

**Development of a model for self-care support of diet and the
gut in the routine care of children with cystic fibrosis**

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Submitted in accordance with the requirements for the degree of
Doctor of Philosophy

The University of Leeds
Faculty of Medicine and Health
School of Healthcare

June 2022

The candidate confirms that the work submitted is her own, except where work which has formed part of jointly-authored publications has been included. The contribution of the candidate and the other authors to this work has been explicitly indicated below. The candidate confirms that appropriate credit has been given within the thesis where reference has been made to the work of others.

The work in Chapter 3 of the thesis has appeared in publication as follows:

Cave, L., Milnes, L.J. and McHugh, G.A. 2019. Self-care support of diet and the gut in the routine care of school-age children with long-term conditions: a systematic integrative review [CRD42019144941]. *PROSPERO: international prospective register of systematic reviews* [online].

Cave, L., Milnes, L.J. and McHugh, G.A. 2021. [Post-print]. Self-care support of diet and the gut in the routine care of school-age children with long-term conditions: An integrative review. *Journal of Child Health Care*.

The candidate was responsible for development and writing of the protocol, development of the search strategy, completion of study selection, data extraction, quality appraisal, data analysis and synthesis, and drafting of the paper.

The contribution of the other authors was to review the work at every stage and specifically, they independently checked study selection, data extraction and quality appraisal as part of a systematic and rigorous approach to conducting the review. They also reviewed the manuscript and approved the final version of the paper submitted for publication.

The work in Chapters 5 and 6 of the thesis was presented by the candidate at the European Cystic Fibrosis Conference 2021 and North American Cystic Fibrosis Conference 2021 respectively:

Cave, L., Milnes, L.J. and McHugh, G.A. 2021. Developing a model for self-care support of diet and the gut in the routine care of children with cystic fibrosis: a qualitative study. *Journal of Cystic Fibrosis*. **20**(S1), p.S94.

Cave, L., Milnes, L.J. and McHugh, G.A. 2021. Children's perspectives on learning to look after their diet and gut: a multicentre qualitative study. *Journal of Cystic Fibrosis*. **20**(S2), p.S152.

The candidate drafted the abstracts. The other authors reviewed them and approved the final versions.

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Acknowledgements

I would like to start with a huge thank you to everyone who has contributed to this research, undertaken as part of a Clinical Doctoral Research Fellowship funded by Health Education England/ National Institute for Health and Care Research (ICA-CDRF-2018-04-ST2-017).

A special heartfelt thanks goes to all the study participants who generously gave their time and shared their insights with such enthusiasm, and to the recruiting dietitians for their commitment and willingness to ensure the study progressed during the pandemic.

Huge thanks to my academic supervisors, Dr Linda Milnes and Professor Gretl McHugh for their amazing support and constant encouragement, to my clinical supervisors Dr Alistair Duff and Mrs Carolyn Patchell for helping me find a balance clinically, to the members of the project advisory group for their ongoing support and wisdom and to the members of Leeds Young Research Owls for their time and expertise.

None of this would have been possible without the continuous steadfast support of my husband Ben and our three beautiful children – thank you all so much.

And finally, I would like to dedicate this work to my wonderful patients and their families, who are my inspiration.

Abstract

Background: Key UK health policy advocates provision of self-care support (SCS) for children with long-term conditions such as cystic fibrosis (CF).

However, there is insufficient evidence regarding what SCS should consist of and how it can be delivered as part of routine care. This gap between policy and practice has resulted in a lack of support for children with CF, in gradually learning how to look after their diet and gut, both key components of their CF care.

Aim: To develop a model (conceptual framework) that encapsulates how SCS of diet and the gut as a complex intervention, could theoretically work in the routine care of pre-adolescent children with CF.

Methods: A three-phase multi-method pragmatic study was conducted, guided by the development phase of the Medical Research Council framework and complemented by the Behaviour Change Wheel. Phase one was an evidence synthesis of 27 studies using an integrative review methodology. In phase two, a qualitative descriptive study, the perspectives of 58 key stakeholders (preadolescent children with CF, parents and children's CF dietitians) were explored in semi-structured interviews, and analysed using Framework. In phase three, findings from phases one and two were integrated and further developed through modelling, and a draft model evaluated.

Findings: A model for a theory- and evidence-based digital behavioural intervention is presented, that accounts for implementation of the intervention in the context of routine care and is based on achieving stakeholder-valued outcomes. The intervention would aim to gradually build children's knowledge, understanding, skills and confidence, maintain their motivation, and provide access to peer support and peer-to-peer learning.

Conclusions: The findings inform recommendations for practice and the model informs future co-production and testing of a prototype of the SCS intervention.

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Abbreviations

BCTs	Behaviour Change Techniques
CF	Cystic Fibrosis
COM-B	Capability Opportunity Motivation – Behaviour
GI	Gastro-intestinal
HCP(s)	Health Care Professional(s)
HEMT	Highly Effective Modulator Therapies
IR	Integrative Review
LTC	Long Term Condition
MDT	Multidisciplinary Team
MMAT	Mixed Methods Appraisal Tool
PAG	Project Advisory Group
PERT	Pancreatic Enzyme Replacement Therapy
PPI	Patient and Public Involvement
SCS	Self-Care Support
TDF	Theoretical Domains Framework
T1DM	Type 1 Diabetes

Chapter 1

Background

1.1 Introduction

This chapter provides an overview of the background to the study. It begins by introducing cystic fibrosis (CF) as the condition under study, and the importance of nutrition in CF. It then outlines the motivation for doing the research and why the topic of self-care support in preadolescence became the focus. The chapter then outlines how preliminary work undertaken by the researcher, in addition to an overview of existing evidence, informed the study. It highlights the unique context in which the PhD study was conducted and concludes with the overall study aim and an outline of the chapters that form this thesis.

1.2 Cystic Fibrosis

Cystic Fibrosis (CF) is the most common genetic condition in the UK. It affects more than 10,800 people (of whom over 4,200 are aged under 16 years) (CF Trust, 2021). It is a long-term, progressive condition, for which there is currently no cure. With early diagnosis through national newborn screening, and advances in care and daily treatments, life expectancy is increasing. The median age of survival in 2020 was 50.6 years, compared to 35.2 years in 2007 (CF Trust, 2009; 2021). Whilst this is to be celebrated, the median age at death in 2020 was just 36 years (CF Trust, 2021).

People with CF of all ages perform complex and time-consuming daily treatments, mainly to stay well and minimise progression of CF, but also to treat symptoms. Burden of treatment, together with limitations to social and physical activities and school or work life, are reported by children and adults with CF to have a substantial impact on their daily lives (McCarrier et al., 2020).

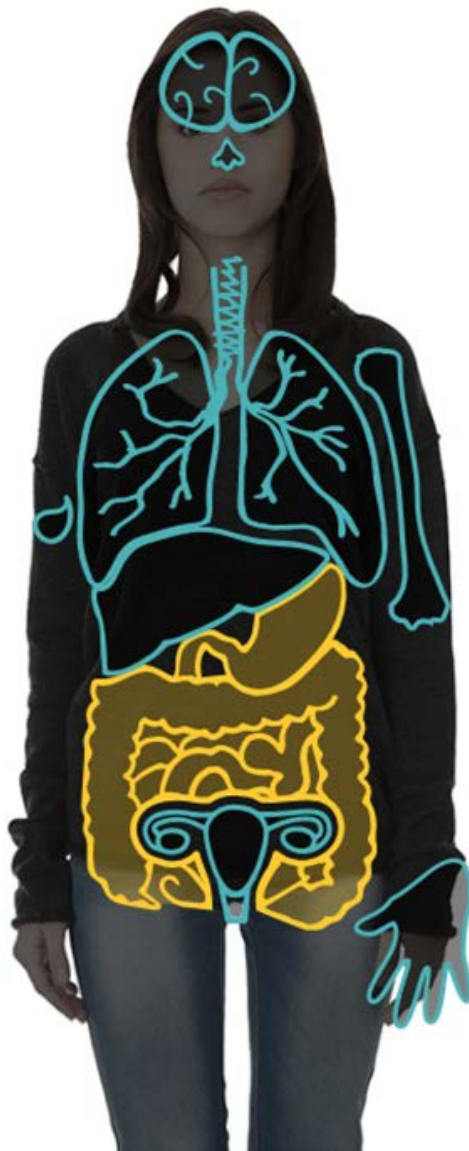
CF is caused by inheriting two mutations of the CF Transmembrane Regulator (CFTR) gene, that encodes the CFTR protein. The consequence is a deficiency or absence of functional CFTR protein which disables the movement of ions across epithelial cells, causing abnormal fluid transport (Li and Somerset, 2014). This results in a build-up of thick and dehydrated secretions (e.g., mucous) which cause inflammation and obstruction in multiple organs (Patchell

and Stead, 2020). There are over 2,000 known CF-causing mutations, which vary in how they affect the CFTR protein (Elborn, 2016). Individuals with CF therefore differ in how CF affects their body and the variety and severity of symptoms they experience (CF Trust, 2022).

1.3 Importance of nutrition and effectively managing the gut

As shown in Figure 1-1, CF affects multiple parts of the body but particularly the lungs, gastrointestinal (GI) tract (or 'gut') and pancreas (Bolia et al., 2018).

Figure 1-1: How CF affects the body



(Reproduced with permission of the CF Trust)

In recent decades, the focus of clinical management of CF has been the lungs (Freedman and Schwarzenberg, 2016), as progressive lung disease is a major cause of morbidity and mortality (Elborn, 2016). Together with lung disease, malnutrition is a frequent feature and comorbidity of CF (Turck et al., 2016). The causes of malnutrition in CF are multifactorial, but may be summarised as:

- Inadequate nutritional intake (energy, macronutrients and micronutrients) due to poor appetite, feeding behaviour problems
- Increased energy losses and nutrient deficits due to malabsorption, losses in the stools and sputum, vomiting following coughing or reflux, complications of CF such as CF-Liver Disease and CF-Diabetes
- Increased energy needs due to lung infections, inflammation, increased work of breathing
(Culhane et al., 2013).

Nutritional management is therefore an essential component of multidisciplinary CF care (Collins, 2018), with optimal nutritional status in childhood associated with improved quality of life (Shoff et al., 2013), improved lung function, growth (height as an adult) and survival (Steinkamp and Wiedemann, 2002; Peterson et al., 2003; Yen et al., 2013).

Approximately 85-90% of people with CF are classed as pancreatic insufficient (PI) (Munck, 2010), where their pancreas is unable to release enzymes critical to the digestion and absorption of fat, fat-soluble vitamins and to a lesser extent protein (Culhane et al., 2013). People of all ages with CF who are pancreatic insufficient, need to take pancreatic enzyme replacement therapy (PERT) with all fat-containing foods and fluids, with doses varying according to the fat content (Bolia et al., 2018). In addition, PERT requirements, both doses and timing, are individual. Optimal PERT, alongside support to eat a normal to high energy (high fat) diet and fat-soluble vitamin supplementation, to meet individual needs, is facilitated at regular reviews by an experienced CF dietitian. The aim of reviews are to optimise nutritional status, achieve control of malabsorption and maintain normal fat-soluble vitamin levels (Patchell and Stead, 2020).

In addition to the pancreas, there are other factors in the gut that contribute to maldigestion and malabsorption in CF, including thick viscous mucous forming a physical barrier to absorption, disturbances in bicarbonate, gastric acid and

bile acid secretion and abnormal gastric and intestinal motility (UK CF Trust Nutrition Working Group, 2016).

In recent years, there has been increased recognition of the need to address the effects of CF on the gut. For example, in 2014 the US CF Foundation launched a training programme for gastroenterologists to develop expertise in CF (Freedman and Wilschanski, 2017). With improved life expectancy, attention is turning to the burden of GI symptoms such as wind, stomach cramps/pain, nausea and bloating, which are highly prevalent in CF and impact daily life (Smith et al., 2020). Relief of GI symptoms was identified as a top 10 research priority for CF in a James Lind Alliance Priority Setting Partnership (JLA PSP) (Rowbotham et al 2018). In a follow-up study, adults and children with CF reported modification of diet and PERT were key in relieving symptoms (Smith et al., 2020). Furthermore, diet and PERT are viewed as a point of control by people with CF (Cave and Milnes, 2020), in what is otherwise an intensely monitored and treated condition.

1.4 Motivation for doing the research

(Written with the researcher in the first person).

I am a specialist children's dietitian. I have worked for over ten years in the care of children with CF and their families at a specialist regional CF centre. I have repeatedly observed a gap in clinical practice and the impact this has on children and young people. There appears to be little support for children to begin to understand what is happening with regards to their diet and gut and how they can be involved in looking after this aspect of their CF.

The gap was most readily observed with children in early adolescence when discussing 'moving towards independence' questionnaires (Leeds Teaching Hospitals NHS Trust, 2008). The questionnaires were a means of assessing children's readiness to be more independent as they transitioned from primary to secondary school. Questions assessed understanding and knowledge of various aspects of CF, including diet and PERT, and how they felt CF affected their life. When discussing the answers children provided, there were frequent gaps, errors and misconceptions, with children often having little or no involvement in their care (i.e., everything was being done to/ for them), and experiencing difficulties e.g., with taking PERT at school. It was clear that more

support was needed in addition to this one-off combined assessment-education session, particularly when I further observed the impact this had. As children moved through adolescence, they were often insufficiently prepared to take over the responsibility in looking after their diet and gut, from their parents/carers. As a consequence, optimal nutrition and PERT adherence during adolescence were often compromised. This negatively impacted nutritional status, and when it persisted, resulted in deterioration in lung function and stunted growth. Such observations are common (Peterson et al., 2003; Eakin et al., 2011; Connett, 2016), with adolescence viewed by healthcare professionals (HCPs) as a period of 'damage limitation'. However, for some, the damage sustained during the adolescent years is great and contributes to a reduced quality of life and premature death in adulthood (Schiff et al., 2021).

My motivation for doing the research was therefore to explore how the observed gap in support could be addressed. Consideration of the type of support to focus on, self-care or self-management, now follows.

1.5 Self-care and self-care support

The terms 'self-care' and 'self-management' are often used interchangeably in the healthcare literature, though the following definitions illustrate how they differ:

- Self-care: the broad range of activities carried out to *live well with a long-term condition* (LTC) (Kirk et al., 2010)
- Self-management: activities related to managing *the LTC well* (Morgan et al., 2017) such as monitoring the condition, managing symptoms and taking medications (Kirk et al., 2010).

Self-care is more comprehensive, and encompasses self-management, health promotion (Bee et al., 2018), and general wellbeing, rather than being medically oriented/ focused on the physical dimension of health (Pelicanand et al., 2013).

Support for self-care (or self-care support (SCS)) refers to enabling individuals to perform self-care and may be provided by family, HCPs (Matarese et al., 2018) or peers, in a variety of ways including information provision, skills training, support networks (Kirk and Prymachuk, 2016).

SCS rather than self-management support is more appropriate for children living with LTCs as it attends to the combination of their developmental, psychosocial and healthcare needs whilst growing (Pelicand et al., 2013). The proactive health promotion element of SCS may also minimise occurrence of symptoms (Chiron et al., 2016), therefore negating their treatment and reducing overall treatment burden.

Having decided that SCS should be the focus for exploration, the next consideration was whether SCS should be targeted prior to adolescence in CF.

1.6 Targeting SCS prior to adolescence

Adolescence is a period of transition from childhood to adulthood, beginning at 10 years of age (WHO, 2022), in which there is rapid physical, psychological and social developmental change (Segal, 2008). The challenge of navigating these multiple changes alongside a considerable treatment burden, often results in suboptimal self-care/treatment adherence during adolescence in CF (Modi et al., 2008; Bucks et al., 2009; Faint et al., 2017) and other childhood-onset LTCs (White et al., 2016; Cameron et al., 2018).

Sawicki et al. (2015) identified early initiation and repeated practice of self-care skills as a facilitator of adherence in adolescents with CF. Other authors concur that interventions seeking to gradually build knowledge, skills and confidence prior to adolescence, may provide a solid grounding in self-care skills to take forward into adolescence (Christian and D'Auria, 2006; Downs et al., 2006; Streisand and Mednick, 2006). Furthermore, Kelo et al. (2011, p.2097) state the basis for lifelong self-care is created at the school age of 6-11 years, when self-care abilities and habits are established. Despite this, Kelo et al. (2011) highlight the dearth of studies involving 6-11 year olds. Likewise, Sullivan-Bolyai et al. (2016) and Velasco et al. (2020) indicate how there has been little attention on interventions targeting the pre-teen age group and children's direct involvement. Though these authors are referring to type 1 diabetes (T1DM), there is similarly under representation of children with CF in this age group.

In clinical practice, children often start to ask questions about their CF from six years of age and are beginning to make independent food choices. Gall et al. (2006) also report the age of seven years as a developmentally appropriate age for children to begin developing independence. Conducting research to further

explore needs and preferences for SCS, directly with primary-school age children aged 6-11 years, was therefore warranted.

1.7 Preliminary work

To inform the PhD study, preliminary work was undertaken by the researcher, with children with CF and their parents, adults with CF and CF dietitians. It shaped the focus of the study and situated it as part of a programme of work.

1.7.1 Patient and Public Involvement (PPI)

As part of a formal PPI consultation (outlined further in section 2.11) to begin understanding children and parents needs for SCS:

- Children with CF aged 7-11 years reported wanting to be independent with their PERT when they transitioned from primary to secondary school
- Their parents wanted more professional support for their child in preparing for this change in responsibility (Cave et al., 2016).

In a further PPI activity, children aged 8-10 years expressed feelings of frustration that their parents were asked about GI symptoms rather than themselves, when they were the ones experiencing the symptoms. The children wanted opportunities to play a more active role in their care, for the adults to trust their accounts and use language they could understand, as *'all the talk is about our bodies and what we need to do to stay good'* (child with CF, male, aged 9 years).

1.7.2 Study conducted with adults with CF

In an interpretative phenomenological analysis study the researcher conducted, adults with CF (n=10) reported having unmet information needs as they were growing up (Cave and Milnes, 2020). Most felt that knowing things earlier in childhood, to begin making sense of how eating and taking enzymes was connected with them gaining weight, growing and having better lung health, did or would have motivated them to keep taking their enzymes in adolescence. In the same study, adults urged CF teams, and particularly dietitians, to do more to directly involve children in their care and encouraged children with CF to not only be involved in looking after their diet and gut, but begin to take control of this aspect of their CF.

1.7.3 Informal discussions about provision of SCS by children's CF dietitians across the UK

The researcher enquired about the provision of SCS (as defined above) by children's CF dietitians. Provision of SCS was found to vary widely across CF centres; it was not structured and was often only delivered in response to the presentation of a problem in busy clinics or during hospital admissions.

As the majority of children are diagnosed with CF through newborn screening, there is an opportunity for the findings of this research to be applied in practice, with dietitians delivering individualised SCS as part of routine care throughout childhood.

1.8 Brief overview of existing evidence to inform the study

A brief review of current guidelines, policy, theory and research evidence on SCS was conducted prior to commencing the study, in addition to a comprehensive integrative review conducted as phase one of the study (Chapter three).

Clinical practice guidelines did not specifically mention SCS. The UK best practice guidelines on nutritional care in CF (UK CF Trust Nutrition Working Group, 2016) highlighted the importance of nutritional knowledge in children with CF and their families and that school-aged children should be encouraged to learn about their PERT, but no further details were provided on either point. In the European and Australian-New Zealand evidence-based guidelines on nutritional care in CF (Turck et al., 2016 and Saxby et al., 2017), regular patient and family education was advocated but only in terms of promoting weight gain and growth.

Key UK health policy stated the importance of SCS for children with LTCs such as CF (Department of Health and Department for Education and Skills, 2004), committed to investment in evidence-based approaches that build people's confidence and skills to self-care/self-manage (NHS England, 2014), and positioned supported self-management (defined in the same way as SCS in section 1.5) as a key component of personalised care (NHS England, 2019). However, these policies stopped short of detailing what SCS should consist of and how it can be delivered as part of routine care.

Three relevant systematic reviews, the first, by Kirk et al. (2013), examined the extent to which SCS interventions (and their components) had a beneficial effect on health-related outcomes in children aged 0-16 years with the LTCs asthma (n=10 studies), CF (n=2) and diabetes (n=1). SCS interventions that targeted children, used e-health or group-based methods and were delivered at home or in community settings were particularly effective (Kirk et al., 2013). Furthermore, there was no evidence that interventions focussed on parents alone or delivered only in hospital settings were effective. Kirk et al. (2013) highlighted the need for:

- Well-designed trials that test interventions with an underlying theoretical basis (i.e., that suggests the nature and content of the intervention and the appropriate outcomes to measure)
- Interventions to be developed with children and parents (including choosing outcome measures that are relevant to them)
- Assessment of implementation issues to inform transfer of interventions into clinical practice.

In the second systematic review on SCS for children with LTCs, Bee et al. (2018) examined SCS interventions that could reduce health service utilisation and costs without compromising outcomes for children aged 0-18 years with a long-term physical or mental health condition. They found that SCS interventions that included the child, and delivered some content to an individual or individual family had a small but positive effect on quality of life (Bee et al., 2018). Of the 97 studies, the majority included children with asthma (n=66), with diabetes (n=6) and CF (n=0). Bee et al. (2018) concluded:

- Identification of optimal models of SCS was challenged by the limited size and scope of the evidence base
- Further research is needed to confirm which intervention characteristics optimise patient- and service-level effects and to determine potential differences in the shorter- and longer-term effects of SCS across a wider range of LTCs
- New evidence-based models of SCS need to be co-developed with patients and their families.

The third review, was a Cochrane systematic review of the effects of self-management education interventions for CF (n=4) on a range of health outcomes in individuals of all ages (Savage et al., 2017). The review found that due to the limited quantity and quality of interventions included, there was insufficient evidence on the use of self-management education for CF in routine clinical practice. Savage et al. (2017) concluded:

- Further well-designed trials of interventions with a strong behavioural component are needed to evaluate intervention types, content, format, duration and delivery (settings, mode and personnel), with consistency in the outcomes measured
- Investigation of the long-term effects on lung function and nutritional growth are needed.

Whilst these three reviews provided some useful evidence to build on, this was limited with respect to consideration of SCS of diet and the gut in children as the studies included in the first two systematic reviews predominantly focused on children with asthma (and therefore did not consider diet and the gut) and of the four studies included in the Cochrane review, one focused on airway clearance and another on adults.

Other research evidence has focused on supporting self-management in children with LTCs. As self-care incorporates self-management, this evidence contributed in part to the evidence base informing the current study. In Modi et al.'s (2012) conceptual framework, self-management behaviours were central, and were impacted by the context of care, considered on an individual, family, community and healthcare system basis. Although the framework focused on self-management to support treatment adherence, it drew attention to the multi-level influences and to modifiable factors that influence behaviours and could potentially be targeted in an intervention.

Saxby et al. (2019), in a systematic review, identified key components of educational interventions to help children learn about managing their LTC. These included a structured and sequenced curriculum, reinforcement of learning, active participation of the child, collaborative learning through group activities and multiple opportunities to learn and practice over time. Though these components were identified chiefly from education interventions in

asthma, they provided insight into potential intervention characteristics. In another review of self-management interventions (n=78), Sattoe et al. (2015) provided a useful outline of intervention content and formats, together with a framework for the selection of appropriate (content-based) outcomes. This review had no restriction on included study designs and therefore dealt comprehensively with what constitutes self-management support across a broad range of LTCs in people aged 7-25 years. The included studies were mostly aimed at the medical management of asthma and diabetes, though intervention content in over half of the studies extended to SCS. The authors similarly highlighted the need for more interventions with an underlying theoretical basis, the adoption of appropriate outcome measures and recommended qualitative research to gain more insight of the context and working mechanisms of interventions.

In summary, the existing evidence base to inform the current study was limited, particularly regarding SCS of diet and the gut. However, there were several elements to build on, to begin filling the gap between policy advocating SCS and the what and how of achieving this in clinical practice. Notably the need to co-develop a theoretically informed and evidence-based SCS intervention with children and parents, paying close attention to the context in which the intervention would be delivered/ implemented in practice and the choice of relevant outcomes.

1.9 Context of the study

Phases two and three of the PhD study were conducted at a time of unique circumstances. First, there was a coronavirus (Covid-19) pandemic. People with CF were defined as clinically extremely vulnerable, with their daily lives significantly impacted by shielding programmes in addition to regional and national lockdowns. Second, from August 2020, NHS England began the roll-out of the next generation of potentially life-changing drugs, highly effective modulator therapies (HEMT), targeting the most common CF mutations and signalling a new era for CF care (Dave et al., 2021). The full impact of HEMT is as yet unknown (Sergeev et al., 2020), but studies on an early HEMT have suggested that increased fat absorption and decreased gut inflammation may contribute to weight gain (Stallings et al., 2018), with increases in the prevalence of overweight and obesity posing a new challenge for the CF

population (Gabel et al., 2021). This is prompting a move away from a reliance on energy-dense, nutrient-poor foods and a greater emphasis on diet quality (Sutherland et al., 2018), with dietitians recommended to adopt a more holistic, individualised approach to diet and PERT (McDonald et al., 2020).

These factors effected the study in numerous ways and are discussed as appropriate throughout the thesis.

1.10 Overarching study aim and objectives

The overarching study aim was to develop a theory and evidence-based model (conceptual framework) for SCS of diet and the gut for pre-adolescent children with CF, in collaboration with key stakeholders: children and parents/ carers who would be receiving SCS and dietitians who would be delivering SCS in routine care. Developing the model in this way may capture what is most relevant to accommodating changing needs of individual children with CF as they grow towards independence, in the context of routine dietetic care.

To address this aim, the objectives of the research were:

Objective 1: To develop a theoretical framework* for SCS in the dietary care of children with CF by identifying, describing and critically evaluating existing theory and evidence for SCS of diet and the gut in children with LTCs

Objective 2: To explore the perspectives of key stakeholders in the routine dietetic care of children with CF to further develop the theoretical framework* for SCS

Objective 3: To develop a model (conceptual framework*) of SCS in the dietetic care of children with CF, including the relationship to outcomes identified as most important by key stakeholders.

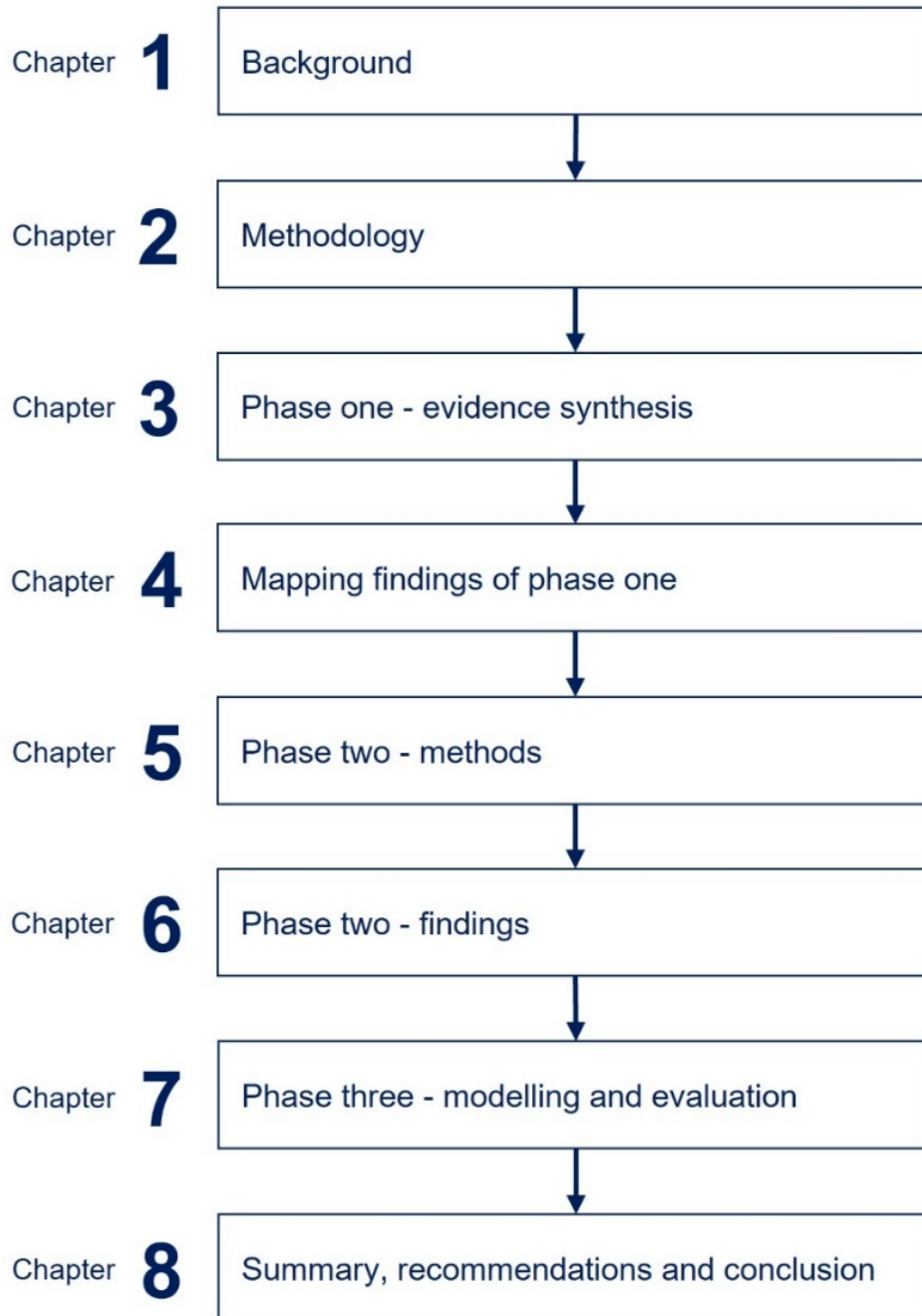
The output of the study - a model (conceptual framework) – will encapsulate how SCS of diet and the gut as a complex intervention, could theoretically work, by putting together all ‘active components’ (Sermeus, 2015). This will inform recommendations for practice and future testing of a prototype of the SCS intervention in the context of routine care.

*The terms ‘theoretical framework’ and ‘conceptual framework’ are often used interchangeably in the research literature, but they differ. A theoretical framework describes the *what* of the study, the broad relationship between

components taken from the reviewed literature and/or data collected; a conceptual framework is founded on the theoretical framework but is more specific – it is an integration of findings by the researcher that specifies the components to focus on for practice, the *why* and the *how*, in consideration of context (Ravitch and Riggan 2017).

1.11 Structure of the thesis

The thesis is organised into eight chapters (Figure 1-2). Chapter 2 sets out the methodological decisions which informed the choice of design and methods for each of the three study phases. Chapter 3 describes phase one, an integrative review, the findings of which are mapped to a behaviour change framework in chapter 4 and inform chapter 5. Chapter 5 details the working methods of phase two, an in-depth interview study, the findings of which are presented and discussed in chapter 6. Chapter 7 describes how phase two findings are mapped to a behaviour change framework and integrated with chapter 4 findings, to develop phase three's model. Finally, chapter 8 provides an overview of the thesis, summarising implications for clinical practice and recommendations for future research.

Figure 1-2: Order of chapters

Chapter 2

Methodology

2.1 Introduction

This chapter provides an overview of the research approach of the study, together with the design and working methods for each of the three study phases, with rationale for the choices made. It begins with outlining the philosophical assumptions that underpin the research. Next, the decisions taken in the choice of a methodological framework, and design and methods for the study phases, are considered in turn, with strengths and limitations discussed. The chapter then outlines the ethical and quality considerations for the study and concludes with study management and patient and public involvement.

2.2 Underpinning philosophy

Critical reflection by the researcher on the perspectives and experiences brought to the research and philosophical standpoints (ontological and epistemological), led to consideration of critical realist, interpretivist and pragmatic approaches. With a critical realist approach, reality is only accessible through individual participants' own perceptions and interpretations; whilst an interpretivist stance explores individual participants' interpretation of their experience, or reality, in context, more so than the reality itself (Ormston et al., 2014). Both of these approaches resonated with the researcher, however, as the PhD study aimed to advance understanding of experience, or reality, of what is useful in a given situation/ context at a given time (Duram, 2010), a predominantly pragmatic stance was adopted. Pragmatism is not committed to any one system of philosophy and reality (Creswell and Poth, 2018). It focuses attention on application - what works to find practical solutions to problems, with flexibility to choose the most appropriate approach and methods to address the research questions (Ormston et al., 2014). A pragmatic stance aligns with the study being applied research, in that it sought practical solutions to a recognised problem/gap in clinical practice and the findings will be applied back to practice, with the practical solutions relevant to stakeholders and the context of real-world practice (Glasgow, 2013).

2.3 Methodological framework

To meet the study aim (section 1.10), the recommendations of several studies outlined in section 1.8 were drawn on. First, Kirk et al. (2013) and Sattoe et al. (2015) highlighted the need for interventions to have an underlying theoretical basis. Second, Bee et al. (2018) recommended that new evidence-based models of SCS follow standard frameworks for the development of complex¹ interventions and draw on relevant behavioural models. Two theory and evidence-based intervention development frameworks were therefore considered for the current study: intervention mapping (IM) (Bartholomew-Eldridge et al., 2016; Fernandez et al., 2019) and the Medical Research Council (MRC) framework (Craig et al., 2006).

Both frameworks set out a systematic approach for phased development of a complex intervention and consideration of implementation from the start. The IM framework appears more comprehensive, technical and prescriptive, and is used more for health promotion/ public health and community-based interventions; the MRC framework has little detail in the development phase, but is not prescriptive, and is used more in health care (O’Cathain et al., 2019a, 2019b; Brewster et al., 2021). The MRC framework was chosen over the IM framework, due to its more frequent use in clinical settings and because it encouraged a pragmatic approach to intervention development (Craig and Petticrew, 2013).

The key activities outlined in the MRC development phase (Craig et al., 2006) were:

- Identifying the evidence base
- Identifying/ developing theory
- Modelling process and outcomes.

¹ An intervention might be considered complex because of properties of the intervention itself, such as the number of components involved, the range of behaviours targeted, expertise and skills required by those delivering and receiving the intervention, the number of groups, settings, or levels targeted, or the permitted level of flexibility of the intervention or its components (Skivington et al., 2021, p2)

The limitation of little detail in the development phase was addressed by adding elements proposed by Bleijenberg et al. (2018). Specifically, as part of the activity 'identifying/ developing theory', the needs, preferences and capacities for SCS of both recipients and providers were explored, along with current practice and context. An existing framework of theories on behaviour change was also identified to inform the intervention (O'Cathain et al., 2019a). These additions ensured this early development phase was more comprehensive, which was important in determining how the intervention could theoretically work and may improve the likelihood that a later developed prototype of the intervention, when tested, would be well-adopted, effective and fitted to the delivery context, therefore minimising research waste (Hoddinott, 2015; Bleijenberg et al., 2018).

This comprehensive approach was reflected in an update of the MRC framework – commissioned jointly by the MRC and the NIHR, expected in 2019 but published in September 2021 (Skivington et al.), with more emphasis on engagement with stakeholders, consideration of context and articulation of the underlying theory, including the interventions key components and mechanisms of action.

2.4 Overview of the study phases

The development phase of the MRC framework (Craig et al., 2006), provided an overarching flexible structure to the study and guided the choice of design and methods for the study phases.

The three key activities outlined in the development phase (listed above) informed the three research objectives (section 1.10), and these in turn, aligned with three study phases:

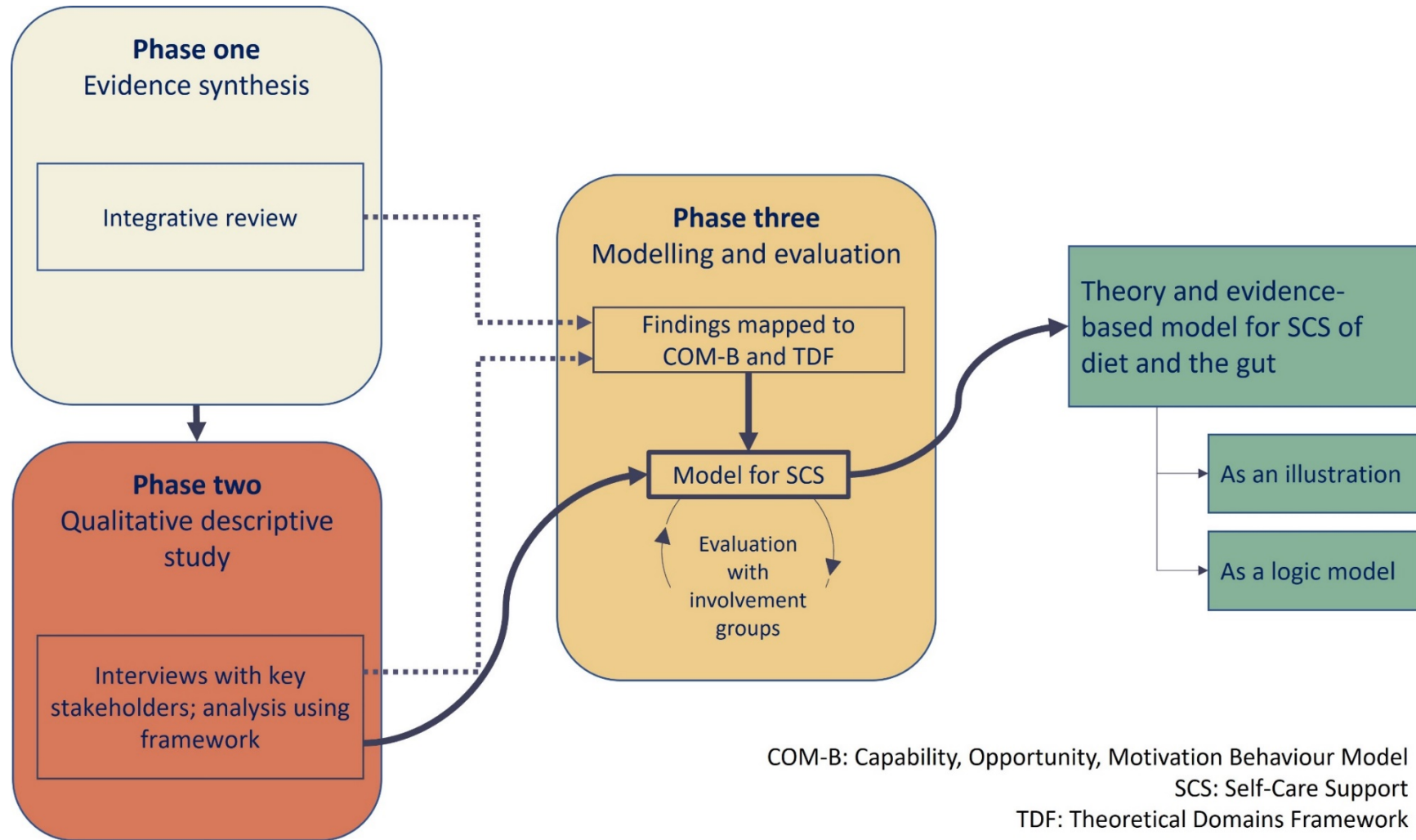
Phase one: Evidence synthesis

Phase two: Qualitative descriptive study – an in-depth exploration of key stakeholders' perspectives

Phase three: Modelling and evaluation.

Figure 2-1 illustrates the three study phases and how they relate to one another.

Figure 2-1: Outline of the study



A detailed overview of the research design and working methods chosen for each of the three study phases now follows.

2.5 Phase one: Evidence synthesis

To meet the objective for phase one, a means of identifying, describing and critically evaluating existing theory and evidence was needed. Moreover, synthesis was essential to integrate the findings into a theoretical framework for SCS of diet and the gut and identify knowledge gaps for practice and future research (Knafl and Whitemore, 2017), and to inform the primary research in phase two.

In considering SCS as a complex intervention for future implementation in routine care, the synthesis questions needed to address not only what works (intervention effectiveness), but what works for whom in what circumstances (intervention effectiveness and contextual moderators) (Pawson et al., 2005). From a preliminary search of the literature, combining these several questions, potentially relevant studies adopted a diverse range of qualitative, quantitative and mixed methods research designs. Synthesis methods that could accommodate this diversity in a single study were therefore considered (Table 2.1). An integrative review (IR) was chosen to be able to understand and define what SCS of diet and the gut consists of and what is already known (and not) about SCS as an intervention.

Table 2.1: Types of syntheses considered for phase one

Type of synthesis	Aim	Sampling frame	Features
Scoping review	Identify knowledge gaps, map/clarify key concepts (Munn et al., 2018)	Can include primary research with diverse designs and theoretical literature; unit of analysis depends on the focus but could be a concept (Schick-Makaroff et al., 2016)	Summarises range of evidence available; no synthesis undertaken or assessment of methodological quality (Peters et al., 2015)
Mixed studies / mixed methods systematic review	Effectiveness and experience of Ix's - whether interventions work and how (Stern et al., 2020)	Can include primary research with diverse designs	Synthesis can be quantitative and/or qualitative, and simultaneous or consecutive, depending on the

Type of synthesis	Aim	Sampling frame	Features
			review questions (Stern et al., 2020); Can contribute to theory development and refinement and understanding of how Ix's interact with contexts to affect outcomes (Leeman et al., 2015)
Realist review	Investigates what works, for whom, under what circumstances, why and how (explains the mechanisms by which an Ix works (or not)) (Pawson et al., 2005)	Can include primary research with diverse designs; unit of analysis is the programme theory (ideas and assumptions underlying what the Ix is trying to achieve and how) (not the Ix itself) (Schick-Makaroff et al., 2016)	All study types in a single synthesis; focuses on theory development and refinement; explores how different contexts might result in different mechanisms and outcomes; involves high engagement with stakeholders (Rycroft-Malone et al., 2012)
Integrative review	Provides a comprehensive understanding of a topic (Oermann and Knafel, 2021); identify knowledge gaps, conceptualisation of a topic/ identify key constructs (Torraco, 2016)	Can include primary research with diverse designs and theoretical literature (Whittemore et al., 2014)	All study types in a single synthesis; Can contribute to theory development/ produce theoretical frameworks (Elsbach and van Knippenberg, 2020)

Ix: intervention

The diverse sampling frame of an IR (both primary research evidence and/ or theory) provides a challenge for analysis and synthesis, yet has the potential to comprehensively capture the breadth and depth of the topic (Whittemore, 2005). IRs also have the advantage of accommodating diverse data sources, for example, bibliographic databases and grey literature. For the current study, it was likely that some relevant literature would not be published in peer-reviewed journals, for example, SCS initiatives tried in clinical practice but only reported as conference proceedings. Inclusion of grey literature was therefore appropriate and sought to minimise publication bias (Olsen, 2013).

