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EMPOWERMENT IN CHILDREN WITH CYSTIC FIBROSIS

Section A:

Facilitators of and barriers to empowerment in children and young people with cystic fibrosis: A meta-synthesis of the qualitative literature

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'Thriving alongside CF': Developing A Grounded Theory of Empowerment in Children & Young People with Cystic Fibrosis During Key Life Transitions

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Summary of the Major Research Project

Section A

This is a literature review of patient empowerment in children and young people (CYP) with cystic fibrosis (CF), using a meta-synthesis of 17 qualitative studies. Thematic synthesis identified analytic themes: ‘relational support’, ‘information and understanding’, ‘feeling heard and respected’, ‘mastery and competence’, appeared to facilitate empowerment; ‘prejudice and assumptions’ was identified as a potential barrier. ‘Navigating being different’ appeared to both influence and be influenced by empowerment. Findings provide an initial understanding of factors influencing empowerment in CYP with CF. Potential clinical and research implications are considered.

Section B

This is an empirical paper presenting a grounded theory study that aims to develop a preliminary theory of empowerment in CYP with CF. Seven young people with CF, five parents and four professionals were interviewed. The emerging model suggests that ‘thriving alongside CF’ may be supported by interactions between ‘having a team’ and ‘taking charge and having a voice’, leading to ‘being able to just be a child/getting on with life’. ‘Concealing self’ may get in the way of ‘thriving alongside CF’. These processes occur within wider medical and developmental contexts. Study limitations, clinical and research implications are discussed.

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Part A

Abstract

Background

Health services are increasingly recognising the importance of patient empowerment. Little is known about empowerment in children and young people (CYP) with long term conditions. Recent medical advances have dramatically increased life expectancy of CYP with cystic fibrosis (CF). Patient empowerment may be particularly important in CF due to high treatment burden and limited peer support opportunities.

Aim

To conduct a meta-synthesis of qualitative literature pertaining to empowerment in CYP with CF.

Method

A systematic search of PsycInfo, Medline, CINAHL and ASSIA databases was conducted. Identified studies were critically appraised and data analysed using thematic synthesis.

Results

Seventeen studies met inclusion criteria, though none explicitly explored empowerment. Thematic synthesis identified six analytic themes: **relational support, information and understanding, feeling heard and respected**, and **mastery and competence**, appeared to facilitate empowerment; **prejudices and assumptions** was identified as a potential barrier. **Navigating being different** appeared to both influence and be influenced by empowerment.

Conclusion

Findings provide an initial understanding of factors influencing empowerment in CYP with CF. Potential clinical implications were suggested and the need for further research

highlighted, particularly relating to developmental influences and factors unique to CF which are not adequately addressed in existing patient empowerment models.

Key words: Cystic Fibrosis, Empowerment, Adolescents, Children

Introduction

Empowerment

The concept of empowerment has been used in a wide range of contexts, including community work, education, health and social care (Rowlands, 1995). Rappaport (1984) defined empowerment broadly as “a mechanism by which people, organizations, and communities gain mastery over their affairs” (p.3). In relation to health, the concept has long been applied to health promotion approaches and, more recently as a strategy for the management of long term conditions (LTCs), primarily amongst adult patients (Cerezo, Juvé-Udina, & Delgado-Hito, 2016). However, ‘patient empowerment’ has proved particularly difficult to define, and has been found to overlap with numerous other concepts including self-efficacy, self-determination, self-management and autonomy (e.g. Bravo et al., 2015)

Most literature on patient empowerment has been written from a nursing perspective (e.g. Coyne, 2006; Gibson, 1991), whilst psychological theories of empowerment come largely from community psychology, e.g. Rappaport (1984) and Zimmerman (1995). The current review will use the conceptual definition of patient empowerment employed in the Jørgensen et al. (2018) study which explored facilitators of empowerment in the context of adults who had undergone cancer treatment. This definition was based on Rappaport (1984) and Zimmerman's (1995) work, defining the state of empowerment (a sense of having mastery or being in control) and the process of empowerment (whereby people build motivation, skills and knowledge in order to take ownership of their situation).

Patient empowerment is increasingly being recognised both nationally and internationally as a mechanism for improving health outcomes, health system performance and satisfaction with health care (World Health Organization Regional Office for Europe, 2013). Previous research involving adult patients with a range of LTCs has found that empowering patients increases adherence and promotes positive health outcomes (Prigge, Dietz, Homburg, Hoyer, & Burton, 2015; Yeh, Wu, & Tung, 2018). A number of previous qualitative reviews have explored patient empowerment in the context of specific patient groups, e.g. adults in remission from cancer (Jørgensen et al., 2018) or adults in the advanced stages of a life-limiting illness (Wakefield et al., 2018). Other previous qualitative reviews took a particular focus, e.g. the impact of technology on empowerment across adult patients with a range of LTCs (Calvillo, Román, & Roa, 2015).

Facilitators of patient empowerment in adults have been found to include access to information, feeling respected, having an active role, good communication between patients and healthcare professionals, support from being in a group and religion/spirituality (Bravo et al., 2015; Jørgensen et al., 2018). Many previous reviews focusing on patient empowerment have explicitly excluded children and young people (CYP) (e.g. Bravo et al., 2015).

There is now growing interest in patient empowerment in adolescents and a scale of empowerment has recently been developed for use with CYP with LTCs (Acuña Mora et al., 2018). There is some evidence to suggest a correlation between empowerment and quality of life in CYP with LTCs including diabetes and congenital heart disease (Acuña Mora et al., 2019). However, this was based on models of patient empowerment developed from research with adult patients with a range of LTCs and may not reflect the empowerment experiences of CYP growing up with LTCs.

To the author's knowledge there have been no previous systematic reviews of empowerment in CYP with LTCs. Empowering CYP with LTCs is essential for future engagement and has the potential to impact significantly on long term health outcomes. This is particularly important in cystic fibrosis (CF) due to the significant treatment burden and the inability of people with CF to meet in person to access peer support due to risks of cross-infection (Cystic Fibrosis Trust, 2019).

Cystic Fibrosis

CF is a multisystemic condition with a high treatment burden; daily therapies typically include chest physiotherapy, nebulised medication, pancreatic enzymes, nutritional supplements and antibiotics (Malone et al., 2017). CF is the most common life-limiting genetic condition affecting white populations (Williams & Barker, 2010). Approximately one in every 2,500 babies born in the UK will have CF (Cystic Fibrosis Trust, 2019). Fifty years ago it would be unlikely for a child with CF to live to their 10th birthday (Coulthard, 2018). With universal screening, earlier diagnosis, new understandings and treatments there has been a dramatic increase in life expectancy with over half of people with CF living beyond the age of 47 (Cystic Fibrosis Trust, 2019).

Although people with CF are now living longer they are often affected by significant morbidity (Coulthard, 2018). Improved survival means that people with CF and their families have to cope with the demands of living with the condition longer, and managing increasingly complex regimens (Sawicki et al., 2013). While there is variation in disease

severity and progression, many individuals require frequent inpatient treatment alongside complex home treatment regimens throughout their lives.

Although there is some disagreement within the literature, it appears that the additional psychosocial challenges associated with having CF increase the risk of experiencing psychological distress (Cruz, Marciel, Quittner, & Schechter, 2009). Psychological adjustment and health-related behaviours in CYP with CF have been found to be impacted by different psychosocial factors at different stages of development (Ernst, Johnson, & Stark, 2011). It is likely that factors influencing empowerment will also vary according to age and developmental stage.

