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Original Research

Consent for newborn screening: screening professionals' and parents' views



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ABSTRACT

Objectives: Expansion of newborn bloodspot screening (NBS) within England, which practices an informed consent model, justified examining acceptability and effectiveness of alternative consent models.

Study design: Qualitative focus groups.

Methods: Forty-five parents and 37 screening professionals (SPs) participated. Data were analysed using thematic analysis.

Results: Parents and SPs initially appeared to have differing views about appropriate consent models. Most parents accepted assumed consent, if adequately informed; however, once aware of bloodspot storage, informed consent was wanted. SPs valued informed consent, but acknowledged it was difficult to obtain. Both samples wanted parents to be informed but were unclear how this could be achieved. Most parents felt NBS was not presented as optional.

Conclusion: The simultaneous exploration of parents and SPs views, in real time is original. This rigour avoided the reliance on retrospective accounts which make it difficult to establish how decisions were made at the time. It is also unique in providing pre-interview consent models to drive the depth of data. It was rigorous in member checking. Findings suggested a preference for full disclosure of all information with some parents valuing this more than choice. Both samples queried whether current consent was sufficiently informed and voluntary. Results suggest differing tolerances of consent type if screening is solely for diagnostic purposes vs bloodspot storage. Results highlight the need for caution when examining consent model preferences without also checking knowledge, as opinions may be based on incomplete knowledge. Future research is needed to examine efficacy of proposed changes.

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Introduction

Newborn screening involves taking samples of blood from a baby's heel (the bloodspot test) to screen for serious conditions in babies within the first days of life. Early identification enables timely treatment, leading to improved prognosis and quality of life. However, consent for newborn bloodspot screening (NBS) is complex. Consent models differ internationally ranging from mandatory screening—the state decides to screen; 'opt out models'—the state recommends but parents can decline to voluntary informed consent—the state recommends, but parents indicate willingness for screening to be performed. Where consent is needed, parents are deciding on behalf of their newborn—known as proxy consent. The information which needs to be understood is novel and vast; there are numerous rare diseases (9 conditions in England) which are unknown to many,¹ there are a range of possible screening results: diagnosis, inconclusive diagnosis, carrier, suspected carrier, false positive and normal. The results can have implications for family members' genetic risk and paternity, and bloodspots may be stored for anonymised research. Finally, consent is taken 5 days after birth when parents are tired and processing volumes of information regarding their child. Concerns have been raised about the validity of consent for NBS in practice as uptake is high in most countries,^{2–7} yet parental awareness and knowledge levels are low.^{8–12, 30, 34, 35, 36, 37, 38, 39, 40}

NBS in England is promoted as only performed after receiving valid informed consent from parents¹³ but is offered to parents routinely. Midwives inform parents and distribute a screening booklet. However, this high standard of consent can be difficult to achieve as outlined above. Two Health technology assessment (HTA) studies have reported concerns regarding whether parents in England were adequately informed before screening.^{15,16} If decisions about screening are not sufficiently informed, consent validity is questionable. This study sought to examine parent and screening professional (SP) preferences for different NBS consent models.

Methods

Participants

As previous work¹⁷ evidenced variation in Newborn screening programme (NBSP) communication practice which will affect views of consent, all Senior Quality Assurance Managers in England were sampled to capture maximum practice variation ($n = 6$). Through these a regional quality assurance manager ($n = 1$), hospital screening coordinators ($n = 13$), community midwives ($n = 14$) and hospital midwives ($n = 3$) were sampled proportional to NBS involvement and purposively to capture variance in experience and regional coverage. The SP sample ($N = 37$) covered 16 trusts in rural and urban areas of England. All regions of England were represented.

Parents were sampled across the screening pathway: parents-to-be ($n = 14$), parents 'screened waiting for result'

Research in context

Evidence before this study

Countries vary in the consent model used. In countries with informed consent models, concerns exist about the validity of consent. Although all countries value informing parents about screening, there is a lack of an effective model. The increase in the number of conditions included in newborn screening raised questions about what consent model is most appropriate.

Added value

This is one of the first studies to explore the views of parents and screening professionals simultaneously and in real time. Previous research has either separated these samples, therefore, because newborn bloodspot screening (NBS) is constantly evolving, parents and health professionals may be providing views on different forms of NBS, or it has relied on retrospective parental accounts or subsamples of parents affected or professional groups. This 360 design has enabled contrasts to be made between participants and the unpicking of what is driving opinions. Furthermore, its novel design of a pre-interview contemplation exercise enabled more in-depth data to be gathered, enabling the team to understand views which are likely to drive responses to NBS. The design highlighted the risk of creating policy on research without a clear idea of participants knowledge and the damage to public trust that can be done when parents become aware of key screening information after screening has been performed. The above not only adds to the evidence around optimum prescreening information provision but also can help explain why parents may have such negative responses to positive screening results.

Implications of all available evidence

Both parents and screening professionals value parents being informed so highly that it drove their preferences for different consent models. Indeed information was more valued than choice for some.

Parents hold newborn screening in high regard, but this may be based on a belief of screening confirming a state of health and are often are unaware of their choice or of other potential outcomes or bloodspot storage. This high regard and the use of leaflets may be driving low parental engagement with screening information, whereby screening is performed because of parental trust in the NHS.

Designing materials which prioritise information about choice and bloodspot storage may increase parental engagement with information and help to achieve the goal of parents feeling sufficiently informed.

($n = 6$), parents who received normal results ($n = 15$), parents of children with a metabolic condition ($n = 8$ [Phenylketonuria (PKU); $n = 4$, medium chain acyl-CoA dehydrogenase deficiency (MCADD); $n = 2$, Glutaric Aciduria Type 1 (GA1) $n = 2$]), parents whose child had a repeat bloodspot sample ($n = 2$). Sampling was conducted across the pathway as retrospective recall designs are problematic, and immediacy recall designs have been advocated in NBS.²⁰

Recruitment of postresult parents occurred nationally via laboratories because of the rarity of conditions, final sampling occurred via social media and parent groups to enhance geographical and service experience diversity in our sample. Recruitment of parents before receipt of results occurred in the North West of England via antenatal clinics and community midwives. Local family centres were used to reach more diverse parents. Exclusion criteria: Parents whose child had died or was born prematurely, who had NBS performed >180 days ago, or where multiple abnormalities were identified, who lacked capacity to consent. We sought to ensure all participants who had received positive results did so in the previous year to minimise recall bias and control for communication practice changes which have weakened previous studies.¹⁶ Table 1 below displays participants' sociodemographic characteristics.

Interviews

Parents were given outlines of alternative consent models before interview so they could consider their views before the interview. The semistructured interview schedule was designed by health psychologists with expertise in qualitative methods and NBS communication. It was developed from a realist review,¹⁵ guided by a scientific advisory panel, parent reference group and NBS advisors, and was piloted with two parents.

Participants were asked about their views on consent models with prompts directing them to discuss factors which might affect acceptability or recipients' understanding within each model. Parents were also asked how they were informed antenatally and reflected on their understanding (data reported elsewhere¹⁵). Interviews were audio recorded and transcribed professionally. SPs participated in telephone interviews to facilitate their participation.^{17,21} Parents chose face-to-face or telephone interviews.

'Member checking', a form of validation in qualitative research,²² was conducted by providing participants with project summaries and inviting them to participate in focus groups or telephone interviews. Representatives of laboratories, other screening programmes and the Royal College of Midwives were also invited to participate to explore the reach of the results creating a total SP sample of 17 for this phase.

