by the evidence that children with long-term health conditions may begin to engage (informally) in self-management around this age.[8] Papers including samples described as CYP without further specificity around age. Papers involving samples primarily encompassing CYP with a reported mean age between 8 and 25 years. Papers including children and/or young people with long-term conditions that reported findings for CYP with SCD.	Only included participants over 25 years old. Mean age >25 years.
Intervention	We defined self-management as purposeful performance of specific learned tasks, activities and behaviours to manage the medical, psychosocial and life impact of a chronic illness.[6-8] And we defined a self-management intervention as any intervention, programme or approach designed to develop the ability of and/or support CYP with SCD to manage their long-term health condition through education, training and support to develop their knowledge, skills or psychosocial resources. All self-management interventions that target CYP with SCD. All types/forms of self-management intervention—health, social care and educational interventions—designed to support or facilitate CYP with SCD to take control of and manage their condition, promote their capacity for self-care and/or maintain and enhance their physical and/or mental health.	Interventions in which children and/or young people are not actively engaged and/or remain passive recipients of knowledge or instructions. Self-management interventions that target professionals, parents/carers/siblings or families as a whole, without a distinct component(s) for CYP. Papers only describing the development of an intervention. Papers where the effects of the self-management intervention cannot be distinguished from broader interventions for SCD.
Comparator	Usual care Another self-management intervention(s) No comparator	
Outcome	No restrictions	

Study design	Primary research, including qualitative, mixed-methods and quantitative studies of all designs. Evaluations and discursive articles about SCD self-management interventions. Conference papers that present detailed information about the intervention, research methods and outcome(s).	Dissertations/theses Secondary research (secondary data analysis and literature reviews) Case studies Posters/conference abstracts and proceedings

Abbreviations: CYP, children and young people; SCD, sickle cell disease.

Study selection

All citation records from the database and manual searches were exported into EndNote 20® and deduplicated. Records were independently screened in Covidence® by two reviewers (B. A. P. and S. K.). Any disagreements were adjudicated by the third author (K. M. A.). Reasons for excluding papers were noted.

Quality assessment

Included papers were appraised for methodological quality independently by B. A. P. and S. K. using design-specific standardized critical appraisal instruments from the JBI System for the Unified Management, Assessment and Review of Information (SUMARI).[24] For each of the appraisal criteria, the individual appraisers assessed whether the article met each quality criterion ('yes'), if it failed to meet the criterion ('no'), if insufficient information was presented to assess the specific criterion adequately ('unclear') or if the criterion was not applicable due to the study type, design and/or intervention ('N/A') (Tables 3–7). The total number of appraisal criteria met determined the overall quality of a paper. Papers that met three-quarters or more of the appraisal criteria were considered to be of high quality, between half and three-quarters as moderate quality and less than half as poor quality (Tables 3–7). The quality appraisal did not impact study inclusion or exclusion but was considered in the synthesis.

Data extraction

A typology from previous studies on self-management interventions guided data extraction on intervention characteristics.[25,26] Data were extracted on the following dimensions: study aims and objectives, study location, publication year, research design, participant demographics, intervention characteristics and all outcomes measured for effectiveness and acceptability, as well as reported feasibility and implementation issues. For quantitative studies, extracted outcomes comprised descriptive and/or inferential statistical results and themes from the analysis of open-ended survey questions. For the qualitative studies, data extracted included themes with corresponding quotes. One reviewer (B. A. P.) undertook data extraction, whilst a second reviewer (S. K.) independently extracted data from 10% of all the included papers as a measure to ensure quality control and consistency. Additional information was located and extracted from published protocols/formative papers linked to the included studies. See Supporting Information: File 2 for the detailed data extraction domains.

Data synthesis

Results from the included papers were synthesized narratively in line with the JBI approach. As only one qualitative research paper was included, it was impossible to follow the JBI convergent integrated approach for mixed-method review. The characteristics of the different interventions were examined and summarized in relation to their effectiveness, acceptability and feasibility for implementation in 'real life' settings.

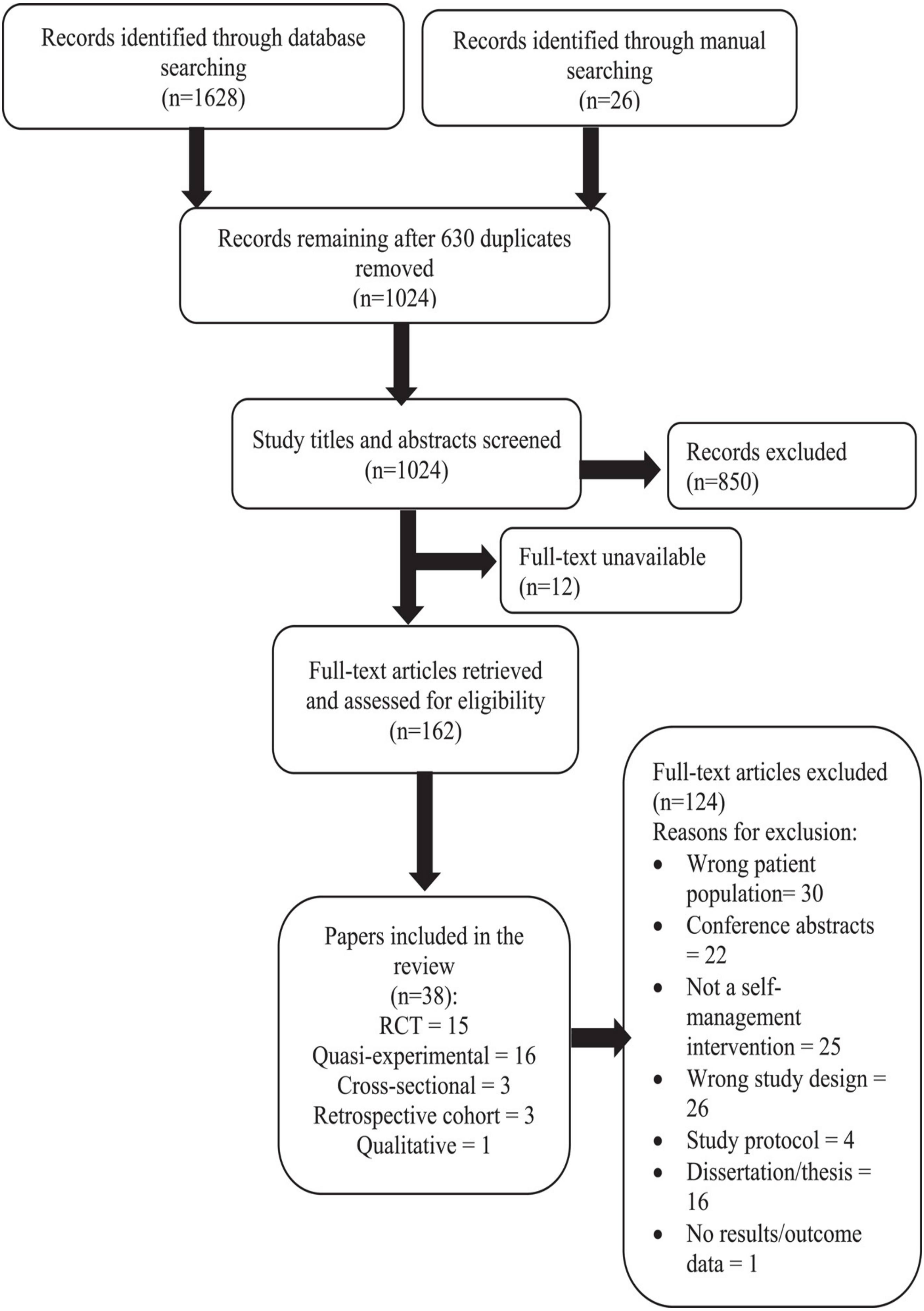
Patient and Public Involvement (PPI)

The review team worked with a PPI group of three young people with SCD recruited from a patient support group. They were trained and involved at every review stage to ensure that patient perspectives (and concerns) were reflected in the review. They commented on the search strategy and the data extraction dimensions developed by the reviewers, helping to identify additional search terms and quality criteria. In addition, they highlighted issues for discussion and recommendations after reviewing the extracted and synthesized data.

RESULTS

A total of 1654 records were imported into EndNote®. After deduplicating the records, 1024 were screened in

Covidence®; 38 (representing 32 distinct studies) met all the inclusion criteria and were included in the final review (Figure 1). Of these, 37 were quantitative studies[27-63] and 1 was a qualitative study.[64]



Enlarge this image.

Study and sample characteristics

Table 2 presents a summary of the included studies. The 38 articles published between 1987 and 2021 were predominantly from the United States (*n* = 34, 89.5%). More than half (*n* = 26, 68.4%) were published within the last 5 years. There was a preponderance of relatively small-scale (e.g., single-site, small-sample) feasibility/pilot studies (*n* = 21)[28,34-36,40,44,46-51,55,57,58,60-64] whose primary objective was to report effectiveness data, acceptability data and/or assess the feasibility and trial design of interventions. The majority (*n* = 31) of the included studies used a randomized[30,31,37,42-44,46-50,54,55,58,64] or non-randomized comparative study design[27,28,32,38,39,41,45,51,52,56,57,59-63] in which self-management interventions were either compared with standard care[37,42,45,47-49] or specific control interventions.[30,36,44,46,54,55,58]

Table 2 Study and participant characteristics

References	Country	Study design	Target population	CYP participant characteristics (number, mean age, % ethnicity, % gender, % SCD diagnosis)	Intervention name and type	Control	No. of study sites	Attrition rate (follow-up) (%)
Adegbolagun et al.[28]	Nigeria	Noncontrolled, pre-post experimentala	CYP	*N* = 20; 19.8 years; 100% HbSS; gender and ethnicity not specified	Behavioural	N/A	1	0
Abd Elaziz and Abd Elghany[27]	Egypt	Noncontrolled, pre-post experimental (full trial)	CYP-parent dyads	*N* = 100; 12 ±3.12 years; 55% males; 100% HbSS; ethnicity not specified	Educational, skill training	N/A	2	0
Allemang et al.[29]	Canada	Cohort—retrospective	CYP	*N* = 110; 18.1 years. Gender, diagnosis and ethnicity not specified	Educational, behavioural	N/A	1	14
Barakat et al.[30]	USA	RCT	CYP &family	*N* = 53 (27 IG, 26 CG); 14.24 years IG, 14.10 years CG; 52.9% males IG, 30% males CG; 100% African American IG, 95% African American CG	PAIN (educational, behavioural)	Attention control: DISEASE ED (educational)	1	40

Broome et al.[31]	USA	RCT	CYP	*N* = 67 (35 IG children, 32 IG adolescents); 9.2 years IG children; 15.3 years IG adolescents; 51% males IG children, 65% males IG adolescents; 100% African American. No data provided on CG	Educational, behavioural, social support	Attention control (picnic, trip to a museum)	Not specified	48
Crosby et al.[33]	USA	Cross-sectionalb	CYP &parents	*N* = 43; 12.8 ±3.98 years; 39.5% males; 79% HbSS; 100% African American	Take-Charge Programme (skill training)	N/A	1	O
Crosby et al.[34]	USA	Cross-sectionala	CYP	*N* = 5; 13–24 years. Gender, diagnosis and ethnicity not specified	iManage (skill training, social support	N/A	1	0
Crosby et al.[35]	USA	Cross-sectionala	CYP	*N* = 22; 18.8 years; 55% males; 100% African American. Diagnosis not specified	Chronic Disease Self-Management Programme (CDSMP) (educational, skill training)	N/A	1	36
Crosby et al.[36]	USA	RCTa	CYP	*N* = 58 (27 IG, 31 CG); 16.7 years IG, 16.3 years CG; 52% females IG, 54% females CG; 46% HbSS IG, 63% HbSS CG; 100% African American	SC Thrive (educational, skill training)	Attention control: SCHealthED (educational)	1	9
Crosby et al.[64]	USA	Qualitative	CYP	*N* = 19; 17.1 years; 58% females; 53% HbSS; 100% African American	SC Thrive (educational, skill training)	N/A	1	N/A
Cozzi et al.[32]	USA	Noncontrolled, pre-post experimental	CYP	*N* = 8; 10–20 years; 63% females; 88% HbSS; ethnicity not specified	Educational, behavioural	N/A	1	0

Daniel et al.[37]	USA	RCT	CYP &family	*N* = 83; 8.29 years; 50% males; 59% HbSS; ethnicity not specified	Family Taking Control (educational, skill training)	Delayed intervention control	2	27
Dobson and Byrne[38]	USA	Noncontrolled, pre-post experimental	CYP	*N* = 20; 8.4 years; 60% males; 95% HbSS; 75% African American	Behavioural	N/A	1	0
Dobson[39]	USA	Noncontrolled, pre-post experimental	CYP	*N* = 20; 8.4 years; 60% males; 95% HbSS; 75% African American	Behavioural	N/A	1	0
Estepp et al.[40]	USA	Cohort—retrospectivea	CYP	*N* = 83; 13.9 years; 53% females; 91% HbSS; ethnicity not specified	Scheduled Instant Messaging Over the Network (SIMON) (behavioural)	N/A	1	32
Fouda et al.[41]	Egypt	Noncontrolled, pre-post experimental	CYP	*N* = 50; 14.4 ±1.15 years; 54% males. Gender and ethnicity not specified	Educational, skill training	N/A	2	0
Gil et al.[42]	USA	RCT	CYP	*N* = 49 (25 IG, 24 CG); 11.9 ±3 years; 63% males; 67% HbSS; 100% African American	Behavioural	Standard care	1	0
Gil et al.[43]	USA	RCT	CYP	N = 49 (25 IG, 24 CG); 11.9 ±3 years; 63% males; 67%	Behavioural	Standard care	1	0
Green et al.[44]	USA	RCTa	CYP &parents	*N* = 28; 14.3 years; 43% females; 50% Hispanic. IG and CG are not specified.	HABIT (educational, behavioural, skill training)	Two educational brochures	2	11
Hazzard et al.[45]	USA	Noncontrolled, pre-post experimental	CYP	*N* = 47. Other characteristics not specified	STARBRIGHT World (educational, social support)	Usual education programme	1	0

Hood et al.[46]	USA	RCTa	CYP	*N* = 58 (27 IG, 31 CG); 16.7 years IG, 16.3 years CG; 52% females IG, 54% females CG; 46% HbSS IG, 63% HbSS CG; 100% African American	iManage (behavioural, skill training, social support)	Attention control SCHealthED (educational)	1	0
Kaslow et al.[47]	USA	RCTb	CYP &family	*N* = 39 (IG = 20, CG = 19); 10.3 years; 62% females; 69% HbSS; 100% African American	Educational, behavioural	Treatment as usual	1	34
Ketchen et al.[48]	USA	RCTb	CYP &parents	*N* = 56; 12.7 years IG, 12.18 years CG; 50% males IG, 53% males CG; 100% African American. Diagnosis not specified	STARBRIGHT World (educational, behavioural, social support)	Wait-list	3	34
McClellan et al.[49]	USA	RCTa	CYP &parents	*N* = 19 (9 IG, 10 CG); 13.4 ±2.9 years; 68% females; 100% African American. Diagnosis not specified	Behavioural	Wait-list	1	0
Palermo et al.[50]	USA	RCTa	CYP &parents	*N* = 25 (15 IG, 10 CG); 14.8 years; 64% females; 64% HbSS; 64% African American	Web-based Management of Adolescent Pain (Web-MAP) (behavioural, skill training)	Internet-delivered pain education	9	20
Phillips et al.[51]	USA	Noncontrolled, pre-post experimentala	CYP &parents	*N* = 60; 7.8 years; 53% males. Diagnosis and ethnicity not specified	Voice Crisis Alert V2 (educational, behavioural, skill training)	N/A	1	20
Rodgers-Melnick et al.[52]	USA	Noncontrolled, pre-post experimental	CYP	*N* = 30; 21.04 years; 50% males; 63% HbSS; 100% black ethnicity	Build, Educate, Advance, Transition in Sickle cell disease (BEATS) (educational, behavioural, skill training)	N/A	1	18% attended 0/4 sessions

Saulsbery et al.[53]	USA	Cohort—longitudinalb	CYP	*N* = 53; 14 years; 62% males; 66% HbSS; 100% African American	Sickle Cell Transition E-Learning Programme (STEP) (educational, behavioural)	N/A	1	26
Schatz et al.[54]	USA	RCT	CYP	*N* = 48 (23 IG, 25 CG); 12.3 ±2.2 years CG, 13.8 ±2.7 years IG; 43% males CG, 39% males IG; 78% HbSS CG, 74% HbSS IG; 96% African American CG, 91% African American IG	Behavioural	Wait-list standard care	2	4
Schwartz et al.[55]	USA	RCTa	CYP &family	*N* = 49 (25 IG, 24 CG); 14.32 years IG; 56% females IG; 88% HbSS IG; 100% African American	Behavioural	Disease education attention control	1	0
Sil et al.[57]	USA	Noncontrolled, pre-post experimentala	CYP	*N* = 57; 13.33 years; 61% females; 73.7% HbSS; 100 non-Hispanic black	Comfort Ability Programme for Sickle Cell Pain (CAP for SCP) (educational, behavioural)	N/A	3	53
Sil et al.[56]	USA	Noncontrolled, pre-post experimental	CYP	*N* = 57; 13.4 years; 56.4% females; 68-1% HbSS; 98% African American	Behavioural, skill training	N/A	1	38
Smaldone et al.[58]	USA	RCTa	CYP &parents	*N* = 28; 14.3 years; 43% females; 50% Hispanic. IG and CG are not specified.	HABIT (educational, behavioural, skill training)	Two educational brochures	2	11
Smith et al.[59]	USA	Noncontrolled, pre-post experimental	CYP	*N* = 32. Other characteristics not specified	Virginia Commonwealth University Transition Intervention Programme (educational, skill training)	N/A	1	0

Treadwell and Weissman[60]	USA	Noncontrolled, pre-post experimentalb	CYP &parents	*N* = 11; 11.6 years; 100% African American. Gender and diagnosis not specified	Desferal Day Camp (educational, behavioural, social support)	N/A	1	0
Viola et al.[61]	USA	Noncontrolled, pre-post experimentala	CYP	*N* = 24; 20.8 ±2–3 years; 45.8% females; 75% HbSS; 87.5% black or African American	Students Helping Individuals Facilitate Transition (SHIFT) (educational, social support)	N/A	1	0
Wihak et al.[62]	USA	Noncontrolled, pre-post experimentala	CYP &parents	*N* = 8; 10–17 years; 100% black. Diagnosis and gender not specified	Comfort Ability Programme for Sickle Cell Pain (CAP for SCP) (educational, behavioural)	N/A	1	0
Yoon and Godwin[63]	USA	Noncontrolled, pre-post experimentalb	CYP	*N* = 22; 10.86 ±2.58 years; gender, ethnicity and diagnosis not specified	Sickle Cell Slime-O-Rama Game (educational)	N/A	1	0

Abbreviations: CG, control group; CYP, children and young people; HbSS, haemoglobin SS genotype; IG, intervention group; *N*, number; N/A, not applicable; RCT, randomised controlled trial; SCD, sickle cell disease. a Feasibility study.

b

Pilot study.

Most studies (*n* = 24) evaluated self-management interventions targeting CYP, ten targeted CYP-parent dyads [27,33,44,48-51,58,60,62] and four targeted family units.[30,37,47,55] Most studies (*n* = 23) included self-selected convenience samples, with sizes ranging from 5[34] to 110[29] participants recruited mainly from single sites. Nine studies recruited participants from multiple settings.[27,37,41,44,48,50,54,57,58] CYP participants were aged between 6 and 24 years, predominantly African American and had homozygous SCD (i.e., HbSS genotype).

Methodological quality of included studies

Overall, the body of evidence was of moderate methodological quality (Tables 3–7). Several of the randomised controlled trial (RCTs) (*n* = 7) were assessed as being of poor quality. Four were feasibility RCTs with poor reporting quality. Only four RCTs were rated as high quality, three of which were feasibility/pilot RCTs. The RCT studies lacked details of randomization methods, group allocations, blinding of assessors, group comparativeness and statistical analysis methods. The limitations of the quasi-experimental studies were their short follow-up length, the multiplicity of outcome measurements and the reliability of the outcome measurement tools used. The cohort and cross-sectional studies failed to report on how confounding factors were managed in the analysis. Only two quantitative study article[46,64] sreported sample size calculation or provided reasons for the chosen sample sizes. While all articles reported the number of participants lost to follow-up, none of the full trials reported conducting an intention-to-treat analysis.

Table 3 Methodological quality of included RCTs

Quality criterion	Green et al.[44]	Barakat et al.[30]	Broome et al.[31]	Crosby et al.[36]	Daniel et al.[37]	Gil et al.[42]	Gil et al.[43]	Hood et al.[46]	Kaslow et al.[47]	Ketchen et al.[48]	McClellan et al.[49]	Palermo et al.[50]	Schatz et al.[54]	Schwartz et al.[55]	Smaldone et al.[58]
Was randomization used for assignment of participants to treatment groups?	Yes	Unclear	Unclear	Yes	Yes	Unclear	Unclear	Unclear	Unclear	Yes	Unclear	Yes	Yes	Unclear	Unclear
Was allocation to treatment group concealed?	Yes	Unclear	Unclear	Unclear	Yes	Unclear	Unclear	Unclear	Unclear	Yes	Unclear	Yes	Yes	Unclear	Yes
Were treatment groups similar at the baseline?	Unclear	Yes	Unclear	Yes	Yes	Yes	Unclear	Unclear	Yes	Yes	Unclear	Unclear	Unclear	Unclear	No
Were participants blind to treatment assignment?	N/A	N/A	N/A	N/A	N/A	N/A	N/A	N/A	N/A	N/A	N/A	N/A	N/A	N/A	N/A
Were those delivering treatment blind to treatment assignment?	N/A	N/A	N/A	N/A	N/A	N/A	N/A	N/A	N/A	N/A	N/A	N/A	N/A	N/A	N/A
Were outcomes assessors blind to treatment assignment?	Unclear	Unclear	Unclear	Unclear	Unclear	Unclear	Unclear	Unclear	Unclear	Unclear	Unclear	Unclear	Unclear	Unclear	Unclear
Were treatment groups treated identically other than the treatment of interest?	Yes	Unclear	Yes	Yes	Yes	Yes	Yes	Unclear	Yes	N	Unclear	Unclear	Unclear	Unclear	Unclear
Was follow-up completed and if not, were differences between groups in terms of their follow-up adequately described and analysed?	Unclear	N	Yes	N	Yes	Unclear	Unclear	Unclear	Yes	Yes	Yes	N	Unclear	N	Unclear
Were participants analysed in the groups to which they were randomized?	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Unclear	Yes	Yes	Unclear	Unclear	Yes	Unclear	Yes

Were outcomes measured in the same way for treatment groups?	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Unclear	Yes	Yes	Unclear	Yes	Yes	Unclear	Yes
Were outcomes measured in a reliable way?	Unclear	N	Yes	Unclear	Unclear	Unclear	Unclear	Yes	Yes	Unclear	Unclear	Yes	Unclear	Unclear	Yes
Was appropriate statistical analysis used?	Yes	Yes	Yes	Yes	Yes	Unclear	Unclear	Yes	Yes	Unclear	Unclear	Yes	Unclear	Unclear	Yes
Was the trial design appropriate, and any deviations from the standard RCT design (individual randomization, parallel groups) accounted for in the conduct and analysis of the trial?	N/A	N/A	N/A	N/A	N/A	N/A	N/A	N/A	N/A	N/A	N/A	N/A	N/A	N/A	N/A
Overall quality	*Moderate*	*Poor*	*Moderate*	*Moderate*	*High*	*Poor*	*Poor*	*Poor*	*High*	*High*	*Poor*	*Poor*	*Moderate*	*Poor*	*High*

Abbreviations: N, number; RCT, randomised controlled trial

Table 4 Methodological quality of included quasi-experimental studies

Quality criterion	Hazzard et al.[54]	Abd Elaziz and Abd Elghany[27]	Adegbolagun et al.[28]	Cozzi et al.[32]	Crosby et al.[34]	Dobson and Byrne[38]	Dobson[39]	Fouda et al.[41]	Phillips et al.[51]	Rodgers-Melnick et al.[52]	Sil et al.[56]	Sil et al.[57]	Smith et al.[59]	Treadwell and Weissman[60]	Viola et al.[61]	Wihak et al.[62]	Yoon and Godwin[63]
Is it clear in the study what is the ‘cause’ and what is the ‘effect’? (i.e., there is no confusion about which variable comes first)	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Unclear	Yes	Yes	Yes	Yes