Developmental perspectives

Patient empowerment may be more complex in CYP due to the roles played by parents in the management of LTCs in childhood and the influence of peers (Kirk et al., 2013). There are likely to be interactions between CF and all stages of development (Ernst et al., 2011).

Diagnosis often occurs soon after birth during the initial bonding stage which may impact on attachment relationships. During early childhood, issues may arise related to interactions between developing self-regulation and CF regimens. For example developing language abilities, cognitive development in areas such as understanding causality and experimenting with ability to control the environment may lead to battles over mealtimes or completing physiotherapy (Ernst et al., 2011).

The school years are characterized by increasing cognitive ability and increasing emphasis on peer relationships. As cognitive and language skills develop, CYP become more able to communicate their own health-related beliefs and expectations. According to Erikson's psychosocial development model (1959), the key conflict for resolution during this stage is industry versus inferiority. Through processes such as comparing themselves with peers and being recognised by parents and teachers, CYP begin to develop a sense of identity and competence. During this stage 'being different' from peers can be particularly challenging.

Historically CYP with CF benefited from peer support through residential events. This helped to reduce isolation, protected against stigmatisation and increased quality of life through the sharing of adaptive coping strategies. In the mid-1990s, the dangers of cross-infection were recognised, resulting in strict infection control policies, including guidance that patients should not ever meet in person (Ernst et al., 2011).

During adolescence, CYP typically begin to develop a sense of independent identity, spend proportionally more time with peers than family, develop romantic relationships and plan for the future (Erikson, 1959; Taylor, Gibson, & Franck, 2008). For CYP with CF this period of rapid developmental change often coincides with a worsening of CF symptoms such as cough and fatigue, increased frequency of pulmonary exacerbations (episodes of intense disease activity) and associated increases in treatment burden (Hegarty, MacDonald, Watter, & Wilson, 2009). At a time when CYP would otherwise be becoming more independent and spending time with friends, deterioration of their health may cause them to become isolated and more dependent on family, needing to stay at home or being admitted to hospital (Iles & Lowton, 2008).

Summary

Much of the literature on patient empowerment has focused on adults with LTCs. There is growing interest in CYP patient empowerment (Acuña Mora et al., 2018), though much of the existing research has attempted to map understandings of adult patient empowerment onto CYP. This is likely to overlook the potential complexities of patient empowerment in CYP relating to developmental factors and systemic influences such as parents, teachers and peers. Empowerment may be particularly important for CYP with CF due to high treatment burden and potential for isolation.

As little research has been conducted in this area and the review sought to develop a broad understanding of the complexities of the experiences of CYP with CF in relation to empowerment, only qualitative studies were included (Flemming, José Closs, Hughes, & Bennett, 2016).

Aim

To conduct a meta-synthesis of the qualitative literature that elucidates experiences of CYP with CF in relation to empowerment facilitators and barriers.

Method

This review was guided by the ENTREQ (enhancing transparency in reporting the synthesis of qualitative research) framework (Tong, Flemming, McInnes, Oliver, & Craig, 2012),

which includes a checklist designed to improve the quality of reviews using meta-synthesis in the context of health research.

A review protocol was developed prospectively and registered on PROSPERO, an international register of systematic reviews, used by researchers conducting reviews in the field of health. Prospective registration of systematic reviews helps avoid duplication, increases transparency and encourages methodological rigor (Stewart, Moher, & Shekelle, 2012).

Meta-synthesis refers to systematic approaches to reviewing qualitative research. Several methods exist for conducting meta-synthesis (Dixon-Woods et al., 2006). Thematic synthesis (Thomas & Harden, 2008) was selected for this review as it was developed to address questions about people's perspectives and experiences, and consists of a clear set of steps:

- Preparatory phase – searching the literature; screening and assessing the quality of the papers; extracting data from the selected literature.
- Thematic synthesis: a) initial coding of text; b) developing descriptive categories; c) generating analytical themes

In order to promote rigour, a bracketing interview was undertaken to consider how the researcher's prior experiences, biases and assumptions may impact on the thematic synthesis and the NVivo 12 software package was used to maintain a clear audit trail of the development of descriptive and analytical themes.

The rest of this review will use the headings suggested by Thomas and Harden (2008).

Preparing for thematic synthesis

Search strategy

The search strategy was guided by the SPIDER (Sample, Phenomenon of Interest, Design, Evaluation, Research type) framework – a tool developed to facilitate rigour in reviews of qualitative and mixed method research (Cooke, Smith, & Booth, 2012). Two domains were identified to combine within the search strategy:

- Sample – children and young people with cystic fibrosis
- Phenomenon of Interest - patient empowerment

In line with Cochrane guidance (Malone et al., 2017), terms for ‘children and young people 0-19 years’ were not used to limit the search, in order to maximise relevant results. Similarly, a decision was made not to include terms for ‘qualitative research’ as there are numerous qualitative methods that may be described by authors in a multitude of ways (Barroso et al., 2003). Search terms (Table 1) were generated from existing research and theoretical literature relating to patient empowerment (Acuña Mora et al., 2019; Bravo et al., 2015; Jørgensen et al., 2018; Wakefield et al., 2018). Various combinations of concept heading and search terms were trialled before settling on a broad search strategy. PsycInfo, Medline, CINAHL and ASSIA databases were selected for their relevance to the field and searched from inception to March 2019.

Table 1				
<i>Search terms</i>				
<u>Search Terms</u>	<u>PsycInfo</u>	<u>Ovid Medline</u>	<u>CINAHL</u>	<u>ASSIA</u>
Attitude to Health/		X		
CF				X
CF Key Words	X	X		
CF Key Words in abstract			X	X
CF Mesh terms	X	X		
collaborative care			X	X
collaborative care.mp.	X	X		
confidence			X	
confidence.mp.	X			
control.mp.	X			
cystic fibrosis			X	X
cystic fibrosis.mp.	X			
Cystic Fibrosis/px [Psychology]		X		
Decision Making/		X		
Delivery of Health Care, Integrated/		X		
empowerment			X	
Empowerment Key Words	X	X		
Empowerment Key Words in abstract			X	X
Empowerment Mesh terms	X	X		
Empowerment MM			X	
exp "Internal External Locus of Control"/	X			
exp Autonomy/	X			
exp Client Attitudes/	X			
exp Client Education/	X			
exp Client Participation/	X			
exp Cystic Fibrosis/	X			
exp Empowerment/	X			
exp Health Literacy/	X			
exp Interpersonal Control/	X			
exp Self-Care Skills/	X			
exp SELF-CONFIDENCE/	X			
exp Self-Determination/	X			
exp Self-Efficacy/	X			
exp Self-Management/	X			
integrated care			X	X
integrated care.mp.	X	X		
Internal-External Control/		X		
mastery			X	X
mastery.mp.	X	X		
mucoviscidosis			X	X
mucoviscidosis.mp.	X			
patient activation			X	X

patient activation.mp.	X	X		
patient autonomy			X	X
patient autonomy.mp.	X	X		
patient centered care			X	X
patient centered care.mp.	X	X		
patient centred care			X	
patient education			X	X
Patient Education as Topic/		X		
patient education.mp.	X	X		
patient empowerment			X	X
patient empowerment.mp.	X	X		
patient engagement			X	X
patient engagement.mp.	X	X		
patient focused care			X	X
patient focused care.mp.	X	X		
patient involvement			X	X
patient involvement.mp.	X	X		
patient participation				X
patient participation.mp	X	X		
Patient Participation/px [Psychology]		X		
patient preference			X	X
patient preference.mp.	X	X		
Patient Preference/px [Psychology]		X		
Patient-Centered Care/		X		
perceived control			X	X
perceived control.mp	X	X		
Personal Autonomy/		X		
Power (Psychology)/		X		
self advocacy			X	X
self advocacy.mp.	X	X		
self care			X	X
self care.mp.	X	X		
Self Care/px [Psychology]		X		
Self Concept/		X		
self efficacy			X	X
self efficacy.mp.	X	X		
Self Efficacy/		X		
self-advocacy			X	
self-care			X	
self-efficacy			X	
shared care			X	X
shared care.mp.	X	X		
shared decision making			X	X
shared decision making.mp.	X	X		