Data analysis

Data were analysed using thematic analysis which seeks and reports patterns inherent in data sets.²³ A realist stance was adopted; the data were seen as reporting participants' actual reality and experiences.²³ A deductive, manifest level²⁴ approach used the different consent models to frame the analysis.²⁵ Aspects of grounded theory which enhance rigour

Table 1 – Sociodemographic characteristics of sample ($n = 45$).

Parents' sociodemographic characteristics	N (%)
Age in years	
18–25	5 (11.1)
26–30	8 (17.8)
31–35	15 (33.3)
36–40	17 (37.8)
Ethnicity	
White British	26 (57.8)
White European	6 (13.3)
Black African	3 (6.7)
Pakistani	3 (6.7)
White Irish	2 (4.4)
Black Caribbean	1 (2.2)
North African	1 (2.2)
British Chinese	1 (2.2)
Mixed	1 (2.2)
White Other	1 (2.2)
Primary language	
English	35 (77.8)
Urdu	3 (6.7)
French	2 (4.4)
Polish	2 (4.4)
German	1 (2.2)
Romanian	1 (2.2)
Ukrainian	1 (2.2)
Employment status	
Employed full-time	25 (55.6)
Employed part-time	7 (15.6)
Full-time parent	3 (6.7)
Full-time student	2 (4.4)
Self-employed	5 (11.1)
Maternity leave	3 (6.7)
Highest educational attainment	
Degree or higher degree	30 (66.7)
College level or equivalent	13 (28.9)
High school or equivalent	2 (4.4)
Number of children	
Pregnant with first child	12 (26.7)
Parent of one child	11 (24.4)
Parent of two or more children	22 (48.9)

were used such as iterative data generation and analysis, constant comparative analysis,²⁶ and detailed line-by-line coding. Deviant cases were sought to test the analysis. Coding continued until the analysis described the data set holistically and saturation had been achieved. Analysis was conducted within Nvivo.

Results

Parents' preferences for different consent models are shown in Table 2.

Although there was variation in parents' consent model preferences, they unanimously felt parents should be as fully informed about NBS as possible.

It's about being involved and being made aware as to what is happening, what the options are, what are the advantages,

Table 2 – Parents’ consent preferences from pre-interview tool.

How much choice	Number of parents
(a) Parents can choose if their baby will be screened or not	9
(b) Parents can choose if their baby will be screened or not AND if they say yes, they can choose which conditions they would like their baby to be screened for	7
(c) Healthcare professionals will assume parents want their baby to be screened. If parents do not want their baby screened, they will have to explain the reason why to a healthcare professional	19
(d) Parents do not have any choice about screening as screening is compulsory for all babies	10

disadvantages, effects, that kind of thing [...] being fully informed as to what’s happening to my child. And for my husband as well, not just for me.

(#5, mother, English, 2 children, normal result)

The mention of the husband’s needs here highlights how this is a family decision and both parents need to be informed, which was also present in the health professional data.

Generic consent

Generic consent is a model where parents agree to a process in a broad way (such as biobank research). Although generic consent was not widely supported by parents for the current number of NBS conditions, SPs felt further expansion may necessitate this model:

...[if we were] testing for 20 conditions I think it would have to be a case of looking at a different way of consenting and perhaps examining more closely something like a generic consent.

(#19, trust screening coordinator)

These sentiments were echoed by parents who reflected on the amount of potentially relevant information if NBS was expanded and questioned whether it was desirable or necessary for them to receive this if the National Health Service (NHS) had decided the conditions were suitable for inclusion in NBS, illustrating the trust parents place in the NHS. However, SPs still preferred parents to be fully informed before screening taking place.

Informed consent

Most SPs preferred voluntary informed consent, seeing it as central to their practice:

I think it’s important in everything we do [...] it’s their baby and they’ve got a right to say what tests they want for themselves and their children.

(#22, trust screening coordinator)

For parents who valued consent, their perceived responsibility to act in their child’s best interest was important. For one parent, having a choice about NBS appeared to foster empowerment and confidence as a mother compared with her prior NBS experiences when screening was not perceived as optional:

I had a choice and that made me feel good because it made me feel like a parent, whereas with the other two children I felt like I had no choice and [the midwife] just came and did it

(#34, mother, Polish, needs interpreter, third child, screened no result)

There was widespread acknowledgement that informed consent was difficult to obtain in practice as both samples reported little engagement with information in part because of an overall positive view of NBS:

They don’t want to read the leaflets, they don’t want to be told about it, but they’re happy to have any tests done on their baby for the health of their baby.

(#30, community midwife)

Parents had a different view on engagement and the effect that the leaflet had in achieving consent. They viewed the use of leaflets as reducing their ability to engage with the process. Indeed many parents’ accounts of what they would want mirrors what is meant to occur in informed consent:

But for it to be offered, so this is what we offer; would you like to have that? And then maybe that should be documented as you... so not just having the leaflet and that’s you giving your consent because you’ve been given a leaflet

(#1, mother, English, second child, negative result)

The need to truly achieve informed consent was also voiced by SPs:

.... consent needs to be obtained as well for obvious reasons but I think it needs to be done properly not just like, oh, are you happy to have this?

(Community midwife—London; interview)

Indeed only one parent, a health professional, felt they had made an informed choice about NBS. Early within the health professional focus group, they brought up the need to make it clear to parents that screening was optional. This was also raised spontaneously in the parent focus groups in reaction to in introduction provided about NBS is the fact that screening is optional:

I mean, if, right now, there is a choice, I think it should be... parents should be aware of that, and I don't think they are.

(mother, English, 2 children normal results, focus group)

Assumed consent/opt-out

SPs were generally not in favour of assumed consent because of concerns that parents may not be fully informed:

You've got to maintain your professionalism and in something like screening if you just have an opt-out policy, you are, kind of, doing your own profession an injustice by not giving all the information that you should be giving to the parent.

(#3, Senior Quality Assurance Managers)

Thus, preferences were embedded in how they drove information provision. Parents, however, thought this model would increase information provision and engagement:

...at least you would know whether they'd been informed or not [...] because they're talking to the healthcare professional on why they don't want it.

(#15, father, second child, GA1 diagnosis second child)

A similar response was provided by a mother who received a PKU result. Parents, who had not received a diagnosis, however, also felt parents should be aware of screening implications. They were happy with SPs directing care but wanted to be fully informed. Several SPs thought routine offering of NBS created an assumed consent scenario, yet the importance of fully informing parents was also dominant in their narratives:

I think there's an element of that, to be honest with you, that we already do assume [...] Yeah, we do actually, because we diary it in, that's a routine sort of midwifery...see them on the day they come out of hospital, you then see them on day five for bloodspot and weight...

(#12, trust screening coordinator)

Mandatory screening

Mandating screening was not supported by any SPs who were concerned about the vulnerability they faced as professionals by collecting a sample without parents' consent. This was incongruent to midwifery practice's ethos of providing choice.

So while most things its informed consent such as the Down's Syndrome screening and then to have a mandatory consent for another part of the screening programme it seems to be that the principles are seen to be a little bit conflicting.

(#19, trust screening coordinator)

This resistance to mandatory screening and comparison with screening programmes such as Down's syndrome occurred across the SPs data set. They also worried that

compulsory screening may trigger parents to resist NBS and other services:

In all my time in 12-14 years of being a midwife I've never had one person saying no, but potentially I think you could do if it was compulsory.