Were the participants included in any comparison similar?	No	N/A	N/A	N/A	N/A	N/A	N/A	N/A	N/A	Unclear	Yes	N/A	N/A	Yes	N/A	N/A	N/A
Were the participants included in any comparison receiving similar treatment/care, other than the exposure of intervention of interest?	Unclear	N/A	N/A	N/A	N/A	N/A	N/A	N/A	N/A	Unclear	Yes	N/A	N/A	Unclear	N/A	N/A	N/A
Was there a control group?	Yes	No	No	No	No	No	No	No	No	No	No	No	No	No	No	No	No
Were there multiple measurements of the outcome both pre and post the intervention?	No	No	No	Unclear	Yes	No	No	No	No	No	No	No	No	No	No	No	No
Was follow-up completed and if not, were differences between groups in terms of their follow-up adequately described and analysed?	Unclear	Yes	Unclear	Unclear	Unclear	Yes	Unclear	Yes	No	No	No	No	Unclear	Unclear	Yes	Unclear	Yes
Were the outcomes of participants included in any comparison measured in the same way?	Yes	N/A	N/A	N/A	N/A	N/A	N/A	Unclear	N/A	Unclear	No	N/A	N/A	Yes	N/A	N/A	N/A
Were outcomes measured in a reliable way?	Unclear	Unclear	Yes	Yes	Yes	Unclear	Unclear	Unclear	Yes	Unclear	Unclear	Yes	No	Unclear	Yes	unclear	No

Was appropriate statistical analysis used?	Yes	Yes	Yes	Yes	Yes	Yes	Unclear	Yes	Yes	Yes	Yes	Yes	Unclear	Yes	Yes	Unclear	Yes
Overall quality	*Moderate*	*Moderate*	*Moderate*	*Moderate*	*High*	*Moderate*	*Poor*	*Moderate*	*Moderate*	*Poor*	*Moderate*	*Moderate*	*Poor*	*Moderate*	*High*	*Poor*	*Moderate*

Table 5 Methodological quality of included cohort studies

Quality criterion	Allemang et al.[29]	Estepp et al.[40]	Saulsberry et al.[53]
Were the two groups similar and recruited from the same population?	Yes	N/A	N/A
Were the exposures measured similarly to assign people to both exposed and unexposed groups?	N/A	N/A	N/A
Was the exposure measured in a valid and reliable way?	N/A	No	Yes
Were confounding factors identified?	Unclear	No	No
Were strategies to deal with confounding factors stated?	Unclear	No	No
Were the groups/participants free of the outcome at the start of the study (or at the moment of exposure)?	N/A	Yes	Yes
Were the outcomes measured in a valid and reliable way?	Yes	Unclear	No
Was the follow-up time reported and sufficient to be long enough for outcomes to occur?	Yes	Yes	Unclear
Was follow-up completed, and if not, were the reasons to loss to follow-up described and explored?	Yes	Yes	Yes
Were strategies to address incomplete follow-up utilized?	N/A	N/A	N/A
Was appropriate statistical analysis used?	Yes	Yes	Yes

Overall quality	*Moderate*	*Moderate*	*Moderate*

Table 6 Methodological quality of included cross-sectional studies

Quality criterion	Crosby et al.[33]	Crosby et al.[34]	Crosby et al.[35]
Were the criteria for inclusion in the sample clearly defined?	Yes	Yes	Yes
Were the study subjects and the setting described in detail?	Yes	Yes	Yes
Was the exposure measured in a valid and reliable way?	Unclear	N/A	Yes
Were objective standard criteria used for measurement of the condition?	Yes	N/A	Yes
Were confounding factors identified?	No	N/A	No
Were strategies to deal with confounding factors stated?	No	N/A	No
Were the outcomes measured in a valid and reliable way?	Unclear	Yes	Unclear
Was appropriate statistical analysis used?	Yes	Yes	Yes
Overall quality	*Moderate*	*High*	*Moderate*

Table 7 Methodological quality of included qualitative study

Quality criterion	Crosby et al.[64]
Is there congruity between the stated philosophical perspective and the research methodology?	N/A
Is there congruity between the research methodology and the research questions or objectives?	N/A
Is there congruity between research methodology and the methods used to collect data?	N/A
Is there congruity between the research methodology and the representation and data analysis?	No
Is there congruity between the research methodology and the interpretation of results?	No
Is there a statement locating the researcher culturally or theoretically?	No
Is the influence of the researcher on the research, and vice-versa addressed?	No

Are participants and their voices adequately represented?	Unclear
Is the research ethical according to current criteria or for recent studies, and is there evidence of ethical approval by an appropriate body?	No
Do the conclusions drawn in the research report flow from the analysis and interpretation of the data?	Unclear
Overall quality	*Poor*

While studies reported differences in the characteristics of those who completed the study and those who dropped out, they failed to report how these had been factored into their analysis. Some RCTs only reported data on the intervention groups. In some studies, not all outcomes were measured for the comparison groups or intervention delivery and the timing of outcome assessments was variable between and within the groups. Several studies assessed outcomes using nonvalidated tools, or there were missing outcome data. Eleven papers reported high follow-up attrition rates (Table 2). Many studies experienced recruitment challenges, achieving less than 50% of their target sample size. Commonly reported reasons for recruitment difficulties included people's lack of interest in the research, time constraints, inconvenience of intervention format and delivery approaches and perceived lack of intervention benefits.

Description of self-management interventions

Thirty-two self-management interventions were reported in the 38 included articles (Table 8). The majority (*n* = 22) combined different types of self-management support,[28-32,34-39,41,44,45,51,52,57-60,62,64] but all had an educational component on general information about SCD or combined this with more specific information about pain, pharmacological treatments, self-care, clinic appointments, school and academic support, medical insurance and social care supports. Twenty-eight interventions included skills training programmes (self-assessment and planning, goal-setting, problem-solving, coping and interpersonal communication)[27-39,41-44,47-49,51-61,64] and 15 were cognitive-behavioural therapy programmes.[28,30-32,38,39,42,43,49,50,54-57,62] Several interventions included elements to facilitate peer and social interactions and support.[34,37,45,46,48,52,57,60,61] Others[29,33,40,44,58-61] focused on promoting medical adherence (i.e., medication adherence and clinic attendance). Several interventions were referred to by names and/or acronyms that reflected their intent or objective(s) (see Table 2).

Table 8 Intervention characteristics

References	Intervention content	Theoretical framework	Format/delivery	Duration	Location	Facilitators	Intervention development (PPI)

Abd Elaziz and Abd Elghany[27]	SCD education and self-care skill training	Not specified	In-person, group-based, Face-to-face teaching (group discussions, brainstorming, role play, demonstration &redemonstration), printed materials	3 daily sessions, 20–30 min/session	Outpatient	Research staff (nursing background)	Developed by researchers with no PPI or CYP involvement.
Adegbolagun et al.[28]	SCD education, pain management, skill training (activity scheduling, relaxation techniques, attention diversion, effective communication), general self-management,	Not specified	In-person, group-based, Face-to-face teaching	5 weekly sessions. Duration of each session not indicated	Outpatient	Research staff (psychology background)	Adapted from a previous study. Adaptation done by researchers with no PPI.
Allemang et al.[29]	SCD education, self-management counselling (medication appointment adherence), health and social system navigation support	Model for improvement (a quality improvement model)	Face-to-face teaching, telephone and e-mail support, printed resources	120–180 min/in-person sessions. The overall duration was 12 months	Outpatient, remote (phone call, e-mail)	Transition navigator (social worker)	Developed based on survey on the facilitators and barrier to successful transition from young people with SCD. Programme content and format developed with input from clinicians. No PPI or CYP involvement.

Barakat et al.[30]	Knowledge and skill training for pain management (relaxation, guided imagery, positive coping self-statements)	Not specified	In-person, family-based, Face-to-face teaching, digital resources (guided imagery audiotapes), phone calls	4 sessions, the first 3 sessions were 2 weeks apart with a booster session 1 month apart. 90 min/sessions	Home, phone calls	Psychology students &psychologists	Not specified
Broome et al.[31]	SCD education, cognitive-behavioural skill training (relaxation, distraction breathing, guided imagery), art therapy (drawing, murals, art, interaction) to express feelings about pain and develop social skills	Not specified	In-person, group-based, Face-to-face teaching, printed materials, digital resources (videotapes, audiotapes)	4 sessions over 6 weeks. Duration of each session not indicated.	Outpatient	Clinical nurse specialists, art therapist	No PPI or CYP involvement.
Crosby et al.[33]	Self-assessment of barriers to treatment adherence (medication and appointments), action planning, problem-solving skills	Not specified	Voice automated interactive web-based assessment tool	Not specified	Remote (web-based)	Self-guided	CYP with SCD and their parents involved in the second phase (identification of content domains) of development.

Crosby et al.[34]	Pain monitoring/tracking, skill training (goal setting, goal evaluation), social support (group formation and competition)	Not specified	Digital, Mobile app, individual-based	6 weeks	Web-based (digital)	Self-directed	3-phased development process (survey, design and usability testing). CYP involved in all the development phases
Crosby et al.[35]	SCD education, action planning and problem-solving skill training	Not specified	In-person, group-based, face-to-face teaching	6 weekly sessions, 150 min/session	Outpatient	Lay leaders	Pre-existing programme (Stanford Chronic Disease Self-Management Programme) without adaptation for CYP with SCD.
Crosby et al.[36]	SCD education, skills training (communication, problem-solving, pain, mood, mind and health management)	Not specified	In-person, group-based and remote (Zoom &mobile app) face-to-face teaching (brainstorming, video vignettes, modeling, role-play, rehearsal, group exercises, group discussions), e-handouts	6 weekly sessions (first 3 in-person and last 3 via Zoom) plus optional booster session after 2 weeks, 90 min/session	Outpatient, web-based	Psychology student, psychologist	3-phased development process (survey, design and usability testing). CYP involved in all the development phases.

Crosby et al.[64]	SCD education, skills training (communication, problem-solving, pain, mood, mind and health management)	Not specified	In-person, group-based, face-to-face teaching (brainstorming, video vignettes, modelling, role-play, rehearsal, group exercises, group discussions), e-handouts, web-based (individual-based)	6 weekly sessions (first 3 in-person &last 3 via Zoom) plus optional booster session, 90 min/session	Outpatient, web-based	Psychology student, psychologist	3-phased development process. (survey, design and usability testing). CYP involved in all the development phases.
Cozzi et al.[32]	behavioural skill training for pain management (breathing techniques, electromyographic and thermal biofeedback)	Not specified	In-person, individual-based, telephone-based, Face-to-face teaching, audiotapes and phone calls	13 weekly sessions, 20–30 min/session	Outpatient, home	Clinical psychologist and prerecorded audiotapes	Pre-existing intervention adapted for CYP with SCD but no PPI or CYP involvement in the adaptation process.
Daniel et al.[37]	SCD information, school support information, problem-identification and problem-solving skills	Problem-solving	Face-to-face (role-play, problem-solving) telephone teaching	4 weekly sessions plus 3 booster phone call sessions. 75–90 min/in-person session &30 min/booster session	Outpatient, telephone	Psychology students	Intervention materials were developed based on focus group feedback and reviewed by a focus group of parents and CYP with SCD.
Dobson and Byrne[38]	guided imagery education and skill training	Cognitive-behavioural theory &Bandura self-efficacy theory	In-person, individual-based, Face-to-face teaching, audiotapes	One session, 15–45 min	Outpatient	Child life specialist	Not specified.

Dobson[39]	guided imagery education and skill training	Cognitive-behavioural theory &Bandura self-efficacy theory	In-person, individual-based, Face-to-face teaching, audiotapes	One session, 15–45 min	Outpatient	Child life specialist	Not specified.
Estepp et al.[40]	Daily text reminders about taking hydroxyurea. Participants can reply to the system	Not specified	Telephone-based, individual-based	12 months	Remote (telephone-based)	Automated	Text message content customized for each CYP participant. No PPI or CYP involvement in the intervention development. Intervention developed by clinical informatics staff.
Fouda et al.[41]	SCD education, self-care practices/skill training	Not specified	Face-to-face teaching (group discussion, role-play), printed materials	6 sessions over 3 days, 30–60 min/session	Outpatient	Research staff (nursing background)	Developed by researchers based on the assessed needs of CYP with SCD. No PPI or CYP involvement.
Gil et al.[42]	Cognitive behavioural skill training for coping with pain (breathing, relaxation, guided imagery, calming self-statements)	Not specified	Face-to-face, individual-based, in-person teaching, prerecorded audiotapes for self-directed practice, homework assignments, phone calls	2 sessions. First session 45 min, second session a week later to review skills taught/learned	Outpatient, remote (telephone)	Research staff (psychology background) plus self-directed practice for 1 week	Modelled after one used in an SCD adult population. PPI or CYP involvement not specified.

Gil et al.[43]	Cognitive behavioural skill training for coping with pain (breathing, relaxation, guided imagery, calming self-statements)	Not specified	Face-to-face, individual-based, in-person teaching, prerecorded audiotapes for self-directed practice, homework assignments, phone calls	In-person session—45 min. Weekly telephone contacts for 4 weeks	Outpatient, remote (telephone)	Research staff (psychology background) plus self-directed practice for 1 month	Modelled after one used in an SCD adult population. PPI or CYP involvement not specified.
Green et al.[44]	SCD and hydroxyurea education, adherence barriers identification, action planning, problem-solving skills, automated medication text reminders	Not specified	Face-to-face teaching, text messages, phone calls, printed brochures	5 sessions, 60–90 min/session	Outpatient, remote	Community health workers	Not specified.
Hazzard et al.[45]	SCD education, recreational activities (games, arts, crafts) to enhance coping and social support		Face-to-face teaching, web-based (video conferencing, chatrooms, e-games, arts, crafts	Up to the time of hospital discharge	In-patient, web-based	self-guided. Healthcare professionals present educational activity (15–45 min).	Pre-existing programme with the educational component adapted for CYP with SCD. Adapted component reviewed by a team of clinicians. No PPI or CYP involvement in the adaptation process.

Hood et al.[46]	Pain monitoring/tracking, skill training (goal setting, goal evaluation), social support (group formation and competition)	Not specified	Digital, Mobile app, individual-based	6 weeks	Web-based (digital)	Self-directed	3-phased development process (survey, design and usability testing). CYP involved in all the development phases.
Kaslow et al.[47]	SCD education, pain management techniques, problem-based coping practices	Stress-coping-adjustment framework	In-person, family-focused, Face-to-face teaching (role-play, games), homework, printed materials	6 weekly sessions, 60 min/session	Outpatient	Psychology students	Input from clinicians and a SCD family member. No CYP involvement.
Ketchen et al.[48]	SCD education, skill training (interpersonal communication, self-expression, distraction-effective elevation)	Not specified	Web-based (video conferencing, chatrooms, e-recreational activities), a CD-ROM game, homework	6 weeks intervention	In-patient, home, web-based	Self-guided. Weekly reminder phone calls from the research team	Pre-existing programme used without adaption for CYP with SCD. Programme development not identified.
McClellan et al.[49]	Cognitive-behavioural coping skills training for pain management (deep breathing, guided imagery, relaxation)	Not specified	In-person, family-based, face-to-face teaching (participatory activities and demonstrations), printed materials, e-modules, telephone calls, e-mails	One 120 min in-person session plus digital engagement over 2 months and weekly telephone calls within the month	Outpatient, web-based	Psychologists, self-directed	Not identified

Palermo et al.[50]	Cognitive and behavioural skill training for pain management (relaxation techniques, healthy lifestyles, operant strategies for adaptive behaviours), goal setting	Cognitive behavioural theory	Web-based, family-based, e-modules (presented through videos, vignettes, activities, illustrations), e-mail contact	8 weekly e-modules	Web-based	Self-directed	A generic pre-existing programme used without adaption for the study population. Intervention development not detailed.
Phillips et al.[51]	SCD education, pain tracking/monitoring, pain management behaviour modification	Paediatric self-management model	Digital, mobile app	Intervention duration—12 weeks	Web-based	Self-guided	CYP with SCD and their parents or caregivers reviewed the intervention and it was revised based on the feedback.
Rodgers-Melnick et al.[52]	SCD education, pain management, medication management, skill training (relationship-building), peer support	Not stated	In-person, group-based, group-drumming, songwriting, music production/creation, printed action plans,	4 sessions every 3 months for 12 months	Outpatient	Multidisciplinary team (haematologists, nurse practitioner, social worker, paly patient navigator, music therapist)	Incorporated resources (song stories) developed with CYP with SCD.

Saulsberry et al.[53]	SCD information and transition skills	Not specified	Web-based, individual-based, e-modules, videos, quizzes	6 modules, completed within 1 year	Outpatient	Healthcare professionals (nurse-led)	Pre-existing intervention adapted for CYP with SCD. Adaption involved a team of clinicians and hospital administrative staff. No PPI or CYP involvement.
Schatz et al.[54]	Cognitive and behavioural skill training for pain management (deep breathing, relaxation, guided imagery)	Not specified	In-person, individual-based, face-to-face teaching plus home-based practices and skill acquisition using a mobile app, written materials, telephone calls	One in-person session 45–60 min. 8 weeks of engagement with the mobile app	Outpatient, home, telephone	Psychology students. Self-directed	Modelled from a pre-existing intervention developed for SCD adult population. No PPI or CYP involvement in the adaption.
Schwartz et al.[55]	Cognitive and behavioural skill training for pain management (deep breathing, relaxation, guided imagery, positive self-statements)	Not specified	In-person, individual-based, face-to-face teaching plus home-based practices using prerecorded audiotapes	4 sessions, sessions 1–3 occur 2 weeks intervals, session 4 a month after 3. Each session is 90 min	Home and telephone-based	Psychology students and psychologists	Guided imagery content chosen by CYP participants but no PPI or CYP involvement in the development.

Sil et al.[57]	Psychological and behavioural pain management skill training	Dynamic adaption process model	In-person, group-based, face-to-face teaching (group discussion), videotapes, workbook exercises	3 sessions, 60 min/session over duration of hospitalization	In-patient	Psychologists	Pre-existing programme with the educational component adapted for CYP with SCD. Adapted component reviewed by a team of clinicians. No PPI or CYP involvement in the adaption process.
Sil et al.[56]	Cognitive and behavioural education and skill training for pain and illness management (education about pain, behavioural strategies for parents. Relaxation training, activity pacing, cognitive restructuring techniques, healthy lifestyle habits)	Cognitive-behavioural theory	In-person, individual-based, face-to-face teaching and skill acquisition	8 bi-weekly sessions, 45–60 min/session.	Outpatient	Psychologists	Intervention personalized for each CYP-parent dyad but no PPI or CYP involvement in the development.

Smaldone et al.[58]	SCD and hydroxyurea education, adherence barriers identification, action planning, problem-solving skills, automated medication text reminders	Not specified	Face-to-face teaching, text messages, phone calls, printed brochures	5 sessions, 60–90 min/session	Outpatient, remote	Community health workers	Not specified.
Smith et al.[59]	SCD education, information about transition policy, culture of adult medicine, school and work support and self-advocacy skill training	Not specified	In-person, group-based, individual-based, face-to-face teaching	3 years	Outpatient	Multidisciplinary team (medical providers, educational coordinator, clinical psychologist, social worker, transition coordinator)	Not specified.
Treadwell and Weissman[60]	Medication (deferoxamine) information and management, general information (genetics, nutrition), social support	Not specified	In-person, group-based, face-to-face teaching, games, visit to a theme park	4 day-camp	Community	Lay leaders	Not specified
Viola et al.[61]	Educational, skill training and social support mentorship programme focused on transition	Social-ecological model of adolescent and young adult readiness to transition (SMART)	Web-based, individual-based communication via video conferencing and text messaging	Monthly video calls and weekly text messaging for 6 months. Video call last 25–60 min.	Web-based	Medical students	Content informed by formative need assessment of CYP population.

Wihak et al.[62]	Cognitive and behavioural pain management training (breathing, relaxation, guided imagery), skill training (building confidence and positive thoughts)	Not specified	In-person, individual-based, face-to-face teaching, audio-visual resources, workbook	One-off, 150 min	Outpatient, in-patient	Multidisciplinary team (haematology nurse, child life provider, social worker)	PPI in the 4-phased adaptation process of a pre-existing programme. PPI included clinicians, patient advocate and community consultant. CYP involvement not specified.
Yoon and Godwin[63]	SCD information, self-care and pain management	Lieberman's theory on interactive health games and health outcomes in children	CD-ROM, individual-based, e-game	3 rounds, one-off. Intervention duration not indicated	Outpatient	Self-directed	Pre-existing programme with the educational component adapted for CYP with SCD. Adapted component reviewed by a team of clinicians and no PPI/CYP involvement.

Abbreviations: CYP, children and young people; SCD, sickle cell disease.

Format and delivery methods

Twelve interventions were delivered both in-person and remotely,[29,30,32,37-39,42-44,47-49,54,58,62] combining lectures, group activities, printed materials, prerecorded audio-visuals, telephone calls and online features.Combining different delivery methods aimed at promoting flexibility, convenience and financial accessibility, as well as including methods to engage young people. Twelve interventions were technology-based (e.g., mobile apps, virtual chatrooms, text messaging, e-modules, electronic or online games, e-diaries).[33,34,36,44-46,48-51,53,54,58,61,63,64] Some of these interventions included symptom monitoring and tracking programmes aimed to change and reinforce behaviours believed to promote medical adherence and pain management.

Most interventions were delivered on an individual basis, with only eight interventions (21.1%) being group-based.[27,28,35,37,52,57,59,60] Intervention duration ranged from 45 min[38,39] to 36 months.[59] Interventions were delivered as one-off sessions, multiple sessions or were self-guided. Most were either delivered at a hospital (*n* = 16)[27,28,31,38,39,41-43,45,47,52,53,55,56,59,63] or at multiple locations (*n* = 12) (internet, hospital, telephone or community/home[29,32,36,37,44,48,49,54,57,59,62,64]). One intervention was delivered at a camp.[51]

Most interventions (*n* = 29, 76.3%) were delivered by the researchers themselves or health and social care staff or students, who were predominantly from a psychology background. Only two interventions were delivered by lay counsellors.[35,60] Some studies explicitly detailed the training and supervision of interventionists and measures to maintain and monitor intervention consistency and fidelity.[30,37,44,47,54,55,58,61]

Intervention development

Several articles (*n* = 13) explicitly stated the frameworks underpinning the interventions' development or implementation. These were predominantly psychosocial and behavioural change theories (Table 8). Only one intervention was underpinned by a self-management model[48] and none by the self-care management model for SCD.[65] Information on intervention sources or development methods was provided for 27 (84.4%) of the 32 interventions. Thirteen were new interventions developed for the study,[27,29,33,34,36,37,41,46,47,51-53,59,64] six were adapted from existing generic (noncondition specific) self-management programmes,[33,40,41,48,49,56] four were self-management existing programmes for SCD,[42,43,54,63] and three were generic self-management interventions implemented without adaptation for SCD.[45,48,50]

Some studies reported involving different lay and professional stakeholders in intervention development or adaptation. However, only five (18.5%) studies involved CYP with SCD in developing their self-management programmes.[33,34,36,37,46,51,61,64] Of these, one intervention described a co-creation process where CYP were engaged throughout the development stages.[34,36,46,64] In the remaining four studies, CYP with SCD appear to have been involved at the initial needs assessment stage[61] or the review stage following the intervention development.[33,37,51] CYP involvement appeared limited to intervention content.

In all the studies where race and ethnicity were identified, the SCD population were of African American background. There appeared to be an assumption that participants would be of low socioeconomic status and have strong family systems due to their racial and ethnic identity. These assumptions influenced the intervention format, delivery and target population, with attempts to make interventions financially accessible and/or involve family relatives. Some studies also highlighted the black culture of the SCD population and included black music, sports celebrities, mentors, navigators, interventionists and content from black TV shows to make them ‘culturally sensitive’.[37,47,49,55,61,62] The agency of CYP, however, appeared to be overlooked, with most interventions developed with minimal or no CYP involvement (see Table 8) to tailor them to the values, expectations and hopes of the CYP. All the interventions were individual- or family-focused, aimed at knowledge, skill acquisition, reinforcement and/or behaviour change, underpinned predominately by psychological models (see Table 8). There was little or no consideration for CYP's wider social contexts like school and/or workplace.

Outcomes measurement

Thirty-one studies assessed the effectiveness of self-management interventions using multiple outcome measures (Table 9). Fifteen were feasibility/pilot studies that assessed and reported intervention effectiveness as primary outcomes.[28,35,36,40,44,46-48,53,57,58,60,61,63,64] The most commonly assessed outcomes were disease knowledge, coping, self-efficacy, quality of life, family and social functioning and support, self-management confidence and behaviours, healthcare utilization, treatment adherence, parent–child relationship and transition readiness and success. Twenty-four papers reported on intervention acceptability and feasibility and/or implementation issues. No studies provided data on cost-effectiveness or intervention development and/or implementation costs.