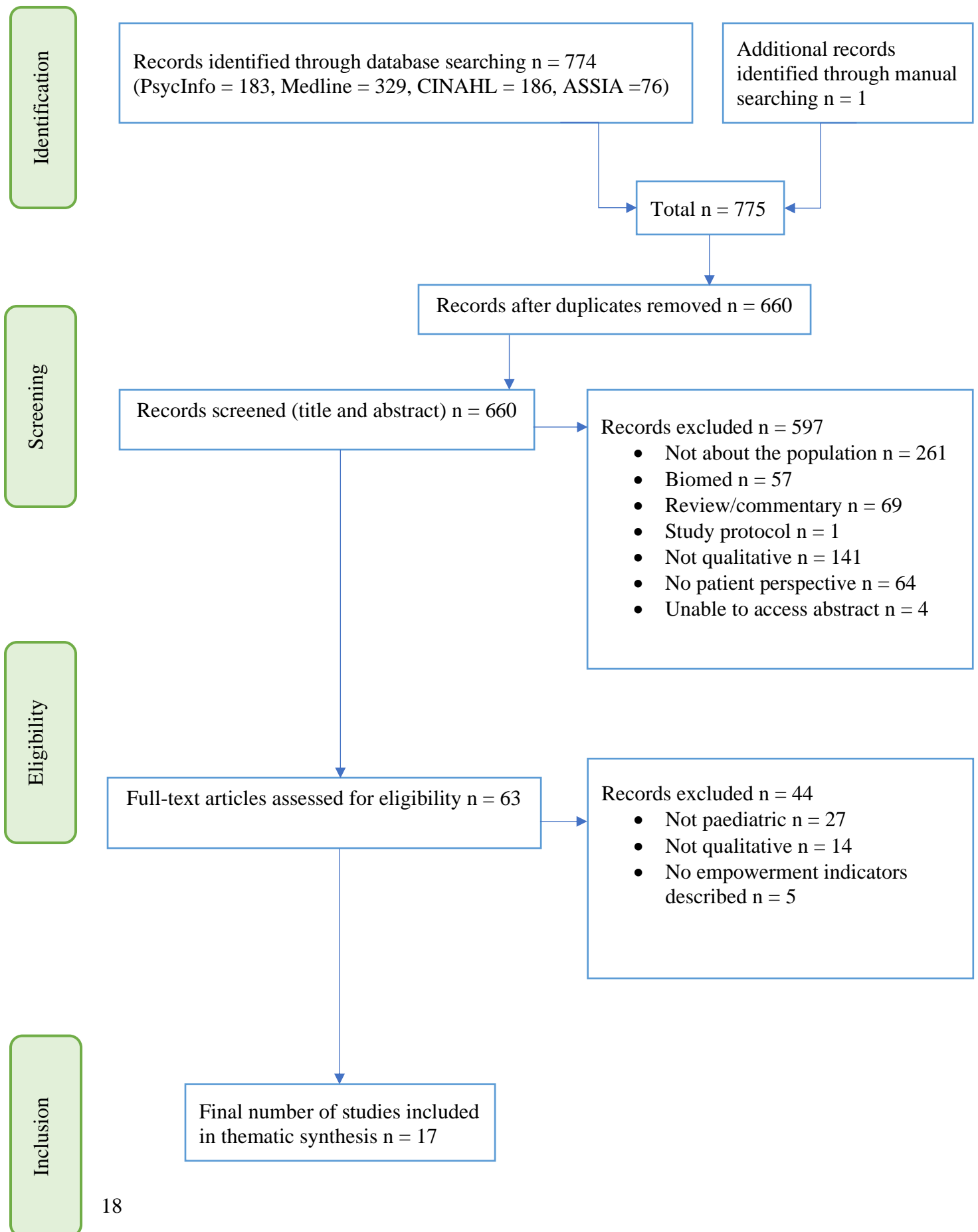
Inclusion criteria

Studies were included where (a) participants were CYP with CF (i.e. ≤ 18 years) or explicitly under the care of paediatric CF services; (b) the participants' perspectives were explored; (c) a qualitative research design was used; and, (d) at least one empowerment indicator was described. Based on the conceptual definition of empowerment described above, these indicators were: feeling in control, having mastery, being in charge, having influence, having agency, or having autonomy. Due to the significant role of family context plays in the lives of CYP, studies that reported perspectives of other family members in addition to the perspectives CYP with CF were included.

Screening

The screening process is shown in Figure 1. Duplicate articles were removed and remaining studies screened for relevance of title and abstract. Where there was uncertainty, full texts were obtained for further review and reasons for exclusion documented.

Figure 1: PRISMA diagram



Quality assessment

Assessing quality in qualitative research is complex. Qualitative methods have a range of different philosophical underpinnings, and are varied and broad. There is much debate as to whether qualitative studies should undergo quality assessment as part of the review process (Carroll, Booth, & Lloyd-Jones, 2012). As there are no widely accepted methods for excluding qualitative studies from reviews on the basis of quality, Thomas and Harden (2008) suggested the solution of sensitivity analysis. In this approach, the possible impact of study quality on the reviews' conclusions are considered after the thematic synthesis by examining the relative contribution of each study to final themes and recommendations.

Study quality was assessed using a tool for appraising qualitative research developed by the Critical Appraisal Skills Programme [CASP] (Public Health Resource Unit, 2006) and the criteria for assessing research conducted with children used by Thomas et al. (2003) (Appendices A-B). Whilst numerous quality assessment tools could have been used, the CASP checklist was selected because it has a strong methodological focus (Leung, 2015) covering clarity and appropriateness of the research question and design, appropriateness of the recruitment strategy, data collection and analysis, issues of reflexivity, ethics, levels of support and evidence for claims and the overall contribution of the research. Additional criteria used by Thomas et al. (2003) were used to supplement the CASP checklist as it was felt important to consider quality issues specific to research with CYP.

Meta-synthesis process

As detailed below, the meta-synthesis followed the steps outlined by Thomas and Harden (2008).

Extracting data from the selected articles

Characteristics of studies meeting the inclusion criteria were entered into a template along with the empowerment indicators described by the study. Following the approach of Thomas and Harden (2008), all data labelled as ‘results’ or ‘findings’ was extracted digitally and imported verbatim into NVivo 12 software for qualitative analysis.

