

Editor: **Simon Langley-Evans**

Journal of **Human Nutrition** and **Dietetics**

VOLUME 33 • ISSUE 2 • APRIL 2020

- Editorial
- Gut Health
- Older People
- Infants and Children
- Professional Practice

Journal of Human Nutrition and Dietetics

The Official Journal of the British Dietetic Association

Editor-in-Chief

Professor Simon Langley-Evans
Deputy Head of School of Biosciences
University of Nottingham,
UK.
E-mail: simon.langley-evans@nottingham.ac.uk

Associate Editors

S Burden, University of Manchester, UK
C Green, Nutricia, The Netherlands

Editorial Board

A. Anderson, *Centre for Public Health Nutrition Research, University of Dundee, UK*
T. Baranowski, *Baylor College of Medicine, USA*
T. Burrows, *University of Newcastle, Australia*
L. Chen, *University of California, USA*
J. Coad, *Massey University, New Zealand*
C. Collins, *University of Newcastle, Australia*
P. Collins, *Griffith University, Australia*
K. Davison, *Simon Fraser University, Canada*
J. Harvey, *University of Vermont, USA*
M. Hickson, *Plymouth University*
M. Kiely, *County College Cork, Ireland*
I. Lemieux, *Quebec Heart Institute, Laval University, Canada*
S. Lennie, *University of Newcastle, UK*
A. Madden, *School of Health and Emergency Professions, University of Hertfordshire, UK*
G. Mailhot, *Université Montréal, Canada*
D. Mellor, *Aston University*
A. O'Sullivan, *Institute of Food and Health, University College Dublin, Ireland*
Y. Probst, *University of Wollongong, Australia*
A. Roefs, *Faculty of Psychology, Maastricht University, The Netherlands*
K. Whelan, *Kings College London, UK*
L. Williams, *Department of Oncology, University of Sheffield, UK*

Aims and editorial policy

Journal of Human Nutrition and Dietetics is an international peer reviewed journal publishing papers in applied nutrition and dietetics. Papers are therefore welcomed on:

- Clinical nutrition and the practice of therapeutic dietetics
- Public health nutrition and nutritional epidemiology
- Health promotion and intervention studies and their effectiveness
- Food choice and the psychology of eating behaviour
- Food intake and nutritional status
- Sociology of food intake

Further information on this journal can be accessed at wileyonlinelibrary.com/journal/jhn
The Publisher, British Dietetic Association and

Editors cannot be held responsible for errors or any consequences arising from the use of information contained in this journal; the views and opinions expressed do not necessarily reflect those of the Publisher, British Dietetic Association and Editors, neither does the publication of advertisements constitute any endorsement by the Publisher, British Dietetic Association and Editors of the products advertised.

Journal of Human Nutrition and Dietetics © 2020 The British Dietetic Association. All rights reserved. No part of this publication may be reproduced, stored or transmitted in any form or by any means without the prior permission in writing from the copyright holder. Authorization to photocopy items for internal and personal use is granted by the copyright holder for libraries and other users registered with their local Reproduction Rights Organisation (RRO), e.g. Copyright Clearance Center (CCC), 222 Rosewood Drive, Danvers, MA 01923,

USA (www.copyright.com), provided the appropriate fee is paid directly to the RRO. This consent does not extend to other kinds of copying such as copying for general distribution, for advertising or promotional purposes, for creating new collective works or for resale. Permissions for such reuse can be obtained using the RightsLink "Request Permissions" link on Wiley Online Library. Special requests should be addressed to: permissions@wiley.com

The *Journal of Human Nutrition and Dietetics* is published by Blackwell Publishing Ltd: 9600 Garsington Road, Oxford OX4 2DQ, UK. Tel: +44 1865 776868; Fax: +44 (0)1865 714591. Blackwell Publishing was acquired by John Wiley & Sons in February 2007. Blackwell's programme has been merged with Wiley's global Scientific, Technical and Medical business to form Wiley Blackwell.

Production Editor: Devvie Rose Miranda (email: jhn@wiley.com)

EDITORIAL

Recommendations for dietetics in mental healthcare

The poor physical health of people with mental illness is increasingly being recognised as a critical area of health inequality across modern-day society⁽¹⁾. This is perhaps best characterised by the severely reduced life expectancy observed among these populations because people with mental health conditions (across a broad spectrum of diagnoses) die 10–30 years younger than the general population, primarily as a result of physical health causes (as opposed to mental health causes)^(2,3).

The 2019 World Congress of Psychiatry saw the official launch of the report from the Lancet Psychiatry Commission on ‘protecting physical health in people with mental illness’⁽¹⁾. The commission incorporated 42 international expert researchers and clinicians from a range of relevant professions, collaborating together to better define this growing problem, as well as to present new opportunities for addressing the health disparities facing people with mental illness when continuing to provide appropriate mental health recovery.

The Commission’s report showed that cardiovascular and metabolic diseases comprise many of the physical comorbidities that apply to a broad spectrum of mental illnesses, taking into consideration that individuals with depression, anxiety disorders, bipolar disorder, psychotic disorders and substance use disorders have a 1.4–2-fold greater risk of cardiometabolic conditions compared to the general population. Furthermore, the Commission identified a broad range of ‘lifestyle factors’ that contribute towards the elevated risk of cardiometabolic diseases, as well as the lifelong burden of poor physical health, as associated with mental illness.

The dietary risks of people living with mental illness were clearly described, with a greater frequency of excessive and unhealthy dietary intakes compared to people without mental illness^(1,4). The reasons for this are often multifactorial and include an increased appetite and disordered eating behaviours, a preference for highly processed convenience and fast foods, low motivation, and low confidence with food preparation⁽⁵⁾. These behaviours appear to be driven by psychotropic medication, particularly antipsychotic medication, positive and negative symptoms of mental illness, and impaired cognition and characteristics associated with the mental illness (e.g. financial and social constraints).

The evolving role for dietitians with respect to improving physical health in people with mental illness has

already been described in a previous issue of this journal⁽⁵⁾; however, although this was generally accepted by the dietetic community, recognition from the psychiatric community was limited. Dietitians were not seen as part of routine care, appearing inconsistently across mental health services. Nonetheless, the inclusion of nutrition and dietetics in the recent Lancet Psychiatry Commission marks the culmination of a paradigm shift where dietary interventions delivered by dietitians are now seen as a core prevention and management strategy for physical health in people living with mental illness.

A recent example of how guidelines have led to change in dietetic support within conventional healthcare services has been described in a previous issue of this journal. Whiteley *et al.* (2019) explored the effect of the introduction of the National Institute for Clinical Excellence (NICE) Guidelines for Epilepsies: Diagnosis and Management on dietetic services, finding a 647% increase in the number of patients being treated with a ketogenic diet for epilepsy, as well as a 77% increase in the number of centres offering a ketogenic diet, in the U.K. and Ireland, within the 7-year period⁽⁶⁾.

We now are optimistic that the introduction of the Commission will provide a strong basis for working towards dietetic guidelines and incorporating these within mental health services; specifically by encouraging health commissioners and authorities to ensure that mental healthcare is equipped with appropriate access to dietetics support and, subsequently, by ensuring that people with mental illness receive adequate physical health care from dietitians, to confront dietary and eating behaviour risks.

It is also important for dietitians to recognise the overarching strategies for the management of physical health in people with mental illness, as well as current recommendations and emerging strategies for dietary interventions. Comprehensive physical health care in people with mental illness encompasses metabolic monitoring, best practice psychotropic prescribing (minimising antipsychotic polypharmacy, as well as prescribing psychotropics with lower metabolic side effects as first line treatments), utilising metformin in a preventative approach, adding additional comorbidity medications as necessary, providing access to nicotine replacement therapy, and providing access to lifestyle intervention early in the course of treatment. To address this in further detail, Part 4 of the Commission comprehensively describes the evidence for

lifestyle interventions in people with mental illness. In particular, the Diabetes Prevention Program can be used as an example of gold-standard lifestyle intervention ⁽⁷⁾, by ensuring the following principles are followed: (i) using behaviour change theory, including specific and measurable behavioural goals, and self-monitoring; (ii) delivery by staff with professional expertise in nutrition and dietetics or exercise; (iii) offering supervised exercise sessions ≥ 2 per week; (iv) familiarising mental health staff with lifestyle intervention; and (v) using peer-support to assist with engagement and adherence.

In summary, the Lancet Psychiatry Commission has shown that the health inequalities experienced by people living with mental illness are driven by a range of factors, with dietary intake being a previously overlooked risk factor. Nonetheless, recognition from the psychiatric community of the importance of dietetics is a key step forward towards evidence-based guidelines and the subsequent implementation of dietetic interventions for mental healthcare.

S. B. Teasdale¹  & J. Firth^{2,3} 

¹School of Psychiatry, UNSW Sydney, Kensington, NSW, Australia; ²NICM Health Research Institute, Western Sydney University, Westmead, NSW, Australia; ³Division of Psychology and Mental Health, Faculty of Biology, Medicine and Health, University of Manchester, Manchester, UK
E-mail: scott.teasdale@outlook.com

References

1. Firth J, Siddiqi N, Koyanagi A *et al.* (2019) The Lancet Psychiatry Commission: a blueprint for protecting physical health in people with mental illness. *Lancet Psychiatry* **6**, 675–712.
2. Walker ER, McGee RE & Druss BG (2015) Mortality in mental disorders and global disease burden implications: a systematic review and meta-analysis. *JAMA Psychiatry* **72**, 334–341.
3. Chesney E, Goodwin GM & Fazel S (2014) Risks of all-cause and suicide mortality in mental disorders: a meta-review. *World Psychiatry* **13**, 153–160.
4. Teasdale SB, Ward PB, Samaras K *et al.* (2019) Dietary intake of people with severe mental illness: systematic review and meta-analysis. *Br J Psychiatry* **214**, 215–259.
5. Teasdale SB, Samaras K, Wade T *et al.* (2017) A review of the nutritional challenges experienced by people living with severe mental illness: a role for dietitians in addressing physical health gaps. *J Hum Nutr Diet* **30**, 545–553.
6. Whiteley VJ, Martin-McGill KJ, Carroll JH *et al.* (2019) Nice to know: impact of NICE guidelines on ketogenic diet services nationwide. *J Hum Nutr Diet*. [epub ahead of print 20 Aug 2019, doi: 10.1111/jhn.12697]
7. Diabetes Prevention Program (DPP) Research Group (2002) The Diabetes Prevention Program (DPP) description of lifestyle intervention. *Diabetes Care* **25**, 2165–2171.

GUT HEALTH

The lived experience of adults with cystic fibrosis: what they would tell their younger selves about the gut

L. Cave^{1,2}  & L. J. Milnes²

¹Children's Nutrition and Dietetics, Leeds Teaching Hospitals NHS Trust, Leeds, UK

²School of Healthcare, University of Leeds, Leeds, UK

Keywords

cystic fibrosis, gut, lived experience, patient perspective, self-management.

Correspondence

L. Cave, Leeds Regional Paediatric Cystic Fibrosis Centre, A Floor Clarendon Wing, Leeds Children's Hospital, Leeds General Infirmary, Leeds, LS1 3EX, UK.

Tel.: +44 (0)113 3922479

Email: l.cave@nhs.net

How to cite this article

Cave L., Milnes L. J. (2020) The lived experience of adults with cystic fibrosis: what they would tell their younger selves about the gut. *J Hum Nutr Diet.* **33**, 151–158
<https://doi.org/10.1111/jhn.12703>

This is an open access article under the terms of the Creative Commons Attribution License, which permits use, distribution and reproduction in any medium, provided the original work is properly cited.

Introduction

Cystic fibrosis (CF) is a multisystem condition that predominantly affects the respiratory and gastrointestinal (GI) systems⁽¹⁾. In recent years, the focus in clinical practice has been on managing complications of the respiratory system⁽²⁾ as respiratory failure secondary to progressive lung disease is the most common cause of death⁽³⁾. However, there are now calls to redress the balance and consider the non-respiratory manifestations of CF, in particular, the GI system (or 'gut')⁽⁴⁾, with relief of

Abstract

Background: Little is known about adults' experience of living with cystic fibrosis (CF) specifically in relation to the gut. However, their unique perspectives may be meaningful to children with CF and inform the understanding and practice of dietitians. The present study aimed to explore adults' lived experience of the CF gut and how they learnt to manage the gut as they were growing up.

Methods: Semi-structured interviews were conducted with adult inpatients ($n = 10$). Interviews were audio-recorded, transcribed verbatim and accounts analysed using interpretative phenomenological analysis.

Results: Three super-ordinate themes were identified: *taking Creon, the learning process* and *this much I (now) know*. Participants accounts of how CF affects the gut predominantly focused on taking Creon (pancreatin, Mylan). Various strategies were employed for coping with peer responses to taking Creon at school. Several participants reached adulthood before they understood and/or accepted that taking Creon consistently needed to be normal for them. Knowledge and understanding developed over time, with 'CF experience' and was shaped by family, CF care teams and other children with CF. All had unmet information needs when growing up. Having key explanations earlier, to make connections between eating, taking Creon, gaining weight and growth, did or would have helped most participants. Participants urged children to be assertive, ask questions and not only be involved in managing their diet and gut, but also begin to take control of this aspect of their CF.

Conclusions: Supporting development of knowledge, skills and confidence to manage diet and the gut needs to be integral to care throughout childhood.

GI symptoms identified as a research priority for CF⁽⁵⁾. This is because effective management of the gut, through control of symptoms and optimising nutritional status, has a major impact on quality of life⁽⁶⁾ and survival⁽⁷⁾.

Previous research has explored adults experience of living with CF^(8–12), although not specifically in relation to the gut. Where this has been carried out in other chronic GI-related conditions, patients' perspectives have helped inform clinicians understanding and practice^(13–15).

The present study therefore aimed to explore adults lived experience of the CF gut, how they made sense of

their experiences and how they learnt to manage the gut as they were growing up. Their unique insights will be shared with children with CF. This is because young people have previously identified information from peers with the same long-term condition to be particularly relevant to them and their experiences⁽¹⁶⁾. Through this specialist type of peer support, children may feel connected and better supported to make choices around self-care⁽¹⁷⁾. This is important as children begin to gain independence, for example, in making food choices and adjustments to their accompanying pancreatic enzyme replacement therapy (PERT). The study findings will also inform dietitians understanding of patient needs and how patient-centred care can be enhanced⁽¹⁸⁾ with children with CF and their families.

Materials and methods

Design

This qualitative study adopted the approach of interpretative phenomenological analysis (IPA) because this was appropriate for gathering in-depth accounts of individuals lived experiences and exploring the meanings they attribute to these experiences⁽¹⁹⁾.

The study design, method and setting for data collection were chosen after consultation with four patients attending a single regional adult CF centre; in addition, one patient reviewed and provided valuable feedback on all study documentation (all nonparticipants).

Ethical approval was granted by South West-Exeter Research Ethics Committee, reference: 17/SW/0042.

Sampling and recruitment

Purposive sampling was conducted at a single regional adult CF centre in the UK. A sample size of 8–10 potential participants was aimed for because IPA studies are concerned with a concentrated focus on a small number of individuals, typically less than 10⁽¹⁹⁾.

The lead dietitian in the direct clinical care team identified eligible participants from electronic medical records based on the criteria in Table 1.

The lead dietitian approached and invited potential participants to take part in the study when they were part way through an admission and were clinically stable. All were provided with written study information and the opportunity to ask questions. Written informed consent was obtained by the first investigator immediately prior to participation.

Data collection

Ten potential participants were approached and all agreed to participate (five females and five males; aged 19–30

Table 1 Eligibility criteria for potential participants

Eligibility	Rationale
Inclusion	
Adults diagnosed with cystic fibrosis (CF) in early childhood	Adults asked to reflect on their experience of growing up with CF
Aged 20–30 years	Upper age limit of 30 years so they all had experience of the same 'high fat, high calorie CF diet' and used the same preparation of PERT that is currently available to children; lower age limit of 20 years so they were out of their teens
Pancreatic insufficient (PI), so required pancreatic enzyme replacement therapy (PERT)	To be able to explore their experience of managing PERT, how this changed over time and how they developed their knowledge and understanding (Approximately 85%–90% of people with CF are PI. In clinical practice, the majority of children find it challenging to manage PERT, particularly as they move into adolescence)
Admission for routine intravenous antibiotic therapy (IVAT) between May and July 2017	This was the preferred setting chosen by patients over the 3-month period for data collection
Exclusion	
Approximately continuous IVAT over the previous year	Severely affected by CF and possibly too unwell to participate
On the active transplant list	
Pregnant	Already committing extra time for close monitoring

years) (Table 2). Single one-to-one semi-structured interviews were conducted by the first investigator throughout May and June 2017. Interviews were up to 1 h in duration and conducted in patients own rooms on the ward, with only the participant and the researcher present. A topic guide (Table 3) was used flexibly. Interviews were audio-recorded, transcribed verbatim and transcripts annotated with field notes made after each interview. The first two interviews enabled pilot testing of the topic guide; these two interviews were included in the final analysis, as no changes were required. Where possible, data were collected and analysed concurrently.

Data analysis

Anonymised transcripts were analysed systematically using IPA, following the process described by Smith *et al.*⁽¹⁹⁾ In summary, a transcript was read and re-read and exploratory comments added to describe content, use of

Table 2 Characteristics of participants ($n = 10$)

Pseudonym	Gender	Age (years)	Age at CF diagnosis
Anne	F	30	3 months
Christine	F	21	4 months
David	M	28	1 month
Emma	F	19	1 month
Jane	F	27	At birth
John	M	24	13 months
Mark	M	27	At birth
Philip	M	27	2 years
Steven	M	24	3 weeks
Susan	F	28	18 months

CF, Cystic fibrosis; F, female; M, male.

language and concepts. Connections and patterns between exploratory comments were mapped to identify emergent themes and these clustered to form themes. This process was then repeated for each transcript and patterns identified across the sample as a whole. Themes derived from the data (LC) were independently checked (LM) and a high degree of agreement taken as validation of identified themes.

Results

Three themes were inferred: taking Creon (pancreatin, Mylan), the learning process and 'this much I (now)

Table 3 Interview topic guide

Topics for discussion	
	Experience of CF and gut involvement for them
	Experience as a child of eating, taking Creon, managing their gut (prompts: experience at school, with friends, on trips; coping strategies)
	How experiences have changed over time (insights of their journey) (prompts: attitudes, knowledge, understanding, behaviours, motivation)
	Facilitators
	Something or someone that particularly helped them make sense of how CF affects their gut
	(any lightbulb moment/specific event that made things click in terms of their understanding)
	(prompts: any appropriate, relevant support and guidance received – from HCPs/family/friends (with or without CF))
	Barriers
	Anything they wish they were told as a child – if so, when, how and by whom
	Insights/explanations they feel would be important to share with a child with CF ('top tips' to pass on)
	Anything else they would like to add

CF, cystic fibrosis; HCP, healthcare professional.

know'. Within these themes, several subthemes emerged, each supported by exemplar quotations from participants. These are presented in the narrative below and in Table 4.

Taking Creon

Participants' accounts of how CF affects the gut, predominantly focussed on taking Creon (the most widely available brand of PERT in the UK). A wide range of views were expressed about the continual need for Creon and the tension this could create.

Coping with peer responses

Taking Creon was described as the most visible part of having CF when at school. Peer responses were on a spectrum, from bullying and accusations of being a drug addict, through to friends giving reminders to take it. Peers at primary school were generally accepting but, at secondary school, they more readily recognised difference and questioned it. Various coping strategies were adopted, including not taking Creon at all, taking it out of sight of others, not eating or choosing foods that required less or no Creon. Although some participants felt embarrassed about getting their Creon 'out', others cautioned against hiding it and advised either ignoring what peers say, or giving a simple explanation to close down further questioning.

'If anyone asked, I explained why it was needed. I'd just tell them they are enzymes and you make them in your body but I don't, so I just need to have them when I have some food so my body can break down the food and absorb all the nutrients out of it' (Mark)

This coping strategy relied on them having an explanation they understood and felt confident to share, though as adults this strategy was reported as the most widely adopted.

'This is our normal'

All participants reported that as they grew older and the course of CF progressed, they realised how important Creon was and accepted that taking it was a normal part of life for them. For several, this realisation came as a result of severe weight loss and they described almost giving up fighting against it.

'Cos obviously I wasn't taking them (Creon) my food wasn't digesting or anything, so I was just losing weight, losing weight. I ended up, it got that bad I had to have a feeding tube put in and that's when I realised, it'd be better just to take them' (Philip)

For some, it was only when they started taking Creon that they realised what it did because symptoms of

Table 4 Themes and subthemes supported by exemplar participant quotations

Theme and subthemes	Participant quotations
Taking Creon	
Coping with peer responses	<ul style="list-style-type: none"> • [At primary school] 'It was just realising you're a bit different from all the other kids. In secondary school, obviously that's when they did ask more questions, they understood then, 'well we don't have to take medicines when we eat, so why do you?' (Christine) • [At secondary school] 'A couple of people clocked me taking my tablets (Creon) and I started getting bullied. So, I kind of stopped taking them and that upset my stomach a lot, but I didn't care I just sort of suffered it out through the daytimes at school' (Susan)
'This is our normal'	<ul style="list-style-type: none"> • 'It was just a normal part of my life to take Creon. From quite an early age I was trusted to do it' (Jane) • 'Everyone's different. It's finding out what you need, what works best for you and just always remembering to have it' (Steven)
The learning process	
Family	<ul style="list-style-type: none"> • 'In my family... I didn't grow up with routine... whenever I went in [for a hospital admission] it was brilliant 'cos I got up to date with all my meds, I got used to everything' (David) • 'The dietitians were telling my mum what foods are high in fat and then I learnt from her... I relied on my mum a lot to know how many (Creon) I needed to have' (Christine)
CF team	<ul style="list-style-type: none"> • 'If you've got questions – just ask – don't sit there quietly thinking you don't want to ask because you don't want to get into trouble for not knowing the answer; that's what I did, I always wondered about stuff and then I was never asked' (Anne) • 'I moved to X (a specialist CF centre) and it was completely different to what I was used to. I felt more involved, definitely. Rather than them telling me stuff, they were asking me stuff and including me with decisions' (Steven)
Other children with CF	<ul style="list-style-type: none"> • [Children with CF now segregated] 'They'll have no-one really that they can relate to... they may want some reassurance that they're not the only one going through it and ask questions on how you deal with things' (Susan) • 'Cross-infection wasn't in then... (during admissions) there was a lot of CFs and diabetics together – we had take-away nights and could talk and ask each other questions' (Anne)
'This much I (now) know'	
Knowing things earlier	<ul style="list-style-type: none"> • 'I had it explained and explained to me (why I needed to take Creon) but I never listened. I did know about it, I just pretended not to, just silly really' (Philip)
Making connections	<ul style="list-style-type: none"> • 'I was always told I need to keep gaining weight to make me grow, and 'cos I wanted to be tall, that always made me motivated to eat more... and it was always said eating gives you energy and you'll be able to do more and 'cos I love doing sport, it was always important for me to eat' (Emma) • 'If I knew taking Creon contributed to growth, I'd probably have taken it more, as I would have liked to be taller... but not knowing that, I just thought it (Creon) was something and nothing, so I just shrugged them off' (John)
Thinking longer-term	<ul style="list-style-type: none"> • 'Explain what's happening - for the long-term benefits it'll bring – that it's keeping them well' (Mark) • [To a child with CF] 'Get involved – be in control - do what you need to do and you've got a bright future ahead of you' (David)

CF, cystic fibrosis.

malabsorption were normal for them. Many reported that it was a process of trial and error to find the doses and timing of Creon that suited their individual bodies. Without exception, all participants stressed that taking Creon consistently is essential and offered encouragement to children to build this in as normal.

The learning process

All participants shared the view that they had developed their knowledge and understanding as a gradual accumulation of learning over time, with 'CF experience'.

Family

Family support ranged from taking complete control through to participants having to deal with CF on their own from an early age. Where parents took control and

did everything for them, this appeared to hinder development of their own understanding. Several participants described intermittent or erratic family involvement and felt they did better when they were an inpatient. A lack of explanations from their families left them unaware of why treatments were needed and this affected adherence, particularly to Creon. Others reported having supportive families, where it appeared that they were dealing with it together and information was shared about how to manage diet and Creon.

'The way my family approached it... I never felt it was an illness when I was younger, it was always just "ah this is what I need to do", just everyday' (Emma)

All participants expressed how the type of family support they received shaped their understanding and confidence. Participants urged parents of children with CF to

provide time, space and encouragement to develop independence, whilst also providing a safety net.

Cystic fibrosis team

Most participants did not feel included at clinic visits when they were a child and urged CF teams to do more to directly engage and involve children. Several described only the adults speaking and they were left feeling 'why am I here?'

'Children's clinic was always waiting round a lot. They'd obviously do whatever checks they needed to and then it would just be them talking to my mum. I was just always waiting to leave instead of them talking to me' (John)

There were exceptions, particularly if they attended a specialist paediatric CF centre, with some reporting their CF team an asset in informing their learning. Participants acknowledged the need for children to be assertive, to actively seek support and ask questions. Some expressed regret that they had not done so themselves and urged children to be braver than they had been.

Other children with cystic fibrosis

Participants described how other children with CF were key contributors to their learning. All acknowledged that there was no strict segregation (to reduce the risk of cross-infection) when they were children and several expressed concern that children now will be even more isolated than they were.

This much I (now) know

All participants reflected that as children growing up with CF, they had some unmet information needs. This varied from needing information on everything, through to wanting explanations on why specific vitamins were needed.

Knowing things earlier

Most participants felt it would have made a difference to have had things explained earlier, in terms they could understand, with things revisited at regular intervals and in greater depth as they grew older. They described how this would have given them more confidence in what was happening.

'I never got it explained so I grew up thinking "well, I don't actually know why I'm doing this, so I might as well not do it" ... I wish I was told everything, appropriate for the age you are, in language you can understand and then as you grow up, just that little bit more information, just gradually' (Susan)

However, some acknowledged they were not or would not have been ready to take on the information. Only a few felt they got the explanations they needed at an appropriate level and age; though as children, these participants were confident to ask questions and felt supported within their family units.

Making connections

Most participants described themselves as scrawny or skinny children, some small in stature, who on the whole, had large appetites and good lung function. Some reported having the information they needed to make necessary connections between eating, taking Creon, gaining weight and growing. Several highlighted how it had been helpful to link eating well with having energy to be active (e.g. to play out with friends and play sport).

'Food plus Creon equals weight ... means less admissions and more time with your friends' (David)

Others felt that having this information would have helped them to make sense of things and given them an incentive to eat more and improve their Creon adherence. Several participants had not made the connection between weight and lung function until they were adults. Some felt it would have been helpful to know this earlier, whereas others felt it would have put even more pressure on eating and gaining weight and so were glad to have not known sooner.

Thinking longer-term

Although some participants described embracing a high-energy, high-fat 'CF diet' and having no wish to change this, several described how their diets had changed over time to become more balanced, as a result of thinking more about their long-term health, not just their CF.

'I always remember dietitians saying about calories and needing to eat more fat ... "cos people didn't live as long it didn't matter ... but now it should be about how important nutrition is generally, for your health as a whole' (Jane)

These participants expressed how children with CF need to be motivated to begin looking after their diet and gut in ways that relate positively to the future and goals for their adulthood.

Discussion

Through in-depth interviews, adults talked at length about their lived experiences of CF and what had shaped their knowledge and understanding of the CF gut. They were enthusiastic about sharing their insights with children with CF and optimistic about children's futures.

They suggested how families and CF teams could promote children's learning and expressed concern at the current lack of peer support.

As reported previously^(11,20,21), various coping strategies were adopted for taking Creon, (particularly at secondary school), to reduce feelings of difference from their non-CF peers. However, participants advised children with CF to not hide their Creon but, instead, to offer a simple explanation to peers as to why it is needed. This has implications for practice as children clearly need support in preparing for transition to secondary school not only in increasing their independent Creon taking, but also in being equipped with the knowledge, skills and confidence to relay explanations to peers.

Knowledge and understanding developed gradually as an accumulation of learning over time, as described previously⁽⁸⁾. Families, the CF team and peers with CF played important roles in informing participants learning. The role of families in passing on information about diet and Creon was essential to their understanding and confidence and impacted on their Creon adherence. The link between family functioning and treatment adherence in CF has been identified previously^(22–25), with families that find a balance between cohesion and flexibility reporting higher rates of adherence⁽²²⁾. In the present study, participants in supportive family units were, as children, confident to ask questions and seek the explanations they needed from the CF team; however, all participants urged children to be assertive, seek support and be actively involved in their dietary care.

Advice to the CF team strongly resonated with that highlighted previously^(26,27), with a call for a more child-centred approach to dietary consultations, where children can actively participate rather than feel marginalised, and discussions use terms relevant and meaningful to children's daily lives.

Interaction with other children with CF made a significant contribution to participants learning, supported their interactions with the CF team and reduced feelings of difference and isolation. This was congruent with findings from other studies^(21,28,29). With strict segregation now in force, virtual peer interaction may play an important role^(30,31), although creating online platforms suitable for primary school age children remains a challenge.

As reported previously⁽³²⁾, all participants had some unmet information needs when growing up. As a consequence, several described a decline in adherence during adolescence. This may impact on health outcomes such as growth and lung function⁽³³⁾ and contribute to reduced survival in adulthood. Information needs must therefore be addressed throughout childhood as part of developing a solid grounding in self-management skills to take forward into adolescence^(34,35).

Study strengths and limitations

All participants attended the same single regional adult CF centre. However, they shared experiences of attending different regional and shared-care paediatric CF centres across the UK and were therefore exposed to different service provision when growing up with CF.

The present study focussed only on adults who were pancreatic insufficient. The experiences of adults who are pancreatic sufficient warrant full consideration in their own right and could be the focus of a future study.

Participants may have responded differently if interviewed in a different setting. Although they appeared relaxed in their own rooms on the ward, the study captured how they made sense of their experiences at one point in time, in the context of a single one-off interview. The data generated were a co-production between participant and researcher⁽³⁶⁾ and the findings are an interpretation of participants' accounts by LC and checked by LM. There was insufficient time for participants to feedback on the derivation and identification of themes; however, a journal was kept throughout by LC to reflect on how prior knowledge and experience as a children's CF dietitian may impact on the data.

Conclusions

Supporting children with CF to develop their knowledge, skills and confidence to manage diet and the gut needs to be integral to care throughout childhood. Targeting this support in preadolescence may equip children to effectively manage the demands of this aspect of CF care during adolescence. The findings of the present study inform a current programme of work identifying how this support can be delivered as part of routine dietetic practice and how families, CF teams and peer support may enhance this.

ACKNOWLEDGMENTS

We thank all of the participants for sharing their experiences; the four patients who were consulted about their preferred study design and method and setting for data collection; the patient who reviewed and fed back on study documentation; the lead dietitian for recruiting participants; and Dr Helen White for her support and guidance.

Conflicts of interests, source of funding and authorship

The authors declare that they have no conflicts of interest.

Laurie Cave was funded by a National Institute for Health Research (NIHR) Masters in Clinical Research Studentship for this research project. This publication presents independent research. The views expressed are those of the authors and not necessarily those of the NHS, the NIHR or the Department of Health and Social Care.

LC and LM were responsible for the study design. LC performed the data collection. LC and LM performed the data analysis. LC and LM prepared the draft of the manuscript. LM supervised the study. All authors critically reviewed the manuscript and approved the final version submitted for publication.

Transparency declaration

The lead author affirms that this manuscript is an honest, accurate and transparent account of the study being reported. The reporting of this work is compliant with COREQ guidelines⁽³⁷⁾. The lead author affirms that no important aspects of the study have been omitted and that any discrepancies from the study as planned have been explained. The dataset and study documentation are available at: <https://doi.org/10.5518/609>.

References

- Bishay LC & Sawicki GS (2016) Strategies to optimize treatment adherence in adolescent patients with cystic fibrosis. *Adolesc Health Med Ther* **7**, 117–124.
- Freedman SD & Schwarzenberg SJ (2016) Beyond the lungs: the new GI frontier. *J Cyst Fibros* **15**, 409–410.
- Elborn SJ (2016) Cystic fibrosis. *Lancet* **388**, 2519–2531.
- Cystic Fibrosis Trust (2016). News of the Trust's latest research. Available at: <https://www.cysticfibrosis.org.uk/news/chewing-the-fat> (accessed June 2017).
- Rowbotham NJ, Smith S, Leighton PA *et al.* (2018) The top 10 research priorities in cystic fibrosis developed by a partnership between people with CF and healthcare providers. *Thorax* **73**, 388–390.
- Shoff SM, Tluczek A, Laxova A *et al.* (2013) Nutritional status is associated with health-related quality of life in children with cystic fibrosis aged 9–19 years. *J Cyst Fibros* **12**, 746–753.
- Yen EH, Quinton H & Borowitz D (2013) Better nutritional status in early childhood is associated with improved clinical outcomes and survival in patients with cystic fibrosis. *J Pediatr* **162**, 530–535.
- Huyard C (2008) Exploring one's own human condition: adults affected by cystic fibrosis. *Qual Health Res* **20**, 535–544.
- Badlan K (2006) Young people living with cystic fibrosis: an insight into their subjective experience. *Health Soc Care Community* **14**, 264–270.
- Gjengedal E, Rustoen T, Wahl AK *et al.* (2003) Growing up and living with cystic fibrosis: everyday life and encounters with the health care and social services – a qualitative study. *Adv Nurs Sci* **26**, 149–159.
- Lowton K & Gabe J (2003) Life on a slippery slope: perceptions of health in adults with cystic fibrosis. *Sociol Health Illn* **25**, 289–319.
- Tracy JP (1997) Growing up with chronic illness: the experience of growing up with cystic fibrosis. *Holist Nurs Pract* **12**, 27–35.
- Sammut J, Scerri J & Xuereb RB (2015) The lived experience of adults with ulcerative colitis. *J Clin Nurs* **24**, 2659–2667.
- McCormick JB, Hammer RR, Farrell RM *et al.* (2012) Experiences of patients with chronic gastrointestinal conditions: in their own words. *Health Qual Life Outcomes* **10**, 25.
- Fletcher PC, Schneider MA, Van Ravenswaay V *et al.* (2008) I am doing the best that I can!: Living with inflammatory bowel disease and/or irritable bowel syndrome (part II). *Clin Nurse Spec* **22**, 278–285.
- Milnes LJ, McGowan L, Campbell M *et al.* (2013) A qualitative evaluation of a pre-consultation guide intended to promote the participation of young people in asthma review consultations. *Patient Educ Couns* **91**, 91–96.
- National Endowment for Science, Technology and the Arts (NESTA). (2013). People helping people: peer support that changes lives. Available at: <https://www.nesta.org.uk/report/people-helping-people-peer-support-that-changes-lives/> (accessed July 2019).
- Sladdin I, Chaboyer W & Ball L (2018) Patients' perceptions and experiences of patient-centred care in dietetic consultations. *J Hum Nutr Diet* **31**, 188–196.
- Smith JA, Flowers P & Larkin M (2009) *Interpretative Phenomenological Analysis: theory, Method and Research*. London: SAGE.
- Jessup M & Parkinson C (2010) 'All at sea': the experience of living with cystic fibrosis. *Qual Health Res* **20**, 352–364.
- Christian BJ & D'Auria JP (1997) The child's eye: memories of growing up with cystic fibrosis. *J Pediatr Nurs* **12**, 3–12.
- White T, Miller J, Smith GL *et al.* (2009) Adherence and psychopathology in children and adolescents with cystic fibrosis. *Eur Child Adolesc Psychiatry* **18**, 96–104.
- Everhart RS, Fiese BH, Smyth JM *et al.* (2008) Family functioning and treatment adherence in children and adolescents with cystic fibrosis. *Pediatr Allergy Immunol Pulmonol* **27**, 82–86.
- Berge JM & Patterson JM (2004) Cystic fibrosis and the family: a review and critique of the literature. *Fam Syst Health* **22**, 74–100.
- DeLambo KE, Levers-Landis CE, Drotar D *et al.* (2004) Association of observed family relationship quality and problem-solving skills with treatment adherence in older children and adolescents with cystic fibrosis. *J Pediatr Psychol* **29**, 343–353.

26. Savage E & Callery P (2007) Clinic consultations with children and parents on the dietary management of cystic fibrosis. *Soc Sci Med* **64**, 363–374.
27. Savage E & Callery P (2005) Weight and energy: parents' and children's perspectives on managing cystic fibrosis diet. *Arch Dis Child* **90**, 249–252.
28. Tierney S, Deaton C, Jones A *et al.* (2013) Liminality and transfer to adult services: a qualitative investigation involving young people with cystic fibrosis. *Int J Nurs Stud* **50**, 738–746.
29. Berge JM, Patterson JM, Goetz D *et al.* (2007) Gender differences in young adults' perceptions of living with cystic fibrosis during the transition to adulthood: a qualitative investigation. *Fam Syst Health* **25**, 190–203.
30. Moola F (2018) The complexities of contagion: the experience of social isolation among children and youth living with cystic fibrosis in Canada. *J Child Health Care* **22**, 4.
31. Kirk S & Milnes L (2016) An exploration of how young people and parents use online support in the context of living with cystic fibrosis. *Health Expect* **19**, 309–321.
32. Sawicki GS, Sellers DE, McGuffie K *et al.* (2007) Adults with cystic fibrosis report important and unmet needs for disease information. *J Cyst Fibros* **6**, 411–416.
33. Eakin MN, Bilderback A, Boyle MP *et al.* (2011) Longitudinal association between medication adherence and lung health in people with cystic fibrosis. *J Cyst Fibros* **10**, 258–264.
34. Christian BJ & D'Auria JP (2006) Building life skills for children with cystic fibrosis: effectiveness of an intervention. *Nurs Res* **55**, 300–307.
35. Downs JA, Roberts CM, Blackmore AM *et al.* (2006) Benefits of an education programme on the self-management of aerosol and airway clearance treatments for children with cystic fibrosis. *Chron Respir Dis* **3**, 19–27.
36. Mason J (2002) *Qualitative Researching*, 2nd edn. London: SAGE.
37. Tong A, Sainsbury P & Craig J (2007) Consolidated criteria for reporting qualitative research (COREQ): a 32-item checklist for interviews and focus groups. *Int J Qual Health Care* **19**, 349–357.

GUT HEALTH

Inducing remission in paediatric Crohn's disease using nutritional therapies – A systematic review

L. McVeigh¹ & A. Payne²

¹Nutrition and Dietetics Department, Bristol Royal Hospital for Children, Bristol, UK

²Faculty of Health & Human Sciences, School of Health Professions, The University of Plymouth, Plymouth, UK

Keywords

Crohn's disease, remission, children, paediatric, nutrition, diet.