To ensure the IR was structured (Dhollande et al., 2021) and conducted systematically, the methodological guideline described by Whitemore and Knafli (2005) was followed. Strategies adopted to ensure rigour included:

- Development of a comprehensive search strategy
- Independent review of the selection process, data extraction, quality appraisal, data analysis and synthesis
- Clear detailed reporting of the whole IR process (Lubbe et al., 2020).

IRs are reported to lack transparency (Snyder et al., 2019). To address this, a protocol was developed and registered with PROSPERO (CRD42019144941) (Cave et al., 2019) and reporting followed PRISMA guidance (Page et al., 2021). A further limitation of IRs is that they can lack synthesis and present findings as descriptive summaries (Snyder et al., 2019). To overcome this, true integration of the findings was sought and is described in section 3.2.4.

Appraising the quality of diverse study designs included in the IR presents a major challenge (Whitemore et al., 2014). Two critical appraisal tools were considered. First, the Critical Appraisal Skills Programme (CASP), which has separate checklists for each study design, for example, randomised controlled trials (RCTs) and qualitative studies (CASP, 2018), but no checklist for mixed methods studies. Second, the Mixed Methods Appraisal Tool (MMAT), which is a single tool with criteria to apply to common study designs, including mixed methods studies (Hong et al., 2018). The MMAT was chosen over CASP to negate use of different checklists for each of the study designs and because it specifically considered mixed methods studies. Strengths of MMAT included it being an easy to use validated tool with comprehensive guidelines, allowing concomitant appraisal of core methodological criteria for five types of study design, with clear rating criteria for each item (Hong et al., 2019). Its limitations were that it was restricted to five core criteria for each type of study design, so possibly lacked the depth necessary for comprehensive appraisal (Crowe and Sheppard, 2011), and was not useful for theoretical papers because there were no criteria related to philosophical assumptions. However, it suited the overall pragmatic stance of the study in terms of utility and best fit for purpose (Heyvaert et al., 2013).

2.6 Phase two: Qualitative descriptive study

To meet the objective for phase two, a qualitative exploratory design was appropriate as little is known about the topic of SCS of diet and the gut (LoBiondo-Wood et al., 2018). To gain an in-depth understanding of SCS to further develop the theoretical framework for SCS, perspectives were sought of those involved in the routine dietetic care of children with CF: children with CF and parents/ carers who would be receiving SCS and children's CF dietitians who would be delivering SCS.

2.6.1 Approach

Key approaches that were considered for this research and reasons why they were not used are summarised in Table 2.2.

Table 2.2: Qualitative approaches considered for phase two

Qualitative approach	Features	How it could be applied	Why rejected
Phenomenology	Understand a phenomena, how people interpret their lives and make meaning of what they experience (Cohen et al., 2000).	Could explore individual stakeholders lived experience of SCS and what meaning that had for them ('what' they experienced and 'how' they experienced it). This would have provided a description of the common components of SCS as experienced by stakeholders.	Individual stakeholders would all need to have experienced SCS.
Grounded theory	Construction of theory grounded in data collected during the research process. Data are collected and analysed and the concepts derived form the basis of subsequent data collection (Corbin and Strauss, 2015).	It would generate a theory that was grounded in data from stakeholders who had experienced SCS. This would have provided understanding of how stakeholders experienced SCS, what was central to SCS, what influenced it and what the outcomes were. The theory could have been presented as a model.	Individual stakeholders would all need to have experienced SCS. Ideally, the researcher would not have background knowledge to a level that allows tentative conceptual and theoretical links to form (Cutcliffe, 2000), though as an experienced clinician and having completed an IR in

Qualitative approach	Features	How it could be applied	Why rejected
			phase one, this would be difficult. Generating a new theory was not necessary as there were existing theories to draw on.
Case study	Develop an in-depth description and explanation of a single case or multiple cases (Yin, 2018).	Could explore and gain an in-depth understanding of SCS as a case within the context of routine care and how it affects outcomes, at one or multiple sites, using multiple data sources e.g., non-participant observations, interviews, workshops (Yin, 2018). It would increase understanding of when SCS, its implementation and/ or the context may need to be modified to enhance fit and therefore achieve intended outcomes (Glasgow, 2008).	Would require well-defined SCS to be implemented at a site(s) (or could be compared to a site with no SCS to understand needs for SCS and how it could be implemented by busy clinicians in their routine practice) (Keen, 2006). Could be an option when SCS is well-defined and due to be implemented in practice i.e., an option for the future rather than the current study.

A qualitative descriptive approach was chosen as a rich description of SCS was sought: the what, how, where, who and why of SCS, first hand from stakeholders (Neergaard et al., 2009). With this approach, the researcher stays close to the data gathered, throughout analysis and presentation (Neergaard et al., 2009) and seeks to ensure their own interpretations are transparent (Sandelowski, 2000). This was suitable for informing development of a stakeholder-centred SCS intervention and in decreasing the likelihood that competing explanations may be responsible for the relationship between intervention components and outcomes (Sullivan-Bolyai et al., 2005), therefore enhancing the internal validity of the study (Sandelowski, 1996).

A limitation of the qualitative descriptive approach is that it is less well defined in comparison with other qualitative approaches that have a specific theoretical underpinning (Smith and Bekker, 2008), such as those outlined in Table 2.2.

However, choice of this approach was congruent with the overall pragmatic stance of the study (Sandelowski, 2000). It ensured choice of data collection and analysis methods were best suited to answering the study questions (Smith and Bekker, 2008) and the knowledge gained had a practical application (Moisey et al., 2022). Whilst this inherent flexibility and utility may be regarded as strengths of the qualitative descriptive approach, it can lead to a lack of rigour (Doyle et al., 2020). Strategies to address this are outlined in section 2.9.

2.6.2 Methodological considerations of conducting research with children

Before providing an overview of the considerations at each stage of the qualitative descriptive study, key methodological issues in conducting qualitative health research with children are now outlined, with key ethical issues discussed in section 2.8.2.

First, the choice of methods will be shaped by how childhood is conceptualised (Punch, 2002). If childhood and adulthood are considered as different cultures, adult researchers may try to understand the child's perspective and the way they view the world, by managing the culture gap between children and adults (Kirk, 2007). For example, by attending to how they seek to develop rapport (including with adult gatekeepers such as parents), taking care not to impose their own views and interpretations, being conscious of the language used, offering a choice of research setting, reassuring children that there are no right and wrong answers (Punch, 2002), use of reflexivity to increase awareness of assumptions about childhood and how this may influence the research process (Harden et al., 2000), and involvement of children in the design and conduct of the research (Davies et al., 2019).

Second, the choice of methods will be shaped by the heterogeneous nature of childhood and therefore consideration of the individual child (Kirk, 2007). For example, their age/ developmental stage, gender, ethnicity; also children's articulation may vary between research topics (Huang et al., 2016). Use of activities/ tools/ techniques to maximise children's engagement in the research process needs to be critically reflected on, including how they match the study questions and account for differences in participants needs (Fargas-Malet et al., 2010); and involving children in decisions about what to use and how, is recommended (Huang et al., 2016).

Reflection on these issues, together with completion of formal training in conducting research with children, informed the choice of methods and approach taken with children, and thread throughout the following overview of the qualitative descriptive study.

2.6.3 Considerations for sampling and recruitment

Purposive sampling was selected as the most appropriate sampling strategy, where potential participants were deliberately chosen because they were key stakeholders in the routine dietetic care of children with CF, and a detailed exploration of their perspectives was central to meeting the objective for phase two (Ritchie et al., 2014). Choosing members of a sample because they had direct experience of either receiving or providing routine CF dietetic care, including SCS (or not), ensured they were able to share insights of most relevance in understanding what could work well (or less well) regarding SCS of diet and the gut as part of routine care (Bradshaw et al., 2017). Other sampling strategies such as theoretical, where sample selection is decided in stages based on developing an emerging theory, or convenience, where selection is made purely on the basis of who is available, were not appropriate. This is because iteratively generating a new theory was not necessary (as outlined above for a grounded theory approach) and selecting only who was available would limit what insights, and therefore the depth of understanding that could be gained (Ritchie et al., 2014).

The sampling frame for children with CF was considered in terms of:

- Gender and the primary school age range of 6-11 years - in order to explore how the needs and preferences for SCS of boys and girls may change as they grow older.

The lower limit of six years was chosen as in clinical practice, children often start to ask questions about their CF at around six years of age and this age was agreed as appropriate by members of the project advisory group (PAG) (section 2.10). The upper limit of 11 years was chosen to coincide with the age at which children transition from primary to secondary school in the UK.

- Receiving care at multiple specialist CF centres – in order to:

(a): Maximise recruitment success

In a previous interview study with children with CF and their parents, reasons for declining participation included being too busy juggling treatments and daily life, not wanting to expose children to talking about CF and being involved in other CF research (Savage and Callery, 2005). Each of these reasons were relevant at the time of this PhD study, particularly children's involvement in clinical trials of new HEMT.

(b): Understand variation in context across the centres

The SCS intervention will need to work across very busy centres and variation between centres will draw attention to features that may otherwise go unnoticed/ have not been considered if at a single centre, to generate insights into what is (and is not) significant for the intervention to work (Benzer et al., 2013).

Three children's specialist CF centres, that will be referred to as centres A, B and C, were chosen because they are three of the largest CF centres in England, each with a caseload of over 300 children.

(Note: the CF centre at which the researcher is a clinician was not included, due to knowing the patients and continuing to be a member of their direct clinical care team).

The sampling frame for children's CF dietitians was considered in terms of:

- Bringing experience of working with children with CF
- Providing care at multiple specialist CF centres – in order to explore current routine dietetic practice and provision of SCS.

To assess when to stop purposive sampling, data saturation was sought. This was judged to occur when additional data were not generating new information or contributing further insights to answer the research questions (Saunders et al., 2018). There has been much debate in the qualitative literature about the concept of saturation (e.g., Morse, 2015; Guest et al., 2020), particularly concerning what it is and how it is recognised, and the underlying assumptions these decisions entail. In the current study, a broad assessment of data saturation was adopted, as recommended by Vasileiou et al. (2018), in which the sample composition and size were based on an assessment of data

adequacy i.e., when there was an adequate depth and variety of data, including contradictory data, to sufficiently answer the study questions.

2.6.4 Data collection

Data collection methods that were compatible with the qualitative descriptive approach and were considered to meet the objective for phase two, included focus groups and individual interviews (Colarafi and Evans, 2016). Focus groups are moderated discussions to obtain perceptions on a specific topic in a non-threatening environment (Krueger and Casey, 2009). Through collective conversation and interaction of group members, a wider discussion of perspectives may be gathered than in individual interviews, as participants explore and clarify their views, generate their own questions and pursue their own priorities (Kitzinger, 2006). However, focus groups would only have been possible if conducted virtually, as children with CF cannot be brought together in person due to the risk of cross-infection (NICE, 2017). Interviews were chosen over focus groups to allow gathering of in-depth descriptions of individuals' experiences, views, needs and preferences, and exploration of the factors underpinning individuals' accounts (Yeo et al., 2014). Furthermore, individual interviews were the preferred method of data collection reported by children with CF and their parents in a PPI consultation conducted prior to the study (section 2.11).

Of the three types of interviews (structured, semi-structured and unstructured), semi-structured were chosen. This was because they provided a balance between having some structure (in terms of having a pre-determined set of topics to address the study questions, that could be approached in any order as appropriate (section 5.2.3.1)), but also scope to prompt and further probe interviewee's responses (Green and Thorogood, 2018). This flexibility enabled exploration of understandings and issues raised by interviewees (Braun and Clarke, 2013) as well as generation of ideas and potential solutions (Yeo et al., 2014). Interviews provided access to interviewees accounts i.e., what they say rather than what they do (Green and Thorogood, 2018), and the data gathered was a 'snapshot' at one point in time, however these potential shortcomings did not undermine their value in the current study.

Before outlining consideration of the mode of interviews and setting, attention turns to the practicalities of interviewing children.

Practicalities of interviewing young children

Establishing a rapport is critical for interviewing children and is aided by allowing children to discuss what is important to them (Spratling et al., 2012). Starting with close-ended questions on topics they can speak freely about, for example, everyday activities, before moving onto open-ended questions also helps to develop rapport (Irwin and Johnson, 2005). Activities can assist children to describe their experiences or sustain interest (Huang et al., 2016), though different children will prefer different activities (Punch, 2002). Though the child's lead should be followed, providing more structure and direction to the interview may be needed compared to interviewing adults, such as the use of verbal prompts (e.g., 'tell me more about that') or follow-up questions (Gibson, 2012). Adapting to meet children's individual developmental and language needs is key, such as the phrasing and pacing of questions (Rogers et al., 2021). For more abstract questions, using words denoting actions will make it easier for children to answer (Kortesluoma et al., 2003), for example, 'what do you do if someone at school asks you why you take Creon (PERT)?' Face-to-face interviews allow the researcher to observe and respond to non-verbal behaviours of the child, for example, looking away or becoming silent (Rogers et al., 2021). The researcher's use of non-verbal behaviours is also important, for example, maintaining eye contact and head nods, to indicate interest and active listening (Fargus-Malet et al., 2010). Finally, reducing the power imbalance between adult interviewer and child interviewee where possible is key, for example, encouraging the child to ask questions, having some control over the recording, sitting at the same level (Rogers et al., 2021).

Mode of interviews and setting

In a PPI consultation conducted prior to the study (section 2.11), children with CF and their parents had reported their preferred mode and setting for data collection as face-to-face interviews in a private room at their CF centre or in their own home. This would enable the researcher to spend time initially building rapport and have greater scope to actively engage each child in a range of activities. This was particularly pertinent given the abstract nature of

self-care and SCS; having a choice of activities relevant to the topic could help stimulate discussion and make the interviews more interesting and fun (section 2.11). With face-to-face interviews, the interviewer can demonstrate continued interest and attention (Irvine et al., 2013), yet also readily observe for any signs of discomfort, distress or dissent (O'Reilly and Dogra, 2017).

Another consideration was whether to interview children and their parent/ carer individually or together. Interviewing them individually may enable differences in perspectives and priorities to be expressed, as found previously by Savage and Callery (2005). Prior to the study, children with CF and their parents had requested that this choice was offered to families. Also, as children in the target age range were young, they suggested children choosing to be interviewed individually have the option of their parent/ carer remaining present. This may be comforting to some children, but it may also affect the child's ability to speak freely e.g., if the child does not want to reveal information in front of their parent/ carer (Spratling et al., 2012). With the parent/ carer present, they could potentially dominate (so the child's contribution was limited) or lead the child's response (O'Reilly and Dogra, 2017). Conversely, parents could compliment children's contributions positively, for example, through interjecting with cues or non-directive prompts that help their child express their views (Gardner and Randell, 2012), or 'scaffolding' their child's responses by supplementing with explanations or additional context (Irwin and Johnson, 2005). Parents may also evoke shared memories and experiences that may not be accessible if the child was interviewed alone (Gardner and Randell, 2012). Another consideration was the feedback from parent members of the PAG (section 2.10). They felt it would be important for parents/ carers to be able to speak openly and freely, without their child present. Each of these factors were considered and a decision taken to offer all children the choice of being interviewed individually, with or without a parent/ carer present, or jointly with their parent/ carer, and the researcher to reflect on how this affected the quality of interview data gathered.

For dietitians, telephone interviews were the chosen mode. This enabled inclusion of dietitians at CF centres that were geographically scattered across England, for whom interviews could be arranged at times to fit around their busy clinical work (i.e. without impacting on their NHS time). In telephone interviews, the absence of visual cues may impact rapport building (Novick, 2008), though

does not necessarily preclude probing and in-depth discussion (Sturges and Hanrahan, 2004). However, greater care with phrasing, intonation and clarification of questions may be needed for the quality of data collected to be comparable with data from face-to-face interviews (Irvine et al., 2013).

2.6.5 Data analysis

Data analysis methods used with the qualitative descriptive approach typically include thematic analysis (Moisey et al., 2022), whereby patterns (themes) are identified to describe data in rich detail (Braun and Clarke, 2006). From the broad range of methods that utilise thematic analysis, Framework (Ritchie and Spencer, 1994; Spencer et al., 2014) was chosen as the most appropriate. This was because it could be used at first deductively, addressing pre-determined topics in the interview topic guide (outlined in section 5.2.3 and informed by findings from the IR), but also inductively in response to issues raised by interviewees as important (Pope et al., 2000).

Use of Framework was consistent with the overall pragmatic stance of the study as it is a flexible tool not aligned with a particular philosophical viewpoint or theoretical approach (Gale et al., 2013). It provided a structured and rigorous process for analysing a large (fairly homogenous) dataset of semi-structured interview transcripts. Strengths included being:

- Systematic: a series of interconnected stages (described in section 5.2.4) guided the process (Smith and Firth, 2011)
- Comprehensive: allowed full review of all the data gathered (Main, 2019)
- Transparent: there is a clear audit trail of how data was handled and findings derived (this facilitated sharing and checking analysis with supervisors and the PAG to enhance rigour (Ward et al., 2013)).

Crucially, Framework allowed for both theme-based and case-based analysis (Ward et al., 2013) so accounts from children, parents and dietitians could be compared, with both context and content preserved (Main, 2019). The method has been used successfully in a previous study, to compare the views of children, parents and HCPs in the development of a self-management support intervention (Waite-Jones et al., 2018).

Limitations included being labour intensive and time consuming (Gale et al., 2013). There is also a risk of the focus being on the process rather than the

outcome, though this can be addressed through the researcher adopting a reflexive and critical approach (Main, 2019).

Though data collection and analysis have each been outlined in turn, the two proceeding concurrently is valuable in allowing emerging themes to be reflected on with subsequent participants (Kendall et al., 2009) and new data informing the analytic processes (Thorne, 2000).

2.7 Phase three: Modelling and evaluation

To develop a model (conceptual framework) of SCS of diet and the gut (and therefore meet objective three), a process was needed to integrate the theoretical frameworks generated in phases one and two and move towards more specific propositions about how SCS as a complex intervention could work in practice.

As self-care of diet and the gut involves repeated daily behaviours, development of a behavioural-focused intervention was indicated. The first step in the process was therefore further exploration of the findings from phases one and two to determine which theoretical components to target for self-care/ SCS behaviours to occur. To guide this process, several behaviour change theories/ models were considered. These included the theory of planned behaviour (Ajzen, 1991), as this had been used previously to understand fruit and vegetable eating behavior in primary school children (Duncan et al., 2014); and the transtheoretical model of behaviour change (Prochaska and Velicer, 1997), as this has been widely used for health promotion and health education interventions targeting dietary behaviours (Spencer et al., 2007). However, the behaviour change wheel (BCW) (Michie et al., 2014) was chosen over these. This is because it was developed from a synthesis of 19 behaviour change frameworks to overcome limitations in comprehensiveness, for example, it incorporates the context in which a behaviour occurs (Michie et al., 2011).

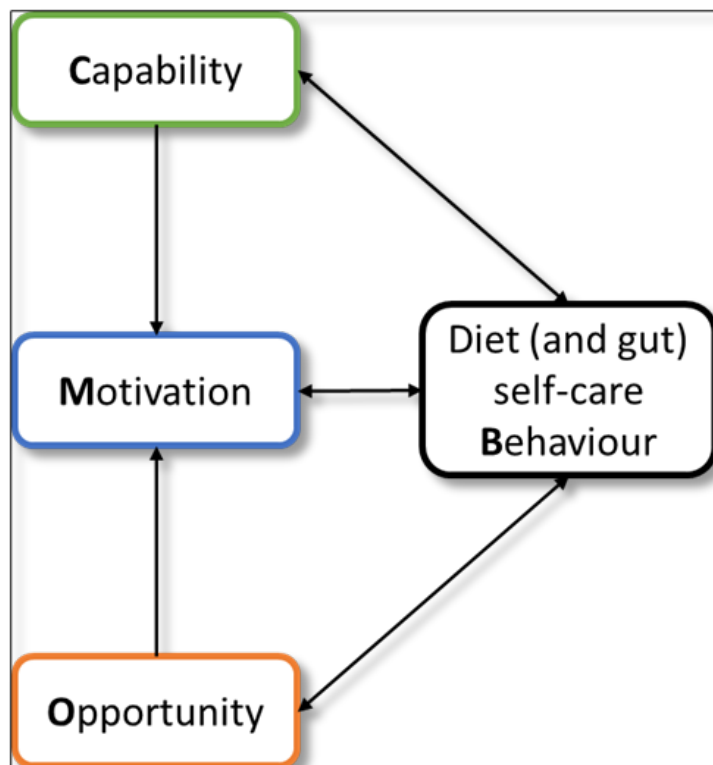
The BCW is comprehensive and pragmatic (Combes et al., 2021) and can be used to complement the development phase of the MRC framework (Staniford and Schmidtke, 2020). It has been widely used to understand and develop interventions for self-care/ self-management of LTCs (Brewster et al., 2021), including CF (Arden et al., 2021), as well as diet (e.g., Rohde et al., 2019), particularly in childhood weight management (e.g., Curtis et al., 2015). There

are, however, few examples of its application in interventions that directly target children (Koripalli et al., 2022).

At the hub of the BCW is the Capability, Opportunity, Motivation-Behaviour (COM-B) model, which recognises that behaviour is part of an interacting system (Figure 2-2) and for a behaviour to occur, there must be:

- Capability to perform the behaviour – can be either physical (e.g., having the skills) or psychological (e.g., having the knowledge)
- Opportunity for the behaviour to occur – can be physical (e.g., time) or social (e.g., social cues)
- Motivation to do the behaviour (rather than not, or engage in a competing behaviour) – can be reflective (e.g., planned) or automatic (e.g., habit) (Michie et al., 2014).

Figure 2-2: The COM-B model

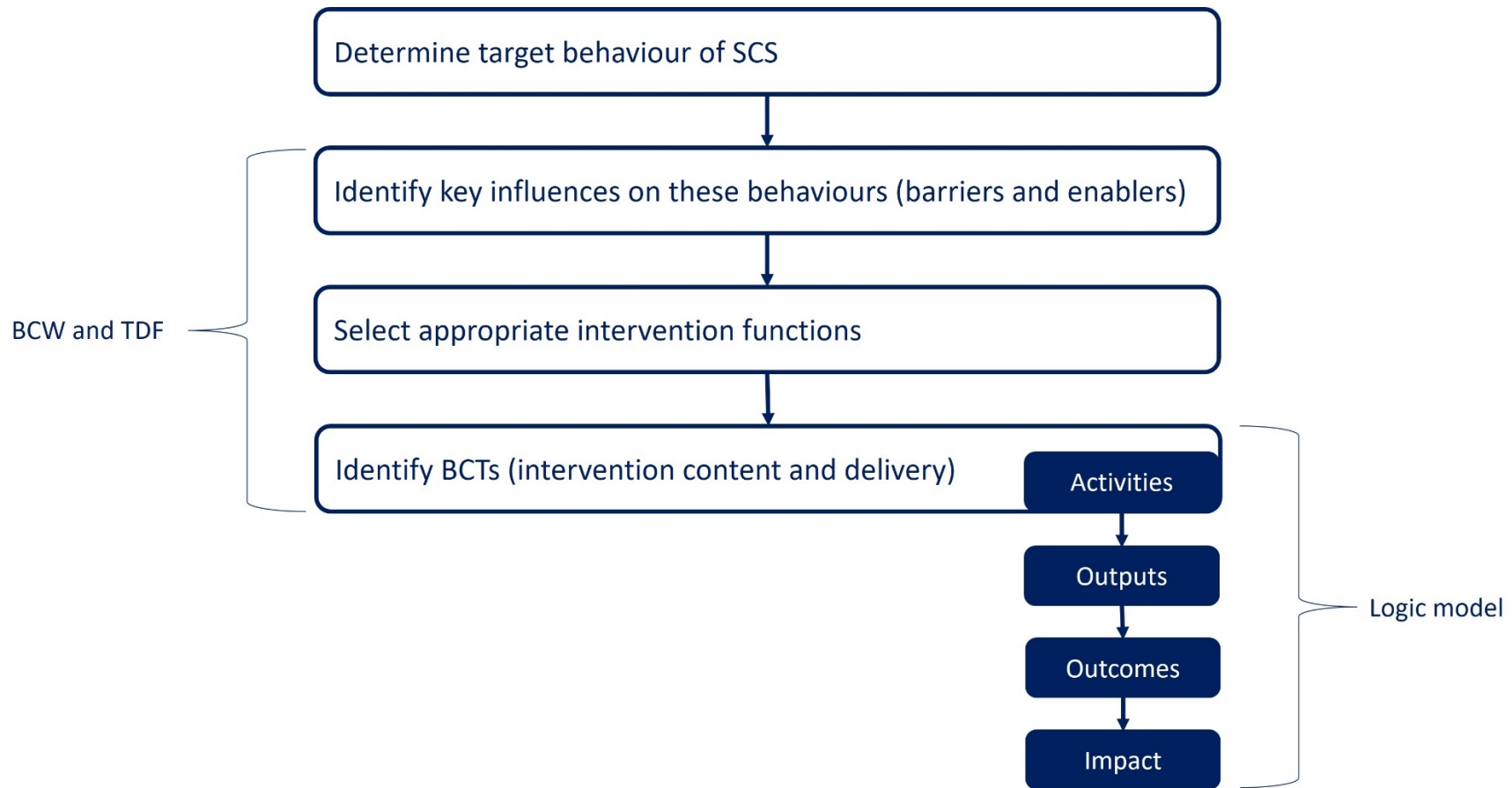


The COM-B model was used alongside the more finely detailed Theoretical Domains Framework (TDF) (Cane et al., 2012). (Appendix A on page 200 illustrates the links between COM-B components and TDF domains). Using the COM-B and TDF enabled the researcher to identify factors influencing (enabling or presenting a barrier to) individual and collective self-care/ SCS behaviours, in

the context in which they occur (Atkins et al., 2017). Such influencing factors could then be targeted to bring about the desired change in behaviour, through identifying appropriate intervention functions and behaviour change techniques ((BCTs), the 'active ingredients' of the intervention) using the BCT taxonomy (Michie et al., 2013).

Next, findings were combined in a draft model (conceptual framework) for SCS of diet and the gut as an intervention: its content, forms of delivery and outcomes, for evaluation with stakeholder groups (therefore addressing a lack of stakeholder involvement, highlighted as a limitation of the BCW (O'Cathain et al., 2019b)). Evaluation of the draft model with children and parents was based on advice from the CF Trust (the leading charity for CF in the UK), regarding what had worked well previously (Brownlee, 2019; 2021) and was conducted online, with the CF Trust facilitating throughout. For dietitians, evaluation was planned to take place in person at one of the bi-annual CF Specialist Group meetings. Following evaluation, a final model (conceptual framework) was developed as a logic model. A logic model approach was chosen as the best fit for summarising and communicating how SCS as a complex intervention could theoretically work (Baxter et al., 2014). This is because logic models illustrate the potential links between inputs (intervention resources), specific activities (BCTs), outputs (effects of the activities), specific short and longer-term outcomes and potential impacts (Allmark et al., 2013), so their development can follow on directly from using the BCW and TDF (Figure 2-3)

Figure 2-3: How the BCW and TDF link through to the logic model



BCTs: Behaviour Change Techniques; BCW: Behaviour Change Wheel; SCS: Self-Care Support; TDF: Theoretical Domains Framework

Logic models have been criticised for inadequately describing the dynamic nature and complexity of complex interventions, notably their interaction with context (Mills et al., 2019). To try to overcome this, an attempt was made to capture learning about the intervention-implementation-context of routine care (Mills et al., 2022). Another limitation of logic models is that they can be viewed as the end of a process, rather than a starting point (Peyton and Scicchitano, 2017). The intention in this study was that the logic model would represent the end point of the PhD, to be revisited and further refined at future stages in a programme of work. It would also ultimately inform future evaluation of the intervention.

In the next section, ethical considerations for the study are discussed.

2.8 Ethical considerations

Specific ethical considerations were raised for phase two.

2.8.1 Ethics and research governance approval

NHS ethics and Health Research Association (HRA) governance approvals were required for phase two, as participants were children with CF (and parents) receiving care at one of three specialist CF centres located at three NHS organisations in England.

An IRAS (Integrated Research Application System) form was completed for research involving qualitative methods only, with University of Leeds as the Sponsor. When a favourable opinion from Yorkshire and The Humber - Leeds West Research Ethics Committee (20/YH/0170) and HRA approval had been received, localised Organisation Information Documents and Schedule of Events documents were completed for each of the three participating NHS organisations, to confirm their capacity and capability to undertake the research activities. A non-substantial amendment was needed to appoint the specialist dietitians at each of the three centres as Principal Investigators (rather than local collaborators) as recommended in the HRA approval letter. Finally, a NHS to NHS proforma was completed to obtain a letter of access/ honorary research contract for the researcher to be able to attend each of the centres.

In addition, the study was eligible for inclusion in the NIHR Clinical Research Network (CRN) Portfolio. This enabled access to NIHR CRN support in monitoring progress with recruitment.

Impact of Covid-19

There were delays of between three and seven months in obtaining confirmation of capacity and capability at the participating NHS organisations due to prioritisation of Covid-19 studies.

2.8.2 Key ethical considerations

Key ethical considerations and how these were addressed, are summarised in Table 2.3. They are based on research ethics guidance (ESRC, 2018; ERIC, 2020; BPS, 2021; SRA, 2021) and training completed by the researcher in Good Clinical Practice (GCP), University research ethics and ethical research with children.

Table 2.3: Key ethical considerations for phase two

Ethical consideration	Steps taken
Gaining access/ identification of children as potential participants	Only the specialist children's dietitians at each of the three CF centres (who were members of the child's direct clinical care team) accessed patient records to check which children met the eligibility criteria (outlined in Table 5.1)
Obtaining informed consent/ assent	<p>The following were obtained immediately prior to interviews:</p> <ul style="list-style-type: none"> - Written informed assent directly from the child (Oulton et al., 2016) together with parental consent for the child - Written informed consent of parents and dietitians. <p>[As described in section 2.11, the patient information sheets for ages 6-8 years and 9-11 years and assent form were developed with children of the same age as potential participants, so they used language children could understand (Huang et al., 2016)].</p> <p>It was emphasised that participation was completely voluntary and that they could decline to take part or withdraw themselves and their data from the study at any time without having to give a reason, and for families, without detriment to the child's care. If children chose to participate, they were observed for verbal and non-verbal cues that they were not willing participants, as they might have felt pressured to continue if their parent/ carer wished/ insisted they do.</p> <p>To facilitate this, children were provided with a yellow and red card and encouraged to hold up the yellow card if they wished to pause e.g., to skip a question, and the red card if they wanted to stop the interview.</p>

Ethical consideration	Steps taken
Potential for distress	Though the topic of SCS was not outwardly intrusive or sensitive, a participant may have become upset when discussing their experiences and needs. Should this have happened, they would have been asked if they wished to pause or end the interview (and if they wanted to discuss any issues raised with a trusted member of their CF care team).
Confidentiality and anonymity	<p>Complete confidentiality was aimed for but could not be guaranteed; if participants had disclosed information about potential harm/ risk to themselves or others, the researcher would of shared the disclosure with the named person responsible for safeguarding at the respective CF centre. This limit to confidentiality and how it would be managed was highlighted to participants (Kirk, 2007).</p> <p>Anonymity could be fully assured. The University of Leeds Protocol on the protection, anonymisation and sharing research data (2019) was followed, which incorporates the Data Protection Act 2018.</p> <p>Details regarding the collection, storage and use of personal information during and at the end of the study were included in a University of Leeds data management plan that was regularly reviewed.</p>

The next section in this chapter outlines the quality considerations for the study.

2.9 Quality considerations

To enhance the quality or ‘trustworthiness’ of the research process and subsequently the data gathered in phase two using a qualitative descriptive approach, the following criteria, based on the seminal work of Lincoln and Guba (1985) were applied: credibility, dependability, confirmability and transferability (Bradshaw et al., 2017).

Credibility

Credibility refers to the truth value of the findings, whether (or not) they represent the original data and accurately present participants’ perspectives (Noble and Smith, 2015). This was achieved by:

- Having a clear audit trail of the analysis process (section 5.2.4), and checking interpretations with supervisors and the project advisory group
- Repeatedly revisiting the data to ensure emerging themes remained close to participants accounts

- Checking whether emerging themes resonated with subsequent participants
- Presenting rich descriptions of participants perspectives and their context, supported by quotations from participants (section 6.2.3)
- Researcher reflexivity throughout (see below).

However, interview transcripts were not shared with individual participants to verify their accuracy (member checking) (Bradshaw et al., 2017) as this was thought to be too burdensome.

Dependability

Dependability refers to the consistency in which the study was undertaken across participants, and over time (Morrow, 2005). This was demonstrated through detailed description of all study procedures used to generate the findings, and decisions taken throughout the study, for example, in managing the impact of Covid-19. This included the use of standardised information for recruiting dietitians (section 5.2.2) and interview topic guides (section 5.2.3) (Colorafi and Evans, 2016).

Confirmability

Confirmability refers to the findings being clearly derived from the data and requires exploration of any researcher bias or subjectivity. It was addressed by:

- Researcher reflexivity throughout, for example, to increase awareness of, and account for, the researcher's prior experience and perspectives as a clinician and researcher (key learning points shared in section 5.2.5)
- Transparent reporting of all study procedures and decisions taken (an audit trail as for dependability)
- Independent checks by supervisors at each stage of the analysis process
- Inclusion of direct quotations from participants (section 6.2.3) (Bradshaw et al., 2017).

Transferability

Transferability refers to the extent to which the reader can generalise the findings of the study to their own context (Morrow, 2005). This was aided by:

- Proving information about the researcher and the research context (Morrow, 2005)
- Presenting detailed information regarding sampling (section 5.2.1)
- Describing characteristics of participants (section 6.2.1)
- Presenting rich descriptions
- Suggesting how the findings may be tested further by other researchers (Colorafi and Evans, 2016).

In addition to these criteria, reflexivity is an essential component in enhancing quality (Korstjens and Moser, 2018).

Reflexivity

Reflexivity refers to the researcher undertaking critical self-reflection on their own biases, assumptions, preferences, preconceptions, and their relationship with the participants, and how these could have influenced the research and its findings (Morse, 2018). A reflexive journal was kept throughout the study to support this process. In addition, identification of the researcher as an experienced clinician researching in her practice area, and motivations for doing this, were explicitly stated (section 1.4).

Finally, to aid transparency and comprehensive reporting, several checklists were used throughout the study phases. These were the PRISMA checklist (Page et al., 2021) in phase one, the COREQ checklist for interviews (Tong et al., 2007) in phase two, and the TIDieR checklist for better reporting of interventions (Hoffman et al., 2014) in phase three.

This chapter concludes with a discussion about study management and how patient and public involvement was woven throughout the study.

2.10 Roles and responsibilities of the study management group

A Project Advisory Group (PAG) was convened for the study. The group provided expertise and advice and fulfilled the following responsibilities:

- Oversaw the study
- Monitored progress against the study timetable
- Helped identify, and suggest solutions for, any issues or concerns that arose during the study to ensure milestones were achieved.

Specific examples of input included reviewing the draft protocol, reviewing the recruitment plan and assessing progress midway through the recruitment period, checking analysis and interpretation of findings, and reviewing plans for dissemination.

The group comprised two parents of children with CF, a Lead Paediatric CF Consultant, a Clinical Specialist Paediatric CF Dietitian and a Professor of Family and Child Health with expertise in self-care of childhood LTCs.

The parent members brought lived experience of caring for a child with CF, of supporting their child's journey towards independence in self-care and of participating in dietetic consultations. The researcher supported the parents involvement, for example, through providing an induction pack (including the 'Starting Out' guide (NIHR INVOLVE, 2017), a research jargon buster, information regarding training/ learning opportunities), offering pre-meetings to discuss meeting agendas and content, having regular contact via email, reimbursing expenses and providing payment for their time and expertise.

2.11 Patient and Public Involvement (PPI)

The following aspects of the research process actively involved patients (children with CF), parents/ carers of children with CF and members of the public.

Prior to the study

To inform the funding application for the Fellowship, a formal PPI consultation was completed with six families (six children with CF and seven parents) attending a Specialist CF Centre. They shared insights of their needs and priorities for SCS and advised on the acceptability, design and conduct of the proposed study, for example, method of data collection (including mode, setting, and what would make it fun to take part). As a result of their involvement, the study aims and outcomes gained clarity and the study conduct was changed to be more accessible and engaging to children with CF and their families. In addition, one parent continued their involvement by becoming a member of the PAG.

During the study

Development of the participant-facing documentation for children: two participant information sheets, for age ranges 6-8 years and 9-11 years, and an assent form, was informed by Ford et al., (2007) and guidance from the NIHR Generation R Alliance Young Person's Advisory Group (2019). The documents were then reviewed by:

- Five members of a PPI group (the Young Person's Advisory Group (YPAG*) at a Children's Hospital)
- Three children with CF attending a Specialist CF Centre (who met the same eligibility criteria as children targeted in the study)
- Two members of the public (children in the same age range as children targeted in the study).

This was to ensure information provided to children was easy to understand and interesting.

Study invitations, participant information sheets and consent forms for parents/ carers were reviewed by the two parent members on the PAG.

Collectively, feedback received on the language used, clarity, layout and design, informed several improvements to the documentation. For example, some words were changed to reflect the words children use or hear and understand, such as replacing 'enzymes' with 'Creon'; a sentence was added to explain why their real name would not be used; some questions on the assent form were combined and simplified; the colours were liked and it was highlighted that printing them in colour would add to their appeal.

Further input during the study included:

- Feedback on the interview topic guide and appropriateness of interview techniques for children, provided by the PPI group (YPAG)
- Evaluation of the draft model by two involvement groups (one for children with CF and parents, and one for youth members) organised by the CF Trust.

The researcher provided timely feedback to participants regarding how their contributions shaped the research (Preston et al., 2022) and provided payment to YPAG members for their time and expertise.

*YPAG brought knowledge and experience of assisting researchers to improve design and conduct of studies involving children. Interaction with the group was facilitated by a Senior Research Nurse at the Children's Hospital.

2.12 Chapter summary

This chapter has presented how the study has an overall pragmatic stance, with methodology guided by the development phase of the MRC framework and BCW. It has provided a rationale for the design and working methods chosen for each of the three study phases. It has also outlined how ethical and quality considerations and the input of the PAG and various PPI has shaped the study.

The following chapter will describe the process of conducting phase one of the study - an evidence synthesis using an IR methodology.

Chapter 3

Phase One: Evidence Synthesis

3.1 Introduction

This chapter presents phase one of the study, a synthesis of existing theory and evidence for SCS of diet and the gut in children with LTCs, to develop a comprehensive understanding of what is known about the topic and identify gaps in knowledge (Noble and Smith, 2018). Having chosen an IR design to meet the objective for phase one and described the rationale (Chapter 2, Methodology), this chapter describes how the methodological guideline of Whitemore and Knafl (2005) was applied. It then presents the findings as a thematic framework and narrative and concludes with a discussion of the findings, which informed phases two and three of the study.

A protocol was developed and registered on PROSPERO, an international prospective register of systematic reviews (CRD42019144941) (Cave et al., 2019) and the findings published (Cave et al., 2021). Published tables as online supplemental material are included in the appendices and are referred to throughout the chapter. Furthermore, the chapter details how the IR was updated following publication, with additions to the tables highlighted as appropriate.

3.2 Methods

The description and critique of the procedures undertaken follow the five IR stages outlined by Whitemore and Knafl (2005):

- Problem identification
- Literature search
- Data evaluation
- Data analysis
- Presentation (section 3.3).

In addition, at each stage the relevant items in the 2020 PRISMA checklist are addressed to enhance the transparency, completeness and accuracy in reporting (Page et al., 2021).

3.2.1 Problem identification

The IR was the first phase in a three-phase study seeking to explore the research problem described in chapter one: to begin addressing the evidence gap between policy advocating SCS and the what and how to deliver SCS of diet and the gut in the routine clinical care of children with CF.

The IR built on the findings of two systematic reviews on SCS for children with LTCs (Kirk et al., 2013; Bee et al., 2018) and a Cochrane review on self-management education for CF (Savage et al., 2017) discussed in section 1.8. However, it differed from the existing reviews in three ways:

- a) In addition to quantitative studies, inclusion of qualitative and mixed methods studies was considered
- b) The inclusion of theoretical literature, as appropriate
- c) The focus was on SCS of diet and the gut.

The aim of the IR was to obtain a comprehensive understanding of what is already known about SCS of diet and the gut, by identifying, critically appraising and synthesising current evidence/ theory from other childhood-onset physical LTCs with a diet and/ or GI-related component of care. This may have led to identification of an existing theoretical framework that could be adapted for CF, with the evidence gathered in phase two seeking to refine or challenge this theory. Or, inform development of a theoretical framework for SCS of diet and the gut in children with CF, to meet objective one of the study.

The IR addressed two questions:

- (1) What is SCS of diet and the gut for school-age children with LTCs?
- (2) What models of SCS have worked, when and how, in the routine care of school-age children with LTCs, including enablers for and barriers to, delivery and uptake?

The first question sought to define what SCS of diet and the gut consists of, as reported in the literature. The second question sought to identify existing models of SCS that have worked (or not), the characteristics of these and what helped or hindered their delivery and uptake in routine care.

3.2.2 Literature search

Selection criteria

The selection criteria were developed iteratively using the SPIDER (Sample, Phenomenon of Interest, Design, Evaluation, Research type) framework (Cooke et al., 2012). This framework was chosen over other frameworks such as PICO (Schardt et al., 2007) as its components were more relevant to the review questions and aided inclusion of search terms relevant to retrieval of not only quantitative, but also qualitative and mixed methods studies (Amir-Behghadami, 2021).