Thematic synthesis

Coding text and developing descriptive themes

Data was analysed inductively. Descriptive codes were assigned to sections of text through the process of line-by-line coding, resulting in a total of 53 initial codes. Initial descriptive codes were then compared and grouped into a hierarchical tree structure. New codes were created to capture the meaning of groups of initial codes, resulting in a tree structure with several layers of descriptive themes. For example, some of the text that was initially coded as ‘relationship with health care professionals’ from one study (Nuttall & Nicholes, 1992) was: *“I’m happier if I can negotiate and be involved and knowledgeable about my care; I know my body best.”* This use of line-by-line coding enabled the *translation* of concepts from one study to another (Thomas & Harden, 2008).

Generating analytical themes

Whilst the descriptive themes adhered closely to the original findings of included studies, generation of analytical themes aimed to *move beyond* the findings of the primary studies to generate new concepts, understandings and hypotheses. As per Thomas and Harden (2008) ‘going beyond’ the content of the original studies was achieved by using the descriptive themes that emerged from the inductive analysis to provide answers to the more abstract review question.

Facilitators of empowerment, and barriers to empowerment for CYP with CF were *inferred* from the views of CYP and the interpretations provided by researchers in original studies. Generation of analytic themes was done independently by the author and discussed in supervision and with research peers. Through discussion, more abstract and analytical themes began to emerge. Facilitators of empowerment, barriers to empowerment, and implications for clinical practice and research were examined again in light of these increasingly analytic themes and changes made as necessary. This process was repeated until themes were sufficiently abstract to describe and/or explain the initial descriptive themes, inferred facilitators and barriers and the clinical and research implications.

Results

The database search identified 774 papers, of which 16 were relevant for inclusion. An additional paper was identified through searching reference sections. Table 2 summarises the 17 included studies.

This review aimed to include enough information about the original studies to enable the reader to understand the context in which the findings were developed. The results section will start with an overview of included studies. A methodological critique of the studies will then be provided, followed by the thematic synthesis.

Table 2

Summary of studies

<u>Author, year, country</u>	<u>Aims</u>	<u>Participants</u>	<u>Data collection</u>	<u>Qualitative analysis</u>	<u>Empowerment indicators described (Jørgensen et al., 2018)</u>
Ayers et al. (2011) UK	To understand the nature and treatment of needle-related distress in children with CF	14 child-parent dyads. Children were 5 males, 9 females aged 7-17yrs with CF. All participants White European. Frequency of needle procedures ranged from 1-6 times a year, no other indicators of CF severity reported.	Interviews	Thematic analysis (Boyatzis, 1998)	Feeling in control, having influence
Barker et al. (2012) USA	To investigate perspectives on the support adolescents received from friends / family in CF management	24 adolescents with CF; mean age 15.7 years, 50% female, 17% Hispanic no other ethnicity data reported, mean FEV1 ^a % 73.6% ±24.1,	Interviews	Template analysis (King, 1999)	Feeling in control, being in charge, autonomy
Christian & D'Auria (1997) USA	To explore adolescents' conceptualisations of CF & related life events	20 12-18yr olds with CF; 12 female, 8 male; 17 described as white, 3 as Native American; CF severity: 6 mild, 14 moderate	Interviews	Grounded theory (Strauss & Corbin, 1990)	Feeling in control, having influence, agency

Christofides et al. (2016) Canada	To explore research decision processes, particularly in children with CF who are extensively involved in research	19 8-18yr olds with CF (12 female, 7 male); ethnicity & CF severity not reported	Interviews	Thematic analysis (Braun & Clarke, 2006)	Feeling in control, having influence, being in charge, agency
Dashiff et al. (2013) USA	To describe the experience of CF-related diabetes (CFRD), and the relationship of adolescent disease self-management with parental support.	10 15-19yr olds (30% female, 70% male), 90% described as Caucasian (non-Hispanic), 10% Black, CF severity not described. Included parent perspective.	Mixed methods: Interviews; quantitative questionnaires	Qualitative description (Sandelowski, 2000)	Being in charge, having mastery, agency, autonomy
D'Auria et al. (1997) USA	To understand the middle-childhood experience of CF as a chronic illness	20 children aged 6-12yrs with CF, 12 males 8 females, all Caucasian, CF severity not reported.	Interviews	Grounded theory (Strauss & Corbin, 1990)	Having influence, agency
Durst et al. (2001) USA	To understand CF patients' common themes and emotional responses, post lung-transplant	19 adolescent transplant recipients with CF. Mean age at transplant 15.7 ± 2.7 . Time from transplant to interview 1-58 months. 11 female, 8 male. 15 Caucasian, 3 Latino, 1 African American; no other indicators of CF severity reported	Interviews	Ethnographic study design – no further details of data analysis provided	Feeling in control

Foster et al. (2001) UK	To explore the impact of CF and treatment on patients and first-degree relatives	8 10-18yr olds with CF (5 female, 3 male). 8 parents, 8 siblings. Ethnicity not reported. FEV1 ^a 38-114%	Interviews	Grounded theory (Glaser & Strauss, 1967)	Being in charge, agency or autonomy
Moola & Faulkner (2014) Canada	To describe attitudes to illness and how illness narratives affect attitudes towards physical activity in children with CF. Part of a broader study (Moola et al. 2011) evaluating a physical activity counselling intervention.	2 adolescents with CF aged 16 & 17yrs (1 female, 1 male); ethnicity not reported; CF severity: moderate FEV1 ^a = 85%	Multiple interviews. Part of a wider intervention study.	Thematic analysis (Braun & Clarke, 2006); Case study analysis (Stake, 2000)	Feeling in control, having mastery, being in charge, having influence, agency, autonomy
Moola et al. (2012) Canada	To explore perceptions of exercise in young people with CF	14 young people with CF aged 11-17yrs (10 female, 5 male) 13 described as Caucasian, 1 as Black, 1 as East Indian. CF severity not reported.	Interviews	Grounded theory (Charmaz, 2003)	Having mastery, agency
Nuttall & Nicholes (1992) USA	To describe problems arising in adolescents with CF related to maternal concerns, hospital/ home care, and healthcare	20 adolescents with CF; 11 female, 9 male; mean age 17.9yrs; 98% Caucasian; CF severity described as 'moderate advanced'	Interviews	Content analysis (no reference provided)	Feeling in control, being in charge, having influence, agency or autonomy

	professionals' role in promoting care				
Pizzignacco & de Lima (2006) Brazil	To identify the routines of CYP with CF and situations that can affect these routines	8 7-18yr olds with CF; gender, ethnicity, & CF severity not reported	Ethnographic design: Interviews, field diary notes & patient chart review	Thematic analysis - no further details of data analysis provided	Feeling in control, being in charge, having influence, agency or autonomy
Savage & Callery (2007) Ireland	To explore children and parents' perspectives on communication between children, parents & professionals about CF diet.	32 children with CF aged 6-14yrs (18 female, 14 male); ethnicity not reported; CF severity: children in sample ranged from below 3rd to above 97th percentile for height & weight. Included parent perspective.	Interviews; observations of clinical consultation	Discourse analysis (Potter, 1997)	Feeling in control, being in charge, having influence, agency or autonomy
Sawicki et al. (2015) USA	To explore facilitators and barriers to adherence with chronic CF therapies; perspectives of adolescents and their parents.	18 adolescents with CF aged 16-21yrs (10 female, 8 male). Ethnicity not reported. Mean FEV1 ^a = 91% (range 38-127%), Mean BMI 21.9 (range 18.7 - 24.6)	Interviews	Thematic analysis? - no further details of data analysis provided	Feeling in control, having mastery, being in charge, having influence, agency, autonomy
Williams et al. (2007a) UK	To explore the nature and variation in roles of family members, and how responsibility is	32 children with CF aged 7-17yrs (18 female, 14 male); ethnicity not reported; CF severity reported as: 16 mild FEV1 ^a	Interviews	Framework analysis (Ritchie & Spencer, 1994)	Feeling in control, having mastery, being in charge, having influence, agency, autonomy