(Senior community midwife; interview)

Indeed parents repeatedly stated that they would not decline screening but wanted the choice.

...even though I would have chosen to have had it done, I think that having the choice there is quite important.

(Mother; English; pregnant; interview)

Yet most parents reported NBS had been presented without a choice '... it wasn't, do you want your child to have this test; it was she is having it' (#12, mother, two children, PKU result second child). Many parents contrasted their awareness of choice in NBS to antenatal screening:

... it should be down to the parents, just like the Down's Syndrome test is completely optional and up to yourself.

(#18, Father, PKU result)

Thus, although SPs earlier report a need for all screening to be seen as optional, parents are noting a difference in actual choice in screening programmes. This is despite all screening information being included in one booklet.

Some parents, however, supported mandatory screening

... they should need to have it done. You should want your child... you should want to know that your child is healthy

(#32, mother, first child, pregnant sample)

There is a clear moral imperative suggested by the repeated use of 'should' based on a view that screening is confirming a baby's health, rather than potentially identifying disease. Illustrating that even parents involved in this study had not received or assimilated information as to why choice was important. Indeed, the parent focus group discussion centred around there being 'no risks for the baby' (#1, mother). This may be because of the way they are categorising risk, as one mother explained a different view based on culture:

... someone from my culture, taking my baby's blood is really, really important, it's the life of my baby. [...] So by giving at least the information and the consent form, at least you are putting me at ease.

(Mother #3, French, 3 children, one identified as sickle cell carrier; focus group)

When these parents were asked about choices about bloodspot storage, their views changed dramatically, triggering concerns about trust:

Mother #3: For me, I'm angry that they are using my kid's blood without me even knowing. Even though I know that I will never say no, but doing it almost in a hiding way, it's not really right. Then you are regretting that you have given the consent from, or you have the right to say no. Yeah, honesty is just enormous. It's like I take something from you without you knowing.

Importantly, even when these parents supported mandatory screening, they wanted complete information and believed 'It doesn't cost them anything to tell you verbally and to give you one set of information.' (mother, English, two children, focus group) and talked of parent rights. Of concern, this parent thought some information was purposively withheld:

I don't think anybody likes to have to sit down with somebody and say these are the nine things that potentially could be wrong with your child. They don't want to do that. So on the day they come and take the test, they say, oh, we're going to test for some things, but it will be okay.

(Mother, English, 2 children, focus group)

These data highlight how parents recall what health professionals tell them and place trust in the healthcare system. If they later find out information which is incongruent with the original information, this can have implications on willingness to trust future messages.

Discussion

This study examined the acceptability of different NBS consent models. While parents' preferences varied, they unanimously valued being fully informed with some valuing information more than choice. Indeed, throughout all accounts was a clear desire to ensure parents are informed and their preference for consent model was driven by which model would achieve this. SPs predominantly favoured voluntary informed consent as did parents when aware of bloodspot storage. That parents consistently value choice in regard to bloodspot storage,^{7,28} fits with previous research. Thus if NBS includes bloodspot storage, as in England, a voluntary informed consent model would be supported, fitting with existing health professional guidance.²⁷

The change in parents' views once aware of bloodspot storage highlights the danger of basing consent policy on research where parents may have incomplete knowledge when indicating preferences. Our data illustrate how even research-engaged parents were unaware of bloodspot storage. Indeed, SPs reflected on the challenges of obtaining valid informed consent in current practice, and with the exception of one parent, parents did not feel it was their choice. Importantly, the data highlighted the needs for the couple to be informed not just the mother which fits with previous research outlining the familial influences on screening decisions,³⁰ but also research indicating fathers often feel left out of such communication.³¹

Learning key NBS information after screening appeared detrimental to trust and potentially the public view of NBS. Research shows that communicating clearly about bloodspot

storage can increase parental engagement with information without reducing screening uptake.⁷ Our data suggest lack of parental engagement may, in part, be because of the overall regard parents have of the health benefits of NBS. It may be the high regard which not only drives lack of engagement, but also high levels of trust which in turn makes the outcome more profound when parents realise they did not have full information. This may explain previous research showing that parents are unprepared for screening outcomes and this reduces trust. Redeveloping information models which drive parental engagement, potentially focussing on parental choice and bloodspot storage, may improve how informed parents are, which appears a universal desire.

Support for mandatory was largely driven by a wish to ensure that children did not lose the benefit of being identified and receiving timely treatment; in line with previous literature.^{11,32} However, there is no indication either in this study or the international literature¹⁵ that voluntary informed consent results in parents declining testing.^{7,33} Also as per previous research, the advocates of mandatory screening focused on the identification of children with conditions,³² crucially our data picked up another driver—a misbelief that screening would confirm their child was healthy. This has been found in other areas of genetics as a misattributed drive for engagements with testing. The same parents showed no awareness of carrier identification or bloodspot storage again highlighting the need for consent to explore consent preferences and knowledge.

In our interviews, some parents explicitly said they would not like mandatory screening, and SPs did not feel comfortable with this model. SPs also felt they would need more training and be more knowledgeable about NBS to ensure that they were confident in delivering the information. Previous research has highlighted the challenges of achieving this and argued for a significant increase in midwives.³⁴

The generic consent model³⁵ uses analogies with communication and consent models for biobanking and genetic profiling, both situations where the potential information to convey is vast and evolving, prohibiting in-person communication in terms of time, money and potential utility for the recipient of each piece of information. There are caveats with this model, however, that parents should be enabled to ask questions and additional information to be provided by those who need it, requiring an in-person and tailored approach. In this sample, parents saw it as a possible future model to avoid being overwhelmed with information as found in other studies,¹⁶ which have argued that providing too much information can be counterproductive to the goal of informed consent.

Strengths and limitations

This work is original in combining the views of those taking consent with parents, simultaneously capturing the broadest range of screening experience in real time. This rigour avoided the reliance on retrospective accounts. It is also unique in using a pre-interview consideration exercise to drive the depth of data, capturing opinions more likely to be those which drive reactions, rather than the quick response interviewees may give to be 'a good interviewee' which are more

likely to parrot dominant models. Rigour was furthered by member checking. Aspects of grounded theory which enhance rigour were also used.

Those who do not speak fluent English were supported in participating. We successfully recruited fathers, young parents, and those with lower education achievement who are commonly underrepresented in the research¹⁶ and may have different communication needs. The parent sample is large (N = 45), especially in comparison to previous in-depth interview studies, but more importantly, it captures viewpoints across the screening pathway and ensured that within each subsample, something substantive could be said about consent model preferences. One consideration is that we used an 'opt-in' model of consent for this research. This is generally seen as less likely to lead to coercive but means that participants had to be proactive to participate. Our sample is likely to be skewed towards participants who are functioning at a level which enables them to read research information and find the time to be interviewed. Although this is commonly seen as a limitation, in this setting, it means our sample is more likely to include parents who read and act on information given to them. Therefore, the findings related to gaps in knowledge are of more concern. However, for the same reasons caution should be taken in assuming that parents who have not engaged with the research information would equally value NBS information as found in this study.