Correspondence

L. McVeigh, Nutrition and Dietetics Department, Bristol Royal Hospital for Children, Bristol BS2 8BJ, UK.

Tel.: +44 0117 34 28802

E-mail: lauren.mcveigh@nhs.net

How to cite this article

McVeigh L., Payne A. (2020) Inducing remission in paediatric Crohn's disease using nutritional therapies – A systematic review. *J Hum Nutr Diet.* **33**, 170–186
<https://doi.org/10.1111/jhn.12714>

Abstract

Background: Exclusive enteral nutrition (EEN) is known to be a safe and effective treatment option for managing active Crohn's disease (CD) in children, although no uniform protocol exists. The aim of this systematic review was to evaluate and compare the clinical effectiveness of aspects of EEN protocols to ascertain whether an optimum regimen can be identified.

Methods: A systematic search of the Cochrane Library, PubMed, MEDLINE, EMBASE, CINAHL and AMED was conducted for studies published between 1998 and 2018 that examined paediatric patients being treated with an enteral nutrition protocol to induce remission. Studies that included patients receiving concurrent medication for active disease were excluded. Quality assessment was performed using separate tools for randomised controlled trials, cohort studies and for studies without a control group.

Results: Sixteen studies met the inclusion criteria. Of these, six found insufficient evidence to support use of a specific formula. One study examined the route of EEN, finding no difference between oral or nasogastric tube administration with respect to inducing remission. Three examined the use of partial enteral nutrition to induce remission, although conflicting results were seen. No studies explored the effect of length of treatment or energy prescription on remission rates

Conclusions: An optimum enteral nutrition protocol for inducing remission cannot be identified. Further focused research is required by well designed, adequately powered prospective clinical trials to examine aspects of enteral feeding protocols that remain uncertain, including the use of partial enteral nutrition as a potential alternative to EEN.

Introduction

Crohn's disease (CD) is a chronic, inflammatory bowel disease (IBD) that mostly affects the gastrointestinal tract and is characterised by periods of disease exacerbation or remission. CD can affect any part of the gastrointestinal tract, although it typically involves the distal ileum or colon. Affected areas are normally interspersed with sections of relatively normal tissue, although inflammation can extend through the epidermis, leading to deep ulceration and fissuring of the mucosa. With the disease focus being on the gastrointestinal tract, unsurprisingly, nutrition has been shown

to be involved in several aspects of CD, from the aetiology of disease, to inducing remission and disease maintenance ⁽¹⁾. Disease symptoms vary between individuals and will often be dependent on the site of disease activity, although they can include abdominal pain, diarrhoea, weight loss and rectal bleeding. CD can also be complicated by the development of intestinal obstruction, fistulae or perianal disease, stricture and, occasionally, perforation of the gastrointestinal tract ⁽²⁾. Because CD is an incurable autoimmune condition, the aim of treatment is primarily directed at inducing remission, preventing relapse, controlling symptoms and maintaining quality of life ⁽³⁾.

Epidemiological studies have demonstrated a significant increase in cases of CD in the past 30 years⁽⁴⁾ and it is considered that up to one-third of patients are diagnosed before the age of 21 years. Treatment aims are ultimately the same in both adult and paediatric patients; however, in paediatric cases, a unique aspect requiring further management is the debilitating affect that poorly controlled CD can have on linear growth and pubertal development⁽⁵⁾, as well as impairment on the bone formation process leading to suboptimal bone growth^(6,7). As a result of this and the high rate of malnutrition noted at diagnosis⁽⁸⁾ nutritional intervention is an essential component of any management programme. However, controversy exists surrounding the optimal treatment to induce remission in active disease and the role that nutritional therapies might play.

Corticosteroids have a long-standing use in the treatment of CD and there is universal agreement that they can be highly effective to control active disease⁽⁹⁾, although their use is associated with numerous adverse effects, particularly in children. Acutely, this includes the development of Cushing's syndrome, although the greatest concerns relate to its long-term usage and associations with growth impairment and osteoporosis. Therefore, the use of nutritional therapies that do not have such associated problems can provide a uniquely different and beneficial approach.

Utilising nutrition as a primary therapy in CD was first described in the early 1970s and involved providing complete bowel rest alongside parenteral nutrition (PN) to ensure that nutritional status was maintained. Evidence demonstrates good rates of remission in adults⁽¹⁰⁾, although no studies have examined its use in children. Today, PN continues to have some use, although it is typically reserved for patients with severe malnutrition or in an acute inflammatory phase preoperatively. For the majority of patients, a safer and more practical option of exclusive enteral nutrition (EEN) is used, which is now a well established therapeutic approach involving delivery of a liquid nutritional formula enterally, at the same time as excluding all solid food and other forms of nutrition. In adults, several systematic reviews have suggested that there is evidence, albeit weak, that EEN is less effective than conventional treatment with corticosteroids^(11,12). However, the results from individual studies in children are more supportive of EEN^(13–16). For example, the results from the GROWTH-CD study demonstrated that EEN was more effective at inducing remission than corticosteroids and also offered greater benefits in terms of improved growth⁽¹⁵⁾. These results have been further strengthened by several systematic reviews^(17–20), including a very recent meta-analysis of 18 paediatric studies⁽¹⁹⁾ showing no added benefit of corticosteroids over

EEN, suggesting that EEN is in fact superior in improving short-term mucosal inflammation. Consequently, in children who have not finished growth, current European guidelines recommend that EEN should be the first-line treatment in the induction of remission⁽²¹⁾. However, although EEN is an effective treatment for CD, it is not yet clear why or how it works and there appears to be variable uptake of it as a treatment, with corticosteroids commonly being used as first-line therapy in children. This may possibly be a result of wide variation in the protocols used⁽²²⁾, which is leading to inconsistent remission rates achieved in research and in clinical practice. If an optimum protocol could be identified and applied in practice, a better outcome with consistency of positive results could be achieved, with reduced use of steroid medication, along with its associated complications.

The aim of this systematic review is to evaluate and compare the clinical effectiveness of enteral nutrition regimens and protocols used to treat active paediatric Crohn's disease, measured by whether clinical remission is reached. More specifically, this included comparing and evaluating the clinical effectiveness of elemental, semi-elemental, polymeric and specific-nutrient polymeric formulas; different durations of EEN regimens; and different calorific prescriptions of EEN or partial enteral regimens.

Materials and methods

Protocol

The protocol for this systematic review was registered with PROSPERO (registration number CRD42018100463) describing the question and proposed methodology.

Search strategy

The systematic review followed the PRISMA-P guidelines⁽²³⁾ with PubMed, MEDLINE, EMBASE, CINAHL, AMED and the Cochrane Database of Systematic Reviews being searched using the medical subject headings (MeSH) and keywords relating to diet and Crohn's disease (Table 1). Although the population group of interest was paediatric patients, this term was not applied in the search to ensure that potentially relevant literature was not missed. It had been noted in an initial search that authors do not always specify the population group in the title or abstract. Boolean operators were used to help connect and define the search terms, whereas truncation symbols were used to broaden the search, looking for word variations. Search results were limited to full-text papers in English published between January 1998 and June 2018 when the final search was

Table 1 PICO criteria and search terminology used

PICO heading	Keywords/ variations	Example search strategy applied: MEDLINE (Ovid) Searched 14 February 2018
Population	Paediatric Pediatric Child or Children Adolescents	Not applied
Population	Crohn's disease	1 Crohn Disease/
Intervention	Diet	2 Inflammatory Bowel diseases/
	Liquid diet	3 Crohn*.tw
	therapy	4 Inflammatory bowel disease*.tw
	Enteral nutrition	5 IBD 6 1 or 2 or 3 or 4 or 5 7 Enteral and Nutrition\$ or Diet\$ or feed\$ or formula\$.tw 8 Enteral nutrition/ 9 Diet/ 10 7 or 8 or 9 11 6 and 10 12 Limit 11 to English language
Control	Passive – placebo or no control Active – EN treatment variation, alternative medication	Not applied
Outcome	Clinical remission Anthropometric measurements	Not applied

EN, enteral nutrition; IBD, inflammatory bowel disease; PICO: P – Patient, problem or population; I – Intervention; C – Comparison, control or comparator; O – Outcome(s) (eg. pain, fatigue, nausea, infections, death).

completed. The electronic database search was supplemented by searching for grey and unpublished literature through Mednar, Open Grey, Google Scholar and the WHO international clinical trials registry platform. Eligible papers were cross-checked for references and forward citation searching was undertaken. The search was rerun in June 2019 prior to publication with no further relevant records identified at this time.

Inclusion and exclusion criteria

The criteria used to assess eligibility for inclusion were: (i) any experimental, Quasi-experimental study or observational study; (ii) any child under 18 years old with active Crohn's disease at the time of study baseline; (iii) studies examining exclusive enteral nutrition protocols or any other enteral nutrition protocol being used for induction of remission; and (iv) studies that had accessible

full-text articles written in English. The primary outcome measure was rate of remission achieved. Because, at present, there is no universally agreed method or standard for assessing disease activity in paediatric patients, remission was regarded as that defined by the study authors, on an intention-to-treat basis. However, the method of assessing remission was examined as part of the assessment into the overall quality of the study, with nonvalidated tools being considered as poorer quality. Studies including patients with indeterminate colitis or pre-existing complications of disease were excluded, as were patients receiving diet therapy in conjunction with either parenteral nutrition or another medication to treat active disease. Studies that only compared EEN with corticosteroids were not included because they did not meet the remit of this current systematic review. Duplicate studies, case reports/studies or review articles and studies not meeting the above inclusion criteria were also excluded.

Study selection

Data were managed using 'EndNote' citation manager (Clarivate Analytics, Philadelphia, PA, USA) with all of the results being imported into the manager and duplicates removed. Titles were examined initially for relevancy then the remaining abstracts screened on the basis of the above eligibility criteria. If there was uncertainty regarding appropriateness for inclusion from the title and abstracts, then the full text was obtained. Sixteen studies remained after this process, which are included in the current review. Figure 1 displays the search strategy. One independent reviewer completed this process.

Data extraction and analysis

Articles identified for inclusion were read and relevant data extracted for synthesis, including: (i) study details (Author, study design, sample size, follow-up duration); (ii) participant details [age, inclusion/exclusion criteria, Paediatric Crohn's disease assessment index (PCDAI) or clinical/endoscopic definitions of CD activity, new or relapsed disease, disease location]; (iii) intervention details (type of formula or diet intervention, mode, duration, dosage; intervention comparator); and (iv) outcome characteristics (type of measurement/definition of remission).

To evaluate study quality, because there is no single obvious candidate tool for assessing quality of all study types, quality assessment was performed using different tools depending on study type: (i) the Cochrane Risk of Bias Tool was used for randomised controlled trials; (ii) the Newcastle-Ottawa Scale was used for cohort studies; and (iii) the Institute of Health Economics quality

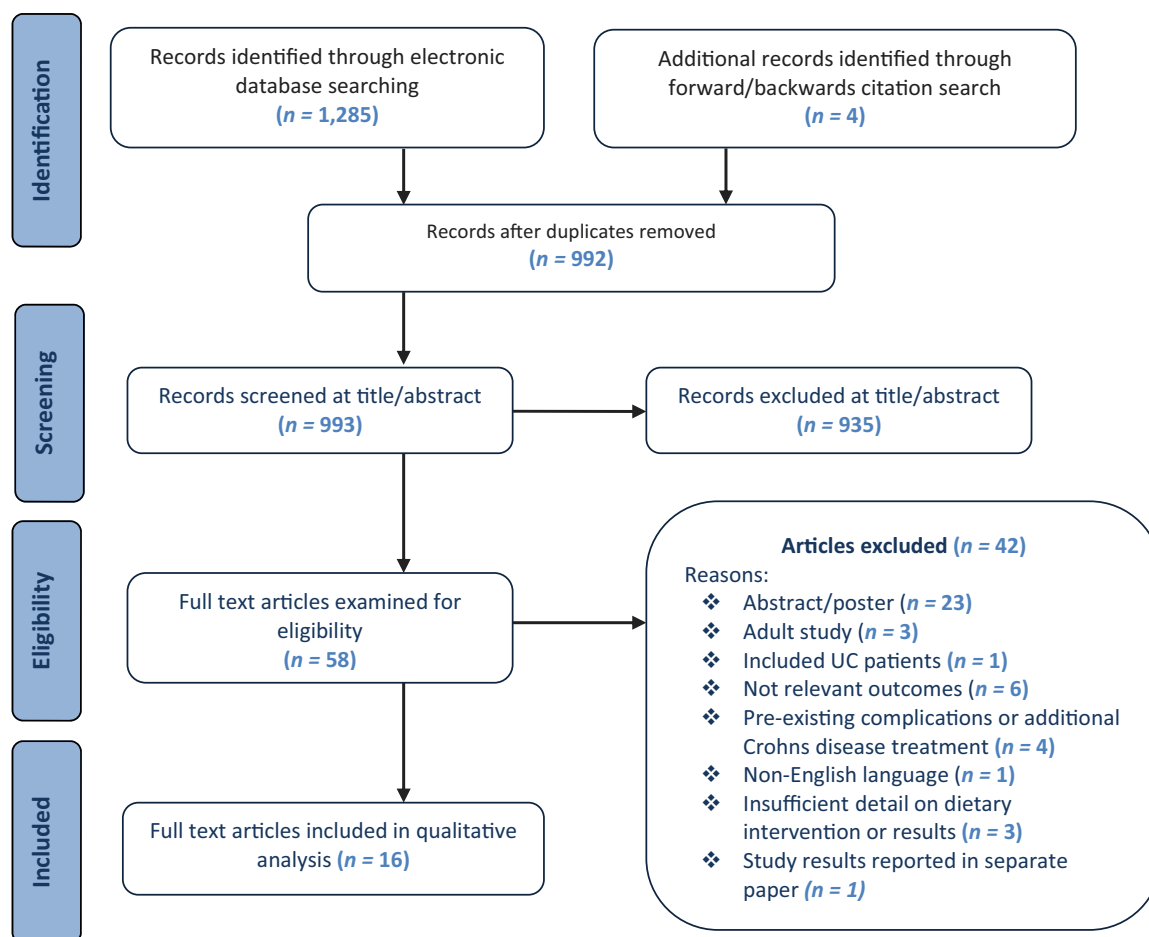


Figure 1 PRISMA diagram. UC, ulcerative colitis.

appraisal checklist was used for studies without a control group^(24–26). Because these tools only assess the quality in individual study types, and consequently do not take into consideration the inherent flaws and design specific risk of bias between the study types, additional elements related to study design were also examined and considered to supplement the tools listed. An overall quality rating was then given to each study, as shown in Table 2. The results from individual quality assessment tools are provided in the Supporting information (Tables S1–S4).

Although several randomised controlled trials (RCTs) were identified, there were insufficient data available overall to enable meta-analysis to be undertaken as a result of the considerable variation in study design.

Results

In total, 16 studies met the inclusion criteria, with the key findings being summarised in Table 3. The majority of the studies identified were of a retrospective design, including three cohort studies^(27–29) and eight

uncontrolled cohort/case series studies^(30–37). The remaining five studies were prospective studies and included four RCTs^(38–41) and one uncontrolled study⁽⁴²⁾. Overall, the quality of studies included in this systematic review was weak, with 11 studies being rated as poor or very poor quality once study type was taken into consideration, with noncontrolled studies receiving a lower overall rating. Three studies were considered to be of moderate quality and one identified as good/high quality overall.

Participant and study characteristics

The study participants were all aged under 18 years and came from a variety of countries, with six studies based in the UK, five studies throughout the rest of Europe, two studies from Asia, one from Australia and one from North America. All enteral diets were administered via nasogastric tube (NGT) or orally. Four studies included only newly diagnosed patients^(29,30,36,41) whereas the others included both newly diagnosed and relapsed patients. Disease activity and remission were based on the PCDAI⁽⁴³⁾ in most cases with remission assessed as

Table 2 Overall quality grade given to each study

Author	Quality score or risk of bias rating from assessment tool	Other weaknesses/risk of bias noted from critical appraisal*	Overall level of quality (high, moderate, low or very low)
Akobeng <i>et al.</i> ⁽⁴⁰⁾	Moderate risk of bias	Small sample size Study duration potentially too small to allow for effect	Low
Berni-Canani <i>et al.</i> ⁽²⁹⁾	Moderate quality	Only included patients who completed 8 weeks of EEN Small numbers in each subgroup of formula type Incomplete reporting of results (no numerical data)	Very low
Buchanan <i>et al.</i> ⁽³⁰⁾	Low quality	No control group Total remission calculation includes patients who started CS treatment during EEN period	Very low
Day <i>et al.</i> ⁽³¹⁾	Moderate quality	No control group	Low
De Bie <i>et al.</i> ⁽³⁷⁾	Moderate quality	No control group Non validated method of assessing remission	Very low
Fell <i>et al.</i> ⁽⁴²⁾	Moderate quality	No control group	Low
Gavin <i>et al.</i> ⁽³²⁾	Moderate quality	No control group Non validated method of assessing remission	Very low
Grogan <i>et al.</i> ⁽⁴¹⁾	Low risk of bias		High
Gupta <i>et al.</i> ⁽³⁵⁾	Moderate quality	No control group	Low
Johnson <i>et al.</i> ⁽³⁸⁾	Moderate risk of bias		Moderate
Kim <i>et al.</i> ⁽³³⁾	Moderate quality	No control group	Low
Knight <i>et al.</i> ⁽³⁶⁾	Moderate quality	No control group	Low
Ludvigsson <i>et al.</i> ⁽³⁹⁾	High risk of bias	Deviation from study protocol in 1/3 of patients which was documented but not adjusted for Small sample size	Low
Rodrigues <i>et al.</i> ⁽²⁷⁾	Moderate quality		Moderate
Rubio <i>et al.</i> ⁽²⁸⁾	Moderate quality		Moderate
Sigall-Boneh <i>et al.</i> ⁽³⁴⁾	Moderate quality	No control group	Low

CS, Crohn's corticosteroids; EEN, exclusive enteral nutrition.

*Downgraded – 1 for each weakness noted, +1 if Randomised controlled trial.

PCDAI <10. However, three studies used currently non-validated tools to assess clinical remission, which was taken into consideration when assessing the overall quality rating of the study^(27,30,37) Eleven studies assessed specific elements of enteral nutrition protocols, with six examining the effectiveness of different formula types^(27,29,39–42), one investigating the effectiveness of different routes of EEN administration⁽²⁸⁾ one looked at energy intakes/prescriptions in relation to clinical response⁽³²⁾ and three looked at use of a partial EN protocol^(34,35,38) The remaining five studies included in the review examined EEN in general^(30,31,33,36,37) No studies were identified that specifically examined the affect of varying protocol lengths on induction of remission.

Formula type

Five RCTs specifically investigated the effectiveness of different enteral formulas with respect to inducing remission in active disease, which included elemental, polymeric, semi-elemental or 'specialised formulas'. Three compared elemental and polymeric formulas^(27,39,41) and none found

any significant difference in remission rates between formula types when analysed on an intention-to-treat basis. One trial not only primarily compared the effectiveness of EEN with corticosteroids, but also undertook a sub-analysis comparing Elemental, semi-elemental and polymeric formulas and similarly found no difference in efficacy of inducing remission between formula types⁽²⁹⁾ Patients in this study had repeat endoscopic assessment after 8 weeks of treatment and all patients showed significant improvements in both their endoscopic and histological score ($P = < 0.001$) when on EEN, although no difference was seen between formulas. A study comparing a glutamine enriched formula with a standard polymeric formula similarly found no difference in levels of remission or anthropometric measures although interestingly, a greater decline in mean PCDAI score was actually seen in the standard polymeric group ($P = 0.002$)⁽⁴⁰⁾ Eight further studies specified the type of formula used with their EEN protocol, although no control group was provided for comparison and, apart from one study⁽⁴²⁾ the studies did not specifically aim to examine the efficacy of formula type. The three formula types used were elemental, standard polymeric and a

Table 3 Details of included studies

Author	Intervention/study design	Formula	Duration	Energy prescription	Outcomes of interest	Sample size	Control group?	Results
Akobeng <i>et al.</i> (40)	RCT comparing intervention using G-PF formula or standard PF when treating children with active CD with EEN	Specialised versus standard polymeric	4 weeks	Not specified	Clinical remission assessed by PCDAI (<10) Weight gain (kg)	n = 9 (G-PF) n = 9 (PF)	n = 9 (PF)	No significant difference in remission between groups (44% to 55%) No significant difference in mean weight gain between groups
Berni-Canani <i>et al.</i> (29)	Retrospective cohort study of children with newly diagnosed CD allocated to three different enteral formulas (polymeric, peptide, and elemental) or corticosteroid treatment (CS – control)	Polymeric versus peptide versus EF	8 weeks	50–70 kcal kg ⁻¹	Clinical remission rates assessed by PCDAI (<10) Endoscopic and histological remission Nutritional status assessed by serum iron and albumin levels and growth Occurrence of adverse events	n = 37 (EEN)	n = 10 (CS)	No significant difference in clinical remission between formulas or between EEN and CS Significant change in histological score ($P = < 0.001$) and height gain ($P = < 0.05$) only seen in EEN group (no difference between formulas) Significantly more adverse reactions with CS ($P = 0.003$)
Buchanan <i>et al.</i> (30)	Prospective uncontrolled cohort study/ case series examining effect of disease phenotype on efficacy of EEN	Specialised (Modulen)	8 weeks	EAR for age or more if underweight (mean 107% EAR)	Clinical remission assessed by global patient assessment Median weight, BMI and height Z-score changes	n = 110	No	Phenotype did not influence efficacy of EEN 80% achieved clinical remission when assessed by GPA Significant change in weight and BMI median Z-score ($P = < 0.01$) in those who achieved remission (not analysed in full group) No difference in remission rates between oral or NGT group
Day <i>et al.</i> (31)	Retrospective uncontrolled cohort study/ case series examining use of EEN in Australian children	Specialised (Modulen)	6–8 weeks	EAR for actual or ideal bodyweight	Clinical remission assessed by PCDAI < 10 Change in PCDAI score Weight change	n = 27	No	Overall 70% achieved clinical remission (PCDAI < 10) Significant improvement in PCDAI in all those who completed duration of EEN (n = 24) Significant weight gain in those who completed duration of EEN Significant improvements in inflammatory markers in those who completed the duration of EEN
De Ble <i>et al.</i> (37)	Retrospective uncontrolled cohort study/ case series examining use of standard EEN protocol in the Netherlands	Polymeric	6 weeks	110–120% EAR	Remission assessed by a pattern recognition model BMI Z-score	n = 58	No	53% complete remission, 20% partial response and 14% with no response Significant improvement in BMI Z-score in patients achieving remission ($P < 0.001$)

Table 3 Continued

Author	Intervention/study design	Formula	Duration	Energy prescription	Outcomes of interest	Sample size	Control group?	Results
Fell <i>et al.</i> (42)	Prospective uncontrolled cohort study/ uncontrolled pre/post design assessing mucosal healing induced by specialised oral polymeric EEN	Specialised (Modulen)	8 weeks	Not specified	Clinical response assessed by PCDAI (<10) Change in PCDAI score Endoscopic response % weight gain	n = 29	No	Significant decline in PCDAI in all patient ($P < 0.00001$). Median drop of 15 points 79% in clinical remission by week 8 (PCDAI < 10) Significant improvement in median colonoscopy score in paired analysis of 26 patients Mean weight gain of 3.2 kg and mean weight SD score increase of 0.42 ($P < 0.001$)
Gavin <i>et al.</i> (32)	Retrospective uncontrolled cohort study/ case series examining energy intakes of children treated with EEN	Specialised (Modulen)	6–8 weeks	EAR for age initially	Energy intake as % of EAR Weight gain (kg) and % weight change Change in CRP as marker of inflammation	n = 40	No	78% remission (CRP < 2 mg L ⁻¹) Significant improvement in median CRP showing biochemical remission in 78% of patients ($P = < 0.0001$) Median energy consumption 117% EAR, only 18% consumed EAR for age Median weight gain of 3.8 kg/ 11% No correlation between energy intake, remission and weight gain
Grogan <i>et al.</i> (41)	RCT comparing polymeric and elemental formula (EF) as EEN	Polymeric versus elemental	6 weeks	Not clear	Clinical remission assessed by PCDAI (<10) Weight Z-score	n = 21 (PF)	n = 20 (EF)	71% polymeric, 70% elemental achieved remission Significant improvement in mean weight Z-score in both groups No significant difference in outcomes between groups when analysed on ITT Significant improvement in PCDAI score ($P = < 0.0001$) Significant improvement in mean weight Z-score ($P = < 0.03$) Significant improvement in inflammatory markers ESR, CRP and Alb ($P = < 0.0001$, $P = 0.02$, $P = < 0.03$) Overall 65% achieved clinical remission by week 8
Gupta <i>et al.</i> (35)	Retrospective uncontrolled cohort study/ case series examining efficacy of novel EN protocol (not exclusive) at inducing remission	Variable (polymeric, elemental and peptide)	8–12 weeks	80–90% requirements assessed by Harris-Benedict equation	Clinical remission assessed by PCDAI (<10) Weight Z-scores Changes in inflammatory markers	n = 43	No	Significant improvement in PCDAI score ($P = < 0.0001$) Significant improvement in mean weight Z-score ($P = < 0.03$) Significant improvement in inflammatory markers ESR, CRP and Alb ($P = < 0.0001$, $P = 0.02$, $P = < 0.03$) Overall 65% achieved clinical remission by week 8

Table 3 Continued

Author	Intervention/study design	Formula	Duration	Energy prescription	Outcomes of interest	Sample size	Control group?	Results
Johnson <i>et al.</i> ⁽³⁸⁾	RCT comparing Partial EN (providing 50% EAR) with EEN	Elemental	6 weeks	50% EAR in PEN 100% EAR in EEN (increased to alleviate hunger)	Clinical remission assessed by PCDAI (<10) Changes in inflammatory markers Weight, weight/height SDS	n = 26 (PEN)	n = 24 (EEN)	Significant improvements in PCDAI score and weight/height gain in both groups ($P = < 0.0001$) But remission in 15% with PEN and 42% EEN (significant difference) Significant improvement in ESR and Alb in EEN group only No difference in mean weight or weight/height SDS between groups
Kim <i>et al.</i> ⁽³³⁾	Retrospective uncontrolled cohort study/ case series examining use of standard EEN protocol	Elemental	6 weeks	Not specified	Clinical remission assessed by PCDAI (<10)	n = 66	No	88% remission on ITT basis
Knight <i>et al.</i> ⁽³⁶⁾	Retrospective uncontrolled cohort study/ case series examining long term outcome of EEN	Elemental	8 weeks	Not specified	PCDAI	n = 44	No	90% remission 73% required NGT
Ludvigsson <i>et al.</i> ⁽³⁹⁾	RCT comparing polymeric and elemental formula (EF) as EEN	Polymeric versus elemental	6 weeks	0–10 kg = 100 kcal kg ⁻¹ , 10–20 kg = 50 kcal kg ⁻¹ , >20g = 20 kcal kg ⁻¹	Clinical remission assessed by PCDAI (<10 or decrease by 40%/15 points) Weight gain	n = 18 (PF)	n = 17 (EF)	82% Polymeric, 69% Elemental entered remission Significant improvements in PCDAI score ($P = < 0.001$) in both groups Significantly greater weight gain in polymeric group even when adjusting for lower volume/kcal consumed with elemental group
Rodrigues <i>et al.</i> ⁽²⁷⁾	Retrospective cohort study comparing polymeric with elemental formula (EF) and affect on adherence to EEN	Polymeric versus elemental	6 weeks	EAR for age, adjusted for hunger	Remission (? How assessed) Adherence to EEN Need for NGT	n = 45 (PF)	n = 53 (EF)	No significant difference in remission rates between groups (51 versus 64%) No significant difference between those completing course However, significantly more required NGT with Elemental ($P = 0.02$)

Table 3 Continued

Author	Intervention/study design	Formula	Duration	Energy prescription	Outcomes of interest	Sample size	Control group?	Results
Rubio <i>et al.</i> (28)	Retrospective cohort study comparing continuous NG feeding with fractionated oral feeding in EEN	Specialised (Modulen)	8 weeks	EAR for age	Clinical remission assessed by PCDAI (<10) Change in PCDAI score Weight gain and height gain	n = 61 (NGT)	n = 45 (oral)	Both groups showed significant improvements in PCDAI score, weight gain and inflammatory markers (ESR, CRP, Alb) all $P < 0.001$ 75% remission in oral group compared to 85% remission in NG group but not significant Greater weight gain in NGT group ($P < 0.05$)
Sigall-Boneh <i>et al.</i> (34)	Retrospective uncontrolled cohort study/ case series examining effectiveness of partial EN with a specialised Crohn's disease exclusion diet	Polymeric	6 weeks	50% EAR for age	Clinical remission assessed by PCDAI (<7) Changes in inflammatory markers	n = 47	No	Significant improvement in PCDAI ($P < 0.0001$) Significant improvement in ESR and CRP ($P < 0.0001$) but not Alb 72% entered remission although significantly more successful in those with mild-moderate disease compared to severe disease

Alb, albumin; BMI, body mass index; CRP, C-reactive protein; CS, Crohn's corticosteroids; CD, Crohn's disease; EAR, estimated average requirements; EEN, exclusive enteral nutrition; EF, elemental feeds; ESR, erythrocyte sedimentation rate; G-PF, glutamine-enriched polymeric formula; GPA, global patient assessment; ITT, intention-to-treat; NGT, nasogastric tube; PCDAI, Pediatric Crohn's Disease Activity Index; PEN, partial enteral nutrition; PF, polymeric formula; RCT, randomised controlled trial; SDS, standard deviation score.

specialised polymeric formula, Modulen IBD® (Nestle, Vevey, Switzerland) which is rich in transforming growth factor-beta 2 (TGF-β2), which has been reported to play a role in anti-inflammatory responses and autoimmunity⁽⁴⁴⁾ and so it is claimed to be more effective at inducing remission. Variable rates of remission were seen between the formulas (51–90%), as shown in Fig. 2.

Duration of treatment

No study specifically aimed to examine the affect of treatment duration on remission rates, although all studies did state the duration that treatment was prescribed for. This was either 6 or 8 weeks, apart from one study that prescribed treatment for 4 weeks⁽⁴⁰⁾ The 4-week EEN protocol showed the lowest percentage of remission achieved (44%), with the highest rates of remission noted in a study applying an 8-week protocol⁽³¹⁾ in which up to 90% of patients entered remission, although it was noted that the mean time until remission was only 6 weeks, with patients having been assessed every week. This was supported by the results obtained by Kim *et al.*⁽³³⁾ who also found positive results on a 6-week protocol, with 88% of patients entering remission by this point. Unfortunately, a direct comparison of remission rates grouped to treatment duration was not possible, with all studies differing too significantly in other aspects of EEN protocol design.

Route of delivery

Only one study examined whether the route of administration of EEN affected clinical outcome, retrospectively comparing a group of patients who received Modulen IBD® orally with those who had it via continuous NGT⁽²⁸⁾ No significant difference was seen between groups in remission rates although patients receiving their EEN via continuous NGT gained significantly more weight ($P < 0.05$) than those taking it orally, despite there being no difference in quantity of formula consumed. No assessment was made on level of activity and therefore energy expenditure between the groups.

Energy provision

Only one study specifically aimed to examine the effect of energy provision from EEN, although, unfortunately, no attempts were made to directly compare energy intake with remission rates⁽³²⁾. However, they did note that only 18% of patients consumed formula equal to the estimated average requirements (EAR) for energy for age, which was the initial target prescription. The remaining 82% of patients all consumed above the EAR, with the median intake being 117% EAR for age. Within the other studies identified, one study routinely prescribed 110–120% EAR for age⁽³⁷⁾ whereas another used a calculation that appears to provide less than the current EAR

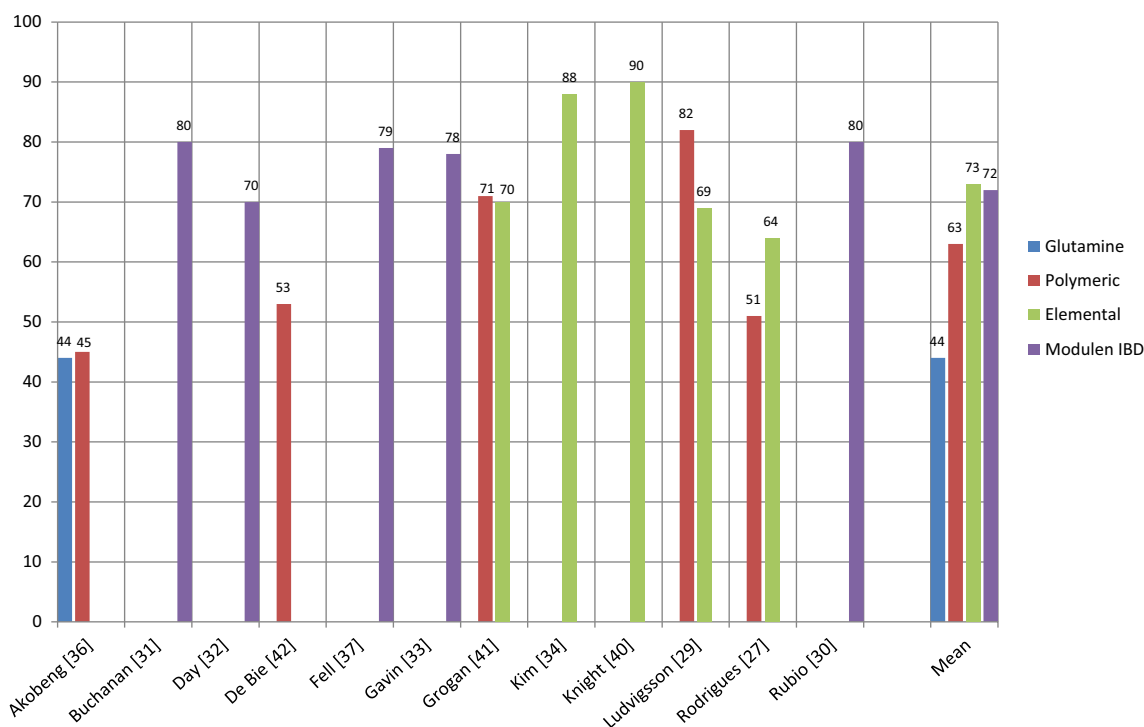


Figure 2 Comparison of formula type showing the percentage of patients achieving remission.

guidelines (SACN, 2011), up to 800 calories less per day in older children⁽³⁹⁾ The majority of other studies stated that the initial volume of formula was calculated with respect to the EAR for age, although it was increased to alleviate hunger or if the patient was underweight. Because there were too many variables between studies, it was not possible to correlate energy intake with remission to determine conclusively whether the volume of formula prescribed influences remission rates.

Exclusivity

Three studies applied a partial enteral nutrition (PEN) protocol as a method to induce remission although all differed in the percentage of formula prescribed, formula type used and whether additional dietary restrictions were applied to the solid food allowed; thus, as a result, a direct quantitative comparison was not possible. Only one of the three studies was an RCT⁽³⁸⁾ which found a significantly lower remission rate of 15% in patients prescribed 50% of their EAR for age as a liquid formula plus food compared to those prescribed 100% of EAR as exclusively liquid formula diet (42%; $P = 0.035$). Differing results were obtained in a study by Sigall-Boneh *et al.*⁽³⁴⁾ who similarly prescribed 50% of energy intake as liquid formula but requested that patients follow a structured 'Crohn's disease exclusion diet' with specific foods excluded as opposed to an unrestricted one. In their study, higher rates of remission were noted, with 72% of patients entering remission assessed by the PCDAI, although patients with mild-moderate disease were more likely to respond than those with severe disease ($P < 0.05$)⁽³⁴⁾ The final study reviewed applied a higher percentage of the EAR as liquid formula with patients being prescribed 80–90% EAR, infused via NGT overnight and allowed the remaining 10–20% EAR from solid food during the day. Overall, 65% achieved clinical remission by week 8, with a further 22% demonstrating a clinically significant response⁽³⁵⁾

Discussion

Exclusive enteral nutrition therapy has been suggested as an effective treatment option in both adult and paediatric patients to control the extent of inflammation for many years, although significant variation exists in the protocols applied and rates of remission seen between studies. The aim of this systematic review was therefore to evaluate and compare the clinical effectiveness of aspects of EEN protocols to ascertain whether an optimum regimen can be identified. Published research over a 20-year period up to March 2019 has been reviewed, giving a unique insight

into the influence of aspects of EEN with respect to inducing remission in paediatric patients, as discussed further below. Considerable heterogeneity was found between studies and thus it has not been possible to perform a meta-analysis of data to identify an optimum feeding protocol.

As yet, the exact mechanisms of the action of EEN are not known. This may partly explain the heterogeneity seen between the studies. Both nutritional and anti-inflammatory mechanisms are probably involved and there are several hypothesised mechanisms underlying the proposed benefits of EEN in CD, including a reduction of antigenic load, reduced gut activity or provision of 'bowel rest', nutritional rehabilitation effects, anti-inflammatory effects or modulation of immune system and gastrointestinal flora⁽⁴⁵⁾. A recent study by Ashton *et al.*⁽⁴⁶⁾ provides a good review of the evidence surrounding these proposed mechanisms and some of the theories are discussed below in the context of the objectives and findings of the present systematic review.

Formula choice

Elemental feeds (EF) were traditionally the formula of choice for EEN stemming from early observations that an amino acid formula given to malnourished patients with poorly controlled disease for nutrition support could also induce remission⁽⁴⁷⁾. Consequently, there was an assumption that the therapeutic effects observed were a result of the absence of the whole protein in the diet and that dietary antigen stimulation might promote the inflammatory process. Providing EEN as an elemental formula would limit antigen exposure and thereby reduce inflammation, with data existing to support this suggestion^(48,49). That being said, the evidence is variable and a recent Cochrane review concluded that there was insufficient evidence to support the use of EF over other formulas⁽²⁰⁾. The Cochrane review only included one paediatric study⁽⁴¹⁾ but, by widening the inclusion criteria in the present systematic review, it was possible to examine additional paediatric studies, similarly suggesting no difference between elemental or polymeric formulas with respect to inducing remission. However, this alone is insufficient evidence to argue against the antigen theory and exclusive feeding from an artificial, manufactured liquid formula could still potentially limit intake of an unknown antigen from a 'normal diet'.

It is also worth noting that the evidence reviewed is less supportive of the use of elemental diets over a standard polymeric formula (PF) when examining additional outcome measures such as weight gain and compliance. Two

studies in this systematic review noted greater improvements in weight gain with a standard PF compared to an EF, even after adjustment for energy intake^(39,41) and it is suggested that the high osmolality of EF increased stool output and thereby energy loss. Compliance is also an important factor to consider when looking at EEN and, theoretically, the provision of a more palatable liquid formula may improve compliance. Although, from the studies examined, no difference in overall compliance was noted between EF and PF, significant numbers of patients (55–100%) required EF via a NGT. The use of NGTs has been shown to be associated with patient's negative feelings towards EEN⁽⁵⁰⁾ and so the provision of a more palatable formula may facilitate increased acceptance and overall uptake of EEN therapy.