	transferred from the parent/family to the child, and what factors aid or hinder this	> 70%, 9 moderate 51-69%, 8 severe 20-50% Included parent perspective.			
Williams et al. (2007b) UK	To explore child and parent accounts of the difficulties of adhering to chest physio for CF, and identify strategies used by families to overcome these.	32 children with CF aged 7-17yrs (18 female, 14 male); ethnicity not reported; CF severity: 15 mild FEV ₁ ^a > 70%, 9 moderate 51-69%, 8 severe 20-50%. Included parent perspective.	Interviews	Framework analysis (Ritchie & Spencer, 1994)	Feeling in control, having mastery, being in charge, having influence, agency or autonomy
Williams et al. (2009) UK	To describe the idea of normality: forms, meaning and importance.	32 children with CF aged 7-17yrs (18 female, 14 male); ethnicity not reported; CF severity: 15 mild FEV ₁ ^a > 70%, 9 moderate 51-69%, 8 severe 20-50%	Interviews	Framework analysis (Ritchie & Spencer, 1994)	Feeling in control, having mastery, being in charge, having influence, agency or autonomy

^a FEV₁ (forced expiratory volume in 1 second) is a measure of CF disease severity. An FEV₁ of 100% means that lung function is as would be expected for a person of the same age and height who doesn't have CF. FEV₁ ≥ 85% is considered normal; 70-84% indicates mild lung disease, 41-69% indicates moderate lung disease; ≤ 40% suggests severe lung disease (UK Cystic Fibrosis Registry, 2013).

Characteristics of studies and participants

Studies were published between 1992-2016 in peer-reviewed journals from a range of disciplines including medicine, nursing and psychology. Despite a thorough search, not limited by date, no studies published outside of the above date range were found to meet eligibility criteria. Studies took place in a variety of geographical and cultural contexts including USA, Canada, Brazil, UK and Ireland.

A total of 324 CYP were included in the 17 studies. Participants ranged in age from 6-21, and were predominantly white, though there were representatives from black and Asian communities. Studies included CYP whose CF ranged from mild to severe.

The majority of studies employed purely qualitative methods (n=16), the remainder used mixed methods. All studies used interviews as the primary qualitative data collection method. A minority additionally used field diary notes and patient chart review (n=1) or clinic observations (n=1). A wide range of data analysis methods were used including: thematic analysis (n = 5), grounded theory (n=4), framework analysis (n=3), template analysis (n=1), content analysis (n=1), discourse analysis (n=1), qualitative description (n=1) and ethnography (n=1).

Focus of studies

All included studies described at least one empowerment indicator as defined by Jørgensen et al. (2018). Within this, studies covered a broad range of topics, including identity, decision-making, adherence, the role of family and friends, and communication with healthcare

professionals. The majority of studies focused on a specific issue, e.g. needle-related distress, CF-related diabetes, or coping post-lung transplant. Other studies explored the experience of growing up with CF more broadly.

Quality assessment

As described above, the quality of studies was assessed using the CASP qualitative assessment checklist, supplemented by criteria designed for assessing research conducted with CYP. Appendix C provides an overview of key strengths and limitations of all studies. An overview of quality assessment ratings is provided in Appendix D.

Aims and design

All studies provided a clear statement of aims. Whilst specific aims and objectives varied between studies, all aimed to provide rich descriptions of complex phenomena and thus qualitative research methodology was appropriate.

There was significant variation in the level of detail in which study design was reported. Only three studies (Williams et al., 2009; Williams et al., 2007b, 2007a) reported actively involving CYP in the design and conduct of the study. Few studies provided a detailed rationale for choice of specific qualitative method.

Participants and sampling

Unlike quantitative research, there is little consensus on what constitutes adequate sample size in qualitative studies. Sample size ranged from a small case series involving two

participants to large scale qualitative studies involving 32 participants. Studies varied in the reporting of demographic variables, though all reported age and gender.

Most studies clearly reported an appropriate recruitment strategy (generally convenience or purposive sampling). Most studies acknowledged the role clinicians played in recruitment. Few authors reflected on how many and why potential participants choose not to participate.

Ethical considerations

The majority of studies (n=14) reported obtaining appropriate ethical approvals. All but two studies (Durst et al., 2001; Moola & Faulkner, 2014) described procedures for obtaining informed consent/assent. Only a minority of studies explicitly referred to confidentiality and none reported procedures for responding to any participant distress relating to the research.

Data collection and analysis

Studies varied in the level of detail reported regarding data collection processes. Twelve noted the use of topic guides and gave an overview of topics. The majority of studies reported details of audio-recording and transcription.

Reporting of details relating to the process of qualitative analysis varied between studies, though most described and clearly referenced specific forms of analysis. Most studies clearly evidenced using appropriate methods for ensuring data analysis was grounded in the views of children, though few authors made reference to data saturation.

Reflexivity

Researcher sensitivity to the ways in which the researcher and research process shape the data, including personal characteristics, prior experiences and assumptions, is referred to as reflexivity (Mays & Pope, 2000). This was a weakness across studies, as only three studies (Moola et al. 2012; Sawicki et al. 2015; Williams et al. 2009) demonstrated reflexivity. Moola et al. (2012) described using bracketing techniques to become cognizant of preconceived assumptions, while Sawicki et al. (2015) included some discussion of the potential for bias. Williams et al (2009) was the only study to describe external validation.

Validity of study findings

Although all studies were explicit in reporting findings there was variation in levels of evidence provided. Some authors discussed credibility of findings, but none reported respondent validation. Approximately two thirds of studies specified that data was analysed by more than one researcher; inter-rater reliability was reported where this was consistent with the data analysis method (e.g. template or content analysis).

Although not the focus of the current review, approximately half the studies included parent perspectives, adding to the validity of findings. Other studies demonstrated triangulation of data through use of life event lines (Christian & D'Auria, 1997; D'Auria et al., 1997), reviewing patient notes and field notes (Pizzignacco & de Lima, 2006) or clinic observations (Savage & Callery, 2007).

Williams et al. (2007b) and Williams et al. (2009) added to the validity of findings through attention to negative cases (Mays & Pope, 2000). Williams et al. (2009) sought external validation through the employment of two researchers not otherwise involved with the study.

Thematic synthesis

The thematic synthesis resulted in the generation of six analytical themes (highlighted in bold). Facilitators of empowerment appeared to include having **relational support**, having **information and understanding, feeling heard and respected**, and experiencing **mastery and competence**. In contrast the **prejudice and assumptions** of others could be a barrier to empowerment, whilst **navigating being different** appeared to both influence and be influenced by empowerment.