Inclusion and exclusion criteria are outlined in Table 3.1, with additional rationale as follows:

- The age range of children was wider than the target age range for the SCS intervention (6-11 years), as there are a dearth of studies specifically targeting 6-11 years, however, the mean age was capped at 16 years, so that the focus of selected studies did not extend beyond mid-adolescence and into late-adolescence/ young adulthood (WHO, 2022)
- The focus was on children with a physical LTC with a diet or GI-related component of care. Several exemplar conditions, in addition to CF, were also included, that require lifelong dietary treatment/ modification:
 - T1DM - requires the dose of insulin to be matched to the amount of carbohydrate, this is similar to CF in which the dose of enzyme is matched to the amount of fat
 - Coeliac Disease - requires a strict lifelong adherence to a gluten-free diet (White et al., 2016)
 - Phenylketonuria – like CF, this is a rare genetic condition; it requires a restriction in protein intake to minimise intake of the amino acid phenylalanine (Belanger-Quintana et al., 2011)
 - Inflammatory Bowel Disease – this includes Crohn’s disease and ulcerative colitis, which like CF, are characterised by chronic inflammation of the gut; diet is modified to improve symptoms and prevent relapse (Halmos and Gibson, 2015).

Table 3.1: Inclusion and exclusion criteria

(Published as S1 in Cave et al. (2021))

Criteria	Inclusion	Exclusion
Sample: participants and conditions	Focus on <ul style="list-style-type: none"> Children of compulsory school age (4-16 years old), or Child-parent/ carer dyads 	<ul style="list-style-type: none"> Mean age of children reported as under four years of age or over 16 years of age Focus on parents/ carers only
	Children with <ul style="list-style-type: none"> Any physical LTC with a diet or GI-related component of care, or Any of the following: CF, T1DM, Coeliac Disease, Phenylketonuria or Inflammatory Bowel Disease 	<ul style="list-style-type: none"> Children with non-physical LTCs e.g., mental health conditions No specific diet/ GI component
Phenomena of Interest	<ul style="list-style-type: none"> Engagement or active involvement of children in some aspect of self-care of diet and/ or the gut, having received some type of SCS (no restrictions regarding the format or mode of delivery of SCS) Enablers for and barriers to, delivery and uptake of SCS 	<ul style="list-style-type: none"> Focus on perspectives/ perception of self-care only Children's involvement in self-care of their parents or siblings LTC Transition from child to adult health services (except where a study relates to supporting increased independence for self-care pre-transition)
Design	<ul style="list-style-type: none"> No restriction - qualitative, mixed methods and quantitative studies of all designs 	
Research type	<ul style="list-style-type: none"> Published literature and grey literature sources such as conference proceedings, dissertations and theses 	<ul style="list-style-type: none"> Editorials, commentaries/ opinion papers and protocols

CF: cystic fibrosis; GI: gastrointestinal; LTC: long-term condition; SCS: self-care support; T1DM: type 1 diabetes

Search strategy

A wide range of information sources were searched to identify studies. These included the following bibliographic databases relevant to healthcare: CINAHL, MEDLINE, Embase, PsycINFO, Scopus, Web of Science, the Cochrane Library, and OpenGrey, as a source of grey literature (inclusion discussed in section 2.5). The registers ISRCTN and ClinicalTrials.gov were also searched to identify possibly relevant ongoing and completed studies.

Each source was searched with the date range of 1st January 1990 to 31st July 2020, to reflect the development of policy and research in self-care/ self-management of LTCs since the 1990s (Lorig and Holman, 2003). Searches were adapted for the different databases, included use of database specific subject headings and free-text terms and variations relating to: diet/gut self-care; children; LTCs. An example of the full line-by-line search strategy run in MEDLINE is included as Appendix B.

Supplementary search strategies included citation searching (in Scopus and Web of Science), targeted author searches and hand searching reference lists of included studies and review articles on the same or similar topics. Searches were limited to studies published in English as translation services were not available to the researcher.

The search strategy was reviewed by an experienced information specialist at the University library. Their input facilitated a balance to the searches, in them being sensitive yet specific.

Study selection

Search results were collated, uploaded into EndNote X8 (Thomson Reuters, 2016) and duplicates removed. A two-stage screening process was then completed. In stage one, titles and abstracts were screened. Studies that met or potentially met the inclusion criteria, were taken forward to stage two, in which the full texts were retrieved and assessed in detail against the inclusion criteria. Where required, missing or additional information to clarify eligibility was sought via companion papers or email requests to authors (Hong et al., 2018). Both stages were led by the researcher, with independent review by supervisors LM and GM. Any differences in opinion were resolved through discussion until consensus was reached.

3.2.3 Data evaluation

Data extraction

Data were extracted from included studies using a pre-piloted data extraction table in Microsoft Excel. The data extracted included specific details about the research aim, participants (including LTC, age and number of participants), study design and methods, SCS intervention/ exposure (including setting),

theoretical basis, outcomes assessed, key findings and supporting data for quality appraisal. Data were extracted by the researcher, 10% of the full extraction checked for accuracy and completeness by LM and GM and any discrepancies resolved through discussion.

Quality appraisal

The methodological quality of included studies was assessed using the validated critical appraisal tool MMAT (version 2018) (Hong et al., 2018) in Microsoft Excel, with the rationale described in section 2.5.

Use of the MMAT for each of the included studies comprised of (1) completing screening questions to assess whether appraisal using MMAT was appropriate, and (2) selecting the appropriate category from five study designs (qualitative, quantitative (randomised controlled trials, non-randomised or descriptive) or mixed methods) and responding yes/ no/ can't tell to the five core appraisal criteria for that study design. The researcher conducted the quality appraisal, with 10% of the full assessment independently reviewed by LM and GM and differences in the assessment of two studies resolved through discussion.

3.2.4 Data analysis

Data reduction, display and comparison

To combine the data in a single synthesis, the quantitative data (extracted from quantitative studies and the quantitative component of mixed methods studies) was converted into textual descriptions to facilitate integration with extracted qualitative data. Assembled data were then organised into categories based on similarity in meaning, and displayed in a matrix. This enabled comparison of data and identification of patterns, variations and relationships within and across the studies (Whittemore and Knafl, 2005).

Conclusion drawing and verification

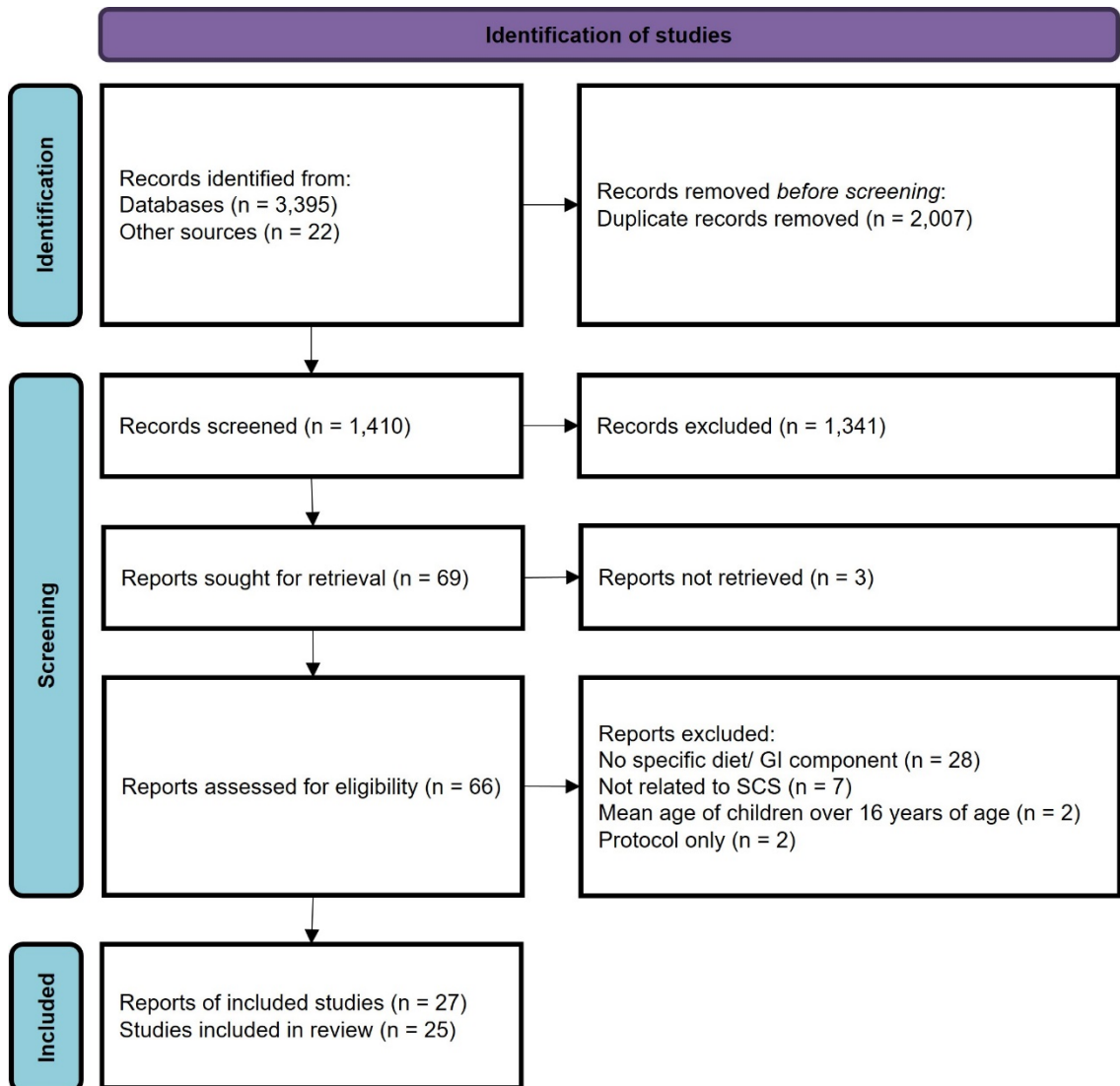
The researcher conducted the analysis and integrated findings to develop a set of themes. These were checked against the primary source data for accuracy and confirmability. Themes were confirmed following discussion with LM and GM and presented as a thematic framework and narrative summary.

3.3 Findings

Presentation stage

The search identified 3,417 records; 2,007 records were excluded before screening. 1,410 records were screened, with 1,341 excluded after titles and abstracts. Three potentially eligible reports were not retrievable, but 66 reports were retrieved for detailed evaluation. 39 reports did not meet the inclusion criteria, chiefly because a diet/GI component was not specified or it was not related to any type of SCS. A total of 25 studies (from 27 reports) were included in the review. The selection process is summarised in Figure 3-1 (Page et al., 2021).

Figure 3-1: PRISMA flow diagram



Note: a 'report' could be a journal article, conference abstract, study register entry, dissertation or any other document providing relevant information.

Updated search findings

The review was updated (see Table 3.2) on 31st March 2022 by re-running the searches using the same procedures described above. The search identified 415 records; 291 records were excluded before screening. One hundred and twenty four records were screened, with 79 excluded after titles and abstracts. One potentially eligible report in an Italian journal was not retrievable, however, 17 full reports were retrieved for detailed evaluation. Fifteen reports did not meet the inclusion criteria. Reasons for not meeting the inclusion criteria were the same as the initial search, with the addition of: not engaging/ actively

involving children (n=5). The search provided a further two studies (from two reports) for inclusion; consequently, a total of 27 studies (from 29 reports) were included in the updated review.

Table 3.2: Updated search findings

Search activity	Results
Date range	1st August 2020 – 31 st March 2022
Records identified (after duplicates removed)	124
Records screened	45
Full-text screening	17
Studies identified for inclusion	2
Total (with 25 from the initial search)	27

In the updated search, there was a significant increase in focus on supporting self-management via technology in T1DM, with knowledge and skills training in the use of continuous glucose monitoring and insulin pumps, though not with any clear inclusion of a diet component alongside.

3.3.1 Description of included studies

Characteristics of the included studies are summarised in Appendix C.

Overall, the included studies involved children and adolescents aged 2-19 years, predominantly with T1DM (n=13) and CF (n=9), followed by Coeliac Disease (n=2), Phenylketonuria (n=2), concurrent Coeliac Disease and T1DM (n=1) and Inflammatory Bowel Disease (IBD) (n=0). Four of the CF studies were sourced from reports in the grey literature, and through contacting authors, extensive additional information was obtained (Bell, 2004; Culhane, 2013; Owen et al., 2013 and Boon et al., ahead of inclusion of their 2020 publication). Identification of patient education interventions to improve self-management was previously rated as a research priority for IBD (Dibley et al., 2017), so it was perhaps surprising that no studies involving children and adolescents with IBD met the inclusion criteria.

The studies were conducted in Europe (n=13), North America (n=11), Australia (n=1), Brazil (n=1) and Israel (n=1), and mainly reported between 2013 and 2019 (n=16). Of the 27 studies, 20 were quantitative, five mixed methods and two qualitative designs. The majority of studies (n=21) provided models of SCS, whilst the others (n=6) informed the context of SCS. Several studies presented only limited detail about the diet/GI-related component of care (Cottrell et al., 1996; Nabors et al., 2014; Cooper et al., 2018; Fiallo-Scharer et al., 2019; D'Souza et al., 2021), the dietary self-care programme (Austin et al., 2011; Austin et al., 2013) or routine support (Rankin et al., 2018a). However, whilst this was noted as a limitation (see section 3.4), these studies were included as they could potentially contribute to answering the review questions.

3.3.2 Quality appraisal

A summary of the quality appraisal findings using MMAT¹ are presented in Appendix D. The following narrative addresses key aspects of the quality appraisal, based on the MMAT criteria for each of the five study designs.

There were nine RCTs: Cottrell et al., 1996; Stapleton, 2001; Davis et al., 2004; Stark et al., 2009; Spiegel et al., 2012; Coates et al., 2013; Christie et al., 2016; Price et al., 2016; Fiallo-Scharer et al., 2019. The MMAT criterion for this category focussed around the assessment of biases e.g., selection, attrition, detection and reporting bias (Mansournia et al., 2017). With the exception of Price et al. (2016), there were many examples of insufficient information, leading to a 'can't tell' response, with study conclusions potentially prone to bias.

There were three non-randomised studies: Singh et al., 2000; Revert et al., 2018; D'Souza et al., 2021. Key criteria for this category were assessment of attrition bias and accounting for confounding factors. D'Souza et al. (2021) rated poorly for both, so an association may be masked or falsely demonstrated between the education module and outcomes (Skelly et al., 2012).

¹ As the screening questions for Bell (2004) could not be answered, appraisal using MMAT was not appropriate for this study

There were seven descriptive studies: Austin et al., 2011 and 2013; Culhane, 2013; Owen et al., 2013; Nabors et al., 2014; Witalis et al., 2017; Fishman et al., 2018; Meyer and Naveh, 2021. As these were mainly survey studies, the key criterion was assessment of response bias (Jones et al., 2013). This was high for Nabors et al. (2014), as only 68/129 (52%) of children attending the 2012 camp completed the survey, therefore children's learning due to the camp could not be fully assessed.

There were five mixed methods studies: Kyngas et al., 1998; Froisland and Arsand, 2015; Cooper et al., 2018; Connan et al., 2019; Boon et al., 2020. Using MMAT, these studies required separate assessment of the qualitative and quantitative components, and then a mixed methods criteria that mainly focused on integration of the components. All criteria were met in the qualitative component of the five studies. Where the quantitative component was non-randomised, confounders were not accounted for by Froisland and Arsand (2015), and with a descriptive quantitative component, there was insufficient information to determine responses to any of the criteria for Kyngas et al. (1998). For the mixed methods criteria, studies did successfully integrate qualitative and quantitative components, with outputs of the integration adequately reported, though Kyngas et al. (1998) only displayed results together, with no integration or rationale for adding the quantitative component.

Finally, there were two qualitative studies: Sparapani et al., 2017; Rankin et al. 2018a and 2018b. These met all of the MMAT criteria, however, neither the studies nor MMAT considered reflexivity (the importance of which was discussed in section 2.9), though this is included in the CASP qualitative checklist (CASP, 2018).

In addition to the MMAT criteria, other methodological strengths and limitations are highlighted throughout section 3.3.4. For example, regarding the variable and often short duration of interventions, and follow-up post intervention. Whilst several studies had follow-up of two years to explore the medium-longer-term effects of SCS (Stark et al., 2009; Coates et al., 2013; Christie et al., 2016; Price et al., 2016), other studies had no follow-up to examine if knowledge acquired or skills learnt were employed after the intervention ended (Davis et al., 2004; Nabors et al., 2014). However, having adequate follow-up is important, not only in assessing outcomes but also the full impact of the

intervention (intended and unintended), to inform future decisions regarding intervention development and implementation (Clarke et al., 2019).

Also, in some of the studies that used usual care as a comparator, there was wide variation in the complexity and duration of education offered in usual care (Christie et al., 2016; Price et al., 2016), with some usual care close to what was delivered in the intervention (Stark et al., 2009; Spiegel et al., 2012), making it difficult to observe a change in outcome as a result of the SCS intervention.

Despite various methodological limitations across the studies, they were generally rated fair to good using MMAT. A decision was therefore taken by the researcher, in agreement with supervisors LM and GM, that no studies would be excluded based on the findings of the quality appraisal. However, study limitations were taken into consideration in the synthesis, in judging the strength of evidence for developing themes.

3.3.3 Overview of the findings as a thematic framework

Through synthesis of all of the included studies, six themes were identified. Two themes related to the first review question and four to the second review question. An overview of these themes is presented in Table 3.3 and detailed in the narrative below.

Table 3.3: Thematic framework for phase one

SCS of diet and the gut for school-age children with LTCs is	
Support in developing and applying specific knowledge and skills	Knowledge and skills development, essential through to advanced, includes understanding how treatments such as PERT and insulin work and how to administer, skills such as reading food labels, estimating portion sizes, self-monitoring. F: visual; B: numeracy skills
	Practice/ repetition over time to develop confidence in applying knowledge and skills
Practical help with incorporating the demands of self-care into everyday life	Support to keep going with daily self-care, includes physical access to foods, autonomy support from HCPs and parents. Includes helping child/ adolescent to address factors affecting motivation to self-care, facilitating gradual transfer of responsibility for self-care over time. F: individualised
Models of SCS were more successful	
When starting early and keeping it going	Early in disease course/ early childhood (pre-teens). Ongoing stepped approach, regular reinforcement (leading to development of habits/ routines?)
(How) Being flexible	Variety of formats and modes of delivery, incorporated into routine care rather than as an optional extra. Flexibility to tailor to individual needs. F: visual, interactive (not passive); B: time, needs service and system level buy-in
When success is measured by choosing appropriate outcomes	Current overreliance on clinical (reflective of medium-long-term changes); more outcomes and outcome measures that are patient-reported/ relevant to SCS are needed.

F: facilitators; B: barriers

3.3.4 Narrative of the findings

In the following narrative, the six themes in the thematic framework are presented in detail, to answer, in turn, the two review questions.

3.3.4.1 First review question: What is SCS of diet and the gut for school-age children with LTCs?

Support in developing and applying specific knowledge and skills

There was consistency across the studies in supporting children and adolescents' capability to self-care, beginning with development of essential, through to more advanced, knowledge and skills, with repeated opportunities to practice and develop confidence in their application.

Essential knowledge and skills

The common starting point for school-age children in many of the studies, was knowing how to identify which foods contained fat, carbohydrate, protein or gluten, as this set the foundation for selecting foods to eat, or restrict or avoid completely, as appropriate to the LTC (Singh et al., 2000; Owen et al., 2013; Christie et al., 2016; Sparapani et al., 2017; Fishman et al., 2018; Boon et al., 2020). Further knowledge was required to estimate how much fat, carbohydrate, phenylalanine or protein was contained in foods (Coates et al., 2013; Owen et al., 2013; Witalis et al., 2017), possibly through developing the skill of reading and interpreting food labels (Bell, 2004; Culhane, 2013; Rankin et al., 2018a). However, as this skill also relied on knowing how to estimate portion sizes, Spiegel et al. (2012) encouraged dietitians to support children and adolescents to repeatedly practice estimating portion sizes using real food and food models, alongside measuring actual portions. An alternative approach, in which adolescents with T1DM took photos of their own foods using an app (Froisland and Arsand, 2015) had the advantage of facilitating estimation of both carbohydrate content and portion sizes, and with further development the app may be a useful tool for SCS.

Advanced knowledge and skills

More advanced knowledge was required to begin making sense of the complex relationships between carbohydrates, blood glucose and insulin (Coates et al., 2013; Froisland and Arsand, 2015; Christie et al., 2016); fats, absorption and PERT (Boon et al., 2020); food containing gluten and absorption (Connan et al., 2019). In several studies, promoting a visual understanding of these relationships through use of an app (Froisland and Arsand, 2015), an interactive e-learning module (Connan et al., 2019) or a video game (Sparapani et al.,

2017), helped children and adolescents to make sense of what was happening inside their bodies as a result of Coeliac Disease and T1DM.

In many of the studies, the advanced skill of self-monitoring enabled recognition and management of GI symptoms (Cottrell et al., 1996; Stapleton, 2001; Bell, 2004; Culhane, 2013) hypo and hyperglycaemia (Nabors et al., 2014) and also tracking adherence to daily goals (Stapleton, 2001). Some studies combined building knowledge on how PERT or insulin works, with skills on administering and understanding what happens if too little or too much is taken (Bell, 2004; Davis et al., 2004). Further to this, studies focused on the advanced skill of titrating the dose of PERT to fat intake (Bell, 2004; Owen et al., 2013; Revert et al., 2018) and the dose of insulin to carbohydrate (Coates et al., 2013; Christie et al., 2016; Price et al., 2016). Only one study (Rankin et al., 2018a), together with a companion paper to Coates et al. (2013), (Chaney et al., 2010), highlighted poor mathematical comprehension as a barrier to performing this complex self-care task. Children adopted strategies to limit the need for complex maths skills such as choosing foods with carbohydrate values they could remember or using mobile phones to contact their parents about carbohydrate contents (thus remaining reliant on their parents) (Rankin et al., 2018a). In the trial of an app for CF, the enzyme dose calculation was the most used function by both children and their parents (Boon et al., 2020). This highlights numeracy as a vital core skill for self-care of diet and the gut in T1DM and CF.

Practical help with incorporating the demands of self-care into everyday life

Across many of the studies, SCS consisted of practical help for children and adolescents to have sustained opportunities and motivation to perform daily self-care. In some studies, performing daily self-care relied on the creation of supportive physical environments, in which there was availability of planned foods for children with CF (Stapleton, 2001) and low protein foods for adolescents with Phenylketonuria (Singh et al., 2000). Children and adolescents' ability to perform self-care also varied with the social interactions and support received from HCPs, parents, and friends (Kyngas et al., 1998; Singh et al., 2000; Stapleton, 2001; Spiegel et al., 2012; Austin et al., 2013;

Froisland and Arsand, 2015; Witalis et al., 2017; Rankin et al., 2018b; Revert et al., 2018; Boon et al., 2020).

In summary, across these studies, findings suggest HCPs had roles in directly supporting children and adolescents, supporting their parents and potentially supporting their friends.

HCPs supporting children's growing autonomy

Adolescents who felt their HCPs understood their dietary self-care challenges, accepted them as they were, provided them with choices and rationale about dietary self-care, were more motivated toward dietary self-care and had greater self-confidence to self-care over time (Austin et al., 2013). Equally, children and adolescents valued HCPs who gave tailored advice and timely feedback (Froisland and Arsand, 2015; Boon et al., 2020), particularly where this was simple practical advice relevant to their immediate situation, so they could take action and be in charge (Froisland and Arsand, 2015).

Kyngas et al. (1998) findings highlighted the need for HCPs to ensure consultations are not dominated by disease-monitoring activities such as blood tests, to permit time for discussion of perceived barriers to self-care and how to integrate self-care into their daily lives. In other studies, this extended to discussing other factors that may affect motivation to self-care, such as attitudes and beliefs in adolescents with Phenylketonuria (Singh et al., 2000), goal setting in children with T1DM (Nabors et al., 2014) and emotions around food intake in children with T1DM (Sparapani et al., 2017). Motivational messages from dietitians were valued if these were personalised, rather than generic (Floch et al., 2020) and improvements in HbA1c were largely due to resources that addressed patient-specific barriers to motivation (Fiallo-Scharer et al., 2019), underlining the importance of individualised patient-centred care.

HCPs supporting parents

Across several studies, HCPs provided practical support to parents in positively accepting and encouraging their child's growing independence (Witalis et al., 2017). They facilitated a balance of parents not exerting too much pressure or control (Kyngas et al., 1998; Austin et al., 2011) or having too little involvement, as adolescents with T1DM who collaborated more with their parents had better

metabolic control (Spiegel et al., 2012). In addition, the value of an app to adolescents with T1DM was greater with parental support (Cooper et al., 2018).

Across some of the studies, the importance of asking children directly about their views/ goals/ barriers for self-care/ management, rather than obtaining by proxy from parents, was identified. For example, parents recognised some of the unachieved goals and identified self-management barriers to target were inappropriate for their children (Stapleton, 2001; Fiallo-Scharer et al., 2019). Further to this, HCPs facilitating negotiation and setting of joint goals (Kyngas et al., 1998) or allowing parents and children time to discuss similarities and differences in their chosen self-care goals (for the child), may improve understanding and lead to greater family collaboration (Nabors et al., 2014).

HCPs supporting friends and peers

In one study, Rankin et al. (2018b) suggested HCPs could assist small friendship groups, to enable close friends of children with T1DM to provide emotional and practical support at school in the form of monitoring and prompting self-care tasks and practically helping perform tasks. Furthermore, they suggested HCPs consider working closely with schools to increase awareness and understanding of the need for self-care tasks among school peers, to help reduce stigma and normalise performance of self-care throughout the school day. However, further research is needed to explore the perspectives and experiences of friendship groups and class/school peers.

3.3.4.2 Second review question: What models of SCS have worked, when and how, in the routine care of school-age children with LTC's (including enablers for and barriers to, delivery and uptake)

Four themes related to the second review question: starting early, keeping it going, being flexible, and choosing appropriate outcomes.

Starting early

Across the included studies, models of SCS were more successful when started early in the disease course or in early childhood (as appropriate to the LTC). Starting SCS early on in the disease course (Stapleton, 2001; Revert et al., 2018; Boon et al., 2020; D'Souza et al., 2021) negated having to change established behaviours and reverse poor metabolic control. Significant

challenges were encountered where there was a wide variation in how long study participants had been diagnosed with their LTC or had already been performing dietary self-care. For example, between one and 17 years since diagnosis for participants with T1DM (Coates et al., 2013), between one and 13 years since diagnosis for participants with coeliac disease (Meyer and Naveh 2021), between one and 11.7 years since starting a gluten-free diet (Connan et al., 2019), and between 6 months and 9 years already carbohydrate counting (Spiegel et al., 2012). In the study by Austin et al. (2011 and 2013), adolescents who completed questionnaires in both years had a more recent diagnosis of T1DM than those who dropped out. In the study by Christie et al. (2016), participants with the highest HbA1c (poorer metabolic control) were less likely to attend group education sessions; in addition, significantly more children (8-12 years) attended, compared with teenagers (13-16 years) (n= 62, 64% vs n= 42, 44% respectively). This finding was consistent across several studies, where children in the younger age groups were the more receptive and keener to learn (Bell, 2004; Culhane, 2013; Owen et al., 2013; Fishman et al., 2018).

Keeping it going

Across the included studies, models of SCS were more successful when the intervention or exposure to SCS was of longer duration, as a minimum between six and 12 months (Owen et al., 2013; Cooper et al., 2018; Fiallo-Scharer et al., 2019; Boon et al., 2020), to over three years (Revert et al., 2018). Provision of ongoing input, with regular reiteration of topics to reinforce knowledge and skills (Owen et al., 2013), correct misconceptions, misinformation or fill gaps in information deficits (Culhane, 2013), together with further top-up education, enabled tailoring of SCS to meet specific and changing needs of children over time. This was not possible where SCS was delivered over a short duration, for example, as an intensive five-day block (Singh et al., 2000; Nabors et al., 2014; Price et al., 2016) or as a very brief intervention, for example, two six-hour group sessions (Cottrell et al., 1996) or viewing a CD-ROM for approximately 30 minutes (Davis et al., 2004). Interventions of such short duration produced at best, short-term gains, but the effects were not sustained. For example, adolescents improved metabolic control (as reduced dietary phenylalanine intake) progressively returned to baseline levels over one-year (Singh et al., 2000).

Whilst provision of regular ongoing SCS may enable behaviours leading to the formation of self-care habits and routines, this was poorly addressed in the included studies.

Being flexible

Models of SCS across the included studies employed various formats and modes of delivery, in a range of settings, to accommodate differing needs and preferences. Study participants engaged well when SCS utilised a range of interactive (rather than passive) and practical learning opportunities (Stapleton, 2001; Bell, 2004; Spiegel et al., 2012; Owen et al., 2013; Price et al., 2016; Connan et al., 2019; Boon et al., 2020; Meyer and Naveh, 2021). For example, adolescents with T1DM practised carbohydrate counting in practical cookery sessions (Price et al., 2016), children with CF learnt the fat content of foods through doing hands-on labelling activities and matching pair games (Owen et al., 2013) and adolescents with coeliac disease planned actions needed for eating out in simulation activities (Meyer and Naveh, 2021).

With regards to settings, SCS interventions that were integrated into routine care (Bell, 2004; Culhane, 2013; Revert et al., 2018; Fiallo-Scharer et al., 2019), either in a group-based format or on an individual basis were challenging, because of time constraints in busy clinics. However, further significant challenges were encountered when SCS was delivered as an optional extra. For example, in groups in the clinic setting but independent to regular outpatient clinic (Coates et al., 2013; Christie et al., 2016), HCPs were trying to organise and deliver sessions in addition to their usual workload. This was often following little or no training and with a lack of practice time or ongoing supervision. Children and families also had competing demands, for example, school and work commitments. Staff also reported that the pressure on hospital clinic facilities was too great (Christie et al., 2016). In a 10-week home-based programme, though carers enjoyed helping their child learn and learning themselves, some carers reported being too busy to easily fit in daily recording and weekly paper-based exercises with their child (Stapleton, 2001). In addition, given the choice of completing the ADNAT app at home or in clinic, the majority of adolescents chose clinic (Cooper et al., 2018), though this relied on having access to wifi in clinics and for individual sessions with a dietitian, incorporating

these as part of outpatient clinic visits rather than separate home visits was preferred by children and the dietitian (Owen et al., 2013).

In summary, across the included studies, it was evident that integrating SCS into routine care required organisational commitment, with prioritisation and active support of HCPs at a service level (Christie et al., 2016; Cooper et al., 2018; Revert et al., 2018).

Choosing appropriate outcomes

Evaluating success of the models of SCS relied on the choice of appropriate outcomes, and measures of these outcomes. Many of the included studies adopted outcomes and measures commonly used in clinical practice. For example, HbA1c as a measure of the outcome glycaemic control (Spiegel et al., 2012; Coates et al., 2013; Froisland and Arsand, 2015; Christie et al., 2015; Price et al., 2016; Cooper et al., 2018; Fiallo-Scharer et al., 2019; D'Souza et al., 2021) and weight or BMI z-score as a measure of the outcome nutritional status (Cottrell et al., 1996; Stark et al., 2009; Coates et al., 2013; Owen et al., 2013; Revert et al., 2018; Boon et al., 2020). However, whilst such measures are valid, these outcomes may not be sensitive enough to demonstrate clinically meaningful change or sustained behaviour change over the short duration of SCS interventions observed in the majority of included studies. Other outcomes were sensitive in demonstrating the effects of intervention activities. For example, condition-related knowledge and/or skills (Cottrell et al., 1996; Singh et al., 2000; Stapleton, 2001; Bell, 2004; Davis et al., 2004; Culhane, 2013; Owen et al., 2013; Froisland and Arsand, 2015; Connan et al., 2019; Boon et al., 2020), however, validated measures of these outcomes are needed. Perhaps surprisingly (for studies related to SCS of diet and the gut), few included patient-reported outcomes, such as control of symptoms or quality of life, the exceptions being Cottrell et al., 1996; Christie et al., 2016; Price et al., 2016; Fiallo-Scharer et al., 2019; D'Souza et al., 2021; with all using valid measures for the outcome quality of life, and one study using valid measures for both control of symptoms and quality of life (Boon et al., 2020).

The limited choice of outcomes in the included studies may, in part, reflect how the majority of SCS interventions lacked a theoretical basis to their development. Only four of the 21 intervention studies reported using an

underlying theory or model of behaviour change (Singh et al., 2000; Stapleton, 2001; Price et al., 2016; Cooper et al., 2018). However, more encouragingly, nine of the 21 intervention studies (Stapleton, 2001; Coates et al., 2013; Christie et al., 2016; Price et al., 2016; Cooper et al., 2018; Connan et al., 2019; Fiallo-Scharer et al., 2019; Boon et al., 2020; D'Souza et al., 2021) reported involvement of patients and families in their development, to increase their relevance and acceptability.

3.4 Discussion of phase one

The aim of this IR was to identify, critically appraise and synthesise evidence from primary studies on SCS of diet and the gut in school-age children with LTCs. Synthesis of the 27 eligible studies identified six themes that collectively contribute new understanding of what SCS of diet and the gut consists of, together with key requisites for models of SCS in the context of delivery and uptake in routine care.

SCS of diet and the gut throughout the school-age years was found to be complex and dynamic, yet on a continuum as the child grows. It included supporting stepwise development and application of a specific knowledge and skill set. This may be facilitated by the use of age/ developmental stage competency checklists (Bell, 2004; Culhane, 2013; Fishman et al., 2018), such as those currently used in UK practice for children with T1DM aged 6-18 years (Thornton et al., 2016) and in the USA for children and young people with CF aged 10-25 years (CF Foundation, 2021). In a survey of CF HCPs, mean age estimates for when children with CF might master specific self-care tasks related to diet and enzymes, ranged from 6-12 years in 50% of HCPs (Patton et al., 2005). Whilst an update of this study that includes current self-care behaviours would be helpful, it suggests that an age/ stage checklist for diet and the gut in CF could mirror T1DM in beginning at 6 years of age. Emphasis on numeracy training as a core skill is required, with a means of assessment and provision of tailored support as appropriate (Moosa and Segal, 2011; Mulvaney et al., 2013), though use of emerging technology such as continuous glucose monitoring and insulin pumps will in part reduce this need in T1DM.

Visual resources may also be required for children and adolescents to begin making sense of complex relationships between food components, their

absorption and effect on the body, as these are abstract concepts for school-aged children to understand. The SEREN magnetic storyboard is an example of such a resource (D'Souza et al., 2021), and although currently used in practice with children to demonstrate the differences in the body with and without T1DM, this interactive storyboard with the GI tract and pancreas could also be useful in CF. As images can be interpreted in many ways, involving children in the design of visual resources will be essential to ensure images are meaningful to them and evoke positive emotional responses (Houts et al., 2006). Further studies are also needed combining use of visual resources with hands-on practical experience, as this may have more impact than visualisation alone. For example, Evans et al. (2009) suggest engagement with a low-protein recipe book and accompanying DVD of children with Phenylketonuria preparing the recipes, may have been greater than approximately half of the targeted audience (n=105), if combined with hands-on low-protein cooking workshops.

SCS of diet and the gut also included providing practical help with incorporating the demands of self-care into everyday life. This encompassed attention to the fine detail, to enable both proactive and responsive tailoring of support. Further work is needed to identify how this collaborative approach can be implemented in routine care (Boon et al., 2020). For example, regarding the change in practice behaviours and attitudes needed by HCPs to provide this broad range of support, and the training and support this necessitates (Kennedy et al., 2014). In a study by Sullivan-Bolyai et al. (2014), parents and teenagers with T1DM wanted HCPs to be less focussed on numbers (blood glucose levels) and pay attention to the teenager as a whole. This corroborates the earlier findings of Kyngas et al. (1998) whilst also highlighting how having choices, approaching problem-solving jointly, and discussing factors such as emotions and frustrations affects motivation to perform daily self-care. The included studies did not address sustaining daily self-care through development of routines and habits. However, when treatment burden is high, routine is key to motivation (Calthorpe et al., 2020). More research is needed on supporting cues for habit formation throughout childhood and whether self-care behaviours established during childhood can be maintained despite challenges (such as lack of time, competing demands) during adolescence and through to adulthood (Lally and Gardner, 2013; Hoo et al., 2019).

In the included studies, models of SCS were more successful when started early on in the disease course, and were ongoing, to allow tailoring to changing needs and priorities over time. More studies are required examining the effect of SCS delivered soon after diagnosis, as behaviours are being established. In addition, as food and eating are an integral part of everyday life and are often a focal point for social interactions (Singh et al., 2000), this means targeting the learning needs and preferences of young children, which could be explored through further qualitative studies. For models of SCS to be ongoing and remain relevant, there is the further challenge of continual adaptation and adjustment, for example, with advances in treatments, such as the next generation of HEMT in CF and emerging technologies such as insulin pumps in T1DM. Perhaps this is only possible if SCS is embedded within routine care and can evolve as part of it - as in the included studies - models of SCS were more successful when integrated into routine care, rather than being an optional extra. To offset the additional burden of delivering SCS as part of routine care, a wraparound package for SCS is needed, with flexibility in terms of what SCS resources/ activities can be accessed when and how (such as interactive e-learning, mobile apps (Day, 2020), hands-on activities, learning through fun play sessions (La Banca et al., 2020)), to meet individual needs at any one time. Engagement with any particular resource may also be short-lived, for example, Boon et al. (2020) found that regular app usage declined over six-months as knowledge of PERT doses with stable diets was gained and applied.

The complexity of SCS was further compounded by the need for a whole system approach, where there is strong leadership and organisational support for HCPs to implement SCS in routine care, as reported previously (Taylor et al., 2014). This also requires buy-in and prioritisation of SCS within multidisciplinary care teams. To enable this, further studies are needed to identify how system constraints such as limited consultation times and workload pressures can be adjusted (Eaton et al., 2015).

The choice of outcomes in the included studies reflected more of a focus on self-management than self-care, with most being medium- to long-term, and with outcome measures routinely used in clinical practice. Whilst NICE (2015) recommends HbA1c as a measure of glycaemic control, there are difficulties achieving HbA1c targets in practice, so it is perhaps not surprising that positive

and sustained change in HbA1c in research is seldom achieved. Higher mean HbA1c levels in children and young people continue to be associated with being female, longer duration of diagnosis, living in a more deprived area, and Black, mixed or Asian ethnicity (Royal College of Paediatrics and Child Health, 2021). HbA1c and weight/ BMI z-score will also be affected by factors such as illness, puberty, change in activity levels and stress, but as discussed in section 3.3.2, these (confounding) factors were often unaccounted for in the included studies. Six of the included studies chose the patient-reported outcome (PRO) quality of life, with measurement using the generic and condition-specific modules of the validated Pediatric Quality of Life Inventory (Varni, 2022). Whilst use of PROs and patient-reported outcome measures (PROMs) in children with LTCs is encouraged, more work is needed to understand which PROs are appropriate, how to select and interpret the results of validated PROMs and which PROMs detect meaningful change over time (Ronen, 2016).

To facilitate development of the evidence base, studies need to state the rationale for selection of outcomes, including PROs that children and their families have reported as relevant to them (Kirk et al., 2013; Ye et al., 2017; De Wit et al., 2020) as well as other relevant stakeholders, such as HCPs (Boger et al., 2015). This is so that intervention content is specified in terms of how the desired outcomes may be achieved and measured (Coster, 2013) and understanding increased regarding what content/ mechanisms of action lead to success (or not) of interventions in the context in which they are implemented. This emphasises the importance of intervention studies describing an underlying theory (O’Cathain et al., 2019a), yet this was only reported in four of the 21 intervention studies included in the IR. Similar findings have been reported previously, for example, in a review by Pals et al. (2020), there was limited description and application of theories across interventions targeting children aged 7-13 years with T1DM.

Further work is needed to develop theory-based models of SCS that accurately reflect the broad range of activities involved in self-care, that incorporate health promotion, identify outcomes that capture that breadth (Feillet et al., 2010) and measure shorter, as well as medium and longer-term effects of SCS.

Strengths and limitations of the IR

In this review, a range of information sources were used to identify primary research of all study designs, in both published and grey literature. Whilst this allowed exploration of multiple aspects of SCS of diet and the gut, analysis and synthesis of data from such a diverse range of studies is complex and can introduce bias and inaccuracy (Whittemore and Knafl, 2005). To combat this, a systematic and rigorous approach was adopted, with the researcher leading, and independent review by supervisors at each stage.

The methodological quality of some of the included studies (as presented in section 3.3.2) is a limitation. Use of the MMAT (Hong et al., 2018) may also be a limitation, as although it was convenient to use a single tool for quality appraisal, it was less detailed than the CASP checklists (CASP, 2018) or alternatives such as the Cochrane tool for assessing risk of bias in RCTs (Higgins et al., 2011). There were also many 'can't tell' responses, though this was due to insufficient reporting of information by authors of the primary studies. The inclusion of only English language studies, is a limitation as some relevant non-English studies may have been omitted. Furthermore, several included studies presented only limited descriptions of the interventions or study context and the findings in turn represent interpretation by the researcher of what was reported.

Some of the included studies were conducted some time ago, but where possible, their findings have been supported by more contemporary studies. Two of the CF studies were conducted prior to segregation to minimise the risk of cross-infection, however children with CF can no longer come together in-person for workshops (Stapleton, 2001) or groups (Stark et al., 2009). Two of the CF studies also focused on promoting intake of high calorie, high-fat foods (Bell, 2004; Stark et al., 2009), however as outlined in section 1.9, an individualised approach is now more appropriate.

The protocol for the review stated that analysis of a subgroup: studies involving primary school-age children (4-11 years), would be undertaken if possible. However, as only two studies (Stapleton, 2001; Bell, 2004) exclusively involved children in this age range, the researcher was unable to conduct the analysis as part of this current review.

3.5 Chapter summary

This chapter has presented phase one of the study, in which existing evidence for SCS of diet and the gut in children with LTCs has been identified, critically appraised and synthesised to address the two review questions. Findings have been presented as six themes and discussed in the context of the wider literature.

No existing theory or model for SCS of diet and the gut in children was identified for possible adaptation for CF. The review findings have therefore collectively informed early development of a theoretical framework for SCS of diet and the gut in children with CF, to take forward for further development in phase two (chapters five and six). Prior to this, in the following chapter, further understanding of the findings was sought through the process of mapping to a behaviour change framework.

Chapter 4

Mapping findings of Phase One

4.1 Introduction

Having chosen the BCW over other behaviour change theory and described the rationale (Chapter 2, Methodology), this chapter presents the first part of phase three in which the findings of the IR were mapped to the COM-B model and TDF to understand which theoretical components were targeted in SCS of diet and the gut. The chapter then presents and discusses the findings, which inform phase two (which follows, in Chapter 5) and the second part of phase three (Chapter 7).