Conclusion

Consent model preferences seem to have been driven by a desire to ensure parents are informed. Consent models believed to achieve this are preferred, and acceptance of other models is caveated with a need to ensure parents are still informed. Thus, while people may tolerate changes to consent models, parents being insufficiently informed is not tolerated. Both samples queried whether currently consent was sufficiently fully informed. Results suggest differing tolerances of consent if screening is solely for diagnostic purposes vs bloodspot storage. Findings suggest a need for caution when examining participants' preferences for consent models without checking knowledge.

Author statements

Ethical approval

Ethical approval for the health professionals' interviews was obtained from the University of Manchester Research Ethics Committee (reference: 13198). Ethical approval for the parents' interviews was obtained from the NHS West Midlands Edgbaston Research Ethics Committee (REC reference: 13/WM/0438). R&D approval was gained from trusts for both samples.

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Competing interests

The authors have no conflicts of interest relevant to this article to disclose.

Data sharing statement

Deidentified individual participant data will not be made available.

Related study documents will be made available upon request to corresponding author.

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Short Communication

Sexual orientation and poor psychological health: a population-based study

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ABSTRACT

Objectives: The purpose of this study was to analyse associations between sexual orientation and poor psychological health.**Study design:** This was a cross-sectional study.**Methods:** The 2012 public health survey in Scania, Southern Sweden, is a cross-sectional population-based study with 28,029 participants aged 18–80 years. Logistic regression analyses were performed.**Results:** The prevalence of poor psychological health (measured using the General Health Questionnaire [GHQ]-12) was 16.3% among men and 22.4% among women. Bisexual men and women had significantly higher odds ratios of poor psychological health throughout the multiple analyses than heterosexual individuals. In contrast, the odds ratios of poor psychological health among gay men and lesbian women were not significantly higher.**Conclusions:** Respondents with bisexual orientation have increased risk of poor psychological health.© 2019 The Authors. Published by Elsevier Ltd on behalf of The Royal Society for Public Health. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

Sexual minorities have poorer health than heterosexual individuals in many respects, including higher levels of all-cause mortality,¹ morbidity and indicators of morbidity.² Sexual minorities generally also have increased risk of poor psychological health compared with heterosexual in-

dividuals,³ including anxiety and depression,⁴ most mental health disorders, substance use disorders and suicidality.³

Poor psychological health among sexual minorities compared with that among heterosexual individuals may be caused by factors such as formal legal sanctions and also

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informal factors such as victimisation⁵ and internalised negative feelings.⁶ Sexual minorities have traditionally been marginalised and thus exposed to increased stress (i.e., the minority stress hypothesis). Stress, defined as an imbalance between resources and requirements, may lead to poorer health. Resources include both social and material resources. Social capital, which includes both social participation in activities in society and generalised trust in other people, may be regarded as an important social resource. Socio-economic status (SES) and the ability to pay bills may be regarded as indicators of material resources. The general psychological health of sexual minorities has not been investigated previously in population-based studies in the southernmost part of Sweden; thus, the present study is important.

The aim of this study was to investigate the association between sexual orientation and poor psychological health measured by the General Health Questionnaire (GHQ)-12.

The study population consists of respondents to the cross-sectional public health survey conducted in 2012 in Scania, Southern Sweden, based on a random sample of adults aged 18–80 years, stratified by sex and geographic location, extracted from the public population register of residents. A total of 28,029 respondents returned the questionnaire between August and October 2012 (51.7% response rate). Owing to internally missing values for each of the included variables, only 22,357 respondents were analysed in this study (79.8% of all respondents). Ethical approval was granted by the Ethics Committee in Lund, Sweden.

Self-reported psychological health measured with the GHQ-12 questionnaire includes twelve questions. The first eight questions, which include the item ‘Have you been able to concentrate on what you have been doing during the past few weeks?’ and also address sleeping problems, feeling of usefulness, capability of making decisions, feeling tense, capability to enjoy normal activities, capability to overcome difficulties and feeling of happiness during the past weeks, have four mutually excluding answers: ‘more/better than usual’, ‘as usual’, ‘less than usual’ and ‘much less than usual’. The items were dichotomised with the first two alternatives depicting ‘good’ and the two latter ‘poor’ psychological health. The remaining four questions include the item ‘Have you felt unable to deal with your own personal problems during the past few weeks?’ and questions regarding feeling unhappy and depressed, self-confidence and feeling worthless during the past weeks. These four items had the following answers: ‘not at all’, ‘no more than usual’, ‘more than usual’ and ‘much more than usual’, which were also dichotomised by grouping the first two responses as ‘good’ psychological health and the latter two alternatives as ‘poor’ psychological health. If an individual scored three or more of the twelve dichotomised GHQ-12 items as ‘poor’, this depicted overall poor psychological health; a score of two or less was deemed as overall ‘good’ psychological health.⁷

Sexual orientation was defined by the item ‘Do you regard yourself today as 1) heterosexual, 2) bisexual, 3) homosexual (exact wording used in the questionnaire [rephrased as gay or lesbian in this article]), 4) other?’.

Age was included as a continuous variable.

Participants were dichotomised as born in Sweden or born in a country other than Sweden.

Marital status was defined according to four alternatives as follows: (1) married/cohabiting, (2) unmarried, (3) separated/divorced or (4) widowed.

SES was defined by occupation, employment and insurance categories, including high-level non-manual employees, medium-level non-manual employees, low-level non-manual employees, skilled manual workers, unskilled manual workers, self-employed/farmers, early retirees/sick leave (younger than 65 years), unemployed, students, pensioners (older than 65 years), unclassified/homeworker.

Economic stress was assessed using the item ‘How often in the past 12 months have you had difficulties in paying your bills?’. The responses were as follows: (1) every month; (2) approximately half of the months of the year; (3) maybe once; or (4) never.

Social participation was assessed by participation in activities of formal and informal groups of society (study circle/course at workplace, other study circle/course, union meeting, meeting of other organisations, theatre/cinema, arts exhibition, church, sports event, letter to the editor of a newspaper/journal, demonstration, night club/entertainment, big gathering of relatives, private party). If three or fewer activities were reported during the past year, social participation was defined as low.

Generalised trust in other people was assessed using the item ‘Generally, you can trust other people’ with the following alternatives: ‘do not agree at all’, ‘do not agree’, ‘agree’ and ‘completely agree’. This variable was dichotomised with the two first alternatives as ‘low’ trust and the latter two as ‘high’ trust.

The prevalence of poor psychological health, sexual orientation and the other variables was calculated, stratified by sex (not shown in Table 1). Bivariate analyses of odds ratios with 95% confidence interval of poor psychological health according to sexual orientation and the other variables were calculated (not shown in Table 1). Multiple analyses (odds ratios with 95% confidence intervals) of the association between sexual orientation to poor psychological health were calculated adjusting for the following (Table 1): age (model 1); age, born in Sweden and marital status (model 2); age, born in Sweden, marital status, SES and economic stress during the past year (model 3); age, born in Sweden, marital status, SES, economic stress during the past year, social participation and generalised trust in other people (model 4). Statistical analyses were carried out using the SPSS software, version 24.0.