Themes, associated subthemes and the studies contributing to each theme are described in Table 4, while representative quotations are shown in Table 5.

In some cases, there was a direct connection between the data and empowerment; for others, further synthesis was required. In contrast to quantitative approaches to data synthesis, the number of contributing studies is not necessarily representative of the strength of the theme (Dixon-Woods et al., 2006).

Themes identified through the review process as appearing to facilitate empowerment will be reported first, followed by themes relating to potential barriers to empowerment. Themes

relating to processes that both influence and are influenced by empowerment will then be described.

Table 4

Table of studies contributing to themes

<u>Theme</u>	<u>Subtheme</u>	<u>Contributing studies</u>
Relational support	Treatment related support	Ayers, Muller, Mahoney, & Seddon, 2011; Barker et al., 2012; Dashiff, Suzuki-Crumly, Kracke, Britton, & Moreland, 2013; Moola & Faulkner, 2014; Nuttall & Nicholes, 1992; Sawicki et al., 2015; Williams et al., 2009, 2007b; Williams, Mukhopadhyay, Dowell, & Coyle, 2007a
	Emotional support	Ayers et al., 2011; Christian & D’Auria, 1997; D’Auria et al., 1997; Dashiff et al., 2013; Durst et al., 2001; Moola & Faulkner, 2014; Moola et al., 2012; Nuttall & Nicholes, 1992; Sawicki et al., 2015; Williams et al., 2007a, 2007b; Barker et al., 2012; Christofides et al., 2016; Foster et al., 2001; Pizzignacco & de Lima, 2006; Savage & Callery, 2007; Williams et al., 2009
Information and understanding	Knowledge about CF	Christian & D’Auria, 1997; Christofides et al., 2016; D’Auria et al., 1997; Dashiff et al., 2013; Moola & Faulkner, 2014; Moola et al., 2012; Nuttall & Nicholes, 1992; Pizzignacco & de Lima, 2006; Williams et al., 2007b Ayers et al., 2011; Dashiff et al., 2013; Durst et al., 2001; Moola &

	Learning from experience	Faulkner, 2014; Sawicki et al., 2015; Williams et al., 2007b
	Feeling heard and respected	Ayers et al., 2011; Nuttall & Nicholes, 1992; Savage & Callery, 2007; Sawicki et al., 2015; Williams et al., 2009, 2007a
	Mastery and competence	Christian & D'Auria, 1997; D'Auria et al., 1997; Durst et al., 2001; Nuttall & Nicholes, 1992; Savage & Callery, 2007; Sawicki et al., 2015; Williams et al., 2009, 2007b, 2007a
	Prejudice and assumptions	
	Bullying	Barker et al., 2012; Christian & D'Auria, 1997; D'Auria et al., 1997; Foster et al., 2001; Pizzignacco & de Lima, 2006; Williams et al., 2009
	Differential treatment	Christian & D'Auria, 1997; Durst et al., 2001; Foster et al., 2001; Moola et al., 2012
	Navigating being different	
	Becoming aware of difference	Christian & D'Auria, 1997; D'Auria, Christian, & Richardson, 1997; Moola, Faulkner, & Schneiderman, 2012; Sawicki, Heller, Demars, & Robinson, 2015; Williams, Corlett, Dowell, Coyle, & Mukhopadhyay, 2009
	Deciding if and who to tell	Barker, Driscoll, Modi, Light, & Quittner, 2012; Christian & D'Auria, 1997; Christofides, Dobson, Solomon, Waters, & O'Doherty, 2016; D'Auria et al., 1997; Pizzignacco & de Lima, 2006; Sawicki et al., 2015; Williams et al., 2009; Williams, Mukhopadhyay, Dowell, & Coyle, 2007
		Christian & D'Auria, 1997; D'Auria et al., 1997; Durst et al., 2001; Foster et al., 2001; Moola & Faulkner, 2014; Williams et al.,

Perceived threats to 'normality'	2009, 2007; Dashiff, Suzuki-Crumly, Kracke, Britton, & Moreland, 2013
Competing priorities	Durst et al., 2001; Moola & Faulkner, 2014; Moola et al., 2012; Nuttall & Nicholes, 1992; Williams et al., 2009, 2007b
Positive aspects of difference	Christofides et al., 2016; Durst et al., 2001

Table 5

Representative quotations

Themes	Subthemes	Exemplary quotations
Relational support	Treatment related support	<i>"[Mom] keeps telling me to do it whether I want to or not, she knows that it's going to help me so it's pretty supportive."</i> (Barker et al., 2012, p. 500)
	Emotional support	<i>"I think it's really helpful if you could talk to somebody else that had CF. You know it's like a CF person. I think that is one of the most helpful things that has helped me, those CF camps that we can't, we can't have them anymore ... I learned a lot from them."</i> (Christian & D'Auria, 1997, p. 9).
		<i>"She [mum] usually just holds my hand and cuddles next to me . . . I know she's there and it's just better"</i> (Ayers et al., 2011, p. 336)
Information and understanding	Knowledge about CF	<i>"Get personally involved; don't be afraid just because we may not be around too long."</i> (Nuttall & Nicholes, 1992, p. 206)
		<i>"I didn't take my pills at school because that was when I first started, 'cause I was like "Why do you take your pills?" And I didn't really know what to say to them because I was younger"</i> (Christian & D'Auria, 1997, p. 6)
		<i>"I don't know very much. I know it's more or less similar to Down's Syndrome. It's from the same</i>

		<p><i>chromosome, I think it is, but I really don't know for sure" (Pizzignacco & de Lima, 2006, p. 572)</i></p> <p><i>"I think it's really helpful if you could talk to somebody else that had CF...those CF camps that we can't, we can't have them anymore ... I learned a lot from them" (Christian & D'Auria, 1997, p. 9)</i></p> <p><i>"I like being healthy. It makes me feel good and I did not know before that I would feel this way about being healthy. Once I did, I felt good, I felt great, and once you do that, you do not want to go back." (Moola & Faulkner, 2014, p. 32)</i></p>
Feeling heard and respected		<p><i>"I'm happier if I can negotiate and be involved and knowledgeable about my care; I know my body best." (Nuttall & Nicholes, 1992, p. 204)</i></p>
Mastery and competence		<p><i>"I'm competent and responsible; I've been caring for myself all my life." (Nuttall & Nicholes, 1992, p. 203)</i></p> <p><i>"I could go to people's houses and stay over because I knew how to do my physio. I didn't have to come home and get my mum to do it for me" (Williams et al., 2009, p. 1450)</i></p>
Prejudice and assumptions	<p>Bullying</p> <p>Differential treatment</p>	<p><i>"All the kids, you know, saying "ewwr".... On the bus" (Christian & D'Auria, 1997, p. 6)</i></p> <p><i>"I do not want to be treated differently . . . so what, I have CF. One of my cousins, he went easy on me in sports and I got really mad." (Moola et al., 2012, p. 53)</i></p> <p><i>"...my mom never wants me to leave the house...but I did not go through transplant to live my life in a bubble" (Durst et al., 2001, p. 30)</i></p>
Navigating being different	<p>Becoming aware of difference</p> <p>Deciding if and who to tell</p> <p>Perceived threats to 'normality'</p>	<p><i>"I didn't know what it was, 'cause I didn't even know I was sick" (Christian & D'Auria, 1997, p. 6)</i></p> <p><i>"I'm not going to tell my coach I have it. So he won't treat me any different than the other kids." (Christian & D'Auria, 1997, p. 7)</i></p> <p><i>"it's quite hard when I've got friends staying or something, it's like I have to go through to the spare room do my breathing stuff, whereas if I didn't have CF I wouldn't have to, I just hate having to do all the stuff I have to do." (Williams et al., 2007b, p. 1100)</i></p>