4.2 Methods

As described in section 3.3.1, the majority of studies included in the IR (n=21) provided models of SCS, whilst other studies (n=6) informed the context of SCS for school-age children with LTCs.

First, to understand more about intervention content, data were extracted from the reports of the 21 intervention studies and mapped onto the COM-B components/ TDF domains that they targeted. For example, in the study by Boon et al. (2020), the intervention (a mobile app for children with CF) included:

- Food recording to develop the **skill** of matching the fat content with enzyme dose (using the enzyme dose calculation support)
- Educational games to increase **knowledge** about nutrition
- A diary for recording GI symptoms (develop skill of self-monitoring = **behavioural regulation**)
- Receiving messages from HCPs (opportunity for tailored feedback = **social influence**)

with reported benefits including increased confidence and self-efficacy (= **beliefs about capabilities**) (Floch et al., 2020).

Therefore, each of these components were assigned to the corresponding TDF domains (highlighted in bold).

Second, to understand the contextual factors influencing delivery and uptake of SCS, enablers and barriers were extracted from all 27 included studies, and mapped onto the appropriate COM-B components/ TDF domains. For example, Cooper et al. (2018) in evaluating the feasibility of integrating a self-care app into adolescents T1DM care, reported:

- Barriers (mapped to the opportunity – physical component)
 - access to Wi-Fi in clinics
 - access to technology support and iPads
 - time taken for adolescents to complete and HCPs to review the needs assessment tool
- Enablers (opportunity – social)
 - parental support
 - tailored education and support from HCPs
- Enablers (motivation – reflective component; professional role and identity TDF domain)
 - lead clinician support and buy in from the MDT.

4.3 Findings

This section details what the extracted data looked like and how it was interpreted.

The findings from characterising the intervention content across the 21 studies are presented in Table 4.1. All of the studies addressed capability to perform self-care/ self-management behaviours, through developing knowledge (n=21) and skills (n=19), such as interpreting food labels and carbohydrate counting. Within the remainder of the capability component, only two studies targeted the TDF domain of interpersonal skills (Christie et al., 2016 and Cottrell et al., 1996, for communicating with HCPs), and two studies the memory, attention and decision making processes domain (Meyer and Naveh, 2021, for decision making skills and Bell, 2004, for remembering to take medications). However, intervention content of 11 studies targeted the behavioural regulation TDF domain, by addressing self-monitoring, problem solving and action planning.

There was far less consistency across studies in addressing opportunity for self-care/ self-management behaviours to occur and motivation to do the behaviours, though seven of the nine TDF domains within the opportunity and

motivation components were represented. Furthermore, SCS interventions with content that targeted multiple TDF domains across all three COM-B components (capability, opportunity and motivation) were descriptively the more effective/ successful (Boon et al., 2020; Cooper et al., 2018; Fiallo-Scharer et al., 2019; Frøisland and Årsand, 2015; Stapleton, 2001).

Success was further qualified by examination of context-specific enablers for and barriers to delivery and uptake of SCS, presented in Table 4.2.

Table 4.1: COM-B components and TDF domains targeted by the SCS interventions

Study	COM-B components and TDF domains													
	CAPABILITY					OPPORTUNITY		MOTIVATION						
	Phys.		Psych.			Phys.	Soc.	Reflective					Automatic	
	S	K	CIS	MAD	BR	EN	SI	P/S ID	B Cap	O	B Con	G/I	R	E
Bell (2004)	✓	✓		✓	✓									
Boon et al. (2020)	✓	✓			✓		✓		✓				✓	
Christie et al. (2016)	✓	✓	✓						✓		✓	✓		
Coates et al. (2013)	✓	✓												
Connan et al. (2019)		✓												
Cooper et al. (2018)	✓	✓					✓		✓			✓		
Cottrell et al (1996)	✓	✓	✓		✓									
Culhane (2013)	✓	✓			✓								✓	
Davis et al. (2004)	✓	✓												
D'Souza et al. (2021)	✓	✓										✓		
Fiallo-Scharer et al. (2019)	✓	✓			✓		✓		✓		✓			
Frøisland and Årsand (2015)	✓	✓			✓		✓		✓		✓			
Meyer and Naveh (2021)	✓	✓		✓	✓									
Nabors et al. (2014)	✓	✓			✓							✓		✓
Owen et al. (2013)	✓	✓											✓	
Price et al. (2016)	✓	✓			✓									
Revert et al. (2018) *		✓										✓	✓	
Singh et al. (2000)	✓	✓				✓					✓			

Study	COM-B components and TDF domains													
	CAPABILITY					OPPORTUNITY		MOTIVATION						
	Phys.		Psych.			Phys.	Soc.	Reflective					Automatic	
	S	K	CIS	MAD	BR	EN	SI	P/S ID	B Cap	O	B Con	G/I	R	E
Spiegel et al. (2012)	✓	✓					✓							
Stapleton (2001)	✓	✓			✓	✓			✓			✓	✓	
Stark et al. (2009)	✓	✓			✓							✓		

Key for the COM-B components: Phys.: physical; Psych.: psychological; Soc.: social.

Key for the 14 TDF domains: S: skills; K: knowledge; CIS: cognitive and interpersonal skills; MAD: memory, attention and decision making processes; BR: behavioural regulation; EN: environmental context and resources; SI: social influences; P/S ID: professional/ social role and identity; B Cap: beliefs about capabilities; O: optimism; B Con: beliefs about consequences; G/I: goals/intentions; R: reinforcement; E: emotion.

*Development of 'skills' was reported, but no detail provided regarding what skills

Table 4.2: Enablers for and barriers to delivery and uptake of SCS mapped to COM-B components and TDF domains

Study	COM-B components and TDF domains													
	CAPABILITY					OPPORTUNITY		MOTIVATION						
	Phys.		Psych.			Phys.	Soc.	Reflective					Automatic	
	S	K	CIS	MAD	BR	EN	SI	P/S ID	B Cap	O	B Con	G/I	R	E
Austin et al. (2011 & 2013)							E	E						
Bell (2004)						B + E								
Boon et al. (2020)		E				B	E		E					
Christie et al. (2016)	B	E				B	B	E						
Coates et al. (2013)	B + E						B							
Connan et al. (2019)														
Cooper et al. (2018)						B	E	E						
Cottrell et al (1996)						B								
Culhane (2013)						B								
Davis et al. (2004)														
D'Souza et al. (2021)	E													
Fiallo-Scharer et al. (2019)						E	E							
Fishman et al. (2018)														
Frøisland and Årsand (2015)		E					E		E					
Kyngas et al. (1998)					E	E	E	E				E		
Meyer and Naveh (2021)									E					
Nabors et al. (2014)							E		E					
Owen et al. (2013)						B + E	E							

Study	COM-B components and TDF domains													
	CAPABILITY					OPPORTUNITY		MOTIVATION						
	Phys.		Psych.			Phys.	Soc.	Reflective					Automatic	
	S	K	CIS	MAD	BR	EN	SI	P/S ID	B Cap	O	B Con	G/I	R	E
Price et al. (2016)	E													
Rankin et al. (2018a)					E		B + E	E						
Rankin et al. (2018b)	B					E	B + E	E				E		
Revert et al. (2018)	E					B + E	E	E						
Singh et al. (2000)						B	B + E							
Sparapani et al. (2017)		E									E			E
Spiegel et al. (2012)		E				E	E							
Stapleton (2001)						B + E	E							
Stark et al. (2009)						B	E							
Witalis et al. (2017)						B			E					B

COM-B components and TDF domains as in Table 4.1 above; B: barrier; E: enabler.

The majority of barriers to and enablers for delivery and uptake of SCS corresponded with the opportunity component of COM-B. This is perhaps not surprising given that this component is external to the individual (compared with the capability and motivation components which act at the level of the individual), linking with the TDF domains of environmental context and resources, and social influences. For example, key physical barriers were lack of time to deliver SCS and an increased workload for HCPs (chiefly dietitians). The key physical enabler overcame these barriers by integrating delivery of SCS into routine care. For the social TDF, key barriers were competing demands for families and HCPs, however, the main enablers were tailored support and feedback from HCPs and parental and/or peer support, suggesting a change in approach/ focus of input with the limited time available may be beneficial. Related to these influencing factors, key enablers in the capability and motivation components were increasing knowledge and skills training for HCPs (dietitians) to deliver SCS, practice of skills to build confidence in delivering SCS and buy-in and support from the wider MDT. Each of these influencing factors therefore suggest a requirement for structural change/ investment for SCS to occur.

4.4 Discussion

The aim of this first part of phase three was to gain understanding of which theoretical components were targeted in the studies included in phase one's IR. This aim was achieved by exploring content of the SCS interventions (n=21 studies) and context of delivery and uptake of SCS (n=27 studies).

The findings on intervention content were congruent with the wider literature. For example, regarding capability, building knowledge in particular, but also skills, has been the focus of a broad range of health interventions (Beard et al., 2022). However, less attention has been paid to content important in maintaining self-care behaviours, such as the development of interpersonal skills, memory and decision making, and self-regulatory skills (Stanton-Fay et al., 2021), though the combination of self-monitoring with other self-regulatory skills has been identified as effective content in dietary interventions (Michie et al., 2009).

Variability in addressing opportunity and motivation components has been reported previously (e.g., Johnson et al., 2018), though together with capability, have been shown to be relevant for interventions targeting self-care behaviours (Richardson et al., 2019). As all components of the COM-B model and multiple TDF domains were represented across the combined studies, further exploration is needed in phase two to identify which of the TDF domains are most important to target in SCS of diet and the gut.

Through extraction of enablers and barriers to the delivery and uptake of SCS, further insight of the context was gained, however, it was more relevant to understanding implementation of a SCS intervention rather than the factors influencing self-care/ SCS behaviours. Despite this, the mapping was useful for two reasons. First, it highlighted the importance of exploring influencing factors with stakeholders in phase two, to be able to identify what needs to change for the target SCS behaviours (preferred outcomes of SCS) to occur and therefore inform development of the SCS intervention. Second, it suggested that implementation of the SCS intervention may need an intervention in itself, directed at HCPs (dietitians) and system level behaviours. This concurs with previous studies of implementing interventions (e.g., Haighton et al., 2021), including in children's health (Alexander et al., 2014), self-care (Nelson et al., 2020) and nutrition education (Gianfrancesco and Johnson, 2020).

Strengths and limitations

The depth of the mapping was limited by the detail provided in each report or companion paper(s) where these were available. The process of extracting and assigning data on intervention content and enablers and barriers to COM-B components/ TDF domains required judgement and interpretation by the researcher. This may have introduced bias, however, completion of training in the use of the BCW, and regular discussions with supervisors LM and GM, helped the researcher navigate this process.

4.5 Chapter summary

This chapter has presented the first part of phase three, in which findings of the IR were mapped to COM-B components and TDF domains. Gaining understanding of intervention content suggested the developing SCS intervention will need to be multi-component, targeting multiple TDF domains

across all COM-B components. Exploring implementation of SCS also suggested the developing SCS intervention will need to be multi-level, targeting (at its simplest) children and dietitians. The findings directly inform the following chapter which presents how phase two was conducted.

Chapter 5

Phase Two: Qualitative Descriptive Study - Methods

5.1 Introduction

Having chosen an exploratory qualitative descriptive design to meet the objective for phase two and described the rationale (Chapter 2, Methodology), this chapter outlines how the chosen methods for phase two were applied. It includes description of the study procedures and decisions taken at each stage, with their rationale, together with clear reporting of the adaptations needed and decisions taken in response to Covid-19.

The chapter begins with sampling and recruitment, followed by data collection through semi-structured interviews, analysis using Framework, and concludes with researcher reflexivity. Presentation and discussion of phase two findings then follows in Chapter 6.

5.2 Methods

To explore the perspectives of those involved in the routine dietetic care of children with CF: children, parents/carers and children's CF dietitians, a qualitative descriptive approach was chosen (as discussed in section 2.6.1), with the aim of answering the following questions:

- What could SCS of diet and the gut consist of?
- How could SCS be delivered as part of routine care?
- What outcomes would be most important to achieve?

To ensure the methods were reported in sufficient detail, appropriate items in the COREQ checklist for interviews were addressed (Tong et al., 2007), with the aim of increasing transparency.

5.2.1 Sampling

5.2.1.1 Purposive sampling

Purposive sampling was selected as the most appropriate sampling strategy (section 2.6.3). It facilitated approaching a diverse population of children with CF, in terms of gender, age range of 6-11 years and receiving care at multiple

specialist CF centres, as outlined in section 2.6.3. The eligibility criteria are outlined in Table 5.1.

Table 5.1: Eligibility criteria for phase two study

Participants	Inclusion criteria	Exclusion criteria
Children's CF Dietitians	<ul style="list-style-type: none"> - Dietitian at one of the 24 children's specialist CF centres in England (CF Trust, 2021) - To have had regular contact with children with CF for at least one year 	<ul style="list-style-type: none"> - Dietitian at a children's network/ shared-care CF centre (i.e., not a specialist CF centre) - Less than one years' experience of working with children with CF
Children with CF	<ul style="list-style-type: none"> - Children with CF who are pancreatic insufficient (PI) (and so require PERT) - Primary school-age 6-11 years - Receiving care at one of three children's specialist CF centres: A, B or C, for >one year - Clinically well enough to participate in an interview* 	<ul style="list-style-type: none"> - Children with CF who are pancreatic sufficient (PS) (do not require PERT and therefore have no experience of using it) - Children outside the target age range: <six years and >11 years - Children receiving care at specialist CF centres at different locations in England or for <one year at targeted centres
Parents/ carers of children with CF	<ul style="list-style-type: none"> - Parents/ primary caregivers of children meeting the above inclusion criteria 	<ul style="list-style-type: none"> - Parents/ primary caregivers of children meeting the above exclusion criteria

*This criterion was based on the number of courses of intravenous antibiotics needed over the previous one year as a measure of disease severity

5.2.1.2 Sample size

As data collection and analysis were concurrent, sampling continued until there was data saturation, as discussed in section 2.6.3. It was estimated that interviews with approximately 15 participants from each of the three stakeholder groups may be needed to achieve saturation (Green and Thorogood, 2018). This estimated number was supported by a recent systematic review, in which Hennink and Kaiser (2022) identified that in studies with relatively homogenous study populations, saturation may be achieved with 9-17 interviews. The following tentative sample sizes were therefore planned for:

Children's CF Dietitians - target of recruiting one dietitian at 15-20 of the 24 children's specialist CF centres in England (as listed in the CF Trust 2019 registry report (CF Trust, 2020)).

Children with CF and their parents/carers - target of recruiting 15-20 children and 15-20 parents/ carers i.e., up to seven children and seven parents/ carers at each of the three specialist CF centres.

5.2.2 Recruitment

The recruitment plan was based on feedback from the initial PPI consultation and YPAG (section 2.11) and was discussed and agreed with the recruiting dietitians at each of the three specialist CF centres. In addition, the PAG reviewed the recruitment plan prior to commencing recruitment and assessed progress with recruitment midway through the recruitment period.

Participant-facing documents were developed and reviewed with the input of various PPI, as described in section 2.11. Example documentation is included as Appendix E.

Impact of Covid-19

In relation to review of participant-facing documentation:

- Two in-person meetings of the researcher with YPAG, were replaced with email feedback and an online group discussion
- In-person meetings with children with CF when attending a Children's Specialist CF Centre and a meeting with PAG were all replaced with email feedback.

5.2.2.1 Recruitment procedures

Children's CF Dietitians

The Standards for CF Care (CF Trust, 2011) specify a commitment for all specialist CF centre dietitians to be members of the specialist group of their professional body, the British Dietetic Association (BDA). Following confirmation of study approvals (described in section 2.8.1), children's CF dietitians were invited to participate via a group email from the BDA CF Specialist Group. Interested dietitians made contact via email or telephone using the details included in the dietitian participant information sheet and interviews were arranged at times convenient for them i.e., to fit around their clinical work and not impact on their NHS time. Recruitment took place between 1st September 2020 and 16th April 2021.

Impact of Covid-19

A presentation explaining the study to BDA CF Specialist Group members had been planned for the May 2020 group meeting, however, due to Covid-19, the meeting was cancelled. The next group meeting did not take place until December 2020, at which point, a short presentation by the researcher served as a study reminder. The email invitation was also recirculated several times by the BDA, as many dietitians were temporarily deployed to other areas to assist in managing the surge in hospitalised Covid cases.

Children with CF and their parents/ carers

Following confirmation of study approvals (described in section 2.8.1), the recruiting children's dietitians at each of the three specialist CF centres (who were members of children's direct clinical care teams), accessed children's medical records/ electronic patient records to compile a list of who met the eligibility criteria in Table 5.1. Study packs which included a study invitation and participant information for parents and children (aged 6-8 years or 9-11 years as appropriate) were posted by the researcher to the three specialist CF centres. The recruiting children's dietitians then added children's names and addresses to the pre-stamped study packs and posted these on to all eligible children and their parents.

Interested families could contact the researcher directly to discuss the study, using the contact details provided in the participant information sheets. Families were also approached by the recruiting dietitians at the child's next clinic appointment, to receive a study reminder/ invitation to take part. Again, interested families could contact the researcher directly, or give their dietitian permission to share their contact details with the researcher via secure email (nhs.net), who then contacted them to discuss the study and if they wished to take part, an interview was arranged.

Being telephoned by a researcher, to provide potential participants with an opportunity to discuss and ask further questions regarding the study, had been carried out previously in other studies conducted at each of the three CF centres. As identifying, approaching and inviting potential participants to take part in the study was extra to the recruiting children's dietitians' workload, this process was discussed and jointly agreed with all the dietitians as the most practical option. Recruitment information was provided for dietitians, so that the process across the three CF centres was as standardised as possible (Appendix F). Recruitment took place at each specialist CF centre between: centre A: 23rd September 2020 – 30th April 2021; centre B: 12th October 2020 – 31st March; centre C: 24th January 2021 – 28th May 2021.

Impact of Covid-19

During the recruitment dates, there were interruptions to the postal service due to Covid. This caused delays in study packs reaching the three CF centres for dietitians to add names and addresses, and then further delays in the packs reaching families. For example, some families were only receiving post one or two days per week.

Despite being granted access to the three CF centres, it was not possible for the researcher to visit centres in person due to travel restrictions and/ or additional infection prevention and control procedures. This prevented the researcher being available in clinics to meet potentially interested families and discuss the study as appropriate.

Between November 2020 and January 2021, there was a pause in recruitment at centres A and B as recruiting dietitians were helping manage the surge in

hospitalised Covid cases. However, regular communication was maintained throughout that time and recruitment plans revised accordingly.

5.2.2.2 Consent

Children's CF Dietitians

When an interview slot was confirmed, the researcher emailed a consent form to the dietitian. At the pre-arranged time, the dietitian was telephoned and there was an opportunity to ask questions and discuss the study further, referring to the participant information sheet. If the dietitian wished to proceed, the consent form was discussed, completed, signed and emailed back to the researcher immediately prior to the interview. Immediately after the interview, the researcher signed the consent form and emailed a copy to the dietitian for them to keep together with the participant information sheet.

Children with CF and their parents/ carers

When interview slots had been arranged, families chose to receive the study forms and an activity pack by post or email, and the researcher emailed a link for the video call. At the pre-arranged time, the family joined the video call and there was an opportunity to ask questions and discuss the study further, referring to the participant information sheets. If they wished to proceed, the child's assent and parental consent were discussed and obtained for the child's interview, together with consent for the parent/ carer's interview. The child and parent/ carer were asked to sign two copies of the assent and consent forms. One copy of each was kept by the family along with the participant information sheets and the other copy returned to the researcher using the stamped addressed envelope provided.

Only one child was unsure about proceeding with the interview. As obtaining their assent was not possible, an interview was conducted only with their parent.

5.2.3 Data collection

Data were collected in single semi-structured interviews (rationale described in section 2.6.4).

5.2.3.1 Topic guides

Topic guides with open-ended questions and prompts were used with each stakeholder group. Using a topic guide ensured consistency in data collection by defining the key areas to be explored, yet it allowed flexibility to respond to issues raised as personally salient to each individual participant (Arthur et al., 2014).

Topic guides were initially developed based on the findings of the IR, with feedback from the YPAG. Topics centred around meeting the following objectives:

- To explore their experience of what currently happens in routine CF dietetic care/ clinic consultations
- To explore understanding of and capacity for self-care and SCS of diet and the gut
- To identify needs, preferences and goals for SCS
- To identify what outcomes of SCS matter most (in the short and longer-term)
- To explore perceptions of barriers and facilitators for delivery and uptake of SCS being provided as part of routine care.

The topic guide used with children (Appendix G) was developed by considering the practicalities of interviewing children presented in section 2.6.4. For example, the use of yellow and red cards was a means of giving children a voice, if they felt too shy to say they would like to skip a question or stop the interview. This attempted to reduce the power imbalance between adult interviewer and child interviewees.

Rather than a formal pilot, the topic guides were reviewed after the first two interviews with each stakeholder group. The review was essential for assessing if the data being collected was the data needed to address the phase two study questions. Although the review suggested this goal had been met, minor changes were made. For example, for the dietitian and parent topic guides, the 'warm up' was reworked and topics slightly reordered to improve the flow of the interview.

5.2.3.2 Interview procedures

As advised by members of the PAG, rather than interviews being conducted with stakeholder groups (children, parents/carers and children's CF dietitians) in consecutive blocks (as per the original plan), interviews with participants from all stakeholder groups were mixed throughout the nine-month data collection period. This allowed exploration and development of emerging themes with a mix of subsequent participants. It also maximised the recruitment period for all potential participants with a buffer against e.g., the disruptions caused by Covid, the Christmas holidays, staff absence and staff winter pressures.

Children's CF dietitians

The length of telephone interviews ranged between 38 and 70 minutes (average 52 minutes) and were conducted with dietitians at a time to fit around their clinical commitments. The majority chose to be interviewed when working from home. However, one dietitian was interviewed in a shared space in her hospital environment and one dietitian as she walked home from work.

Interviews with children and parents

Face-to-face interviews via video call with children and parents ranged between 42 and 71 minutes (average 58 minutes).

The option of conducting interviews in-person with the researcher visiting the child's home or in a quiet private room at their CF centre, for example, as part of a planned outpatient clinic visit, was offered but not taken up due to Covid restrictions.

Children

Six of the 20 children chose to be interviewed individually, with their parent present in the background; 14 were interviewed jointly with their parent(s).

An activity pack, posted or emailed to children in advance of the interview, contained a range of activities for children to choose from and complete whilst talking if they wished. The pack included drawing and colouring sheets, wordsearches, mazes and quizzes (all with a food theme), as well as a food sorting card game (examples of activities are included as Appendix H). As discussed in section 2.6.4, these activities were used to help build rapport, maintain engagement and aid discussions e.g., around children's understanding

of PERT, i.e., completed activities were not considered research 'data' in themselves (Harden et al., 2000). Several younger children acted on the suggestion to take a photo or draw a picture to bring to the interview, and through asking children what meaning these had for them, was also helpful in building rapport (Spratling et al., 2012).

Parents

Interviews with individual parents followed on directly from individual interviews with their child (and continued after 10 of the 14 joint interviews). The child would go off to play or complete schoolwork, leaving the parent to speak openly and freely (as had been requested by the parent members on the PAG). However, some parents were also cooking dinner or needing to feed an infant and/ or entertain a toddler during the interview. Parents who had been in the background for their child's interview, were able to add further detail to some of their child's answers and provide more description of the context.

Where children and parents chose to be interviewed together, onus was placed on the child answering questions first, yet parents provided useful 'scaffolding' as described in section 2.6.4.

All interviews were recorded using an encrypted audio-recording device. Field notes were taken during interviews e.g., key words for important aspects and as reminders to return to for further exploration, and immediately post interviews to capture contextual information and reflections that may assist analysis (Phillippi and Lauderdale, 2018).

5.2.4 Data analysis

Data was analysed using Framework (rationale described in section 2.6.5). Analysis was an iterative process, undertaken partly during data collection to inform future interviews, and fully, after data collection was completed. NVivo12 qualitative analysis software was used to assist with storing, organising, retrieving and managing the data (QSR, 2018).

The following stages of Framework (Spencer et al., 2014) were followed, though the process was not linear:

- Familiarisation

- Construction of an initial thematic framework
- Indexing and sorting
- Charting
- Interpretation

5.2.4.1 Familiarisation

Audio-recorded interviews were transcribed verbatim by a University of Leeds approved transcription service. Each transcript was read and re-read whilst listening to the corresponding audio-recording. Changes were made to the transcripts as appropriate to ensure they were accurate, e.g., words that had been missed or were incorrect, were filled in or amended. Field notes made during and immediately after each interview were reviewed and added to the transcripts and all personal identifiers removed to allow future sharing of transcripts. Preliminary ideas and impressions of the data were noted (Gale et al., 2013). This immersion in the data enabled familiarity with the content of whole interviews, and was important in informing exploration of preliminary ideas and emerging issues (all feeding into the initial framework) as further interviews were conducted.

5.2.4.2 Construction of an initial framework

Next, a framework: a set of codes (labels) organised into categories, was needed to allow reorganisation of the dataset to support answering the study questions (Gale et al., 2013). To identify an initial framework, topics from the interview topic guide, together with the noted preliminary ideas and emerging issues were organised under the headings of the phase two study questions. It was then applied to an arbitrary selection of six full transcripts (two from each stakeholder group, one early and one later on in the data collection), following their line by line open coding. The process of open coding is illustrated in Table 5.2 which contains an excerpt of one of the selected transcripts.

Table 5.2: Table illustrating the process of open coding of transcripts

Codes		Excerpt of transcript	Notes and ideas
Child-centred	224	Yes, so personally I really feel that talking to the child	? Of similar importance to other dietitians
	225	themselves and not the parent is really important and I	
	226	also think just doing it so that they are engaging with an	What's engaging at different ages/ stages?
	227	activity that's age-appropriate works better. Yes, so I did	
	228	the teaching of the body so I often do, not often but	Refers+ to education and teaching as part of role
	229	occasionally do, to kids of about eight, nine, ten (years),	Fits target age range
Specific content	230	how enzymes work in the body and just sort of travel	Overlap of 'what' and 'how'
	231	down through the GI tract and kind of that's something	
	232	that tends to stay with them because it's visual. I think	Visual = more memorable
	233	things that are visual at that age seem to work. I do feel	
	234	it's, yes, it's something that then has to be followed up	Revisit and build on
	235	because I think sometimes and again many of our	
	236	families have quite chaotic lives or very busy lives and	Competing priorities
Part of dietitian role	237	the reliance is on us dietitians to empower those kids	Uses 'empower' again
	238	and it isn't always the parents that can do it. Some	
	239	parents do rely on us to do that as well. So I think	? Expectation of dietitian role by parents
Barrier - delivery	240	the constraints are the time often, they are, yes.	Not having time within routine clinic

Numbers are line numbers as they appeared in the transcript; highlighted text was text considered useful in answering the study questions. In the left-hand margin, the transcript content was described with a code. In the right-hand margin, more detailed notes and ideas were recorded e.g., questions to consider.

Codes that were related/ linked were then grouped together into categories and descriptions added to clarify what each code consisted of. The example below (Table 5.3) shows a category in the initial framework.

Table 5.3: Example of the category ‘features of SCS’ in the initial framework

(later relabelled as ‘characteristics of SCS of diet and the gut’)

CODE	DESCRIPTION
Features of SCS	
Child-centred	<i>Directly engages the child/ encourages child involvement, responsive to child’s questions</i>
Parent-friendly	<i>Aids parents in supporting child, a refresher for parents</i>
Guides/prompts dietitians	<i>Provides a structure – what’s appropriate for child to learn when and how, links to appropriate activities and resources</i>
Flexible to tailor to individual	<i>Appropriate to age and developmental stage, match to priorities/needs and learning style preferences of the child/ family at a particular time, can dip in and out</i>
Ongoing stepped approach	<i>Regular revisiting of topics - assessing understanding and topping up, reinforcement</i>
School-friendly	<i>Aligns with school curriculum, increases awareness of staff (and peers) of child’s needs whilst at school</i>

As identifying a framework was subjective, several excerpts of the open coding and the initial framework were shared, first with supervisors LM and GM, and second, in a meeting of the PAG to discuss whether group members had different interpretations of the text or the codes assigned. Gaining their perspectives was helpful in checking out assumptions and decisions made about the most relevant and important issues. The initial framework was then used to code more transcripts and the framework repeatedly refined as more data was added.

5.2.4.3 Indexing and sorting

All anonymised transcripts were formatted, imported into NVivo12 and the refined framework systematically applied to each transcript. This was a lengthy process as it involved sifting through each transcript and selecting and applying

appropriate codes to interesting segments of text. The process led to further refinement of the framework as codes were added to for greater clarity, some codes collapsed and merged and overlapping codes highlighted to capture connections and complexities. Supervisors LM and GM independently coded three anonymised transcripts using the refined framework and following discussion, there was consensus that the framework captured all of the data and that the study questions were being fully addressed.

5.2.4.4 Charting

When all the data had been coded using the framework, it was auto-summarised in an NVivo generated matrix (chart) for each category, with one row per participant and one column per code. Data were then exported from NVivo12 to Microsoft Excel and manually summarised in each cell of the matrix i.e., it was reduced further whilst retaining its original meaning (Gale et al., 2013). Illustrative quotes were also highlighted. Use of NVivo12 enabled summaries to be easily cross-checked against the original transcripts and following some further merging of codes, a final thematic framework was generated (presented in section 6.2.2). The example below (Table 5.3) is an excerpt of a chart entry for the category 'SCS of diet and the gut – what it is - characteristics'.

Table 5.4: Excerpt of charting for the category: characteristics of SCS of diet and the gut

	Child-centred	Parent-friendly	Guides/ prompts dietitians	School-friendly	Flexible to tailor to individual	Ongoing stepped approach	Other
INT-P03	Direct involvement of child in conversations may lead to them taking a little more responsibility	Sets out expectations of what will happen when		Increase awareness and understanding of staff and peers of child's needs with respect to eating and PERT	Range of resources/ activities needed to suit learning style preferences of the child/ family	A grounding/ foundation from which to progress	Target age range. P03: I think that if they start sort of at seven (years), taking a little bit more on, it's so gradual, by the time they get to 11 (years), that you haven't noticed that big, oh gosh, I haven't done anything, and now he doesn't know what he's doing and it's really panicked him
INT-D11		Enables parents to learn themselves/ serves as a refresher	Provides a structure for delivering consistent and comprehensive support	Complimentary resource for schools. D11: In CF, food and Creon is probably the only aspect that the schools will need to be engaged with really. Occasionally they need to do some physiotherapy in school, but for all of them it will be about food and Creon won't it?	Appropriate to age. SCS intervention needs to be flexible enough to meet needs of an increasingly diverse CF population.	Assessing understanding is important. D11: We don't have a formalised structure for it (SCS) as such, but it's a drip, drip effect that we do throughout our time working with the children and families	

Key: Summaries in black, quotations in grey, interpretations in green; INT-P03: interview with parent no.3; INT-D11: interview with dietitian no.11.

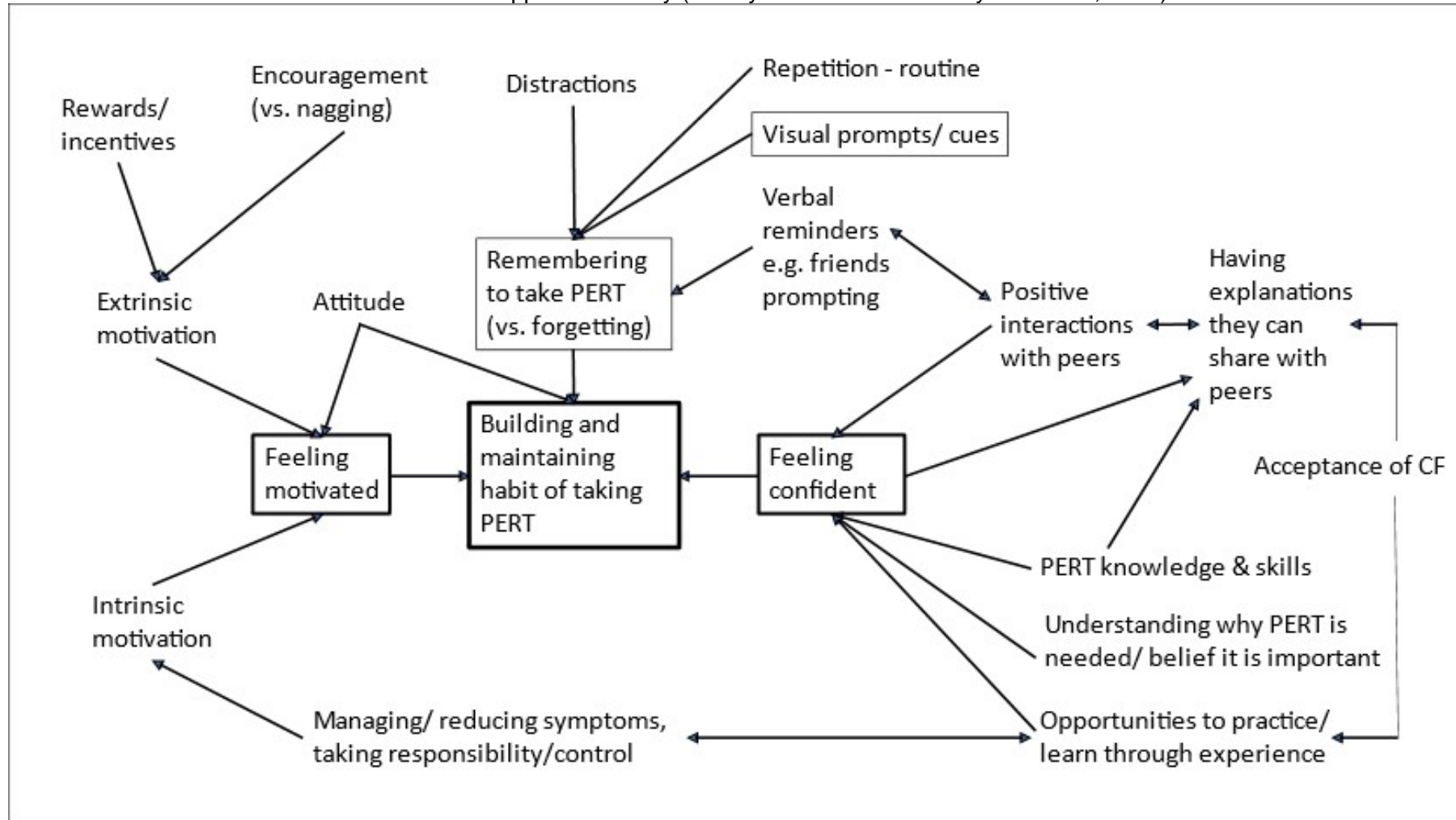
5.2.4.5 Interpretation

Summarised data in each matrix (one matrix for each of the five categories) was reviewed by searching for patterns of similarity and difference within and between participants for each of the codes. Themes were developed to bring together key characteristics of the data as a whole, across categories, and go beyond description, to consider underlying explanations within the data. This process was helped first by mapping on paper to visualise the different connections between codes/ categories, and second through discussions of ideas with supervisors LM and GM, clinical supervisor AD, and members of the PAG. The 'messiness' of this final stage is difficult to illustrate, however the figure below (Figure 5-1), an excerpt of a map exploring potential linkages, demonstrates attempts to fit multiple layers of the data together.

Themes are presented as a narrative in section 6.2.3, whilst the conclusion to this chapter, researcher reflexivity, now follows.

Figure 5-1: Excerpt of a map exploring the interplay of factors in building the habit of taking PERT

Wherein the role of motivation and confidence appear to be key (initially based on a model by Yeh et al., 2019)



5.2.5 Researcher reflexivity

In this section, selected notes from my reflexive journal are shared to offer additional insight of the study processes and procedures.

5.2.5.1 Reflections on Recruitment

There were a number of challenges with the recruitment of participants. Email invitations needed to be recirculated several times as children's CF dietitians were covering adults/ other clinical areas due to Covid and not able to regularly check email. Dietitians who participated reported being grateful for the reminder, but I was acutely aware that everyone was working at maximum and that inviting study participation during a pandemic could be overburdening them. Several dietitians kindly emailed to say they would like to participate but were unable due to lack of time.

Due to travel restrictions, I was unable to visit the CF centres and be available in clinics to meet potentially interested families in-person, and the rest of the MDT, to discuss the study. It is difficult to gauge what impact this had, but if I had met families beforehand, perhaps the sample would have included some six-year-olds; meeting the MDT may have also increased sign-posting to the study.

I was reliant on the recruiting dietitians at the three CF centres, yet as they were reviewing patients by telephone and online rather than in clinic as was pre-Covid practice, this made it more difficult for them to discuss the study with potential participants. I was aware that regular communication and my providing ongoing support would be essential, but this played an even greater role in maintaining study momentum throughout the pandemic.

The delay in opening specialist CF centre C to recruitment was stressful and the addition of another fourth centre was considered. However, staggering the recruitment dates at centres actually prevented me from being overwhelmed. It was a considerable time commitment in following up potentially interested families and the majority of interviews were rescheduled at least once due to families having other commitments that needed to take priority. The effort to recruit participants was exacerbated because of the restrictions of Covid but the

disruption to usual attendance of clubs and sport after school and parents working from home may have aided recruitment. Having a dedicated study telephone to send text reminders was useful as this felt less intrusive.

It is difficult to assess what impact conducting the interviews via video call may have had on families' response rate. Recruiting dietitians did not feedback that digital exclusion was a barrier to recruitment. Participants joined the video call on a mix of personal computers, tablets and smartphones, just as they were doing for clinic reviews, school and work calls, and whilst at times there were some connection difficulties, there didn't appear to be any training or support needed with access.

Only one child did not assent to be interviewed, but on interviewing their parent, it became clear that the child did not know he had CF. I was therefore grateful that a potentially challenging situation had been averted.

5.2.5.2 Reflections on data collection

There were many points for reflection throughout data collection. Pausing to review the topic guides was useful for reflecting on what had gone well and not so well e.g., how I was phrasing questions, what I had missed following up on, consideration of the tone and pace. It also revealed the importance of not scheduling interviews when I would be tired or hungry.

Due to interruptions to the postal service, study forms and activity packs did not arrive with some families prior to interviews, causing interviews to be rescheduled, or at all, and I did not receive some completed returned study forms. Receipt of study forms and activity packs by families via email worked well, except where they were unable to print out activities. There were occasions where the parent had kept back the child participant information sheet, so the child was not aware of the study. The process of assent was therefore even more important. In contrast, some families had already completed study forms and posted them back to me prior to the interviews; the assent and consent processes were then revisited prior to commencing interviews.

Not having the opportunity to meet children prior to interviews necessitated being more creative in establishing rapport and maintaining engagement. When interviewing children, I drew on my ten years of experience in working with

children with CF as a clinician e.g., in adapting questions to be developmentally appropriate for each child, demonstrating genuine interest and encouraging children to talk about what was important to them. I also drew on training completed in conducting research with children and depth interviewing. Nevertheless, conducting the interviews was exhausting. High concentration was needed for continuous active listening, vigilance for cues and keeping track of points raised.

Use of the activity packs worked well for children under 11 years¹, with children and parents requesting they be included in the SCS intervention. Children particularly liked the empty plate activity (Appendix H) because it was drawn by an adult with CF who had kindly agreed for it to be used in the study. For each of the puzzles, there were four options that were increasingly more difficult and they liked choosing which one was right for them. Many of the children kept the yellow and red cards close to them. It would have been useful to know if children felt these gave them a voice and whether these did contribute to reducing the power imbalance.

At the beginning of interviews, assurance was given that there were no right or wrong answers. This proved to be important information to relay as it was repeated back/ spoken of many times during the interviews. Some interviews were emotionally challenging as parents when interviewed on their own described difficulties e.g., with feeling isolated. I felt less equipped to deal with these situations in my role as researcher (rather than my more usual role as clinician). When interviewing siblings from different age groups (C12A and B), I thought they may have had different perspectives to share compared with individual children, but this was unfounded, perhaps as many of the other children also had siblings with CF.

I was aware there may be aspects of data collection that I took for granted as a clinician researcher – or that families took for granted in sharing their insights with me – conversely, did this mean we could have a more in-depth discussion? I took care to seek clarification, check what they understood 'x' to be, what

¹ 11-year-olds were not interested and so further work is needed regarding what hands-on activities would engage them

meanings they attributed to words/ actions and asked for examples wherever possible. At several points throughout each interview, I checked if they were happy to keep going/ go ahead with the next part of the interview.

Regarding their perception of me, as the interviews were not conducted in the clinical environment, they may have viewed me more as a researcher than a clinician. Children were used to having video calls due to Covid (e.g., for completing school lessons), they were in the comfort and familiarity of their home environment and I was readily shown inside cupboards and fridges. We could see each other, but as I was not in their physical space, this may have contributed to less of a power imbalance.

5.2.5.3 Reflections on data analysis

There were a few points for reflection throughout data analysis. For some of the interview transcripts, it was noted that the audio quality had been poor and it was a lot of work adding to and correcting transcripts to ensure they were accurate.

Conducting Framework was very time-consuming and it took double the amount of time I had planned, despite being able to draw on the experience of my supervisors in using this approach. Preparing how to explain to PAG how I had moved from the interview data to begin developing an initial framework really helped me understand what I was doing. A sample of the analysis and interpretations were also to be shared with YPAG. However, the technology failed and it was not possible to reschedule their input.

A large volume of data was generated through interviews with 58 participants, so I used NVivo12 to help organise and manage it. It was time consuming learning how to use the software and frustrating that I could not 'see' the data in the same way as if using paper charts displayed across a wall i.e., it made the process of data analysis less tactile. However, these disadvantages were outweighed later on by being able to auto-summarise each chart, and the easy retrieval of data when linking back to the original transcripts.

5.3 Chapter summary

This chapter has presented application of the working methods for phase two. It detailed how participants were recruited and how data was generated and analysed. In the following chapter, the findings of phase two are presented and discussed.

Chapter 6

Phase Two: Qualitative Descriptive Study - Findings

6.1 Introduction

This chapter follows on directly from Chapter 5, providing an overview and discussion of the findings of phase two. It begins with presenting the characteristics of study participants, followed by an overview of the findings as a thematic framework and then as a narrative, with exemplar quotations from participants to illustrate how themes were grounded in the data. It concludes with a discussion of the findings in relation to answering the phase two study questions and how these informed development of the model (conceptual framework) in phase three (Chapter 7).