Table 9 Summary of the effectiveness of interventions on outcomes

References	Outcome	Measure	Result or finding (d, day; w, week; m, month)	Study quality rating
Abd Elaziz and Abd Elghany[27]	Knowledge, Pain level, Pain Coping Strategies, Self-Care Practices	Verbally administered Numerical Rating Scale for pain assessment. Other outcomes were measured using checklists designed for the study.	*3 d*: Improvement in outcomes (p <.001)	Moderate

Adegbolagun et al.[28]	Anxiety, Depression, Coping Strategies, Quality of Life (QOL), Knowledge	Hospital Anxiety and Depression Scale, Coping Skills Questionnaire for Sickle Cell Disease-Adult Version, SF-36, Knowledge Questionnaire (designed for the study)	*1 w*: Reduction in anxiety (*p* <.001) and depression (*p* <.02). Improvement in active and passive coping skills (*p* <.004), affective coping skills (*p* = .58), self-reported knowledge, the practice of coping skills and confidence (*p* <.0001)	Moderate
Allemang et al.[29]	Lost to Follow-up, Appointment Attendance Rate, Medication Adherence, Hospitalization	Medical Records and self-reported checklists	*12 m*: Reduction in the proportion of transition patients lost to follow-up (*p* = .00034). Improvement in appointment (*p* = .096), medication adherence (*p* = .047) and hospitalization rates.	Moderate
Barakat et al.[30]	Pain (presence, healthcare use, medication), School Attendance, Health-Related Hindrance, Coping Strategies, Knowledge, Teen and Family Physical and Psychosocial Well-being, Family Cohesion	Pain Diary (designed for the study), Health-related Hindrance Inventory (designed for the study), Coping Strategies Questionnaire, SCD Knowledge Questionnaire, SCD Transition Knowledge Questionnaire (designed for the study), Disease Self-Efficacy Scale (adapted for this study from a cancer-specific scale), Child Health Questionnaire	*10 w, 12 m*: No difference in outcomes between groups	Poor

Broome et al.[31]	Coping	School-agers' Coping Strategies Inventory, Adolescent Coping Orientation for Problem Experiences	*6 w, 12 m* (children): No difference in the frequency of use of coping strategies among the groups. Increase in the number of strategies used and rated effective over time in the intervention groups (*p* <.0003). Nonsignificant reduction in clinic and ER visits and admissions over time. *Last session, 12 m* (adolescents): increase In the frequency of use of coping strategies (*p* = .018). decrease in ER and clinic visits (*p* = .05) and admissions (*p* = .03)	Moderate
Crosby et al.[33]	Clinic Attendance, Hydroxyurea Adherence	Barriers to Care Questionnaire, Web-based Assessment Tool (Take Charge Programme), Medical Record Review	No real data on the impact of the intervention reported	Moderate
Crosby et al.[35]	General and Disease-Specific Self-Efficacy, Self-Management Behaviours, Health Status, QOL, ER Visits, Patient Activation	SCSES, Self-Efficacy for Managing Chronic Disease Scale, Transition Readiness Assessment Questionnaire, National Health Interview Survey, Patient Activation Measure, Pediatric QoL Inventory	*6 w, 3 m, 6 m*: significant improvement in general self-efficacy (*p* = .015) but not in disease-specific self-efficacy (*p* = .293), self-management behaviours (*p* = .39)), health status (*p* = .30), quality of life (*p* = .17) and ER visits (*p* = .59). Medium effect size was noted for patient activation (*p* = .70).	Moderate
Crosby et al.[64]	Health-Related Quality of Life (HRQOL), Self-Efficacy, Self-Management Skills, Knowledge, Health Motivation	Patient Activation Measure, Transition Readiness Questionnaire, Disease-Specific Knowledge Questionnaire, Treatment Self-Regulation Questionnaire, Paediatric QoL Inventory SCD Module	*2 w*: Higher scores in quality of life (*p* = .01). Medium effect size in self-efficacy (*p* = .90). No difference in health motivation, self-management skills and knowledge (*p* >.05).	Moderate

Cozzi et al.[32]	Self-Concept, Pain, Anxiety, In-patient Hospital Visits, Emergency Room (ER) Visits	State-Trait Anxiety Inventory for adults or children, Tennessee Self-Concept Scale for adults, Piers-Harris Children's Self-Concept Scale, Human Drawings Test	*13 w*: Nonsignificant change in in-patient and emergency room visits and self-concept. Improvement in anxiety for children ($p <.01$) and adolescents ($p <.05$); reported headaches as crises symptoms ($p <.01$); reported pain medication days ($p <.001$); reported pain intensity ($p <.001$) and self-treated crises ($p <.05$).	Moderate
Daniel et al.[37]	HRQOL, School Attendance, Access to School Resources	Haematology/Oncology Psycho-Educational Needs Assessment, Paediatric QOL Inventory, Woodcock-Johnson III, Wechsler Abbreviated Scale of Intelligence, Social Problem-Solving Inventory-Revised Short Form	*4 w, 6 m*: No impact on all the outcomes.	High
Dobson and Byrne[38]	Self-Efficacy, School Attendance, Analgesic Use, ER Visits, Pain Intensity	Pain Assessment Tool (investigator-adapted), Sickle Cell Self-Efficacy Scale (SCSES), Pain Diary (designed for the study)	*1 m*: improvement in self-efficacy score ($p = .000$) and pain intensity ($p = .003$), nonsignificant reduction in missed school days, use of analgesics and acute care visits.	Moderate
Dobson[39]	Disease-specific Self-Efficacy	SCSES	*1 m*: Greater disease-specific self-efficacy ($p <.000$)	Poor
Estepp et al.[40]	Hydroxyurea Adherence, Hospitalization Rates, Clinic Visits	Medication Possession Ratio (days medication in family's possession/days prescribed medication), Haematologic Laboratory Parameters (HbF levels, mean corpuscular volumes, bilirubin levels, absolute reticulocyte counts), Medical Record Review	*3–12 m*: Significant improvement in haematological parameters ($p <.03$) and ER attendance ($p = .0013$). Nonsignificant improvement in hospitalization rates ($p = .79$) and medication possession ratio ($p = .99$). A change in participant's medication possession ratio was the only predictor of improved HbF levels after intervention initiation	Moderate

Fouda et al.[41]	Knowledge, QOL, Self-Care Practices	Paediatric QOL Inventory. Other outcomes were measured using checklists designed for the study	*3 d, 3 m*: improvement in all outcomes ($p \leq .05$). Positive correlation between knowledge and QOL and reported self-care practices and QOL ($p \leq .001$)	Moderate
Gil et al.[42]	Pain Sensitivity, Coping Strategies	Forgione-Barber Focal Pressure Stimulator, Coping Strategies Questionnaire for SCD	*1 w*: Lower negative thinking scores ($p < .05$) but nonsignificant interaction effects for coping attempts and illness-focused strategies. Less pain during low levels of laboratory pain stimulation ($p < .02$). positive and significant correlation between improvement in coping strategies (negative thinking) and improvement in pain sensitivity ($p < .01$)	Poor
Gil et al.[47]	Depression, Anxiety, Pain (sensitivity, sleep quality, functionality, healthcare contacts) Coping Strategies	Children's Depression Inventory, Revised Children's Manifest Anxiety Scale, Pain Diary, Coping Strategies Questionnaire, Structured Pain Interview	*2–3 w, 1 m*: nonsignificant improvement in all outcomes ($p > .05$)	Poor
Green et al.[44]	Hydroxyurea Adherence	Prescription Refill Adherence, Morisky Self-Report Scale, Foetal Haemoglobin (HbF) level	*1–6 m*: Nonsignificant improvement in pharmacy refill ($p = .33$) and self-reported medication adherence. Less decrease in Personal best HbF in the intervention group compared to controls ($p = .009$). Controlling for intervention month and group assignment, the intervention group, progressed to Personal best by 2.3% per month during Months 0–4 ($p = .30$).	Moderate
Hazzard et al.[45]	Knowledge, Perceived Social Support, Coping Strategies	How Much Do I Know About SCD Scale, Perceived Social Support-Friends Scale, Kidcope	*At hospital discharge*: Significant effect on all outcomes ($p < .001$).	Moderate

Hood et al.[46]	Self-Efficacy, Self-Management (goal and confidence)	Transition Readiness Assessment Questionnaire, Patient Activation Measure	*6 w*: Nonsignificant improvement in self-efficacy ($p > .05$). improvement in self-management confidence, goal-setting and progress tracking but the significance of improvement is not indicated	Poor
Kaslow et al.[47]	Knowledge, Internalizing and Externalizing Behaviours, Family/Social Functioning and Support	Children's Depression Inventory, Family Adaptability and Cohesion Evaluation Scale, Child Behaviour Checklist, Sickle Cell Disease Knowledge Test (designed for the study)	*6 w*: Improvement in outcomes after the last session ($p < .01$). *6 m*: Improvement in CYP's knowledge sustained. Improvement in parental knowledge and internalizing behaviours reduced ($p < .05$)	High
Ketchen et al.[48]	Knowledge, Health promotion Activities, QOL, Depression, Parent-Child Relationship Quality	How Much Do I Know About SCD Scale, Health Promotion Activities Checklist (designed for the study), Paediatric QOL Inventory, Children's Depression Inventory, Interaction Behaviour Questionnaire	*At hospital discharge, 2 m*: No significant effect on knowledge, health promotion and depressive symptoms. Significant improvement in QOL ($p = .05$) and parent-child relationship ($p = .02$))	High
Rodgers-Melnick et al.[52]	Self-Efficacy, Trust, Knowledge, ER Visits, Hospitalization	SCSES, Seidman Sickle Cell Knowledge Quiz (designed for the study), Wake Forest Trust in Medical Profession Scale	*3–12 m*: Significant improvement in knowledge ($p = .0002$). No effect on self-efficacy ($p = .37$), trust ($p = .08$), ER visits ($p = .33$) and hospitalization ($p = .2$)	Poor

Saulsberry et al.[53]	Knowledge, Self-Management Confidence	Disease Knowledge Tool, Self-Management Skills Checklist (designed for the study)	*12 m*: Median self-management confidence score was 8 (range 5–10), and the median knowledge assessment score was 79 (range 37–100). A positive correlation between the number of modules completed and the disease knowledge score ($p = .003$). No correlation was found between the number of modules completed and the self-management confidence ratings ($p = .945$).	Moderate
Schatz et al.[54]	Coping Strategies, Pain (frequency, intensity, functionality)	Coping Strategies Questionnaire for SCD, Daily Pain and Activity Diary (electronic)	*2 m*: Increase in self-reported use of cognitive-behavioural skills ($p = .03$) and beliefs in pain controllability ($p = .02$).	Moderate
Sil et al.[57]	Knowledge, Pain Coping Efficacy	Knowledge of Psychological Interventions for Pain, Child Pain Coping Self-Efficacy Scale	*End of each session*: Only significant improvement postintervention for session 1 (<.01). No significant difference in coping scores and self-efficacy remained relatively similar across all sessions	Moderate
Sil et al.[56]	Healthcare Utilization, Treatment Adherence, Pain Intensity, Functional Disability, Pain Coping Efficacy	Medical Record Review, Numeric Rating Scale (designed for the study), Functional Disability Inventory, Pain Coping Questionnaire	*Every 2–3 sessions, 8 w*: No significant improvement in healthcare utilization ($p >.17$). 31.6% of participants in the treatment group reported a statistically significant decrease in pain intensity, functional disability and improved coping efficacy.	Poor

Smaldone et al.[58]	Hydroxyurea Adherence, HRQOL, Youth-Parent Self-Management Responsibility Concordance	Paediatric QOL Inventory, Paediatric QOL SCD Module, Sickle Cell Family Responsibility Scale (adapted from the Diabetes Family Responsibility Questionnaire)	*3 m*: Improved dyad self-management responsibility concordance (3.5 points, 95% CI: −0.2, 7.1). *6 m*: Improved generic HRQOL (9.8 points, 95% CI: 0.4, 19.2), No difference in QOL	Moderate
Smith et al.[59]	Transition Readiness, Disease-Related Self-Efficacy, Disease-Related Stress, Feelings about Transition, Transfer Success, Patient Retention	Transition Intervention Programme-Readiness For Transition Assessment Tool (designed for the study), SCSES, Feelings-Sickle Cell Transfer Questionnaire, Sickle Cell Stress Scale-Adolescent (designed for the study)	*36 m*: Improvements in all outcomes but statistical/clinical significance not indicated	Poor
Treadwell and Weissman[60]	Treatment Compliance, Knowledge, Perceived Social Support, Parent-Child Responsibility for Medication Management	24-h recall interview, Multi-Choice Questionnaires (designed for the study), Perceived Competence Scale for Children	*2 w*: Nonsignificant improvement in outcomes	Moderate
Viola et al.[61]	Transition Readiness, Disease-Related QOL, Disease-Related Self-Efficacy, Health Literacy, Knowledge	Sickle Cell Transition Intervention Programme-Readiness for Transition, Adult Sickle Cell QOL Measurement System, SCSES, Morisky Medication Adherence Scale	*6 m*: Significant improvement in transition readiness ($p < .01$) and self-efficacy ($p = .002$). Nonsignificant improvement in QOL, health literacy and knowledge	High
Yoon and Godwin[63]	Knowledge, Self-Management Confidence	Knowledge checklist (designed for the study), 100 mm horizontal Visual Analogy Scale	*0 d*: improvement in knowledge ($p = .010$) and self-management confidence ($p = .001$)	Moderate

Abbreviations: CYP, children and young people; HRQOL, health-related quality of life; QOL, quality of life; SCD, sickle cell disease.

Outcomes were measured at different time points between the final intervention session and 12 months postintervention implementation (Table 9). Only four studies assessed outcomes over a longer period.[30,31,52,60] Three studies measured several CYP-focused outcomes via proxy reports.[27,51,58] These studies also employed measurement tools not validated for CYP. Further, several studies involving self-guided interventions did not assess engagement levels, and only two[48,53] accounted for the connection between engagement and outcomes. No studies reported if CYP with SCD participated in deciding how self-management interventions' effectiveness, acceptability and feasibility should be evaluated.

Different validated and unvalidated generic and SCD-specific outcome measurement tools were used (Table 4). Most tools had not been formally evaluated for use with CYP with SCD, and different tools were used to measure the same outcomes across the papers (Table 9). Only three papers[44,58,60] reported the measurement of physiological parameters (Table 9). Most of the studies reporting intervention effectiveness failed to report on the effect size and/or 95% confidence intervals of the results. The evidence below must, therefore, be considered in the context of these limitations. Table 9 summarizes the effectiveness data.

Effectiveness

Seventeen studies (20 articles) evaluated the impact of self-management programmes on psycho-behavioural outcomes (self-efficacy, anxiety, depression and coping).[28,30-32,34,36,38,39,42,43,45-48,52,54,57-59,61] Findings from five studies associated self-management interventions with statistically significant positive impacts on self-efficacy.[28,38,39,61,64] In one study, improved self-efficacy was correlated with improved self-management confidence and behaviours.[46,64] Similarly, five studies reported significant improvements in coping and the use of positive coping strategies.[27,28,45,54,56] Coping outcomes were reported primarily for pain. Mixed findings were reported regarding self-management interventions' impact on pain frequency and intensity.[30,37,42,43,49,54-56] Only one study reported significant improvement in anxiety and depression postintervention.[28] The effective interventions for psycho-behavioural outcomes were predominantly in-person, hospital-based and multisessional.

The overall strength of the evidence base for the psycho-behavioural outcomes limits confidence in the results, particularly since 16 of the 20 articles were of moderate to poor methodological quality. Only one high-quality study associated the self-management intervention (e-mentorship by medical students) with significant improvements in self-efficacy.[61] Furthermore, effectiveness appeared to reduce overtime for self-efficacy and coping.[30,31,47,52]

Eight studies of mixed quality examined the impact of self-management interventions on healthcare utilization which included emergency room visits, in-patient admission duration and/or hospitalization rate.[29,30,32,35,38,40,52,56] Only a 12-month CYP-personalized transition programme delivered by a designated social worker reported a statistically significant reduction in hospitalization rate at 12 months follow-up.[29] This study involved the largest number of participants (*n* = 110); however, outcome measurement was based on a retrospective record review, which would not have included hospitalizations outside the study centre.

Health-related quality of life was measured in eight studies.[28,35-37,41,48,58,61] Only three studies of moderate quality reported statistically significant improvement in quality of life.[28,41,64] However, in two, improvement was only observed in the social domain.[28,64] The high-quality studies identified no significant improvement in quality of life.[37,61]

Overall, our greatest confidence is in the evidence of improvements in SCD knowledge and social outcomes, where studies of predominantly moderate methodological quality reported positive outcomes (Table 9). Sixteen studies of strong to moderate quality reported a statistically significant impact of self-management interventions on these outcomes.[27,29,36,38,40,41,44,45,47,48,52,57-61,63] Eight studies found that educational programmes on SCD and pain improved knowledge and understanding.[27,40,47,52,57,60,63,64] Six studies found a positive association between improved knowledge and understanding and self-management confidence, behaviours and practices.[27,41,46,53,63,64] Similarly, six studies also found improvements in social functioning and perceived social support.[38,45,47,48,59,60] Although evidence on knowledge and social outcomes is based upon high-quality studies, many used different unvalidated measurement tools and had small sample sizes and short-term follow-ups.

Six studies measured medical adherence as medication adherence, CYP responsibility for treatment, clinic attendance or transition success.[29,40,44,58,60,61] Four studies reported significant improvements in these outcomes.[29,44,58] The effective interventions were behavioural and skills training programmes delivered by a designated social care provider[29,44,58] and an educational programme delivered by lay counsellors.[60] None of the technological interventions using treatment reminders improved treatment adherence.[40,61]

Acceptability and engagement

Ten RCTs,[30,37,43,44,46,47,49,50,55,58] eight quasi-experimental studies,[28,32,45,51,52,57,61,62] three cross-sectional studies,[33,35] two cohort studies[40,53] and one qualitative study[64] reported on participants' satisfaction and engagement with the interventions. Levels of satisfaction and engagement were measured by survey and were assessed only for participants who completed the interventions. Studies reported high levels of satisfaction and acceptance, with those associated with the highest satisfaction levels being interventions that were delivered in person or remotely;[28,30,32-37,44,47,51,55,61,62,64] involved peers with SCD or parents[32,34,45-47,52] and supported pain coping skills and strategies,[30,32,34,46,47] SCD knowledge and understanding,[30,34,46,47,52] advocacy skills[46] and direct interaction with care providers.[40,61] Participants valued interventions that were engaging, interactive, motivating and enjoyable.[33-37,46,52,64]

However, participants' engagement with digital interventions was reported to be variable, decreasing over time. Engagement with interventions was reported to be higher amongst parent participants than CYP for the interventions that involved CYP-parent dyads.[44,51,58] In addition, engagement levels were reported to be higher among younger CYP participants than in the older CYP cohort in the same studies.[34,43,46,47]

Some studies reported on CYP participants' feedback regarding the negative aspects of the interventions and their preferences.[44,47,50,58,61,64] Negative aspects included the burden of home-based self-directed activities,[47] the lack of mobile phone-based technology interventions,[50,64] the frequency of reminder notifications and the surveillance features of some interventions.[44,50,58,61] Interestingly, participants recommended incorporating in-person sessions for interventions involving virtual platforms for social interaction and strengthening peer support.[61,64] Some studies described how the professionals delivering the interventions[31,61] reported improved communication and relationships with CYP and improved understanding of their challenges.

Evidence related to self-management intervention implementation

The main challenges reported for intervention implementation related to study recruitment and retention and provider (interventionist) availability. Eleven studies reported high follow-up attrition rates, which ranged between 32%[40] and 53%[57] (Table 2). Participants' responses for not completing the intervention and/or outcome assessments included the burden of completing outcome measures,[44] loss of interest,[37,52,54] dislike of intervention format,[50,54] acute illness and hospitalization,[35,48,57] and transport and time constraints.[48,56] Some eligible participants were excluded or lost to follow-up due to the intervention times being limited to the clinic's working hours. Barriers to participation and engagement in the m-Health interventions included device and technical malfunctions.[46,49,54] Implementation facilitators included patient navigation systems, study schedule flexibility, parental, family and clinical staff involvement, third-sector organization partnership and accessible m-Health programmes.[31,34,37,46,48,51,52] The only healthcare system implementation issue reported was provider availability[57,59] when competing for clinical demands and emergency consultations often interfered with intervention delivery. Training each multidisciplinary team member to deliver self-management interventions was reportedly facilitative. Several studies made recommendations to facilitate future intervention acceptability and engagement (Table 10).

Table 10 Recommendations for future self-management interventions

Self-management programmes/interventions need to:	m-Health self-management interventions need to:

•Be personalized to CYP's goals and values.[34,45,46] •Be family-oriented.[30] •Use multiple formats and delivery approaches to enhance flexibility, convenience, acceptability, usefulness and engagement.[37,50] •Include components to facilitate communication with health and social care professionals.[50] •Develop the broad range of knowledge and skills for managing SCD.[50] •Support older CYP's capacity to navigate psychological, relational, educational, vocational, economic and health and social care system challenges.[29,30,43,47,64] •Be integrated within the wider healthcare system.[46] •Involve schools.[47] •Designate a lead for self-management in the multidisciplinary team to support CYP.[30,59,61,62]	•Employ user-centred design.[51] •Mitigate risks to privacy.[46] •Consider time, resources, monitoring and technology maintenance.[46] •Include features to allow CYP to self-assess their progress and intervention impact.[51] •Help CYP communicate health, illness and self-management information with health care professionals by enabling them to capture visual feedback in real-time to share.[51]

Abbreviations: CYP, children and young people; SCD, sickle cell disease.

DISCUSSION

The development and evaluation of self-management interventions to support CYP with SCD is a growing area of research, particularly in the United States, with 50% of the included papers being published in the last 5 years. This paper reports the first comprehensive review of the evidence on their effectiveness and acceptability. In terms of their characteristics, the interventions developed are wide-ranging in relation to their purpose, content, format and delivery methods. However, only a third of these (10/32) demonstrated statistically significant improvements in the outcomes measured.[27-29,38,39,41,45,47,54,63] These interventions were delivered in-person, involved educational components, skills training for pain management and medical adherence and/or promoted family and/or peer support; and reported improvements in knowledge, social functioning and/or medical adherence. However, it is unclear if these improvements are maintained over time. Similar to the findings of previous reviews on CYP with chronic conditions,[66,67] the majority of the included interventions were focused on medical management (i.e., disease-specific and healthcare-related) and/or emotional management (i.e., feelings and intrinsic characteristics). This notwithstanding, no clear effects were found in favour of any interventions aimed at symptom reduction.

Only a third of the technological interventions (4/12) showed effectiveness. These findings are consistent with the findings of other reviews in which limited data was found to support the effectiveness of mobile or web-based applications for self-management of adolescents with a chronic illness[68] and for treatment adherence among CYP and adults with SCD.[19,21] There was limited evidence for interventions improving healthcare utilization or quality of life. Conflicting and inconsistent evidence for the effectiveness of self-management interventions on quality of life in CYP with chronic illnesses has been found and reported by others.[66] In addition to previous research on the effectiveness of self-management for CYP with chronic illnesses,[66] this review revealed that Interventions that combined technological platforms and in-person group-based approaches and involved family and care providers appeared to be more acceptable to CYP.

However, confidence in these results is limited by the methodological quality of the included papers. Most interventions are in the early stages of evaluation, and most papers report findings from retrospective observational studies, single-arm pre- and poststudies or small feasibility/pilot trials. Several studies were characterized by high follow-up attrition rates and demonstrated significant potential risk for bias. There were inconsistencies and wide variations in the outcome measurements used across the studies, with some unvalidated tools. This reflects the current state of limited availability of validated health-related outcome tools for CYP with SCD specifically and the SCD population in general. Further, none of the studies compared the different types of intervention components or examined if specific self-management intervention components are more or less effective and for which patient and health and social setting characteristics, which would allow for creating targeted interventions.

Consequently, more robust definitive RCTs conducted across multiple and different settings are needed. Future trials should be methodologically robust in terms of randomization, larger sample size and power calculations, blinding and outcome measures (i.e., use of age-appropriate validated tools, effect size calculations, clinical significance indications and longer follow-up periods). Qualitative studies should be nested within future trials to help understand intervention effectiveness and acceptability. This is particularly important as patient-reported outcomes measures (e.g., quality of life, psychosocial well-being) are highly variable and qualitative methods will provide additional explanatory information that can facilitate the interpretation of trial outcomes. More economic evaluations of interventions are also needed. In addition, more research is needed to develop and validate health-related outcome measurement tools for CYP with SCD to support intervention and programme evaluations.