Although the exact relationship is yet to be clarified, recent evidence now indicates that anti-inflammatory effects are primary components of the aetiology of EEN. TGF- β 2 is a bioactive substance present in both human and bovine milk that has been shown to play a critical role not only in the development of tolerance, prevention of autoimmunity, but also in anti-inflammatory responses^(51,52). For this reason, there are claims that Modulen IBD® could be superior to other formulas. Although the precise levels have not been measured, because the sole source of protein in Modulen IBD® is casein, it is assumed to be rich in TGF- β 2. The studies that used Modulen IBD® demonstrated good rates of remission (75–85%)^(28,30,31) although, because no papers were identified that compared Modulen IBD® directly with other formulas, it is not possible to presume its greater efficacy. *In vitro* models have also suggested that a standard PF also has anti-inflammatory effects upon the gastrointestinal mucosa⁽⁵³⁾, perhaps because they also usually contain protein derived from bovine milk. The rates of remission seen in the studies using standard PF were more variable, with some finding rates comparable to Modulen IBD®^(39,41) whereas others showed rates of only 50–53%^(37,42) although it is worth noting that, as a result of high dropout rates in one study, a remission of 71% was achieved when the data were analysed per protocol⁽³⁷⁾.

Energy provision

Calculating the nutritional requirements of patients starting nutritional therapy is clearly of clinical relevance but, unfortunately, there is little published guidance available to date to address this with confidence. Patients presenting in a malnourished state often have reduced energy requirements^(54,55) but, in active CD, as a result of additional demands placed on the body

by the inflammatory process, it is likely this reduction does not occur. Although the current consensus is that EEN should provide at least 100% EAR for age⁽²¹⁾, these values originate from data on healthy, well-nourished children, and so they could be inadequate for those children in an inflammatory state with altered digestion and absorption capacity. Only one study was identified in the present review that specifically examined the calorific intake of patients being treated with EEN⁽³²⁾. They supported the assumption that patients with active disease have requirements above EAR for age, noting that, when children were encouraged to take sufficient formula to feed their appetite, 82% consumed above the EAR, with a median intake of 118% EAR for age. However, this was a purely observational study. No significant difference was found when correlating energy intake and percentage weight gain, although the study did not examine energy intake with remission, and so it is not possible to draw conclusions regarding whether the energy consumed affected remission rates⁽³²⁾. Theoretically, suboptimal dosing has the potential to affect the efficacy of EEN and it may be prudent to adopt an initial target above the EAR and carry out careful regular monitoring of hunger, weight gain, tolerance and physical activity levels to allow the advised energy prescription to be titrated according to response. There is a need for more detailed and complete studies of how energy expenditure, and therefore energy requirements, may differ between patients who can vary markedly due to the differences in disease severity and phenotype, as well as the presenting degree of malnutrition.

Duration of treatment

The duration of prescribed EEN varies substantially, with an international survey of gastroenterology units demonstrating a range of 2–12 weeks⁽⁵⁶⁾. The majority of units surveyed, however, used a 6–8-week protocol, which corresponds with the results of this systematic review. The results reviewed suggest that this is likely to be sufficient for the majority of patients, although it should be acknowledged that a longer course may potentially increase efficacy in certain patients, such as those in a severely malnourished state or with colonic disease. Potential long-term benefits should also be considered, such as sustained remission and better growth if the induction course of EEN is longer. It is likely that the adequate length of treatment will vary between patients, and also that the time period prescribed may need to be a compromise between ensuring compliance with maximum efficacy. Focused evaluations of EEN over different time periods are clearly required.

Exclusivity

Adherence to EEN is difficult and, as mentioned previously, this can have significant psychosocial consequences on patients. Many gastrointestinal units report allowing some additional nutritional components (e.g. hard boiled sweets) to the liquid formula to support compliance, despite the proposed model of exclusivity⁽⁵⁶⁾. These small additions have not been evaluated formally, although three studies were identified in this review that did specifically challenge the theory of exclusivity by prescribing a specified amount of liquid formula alongside solid diet as an alternative treatment method. However, unfortunately, mixed results were seen. In the strongest of these studies, using 50% of EAR with formula in addition to a free diet was shown to have some clinical benefit in terms of improved well-being and weight gain, although it did not suppress intestinal inflammation, with only 15% of patients entering remission by the end of therapy⁽³⁸⁾. It is worth noting, however, that only patients with moderate-severe disease were included, and a subsequent study treating patients mostly with mild-moderate disease found much higher remission rates (70%) on a PEN protocol similarly providing 50% EAR as liquid formula and 50% as free diet⁽³⁴⁾.

There are other possible reasons behind the stark differences in remission rates found. For example, there are growing suggestions that the gut microbiota might play a significant role in the development of CD in susceptible individuals, with several studies reporting altered gut microflora eliciting an inflammatory response of the intestinal mucosa in subjects with an underlying immunological mucosal defect^(57–59). There is also an increasing body of evidence suggesting that EEN allows modification of gut microflora to a more favourable state, leading to the functional changes that allow inflammation to be controlled^(60–63).

The poor remission rates (15% in PEN group) found by Johnson *et al.*⁽³⁸⁾ which suggest that the efficacy of nutritional therapy is reduced when a free diet is permitted alongside PEN, lend support to the theory that the ongoing consumption of certain dietary components found in a free diet could negatively alter the microbiome, or at least prevent its modification to a more favourable state. Expanding on this theory, rather than allow a 'free diet' alongside PEN, Sigall-Boneh *et al.*⁽³⁴⁾ applied a dietary intervention that reduced exposure to certain dietary components previously shown to induce inflammation or change the microbiome in animal/*in vitro* models^(64,65). The Crohn's disease exclusion diet (CDED) was prescribed to patients alongside 50% EAR from a polymeric liquid and involved the avoidance of animal fat, dairy products, gluten and emulsifiers for

6 weeks, finding a higher rate of remission (72%), especially in those with mild–moderate disease. A newly published RCT (July 2019) by the same research group adds further support to use of this novel nutritional therapy. Levine *et al.*⁽⁶⁶⁾ demonstrate that, although there was no significant difference in the number of patients entering remission by week 6, the CDED with PEN ($n = 40$) was better tolerated than EEN ($n = 34$), with a significantly lower dropout rate. It was also suggested that the CDED could be superior to EEN in terms of sustaining remission, with a significantly greater proportion of the CDED group having a PCDAI <10 at week 12 compared to the EEN⁽⁶⁶⁾. Alternative dietary treatment options to EEN are certainly a growing area of interest, with another research team having also recently published their findings after examining a completely solid food-based diet as opposed to PEN alongside a specialised diet. The CD-TREAT was designed to replicate the nutrients and composition of EEN and thereby hypothesised to mimic its effect on the gut microbiome⁽⁶⁷⁾. Although the diet was only applied to a very small paediatric group with active Crohn's disease ($n = 5$), the results obtained are promising, with 80% having a clinical response, and examination of the faecal microbiome composition in healthy adults showed that the diet induced similar effects to EEN and was easier to comply with.

Interestingly, the final study identified in the systematic review examining the efficacy of PEN therapy also found reasonable levels of remission (65%) despite allowing a free diet⁽³⁵⁾. However, the key difference in their PEN protocol was that free diet should have provided only 10–20% requirements, with the remaining 80–90% coming from the prescribed liquid diet. It may be that the greater anti-inflammatory effects seen could be attributed to a simple 'dose effect'. By providing a greater proportion of the diet as liquid, the biological effect on inflammatory process and microbiome alterations were maintained despite the intake of a small quantity of unrestricted whole foods.

Limitations of evidence

Unfortunately, given the limitations of the available evidence in terms of sample size, study design, methodological quality and risk of bias in individual trials identified, the interpretation of the findings outlined above should be made with caution. Heterogeneity was also unfortunately seen in all aspects of the studies, from the inclusion criteria used, through to how the results were expressed or presented in the final written document, making the interpretation of the data more difficult. One of the key limitations of the available evidence is that, at present, there is no agreed gold standard or

defined method for assessing remission. Studies that use endoscopic assessment at the end of treatment will show a more truthful representation of reduction of inflammation, although, because this is costly and invasive for patients, it is rarely used to assess the early treatment response. Nevertheless, its use in disease monitoring is increasing as a result of the more recent focus on mucosal healing⁽⁶⁸⁾, and so, in the future, it may be used more commonly to assess treatment outcomes. The PCDAI is perhaps the most widely accepted and evaluated quantitative measure to assess disease activity⁽⁶⁹⁾, although it is not without its limitations, such as using a greater proportion of subjective measures to score disease activity, which are more prone to bias. It has also been suggested that it only has a fair to poor correlation with endoscopic assessment of the level of mucosal healing⁽⁷⁰⁾. Many studies also did not limit inclusion to the first exacerbation of disease and the significance of this is not clear; only one group conducted a sub-analysis of remission rates between newly diagnosed patients and those with long-standing disease, finding greater efficacy of EEN in those with first presentation of disease⁽³¹⁾. The inclusion of patients with existing disease introduced additional sources of confounding. Although the inclusion criteria for this systematic review was that patients should not receive additional treatment medication during their course of nutritional therapy, medication given as a prophylactic dose was allowed, and this differed across the studies and treatment groups in frequency/dose, making it difficult to relate any observed outcomes to dietary treatment alone.

Conclusions

Although extensive research is available examining the effectiveness of EEN, a limited number of systematic reviews have been conducted specifically in the paediatric population with Crohn's disease, and none in the past 10 years. This is also the first systematic review to examine the various elements of EEN regimens in depth as opposed to EEN versus corticosteroid treatment. From the 16 research studies identified, the results continue to support EEN being an effective treatment option for paediatric patients with active Crohn's disease, although, unfortunately, it has not been possible to further our understanding of what specific aspects of EEN protocols could improve remission rates. An elemental formula appears to have no substantial benefit over the more palatable and cheaper polymeric formula, supporting the conclusions drawn from systematic reviews of research in adult populations. However, a cautious interpretation of the findings is still needed given the small number of studies, methodological weaknesses, poor reporting and a role of bias with

respect to influencing the results. Further focused research is required using well designed and adequately powered, randomised prospective clinical trials to address whether specialised formulas, such as Modulen IBD® are superior to other formulas, as claimed by the manufacturers, as well as other aspects of EEN protocols. There are some early indications that PEN may be a viable alternative in patients where the exclusion of all solid food is considered unmanageable, although additional research is required to develop this idea further and determine the optimum ratio of liquid diet to solid food, as well as whether certain dietary components/specific foods need to be excluded for optimum efficacy. However, because our understanding of the pathology of Crohn's disease remains limited, until we know more about this, as well as how nutritional therapies work, it will be difficult to formulate meaningful research to ascertain the best way of applying either EEN or PEN to patients.

Conflict of interests, source of funding and authorship

The authors declare that they have no conflicts of interest. This paper represents self funded independent research as part of a master's thesis. Both authors participated in the protocol development. LM conducted the literature search, and screened and extracted the data from the literature, with AP reviewing for completeness. LM wrote the first draft with contributions from AP, with subsequent drafts also being reviewed and commented on. Both authors approved the final version of the manuscript submitted for publication.

Transparency declaration

The authors affirm that this manuscript is an honest accurate, and transparent account of the study being reported. The authors affirm that no important aspects of the study have been omitted and that any discrepancies from the study as planned have been explained. The reporting of this work is compliant with PRISMA guidelines and registered with PROSPERO (registration number CRD42018100463).

References

1. Penagini F, Dillillo D, Borsani B *et al.* (2016) Nutrition in pediatric inflammatory bowel disease: from etiology to treatment. A systematic review. *Nutrients* **8**, E344.
2. Stange EF, Travis SP, Vermeire S *et al.* (2006) European evidence based consensus on the diagnosis and management of Crohn's disease: definitions and diagnosis. *Gut* **55**(Suppl 1), i1–15.

3. Travis SP, Stange EF, Lémann M *et al.* (2006) European evidence based consensus on the diagnosis and management of Crohn's disease: current management. *Gut* **55**(Suppl 1), i16–35.
4. Benchimol EI, Fortinsky KJ, Gozdyra P *et al.* (2011) Epidemiology of pediatric inflammatory bowel disease: a systematic review of international trends. *Inflamm Bowel Dis* **17**, 423–39.
5. Gasparetto M & Guariso G (2014) Crohn's disease and growth deficiency in children and adolescents. *World J Gastroenterol* **20**, 13219–13233.
6. Pappa H, Thayu M, Sylvester F *et al.* (2011) Skeletal health of children and adolescents with inflammatory bowel disease. *J Pediatr Gastroenterol Nutr* **53**, 11–25.
7. Schmidt S, Mellström D, Norjavaara E *et al.* (2012) Longitudinal assessment of bone mineral density in children and adolescents with inflammatory bowel disease. *J Pediatr Gastroenterol Nutr* **55**, 511–518.
8. Sawczenko A & Sandhu BK (2003) Presenting features of inflammatory bowel disease in Great Britain and Ireland. *Arch Dis Child* **88**, 995–1000.
9. Jewell DP (1989) Corticosteroids for the management of ulcerative colitis and Crohn's disease. *Gastroenterol Clin North Am* **18**, 21–34.
10. Ostro MJ, Greenberg GR & Jeejeebhoy KN (1985) Total parenteral nutrition and complete bowel rest in the management of Crohn's disease. *J Parenter Enteral Nutr* **9**, 280–287.
11. Zachos M, Tondeur M & Griffiths AM (2007) Enteral nutritional therapy for induction of remission in Crohn's disease. *Cochrane Database Syst Rev* **1**, CD000542.
12. Thomas AG, Taylor F & Miller V (1993) Dietary intake and nutritional treatment in childhood Crohn's disease. *J Pediatr Gastroenterol Nutr* **17**, 75–81.
13. Connors J, Basseri S, Grant A *et al.* (2017) Exclusive enteral nutrition therapy in paediatric Crohn's disease results in long-term avoidance of corticosteroids: results of a propensity-score matched cohort analysis'. *J Crohn Colitis* **11**, 1063–1070.
14. Cohen-Dolev N, Sladek M, Hussey S *et al.* (2018) Differences in outcomes over time with exclusive enteral nutrition compared to steroids in children with mild to moderate Crohn's Disease: results from the GROWTH CD study. *J Crohns Colitis* **28**, 306–312.
15. Lafferty L, Tuohy M, Carey M *et al.* (2017) Outcomes of exclusive enteral nutrition in pediatric crohn's disease. *Eur J Clin Nutr* **71**, 185–191.
16. Heuschkel RB, Menache CC, Megerian JT *et al.* (2000) Enteral nutrition and corticosteroids in the treatment of acute Crohn's disease in children. *J Pediatr Gastroenterol Nutr* **31**, 8–15.
17. Dziechciarz P, Horvath A, Shamir R *et al.* (2007) Meta-analysis: enteral nutrition in active Crohn's disease in children. *Aliment Pharmacol Ther* **26**, 795–806.
18. Swaminath A, Feathers A, Ananthakrishnan AN *et al.* (2017) Systematic review with meta-analysis: enteral nutrition therapy for the induction of remission in paediatric Crohn's disease'. *Aliment Pharmacol Ther* **46**, 645–656.
19. Yu Y, Kang-Chen C & Jie C (2019) Exclusive enteral nutrition versus corticosteroids for treatment of pediatric Crohn's disease: a meta-analysis. *World J Pediatr* **15**, 26–36.
20. Narula N, Dhillon A, Zhang D *et al.* (2018) Enteral nutritional therapy for induction of remission in Crohn's disease. *Cochrane Database Syst Rev* **4**, CD000542.
21. Ruemmele FM, Veres G, Kolho KL *et al.* (2014) Consensus guidelines of ECCO/ESPGHAN on the medical management of pediatric Crohn's disease. *J Crohns Colitis* **8**, 1179–207.
22. Day AS, Whitten KE, Sidler M *et al.* (2008) Systematic review: nutritional therapy in paediatric Crohn's disease. *Aliment Pharmacol Ther* **27**, 293–307.
23. Moher D, Shamseer L, Clarke M *et al.* (2015) Preferred reporting items for systematic review and meta-analysis protocols (PRISMA-P) 2015 statement. *Syst Rev* **4**, 1.
24. Higgins JPT, Thomas J, Chandler J *et al.* (editors) (2019) *Cochrane Handbook for Systematic Reviews of Interventions* (2nd edn). Chichester: John Wiley & Sons.
25. Wells G, Shea B, O'Connell Det *et al.* (2008) The Newcastle-Ottawa Scale (NOS) for assessing the quality of nonrandomised studies in meta-analyses. http://www.ohri.ca/programs/clinical_epidemiology/oxford.asp
26. Guo B, Moga C, Harstall C & *et al.* (2016) A principal component analysis is conducted for a case series quality appraisal checklist. *J Clin Epidemiol* **69**, 199–207.e2.
27. Rodrigues AF, Johnson T, Davies P *et al.* (2007) Does polymeric formula improve adherence to liquid diet therapy in children with active Crohn's disease? *Arch Dis Child* **92**, 767–70.
28. Rubio A, Pigneur B, Garnier-Lengline H *et al.* (2011) The efficacy of exclusive nutritional therapy in paediatric Crohn's disease, comparing fractionated oral vs. continuous enteral feeding. *Aliment Pharmacol Ther* **33**, 1332–9.
29. Berni Canani R, Terrin G, Borrelli O *et al.* (2006) Short- and long-term therapeutic efficacy of nutritional therapy and corticosteroids in paediatric Crohn's disease. *Dig Liver Dis* **38**, 381–387.
30. Buchanan E, Gaunt WW, Cardigan T *et al.* (2009) The use of exclusive enteral nutrition for induction of remission in children with Crohn's disease demonstrates that disease phenotype does not influence clinical remission. *Aliment Pharmacol Ther* **30**, 501–507.
31. Day AS, Whitten KE, Lemberg DA *et al.* (2006) Exclusive enteral feeding as primary therapy for Crohn's disease in Australian children and adolescents: A feasible and effective approach. *J Gastroenterol Hepatol* **21**, 1609–1614.

32. Gavin J, Anderson CE, Bremner AR & et al (2005) Energy intakes of children with Crohn's disease treated with enteral nutrition as primary therapy. *J Hum Nutr Diet* **18**, 337–342.
33. Kim HJ, Kim Y, Cho JM, et al (2016) Therapeutic efficacy of oral enteral nutrition in pediatric Crohn's disease: a single center non-comparative retrospective study. *Yonsei Med J* **57**, 1185–1191.
34. Sigall-Boneh R, Pfeffer-Gik T, Segal I *et al.* (2014) Partial enteral nutrition with a Crohn's disease exclusion diet is effective for induction of remission in children and young adults with Crohn's disease. *Inflamm Bowel Dis* **20**, 1353–1360.
35. Gupta K, Noble A, Kachelries KE *et al.* (2013) A novel enteral nutrition protocol for the treatment of pediatric Crohn's disease. *Inflamm Bowel Dis* **19**, 1374–1378.
36. Knight C, El-Matary W, Spray C *et al.* (2005) Long-term outcome of nutritional therapy in paediatric Crohn's disease. *Clin Nutr* **24**, 775–9.
37. de Bie C, Kindermann A & Escher J (2013) Use of exclusive enteral nutrition in paediatric Crohn's disease in The Netherlands. *J Crohns Colitis* **7**, 263–270.
38. Johnson T, Macdonald S, Hill SM *et al.* (2006) Treatment of active Crohn's disease in children using partial enteral nutrition with liquid formula: a randomised controlled trial. *Gut* **55**, 356–361.
39. Ludvigsson JF, Krantz M, Bodin L *et al.* (2004) Elemental versus polymeric enteral nutrition in paediatric Crohn's disease: a multicentre randomized controlled trial. *Acta Paediatr* **93**, 327–35.
40. Akobeng AK, Miller V, Stanton J, et al (2000) Double-blind randomized controlled trial of glutamine-enriched polymeric diet in the treatment of active Crohn's disease. *J Pediatr Gastroenterol Nutr* **30**, 78–84.
41. Grogan JL, Casson DH, Terry A *et al.* (2012) Enteral feeding therapy for newly diagnosed pediatric Crohn's disease: a double-blind randomized controlled trial with two years follow-up. *Inflamm Bowel Dis* **18**, 246–253.
42. Fell JME, Paintin M, Arnaud-Battandieri F *et al.* (2000) Mucosal healing and a fall in mucosal pro-inflammatory cytokine mRNA induced by a specific oral polymeric diet in paediatric Crohn's disease. *Aliment Pharmacol Ther* **14**, 281–289.
43. Hyams JS, Ferry GD, Mandel FS *et al.* (1991) Development and validation of a pediatric Crohn's disease activity index. *J Pediatr Gastroenterol Nutr* **12**, 439–447.
44. Triantafyllidis J, Asteria Stamataki A, Sklavaina M *et al.* (2006) Beneficial effect of a polymeric feed, rich in TGF-beta, on adult patients with active crohn's disease: a pilot study. *Anna Gastroenterol* **19**, 66–71.
45. Wedrychowicz A, Zajac A & Tomasik P (2016) Advances in nutritional therapy in inflammatory bowel diseases: review. *World J Gastroenterol* **22**, 1045–1066.
46. Ashton JJ, Gavin J & Beattie RM (2019) Exclusive enteral nutrition in Crohn's disease: evidence and practicalities. *Clin Nutr* **38**, 80–9.
47. Voitk AJ, Echave V, Feller JH *et al.* (1973) Experience with elemental diet in the treatment of inflammatory bowel disease. Is this primary therapy? *Arch Surg* **107**, 329–333.
48. Gjaffer MH, North G & Holdsworth CD (1990) Controlled trial of polymeric versus elemental diet in treatment of active Crohn's disease. *Lancet* **335**, 816–819.
49. Verma S, Brown S, Kirkwood B *et al.* (2000) Polymeric versus elemental diet as primary treatment in active Crohn's disease: a randomized, double-blind trial. *Am J Gastroenterol* **95**, 735–739.
50. Gailhoustet L, Goulet O, Cachin N *et al.* (2002) Study of psychological repercussions of 2 modes of treatment of adolescents with Crohn's disease. *Arch Pediatr* **9**, 110–116.
51. Triantafyllidis JK, Vagianos C & Papalois AE (2015) The role of enteral nutrition in patients with inflammatory bowel disease: current aspects. *Biomed Res Int* **2015**, 1–12.
52. Beattie RM, Schiffrin EJ, Donnet-Hughes A *et al.* (1994) Polymeric nutrition as the primary therapy in children with small bowel Crohn's disease. *Aliment Pharmacol Ther* **8**, 609–615.
53. de Jong NS, Leach ST & Day AS (2007) Polymeric formula has direct anti-inflammatory effects on enterocytes in an in vitro model of intestinal inflammation. *Dig Dis Sci* **52**, 2029–2036.
54. Weyer C, Walford RL, Harper IT *et al.* (2000) Energy metabolism after 2 y of energy restriction: the biosphere 2 experiment. *Am J Clin Nutr* **72**, 946–953.
55. Lindmark L, Bennegård K, Edén E *et al.* (1984) Resting energy expenditure in malnourished patients with and without cancer. *Gastroenterology* **87**, 402–408.
56. Whitten KE, Rogers P, Ooi CY *et al.* (2012) International survey of enteral nutrition protocols used in children with Crohn's disease. *J Dig Dis* **13**, 107–112.
57. Sartor RB (2004) Therapeutic manipulation of the enteric microflora in inflammatory bowel diseases: antibiotics, probiotics, and prebiotics. *Gastroenterology* **126**, 1620–1633.
58. Saunier K & Dore J (2002) Gastrointestinal tract and the elderly: functional foods, gut microflora and healthy ageing. *Dig Liver Dis* **34**(Suppl 2), S19–24.
59. Martin HM, Campbell BJ, Hart CA *et al.* (2004) Enhanced *Escherichia coli* adherence and invasion in Crohn's disease and colon cancer. *Gastroenterology* **127**, 80–93.
60. Lionetti P, Callegari ML, Ferrari S *et al.* (2005) Enteral nutrition and microflora in pediatric Crohn's disease. *J Parenter Enteral Nutr* **29**(4 Suppl), S173–S175; discussion S5–8, S84–8.
61. MacLellan A, Moore-Connors J, Grant S *et al.* (2017) The impact of exclusive enteral nutrition (EEN) on the gut microbiome in Crohn's disease. A Review. *Nutrients* **9**, 0447.

62. Gatti S, Galeazzi T, Franceschini E *et al.* (2017) Effects of the exclusive enteral nutrition on the microbiota profile of patients with Crohn's disease: a systematic review. *Nutrients* **9**, 832.
63. Quince C, Ijaz UZ, Loman N *et al.* (2015) Extensive modulation of the fecal metagenome in children with Crohn's disease during exclusive enteral nutrition. *Am J Gastroenterol* **110**, 1718–1729, quiz 30.
64. Martinez-Medina M, Denizot J, Dreux N *et al.* (2014) Western diet induces dysbiosis with increased *E. coli* in CEABAC10 mice, alters host barrier function favouring AIEC colonisation. *Gut* **63**, 116–124.
65. Swidsinski A, Ung V, Sydora BC *et al.* (2009) Bacterial overgrowth and inflammation of small intestine after carboxymethylcellulose ingestion in genetically susceptible mice. *Inflamm Bowel Dis* **15**, 359–364.
66. Levine A, Wine E, Assa A *et al.* (2019) Crohns disease exclusion diet plus partial enteral nutrition induces sustained remission in a randomised controlled trial. *Gastroenterology* **157**, 440–450.
67. Svolos V, Hansen R, Nichols B *et al.* (2019) Treatment of active Crohn's disease with ordinary food-based diet that replicates exclusive enteral nutrition. *Gastroenterology* **156**, 1354–1367.
68. Rameshshanker R & Arebi N (2012) Endoscopy in inflammatory bowel disease when and why. *World J Gastrointest Endosc* **4**, 201–211.
69. Kundhal PS, Critch JN, Zachos M *et al.* (2003) Pediatric Crohn disease Activity Index: responsive to short-term change. *J Pediatr Gastroenterol Nutr* **36**, 83–89.
70. Zubin G & Peter L (2015) Predicting endoscopic Crohn's disease activity before and after induction therapy in children: a comprehensive assessment of PCDAI, CRP, and fecal calprotectin. *Inflamm Bowel Dis* **21**, 1386–1391.

Supporting information

Additional supporting information may be found online in the Supporting Information section at the end of the article.

Table S1. Cochrane RoB assessment for RCT's.

Table S2. Newcastle Ottawa Scale for Cohort studies.

Table S3. Quality assessment of case series and studies without comparator arms.

OLDER PEOPLE

The use of finger foods in care settings: an integrative review

M. Heelan,¹  J. Prieto,¹  H. Roberts,²  N. Gallant,¹ C. Barnes³  & S. Green⁴ 

¹School of Health Sciences, University of Southampton, Southampton, UK

²Faculty of Medicine, University of Southampton, Southampton General Hospital, Southampton, UK

³Research and Improvement Team, St Marys Community Health Campus, Portsmouth, UK

⁴Faculty of Health and Social Sciences, Bournemouth University, Bournemouth, UK

Keywords

adults, care setting, finger foods, integrative review.

Correspondence

M. Heelan, School of Health Sciences, University of Southampton, University Road, Southampton, SO17 1BJ, UK.

Tel.: +44 07708943678

E-mail: a.r.heelan@soton.ac.uk

How to cite this article

Heelan M., Prieto J., Roberts H., Gallant N., Barnes C. & Green S. (2020) The use of finger foods in care settings: an integrative review. *J Hum Nutr Diet.* **33**, 187–197
<https://doi.org/10.1111/jhn.12725>

Abstract

Background: Reduced food intake is prevalent in people in residential and hospital care settings. Little is known about the use of finger foods (i.e. foods eaten without cutlery) with respect to increasing feeding independence and food intake. The Social Care Institute for Excellence (Malnutrition Task Force: State of the Nation, 2017) recommends the use of finger foods to enable mealtime independence and to prevent loss of dignity and embarrassment when eating in front of others. The aim of this review is to identify and evaluate the existing literature regarding the use and effectiveness of finger foods among adults in health and social care settings.

Methods: An integrative review methodology was used. A systematic search of electronic databases for published empirical research was undertaken in October 2018. Following screening of titles and abstracts, the full texts of publications, which investigated outcomes associated with the provision of finger foods in adult care settings, were retrieved and assessed for inclusion. Two independent investigators conducted data extraction and quality assessment using Critical Appraisal Skills Programme checklists. Thematic analysis was used to summarise the findings.

Results: Six studies met the inclusion criteria. Four themes were identified: Finger food menu implementation; Importance of a team approach; Effect on nutrition; and Influence on wellbeing. Study designs were poorly reported, with small sample sizes.

Conclusions: There is some evidence that the provision of finger foods may positively affect patient outcomes in long-term care settings. There is a paucity of research evaluating the use of a finger food menu in acute care settings, including economic evaluation. Future high quality trials are required.

Introduction

The ageing population living with multiple co-morbidities (e.g. dysphagia, stroke and dementia) is increasing ⁽¹⁾. Older people, particularly those living in residential care settings and those admitted to hospital, are at risk of reduced oral food intake and malnutrition ⁽²⁾. Being under-nourished can cause a loss of muscle mass and weakness, together with other physiological effects, including an increased susceptibility to infection and

delayed wound healing ⁽¹⁾. It can impact on mental wellbeing and lead to reduced quality of life as a result of an increased dependence on others ^(3,4). Malnutrition is associated with increased costs to the National Health Service (NHS) as a result of extended and more frequent hospital stays and multiple general practitioner visits ⁽⁵⁾.

Reduced food intake in institutional care can be the result of a number of complex factors, including the environment and the patient ⁽⁶⁾. Environmentally, staff shortages reducing access to mealtime assistance, limited

choice, unappealing food and mealtime interruptions can lead to a patient refusing food. Patient factors relating to eating difficulties can be associated with older age⁽⁷⁾, as well as specific diseases such as dementia or stroke. People with dementia experience change in cognition, which can cause difficulties in recognising food or cutlery, uncoordinated transfer of food from the plate to the mouth and distraction during the mealtime task⁽⁸⁾. People after stroke experience physical changes such as hemiparesis, limb apraxia or visual disturbances, which can cause difficulty in manipulating cutlery or transferring food from the plate to the mouth^(9–11), alongside embarrassment when eating in view of others⁽¹²⁾.

The need to improve food intake in care settings has been acknowledged internationally, resulting in the publication of guidelines⁽¹³⁾. Guidelines include various proposed strategies to improve intake in older adults and particularly adults with dementia; however, little is known about the effectiveness of these strategies with respect to improving oral intake^(14–16). Evidence-based recommendations for healthcare promote the provision of adequate support for people who are unable to eat independently⁽¹⁷⁾ and offering food that is appropriate for the person, using a food first approach⁽¹³⁾. Despite this, relatives of older people frequently report an inadequate amount of appropriate food and lack of support for people unable to feed themselves^(18,19). The European Society for Clinical Nutrition and Metabolism (ESPEN) clinical recommendations⁽²⁰⁾ suggest using finger foods for older adults because of their limited cost and low risk, although the supporting evidence for this intervention is sparse.

For the purpose of this integrative review, finger foods are defined as foods presented in a form that is easily picked up with the hands and transferred to the mouth without the need for cutlery. Finger foods are considered easier to eat because they do not require manipulation with cutlery⁽²¹⁾. Typically, a finger food menu includes small sandwiches, pieces of quiche, cut up vegetables and cake slices or foods presented in bite sized portions, for people managing regular textured foods⁽²²⁾.

The Social Care Institute for Excellence⁽²³⁾ recommends the use of finger foods to enable mealtime independence and to prevent any loss of dignity and embarrassment when eating in front of others⁽²⁴⁾. For people after stroke or with cognitive impairment, finger foods have the potential to support participation and to increase independence at mealtimes^(20,25,26). The potential benefits of using finger foods are an enhancement of nutritional intake and maintenance of weight^(20,27). Additionally, finger foods are described as a more flexible approach to dining⁽²⁸⁾. They can be used as a portable alternative to a plated meal and can be eaten 'on the go'⁽²¹⁾.

No previous high quality reviews have purposefully addressed the use of a finger food menu with older adults in care settings. NHS hospital trusts have implemented finger foods as part of a multimodal approach to nutritional intervention, without evidence showing that they singularly have a positive impact on patients⁽²⁹⁾. Locating and reviewing the literature to identify which finger foods are most appropriate and which groups would benefit, as well as the cost effectiveness of the intervention, would inform future research and support clinical practice, guiding decisions regarding resource allocation. Therefore, the aim of this review was to locate and synthesise empirical published literature on the use of finger foods in adults in care settings.

Materials and methods

An integrative review methodology allows the full understanding of a phenomenon⁽³⁰⁾. It supports the objective critique and summary of selected quantitative and qualitative research studies, as opposed to a systematic review, which addresses a distinctive clinical question and evaluates the effectiveness of an intervention⁽³¹⁾. This integrative review follows the five steps outlined by Souza *et al.*⁽³²⁾: definition of the guiding question, a detailed and systematic search of the literature, data extraction, critical analysis of included publications, and interpretation and synthesis of results.

Selection criteria

Eligible studies were selected through predefined inclusion criteria developed using the PICOST tool (Population, Intervention, Comparator, Outcome, Setting, Type)⁽³³⁾. Studies were included if (i) the sample population included adults aged 18 years or above; (ii) the study involved use of finger foods, including an increase in finger foods offered; (iii) any comparator was present, or none at all; (iv) any subsequent outcomes were used; (v) the study was conducted in any institutional setting (e.g. long-term care centres, assisted living residence, residential homes, nursing homes, acute hospital ward); and (vi) the study was an example of empirical research. Review publications were not included because the aim was to find empirical evidence (Table 1).

Search strategy

Databases were searched using a wide range of predefined search terms developed with the assistance of a medical librarian and combined using Boolean operators (And/Or/Near) and MeSH (Medical Subject Heading) terms. Full search terms are shown in Data S1. This aimed to

Table 1 PICOST table

Category	Criteria
Population	Adults, 18 years or over
Intervention	Any use of a finger food menu including an increase in finger foods offered or complete change of the menu
Comparator	Studies included if any comparator present, or none at all
Outcome	Any subsequent outcomes
Setting	Any institutional setting (e.g. long-term care centres, assisted living residence, residential homes, nursing homes, acute hospital ward)
Type	All types of empirical research included

retrieve the widest scope of publications possible across different platforms. In addition, reference lists of selected publications were searched. In attempt to review the most robust publications, the grey literature was not included in this search.

Databases searched to October 2018 included MEDLINE, EMBASE, CINAHL Plus® with Full Text (1937–2018), Psych INFO (1880–2018), Web of Science, Cochrane and Ahmed. No language restrictions were placed during the search. Search terms included: adult, patient, elderly, senior, geriatric, dementia, Alzheimer's, neurocognitive impairment, neurocognitive decline, finger food, buffet, utensil less, menu modification, mealtime intervention, dementia diet and eating with hands or fingers.

The inclusion criteria were used by two investigators (MH and NG) to screen title and then abstracts initially. Full texts of publications that appeared to be relevant were retrieved for further consideration by three investigators (MH, NG, SG).

Data extraction and quality

Selected publications were read multiple times to ensure familiarity. Data were extracted using a preprepared and piloted instrument based on the data extraction table by Souza *et al.*⁽³²⁾. Studies were appraised using the appropriate Critical Appraisal Skills Programme (CASP) tool for the study design⁽³⁴⁾. This tool supports systematic evaluation of published papers, considering validity, credibility, relevance and results of papers⁽³⁴⁾. The results of the CASP tool (Data S2) were discussed and agreed with multiple authors (SG and MH). None of the publications included met all of the criteria assessed by the CASP appraisal form. However, it was not possible to assess whether the publications omitted these key components or whether it was simply not reported by the authors, despite attempts to contact the authors.

Data synthesis

Primary data sources were coded, categorised and synthesised using a systematic approach in accordance with guidelines for preparing an integrative review⁽³⁰⁾. As a result of the small number of publications found, it was not necessary to subgroup papers. Initial codes were derived inductively from publications, using descriptive codes to simplify and sort data into manageable data forms. Next, these descriptive codes were displayed in a visual matrix to observe patterns and themes. Codes were analysed iteratively by clustering descriptive codes into overarching themes and comparing and contrasting codes. These overarching themes were discussed and agreed with the other authors. All relevant studies identified were included in the thematic analysis regardless of quality.

Results

Descriptive findings

Six publications were included in the final selection. Figure 1 summarises the selection process using the PRISMA (Preferred Reporting Items for Systematic review and Meta-Analysis) flow diagram, including reasons for exclusion.

Table 2 provides a summary of the publication characteristics. Publications reported studies undertaken in long-term care settings in the USA^(35,36), the UK^(24,27,37) and France⁽³⁸⁾. None described the use of a finger foods in acute care settings. Study designs varied, including observational studies^(36–38), a pilot study⁽³⁵⁾, a case study⁽²⁴⁾ and a retrospective study⁽²⁷⁾, although they did not include randomised controlled trials. Sample sizes were generally small, ranging from six participants⁽²⁴⁾ to 114 participants⁽³⁸⁾ using a range of outcome measures.

All participants included had a diagnosis of dementia or other psychiatric conditions. Participants presented with a range of physical and cognitive eating difficulties, which were attributed to their cognitive impairment. These included difficulties using utensils^(24,27,35–37); for example, poor hand or finger control, tremor and limited concentration or high level of distractibility⁽³⁶⁾.