6.2 Findings

This section details what the gathered data looked like and how it was interpreted.

Between September 2020 and May 2021, semi-structured interviews were conducted with a total of 58 participants: 18 children's CF dietitians, 20 children and 20 parents.

6.2.1 Characteristics of participants

Children's CF dietitians

All but one of the 18 dietitians interviewed was female. Other characteristics are summarised in Table 6.1. The size of the specialist CF centres at which the dietitians worked, varied considerably, with as few as 37 children cared for, through to 340 children. Although the numbers of children receiving full-care versus shared-care at each specialist centre was not recorded, the dietitian: child ratios were inconsistent across the centres, suggesting some centres were markedly understaffed at the time of interview. Dietitians' experience in children's CF ranged between one and 30 years, though many had several years prior experience in paediatrics and several worked across both children's and adults CF.

Table 6.1: Characteristics of participants: Children's CF dietitians

Dietitian code	Size of specialist CF centre approx. no. of children seen	Number of Dietitian WTEs for children's CF (current number due to maternity leave)	Experience in children's CF (no. of years)	Age range (years)
D01	~230	2.0 (1.0)	20	45-54
D02*	40	0.2	10	35-44
D03	~170	1.0	2	55-64
D04*	37	0.28	12	45-54
D05	310	1.4 (0.8)	5	45-54
D06	~200	1.6-2.0	13	45-54
D07	75	0.8	6.5	25-34
D08	220	1.1	25	45-54
D09	340	3.0	13	35-44
D10**	~210	1.0	5	25-34
D11	310	1.5	30	55-64
D12	50	0.5	13	35-44
D13	67	0.5	10	35-44
D14*	78	0.6	6	35-44
D15	187	1.0	20	45-54
D16	325	1.7	2	25-34
D17	~190	1.0	9	35-44
D18	~145	1.2	1	55-64

* Works across children's and adults CF;

** Usually works in adult CF

Following discussions with supervisors LM and GM, interviews were halted after 18 were completed, as it was felt that data saturation assessed in terms of data adequacy (as described in section 2.6.3) had been achieved.

Children with CF and their parent(s)

Similarly, with children and parents, interviews were halted after 20 interviews. Of the 20 children interviewed, seven were receiving care at centre A, six at centre B and seven at centre C. Characteristics are summarised in Table 6.2.

There were more girls than boys, 12 and eight respectively. All but one of the children reported their ethnicity as white. This reflected the profile of CF, with 93% of the UK CF population currently reported as white (CF Trust, 2021). The children's ages ranged between 7.3 and 11.9 years (mean 8.8 years). The two older children, aged 11.5 and 11.9 years had already transitioned to secondary school. Only one child was gastrostomy fed and three regularly used oral

nutritional supplements, indicating how the majority of children had a good nutritional status.

Fifteen children had been diagnosed with CF through new-born screening (NBS). If more children had initially presented with meconium ileus (MI), greater gut involvement in their CF would have been expected (Sathe and Houwen, 2017). Only a quarter of the children had required intravenous antibiotics (IVAT) in the year prior to their interview. With the exception of one child, who was on a demanding treatment regime for Non-Tuberculosis Mycobacteria, this suggests children were well at the time of interview. Several children had begun HEMT; though this was discussed during interviews as appropriate, the specific HEMT taken by each child was not recorded. The socioeconomic status of families was not captured, which was an omission. In retrospect this should have been captured as low socioeconomic status is associated with worse clinical outcomes e.g., reduced growth and lung function (Taylor-Robinson et al., 2015) and may impact a family's capacity to self-care.

Table 6.2: Characteristics of participants: Children with CF and their parent(s)

Child					Parent/ Carer	
Code	Age (years)	Gender	Age at diagnosis, how diagnosed (NBS/ MI/ other)	No. of courses of IVAT over past year	Code	Relationship to child**
C01	10.1	Female	3 weeks, NBS	Nil	P01	Mum
C02	10.7	Female	4 weeks, NBS	Nil	P02	Mum
C03	7.6	Male	3 weeks, NBS	1	P03	Mum
C04	11.5	Male	5 weeks, NBS	1	P04	Mum
C05*	7.4	Male	4 weeks, NBS	Nil	P05	Mum
C06	11.9	Female	4 weeks, NBS	Nil	P06	Mum
C07	9.0	Female	2 weeks, MI	Nil	P07	Mum and Dad
C08	9.6	Female	At birth, MI	2	P08	Mum
C09	7.6	Female	4 weeks, NBS	Nil	P09	Dad and Mum
C10	9.9	Male	3 weeks, NBS	1	P10	Mum

Child					Parent/ Carer	
Code	Age (years)	Gender	Age at diagnosis, how diagnosed (NBS/ MI/ other)	No. of courses of IVAT over past year	Code	Relationship to child**
C11	10.6	Male	3 weeks, NBS	Nil	P11	Mum
C12A and C12B	11.3 7.3	Female Female	3 weeks, NBS 3 weeks, NBS	Nil Nil	P12	Mum and Dad
C13*	8.3	Female	2 weeks, NBS	1	P13*	Mum
C14	8.7	Female	6 weeks, NBS	Nil	P14	Mum
C15	8.6	Male	6 weeks, NBS	Nil	P15	Mum
C16	8.7	Male	At birth, other	Nil	P16	Mum
C17	11.3	Male	2 years, other	Nil	P17	Mum
C18	8.8	Female	3 weeks, NBS	Nil	P18	Mum
C19	7.3	Female	18 months, other	1	P19	Mum
C20	8.6	Male	3 weeks, NBS	Nil	P20	Mum
C21	9.1	Female	3 weeks, NBS	Nil	P21	Dad

IVAT: intravenous antibiotics NBS: new-born screening; MI: meconium ileus; *Declined to take part; **Where both parents participated, the main contributor appears first.

6.2.2 Overview of the findings as a thematic framework

The final thematic framework used to organise the data (as described in section 5.2.4) is presented in Table 6.3. The framework captured the richness and depth of data gathered in the interviews and structured it to answer each of the phase two study questions.

As shown in Figure 6-1, the framework consisted of five categories. The first two categories addressed 'what is SCS of diet and the gut?', by defining its characteristics and content. The third category addressed 'how can SCS be delivered as part of routine care?', with the fourth category attending to the potential barriers and facilitators for delivery and uptake as part of routine care. Finally, the fifth category addressed 'what outcomes would be most important to achieve?'

Figure 6-1: The final thematic framework

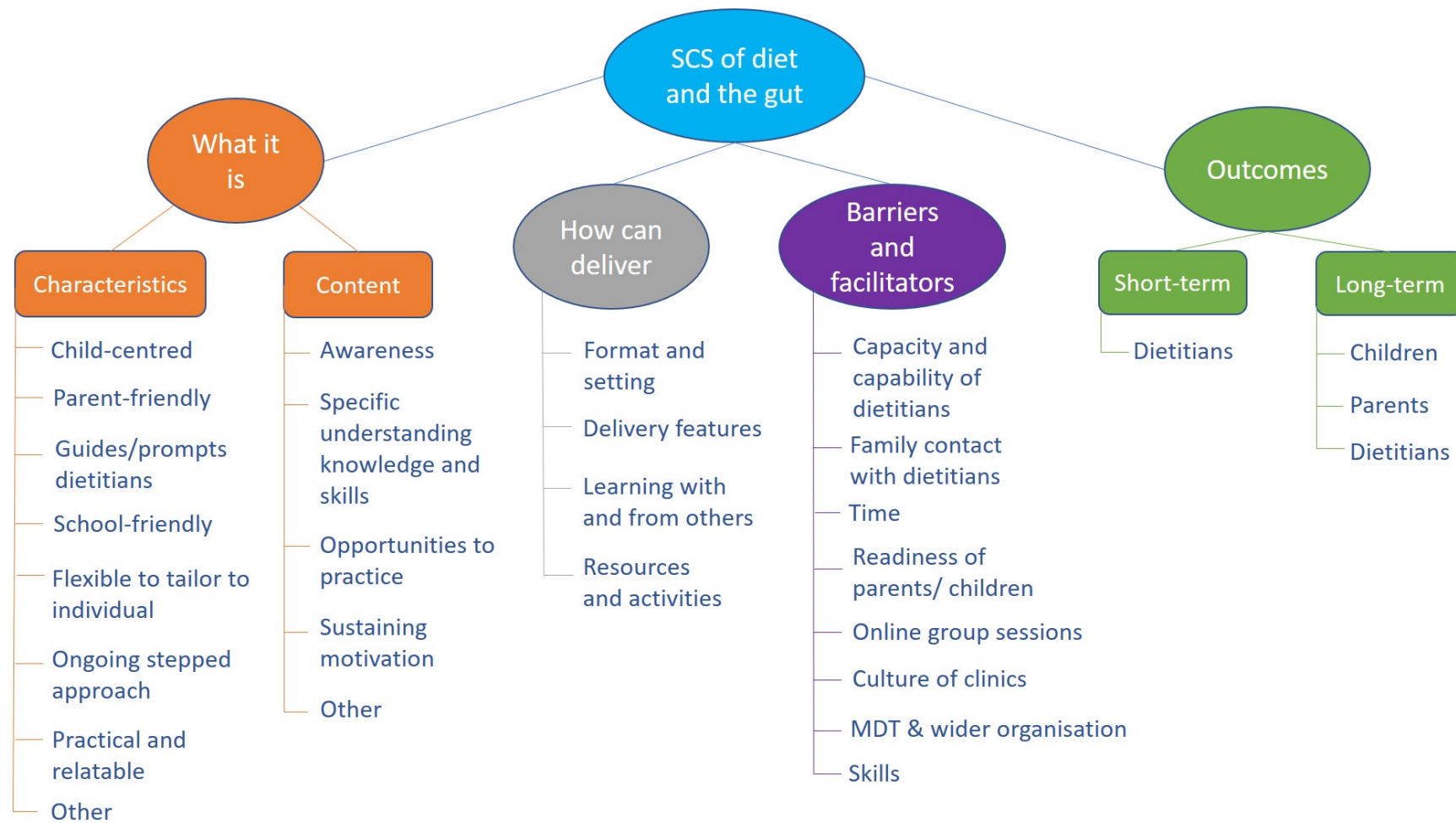


Table 6.3: Thematic framework for phase two

SCS of diet and the gut: what it is - characteristics	
Child-centred	Directly engages/ involves the child in learning to look after their diet and gut; uses child-accessible language; responsive to child's questions/ curiosity
Parent-friendly	Aids parents in supporting their child; enables parents to learn themselves/ serves as a refresher; sets out expectations of what will happen when (the trajectory for supported gradual learning)
Guides/prompts dietitians	Provides a structure (a focus) for delivering consistent and comprehensive support – what's appropriate for children to learn when and how, with links to access resources and appropriate activities; strategies/ techniques to facilitate child and parent learning
School-friendly	Aligns with school curriculum; complimentary resource for schools to increase awareness and understanding of staff (and peers) of child's needs with respect to eating and PERT whilst at school [to link in with CF Trust school resources]
Flexible to tailor to individual	Appropriate to age and developmental stage; can be matched to differing priorities/ needs/ interest and learning style preferences of the child/ family at a particular time (includes if diet/ gut/ weight an issue for them); can dip in and out (it's extra support that is there, with no pressure to have to engage with it or complete it in full)
Ongoing stepped approach	Regular revisiting of topics - assessing understanding (rather than making assumptions about what is understood or not) and topping up ('drip feeding'); reinforcement; a grounding/ foundation from which to progress
Practical and relatable	'Doing' things, trying things out; gives access to options/ increases awareness - to enable increased choices (you don't know what you don't know); resonates with/ people able to readily identify with/ care about the content
Other	Target age range – preadolescence vs adolescence; health promotion element - pro-active/ anticipatory

Table 6.3: Thematic framework for phase two : continued

SCS of diet and the gut: what it is - content	
Awareness	Need for PERT with foods; reminding/ asking parents/ carers for PERT; what signs/ symptoms to report to parents/ CF team (what to look out for, what different feelings in their body might mean)
Specific understanding, knowledge and skills	<p>Understanding and knowledge:</p> <p>Importance of taking PERT, why they need PERT, what it is and how it works; what happens if take too much or too little PERT; which foods contain fat and how much and types of fat – sources of ‘good’ vs ‘bad’ fats; explanations they can share with peers.</p> <p>Different parts of the gut/ body; why may experience GI symptoms; benefits of eating a wide range of foods (including for long-term health); food choices that are right for them (including how this may change if they are unwell and how what’s ‘healthy’ for them (at a particular time) might not be healthy for others); how to have extra calories in a balanced way; importance of fluids and how much they need; fibre, salt, calcium, iron</p> <p>Skills:</p> <p>Swallowing tablet/ capsule preparations; maths/numeracy; reading and interpreting food labels; administering PERT (set doses, timing); titrating PERT to fat (using a ratio or ‘broad brush’ low-medium-high approach) (including for home-cooked foods and when eating out); self-monitoring e.g., recognising GI symptoms and helping begin to problem solve/ manage; taking part in decision making; food preparation</p>
Opportunities to practice	Time and space to repeatedly practice applying knowledge, practice skills and learn from mistakes (learn from experience) (with parent checking/ supporting); providing appropriate prompts/ cues for this age group (e.g., to facilitate remembering to take PERT); practice over time develops confidence
Sustaining motivation	Support to keep going with daily self-care; discussing/ addressing perceived or actual barriers/ challenges to self-care; facilitating formation of habits/ routines (including developing habit of remembering to take PERT); positivity and encouragement (rather than always – ‘could do/ be better’); supporting transfer of responsibility for self-care over time
Other	Addressing changes (re-education) with/ impact of HEMT (e.g., increased appetite, increased weight, stopping oral nutritional supplements, changing balance of diet, possible reduction in PERT requirement etc.); need something specifically for siblings and close friends?

Table 6.3: Thematic framework for phase two : continued

SCS of diet and the gut: how it can be delivered	
Format and setting	Where/ when: (1:1) incorporated into outpatient clinic (+/- annual assessment) with option to take home if appropriate; during hospital admission/ lengthy trial visits; if possible/ appropriate during home visit/ school visit/ (within the community?); at home; in groups online
Delivery features	Visual (simple pictorial for younger ones) (includes showing what a 'healthy' diet looks like); interactive (not passive) i.e., with feedback/ facilitated by an HCP/ another person; fun, creative, learning through play – games and (short learning burst) bite-sized activities; knowledge/ info exchange; online resource hub to access anytime – central point of access (c/o the CF Trust?); 'who' delivers – dietitians/ Dietetic Assistants/ CFNSs or dietitians with another member of MDT e.g., physiotherapist, social worker, psychologist (depending on topic being delivered)
Learning with and from other children with CF/ other parents of children with CF*	Feeling connected to others with CF/ affected by CF; able to ask questions they wouldn't ask people without CF; video library e.g., children sharing filmed demos/ sharing explanations (how it makes sense to them), how they manage different things; families sharing (lived experience of) what works well/ food suggestions and recipe ideas; role modelling/ leading by example – showing them how/ being inspired
Resources and activities	(Examples of) resources/ activities currently using; resources previously used; resource ideas (e.g., recipe bank, animations); activity ideas (e.g., in groups - cook-a-longs, group games, quizzes); items in activity pack

*Whose children are the same age/stage or a little older/further on

Table 6.3: Thematic framework for phase two : continued

SCS of diet and the gut: potential barriers and facilitators for delivery and uptake as part of routine care		
	BARRIERS	FACILITATORS
Capacity and capability of dietitians	Limited willingness to engage in SCS (e.g., dietitian viewing SCS as chiefly role of parent); dietitian approach – quite prescriptive/ authoritarian (e.g., focus = sorting out issues); possible future reduction in CF dietetic time due to HEMT/ reduced capacity to deliver additional support in an underfunded/ short staffed service; variability in practice and emphasis across specialist and shared-care centres	Dietitian viewing SCS as inherent to their role; dietitian approach of enabling; bringing knowledge and experience of what children can do at different ages; ability to demonstrate making (more/ most) effective use of contact time; (need) knowledge, skills and confidence to deliver SCS; training (possible formats: video, groups of dietitians face-to-face or online (practical discussions, sharing experiences) alongside a user 'how to deliver' guide); supervision/ support to deliver (e.g., from specialist centres or more experienced dietitians)
Family contact with dietitians	Limited contact with dietitian e.g., when at a shared-care centre/ usually only see dietitian briefly unless there is an issue; family feeling additional support/ SCS from dietitian/ MDT not needed (or not sure what SCS from the dietitian could look like)	More interesting/ useful when do have contact with dietitian? (less repetitive/ boring/ more fun); families and dietitians know each other well (relationships built up through seeing each other regularly over long periods of time); parent/ child able to ask dietitian questions in and outside of clinic (e.g., by phone, email) and contact with dietitian via video call liked
Time	Time constraints/ lack of opportune times for delivery or uptake; competing demands on family's time; other aspects of CF needing to take priority; with HEMT, families spending less time in hospital	Setting expectations/ planning in advance; utilise gaps between HCPs in outpatient clinic +/- if appropriate, include at annual assessment; utilise time freed up (in clinic) by children being more well on HEMT; resources to access anytime

Table 6.3: Thematic framework for phase two : continued

SCS of diet and the gut: potential barriers and facilitators for delivery and uptake as part of routine care		
	BARRIERS	FACILITATORS
Preparedness/ readiness/ concerns of parents	Reduced preparedness/ readiness of parents to begin 'letting go'/ concern re: transfer of responsibility/ willingness to engage or support child's involvement; parents focus on what they need to know at the time (a coping strategy for dealing with CF); not wanting child to get it wrong or grow up so fast or appear different	Prompts/ supports parents: with gradual changes/ transfer of responsibilities for self-care (including use of language/ approaches to try), in supporting their children; parent approach helps child to give it a go; agreeing in advance the roles, tasks and responsibilities for self-care/ SCS adopted by children, parents/ carers and dietitians
Willingness/ readiness/ interest of children	Reduced willingness/ readiness of children to discuss CF/ begin taking on more responsibility/ ownership of certain self-care tasks; reduced willingness/ readiness to engage as not wanting to appear different; little enthusiasm/ ambivalence about attending online group sessions; no/ little interest in understanding 'why'; character of child e.g., shyness inhibits direct engagement/ involvement	Not described
Online group sessions	Concerns re: information governance/ safety aspects; no/ limited access to (IT) equipment/ connectivity (data)/ reliable platforms or for cook-alongs, to cooking equipment and ingredients; child/ parent not wanting child to make a friend(s) because they are not allowed to meet in-person; preference for parents to meet face-to-face; previous negative experience puts them off; only those already interested/ engaged attend	Increased familiarity with IT/ use of online platforms (including as part of CF care) as a result of COVID; sessions structured, facilitated and provide a secure safe space; provides an opportunity for peer support - to meet/ talk to: children around the same age about normal things, not necessarily to do with CF, or parents (perhaps at specific times) with children around the same age/ stage or a little older/ further on;

Table 6.3: Thematic framework for phase two : continued

SCS of diet and the gut: potential barriers and facilitators for delivery and uptake as part of routine care		
	BARRIERS	FACILITATORS
		negates need for travel, parking, child care; could be delivered regionally or centrally (e.g., c/o CF Trust) to children nationally
Accessibility	[Overlap with online group sessions]	Available in different languages (as and when required)
Culture of clinics	Current practice is to meet the CF Trust standards of care/ (deep-rooted) expectations of what happens in clinic/ (high) frequency of clinics; current practice is to learn through talking; clinic visits can be boring and lengthy (and children tired and disengaged as a result); (medical) terminology used is not appropriate/ helpful; limited direct engagement/ involvement of children/ parent(s) speaking on child's behalf; focus on weight	Flexibility to take the time needed to see patients; ethos of actively involving children
MDT and wider organisation	Not described	Having support/ awareness of MDT; aligning/ overlap delivery with other MDT members; (complimentary/) resources accessible to other MDT members to help with consistency of messages re: diet/ gut across MDT and between specialities e.g., DM; seen as part of the service – have organisational buy-in and support
Skills	Poor literacy and/ or numeracy skills of children/ parents	Not described

Table 6.3: Thematic framework for phase two : continued

SCS of diet and the gut: outcomes	
Short-term	
Children	Not described
Parents	Not described
Dietitians	<ul style="list-style-type: none"> • Increase direct engagement with children • Provision of information that's more accessible to children and families • More pro-active (anticipatory/ preventative) approach <p>Dietitian outcomes for children</p> <ul style="list-style-type: none"> • Increase knowledge and understanding
Long-term (at the point of transition from primary to secondary school)	
Children	<ul style="list-style-type: none"> • To be independent and confident with PERT (includes being able to relay simple explanations to peers/ people they don't know and being in the habit of remembering to take their enzymes) so have an increased sense of control • Knowledge and confidence to choose foods that are right for them (includes how to have extra calories in a balanced way) • Meeting and talking to/ learning with/from other children with CF
Parents	<ul style="list-style-type: none"> • Knowledge and confidence to gradually hand over responsibility for certain self-care tasks and support their child's increasing independence • Able to access more support if wanted, particularly from early doors (includes peer support (at specific times), online resource hub etc.) <p>Parent outcomes for children</p> <ul style="list-style-type: none"> • To be independent and confident with PERT (includes being able to relay simple explanations to peers/ people they don't know and being in the habit of remembering to take their enzymes) so have an increased sense of control, but includes when outside the home or usual routines • Be independent with choosing foods and have confidence to try new foods • Increased involvement of child in own care and gradually taking more responsibility e.g., able to recognise and help to problem solve/ manage GI symptoms • Meeting and learning with/ from other children with CF <p>Parent outcomes for dietitian</p> <ul style="list-style-type: none"> • Reduced pressure on parents re: weight; shift focus (from weight) to overall wellbeing of the child • Help build and reinforce child's confidence <p>Parent outcomes for school</p> <ul style="list-style-type: none"> • Increased knowledge and understanding
Dietitians	<ul style="list-style-type: none"> • Able to have more open and honest discussions with children (and continuing as they grow) • Enabling children's increased self-confidence and independence • Empowering children to make decisions that are right for them

Table 6.3: Thematic framework for phase two : continued

SCS of diet and the gut: outcomes	
	<ul style="list-style-type: none"> • Encouraging improved quality of children’s diets • Standardised (consistent) content and delivery across UK CF centres • Ability to demonstrate making most effective use of dietitian contact time <p>Dietitian outcomes for children</p> <ul style="list-style-type: none"> • To be independent and confident with PERT (includes being able to relay simple explanations to peers/ people they don’t know and being in the habit of remembering to take their enzymes) so have an increased sense of control, but includes when outside the home or usual routines • Increase knowledge and understanding of the gut in CF, recognise GI symptoms and communicate this/ begin to help problem solve/ manage GI symptoms • No or reduced GI symptoms (particularly in children for whom there is a lot of gut involvement in their CF) • Increase knowledge and understanding of diet in CF and confidence to choose foods that are right for them • Achieve optimal nutritional status/ growth and positive impact on overall health • As child grows, and goes into teens, able to use the CF MDT effectively as a resource – able to ask questions/ have more open and honest discussions with (parents)/ dietitian/ MDT around what they are doing (/not doing) and what support they need • Begin to engage in decisions that concern them <p>Dietitian outcomes for parents</p> <ul style="list-style-type: none"> • Increased knowledge and understanding of gut in CF • Encouraging improved quality of children’s diets

CFNS: CF nurse specialist; DM: diabetes; HCPs: healthcare professionals; HEMT: highly effective modulator therapies; MDT: multidisciplinary team.

6.2.3 Narrative of the findings

Through rigorously conducting the interconnected stages of Framework, a thematic framework was developed for phase two. In the following narrative, key themes from the framework are presented in detail, supported by exemplar quotations from participants.

The narrative is structured to answer, in turn, the phase two study questions:

- What could SCS of diet and the gut consist of?
- How could SCS be delivered as part of routine care?
- What outcomes would be most important to achieve?

6.2.3.1 What could SCS of diet and the gut consist of?

In answering this question, key themes were being child-centred, adopting an ongoing stepped approach and establishing health-promoting behaviours.

Child-centred

The findings suggest an essential component of SCS is dietitians involving and engaging the child directly in learning to look after their diet and gut, and supporting the parent to support their child. This requires a shift from paternalistic to child-centred care for children to begin making sense of what is happening, by positioning sense-making from the child's perspective (Coyne et al., 2016).

For children to construct understanding of what is happening, more child-accessible language is needed. For example, to help children understand what signs and symptoms to look out for and begin making the connection as to why they might be experiencing symptoms, including in relation to their PERT:

'Children like you (addressing C08) or other kids might like to be able to recognise themselves when they go to the loo, the clues about if they've had too much or too little Creon (PERT), that's probably something that's quite important to learn... To try and spot the patterns so that you can look after it yourself' (Parent, P08).

Constructing understanding of what is happening can be challenging because of the largely abstract nature of the topic (e.g., 'fattiness' and size of the meal versus the actual fat content) at a concrete stage of their cognitive development (Brouse and Chow, 2009). There is also a requirement for children to understand and feel confident with numbers when learning to be independent with PERT.

'You do have to have quite good maths skills in my opinion' (Child, C04, boy, 11 years).

The current CF service model presents a barrier as there are very few resources available and nothing is happening (in clinic or elsewhere) to help children learn, other than through talking and this is often mostly amongst the adults. Clinic visits for children can be lengthy, boring and repetitive:

'Sometimes it's a bit boring whilst my mum talks. And my dad. I'm just, like, bored waiting for them to stop. But if I have a question, I'm like, waiting for ages' (Child, C20, boy, 8 years).

'We tend to talk to the parents a lot, and obviously then the children get to 10 or 11 (years) and don't really know what they're doing, so they don't become that independent with their care... It must be really frustrating for patients and parents when we just do the same thing over and over again' (Dietitian, D16).

The dietitian's approach could also present a barrier. Though some dietitians saw SCS as inherent to their role and were already delivering (ad-hoc) SCS in a child-centred way, other dietitians appeared to be delivering narrower self-management support. They described a focus on 'sorting out issues'. This corresponded with reports by several parents, that the dietitian was only seen briefly in clinic, unless there was an issue, and there was very little, if any, involvement of the child.

'So the dietitian part (of the clinic visit), unless he's had a bad tummy, it's a very small part of the clinic. And it's over as quick as we can. It's hi [name of dietitian], yes, everything's fine, thanks a lot, bye. Or, actually we've got some issues we need to talk about... So, when it comes to dealing with the dietitian, it's me that deals with it' (Parent, P03).

Brief and focussed contact with the dietitian led several parents to express uncertainty as to what SCS from the dietitian could look like. Other parents viewed this scenario as a missed opportunity for education and, for example, exploring the practicalities and children's progress with performing daily self-care tasks:

'And it's always been keeping it (PERT) simple to help your tummy work, help you absorb the fat in your food, but to actually move it onto the next level of how the pancreas releases enzymes and yours are getting stuck in thick mucus...we've not gone...and I think we probably should, especially with the leap between primary and secondary school is so great and they'll be learning about that kind of thing in lessons... Also, it is that discussion about how things are going, 'what stage are you at with looking after yourself? do you understand about your Creon?' And [name of dietitian] does do a lot of 'what did you have last night and how many Creon did you have with that?' But to go to the next stage of 'did Mum have to remind you or are you big enough to be doing that yourself now?' And having a target for what to be doing by next clinic, that would be useful' (Parent, P10).

Differences between dietitians experiences such as when they had completed their dietetic training or how long they had worked in children's CF, did not appear to influence the contrasting approaches. However, it was clear that more positivity and encouragement is needed to maintain children's motivation to keep doing daily self-care, rather than the message being 'could do/ be better', as this dietitian describes:

'That's quite often what children hear, that it's never enough, even if their weight's okay and their bowels are okay, they're not doing something right. And I think that's just us... always searching to be helpful and give advice, trying to fine tune things all the time... and tweak things or make sure everyone's doing as best they can' (Dietitian, D09).

In contrast, a facilitator of SCS is that families and dietitians know each other well. Relationships are built up through seeing each other regularly over long periods of time and provide a strong foundation for SCS. With appropriate training, supervision, access to resources (and the guidance of a 'road map – see below), there is an opportunity for dietitians to consistently deliver child-centred SCS.

Ongoing stepped approach

The findings suggest another essential component of SCS is having a 'road map' for gradual ongoing learning, whereby the possible roles and responsibilities for self-care/ SCS adopted by children, parents and dietitians are planned in advance, with expectations set out regarding what may happen when. This would include approximate targets for milestones and choices for how milestones are achieved. A parent summarised this as:

'A programme that would have like a year-by-year stepped approach to being more independent, with learning about foods and gut and Creon (PERT)' (Parent, P04).

The proposed target age range of 6/7-11 years was thought appropriate, for being able to respond when children start asking questions about CF and in gradually preparing for transition from primary to secondary school/ transition to adolescence.

'I think that if they start sort of at seven (years), taking a little bit more on, it's so gradual, by the time they get to 11 (years), that you haven't

noticed that big, oh gosh, I haven't done anything, and now he doesn't know what he's doing and it's really panicked him... These are skills for life... and starting them when they're younger... putting the good foundations in, when the challenges come up, they're (children are) going to be more robust to cope with it' (Parent, P03).

The 'road map' could prompt dietitians not only regarding what to deliver when and how with children, but also when and how to facilitate gradual transfer of responsibility for self-care tasks, from parents to children. For example, in providing practical strategies and approaches to try. Parent interviewees varied in their willingness and/ or readiness to begin 'letting go', with some promoting and encouraging their child to take on more responsibility, whilst others described wanting to protect and shield their child. This impacted children in different ways, with some able to try things out, and others quite unaware that anything needed to change, as exemplified by these dietitians :

'Some parents are more relaxed about it and I think the child picks up on that and is therefore willing to give it a go, and can learn what works for them' (Dietitian, D02).

'It's very difficult for the ones (children) whose parents just take onboard all of the responsibility... Because... it's as if it's not their condition, it's just something that their parents manage and I think you can run risk then of them disassociating from it, always thinking I don't need to do anything with this, this is something that is done to me or for me. And then they're often very stuck when it comes to transition' (Dietitian, D13).

A road map would need to address this variability in practice, to support parents at whatever point of readiness they are. This is because parent support of their child will be essential for the child learning and performing self-care. Several parents wanted structured stepwise support with the 'handover' process, for example:

'It's at what point and how do I let go and how do I allow him (referring to C11), give him the rope to get it wrong, essentially?... having that objectivity of somebody to say, you know, maybe try this or maybe try it that way is helpful... It's not saying the wrong thing when they're really fed up about having to take tablets...for those different stages that they go through, what can we say and do to make it better for them and what can we say and do to develop their independence' (Parent, P11).

There were a range of examples of dietitians already delivering SCS to parents and children in the target age range, but in an ad-hoc way. Having a roadmap (structure) could increase consistency and breadth of delivery across centres and over time.

Participants accounts suggested that interaction with/ facilitation of activities by a dietitian or another person was important for children's learning and also for assessing children's understanding (particularly of *why* they are being asked to do things). A regular revisiting of topics and topping up (described by several dietitians as 'drip feeding'), alongside children having opportunities to repeatedly practice over time was key to the 'stepped' approach, and would mirror current practice in T1DM:

'The more we do early on, but do it repeatedly I think is quite important, to do something but then to revisit and build on, going into further depths' (Dietitian, D06).

'Diabetes do a number of bite size sessions where they're just targeting a small aspect of care in very short hits and they're very good at checking back to ensure there's understanding' (Dietitian, D11).

Descriptions of children practicing (with their parent(s)/carer(s) supporting), enabled learning through trial and error and helped them develop their confidence. This learning through experience was also recalled as important by adults with CF in an earlier study conducted by the researcher (outlined in section 1.7.2):

'It does get much more easier. I think when you've done it for quite a long time, I think you get a bit more confident doing it' (Child, C09, girl, 7 years).

Aligning ongoing learning with the school curriculum (i.e., with what they will be learning at school) was suggested by many of the children, parents and dietitians, particularly as a way of ensuring learning is appropriate to children's ages/ developmental stages. A complimentary resource for school is also needed, for the child's peers and all staff who come into contact with the child. This is to increase awareness and understanding of (i) CF and (ii) the children's needs with respect to eating and PERT whilst they are at school:

'When [name of C18] started (school), you say, 'oh, she has CF', and they instantly just think lungs and they don't know much about the digestive side of things at all. That was a real difficult first couple of years... With CF, a lot of the main things happen at home, like the treatment, the physio, but the Creon and things like her food and her tummy, that all happens throughout the day. So that's like a key thing that all the children pick up on, is the Creon and 'why do you take that?' (Parent, P18).

'It's good in my school because all my teachers understand why I have to have Creon' (Child, C07, girl, 9 years).

Though dietary advice for CF is changing (particularly with the introduction of HEMT), and choosing different (healthier) foods may enable children to feel less different to their peers, the requirement for PERT will, however, remain, as may the need for higher calorie foods at certain times, for example during an acute exacerbation. Children gradually learning to look after their diet and gut, will therefore remain relevant, and could be optimised within the more supportive environment of primary school, prior to the step up in responsibility and independence needed at secondary school.

Establishing health-promoting behaviours

A key component of SCS that appeared to be missing in the descriptions of current dietetic care, was that of health promotion. However, the need for this proactive component to be integral to ongoing learning, was expressed by many parents and dietitians. For example, dietitians providing anticipatory guidance may prevent problems occurring, rather than children and parents receiving information only in response to a problem, such as a blockage in the gut (which requires treatment, and possibly a hospital admission). Another example was planning ahead for possible changes with starting HEMT, as described by a dietitian and parent:

'It worries me about all the body image stuff, the kids being really thin and then suddenly they're going to...well, they might put weight on (with starting HEMT) and how they cope with all of that. How we manage that really and prepare them for that' (Dietitian, D03).

'We are preparing [name of C11] for when he starts [name of next HEMT] that we've probably got to reduce some of the junk food and the high calorie stuff... we have started to think about how do we get healthier fats... [name of dietitian] has provided some excellent materials for us, recipes and those sorts of things and the types of food to be eating' (Parent, P11).

Anticipatory guidance could facilitate development and maintenance of routines and habits. Allied to this, the provision of prompts or visual cues appeared to be a key gap to fill for this age group in developing the habit of remembering to take PERT. Any prompt/ cue would need to be something the child readily identifies with, cares about or provides some sort of reward for performing the behaviour of remembering PERT. Friends (healthy peers) may also play a role in prompting (also a finding of the IR), for example:

'If I'm honest, completely honest, I'm not that great at remembering to take it (PERT). I have one friend who has been my friend since we were three (years old) so she usually reminds me to take it because she's grown into that habit as well!' (Child, C06, girl, 11 years).

A barrier when taking PERT is a habit and is automatic, was also highlighted (as it was in the study with adults with CF (section 1.7.2)):

'Obviously because they've done it for so long (taken PERT), it's just second nature. And then it's 'did I have it? did I not have it?', that way it's a bit tricky' (Parent, P12).

Further exploration of these findings, to propose how the content of the SCS intervention could provide practical solutions to these challenges, is detailed in phase three (Chapter 7). Whilst themes important in answering the second question of phase two now follows.

6.2.3.2 How could SCS be delivered as part of routine care?

In answering this question, key themes were low intensity delivery and learning with, and from, other children with CF.

Low intensity delivery

Preferences for where and when SCS is received/ delivered differed markedly between families and dietitians, but all were low intensity. For children and parents, SCS was a mix of at home online, mostly in groups and/ or one-to-one as part of an outpatient clinic review. However, for dietitians, there was a preference for online in groups and one-to-one review during hospital admissions. Looking at these preferences more closely revealed the following. Incorporating short activities into outpatient clinic would provide an opportunity for children to receive immediate feedback and/or further explanation. Gaps

between reviews by different HCPs could be utilised (note in CF clinic, the family stays in the same room for the whole visit and HCPs rotate), for example:

'We did a questionnaire to see what families wanted and they did want something to do in the downtime between professionals... rather than us just hitting them with loads of questions, they would answer those in that downtime so that in the time (we had together) we were able to discuss them and then potentially, do a bit more education with them' (Dietitian, D17).

However, some children and parents reported they can be too tired and/ or disengaged due to the length of clinic, or dislike being in clinic for longer than is absolutely necessary, therefore home was the favoured setting for SCS. One parent highlighted the importance of it being encouraged and signposted in clinic, to make it more likely to happen at home. The dietitians' preference for online or during admissions appeared to be chiefly explained by clinic visits already being busy and time pressured. However, with some forward planning, clinic may be possible, for example:

'I think sometimes we get carried away with 'oh, I have to check this, I have to check that'. But most patients are pretty good at telling you if there's something wrong, and if you had planned that at some point, we would use a resource and get them working on it, I think that could be done in a regular (clinic) slot' (Dietitian, D10).

It was also acknowledged by dietitians that although hospital admissions were an opportunity to spend more time with children and parents, and run through things in more detail, less children were now being admitted to hospital, so there would be more reliance on delivering SCS in an outpatient setting or at home.

As part of a low intensity delivery, it was requested that all resources are together in one place, in an online hub, so they can be accessed at any time, including during clinic. The idea of a hub fitted with an overall preference for SCS being support that is there if you want it, with no pressure to have to engage with it in full. However, the resource hub could reinforce key learning for children (and parents). A wide range of age/ developmental stage-appropriate resources/ task-based activities would be needed to tailor to individuals:

- To match differing needs/ priorities/ interests and learning style preferences of the child/ family at any one time, and over time
- Having choices/ options for how to learn was reported as important
- The CF population is becoming increasingly diverse (e.g., children's nutritional status ranges from <0.4th centile through to >98th centile, some are taking HEMT whilst others are not eligible or not able to tolerate HEMT)
- Interest in any one resource/ activity may be short-lived, as described by this young boy:

'Well, if they're anything like me, they have phases of what they like, they go mad over something they like for a little bit of time and then they'll change' (Child, C11, boy, 10 years).

Participants accounts revealed a preference for resources/ activities to enable learning through play and be fun. Visual learning appeared to be key. For example, many of the children reported how they had loved watching and re-watching the animation 'Oli and Nush' (CF Trust, 2010; targeted at children with CF aged approximately six years). The element of humour in this animation was also mentioned by several children as important for their engagement.

Having accessible resources could enable parents to learn themselves or serve as a refresher. It was acknowledged by many dietitians that high intensity education is delivered to parents at diagnosis, and in the following one to two years, but is then rarely revisited. A dietitian clearly expressed the impact this may have:

'We tell our parents so much early on, but do we go through it again? No, probably not. I think then it can be difficult for the parents to say 'well, actually, my child's six (years) and I have no clue why we're doing these things'. I don't think that's a very easy conversation for a parent to raise with us. So actually, this is almost a way of doing a refresher with them in a gentle and supportive way without challenging them or exposing them in a way that makes them feel uncomfortable' (Dietitian, D11).

All members of the MDT being able to access the resources may increase consistency of messages about diet and the gut, across the team and across specialities such as diabetes. This may also reinforce key learning. Aligning/ overlapping delivery with other members of the MDT, where appropriate, may

also present families with a more joined up, 'package' of care. These findings were in agreement with those of the IR, whereby SCS needs to be seen as part of the service, with MDT and organisational buy-in and support.

Learning with, and from, other children with CF

Learning together with other children with CF in groups online was a desire of almost all of the children, with the majority of children having never met anyone else with CF. This would provide an opportunity to talk to other children with CF, and ask questions they wouldn't ask people without CF. A parent shared this insight:

'We had this conversation the other night... he was saying, 'oh, it's rubbish having to take tablets every day, it's rubbish having to do physio every day, it's rubbish having to think about taking Creon when you have a snack, you have to think about everything', and I said, 'oh, I know, we understand'. He said, 'yes, you and Dad say you understand but you can't, really understand'. I said, 'no, I get that, we can only try and understand', and we were having that conversation about possibly speaking to other people in the same situation' (Parent, P17).

Learning from other children with CF - peer-to-peer, was also sought e.g., to share explanations of how things make sense to them and what works well for them. From their descriptions, it appeared children would be more likely to take information on, from someone who knows exactly what it is like, who has personal lived experience of CF. For example:

'As a child, it's sometimes good to have another child (with CF) explaining it, not just adults (without CF) going on about it' (Child, C17, boy, 11 years).

The increased and widespread use of online platforms, including for school and as part of CF care, as a result of COVID-19, would greatly facilitate this:

'(Having group sessions was) something we'd just sort of written off before the pandemic because we thought, we can't get people together, it's too difficult. I think we thought they wouldn't tolerate online stuff, but of course everybody's tolerating online stuff all the time and it's getting better and better' (Dietitian, D08).

Only one child reported not wanting to join an online group. This was because he didn't want to make a friend that he couldn't then meet in-person.

An example of peer-to-peer learning suggested by a child and liked by other children in subsequent interviews, was as follows. A short series of YouTube videos could be prepared, in which different children share how they explain to peers about why they need to take PERT. A child at the point of working this out, could then watch the videos and select the explanation that makes sense for them and use this themselves.

Similarly, a parent's suggestion of being able to access a video reference library was keenly taken up by other children and parents in later interviews; it added to a previous idea of sharing meal and snack suggestions, food swaps and recipe ideas in a recipe bank, with children posting videos of them cooking recipes to camera. Dietitians also liked this suggestion, for example:

'Now, more than ever, our parents and our kids with CF need more guidance on what they should be eating. Whereas, in the past, you know, we've just said, eat high fat, eat what you want, blah, blah, blah, now the messages are slightly changing... and they are asking us for examples of what a healthy diet looks like for a child with CF. Literally, this is what the plates should look like. And I think that that would be a perfect form of education to come online, peer to peer' (Dietitian, D12).

There were also ideas for facilitated online group activities including cook-alongs and group games. For example:

'Working out (maths) games, be like a detective solving a mystery or special agents on an important mission ... have a story in which you get to choose what happens' (Child, C16, boy, 8 years).

Other ideas suggested by children and parents included treasure hunts, quizzes, a virtual trip to their favourite restaurant (what's on the menu?) or to the supermarket, or a creative session where they invent some smoothies. In addition, being able to relate to and be inspired by someone with CF was seen as important, though there currently appears to be a dearth of role models for young girls, for example:

'Our kids need some positive role models; they need someone to look up to... There's Ben Mudge, he's amazing and he might be one to give some of the young boys a bit of confidence, because he was a scrawny little squirt and now, he's a Thor impersonator... but there's no one for our little girls to look up to' (Parent, P14).