The lack of, or where sought, the minimal involvement of CYP in the intervention development and implementation and study design and conduct is a limitation of the included studies. This may explain why several interventions included features that CYP found burdensome and unhelpful, as well as problems with recruitment, retention and engagement. A recent review suggests that involving CYP in designing and delivering self-management interventions improves effectiveness.[69] Ignoring the agency of CYP, particularly from minority backgrounds like CYP with SCD, also has the potential to create and deepen health disparities and inequalities. This supports embedded co-production to ensure self-management intervention design and implementation are consistent with CYP's needs, voices, experiences and expectations rather than prioritizing medical adherence.

Further, interventions focused on role management (i.e., social domains and social participation) were limited. Despite the importance of the social context on self-management,[70,71] the included studies overlooked home, school and/or work environments. This is despite several research studies reporting the difficulties of engaging in SCD self-management practices in these settings.[9,11,13-15] The lack of accommodation for SCD self-management practices in the home, school and workplace has been highlighted, including how school policies and practices contradict self-management.[9,11,13-15] At the same time, self-management practices may be (dis)regarded as interference and perceived as disruptive and stigmatizing by others and CYP themselves.[9-11,13-15,72] Future interventions should, therefore, consider how families and school and/or work contexts can support CYP's ability to self-management.

Including CYP with SCD from diverse backgrounds or developing interventions inclusive of diverse cultures is important. However, no study included CYP with SCD from Eastern Mediterranean, Middle Eastern and Southeast Asian backgrounds. Indeed, SCD appeared to be ‘problematised’ as a ‘black illness’ in the included studies. Future research should, therefore, strive to involve CYP with SCD from non-Black backgrounds. This could be achieved through cross-country, cross-institutional and cross-centre research collaborations; development of interventions that are diverse (in terms of language and content); targeted recruitment of non-Black populations and more non-US-based studies.

Overall, the highlighted limitations mean that there is limited evidence for any single self-management programme or programme components in relation to promoting health and social outcomes for CYP with SCD. However, we found effective and acceptable interventions appear to include those that provide support from peers and trusted adults (e.g., parents, teachers, relatives and care providers). While the ultimate goal for self-management is to increase CYP with SCD autonomy and independence in managing their illness, they depend on others for encouragement, communication and treatment support. Adults (e.g., parents, relatives, care providers, teachers and adults with SCD) can provide information on how CYP can independently handle health and social care needs, while peers can learn from and support one another by sharing personal experiences. A recent review highlights the pivotal role peers and adults play in supporting the adoption and promotion of self-management strategies in CYP with chronic conditions.[69] Future interventions should, therefore, include components that facilitate peer and significant adult support. The involvement of peers, families, care providers and teachers is particularly important because CYP with SCD have been found to experience significant stigma in relation to their illness management, even from families, care providers and teachers due to their illness and race/ethnicity.[9,11,72,73]

STRENGTHS AND LIMITATIONS

This is the first review of self-management interventions for CYP with SCD that synthesizes the evidence from the range of programmes developed, the range of outcomes assessed and the diversity of evaluative study designs. Including young people with SCD in the review's conduct has enhanced the credibility of the process. However, there are some limitations. Only peer-reviewed articles published in English were included, posing a potential publication bias. It was impossible to synthesize study results using meta-analysis due to the heterogeneity of the interventions and outcome measures. As previously described, the robustness of the included studies limits the confidence that can be placed on the results. In addition, the cost-effectiveness or cost-benefits of interventions has not been evaluated, and there is a lack of qualitative research to inform understanding of intervention effectiveness and acceptability.

CONCLUSION

Findings generated from this systematic review provide a critical summary of the characteristics, effectiveness, acceptability and feasibility of self-management interventions for CYP with SCD. This review found that interventions that promoted peer, parent/family and care provider support and combined technology and in-person group methods appear to be associated with the effectiveness and acceptability to CYP. However, there is limited evidence for any single self-management programme or programme components in relation to promoting health/social outcomes for CYP with SCD. Future studies need to be robust in design, conducted across multiple and different settings and collect long-term outcome data to assess the effectiveness, acceptability, cost-benefits and generalizability of interventions across populations and settings. In addition, it is important that more validated health-related outcome tools are developed for CYP with SCD; CYP are involved in intervention development, study design and implementation; CYP participants from non-Black backgrounds are included and that attention is paid to the wider context of CYP's lives when designing and evaluating self-management interventions.

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CONFLICT OF INTEREST

The authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

Data sharing does not apply to this article as no data sets were generated or analysed during the current study.

DETAILS

Subject:	Intervention; Journals; Databases; Fatigue; Selfmanagement; Sickle cell disease; Chronic illnesses; Young adults; Social functioning; Acceptability; Design; Performance evaluation; Advisors; Pediatrics; Appraisal; Children; Social support; Research methodology; Feasibility; Systematic review; Social skills; Age; Psychosocial factors; Social environment; Effectiveness; Skill development; Medical personnel; Design standards; Social interactions; Qualitative research; Management; Research projects; Social education; Youth

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Developing a patient care pathway for emotional support around the point of multiple sclerosis diagnosis: A stakeholder engagement study

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ABSTRACT (ENGLISH)

Background

Diagnosing multiple sclerosis (MS) can be a lengthy process, which can negatively affect psychological well-being, condition management, and future engagement with health services. Therefore, providing timely and appropriate emotional support may improve adjustment and health outcomes.

Purpose

To develop a patient care pathway for providing emotional support around the point of diagnosing MS, and to explore potential barriers and facilitators to delivery and implementation.

Methods

Focus groups were conducted with 26 stakeholders, including 16 people living with MS, 5 carers/family members and 5 professionals working with people living with MS (3 MS nurses, 1 psychiatrist, and 1 charity staff member). Discussions were audio-recorded, transcribed verbatim and analyzed using framework analysis.

Results

Participants suggested that a patient care pathway should include comprehensive information provision as a part of emotional support at diagnosis, and follow-up sessions with a healthcare professional. Barriers including increasing staff workloads and financial costs to health services were acknowledged, thus participants suggested including peer support workers to deliver additional emotional support. All participants agreed that elements of a care pathway and embedded interventions should be individually tailored, yet provided within a standardized system to ensure accessibility.

Conclusions

A patient care pathway was developed with stakeholders, which included an embedded MS Nurse support intervention supplemented with peer support sessions. Participants suggested that the pathway should be delivered within a standardized system to ensure equity of service provision across the country.

Patient or Public Contribution

This research was conceptualized and designed collaboratively with Nottingham Multiple Sclerosis Patient and Public Involvement and Engagement (PPIE) group members. One member is a co-author and was actively involved in every key stage of the research process, including co-design of the pathway and research protocol, data collection (including presenting to participants and moderating group discussions), analysis and write-up. Authors consulted with PPIE members at two meetings (9 and 11 PPIE attendees per meeting) where they gave feedback on the research design, findings and the resulting pathway. People living with MS and carers of people with MS were included in the focus groups as participants.

FULL TEXT

INTRODUCTION

Multiple sclerosis (MS) is a chronic neurological condition, often diagnosed in mid-adulthood, and is the most common cause of nontraumatic neurological disability in working-age adults.[1] Symptoms of MS, which may include ‘visible’ (e.g., dexterity and mobility problems) and ‘invisible’ symptoms (e.g., fatigue, cognitive problems),[2] result from inflammation and demyelination of the central nervous system.[3,4] An estimated 2.8 million people live with MS worldwide, with around 107,000 new diagnoses each year[1]—a frequency that emphasizes the importance of delivering diagnostic news which meets the needs of an increasing clinical population. However, as there is no single, simple diagnostic test, diagnosing MS can be a lengthy process for individuals, which can cause confusion, relief, distress, and frustration.[5–7]

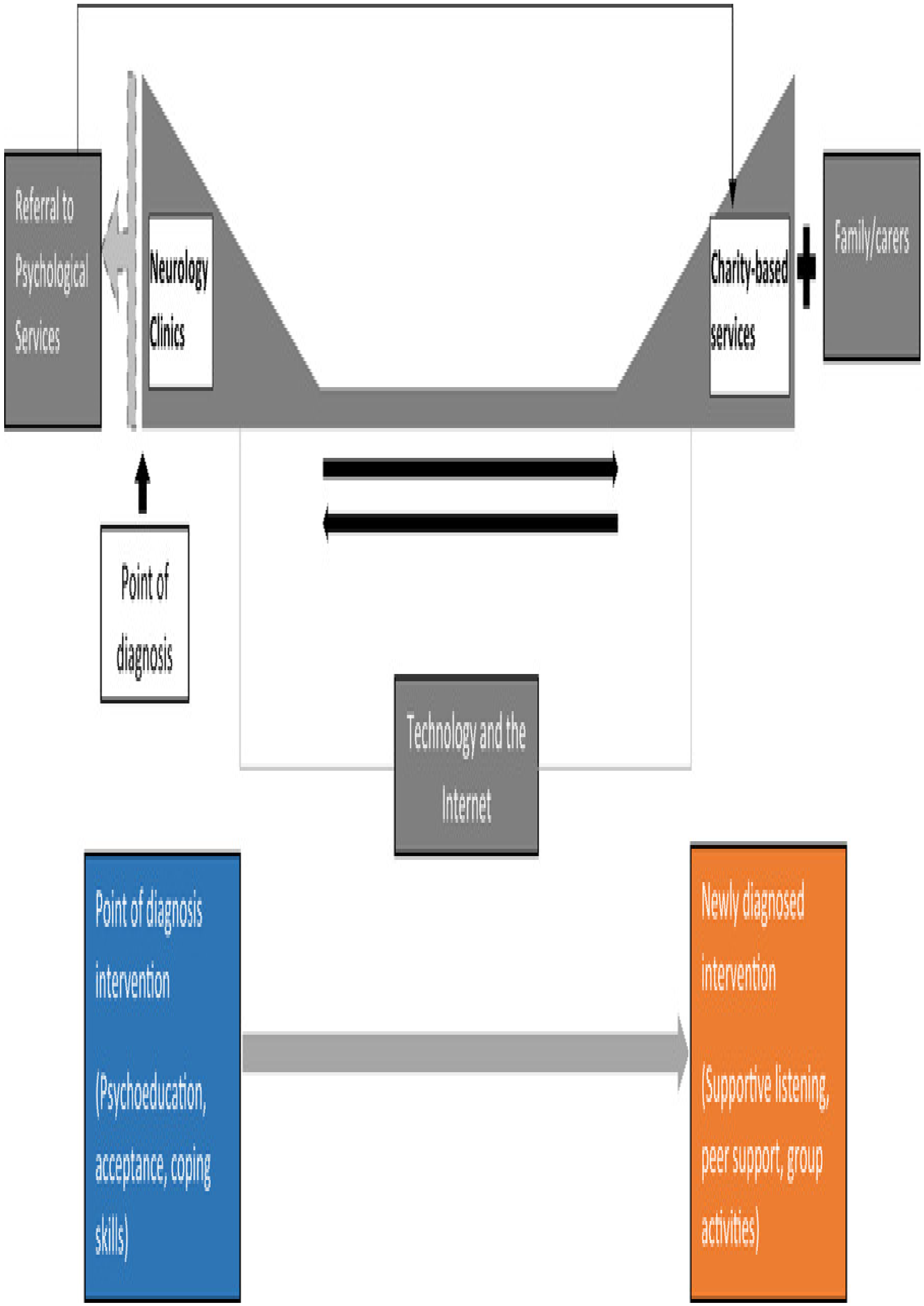
The general well-being of people with MS can be impacted by how they adapt to their changing health circumstances.[8] Psychological adjustment refers to the process of adapting to circumstances such as chronic disease and associated treatment,[9] whereby the individual aims to maintain equilibrium between competing environmental demands and the resulting stress. The way in which the period surrounding diagnosis is managed may determine how successfully a person adjusts to MS, influencing future perceptions of their condition, and may affect subsequent engagement with services.[6] The prediagnosis period in which symptoms are investigated can be particularly distressing due to the perceived uncertainty while awaiting diagnosis.[6] Qualitative evidence shows feelings of being misunderstood before the legitimization of individuals' condition by confirmed diagnosis, such that diagnosis produced feelings of devastation which conflicted with relief at being able to explain symptoms that were previously disbelieved by others.[6] Therefore, ensuring that people receive comprehensive information and support from the beginning of their lifelong MS journey may be crucial to facilitating positive psychological adjustment, while improving treatment outcomes and long-term management.

A recent meta-synthesis showed that many newly diagnosed people with MS had unmet emotional and informational needs during their diagnosis period.[10] In a qualitative study of experiences of adjusting to early-stage MS, many participants described feeling fear at being given a diagnosis and feeling overwhelmed by thoughts of impending doom.[5] However, they also felt that seeking positive, optimistic information increased their ability to accept the diagnosis and their perceived control over MS, while social support was regarded as critical for their adjustment.[5] Moreover, other qualitative research has suggested that providing adequate information about their condition and its treatment options at diagnosis may reduce feelings of anxiety and uncertainty.[6] This suggests that providing positively framed information coupled with social support may be key to facilitating successful psychological adjustment and that informational support is considered a part of emotional support.[5,6] Similarly, a recent metareview of systematic reviews on adjustment to MS suggested that professional support, information provision, continuity of care and peer support are factors throughout the diagnostic process which were linked to better psychological adjustment.[11]

The value of providing accessible information, suitable advice and support at diagnosis is well recognized.[12,13] However, current literature demonstrates that poor support and information provision has continued for people with MS throughout their diagnosis period[10,11] and should be part of emotional support.[5,6] A survey of people with MS in the United Kingdom identified that information provision at diagnosis was inconsistent.[14] Inadequate information provision has persisted over time and appears to be a common issue across Europe.[6,7,10,11,15] Findings from a meta-review showed that there are no adequate emotional support interventions that specifically target individuals newly diagnosed with MS.[11] Most resources dealt with providing information about MS (its causes, symptoms and treatment options), rather than broader emotional support including advice around living with MS. Furthermore, there are no established care pathways in the United Kingdom that include emotional support around MS diagnosis, and no referral systems to seamlessly incorporate wider ‘third-sector’ or charity-based support resources or services.[10]

The healthcare charity sector is an important source of support for many people with specific health issues. In the United Kingdom, the MS Society (www.mssociety.org.uk) and MS Trust (www.mstrust.org.uk) are trusted resources for many people with MS. Shift.MS (www.shift.ms) offers users peer support through its social network to connect with others with MS, helping people to make sense of their diagnosis and adapt to life with MS. These charities and support groups serve a useful adjuvant to the standard care patients receive from the National Health Service (NHS; e.g., providing emotional and social support) and in some cases, offer support that no one else provides (e.g., help with insurance, support for carers, social connection). In the UK context, however, the NHS and the charity providers for people with MS are somewhat disjointed, resulting in patients not receiving the best care they can receive. To address this gap, we aimed to co-construct a care pathway to provide emotional support to people around the point of MS diagnosis that linked NHS services with those provided by the charity sector.

The importance of theory in developing and evaluating complex interventions is well established.[16] Here, we propose an initial pathway that depicts the theoretical framework for providing emotional support around the point of MS diagnosis (Figure 1). This initial pathway was informed by reviews of literature,[10,11] pertinent theory,[17] Patient and Public Involvement and Engagement (PPIE), clinical experience and service realities. It proposes a timely referral system to charity-based services for bridging the gap between services the clinic and relevant charities can provide around the time of the diagnosis. However, this pathway is preliminary and needs further exploration to determine its utility and acceptability. Moreover, exploring the views of key stakeholders enables the production of experience-based co-developed interventions that empower and engage service users.[18,19]



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This study aimed to develop a patient care pathway to provide emotional support around the point of MS diagnosis (i.e., prediagnosis when investigations are underway when the diagnosis is given, and in the weeks postdiagnosis).

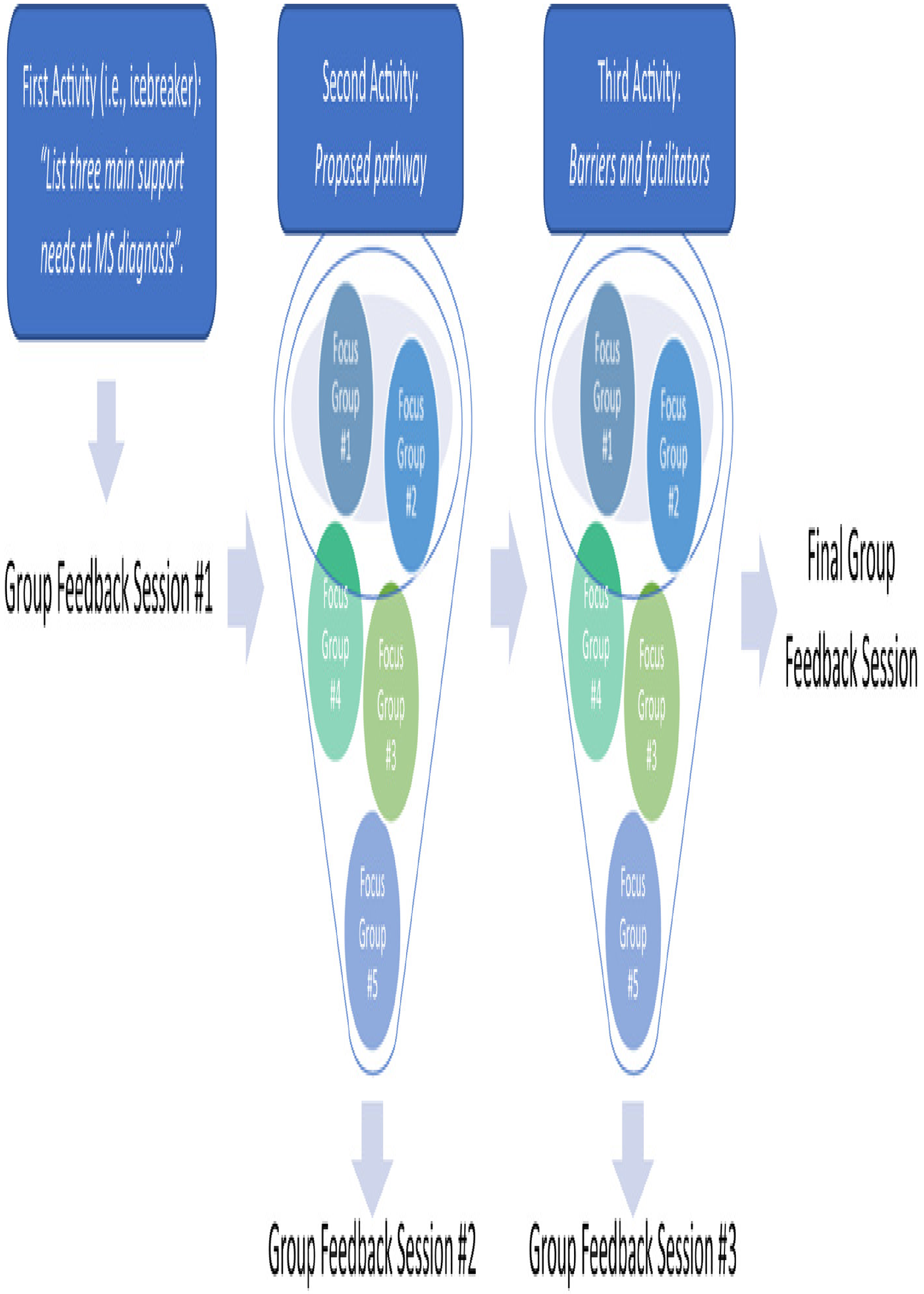
The secondary aim was to explore potential barriers and facilitators to the delivery and implementation of this pathway. We followed a person-centred,[19] experience-based co-design approach[18] allowing key stakeholders (i.e., service users and service providers) to inform pathway design *collaboratively* through group discussions, to identify sustainable changes that meet patients' needs. The present study is part of a wider research project aiming to develop and evaluate an intervention to support individuals around MS diagnosis (Providing Emotional Support around the Point of MS Diagnosis—PrEliMS Study[20]).

METHOD

To ensure the quality of reporting and transparency, we followed the consolidated criteria for reporting qualitative studies (COREQ) and the reporting recommendations for qualitative methods in communication and patient education research.[21,22]

Design

A qualitative, multistakeholder engagement study design was used to gain different perspectives from key stakeholders. The engagement involved consulting with stakeholders through different research activities. This consisted of one activity and two discussions, each addressing a different open question, across five individual focus groups and whole-group feedback sessions following each discussion (Figure 2). All stakeholders participated in the focus groups, and one representative from each group summarized their groups' reflections within whole-group feedback.



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Participants

We refer to stakeholders as individuals targeted by the proposed pathway or involved in its development and/or delivery. Inclusion criteria for each stakeholder category are provided in Table 1. All participants were required to be above 18 years, able to give informed consent and communicate in English. People with MS and carers/family members were recruited through MS charities, local MS support groups and from our local PPIE database. Health professionals and charity staff were recruited through charities and professional networks of the research team. Participants were a self-selected convenience sample, weighted towards people living with MS, in line with the experience-based co-design approach.[18]

Table 1 Inclusion criteria for each stakeholder group

Stakeholder group	Inclusion criteria
People with MS	People referred for possible MS diagnosis (i.e., the period just before receiving a formal diagnosis of MS by a neurologist up to 5 years), or newly diagnosed with MS (up to 5 years postdiagnosis) or have been diagnosed longer than 5 years ago who could comment on their past experiences. We used a 5-year period because, for some people with MS, the uncertainty around diagnosis lasted for this long, and our PPIE group felt that it was important to capture those who were within this period.
Carers/family members	Relative/carer of a person with MS. Carer is defined as a relative or friend providing informal care for someone with MS.
Health professionals	Clinicians (e.g., neurologists, MS nurses, psychologists, occupational therapists) working with people with MS.
MS charity staff/volunteers	People working or volunteering within MS charities.

Abbreviations: MS, multiple sclerosis; PPIE, Patient and Public Involvement and Engagement.

Procedure

Participants were invited to a local community centre to participate in a day-long event involving planning a patient care pathway for people with MS at the point of diagnosis. Before discussions, informed consent and relevant demographic information were obtained from all participants. The agenda for the day was co-created with our research team, which included a person with MS. Five semistructured focus groups of five to six participants (representing different stakeholder categories) sat at tables arranged Cabaret-style for ease of discussions, moderated by five research-active clinicians (a consultant neurologist, occupational therapist, clinical psychologist and two trainee psychologists; one moderator per focus group).

The event began with a presentation of what is already known about emotional support at MS diagnosis and the study aims. The first activity (‘What do you think are the three main things that need to be in place to support emotional wellbeing around the point of MS diagnosis?’) was used as an icebreaker. Participants wrote ideas on little sticky-notes individually, which guided reflections for the second discussion. Participants were provided with a diagram outlining a proposed care pathway designed based on literature, theory, clinical experience of research-active clinicians and a consultation meeting with nine PPIE members recruited from the Nottingham Multiple Sclerosis Patient and Public Involvement groups (Figure 1). Participants were invited to use the model to initiate the second discussion, exploring participants' suggestions for elements to add/remove from the pathway. Moderators used open-ended questions to probe for suggested changes, additions, the timing of various activities, who might deliver support and how/when it might be offered. The third discussion considered possible barriers and facilitators to delivery and implementation. After each round of discussion in focus groups, lasting approximately 45 min each, participants engaged in 30-minute feedback sessions with the whole group, led by a PPIE member. Finally, participants were invited to make additional comments towards the end of the discussions and were debriefed.

Data analysis

Discussions were audio-recorded and transcribed verbatim, omitting participant-identifiable information and analysed using framework analysis.[23,24] The analysis primarily applied a deductive-inductive approach to address our research aims. Thus, we anticipated that themes such as information, peer support and individualization would be relevant due to their prevalence in the extant literature (i.e., the deductive approach). The data were also examined inductively to allow unexpected responses or new insights.[23]

Transcripts were coded on NVivo (Version 12) by the first author, to classify the data and allow for systematic comparisons across discussions. A working analytical framework was developed after coding one focus group transcript for both discussions and whole group feedback sessions, reviewed by two other authors. Codes were grouped into categories, with new codes added as subsequent transcripts were indexed by the first author. Discussions with the rest of the research team were held to sense-check and modify the coding scheme. Any disagreements were resolved by discussion until consensus was reached, or arbitrated if necessary, by the wider research team. Yardley's[25] evaluative characteristics for good qualitative research were followed to ensure credibility and rigour.