Quality assessment

Assessment using the CASP case control critical appraisal tool indicated that two publications reporting quantitative findings were of low quality^(27,35). Soltesz and Dayton⁽²⁷⁾ used a control group that differed from the intervention group with respect to key characteristics. The control group comprised 11 residents consuming a modified pureed diet, and an intervention group of 43 residents with no swallowing difficulties eating a normal diet. In

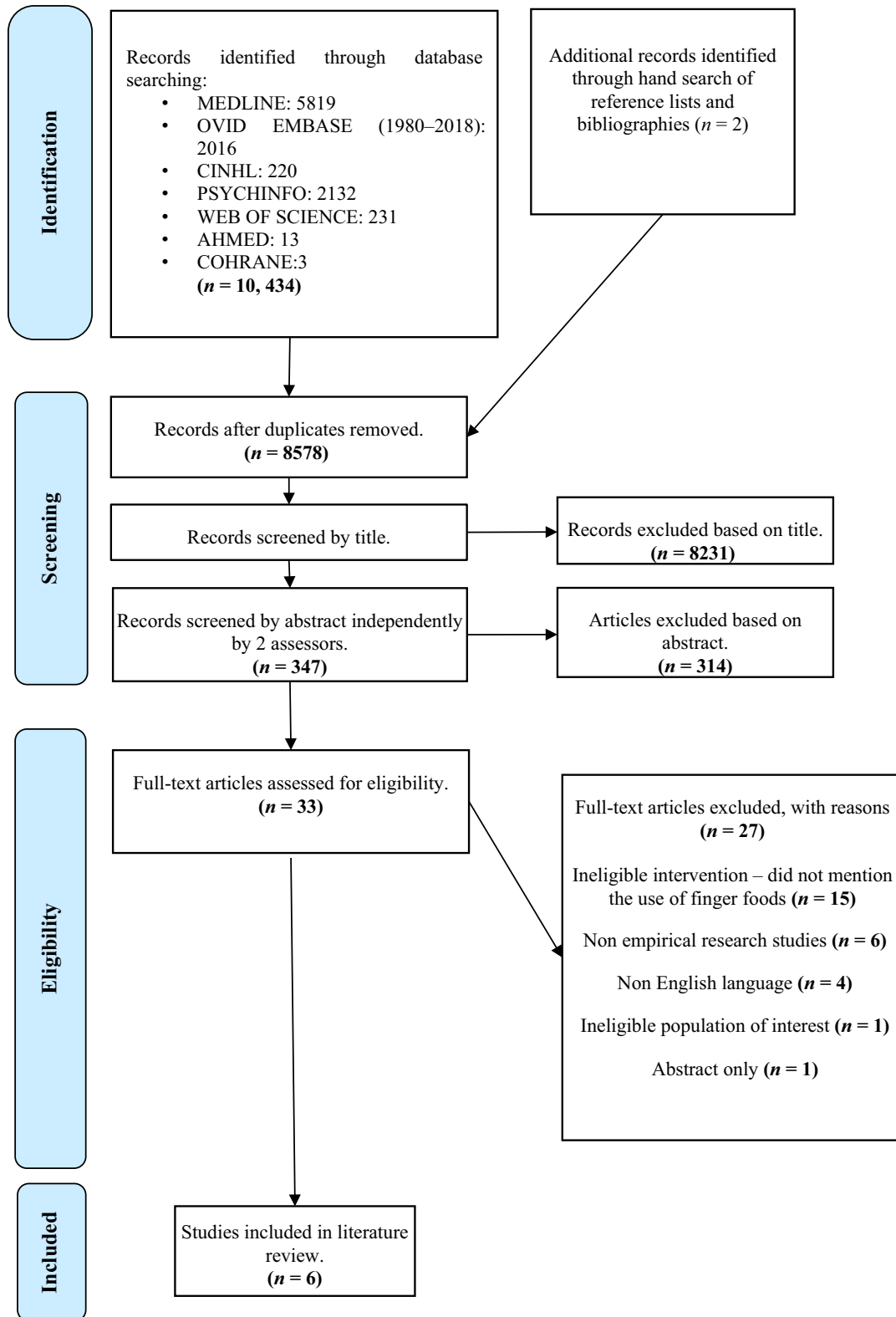


Figure 1 PRISMA flow diagram.

Table 2 Summary of publication characteristics

Author (Year)	Study design	Setting	Population (Number)	Intervention	Comparator	Outcomes	Assessment method	Results
Barratt <i>et al.</i> ⁽²⁴⁾ (2001)	Case study	Specialist care ward, UK	Females with dementia. (6)	Finger food menu	Standard menu	<ul style="list-style-type: none"> Well-being Independence Weight Costs 	Observations with dementia care mapping (DCM); Change in weight	<ul style="list-style-type: none"> 50% participants maintained weight, 33% gained weight, 17% lost weight. Mean weight gain of 0.4 kg with finger food menu DCMI scores showed 100% of participants increased levels of independence and well-being with finger food menu £1 increase in food costs per patient per week
Ford ⁽³⁷⁾ (1996)	Observational study	Behavioural health unit, UK	Psychiatric patients (10)	Finger food menu, five small meals per day.	No comparator	<ul style="list-style-type: none"> Food intake Weight Quality of Life (QOL) Self-esteem 	Observation methods (not fully described); Assessment of abnormal involuntary movement	<ul style="list-style-type: none"> 70% of participants increased food intake and gained weight. Mealtimes reported to be more enjoyable for 100% participants, families and staff Quality of life and self-esteem increased by helping patients gain some independence
Jean ⁽³⁵⁾ (1997)	Pilot study	2 nursing and rehabilitation homes, USA	6 with Alzheimer's Disease; 6 without Alzheimer's Disease (12)	Finger food menu	Standard menu	<ul style="list-style-type: none"> Weight Food intake Independent feeding skills 	Percentage of food eaten (0%, 25%, 50%, 75%, 100%); Scale to measure feeding dependence (dependent feeding, hand over hand cueing, independent with tray set up, total independence)	<ul style="list-style-type: none"> 83% of participants maintained or gained weight, 17% continued to lose weight post finger food menu introduction, 100% participants increased percentage of food eaten 100% participants became more independent with feeding skills 25% participants no longer required high calorie and protein supplements, suggesting cost savings
Nangeroni and Pierce ⁽³⁶⁾ (1985)	Observational study	Intermediate care unit for psychogeriatric patients, USA	Psychogeriatric patients (22),	Soft textured finger foods added to menu.	No control	<ul style="list-style-type: none"> Independence with feeding Quality of life 	Not described	<ul style="list-style-type: none"> Nil numerical results displayed. Generally positive responses and increased palatability with finger foods Participants enjoyed wider variety of food Less eating assistance required

Table 2 Continued

Author (Year)	Study design	Setting	Population (Number)	Intervention	Comparator	Outcomes	Assessment method	Results
Pouyet <i>et al.</i> (38) (2014)	Observational study	8 Nursing homes, France	Residents with Alzheimer's dementia (114)	Puree finger food, with and without sauce, 1 or 2 layers and 2 different shapes.	Puree finger food with different presentations	<ul style="list-style-type: none"> Participant choice 	Staff questionnaire	<ul style="list-style-type: none"> No conclusions regarding effectiveness of finger food 92% participants trialled finger foods, suggesting well accepted Finger foods with sauce and visual contrast chosen first Shape of finger food was not shown to exert an influence on food attractiveness, but were relatively easy to pick up, appealing and attractive for the subjects who participated in this study
Soltész and Dayton (27) (1995)	Retrospective study	Alzheimer's care centre facility, USA	Residents with Alzheimer's dementia (54)	Increased number of finger foods on solid food menu for 6 months	Puree diet	<ul style="list-style-type: none"> Weight food intake for breakfast, lunch and dinner calculated from % consumed 	Mean weight over one month for each resident; % of meal consumed recorded in medical chart	<ul style="list-style-type: none"> No significant change in weight for control and intervention group. Significant increase ($P < 0.05$) in food consumption overall and for some meals with intervention Anecdotal report that 'generally cost no more money', staff, retraining or food purchase

addition, confidence intervals were not provided for key outcomes, giving no indication of variability⁽²⁷⁾.

In the study by Jean⁽³⁵⁾, participants acted as their own control groups, in a pre–post study design. No confounding factors were reported, making it difficult to attribute maintenance or increase in weight to the finger food menu intervention⁽³⁵⁾. Additionally, Jean⁽³⁵⁾ presented results using only descriptive statistics, which makes it difficult to generalise the results and places external validity at risk. Based on the CASP case control checklist, the study by Pouyet *et al.*⁽³⁸⁾ satisfied most of the criteria of the three studies, although it was the only study of its kind reporting on attractiveness of pureed finger foods, limiting the external validity.

The studies employing a qualitative methodology were assessed as low quality^(24,36,37). Ford⁽³⁷⁾ did not report sufficient detail of the study methodology or findings. Barratt *et al.*⁽²⁴⁾ and Nangeroni and Pierce⁽³⁶⁾ did not adequately consider the researcher and participant relationship, or ethical considerations, and included unclear statements relating to findings and credibility. The limited information regarding the recruitment strategy, or the reasons for the populations recruited, makes it difficult to establish a target sample for all of the studies.

Meta synthesis

Four main themes were identified inductively through thematic analysis: (i) Finger food menu implementation; (ii) Importance of a team approach; (iii) Effect on nutrition; and (vi) Influence on well-being.

Finger food menu implementation

Included publications defined finger foods as food that did not require cutlery^(27,37,38), or could be eaten easily with the hands^(24,36,38). Generally, finger foods offered were considered appropriate for residents eating normal or regular textured foods⁽²²⁾ and with no evidence of oropharyngeal dysphagia. However, Barratt *et al.*⁽²⁴⁾, Nangeroni and Pierce⁽³⁶⁾ and Pouyet *et al.*⁽³⁸⁾ used softer foods and pureed forms of finger food⁽³⁸⁾ to support older people with dysphagia or difficulties chewing. Pouyet *et al.*⁽³⁸⁾ showed that the pureed finger foods were generally well accepted by adults with Alzheimer's disease, with reports that shape was not an influence on food attractiveness⁽³⁸⁾. However, Pouyet *et al.*⁽³⁸⁾ did consider shape as being important to support manipulation with the hands.

Details of the specific implementation of a finger foods varied. In two studies, finger foods were offered alongside the normal menu to increase variety of food offered^(27,36). Soltesz and Dayton⁽²⁷⁾ added extra finger foods to the existing menu; however, the overall number of finger

foods increased minimally, leading to difficulties comparing the control and intervention group. This contrasts with other publications, where a finger food menu was developed to replace the standard menu offered over lunch and dinner times^(24,35), or offered as smaller, more frequent meals⁽³⁷⁾. None of the publications reported difficulties with intervention fidelity and suggested that no additional staff or additional food items were required⁽²⁷⁾. Success with using finger foods was supported using simple and easy foods for staff to make⁽³⁵⁾.

The cost of implementing a finger food menu was considered by Barratt *et al.*⁽²⁴⁾, Soltesz and Dayton⁽²⁷⁾ and Jean⁽³⁵⁾. However, none reported a robust economic evaluation, resulting in conflicting results. Soltesz and Dayton⁽²⁷⁾ suggested that the implementation of a finger food menu cost no more than the provision of standard foods and Jean⁽³⁵⁾ suggested that high energy and protein supplements were discontinued in 25% of participants receiving a finger food menu, giving a cost saving. Conversely, in a later study Barratt *et al.*⁽²⁴⁾ described an increase on cost per person to implement the finger food menu.

Importance of a team approach

Collaboration between clinical and catering teams to support the provision of a finger food menu was a common theme arising in three studies^(24,27,35). Despite catering services often being perceived as nonclinical services, their involvement ensured that food was presented in such a way that patients could access allowed observable changes in clinical outcomes⁽²⁴⁾. In studies showing increased costs for providing finger foods, agreement between budget holders (often clinical managers, commissioning services and catering teams) is required to justify the need for this intervention⁽²⁴⁾. Staff training in understanding the need and rationale of finger foods was one approach influencing the maintenance and success of implementing the intervention across departments^(35,37).

Barratt *et al.*⁽²⁴⁾ and Soltesz and Dayton⁽²⁷⁾ described collating feedback from the clinical and catering team to support the development and implementation of the finger food; however, little detail was given about the changes made and how these data were collected.

Effect on nutrition

Nutritional outcomes were measured in only three studies by assessing food intake via food chart reviews, plate waste observations and changes in weight^(35,37). Increased nutritional intake and weight maintenance during the finger food menu intervention period were demonstrated in all three studies^(27,35,37). A full description of the menu offered with nutritional values was not provided; therefore, although there was an increase in weight of food

consumed, the nutritional value of the foods eaten could not be evaluated. Ford⁽³⁷⁾ suggested that changes in nutritional status could affect medical status; however, an explanation regarding how medical status will change was not included.

Influence on well-being

The fourth theme describes the improvement in well-being during the implementation of finger foods, which was reported in all publications. Well-being was measured formally by Barratt *et al.*⁽²⁴⁾ using dementia care mapping. Barratt *et al.*⁽²⁴⁾ demonstrated an increase in the mean well-being scores of residents who were offered a finger food menu that was maintained 6 weeks after the introduction. However, the small sample size used by Barratt *et al.*⁽²⁴⁾ and the pre–post study design limits the control of confounding variables in the complex long-term care setting and makes it difficult to attribute these findings wholly to the food offered.

An increased independence with eating for people choosing to eat finger foods was described in three studies^(24,35,36), despite variation in the outcome measures used. Barratt *et al.*⁽²⁴⁾ observed an increase in the mean percentage of observations recorded as ‘independent feeding’ over lunchtime meals. This contrasts to Jean⁽³⁵⁾ who created a scale demonstrating that three of 12 residents became fully independent eating their meal when offered finger foods, despite being fully dependent with feeding during the baseline measure. Nangeroni and Pierce⁽³⁶⁾ did not provide details of how independence was measured. Within these studies, the blinding or reflexive views of the researcher were not described, which increases the risk of bias and makes it difficult to distinguish whether this would lead to a reduced requirement for support by staff and visitors⁽²⁴⁾.

Discussion

The aim of this integrative review was to locate and synthesise empirical published literature on the use of finger foods for adults in care settings, aiming to inform future research and to support clinical practice and policy decisions.

The lack of high quality trials identified suggests that the use of a finger foods with adults is yet to be robustly evaluated. There is some evidence to demonstrate an improvement in relevant outcomes, such as food intake, although this was shown in studies that lacked a control, making it difficult to ascertain the cause of the effect shown. The variation in interventions provided across these publications provides additional challenges when comparing outcomes. However, this does highlight the need for a pragmatic approach to future research, considering all of the stakeholders involved. A study by Cluskey and Kim⁽³⁹⁾ undertaken in the USA suggested that finger

foods are judged by healthcare professionals, working in long-term care settings, as being beneficial for residents, cheap and easily implemented in institutions. The limited adverse effects and expense to provide these types of foods means that their use continues to remain in clinical guidelines on nutrition and hydration in geriatrics⁽²⁰⁾.

Despite guidelines suggesting that finger foods could be used to support people with other conditions, such as stroke⁽²⁵⁾, all studies focussed on people with cognitive impairment. Ford⁽³⁷⁾ acknowledged the potential of using a finger food menu to support older adults with a wide range of eating difficulties, including mental health or physical difficulties. An increase in food intake in people with cognitive impairment has been shown in other studies with different presentations of food. In a cross-over, randomised controlled trial undertaken in a nursing home, Young *et al.*⁽⁴⁰⁾ demonstrated an increased energy intake when high carbohydrate foods were offered in place of a usual meal, which was not fully described. Although that study did not aim to evaluate the use of finger foods, it was noted that many of the high carbohydrate foods could be defined as finger foods, such as bread with jam, hard-boiled egg, muffins and slices of cheese. In addition, a greater severity of cognitive deficit and atypical motor behaviour was associated with greater intervention success⁽⁴⁰⁾. Young *et al.*⁽⁴⁰⁾ acknowledged that, in this trial, people with nutritionally controlled diabetes were excluded. This highlights that the suitability for a finger food diet would need to be assessed individually because the nutritional content and presentation may not meet some people’s dietary needs.

None of the research studies in this integrative review conducted a well described economic evaluation to assess the benefits of individual interventions and to evaluate the best use of available resources alongside highest patient satisfaction⁽⁴¹⁾. It is important to reflect the true direct and indirect costs of healthcare interventions, particularly when implementing a change in practice⁽⁴²⁾.

Interestingly, none of the studies in this review included or explored the views of staff, carers or the recipient of the finger foods, despite suggestions that they may have positive benefits on quality of life and well-being. A conference abstract, with no associated published paper, was identified that used a survey methodology to explore the experiences of residents, caregivers and relatives with respect to providing a finger food menu in a nursing home⁽⁴³⁾. It appears that further in depth research investigating the experience of residents, caregivers and relatives could give further information on the acceptability of this menu⁽⁴³⁾ to support effective and efficient service delivery⁽⁴⁴⁾.

The findings of this review are in agreement with broader reviews on nutritional interventions. Abdelhamid

et al.⁽⁴⁵⁾ and Malerba *et al.*⁽⁴⁶⁾ suggest positive outcomes for the use of finger foods, although there is a further need for high quality investigations and well powered randomised control trials. The review by Abdelhamid *et al.*⁽⁴⁵⁾ focussed on interventions to support food intake in people with dementia and included two studies that classified the use of finger foods as a direct dietary intervention^(27,35). Addressing the use of multiple dietary interventions meant that the review did not focus specifically on the use of finger foods and limited the range of publications found. However, two studies⁽²⁷⁾ were also included in this integrative review and, interestingly, no studies published later than 2016 were found. The descriptive review by Malerba *et al.*⁽⁴⁶⁾, in France, commented on the use of finger foods for people with dementia in community and home settings. Malerba *et al.*⁽⁴⁶⁾ suggests beneficial outcomes relating to the use of finger foods; for example, a reduced workload of carers, increased independence and individualised care for people with dementia. Despite useful results, the review did not show a systematic approach to searching the literature or provided a quality critique of the publications included.

Strengths and limitations to integrative review

The range of study designs included in the present review, as well as the synthesis of quantitative and qualitative data, adds a level of complexity to the review and therefore can introduce bias⁽³⁰⁾. To ensure the quality of this review, rigorous systematic approaches were used throughout. To reduce bias, two reviewers (MH and NG) screened 347 abstracts for inclusion and discrepancies were dealt with through discussion. The full texts were chosen following discussion with the other investigating authors.

Conclusions and future recommendations

The findings obtained suggest that the use of finger foods may increase nutritional intake and enhance independence and well-being for adults with cognitive impairment in long-term care settings. However, the low quality of the studies included does not provide robust evidence for the effectiveness of using these types of foods in care settings. Therefore, the results should be interpreted with caution.

The review highlights key considerations with respect to implementing a finger food menu within care settings, as well as a particular need to focus on the use of this menu in hospital settings. Further research is required to determine whether this intervention is cost effective, feasible and acceptable to be used in acute care settings for older adults.

Acknowledgments

The authors thank Vicky Fenerty, Research Engagement Librarian, for her support in developing the search strategy.

Conflict of interests, source of funding and authorship

The authors declare that they have no conflicts of interest.

This review was completed as part of a clinical doctoral research fellowship for the first author (MH). The research fellowship is funded through an educational grant by Medirest, a division of Compass Group UK and Ireland; however, this company had no input with respect to the study design or analysis.

MH, SG, HR and CB were involved in the design of the review, developing the protocol extracting and appraising study methodologies. MH and NG had a lead role in identifying articles for inclusion. MH, SG, JP, HR and CB contributed to data analysis and the drafting of the paper. All authors named in the paper agreed on the final version of the manuscript submitted for publication.

Transparency declaration

The lead author affirms that this manuscript is an honest, accurate and transparent account of the study being reported. The reporting of this work is compliant with PRISMA3 guidelines. The lead author affirms that no important aspects of the study have been omitted and that any discrepancies from the study as planned have been explained.

References

1. Wilson L (2017) Malnutrition task force: State of the Nation. Available at: http://www.malnutritiontaskforce.org.uk/wp-content/uploads/2017/10/AW-5625-Age-UK-MTF_Report.pdf (accessed 27/03/2019).
2. Elia M (2015) *The Cost of Malnutrition in England and Potential Cost Savings from Nutritional Interventions*. Redditch: British Association for Parenteral and Enteral Nutrition.
3. Altman KW, Yu G-P & Schaefer SD (2010) Consequence of dysphagia in the hospitalized patient: impact on prognosis and hospital resources. *Arch Otolaryngol Head Neck Surg* **136**, 784–789.
4. Forsey A (2018) Hidden hunger and malnutrition in the elderly. Available at: <http://www.frankfield.co.uk/upload/docs/Hidden%20hunger%20and%20malnutrition%20in%20the%20elderly.pdf> (accessed 04/06/18).
5. Guest JF, Panca M, Baeyens J-P *et al.* (2011) Health economic impact of managing patients following a community-based diagnosis of malnutrition in the UK. *Clin Nutr* **30**, 422–429.
6. Mudge AM, Ross LJ, Young AM *et al.* (2011) Helping understand nutritional gaps in the elderly (HUNGER): a

- prospective study of patient factors associated with inadequate nutritional intake in older medical inpatients. *Clin Nutr* **30**, 320–325.
7. Pu D, Murry T, Wong MC *et al.* (2017) Indicators of dysphagia in aged care facilities. *J Speech Lang Hear Res* **60**, 2416–2426.
 8. Chang CC & Roberts BL (2008) Feeding difficulty in older adults with dementia. *J Clin Nurs* **17**, 2266–2274.
 9. Intercollegiate Stroke Working Party (2016) National clinical guideline for stroke. Available at: [https://www.strokeaudit.org/SupportFiles/Documents/Guidelines/2016-National-Clinical-Guideline-for-Stroke-5t-\(1\).aspx](https://www.strokeaudit.org/SupportFiles/Documents/Guidelines/2016-National-Clinical-Guideline-for-Stroke-5t-(1).aspx) (accessed 15/06/18).
 10. Poels B, Brinkman-Zijlker H, Dijkstra P *et al.* (2006) Malnutrition, eating difficulties and feeding dependence in a stroke rehabilitation centre. *Disabil Rehabil* **28**, 637–643.
 11. Wu H-S & Lin L-C (2015) Comparing cognition, mealtime performance, and nutritional status in people with dementia with or without ideational apraxia. *Biol Res Nurs* **17**, 199–206.
 12. Medin J, Larson J, Von Arbin M *et al.* (2010) Striving for control in eating situations after stroke. *Scand J Caring Sci* **24**, 772–780.
 13. Department of Health, Jeffrey D (2014) The Hospital Food Standards Panel's report on standards for food and drink in NHS hospitals. Available at: https://assets.publishing.service.gov.uk/government/uploads/system/uploads/attachment_data/file/523049/Hospital_Food_Panel_May_2016.pdf (accessed 24/05/19).
 14. Council of Europe (2003) Resolution ResAP (2003) 3 on food and nutritional care in hospitals. Available at: <https://wcd.coe.int/ViewDoc.jsp?xml:id=85747> (accessed 10/04/2019).
 15. Herke M, Fink A, Langer G *et al.* (2018) Environmental and behavioural modifications for improving food and fluid intake in people with dementia. *Cochrane Database Syst Rev* **7**, CD011542.
 16. Liu W, Cheon J & Thomas SA (2014) Interventions on mealtime difficulties in older adults with dementia: a systematic review. *Int J Nurs Stud* **51**, 14–27.
 17. NICE (2017) Nutrition support for adults: oral nutrition support, enteral tube feeding and parenteral nutrition. Available at: <https://www.nice.org.uk/guidance/cg32> (accessed 18/11/17).
 18. Age Concern (2006) *Hungry to be Heard. The Scandal of Malnourished Older People in Hospital*. England: The Social Care Institute for Excellence.
 19. Care Quality Commission (CQC) (2015) Adult inpatient surveys. Available at: <http://nhssurveys.org/surveys/425> (accessed 12/11/18).
 20. Volkert D, Beck AM, Cederholm T *et al.* (2019) ESPEN guideline on clinical nutrition and hydration in geriatrics. *Clin Nutr* **38**, 10–47.
 21. Crawley H & Hocking E (2011) *Eating Well: Supporting Older People and Older People with Dementia. A Practical guide*. Hertfordshire: The Caroline Walker Trust.
 22. IDDSI Committee (2016) IDDSI Framework evidence statement. Available at: <http://iddsi.org/Documents/IDDSIFramework-EvidenceStatement.pdf> (accessed 08/05/18).
 23. The Social Care Institute for Excellence (2009) *Nutritional Care and Hydration*. Great Britain: Social Care Institute for Excellence.
 24. Barratt J, Gatt J, Greatorex' B *et al.* (2001) Using finger food to promote independence, well being and good nutrition in people with dementia. *PSIGE Newsllett* **77**, 26–31.
 25. BDA Association of UK Dieticians (2017) The nutritional and hydration digest. Improving outcomes through food and beverage services. Available at: <https://www.bda.uk.com/publications/professional/NutritionHydrationDigest.pdf> (accessed, 24/05/19)
 26. Volkert D, Chourdakis M, Faxen-Irving G *et al.* (2015) ESPEN guidelines on nutrition in dementia. *Clin Nutr* **34**, 1052–1073.
 27. Soltesz KS & Dayton JH (1995) The effects of menu modification to increase dietary intake and maintain the weight of Alzheimer residents. *Am J Alzheimers Dis* **10**, 20–23.
 28. Burbidge D (2013) Food for thought: Facilitating independence with finger foods. Available at: http://journalofdementiacare.com/dementia_finger_foods_menu/ (accessed 18/09/17).
 29. James J, Gilby L & Pettit J (2017) Transforming nutrition and hydration for people with dementia in hospital - The NOSH project at Imperial College Healthcare NHS Trust. *Age Ageing* **46**, i1–i22.
 30. Whittemore R & Knafk K (2005) The integrative review: updated methodology. *J Adv Nurs* **52**, 546–553.
 31. Noble H & Smith J (2018) Reviewing the literature: choosing a review design. *Evid Based Nurs* **21**, 39–41.
 32. Souza MTD, Silva MDD & Carvalho RD (2010) Integrative review: what is it? How to do it? *Einstein (São Paulo)* **8**, 102–106.
 33. Aveyard H (2010) *Doing a Literature Review in Health and Social Care: A Practical Guide*. England: McGraw-Hill.
 34. CASP (2017) CASP Checklists. Available at: <http://www.casp-uk.net/casp-tools-checklists> (accessed 19/09/17).
 35. Jean L (1997) "Finger food menu" restores independence in dining. *Health Care Food Nutr Focus* **14**, 4–6.
 36. Nangeroni J & Pierce P (1985) A geriatric nutrition program designed to improve independence in feeding. *Hosp Community Psychiatry* **36**, 666–667.
 37. Ford G (1996) Putting feeding back into the hands of patients... 'utensil-less' diets for patients with acute psychiatric conditions. *J Psychosoc Nurs Ment Health Serv* **34**, 35–39.
 38. Pouyet V, Giboreau A, Benattar L *et al.* (2014) Attractiveness and consumption of finger foods in elderly Alzheimer's disease patients. *Food Qual Prefer* **34**, 62–69.

39. Cluskey M & Kim YK (2001) Use and perceived effectiveness of strategies for enhancing food and nutrient intakes among elderly persons in long-term care. *J Am Diet Assoc* **101**, 111–114.
40. Young KW, Greenwood CE, Van Reekum R *et al.* (2005) A randomized, crossover trial of high-carbohydrate foods in nursing home residents with Alzheimer's disease: associations among intervention response, body mass index, and behavioral and cognitive function. *J Gerontol A Biol Sci Med Sci* **60**, 1039–1045.
41. Brent RJ (2004) *Cost-Benefit Analysis and Health Care Evaluations*. Cheltenham, UK: Edward Elgar Publishing.
42. McMahon A & Sin C (2013) Introduction to economic assessment. *Nurs Manage* **20**, 32–38.
43. Tuinier K, Westrate W, Hoogendoorn *et al.* (2014) Finger food. Intervention for patients with eating difficulties. In 43rd Biennial Convention Las Vegas, Nevada, USA STTI.
44. Collins J, Huggins CE, Porter J *et al.* (2017) Factors influencing hospital foodservice staff's capacity to deliver a nutrition intervention. *Nutr Diet* **74**, 129–137.
45. Abdelhamid A, Bunn D, Copley M *et al.* (2016) Effectiveness of interventions to directly support food and drink intake in people with dementia: systematic review and meta-analysis. *BMC Geriatr* **16**, 26.
46. Malerba G, Pop A, Rivasseau-Jonveaux T *et al.* (2015) Feeding a patient with neurocognitive impairment in the hospital and at home? Convenience of finger-food. *Nutrition Clinique et Metabolisme* **29**, 197–201.

Supporting information


Additional supporting information may be found online in the Supporting Information section at the end of the article.

Data S1. Search strategy for CINAHL Plus with Full Text (Cumulative Index of Nursing and Allied Health Literature).

Data S2. CASP checklist questions with comments.

INFANTS AND CHILDREN

Retinol status and associated factors in mother–newborn pairs

D.S. Bezerra,¹  K.D.S. Ribeiro,² M.S.R. Lima,³ J.F. Pires Medeiros,³ A.G.C.L. da Silva,² R. Dimenstein³ & M.M. Osório⁴

¹Department of Nutrition, School of Health Sciences of Trairi – Federal University of Rio Grande do Norte, Santa Cruz, Rio Grande do Norte, Brazil

²Department of Nutrition – Federal University of Rio Grande do Norte, Natal, Rio Grande do Norte, Brazil

³Department of Biochemistry, Bioscience Centre – Federal University of Rio Grande do Norte, Natal, Rio Grande do Norte, Brazil

⁴Program in Nutrition – Federal University of Pernambuco, Recife, Pernambuco, Brazil

Keywords

vitamin A, serum, umbilical cord, colostrum, high-performance liquid chromatography.

Correspondence

D. S. Bezerra, Department of Nutrition, School of Health Sciences of Trairi – Federal University of Rio Grande do Norte, St Teodorico Bezerra, 2-122, Santa Cruz, Rio Grande do Norte, 59200-000 Brazil.

Tel.: +55 84 32912411

Fax: +55 84 20201657

E-mails: dani_querubim@yahoo.com.br; daniellesbezerra@yahoo.com.br

How to cite this article

Bezerra D.S., Ribeiro K.D.S., Lima M.S.R., Pires Medeiros J.F., da Silva A.G.C.L., Dimenstein R. & Osório M.M. (2020) Retinol status and associated factors in mother–newborn pairs. *J Hum Nutr Diet.* **33**, 222–231

<https://doi.org/10.1111/jhn.12707>

Abstract

Background: Vitamin A deficiency is still considered to be a nutritional problem during pregnancy, lactation and early childhood. The present study aimed to assess the vitamin A status of women and their newborns in the Brazilian Northeast and to determine the association between retinol in the maternal serum, umbilical cord blood and colostrum.

Methods: Vitamin A status in 65 pairs of women and newborns was assessed from samples of the mother's serum, umbilical cord serum and colostrum using high-performance liquid chromatography. The inadequacy of the vitamin A status of mothers and infants was identified if the retinol values were $<0.7 \mu\text{mol L}^{-1}$ in maternal serum or umbilical cord blood or $<1.05 \mu\text{mol L}^{-1}$ in colostrum.

Results: The prevalence of inadequate maternal vitamin A status was 21.5% (95% CI: 11.5%–31.5%) and 13.8% [95% confidence interval (CI) = 5.4%–22.2%] based on maternal serum and colostrum, respectively. Among newborns, 41.5% (95% CI = 29.3%–53.5%) presented a low status of vitamin A based on cord serum. Multiple linear regression analysis identified that maternal serum retinol is a predictor of umbilical cord retinol ($P = 0.005$). Retinol in maternal serum was lower in mothers who were less educated ($P = 0.04$) and colostrum retinol was higher in older ($P = 0.04$) and multiparous ($P = 0.002$) mothers.

Conclusions: Vitamin A deficiency is a common problem among mothers attended in public hospitals in Northeast Brazil and maternal retinol concentrations are associated with retinol status in newborns. Maternal age, parity and educational level were related to the maternal vitamin A status.

Introduction

Vitamin A (VA) plays essential but distinct roles regarding the functions it performs, including the vision, normal cell differentiation, embryonic growth and development, skeletal growth, immune function, and reproduction⁽¹⁾. As a result of the importance of VA to human health, vitamin A deficiency (VAD) in pregnancy and early childhood is a concern worldwide, especially in developing societies such as Brazil. Globally, estimates

based on the serum retinol indicator suggest that, in the period 1995–2005, VAD affected 33.3% (190 million) of children below the age of 5 years, and that approximately 15.3% (19.1 million) of pregnant women also had VA concentrations considered deficient⁽²⁾.

In Brazil, VAD is considered to be a moderate public health problem⁽²⁾. Children aged under 5 years (17.4%) and women (12.3%) have low serum VA levels ($<0.70 \mu\text{mol L}^{-1}$). The highest prevalence of low VA levels in children under 5 years was found in the

Northeast (19.0%) and Southeast (21.6%) regions of the country, although there were no statistical differences between macroregions when evaluating women⁽³⁾. In the Brazilian Northeast context, many studies^(4–9) have reported a high probability of this kind of deficiency occurring among women attending public hospitals in this region, which suggests that DVA can result in serious prejudice for the health of women and their babies in the initial days after childbirth. Faced with these circumstances, it is important that studies assess the VA status of populations at risk of developing VAD, such as those in the Brazilian Northeast. Accordingly, the present study aimed to analyse the vitamin A nutritional status in women and their newborns in the Brazilian Northeast from different perspectives (maternal serum, umbilical cord serum and colostrum milk), identify sociodemographic and obstetric risk factors, and determine possible associations between variables.

Materials and methods

Population and study characterisation

The present study is a cross-sectional study conducted with pairs of mothers and children delivered at a children's hospital: Ana Bezerra University Hospital, located in Santa Cruz municipality – Rio Grande do Norte (RN), Brazilian Northeast. This municipality is located 115 km from Natal, capital of the State of RN. The territorial area of Santa Cruz is 624.36 km², with an estimated population of 39 355 inhabitants, and a demographic density of 57.33 inhabitants per km². The population lives predominantly in the urban area and the Municipal Human Development Index is 0.635⁽¹⁰⁾.

Sample calculation

To calculate the sample size, the total number of individuals required to obtain a reliable estimate of the population mean was used. The basic parameters comprised the critical value that corresponds to the desired confidence level, the expected SD for the variable studied and the maximum permissible error.

Assuming that SDs for retinol in the maternal milk (colostrum), in the maternal serum and in the serum of the newborn (umbilical cord) would not exceed 1.01 $\mu\text{mol L}^{-1}$ ⁽¹¹⁾, 0.38 $\mu\text{mol L}^{-1}$ ⁽¹¹⁾ and 0.93 $\mu\text{mol L}^{-1}$ ⁽¹¹⁾, respectively, it therefore would be necessary to recruit at least 63 mother–children pairs because this was the number estimated to meet the breast milk retinol variable, substrate with greater variability between sample types. The maximum estimated error would be 0.25 $\mu\text{mol L}^{-1}$ at a significance level of 5%.

Selection of participants

The selection of participants (year 2014) of the study was carried out at the moment of their admission for delivery, according to the spontaneous demand of the maternity hospital. The inclusion criteria adopted were: women aged 18–40 years of low obstetrical risk. As exclusion criteria for the mothers, the parameters considered were: women who had multiple fetuses or used vitamin supplements containing VA during pregnancy or did not have colostrum ejection at the time of collection. Regarding the children, premature babies (less than 37 weeks) and their respective mothers were not included as part of the survey.

Biochemical evaluation of retinol

To determine the serum concentrations of retinol, a 5-mL blood sample was drawn from the parturients via brachial venipuncture at the moment women were being prepared for the delivery (i.e. when collecting samples for Venereal Disease Research Laboratory and HIV tests). The neonates then underwent umbilical cord blood milking, immediately after delivery. After being collected, blood samples were centrifuged (at 1073 x *g* per 10 min) to separate and extract the serum.

An aliquot (1 mL) of the colostrum was collected in the first morning after delivery, after a night fasting, by manual pumping of one single breast, at time intervals of at least 2 h between the last breastfeed and the sample collection.

All samples (mother's serum, umbilical cord serum and colostrum) were stored in polypropylene tubes after being sterilised and covered with aluminum paper for protection against the light. Then, the samples were frozen at $-20\text{ }^{\circ}\text{C}$ and shipped once a week to the Food and Nutrition Biochemistry Lab of the Federal University of Rio Grande do Norte, in the city of Natal-RN.

For the analysis of retinol, the serum samples were extracted by adapting the method described by Ortega *et al.*⁽¹²⁾. For 1 mL of serum, 1 mL of ethanol 95% (Vetec; Sigma-Aldrich, St Louis, MO, USA) was used to precipitate the proteins, followed by three steps of extraction with 2 mL of hexane (Merck, Kenilworth, NJ, USA) added in each step. After adding hexane each time, the samples were agitated for 1 min and centrifuged at 1073 x *g* for 10 min. Then, the resulting hexane sample was removed to another tube. An aliquot of 3 mL of the hexane phase was evaporated under a nitrogen atmosphere in water bath at 37 $^{\circ}\text{C}$. To extract retinol from the colostrum, a saponification step was added to the above-mentioned method with 1 mL of potassium hydroxide 50% v/v (Vetec)⁽¹³⁾.

After this process, all extracts resulting from samples of serum and colostrum were dissolved again in 500 μL of methanol (Vetec) with purity grade for high-performance liquid chromatography and 20 μL was applied to the chromatographer (Shimadzu Corp., Kyoto, Japan) to quantify the existing retinol. The mobile phase used for the analysis of retinol was methanol at 100% in an isocratic system, with a 1 mL min^{-1} flow and a retention time of 4.2 min. The wavelength adopted for the monitoring of absorbance was 325 nm. The identification and quantification of retinol in the samples were established by comparing the respective peak area obtained in the chromatogram with the retinol standard area (Sigma-Aldrich). The concentrations of the standards were confirmed by the extinction coefficient specific to retinol in absolute ethanol (ϵ 1%, 1 $\text{cm} = 1780$ at 325 nm).

The retinol found in the colostrum, in the maternal serum and in the umbilical cords of the newborns is presented in $\mu\text{mol L}^{-1}$ and is used to express the VA nutritional status of the mothers and neonates. It is known that colostrum has more retinol than mature milk⁽¹⁴⁾; however, there is no defined cut-off point to identify an inadequate status of vitamin A in the colostrum. Accordingly, it was assumed that the retinol concentrations in colostrum lower than the cut-off point defined for mature milk (1.05 $\mu\text{mol L}^{-1}$)⁽¹⁵⁾ would indicate sub-clinical vitamin A deficiency (DVA). When considering the maternal serum, retinol concentrations less than 0.7 $\mu\text{mol L}^{-1}$ were considered indicative of VAD^(16,17). Regarding the serum of the umbilical cords of newborns, retinol concentrations less than 0.7 $\mu\text{mol L}^{-1}$ were classified as indicating a low status⁽¹⁸⁾.

Variables of interest

To characterise the sample, sociodemographic information on the women's prenatal period, delivery and clinical history was obtained by interviewing the puerperae, in addition to information collected from their hospital medical records and also from their prenatal follow-up cards.

Among the variables possibly related to the immediate causes of the nutritional status of the studied group, the details included were biological/obstetric (anthropometric gestational nutritional status and parity) and sociodemographic information (mother's age range, number of individuals per household, mother's education level and family income).

The classification of the women's gestational anthropometric status was made according to the body mass index for the gestational ages, as proposed by Atalah *et al.*⁽¹⁹⁾ and adopted by the Brazilian Ministry of Health⁽²⁰⁾. The anthropometric assessment related to their pregnancy was

based on the information of their last antenatal care visit. For anthropometric classification of the neonates, those who weighed less than 2500 g were considered to be in a low weight condition at birth⁽²¹⁾. Cases of maternal anaemia during pregnancy were identified from haemoglobin ($<11 \text{ g dL}^{-1}$) and haematocrit ($<33\%$) results⁽²²⁾. All of the collected information was registered in questionnaires and precoded and pretested forms.