'A lot of how they learn is through knowing what other people have said. You know, you do have these, kind of, icons and they say, oh this person...he does all this climbing up mountains and he does all this weightlifting and he's never had a Scandishake (an oral nutritional supplement) in his life and this is the diet he follows and...you know, sometimes we just go along with that because I think, well great, that person's really inspiring you and role modelling is so important in children' (Dietitian, D09).

Along with role models, children and parents also appealed for more positivity, as part of raising an overall awareness about CF. This was summed up by a young girl as:

'People who don't have CF, they say, 'oh, they (people with CF) have got a really hard life', but we don't, we've just got to get on with it' (Child, C14, girl, 8 years).

Several of the findings outlined are explored further in phase three (Chapter 7). Findings important in answering the third and final question of phase two will now be described.

6.2.3.3 What outcomes would be most important to achieve?

Of the phase two study questions, this was the more difficult question to answer. As delivery of SCS is currently at best, ad-hoc, asking children, parents and dietitians what they would like the results of SCS to be, required free and creative thinking. As shown in the outcomes category of the thematic framework (Table 6.3), it was not possible for children and parents to describe their preferred short-term outcomes, however, those expressed by dietitians directly align with being child-centred and adopting a more proactive approach.

When asking about preferred long-term outcomes, the researcher defined 'long-term' as the point at which children transition from primary to secondary school i.e., to coincide with the end point of the SCS intervention. This was more tangible, with all participants able to express their preference. Several of the reported outcomes overlapped. For example, there was agreement between children, parents and dietitians that key outcomes for children were to be independent and confident with their PERT, be confident to choose foods that are right for them, and have the opportunity to learn with and from other children with CF.

For parents and dietitians, their choice of outcomes included not only outcomes for themselves and children, but also for each other (Table 6.3). Only dietitians spoke of incorporating clinical outcomes routinely measured in routine practice.

Two key outcomes were taken forward as part of the modelling of SCS in phase three (Chapter 7), whilst discussion of phase two findings follows below.

6.3 Discussion of phase two findings

This section presents a detailed discussion of the findings through providing a summary of the main findings and then discussing them in relation to the wider literature.

6.3.1 Introduction

The aim of this qualitative descriptive study was to develop an in-depth understanding and rich description of SCS of diet and the gut, through exploring the perspectives of key stakeholders in the routine dietetic care of children with CF: children with CF and parents/carers who would be receiving SCS and children's CF dietitians who would be delivering SCS. This aim was met through conducting semi-structured interviews with 20 children, 20 parents and 18 dietitians at multiple centres and rigorous analysis of the gathered data using Framework. The findings enabled further development of the theoretical framework initiated in phase one, therefore meeting the objective for phase two.

6.3.2 Summary of main findings

Themes that were key in answering the phase two study questions, that feed forward to inform the phase three modelling (Chapter 7) were as follows.

Essential components of SCS of diet and the gut were:

- Being child-centred
- Adopting an ongoing stepped approach
- Establishing health-promoting behaviours.

Delivery of SCS as part of routine care comprised:

- Low intensity delivery
- Learning with, and from, other children with CF.

Outcomes agreed as important for children to achieve (at the point of transitioning from primary to secondary school) included:

- To be independent and confident with PERT
- To be confident to choose foods that are right for them
- To have the opportunity to learn with, and from, other children with CF.

Discussion of these findings now follows, including how they relate to the wider literature.

6.3.3 Review of the findings in relation to the wider literature

SCS of diet and the gut by dietitians was found to require a change in focus from the current adult-orientated care of families, to care that is more child-centred. In agreement with Ford et al. (2018), this does not diminish the importance of the child's family, but rather argues that children be able to play a more active role in their care (if they wish to do so) as they are growing up. To enable this, it was found that more child-accessible language is needed for children to begin making sense of what is happening. Across the study, suggestions for accessing, understanding and using health-relevant information encompassed the three dimensions of health literacy defined by Nutbeam (2008) and summarised by Harris et al. (2015, p.3) as:

- Functional - the ability to understand written information and numeracy
- Interactive - the ability to communicate health needs and interact to address health issues
- Critical - the ability to assess the quality and relevance of information and advice to one's own situation.

Whilst there has been a narrow focus on promoting children's functional health literacy (Fairbrother et al., 2016), no resources were currently available to assist with numeracy, identified in phase two as well as the IR, as a core self-care skill in CF. There were also no resources addressing children's interactive health literacy (e.g., to enable them to relay explanations to peers about PERT or report observation of GI symptoms to their parents) or critical health literacy (e.g., in comparing the relevance of healthy eating messages at school with what their bodies need to stay well with CF). An added complexity was the abstract nature of much of the content of SCS of diet and the gut. In the study,

learning through a variety of play-based activities and/or visually were key suggestions for overcoming this. Further research is needed to explore how activities/ resources appropriate for age/ developmental stage across middle childhood (6-11 years) can incorporate all three dimensions of health literacy, whether these are more accessible if play-based and/or visual, and what impact their use has on self-care behaviours (Abrams et al., 2009).

A significant barrier to delivery of child-centred care was the current CF service model, particularly the structure/ process of clinic and the culture of clinics. This finding was in agreement with MacDonald et al. (2015), in which the repetitive order and ‘tick box’ nature of CF clinic was observed. The authors also highlighted how a change to patient-led interactions assumes that (adult) patients would be willing to take responsibility for raising issues. This is also relevant to the current study in that proposing a shift to child-centred care assumes the child may be willing/ ready to (i) discuss CF and (ii) begin learning, and gradually taking ownership, of certain self-care tasks. However, child-centred care as proposed here, is at the level of the individual child, with the SCS intervention needing the flexibility to both assess readiness and then tailor support as appropriate to the child’s individual needs and preferences.

To enable this, a ‘road map’ for SCS of diet and the gut was found to be essential. This was also a finding of the IR and is already a requirement for children and young people with diabetes (NICE, 2022). The roadmap could dovetail the expectations set out in advance by CF centres regarding transition to adult services (Skov et al., 2018), many of which begin at 11-12 years and are based on the Ready, Steady, Go transition programme (Connett and Nagra, 2018).

Findings suggested targeting SCS for children throughout pre-adolescence was appropriate, in agreement with the literature that informed the current study (section 1.8). The need for bite-sized content, again mirrored current practice in diabetes (Diabetes UK, 2022). The approach of ‘elicit-provide-elicit’¹ for information-giving, commonly used in motivational interviewing (Duff and Latchford, 2010), also appeared to be integral to stepwise support. Some

¹ asking what they already understand - share some information – and ask again

dietitians reported already using motivational interviewing, however they worked across children's and adult CF; those working only with children may therefore require additional training and practice in applying this technique (Duff and Latchford, 2013).

Having a structured and stepwise curriculum and repeated opportunities to practice over time were also found to be key components of self-management support by Saxby et al. (2019) (discussed in section 1.8). However, the authors highlighted the need to develop practical guidance for HCPs in how to do this. Similarly, guidance is needed on how HCPs can assist in the process of parent-to-child transfer of self-care responsibilities (Nightingale et al., 2019). As these dual functions are proposed for the road map in the current study, further work will be needed to develop this intervention component.

The findings suggested that a complimentary resource for school is needed (despite there already being a resource pack available for primary-school aged children with CF (CF Trust, 2016)). This is in agreement with Gathercole (2017), who found that children's needs can be obscured in the school setting, with teachers having a low awareness and understanding of CF. In a study of direct support to primary-school aged children with T1DM, staff were ideally placed to aid children's increasing confidence and independence with self-management skills (Marshall, 2017). More research is needed to be able to replicate this supportive school environment for children with CF, as repeated performance of daily self-care could facilitate development of habits, ahead of transition to secondary school. Indeed, Gardner (2015) highlights that the unchanging context of e.g., eating a school meal, provides a consistent prompt/ cue for habits (as a cue-response), to remain stable over time. This was highlighted by parents and dietitians in their added detail to the preferred outcome for children, i.e., to be independent and confident with PERT, *when outside the usual routines*. This referred to maintaining the habit of remembering to take PERT when the prompt/ cue is not present. However, given that children and parents identified a need for provision of prompts/ cues, these may need to be a continuation of what is used in primary school, such as a feature of an app, when they transition to their new secondary school environment, to trigger the response of taking PERT. This also raises the question of whether an additional resource is needed for the start of secondary school, so that staff have greater

awareness and are able to support initiation (and maintenance) of this key self-care behaviour.

With regards to delivery of SCS, clear requirements were that it is flexible and low intensity. Being able to access 'on demand' resources in an online hub was an unanimous request. This would provide opportunity for repeatedly reinforcing learning over time, the importance of which has been demonstrated by Dashiff et al. (2013). In addition, it would provide opportunity for parents to top up their learning. In a study by Bregnballe et al. (2017), parents reported difficulties in answering their child's questions about CF, emphasising the need to re-educate parents as information was not regularly repeated. With the current introduction of HEMT, a 're-education' or at least support for families in changing long-established eating behaviours will be necessary, and was an aspect of SCS referred to by many parents and dietitians in the current study.

Another collective request regarding delivery of SCS was for children to learn with, and from, other children with CF. Most of the children reported having no interaction with other children with CF, but this was wished for. In previous studies, though predominantly with adolescents and young adults with CF, online contact provided social, emotional and practical support (Kirk and Milnes, 2015), reduced experience of isolation (Moola, 2018), and inspired and motivated performance of self-care (Perkins et al., 2021). As the Covid-19 pandemic has accelerated widespread use of digital platforms, perhaps now is an opportune time to further explore children connecting and being able to learn together in facilitated online group activities, as a part of standard care. Further work is needed regarding exactly how, peer support, defined as 'the giving of assistance and encouragement by an individual considered equal' (Dennis, 2003, p.323), can be provided and accessed by pre-adolescent children (Waite-Jones and Swallow, 2018).

With regards to peer-to-peer learning, there are some existing models to draw on. For example, DigiBete, a self-management education platform for children with T1DM, includes videos of children with T1DM and their families sharing their experiences and practical suggestions for managing diabetes well (DigiBete, 2022). There is also CFHealthHub, a self-care education and behaviour change platform for adults with CF, which incorporates peer descriptions of successful self-care ('talking heads' videos) to support behaviour

change (Arden et al., 2021). However, in the CFHealthHub pilot, whilst videos were generally well received, some adults expressed concern about negatively comparing themselves with others who were more or less healthy (Kirkpatrick et al., 2017). In the current study, only one parent expressed this view, though this may not have occurred to the children, as most had no experience of interacting with others with CF and they were well.

Online group sessions could also become a part of standard care for parents, at specific times. For example, when their child is first diagnosed, and prior to starting primary school and secondary school. This could be with parents whose children are the same age/ stage or a little older/ further on, to create a sense of community. Parents sharing experiences may reinforce learning and contribute to increasing confidence (McDonald et al., 2013) e.g., in beginning to hand-over responsibility for certain self-care tasks. As for children, parents having the option of accessing peer support, would need to be organised and facilitated by CF centres (at least initially). This is so that it is not another duty for parents to take on, a finding similarly reported by parents of adolescents regarding app-based peer support (Akre et al., 2020).

6.3.4 Strengths and limitations of phase two

The design and conduct of this qualitative descriptive study was informed by, and benefitted from, extensive PPI. For example, from first reporting their preferred method and mode of data collection, to reviewing study packs for families, through to checking analysis and interpretation of the gathered data. This contributed to the study progressing at each stage, despite the Covid-19 pandemic.

Families who chose to participate may have had an existing interest in diet, and/ or be more open to discussing support. Conducting interviews face-to-face via video call rather than in-person, may have affected the quality of the gathered data. Some authors report a reduced richness/ depth with video calls (Johnson et al., 2021), though others report comparable findings across the two modes (Irani, 2019; Krouwel et al., 2019). The quality may have been reduced where participants felt rushed or were interrupted/ distracted e.g., several parents were cooking dinner, feeding an infant and entertaining boisterous toddlers. Similarly, in the telephone interviews with dietitians, one interview was squeezed in

between clinics, another over lunch, and one whilst the dietitian was walking home from work.

There did not appear to be any difference in the data gathered from children interviewed individually, compared with those interviewed jointly with their parent(s). However, children interviewed individually all chose to have a parent present in the background. The gathered data may have been different if more dads had participated. In many of the interviews, the child and mum referred to the dad playing a large role in caring for the child's diet and gut. Further exploration of the fathers' input is therefore needed, as suggested previously (Swallow et al., 2011). The gathered data may also have differed with the participation of more children diagnosed with CF via meconium ileus (and therefore greater gut involvement in their CF (Sathe and Houwen, 2017)). [This could be the focus of future work, e.g., in the development of a SCS resource, whereby insights are shared from children with moderate to severe gut involvement]. Accounts of what usually happens in routine CF dietetic care/ clinic consultations may have been obscured, as due to Covid-19, some children had not attended clinic in-person for almost a year and found it difficult to remember.

A study strength was the lengthy, but comprehensive and rigorous analysis using Framework. The effect that the researcher's clinical experience in CF may have had on gathering, analysing and interpreting the data, was addressed through critical reflection, supervision, independent review (of transcripts, analysis and interpretation) by supervisors, review of analysis and interpretation by PAG, and presentation of the findings to PAG. The researcher kept a reflexive journal in which personal, professional, and methodological issues were recorded (excerpts shared in section 5.2.5), and these were discussed in supervision.

6.4 Chapter summary

This chapter has presented and discussed the findings for the phase two study questions. These findings feed forward to inform the phase three modelling in the following chapter, and indicate that delivery of the SCS intervention will need to be at (at least) two-levels:

- (i) The SCS intervention targeting learning and behaviours of children
- (ii) An intervention for dietitians to be able to implement the SCS intervention (i.e., targeting the behaviours of dietitians).

Chapter 7

Phase Three: Modelling and Evaluation

7.1 Introduction

This chapter presents phase three of the study, in which findings from phases one and two (presented in chapters three and six respectively) were integrated to develop a model (conceptual framework) of SCS of diet and the gut for children with CF, to meet the objective for phase three. It begins by outlining the process of developing a model and then describes how the findings, as a draft model in the form of an illustration, were evaluated. It concludes with presentation of the revised illustration (to be used in disseminating study findings), and presentation and discussion of the final model (conceptual framework), in the form of a logic model.

7.2 Methods

This section begins by describing application of the COM-B and TDF. This differed to the mapping described in Chapter 4. This was because in the interviews, the researcher asked participants in each stakeholder group, what outcomes of SCS would be most important to achieve. In terms of planning an intervention pathway to the preferred outcome (target behaviours that the SCS intervention will aim to change), the researcher was then able to work backwards from the outcome, to identify what needs to be in the intervention for the outcome (target behaviours) to occur. The stepwise process of this 'behavioural analysis' will now be described, but in summary, it involved:

- Specifying the target behaviours to be changed
- Identifying the key influences on these behaviours (barriers and enablers) (i.e., the context in which these behaviours occur)
- Selecting intervention components (intervention functions)
- Selecting potential BCTs that address the influences, to promote sustained self-care behaviours (Michie et al., 2014).

As discussed in section 6.3, delivery of SCS as part of routine care will need to address target behaviours at multiple levels (child, parent, dietitian and possibly school). However, the modelling and evaluation phase described in this chapter

focuses on the target behaviours of (i) children receiving the SCS intervention, and (ii) dietitians implementing the SCS intervention.

To model these two levels of the intervention, a separate intervention pathway was needed for target behaviours of (i) children, and (ii) dietitians (which could later merge if behaviours were influenced by similar factors/ similar BCTs were chosen).

7.2.1 Behavioural analyses

7.2.1.1 Children

Specifying the target behaviours to be changed

As presented in section 6.2, a key preferred outcome (target behaviour) of SCS reported by all children, and parents and dietitians for children, was to be independent with PERT at the point of transitioning from primary to secondary school. This was therefore chosen as a specific target behaviour, however, this target behaviour is not one behaviour, but rather a set of behaviours. As children may find some behaviours more difficult than others and each behaviour may be influenced by different factors (different barriers and enablers for each), alternative strategies may be needed to help children learn how to e.g., estimate the fat content of foods versus relay explanations to peers.

Identifying the key influences on these behaviours

Data regarding the set of behaviours and their influences (barriers and enablers) were extracted from the full dataset of phase two and are listed in Table 7.1 under the heading of 'text description', and mapped onto the corresponding COM-B components and TDF domains (Table 7.1). This was performed by the researcher and independently reviewed by the researcher's mentor, an expert in behavioural analysis.

Table 7.1: Behavioural analysis for the target behaviour of children with CF being independent with PERT

Text description	COM-B components and TDF domains														Intervention functions	Possible BCTs (numbers as they appear in BCTTv1 (Michie et al., 2013))
	C					O		M								
	Phy	Psychological				Phy	Soc	Reflective					Auto			
	S	K	CIS	MA D	BR	EN	SI	P/S ID	B Cap	O	B Con	G/I	R	E		
Build knowledge, understanding, and skills* Includes E : Knowledge of explanations to relay to peers re: why they need to take enzymes	✓	✓									✓				Training Education Modelling	5.1 Information about health consequences; 8.7 Graded tasks; 4.1 Instruction on how to perform the behaviour; 6.1 Demonstration of the behaviour; 8.1 Behavioural practice/ rehearsal
B : Forgetting to take enzymes at the right time** E : Having enzymes readily accessible – with them rather than e.g., having to go to the school office				✓		✓									Training Environmental restructuring Enablement	7.1 Prompts/ cues; 12.1 Restructuring the physical environment; 12.5 Adding objects to the environment; 1.4 Action planning; 3.2 Social support (practical)
Self-monitoring GI symptoms, problem solving capability etc.					✓										Education Training Modelling Enablement	2.3 Self-monitoring of behaviour; 1.2 Problem solving; 1.4 Action planning; 8.7 Graded tasks
Time and repeated opportunities to practice and develop self-						✓			✓						Education Training Environmental restructuring Modelling	4.1 Instruction on how to perform the behaviour; 8.1 Behavioural practice/ rehearsal; 8.7 Graded

Text description	COM-B components and TDF domains														Intervention functions	Possible BCTs (numbers as they appear in BCTTv1 (Michie et al., 2013))
	C					O		M								
	Phy	Psychological				Phy	Soc	Reflective					Auto			
	S	K	CIS	MA D	BR	EN	SI	P/S ID	B Cap	O	B Con	G/I	R	E		
confidence and a sense of control															Enablement	tasks; 7.1 Prompts/cues; 6.1 Demonstration of the behaviour; 3.2 Social support (practical); 1.2 Problem solving; 8.3 Habit formation
E: Support from family, friendship groups, peers with and without CF, CF MDT, teachers							✓								Modelling Enablement	3.2 Social support (practical); 3.3 Social support (emotional); 6.1 Demonstration of the behaviour; 9.1 Credible source
B: Issues around stigma and social identity – fear of appearing different, not wanting to take enzymes in front of others E: Feeling empowered							✓	✓						✓	Modelling Enablement	3.2 Social support (practical); 3.3 Social support (emotional); 6.1 Demonstration of the behaviour; 9.1 Credible source
E: Belief it is worth doing – that it is right for them (want and need to do it)									✓	✓	✓	✓			Education Enablement	5.1 Information about health consequences; 1.1 Goal setting (behaviour); 1.4 Action Planning; 2.2 Feedback on behaviour; 2.7 Feedback on

Text description	COM-B components and TDF domains														Intervention functions	Possible BCTs (numbers as they appear in BCTTv1 (Michie et al., 2013))	
	C					O		M									
	Phy	Psychological				Phy	Soc	Reflective					Auto				
	S	K	CIS	MA D	BR	EN	SI	P/S ID	B Cap	O	B Con	G/I	R	E			
E: Having targets and goals around this																	outcome(s) of the behaviour
E: Reinforcement to build routines and habits for taking PERT (reflective → automatic over time)													✓		Training Environmental restructuring	6.1 Demonstration of the behaviour; 8.1 Behavioural practice/rehearsal; 2.3 Self-monitoring of behaviour; 2.2 Feedback on behaviour; 2.7 Feedback on outcome(s) of the behaviour; 10.10 Reward (outcome); 8.3 Habit formation	

Phy: physical

Key for the 14 TDF domains: S: skills; K: knowledge; CIS: cognitive and interpersonal skills; MAD: memory, attention and decision making processes;

BR: behavioural regulation; EN: environmental context and resources; SI: social influences; P/S ID: professional/ social role and identity; B Cap: beliefs about capabilities; O: optimism; B Con: beliefs about consequences; G/I: goals/intentions; R: reinforcement; E: emotion.

B: barrier; E: enabler. * Includes knowledge of *why* enzymes are needed, how enzymes work and how to take them, knowledge of which foods contain fat and how to estimate fat content of foods (e.g., through skill of reading and interpreting food labels) and portion sizes, knowledge and skill of titrating enzyme dose to fat intake; ** From the interviews, this was particularly relevant to the target population.

Selecting intervention functions

Intervention functions are broad categories contained within the BCW which describe how the SCS intervention would aim to change behaviour (Michie et al., 2014). The next step was to select intervention functions that linked to the COM-B components and TDF domains identified as key influences on the set of target behaviours, using the guidance of Michie et al. (2014).

This step and the following step required judgement as to what would be most relevant for the context and was based on the researcher repeatedly returning, and remaining close to, the findings of phase two and the mapping findings of phase one. In addition, the BCW APEASE criteria (Affordability, Practicability, Effectiveness, Acceptability, Side effects, Equity; Michie et al., 2014) were used to assess which intervention functions (and BCTs in the next step) were pragmatic choices based on the resources available. Of these criteria, the most helpful were:

- Practicability e.g., could an intervention function be delivered as part of routine care?
- Acceptability e.g., from the interviews, could an intervention function be considered appropriate across the different stakeholder groups?

Another key factor was accessibility i.e., could an intervention function be accessed by every child for whom it would be relevant or of potential benefit (Michie et al., 2014).

Intervention functions considered possible (Table 7.1) included:

- Education - increasing knowledge or understanding
- Training - developing skills
- Environmental restructuring - changing the physical or social context
- Modelling - providing an example for people to aspire to or imitate
- Enablement - increasing means/ reducing barriers to increase capability (beyond education and training) or opportunity (beyond environmental restructuring).

Selecting BCTs

The next steps were to identify intervention content by selecting which BCTs best served the possible intervention functions and which form of delivery would be appropriate for future implementation of the SCS intervention (Dombrowski et al., 2016).

As outlined in section 2.7, BCTs are the proposed active ingredients (mechanisms of change) within the intervention. Using the 93-item BCT taxonomy BCTTv1 (Michie et al., 2013), a list of BCTs were drawn up (guided by the standardised BCT definitions rather than their labels), for each intervention function. The list was then reduced by considering the most frequently used BCTs (Michie et al., 2014), by using judgement, informed as detailed in the step above and consulting the online Theory and Techniques Tool, which links BCTs with their proposed mechanisms of action (Human Behaviour Change Project, 2018).

Several of the same BCTs were appropriate for different intervention functions, however, a total of 19 BCTs were identified (Table 7.1): 1.1 Goal setting (behaviour), 1.2 Problem solving, 1.4 Action planning, 2.2 Feedback on behaviour, 2.3 Self-monitoring of behaviour, 2.7 Feedback on outcome(s) of the behaviour, 3.2 Social support (practical), 3.3 Social support (emotional), 4.1 Instruction on how to perform the behaviour, 5.1 Information about health consequences, 6.1 Demonstration of the behaviour, 7.1 Prompts/ cues, 8.1 Behavioural practice/ rehearsal, 8.3 Habit formation, 8.7 Graded tasks, 9.1 Credible source, 10.10 reward (outcome), 12.2 Restructuring the physical environment, 12.5 Adding objects to the environment.

Having proposed which BCTs to include in the child-targeted level of the SCS intervention (i.e., the intervention content), the final consideration was the form of intervention (the BCTs) delivery. The TIDieR checklist (Hoffman et al., 2014) and additional elements proposed by Dombrowski et al. (2016), were used to ensure all relevant intervention features were detailed:

- Who delivers: dietitian (trained in intervention delivery and facilitation)
- How: mode of delivery: face-to-face and remote; delivery method: individual, groups (online); delivery route: audio, text, visual, hands-on

- What materials: wide range of resources in an online hub e.g., video library (e.g., animations of how PERT works, filmed demonstrations by children showing how to administer PERT and sharing explanations they relay to peers), recipe bank, games, activities etc.
- Where: face-to-face in hospital outpatient clinic or online at home
- When and how much: low intensity but ongoing, bite-sized contacts over age range of 6-11 years, tailored to individuals (frequency and spacing of contacts and core BCTs for the intervention to be effective need to be determined)
- Delivery style of the intervention: child-centred.

7.2.1.2 Dietitians

The process detailed above for children, was repeated using the same steps, to address a key preferred outcome (target behaviour) of dietitians that would also be required to implement the SCS intervention. This was directly involving and engaging children (in learning to look after their diet and gut). The process is summarised in Table 7.2.

Eleven BCTs were chosen, that could be delivered as follows:

- Who delivers: dietitian
- How: face-to-face and remote group workshops; individual and/ or group follow-up support and supervision (practical discussions and sharing of experiences e.g., in how dietitians are using different resources/ task-based activities to promote and maintain children's engagement), SCS champions could be nominated at specialist CF centres across different regional CF networks in the UK, to act as role models/ mentors for dietitians working at both specialist and shared-care CF centres
- What materials: a 'how to deliver' (the SCS intervention) written guide with accompanying online training module
- Where: if face-to-face, could coincide with regional CF network meetings; if online, could access at the hospital or at home
- When and how much: this would need to be determined, also level of tailoring
- Delivery of the intervention: standardised.

Table 7.2: Behavioural analysis for the target behaviour of dietitians directly involving and engaging children in learning to look after their diet and gut

Text description	COM-B components and TDF domains														Intervention functions	Possible BCTs (numbers as they appear in BCTTv1 (Michie et al., 2013))
	C					O		M								
	Phy	Psychological				Phy	Soc	Reflective					Auto			
	S	K	CIS	MA D	BR	EN	SI	P/S ID	B Cap	O	B Con	G/I	R	E		
Knowledge and understanding of what to do, why, when and how E: A 'roadmap' and access to resources to aid engagement		✓													Education	4.1 Instruction on how to perform the behaviour; 5.3 Information about social and environmental consequences; 7.1 Prompts/ cues
Communication skills (including using child-accessible language), group facilitation skills	✓														Training	4.1 Instruction on how to perform the behaviour; 6.1 Demonstration of the behaviour; 8.1 Behavioural practice/ rehearsal
B: Insufficient staffing, time constraints/ competing demands/ current clinic culture; B: Lack of time/ resources for training						✓									Environmental restructuring Enablement	1.4 Action planning; 12.1 Restructuring the physical environment; 12.2 Restructuring the social environment; 3.2 Social support

Text description	COM-B components and TDF domains														Intervention functions	Possible BCTs (numbers as they appear in BCTTv1 (Michie et al., 2013))
	C					O		M								
	Phy	Psychological				Phy	Soc	Reflective					Auto			
	S	K	CIS	MA D	BR	EN	SI	P/S ID	B Cap	O	B Con	G/I	R	E		
E: Ongoing support/supervision							✓						✓		Enablement Modelling	3.2 Social support (practical); 6.1 Demonstration of the behaviour; 9.1 Credible source
B: Feeling out of their comfort zone; lacking confidence E: Belief they can do it, that it is important/ a valuable part of their professional role and identity								✓	✓					✓	Education Modelling Enablement	6.1 Demonstration of the behaviour; 1.2 problem solving; 2.2 Feedback on behaviour; 2.7 Feedback on outcome(s) of the behaviour

Phy: physical; TDF domains as table above.

7.2.2 Draft model as an illustration

Next, the findings of these behavioural analyses, together with other key findings of phase two, were combined in a draft model for evaluation via online discussion groups (outlined in section 7.2.3 below).

The original plan had been to share the draft model visually, in the form of a logic model. However, as the study progressed, the researcher, with the agreement of supervisors LM and GM, chose to share the key findings visually in the form of an illustration, using the language of interviewees. This was consistent with the qualitative descriptive approach, in that the findings remained close to the data (Neergaard et al., 2009). It sought to increase the accessibility of the findings so that group members, including children, could understand and interpret the findings to offer their feedback, therefore aligning with the study finding of needing to communicate information in a more engaging and child-centred way.

The researcher worked closely with an illustrator, who developed an initial rough 'sketch' to share with the groups. The sketch (as opposed to a fully refined illustration) permitted group members to more easily comment on any changes they felt necessary.

7.2.3 Evaluation

Evaluation was planned through structured feedback sessions with dietitians via a CF Specialist Group meeting and with children and parents at involvement groups via the CF Trust, with the choice of evaluation method and rationale outlined in section 2.7.

7.2.3.1 Dietitians

Unfortunately, evaluation of the draft model with dietitians was delayed due to repeated postponement of the Specialist Group meetings, and is currently outstanding.

7.2.3.2 Children and parents

The convening of two involvement groups online, one for children aged 6-11 years and parents, and one for adolescents over 12 years, was organised by

the involvement team at the CF Trust. Recruitment to the groups to review the study findings was posted on the involvement section of the CF Trust website for six weeks. For those who expressed an interest, the involvement manager emailed the sketch, a study summary and key questions prepared by the researcher, one week before the event. A reminder was also sent one day before, with a link to a jamboard (digital whiteboard) for group members to post ideas and suggestions during the sessions or up to 48 hours later.

Questions centred around the clarity of the model, as a way of understanding the study findings, and particularly whether the findings resonated with group members. The aim of the evaluation was therefore confirmability of findings, to enhance rigour (as discussed in section 2.9), together with exploration of how group members envisaged the model could be used (Baxter et al., 2014).

Questions were as follows:

- Do the findings make sense?
- Based on your experience, are any changes needed? e.g., do you think anything is missing or is there something that could be taken out?
- How would you like to see the findings taken forward?

The feedback received from the involvement groups (outlined in section 7.3.1) informed revisions to the illustration and also development of a logic model.

7.2.4 Logic model for SCS of diet and the gut

Having chosen a logic model approach and described the rationale (Chapter 2, Methodology), the iterative development of the logic model in relation to achieving the outcomes in the behavioural analyses above, will now be outlined.

The researcher began by working backwards from the child-level outcome of the SCS intervention, adding to the intervention pathway the outputs, activities (active ingredients (BCT's)), inputs (intervention resources), and assumptions (the contextual barriers and enablers) identified in the behavioural analysis (Davidoff et al., 2015). Impacts were then added. In this way, the model communicated what the intervention is and how it could work (Funnell and Rogers, 2011). An additional dietitian-level of the intervention was then included to communicate implementation of the intervention as part of routine care, together with barriers and enablers to delivery and uptake. The process was

performed by the researcher following input from an expert advisor on logic models, and independently reviewed by supervisors LM and GM.

7.3 Findings

This section provides an overview of the findings of the evaluation and presents the overall study findings as:

- The revised illustration, that will be used in disseminating study findings
- The logic model, that will be used at future stages in the programme of work.

7.3.1 Evaluation

On the same day that the researcher met with the involvement groups, the CF Trust had a live lunchtime session to discuss the rollout of a new era HEMT for 6-11 year olds. It also launched the 'Research Priorities Refresh' initiative that encouraged the whole CF community, including clinicians and researchers to add their priorities for future CF research (five years on from the JLA PSP). This timing was unfortunate, in that children and parents possibly interested in reviewing the study findings, may have already participated in these events via the CF Trust. Four parents (of 10 who had expressed an interest) and three adolescents (of seven) did attend the involvement group sessions. However, as no children attended, the same information was also shared with three children in the target age range of 6-11 years that attended the specialist CF centre at which the researcher is a clinician.

Overall, the draft (sketch) illustration (Figure 7-1) was well received, with general agreement that the findings resonated with their experiences. The two younger children (both aged eight years), commented that it was a lot to look at and were quite overwhelmed, but they liked that the story of the study findings was visual.

'I like how it's telling the story with pictures and some words' (boy, aged 8 years)

There was a lot of discussion about the language used, particularly whether children understand what is meant by 'diet and gut'. The younger children only understood these terms when their parents had translated them into 'food and

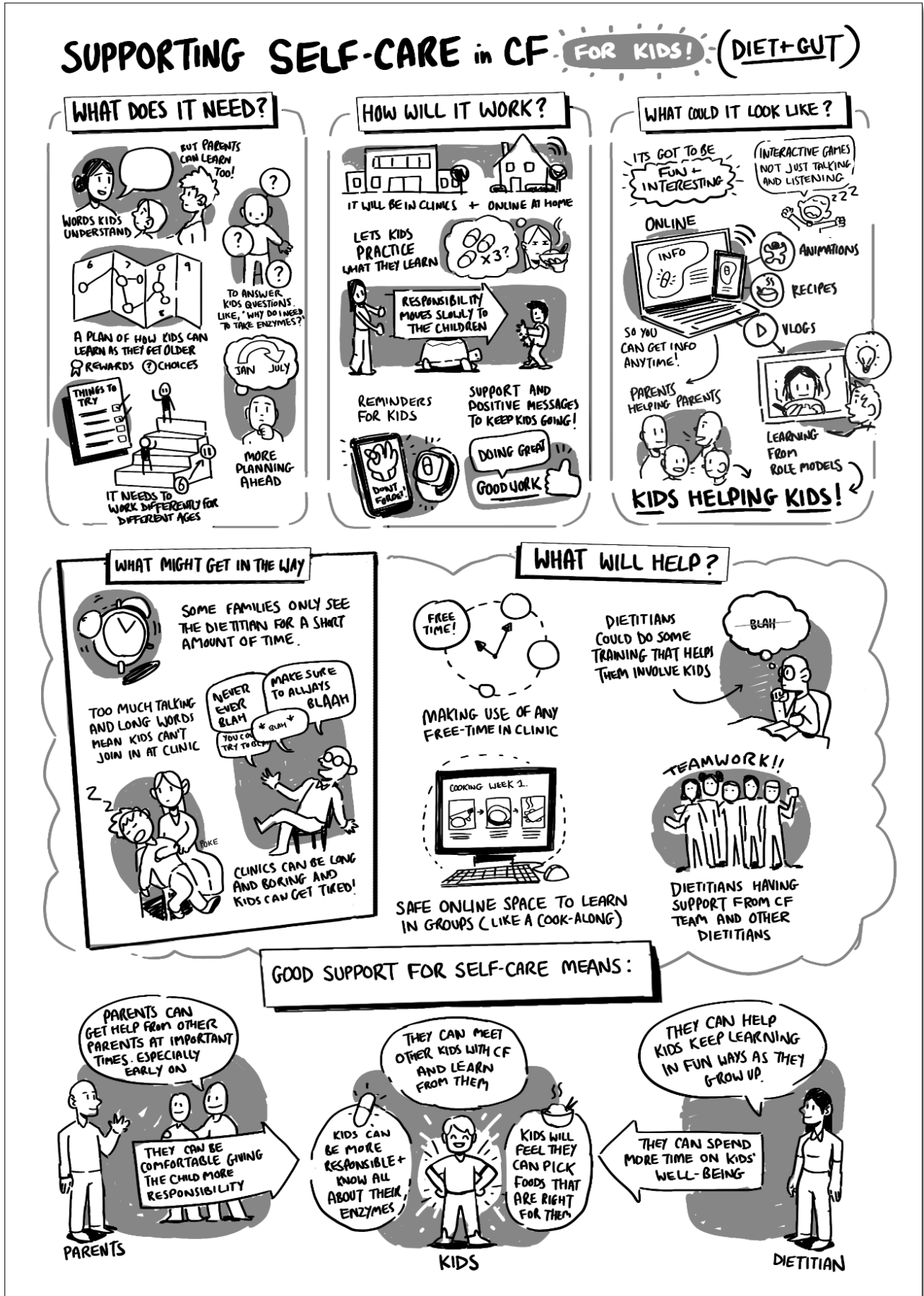
tummy'. When an 11-year-old was asked what she thought was meant by the terms 'self-care', her response was 'looking after yourself'; 'well-being' – 'feeling healthy'; 'gut' – 'my intestines' – 'I learnt about this at school a long time ago' (girl, aged 11 years).

A parent offered the following explanation and suggestion:

'Gut' is rarely used in discussions at home or in clinic, it's more usually referred to as 'tummy' but maybe this is one of the areas that needs exploring through the self-care programme as it's not all about the stomach but more about the gut overall. I think changing to "food and tummy" makes it more accessible to the younger readers but keeping as "diet and gut" seems more appropriate and maybe would just need clarification that the reader understood its meanings' (Parent)

It was agreed that for dissemination of the study findings, an explainer for younger children could accompany the illustration. It was also agreed that a key part of taking the findings forward would be co-developing intervention resources with children in the target age range, so that language would be appropriate for different ages/ developmental stages.

Figure 7-1: Draft model as a sketched illustration



With reference to specific findings represented in the draft illustration, children and adolescents commented on how helpful they had found one of the few, though widely used resource, the animation Oli and Nush (CF Trust, 2010). They remarked that having more information in this format would be beneficial. Just as in the interviews, the importance of also seeing real people, rather than just animated characters, sharing their learning was highlighted (e.g., as top tips), as was the use of humour. Findings that could be more clearly depicted included parents being able to 'top up' their learning, children having prompts to help develop habits, and children learning through play. The 'steps' were liked, though as it was suggested that activities be available in clinic to match the steps, this also needed to be clearer (i.e., that activities/ resources could be incorporated into clinic visits and available online to match the 'roadmap'). Additionally, to spark initial interest in children accessing/ engaging with resources, a parent suggested having fun trailers as screen savers in clinic. 'Planning ahead' and the tortoise signalling slow transfer of responsibility was liked, as was 'positive messages', though this sparked discussion around the need for a change of approach by many dietitians, with less focus on weight and more thought about the child as a whole. One adolescent commented:

'It can be really hard to gain weight, even when you're trying and trying. We don't need more pressure and stress. Dietitians need to relax about it!' (girl, aged 17 years).

A parent also commented that the diet aspect of her daughter's CF had often been medicalised but that eating should be a pleasure, with the dietitian encouraging enjoyment of foods. As it had been in the interviews, it was clear in the evaluation sessions, that a lot of work is needed for dietitians to change their focus from support of self-management towards self-care.

Regarding barriers to SCS represented in the draft illustration, it was suggested that the adults talking about the child rather than directly to the child could be more clearly depicted e.g., over the top of the child's head; and for the enablers of SCS, the dietitian could be shown directly engaging the child and them learning from each other. With respect to outcomes (good SCS means ...) having the child at the centre (happy and glowing) was very much liked,

however, more emphasis was needed on the child taking charge and feeling confident, and for the dietitian to be shown as an 'enabler' rather than 'fixer'.

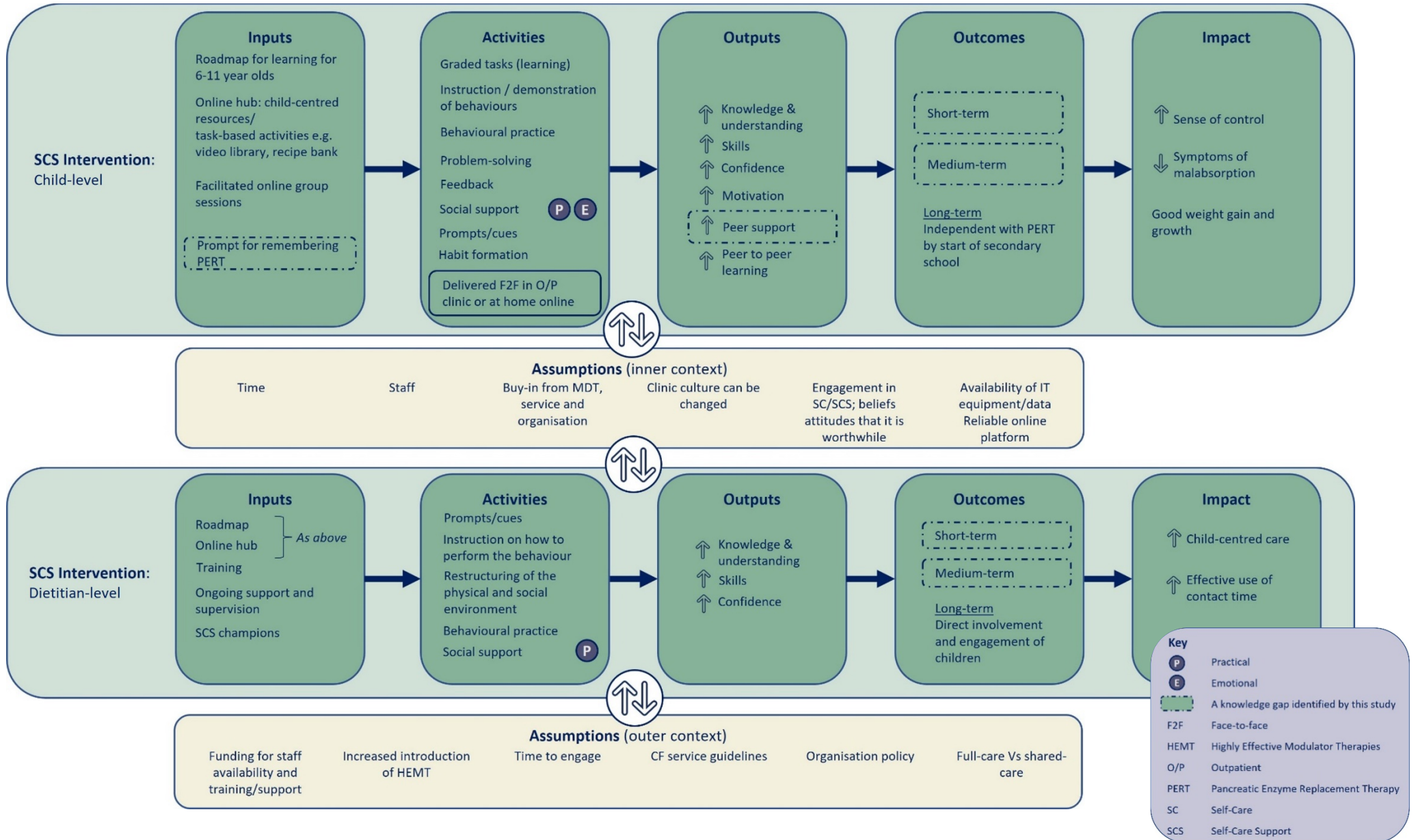
7.3.2 Overall study findings

Based on the feedback, the illustration was revised (Figure 7-2) and a logic model developed (Figure 7-3). In terms of the proposed taxonomy of logic models (Rehfuess et al., 2018), the logic model most closely resembles a systems-based type, as it depicts the interaction between the intervention, implementation and context.