The prevalence of poor psychological health defined with the GHQ-12 instrument was 16.3% among men, 22.4% among women and 19.3% for both sexes in total. A 96.8% proportion of men, 94.7% of women and 95.8% for both sexes in total reported their sexual orientation as heterosexual. Bisexual orientation was reported by 1.3% of men and 2.2% of women. A 1.4% proportion of the men were gay, and 0.8% of the women were lesbian. ‘Other’ sexual orientation was reported by 1.1% of men and 0.9% of women. Poor psychological health was significantly associated with bisexual and other sexual orientation compared with heterosexual individuals, younger age groups, those born outside Sweden, unmarried and divorced, those on sick leave, unemployed, students, those with economic stress during the past year and those with low generalised trust in other people among both men and

Table 1 – Adjusted odds ratios (ORs) and 95% confidence interval (CI) of poor psychological health according to sexual orientation. Public health survey in Scania 2012 (N = 22,357).

Sexual orientation	Model 1 ^a	Model 2 ^b	Model 3 ^c	Model 4 ^d
	OR (95% CI)	OR (95% CI)	OR (95% CI)	OR (95% CI)
Men				
Heterosexual	1.00	1.00	1.00	1.00
Bisexual	1.84 (1.27–2.67)	1.68 (1.16–2.44)	1.63 (1.10–2.40)	1.66 (1.12–2.45)
Gay	1.09 (0.73–1.61)	1.06 (0.72–1.58)	1.07 (0.71–1.62)	1.11 (0.73–1.68)
Other	1.97 (1.32–2.95)	1.37 (0.89–2.12)	1.37 (0.89–2.12)	1.37 (0.88–2.13)
Women				
Heterosexual	1.00	1.00	1.00	1.00
Bisexual	2.16 (1.68–2.79)	2.11 (1.63–2.73)	1.86 (1.42–2.44)	1.77 (1.34–2.33)
Lesbian	0.77 (0.46–1.28)	0.79 (0.47–1.31)	0.70 (0.41–1.21)	0.71 (0.40–1.26)
Other	2.26 (1.51–3.40)	2.05 (1.36–3.09)	1.77 (1.14–2.76)	1.61 (1.03–2.51)

^a Adjusted for age.

^b Adjusted for age, country of birth and marital status.

^c Adjusted for age, country of birth, marital status, socio-economic status (SES) and economic stress during the past year.

^d Adjusted for age, country of birth, marital status, socio-economic status (SES), economic stress during the past year, social participation and generalised trust in other people.

women. In addition, women in unskilled blue-collar jobs also had poorer psychological health than women with non-manual jobs in higher positions. Pensioners (older than 65 years) of both sexes and widows had significantly lower odds ratios of poor psychological health (not shown in Table 1).

Table 1 shows that men and women with bisexual orientation had significantly higher odds ratios of poor psychological health than heterosexual individuals throughout the age- and multiple-adjusted analyses. In contrast, gay men and lesbian women did not have statistically significant odds ratios of poor psychological health throughout the age-adjusted and multiple-adjusted analyses. These patterns remained even after adjustments for SES, economic stress and social capital. Men with other sexual orientation had significantly poorer psychological health than heterosexual men, but only in the age-adjusted model in Table 1 (model 1), while women with other sexual orientation retained higher odds ratios of poor psychological health than heterosexual women throughout the multiple analyses.

Discussion

The psychological health of sexual minorities should be investigated in population-based studies rather than limited subpopulations with questionable representativeness. The GHQ-12 instrument may be a relevant indicator of psychological health in this context, but the association between sexual orientation and GHQ-12 has scarcely been investigated previously.

The poorer psychological health of bisexual individuals, but not of gay men or lesbian women, than that of heterosexual individuals may be interpreted in terms of juridical and social conditions and the development of these over recent years. In Sweden, registered partnership for same-sex couples was granted in 1995 and marriage in 2009. A US study has shown that same-sex marriage state policies were associated with a reduction in the proportion of high-school student reports of suicide attempts between 1999 and 2015.⁸ The

absence of differences in psychological health between gay men, lesbian women and heterosexual individuals may be interpreted partly in this context. The bisexual group is plausibly more vulnerable because of less change in attitudes towards their sexual orientation.⁴ One conclusion is that the results of this study may not be generalised directly to other countries. Instead, the results should be related to the juridical and sociocultural situation in the specific country under study.

Strengths and limitations

The major strength of this study is that this is a large population-based study. Relevant confounders were also included in the analyses. Limitations include the moderately low participation rate, which reflects the international trends and Western levels. A previous study comparing the respondent population with the general population in Scania based on official register data with regards to age, sex and education concluded that the risk of selection bias was acceptably moderate to low.⁹ The GHQ-12 instrument is well established, and the validity and reliability may be expected to be the same as in other Western populations.⁷ Poor psychological health according to the GHQ-12 instrument has been shown to be associated with suicide attempt and suicide thoughts in the 2012 questionnaire study population in Scania. The reliability of the instrument has not been investigated in Scania. This high validity and reliability also hold for the item regarding sexual orientation.¹⁰ The other items are also well established and valid, although self-report in general, including recall bias and social desirability bias in particular, may have resulted in misclassification to some extent. The study is cross-sectional and thus reflects associations.

Bisexual men and women, but not gay men or lesbian women, had significantly higher odds ratios of poor psychological health throughout the multiple analyses than heterosexual individuals. Studies investigating sexual orientation should be population-based and may include GHQ-12 as an indicator of psychological health. Results

should be interpreted in relation to the juridical and socio-cultural context.

Author statements

Ethical approval

Ethical approval was granted by the Ethics Committee in Lund, Sweden.

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Competing interests

There are no competing interests.

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Original Research

Strengthening the maternal and child health system in remote and low-income areas through multilevel governmental collaboration: a case study from Nujiang Prefecture in China

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ABSTRACT

Objective: The aim of the study was to assess the effects of the maternal and child health (MCH) system strengthening through multilevel governmental collaboration in Nujiang Prefecture, China.

Study design: A case study design was applied.

Methods: Guided by the logical framework of the Nujiang MCH Comprehensive Intervention Project, national, provincial, and prefecture government sectors jointly implemented comprehensive health system strengthening (HSS) interventions in Nujiang Prefecture. In this case study, we conducted the end point surveys (November 2015 and January 2016) with 33 local MCH facilities. We also interviewed 35 MCH providers, government officials, leaders of MCH facilities, and project specialists. The thematic framework method was used to analyze the interview data, and descriptive analysis was performed to analyze the survey data.

Results: The three levels of governmental collaboration contributed to increased government investment in the local MCH system and ensured the successful implementation of the project. Participatory training methods and appropriate HSS interventions tailored to the local context were crucial to improve MCH providers' knowledge and skills, with the proportion of qualified MCH providers increasing from 70% in 2011 to 96% in 2015. Owing to this increase in knowledge and the increase in needed equipment, more MCH hospitals could provide inpatient obstetric services, and more town health clinics were capable of providing basic MCH services. The development of a reimbursement policy tailored to the local context promoted in-hospital delivery. At the conclusion of the project, percentages of antenatal care, in-hospital delivery, and newborn screening increased by 20.71%, 18.12%, and 278.62%, respectively. Growth monitoring coverage for children younger than three

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years remained stable at around 90%. However, the MCH system was negatively impacted by the workforce shortage. Those shortages were caused by a lack of positive recruitment and retention and incentive policies.

Conclusions: Implementation of comprehensive HSS interventions through multilevel governmental collaboration improves the MCH system in remote and low-income areas.