RESULTS

The stakeholder sample consisted of 16 people with MS (68.75% relapsing-remitting, 18.75% secondary progressive, 12.5% primary progressive) with an average illness duration of 8.6 years (SD = 6.6); 5 carers/family members of a person with MS (4 partner/spouse; 1 parent) and 5 healthcare professionals and charity staff (3 MS nurses, 1 psychiatrist and 1 MS Society staff member) with a range of 10–33 years experience working with MS (*M* = 17.8; SD = 10.3). Table 2 describes further demographics.

Table 2 Participant demographics per stakeholder group

Demographics (*n*, %)	People with MS (*n* = 16; 62%)	Carers/family (*n* = 5; 19%)	Healthcare professionals and charity staff* (*n* = 5; 19%)	Overall** (*n* = 26)
Age				
21–30	1 (6.25%)	0 (0.0%)	0 (0.0%)	1 (4.0%)
31–40	4 (25.0%)	0 (0.0%)	1 (20.0%)	5 (20.0%)
41–50	4 (25.0%)	0 (0.0%)	0 (0.0%)	4 (16.0%)
51–60	3 (18.75%)	1 (20.0%)	2 (40.0%)	6 (24.0%)
60+	4 (25.0%)	4 (80.0%)	1 (20.0%)	9 (36.0%)
Gender				
Man	5 (31.25%)	4 (80.0%)	2 (40.0%)	11 (42.3%)
Woman	11 (68.75%)	1 (20.0%)	3 (60.0%)	15 (57.7%)
Education level				

GCSE	6 (37.5%)	0 (0.0%)		6 (28.6%)
A Level	3 (18.75%)	0 (0.0%)		3 (14.3%)
Degree	4 (25.0%)	4 (80.0%)		8 (38.1%)
Higher degree	3 (18.75%)	1 (20.0%)		4 (19.0%)
Employment status				
Not employed	2 (12.5%)	0 (0.0%)		2 (9.5%)
Employed full-time	1 (6.25%)	1 (20.0%)		2 (9.5%)
Employed part-time	4 (25.0%)	1 (20.0%)		5 (23.8%)
Retired	8 (50.0%)	3 (60.0%)		11 (52.4%)
Voluntary part-time	1 (6.25%)	0 (0.0%)		1 (4.8%)

*One professional did not disclose age. **Education level and employment status information was not collected from healthcare professionals and charity staff.

Following framework analysis, 31 subthemes were interpreted (presented in italics) relating to the development of an emotional support pathway, organized into 5 superordinate categories (Table 3).

Table 3 Categories and subthemes identified

Category	Subthemes
Information	Condition management
	Services
	Accuracy and reliability
	Social network and employers
	Practical information
	Overload
	Format

	Cooling off period
	Framing
Individualization	Person-centred approach
	Toolbox of options
	Patient experience
	Family involvement
	Timing
Standardization	Accessibility
	Postcode lottery
	Technology integrating services
	Evaluation
	Hub
Professional role	Personal attributes
	Duties
	Training
	Value of human connection
	Governance
	Limitations
Voluntary peer support role	Shared experiences
	Benefits to peer supporter
	Savings
	Sustainability
	Befriender

Information

Participants across individual focus groups felt that a package of adequate, relevant and appropriate information should be provided at the point of diagnosis, for example for *condition management* and signposting to *services* and MS charities. Participants agreed that *practical information* regarding responsibilities such as contacting the UK Driver and Vehicle Licensing Agency (which issues drivers' licences) and general advice for daily life should be given to people with MS, and their *social network and employers*. Participants suggested that verbal information should be accompanied by different *formats*, such as hard-copy booklets to keep and refer to later. Most participants discussed that information should come from a reliable source to avoid receiving inaccurate information (*accuracy and reliability*), as participants in two focus groups opined ‘don't google’ (Groups 1 and 2). Overall, participants felt that information and diagnostic news should be positively framed with hope (*framing*):

I think the biggest thing I could have found out at the start was someone saying, ‘you're not alone, there is support there’ (Woman with MS, Group 2)

However, participants recommended that clinicians should strike a balance between giving sufficient information—which they felt was an important element of emotional support facilitating ongoing psychological adjustment—and avoiding *overload*. One participant stated that they felt bombarded with medical information at diagnosis: ‘You just can't assimilate it’ (Woman with MS, Group 1). To overcome this issue, four focus groups discussed a *cooling off period*, whereby patients should be given time to process their MS diagnosis, for example allowing a 2-week interval between diagnosis and a follow-up session with a healthcare professional, such as an MS Nurse.

Individualization

Focus groups elaborated on the *timing* of the pathway's elements, which was also repeated during whole-group feedback. They felt it should depend on the patient's unique needs and preferences. Furthermore, participants felt that it was essential that a patient care pathway provides an individualized, *person-centred approach* to account for their personal circumstances, such as *family involvement*, ‘because what suits one person might not [suit] another’ (Carer, Male, Group 3). Participants reflected on their varied *patient experience* and suggested that adjustment is a different process for every person, thus the diagnosis procedure ‘must be fluid’ (Carer, Male, Group 5). Therefore, participants felt that individuals should have access to a *toolbox of options* via professional contact, with alternatives based on need and personal choices, such as topic-based information or referrals:

keep it as simple as you can, but say ‘here are some things that may be available if you need it’ (Man with MS, Group 2)

Standardization

While participants emphasized the importance of individualization, they agreed that some standardization is required to ensure *accessibility. Postcode lottery* should be avoided, whereby a person's geographical location in the country can determine the service they receive. Therefore, participants suggested using *technology* (e.g., the Internet, videoconferencing) to standardize service delivery throughout the pathway to be accessible for everyone. However, some participants felt that this would not be appropriate for all, arguing that some people avoid the Internet or would benefit more from in-person sessions rather than telephone calls. Again, this is linked with individualization and applying a person-centred approach throughout.

To avoid geographical location-based limitations, participants felt that *integrating services* through a ‘holistic approach’ (Woman with MS, Group 4) would facilitate collaboration between the NHS and other organizations, such as charities, to reach out to people in rural areas with fewer services. For example, participants suggested creating a *hub* to direct patients to different services or professionals. Finally, participants discussed the importance of *evaluation* of the pathway, because ‘when the NHS has got evidence, they're compelled to act’ (MS Nurse, Female, Group 5), to ensure its sustainability from a resource perspective.

Professional role

All focus groups felt strongly that a paid healthcare professional should deliver part of the intervention. Most agreed that this should be an MS Nurse, while a smaller selection of participants wondered whether it would save costs to develop this role for less qualified staff. *Personal attributes* of the professional were discussed, whereby participants described essential qualities such as expertise, experience and understanding of MS: ‘somebody who's got that appropriate knowledge—whoever that is’ (Man with MS, Group 1).

Duties of the role included referrals and triaging for emotional support according to clinical need, including an ‘individual assessment of their needs in the first instance’ (MS Nurse, Female, Group 5). Participants agreed that professionals should give patients time to ask questions and listen to concerns at diagnosis, initiating the provision of emotional support. Many felt that this professional should be present at diagnosis, alongside the neurologist delivering the news, as an MS champion and advocate. Groups agreed that the professional should be introduced in person to build a relationship for personalized emotional support, emphasizing the *value of human connection*:

I would have liked there to have been an MS nurse with us, but then took us aside and said like, ‘you've been told [about your MS diagnosis]—what are your questions?’ (Woman with MS, Group 2)

However, *limitations* of the role included the barrier of recruiting and retaining enough staff. Two focus groups discussed the demands of nursing roles, with long hours and large workloads, which would likely increase with extra responsibilities accompanying delivering a new intervention within the pathway. To overcome these barriers, participants felt the role would need appropriate *governance* to set parameters for the role, with *training* and supervision to quality-check support, hence its requirement for remuneration and professional accountability:

if you're properly trained by a body, and are affiliated to do it, then you're alright. It is protection. (Man with MS, Group 3)

Furthermore, participants proposed outsourcing and delegating duties to a voluntary support worker role, to deliver further aspects of emotional support via a ‘parallel service’ (Woman with MS, Group 5), offsetting the burden of nurses' long hours and increasing workload.

Voluntary peer support role

Some participants across groups felt that peer supporters could be paid with ‘savings that these programmes will achieve’ (Woman with MS, Group 4), facilitating their remuneration, yet most referred to this role as voluntary to save costs for the NHS. In addition, groups suggested that *savings* could be made across wider NHS capacity. Developing a role for peers also living with MS could mitigate the barrier of not having enough time or staff, as another participant noted, ‘time is precious for medical professionals in the NHS’ (MS Nurse, Female, Group 2), which may not be an issue for volunteers by virtue of them volunteering time. Contrastingly, the *sustainability* of voluntary roles was questioned:

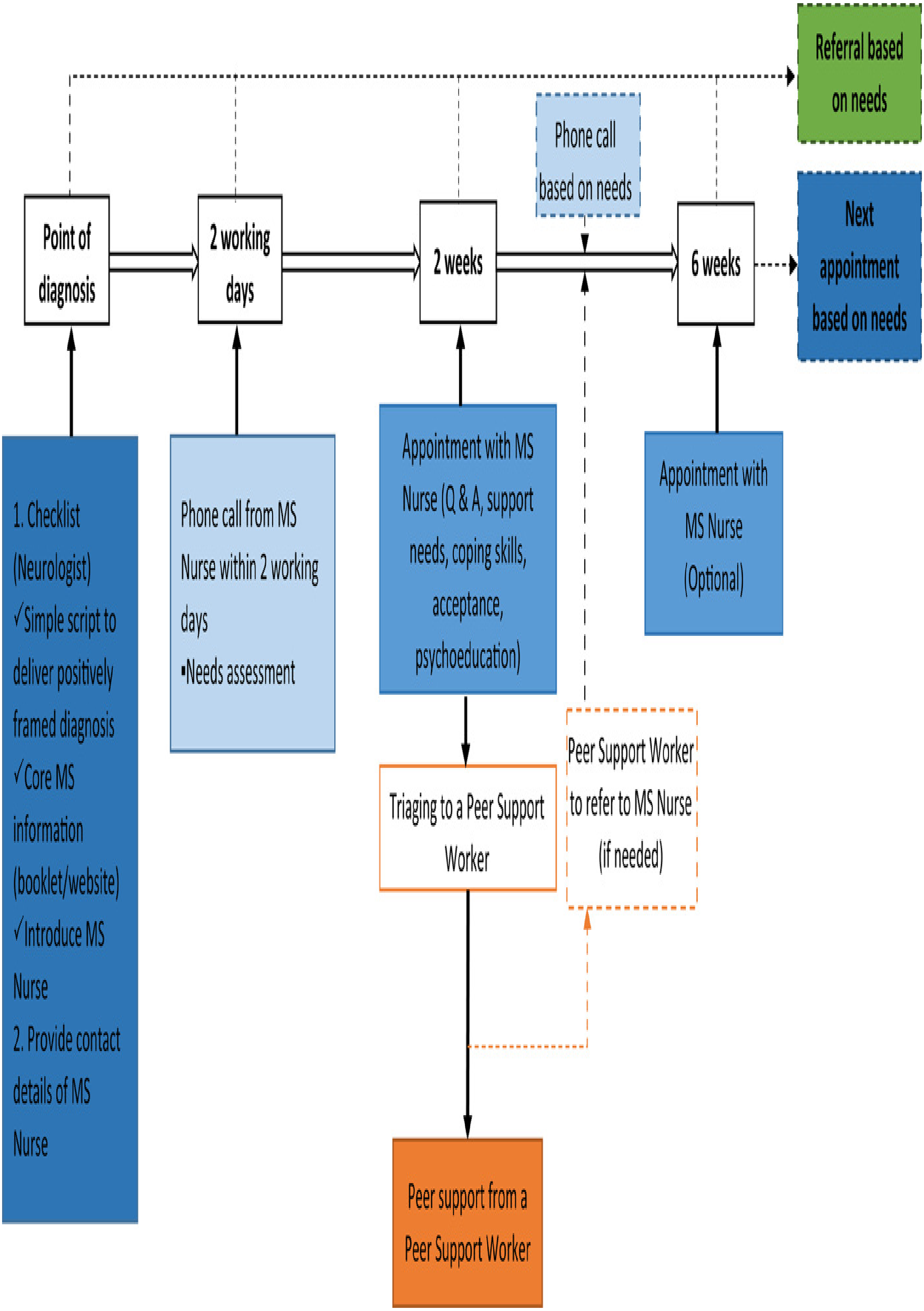
When you volunteer, sometimes you can get dragged into doing so much that actually it's a detriment to yourselves. (Charity staff, Female, Group 3)

Therefore, finding appropriate volunteers who are committed to the intervention's duration with each patient was presented as another barrier, which participants suggested could be mitigated by training and ongoing support. A further facilitating factor was discussed: *benefits to peer supporter*. Participants felt that people with MS might find this role fulfilling and thus remain enthusiastic and committed to ‘giving something back’ (Man with MS, Group 4), obviating the need for payment, particularly for people who might be transitioning from employment to retirement:

if they had to give up work because of their condition, this might be a way that they can still feel valuable. (Man with MS, Group 3)

Groups unanimously agreed that the crucial facilitator of peer support within a pathway was the contribution of *shared experiences*. Many participants felt that individuals living with MS would be more relatable and credible, as most clinicians cannot understand the subjective experience of MS. Therefore, sharing relevant experiences may help patients to problem-solve, which many felt was vital for emotional support. Moreover, participants suggested that the peer supporter could act as a *befriender* to listen and empathize because they felt that sometimes patients need more emotional support than a healthcare professional can provide. Therefore, peers ‘on the same wavelength’ (Woman with MS, Group 5) could be instrumental during the process of adjustment around the point of diagnosis.

A patient care pathway was subsequently created by the research-active clinicians present during the stakeholder engagement discussions, in light of participants' reflections. This was then presented to a group of seven people with MS and four carers, who helped us to refine the pathway. Figure 3 depicts the co-constructed pathway.



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DISCUSSION

Stakeholder focus groups and whole-group discussions informed the development of a blueprint for a co-constructed ‘point of MS diagnosis’ patient care pathway to provide emotional support around the point of diagnosing MS (Figure 3). Many participants agreed that the proposed pathway should involve MS Nurses at the forefront, who should be introduced to the patient early during discussions about the diagnosis. Groups also agreed that adequate, appropriately delivered information is a key element of emotional support, equipping patients with reliable advice for dealing with practical issues, which should be positively framed to instil hope. The addition of peer support volunteers to assist during the pathway was suggested and accepted by many participants. This may also improve the health and well-being of the volunteers themselves.[26] However, some participants worried about the role's sustainability, whereby volunteers may not remain committed without payment. Furthermore, there are valid debates about volunteer remuneration[27] that are beyond the scope of this paper, but nonetheless need to be considered.

In the United Kingdom, our MS services are embedded within a nationalized healthcare system (NHS) funded by the state.[28] While this affords equality of care, it poses certain funding and service constraints (with overstretched and understaffed services). This is one of the reasons why we explored who would be best placed to offer emotional support services and considered the potential involvement of a volunteer workforce. Thus, issues of sustainability could be mitigated by offering patients combined input from MS Nurses and peer supporters. As such, this could offset barriers regarding limited resources, such as time and funding, while containing the voluntary role within certain boundaries to ensure that the volunteers are not overburdened or work outside of their expertise. Indeed, while participants suggested that retirees with MS may be well-suited, recruitment and retention may be difficult, especially if volunteers are limited by the progression of MS symptoms. Alternatively, there have been several models of such peer-support work in the mental health arena, which have shown great value,[29–31] and the NHS has created job descriptions that have been mapped onto NHS pay scales. Therefore, such models could be adopted in MS services.

Our findings support the existing research literature, particularly around the timing of commencing the pathway and providing the embedded support intervention. Participants felt that after delivering news of the diagnosis, further support should be delayed to allow the individual time to process the diagnosis. Moreover, participants felt they needed timely information regarding reliable sources of information. This echoes the line of argument resulting from a recent meta-synthesis.[10] Others also recommend that professionals delivering diagnoses of neurological conditions (including MS) should assess and respond to patients' information preferences with empathy and provide more time for questions,[32] which our findings support. Indeed, while individuals may have different information needs due to personal circumstances, several studies have highlighted inadequate information provided.[6,14,33–35] Reflecting Miller's[36] theory of coping with stress-provoking situations, a study proposed that people with MS could be categorized into ‘monitors’, who seek information to manage their MS, or ‘blunters’, who avoid information as they believe it increases anxiety.[33] Another study found that a significant proportion of respondents declined further advice (‘blunters’), suggesting that the impact and severity of one's MS affects their need for information.[14] Our results also support the provision of comprehensive, individualized information which may benefit longer-term emotional adjustment. Also, our findings urge that information provided must be given to individuals at the time they need it, ensuring they have time to digest new information to improve experiences around diagnosis. Nevertheless, an examination regarding the type of information people with MS find helpful/unhelpful and how it might be incorporated as part of emotional support would improve the usefulness of the findings. Additionally, further qualitative research could explore how to support those who specifically undergo a prolonged period of diagnostic investigations during prediagnosis, as this may present unique challenges. The present study focused on the development of the patient care pathway around MS diagnosis *in general*, including prediagnosis, diagnosis and the immediate postdiagnosis period.

Strengthening the study, we followed Yardley's[25] guiding principles for good quality qualitative research: context sensitivity, commitment and rigour, transparency and coherence and impact and importance. Commitment considers topic engagement and data immersion which is supported by our evidence-based approach, strengthening context sensitivity. To ensure the rigour of the findings, different stakeholder groups collaborated to achieve multilayered insights and whole-group feedback triangulated responses from focus group discussions.[25] While at least two researchers analysing data separately before reaching a consensus has been recommended,[37] the concept of interrater reliability in qualitative research has been regarded as meaningless and cannot exclude subjective interpretations.[25] However, two coders may have further improved rigour and transparency to reach a consensus without the need to calculate interrater reliability. In the interest of transparency, the researcher responsible for the initial coding of transcripts was absent from the event, thus missing an opportunity to become immersed in the process and begin familiarization during data collection.[23] To mitigate this limitation, they listened to recordings multiple times to aid familiarization and received input from team members present at the event when developing the coding scheme to enhance collaboration during analysis. Considering coherence, adopting framework analysis based on a person-based co-design was appropriate for our aims, as framework analysis is particularly suitable when study aims are clearly defined at the onset.[23]

Furthermore, the present study aimed to maximize impact and utility[25] by adhering to the Medical Research Council's guidance for complex intervention implementation, and by developing relevant theory from literature and stakeholder engagement before commencing the lengthy evaluation process.[16] Thus, by identifying important active ingredients based on the person- and experience-based co-design approaches, the resulting pathway and embedded support intervention are more likely to be sustainable whilst meeting patients' needs, maximizing the likelihood of acceptability during subsequent evaluation within a full feasibility trial.[19] Moreover, context-sensitivity is increased by eliciting perspectives of different stakeholder groups, illuminating potential barriers and facilitators to inform implementation within healthcare settings.[25] While research-active clinicians acting as moderators in the focus groups may have biased discussions (e.g., reflecting professional disciplines), all have varied clinical backgrounds offering diverse perspectives.

Recognizing the need for further emotional support around MS diagnosis, we have developed a co-constructed patient care pathway for emotional support provision around MS diagnosis via multistakeholder engagement. Our stakeholders felt that information provision coupled with emotional and social support (individualized MS Nurse support, peer support) would improve adjustment to diagnosis, reflecting prior research.[5–7,10–13,15] Participants also suggested that emotional support should be delivered within a standardized system, including those who may miss out due to sparse services in their location, and/or have difficulties accessing technology and the Internet. However, barriers were noted, including financial costs to the NHS and demand on nurses' limited time, which could be mitigated by enlisting peer support workers to facilitate ongoing emotional support.

As part of the PrEliMS programme, a point of diagnosis intervention was developed using this pathway and is currently being tested in a feasibility trial, and a definitive trial evaluating the clinical and cost-effectiveness of this pathway is planned. Future research could consider whether matching patient participants to peer supporters (e.g., across specific demographic variables) would be beneficial, as patient participants may have different priorities across life stages (such as work, raising children and retirement), and research should explore how the peer support worker role in mental health services can be adapted for MS services.

AUTHOR CONTRIBUTIONS

Tierney Tindall: Data curation; formal analysis; methodology; writing –original draft; writing –review and editing. **Gogem Topcu**: Conceptualization; methodology; investigation; formal analysis; writing –original draft; writing –review and editing; supervision; project administration; funding acquisition. **Shirley Thomas**: Methodology; formal analysis; writing –review and editing; supervision. **Clare Bale**: Conceptualization; investigation; writing –review and editing. **Nikos Evangelou**: Conceptualization; investigation; writing –review and editing; funding acquisition. **Avril Drummond**: Conceptualization; investigation; writing –review and editing; funding acquisition. **Roshan das Nair**: Conceptualization; investigation; writing –review and editing; funding acquisition.

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CONFLICTS OF INTEREST

R. d. N. is the Chair of the NIHR Research for Patient Benefit East Midlands Research Advisory Committee; he has received funding to prepare and deliver lectures on cognitive rehabilitation in multiple sclerosis from Novartis and Biogen. N. E. is a member of the advisory board for Biogen, Merck, Novartis and Roche; he has received grant income from the MS Society, MRC, PCORI and NIHR. The remaining authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available on request from the corresponding author. The data are not publicly available due to privacy or ethical restrictions.

ETHICS STATEMENT

Ethical approval was obtained from the University of Nottingham Faculty of Medicine and Health Sciences Ethics Committee (193-801). Participants gave informed consent to participate and for anonymized results to be disseminated including through publication. We confirm all personal identifiers have been removed or disguised so the patient/person(s) described are not identifiable and cannot be identified through the details of the story.

DETAILS

Subject:	Intervention; Working conditions; Medical diagnosis; Patients; Workloads; Relatives; Facilitators; Charity; Psychological factors; Health status; Diagnosis; Nurses; Caregivers; Peer tutoring; Support workers; Charities; Health services; Information sharing; Multiple sclerosis; Access; Emotions; Citizen participation; Support groups; Public involvement; Stakeholders; Co-design; Clinical outcomes; Medical personnel; Social support; Clinics; Focus groups; Adjustment; Data collection; Health professional-Patient communication; Emotional support; Research design; Psychological well being; Qualitative research
Business indexing term:	Subject: Stakeholders
Location:	United Kingdom--UK
Identifier / keyword:	diagnosis; emotional support; multiple sclerosis; peer support; qualitative; stakeholder engagement
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Access to novel drugs and therapeutics for children and youth: Eliciting citizens' values to inform public

funding decisions

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ABSTRACT (ENGLISH)

Introduction

The unique evidentiary, economic and ethical challenges associated with health technology assessment (HTA) of precision therapies limit access to novel drugs and therapeutics for children and youth, for whom such challenges are amplified. We elicited citizens' perspectives about values-based criteria relevant to the assessment of paediatric precision therapies to inform the development of a child-tailored HTA framework.

Methods

We held four citizen panels virtually in May–June 2021, informed by a plain-language citizen brief summarizing global and local evidence about the challenges, policy and programmatic options and implementation strategies related to enhancing access to precision therapies for Canadian children and youth. Panellists were recruited through a nationally representative database, medical/patient networks and social media. We inductively coded and thematically analysed panel transcripts to generate themes and identify priority values.

Results

The perspectives of panellists (*n*=45) coalesced into four overlapping themes, with attendant subthemes, relevant to a child-tailored HTA framework: (1) Childhood Distinctions: vulnerability, 'fair innings', future potential, family impacts; (2) Voice: agency of children and youth; lived versus no lived experience; (3) One versus Many: disease severity, rarity, equity, unmet need and (4) Health System Governance: funding, implementation inequities, effectiveness and safety. Participants broadly agreed that childhood distinctions, particularly family impacts, justify child-tailored HTA. Dissent arose over whose voice should inform HTA and how such perspectives are best incorporated.

Conclusions

Citizens can offer unique insights into criteria relevant to the development or revision of HTA frameworks to capture holistic, societally responsive dimensions of value attached to unique contexts or populations, including children. Balancing the hopes and expectations of patients and caregivers for access to expensive but potential life-altering therapies against the opportunity costs borne by encompassing health systems is a fundamental challenge that will require rigorous methods to elicit, weigh and reconcile varied views.

Patient or Public Contribution

A patient advocate served on the steering committee of this study and co-authored this article. Key informants for the Citizen Brief included patient advocates and caregivers; a separate patient advocate reviewed the Brief before dissemination. Qualitative and quantitative data were collected from the general public and caregivers of children, with written consent.