Figure 7-2: Overall study findings for SCS of diet and the gut, as an illustration



Figure 7-3: Logic model for SCS of diet and the gut for children with CF



7.4 Discussion of phase three findings

This section provides a summary of the main findings, a discussion of these findings in relation to the wider literature and consideration of the strengths and limitations of phase three.

7.4.1 Introduction

The aim of phase three was to integrate the findings from phases one and two to develop a model of SCS of diet and the gut for children with CF. This phase sought to build on the theoretical frameworks developed in phases one and two and move to a conceptual framework (as defined in section 1.10), that is, to move from theory to specify what the SCS intervention could be and how it could work in practice. This aim was met through conducting detailed behavioural analyses for two levels of the intervention i.e., to address the outcome/ target behaviour at child and dietitian levels. Based on the feedback from an evaluation of a draft model, the illustration was revised and a final model developed, as a logic model, therefore meeting the objective for phase three and the overall study aim (as stated in section 1.10).

7.4.2 Summary of main findings

The target behaviour of children being independent with PERT was found to be a complex set of behaviours. Mapping these behaviours and their influences, revealed that all components of the COM-B model and 13 of the 14 TDF domains were represented. However, there was overlap in the selected intervention functions and BCTs (active ingredients), and their proposed delivery (summarised in section 7.2.1) included:

- ‘Graded tasks’ via a roadmap for learning (as discussed in section 6.3.3), supported with access to child-centred resources/ activities appropriate for ages 6-11 years/ equivalent developmental stage
- ‘Prompts/cues’, ‘behavioural practice/ rehearsal’ and ‘habit formation’ e.g., introduce a prompt for practice and repetition of remembering to take PERT in the context of eating school lunch; this requires ‘restructuring the physical environment’/‘adding objects to the environment’ in introducing a prompt, but also the child having PERT

with them, as arranged in advance 'action planning' with school staff
'social support (practical)'

- 'Information about health consequences', 'social support (emotional)' e.g., sharing reasons for taking PERT and strategies for reducing feelings of difference, via peer support from other children with CF
- 'Instruction on how to perform behaviours', 'demonstration of behaviours', 'credible source' e.g., children with CF showing how to read and interpret a food label, via a video library (peer-to-peer learning)
- 'Problem solving', 'action planning' e.g., via online group games such as choosing what happens next in a story.

A significant finding was that all seven of the TDF domains within the motivation component of COM-B were represented. This highlighted the importance of the SCS intervention incorporating content to target this, as reflected in the IR finding of attention to the fine detail, with both proactive and responsive tailoring of support to maintain daily self-care.

For the target behaviour of dietitians directly involving and engaging children in learning to look after their diet and gut, mapping this smaller set of behaviours and their influences showed that all components of the COM-B model and eight of the 14 TDF domains were represented. From the selected intervention functions and BCTs, their proposed delivery (summarised in section 7.2.1) included:

- 'Instruction on how to perform behaviours', 'prompts/cues' via a roadmap (what to do, why, when and how), supported with access to child-centred resources as above
- 'Action planning', 'restructuring the physical environment', 'restructuring the social environment', this addressed planning performance of the behaviour through e.g., planning ahead (changing the expectation of a clinic visit), making use of any free time in between other HCPs review, adjusting the seating in the clinic room, having a selection of appropriate resources readily available.

Other BCTs addressed training in communication and group facilitation skills, practice to develop confidence, support and supervision (which included

modelling behaviours), and changing the approach/ focus so that the behaviour becomes inherent to their professional role and identity.

The model proposed that the SCS intervention is a multi-component and multi-level behavioural intervention, that will be hosted on a digital platform. It will consist of a wide range of co-produced resources and activities, that can be accessed at home, but also signposted to, accessed and used face-to-face in bite-sized sessions with a dietitian in outpatient clinic. This will be complemented by children having regular opportunities to learn in online groups that are facilitated by a dietitian. A route for children accessing additional peer support needs to be explored, in a future study. A means of providing prompts/ cues for developing the habit of remembering to take PERT is also needed (as there is currently nothing available). This could be via a mobile app, through which hub resources could also be accessed (and features added to the app to aid self-regulation, such as self-monitoring, and positive reinforcement, such as rewards to affirm their efforts/ achievement).

7.4.3 Review of the findings in relation to the wider literature

There is little evidence regarding which BCTs (active ingredients) are most important in modifying diet-related health behaviours in interventions targeting children. However, Brannon and Cushing (2015) suggest modelling, prompting practice and social support to be the most effective, which is consistent with the study findings. In a cross-sectional study of the BCTs used in 105 nutrition-themed mobile apps intended for children aged 12 years and under, the most common BCTs identified were information about health consequences, followed by instructions on how to perform the behaviour (Brown et al., 2022). This is also in agreement with the study findings, as were the BCTs used in a game-based nutrition education app 'Foodbot Factory', aimed at 9-12 year olds, in which children self-assessed their knowledge by helping game characters make dietary decisions (Brown et al., 2020). Though these last two examples were studies concerned with apps promoting healthy eating behaviours, they demonstrate that the study findings and model are congruent with the existing small knowledge base regarding targeting diet-related health behaviours in children.

It was not possible to find any examples of studies that had identified which BCTs are most important in interventions aiming to enhance self-care/ self-management of chronic conditions by children. This was not surprising, given that in a large scoping review of self-care/ self-management interventions for adults (n=233), that included nine chronic conditions, BCTs were found to be rarely used (Riegel et al., 2021). The authors of the review strongly recommended that BCTs be used in future SCS intervention research.

Existing evidence on effective digital behavioural interventions for self-care/ self-management of chronic conditions targeting primary school-aged children is scant, and reflects how this is an under-researched population (Brigden et al., 2020). The most promising interventions have targeted overweight or obesity, through using exercise with active video gaming (Azevedo et al., 2021).

DigiBete (described in section 6.3.3) for children with T1DM, is an example of a self-management intervention comprising a digital platform and app, with resources co-developed with children and their families (DigiBete, 2022). The proposed model for the SCS intervention is therefore very similar and there may be much to learn from DigiBete's successful implementation e.g., how they developed their infrastructure with respect to being digital in the NHS and how they manage an ever-evolving platform content. However, relevance in informing development of the SCS intervention is limited, as the DigiBete intervention was not developed with a theoretical basis, its core content and active ingredients have not been specified (to inform replication efforts) and although adopted by the NHS, it has not been evaluated (Julian, 2021).

Referring specifically to CF, the few examples of digital behavioural interventions targeting primary school-aged children all have a physiotherapy focus. For example, Project Fizzyo (Raywood et al., 2020), comprises an app with activity tracker and optional games. It aims to make airway clearance in 6-16 year olds more fun and engaging, to encourage routine and improve adherence (results of this longitudinal cohort study are awaited). In a recent systematic review (Lopez-Liria et al., 2022), people with CF of all ages were found to engage well and increase their exercise through playing active video games. Notably, supervision by physiotherapists, while games were being played by both children and adults, was a key factor in promoting adherence and motivation. This emphasises the importance of dietitians being encouraging

and positive whilst facilitating online group games, which may necessitate training and practice to build confidence.

Considering the evidence base on effective digital behavioural interventions in adults, interventions that were more extensively theory-based, incorporated multiple BCTs and supplementary modes of delivery (e.g., text messages to promote interaction with the intervention, send motivational messages or provide a cue to action) were found to be more effective (Webb et al., 2010). The authors of this large systematic review (n=85), provided this as rationale for future investment in interventions with these characteristics. This specification was also supported by Hekler et al. (2016) and Michie et al. (2017), who highlighted that this clarity is currently limited.

In relation to implementation of interventions, evidence for BCTs important in implementing interventions for children, by HCPs, could not be found. However, there were examples of studies identifying BCTs for HCPs delivering complex interventions to adults with chronic conditions. For example, HCPs implementing an online self-management tool for adults with T1DM starting an insulin pump (Reidy et al., 2020). BCTs corresponded with nine of the 11 BCTs found in the current study and were an exact match for barriers such as lack of time and resources, and lack of confidence. Other studies also identified similar influencing factors e.g., in limiting primary care nurses provision of support for behaviour change and depth of diabetes nutrition education (Westland et al., 2018; Gianfrancesco and Johnson, 2020). Though these studies were with adults, findings corroborate those of the current study and highlight the challenge of integrating interventions into routine care.

7.4.4 Strengths and limitations of phase three

Using the BCW provided a systematic and comprehensive approach for guiding development of the theory- and evidence-based model (Reidy et al., 2020). It enabled stakeholder insights of the potential barriers and enablers influencing self-care/ SCS behaviours to be incorporated, so that selection of intervention functions and specification of BCTs were tailored to the context. Having received training, the researcher found the BCW guidance (Michie et al., 2014) accessible, the terminology largely easy to understand, and with practice, it became quicker to use, though the process from start to finish was lengthy

(Phillips et al., 2015). Use of the APEASE criteria (section 7.2.1) and the online Theory and Techniques Tool (section 7.2.1) assisted the judgement and inference needed when selecting BCTs (Johnstone et al., 2021). However, some BCTs did not quite capture the activity. For example, 'social support' was selected for the intervention function of modelling, though a BCT of 'experience sharing and learning', as proposed by Agbadje et al. (2020), would have been more accurate.

Evaluation of the draft model with involvement groups led to an improved illustration for dissemination of study findings and informed development of the logic model (Abrehart et al., 2021). However, evaluation with dietitians was delayed and will not be conducted until September 2022 at the earliest. A strength of the logic model, is that it communicates the what and how of the SCS intervention, but also highlights the current gaps in knowledge (e.g., short- and medium-term outcomes) that need to be addressed in future research.

7.5 Chapter summary

This chapter has presented phase three, in which theory from phases one and two were integrated and further developed using the BCW, to propose the specifics of a model for practical application. The model contributes new knowledge regarding what a digital behavioural intervention for SCS of diet and the gut for children with CF may consist of, and how it could work. It is novel in directly targeting children aged 6-11 years.

In the following chapter, recommendations are outlined for how the model may inform clinical practice and future research.

Chapter 8

Summary, recommendations and conclusion

8.1 Introduction

This chapter presents a summary and conclusion to the thesis. It reviews the overarching study aim and objectives outlined in chapter one and how these have been met. It summarises key findings of the study, and strengths and limitations of the study as a whole. It then presents the implications that the study findings have for practice and policy, and outlines recommendations for future research. Finally, it presents an overall conclusion to the PhD study.

8.2 Thesis overview

The overarching aim of this study was to develop a theory and evidence-based model (conceptual framework) for a complex intervention: SCS of diet and the gut in the routine care of pre-adolescent children with CF. To achieve this aim, three study objectives were addressed:

- Objective one: development of a theoretical framework for SCS of diet and the gut for children with CF - through conducting an evidence synthesis using a IR methodology (phase one, chapter three)
- Objective two: further development of the theoretical framework by exploring the perspectives of key stakeholders (preadolescent children with CF, parents and dietitians) – through in-depth interviews as part of a qualitative descriptive study (phase two, chapters five and six)
- Objective three: development of a conceptual framework that includes the relationship to outcomes identified as most important by key stakeholders – through modelling (using a behaviour change framework) and evaluation (phase three, chapters four and seven).

Having outlined how the study aim and objectives were met, a summary of the key findings now follow, which contribute to the knowledge base.

8.3 Contribution to knowledge

This three-phase pragmatic study began by drawing on what was already known about SCS of diet and the gut in school-aged children with LTCs that had a diet/ GI-related component of care. Findings of the IR defined what SCS

of diet and the gut consisted of according to the literature. This included stepwise development and application of specific knowledge and skills, and required visual resources and repeated opportunities to practice and develop confidence. It also included practical help with incorporating the demands of self-care into everyday life, for which sustaining motivation was particularly important. HCPs were also found to have roles in directly supporting children, as well as their parents.

The IR identified key characteristics of successful models of SCS and factors important for implementation. Characteristics were starting early, before having to change established behaviours, and keeping going, with regular top ups. Flexibility was required in how it was delivered, though integration of SCS as part of routine care was found to work better than it being an optional extra. The importance of choosing outcomes appropriate for the intervention content was also highlighted, and reflected how most studies in the IR focussed on support of self-management rather than self-care (as defined in section 1.5).

These findings were further explored, and considerably expanded on, with findings from primary qualitative research. In defining what SCS of diet and the gut consisted of according to key stakeholders, there were three main findings. First, a crucial component was the dietitian directly involving and engaging the child in learning to look after their diet and gut. This requires a shift to child-centred care and would require a change to the current structure and culture of the clinic. Second, an ongoing stepped approach, with an accompanying 'roadmap' was found to be essential. This would set out expectations in advance regarding what may happen when, and how, with regards to children's learning, gradual transfer of responsibility for self-care tasks from parents to children, and how the dietitian could enable and facilitate these processes. Third, health promotion/ proactive anticipatory guidance (though currently largely missing), was found to be an essential component, and could facilitate performance of behaviours as routine and habit.

Regarding how SCS could be delivered as part of routine care, flexibility was again found to be important. There was preference for a low intensity delivery as a mix of online at home, mostly in groups, and individually, in bite-sized sessions incorporated into routine outpatient clinic visits. This would be facilitated by having a wide range of resources and activities in an accessible

online hub. Visual resources and learning through play would be important. Key requirements were also the provision of opportunities for peer support and peer-to-peer learning, which are currently absent. Regarding what outcomes stakeholders felt would be most important to achieve, it was only possible to attain preferred long-term outcomes (as defined in section 6.2.3), but unlike outcomes in the IR, these were relevant to SCS.

These sets of findings from phases one and two were further explored through mapping to the BCW. First, to understand which theoretical components were targeted in SCS, and second, to work backwards from two key outcomes (one at the child level and one at the dietitian level), to plan an intervention pathway for these outcomes (target behaviours) to be achieved. The overall study findings are summarised as an illustration in Figure 7-2 and a logic model in Figure 7-3, and propose that SCS of diet and the gut for preadolescent children with CF is a digital behavioural intervention.

8.4 Strengths and limitations

Strengths and limitations have been outlined for each of the three study phases (sections 3.4; 6.3.4; 7.4.4), however, strengths and limitations for the study as a whole are considered here.

A key strength of this study was the systematic approach applied to develop the model. This was guided by the overarching MRC framework (Craig et al., 2006), with addition of elements (as suggested by Bleijenberg et al. 2018), to determine the needs and preferences of key stakeholders, and examine current practice and context for future implementation. It was also informed by an existing behaviour change framework, the BCW (Michie et al., 2014), as suggested by O’Cathain et al. (2019a), which enabled the researcher to propose the key components (active ingredients) of the SCS intervention to achieve stakeholder-valued outcomes. In view of the in-depth qualitative research (conducted as the ‘addition of elements’ to the MRC framework), an alternative guiding framework could have been the person-based approach (Yardley et al., 2015), used in combination with the BCW (as e.g., Band et al., 2017; Arden et al., 2021). A limitation of the BCW is, however, its limited application to date, in the development of interventions directly targeting children (Koripalli et al., 2022). Though the proposed active ingredients of the

intervention were congruent with the current limited evidence base, testing the model will be essential in determining the applicability of the BCW for child behaviours.

Whilst development of the model was comprehensive, and provides a coherent basis for future development and evaluation of a prototype of the SCS intervention, it was time intensive. The interview topic guides could have been based on the TDF and gathered data coded directly to the appropriate domains, to increase the efficiency of the process (as e.g., Alexander et al., 2014). However, this may have limited the open-ended questions and breadth of exploration, with the analysis becoming entirely deductive and potentially missing issues of importance to participants that did not fit within the domains (McGowan et al., 2020). It would also have been a departure from the exploratory qualitative descriptive approach.

The study met several of the methodological recommendations for advancing self-care research proposed by Jaarsma et al. (2021). These included defining self-care, building on previous work, using theory in developing an intervention (for which the authors suggested the MRC framework and/ or the BCW), describing how the intervention may work, and its key components.

A study strength was the exploration of children's perspectives, directly with children with CF in the target age range of 6-11 years, who are an under represented group in the research literature (Blower et al., 2020). Allied to this, exploration of parents and dietitians perspectives sought to understand and integrate both complementary and contradictory needs and suggestions for SCS (Kendall et al., 2009). As such, this study was the first step towards developing a co-produced SCS intervention (as recommended by Kirk et al. (2013) and Bee et al. (2018)), which may increase the likelihood of its adoption and effectiveness (Clarke et al., 2017). The findings may also have wider application in other childhood LTCs with a diet and/ or GI-related component of care and within CF, beyond dietetic care.

The study has benefitted throughout from the support of the PAG, input of YPAG and contributions from children and families attending the specialist CF centre at which the researcher is a clinician. This increased the accessibility of the study for children and ensured development of the model remained relevant to the needs of children with CF and their families. Furthermore, this PPI will be

maintained beyond this PhD study, with both parent members of the PAG and parents from the involvement groups via the CF Trust, wishing to continue their involvement in future work.

The researcher adopted an open, critical and reflexive approach and has sought to increase rigour with transparency in reporting throughout. In addition, all study documents and the anonymised interview transcripts will be archived as a single dataset for future sharing and possible reuse.

8.5 Implications for practice and policy

Incorporating SCS, as additional support, into routine care will require changes to the current CF service model. This has implications for the current practice of some dietitians, with a change in approach to be more child-centred and encouraging, and a change in focus from support of self-management to self-care. This includes increasing the quality of contacts, with more individualised, practical and comprehensive (more in-depth) support. Unfortunately, these same changes were requested previously by parents of children with CF, in a survey on dietetic care (CF Trust, 2010). Survey findings also included requests for more information to be available (including online), particularly regarding PERT, with parents keen to swap recipes and ideas with other parents. Many of the characteristics and much of the content proposed for the SCS intervention are therefore long overdue. Taken together with the introduction of HEMT, and accelerated use of digital platforms, it appears that the need and timeliness for a SCS intervention have converged, and also coincide with plans for expanded provision of digital self-management support tools (NHS England, 2019).

Irrespective of having an intervention, the changes to practice outlined above to incorporate SCS will require more time, and training for some dietitians. This will therefore need the support of policy makers and commissioners of CF services.

8.6 Recommendations for further research

The model could inform co-design and co-production of a prototype of the intervention, with children and parents who would be receiving SCS, and dietitians who would be delivering SCS in routine care. The prototype could then be tested in a mixed methods feasibility study at multiple CF centres, and incorporate economic considerations. This would continue the stakeholder-

centred focus in developing and refining the intervention, assess the feasibility and acceptability of the intervention, with consideration of contextual and implementation factors, and inform whether, and how, to go on to evaluate the intervention.

Whilst key components (active ingredients) of the SCS intervention have been proposed, which specific BCTs or BCT combinations, will be most effective for children, and in the context of delivery as part of routine care, needs to be determined (Michie et al., 2018). The current gaps in knowledge highlighted in the logic model also need to be addressed, as well as exploration of how outcomes of SCS (both process and final) can be appropriately measured.

8.7 Conclusion

To begin addressing the evidence gap between policy advocating SCS and the what and how to deliver SCS of diet and the gut in the routine clinical care of preadolescent children with CF, a three-phase multi-method study was conducted. The output of this study was a theory- and evidence-based model for a complex intervention, co-developed with key stakeholders. The model adds to the current evidence base by suggesting what SCS of diet and the gut consists of (its content and characteristics), how it can be delivered, and how it could work, to achieve stakeholder-valued outcomes. The model is also novel, in proposing the first digital behavioural intervention for SCS of diet and the gut, targeting primary school-aged children with CF.

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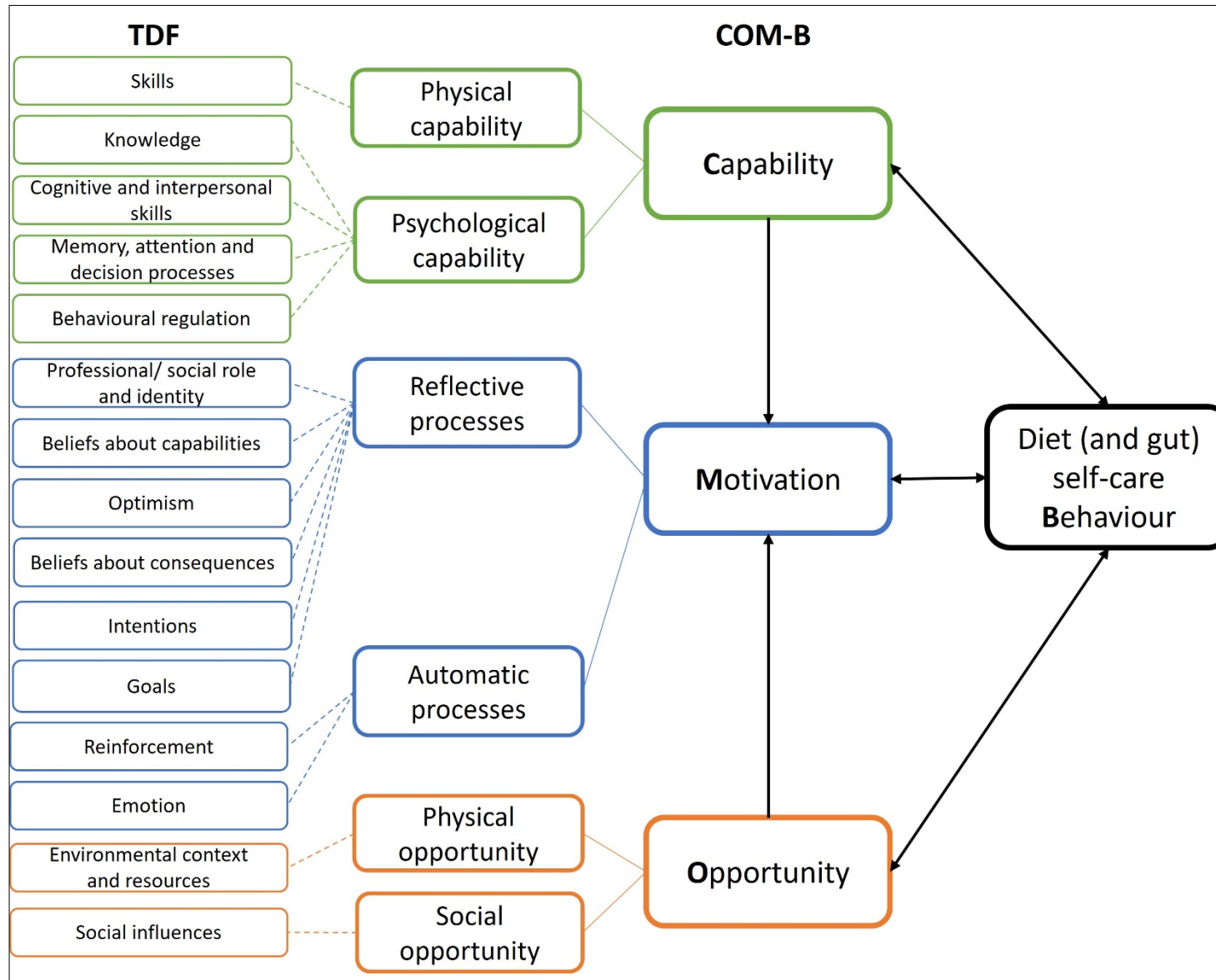
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Appendix A: Links between the COM-B components and TDF domains



Appendix B: Example of the search strategy used in MEDLINE

Published as S2 in Cave et al. (2021).

No.	Searches
1	Child/ or child*.mp.
2	(school age child* or school-age child* or school child*).mp.
3	preadolescen*.mp.
4	Adolescent/ or adolescen*.mp.
5	(teen* or teenager*).mp.
6	youth.mp.
7	(juvenile* or p*ediatric).mp.
8	(young adj (person or people)).mp.
9	((child- adj (parent* or carer* or caregiver*)) or family).mp.
10	Chronic Disease/
11	((chronic* or persist* or ongoing) adj (disease* or condition* or illness*)).mp.
12	((longterm or long term or long-term) adj (disease* or condition* or ill*)).mp.
13	((gastrointestinal or digestive or GI related) adj (disease* or condition* or illness*)).mp.
14	Cystic Fibrosis/ or cystic fibrosis.mp.
15	Diabetes Mellitus, Type 1/
16	Inflammatory Bowel Disease*/
17	Celiac Disease/ or c*eliac disease.mp.
18	Phenylketonuria*/
19	(self care or self-care).mp.
20	Self Care/
21	(self management or self-management).mp.
22	(self help or self-help).mp.
23	(self efficacy or self-efficacy).mp.
24	(collaborati* adj (care or manag*)).mp.

25	((shared or support*) adj (care or manag*)).mp.
26	Diet/ or diet*.mp.
27	nutrition*.mp.
28	gut.mp.
29	((dietary self care or dietary self-care or dietary management) adj support).mp.
30	(coach* or training or program*).mp.
31	(education or skills or information or behavio*r).mp.
32	(engagement or involvement or empowerment or participation).mp
33	(autonom* or responsibility or adapta* or independen*).mp.
34	peer support.mp.
35	(decision* adj (shar* or support* or aid* or making)).mp.
36	(goal set* or problem solving).mp.
37	(plan or action plan* or checklist).mp.
38	1 or 2 or 3 or 4 or 5 or 6 or 7 or 8 or 9
39	10 or 11 or 12 or 13 or 14 or 15 or 16 or 17 or 18
40	19 or 20 or 21 or 22 or 23 or 24 or 25
41	26 or 27 or 28 or 29
42	30 or 31 or 32 or 33 or 34 or 35 or 36 or 37
43	38 and 39 and 40 and 41 and 42
44	limit 43 to (english language and humans and yr="1990-Current")

Appendix C: Characteristics of the studies included in the IR

A version of the table below was published as S3 in Cave et al. (2021). The table below includes the two additional studies in the review update. These are highlighted in grey.

Author (Year), Country	Research aim	Participants (LTC, age, sample size)	Design and methods	SCS Intervention/exposure (including setting)	Key findings
Austin et al. (2013), Canada	Test a model of motivational factors for dietary SC over a 24/12 interval	Adolescents with T1DM aged 11-17 yrs n=289 (n=237 at 24/12 F/U)	Quant descriptive; questionnaires at baseline and 24/12	Routine support/recommended dietary SC programme at 2 DM centres	Autonomy support from HCPs positively predicted autonomous self-regulation and SE, which in turn predicted better dietary SC over 24/12 interval. SE perception was positively related to subsequent parental autonomy support.
Austin et al. (2011), Canada	Examine the relationships between metabolic control, dietary SC and motivation	Adolescents with T1DM aged 11-17 yrs n=289	Quant descriptive; questionnaires at baseline	Routine support/recommended dietary SC programme at 2 DM centres	Overall metabolic control was suboptimal. Better metabolic control was associated with better dietary SC. Interventions should aim to encourage parents to be less controlling. No gender differences.
Bell (2004), UK	Development and evaluation of a nutrition and enzyme education programme for children with CF	Children with CF requiring PERT aged 5-11 yrs and parents	Quant descriptive; questionnaires at baseline and post development	Pen and paper-based 13 module structured education programme; used in O/P clinic and during admissions at n=13 CF centres	Modules used most with 5-10-year-old children - key time as most receptive. When re-evaluated in 2011, UK Dietitians were using individual modules as needed - the 'programme' was never used as it was envisaged.

Author (Year), Country	Research aim	Participants (LTC, age, sample size)	Design and methods	SCS Intervention/ exposure (including setting)	Key findings
Boon et al. (2020), Belgium	Assess effect of a mobile app (MyCyFAPP) for PERT adjustment (+ nutrition education tools and resources) on GI-related QoL in children with CF	Children with CF requiring PERT aged 2-18 yrs n=171	MM; quant non-randomised: 6/12 prospective clinical trial at 6 CF centres in 5 European countries incorporating QoL questionnaires; qual: interviews n=41. No control group.	Use of mobile app for patients and parents (and a professional web tool for HCPs to evaluate patient's data and give feedback) over 6/12	GI-related QoL significantly improved (symptoms reduced), with similar results obtained from patients and parents. Overall median PERT dose did not change. For pts and parents: enzyme dose calculations were used most. For HCPs (particularly dietitians): more precise info obtained prior to consultations allowing for more personalised advice but quality of recorded data decreased over time. The app enables rapid increase in knowledge to implement treatment; regular app usage over a long period is not needed.
Christie et al. (2016), UK	Evaluate effectiveness of a clinic-based structured educational DM Ix (CASCADE) for children and adolescents with poorly controlled T1DM	Children and adolescents with T1DM aged 8-16 yrs Ix n=159, control n=168; parent/carer Ix n=156, control n=169	Quant RCT comparing Ix to usual care at 28 cluster-randomised paediatric DM clinics; 12 and 24/12 F/U post Ix	Manual-based 4 module structured education programme based on behaviour change methods; groups of 3-4 families to attend 1 module/month (2hrs) in clinic over 4/12. Most groups offered during standard clinic times but independent to routine clinic.	No improvement in HbA1c at 12 or 24 months in Ix group; no significant change in other outcomes. Attendance insufficient to demonstrate an Ix effect: only 55/180 (30%) received the full 4 modules. Significant variation in 'usual care' (controls) across sites. Ix may have been more effective if aimed at children with lower HbA1c and earlier in their DM history/soon after diagnosis..

Author (Year), Country	Research aim	Participants (LTC, age, sample size)	Design and methods	SCS Intervention/ exposure (including setting)	Key findings
Coates et al. (2013), UK	Evaluate effectiveness of a structured DM education programme (CHOICE) on glycaemic control and dietary adherence for adolescents with T1DM	Adolescents with T1DM aged 13-19 yrs; Ix n=70, controls n=65	Quant RCT comparing Ix to usual care at 7 hospital sites, parallel design	3-hrly interactive group-based sessions over 4/52 focusing on insulin adjustment to liberate diet and lifestyle; text messaging support at 2, 4 and 5/12; 24/12 F/U post Ix. Clinic setting but delivered independently to regular O/P clinic.	No significant difference in HbA1c between groups at 12/12 (n=57 Ix, 43 control) despite a much more liberal diet in the Ix group but at 24/12 (n=31 Ix, 28 control) HbA1c significantly higher in Ix group. No difference in BMI or hyper and hypoglycaemic episodes. Dietary adherence deteriorated by 12/12 post Ix and in both Ix and controls by 24/12. Single education Ix (of 12hrs) inadequate - need ongoing support/ input following the 4/52 programme ('top up' over time). Also first needs to be delivered shortly after diagnosis.
Connan et al. (2019), Canada	Assess usability of an interactive e-learning module about GFD for children with CD and T1DM and their caregivers	Children with concurrent CD and T1DM mean age 13.5 yrs n=18 and their caregivers n=15	MM; quant descriptive: pre and post knowledge questionnaires, then qual: observation of 60-min usability test of module with think-aloud + semi-structured interviews	Interactive e-learning module on GFD; DM and CD clinics at a single centre	Knowledge test scores increased significantly from pre to post module completion. Pts and caregivers reported module contained lots of useful info for those newly diagnosed with CD, yet patients had been following a GFD for 1-11.7 years. Most participants envisioned module would be best suited for 8-9 yrs and older, and a more game-centred or interactive module would be required for younger children.
Cooper et al. (2018), UK	Evaluate the feasibility of integrating an app (ADNAT App) into UK paediatric DM care	Adolescents with T1DM aged 12-18 yrs n=89 (44 completers and 45 non-completers)	MM; quant non-randomised Ix study at 3 sites; completers (and submitters of ADNAT questionnaires) compared with non-completers (+ survey and focus groups with HCPs)	ADNAT App for 6/12; combines reflective questioning with needs Ax; 6 domains (eating is 1 domain). 3 DM centres in England; participants could choose to complete it at home and/or in clinic.	Large baseline differences in HbA1c and variable rates of change at 6/12; after adjusting for baseline HbA1c and site, completers had a lower post Ix mean HbA1c than non-completers at 6/12. Pt's HbA1c at 6/12 correlated reasonably well with their ADNAT scores.

Author (Year), Country	Research aim	Participants (LTC, age, sample size)	Design and methods	SCS Intervention/ exposure (including setting)	Key findings
Cottrell et al. (1996), USA	Evaluate effectiveness of a self-management training programme for CF	Children with CF aged 8-18 yrs n=20 and their parents (10 children and 10 parents in Ix group and control group)	Quant RCT comparing Ix to usual care at baseline and 6-8/52 post Ix, parallel design; single site	Two 6hr group sessions at CF centre. Manual based. Timeframe between 6-hr sessions not stated. 2/52 self-monitoring and return of records pre and 6-8/52 post Ix.	No significant difference in change in wt, SM behaviours (children and parents), children's adherence or quality of well-being or parents' knowledge between groups at 6-8/52 F/U, though children's knowledge about CF and its management was greater in the Ix group suggesting the Ix could increase children's knowledge in the short-term.
Culhane (2013), USA	Development and evaluation of an in-clinic nutrition education tool for children with CF	Children with CF (all ages) n=205 and their parents	Quant descriptive; baseline and post Ix knowledge questionnaires at a single CF centre	Pre-test of nutrition knowledge followed by education based on identified knowledge deficits. Tailored to age of child. O/P clinic.	Some retention of nutrition knowledge seen at F/U, but annual reinforcement helpful in continuing to educate patients/families. Nutrition checklist continues to be used in practice at annual review. A brief evaluation of BMI trends was conducted 1 year after implementation and an increase in BMI percentiles was found. No further re-evaluation of its use or effectiveness to date.
Davis et al. (2004), USA	Evaluate effectiveness of the STARBRIGHT <i>fitting CF into your everyday life</i> CD-ROM	Children and adolescents with CF; treatment: mean age 13.6 yrs n=25 and wait-list controls: mean age 12.5 yrs n=22	Quant RCT; crossover design: treatment group received Ix, wait-list group served as controls both for time and maturation then received Ix. Pre and post assessment of CF-related knowledge and coping skills using questionnaire and audiotaped vignettes	View CD-ROM for approx. 30 minutes during an O/P clinic visit - 3 modules, 1 on eating	Knowledge improved and small effects found for changes in coping strategies immediately after viewing the CD-ROM in both treatment and wait-list groups. Ix very brief - no F/U so it is not known how long children and adolescents were able to retain this info. Additional studies needed to address whether verbally generated strategies lead to behaviour change and whether the modest improvements are clinically meaningful.

Author (Year), Country	Research aim	Participants (LTC, age, sample size)	Design and methods	SCS Intervention/ exposure (including setting)	Key findings
D'Souza et al. (2021), UK	Evaluate impact of a SM education module on SEREN, a standardised T1DM education programme for children adopted by NHS Wales	Children with T1DM; Ix: mean age 8.4 yrs n=76 and their parents n=80; controls: mean age 8.8 yrs n=56 and their parents n=50	Quant non-randomised Ix study at 12 sites; Ix group (1 yr post-SEREN) compared with control group (1 yr pre-SEREN)	'Diabetes at Diagnosis' standalone module delivered to children newly diagnosed with T1DM. 1-2hr sessions with individual children and families over 6/52 at 12 DM centres in Wales.	Parents scores significantly higher in Ix group for usefulness of resources, ability to manage, coping with diagnosis and understanding of DM; trend towards higher scores for children in Ix group. No difference in children's QoL and no change in overall HbA1c at 6 or 12 months, clinic attendances or hospital admissions between the two groups. Ongoing education needed and a library of group modules subsequently introduced (includes 'moving to year 7'). Children now involved in resource development (only parents for module evaluated).
Fiallo-Scharer et al. (2019), USA	Evaluate impact of an Ix that tailored delivery of T1DM SM resources to families' specific SM barriers	Children with T1DM aged 8-16 yrs n=106 and controls n=108 and their parent(s)	Quant RCT comparing Ix to usual care at 2 sites, parallel design; 12/12 Ix and 12/12 F/U post Ix	PRISM (problem recognition in illness self-management) survey to identify 3 of 5 family SM barriers, received usual care and up to 4 x 75-min tailored SM resource group sessions pre or post routine DM clinic visits	82% of families attended at least half of the group sessions. No overall Ix effect on HbA1c or QoL found; for 13-16yr olds at 1 site, significant improvements in post-Ix HbA1c and mean QoL of parents of 8-12 yr olds found; Ix may benefit specific children, especially those with high baseline HbA1c. Improvements in HbA1c were largely due to resources addressing barriers to motivation to SM.

Author (Year), Country	Research aim	Participants (LTC, age, sample size)	Design and methods	SCS Intervention/ exposure (including setting)	Key findings
Fishman et al. (2018), USA	To create a patient-centred age-appropriate skill acquisition list specific to CD SM in children	Pts with CD mean age 12 yrs n=204 and parents n=155	Quant descriptive; expert group consensus to develop list of tasks specific to CD SM, from which parallel surveys developed for pts and parents re: age at which an 'average child with CD' would master each task	N/A; study included as an enabler of SCS O/P clinic at a single Children's Hospital	The earliest tasks were mastered by a median age of 8 yrs; pts and parents reported similar ages for skill mastery. Many tasks for diet-based SM are mastered at a far younger age compared with general timelines for starting the transition process.
Froisland and Arsand (2015), Norway	Evaluate effect of a mobile-phone-based tool to visualise food intake with regard to empowerment, SE and self-treatment of T1DM	Adolescents with T1DM aged 13-19 yrs n=12	MM; quant non-randomised: pre-post 3/12 pilot of an 1x - 2 smartphone apps - DiaMob app and the Diabetes message system (DMS), consultation midway, then qual: semi-structured interview at end of 3/12	DiaMob app – could use freely but 2 x 3-day recordings were mandatory - took photos of own foods to target evaluation of carbohydrate and insulin dosage, invited to use DMS to send short messages to their providers; consultation midway; 2 O/P clinics	Both apps provided useful support for DM SM. DiaMob promoted visual understanding and more accurate estimation of carbohydrate; adolescents reported improved knowledge and skills, attitudes and self-awareness. DMS - enabled them to take action and be in charge; liked text messages with simple practical advice. HbA1c improved in 7 of the 11 participants who completed the study.
Kyngas et al. (1998), Finland	Explore adolescents' perceptions of how the actions of HCPs, family and friends help or hinder their compliance with T1DM SC	Adolescents with T1DM aged 13-17 yrs n=51	MM: qual (interviews, content analysis) + quant descriptive (questionnaires re: SC compliance with insulin treatment, diet, home monitoring, co-operation with HCPs)	Routine support	Good compliance with SC when: physicians and nurses actions were motivating (ask, listen and take notice of adolescent's opinion; plan SC together), parents motivating or accepting (show interest in them, accept them as they are, provide positive feedback, help solve problems associated with SC and fitting it into everyday life, do not try to take too much control), friends providing silent support and acceptance.

Author (Year), Country	Research aim	Participants (LTC, age, sample size)	Design and methods	SCS Intervention/ exposure (including setting)	Key findings
Meyer and Naveh (2021), Israel	Evaluate effect of a mobile-phone-based tool in promoting GFD SM skills	Adolescents with CD aged 13-18 yrs n=13	Quant descriptive; post Ix online survey via CD social media groups	'Plan My C-Day' app – select actions in three simulation activities to prepare for food-related events (eating out with friends, a meal on holiday, a meal during a school trip)	For over 75% of participants, time to complete the simulation decreased from first to third by an average of 50% indicating ease of use and ease of learning. The wide variety of actions to choose from enabled creation of individually tailored plans. User perceptions of the contribution of the app to users' dietary SM was low and perhaps reflected the experience adolescents already had in these situations (participants diagnosed with CD between 1 and 13 yrs prior to the study).
Nabors et al. (2014), USA	Evaluate children's learning and goal attainment related to change in their SM skills during a T1DM camp	Children with T1DM aged 8-16 yrs n=131 (in 2011) and n=68 (in 2012)	Quant descriptive; cross-sectional surveys: parent report before camp of child goals + child report at end of camp of what they learned	1/52 DM camp in 2011 and 2012; in 2011, parents reported what their child needed to learn + children independently selected goals, in 2012 parent and child shared goal-setting for SM was added.	Children learnt about recognising and managing hypo and hyperglycaemia, eating healthily, improved ability to independently count carbohydrates etc. Boys reported learning more about managing T1DM, whereas girls reported the value of opportunities to express feelings about coping with and managing DM. There was not a strong match between parent goals and children's learning, though joint goal-setting was more successful.
Owen et al. (2013), UK	Evaluate impact of a pilot intensive (joint physiotherapy and) dietetic education Ix on nutrition knowledge and nutritional status in children with CF	Children with moderate to severe CF requiring PERT aged 5-15 yrs n=15	Quant descriptive; baseline and post Ix knowledge questionnaires (+ patient satisfaction survey) at a single CF centre	Nutrition and PERT education; practical activities and 6 individualised teaching sessions of 30-60mins delivered 1-2/12 over 12/12 at home, on ward or in O/P clinic.	Nutrition quiz scores higher post Ix suggesting improvement in knowledge. The majority of children maintained their BMI z-scores within 0.5 of their baseline measurements (note exercise capacity increased). Younger children (5-10yrs) benefited from shorter sessions (20-30mins) and needed regular reiteration of topics to reinforce knowledge. Author suggests targeting 8-11yr olds as most receptive and incorporate sessions into structured O/P clinic visit.

Author (Year), Country	Research aim	Participants (LTC, age, sample size)	Design and methods	SCS Intervention/ exposure (including setting)	Key findings
Price et al. (2016), UK	Determine the effectiveness of the Kids in Control of Food (KICK-OFF) structured intensive education course for T1DM	Adolescents with T1DM aged 11-16 yrs n=198 and controls n=194	Quant RCT with Ax at baseline & F/U at 6/12, 1yr and 2yrs post Ix; cluster-randomised	5-day group education focusing on SM skills for carbohydrate counting and insulin adjustment; usual care and education for control group. 31 DM centres (17 Ix, 15 control), delivered in community setting.	Participation was associated with improved overall QoL within 6/12. No difference in HbA1c between control and Ix groups. At 1 yr and 2yrs post Ix the control group had significantly higher scores for adherence to treatment and at 6/12 and 1 yr, significantly higher SE scores than the Ix group. Authors suggest improved and sustained glycaemic control may require educational interventions delivered from diagnosis as a fundamental component of care and not an 'optional extra'.
Rankin et al. (2018a), UK	Explore barriers and facilitators for taking on DM SM tasks	Children with T1DM aged 9-12 yrs n=24	Qual; interviews; thematic	Routine support; 4 DM centres	Barriers to children taking on new SM responsibilities: over-reliance on parents, lacking maths skills to count carbohydrate and determine insulin doses. Motivators/tipping points to taking on SM responsibilities: alleviate burden on parents, spend time with their friends, preparing to start secondary school.
Rankin et al. (2018b), UK	Identify how children can be better supported by friends and peers to undertake DM SM	Children with T1DM aged 9-12 yrs n=24	Qual; interviews; thematic	DM-related support from friends and peers; 4 DM centres	Informing peers (classmates) about T1DM often resulted in unwanted attention; peers struggled to understand T1DM and could be insensitive and unsupportive. Children had mixed views and ambivalent views about receiving support from other children with T1DM. HCPs could consider ways to assist small friendship groups to undertake monitoring and prompting, practical help with SM tasks and normalising roles.