Statistical analysis

SPSS, version 13.0 (SPSS Inc., Chicago, IL, USA) was used for the statistical analysis. The variables were submitted to Kolmogorov–Smirnov's test to evaluate their coupling to the normal distribution. For the symmetrical distribution variables, the results are presented as the mean (SD); in the cases of asymmetrical distribution, the results are presented as the median (interquartile range).

To evaluate the relationships between retinol concentrations, multiple linear regression analysis with a hierarchical selection of variables was used, for which the theoretical model was constructed using the variables that presented biological plausibility (dependent variable: retinol concentration in umbilical cord serum and independent variables: retinol in maternal serum and retinol in colostrum). In addition, comparison tests were performed for the retinol averages between groups of variables set for the sociodemographic and obstetric conditions of the mother-child group. For this purpose, Student's *t*-tests and analysis of variance or the nonparametric corresponding Mann–Whitney and Kruskal–Wallis test, respectively, were used. In all cases, $P < 0.05$ was considered statistically significant.

Ethical approval

All procedures performed in the present study involving human participants were conducted in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Declaration of Helsinki and its later amendments or comparable ethical standards. The study was approved by the Human Research Ethics Committee of the Federal University of Pernambuco (protocol no. 476/10) and the Clinical Research Commission of the maternity hospital, where the samples were collected. All women provided their written informed consent before participating in the study.

Results

Sample recruitment resulted in 65 pairs of mothers and newborns (59 refused to participate and 52 were

excluded). The participants in the study had a mean (SD) age of 24.8 (5.4) years and 69.2% of them were in the range 21–35 years (Table 1). Assessment of the socio-demographic characteristics determined that most of the participants were married or lived with their partners (81.5%), they were multiparous (70.8%), their household consisted of four or more people (71.9%), they had a low education level (56.3% had only the primary or middle school level) and they had a low income (78.1% had an income up to one average minimum monthly salary of US\$ 243.14 during the studied period). Regarding obstetric characteristics, there was a predominance of vaginal delivery (84.6%) and most of the women (70.8%) had two or more children (parity average = 1.7 children).

According to the mother's anthropometric nutritional status during the gestational period, 38.1% of them had eutrophia. Not all women had registration of haematological exams realised during the gestational period but, among those who had, the results registered ($n = 60$) that more than 86.7% were not anaemic. Regarding newborns, the predominant gender was male (58.5%) and the birth weight of most of them was considered to be adequate (96.9%). Maternal breastfeeding was given in the immediate post-partum period to 87.7% of the neonates.

The mean or median of retinol concentrations and the prevalence of retinol inadequacy status, based on the results of the indicators maternal serum, serum of the newborn (umbilical cord) and colostrum, are presented in Table 2. No relationship was found between retinol concentrations obtained in both colostrum and maternal serum ($r = -0.17$; $P = 0.08$). Retinol concentrations in colostrum and serum of the umbilical cord also were not related ($r = 0.07$; $P = 0.29$). However, there was a significant linear relationship between the averages of retinol found in the maternal serum and the serum of the umbilical cord ($r = 0.35$; $P = 0.002$). The multiple linear regression analysis resulted in a statistically significant model ($F_{1,63} = 8.65$; $P = 0.005$; $r^2 = 0.12$), where maternal serum retinol ($\beta = 0.35$; $t = 2.94$; $P = 0.005$) is a predictor of umbilical cord retinol, with a 12% contribution to retinol level variation in newborns (Table 3).

There was no statistical significance when considering anthropometric nutritional status in the mothers and their newborns via the retinol concentrations in the maternal serum, serum of the umbilical cord and colostrum ($P > 0.05$) (Table 4). Furthermore, there was no significant difference in measurements of central tendency of retinol when the concentrations in biological samples (maternal serum, serum of the umbilical cord and colostrum) were compared among groups with respect to most of the socioeconomic variables (maternal age, marital status, number of individuals per household and family income) ($P > 0.05$); when analysed by educational level,

Table 1 Socio-demographic and obstetric characteristics of mothers and newborns attended in a public hospital in Santa Cruz-RN, Brazilian Northeast

Characteristics	Total ($n = 65$)	%	Mean (SD)
Mother			
Age (years)			24.8 (5.4)
18–20	17	26.2	–
≥21	48	73.8	–
Marital status			
Single	12	18.5	–
Married or living with partner	53	81.5	–
Number of individuals per household			3.5 (1.3)
<4	18	28.1	–
≥4	46	71.9	–
Education level			
Elementary or middle school	36	56.3	–
High school	28	43.8	–
Family income [†]			
≤1	50	78.1	–
>1	14	21.9	–
Parity			1.4 (0.5)
Primiparous	19	29.2	–
Multiparous	46	70.8	–
Type of delivery			
Vaginal	55	84.6	–
Caesarean	10	15.4	–
Haemoglobin (g dL ⁻¹) ($n = 60$)			12.0 (1.2)
<11	08	13.3	–
≥11	52	86.7	–
Haematocrit (%) ($n = 59$)			35.4 (3.1)
<33	08	13.6	–
≥33	51	86.4	–
Gestational nutritional status ($n = 63$) [‡]			
Low weight	14	12.2	–
Eutrophia	24	38.1	–
Overweight	18	28.6	–
Obesity	07	11.1	–
Breastfed another child during pregnancy ($n = 65$)			
Yes	07	10.8	–
No	58	89.2	–
Newborn			
Gestational age, weeks ($n = 63$)	–	–	39.9 (1.4)
Sex ($n = 65$)			
Female	27	41.5	–
Male	38	58.5	–
Birth Weight (g) ($n = 64$)	–	–	3,286 (48.9)
Exclusive breastfeeding ($n = 65$) [§]			
Yes	57	87.7	–
No	08	12.3	–

[†]One minimum Brazilian wage = 243.14 US dollars.

[‡]Body mass index/gestational age.

[§]During the immediate post-partum period (study period).

the results indicated that there was a significant statistical difference between groups in the biological samples of maternal serum ($P = 0.04$) and colostrum ($P = 0.01$). A similar result was found when the retinol concentration

Table 2 Retinol concentrations in the colostrum, maternal serum and serum of the newborn (umbilical cord) attended in a public hospital in Santa Cruz-RN, Brazilian Northeast

Biological material	Retinol ($\mu\text{mol L}^{-1}$)	VAD prevalence (%)
Colostrum	3.10 (1.71 – 5.31) [†]	13.8 [§]
Maternal serum	1.24 (0.58) [‡]	21.5 [¶]
Serum of the newborn	0.75 (0.36) [‡]	41.5 ^{††}

[†]Median (25th to 75th).[‡]Mean (SD).[§]Subclinical VAD = retinol < 1.05 $\mu\text{mol L}^{-1}$.[¶]VAD = retinol < 0.7 $\mu\text{mol L}^{-1}$.^{††}Low status of VA = retinol < 0.7 $\mu\text{mol L}^{-1}$.**Table 3** Multiple regression analysis for umbilical cord serum retinol predictors of newborns attended in a public hospital in Santa Cruz-RN, Brazilian Northeast

Variable	Cord serum retinol			
	r^2	β	t	P
Serum retinol	0.12	0.35	2.86	0.005*
Colostrum retinol	0.12	-0.01	-0.08	0.93

 r^2 , regressive linear multivariate; β , regression coefficient; * $P < 0.05$.

in the colostrum was evaluated according to the parity ($P = 0.002$). In addition, a significant association was identified between educational level and parity ($P < 0.001$), as well as between age and parity ($P = 0.001$), so that older and less educated women had the higher number of children ($P = 0.043$). The mothers who brought these conditions were those who presented the highest values to retinol in the colostrum (Table 4).

Discussion

As a result of the impact of VAD in the health of the populations, especially the mother–child group, the assessment of the VA nutritional status in individuals of communities not yet investigated is very important, particularly in disadvantaged areas such as the municipality of Santa Cruz, Brazilian Northeast. In these regions, the detection of cases of deficiency, especially by means of reliable indicators, becomes essential when deciding upon intervention strategies.

In the present study, despite the average concentration of retinol in the maternal serum indicating adequacy and being compatible with the literature^(23–26), a substantial part of the maternal population presented VAD, highlighting an important risk to the health of women and their infants. To evaluate the VA status in neonates, there is a predominant number of studies in the literature that

use <0.7 $\mu\text{mol L}^{-1}$ as the cut-off point to indicate a low retinol concentration^(11,27–31), and this value was also adopted as the cut-off point in the present study. The average value identified here ratifies the findings of other studies^(23–25,32,33), in which the values found are considered typical for the full-term and healthy newborn, and within the expected estimates for developing countries⁽³⁴⁾. For some studies, part of the explanation for the higher proportion of low values in the developing regions may be a result of the higher rates of preterm deliveries and a low birth weight⁽³⁴⁾. Nevertheless, in the present study, prematurity was considered as one of the exclusion criteria, and a low birth weight was found in only two neonates; therefore, it is not a determining factor for the high prevalence of serum concentrations of retinol below 0.70 $\mu\text{mol L}^{-1}$. Even in countries not known for their VAD, a significant number of children are born with retinol concentrations <0.70 $\mu\text{mol L}^{-1}$; thus, this value is not uncommon, and its use as a deficiency indicator requires more validation⁽³⁵⁾ because it might overestimate the number of inadequacy cases.

The retinol concentrations found in the umbilical cord of newborns were lower than maternal concentrations and the average retinol value in the umbilical cord corresponds to 62.1% of the average concentration of maternal serum retinol, which is consistent with previous investigations^(6,18,24,33). Shirali *et al.*⁽³⁶⁾ found a logarithmic relationship between the concentrations of maternal retinol and retinol in the umbilical cord of the newborns in a study assessing VA serum concentrations in the mother–child binomial of an Indian population, suggesting a saturable transplacental transport of VA. The fetal concentration of VA appears to be stable despite the variations in the maternal status and intake of VA⁽³⁷⁾. In this sense, even if the mothers have adequate VA reserves, such as occurs in developed countries, VA liver stocks in the newborn are found to be physiologically low, considering their requirements for the post-natal period, and VA is only sufficient for the first few days of life⁽³⁸⁾. However, the regulation mechanisms that sustain homeostasis remain obscure, and it is not yet known whether these mechanisms may successfully offset an extreme deprivation or excess of VA⁽³⁷⁾.

When the relationship between the retinol status of these neonates (umbilical cord) and the retinol status of the mothers was verified, it was observed that retinol concentrations in maternal serum are a positive predictor of retinol concentrations in umbilical cord serum. This association corroborates the literature suggesting that, when there is maternal VAD, the risk of the newborn presenting the deficiency is high, and this may have great impact on their immunological system and on child morbidity and mortality rates^(2,6,33).

Table 4 Retinol profile in maternal serum, newborn serum (umbilical cord) and colostrum, according to socio-demographic and obstetric characteristics of mothers and newborns attended in a public hospital in Santa Cruz, Brazilian Northeast (2012–2013)

Characteristics	<i>n</i>	Maternal serum retinol			Newborn serum retinol			Colostrum retinol		
		($\mu\text{mo L}^{-1}$) [#]	SD	<i>P</i>	($\mu\text{mol L}^{-1}$) [#]	SD	<i>P</i>	($\mu\text{mol L}^{-1}$) [#]	25th to 75th	<i>P</i>
Mother age (years) ^{‡,§}										
18–20	17	1.45	0.62	0.96 [¶]	0.77	0.43	0.86 [¶]	2.83	1.59–4.08	0.04 ^{††,*}
≥21	48	1.17	0.55		0.75	0.33		4.48	3.42–5.54	
Marital status ^{¶††}										
Single	12	1.25	0.77	0.96 [¶]	0.76	0.54	0.98 [¶]	2.16	0.98–6.33	0.49 ^{††}
Married or living with partner	53	1.24	0.53		0.75	0.31		3.15	1.98–4.26	
Number of individuals per household ^{¶,††}										
<4	18	1.25	0.58	0.99 [¶]	0.71	0.28	0.49 [¶]	2.11	1.34–4.23	0.32 ^{††}
≥4	46	1.25	0.58		0.78	0.38		3.16	1.98–6.12	
Education level ^{¶,††}										
Elementary and middle school	36	1.12	0.50	0.04 ^{¶,*}	0.76	0.38	0.89 [¶]	3.84	2.50–6.40	0.01 ^{††,*}
High school	28	1.43	0.63		0.77	0.32		2.06	1.08–3.98	
Family income (MBW) ^{¶,††}										
≤1	50	1.20	0.56	0.20 [¶]	0.78	0.35	0.90 [¶]	3.49	2.07–6.12	0.08 ^{††}
>1	14	1.43	0.63		0.77	0.36		1.89	1.01–4.17	
Gestational nutritional status ^{‡§}										
Low weight	14	1.14	0.71	0.47 [‡]	0.80	0.41	0.38 [‡]	2.50	1.42–4.26	0.75 [§]
Eutrophia	24	1.16	0.51		0.65	0.32		3.52	1.77–5.55	
Overweight	18	1.42	0.59		0.84	0.41		2.91	1.10–4.23	
Obesity	07	1.27	0.56		0.78	0.20		2.14	2.09–4.58	
Parity ^{¶,††}										
Primiparous	19	1.32	0.63	0.50 [¶]	0.76	0.35	0.93 [¶]	1.70	0.80–3.76	0.002 ^{††,*}
Multiparous	46	1.21	0.56		0.77	0.36		3.49	2.23–6.35	

[†]One minimum Brazilian wage = 243.14 US dollars.

[‡]Analysis of variance.

[§]Kruskal–Wallis.

[¶]Student's *t*-test.

^{††}Mann–Whitney.

[#]Mean.

*Statistical significance: *P* < 0.05.

However, if the mother has adequate dietary intake or adequate hepatic VA reserves, as well as sufficient milk production and adequate VA concentrations, the newborn's hepatic VA stock may increase during breastfeeding⁽³⁹⁾. This is why the first months of life of a baby are considered as critical for accumulating VA reserves at levels sufficient to prevent VAD in the post-weaning period⁽³⁵⁾. In this context, diagnosing the VA content in the breastmilk provides information about the mother's nutritional status, as well as about the relevant micronutrient that is passed to the baby. For these reasons, the concentration of VA in the human milk is an important indicator and has been used in investigations of VAD in the mother–child group^(40,41).

In the present study, retinol concentrations in the colostrum were compatible with the values in the breast milk found in developing countries (2.1–3.5 $\mu\text{mol L}^{-1}$)^(40,42,43). It is expected that the VA concentrations in the colostrum are usually high^(14,44), which was confirmed in

the results of the present study. This is the main reason explaining why the median value in the colostrum was higher than the cut-off point of 1.05 $\mu\text{mol L}^{-1}$ ^(15,45); however, this does not mean that these values will remain after the subsequent lactation periods are reached because, over time, there is a remarkable drop in the VA levels of the breast milk^(46–48).

When we evaluated the retinol prevalence based on the cut-off point (<1.05 $\mu\text{mol L}^{-1}$) adopted to the colostrum, 13.8% of the maternal population presented values below this cut-off point, which represents an important risk to infant health because VA concentrations in maternal milk of at least 1.05 $\mu\text{mol L}^{-1}$ are required to attend to the metabolic necessities of the offspring but are not sufficient to accumulate reserves in the liver, such that it is considered as the minimum VA concentration to prevent VAD in the first 6 months of the infant's life⁽¹⁵⁾. It is worth noting that this situation is of more concern if it is considered that, as a result of there being no specific cut-

off point to the colostrum, the cut-off point was adopted to the level of mature milk ($1.05 \mu\text{mol L}^{-1}$) to evaluate retinol status even at the beginning of lactation, when VA concentrations are naturally high^(14,44). Accordingly, based on the results, it is possible to speculate that an even greater problem would become more evident in nursing mothers from this region if the cut-off point was increased to take into consideration the natural characteristic of the higher VA concentrations in the initial days after delivery.

In view of all of the above, the diagnosis of the factors that are related to the VA status can help to identify the conditions that possibly modulate retinol in the mothers and may guide preventive measures against VAD for the most vulnerable groups. The literature suggests that some of the nutrition-related factors, as well as the obstetrical and socioeconomic/demographic profile, may have an impact on the concentration of retinol; however, there is no full agreement about these relationships^(6,8,43,49–51). In this sense, even though the results have indicated that retinol in the maternal serum was higher in the group of women who had higher educational level ($P = 0.04$) and it is known that factors such as educational level can interfere in the nutritional knowledge and dietary behaviour⁽⁵²⁾, with a consequent impact upon the nutritional status of the individuals, in the present study, it was observed that the retinol in the colostrum was higher in the group of women with a low educational level ($P = 0.01$), with a stronger statistical significance for the last finding (colostrum), indicating the greatest power of evidence. A relationship was also found between the retinol levels in the colostrum and maternal parity and mother age, which corroborates with the literature with respect to suggesting that parity may have an influence on retinol levels in the milk because previous lactation enables the high mobilisation of retinol reserves and their high rates of transfer to the mammary gland⁽⁵³⁾. In this context, it is worth highlighting that, in the present study, women who had a low educational level had a higher parity ($P < 0.001$), possibly justifying why those women who had a low educational level had higher retinol concentrations in the colostrum, in contrast to the results identified for maternal serum.

Nevertheless, in view of the problems intrinsically related to dietary, demographic and cultural factors, which are typical of economically and socially disadvantaged locations such as the municipality of Santa Cruz, it is considered that the population becomes more vulnerable to the harm of inadequate family planning, educational level and maternal nutritional status, especially with respect to childrens' growth and development, even if the results reported in the present study are apparently

conflicting in some aspects. Moreover, among the limitations of the present study, the differences between the variances could perhaps be compensated for by using a larger sample population than that analysed in the present study. Although the aim of the research is to achieve a retinol status-related representation of two groups that are biologically vulnerable to VAD, the results reported here should not be considered as fully representative because the risk factors for VA deficiency depend on the specific context of the population under investigation, and there are variations in the factors that contribute to VAD between countries, regions and communities. Accordingly, some strategies adopted in some regions of Brazil can still be considered appropriate for combating VAD. In 2005, the Brazilian Ministry of Health expanded the vitamin A supplementation programme, which was exclusively focused on children, aiming to include puerperal women living in the highest risk areas, via single-dose administration of 200 000 international units by an oral route in the immediate post-partum period⁽⁵⁴⁾. However, in June 2016, this measure was discontinued, although research has shown increased concentrations of retinol in breast milk 24 hours after supplementation^(42,55). Therefore, the strategy of prevention and control of vitamin A deficiency for puerperal women will be based on stimulating the consumption of adequate and healthy food, according to the guidelines of the Food Guide for the Brazilian Population⁽⁵⁵⁾.

Additionally, it is also important to extend studies assessing VA status, to refer individuals suspected of having VAD to health services, and to adopt policies improving the life and health conditions of both the woman and child, from the beginning of pregnancy and during the entire lactation period, aiming to prevent this important nutritional deficiency.

Acknowledgments

The authors cordially thank University Hospital Ana Bezerra (HUAB-UFRN) for allowing this study to be carried out; all the women and their newborns who participated in the study on a voluntary basis; and all those involved in collecting the samples and in the biochemical analyses of the biological samples.

Transparency declaration

The lead author affirms that this manuscript is an honest, accurate and transparent account of the study being reported. The lead author affirms that no important aspects of the study have been omitted and that any discrepancies from the study as planned have been explained.

Conflict of interests, source of funding and authorship

The authors declare that they have no conflict of interest.

National Council for Scientific and Technological Development (CNPq) of Brazil (File no. 472021/2011-7) provided funding for the present study. The funder institution had no role in the study design, data collection and analysis, decision to publish or preparation of the manuscript.

DSB was responsible for substantial contributions to the conception and design of the work; acquisition, analysis and interpretation of data for the work; drafting the work and revising it critically for important intellectual content; and final approval of the version to be published. KDSR, MSRL, JFPM and AGCLdS were responsible for substantial contributions to the acquisition and analysis of data for the work; revising it critically for important intellectual content; and final approval of the version to be published. RD and MMO were responsible for substantial contributions to the conception or design of the work and interpretation of data for the work; revising it critically for important intellectual content; and final approval of the version to be published. All authors agree 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 have been appropriately investigated and resolved.

References

- McLaren DS & Kraemer K (2012) Manual on Vitamin A Deficiency Disorders (VADD). *World Rev Nutr Diet* **103**, 1–12.
- World Health Organization (2009) *Global prevalence of vitamin A deficiency in populations at risk 1995–2005: WHO global database on vitamin A deficiency*. Geneva: World Health Organization.
- Brasil (2009) Ministério da Saúde. Centro Brasileiro de Análise e Planejamento. Pesquisa nacional de demografia e saúde da criança e da mulher (PNDS-2006): dimensões do processo reprodutivo e da saúde da criança. Brasília: Ministério da Saúde.
- Ramalho RA, Anjos LA & Flores H (1998) Hipovitaminose A em recém-nascidos em duas maternidades públicas no Rio de Janeiro, Brasil. *Cadernos de Saúde Pública* **14**, 821–827.
- Accioly E & Souza-Queiroz S (2000) Deficiência de vitamina A em embarazadas atendidas em una maternidad pública en Rio de Janeiro, Brasil. *Rev chil Nutr* **27**, 352–357.
- Ramalho RA, Flores H, Accioly E, et al. (2006) Associação entre deficiência de vitamina A e situação sociodemográfica de mães e recém-nascidos. *Rev Assoc Med Bras* **52**, 170–175.
- da Silva Ribeiro K d, de Araújo K f, de Souza H h b, et al. (2010) Nutritional vitamin A status in northeast Brazilian lactating mothers. *J Hum Nutr Diet* **23**, 154–161.
- de Lira LQ, Ribeiro PPC, Grilo EC, et al. (2011) Perfil de retinol no soro e colostro de puérperas atendidas em maternidade pública brasileira e sua associação com características maternas e obstétricas. *Rev Paul Pediatr* **29**, 515–520.
- Gurgel CSS, Grilo EC, de Lira LQ, et al. (2018) Vitamin A nutritional status in high- and low-income postpartum women and its effect on colostrum and the requirements of the term newborn. *J Pediatr* **94**, 207–215.
- Instituto Brasileiro de Geografia e Estatística (IBGE) (2018). <https://cidades.ibge.gov.br/brasil/rn/santa-cruz/panorama> (accessed 16 August 2019).
- Gazala E, Sarov B, Hershkovitz E, et al. (2003) Retinol concentration in maternal and cord serum: its relation to birth weight in healthy mother–infant pairs. *Early Hum Devel* **71**, 19–28.
- Ortega RM, Andres P, Martinez RM, et al. (1997) Vitamin A status during the third trimester of pregnancy in Spanish women: Influence on concentrations of vitamin A in breast milk. *Am J Clin Nutr* **66**, 564–568.
- Giuliano AR, Neilson EM, Kelly BE, et al. (1992) Simultaneous Quantitation and Separation of Carotenoids and Retinol in Human Milk by High-Performance Liquid Chromatography. *Method Enzymol* **213**, 391–399.
- Macias C & Schweigert FJ (2001) Changes in the concentration of carotenoids, vitamin A, alpha-tocopherol and total lipids in human milk throughout early lactation. *Ann Nutr Metab* **45**, 82–85.
- World Health Organization (WHO) (1996) *Indicators for assessing vitamin A deficiency and their application in monitoring and evaluation intervention programmes*. Geneva: World Health Organization.
- West KP Jr (2002) Extent of vitamin A deficiency among preschool children and woman of reproductive age. *J Nutr* **132**, 2857S–2866S.
- Rice AL, West KP Jr & Black RE (2004) *Vitamin A deficiency. Comparative quantification of health risks: global and regional burden of disease attributable to selected major risk factors*. Geneva: World Health Organization.
- Godel JC, Basu TK, Pabst HF, et al. (1996) Perinatal vitamin A (retinol) status of northern Canadian mothers and their infants. *Biol Neonate* **69**, 133–139.
- Atalah ES, Castillo CL, Castro RS, et al. (1997) Propuesta de um nuevo estandar de evaluación nutricional em embarazadas. *Rev Med Chile* **125**, 1429–1436.
- Brasil (2011) Ministério da Saúde. Secretaria de Atenção à Saúde. Departamento de Atenção Básica. Orientações para a coleta e análise de dados antropométricos em serviços de saúde: Norma Técnica do Sistema de Vigilância Alimentar e Nutricional – SISVAN. Brasília: Ministério da Saúde.
- World Health Organization (WHO) (1995) *Physical status: the use and interpretation of anthropometry: Report of a WHO Expert Committee*. WHO-Technical Reports Series; 854. Geneva: World Health Organization.

22. World Health Organization (WHO) (2001) *Iron deficiency anaemia: assessment, prevention and control: a guide for programme managers*. Geneva: World Health Organization.
23. Masters ET, Jedrychowski W, Schleicher RL, et al. (2007) Relation between prenatal lipid soluble micronutrient status, environmental pollutant exposure, and birth outcomes. *Am J Clin Nutr* **86**, 1139–1145.
24. Wang YZ, Ren WH, Liao WQ, et al. (2009) Concentrations of antioxidant vitamins in maternal and cord serum and their effect on birth outcomes. *J Nutr Sci Vitaminol* **55**, 1–8.
25. Beurskens LWJE, Tibboel D, Lindemans J, et al. (2010) Retinol Status of Newborn Infants Is Associated With Congenital Diaphragmatic Hernia. *Pediatrics* **126**, 712–720.
26. Grilo EC, Lima MSR, Cunha LRF, et al. (2015) Effect of maternal vitamin A supplementation on retinol concentration in colostrum. *J Pediatr* **91**, 81–86.
27. Dimenstein R, Trugo NM, Donangelo CM, et al. (1996) Effect of subadequate maternal vitamin-A status on placental transfer of retinol and beta-carotene to the human fetus. *Biol Neonate* **70**, 248.
28. Rondó PH, Abbott R & Tomkins AM (2001) Vitamin A and neonatal anthropometry. *J Trop Pediatr* **47**, 307–310.
29. Galinier A, Périquet B, Lambert W, et al. (2005) Reference range for micronutrients and nutritional marker proteins in cord blood of neonates appropriated for gestational ages. *Early Hum Dev* **81**, 583–593.
30. Barua D, Nasrin B, Nahar K, et al. (2009) Vitamin A Concentration in Cord and Maternal Serum and its Effects on Birth Weight. *Bangladesh J Child Health* **33**, 46–49.
31. Nyuar KB, Ghebremeskel K & Crawford MA (2012) Sudanese women's and neonates' vitamin A status. *Nutr Health* **21**, 45–55.
32. Dallaire F, Dewailly E, Shademani R, et al. (2003) Vitamin A concentration in umbilical cord blood of infants from three separate regions of the province of Quebec (Canada). *Can J Public Health* **94**, 386–390.
33. Deminice TMM, Ferraz IS, Monteiro JP, et al. (2018) Vitamin A intake of Brazilian mothers and retinol concentrations in maternal blood, human milk, and the umbilical cord. *J Int Med Res* **46**, 1555–1569.
34. Allen LH & Haskell M (2001) Vitamin A Requirements of Infants under Six Months of Age. *Food Nutr Bull* **22**, 214–234.
35. Ross AC. (2012) Metabolism of vitamin A in early life. In: Report: WHO technical consultation on vitamin A in newborn health: mechanistic studies. Geneva: World Health Organization.
36. Shirali GS, Oelberg DG & Mehta KP (1989) Maternal-neonatal serum vitamin A concentrations. *J Paediatr Gastroenterol Nutr* **9**, 62–66.
37. Shenai JP. (2012) Vitamin A supplementation in newborns: a conceptual framework for biological plausibility. In: WHO technical consultation on vitamin A in newborn health: mechanistic studies, Geneva: World Health Organization.
38. Underwood BA (1994) Maternal vitamin A status and its importance in infancy and early childhood. *Am J Clin Nutr* **59**, 517S–522S.
39. Sommer A (1995) *Vitamin A deficiency and its consequences: a field guide to detection and control*. 3rd edn, Geneva: World Health Organization.
40. Stoltzfus RJ & Underwood BA (1995) Breast-milk vitamin A as an indicator of the vitamin A status of women and infants. *Bull World Health Organ* **73**, 703–711.
41. Tanumihardjo SA, Russell RM, Stephensen CB, et al. (1816S) Biomarkers of Nutrition for Development (BOND) – vitamin A review. *J Nutr* **1461**(Suppl), 1816S–1848S.
42. da Ribeiro KD, Araújo KF & Dimenstein R (2009) Efeito da suplementação com vitamina a sobre a concentração de retinol no colostro de mulheres atendidas em uma maternidade pública. *Rev Assoc Med Bras* **55**, 452–7.
43. Fares S, Sethom MM, Kacem S, et al. (2016) Retinol and alpha-tocopherol in the colostrum of lactating tunisian women delivering prematurely: Associations with Maternal Characteristics. *Pediat Neonatol* **57**, 120–126.
44. Basu S, Sengupta B & Paladhi PKR (2003) Single megadose vitamin A supplementation of Indian mothers and morbidity in breastfed young infants. *Postgrad Med J* **79**, 397–402.
45. Quadro L, Hamberger L, Gottesman ME, et al. (2005) Pathways of vitamin A delivery to the embryo: insights from a new tunable model of embryonic vitamin A deficiency. *Endocrinology* **146**, 4479–4490.
46. Campos JM, Paixão JA & Ferraz C (2007) Fat-soluble vitamins in human lactation. *Int J Vitam Nutr Res* **77**, 303–310.
47. Sámano R, Martínez-Rojano H, Hernández RM, et al. (2017) Retinol and α -Tocopherol in the Breast Milk of Women after a High-Risk Pregnancy. *Nutrients* **9**, 14.
48. Lima MSR, da Silva Ribeiro KD, Pires JF, et al. (2017) Breast milk retinol concentration in mothers of preterm newborns. *Early Human Dev* **106–107**, 41–45.
49. Mello-Neto J, Rondó PHC, Oshiwac M, et al. (2009) The influence of maternal factors on the concentration of vitamin A in mature breast milk. *Clin Nutr* **28**, 178–181.
50. de Oliveira JM, Oliveira NS & Bergamaschi DP (2009) Concentrações de vitamina A no leite humano e características socioeconômicas e nutricionais maternas: resultados de estudos brasileiros. *Rev Bras Saude Mater Infant* **9**, 11–20.
51. Dimenstein R, de Dantas JC, O, Medeiros ACP, et al. (2010) Influência da idade gestacional e da paridade sobre a concentração de retinol no colostro humano. *Arch Latinoam Nutr* **60**, 235–239.
52. de Vriendt T, Matthys C, Verbeke W, et al. (2009) Determinants of nutrition knowledge in young and

- middle-aged Belgian women and the association with their dietary behavior. *Appetite* **52**, 788–792.
53. Meneses F & Trugo N (2005) Retinol, β -caroteno, and lutein zeaxanthin in the milk of Brazilian nursing women: associations with plasma concentrations and influences of maternal characteristics. *Nutr Res* **25**, 443–451.
54. Brasil (2013). Ministério da Saúde. Secretaria de Atenção à Saúde. Departamento de Atenção Básica. Manual de Condutas Gerais do Programa Nacional de Suplementação de Vitamina A. Brasília: Ministério da Saúde.
55. Brasil (2016). Ministério da Saúde. Secretaria de Atenção à Saúde. Departamento de Atenção Básica. Ofício Circular nº 17–2016 - Encerramento da suplementação de puérperas com megadoses de vitamina A no Programa Nacional de Suplementação de vitamina A. Brasília: Ministério da Saúde.

INFANTS AND CHILDREN

Feeding practices of pre-school children and associated factors in Kathmandu, Nepal

L. Shrestha,¹  A. Parmar,² B. Kulig,¹  O. Hensel¹ & B. Sturm^{1,3}

¹Department of Agricultural and Biosystems Engineering, University of Kassel, Witzenhausen, Germany

²Natural Resources Institute, University of Greenwich, Chatham Maritime, UK

³School of Natural and Environmental Sciences, Newcastle University, Newcastle upon Tyne, UK

Keywords

dietary knowledge, infants' diet, malnutrition, Nepalese mother, socio-economic status.

Correspondence

L. Shrestha, Department of Agricultural and Biosystems Engineering, University of Kassel, Nordbahnhofstrasse 1a, 37213 Witzenhausen, Germany.

Tel.: +495542981685

E-mail: sthaluna@gmail.com; uk027586@uni-kassel.de

How to cite this article

Shrestha L., Parmar A., Kulig B., Hensel O., Sturm B. (2020) Feeding practices of pre-school children and associated factors in Kathmandu, Nepal. *J Hum Nutr Diet.* **33**, 241–251
<https://doi.org/10.1111/jhn.12715>

This is an open access article under the terms of the Creative Commons Attribution-NonCommercial License, which permits use, distribution and reproduction in any medium, provided the original work is properly cited and is not used for commercial purposes.

Introduction

Despite the approximately 50% drop in the global child mortality rate between 1990 and 2016, child malnutrition continues to be a global problem that must be eliminated⁽¹⁾. Malnutrition includes undernutrition (wasting, stunting and being underweight), micronutrient deficiency (vitamins or minerals), being overweight and obesity⁽²⁾. Most overweight children live in developed countries^(3,4). Overweight-induced malnutrition leads to several diseases such as diabetes and various forms of coronary heart disorders. By contrast, children from developing countries

Abstract

Background: In developing countries such as Nepal, many children aged below 3 years do not grow at a sufficiently high rate and are vulnerable to micronutrient deficiencies (e.g. vitamin A). Challenges to child nutrition can result from poverty, unhealthy traditional practices, inadequate caring and feeding practices. The present study aimed to assess the feeding practices of pre-school children and their associated factors.

Methods: A cross-sectional study was carried out in pre-schools located in Kathmandu district between February and March 2018. Three levels in terms of price range (lower, medium and higher level) of pre-schools were selected to reach the mothers of children aged ≤ 3 years. A structured questionnaire was administered to 145 mothers. Descriptive analyses were conducted to observe the characteristics of the population. Multinomial logistic regression analyses were performed to identify the association for the factors of mothers' perception of their current feeding practices.

Results: We found that *dal-bhat/jaulo* was a common complementary food irrespective of socio-economic background. Interestingly, mothers who had received a higher education were significantly less likely to change their feeding practices (odds ratio = 0.118, confidence interval = 0.01–0.94). The mothers that fed a higher quantity porridge to their children showed a high willingness to change the feeding practices.

Conclusions: Poor feeding practices are still an important public health problem in Nepal and were observed to be associated with low socio-economic status, unawareness and a lack of knowledge towards dietary diversity combined with strong beliefs related to social forces and cultures.

mainly suffer from being underweight and stunting, leading to other malnutrition-related health conditions such as diarrhoea and chronic obstructive pulmonary disease^(1,5). An inadequate micronutrient intake leads to one-third of child deaths in Africa⁽⁶⁾ and similar challenges are faced in the south-east Asian countries such as Nepal. Nepal has 2.7 million children aged below 4 years⁽⁷⁾ and 1.6 million children are estimated to suffer from chronic undernutrition and the severe long-term consequences associated with stunting⁽⁸⁾. Malnutrition is the persistent problem, with 35.8%, 27%, 1.2% and 9.7% of children under 5 years of age being stunted, underweight,

overweight and wasted, respectively (data based on the year 2016)⁽⁹⁾. The negative impact of being underweight, overweight or obese during childhood can lead to permanent and irreversible impairment and increases the risk of several chronic diseases because the first 5 years of life are fundamental for the development of organs and bones⁽¹⁰⁾.

Inappropriate feeding practices among infants are one of the major causes of malnutrition in Nepal⁽¹¹⁾. An awareness of the need to feed various types of foods by mothers or caretakers has a significant influence on the health and nutrition of the children⁽⁸⁾. For example, Karkee *et al.*⁽¹²⁾ identified that Nepalese mothers were breastfeeding more (98.0%) and feeding lower (13.4%) levels of infant complementary foods at the age of 22 weeks. This resulted in poor feeding practices at the critical stage (6–24 months), resulting in malnutrition in many children at a vulnerable stage of their growth. It is recommended that children aged 6–23 months must be fed from four or more out of seven food groups daily: [(i) grains, roots, tubers; (ii) legumes and nuts; (iii) dairy foods; (iv) fruits and vegetables; (v) green leafy vegetables and other fruits; (vi) meat, poultry and fish; and (vii) eggs⁽¹³⁾. In Nepal, 53% of children aged between 6 and 23 months do not reach the minimum dietary diversity, and only 36% consume a minimum recommended diet⁽⁹⁾. Traditionally, complementary feeding practices start after the 'Annaprashan' (rice feeding) ceremony. The ritual is normally held in the fifth month of a girl's and the sixth month of a boy's life⁽¹⁴⁾. Even though the time of initiation of complementary foods in the Nepalese

context is in accordance with the recommendations on complementary feeding, the feeding practices and dietary diversity are still falling behind recommendations⁽¹⁵⁾. According to previous studies^(13–15), several factors have been identified to explain the mother's decision with respect to poor feeding practices. The major factors were stated as a mother's education and dietary knowledge, socio-economic conditions, socio-cultural and physiological aspects, family pressure on continuing the traditional feeding practices, and a lack of social support. In the case of developing countries such as Nepal, a key factor affecting all underlying determinants is poverty⁽¹⁶⁾. A low economic background limits access to a good education and health system. Additionally, it reduces the range of affordable products, as well as their quality and quantity. This leads to poor feeding practices and consequently causes child malnutrition. The cultural context, especially at the local level, is also an influential determinant directly affecting the feeding practices⁽¹⁷⁾. Because Nepal is a culturally diversified country, feeding practices might vary according to the geographical region, ethnic groups and cultural taboos⁽¹⁵⁾.