FULL TEXT

INTRODUCTION

The genomic revolution poses challenges to the responsible stewardship of limited healthcare resources and the equitable distribution of benefits. Precision therapies, designed to target patient-level genetic differences that drive disease development and behaviour, hold promise for patients suffering from rare or hard-to-treat diseases, which may lack effective alternative treatment options.[1,2] However, access to and uptake of these therapies is restricted by limited data on clinical effectiveness and by their typically large price tags, posing challenges for traditional health technology assessment (HTA).[3–5] Health systems have yet to develop assessment standards and policies to address the unique evidentiary, economic and ethical challenges presented by precision therapies, constraining the incorporation of best evidence and patient and public values into HTA processes and, by extension, public funding decisions.[4,6–9]

Children who suffer from rare and complex diseases may have the most to gain from precision health technologies, yet they are being left behind at multiple levels of therapy development and access. First, clinical trials in children are limited by small population sizes, complexities of trial enrolment and weak industry interest.[10] Second, submissions to HTA bodies seldom include evidence for paediatric indications, meaning funding recommendations are routinely based on and intended for adult indications. Third, current funding assessments rarely incorporate the unique sociobiology of child health and illness.[11–14] Governments and researchers are responding to the first two constraints by developing unique trial designs, creating disease networks, enacting legislation to compel and incentivize pharmaceutical companies to generate paediatric evidence and formulating policy targeting funding for orphan and rare diseases.[15–22]

The third constraint remains largely unaddressed and stems from a more fundamental problem: the lack of HTA approaches that attend to the unique dimensions of child health and illness.[12,23–26] HTA frameworks set out the principles and methods for systematic weighing of clinical, economic, organizational and patient-related impacts of novel interventions to ascertain value increments compared to existing alternatives, with the extra charge of assessing contextual equity, sustainability and health system implementation impacts. In the context of precision health interventions, evaluations must weigh uncertainties related to limited data on safety and efficacy. When applied to child health technologies, these frameworks face the additional challenge of incorporating the unique evidentiary, physiological, health-system and societal considerations attached to children.[27–31] The absence of a standardized framework for the evaluation and reimbursement of precision health technologies for children thus has significant consequences for children and youth.

Effective citizen engagement and public deliberation have been shown to provide rich and nuanced insights on issues of societal importance.[32–36] Structured and meaningful incorporation of these insights into health policy-making processes helps to legitimize decisions that impact the populace. In many jurisdictions, including Canada, public engagement has been embraced as a component of HTA deliberations, but implementation is variable and best practices for evidence-informed deliberation are still evolving.[37–39] Furthermore, input from *non-patient publics*, particularly youth, is often neglected or underrepresented in HTA processes, which might erode the societal legitimacy of recommendations provided or decisions made. We conducted a series of citizen panels with members of the Canadian public to elicit and prioritize values relevant to HTA for precision child health technologies. This research constituted the initial phase in the development of a value assessment framework for precision therapies for children and youth explicitly informed by societal values.

METHODSStudy design

We convened four citizen panels, based on an established approach developed by the McMaster Health Forum,[40] to identify public values related to HTA for precision child health technologies. The panels were informed by an evidence package and were convened virtually using Microsoft Teams™ between April and June 2021, each lasting 3 h.

An interdisciplinary steering committee provided expert guidance and oversight for the project. The committee consisted of academic paediatricians, a bioethicist, representatives from Health Canada and the Canadian Agency for Drugs and Technology in Health (CADTH), a patient advocate and health services researchers with expertise in genomics, HTA, health systems and policy analysis, deliberative processes and qualitative research methods. The steering committee met six times between November 2020 and September 2021, and was given opportunities to review all materials.

Evidence package

To support informed deliberations, we provided panellists at least 1 week before the panels an evidence package in the form of a citizen brief and complementary videos. We compiled evidence from systematic searches of online evidence synthesis repositories (e.g., Health Systems Evidence) and medical literature databases, and supplemented it with grey literature identified through targeted searches of relevant regulatory and HTA institutional websites. The citizen brief summarized relevant research on the following: (1) context about why precision therapies for children and youth in Canada requires public input; (2) key challenges that exist for accessing precision therapies for children and youth; (3) three elements of a potential comprehensive approach for addressing these challenges and (4) implementation considerations.[41] We presented the evidence in plain language and also conveyed it using a mix of figures, infographics and case studies. We also posed questions for panellists to consider, including those focussed on a child-tailored HTA, that would be used to guide deliberations.

Input from key informants (*n* = 17) helped shape the focus and content of the brief. The key informants included paediatric specialists in rare and chronic conditions, policymakers, leaders of patient advocacy groups, health researchers, industry professionals, representatives from CADTH and Quebec's Institut national d'excellence en santé et services sociaux (INESSS), and regulatory leaders from Health Canada, the FDA and the European Medicines Agency. Four merit reviewers, comprising a health economist, two policymakers and a patient/citizen representative, critically assessed the accuracy, relevance and readability of the brief.

The complementary videos provided general lay information on precision medicine and HTA processes and were publicly available through YouTube™. (Links are found in Supporting Information: Appendix 1, ‘Complementary Videos’.)

Recruitment and panel composition

As a standard for deliberative public engagements (DPEs), we aimed to convene for each panel 14–16 members of the general public who were diverse in terms of gender, age, socioeconomic status, ethnocultural background and geographic residence for two of the four panels. We recruited panellists in collaboration with AskingCanadians™, which has a panel of more than 600,000 Canadians who are affiliated with loyalty programmes in Canada and are representative of all Statistics Canada demographic categories.[42] Through a combination of recruitment through AskingCanadians™ and invitation through clinical and patient networks and social-media advertisements, we aimed to form two population-focussed panels, another composed of caregivers of children with chronic and complex diseases and a last with youth participants aged 16–21.

Data collection and analysis

Two expert facilitators (A. D. and M. G. W.) provided panellists with opportunities to learn from the evidence and others' experiences, share existing and newly informed views and make conclusions based on their values and preferences. The discussions in the two larger groups (one general public and the youth group) took place in an initial plenary, followed by a break-out into two groups to allow freer discussion and concluded with a consolidating plenary where individual groups reported on respective key discussion points. In the two smaller groups, panellists remained together in plenary. Panellists were also encouraged to use the ‘chat’ function. The discussion was guided by questions from the citizen brief, focussing on HTA-related challenges and pertinent approaches to overcome them. We did not aim for consensus among the panellists but rather to identify common ground, differences of opinion and the values underlying different positions. Audio and ‘chat’ exchanges were recorded on Microsoft Teams™, and research team members also took notes and memos throughout the deliberations.

Additionally, online pre- and postdeliberation questionnaires were sent to panellists during each panel asking them to rank 12 HTA-relevant values, which had been identified based on a literature review and prior research.[12,25,43,44] These were as follows: effectiveness; disease severity; safety; unmet need; future potential; costs; impacts on families; rarity; vulnerability; citizen values and preferences and child and youth views.

Each citizen panel recording was transcribed verbatim, cleaned and then verified against the audio-recording. Messages in the ‘chat’ function were included in the data set. Data were anonymized and de-identified to protect panellist confidentiality and were stored in a secure electronic database accessible only by the research team. Following Braun and Clarke's[45] thematic analysis method, transcripts were inductively coded using NVivo12. Three team members (C. L. G., L. W. and M. S.) iteratively created a codebook and independently coded transcripts, employing constant comparative methods to further refine codes, identify patterns and generate themes (See Supporting Information: Appendix 2, ‘Codebook’). Independent coding results were compared in two rounds of examination by the team to resolve discordance and ensure fidelity to the codebook and theme generation. Analysis of the virtual ‘chat’ messages, researcher notes and coding memos all contributed to theoretical interpretations and enabled reflexivity.[46]

RESULTSPanel and panellist characteristics

Table 1 outlines the sociodemographic characteristics of the panellists (*n* = 45). The two general public panels numbered 7 and 14, respectively, the youths numbered 14 and the caregivers 10. While three of the panels were balanced in terms of the number of female and male participants, the caregiver panel was exclusively female. Panellists were from nine of Canada's 10 provinces, with predominant representation from Ontario and Québec, reflecting their relative population sizes within the country. All those with lived experience of accessing paediatric precision therapies were caregivers.

Table 1 Sociodemographic characteristics of panellists, *n* = 45

Characteristics	*n*	%
Sex		
Male	13	29
Female	31	69
Prefer not to disclose	1	2
Age (years)		
16–21	14	31
22–34	7	16
35–49	14	31

50–64	9	20
≥65	1	2
Province of residence		
British Columbia	1	2
Alberta	7	16
Saskatchewan	1	2
Manitoba	1	2
Ontario	18	40
Quebec	10	22
New Brunswick	3	7
Nova Scotia	2	4
Prince Edward Island	2	4
Lived experience		
Yes	12	27
No	33	73
Highest educational level attained		
Elementary school	1	2
High school	9	20
Community college	8	18
Technical school	5	11
Bachelor/postgraduate training/professional degree	22	49
Employment status		
Self-employed	3	7

Full-time employed	17	38
Part-time employed	3	7
Unemployed	4	9
Retired	3	7
Student	12	27
Homemaker	3	7
Income level (Canadian dollars)		
<$20,000	6	13
$20,000–$40,000	8	18
$40,000–$60,000	5	11
$60,000–$80,000	8	18
>$80,000	10	22
Prefer not to disclose	8	18

Perspectives on current approaches to HTA processes for child health technologies

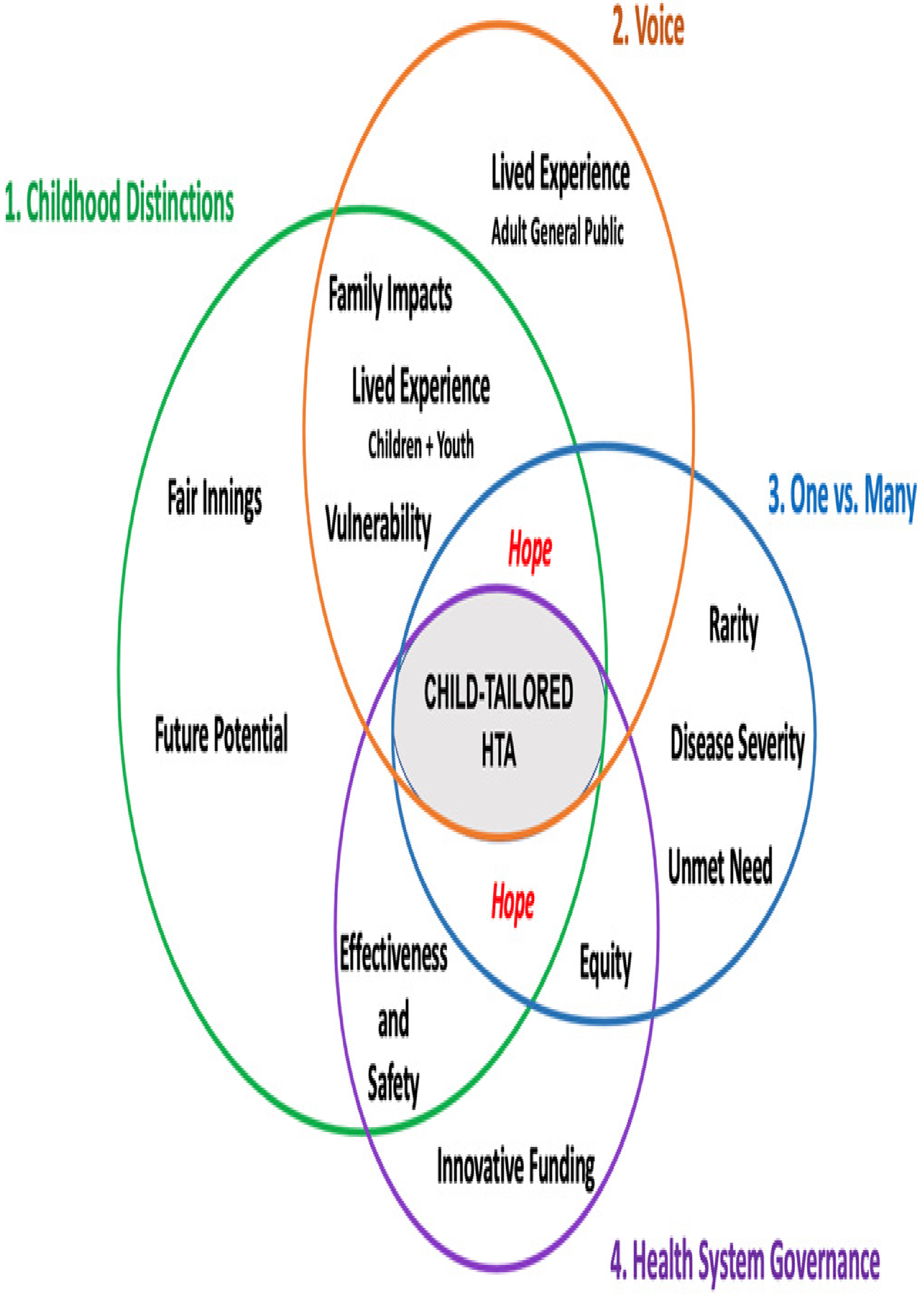
Reflecting on existing core criteria for HTA assessments, panellists broadly agreed that current frameworks do not specifically account for the unique dimensions of child health and illness, nor do they capture family perspectives, generally and specifically for paediatric precision therapies. A range of panellists emphasized that current approaches to measuring effectiveness and adjudicating therapeutic benefit may be inadequate or poorly calibrated to the evolving disease dynamics and treatment paradigms in precision medicine as well as to alternative outcomes that patients and caregivers value. One caregiver of a child with cystic fibrosis asserted that HTA assessment 'gave [a therapy] a negative recommendation because it only improved lung function by 3 to 5%. So that's what it said on paper, but I've been trying to explain to them that maybe 3 to 5% on paper doesn't look like much, but to me that's my child going to a full year of school …not having to miss, you know, 50-something days of school that he had missed previously' (Panellist P4-9). Similarly, some panellists noted a greater need to situate safety considerations within the context of other values—such as disease severity, hope and risk tolerance. As one noted, 'I imagine often the families will have a very large say into what kind of risk they're willing to take' (Panellist P1-2).

Panellists deliberated about costs from both health system and family perspectives, particularly opportunity costs. Several of them highlighted that the full costs associated with a complex or chronic illness, including those related to immediate and long-term disease impacts, are not wholly considered in HTA, with outsized implications for child health due to their care needs. One caregiver shared, 'I have two degrees and I haven't worked since [the two children's] diagnosis because it's a lot of work. So that's lost income for our family and it's also lost tax dollars…' (Panellist P4-3). Some panellists also felt that the nonmonetary costs of illness, including lost schooling or decrements in well-being, need to be included in assessments. Although cost-effectiveness was only implicitly discussed, a number of panellists grappled with the legitimacy of explicit cost-effectiveness thresholds, such as the commonly referred to $100,000 per QALY, in the face of lived realities of disease for children and families. One panellist stated, '…this idea that you can put a price on my child's life, like my daughter is only worth …$100,000 a year …that, you know, just hurts you inside' (Panellist P4-2).

Perspectives on priority values and principles for a child-tailored HTA framework

In thinking about how to create and operationalize a child-tailored HTA framework, panellists queried, weighed and debated the individual values important to such a framework, including those for precision therapies for children and youth. Their perspectives coalesced into four overarching and overlapping themes: (1) Childhood Distinctions—characterized by consensus that children have unique attributes justifying child-tailored HTA; (2) Voice—characterized by disagreement about whose perspectives ought to inform HTA; (3) One versus Many—characterized by the discordance between humanitarian and utilitarian perspectives and (4) Health System Governance—characterized by reflection on structural challenges in equitable access to precision therapies determined by the overarching Canadian health system context.

Figure 1 depicts key interconnections between the overarching themes and subthemes, including 'Hope', which was threaded throughout the discussions.



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Theme 1: Childhood Distinctions

Childhood Distinctions, a cluster of interrelated phenomena which distinguish children and childhood traits, were paramount in both justifying and framing the development of a child-tailored HTA framework for most panellists. Discussion focussed on four closely interlinked subthemes that were seen to set children and youth apart from adults: (a) vulnerability; (b) 'fair innings'; (c) future potential and (d) family impacts.

Vulnerability

A multifaceted concept of childhood vulnerability emerged in panellists' reflections, particularly around the dimensions of physical fragility, cognitive maturity, parental dependence and autonomy within the family. The complex interactions among these dimensions, shaped continuously by age throughout childhood, were reflected in panellists' arguments over how to define, assess and measure vulnerability. The need for proxy voices in medical and policy decision-making contributed to panellists' conception of childhood vulnerability, and disabilities arising from chronic illnesses were seen to complicate these issues. Panellists agreed that society had a collective responsibility to take protective actions for children, as one succinctly explained: 'I think society …seems to view children as being more fragile …with how like children [are] first in a lot of rescue stuff …in general with society we kind of try to prioritize children' (Panellist P3-2). Nevertheless, panellists sometimes grappled with how to define vulnerability given evolving capabilities and appropriate agency for children. One panellist expressed this ambivalence thus: '…making the decisions, it has to be the children when they do have the ability to do so…. I don't know how you would detect the age for that because I think age is really again another debate' (Panellist P3-8).

'Fair innings' and future potential

Although these two subthemes were explored discretely, they were often closely linked during discussions. The concept of 'fair innings' describes the consideration given to those who have experienced, or are expected to experience, comparatively less of a normal span of healthy life.[47] Fair innings, panellists felt, constitute the fair entitlement of children to the same healthy longevity as everyone else in society. They argued that it is equitable and justifiable to prioritize resource allocation to children to avert premature death or diminish long-term physical, cognitive and psycho-social harms imposed by diseases arising during childhood. Relatedly, children were seen to have more future potential. The future potential was described by panellists in both societal and personal terms, as socioeconomic contributions in future adulthood and as prospective personal growth and fulfilment. Panellists overwhelmingly believed that children ought to be prioritized over adults, observing 'as much as …you love your grandmother, they've already lived up to their potential …rather than someone who is 10 years old…' (Panellist P3-13) and '[children] …have a right to have the same amount of time on this Earth as anybody else' (Panellist P4-5).

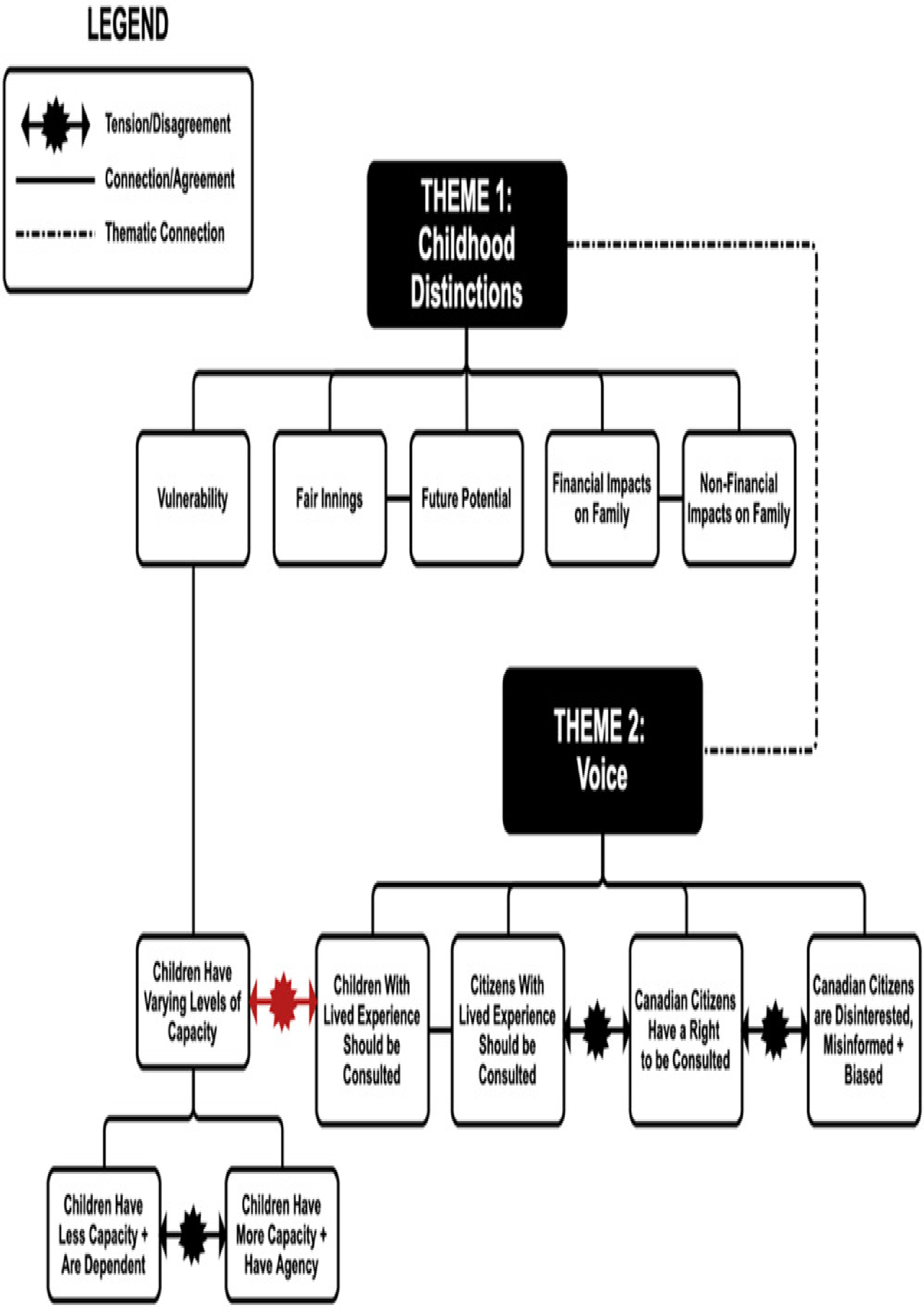
Family impacts

Panellists strongly emphasized that the near-exclusive reliance of children with complex and chronic illnesses on their families marked their distinction from adults. They emphasized that the impacts of illness routinely spill over to the activities and well-being of the entire family, including to siblings. Yet, in conventional HTA processes, impacts on the child are evaluated in isolation. Financial costs, such as travel costs for treatment and lost wages, and nonfinancial costs, such as stress and distress, were equally emphasized by panellists. Caregiver panellists most eloquently shared their insights on family impacts: '…before my daughter was on the modulator, there was two years where she had a really rough time, sometimes we were at clinic every week and you see five or six different practitioners every time' (Panellist P4-3). For this family, and many others, the intensity of treatments for children required an enormous investment of time and energy by all members of the family. Advocacy for their children, whether in the health system to access hard-to-obtain therapies or in the policy sphere to raise awareness among decision-makers, also required substantial effort.

Theme 2: Voice

The issue of which perspectives are most relevant and legitimate as input into funding decision-making for paediatric precision therapies, and therefore whose voices should be heard in medical and evaluative processes, was the central, and most discordant, theme across all panels. Panellists concurred that the public voice, which they considered distinct from the patient voice, was a key input into funding policies, based on shared beliefs in the universality of the Canadian healthcare system. However, they disagreed on the importance of input from those with lived experience and those without. Some panellists without lived experience deferred to the expertise of those with lived experience. As one panellist shared, ‘I think as a [member of the] general population, if you don't understand the severity, the suffering, …the non-financial costs to family of dealing with these diseases and the outcomes and the treatment, it's hard to, you know, prioritize these [values] … when you're weighing other possible considerations’ (Panellist P1-3). Interestingly, caregivers themselves supported including more expansive and removed perspectives in HTA processes, including those without lived experience. Tension was also evident amongst those who thought being a Canadian guaranteed the exercise of voice in a universal healthcare context. Others thought caution was necessary, given misinformation or ignorance about drug regulatory or funding processes, as well as the presence of strong vested interests.

Figure 2 shows the points of tension between subthemes within ‘Childhood Distinctions’ and ‘Voice’ and the connection to children's voices through the subtheme ‘Vulnerability’. A particularly rich discussion is highlighted through the subthemes ‘Children have varying levels of capacity’ and ‘Children with lived experience should be consulted’, within which there was strong discordance in participants' perspectives of children's involvement in clinical decision-making. Participants dissented on the specific age when children might attain capacity, reflecting their personal experiences besides the varying ages of legal consent in provincial jurisdictions. Some panellists argued that some younger children do have the capacity to understand their care and should have the agency to voice their opinions. As one panellist expressed, ‘I've met a number of teenagers and even some kids around 9, 10 years old who would blow you away with their perspectives on the cancer treatment they're going through and the impact it's having on them’ (Panellist P2-1).