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Introduction

In recent years, accessibility and quality of China's health services have improved continuously owing to the rapid development of its health system.¹ However, disparities among various regional health systems remain, including the maternal and child health (MCH) system. The capacity of the MCH system is inadequate in many remote and low-income areas of China, which has led to the exacerbation of unmet health needs and subsequent health problems, such as maternal mortality and child malnutrition.² A study on maternal mortality ratios in 2852 counties of China found that the 2015 maternal mortality ratio in Zanda County, Tibet, western China, was 830.5 per 100,000 live births, which is 38 times higher than the national average for that year.³ Some surveys on child nutrition have shown that the prevalence of stunting in remote and low-income areas of China has ranged from 17.8% to 25.8%, far exceeding the national average.^{2,4,5} A strong MCH system is fundamental to improving MCH outcomes. Therefore, there is an urgent need to improve MCH systems in remote and low-income areas, which commonly has been facilitated by implementing health system strengthening (HSS) interventions.^{6,7}

In remote and low-income areas, it is difficult for the local health sector to strengthen the MCH system owing to the limited workforce, facilities, equipment, and financial resources. Previous studies have shown that collaboration plays an important role in HSS,⁸ however, inadequate attention has been paid to the MCH system. Several studies have reported that successful collaboration among different levels of government can help improve the ability of local MCH facilities to collect and use data, develop MCH policies, provide quality midwifery services, and achieve MCH-related goals.^{9,10,11,12} However, these studies focused only on one specific element of the health system rather than multiple components, such as leadership and governance, service delivery, health workforce, health financing, medical products, and health information. Moreover, none of those studies focused on remote and low-income areas. To fill the gap in this area of focus, there is an urgent need to undertake research to examine whether and how collaboration between multiple levels of the government can improve the MCH system in remote and low-income areas.

Nujiang Prefecture in Yunnan Province of southwest China is a remote, low-income area where the natural environment is challenging, and MCH outcomes are the worst in the country. From 2012 to 2015, three levels of the government

sector (MCH Department of the National Health Commission, Yunnan Provincial Health Commission, and Nujiang Prefecture Government) jointly implemented the Nujiang MCH Comprehensive Intervention Project (hereafter, 'Nujiang Project') that focused on MCH system strengthening, health education, and provision of nutritional supplements.

As part of the Nujiang Project, this study aims to evaluate the effects of MCH system strengthening through multilevel governmental collaboration in Nujiang Prefecture. Using these findings, we advocate the experiences from Nujiang Prefecture be applied to similar remote and low-income areas to develop their MCH systems and improve health outcomes.

Methods

Study design

Based on the theory of change and child health intervention theories, we developed the logical framework of the Nujiang Project, consisting of four components: background, activities, outputs, and outcomes. Guided by this framework, three levels of the government sector jointly implemented the project. Based on the findings of the project demand analysis,² comprehensive HSS interventions, including health workforce training, establishment of a newborn screening network, equipment allocation, and policy making, were developed and implemented in four counties in Nujiang Prefecture (Lushui County, Fugong County, Gongshan County, and Lanping County) from September 2012 to October 2015.

In this study, we used a case study design to assess the effects of comprehensive HSS interventions on the MCH system in Nujiang Prefecture. In November 2015 and January 2016, we conducted end point surveys to collect detailed information on the project implementation process, changes in the MCH system and their associated reasons, and bottlenecks or constraints of the MCH system.

Evaluation framework

We developed the evaluation framework based on the health system theory recommended by the World Health Organization and the HSS experience of Nujiang Prefecture.¹³ The framework consisted of five components: investment, personnel, service, equipment, and leadership/governance. 'Investment' was related to the government's investment in the MCH system in remote and low-income areas. 'Personnel' referred to the local MCH providers' knowledge and skills.

'Service' was related to the provision (measured by the 'coverage rate') of MCH services. 'Equipment' referred to the number and usage of equipment in MCH facilities. Finally, 'leadership/governance' referred to the organization and implementation of the project, the health administrators' capacity to make MCH policies, and the effects and shortcomings of the MCH policies.

Data collection

The data sources of this study included interviews, surveys, and relevant policy documents and data. Multiple data sources helped ensure the internal consistency and reliability of this study.

The qualitative interviews took place in November 2015. Based on individuals' availability and recommendations from the local health sector, we recruited 15 health providers from seven prefecture/county/town/village MCH facilities. We divided these health providers into two groups. Each group discussion lasted about 2 h. The discussion focused on health providers' experiences, attitudes, and perceived benefits gained from the MCH system—strengthening activities. Based on the principle of theoretical saturation,¹⁴ we also conducted in-depth interviews with 20 individuals, including five government officials from Nujiang Prefecture and some counties, nine leaders of MCH facilities, and six national/provincial health specialists responsible for project supervision and training. In these in-depth interviews, we explored MCH investment and policy as well as participants' perspectives on the effects of the project, bottlenecks of the MCH system, and their recommendations for improvement. Each in-depth interview lasted about 1 h. Two professional interviewers with knowledge and skills of qualitative research conducted the interviews together.

In November 2015 and January 2016, we conducted end point surveys (after the intervention). We used the same method and questionnaires from the baseline survey (May and June 2012) to collect data from all local MCH facilities ($n = 33$) including the Prefecture MCH hospital ($n = 1$), county MCH hospitals ($n = 4$), and town health clinics ($n = 28$). The surveys from MCH hospitals mainly collected information on the number and quality of MCH providers and the coverage of main MCH services throughout the prefecture or county. The surveys from town health clinics mainly collected information on the provision of MCH services and equipment allocation.

In January 2016, we also collected policy documents, statistical monitoring data, self-evaluation reports, and publications from Nujiang Prefecture and its four counties.

Data analysis

We performed thematic framework analysis to analyze the interview data. Two professional researchers transcribed all audio recordings verbatim. The interview transcripts were the major data source, while notes taken during the interviews provided additional data. Based on the research objectives and evaluation framework, we identified five research themes: investment, personnel, service, equipment, and leadership/governance. For each theme, we generated codes using words,

sentences, and passages. We also labeled quotes relevant to the codes and/or themes and selected the 'best' quotes to support our findings as reported in the Results section.

For the quantitative data, we performed descriptive analysis to compare the output indicators measured before and after the MCH system—strengthening activities. These indicators included the following: number and density of MCH providers, proportion of qualified MCH providers, number of town health clinics that provided basic MCH services, coverage of antenatal care for at least five visits, percentage of in-hospital delivery, growth monitoring coverage for children younger than three years, and rate of newborn screening. We used EpiData (version 3.0) to input data and SAS (version 9.2) to analyze the data.

Results

MCH investment

The collaboration between the three levels of government contributed to increased government investment in the local MCH system. According to policy documents and data, government investment in the MCH system mainly includes financial subsidies and special investments. Owing to the limited fiscal revenue of local government, financial subsidies to the MCH system were relatively insufficient and unable to meet the needs of health services. Special funds for national basic and major public health projects and the Yunnan Province MCH Project have become important sources of investment in the MCH system. An important feature of the Nujiang Project is the integration of existing special funding. According to the Nujiang Project Plan jointly formulated by the three levels of government, the MCH Department of the National Health Commission and the Health Commission of Yunnan Province should '*set up policy priorities and provide funding support for Nujiang Prefecture to implement the project.*'

During the implementation of the project, the two key aforementioned government sectors fulfilled their responsibilities to support Nujiang Prefecture to strengthen the MCH system. The MCH Department of the National Health Commission included Nujiang Prefecture in the National Newborn Screening Project supported the local health sector in establishing a network for newborn screening. Moreover, the MCH Department of the National Health Commission provided financial support for teacher training, expert supervision, and evaluation. In the meantime, the Health Commission of Yunnan Province selected Nujiang Prefecture as a target area for the Yunnan Province MCH Project. This enabled local MCH providers to receive on-site assistance from provincial health experts and attend on-the-job training at the provincial or national hospitals. Local administrators believed that the three levels of governmental collaboration played a pivotal role in contributing to the development of the MCH system.