Several local infant food formulas, their constituents and method of preparation are presented in Table 1⁽¹⁸⁾. Generally, 'dal-bhat' (pulse-rice) is the primary weaning food, where fruits and vegetables are rarely fed⁽¹⁹⁾. The other supplementary foods given are cow, buffalo and goat milk. Types of semi-solid foods are *jaulo* (rice, turmeric and salt), *chapatti* (home-made breads), *lito* (roasted rice flour; occasionally maize or millet), *ghee* (clarified butter and sugar), *sarbottam pitho* (local market

Table 1 Description of local diet intake of children, their constituents and method of preparation

Serial Number	Local food name	Constituents	Method of preparation
1.	<i>Dal-bhat/jaulo</i>	<i>Dal-bhat</i> : made from a mixture of rice and lentils soup <i>Jaulo</i> : made from rice and turmeric or rice and salt	Rice and lentils are cooked to a semi-solid consistency
2.	<i>Lito</i>	Made from roasted rice flour (occasionally maize or millet), <i>ghee</i> (clarified butter) and sugar	All mixture is stirred into boiling water or milk and cook for a short time
3.	<i>Sarbottam pitho</i>	Two parts pulse – soybeans, small beans, grams and peas (any one of these can be used) One-part whole-grain cereal – maize or rice One part another whole-grain cereal – wheat, millet or buckwheat *All pulses and grains are roasted well (separately) and ground into fine flour and then stored in an airtight container	All mixture (flour) is stirred into boiling water or milk and cook for a short time
4.	Commercial baby foods	Nestle cerelac baby foods – rice, soya bean, sugar, skimmed milk powder (cow's milk), vegetable oils, apple powder (glucose, apple juice concentrate), minerals (calcium carbonate, sodium phosphate, ferric pyrophosphate, zinc sulphate, potassium iodide), contains emulsifier as permitted food conditioner. Contains permitted flavouring. All additives are of plant or synthetic origin	Cerelac is mixed with boiled water
5.	Routine foods	Daily food routine – rice, bread, lentil, pulses, fruits and vegetables, meat, eggs	Only spices are avoided in the baby's food

*is used to emphasise the roasting process of all pulses and grains.

porridge), vegetables, *dal* (pulse soup), eggs and meat⁽²⁰⁾. The National Planning Commission, World Food Program, World Bank, AusAID and UNICEF (2013)⁽²¹⁾ reported that diets comprising starchy staples lack essential micronutrients (vitamins, iron, zinc, magnesium, etc.), therefore contributing to malnutrition.

Despite the existence of many nutritional studies on Nepal, little is known about the feeding practices of pre-school children (6–36 months) located in Kathmandu district. Furthermore, most of the previous studies were conducted in hospital settings. Pre-schools could be a good platform to reach mothers of a mixed socio-economic background and encourage them to vary their feeding practices. The present study therefore intended to assess the feeding practices of mothers and identify the associated factors that can be used as a vital indicator of the overall nutrition and healthy development of pre-school children.

Materials and methods

Study design and setting

In the present study, a cross-sectional survey was conducted between February and March 2018 in Kathmandu valley, Nepal. This design was chosen because it is appropriate for a short period of data collection and to assess the relationship between the selected variables⁽²²⁾.

Nepal has a population of approximately 29 million and is divided into 75 administrative districts. Amongst

these, Kathmandu valley has a total population of 2.8 million (i.e. almost 10% of the overall population). Kathmandu valley is comprised of five Metropolitan cities: Kathmandu Metropolitan City, Lalitpur Metropolitan City, Bhaktapur Municipality, Kritipur Municipality and Madhyapur Thimi Municipality. Kathmandu Metropolitan City, which has approximately 1 million residents⁽²³⁾, was chosen for the present study. Based on the National Population and Housing Census 2011, there are 763 Early Childhood Development and Pre-primary classes available in the Kathmandu valley⁽²⁴⁾. This number was not used to calculate the sample size because it lacks information only for early childhood development.

In the present study, 27 different pre-schools located in Kathmandu Metropolitan City were sampled to recruit mothers from a range of socio-economic backgrounds. Currently, the trend of the pre-schooling education is increasing in the three Kathmandu subdistricts but is still not profoundly activated in other districts of Nepal. Furthermore, different levels of pre-schools are available in Kathmandu: low price range pre-schools charge NRs 550 (\$5), medium price range pre-schools charge NRs 2500 (\$22) and high price range pre-schools (expensive) charge NRs 6000 (\$53) per month on average. The survey plan as split by of pre-schools is presented in Fig. 1. Selecting mothers from pre-schools of all price ranges was relevant with respect to reaching different income group families and gaining a perspective towards differences in feeding practices, their own (healthy) consumption habits and the

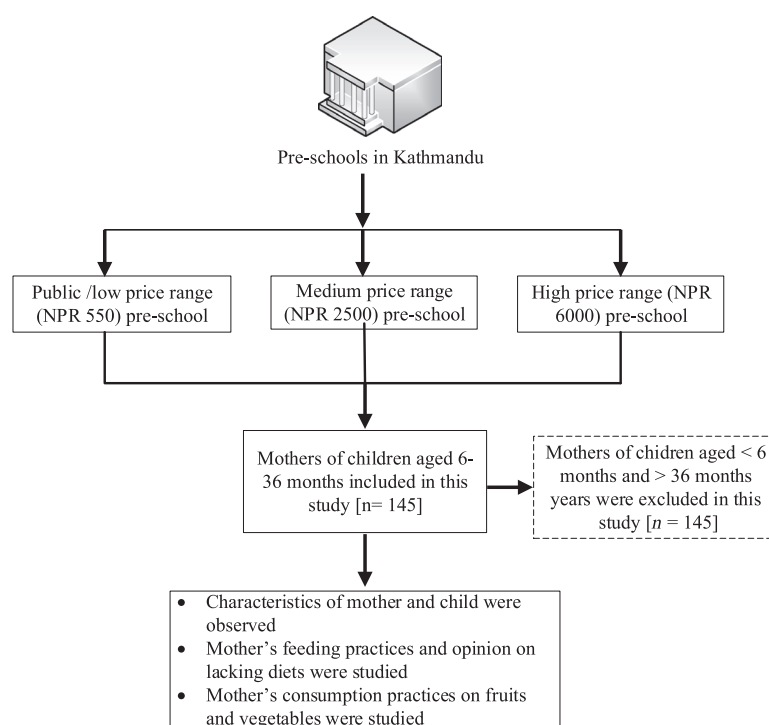


Figure 1 Study setting and design.

influences of social bonding towards their feeding practices.

Sample size and sampling procedure

Mothers with children of an age between 6 and 36 months who had lived in the Kathmandu district for at least 6 months were eligible for the study. Study participants were selected through a random sampling method of the pre-schools available (i.e. who gave permission to use their pre-school premises to take an interview with mothers). With the consent of the chief or head leader of the pre-schools, mothers were pre-informed and invited to participate in the research. Only those who had an interest and the time to participate in the study volunteered. Mothers of children aged more than 36 months were excluded from the study. The final sample comprised of 145 mothers excluding those ($n = 8$) who refused to participate (Fig. 1). The refusal of a mother who was approached was generally because of a lack of interest, a lack of time and the fact that they could not manage the infants during the interview ($n = 8$). The age range of 6–36 months was selected for design reasons relating to feeding practices that are of nutritionally paramount importance for promoting the optimal development and growth of the child.

Data collection tools and procedures

The structured questionnaires were used to collect data; a copy of the questionnaire is provided in the Supporting information (Data S1). Feeding practices were assessed according to the key indicators recommended by World Health Organization⁽²⁵⁾. Outcome variables such as type of complementary feeding adopted by mothers were assessed. Data collectors created a conducive environment by keeping mothers apart and making them comfortable during the data collection to minimise the social desirability bias. A structured interview schedule was used as tool to gather information on factors associated with feeding practices. The questionnaires mainly comprised details regarding household profiles such as age, sex, education level and occupation of household members and household size. Specifically, the factors considered were:

- 1 Characteristics of mothers (age, occupation, residence, number of children, etc.)
- 2 The pattern of complementary foods and the mother's perspective towards lacking diets
- 3 Association between different levels of social life and socio-economic background of the mother
- 4 Association of changing the feeding practices with several factors such as socio-demographic characteristics

5 Mother's fruit and vegetable consumption habits

Quantitative data were designed continuously with the help of a 1–9 scaling score⁽²⁶⁾. An objective rescaling distance was created when performing the analysis (e.g. a 1–3 scaling score).

A pre-tested questionnaire initially developed in English and then translated in the local language (Nepali) was used for data collection. A facilitator and a student assistant who had a background in social work study were involved to conduct all interviews. Prior to data collection, the research assistants (data collectors and supervisors) were trained for 2 days on interview techniques. The questionnaire was pre-tested on 10 subjects in two pre-schools in the study area. The clarity, acceptability and applicability of the procedures were evaluated during the pre-test and used to make any necessary corrections to the questionnaire and the mode of question delivery to the subjects.

Study variables

The dependent variable in the present study is considered to be feeding practices. The tendency of mothers to change their feeding practices was taken as the source to determine the likelihood of adapting appropriate feeding practices. These are indicated by feeding foods in a diversified way, such as animal and plant-based food products. The independent variables were mainly related to the mothers' socio-demographic and economic characteristics [age, education, employment, religion, household size and marital status, child characteristics (age, sex), maternal consumption habits (mother's fruits and vegetable consumption patterns) and health conditions of the children]. The selection of pre-school was organised based on a mother's capability to afford that price level and is a representation of the socio-economic background of the Nepalese population. Each of the variables was measured using a single question.

Statistical analysis

Data were checked for completeness and inconsistencies and then collated using EXCEL (Microsoft Corp., Redmond, WA, USA) before analysis. All data were analysed using SPSS, version 22.0 (IBM Corp., Armonk, NY, USA). Socio-demographic characteristics and the prevalence of complementary feeding practices were analysed using descriptive statistics, with the results being presented as the frequency. Multinomial logistic regression analysis was used to determine the associations of feeding practices and their different factors, such as socio-economic characteristics of mothers and child status. Associations were examined by odds ratios (ORs) for the above-

mentioned indicators to determine the likelihood of changing the feeding practices. The backward elimination method was used, aiming to avoid any statistical bias, by double-checking and following procedures: (i) entering only variables with $P < 0.05$; (ii) testing the backward elimination by also including all potential confounders; and (iii) testing and reporting variables that are highly corrected in a logistic regression model.

Results

Characteristics of mothers and their children

Table 2 provides the characteristics of mothers and their children. All 145 respondents were married. Remarkably, 60% of the mothers were of university-level education followed by a higher secondary degree (37.2%). Some 89% of the mothers were residing in an urban area. The reasons for living in the urban area were cited as marriage (40.8%), better opportunities (24.2%) and to achieve a higher education (21.7%). The extended family structure was found to be represented almost twice as often (60.7%) compared to a nuclear family structure (33.8%). Most mothers had a single child (66.9%) and 75.2% of the children were aged below 24 months. Noteworthy, most mothers selected medium-priced range pre-schools compared to high and low-priced range.

Complementary food practices and mother's perspective towards lacking diets

In Nepal, it is very common practice to introduce complementary food after the rice feeding ceremony. Nevertheless, complementary feeding practices differ greatly between the mothers (Fig. 2a). The findings in the present study indicate staple food (*dal-bhat/jaulo*) to be the most commonly used complementary food (36.10%) among the children. A strong negative correlation was found between the willingness of the mother to change their feeding practices and current feeding by staple foods ($P < 0.05$). Some 32% of the mothers fed their child with homemade porridge known as '*lito*'. It is remarkable that 15.10% of mothers fed their children with commercial food products such as cerelac and only less than half (6.40%) chose a diet based on local products such as *sar-bottam pitho*. Some 10.40% of the children were fed with routine foods (pulses-rice, roots and tubers, fruits and vegetables, meat products).

The food groups lacking the most, according to the mothers' perception, were pulses, fruits and vegetables (fresh and dried), and milk products (Fig. 2b). The participants with a higher education level emphasised the importance of consuming fruits and vegetables for a balanced diet. However, most of the mothers stated that fruit and

Table 2 Mothers' and children's characteristics

Variables	Categories	Frequency (%)
Mothers' age	<21	2 (1.4)
	21–25	46 (33.7)
	26–30	55 (37.9)
	>30	42 (29.0)
Ethnicity	Chhetri	30 (20.7)
	Brahmin	32 (22.1)
	Magar	11 (7.6)
	Tharu	3 (2.1)
	Tamang	22 (15.2)
	Newar	27 (18.6)
	Others	20 (13.8)
Education level	Primary school	4 (2.8)
	Secondary school	54 (37.2)
	Academic/university	87 (60.0)
Origin of residence	Urban	36 (24.8)
	Peri-urban	47 (32.4)
	Rural	62 (42.8)
Current residence	Urban	129 (89.0)
	Peri-urban	16 (11.0)
Reason to migrate	Marriage	64 (40.8)
	Education	34 (21.7)
	Family	21 (13.4)
	More opportunities	38 (24.2)
Occupation	Housewife	70 (48.3)
	Informal sector	51 (35.2)
	Formal sector	14 (9.7)
	Self-employed	10 (6.9)
Number of children	1	97 (66.9)
	2	41 (28.3)
	3	7 (4.8)
Sex of child	Male	76 (52.4)
	Female	69 (47.6)
Child age group	6–12 months	12 (8.3)
	13–24 months	109 (75.2)
	25–36 months	24 (16.6)
Family structure	Own's parent	4 (2.8)
	Extended family	88 (60.7)
	Nuclear family	49 (33.8)
	Alone	4 (2.8)
Type of pre-school	High price range	42 (29)
	Medium price range	92 (63.4)
	Low price range	11 (7.6)

vegetable consumption was lacking in their daily routine. Only a few mothers (5%) considered there to be a lack of meat and meat products in their feeding practices.

The association of level of social life with the economic status of mothers based upon the selection of pre-school

Table 3 shows the association of social bonding with the socio-economic ability of mothers. Other factors, such as education, age and the number of children, were not significantly associated with the different levels of social bonding and thus the results are not presented. In the

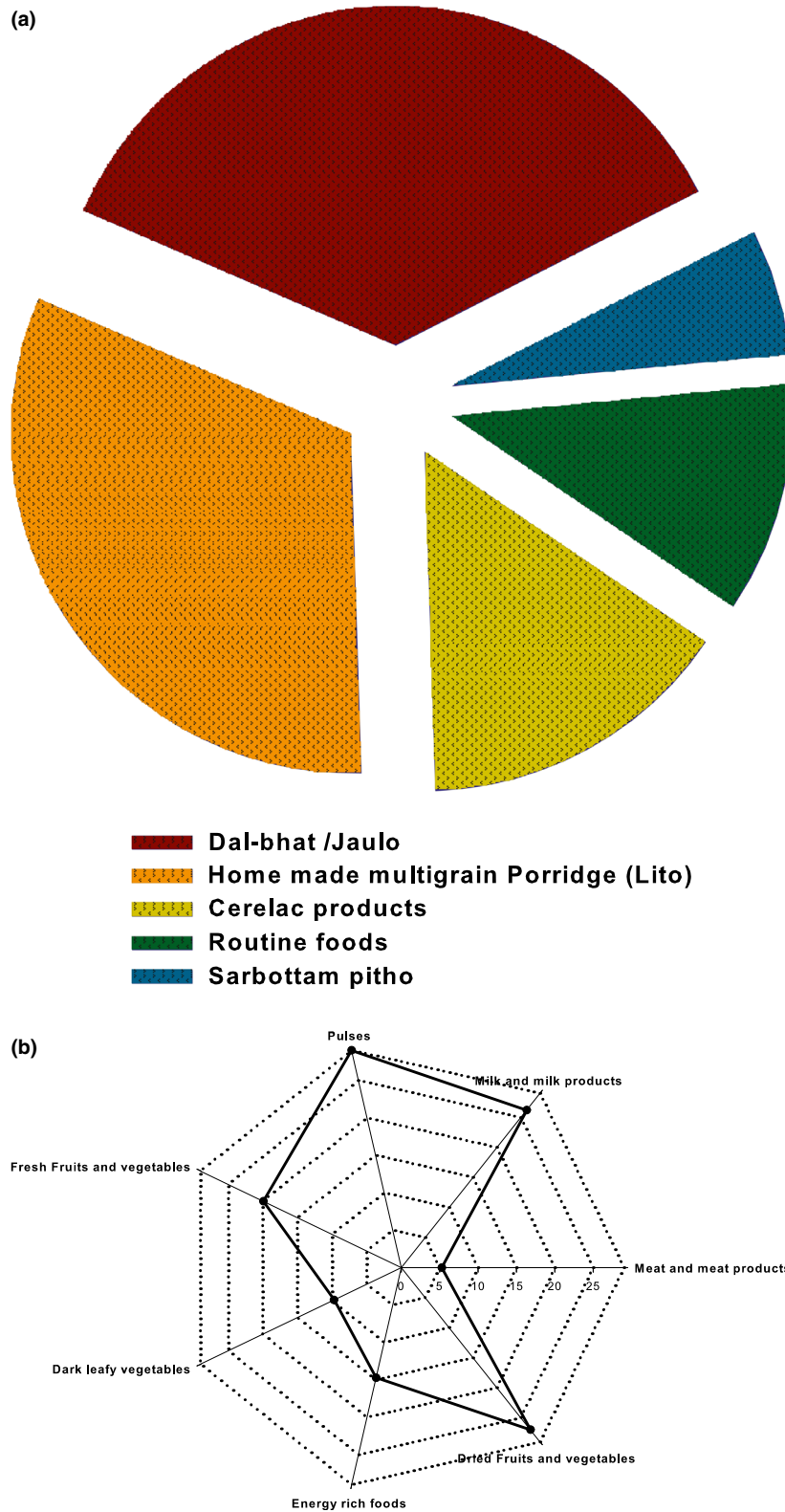


Figure 2 (a) Complementary foods feeding pattern. (b) Food groups lacking in the feeding practices of the children age (6–36 months).

Table 3 Association of social bonding with the mother's socio-economic background

	Not at all			Average			Very much		
	Coefficient	OR (95% CI)	P-value	Coefficient	OR (95% CI)	P-value	Coefficient	OR (95% CI)	P-value
Type of Pre-school									
Lower	2.219	9.20 (2.065–40.99)	0.004	1.749	5.75 (0.832–39.740)	0.076	2.828	16.90 (0.74–385.47)	0.076
Higher	–.049	0.95 (0.336 –2.69)	0.926	0.733	2.08 (0.691–6.275)	0.193	2.971	19.50 (0.90–422.09)	0.058
Medium		1			1			1	

OR, odds ratio; CO, confidence interval.

present study, the mothers who sent their children to a highly priced pre-school (i.e. assuming the mothers from a higher socio-economic background) were less likely to be socially active (OR = 0.95; 95% CI = 0.33–2.69). However, there was no significant difference between

mothers selecting high- and medium-priced pre-schools. Mothers who are sending their children to a low-priced pre-school (i.e. assuming the mothers are from a low socio-economic background) were more likely to engage with the society and have a higher level of social bonding.

Table 4 Mothers' association towards changing their feeding practices with several factors

Variables	Not important OR (95% CI)	Average OR (95% CI)	Very important OR (95% CI)
Education level			
Basic education	0.52 (0.04–6.09)	1.23 (0.10–14.64)	1.89 (0.16 –21.80)
Higher level education	0.83 (0.39–1.78)	1.13 (0.44–2.87)	1.19 (0.55–2.55)
Academic level education	1	1	1
Type of pre-school			
Lower	0.72 (0.12–4.21)	2.500 (0.60–10.29)	0.35 (0.05 –2.15)
Higher	2.04 (0.93–4.46)	0.118 (0.01–0.94)	0.72 (0.12–4.12)
Medium	1	1	1
Opinion on baby health status			
Not healthy	2.63 (0.62–11.16)	5.553 × 10 ⁻⁹ (5.553 × 10 ⁻⁹ to 5.553 × 10 ⁻⁹)	0.36 (0.08–1.56)
Average	1.75 (0.56–5.45)	1.12 (0.25–4.87)	0.53 (0.172–1.68)
Healthy	1	1	1
Age of child (months)			
6–12	0.12 (0.04–1.59)	0.09 (0.01–1.22)	3.66 (0.73–18.33)
13–24	0.09 (0.01–1.41)	0.72 (0.18–2.79)	1.57 (0.57–4.29)
25–36			
Number of children			
1	0.12 (0.01–1.77)	0.148 (0.010–2.236)	3.79 (0.375–38.35)
2	0.09 (0.01–1.41)	0.11 (0.01–1.78)	5.32 (0.49–57.31)
3	1	1	
Routine feeding practices			
Homemade porridge (No)	1.61 (0.59–4.40)	3.47 (1.13–10.95)	0.62 (0.22–1.70)
Homemade porridge (Yes)	1	1	1
Locally available porridge (No)	1.69 (0.45–6.35)	11.66 (1.17–115.90)	0.59 (0.15–2.21)
Locally available porridge (Yes)	1	1	1
Breast milk and dairy foods (No)	0.99 (0.42–2.32)	0.27 (0.06–1.11)	1.01 (0.43–2.37)
Breast milk and dairy foods (Yes)	1	1	1
Commercial market product (No)	0.55 (0.23–1.27)	0.61 (0.19–1.88)	1.81 (0.78–4.20)
Breast milk and dairy foods (Yes)	1	1	1
All type of foods (No)	0.77 (0.289–2.09)	0.62 (0.19–2.03)	1.28 (0.47–3.45)
All type of foods (Yes)	1	1	1

OR, odds ratio; CO, confidence interval.

The association of changing the feeding practices with several factors; fruits and vegetables consumption patterns of mothers

Table 4 shows the factors associated with mother in Kathmandu Metropolitan City changing their feeding practices. Mothers from a high socio-economic background have an average likeness (OR = 0.118; 95% CI = 0.01–0.94) to change their feeding practices. There is a significant difference ($P < 0.05$) between the mothers who had the opinion that their children are healthy, average and not healthy. The mothers that fed more porridge to their children showed a significantly different ($P < 0.05$) willingness to change their feeding practices compared to those that fed medium and low proportions of porridge. The ORs of other factors such as the age of children, number of children and education of mothers were not significantly associated with changing the feeding practices.

The consumption of fruits and vegetables per day by mother is presented in Fig. 3. Interestingly, 71% of mothers consumed at least one or two fresh fruits per day and 75.2% consumed three or four fresh vegetables per day. The consumption of dried fruits and vegetable juice was found to be negligible. However, the consumption of dried vegetables such as *gundruk*, *sinki*, dried broccoli and mushroom was very common.

Discussion

The present study reports the baseline information collected from mothers in Kathmandu Metropolitan City on

their feeding practices towards their children and associated factors. Most participants (60%) had an academic level (bachelor level or above) of education. This is in accordance with the study carried out by Cunningham *et al.* (27) which showed that the educational level in Nepal has substantially progressed during recent decades because the net enrolment rate in elementary education has increased from 66.3% in 1999 to 97% in 2016. Likewise, female participation in educational attainment has also increased dramatically (28). This phenomenon can be observed globally as the role of women has changed over the last decades (29). They are progressively coming closer to being granted equal rights to men regarding education and the range of opportunities.

Most mothers were assumed to be from a middle-class background because they sent their children to a medium price pre-school. In the present study, mothers were not open to answering how high the family income was. Thus, the price level of pre-schools was used as an indirect income indicator based on the assumption that it reflected the affordability.

Furthermore, the extended family structure still exists in large parts of Nepalese society and was found to have the greatest influence on child feeding practices. The children raised within this context were fed mostly staple foods (*dal-bhat/jaulo*). This might be a result of the influence of elders, traditional and religious rituals, and their consumption habits. Mothers from a low socio-economic background were significantly more socially bonded than mothers from a high socio-economic background This

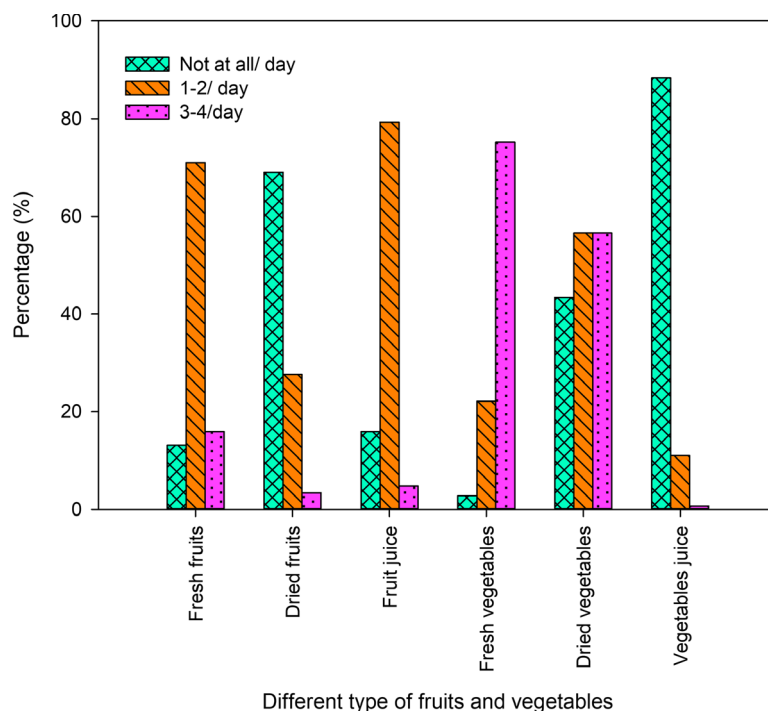


Figure 3 Fresh fruits and vegetable consumption of mothers.

might be why mothers from a low socio-economic background wanted to learn about improving their feeding practices. Moreover, being socially active encourages mothers modifying their feeding behaviour and improving the health status of infants.

Most mothers considered the current diets to be lacking fruits and vegetables (fresh and dried), as well as milk and milk products, rather than meat and meat products. This confirmed that dietary diversification is still lacking for infants among mothers' practices, which aligned well with the study carried out by Cunningham *et al.*⁽²⁷⁾. Those mothers who fed their children with more home-made and locally available porridge were significantly interested in changing their feeding practices to improve the health status of their children. The willingness to change feeding practices increased with a high educational level of the mothers. Thus, it appears that mothers with a higher education have a better understanding of the necessity of balanced infant diets and are more aware of issues related to feeding practices.

The results of the present study show clearly that feeding practices in relation to fruits and vegetables and mothers' consumption were interrelated. Mothers who did not consume fruits themselves did not prioritise feeding fruits and vegetables to their children. These findings agree with the findings of Locks *et al.*⁽¹⁵⁾ in that only some mothers fed their children fruits and vegetables alongside the porridge. This might also be a result of the influence of joint family structures, as well as changing living standards (i.e. moving from the county side to the city). However, education and social bonding were found to have the greatest impact on the mothers' willingness to improve feeding practices and could define what types of food are missing or lacking in infant nutrition. This might explain why the overall child nutrition status has not improved as much would be expected, irrespective of the education and social status of mothers.

Additionally, the respondents in the present study were self-decision makers and mostly willing to change feeding practices to upgrade the nutritional status of their children. The self-decision capability may be a result of the effect of the education level and social interaction motivations. Imdad, Yakoob & Bhutta⁽³⁰⁾ reported that educated mothers place high emphasis on feeding nutrient-rich animal source foods as a diet diversification. Obviously, this might vary again according to ethnicity, residence region and cultural aspects⁽³¹⁾.

Conclusions

An important aspect of the present study was the assessment of primary associations of changing feeding practices with several influencing factors, such as education,

economic conditions and family structure. This helps to understand the mechanisms behind mothers' knowledge on feeding practices and the health status of their children in a real-world context. Mothers were well aware of the fact that the first years of the children's life are crucial for physical and mental development, with a lifelong impact. However, very few mothers had appropriate knowledge regarding the necessary modifications to the dietary intake of children, as well as how to analyse the results by observing health status. These findings reflect the importance of generating awareness of the current situation and the need for improving the knowledge of mothers about child feeding practices, which, in turn, will help to reduce the malnutrition problem in Nepal.

A clear limitation of the present study is that we analysed cross-sectional data; therefore, the findings could only be relevant to the study context and similar settings. The relationship between mothers' fruit and vegetable consumption and that of their children might be associated. To further clarify the cause and effect relationship, longitudinal analyses are needed. In addition, the present study was focused on the mothers and their feeding dietary and consumption patterns, which may evoke a desirability bias. This implies that the need for interventions to increase the dietary diversification patterns, rather than focusing on the traditional way of feeding, should be emphasised. Future studies could include other caregivers and entities, such as grandparents, fathers, schools and children's hospitals, to obtain a broader picture on child feeding practices by assessing the method of feeding, pre-lacteal feeds, colostrum, types of diet, age of complementary food and types of complementary food.

Another limitation was the small sample size, which was a result of the limited time boundary and resources. Hence, there could have been a recall bias. Because the results have been drawn from an urban population, this cannot be generalised to the rural population or other parts of Nepal. Nevertheless, the strength of the present study is that it has determined some important aspect regarding feeding practices and a lack of diversity of diets among Nepalese mothers at pre-schools located in Kathmandu Metropolitan City. Although some indicators identified in the present study provide baseline information, there is still a need to conduct further studies to increase awareness amongst mothers and improve the diets of children aged under 3 years of age.

Acknowledgments

First and foremost, the authors express their sincere thanks to the pre-schools for providing an area to conduct the present study. They are also grateful to all of the mothers for their active participation. Their thanks are

also extended to Ms Pratima Gurung who helped to collect the data.

Conflict of interests, source of funding and authorship

The authors declare that they have no conflicts of interest.

This work was supported by the Katholischer Akademischer Ausländer Dienst (KAAD) as a scholarship and providing the travel grant to the first author.

LS, OH and BS conceived and designed the study. LS conducted the study, collected the data, constructed the database, analysed and interpreted the results, and wrote the manuscript, as supported by BS. BK helped to revise the questionnaire formatting and data analysis. AP read and reviewed all sections of the manuscript. All authors read and approved the final manuscript submitted for publication.

Transparency declaration

The lead author affirms that this manuscript is an honest, accurate and transparent account of the study being reported. The reporting of this work is compliant with STROBE guidelines. The lead author affirms that no important aspects of the study have been omitted and that any discrepancies from the study as planned have been explained.

References

- World Health Organization (2018) World Health Statistics 2018- Monitoring Health for the SDG's (Sustainable development goals) [Internet]. Available at: https://www.who.int/gho/publications/world_health_statistics/2018/en/
- Lamichhane A (2018) Nutritional Assessment of Children Under the Age 6–59 Months of Chepang Community Residing in Kusum Khola Area. Available at: <http://104.236.33.185:8001/jspui/handle/123456789/759>
- Statistisches Bundesamt (2013) *Statistisches Bundesamt Mikrozensus – Fragen zur Gesundheit*, Vol 49. Wiesbaden, Germany. Available at: https://www.destatis.de/GPStatistik/servlets/MCRFileNodeServlet/DEHeft_derivate_00015420/5239001139004.pdf;jsessionid=E2736D29D8600268ACFDF0C9A687D7FC (accessed 4 February 2019).
- Schienkiewitz A, Brettschneider A, Damerow S & Rosario AS (2018) Overweight and obesity among children and adolescents in Germany. Results of the cross-sectional KiGGS Wave 2 study and trends. *Journal of Health Monitoring* 3, 15–22. Available at: <https://doi.org/10.17886/RKI-GBE-2018-022>.
- Grubestic RB (2004) Children aged 6 to 60 months in Nepal may require a vitamin A supplement regardless of dietary intake from plant and animal food sources. *Food Nutr Bull* 25, 248–255.
- ACPF. For Lack of Will: Child Hunger in Africa: African Child Policy Forum (ACPF) [Internet]. Addis Ababa, Ethiopia; 2019. Available at: <https://app.box.com/s/agsfc0xuvp7u6yvez8f6b3etdmgrq29b> (accessed 8 September 2019).
- USAID. Nepal Nutrition Profile. 2014;(July 2014):1–8. Available at: <https://www.usaid.gov/what-we-do/global-health/nutrition/countries/nepal-nutrition-profile> (accessed 16 Jan 2019).
- United Nations Committee for Development Policy. Committee for Development Policy 20th Plenary Session: Vulnerability Profile of Timor-Leste [Internet]. 2018. Available at: un.org/development/desa/dpad/wp-content/uploads/sites/45/CDP-PL-2018-6f.pdf (accessed 16 January 2019).
- Nepal Demographic and Health Survey. Nepal Demographic and Health Survey 2016 Key Indicators Report Ministry of Health Ramshah Path, Kathmandu Nepal New ERA Ministry of Health [Internet]. 2017. Available at: www.DHSprogram.com (accessed 16 Jan 2019).
- UNICEF, WHO, World Bank (2014) Levels and Trends in Child malnutrition. *Midwifery* 4, 154–155. Available at: [https://doi.org/10.1016/S0266-6138\(96\)90067-4](https://doi.org/10.1016/S0266-6138(96)90067-4) (accessed 7 February 2019).
- Osei A, Pandey P, Spiro D *et al.* (2010) Household food insecurity and nutritional status of children aged 6 to 23 months in Kailali District of Nepal. *Food Nutr Bull* 31, 483–494.
- Karkee R, Lee AH, Khanal V & Binns CW (2014) Infant feeding information, attitudes and practices: a longitudinal survey in central Nepal. *Int Breastfeed J* 9, 1–5.
- WHO. Global Strategy on Infant and Young Child Feeding (WHA55 A55/15). World Health Organization [Internet]. 2008;19. Available at: http://whqlibdoc.who.int/publications/2008/9789241596664_eng.pdf?%5Cnhttp://www.unicef.org/progrmme/breastfeeding/innocenti.htm%5Cnhttp://innocenti15.net/declaration.pdf.pdf%5Cnhttp://whqlibdoc.who.int/publications/2010/9789241599757_eng.pdf (accessed 18 January 2019).
- Locks LM, Pandey PR, Osei AK *et al.* (2015) Using formative research to design a context-specific behaviour change strategy to improve infant and young child feeding practices and nutrition in Nepal. *Matern Child Nutr* 11, 882–896.
- Na M, Aguayo VM, Arimond M *et al.* (2017) Trends and predictors of appropriate complementary feeding practices in Nepal: An analysis of national household survey data collected between 2001 and 2014. *Matern Child Nutr* 14, e12564.
- Patel A, Badhoniya N, Khadse S *et al.* (2010) Infant and young child feeding indicators and determinants of poor feeding practices in India: secondary data analysis of

- National Family Health Survey 2005–06. *Food Nutr Bull* **31**, 314–333.
17. Pramod Singh GC, Manju N, Grubestic RB & Connell F (2009) Factors associated with malnutrition among children in Rural Terai of Eastern Nepal. *Asia Pacific J Public Health* **21**, 144–152.
 18. Nidhi KS. (2018) Weaning Practices in Nepal [Internet]. tearfund Learn. Available at: https://learn.tearfund.org/en/resources/publications/footsteps/footsteps_51-60/footsteps_52/weaning_practices_in_nepal/ (accessed 1 December 2018).
 19. Bhandari S & Banjara MR (2015) Micronutrients deficiency, a hidden hunger in Nepal: prevalence, causes, consequences, and solutions. *Int Sch Res Not* **2015**, 276469.
 20. Chapagain RH. (2014) Knowledge and practices of mothers of Infant and young child on complementary feeding (Based on the study done at Kanti Children's Hospital, Kathmandu, Nepal). Available at: https://www.researchgate.net/publication/232237398_KNOWLEDGE_AND_PRACTICES_OF_MOTHERS_OF_INFANT_AND_YOUNG_CHILD_ON_COMPLEMENTARY_FEEDING/citations (accessed 12 September 2018).
 21. National Planning Commission (NPC) CB of S (CBS), World Food Program (WFP), (WB) WB (WB), AusAID, UNICEF. (2013) Nepal Thematic Report on Food Security and Nutrition 2013 [Internet]. Available at: documents.wfp.org/stellent/groups/public/documents/ena/wfp256518.pdf?_ga=1.182793378.52 (accessed 12 September 2018).
 22. Levin KA (2006) Study design III: cross-sectional studies. *Evid Based Dent*. **7**, 24–25.
 23. Panta PR & Dongol D (2009) Kathmandu valley profile. Briefing Paper, Governance and Infrastructure Development Challenges in Kathmandu Valley. Available at: https://www.eastwestcenter.org/fileadmin/resources/seminars/Urbanization_Seminar/Kathmandu_Valley_Brief_f or_EWC___KMC_Workshop__Feb_2009_.pdf (accessed 12 September 2018).
 24. CBS. (2014) National Population and Housing Census 2011 (Tables from Form-II) Government of Nepal National Planning Commission Secretariat. 03. Available at: [http://cbs.gov.np/image/data/Population/Tables fromForm-II/NPHC2011\(TablesfromForm-II\).pdf](http://cbs.gov.np/image/data/Population/Tables fromForm-II/NPHC2011(TablesfromForm-II).pdf) (accessed 12 September 2018).
 25. WHO (2010) *Indicators for Assessing Infant and Young Child Feeding Practices. Part 3 Ctry Profiles*. pp. 1–47. Geneva: World Health Organization.
 26. Krosnick JA & Presser S (2010) Question and questionnaire design. In: *Handbook of Survey Research* [Wright JD & Marsden PV, editors]. San Diego, CA: Elsevier.
 27. Cunningham K, Headey D, Singh A & Rana PP (2017) Maternal and child nutrition in Nepal: examining drivers of progress from the mid-1990s to 2010s. *Glob Food Sec* **13**, 30–37.
 28. Levine S (2019) *Getting in, Dropping out, and Staying on: Determinants of Girls' School Attendance in Nepal*. In: Ullrich H. (eds) *The Impact of Education in South Asia. Anthropological Studies of Education*. Palgrave Macmillan, Cham. pp. 11–36. Springer International Publishing. Available at: http://dx.doi.org/10.1007/978-3-319-96607-6_2 (accessed at 8 August 2019).
 29. Altbach PG, Reisberg L & Rumbley LE (2009) *Trends in Global Higher Education: Tracking an Academic Revolution*. Paris: UNESCO.
 30. Imdad A, Yakoob MY & Bhutta ZA (2011) Impact of maternal education about complementary feeding and provision of complementary foods on child growth in developing countries. *BMC Public Health* **11**, S25.
 31. Manikam L, Lingam R, Lever I *et al.* Complementary feeding practices for south asian young children living in high-income countries: a systematic review. *Nutrients* **10**, 2072–6643.

Supporting information

Additional supporting information may be found online in the Supporting Information section at the end of the article.

Data S1. Questionnaire for mothers of infants aged 6–36 months.

PROFESSIONAL PRACTICE

Exploring the provision of diabetes nutrition education by practice nurses in primary care settings

C. Gianfrancesco¹  & M. Johnson² 

¹Sheffield Diabetes and Endocrine Centre, Sheffield Teaching Hospitals NHSF Trust, Sheffield, UK

²School of Health and Related Research, University of Sheffield, Sheffield, UK

Keywords

diabetes, diet, education, practice nurse, primary care.

Correspondence

C. Gianfrancesco, Sheffield Diabetes and Endocrine Centre, Sheffield Teaching Hospitals NHSF Trust, Herries Road, Sheffield, S5 7LU, UK.
Tel.: +44 0114 271416
E-mail: carla.gianfrancesco@nhs.net

How to cite this article

Gianfrancesco C., Johnson M. (2020) Exploring the provision of diabetes nutrition education by practice nurses in primary care settings. *J Hum Nutr Diet.* **33**, 263–273
<https://doi.org/10.1111/jhn.12720>

Abstract

Background: High-quality nutrition education is recommended as an essential component of diabetes care. In the UK, there has been a gradual shift of inter-professional boundaries with respect to providing nutritional care for people with type 2 diabetes. Only a minority now regularly receive advice from a dietitian. Instead, increased demands for nutrition education are being absorbed by practice nurses. The present study seeks to explore this situation through the views of practice nurses on the services that they provide and the issues they face.