Author (Year), Country	Research aim	Participants (LTC, age, sample size)	Design and methods	SCS Intervention/ exposure (including setting)	Key findings
Revert et al. (2018), France	Evaluate implementation of a therapeutic patient education programme to improve nutritional status of children with CF	Children with CF aged 2-12 yrs n=34 in 2011 and n=44 in 2014	Quant non-randomised; prospective longitudinal cohort at a single CF centre	3yr nutrition education programme for 6-10 and 11-16 yr olds integrated into routine practice alongside intensification of F/U (according to nutritional risk status of individual children)	Nutritional status (median BMI z-scores) improved and mean FEV1 (for children >5 yrs) showed no decline over the 3 yrs. Parents and children acquired skills and autonomy (Note: no details re: skills and no additional information received from authors). The relationship between professionals, pts and parents was strengthened. Programme integrated into routine practice since 2014 and extended to include 1 month -18 yr olds.
Singh et al. (2000), USA	Evaluate effectiveness of an education lx in a camp setting on knowledge, attitudes and beliefs about PKU diet	Adolescent girls with PKU mean age 13 yrs n=13 1st-year campers compared with n=11 in 2nd yr, n=8 yr 3 and n=7 yr 4	Quant non-randomised; pre-post lx questionnaires, 3/7 diet records and blood tests and at F/U at 4, 8 and 12/12	Diet and disease education at a 1/52 metabolic summer camp alongside recreational activities. Study in yr 1 and 3 subsequent yrs of camp.	Short-term effects: significant improvement in metabolic control, knowledge of diet, fewer barriers to complying with PKU diet, less perceived isolation – however, these effects progressively returned to baseline levels over 1yr. The long-term decreased dietary compliance was associated with lack of support, feelings of peer rejection, and increased barriers due to lack of info and availability of Phe-restricted foods.
Sparapani et al. (2017), Brazil	Identify children's learning needs about T1DM, their self-care tasks and their video game preferences	Children with T1DM aged 7-12 yrs n=19	Qual; focus groups (7-9 yrs and 10-12 yrs); content analysis	Routine DM education at 1 DM centre; lx development (video game)	Learning needs: dealing with emotions e.g., around food intake, motivation to eat healthily and be physically active, knowledge of food groups; lack of practical skills to effectively self-care e.g., carbohydrate count, lack of awareness regarding influence of foods on glycaemic control and the function of insulin. Video preferences: to be able to see what happens inside their bodies as a result of T1DM, to learn about what they can and cannot eat and how to easily carbohydrate count.

Author (Year), Country	Research aim	Participants (LTC, age, sample size)	Design and methods	SCS Intervention/exposure (including setting)	Key findings
Spiegel et al. (2012), USA	Determine whether a nutrition education Ix improves carbohydrate counting accuracy and glycaemic control in adolescents with T1DM	Adolescents with T1DM aged 12-18 yrs n=66 who inaccurately count carbohydrates (n=33 Ix, n=33 control group)	Quant RCT comparing Ix to routine care at baseline and 3-4/12 at a single site	Interactive 90-minute nutrition education class with a dietitian/certified DM educator (planned curriculum targeting adolescent's problem areas); completion of 2 x 3-day food records with tel feedback from the dietitian.	At baseline, carbohydrates were under or overestimated. There was no significant difference between the Ix and control groups in carbohydrate counting or HbA1c at 3/12. No association found between duration of carbohydrate counting and accuracy therefore regular re-education needed. Adolescents who collaborated more with their parents had lower HbA1c, therefore continued parental involvement in their DM care needs to be encouraged. More intensive education may be required.
Stapleton (2001), Australia	Development, implementation and evaluation of a nutrition and PERT education and behaviour change programme 'Go and Grow with CF'	Children with CF requiring PERT aged 6-11 yrs n=41 (21 in Ix gp, 20 in control gp) and caregivers of 2-11 yr olds n= 54 (27 in Ix gp, 27 in control gp)	Quant RCT comparing Ix to routine care at baseline, end of Ix and 12/12 post Ix, parallel design; single site	10/52 home-based pen and paper course (children and their caregivers completed weekly exercises each lasting approx. 60 minutes) with introductory and concluding group workshops at hospital and monthly tel calls from dietitian.	Significant improvement in children's knowledge (nutrition and enzymes) at end of Ix, but not at 12/12 F/U. No statistically significant improvements in dietary intake or nutritional status. The apparent lack of a long-term effect of a single exposure to the programme on knowledge suggests that regular, ongoing education and counselling is required by families to reinforce aspects related to the child's stage of development and disease status. Author suggested a preventative approach to nutrition in CF in the form of several behavioural-based programmes throughout the child's life may be advantageous.
Stark et al. (2009), USA	Evaluate efficacy of a behavioural plus nutrition education Ix, 'Be In CHARGE' in children with CF	Children with CF requiring PERT aged 4-12 yrs n=67 and their parent(s)	Quant RCT comparing behavioural plus nutrition education intervention (B+NE) with a nutrition education Ix (NE) at 5 sites, with 5 F/U Ax's up to 24/12 post Ix	7 group sessions over 9/52. Manual-based. Parents and children seen in simultaneous but separate groups. NE component same in NE and B+NE groups. 5 CF centres.	B+NE Ix was more effective than NE at increasing dietary intake and wt over a 9/52 period, however across the 24/12 F/U, both achieved similar outcomes. Authors suggest the dietary info in the NE Ix was very behavioural. [Translated to a 10/52 web-based Ix (instead of face-to-face) targeting parents of children with CF aged 3-10 yrs; trial results awaited].

Author (Year), Country	Research aim	Participants (LTC, age, sample size)	Design and methods	SCS Intervention/ exposure (including setting)	Key findings
Witalis et al. (2017), Poland	Assess patients' and their parents' knowledge and attitudes towards compliance with the PKU diet	Patients with PKU aged 10-19 yrs n=173 (n=140 aged 10-16yrs) and their parents n=110 (+ PKU patients >20 yrs n=45)	Quant descriptive; questionnaires for patients and parents	Routine nutrition education (principles of dietary therapy including individual intakes of Phe and protein, menu planning and Phe, protein and calorie content of foods); 9 metabolic centres	Only 45% of patients knew daily Phe intake recommendations and 27% knew Phe content in the selected foods. Knowledge increased with the child's age, but knowledge was not associated with improved dietary compliance. Consistent, long-term family and individual therapy is required, with practical support for families to promote children's independence in meal selection and positive acceptance and motivation to follow PKU diet.

ADNAT: Adolescent Diabetes Needs Assessment Tool; Ax(s): assessment(s); BMI: body mass index; CASCADE: Child and Adolescent Structured Competencies Approach to Diabetes Education; CD: coeliac disease; CHOICE: Carbohydrate, Insulin, Collaborative Education; CF: cystic fibrosis; DM: diabetes; F/U: follow up; GI: gastrointestinal; GFD: gluten-free diet; HCP(s): health care professional(s); Ix: intervention; MM: mixed-methods; O/P: outpatient; pt(s): patient(s); PERT: pancreatic enzyme replacement therapy; Phe: phenylalanine; PKU: phenylketonuria; QoL: quality of life; Qual: qualitative; Quant: quantitative; RCT: randomised controlled trial; SC: self-care; SE: self-efficacy; SM: self-management; SEREN: Structured Education Reassuring Empowering Nurturing; tel: telephone; T1DM: type 1 diabetes; wt: weight.

Appendix D: Findings of the quality appraisal using MMAT

A version of the table below was published as S4 in Cave et al. (2021). The table below includes the two additional studies in the review update. These are highlighted in grey.

Author (Year)	SQ		1. Qualitative Studies					2. Randomized Controlled Trials					3. Non-randomized studies					4. Quantitative Descriptive Studies					5. Mixed Methods Studies								
	S1	S2	1.1	1.2	1.3	1.4	1.5	2.1	2.2	2.3	2.4	2.5	3.1	3.2	3.3	3.4	3.5	4.1	4.2	4.3	4.4	4.5	5.1	5.2	5.3	5.4	5.5				
Austin et al. (2013)	Y	Y																Y	?	Y	Y	Y									
Austin et al. (2011)	Y	Y																Y	Y	Y	?	Y									
Bell (2004)	N	N																													
Boon et al. (2020)	Y	Y	Y	Y	Y	Y	Y						?	Y	Y	?	Y						Y	Y	Y	?	Y				
Christie et al. (2016)	Y	Y						?	Y	Y	Y	N																			
Coates et al. (2013)	Y	Y						?	Y	N	?	Y																			
Connan et al. (2019)	Y	Y	Y	Y	Y	Y	Y															Y	?	Y	?	Y	?	Y	Y	Y	Y

Key: SQ = screening questions; NA = not applicable; Y = yes; N = no; ? = cannot tell; ?Y = probably yes.

Note: Screening questions and questions for each study design are listed on pages 218 and 219 below.

Author (Year)	SQ		1. Qualitative Studies					2. Randomized Controlled Trials					3. Non-randomized studies					4. Quantitative Descriptive Studies					5. Mixed Methods Studies				
	S1	S2	1.1	1.2	1.3	1.4	1.5	2.1	2.2	2.3	2.4	2.5	3.1	3.2	3.3	3.4	3.5	4.1	4.2	4.3	4.4	4.5	5.1	5.2	5.3	5.4	5.5
Cooper et al. (2018)	Y	Y	Y	Y	Y	Y	Y						?	Y	Y	?	?						?	Y	Y	Y	Y
Cottrell et al. (1996)	Y	Y						?	N	?	?	Y															
Culhane (2013)	?Y	?Y																?Y	?Y	?Y	?Y	NA					
Davis et al. (2004)	Y	Y						?	Y	Y	?	Y															
D'Souza et al. (2021)	Y	Y											Y	?	N	N	?										
Fiallo-Scharer et al. (2019)	Y	Y						?	?	Y	?	?															
Fishman et al. (2018)	Y	Y																Y	Y	Y	?	Y					
Froisland and Arsand (2015)	Y	Y	Y	Y	Y	Y	Y						?Y	?	?	N	?						?	Y	Y	?	?

Key: SQ = screening questions; NA = not applicable; Y = yes; N = no; ? = cannot tell; ?Y = probably yes.

Note: Screening questions and questions for each study design are listed on pages 218 and 219 below.

Author (Year)	SQ		1. Qualitative Studies					2. Randomized Controlled Trials					3. Non-randomized studies					4. Quantitative Descriptive Studies					5. Mixed Methods Studies				
	S1	S2	1.1	1.2	1.3	1.4	1.5	2.1	2.2	2.3	2.4	2.5	3.1	3.2	3.3	3.4	3.5	4.1	4.2	4.3	4.4	4.5	5.1	5.2	5.3	5.4	5.5
Kyngas et al. (1998)	Y	Y	Y	Y	Y	Y	Y											?	?	?	?	?	?	?	?	?	?
Meyer and Naveh (2021)	Y	Y																Y	?	Y	?	Y					
Nabors et al. (2014)	Y	Y																?	?	?	N	?					
Owen et al. (2013)	Y	Y																Y	Y	?	Y	Y					
Price et al. (2016)	Y	Y						Y	Y	Y	Y	Y															
Rankin et al. (2018a)	Y	Y	Y	Y	Y	Y	Y																				
Rankin et al. (2018b)	Y	Y	Y	Y	Y	Y	Y																				
Revert et al. (2018)	Y	Y											Y	Y	Y	?	Y										
Singh et al. (2000)	Y	Y											?	Y	?Y	Y	Y										

Key: SQ = screening questions; NA = not applicable; Y = yes; N = no; ? = cannot tell; ?Y = probably yes.

Note: Screening questions and questions for each study design are listed on pages 218 and 219 below.

Author (Year)	SQ		1. Qualitative Studies					2. Randomized Controlled Trials					3. Non-randomized studies					4. Quantitative Descriptive Studies					5. Mixed Methods Studies				
	S1	S2	1.1	1.2	1.3	1.4	1.5	2.1	2.2	2.3	2.4	2.5	3.1	3.2	3.3	3.4	3.5	4.1	4.2	4.3	4.4	4.5	5.1	5.2	5.3	5.4	5.5
Sparapani et al. (2017)	Y	Y	Y	Y	Y	Y	Y																				
Spiegel et al. (2012)	Y	Y						?	Y	?	N	?															
Stapleton (2001)	Y	Y						?	Y	?	?	Y															
Stark et al. (2009)	Y	Y						N	Y	?Y	N	?															
Witalis et al. (2017)	Y	Y																Y	?Y	?	?	Y					

Key: SQ = screening questions; NA = not applicable; Y = yes; N = no; ? = cannot tell; ?Y = probably yes.

Note: Screening questions and questions for each study design are listed on pages 218 and 219 below.

SCREENING QUESTIONS

S1. Are there clear research questions?

S2. Do the collected data allow to address the research questions?

1. QUALITATIVE STUDIES

1.1. Is the qualitative approach appropriate to answer the research question?

1.2. Are the qualitative data collection methods adequate to address the research question?

1.3. Are the findings adequately derived from the data?

1.4. Is the interpretation of results sufficiently substantiated by data?

1.5. Is there coherence between qualitative data sources, collection, analysis and interpretation?

2. RANDOMIZED CONTROLLED TRIALS

2.1. Is randomization appropriately performed?

2.2. Are the groups comparable at baseline?

2.3. Are there complete outcome data?

2.4. Are outcome assessors blinded to the intervention provided?

2.5. Did the participants adhere to the assigned intervention?

3. NON-RANDOMIZED STUDIES

3.1. Are the participants representative of the target population?

3.2. Are measurements appropriate regarding both the outcome and intervention (or exposure)?

3.3. Are there complete outcome data?

3.4. Are the confounders accounted for in the design and analysis?

3.5. During the study period, is the intervention administered (or exposure occurred) as intended?

4. QUANTITATIVE DESCRIPTIVE STUDIES

4.1. Is the sampling strategy relevant to address the research question?

4.2. Is the sample representative of the target population?

4.3. Are the measurements appropriate?

4.4. Is the risk of nonresponse bias low?

4.5. Is the statistical analysis appropriate to answer the research question?

5. MIXED METHODS STUDIES

- 5.1. Is there an adequate rationale for using a mixed methods design to address the research question?
- 5.2. Are the different components of the study effectively integrated to answer the research question?
- 5.3. Are the outputs of the integration of qualitative and quantitative components adequately interpreted?
- 5.4. Are divergences and inconsistencies between quantitative and qualitative results adequately addressed?
- 5.5. Do the different components of the study adhere to the quality criteria of each tradition of the methods involved?

Appendix E: Examples of participant-facing documentation



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Participant Information Sheet: Parents/ Carers

Study Title: Supporting children with CF in self-care of their diet and gut

We would like to invite you and your child to take part in the above research study. Before you decide whether or not you wish to take part, it is important for you to understand why the research is being done and what it would involve. Please take time to read the following information and discuss it with others if you wish. Please ask if there is anything that is not clear or if you would like more information.

What is the purpose of this study?

This study will explore how dietitians could support primary school children with CF to develop their knowledge, skills and confidence in looking after their diet and gut. They may then be better prepared to self-care and stay in control of this part of their CF during adolescence. This may contribute to them living better, and for longer, as adults.

The purpose of the study is to explore your views about your child receiving ongoing individualised self-care support (SCS) of diet and the gut (digestive system). This includes what you think SCS could be made up of and how it could be provided as part of routine care. The study will run from approx. Sept 2020 to June 2021.

Why have my child and I been asked to take part in this study?

You and your child have been asked to take part because your child has CF, takes enzymes (Creon), is aged 6-11 years and has attended your specialist CF centre for at least one year. This is because we are interested in your experience of what currently happens when you see your dietitian in clinic, including any SCS you already receive. We expect that approx. 20 parents/carers and 20 children with CF will take part in the study (along with approx. 20 dietitians).

What will happen if my child and I take part?

You and your child will each have an interview with Laurie Cave. She is a children's CF Dietitian who is doing the study as part of a PhD at the University of Leeds. Interviews will be face-to-face (in person or via a video call e.g. Skype) or by telephone.

Each interview will last approx. 30-40 minutes. Interviews can take place either in a quiet private room at your CF centre (for example, this could match up with a planned clinic visit) or in your home, at a time convenient for you. [Note travel expenses are available if the interview is at your CF centre].

When preparing for this study, children told us they would want to choose whether their parent/carer stayed with them whilst they were interviewed; parents said it was important to be interviewed without their child so they could speak freely.

The interview will be a bit like a one-sided conversation - you and your child will be asked questions and encouraged to talk in detail about self-care/SCS of diet and the gut. This will include: your experience, understanding, needs and preferences for SCS, what goals matter most to you and what you think might help or get in the way of SCS being provided as part of routine care. For your child's interview, there will be a range of games and activities to play/do if they wish whilst talking.

The interviews will be audio-recorded so they can be written out (transcribed) by a University approved transcriber and the transcripts looked at in detail to answer the study question.

Do my child and I have to take part?

No, taking part is completely voluntary – it is up to you and your child to decide whether or not to take part. If you would both like to take part, before the start of the interview your child will be asked for their assent (agreement to take part) and you will be asked to give your consent for them and for you.

You and your child will be asked to sign two copies of the assent and consent forms and will be given back one copy of each. Please keep these along with this information sheet and your child's information sheet.

What are the possible disadvantages and risks of taking part?

The interview will take up approx. 30-40 minutes of your time.

The topic of SCS is not obviously intrusive or sensitive. Though unlikely, should you or your child become upset during the interview, you will be asked if you wish to pause or end the interview and if you want to discuss any issues raised with a trusted member of your CF team.

What are the possible benefits of taking part?

There are no immediate benefits for those taking part. This is an opportunity to inform and influence development of a model for SCS of diet and the gut. This may directly affect you if it goes on to become part of routine care in the UK.

How will you use information about me and my child?

We will need to use information from you – your name and contact details - for this research study. We will keep this information (your personal data) safe and secure and store it separately to the research data (interview transcripts). All information in the transcripts that could show who you are will be removed (anonymised), and will have a code number instead.

Information will not be shared outside of the study team. However, if you or your child were to disclose information about potential harm/risk to yourself or others, Laurie Cave would need to share the information with the named person responsible for safeguarding at your CF centre.

What are our choices about how our information is used?

You can stop being part of the study at any time, without giving a reason and without it affecting your child's care in any way. If you stop, all of your identifiable data can be withdrawn if you wish. You can find out more about how your personal data is used at: <https://dataprotection.leeds.ac.uk/research-participant-privacy-notice/> and the University's Data Protection Officer can be contacted at dpo@leeds.ac.uk. More general information is available at: www.hra.nhs.uk/patientdataandresearch.

What happens to our data after the study?

Personal data and audio-recordings of interviews will be securely erased within 3 months of the end of the study. Anonymised transcripts and study documentation will be archived at the University of Leeds Research Data Repository (Research Data Leeds) for a minimum of 10 years, so they may be looked at and possibly reused.

We will write our reports in a way that no-one can work out that you took part in the study. All results, including any direct quotations from those taking part in the study, will be anonymised, however, children may choose a made-up name (pseudonym) for themselves. If you wish, you can receive a summary of the study findings or a copy of the published results.

Who is sponsoring this study?

The University of Leeds.

Who has reviewed this study?

All research in the NHS is looked at by an independent group of people - a Research Ethics Committee - to protect your interests. This study has been reviewed and given a favourable opinion by Yorkshire and The Humber - Leeds West Research Ethics Committee on 29/06/20, ethics reference 20/YH/0170.

Contact details

If you are interested in taking part or would like more information about this study, please contact: Laurie Cave, PhD Fellow at the University of Leeds: hc16lac@leeds.ac.uk or 07511 046799. If you have any concerns or wish to make a complaint about the study, please contact Dr Linda Milnes, Associate Professor at the School of Healthcare, University of Leeds at L.J.Milnes@leeds.ac.uk

Thank you for taking the time to read this information sheet

<i>Project title</i>	<i>Document type</i>	<i>Version #</i>	<i>Date</i>
Supporting children with CF in self-care of their diet and gut	Participant Info Sheet: Parents/Carers	1.2	29/06/20

Study ID: 276804

CF Centre ID: XXXXXXXXXX



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Information for Children (6-8 years)

A study about learning to look after your diet and gut

We would like to invite you to take part in a study

What is this study?



We want to find out what primary school children would like to learn about their eating and enzymes (Creon). This is so they can be in charge of this when they go to secondary school.

Why have you asked me to help?

We have asked you to help because you have CF, take enzymes (Creon) and are in primary school.

What will happen if I want to help

You will be interviewed (asked some questions).

This will be done by a dietitian, Laurie Cave, who is the person doing the study.

You will be asked questions like

- What is it like when you visit the dietitian?
- What would be good things to learn and how?



The interview will be at your CF centre or at home. This can be with Laurie visiting or by a video call e.g. via Skype.

There will be games and activities to play/do whilst talking. The interview will be recorded so it can be listened to again.

Study ID: 276804

CF Centre ID:

Do I have to help?

No. It's also OK if you say yes but later change your mind.

What happens at the end of the study?

The findings will be written about. We won't use your real name. This is so no one will know you were in the study, but you can choose a made-up name for yourself. We can send you the findings to read if you like.

What if I have questions?

If there is something you don't understand or would like to ask, you can talk to your parents or dietitian. You or your parents can also contact Laurie Cave, at the University of Leeds: hc16lac@leeds.ac.uk or 07511 046799.



Thank you for reading this information sheet

<i>Project title</i>	<i>Document type</i>	<i>Version #</i>	<i>Date</i>
Supporting children with CF in self-care of their diet and gut	Study Info for Children (6-8yrs)	1.2	29/06/20

Study ID: 276804

CF Centre ID:



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Information for Children (9-11 years)

A study about learning to look after your diet and gut

We would like to invite you to take part in a study

Why are you doing this study?



We want to find out how dietitians could support children to learn how to look after their diet and gut – a little bit at a time as they go through primary school – so they know how to do it themselves when they go to secondary school.

Why have I been asked to take part?

You have been chosen because you have CF, take enzymes (Creon) and are in primary school. We expect about 20 children to take part, as well as about 20 parents and 20 dietitians.

What will I need to do?

Take part in an interview.

This is like having a conversation, but where the researcher, Laurie Cave, will ask you questions and listen to you talking about your experience, views and ideas.

The interview will last about 30 minutes. It can take place at your CF centre or at home, face-to-face with Laurie visiting or by a video call e.g. via Skype.

There will be games and activities to play/do whilst talking.



We will ask you questions about:

- What happens when you see your dietitian in clinic
- What you think would be helpful to learn, and how, so you are ready to do more as you get older e.g. choose foods and know how many enzymes to take

Laurie will record the talking, so it can be written out and looked at to answer the study question.

Do I have to take part?

No - it is your choice whether or not to take part and you can always change your mind.

Is there anything bad about taking part?

It will take about 30 minutes of your time. Sometimes people can get upset when they talk about what they think or how they feel about things. If this were to happen, you could take a rest or stop the interview.

Will the study help me?

The study will not help you right now, but the findings may help us improve future care of children with CF by the dietitian.

What happens at the end of the study?

The findings will be talked and written about. We won't use your real name. This is so no one will know you were in the study, but you can choose a made-up name for yourself. We can send you the findings to read if you like.

What if I have questions?

If there is something you don't understand or would like to ask, you can talk to your parents or dietitian. You or your parents can also contact Laurie Cave at the University of Leeds: hc16lac@leeds.ac.uk or 07511 046799.



Thank you for reading this information sheet

<i>Project title</i>	<i>Document type</i>	<i>Version #</i>	<i>Date</i>
Supporting children with CF in self-care of their diet and gut	Study Info for Children (9-11yrs)	1.2	29/06/20

Study ID: 276804

CF Centre ID: XXXXXXXXXX



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School of Healthcare

Assent (the form that gives your agreement to take part)

A study about learning to look after your diet and gut

Please circle all you agree with:

- | | |
|--|---------|
| I have read (or had read to me) the information about this study | Yes/ No |
| Laurie Cave has explained this study to me | Yes/ No |
| I understand what this study is about | Yes/ No |
| I understand that the interview will be recorded | Yes/ No |
| I have asked all the questions I want | Yes/ No |
| I have had my questions answered in a way I understand | Yes/ No |
| I understand it is OK to stop taking part at any time | Yes/ No |
| I am happy to take part | Yes/ No |

If any answers are 'No' or you don't want to take part, please don't sign your name!

If you **do** want to take part, please write your name and today's date:

Your name: **Date:**

The researcher, who explained this project to you needs to sign:

Name: Laurie Cave **Signature:** **Date:**

Your parent/ guardian will be asked to complete a separate consent form if they are happy for you to take part.

Thank you for your help

<i>Project title</i>	<i>Document type</i>	<i>Version #</i>	<i>Date</i>
Supporting children with CF in self-care of their diet and gut	Assent Form	1.2	29/06/20

Study ID: 276804

CF Centre ID: XXXXXXXXXX

Please record if they are interested or not. If interested, they can contact me, or with their permission, I will contact them to discuss the study further. Please record if they give their permission (by ticking the box) and send me their contact details via nhs.net email.

If they wish to take part, I will then arrange the interviews. The target is to interview up to 7 children and 7 parents/carers at each of the three sites. It may be that we reach this target in response to the first mail out. However, if we do not, a second (and maybe third) mail out will be needed. We can discuss whether this follows on directly from the first 2-month mail out or we take a short break.

A note about the interviews

You'll see from the protocol that children and their parents/carers have several choices to make regarding the interview. For children, interviews will be face-to-face, but can either be in person (at their home or in a quiet private room at their CF centre) or via video call e.g. Skype. The options are the same for parents/carers, but they may also choose a telephone interview if that is more convenient. Children and parents/carers will be interviewed individually, but children can choose if they want their parent/carer to stay with them whilst they are interviewed.

I will offer as much flexibility as possible with the dates and times of interviews, but wherever appropriate, interviews could coincide with a planned clinic visit at their CF centre.

Interviews will last approx. 30-40 minutes and will be audio-recorded. I'll have lots of games and activities for children to play/do (if they would like) whilst talking to me. They will also be invited to take a photo or do a drawing to bring along for the start of the interview.

<i>Project title</i>	<i>Document type</i>	<i>Version #</i>	<i>Date</i>
Supporting children with CF in self-care of their diet and gut	Recruitment Info for Specialist Children's Dietitians	1.1	28/04/20

Appendix G: Interview topic guide for children



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Interview Topic Guide for Children

Study Title: Supporting children with CF in self-care of their diet and gut

Study ID: 276804

Preliminaries

- Names, discuss study referring to child's info sheet, thank child for agreeing to take part
- Remind them it's OK to stop interview at any time or not to answer a particular question. Offer child red and yellow cards - can hold up red card if want to stop and yellow card if want to skip a question
- Check ok with them to audio record the interview. Offer child chance to talk and listen back or the chance to ask questions (be the interviewer) later on
- Give assurance that there are no right or wrong answers. We are just interested to hear about their experiences and what they think and feel about things
- It's OK to say 'don't know' and to say if they don't understand
- Ask if they have any questions.

Tailor the phrasing of questions so they are appropriate for the developmental stage of each child

Warm up

- Invite discussion about a favourite meal
OR the photo they have taken AND/OR the picture they have drawn

Self-care

- Talk through a day - what they do to look after themselves
More specifically - what they do to look after their diet and gut
If appropriate, how this differs when at home and when at school
- What things are most easy/ difficult and why
- Who has helped them learn how to look after their diet and gut
If appropriate, would they like to do more/ same/ less of their own care
- Understanding of 'self-care' and 'self-care support' (SCS)

Routine dietetic care

- Experience of what happens in clinic
- What is helpful and why
- What is not helpful and why
Prompts: who does the talking, how involved are they, can they ask questions
- Interaction with dietitian outside of clinic visits
[note if any aspects described are SCS]

Individual needs for SCS

- What they would like dietitians to help them do
- What they would like to learn (/learn more about)
Prompts: knowledge, skills
If appropriate, when (key times for this learning)

Preferences for SCS

- How they would like to learn
Prompts: target just them or with parent/family member/other;
1:1 face to face or virtually/ virtual group/ self-directed;
paper-based/ visual presentation or interactive/ other;
- Where they would like to learn
Prompts: home/ hospital/ school/ other/ mixture
- What would make SCS good

Goals

- What they would like the **results** of SCS to be – in short and longer term
- Prompts: from adults with CF study – have explanations to share with peers, develop independence, develop self-confidence, improve communication skills with HCPs and parents, peer support
- If appropriate, what they would like the results of SCS to be ultimately

Perceptions of barriers and enablers of SCS in routine dietetic care

- What could get in the way
How to overcome
- What could help
How to make the most of

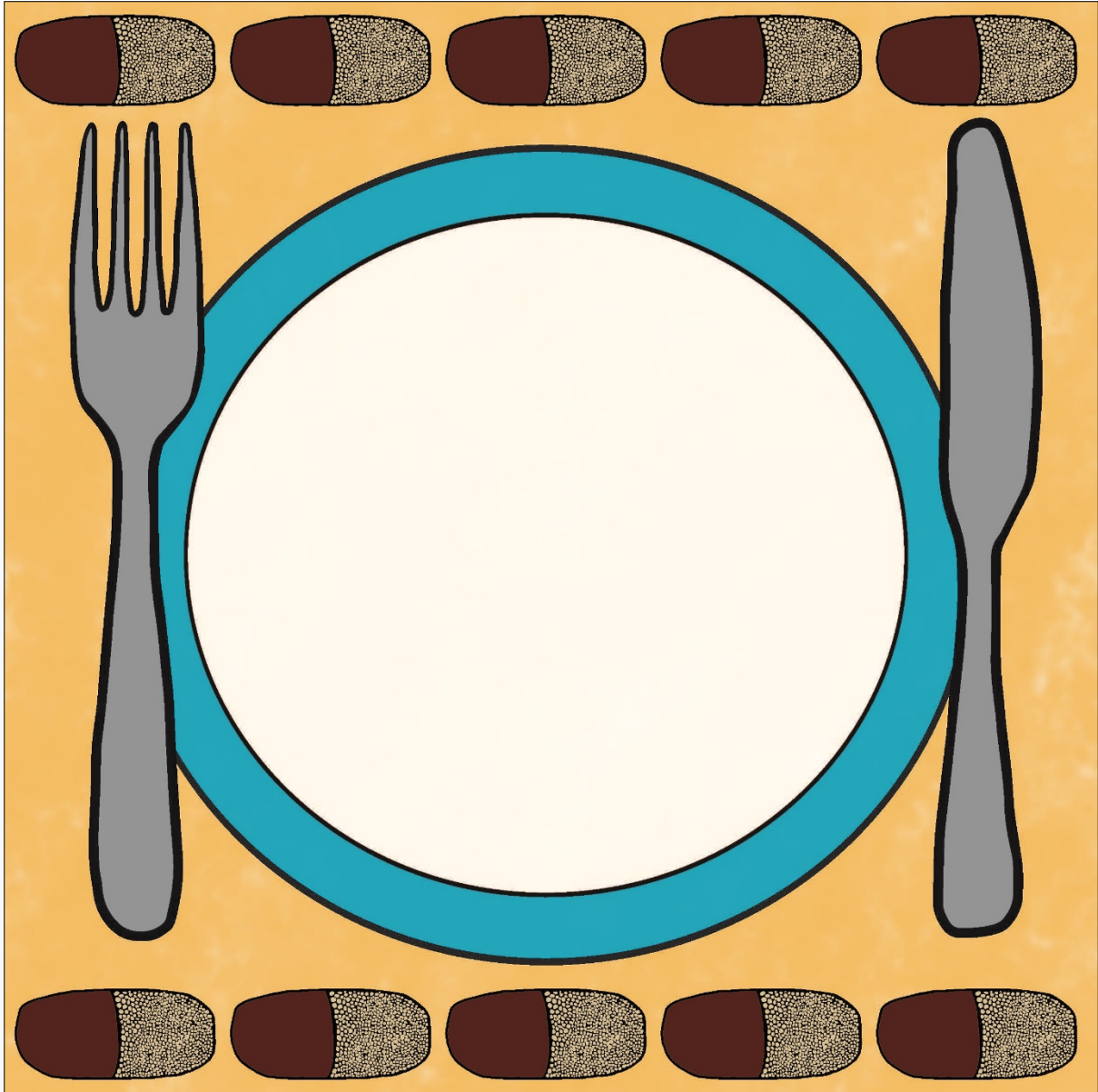
Close

- Anything else they would like to add
- Thank them very much
- Provide info about receiving a summary of study findings
- Would they like to choose a made-up name for themselves and suggest a name for the project (to be voted on by other children and families)?

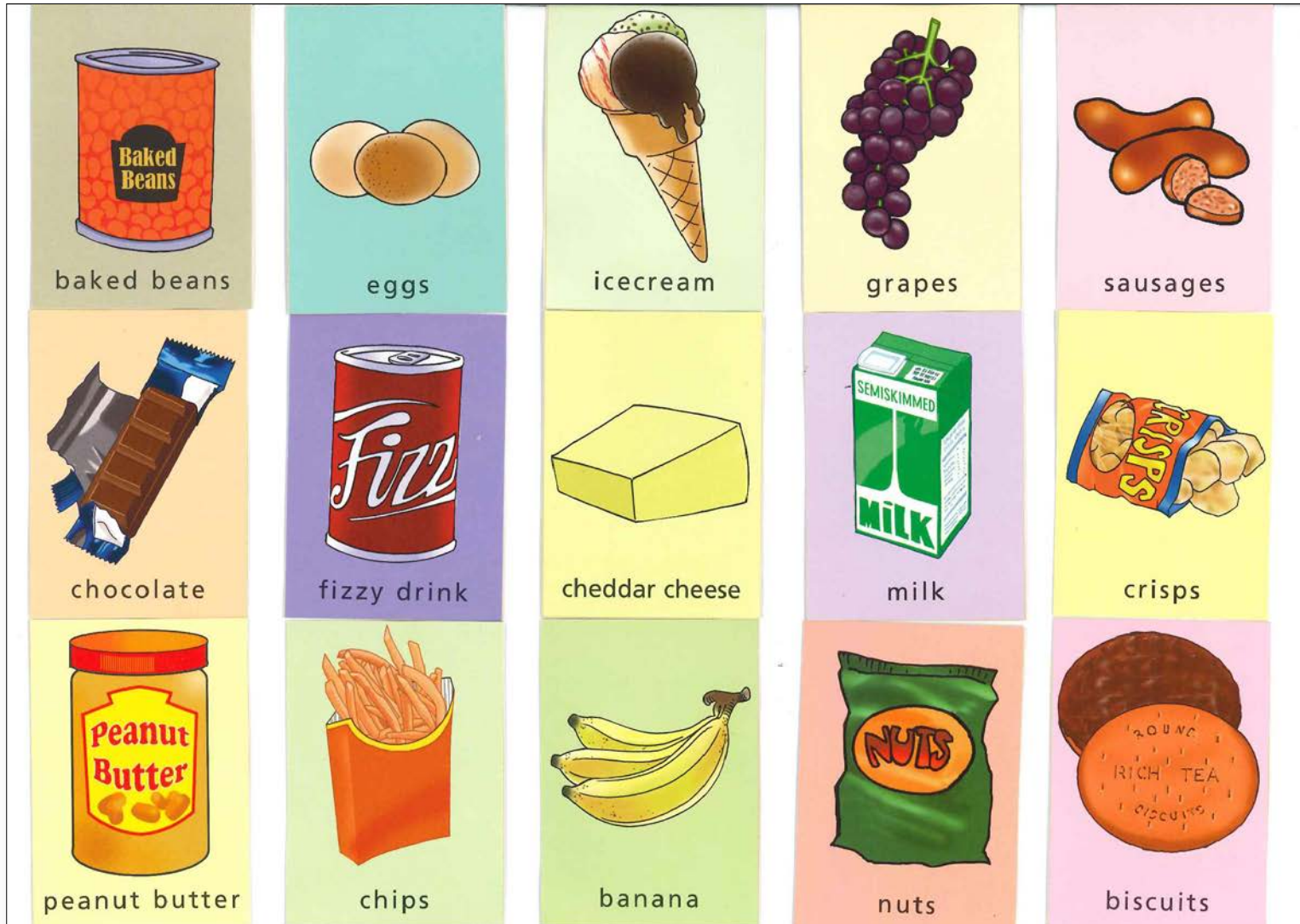
<i>Project title</i>	<i>Document type</i>	<i>Version #</i>	<i>Date</i>
Supporting children with CF in self-care of their diet and gut	Interview topic guide children	1.1	28/04/20

Appendix H: Examples of activities in the children's activity pack

'Empty plate' – children were invited to draw their favourite meal and circle how many Creon capsules they usually take with it. The illustration was shared by an adult with CF, who created 'Benji's Cystic Fibrosis Food Friend' for children with CF aged approximately eight years.



Food sorting game – children were invited to cut out the pictures and sort the foods into three piles: foods that do not need enzymes (Creon), foods that need a little bit of Creon and foods that need a lot of Creon. Food pictures were part of an 'Eatwell Plate' tabletop game produced by Comic Company, copyright permission requested.



Food-themed wordsearches (x4) of increasing difficulty. Copyright permission requested from Buster books.



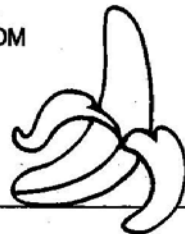
BEGINNER

PUZZLE 1: BREAKFAST

B	E	T	I	U	R	F	M	A	A
C	A	A	E	F	B	U	A	P	H
T	O	K	P	O	S	O	P	A	P
O	T	E	E	H	L	L	S	U	A
A	A	U	R	D	E	H	H	L	S
S	M	O	C	J	B	C	A	S	T
T	O	H	U	R	T	E	T	T	R
M	T	I	O	E	R	C	A	A	I
S	C	W	K	E	G	G	H	N	E
E	N	O	C	A	B	H	O	U	S

APPLE JUICE
BACON
BAKED BEANS
CEREAL
CREPE
EGG
FRUIT

HASH BROWN
KETCHUP
MUSHROOM
PASTRIES
TOAST
TOMATO



INTERMEDIATE

PUZZLE 2: CAKES

B	C	C	S	Y	R	I	A	F	B	T	B
T	C	H	C	U	P	C	A	K	E	R	D
U	S	E	R	E	E	F	F	O	C	A	G
B	P	E	K	I	A	P	P	L	E	T	R
I	I	S	R	L	S	M	A	R	B	L	E
R	E	E	I	O	A	T	B	F	G	L	B
T	C	C	K	D	F	R	M	N	T	E	N
H	C	A	A	A	E	K	I	A	G	W	E
D	L	K	R	G	C	D	C	N	S	E	T
A	E	E	N	R	D	A	O	A	B	K	T
Y	S	I	P	E	O	P	E	W	L	A	A
R	G	C	W	C	S	T	R	T	N	B	B

APPLE
BAKEWELL TART
BATTENBERG
BIRTHDAY
BLACK FOREST
CARROT
CHEESECAKE
CHRISTMAS
COFFEE

CUPCAKE
ECCLES
FAIRY
GINGERBREAD
MARBLE
SPONGE
TEACAKE
UPSIDE-DOWN
WEDDING



ADVANCED



PUZZLE 3 ; WORLD FOODS

A	E	O	J	E	N	I	O	D	E	C	A	M
A	R	E	T	R	U	F	K	N	A	R	F	I
T	L	I	T	U	H	A	C	M	Z	C	D	I
A	G	J	U	J	O	H	L	A	Z	L	R	G
N	A	A	O	A	I	O	A	I	I	U	O	G
O	D	H	G	L	D	K	D	Y	P	B	B	C
R	O	B	A	B	A	G	A	N	O	U	S	H
E	G	D	R	S	A	B	O	I	L	R	A	O
P	A	I	S	S	M	A	J	H	E	R	G	P
E	D	U	O	A	O	T	I	S	N	I	R	S
P	O	M	M	C	F	F	E	U	T	T	O	U
M	A	I	A	O	M	O	F	S	A	O	M	E
S	U	T	U	A	R	K	R	E	U	A	S	Y

- | | | |
|--------------|--------------|-------------|
| BABA GANOUSH | GADO-GADO | POLENTA |
| BHAJI | IMAM BAYILDI | RAGOUT |
| BURRITO | KOFTA | SAMOSA |
| CHOP SUEY | MACEDOINE | SAUERKRAUT |
| DOLMA | MOUSSAKA | SMORGASBORD |
| ENCHILADA | PEPERONATA | SUSHI |
| FEIJOADA | PILAU | TACO |
| FRANKFURTER | PIZZA | |

FIENDISH



PUZZLE 4 ; FRUITS

E	P	G	Q	P	Y	K	N	L	P
C	L	E	M	E	N	T	I	N	E
W	Y	Y	T	S	N	T	L	W	N
E	E	R	M	A	N	G	O	O	I
B	T	R	A	T	N	L	I	D	R
T	E	E	S	S	M	A	O	R	A
W	H	H	T	U	P	T	R	E	T
W	R	C	R	M	A	B	L	G	C
A	L	I	A	A	T	P	E	E	E
N	L	O	W	E	P	R	P	R	N
A	E	A	B	A	P	A	B	N	R
N	O	L	E	M	R	E	T	A	W
A	R	N	R	G	R	P	A	E	P
B	I	Y	R	R	E	B	K	C	A
P	E	E	Y	R	U	W	E	T	A



- | | | |
|------------|-----------|-------------|
| BANANA | KUMQUAT | PEAR |
| BLACKBERRY | LEMON | PINEAPPLE |
| CHERRY | LIME | POMEGRANATE |
| CLEMENTINE | LYCHEE | RASPBERRY |
| DATE | MANGO | SATSUMA |
| ELDERBERRY | NECTARINE | STRAWBERRY |
| GRAPE | ORANGE | WATERMELON |
| KIWI | PEACH | |