Competing priorities	<p><i>“I know that I should do it (activity)—it should be higher on the priority list. The more important things for me are like work and school”</i> (Moola et al., 2012, p. 54)</p> <p><i>“...my mom never wants me to leave the house...but I did not go through transplant to live my life in a bubble”</i> (Durst et al., 2001, p. 30)</p>
Positive aspects of difference	<p>CYP who had undergone lung transplant spoke of their surgical scars as representing ‘strength’, ‘survival’ and a ‘signature point in my life’ (Durst et al., 2001, p. 30).</p>

Relational support

Across studies CYP described support from others including family, friends and professionals as promoting positive coping with the demands of CF and CF treatment, and enabling participation in activities that were important to them. Relational support took a variety of forms which can be broadly divided into tangible support associated with treatment regimens and more general emotional support.

Treatment-related support

In the majority of studies, CYP of all ages implicitly or explicitly referred to the role of parents in providing treatment-related support (Barker et al., 2012; Dashiff et al., 2013; Moola & Faulkner, 2014; Nuttall & Nicholes, 1992; Sawicki et al., 2015; Williams et al., 2009). This took a number of forms, including practical support such as undertaking physiotherapy with or for CYP, preparing medications, arranging medical appointments, and providing treatment reminders and encouragement. In one study (Barker et al., 2012) CYP reported that parents sometimes let them ‘slack off’ their treatments and reported this lack of monitoring as unsupportive.

In several studies CYP also spoke of the role that friends played in providing treatment-

related support (Barker et al., 2012; Christian & D'Auria, 1997; D'Auria et al., 1997).

Many CYP described the high treatment burden associated with CF and made links between receiving support with treatment and being able to do the things that were important to them, e.g. spending time with friends or participating in sport. In one study, CYP identified treatment-related behaviours such as nagging as annoying or unwanted, but were reluctant to describe family members or friends as being 'unsupportive' when they engaged in these behaviours (Barker et al., 2012).

Where studies included a focus on relationships with health care professionals, the importance of consistency and trust was repeatedly articulated (Ayers et al., 2011; Nuttall & Nicholes, 1992; Sawicki et al., 2015; B. Williams et al., 2007b, 2007a)

Emotional support

The importance of emotional support was highlighted by a number of studies across a range of contexts. CYP in Ayers et al. (2011) spoke of the role of parents in providing emotional support during distressing procedures. In other studies (e.g. Dashiff et al., 2013; Moola et al., 2012), CYP referred to emotional support more generally, e.g. the role of family members '*being there and listening*' (Dashiff et al., 2013).

Participants in several studies noted that alongside practical treatment related support, health care professionals often provided essential emotional support (Ayers et al., 2011; Nuttall & Nicholes, 1992; Sawicki et al., 2015; Williams et al., 2007a, 2007b). CYP in several studies described the importance of feeling comfortable talking with their CF team about general life

concerns. CYP in these studies reported that their medical teams were able to understand what their lives were like and empathise with the difficulties of adhering to complex treatment regimens (Sawicki et al., 2015; Williams et al., 2007b). A subgroup of CYP in Nuttall and Nicholes (1992), expressed wanting professionals to be more emotionally involved.

In the Christian and D’Auria (1997) and D’Auria et al. (1997) studies, emotional support provided by ‘good friends’ was found to offer protection from the effects of bullying and difficulties associated with trying to keep CF a secret, such as feeling unable to take medication at school. This was the case both for younger children Christian and D’Auria, (1997) and older children (D’Auria et al., 1997).

Interestingly, in the only study to include transplant recipients, CYP reported improved interactions with class mates and an improved sense of belonging, post-transplant (Durst et al., 2001). It was unclear how participants in this study experienced emotional support from friends prior to transplant and during initial recovery.

In several studies (Christian & D’Auria, 1997; Moola & Faulkner, 2014), participants referred to support from other CYP with CF. Prior to the mid-1990s, CYP were encouraged to socialise through organised events, e.g. summer camps. It became apparent that this practice was potentially dangerous due to infection risks, and as a result, strict infection control guidelines were introduced in order to prevent face-to-face contact between CYP with CF. These studies alluded to the impact of infection control guidelines, which appeared to

have been implemented differently between studies, reflecting differing geographical contexts and years of study publication.

Information and understanding

In many studies, CYP described the impact of the amount of information to which they had access. Having information and understanding appeared to facilitate empowerment by influencing how CYP made sense of their experiences, and how confident they felt in making decisions and explaining CF to others. Information appeared to come both from external sources (e.g. clinic consultations), and CYP's own experiences (e.g. learning from the consequences of medication non-adherence).

Knowledge about CF

Levels of CF knowledge appeared to vary both between and within studies. In several studies, CYP with CF and their peers were found to have difficulty understanding the meaning of CF and how the label could account for their differences (Christian & D'Auria, 1997; Moola & Faulkner, 2014; Pizzignacco & de Lima, 2006).

CYP in a few studies described awareness of not having the knowledge and skills to feel confident in telling their peers about CF (Christian & D'Auria, 1997; D'Auria et al., 1997; Pizzignacco & de Lima, 2006). Some CYP reported the experience of being diagnosed with complications of CF such as CF-related diabetes (CFRD) as “shocking,” “bad,” “hard,” “terrible,” “depressing,” and “agitating”, attributing much of the distress to not understanding the meaning and implications of the news (Dashiff et al., 2013). A lack of information and understanding about CF limited participation, with some CYP avoiding activities because they were unsure what they could do safely or what they might need to do differently (Christian & D'Auria, 1997). Some authors interpreted this as a consequence of professionals

overestimating the scientific knowledge of CYP and their families (Pizzignacco & de Lima, 2006).

In one study CYP reported gaining information through participation in research (Christofides et al., 2016). In other studies CYP described meeting other children with CF at camp gave them a broader perspective of CF and potential implications for their future lives (Christian & D'Auria, 1997; Moola & Faulkner, 2014). Nuttall and Nichole's study (1992) described CYP wanting more information about CF's potential impact on areas such as careers and fertility to enable them to plan ahead.

Learning from experience

Many CYP described learning from experience as a process (Moola & Faulkner, 2014). CYP in a number of studies commented on learning from experiencing the consequences of not following treatment plans, with subsequent increased adherence (Sawicki et al., 2015; Williams et al., 2007b). Some described the development of CF-related comorbidities such as CFRD acting as a 'wake-up call' (Dashiff et al., 2013). Other CYP commented that skipping physio or medication once or twice did not seem to have an impact on their health (Sawicki et al., 2015; Williams et al., 2007b). CYP in one study Durst et al. (2001) seemed to take a particularly proactive approach to the unpredictable nature of transplant, possibly due to their past experiences.