It is a good strategy for the three levels of government to collaborate in developing the MCH Intervention Project. Such multilevel governmental collaboration has led to the MCH Department of the National Health Commission and the Health

Commission of Yunnan Province paying great attention and providing support to Nujiang. (No.1 prefecture administrator)

MCH personnel

In general, the knowledge and skills of MCH providers improved. In this study, MCH workforce training included annual training by national and provincial experts in Nujiang Prefecture, short-term on-the-job training in higher medical institutions of Yunnan Province, and on-site specialist assistance across the county. Considering the relatively low educational level of local MCH providers, the participatory training methods were adopted, and the training course was focused on basic MCH services such as antenatal care, obstetric services, newborn screening, and child growth monitoring. Most interviewees (68%) believed that the trainings were beneficial, enabling local MCH providers to improve professional knowledge. Interviewees also indicated that local MCH providers were more confident in providing MCH services after attending the trainings. The survey data showed that proportion of qualified MCH providers increased from 70% in 2011 to 96% in 2015.

I worked in a national MCH center for three months, through which I advanced my skills. It is really useful for me to understand some basic concepts that were not clear to me in the past. In addition, I can learn about the experiences of other areas. What I have learned can be applied well in my work. (No.5 MCH provider)

The clinical skills of our MCH providers improved greatly and we are able to provide obstetric services with the help of on-site health experts from the provincial MCH hospital. (No.4 leader of a county MCH hospital)

Traditional training has little effect (on improving MCH outcomes) because of the complex content and knowledge....Over the past two years, changes in the training content have shown positive effects. After attending the training, health providers at the county and town health clinics are able to provide obstetric services, while in the past, they could not do that. (No.1 prefecture health administrator)

According to the interviews, we found that innovative training methods were important to ensure the effectiveness of the intervention. The interviewees were satisfied with the participatory training first used in Nujiang Prefecture. They commented that these trainings were better than the traditional class or lecture-based, non-participatory trainings because the training courses were easy to understand and health providers were more enthusiastic to participate.

Participatory training is useful. MCH providers at many town clinics actively participated in the participatory training. It is better than the traditional training.... (No.1 leader of a county MCH hospital)

This training method is very good! It enables us to understand and accept the content more easily. (No.12 health provider)

The Nujiang Project's impact was limited by the challenges of MCH workforce shortage and inadequate incentives for health providers. Some leaders of MCH facilities stated that the workforce shortage made it very difficult to send MCH providers to attend the training. Many voiced that the workforce shortage was the main reason why some MCH facilities were unable to provide basic MCH services. Some interviewees mentioned that owing to the lack of MCH providers, two of five MCH hospitals could not provide inpatient obstetric services and only the prefecture MCH hospital could provide outpatient pediatric services. The quantitative survey data also demonstrated the workforce shortage problem. During the implementation of the project, the total number of MCH providers in Nujiang Prefecture only increased by 5.09%, whereas the number of MCH providers per 1000 population decreased by 2.38%. In addition, some interviewees expressed that MCH providers lacked motivation to attend the training owing to the inadequate incentives. Specifically, MCH providers received the same income after the training as before the training, which did not match their increased skills, knowledge, and workload.

The limitation (of the training) lies in the lack of staff...We find it difficult to select staff to attend the training and our staff are not motivated to attend the training due to the lack of incentives. There is no (income) difference for staff who attend the training or not, or undertake more or less work. (No.1 leader of the prefecture MCH hospital)

There is a general shortage of MCH providers in Nujiang Prefecture...Two MCH hospitals cannot provide inpatient MCH services and only the prefecture MCH hospital has the capacity to provide outpatient pediatric services. (No.1 prefecture health administrator)

MCH services

The provision of MCH services in Nujiang Prefecture improved. Nujiang Prefecture includes one prefecture and four county MCH hospitals. The interview data showed that the number of MCH hospitals providing inpatient obstetric services increased from two to three. The survey data indicated that the number of town health clinics providing basic MCH services also increased. In addition, the rate of newborn screening for metabolic diseases, percentage of in-hospital delivery, and coverage of antenatal care for at least five visits increased significantly from 2011 to 2015. During the same period, the growth monitoring coverage for children younger than three years remained around 90% (Table 1).

Equipment of MCH services

Allocation of equipment for MCH facilities significantly improved. According to the needs of local MCH services, the equipment (see Table 2) for basic MCH services was determined and purchased. The data showed an increase in MCH hospital ownership of all surveyed equipment, except for the lever weight scale. Furthermore, of the 28 town health clinics surveyed, the number of clinics having at least four types of

Table 1 – Coverage rates of basic MCH services in Nujiang Prefecture from 2011 to 2015.

MCH service	Coverage rate ^a (%)					Cumulative growth (%)
	2011	2012	2013	2014	2015	2011–2015
In-hospital delivery	79.46	86.85	90.83	93.61	95.92	20.71
Antenatal care	60.66	79.79	80.14	78.83	71.65	18.12
Growth monitoring for children younger than three years	89.26	96.57	90.14	90.09	89.89	0.71
Newborn screening	Unknown	21.00	63.89	77.30	79.51	278.62

MCH, maternal and child health.

^a Provision of MCH services was measured by the coverage rate, defined as the ratio between the number of people actually receiving a MCH service and the number of people eligible and entitled to receive a MCH service in a specific region.

equipment increased significantly. Local health administrators attributed some of the project's positive effects on increasing MCH capacity to the appropriate allocation of equipment.

Many MCH facilities have purchased necessary medical equipment. We are very grateful to this project (for enabling this), and since its implementation, the service capacity of our MCH facilities has been enhanced. (No.1 prefecture health administrator)

Owing to the expert supervision and the three levels of governmental collaboration, allocation of equipment to MCH facilities was optimized. For example, most of the equipment was allocated to the prefecture MCH hospital in the past, and insufficient attention was paid to the MCH facilities at the county/town/village level. Furthermore, some of the equipment was not used to the fullest potential. The Nujiang Project's group of experts identified these problems in 2013, and it provided feedback to the three levels of government. In response, the government collaborators exerted a joint effort to solve these problems by reallocating medical equipment that was unused or less often used among MCH facilities. By the end of 2015, most of the medical equipment had come into use.

MCH leadership/governance

To improve the MCH system effectively, the three levels of government jointly established a collaboration mechanism that clearly defined their respective responsibilities. They formulated the project implementation plan together; met regularly to discuss strategies for governmental coordination and project implementation; and established an expert group responsible for implementation, supervision, and evaluation of project activities. An important principle of the project was to involve local governmental sectors and MCH facilities in the entire process, from project design to implementation and evaluation. This not only ensured the feasibility and pertinence of the project plan but also greatly mobilized the enthusiasm of local personnel to participate in the project.