Methods: A qualitative approach using semi-structured interviews was employed. Practice nurses were recruited using purposive sampling, and nine were interviewed. Data were analysed using the Framework Method. The Theoretical Domains Framework from the COM-B ('capability', 'opportunity', 'motivation' and 'behaviour') model of behaviour change, as increasingly employed to explore the behaviour of healthcare professionals, was used to further frame the findings.

Results: Practice nurses reported that ongoing diabetes nutrition education only took place at annual review appointments and was limited to 5–10 min. They described how they are expected to take on a more advanced role in diabetes nutrition education than they can provide and are becoming increasingly isolated in this role as a result of a lack of time, practical and informational support, and training standards and provision.

Conclusions: A range of service improvements led by dietitians, which focus on strengthening the working environment and enhancing professional support available for practice nurses who provide diabetes nutrition education, could improve quality of care and health outcomes in people with diabetes within current time restraints.

Introduction

The ongoing provision of dietary advice to people with diabetes (PWD) from diagnosis is recommended by the National Institute for Health and Care Excellence as an essential component of diabetes care⁽¹⁾. To be effective, this should be individualised and provided by healthcare practitioners with specific expertise and competencies in nutrition^(1,2).

As the incidence of diabetes has increased, previous recommendations that all PWD should be referred to a

registered dietitian (RD) for diabetes nutrition education (DNE) at diagnosis and annual review have become unachievable^(3,4) and new models of care have evolved. At diagnosis, DNE is provided within diabetes self-management education (DSME) by trained and quality assured educators including dietitians⁽⁵⁾. Primary care staff, in particular practice nurses (PN), now have a pivotal role in providing ongoing DNE.

Providing nutrition education (NE) for primary prevention has been regarded as a role of primary care teams for decades^(6–8), although NE for chronic disease

management such as diabetes is a more recent development. The role has evolved as the involvement of general practice in diabetes care^(9,10) has proved essential to maintain access to services with the increased incidence of diabetes in the UK^(11,12). Care provision is monitored and incentivised through the audit and the Quality Outcome Framework (QOF)⁽¹³⁾.

The critical role of PNs in the provision of diabetes care is well reported^(14–20), although there are no studies looking specifically at their role in the provision of DNE in the UK. Potential advantages to a model where PNs are the main providers of DNE include the convenience and continuity of care from trusted and influential healthcare professionals^(21–25), although its effectiveness is uncertain⁽²⁵⁾. Having sufficient time is an important factor in bringing about lifestyle change⁽²⁴⁾. However, a lack of available time is reported as being the greatest barrier to DNE⁽²⁶⁾ and general NE in primary care^(6,8). A minimum of 2 h of NE over 6 months is recommended for effective dietary change^(24,27,28) and yet time for nutrition consultations is reported to be falling within the range of 4–25 min^(29,30). The time required to undertake physical checks and tasks for QOF could be contributing to this.

An alternative approach is for PNs to provide only brief interventions, which re-enforce more expert and detailed advice provided by other sources⁽²³⁾. However, this requires primary care to refer PWD to other sources of NE, such as dietetics and DSME and, despite evidence that it is beneficial, referral discussions with PWD are reported to be infrequent and tentative^(26,30,31).

A UK competency framework outlines the minimum competencies required for non-dietitians who provide DNE⁽³²⁾. However, subsequent to the removal of DNE

related QOF indicator DM013, there is less incentive to meet these⁽³³⁾. Subsequently, the provision and uptake of DNE training to PNs remains opportunistic and *ad hoc*^(14,16) and knowledge and skills levels is variable⁽³⁴⁾.

In summary, relevant literature suggests that the rising numbers of PWD in the UK and changes in the way diabetes services are provided has led to PNs now being a main provider of DNE, yet little is known about this aspect of care. Therefore, the present study aimed to explore the views of practice nurses to find out more about the services that they provide and the issues they face.

Materials and methods

Because this is a relatively unexplored topic, a qualitative approach was chosen so that the findings could inform subsequent research^(35,36). Resources were limited as a result of the study being part of a Master's degree carried out by one of the investigators (CG). Therefore, only one professional group was chosen, and all the interviews and analysis were undertaken by the one researcher (CG). Semi-structured interviews were chosen to encourage individuals' own interpretations of the questions and to allow the iterative development of these over the study as new relevant topics emerge⁽³⁷⁾. These also allowed for representation of the diversity of the nurses and their workplace⁽³⁶⁾ as described in Table 1.

Ethical approval was granted by the University of Sheffield Research Management System (URMS 143367).

Setting

The study took place in a UK city with a population of 550 000 and a diabetes prevalence of 6.1%. The provision

Table 1 Description of general practice population and staff

Practice identifier	Participant identifier	Deprivation score* 1 to 5 (1 = lowest level)	Diabetes prevalence* (% of practice with diabetes)	% of patients with optimal blood glucose control*	Total number of patients in practice†	Number of staff GP : nurse : healthcare assistant
City average*			6.1	62		
P1	N1	4	8.1	66	9000	10 : 3 : 3
P2	N2	1	6.5	69	9500	9 : 3 : 3
P3‡	N3, N9	5	8.2	55	6500	4 : 2 : 3
P4	N4	3	7.6	51	8000	6 : 3 : 2
P5	N5	4	9.7	56	2600	1 : 1 : 0
P6	N6	4	7.1	64	8000	8 : 4 : NK
P7	N7	3	8.6	63	7500	7 : 5 : 2
P8	N8	2	7.2	63	18 000	NK : 6 : 2

NK, not known.

*Source⁽⁴⁰⁾.

†Estimated by nurse.

‡P3 employed two of the nurses N3 and N9.

of community dietitians for people with diabetes was 1.5 whole time equivalents.

Sampling

Taking into consideration the limited resource, the aim was to recruit a purposive sample of up to 10 PNs currently providing diabetes care from 88 general practices^(38,39).

To obtain a diverse sample, albeit within a relatively narrow group, a sampling frame that included 58 practices was produced based on demographic and national audit data across target practices available at the time⁽⁴⁰⁾ (see Supporting information, Data S1). From this, a typology of practices was used to select iteratively those agreeing to participate, enabling representation of a diverse range of characteristics⁽⁴¹⁾.

Recruitment

Of the 58 practices, permission to contact PNs was granted from 19 practice managers. Information letters from the Clinical Lead for Diabetes were sent out in two waves to assess response rates and to meet the recruitment target. From the respondents of the first wave, six interviews were arranged and completed. Subsequently, the diversity of the sample was reviewed, and further targeted recruitment then followed. In total, 10 nurses agreed to be interviewed.

Data collection

The interview topic guide is provided in the Supporting information (Data S2). Of the 10 interviews scheduled, nine went ahead with one nurse withdrawing because of a lack of time. Although the sample was small, there appeared to be no new information coming out of the final interviews.

Data analysis

The data were transcribed verbatim, anonymised and imported into NVIVO, version 10 qualitative software package (QSR International, Cambridge, MA, USA). Identifiers were pseudonymised. It was analysed using the Framework Method⁽⁴²⁾ which fits well with the study aims and timescale and provides transparency for the data analysis⁽³⁵⁾. Themes and subthemes were identified following categorisation within the software. During the late stages of analysis, the COM-B ('capability', 'opportunity', 'motivation' and 'behaviour') model of behaviour and associated Theoretical Domains Framework (TDF) shown in Figure 1 was identified as relevant to the category groupings. The model detailed in the Supporting

information (Data S3), recognises that behaviours (B) come about through the interaction of capability, opportunity, motivation (COM) and the TDF has 14 domains each linked to one of the COM components^(43–46). It is used most commonly to inform not only with respect to the implementation of healthcare interventions, but also at any stage of health research⁽⁴⁵⁾. This includes using it at the descriptive and analytical stages of qualitative research as has been done in the present study⁽⁴⁶⁾.

The TDF structure was found to be useful for capturing and presenting relevant data and developing explanatory accounts by providing an understanding of how the categories were related, supporting the meaning of explanations. Questioning the data was also an important part of developing explanations. Considerations included: how outcomes changed under different conditions; the strategies nurses were using in their work and the possible reasons behind these; and the impact of the environmental context they were working in.

Results

Characteristics of the nurses and practices

Of the final sample of nine nurses, all were female. Five were employed as PNs and their experience in this role ranged between one and 10 years (median average of 7 years). Four were employed at the higher grade of Advanced Nurse Practitioner (ANP) with practice nursing experience ranging from 10 to 26 years (median average of 24 years). An overview of the general practice demographics is provided in Table 1. Three of the practices (P1, P3 and P5) had high numbers of patients from black and minority ethnic groups. All of the practices had a higher prevalence of diabetes than the national or city average.

Care provision

In all practices, DNE occurred in one to one diabetes clinic appointments. This was always provided by nurses, although their grade varied between practices. ANP made autonomous decisions about diabetes management and diabetes medications, including insulin initiation. The role of PNs differed in that they followed care plans provided by GPs. Experience of working in diabetes ranged from a few months to 20 years. Newly diagnosed patients were offered on average three diabetes appointments over 3 months, each of 15–30 min in duration. For most with established diabetes, review appointments were offered annually and ranged from 10 to 30 min. The consultation was structured around meeting QOF indicators for diabetes using an electronic template as a prompt. Any remaining time, estimated to be 5–10 min, was spent on education, with nutrition and physical activity mentioned most often.

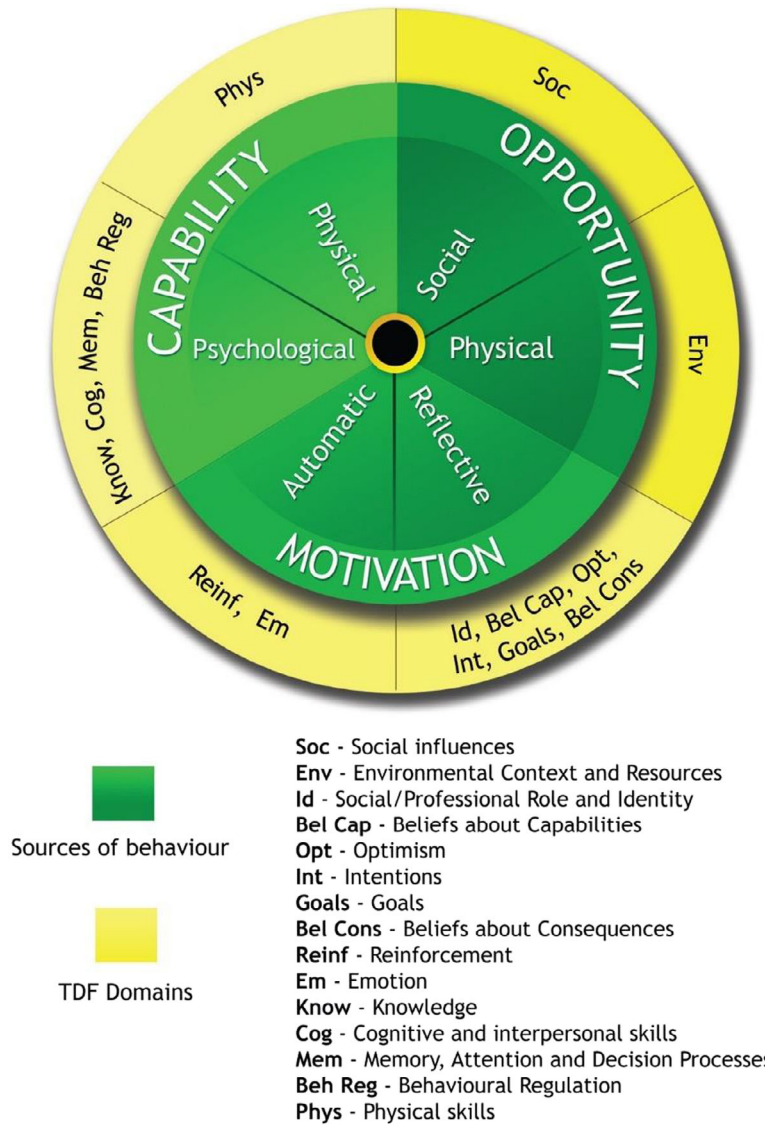


Figure 1 Theoretical Domains Framework linked to COM-B ('capability', 'opportunity', 'motivation' and 'behaviour') components. Reproduced with permission ⁽⁶²⁾.

Themes

Seven domains of the TDF were identified as relevant to this study and used as themes: environmental context and resources; social influences; knowledge; skills; professional role and identity; beliefs about capabilities; and beliefs about consequences.

Environmental context and resources

Nutrition education within the primary care diabetes service
 Despite attempts to free up more nursing time for education by utilising healthcare assistants to perform essential health checks, DNE provision was still estimated to be limited to 5–10 min annually. Meeting service demands

for people with diabetes was mentioned frequently and described as challenging and overwhelming. Nurses with the least support and skills felt the most time pressure when providing DNE.

‘We don’t have enough time. That’s quite important, I think. You know what I have to do in 10 minutes is not enough because that is the whole basis of diabetes when you think about it. It is diet. And if they can’t get that right you know it’s, you’re on a losing battle if you don’t spend time. Because we’ve got so much to do with them’ (N6)

Dietary educational resources were valued, and most nurses wanted a wider range of resources. There was no

consistency across practices in the choice of patient literature used and all preferred to print off these off from websites as required.

Nutrition education outside the primary care diabetes service

Nurses regularly referred PWD to DSME programme DESMOND (<https://www.desmond-project.org.uk/>) and the local weight management service. Positive patient feedback following attendance at these services increased referrals. Dietetic services, however, were rarely utilised. A number of reasons were given for this, including lack of awareness of dietetic service provision and the referral process; strict referral criteria; location of clinics; and high rates of non-attendance.

'I know, you know a few years ago they seemed to be more prominent, the dietitians and then again it's been a while since I've seen anyone and we never really seem to get to find out what services are available and what's no longer available in terms of things like that' (N8)

Social influences

Support from other healthcare professionals, mainly related to medical management, was identified as a main influence on nurses delivering diabetes care. Nurses had higher levels of job satisfaction and self-efficacy when supported by GPs, and Diabetes Teams.

'I feel really happy with the way it's organised here to be honest; I do. I think that's why if we do seem like a reasonably good practice it's because it is well organised and we do have good time for our diabetes patients and I get the debrief with doctor and we've got access to people outside the practice who are experts' (N1)

By contrast, inadequate support from the GPs isolated nurses, which impacted on their confidence.

'Yeah you know if somebody did question something and I was like I don't know but if I knew that I could go to somebody, one of them and ask. I mean we will send them messages and ask and see if they know the answer but a lot of the time they don't. So, then we are looking elsewhere for the answers' (N8)

Some practice staff felt they benefited from shadowing diabetes nurses in clinics, observing DSME and from opportunities to discuss complex cases at practice meetings with diabetes teams. Generally, nurses did not seek

out support specifically relating to DNE as a result of insufficient time and a lack of certainty about where to access it, although ANP were more likely to do so. Nurses with inadequate support expressed feelings associated with isolation, uncertainty and despair.

Knowledge and skills

All nurses made reference to shortfalls in their knowledge and skills to provide DNE. Commonly, nurses felt unable to answer patient questions on diet or provide sufficient detail, including practical suggestions. Having to respond with a 'best guess' had a negative impact on their self-efficacy.

'... when someone asks you how many slices of bread they should have a day and things like that. Because you don't have ongoing education and things sometimes, I think, "Do you know, I've got absolutely no idea". So, it affects your confidence quite a bit that you've got no idea' (N6)

Training in nutrition education

Training levels in diabetes care were highly variable and did not necessarily relate to the grade, position or length of experience of the nurse. No reference was made to the national dietetic competencies for healthcare professionals working in diabetes⁽³²⁾ and of those asked, none were aware of them. National recommendations for nurses to be skilled in DNE were not reflected in available local training. For example, the 12-month PN induction programme contained no education on diabetes or nutrition in relation to chronic disease management.

Although practice management supported requests for training, they did not actively propose any minimum levels of training. One nurse recalled this had been different in the past when dietary advice was briefly made a QOF indicator.

'So what we did then was, because we were saying, we can't legitimately tick that they've had the thing. So that's when I suggested, if I go and do the Blatcham diabetes thing [Diploma in Primary Care Management of Diabetes], because there's quite a big dietary section in that, you were able to give advice. So, we were sort of saying that I could educate other members of staff and would that count for our ...? Yes, we were sort of using me as a suitably qualified health professional' (N3)

Commonly, nurses obtained nutritional information from patient resources and personal experiences. Training by dietitians was helpful but was *ad hoc*, infrequent and felt too advanced to less experienced nurses. Nurses identified a need for tiered levels of formal and informal

training and support, provided locally and regularly, containing a practical element.

Professional role and identity

Aside from diabetes, primary care nurses provided nutrition advice to people of all ages for a range of reasons. Consequently, most nurses perceived providing DNE as their responsibility and did not seek out alternative sources of education for patients. It was suggested that this was also the expectation of patients and GPs. For some, this led to working at a more advanced level than they felt capable of. One exception to this was an ANP who described seeking alternative sources of NE for PWD to supplement what she could provide with her limited time and skills.

'I think I am a bit of a signpost person. So yes, I'll talk to people about healthy eating but I'll also see who else could get involved because I think it's important because they only see me fairly rarely so try and get as many people on board as possible actually' (N1)

With little contact with dietitians, there was a lack of clarity about the dietitian's role especially with less experienced nurses, which affected referrals.

'Probably not for diabetes. You know if there was somebody that had got an oesophageal cancer or something, we would refer them to dietitians for that, but it is really quite specific things that we would do that for' (N9)

Beliefs about capabilities

The awareness of gaps in their nutritional knowledge and skills, negatively influenced nurses' beliefs in their capabilities to provide DNE.

'Sometimes I'm at a loss to be fair. I've had patients that have just sat there and said well I don't like that and I'm not doing that. It's so difficult with some patients, very tricky. I just think and they go out and I just sit there and think 'Oh dear. Where am I going to go with this one?' This is where I want to find somebody, help!' (N4)

Yet other factors appeared to have a more positive impact. The most commonly reported one being the confirmatory messages they received from GPs and PWD indicating that they were the nutrition experts within the practice. Not having the opportunity to compare levels of competency with experts such as dietitians, appeared to

make it more difficult for nurses to reflect objectively on their own practice, also positively influencing belief in their capabilities.

Belief in consequences

All nurses held a strong belief in the impact of dietary change on the physical wellbeing of PWD and this appears to be one of the most significant factors motivating nurses to provide DNE.

'I can think of quite a few people who have lost weight and their diabetes has practically disappeared' (N1)

Discussion

Themes from the data are presented using the relevant domains of the TDF and associated COM-B model to describe the influences on DNE provided by nurses. This is presented in Figure 2. Nurse *capabilities* in NE were negatively affected by a lack of knowledge and skills in this area and a shortage of appropriate training provision. The absence of any mandate for training and competency standards in NE⁽⁴⁷⁾ may in part contribute to this situation. *Opportunities* to provide effective DNE were negatively affected by the high demands on nursing time, inadequate referral rates to alternative sources of NE, and inadequate support from within the primary care team and the wider diabetes specialist team, including dietitians. In many cases, this lack of support led to nurses being isolated, negatively impacting further on capability and opportunity. Unexpectedly, however, their *motivation* to provide DNE appeared to be unaffected by this possibly as a result of the situations that arose from it. Working in isolation led to nurses not being able to recognise what they did not know and compounded their perception of being solely responsible for DNE.

Comparison with the existing literature

This is the first UK study to specifically explore the role of PNs with respect to providing DNE. Similarities were found with other studies that examined primary care NE more broadly for disease prevention and chronic disease management.

The time reportedly spent on NE was similar to that reported in other studies and was significantly less than that suggested as adequate^(27,28). Despite this, there are advantages to receiving NE in primary care, such as improved uptake of care, good relationships, and short frequent appointments^(21–25). This study supports the

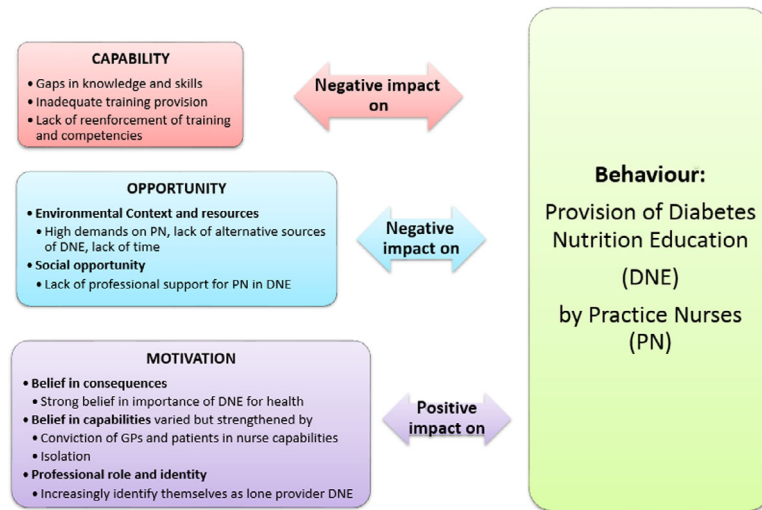


Figure 2 Theoretical Domains Framework domains identified as themes and their impact on Diabetes Nutrition Education.

suggestion that increasing levels of professional support could have the biggest impact on nurse behaviours and improve the quality of care within this current model⁽²⁹⁾. Yet, as identified elsewhere, this can be *ad hoc* and access is inequitable⁽¹⁴⁾.

Feelings of isolation experienced in particular by PNs compared to ANPs are recognised in other studies and are suggested to be a result of the wider range of duties of PNs compared to ANPs and the uncertainty that this brings to their roles⁽⁴⁸⁾. The lack of inter-professional working between nurses and dietitians identified in the present study and found elsewhere appears to worsen the situation for a number of reasons⁽²⁶⁾, including undefined professional roles and no clear pathway for when to refer on. Despite nurses recognising that they should only provide 'basic' care, this situation appears to lead to nurses providing DNE beyond their skill level^(34,49).

It was more likely for PNs to feel their level of knowledge and skills was insufficient for this role compared to ANPs. This diabetes knowledge gap is reported elsewhere^(26,49,50) as a consistent feature of nursing in a range of settings⁽³⁴⁾. The lack of courses was cited as the main obstacle to training; however, other studies highlight more barriers^(16,49,51). QOF incentives may have increased demand for training in DNE in the past, although, with this now removed and no current mandated minimum level of competency, these barriers will remain. Because significant numbers of nurses in primary care reach retirement age⁽⁵²⁾, an increasing proportion of inexperienced PNs and healthcare and physician assistants will provide diabetes care. Competency levels could therefore fall further if training levels remain unchanged.

Nurses' strong beliefs in the benefits of NE on health outcomes were evident in the present study. Subsequent

dissemination of the DiRECT study findings, which demonstrates that diabetes remission is attainable through weight loss, should strengthen this further^(53–55). As a result of DiRECT, services are re-designing diabetes care to introduce a local remission service. The trial used expert dietitians to provide training and ongoing support to dietitians and PNs who delivered the remission service in primary care. Our findings highlight how important the training and support element will be for the continued success of remission services outside of the trial.

Implications for future care

The present study suggests that the quality of DNE for people with type 2 diabetes in the UK could be improved by developing a robust support system for primary care nurses to improve the environment within which a PN provides DNE, as well as the range of support received.

The environmental context could be improved by agreeing an expected level of DNE provision in primary care through a local review of inter-professional roles and boundaries. This would raise awareness among GPs, support PNs to work within these boundaries and identify training and service needs. Resources that provide accessible, up-to-date information on services, referral criteria and processes, educational resources and patient information could improve referral rates and improving consistency of care⁽⁵⁶⁾.

Professional support through improved access to dietitians, including shadowing, responsive e-mail access for troubleshooting, service updates and coaching, could reduce isolation, increase knowledge of sources of NE outside of the practice and be a valuable opportunity for skill development particularly for less experienced nurses^(57,58).

An alternative model which fits with National Health Service strategic plans to make better use of wider primary care workforce is the expansion of dietitians into the primary care team⁽⁵⁹⁾ and this is currently being evaluated⁽⁶⁰⁾. Practice staff could be upskilled⁽⁵⁷⁾ and provide brief nutrition interventions to re-enforce advice from elsewhere, although the burden on the PNs described in the present study would reduce.

Nationally, more consideration could be given regarding how to reinforce training and competency standards. Education and training should continue to be developed in a variety of forms to meet the diverse needs of the primary care team⁽⁵⁷⁾. Innovative approaches, such as webinars, e-learning and the use of videos, as well as innovative face-to-face training, could improve uptake and support translating knowledge into practice^(33,34).

Strengths and limitations

The present study had several strengths. The broad research question was useful for this area that has not been previously researched. The iterative recruitment process led to a diverse sample for the study and, by selecting just one stakeholder, sufficient depth of data were generated to enable the development of explanatory accounts. Using the TDF as a framework for data analysis strengthened the study by providing an understanding of how the themes related to one another and a broader range of factors that impact on behaviour was considered. Although, when using such a model, there is a risk of only reporting data that fit the model (and losing data that don't), care was taken to ensure that all relevant information in the data was captured by at least one domain of the TDF.

The present study comprised a part of a degree and therefore resources were limited to one researcher, with this having the potential to introduce bias and limit the sample size. The Framework Method used was a systematic and thorough approach for the data analysis, which aimed to reduce personal opinion, increase rigor and provide transparency. In the development of explanatory accounts, the data were always revisited to verify or dismiss ideas, and empirical studies also supported the generation of hypotheses. Data saturation appeared to be achieved within the sample recruited. However, if the sample had been larger, it would have been more likely that nurses who regularly referred PWD to dietitians would have been recruited, providing another perspective.

Various factors affected the representativeness of the sample, including the willingness of practices to engage in the study. Practices with a greater interest in diabetes care were more likely to respond positively. With none of the

nurses given time during work to be interviewed, this suggests that participants were enthusiastic and committed to improving diabetes care. Finally, the present study took place in one geographical location in the UK. With local services for diabetes varying, the findings may not be directly transferable to other settings, although the details provided should assist the reader in assessing generalisability⁽⁶¹⁾.

Conclusions

It appears that, over time, with health service changes and increased service demands for diabetes care, there has been a shift in inter-professional boundaries with respect to providing DNE. Structured education is providing DNE for more of those who are newly diagnosed; however, primary care nurses are now one of the main providers of ongoing dietary advice to people with diabetes in the UK.

The present study has identified a number of concerns with the current situation that could be impacting on the effectiveness of ongoing diabetes nutrition education and, consequently, on morbidity and mortality in type 2 diabetes and the rising costs of diabetes. Nurses in the present study had become isolated in this role and this led to an expectation that they take on this role despite insufficient support, resources, time and the skills to do so.

However, the present study hypothesises that significant improvements could be made to the quality of nutrition education by primary care nurses as a result of making improvements to their working environment and the level of professional support through a range of strategies.

Conflict of interests, source of funding and authorship

The authors declare that they have no conflicts of interest.

This study was undertaken as part of a MSc Clinical Research funded by NIHR.

CG designed the study, performed the data collection and analysis, and was the main author of the paper. MJ provided supervision throughout the study and co-authored the paper. All authors critically reviewed the manuscript and approved the final version submitted for publication.

Transparency declaration

The lead author affirms that this manuscript is an honest, accurate and transparent account of the study being reported. The lead author affirms that no important aspects of the study have been omitted and that any discrepancies from the study as planned (University of Sheffield RMS143367) have been explained.

References

- NICE (2015) *Type 2 Diabetes in Adults: Management*. NICE Guidance NG28. London: National Institute for Health and Care Excellence.
- Dyson PA, Twenefour D, Breen C *et al.* (2018) Diabetes UK evidence-based nutrition guidelines for the prevention and management of diabetes. *Diabet Med* **35**, 541–547.
- Nutrition Advisory Committee of British Diabetic Association. (1990) Dietary recommendations for people with diabetes. An update for the 1990s. *Diabet Med* **9**, 189–202.
- NHS England (2012) *2010 Dietitians Workforce Survey report*. Available at: <http://www.dmeg.org.uk/Documents/DietitianWorkForceSurvey2012.pdf> (accessed 8 September 2015).
- NICE (2011) *Diabetes in adults | Quality Standards (QS6)*. NICE. Available at: <http://www.nice.org.uk/guidance/QS6> (accessed 1 September 2015).
- Kushner R (1995) Barriers to Providing nutrition counselling by physicians: a survey of primary care practitioners. *Prev Med (Baltim)* **24**, 546–552.
- Calfas KJ, Zabinski BA & Rupp JA (2000) Practical nutrition assessment in primary care settings: a review. *Am J Prev Med* **18**, 289–299.
- Moore H & Adamson AJ (2002) Nutrition interventions by primary care staff: a survey of involvement, knowledge and attitude. *Public Health Nutr* **5**, 531–536.
- Griffin S (1998) Diabetes care in general practice: meta-analysis of randomised controlled trials. *BMJ* **317**, 390–396.
- Pierce M, Agarwal G & Ridout D (2000) A survey of diabetes care in general practice in England and Wales. *Br J Gen Pract* **50**, 542–545.
- Gatling W (1985) The prevalence of diabetes mellitus in a typical English community. *J R Coll Physicians Lond* **19**, 248–250.
- Diabetes UK (2015) *Diabetes UK: key facts and stats*. 2015 report. Available at: <https://www.diabetes.org.uk/Documents/Positionstatements/FactsandstatsJune2015.pdf>. (accessed 1 December 2015).
- NHS England (2019) *NHS England » 2019/20 General Medical Services (GMS) contract: Quality and Outcomes Framework (QOF)*. Available at: <https://www.england.nhs.uk/publication/2019-20-general-medical-services-gms-contract-quality-and-outcomes-framework-qof/> (accessed 28 May 2019).
- Johnson M & Goyder E (2005) Changing roles, changing responsibilities and changing relationships: an exploration of the impact of a new model for delivering integrated diabetes care in general practice. *Qual Prim Care* **13**, 85–90.
- Campbell SM & Lester H (2008) The experience of pay for performance in english family practice: a qualitative study. *Ann Fam Med* **6**, 228–234.
- Gillibrand W, Taylor J & Hughes JG (2004) Practice nurses' views of their diabetes care. *Pract Nurs* **15**, 144–149.
- Lawton J, Parry O, Peel E *et al.* (2005) Diabetes service provision: a qualitative study of newly diagnosed Type 2 diabetes patients' experiences and views. *Diabet Med* **22**, 1246–1251.
- Beresford C (2011) Patients' perspectives of type 2 diabetes care by practice nurses: a qualitative study. *J Diabetes Nurs* **15**, 391–397.
- Johnson M & Hons RGN (2006) Patient perspective Understanding issues involved in the transfer of diabetes care to general practice: the patient perspective. *Qual Prim Care* **14**, 247–252.
- Daly B, Arroll B, Sheridan N *et al.* (2014) Diabetes knowledge of nurses providing community care for diabetes patients in Auckland, New Zealand. *Prim Care Diabetes* **8**, 215–223.
- van Weel C (2003) Dietary advice in family medicine 1–3. *Am J Clin Nutr* **77**, 1008–1010.
- Vermunt PWA, Milder IEJ, Wielaard F *et al.* (2012) Implementation of a lifestyle intervention for type 2 diabetes prevention in Dutch primary care: opportunities for intervention delivery. *BMC Fam Pract* **13**, 79.
- Moore H, Adamson AJ, Gill T *et al.* (2000) Nutrition and the health care agenda: a primary care perspective. *Fam Pract* **17**, 197–202.
- Sargent GM, Forrest LE & Parker RM (2012) Nurse delivered lifestyle interventions in primary health care to treat chronic disease risk factors associated with obesity: a systematic review. *Obes Rev* **13**, 1148–1171.
- Ball L, Hughes R, Desbrow B *et al.* (2012) Patients' perceptions of nutrition care provided by general practitioners: focus on Type 2 diabetes. *Fam Pract* **29**, 719–725.
- Jansink R, Braspenning J, van der Weijden T *et al.* (2010) Primary care nurses struggle with lifestyle counseling in diabetes care: a qualitative analysis. *BMC Fam Pract* **11**, 41.
- Evert AB, Boucher JL, Cypress M *et al.* (2014) Nutrition therapy recommendations for the management of adults with diabetes. *Diabetes Care* **37**(Suppl 1), S120–S143.
- Duke S, Colagiuri S & Colagiuri R (2009) Individual patient education for people with type 2 diabetes mellitus. *Cochrane database. Syst Rev* CD005268.
- Parry Strong A, Lyon J, Stern K *et al.* (2014) Five-year survey of Wellington practice nurses delivering dietary advice to people with type 2 diabetes. *Nutr Diet* **71**, 22–27.
- van Dillen SME, Noordman J, van Dulmen S *et al.* (2014) Examining the content of weight, nutrition and physical activity advices provided by Dutch practice nurses in primary care: analysis of videotaped consultations. *Eur J Clin Nutr* **68**, 50–56.
- Diabetes UK (2015) *Diabetes education : the big missed opportunity in diabetes care*. 1–8.

32. Diabetes UK Professional Education working (2011) *An integrated career and competency framework for dietitians and front line staff*. Available at: <https://www.diabetes.org.uk/Professionals/Training-competencies/Competencies/> (accessed 8 September 2015).
33. Diabetes UK (2013) *Nutrition and Physical Activity Briefing Document*. Available at: <https://www.diabetes.org.uk/Professionals/Training-competencies/Competencies/> (accessed 8 September 2015).
34. Alotaibi A, Al-Ganmi A, Gholizadeh L *et al.* (2016) Diabetes knowledge of nurses in different countries: an integrative review. *Nurse Educ Today* **39**, 32–49.
35. Mays N & Pope C (1995) Researching the parts other methods cannot reach: an introduction to qualitative methods in health and health service research. *BMJ* **311**, 42–45.
36. Bowling A (2009) *Research Methods in health: Investigating Health and Health Services*, 3rd edn. Maidenhead: Open University Press.
37. Britten N (2006) Qualitative interviews. In: *Qualitative Research in Health Care*. [Pope C & Mays N, editors]. Oxford: BMJ Books.
38. Baker SE & Edwards R *How many qualitative interviews is enough? Expert voices and early career reflections on sampling and cases in qualitative research*. Available at: http://eprints.ncrm.ac.uk/2273/4/how_many_interviews.pdf (accessed 30 August 2019).
39. Ball L, Davmor R, Leveritt M *et al.* (2016) The nutrition care needs of patients newly diagnosed with type 2 diabetes: informing dietetic practice. *J Hum Nutr Diet* **29**, 487–494.
40. Public Health England (2014) *Healthier Lives - Diabetes* [Internet]. Available at: <http://healthierlives.phe.org.uk/> (accessed 1 September 2015).
41. Silverman D (2013) *Doing Qualitative Research*, 4th edn. Thousand Oaks: SAGE Publications.
42. Ritchie J, Spencer L & O'Connor W (2003) Carrying out qualitative analysis. In: *Qualitative Research Practice: A guide for social science students and researchers*. [Ritchie J & Lewis J, editors] Thousand Oaks: SAGE Publications.
43. Cane J, O'Connor D & Michie S (2012) Validation of the theoretical domains framework for use in behaviour change and implementation research. *Implement Sci* **7**, 1–17.
44. Michie S, van Stralen MM & West R (2011) The behaviour change wheel: A new method for characterising and designing behaviour change interventions. *Implement Sci* **6**, 42.
45. Francis JJ, O'Connor D & Curran J (2012) Theories of behaviour change synthesised into a set of theoretical groupings: introducing a thematic series on the theoretical domains framework. *Implement Sci* **7**, 35.
46. Rubin SE, Davis K & McKee MD (2013) New York City physicians' views of providing long-acting reversible contraception to adolescents. *Ann Fam Med* **11**, 130–136.
47. Diabetes UK Professional Education working (2014) *Survey of English CCGs Approaches to Diabetes health professional education*. Available at: <https://www.diabetes.org.uk/Documents/Professionals/Competencies/SurveyofEnglishCCGsApproachestoDiabetesHealthProfessionalEducation.doc> (accessed 8 September 2015).
48. O'Donnell C & Jabareen HWG (2010) Practice nurses' workload, career intentions and the impact of professional isolation: A cross-sectional survey. *BMC Nurs* **9**, 2 (accessed 8 September 2015).
49. Martin L, Leveritt MD, Desbrow B *et al.* (2014) The self-perceived knowledge, skills and attitudes of Australian practice nurses in providing nutrition care to patients with chronic disease. *Fam Pract* **31**, 201–208.
50. Lambe B & Collins C (2009) A qualitative study of lifestyle counselling in general practice in Ireland. *Fam Pract* **27**, 219–223.
51. Renders CM, Valk GD, Griffin SJ *et al.* (2001) Interventions to improve the management of diabetes in primary care, outpatient, and community settings: a systematic review. *Diabetes Care* **24**, 1821–1833.
52. Royal College of Nursing (2011) *Who will care? Protecting employment for older nurses*. Available at: https://www.rcn.org.uk/__data/assets/pdf_file/0008/395378/003849.pdf
53. Leslie WS, Ford I, Sattar N *et al.* (2016) The Diabetes Remission Clinical Trial (DiRECT): protocol for a cluster randomised trial. *BMC Fam Pract* **17**, 20.
54. Lean MEJ, Leslie WS, Barnes AC *et al.* (2019) Durability of a primary care-led weight-management intervention for remission of type 2 diabetes: 2-year results of the DiRECT open-label, cluster-randomised trial. *Lancet Diabetes Endocrinol* **7**, 344–355.
55. Taylor R & Barnes AC (2019) Can type 2 diabetes be reversed and how can this best be achieved? James Lind Alliance research priority number one. *Diabet Med* **36**, 308–315.
56. Gianfrancesco C (2019) *Meeting the nutrition education needs of people with Type 2 Diabetes - NIHR CLAHRC YH. NIHR CLHARC Yorkshire and Humber*. Available at: <http://clahrc-yh.nihr.ac.uk/our-themes/translating-knowledge-into-action/4-projects/meeting-the-nutrition-education-needs-of-people-with-type-2-diabetes> (accessed 1 July 2019).
57. Beckingsale L, Fairbairn K & Morris C (2016) Integrating dietitians into primary health care: benefits for patients, dietitians and the general practice team. *J Prim Health Care* **8**, 372.
58. Beckingsale L, Fairbairn K & Morris C (2016) 'Two working together is so much better than just one': professional support needs of primary healthcare dietitians. *Nutr Diet* **73**, 220–228.
59. BDA Association of UK Dietitians (2017) *Dietitians in Primary Care*. Available at: <https://www.bda.uk.com/profe>

- ssional/influencing/bda_primary_care_paper.pdf (accessed 7 July 2019).
60. BDA Association of UK Dietitians (2011) *Research Brief: Proposal for research to show the impact of dietitians in the multi-disciplinary practice team to inform the development of a competency framework and training package for dietitians in GP practice*. Available at: https://www.bda.uk.com/professional/influencing/summary_or_primary_care_research_aims.pdf (accessed 7 July 2019).
 61. Mays N & Pope C (2000) Assessing quality in qualitative research. *BMJ* **320**, 50–52.
 62. Michie S, Atkins L & West R (2014) *The Behaviour Change Wheel: A Guide to Designing Interventions*. London: Silveback Publishing (accessed 7 July 2019).