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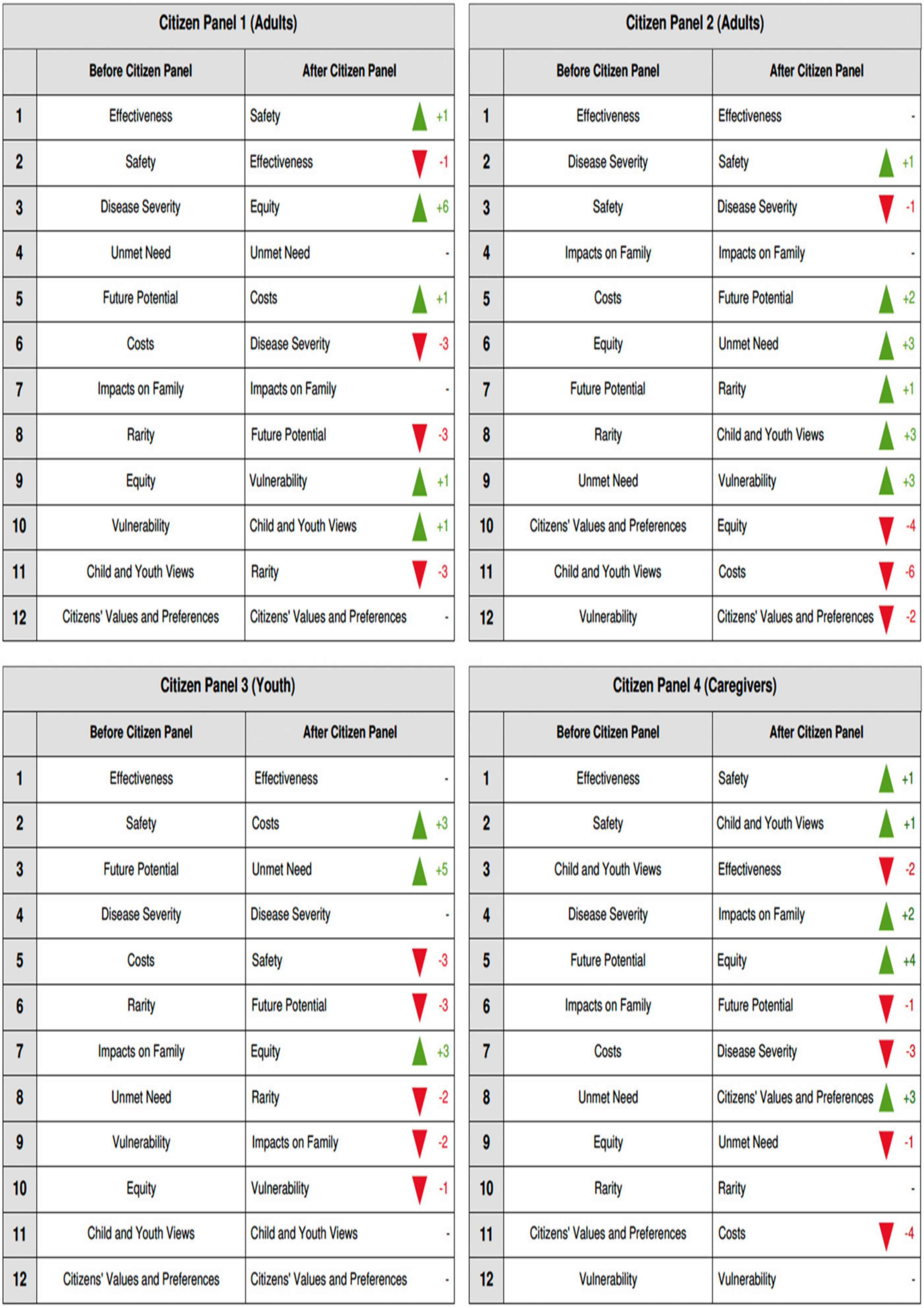
The dissent also manifested in the pre- and postdeliberation ranking of potential HTA values (Figure 3 below). The general public and youth panellists ranked the value of ‘children and youth views’ with low importance for HTA evaluations. As one panellist expressed, ‘honestly, I feel like the child and youth views can probably be taken off [the list of values] completely…’ (Panellist P3-4). Conversely, caregivers not only ranked it highly in the predeliberation poll, third after effectiveness and safety but elevated it further to the second position in the postdeliberation poll.



Citizen Panel 1 (Adults)

	Before Citizen Panel	After Citizen Panel	
1	Effectiveness	Safety	▲ +1
2	Safety	Effectiveness	▼ -1
3	Disease Severity	Equity	▲ +6
4	Unmet Need	Unmet Need	-
5	Future Potential	Costs	▲ +1
6	Costs	Disease Severity	▼ -3
7	Impacts on Family	Impacts on Family	-
8	Rarity	Future Potential	▼ -3
9	Equity	Vulnerability	▲ +1
10	Vulnerability	Child and Youth Views	▲ +1
11	Child and Youth Views	Rarity	▼ -3
12	Citizens' Values and Preferences	Citizens' Values and Preferences	-

Citizen Panel 2 (Adults)

	Before Citizen Panel	After Citizen Panel	
1	Effectiveness	Effectiveness	-
2	Disease Severity	Safety	▲ +1
3	Safety	Disease Severity	▼ -1
4	Impacts on Family	Impacts on Family	-
5	Costs	Future Potential	▲ +2
6	Equity	Unmet Need	▲ +3
7	Future Potential	Rarity	▲ +1
8	Rarity	Child and Youth Views	▲ +3
9	Unmet Need	Vulnerability	▲ +3
10	Citizens' Values and Preferences	Equity	▼ -4
11	Child and Youth Views	Costs	▼ -6
12	Vulnerability	Citizens' Values and Preferences	▼ -2

Citizen Panel 3 (Youth)

	Before Citizen Panel	After Citizen Panel	
1	Effectiveness	Effectiveness	-
2	Safety	Costs	▲ +3
3	Future Potential	Unmet Need	▲ +5
4	Disease Severity	Disease Severity	-
5	Costs	Safety	▼ -3
6	Rarity	Future Potential	▼ -3
7	Impacts on Family	Equity	▲ +3
8	Unmet Need	Rarity	▼ -2
9	Vulnerability	Impacts on Family	▼ -2
10	Equity	Vulnerability	▼ -1
11	Child and Youth Views	Child and Youth Views	-
12	Citizens' Values and Preferences	Citizens' Values and Preferences	-

Citizen Panel 4 (Caregivers)

	Before Citizen Panel	After Citizen Panel	
1	Effectiveness	Safety	▲ +1
2	Safety	Child and Youth Views	▲ +1
3	Child and Youth Views	Effectiveness	▼ -2
4	Disease Severity	Impacts on Family	▲ +2
5	Future Potential	Equity	▲ +4
6	Impacts on Family	Future Potential	▼ -1
7	Costs	Disease Severity	▼ -3
8	Unmet Need	Citizens' Values and Preferences	▲ +3
9	Equity	Unmet Need	▼ -1
10	Rarity	Rarity	-
11	Citizens' Values and Preferences	Costs	▼ -4
12	Vulnerability	Vulnerability	-

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Theme 3: One versus Many

There was a tension between views with humanitarian or utilitarian underpinnings, which emphasized opposing considerations about the individual and the collective as well as equality versus equity. These topics were most prominently articulated in the Youth panel.

Panellists with humanitarian views held that all Canadians deserve equal access to precision therapies in the healthcare system, regardless of their age, geographic location, the rarity of their disease, the price of the therapy and the jurisdictional authority over funding, the more so if the therapy has already been approved for marketing. They believed that no segment of the population should be prioritized over another. Evoking the sanctity of life one panellist argued, '…you can't put a price on someone's life. So definitely every single person deserves equal attention …or treatment. …we can't say this …person is more important than the other one' (Panellist P2-12). Conversely, those expressing utilitarian values centred their perspectives around the notion of achieving the greatest good for the greatest number of people when allocating finite health system resources. Considering the rare nature of diseases that are treated with precision medicine and opportunity costs, a handful of panellists suggested diverting funding away from precision therapies for children and youth and towards medicines and treatments for more prevalent diseases, which may benefit a larger proportion of the population.

Theme 4: Health System Governance

Equity in access was at the heart of panellists' repeated expressions for the Canadian federal government to take a larger or even exclusive role in resolving unequal and variable funding for paediatric precision therapies between provinces. Some panellists even went so far as to suggest that the federal government claims jurisdiction over health care from the provinces. One panellist asserted they '…think what we really need to do in Canada …is …start looking at a constitutional amendment to put more medical in the federal rather than provincial realm' (Panellist P2-10). Federal government-led funding mechanisms and coordination between provincial HTA bodies held appeal for many panellists, all with a view to a more integrated process of drug approval, regulation and funding evaluation. Given the geographical vastness of Canada, to address inequities in access, panellists thought that government could consistently provide travel support for children facing socioeconomic constraints to participate in clinical trials or to obtain already-funded treatments. In acknowledging the limits of the public purse, although, several panellists suggested patient/family out-of-pocket payment models such as co-pay, or payment scaled to income, with provisos to safeguard equity of access.

A caregiver's ability, time and level of medical and legal literacy to advocate government on behalf of their children emerged as a unique factor contributing to inequity. One caregiver observed that 'The steps, the policies, the bureaucracy … that's just another division that I can see forming is that the families that have the financial resources, but also like the [health] literacy resources to do this fighting will have, you know, maybe a better chance than the families who aren't doing this kind of active fighting and pushing' (Panellist P4-2).

Despite panellists' strong opinions on the role of government, and even among caregivers immersed in efforts to access paediatric precision drugs, there was a low level of awareness on federal and provincial HTA processes and their relationships to drug regulation and funding decisions at the provincial level.

Pre- and postdeliberation questionnaires on ranking of HTA values

The dynamic nature of panellists' debates across the four panels was captured by absolute and relative changes in how panellists ranked an a priori list of HTA-relevant values (Figure 3) before and after deliberations. 'Equity' rose in importance in three of the four panels, while 'Cost' had the largest single drop amongst the values. Although panellists had expressed appreciation for being invited to provide their views, 'Citizens' Values and Preferences' ranked unchangingly near or at the bottom of three panels. The rankings of the caregiver panel stood apart from those of other panels, with 'Child and Youth Views' ranked among the highest values alongside therapeutic effectiveness and safety. Notably, by the end of their panel discussion, caregivers ranked 'Child and Youth Views' higher than 'Effectiveness'.

DISCUSSION

Our study illustrates citizens' contributions to an 'upstream' phase of the HTA process, the development of a value framework, focussing on precision therapies for children and youth. We undertook a multi-step approach to preparing an evidence-informed brief and eliciting citizens' views through a method of consultation based on DPE.

Panellists asserted that existing HTA frameworks could and should be revised to incorporate intrinsic, commonly held societal values associated with children and childhood and to better reflect child and family perspectives on the experiences of chronic illness. The biological, physiological and social distinctiveness of children compared to adults—particularly as encapsulated by the values of family impacts, vulnerability, 'fair innings' and future potential—was expressed as the principal reason to develop a child-tailored HTA framework.

Although a child-tailored value framework resonated strongly with our panellists, some important dissenting views emerged. Tensions founded on competing ethical views surfaced, such as the prioritization of health care for a population versus treatment for a few. When discussing vulnerability, panellists debated the delimitation of childhood, the balance of paternalism versus agency and autonomy for children, the reach of rights-based protections for children as positive rights to access health technologies, and distinctions between the pursuit of equality versus equity. Many of the tensions among our panellists arose from dissenting views on age-based prioritization of resources. Youths supported a neutral weighting of age, despite their own age proximity to the paediatric population under discussion.

Dissent was perhaps most pronounced in relation to the issue of representation, or 'voice', in HTA decision-making. Panellists were divided as to the relative importance of those with lived experience. Further, whether and how to incorporate children's voices in evaluating technologies figured prominently in panellists' debates. Differences of opinion were evident in each panel, rather than sharply demarcated between caregivers and noncaregiver panels, as might be expected. Indeed, the most pointed debate about the legitimacy of children's voices arose in the Youth panel, none of whom were caregivers or parents. Finally, assessment rules for child health technologies distinct from those employed for adults were seen by some to clash with population-level decision-making in the context of Canada's universal public healthcare system.

In engaging the nonpatient public, including youth, we identified the wide range of values and the trade-offs important to those who do not have a personal stake in a particular technology funding decision yet are subject to the financial and opportunity costs resulting from it. Values identified by our panellists as particularly salient to child precision health technologies—such as rarity, unmet need and family impacts—have varying resonance in other settings. A UK study using broad preference elicitation methods demonstrated that disease rarity was not prioritized by its citizen respondents.[48] On the other hand, CADTH and INESSS have adapted to the changing pharmaceutical landscape by adopting unmet needs as a criterion for establishing therapeutic value.[49,50] In several European countries, values uncovered through expert deliberation that are not traditionally or formally used in decision-making are framed as contextual or implementation factors. Similarly, in New Zealand's drug funding framework, the individual's connection to family and society is explicitly considered but not formally integrated into reimbursement decisions.[51] As in our study, other research has identified hope as a potential value for extending HTA frameworks, particularly in the context of heightened evidentiary uncertainty and clinical risk.[52]

The potential for significant family impacts in paediatric contexts emerged as a key justification for extended or supplemental HTA frameworks focussed on child health.[53,54] Enabling a specific focus on the unique economic and noneconomic costs that accrue to caregivers and siblings of children with complex or chronic illnesses may allow a more adequate and comprehensive assessment of the value paediatric therapies.[30,55]

We found that most panellists in our study, when prepared with evidence-based resources, were eager and able to present views on evaluating paediatric precision therapies. The pre- and postdeliberation polls suggested that when panellists are given the chance to hear divergent views in a facilitated forum, they may make reasoned changes in judgement. Similarly, a Dutch study using DPE to examine citizens' preferences for inclusions in a social health insurance benefits package found that panellists could articulate a wide range of considerations on a complex topic, learn from the views of others and evolve their opinions through the course of deliberations.[56] As with the panellists in our study, the Dutch panellists prioritized a number of criteria new to conventional HTA frameworks, including 'societal side effects', 'taking away from people' and 'lifestyle'. Motivated by accelerating cancer therapy costs in Canada, DPE has been applied to explore the contentious issue of defunding cancer drugs in British Columbia[57] and to generate recommendations for fairer, more sustainable cancer drug funding for the entire Canadian population.[58]

In the former study, panellists were concerned with defunding and its implications for equity of access and patient choice. In the latter, panellists felt that higher drug costs could be justified through improvements in patients' independence and mental health.

Emerging evidence suggests that DPE may result in improved health outcomes in certain settings.[59,60] In Canada, as elsewhere, the operationalization of DPE in HTA processes remains nascent. CADTH is actively reviewing the integration of deliberative processes into the appraisal stage of its HTA process,[39] while INESSS has recently instituted a flexible evaluation framework that can be adapted to integrate public perspectives.[50] Variability in defining the 'public' remains a key issue for scoping the specific role of participants, however.[61–63] Patients and the public are often viewed interchangeably, and the wider public can be seen to include experts (e.g., clinicians, pharmacists), decision-makers (e.g., government funders, third-party payers), other vested stakeholders (e.g. patient/disease advocates, caregivers) or all citizens of a political jurisdiction. Our study lends support to the greater incorporation of children and youth as a legitimate source of public input for HTA framework development and decision-making. However, implementation issues abound, including how to structure opportunities for participation appropriate to children's evolving developmental capabilities.

Our study has a number of strengths. It is, to our knowledge, the first structured, evidence-informed deliberative process to engage the general public in value assessment framework development for paediatric health technologies, whether for precision therapies or otherwise. Incorporating the opinions of the nonpatient public in DPE is rare, and in HTA framework development, rarer still.[37,60,64] By eliciting, distinguishing and reporting on the varied perspectives of citizens with and without lived experience of rare or hard-to-treat paediatric diseases, it provides a broad canvas of values relevant to child HTA, while also mapping out internal territories marked by the varied, and sometimes conflicting, perspectives of different types of 'publics'. This variation is highlighted by the distinctive perspectives of our youth panellists, who are systematically underrepresented in HTA processes and in DPE more broadly. Finally, our deliberative engagement panels were informed by evidence briefs developed in consultation with a broad range of expert stakeholders, including relevant jurisdictional drug regulatory and HTA bodies, ensuring procedural rigour and optimising the potential for close alignment with real-world policy needs and priorities.

A few limitations of our study are worth noting. Whereas most published experience with DPE to date has been premised on in-person events, the COVID-19 pandemic necessitated a wholly virtual format for panel meetings; panellists may have self-selected to participate on the basis of comfort and technological capability with online meetings. Although a representative national sample was sought, there were no panellists from Canada's northern territories or the province of Newfoundland-Labrador (largely rural regions with sparse populations and limited internet coverage outside capital cities). Consequently, some perspectives on access and equity issues were missed. Participants were predominantly English speakers, so our findings may retain less relevance for regions where different languages or cultures predominate, such as Québec and Nunavut. More generally, our findings should be cautiously applied to other jurisdictions internationally, each of which has its own admixture of sociopolitical and health system realities, with varied bearing on the regulation and reimbursement of health technologies for children and youth.

CONCLUSION

Citizens' perspectives afford rich insights into the range and multi-dimensionality of criteria relevant to decision-making about the allocation of scarce public funds for novel health technologies. They can serve as a key source of values to ground the design of normatively holistic and societally responsive value frameworks for adjudicating the worth of health technologies. Our study adds evidence on values relevant to priority-setting for paediatric precision therapies that can inform the development of child-focused tools and processes for HTA in Canada and comparable health systems internationally.

Future research is warranted to explore and specify the role of citizens in various aspects of HTA processes, including whose voice should be incorporated, how to balance diverse perspectives among varied groups and the optimal ways to operationalize citizen engagement in deliberative approaches to HTA. In the case of precision therapies, particularly for rare diseases, focussing on the distinctiveness of child health is essential, given the preponderance of genetically based diseases in childhood. How to balance the hopes and expectations of patients and caregivers for access to expensive but potential life-altering therapies for relatively rare diseases against the opportunity costs borne by encompassing health systems remains a fundamental challenge. Rigorous methods to elicit, weigh and reconcile views from varied members of society, including children and youth, will prove an essential part of the response to this challenge if decisions about which health technologies we value are to reflect our shared and diverse social values.

AUTHOR CONTRIBUTIONS

Cindy L. Gauvreau, Lisa Wight, Mathushan Subasri, Michael G. Wilson and Avram Denburg conceived and conducted the study. They drafted, edited and reviewed the Citizen Brief and this manuscript. Avram Denburg and Michael Wilson facilitated the citizen panels. Cindy L. Gauvreau, Lisa Wight, Mathushan Subasri prepared and analysed the data. Antonia Palmer, Robin Hayeems, Alysha Croker, Julia Abelson, Brent Fraser, Yvonne Bombard and Charlotte Moore Hepburn were members of the steering committee of this study and provided expert and technical advice. They assisted in key informant and panellist recruitment, reviewed and edited the Citizen Brief and reviewed and edited this manuscript. Antonia Palmer also prepared a video for presentation at the citizen panels.

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CONFLICT OF INTEREST

A. C. is a full-time employee of Health Canada. The opinions expressed in this manuscript are the authors' own and do not reflect the views of Health Canada and may not be understood or quoted as being made on behalf of, or reflecting the position of, the organization with which the author is affiliated. The remaining authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

Data that support the findings of this study are available on request from the corresponding author. The data are not publicly available due to privacy or ethical restrictions.

ETHICS STATEMENT

The Hamilton Integrated Research Ethics Board (HiREB) provided approval for the citizen panels (HiREB project # 13-369). The Hospital for Sick Children provided overall approval for this project (REB #1000069221).

DETAILS

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Robles Arvizu, J.,A., Mann-Jackson, L., Alonzo, J., Garcia, M., Lucero, R. A., Smart, B. D., & Rhodes, S. D. (2023). Experiences of peer navigators implementing a bilingual multilevel intervention to address sexually transmitted infection and HIV disparities and social determinants of health. Health Expectations, 26(2), 728-739. doi:https://doi.org/10.1111/hex.13698

IntroductionSexually transmitted infections (STIs) and human immunodeficiency virus (HIV) disproportionately affect young gay, bisexual and other men who have sex with men (GBMSM) and transgender women of colour. We explored the experiences of community-based peer navigators (‘Community Navigators’) who participated in Impact Triad, a bilingual multilevel intervention developed by our community-based participatory research partnership to reduce STIs and HIV and address social determinants of health (e.g., employment, education, social support and discrimination) among young GBMSM and transgender women of colour.MethodsIndividual in-depth interviews were conducted with 15 Community Navigators who participated in Impact Triad. Themes were identified through constant comparison.ResultsCommunity Navigators' mean age was 31.4 years. Seven were self-identified as African American/Black, 5 as Latine, 2 as multiracial/multiethnic, 1 as Asian American, 10 as cisgender men, 4 as transgender women and 1 as gender nonbinary. Thirteen themes emerged in three domains: (1) key aspects of the Community Navigator role (e.g., desire to serve as a community resource, the importance of being part of the communities in which one was working, the value of having an official role, being connected to other Community Navigators to problem-solving and sustaining intervention aspects long-term); (2) experiences implementing Impact Triad (e.g., engaging community members, meeting prioritized needs, building trust, using social media, increasing awareness and knowledge and challenges related to COVID-19) and (3) lessons learned for future interventions (e.g., facilitating access to broader resources, building additional skills and increasing interactions among Community Navigators).ConclusionInterviews identified important learnings about serving as Community Navigators and implementing Impact Triad that can guide future efforts to address STI/HIV disparities and social determinants of health through community-based peer navigation.Patient or Public ContributionThroughout this intervention trial, our partnership worked collaboratively with a study-specific community advisory board (CAB) comprised primarily of young GBMSM and transgender women of colour. Members of this CAB participated in all aspects of the trial including trial design, intervention development, recruitment and retention strategies, data collection and analysis, interpretation of findings and dissemination.

Corrigendum. (2023). Health Expectations, 26(2), 940. doi:https://doi.org/10.1111/hex.13695

Paisi, M., Allen, Z., & Shawe, J. (2023). New home, new you: A retrospective mixed-methods evaluation of a health-related behavioural intervention programme supporting social housing tenants. Health Expectations, 26(2), 752-764. doi:https://doi.org/10.1111/hex.13700

BackgroundSocial housing tenants are at greater risk of engaging in unhealthy behaviours than the general population. Housing association employees are in an ideal position to contribute positively to their tenants' health. ‘New Home, New You’ (NHNY) is a joint venture between a social housing association, a city council and a community healthcare provider in the South West of England. It was designed with the aim of improving the health and well-being of social housing tenants.ObjectivesThe aim of this retrospective evaluation was to establish whether social housing tenants were benefiting from this health-related behavioural intervention in terms of their mental well-being and health-related behaviours.MethodsThis was a mixed-methods evaluation. The outcome evaluation was a secondary analysis of quantitative data collected during the NHNY project. The process of delivering and receiving the intervention was evaluated using qualitative, semi-structured interviews with housing officers and tenants who had participated in the programme. The development of the intervention was evaluated through a focus group and additional semistructured interviews with key stakeholders. Quantitative data were analysed using the Statistical Package for the Social Sciences. Qualitative interviews were analysed using thematic analysis.ResultsSix key stakeholders and a total of seven housing officers from several teams and seven tenants were interviewed. Of the 1016 tenants who were invited to participate in NHNY, 226 enroled in the programme. For participating tenants, the scope for health-related behaviour change was greatest in relation to eating and smoking. Small positive statistically significant changes in mental health were found between the 6- and 12-month mean score and between the baseline and the 12-month score.ConclusionsThe findings indicate that NHNY may have been beneficial for some participating tenants. Housing officers can have a significant role in promoting health messages and embedding behaviour change among their tenants. Although the programme was implemented as a service improvement rather than a controlled trial and focused on a particular intervention and geographical area, other housing associations may find this evaluation useful for considering whether to adopt some of the principles applied in NHNY in other settings.Patient or Public ContributionA social housing tenant representative provided input on the methodology and methods used to evaluate NHNY, as well as the information sheet.