To enhance the capacity of local MCH administrators to make policies tailored to their local context, the Nujiang Project conducted policy making training. Through the interviews, we found that MCH administrators endorsed the content and methods of this participatory training. When interviewers asked about the policy formulated during the project, four interviewees mentioned the development of a reimbursement policy that provided living and transportation allowances for hospitalized pregnant women and their caregivers. Considering the community's challenges of poor public

Table 2 – Number of MCH facilities owning different types of equipment before and after MCH system strengthening.

Equipment	MCH hospitals (n = 5)			Town health clinics ^a (n = 28)		
	Baseline	End point	Change	Baseline	End point	Change
Lever weight scale	2	1	-1	10	20	10
Horizontal bed	2	5	3	9	28	19
Vertical height gauge	1	4	3	13	24	11
Tape measure	3	4	1	22	28	6
Neonatal resuscitation equipment	2	3	1	–	–	–
Automatic chemistry analyzer	2	5	3	–	–	–
Automatic microelement analyzer	1	3	2	–	–	–
Automatic blood instrument	1	3	2	–	–	–
Children overall quality tester	1	3	2	–	–	–
Fetal heart monitor	3	5	2	–	–	–
Full-field digital mammography system	0	1	1	–	–	–
Hearing screening instrument	2	5	3	–	–	–
Infant radiant warmer	2	3	1	–	–	–
Neonatal jaundice tester	1	2	1	–	–	–

MCH, maternal and child health.

^a According to their needs of practical work, township health clinics were equipped with four types of equipment for child growth monitoring.

transport and low-income level, this policy was effective in promoting women to deliver in the hospital.

The training about contextualized policymaking has had a great effect and will benefit our future work. (No.4 leader of a county MCH hospital)

Every county has its own social reimbursement policies. For example, four counties provide living allowances ranging from ¥200 to ¥400 for each pregnant woman and one county provides 25 kilograms of rice. These policies are effective in increasing in-hospital deliveries and decreasing the under-five mortality rate. (No.1 leader of the prefecture MCH hospital)

Some interviewees revealed that the lack of positive staff recruitment and retention and incentive policies has led to workforce shortages.

Existing staff are leaving, while outsiders are unwilling to join our medical facility because of the poor income and working conditions. (No.2 prefecture health administrator)

Due to the lack of effective incentive policies, we cannot implement performance evaluations for our MCH providers. (No.1 leader of the prefecture MCH hospital)

Discussion

To assess how to develop effective MCH systems in remote and low-income areas, comprehensive HSS interventions were implemented in Nujiang Prefecture. These interventions targeted five key areas: investment, personnel, service, equipment, and leadership/governance. The interventions were undertaken through a unique top-down approach involving multilevel (national, provincial, and prefecture/city) governmental collaboration. As mentioned earlier, such a collaboration is complex and likely to meet challenges or conflicts.^{15,16} In our study, the collaboration of the national, provincial, and prefecture/city governments was successful. This was shown with the increased government investment in the local MCH system. The reason for successful collaboration includes two factors. First, the three levels of government shared a common goal to enhance the MCH system in Nujiang Prefecture, and they jointly established a collaboration mechanism that clarified their respective responsibilities. Second, the project plan was jointly developed by the three levels of government. Previous studies have found that stakeholders at the grassroots level that are not engaged in the project design may resist the implementation of the project.¹⁷ In our study, local governmental sectors and MCH facilities participated together in project design to ensure the successful implementation of the project.

Between 2011 and 2015, coverage of most MCH services in Nujiang Prefecture improved significantly. During this time, the rate of newborn screening nearly tripled, in part owing to the establishment of the newborn screening network and staff training. There was a remarkable increase in the percentage of in-hospital delivery, which was most likely due to the living

and transportation allowances provided for hospitalized pregnant women and their caregivers. Previous studies conducted in low-income areas have shown that low household income is a factor preventing women from going to the hospital to deliver.^{18,19} Our study confirmed that a reimbursement policy incentivizes pregnant women to deliver in the hospital.²⁰ Other reasons for the increased percentage of in-hospital delivery may be related to the expansion of obstetric services at the hospital, the improvement of health providers' knowledge and skills, and the increase of necessary medical equipment at many MCH facilities. However, the various factors affecting MCH coverage are complex. In addition to health system factors, there also are a wide range of social and environmental factors affecting MCH coverage, such as the natural environment, public transport, cultural differences, the population's education and income level, and other socioeconomic factors.^{21,22} The MCH system–strengthening interventions in this study explain a component of the reasons for the change in MCH coverage.

The use of participatory training methods in remote and low-income areas enabled positive outcomes. Participatory training was widely used during the health education activities.^{23,24,25} Evidence suggests that participatory training is better than traditional training methods because it considers an individual's experiences and needs and emphasizes the trainer–trainee interaction. In our study, interviewees commented that the participatory training methods were easy to understand and accept, and these methods made MCH providers enthusiastic to participate in the training. From this experience, we support the use of participatory training, rather than the class or lecture-based training, for MCH providers with a relatively low educational level in remote and low-income areas.

Although the comprehensive HSS interventions improved the service capacity of the MCH system in Nujiang Prefecture, some MCH facilities were still incapable of providing some MCH services. For example, two of the five MCH hospitals could not provide inpatient obstetric services, and only the prefecture MCH hospital was able to provide outpatient pediatric services. Interviews suggested that the workforce shortage was the key barrier for the provision of some MCH services, and it caused many MCH providers to not attend the training. As a result, these providers lost the opportunity to improve their knowledge and skills, further limiting their ability to provide effective MCH services. In summary, workforce shortages significantly affect the service capacity of the MCH system. Possible solutions for this prominent issue include developing positive staff recruitment and retention and incentive policies, providing attractive salaries, and offering staff training opportunities through concerted efforts between national, provincial, and prefecture/city governments as well as local MCH facilities.

This study has a number of limitations. First, we adopted a mixed-methods approach to collect data on the changes of the MCH system, thus reporting bias related to quantitative data may be present because we relied on MCH facilities to complete the questionnaire. Recall bias may have occurred in the interviews because some questions were related to the implementation process of the project, which had happened in the past. Second, given the harsh natural environment,

transportation challenges, local language restrictions, and limited human and financial resources for the project, we did not conduct interviews with women and caregivers receiving MCH services and could not explore the changes in service quality from the perspective of demand side. Finally, all interviews were conducted by two interviewers who were also members of the project team. Although these researchers were skillful interviewers and adhered to the project protocol, their opinions could have influenced interviewees' perceptions about the effects of the HSS interventions.

Conclusion

In conclusion, implementation of comprehensive HSS interventions through multilevel governmental collaboration improves the MCH system in remote and low-income areas. Innovative participatory training methods and appropriate HSS interventions tailored to local contexts contribute to the positive effects of HSS interventions. The participation of local governmental sectors and MCH facilities in project design is conducive to ensuring the successful implementation of the project. However, issues related to workforce shortages and the lack of positive workforce recruitment and retention and incentive policies are constant barriers that must be mitigated to enhance the development of MCH systems in remote and low-income areas.

Author statements

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Ethical approval

This study was approved by Institute of Medical Information and Library Human Research Ethics Committee (HREC) (ref. no. IMICAMS/02/19/HREC). All interviewees provided verbal consent to attend the interviews.

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Competing interests

None declared.

Author contributions

X.L., F.W., and L.W. contributed to study conception and design.

X.L., X.D., and Y.C. collected and analyzed the data.

X.L. drafted the first version of the manuscript.

X.L., F.W., and L.W. revised and reviewed the manuscript.

All authors approved the final manuscript.

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