Supporting information

Additional supporting information may be found online in the Supporting Information section at the end of the article.





Data S1. Study sampling frame (based on data available at the time of the study⁽⁴⁰⁾).

Data S2. Interview topic guide.

Data S3. Theoretical Domains Framework: Domain definitions, links to COM-B components, theoretical constructs and example questions⁽⁶²⁾.

PROFESSIONAL PRACTICE

Priority setting for adult malnutrition and nutritional screening in healthcare: a James Lind Alliance

D. J. Jones,¹  C. Baldwin,² S. Lal,³ E. Stanmore,¹  K. Farrer,⁴ E. Connolly,⁵ C. E. Weekes,⁶ L. Anderson,⁷ J. Murphy,⁸  L. Gillespie,⁹ N. Welsh,¹⁰ M. Ogden,¹¹ M. McDevitt,¹¹ R. Day,¹¹ S. Lynne,¹¹ P. Paulden,¹¹ T. Gronlund¹² & S. T. Burden¹ 

¹Faculty of Biology, Medicine and Health, University of Manchester, Manchester, UK

²Department of Nutritional Sciences, Kings College London, London, UK

³Salford Royal Foundation Trust, Salford, UK

⁴NHS Salford CCG, St James House, Salford, UK

⁵Age UK Salford, Eccles, UK

⁶Guy's & St Thomas' NHS Foundation Trust, London, UK

⁷Buckinghamshire Healthcare NHS Trust, Stoke Mandeville Hospital, Aylesbury Bucks, UK

⁸Bournemouth University, Bournemouth, UK

⁹The Christie NHS Foundation Trust, Manchester, UK

¹⁰Manchester University NHS Foundation Trust, Manchester, UK

¹¹Patient and Carer Involvement/Representatives, Manchester, UK

¹²James Lind Alliance, Southampton, UK

Keywords

healthcare research agenda, James Lind Alliance, malnutrition, nutritional screening, priority setting.

Correspondence

D. J. Jones, Jean McFarlane Building, University of Manchester, Oxford Road, Manchester, M13 9PL, UK.

Tel.: +44 (0)161 306 1508

E-mail: debra.jones@manchester.ac.uk

How to cite this article

Jones D.J., Baldwin C., Lal S., Stanmore E., Farrer K., Connolly E., Weekes C.E., Anderson L., Murphy J., Gillespie L., Welsh N., Ogden M., McDevitt M., Day R., Lynne S., Paulden P., Gronlund T. & Burden S.T. (2020) Priority setting for adult malnutrition and nutritional screening in healthcare: a James Lind Alliance. *J Hum Nutr Diet.* **33**, 274–283
<https://doi.org/10.1111/jhn.12722>

Abstract

Background: Malnutrition is one of the greatest global health challenges of our generation, leading to the increased utilisation of healthcare resources, as well as morbidity and mortality. Research has primarily been driven by industry, academia and clinical working groups and has had little involvement from patients and carers. The project described in the present study aimed to establish a priority setting partnership allowing patients, carers and healthcare professionals an opportunity to influence the research agenda.

Methods: A national survey was conducted to gather malnutrition uncertainties and identify key issues (i.e. areas within scope where an evidence-base is lacking) from those with experience of malnutrition. Uncertainties were analysed according to themes. Similar questions were grouped and summary questions were developed. A second survey was conducted and respondents were asked to choose their 10 most important summary questions. A workshop was conducted to finalise the top 10 research priorities from the most frequently indicated uncertainties on the interim survey.

Results: Overall, 1128 uncertainty questions were submitted from 268 people. The interim survey had 71 responses and a list of the top 26 questions was generated for the workshop. There were 26 questions discussed, ranked and agreed by healthcare professionals, carers and patients at the workshop. The top 10 research priorities were then chosen. These included questions on oral nutritional supplements, vulnerable groups, screening, community care, use of body mass index and technology.

Conclusions: The top 10 research priorities in malnutrition and nutritional screening have been identified from a robust process involving patients, carers and healthcare professionals.

Introduction

Malnutrition in all forms continues to be one of the greatest global health challenges of our generation ⁽¹⁾, with an estimated 462 million adults worldwide being identified as underweight ⁽²⁾. Every country in the world is affected by malnutrition in one form or another and, in the UK, malnutrition affects around 2.65 million people and costs the National Health Service £19.6 billion each year ^(3–5). Malnutrition usually manifests as nutritional deficiencies or excesses causing measurable, adverse effects on tissue or body form and function influencing clinical outcome ⁽⁶⁾. For the purposes of the present study, the term malnutrition will be used to refer to under nutrition only, which includes being underweight or having inadequate vitamins and minerals ⁽²⁾.

Nutritional screening is used in the majority of hospitals in the UK to detect malnutrition. The malnutrition universal screening tool ('MUST'), which is validated and recommended in national guidance ⁽⁷⁾, has been implemented in most hospitals throughout the UK subsequent to its development in 2003 ⁽⁶⁾. A recent survey ⁽⁵⁾ indicated that 29% of patients in hospital are identified as malnourished when assessed using 'MUST' ⁽⁸⁾. Despite the implementation of 'MUST' in hospitals, malnutrition is often unrecognised and is left untreated in many acute and community environments ^(9,10). However, there is evidence demonstrating that, when malnutrition is identified and treated appropriately, there are direct patient benefits in relation to morbidity and mortality shown in a variety of clinical situations and settings ^(11–13).

Patients and carers have had little opportunity to be involved in setting the research agenda in malnutrition ⁽¹⁴⁾ and, in general, healthcare research is often funded by industry, particularly by pharmaceutical companies in healthcare and academics ^(15–17). Healthcare research is often undertaken without taking the needs of patients and healthcare professionals into consideration ^(18–20).

The James Lind Alliance (JLA) is a non-profit making initiative that was established to encourage patients, carers and clinicians to come together to corroborate their joint needs and set research priorities ⁽²¹⁾. This type of collaboration is known within the JLA process as a: Priority Setting Partnerships (PSPs). The aim of these partnerships is to identify uncertainties, or 'unanswered questions', about the effects of a particular treatment or disease and then to prioritise those uncertainties. The JLA process can help to ensure that those who fund health research are aware of what really matters to both patients and clinicians. The priority setting process, which is supported by the National Institute for Health Research (NIHR), is systematic and transparent, and contributes to part of a widening approach to patient and public involvement in research.

The project described in the present study aimed to establish a JLA priority setting partnership to give patients, carers, and healthcare professionals the opportunity to influence research priorities in adult malnutrition.

Materials and methods

The Nutritional Screening and Malnutrition PSP was led by the University of Manchester and managed within JLA structures and processes ⁽²²⁾ and the methods were informed by JLA. A robust process was used to ensure that patients, carers and professionals were brought together from an equal position of power, without any hierarchy, to determine the priorities for research. The project was registered with JLA and followed prespecified JLA processes to establish research questions that were of direct relevance to patients and professionals. A JLA advisor supported and facilitated the process and ensured consistency throughout. The full JLA process included: (i) set-up of a steering group committee and agreement of the scope and protocol (see Supporting information, Appendix S1); (ii) gathering evidence from literature; and (iii) a four step priority setting process (1, gathering uncertainties; 2, organising uncertainties into themes; 3, interim priority setting; and 4, final priority setting) (Figure 1).

Set-up of the steering group committee and project partners

Potential healthcare and charitable partner organisations, which could provide access to a wide range of participants, were identified and invited to be involved in the PSP. Organisations that accepted were partnered and agreed to promote work and progression. Potential steering group members with experience of malnutrition across a broad range of settings, including patients and carers, were also identified and contacted. Steering group members from a healthcare background were identified by purposive sampling, where key organisations and individuals with a specific focus on malnutrition were contacted. A list of these organisations is provided in the Supporting information (Appendix S2). Advertisements were used to purposively recruit patients and patient carers to become steering group members using patient-specific websites, including 'People in Research' (<https://www.peopleinresearch.org/>) and 'Salford Citizen Scientist' (<http://www.citizenscientist.org.uk/>). Patients and carers who were interested, contacted the research team directly and those who became members were offered hourly payments for their time.

A steering group was established and met quarterly throughout the project. To engage members of the public

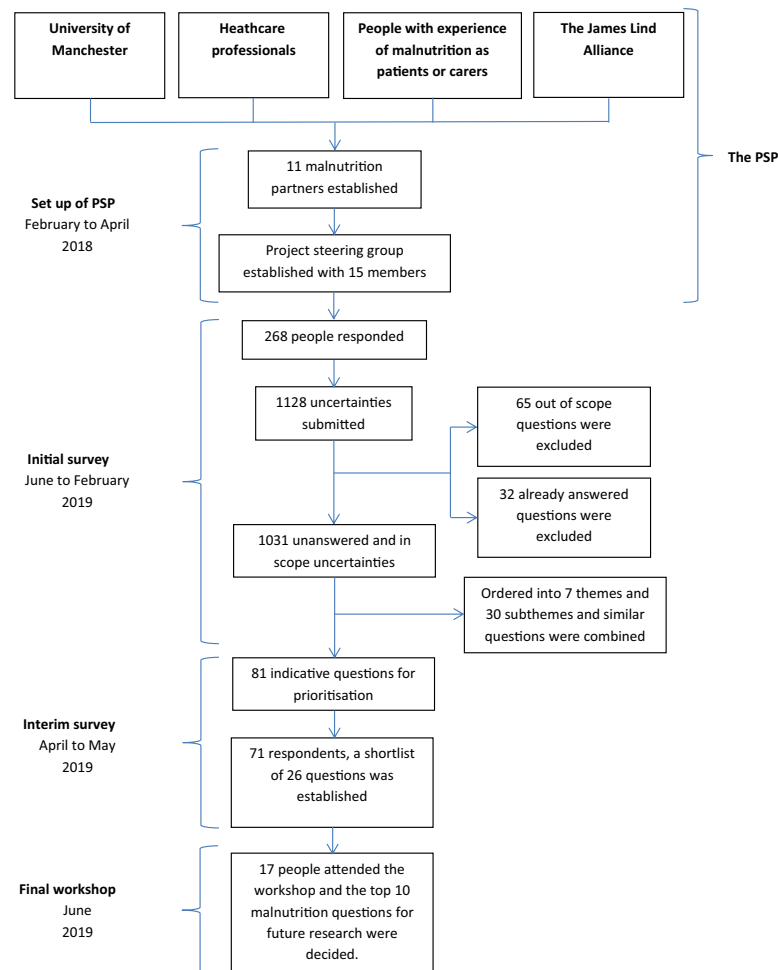


Figure 1 Flow diagram of responses to the surveys and the final workshop. PSP, Priority Setting Partnership.

and partners; A twitter account was set up under the name of @MalnutritionPSP and was used for promoting activities and progress.

Gathering evidence from the literature

In addition to gathering malnutrition uncertainties, this project also gathered data from existing literature and carried out an umbrella review. Details of this are available in the PROSPERO registered protocol CRD42018094702 (https://www.crd.york.ac.uk/prospero/display_record.php?RecordID=94702). This allowed us to understand current evidence and to determine whether the uncertainties submitted had already been answered within the published literature. Uncertainties that were considered to be out of scope, as well as the reasons why, including uncertainties that had already been answered by previous research, are provided in the Supporting information (Appendix S3).

Four step priority setting process

Gathering uncertainties

A survey was created by the steering group members, using the example surveys provided by JLA from previous PSPs. An initial first draft of the survey was created and all steering group members reviewed and commented on the layout, content and wording. In particular, lay members of the steering group were able to provide useful feedback on the clarity, acceptability and comprehension of the survey.

Broad question categories were provided in the survey to guide participants and ease submission of their uncertainties; these broad categories were based on similar categories used by previous PSPs. The exact layout and phrasing of the survey was redefined after a pilot survey involving nine people diagnosed with malnutrition, five malnutrition healthcare professionals and five people with a general interest in malnutrition.

After development and piloting, the survey was distributed nationally to gather 'uncertainties' in malnutrition from those with any kind of experience of malnutrition including patients, carers, healthcare professionals and academics. Partners promoted the survey and we advertised online through 'People in Research', 'Salford Citizen Scientist', 'University of Manchester' and @malnutritionPSP twitter account. Participants included those aged 18 years and over with lived-experience of malnutrition, either as a patient, carer, health and social care professional, nonclinical researcher or representative of a patient group. Respondents were asked about three main areas in malnutrition: Screening and identification; treatment and prevention; and different settings or different patient groups. Respondents could also respond on any other areas they felt were relevant. As a result of a low response rate from patients and carers and because of feedback from some healthcare professionals that the term 'malnutrition' was not necessarily understandable by patient groups, a second version of the survey was created. This version removed the word 'malnutrition' and used 'patient friendly' language, including the phrases 'eating less', 'poor appetite' and 'unplanned weight loss'. The second version was developed and reviewed by the steering group using the same process as the first version. Because of time constraints, we were unable to pilot this version. Both the original version and second version were made available online and as paper-based copies with prepaid reply envelopes.

Organising uncertainties

The analysis of the initial survey data followed methods of data processing of uncertainties from JLA⁽²³⁾. This was a detailed process involving the steering group, where ratification and confirmation were required for each key procedural point to ensure accountability and transparency.

The consultation process (initial survey) produced 'raw' unanswered questions about the diagnosis and effects of treatments for malnutrition. These raw questions were entered into an electronic database verbatim. Data were then assembled, categorised and refined. Questions were first grouped according to themes based on keyword phrases and then similar questions were combined into one 'collated indicative question'. All indicative questions were written in a format that would be clear, amenable to being answered by research design methods, and comprehensible to all. The steering group was informed about questions and themes that were emerging and asked to verify. Once the list of uncertainties had been developed, the steering group ratified by checking off duplicates and ensuring adherence to the prespecified project scope. A clear inclusion and exclusion

criteria was applied to determine any out of scope uncertainties. This criteria was specified *a priori* in the protocol. Systematic reviews and guidelines were identified by the umbrella review and uncertainties were cross-checked with the evidence base.

Indicative questions that had not been adequately addressed by previous research were collated and recorded as an interim list by the data management team and checked and confirmed by the steering group.

Interim priority setting

A second interim survey was created using the list of indicative questions from the initial survey. This survey was a straight forward list of the indicative questions and therefore no layout design or pilot was required. A few sentences, agreed by the steering group, were constructed at the start of the survey to provide brief instructions to users. Participants of this survey were presented with a list of questions and asked to select their 10 most important indicative questions.

The aim of the second survey was to begin prioritisation of identifying uncertainties. The number of times a question was selected by any given participant was recorded, and so questions could be ordered according to popularity. The data were split into two groups: (i) patient, carers and patient representatives and (ii) healthcare professionals, allowing for recognition of the top priorities for each group. Responses were analysed according to frequency and a list of the top questions was generated to go forward to a final workshop.

The survey was only available online as a result of the nature of the survey, which involved a long list of questions. It was considered that this could not be appropriately managed on paper and would be overwhelming to the user. The distribution channels mirrored those used in the first survey. In addition, respondents from the first survey were contacted directly if details had been provided. However, because the responses were anonymous, we were unable to track the number of participants that responded to both surveys.

Final priority setting

The final workshop was arranged and a balance of patients, carers and clinicians was invited to agree the top 10 priorities. The same routes of promotion were used as in the surveys.

The workshop was facilitated by three JLA advisors to ensure transparency, accountability and fairness when discussing the questions. The prioritisation exercise was organised over a full day and participants were provided with the 26 questions in advance. Selection and prioritisation of the top 10 questions during the workshop included discussions, group work and use of question

cards for ranking using; the nominal group technique⁽²⁴⁾. Staff from an independent living service contributed to the process remotely prior to the workshop. This contribution involved participants ranking the questions in order of importance with reasons and then stating their top and bottom three questions. This feedback was integrated into the discussions at the workshop by a representative for each small group discussion.

Ethical approval

The surveys and group work within this PSP were considered to be patient and public engagement activities, which do not require formal ethical approval. However, to ensure the safeguarding of participants, we established standard operating procedures for collecting information. All surveys were answered anonymously by participants. Personal details of participants provided were handled in accordance to the University of Manchester privacy policy and a privacy notice was included on the survey. Respondent's personal details were password protected and stored securely in line with the Data Protection Act and data management policy of the University of Manchester. All data collected from the surveys were stored on an anonymised, electronic database.

Results

Set-up of the steering group committee and project partners

The project's partners were established and agreed between February to April 2018 and included: The British Association for Parenteral and Enteral Nutrition (BAPEN), The British Dietetic Association (BDA), The Malnutrition Action Group (MAG), Macmillan, Hertfordshire Independent Living Service (HILS), Salford Royal NHS Foundation Trust, Age UK Salford, Malnutrition Task Force, Wessex Academic Health Science Network (WAHSN), National Care Association and Care England. The main role of the partners was to support the project and promote the surveys to relevant members of staff and patient groups.

The project steering group included: dietitians ($n = 4$), nutrition nurse specialists ($n = 2$), a voluntary sector representative, a gastroenterologist, nutrition and healthcare professionals and lecturers ($n = 2$), a patient representative, patients with experience of malnutrition ($n = 2$) and carers ($n = 2$).

Gathering evidence from the literature

The umbrella review of the systematic reviews was completed and used alongside the four step prioritisation process.

Four step priority setting process

Gathering uncertainties

The first survey was launched in June 2018 and was open for 8 months. Overall, 1128 uncertainty questions from 268 people were submitted. This including 194 professionals and 74 patients and carers; 86% were female and 78% were White British. The age range (median) was 21–93 (45) years and respondents were located across the UK, plus a few international locations (Table 1).

Organising uncertainties

Of the submitted uncertainties the steering group deemed that 65 were out of scope and 32 had already been answered (Figure 1). The remaining 1031 uncertainties were categorised into seven themes and then into 30 different subthemes (Table 2). Questions with repeating items were grouped together and formed into one

Table 1 Participant characteristics

	Initial survey ($n = 268$)	Interim survey ($n = 71$)
Gender n (%)		
Female	231 (86)	56 (79)
Male	29 (11)	5 (7)
Transgender	0 (0)	1 (1.5)
No response	8 (4)	9 (12.5)
Age (years)		
Range	21–93	19–77
Mean (SD)	46.19 (13.7)	46.41 (12.5)
Location n (%)		
East Midlands	7 (3)	4 (5.5)
East of England	23 (9)	4 (5.5)
London	23 (9)	7 (10)
North East	7 (3)	3 (4)
North West	52 (19)	18 (25)
Northern Ireland	2 (1)	0 (0)
Scotland	10 (4)	2 (3)
South East	66 (24)	12 (17)
South West	25 (9)	5 (7)
Wales	11 (4)	2 (3)
West Midlands	17 (6)	2 (3)
Yorkshire and The Humber	9 (3)	2 (3)
Canada	1 (0.3)	0 (0)
Australia	2 (0.7)	0 (0)
Africa	3 (1)	0 (0)
No response	10 (4)	10 (14)
Ethnicity n (%)		
Asian	11 (4)	3 (4)
Black African	5 (2)	0 (0)
Black Caribbean	1 (1)	1 (1.5)
White British	209 (78)	43 (60.5)
White Irish	9 (3)	4 (6)
Other	24 (9)	10 (14)
No response	9 (3)	9 (12.5)

Table 2 Themes and subthemes for analysis of the submitted uncertainties

7 main themes	30 subthemes	Number of respondent uncertainties
Assessment, diagnosis, symptoms	Improvements in screening	61
	Improvements in community screening	29
	Accuracy of screening	93
	Recognising malnutrition	36
	Screening tools	106
Treatment	Impact of treatment	75
	Treatment cost effectiveness	12
	Treating specific groups	61
	Access to treatment	27
	Treatment type and delivery	85
Prognosis	Evidence of effective care	14
	Impact of malnutrition	20
	patient opinion	11
	Approach for different groups	19
Self-management	Communication	22
	Behaviour change/ nutrition across lifespan	27
	Impact on carers	4
Health services	Staff confidence and training	58
	Follow-up/community care	47
	GP awareness	11
	Standard practices	25
	Assessment in GP surgeries	6
Prevention	Food preferences and access	15
	Food quality and education	40
	Food provision	14
	Prevention programmes	58
Social care, mental health	Dependency issues	6
	Deprived groups	20
	mental health and malnutrition link	20
	Social support	9
	Total number of uncertainties	1031

GP, general practitioner.

indicative question. In total 81 indicative questions were created and put forward for the second interim survey.

Interim priority setting

The interim survey was conducted in April 2019 and asked people to choose which 10 questions in the long list of uncertainties were most important to them. During this survey 71 people responded; of these, 53 were healthcare professionals and 18 were patients and carers and 79% were female and 60.5% were White British. The age range (median) was 19–77 (48) years and respondents were located across the UK (Table 1). The top 15 questions from patients and carers were put forward to the

final list for the workshop. As a result of equal positions of questions selected by healthcare professionals, only the top 13 were selected and taken forward. The next four questions from healthcare professionals were in an equal position at 14. The steering group were asked to determine the order of priority of these four questions and decided that one question was important enough to be carried forward, creating a final list of 26 questions for the workshop.

Final priority setting

The final workshop was held in Manchester in June 2019. The workshop was facilitated by three James Lind advisers and was attended by 17 people from across the UK, including dietitians ($n = 5$), a speech and language therapist, a policy officer for the BDA, a nurse practitioner, a Dietetic Assistant, a Macmillan Project Dietitian, voluntary sector representatives ($n = 2$), patients ($n = 3$) and carers ($n = 2$). Seven people attended the separate meeting for the independent living service, with this including four community dietitians, a registered nutritionist, a member of the administration team and a team member with experience of malnutrition as a carer.

During the workshop, attendees were split into three groups and each group was managed by one of three JLA advisors. The groups separately agreed and ranked the 26 questions for importance. Groups were then mixed up and questions were ranked again. Ranking was recorded from all group sessions and a final overall ranked list was created (Table 3). The workshop finished with a whole group discussion and the final top 10 priorities were decided and agreed (Figure 2).

Discussion

The aim of the partnership was to address the uncertainties of those with experience of malnutrition, and to prioritise the most important uncertainties that should be addressed by research. The partnership brought together people with a wide range of malnutrition experiences, including those working in the area and those affected by the condition. The project was a valuable opportunity for people with lived experience and professionals to work together and shape the research agenda for malnutrition in adults.

Over 300 respondents from across the UK were involved in the two stages of identification and prioritisation of malnutrition uncertainties. Respondents represented those living across the UK and included responses from a wide age range. The majority of respondents were White British, although both stages of the prioritisation process saw representation from Asian, Black African, Black Caribbean and White Irish backgrounds.

Table 3 The top 26 research questions discussed at the final workshop, including final ranking, ranking at the interim priority stage and original theme allocation

Final rank	Interim survey rank		Original theme	Question
	HCPs	Patient/carers		
1	16	11	Treatment	How can early intervention be initiated in vulnerable groups to help prevent malnutrition?
2	23	8	Assessment	What is the best way to carry out screening in the community for effective identification of malnutrition
3	15	51	Prognosis	Are people/patients aware of malnutrition, do they know how to prevent it and do they think screening for malnutrition is important?
4	11	19	Health services	Should multi-agency working be implemented across all care settings to ensure standardised identification and treatment of malnutrition?
5	2	47	Treatment	How useful are nutritional supplements, are there alternatives and should independent research into supplements be carried out, rather than research lead by pharmaceutical companies?
6	9	20	Assessment	Is it appropriate and accurate to use standard BMI ranges to diagnose malnutrition in elderly people, dementia patients, bed-bound patients and patients with fluid imbalances?
7	57	4	Prevention	What other types of community support could be used to prevent malnutrition? For example, surplus from supermarkets and restaurants
8	12	25	Treatment	Is there evidence that treating malnutrition in the community would lower hospital admissions and length of stay?
9	7	36	Treatment	Can technology and electronic records be used to record and improve nutritional treatments and to convey nutritional advice?
10	69	15	Treatment	Should treatment be geared to specific disease states and patient groups (e.g. high-fat diets can be inappropriate in cardiovascular disease)?
11	13	–*	Prognosis	Does social interaction, the use of volunteers and more sociable mealtimes in hospitals improve patient's food intake?
12	62	10	Health services	Would viewing malnutrition as more of a medical condition help with prevention?
13	1	40	Treatment	How beneficial is the food first approach (meeting dietary needs through ordinary foods and fluids), is it used often enough, does it need promoting more and what advice should the food first message convey?
14	36	14	Prognosis	Is malnutrition getting worse over time and is this related to poverty and affordability?
15	4	1	Social care/ mental health	How can we help, treat and support those with mental health issues, or dementia, or those who refuse to eat, and how does their mental wellbeing impact on nutritional status?
16	3	38	Treatment	Is dietitian led treatment more effective in terms of outcomes and should their input increase?
17	6	–*	Health services	Are nutrition telephone consultations with patients as effective as home visits?
18	5	3	Social care/ mental health	What is the extent of malnutrition in deprived/low income/hard to reach groups and what is the best way to provide them with nutritional support, education and good quality food?
19	28	5	Health services	What is the standard practice after malnutrition has been identified and is this carried out routinely and effectively?
20	70	13	Treatment	How to decide if more invasive treatment is required and if certain patients should be prioritised over others?
21	44	6	Prognosis	Can good nutrition, nutritional support and nutritional intervention lead to health improvements and decreased length of stay in hospital?
22	27	7	Self-management	Has the ongoing public health campaign against obesity caused confusion and encouraged disordered eating and can we help people who are struggling with changes in eating behaviour?
23	79	12	Prevention	What is the best healthy eating advice and how should this be communicated to patients at home and people at home who may be at risk of malnutrition?
24	77	9	Self-management	Is decreased appetite in the elderly down to the aging process?
25	8	58	Health services	What are the views of healthcare provider, local authority and commissioners on malnutrition? Are they aware of cutbacks and can commissioners be more engaged to provide quality assurance (CQUIN)?
26	10	2	Assessment	How accurate and valid are current nutritional screening tools?

BMI, body mass index; CQUIN, Commissioning for Quality and Innovation; HCPs, healthcare professionals.

*Question not selected during the interim survey.

- How can early intervention be initiated in vulnerable groups to help prevent malnutrition?
- What is the best way to carry out screening in the community for effective identification of malnutrition?
- Are people/patients aware of malnutrition, do they know how to prevent it and do they think screening for malnutrition is important?
- Should multi-agency working be implemented across all care settings to ensure standardised identification and treatment of malnutrition?
- How useful are nutritional supplements, are there alternatives and should independent research into supplements be carried out, rather than research funded by pharmaceutical companies?
- Is it appropriate and accurate to use standard BMI ranges to diagnose malnutrition in elderly people, dementia patients, bed-bound patients and patients with fluid imbalances?
- What other types of community support could be used to prevent malnutrition?
- Is there evidence that treating malnutrition in the community would lower hospital admissions and length of stay?
- Can technology and electronic records be used to record and improve nutritional treatments and to convey nutritional advice?
- Should treatment be adapted depending on the disease and patient group (e.g. high-fat diets can be inappropriate in cardiovascular disease)?

Figure 2 The top 10 priorities from the malnutrition priority setting partnership. BMI, body mass index.

The final workshop culminated with a list of the top 10 research priorities for malnutrition. The priority considered to be of most importance was ‘early intervention in vulnerable groups to help prevent malnutrition’ and the second most important was asking ‘what is the best way to carry out screening in the community’. This highlights the need to reach vulnerable groups in the community and to be able to identify issues before they develop. This would include: practical ways of measuring nutritional status and body composition validated against criterion measurements⁽²⁵⁾; improvements in communication after discharge⁽²⁶⁾; and adaptation of the current screening methods according to the circumstances⁽²⁷⁾. Improvements in this area have already been made with novel developments in self-screening tools that are more suitable for community use in the voluntary sector domiciliary care and with social care partners. The PaperWeight Armband™ is a public health signposting tool created in Salford and being piloted in five boroughs in Greater Manchester⁽²⁸⁾. Also, new tools have been developed for the earlier identification of malnutrition risk around a conversation for the wider workforce including volunteers and family carers, such as the Patients Association

Nutrition Checklist⁽²⁹⁾ and an interactive version such as the ‘Nutrition Wheel’⁽³⁰⁾. However, further work is imperative in this area to raise awareness, increase recognition of malnutrition, and understand what works for the vulnerable and in the community.

Other topics covered by the top 10 priorities included: oral nutritional supplements, screening, community care, use of body mass index and use of technology (Table 3). These results can now be used to help funders identify important priorities for future research, which are relevant to both healthcare professionals and patients and their carers. This will potentially lead to more valuable research in malnutrition, including screening and assessment, because it will clearly identify research questions that will be of use in clinical practice and also be meaningful to patients, carers and members of the profession of dietetics. There are many examples where research priorities, identified in the top 10 by a JLA PSPs, have resulted in research funding and projects, including Crohn’s PSP, colitis PSP and Palliative care PSP⁽³¹⁾. It is hoped that the valuable work undertaken for this PSP will lead to similar successes and outcomes.

The limitations of this project include the limited numbers of people recruited from diverse ethnic backgrounds who would have been able to read and write in English to complete the surveys. Therefore, the results of this project may be more representative of the White, English-speaking population. Future PSPs should consider engaging with more diverse ethnic groups, including Black, Asian and Minority Ethnic communities, and creating surveys in multiple languages that would also be of benefit to support engagement. In addition to this, there were fewer patients responding to the surveys than healthcare professionals, and we found that there appeared to be a lack of understanding around the risk of malnutrition, malnutrition and the definition of malnutrition. Similarly, fewer patients and carers attended the final workshop, which may have provided a disproportionate representation of the patient and carer populations. It is also worth noting that malnutrition crosses all diseases and so it is difficult to assess patients readily in the way that a PSP for a specific condition may be able to do.

Another limitation was the exclusion of children and adolescents because this was considered to be a separate but just as important issue, which would require its own PSP. Therefore a future PSP could also be considered for childhood and adolescent malnutrition in the UK.

Feedback from this PSP will be provided to all of the patient groups, healthcare professionals and organisations that have been involved and that could also be involved in future funding and research. It is the aim of this PSP to influence the national agenda and so the results will be made available to funding and research agencies to assist with setting research priorities and funding calls on malnutrition and screening.

Conclusions and dissemination

After conducting the JLA PSP, we now have 10 research priorities identified in malnutrition and nutritional screening from a robust process involving both healthcare professionals and patients and carers. It is anticipated that these results will be used nationally to inform the research agenda in malnutrition and nutritional screening in adults.

Acknowledgments

The nutrition team at the University of Manchester would like to acknowledge the hard work, time and commitment dedicated to this project by: the JLA and the project JLA advisors; the project steering group members; the project partners; and the attendees of the final workshop.

Conflict of interests, source of funding and authorship

The authors declare that they have no conflicts of interest. This project was funded by the General and Educational Trust (GET), which is part of the British Dietetic Association (BDA) and was conducted in partnership with James Lind Alliance. DJ was the information specialist, data management specialist and was involved in the writing of the manuscript. CB, SL, ES, KF, EC, CEW, LA, JM, LG, NW, MO, MM, RD, PP, and SL contributed to the development and delivery of the project, as well as the reviewing and writing of the manuscript. SB was principle investigator and project manager, and was involved in the writing of the manuscript. All authors critically reviewed the manuscript and approved the final version submitted for publication.

Transparency declaration

All authors affirm that this manuscript is an honest, accurate and transparent account of the study being reported. The reporting of this work is compliant with PRISMA guidelines. The lead author affirms that no important aspects of the study have been omitted and that any discrepancies from the study as planned with James Lind Alliance and as stated in the protocol have been explained.

References

1. WHO (2018) *The Nutrition Challenge: Food system solutions*. Available at: <https://www.who.int/nutrition/publications/policies/nutrition-challenge-food-system-solution/en/> (accessed 17 June 2019).
2. WHO (2018) *Fact sheet: Malnutrition, Geneva*. Available at: <https://www.who.int/news-room/fact-sheets/detail/malnutrition> (accessed 17 June 2019).
3. Russell CA & Elia M (2008) *Nutritional Screening Survey in the UK in 2008*. Redditch: BAPEN.
4. Elia M (2015) The cost of malnutrition in England and potential cost savings from nutritional interventions. Malnutrition Action Group of BAPEN and the National Institute for Health Research Southampton Biomedical Research Centre. <https://www.bapen.org.uk/pdfs/economic-report-short.pdf> (accessed November 2019).
5. Stratton R, Smith T & Gabe S (2018) *Managing malnutrition to improve lives and save money*. BAPEN. Available at: <https://www.bapen.org.uk/pdfs/reports/mag/managing-malnutrition.pdf> (accessed 17th June 2019).
6. Elia M (2003) *The 'MUST' Report. Nutritional screening of adults: a multidisciplinary responsibility. Development and use of the 'Malnutrition Universal Screening Tool' ('MUST') for adults*. Available at: <http://www.bapen.org.uk/screening-and-must/must/must-report/the-must-report-executive-summary> (accessed 17th June 2019).

7. NICE (2012) *Nutrition support in adults. Quality standard [QS24]*. National Institute of Clinical Excellence. Available at: <https://www.nice.org.uk/guidance/qs24/chapter/Quality-statement-1-Screening-for-the-risk-of-malnutrition> (accessed 17 July 2019).
8. Russell CA, Elia M & on behalf of BAPEN and collaborators (2014) *Nutrition screening survey in hospitals in the UK, 2007–2011*. Available at: <https://www.bapen.org.uk/pdfs/nsw/bapen-nsw-uk.pdf> (accessed 17 June 2019).
9. Barker LA, Gout BS & Crowe TC (2011) Hospital malnutrition: prevalence, identification and impact on patients and the healthcare system. *Int J Environ Res Public Health* **8**, 514–527.
10. Brotherton A, Simmonds N, Stroud M *et al.* (2010) *Malnutrition Matters Meeting Quality Standards in Nutritional Care*. Available at: https://www.bapen.org.uk/pdfs/bapen_pubs/mm-toolkit-exec-summary.pdf (accessed 02 January 2019).
11. Burden ST, Gibson DJ, Lal S *et al.* (2017) Pre-operative oral nutritional supplementation with dietary advice versus dietary advice alone in weight-losing patients with colorectal cancer: single-blind randomized controlled trial. *J Cachexia Sarcopenia Muscle* **8**, 437–446.
12. Capra S, Ferguson M & Ried K (2001) Cancer: impact of nutrition intervention outcome and nutrition issues for patients. *Nutrition* **17**, 769–772.
13. Walton K, do Rosario VA, Pettingill H *et al.* (2019) The impact of home-delivered meal services on the nutritional intake of community living older adults: a systematic literature review. *J Hum Nutr Diet* <https://doi.org/10.1111/jhn.12690>
14. Elia M, Wheatley C & on behalf of BAPEN and collaborators (2014) *Nutritional Care and the Patient Voice: are we being listened to?* Available at: <https://www.bapen.org.uk/pdfs/nutritional-care-and-the-patient-voice.pdf> (accessed 19 June 2019).
15. Tallon D, Chard J & Dieppe P (2000) Relation between agendas of the research community and the research consumer. *Lancet* **355**, 2037–2040.
16. Delaney B (2006) Commentary: is society losing control of the medical research agenda? *BMJ* **332**, 1063–1064.
17. Patsopoulos NA, Analatos AA & Ioannidis JPA (2006) Origin and funding of the most frequently cited papers in medicine: database analysis. *BMJ* **332**, 1061–1063.
18. Chalmers I, Atkinson P, Fenton M *et al.* (2013) Tackling treatment uncertainties together: the evolution of the James Lind Initiative, 2003–2013. *J Roy Soc Med* **106**, 482–491.
19. Chalmers L (2004) Well informed uncertainties about the effects of treatments - How should clinicians and patients respond? *Brit Med J* **328**, 475–476.
20. Partridge N & Scadding J (2004) The James Lind Alliance: patients and clinicians should jointly identify their priorities for clinical trials. *Lancet* **364**, 1923–1924.
21. JLA (2019) *The James Lind Alliance*. Available at: <http://www.jla.nihr.ac.uk/about-the-james-lind-alliance/> (accessed 19 June 2019).
22. JLA (2019) *How does a JLA PSP work?* Available at: <http://www.jla.nihr.ac.uk/about-the-james-lind-alliance/downloads/JLA-PSP-process-final.pdf> (Accessed 24 June 2019).
23. JLA (2019) *Data Processing and verifying uncertainties*. Available at: <http://www.jla.nihr.ac.uk/jla-guidebook/chapter-6/data-processing-and-verifying-uncertainties.htm> (accessed 24 June 2019).
24. Gallagher M, Hares T, Spencer J *et al.* (1993) The nominal group technique - a research tool for general-practice. *Fam Pract* **10**, 76–81.
25. Jones DJ, Lal S, Gittins M *et al.* (2019) Practical measurement of body composition using bioelectrical impedance, air displacement plethysmography and ultrasound in stable outpatients with short bowel syndrome receiving home parenteral nutrition: comparison of agreement between the methods. *J Hum Nutr Diet* **32**, 288–294.
26. Brooks M, Vest MT, Shapero M *et al.* (2019) Malnourished adults' receipt of hospital discharge nutrition care instructions: a pilot study. *J Hum Nutr Diet* **32**, 659–666.
27. Gottchall C, Tarnowski M, Machado P *et al.* (2019) Predictive and concurrent validity of the Malnutrition Universal Screening Tool using mid-upper arm circumference instead of body mass index. *J Hum Nutr Diet* **32**, 775–780.
28. Salford Age UK. *The PaperWeight Armband*. Available at: <http://www.gmhsc.org.uk/invented-in-salford-the-paperweight-armband-set-to-change-thousands-of-lives-in-greater-manchester/> (accessed 7 July 2019).
29. Murphy JL, Aburrow A, Guestini A *et al.* (2019) Concurrent validity of the modified Patients Association Nutrition Checklist (PANC) against the 'Malnutrition Universal Screening Tool' ('MUST') to identify risk of malnutrition in older people living in the community. *Clin Nutr ESPEN* **29**, 248–283.
30. WAHNS (2019) *The Nutrition Wheel*. Wessex Academic Health Science Network. Available at: <https://wessexahsn.org.uk/projects/236/nutrition-wheel> (accessed 22 July 2019).
31. JLA (2019) *Funding research*. Available at: <http://www.jla.nihr.ac.uk/making-a-difference/funded-research.htm> (accessed 15 July 2019).

Supporting information

Additional supporting information may be found online in the Supporting Information section at the end of the article.

Appendix S1. Protocol and scope of the malnutrition priority setting partnership.

Appendix S2. Organisations contacted for identifying and inviting key individuals to become members of the steering group.

Appendix S3. Out of scope uncertainties.