Moniz, S., Karia, A., Ahmad, F. K., & Vindrola-Padros, C. (2023). Stories for change: The impact of public narrative on the co-production process. Health Expectations, 26(2), 919-930. doi:https://doi.org/10.1111/hex.13718

IntroductionInvolving service users in health service design and delivery is considered important to improve the quality of healthcare because it ensures that the delivery of healthcare is adapted to the needs of the users. Co-production is a process used to involve service users, but multiple papers have highlighted the need for the mechanisms and values guiding co-production to be more clearly stated. The aim of this paper was to evaluate the mechanisms and values that guided the co-production approach of the Stories for Change project, which used Public Narrative as part of the co-design process to create change in National Health Service maternity services.MethodsThis study was conducted using a rapid feedback evaluation approach. Semistructured interviews (n = 16) were the main source of data, six of which were maternity service users, with observations (5 h) and documentary analysis also carried out in parallel. RREAL sheets were used for data analysis to organize data based on key topics of interest.ResultsThis study identified three broad mechanisms and values underpinning the co-production approach: creating an open and safe space to share ideas, learning how to tell stories using Public Narrative and having service providers who play a key role in strengthening the health system listen to stories compelling them to action. This study identified the main areas for improvement of the Stories for Change project related to recruitment, the inclusion of participants, the co-design process, the Skills Session and the Learning Event.ConclusionOur study provided a deeper understanding of the co-production approach that addresses the need to uncover the mechanism and values underlying co-production and co-design approaches. This study expands on the literature pertaining to the influence of storytelling in creating meaningful change in health care. We propose a co-design methodology that uses Public Narrative as a model for service user engagement to help inform future healthcare development processes.Patient or Public ContributionThe experiences and perceptions of maternity service users and health professionals informed this evaluation. The project organizers were involved in the manuscript preparation stage by providing feedback, and service users wrote a commentary on the project from the lived experience perspective.

Elliott, M. J., McCarron, T. L., Schick-Makaroff, K., Getchell, L., Manns, B., & Fernandez, N. (2023). The dynamic nature of patient engagement within a canadian patient-oriented kidney health research network: Perspectives of researchers and patient partners. Health Expectations, 26(2), 905-918. doi:https://doi.org/10.1111/hex.13716

IntroductionCanadians Seeking Solutions and Innovations to Overcome Chronic Kidney Disease (Can-SOLVE CKD) is a pan-Canadian health research network that engages patients as partners across 18 unique projects and core infrastructure. In this qualitative study, we explored how research teams integrated patient partners into network research activities to inform our patient engagement approach.MethodsTo capture a breadth of perspectives, this qualitative descriptive study purposively sampled researchers and patient partners across 18 network research teams. We conducted 4 focus groups (2 patients and 2 researchers; n = 26) and 28 individual telephone interviews (n = 12 patient partners; n = 16 researchers). Transcripts were coded in duplicate, and themes were developed through an inductive, thematic analysis approach.ResultsWe included 24 patient partners and 24 researchers from 17 of the 18 projects and all core committees within the network. Overarching concepts relate participants' initial impressions and uncertainty about patient engagement to an evolving appreciation of its value, impact and sustainability. We identified four themes with subthemes that characterized the dynamic nature of patient engagement and how participants integrated patients across network initiatives: (1) Reinforcing a shared purpose (learning together, collective commitment, evolving attitudes); (2) Fostering a culture of responsive and innovative research (accessible supports, strengthened process and product); (3) Aligning priorities, goals and needs (amenability to patient involvement, mutually productive relationships, harmonizing expectations); (4) Building a path to sustainability (value creation, capacity building, sustaining knowledge use).ConclusionsOur findings demonstrate the dynamic and adaptive processes related to patient engagement within a national, patient-oriented kidney health research network. Optimization of support structures and capacity are key factors to promote sustainability of engagement processes within and beyond the network.Patient or Public ContributionThis project was conceived in collaboration with a Can-SOLVE CKD patient partner (N. F.), with lived experience of kidney failure. He also co-designed the study′s protocol, led focus groups and researcher interviews, and contributed to data analysis. L. G. has lived experience as a caregiver for a person with CKD and facilitated patient partner focus groups. The patient partners, both of whom are listed authors, provided important insights that shaped our interpretation and presentation of study findings.

Blomberg, K., & Hälleberg-Nyman, M. (2023). Experiences of human papillomavirus self-sampling by women >60 years old: A qualitative study. Health Expectations, 26(2), 818-826. doi:https://doi.org/10.1111/hex.13707

BackgroundHuman papillomavirus (HPV) self-sampling has shown to be acceptable and feasible across cultures and effective in reaching women who do not participate in regular cervical cancer screening. However, most of these studies have included younger women. There is a lack of knowledge of how older women reason about HPV self-sampling.ObjectiveThe aim of this study was to describe how women (>60 years old) experience the offering of self-sampling of HPV, compared to having a sample collected by a healthcare professional.Design and ParticipantsThe study had a qualitative explorative design. Four focus group discussions were conducted with women 60–69 years old (n = 22). Data were analysed using principles of interpretive description.ResultsFive themes were identified: self-sampling—convenient and without pain, lack of knowledge, worries related to HPV self-sampling, need for information and taking a societal perspective.ConclusionWomen aged >60 years found that HPV self-sampling was convenient and easy to perform. Further, they stressed the importance of being able to remain in the screening programme in advanced age and that self-sampling could be a possible solution. This study also revealed a lack of knowledge among women in this age group regarding HPV infection, how the disease is transmitted and its relation to cervical cancer.Public ContributionWomen who had performed HPV self-sampling participated in the focus group discussion.

Morris, R. L., Giles, S., & Campbell, S. (2023). Involving patients and carers in patient safety in primary care: A qualitative study of a co-designed patient safety guide. Health Expectations, 26(2), 630-639. doi:https://doi.org/10.1111/hex.13673

BackgroundInvolving patients is a key premise of national and international policies on patient safety, which requires understanding how patients or carers want to be involved and developing resources to support this. This paper examines patients' and carers' views of being involved in patient safety in primary care and their views of potentially using a co-designed patient safety guide for primary care (PSG-PC) to foster both involvement and their safety.MethodsA qualitative study using semistructured face-to-face interviews with 18 patients and/or carers in primary care. Interviews were transcribed and analysis was conducted using an inductive thematic approach.ResultsOverall participants expressed enthusiasm for the PSG-PC as a tool to support patients and carers to be involved in patient safety in primary care. However, for some participants being involved in patient safety was seen as taking on the role of General Practitioner and had the potential to add an additional workload for patients. Participants' willingness or ability to be involved in patient safety was influenced by a range of factors including an invisible, often underacknowledged role of everyday safety for patients' interactions with primary care; the levels of involvement that patients wanted in their care and safety and the work of embedding the PSG-PC for patients into their routine interactions with primary care. Participants identified components of the PSG-PC that would be useful to them, in particular, if they had a responsibility for caring for a family member if they had more complex care or long-term conditions.ConclusionInvolving patients and carers in patient safety needs a tailored and personalized approach that enables patients and carers to use resources like the PSG-PC routinely and helps challenge assumptions about their willingness and ability to be involved in patient safety. Doing so would raise awareness of opportunities to be involved in safety in line with personal preference.Patient or Public ContributionPatient and public involvement were central to the research study. This included working in partnership to develop the PSG-PC with patients and carers and throughout our study including in the design of the study, recruiting participants, interpretation of findings.

Ward, A., Jensen, A. M., M.Sc, Ottesen, A. C., M.Sc, & Thoft, D. S. (2023). Observations on strategies used by people with dementia to manage being assessed using validated measures: A pilot qualitative video analysis. Health Expectations, 26(2), 931-939. doi:https://doi.org/10.1111/hex.13719

BackgroundAnalysis of video data was conducted of validated assessments with people with dementia as part of a feasibility control study comparing a lifelong learning service with other dementia services.ObjectiveThe aim was to provide a new perspective on what occurs during the assessment process when using validated measures in research and explore which strategies people with dementia use to manage their participation.DesignVideo recordings were made of pre- and postintervention assessments of people with dementia. An initial pilot analysis of 10 videos of the pre-assessments was conducted.SettingLifelong learning services and other dementia services situated in six municipalities in Northern Denmark took part in this study, with 55 people with dementia participating.ResultsThe themes identified were: ‘State of mind’ and ‘Mental resources’, showing how these aspects influenced the participants' reactions and the strategies they used.DiscussionThe results are discussed in relation to how individual personality traits influence the assessment process and the way a person with dementia will manage the situation.ConclusionThe assessment situation is complex and can be influenced by the strategies adopted by individuals with dementia as they try to manage the assessment process.Patient or Public ContributionPeople with dementia supported the decision-making for the choice of validated measure used within this study.

Pienaar, M., Marais, L., Serekoane, M., Marais, K., Cloete, J., Lenka, M., & Sharp, C. (2023). ‘Being a mother is not child's play’: The capabilities of mothers in a low-resource setting in south africa. Health Expectations, 26(2), 651-661. doi:https://doi.org/10.1111/hex.13689

BackgroundThe importance of a child's first 1000 days has now been widely accepted by the medical fraternity. Yet, we do not know much about caring practices in low-resource settings.AimThis study aimed to investigate the caring capabilities of mothers in a low-resource setting.MethodIn this study, in-depth interviews were conducted with 18 mothers with children aged 30 months or younger to better understand the arrangements, means and ends that inform developmental health in a low-resource setting in South Africa. The study was conducted in a low-income area, the former black township of Mangaung in Bloemfontein. The mothers were recruited via pamphlets, and two interviews followed. Because of Covid-19, interviews took place via mobile phones, in Sesotho, the local language in the area. Trained fieldworkers conducted, translated and transcribed the interviews. We used thematic analysis and the capabilities approach as the theoretical framework to analyse the responses from the mothers.FindingsWe used the following organizing themes: pregnancy and ante-natal care, nutrition, cognitive and physical development, the home environment and access to health care. Although short-term reactions to pregnancy were often negative, the longer-term responses showed that the respondents have agency. Most of them could change their nutrition habits, breastfeed and receive adequate nutrition support from the public health system. Most experienced joy when their children reached milestones (cognitive and others), although they became anxious if milestones were not reached. They emphasized children's play and had dreams for their children's futures. Technology was often mentioned as playing a role in their children's development. A large proportion of the respondents had disrupted homes (because of absent or abusive fathers), but some had stable homes. Most of them showed substantial capability to overcome adverse home environments. The public health system helped them deal with their health problems and their children's health problems, although it also created anxiety in many cases. Our data show how they develop their capabilities and overcome obstacles organically in the face of resource limitations. Despite pregnancies being unexpected and unplanned and fathers being absent, the respondents accepted the pregnancy, adjusted their diets and social behaviour, showed agency by attending primary healthcare facilities and ensured that their children received the required vaccinations. Their extended families played an important role in providing care. Despite the sacrifices, the respondents expressed joy and helped their children function by eating, playing, socializing, learning and using their senses.ConclusionOur sample of mothers have the agency to adapt to the demands of parenthood and childcare and overcome adversity. Our data support the notion that mothers are held disproportionately and unfairly responsible for achieving the first 1000 days ideals. Despite considerable curtailment of their functionings and capabilities, they nevertheless showed agency to ensure their health and their children's health. A holistic approach should consider these findings in designing policy interventions for children's developmental health.Patient and Public ContributionWe used paid fieldworkers to interact with the research participants.

Ruby, E., McDonald, S. D., Berger, H., Melamed, N., Li, J., Darling, E. K., . . . Murray-Davis, B. (2023). Exploring patients' perspectives of gestational diabetes mellitus screening and counselling in ontario: A grounded theory study. Health Expectations, 26(2), 827-835. doi:https://doi.org/10.1111/hex.13708

IntroductionGestational diabetes mellitus (GDM) is associated with adverse perinatal outcomes. Approaches to screening for GDM continue to evolve, introducing potential variability of care. This study explored the impact of these variations on GDM counselling and screening from the perspectives of pregnant individuals.MethodsFollowing a Corbin and Strauss approach to qualitative, grounded theory we recruited 28 individuals from three cities in Ontario, Canada who had a singleton pregnancy under the care of either a midwife, family physician or obstetrician. Convenience and purposive sampling techniques were used. Semi-structured telephone interviews were conducted and transcribed verbatim between March and December 2020. Transcripts were analysed inductively resulting in codes, categories and themes.ResultsThree themes were derived from the data about GDM screening and counselling: ‘informing oneself’, ‘deciding’ and ‘screening’. All participants, regardless of geographical region, or antenatal care provider, moved through these three steps during the GDM counselling and screening process. Differences in counselling approaches between pregnancy care providers were noted throughout the ‘informing’ and ‘deciding’ stages of care. Factors influencing these differences included communication, healthcare autonomy and patient motivation to engage with health services. No differences were noted within care provider groups across the three geographic regions. Participant experiences of GDM screening were influenced by logistical challenges and personal preferences towards testing.ConclusionInforming oneself about GDM may be a crucial step for facilitating decision-making and screening uptake, with an emphasis on information provision to facilitate patient autonomy and motivation.Patient or Public ContributionParticipants of our study included patients and service users. Participants were actively involved in the study design due to the qualitative, patient-centred nature of the research methods employed. Analysis of results was structured according to the emergent themes of the data which were grounded in patient perspectives and experiences.

Janet Delgado Rodríguez, Ramos-García, V., Infante-Ventura, D., Suarez-Herrera, J., Rueda-Domínguez, A., Serrano-Aguilar, P., & María del Mar Trujillo-Martín. (2023). Ethical, legal, organizational and social issues related to the use of scalp cooling for the prevention of chemotherapy-induced alopecia: A systematic review. Health Expectations, 26(2), 567-578. doi:https://doi.org/10.1111/hex.13679

IntroductionScalp cooling (SC) aims to prevent chemotherapy-induced alopecia. The goal of this systematic review is to tackle ethical, legal, organizational and social issues related to SC.MethodsA critical appraisal of the literature was carried out using a systematic review design. MEDLINE, Embase and Web of Science databases were searched up until 2 June 2021. Studies addressing these aspects in English or Spanish were considered. Representatives of both patient associations and professional scientific societies related to the topic participated in the design of the protocol and the review of the findings.ResultsA total of 17 studies were included. Articles were critically appraised using the MMAT and SANRA. Findings were organized into four categories: (1) ethical aspects focused on equal access, gender equity and doctor–patient communication supported by Patient Decision Aids (PtDAs); (2) patient perspective and acceptability; (3) professional perspective and acceptability; (4) organizational aspects focused on accessibility and feasibility.ConclusionCancer patients' expectations when using SC need to be adjusted to reduce the potential distress associated with hair loss. PtDAs could help patients clarify their values and preferences regarding SC. Equal access to technology should be guaranteed.Patient or Public ContributionIn this systematic review, the representatives of the patient associations (Ms. María Luz Amador Muñoz of the Spanish Association Against Cancer AECC] and Ms. Catiana Martinez Cánovas of the Spanish Breast Cancer Federation FECMA]) participated in the review of the study protocol, as well as in the results, discussion and conclusions, making their contributions. In the type of design of these studies (systematic reviews), it is not usual to have the direct participation of patients, but in this one, we have done so, as it is a systematic review that is part of a report of the Spanish Network of Health Technology Assessment Agencies (ETS).

Nampoothiri, V., Mbamalu, O., Surendran, S., Bonaconsa, C., Pennel, T., Boutall, A., . . . Charani, E. (2023). The elephant in the room: Exploring the influence and participation of patients in infection-related care across surgical pathways in south africa and india. Health Expectations, 26(2), 892-904. doi:https://doi.org/10.1111/hex.13715

ObjectiveThe irrational use of antibiotics is a leading contributor to antibiotic resistance. Antibiotic stewardship (AS) interventions predominantly focus on prescribers. This study investigated the influence and participation of inpatients in infection-related care, including antibiotic decision-making, within and across two tertiary hospitals in South Africa (Cape Town) and India (Kerala).MethodsThrough ethnographic enquiry of clinical practice in surgical pathways, including direct nonparticipant observation of clinical practices, healthcare worker (HCW), patient and carer interactions in surgical ward rounds and face-to-face interviews with participants (HCWs and patients), we sought to capture the implicit and explicit influence that patients and carers have in infection-related care. Field notes and interview transcripts were thematically coded, aided by NVivo 12® Pro software.ResultsWhilst observational data revealed the nuanced roles that patients/carers play in antibiotic decision-making, HCWs did not recognize these roles. Patients and carers, though invested in patient care, are not routinely involved, nor are they aware of the opportunities for engagement in infection-related decision-making. Patients associated clinical improvement with antibiotic use and did not consider hospitalization to be associated with infection acquisition or transmission, highlighting a lack of understanding of the threat of infection and antibiotic resistance. Patients' economic and cultural positionalities may influence their infection-related behaviours. In the study site in India, cultural norms mean that carers play widespread but unrecognized roles in inpatient care, participating in infection prevention activities.ConclusionFor patients to have a valuable role in AS and make informed decisions regarding their infection-related care, a mutual understanding of their role in this process among HCWs and patients is crucial. The observed differences between the two study sites indicate the critical need for understanding and addressing the contextual drivers that impact effective patient-centred healthcare delivery.Patient or Public ContributionEthnographic observations and interviews conducted in this study involved patients as participants. Patients were recruited for interviews after obtaining signed informed consent forms. Patients' identities were completely anonymized when presenting the study findings.

Albert, A., Islam, S., Haklay, M., & McEachan, R. R. C. (2023). Nothing about us without us: A co-production strategy for communities, researchers and stakeholders to identify ways of improving health and reducing inequalities. Health Expectations, 26(2), 836-846. doi:https://doi.org/10.1111/hex.13709

IntroductionCo-production with communities is increasingly seen as best practice that can improve the quality, relevance and effectiveness of research and service delivery. Despite this promising position, there remains uncertainty around definitions of co-production and how to operationalize it. The current paper describes the development of a co-production strategy to guide the work of the ActEarly multistakeholder preventative research programme to improve children's health in Bradford and Tower Hamlets, UK.MethodsThe strategy used Appreciative Inquiry (AI), an approach following a five-step iterative process: to define (Step 1) scope and guide progress; to discover (Step 2) key issues through seven focus groups (N = 36) and eight in-depth interviews with key stakeholders representing community groups, and the voluntary and statutory sectors; to dream (Step 3) best practice through two workshops with AI participants to review findings; to design (Step 4) a co-production strategy building on AI findings and to deliver (Step 5) the practical guidance in the strategy.ResultsNine principles for how to do co-production well were identified: power should be shared; embrace a wide range of perspectives and skills; respect and value the lived experience; benefits should be for all involved parties; go to communities and do not expect them to come to you; work flexibly; avoid jargon and ensure availability of the right information; relationships should be built for the long-term; co-production activities should be adequately resourced. These principles were based on three underlying values of equality, reciprocity and agency.ConclusionThe empirical insights of the paper highlight the crucial importance of adequate resources and infrastructure to deliver effective co-production. This documentation of one approach to operationalizing co-production serves to avert any misappropriations of the term ‘co-production’ by listening to service users, stakeholders and other relevant groups, to develop trust and long-term relationships, and build on the learning that already exists amongst such groups.Patient or Public ContributionThe work was overseen by a steering group (N = 17) of individuals, both professional and members of the public with experience in undertaking co-production, and/or with some knowledge of the context of the two ActEarly field sites, who provided regular oversight and feedback on the AI process.

Nathan, S., Hodgins, M., Wirth, J., Ramirez, J., Walker, N., & Cullen, P. (2023). The use of arts-based methodologies and methods with young people with complex psychosocial needs: A systematic narrative review. Health Expectations, 26(2), 795-805. doi:https://doi.org/10.1111/hex.13705

BackgroundArts-based methodologies and methods (ABM) can elicit rich and meaningful data with seldom-heard groups and empower participants in research. Young people with complex psychosocial needs could be better engaged in research using arts-based approaches to overcome communication and literacy issues as well as distrust of those with power, including researchers. A critical review of the use and impact of ABM among this population is timely. The purpose of this review is to synthesize and examine the experience and use of ABM with young people with complex psychosocial needs.MethodsA systematic narrative literature review was conducted with a search of the literature from 2009 to 2021. All abstracts were reviewed independently by two authors and full papers were screened for eligibility against inclusion and exclusion criteria. Data synthesis focused on a descriptive numerical summary and a thematic analysis focused on key patterns across papers relating to the review objectives.Results and DiscussionA total of 25 papers were included. The most common issues of focus were mental health (n = 10) and homelessness (n = 11) and methods using Photovoice (n = 12) and Body Mapping (n = 5). Individual interview data (n = 20) were the most commonly analysed, followed by created works (n = 19). Less than half the studies involved young people in the interpretation of the data collected. Knowledge translation was not described in almost half the studies, with public exhibits (n = 7) and forums with service providers (n = 4) being the most common activities. Key themes across the studies were valued over traditional methods in eliciting data, ABM as an approach to engage these young people in research and the impact of the use of ABM on participants and on key stakeholders through knowledge translation.ConclusionsThe growing field of ABM presents opportunities to enhance research with young people with complex psychosocial needs by promoting meaningful exploration of experiences, engaging participants in research and strengthening knowledge translation. The involvement of young people in the interpretation of data and ensuring that knowledge translation occurs are key areas for future attention.Patient or Public ContributionThe findings of this review will inform future research to improve the engagement of young people with complex psychosocial needs in research and promote power sharing between researchers and research participants.

Ramfelt, K., Boel Andersson Gäre, Andersson, A., & Petersson, C. (2023). ‘It's like a never-ending diabetes youth camp’: Co-designing a digital social network for young people with type 1 diabetes. Health Expectations, 26(2), 662-669. doi:https://doi.org/10.1111/hex.13690

IntroductionLiving with a chronic condition such as type 1 diabetes (T1D) affects everyday life and support from others experiencing a similar situation can be helpful. A way to receive such support is to use an online network where people can connect and share experiences. Research has described the benefits of using such tools for connecting patients. The aim of this study was to describe the co-design of a social network for young people with T1D and to describe their experiences when using this network.MethodsA co-design approach was used, following three steps adapted from Sanders and Stappers (2008). In all, 36 adolescents with T1D participated. Data in the form of recordings and notes from telephone interviews, workshops and focus groups were collected and then analysed using content analysis. Numerical data from the digital platform were also used.FindingsFor the interpersonal values, supporting, learning and relating to emerge, the framework of the network must be appealing and user-friendly. The limits of time and place are eliminated, and there is a possibility for many more to join in.ConclusionCo-design ensures that what stakeholders think is important forms the basis for the design. The interpersonal values that are promoted are ones that only the exchange of lived knowledge and experience can generate. It is complementary to the support that healthcare professionals can offer; thus, this kind of social network is important for improved, coproduced care.Patient or Public ContributionThe participants in the present study were persons living with T1D. They were active co-creators from the start to the end. An adult person with experience of living with T1D was involved as an advisor in the research team when drafting the manuscript.

Manley, K., Saunders, K., Wilkinson, D., Faruqui, R., & Sakel, M. (2023). Co-creating system-wide improvement for people with traumatic brain injury across one integrated care system in the united kingdom to initiate a transformation journey through co-production. Health Expectations, 26(2), 869-881. doi:https://doi.org/10.1111/hex.13712

Background and ObjectiveThere is a need for better integration of services across communities and sectors for people living with traumatic brain injury (TBI) to meet their complex needs. Building on insights gained from earlier pilot work, here we report the outcomes of a participatory workshop that sought to better understand the challenges, barriers and opportunities that currently exist within the care pathway for survivors of TBI.MethodsA diverse range of stakeholders from the acute and rehabilitation care pathway and the health and social care system were invited to participate in a 3-h workshop. The participants worked in four mixed subgroups using practice development methodology, which promotes person-centred, inclusive and participatory action.ResultsThematic analysis identified shared purposes and values that were used to produce a detailed implementation and impact framework for application at both the level of the care interface and the overarching integrated care system. A variety of enablers were identified that related to collective values and behaviours, case management, team leadership and integrated team working, workforce capability, evidence-based practice and resourcing. The clinical, economic, cultural and social outcomes associated with these enablers were also identified, and included patient safety, independence and well-being, reduced waiting times, re-admission rates, staff retention and professional development.ConclusionThe co-produced recommendations made within the implementation and impact framework described here provide a means by which the culture and delivery of health and social care services can be better tailored to meet the needs of people living with TBI. We believe that the recommendations will help shape the formation of new services as well as the development of existing ones.Patient or Public ContributionPatient and public involvement have been established over a 10-year history of relationship building through a joint forum and events involving three charities representing people with TBI, carers, family members, clinicians, service users, researchers and commissioners, culminating in a politically supported event that identified concerns about the needs of people following TBI. These relationships formed the foundation for the interactive workshop, the focus of this publication.

Schackmann, L., Hek, K., Vervloet, M., Koster, E. S., & Liset, v. D. (2023). Provision of and trust in COVID-19 vaccines information: Perspectives of people who have had COVID-19. Health Expectations, 26(2), 806-817. doi:https://doi.org/10.1111/hex.13706