Feeling heard and respected

The importance of being heard and respected, particularly in the hospital setting, was articulated across a number of studies by participants of all ages (Ayers et al., 2011; Nuttall & Nicholes, 1992; Savage & Callery, 2007; Sawicki et al., 2015; Williams et al., 2009). This

was true both for studies focusing on specific issues such as needle-related distress and for studies exploring experiences of growing up with CF more generally.

Many CYP commented on the distress caused when they perceived their voice was not being heard (Nuttall & Nicholes, 1992). They suggested patient care could be improved through increased patient input in care planning (Nuttall & Nicholes, 1992), and believed that they should be the principal person consulted on matters such as diet (Savage & Callery, 2007).

For many older participants it was important to be treated as an adult, seeing the doctor alone or the doctor addressing them rather than their parents; having their social life, roles and commitments accommodated were important indicators of this (Sawicki et al., 2015; Williams et al., 2007a)

Mastery and competence

In many studies CYP made references to the importance of experiencing mastery and being perceived by others as competent. This was the case both in relation to CF care and typical activities of childhood, e.g. performance in sports.

Some CYP described feeling competent in managing CF as becoming easier over time; others spoke of becoming used to it as ‘all I’ve ever known’ (Dashiff et al., 2013). Feeling confident and competent in managing CF regimens appeared to increase independence, enabling CYP

to adopt more 'normal' roles and participate in social activities such as going on trips (Williams et al., 2009, 2007a).

CYP in Nuttall and Nicholes (1992) commented on the importance of being recognised as competent in self-management of CF. Some CYP said having their parents 'on them' all the time regarding CF treatment had a negative impact on their behaviour and relationships; when their parents stepped back and gave them responsibility for their own care they did better (Sawicki et al., 2015). Similarly, in their study of clinic consultations, Savage and Callery (2007) found that CYP felt more competent when professionals used a communication style that encouraged participation rather than telling them what to do, e.g. asking CYP to advise as to which physiotherapy technique they found most helpful. In several studies CYP referred to how increasing independence as they grew older allowed more autonomy in CF management and made it easier for to appear 'normal' to others (Williams et al., 2009, 2007b).

CYP in a number of studies commented on enjoyment of physical activities (Moola & Faulkner, 2014; Moola et al., 2012; Williams et al., 2009, 2007b). The comments of some CYP suggested that these activities provided memorable mastery experiences which resulted in an enduring sense of competence and achievement (Moola et al., 2012). In several studies, CYP emphasized that CF didn't stop them from achieving goals and aspirations (Williams et al., 2009, 2007b, 2007a). For many of the CYP in these studies this appeared to relate to the pursuit of normality and ensuring that personal, hopes, values and aspirations were not disrupted by CF. In contrast, participants in the only study to focus on the experiences of CYP who had undergone transplant, described how post-transplant they were now able to do

ordinary tasks such as walking without breathlessness, housework, and socialising (Durst et al., 2001).

Prejudice and assumptions

In many studies, CYP described the negative impact of other people's responses to differences associated with CF. Both explicit teasing or bullying and well-intentioned differential treatment appeared in some cases to act as barriers to empowerment.

Bullying

In a number of studies CYP spoke of attracting negative attention from peers. This was often described as one of the most stressful aspects of growing up with CF, with some CYP becoming anxious as they anticipated the potential reactions of their peers (D'Auria et al., 1997). CYP described peers highlighting and mocking CF-related differences and treating them with suspicion (Barker et al., 2012; Christian & D'Auria, 1997; D'Auria et al., 1997; Foster et al., 2001; Pizzignacco & de Lima, 2006; Williams et al., 2009). For some this included being labelled as 'druggies' or 'contagious', and bullies encouraging other children to stay away (Christian & D'Auria, 1997; D'Auria et al., 1997; Foster et al., 2001).

Perceived differential treatment

CYP in several studies reported being treated differently by others because of CF. These perceptions of differential treatment included experiencing teachers and coaches using unequal standards to evaluate performance (Christian & D'Auria, 1997), healthy peers being less competitive (Moola et al., 2012) and parents disciplining healthy siblings more harshly (Foster et al., 2001). CYP in the Durst et al. (2001) study described continuing to feel overprotected, post-transplant. Across a number of studies, CYP expressed strong preferences

for adults treating them like any other child, and described feeling frustrated, patronized or guilty when they perceived differential treatment.

Navigating being different

CYP's experiences of navigating differences associated with having CF appeared to influence and be influenced by empowerment.

Becoming aware of difference

CYP in a number of studies described the process of becoming aware of having CF and of the ways that this made them different from other children. For many children becoming aware of 'being different' occurred between 6-8 years (Christian & D'Auria, 1997) and was often reported as being a surprise (Christian & D'Auria, 1997; D'Auria et al., 1997). Younger children in contrast did not reveal any major struggles to mask differences associated with having CF such as taking medication (Williams et al., 2009). Whereas older participants made reference to issues of difference, sameness, and acceptance, and were found to use language that emphasised non-difference (Moola et al., 2012; Sawicki et al., 2015; Williams et al., 2009)

Deciding if and who to tell

CYP in approximately half the studies mentioned deciding if and who to tell that they had CF. Telling others about the CF diagnosis appeared to threaten a normal developmental need to gain social approval and develop friendships (Christian & D'Auria, 1997; D'Auria et al., 1997; Williams et al., 2007b). Many participants described ambivalence about disclosing CF, e.g. wanting to tell but fearing that this could jeopardize their potential to have romantic relationships (Christian & D'Auria, 1997).

Some CYP reported that by not telling teachers and coaches about CF, they could be viewed as equally competent in group activities (Christian & D'Auria, 1997). For others, there seemed to be a more general reluctance to talk about CF (Christofides et al., 2016).

Perceived threats to 'normality'

The 'CF cough', taking medications and physical limitations were reported by CYP in a number of studies as the aspects of CF that most highlighted difference from peers (Christian & D'Auria, 1997; D'Auria et al., 1997; Williams et al., 2009). Williams et al. (2007b) found that physiotherapy threatened CYP's sense of 'non-difference' in three ways: time taken from activities that maintain 'non-different' identity; non-participation in typical childhood activities; and physiotherapy elements perceived as 'embarrassing' and 'disgusting' e.g. spitting.

Sawicki et al. (2015) grouped comments from CYP under 'privacy concerns': wanting to be 'normal', not wanting to be different or disabled, self-consciousness about taking medications at school, and not wanting to take CF-related equipment outside the home. Similarly, participants in Williams et al. (2007a) described being embarrassed undertaking physiotherapy away from home and that joining their peers in typical childhood activities such as sleepovers "wasn't worth the hassle". Interestingly, CYP expressed a sense of belonging and improved interactions with peers post-transplant (Durst et al., 2001).

Competing priorities

The impact of having limited time and a high treatment burden was described by CYP across studies and age groups. CYP in several studies described the challenge of balancing competing priorities within the context of a life-limiting condition (Moola et al., 2012; Nuttall & Nicholes, 1992). Many CYP understood that their life expectancy was shorter than

that of their peers and questioned whether they would ever attain important developmental milestones (Moola et al., 2012). Some older participants in described CF as getting in the way of their goals and career aspirations (Williams et al., 2009). CYP in Durst et al. (2001) reflected on balancing the risks of infection and transplant rejection against ‘trying to have a normal life’, suggesting this continues to be the case, post-transplant.

CYP of all ages reported a sense of inequality and unfairness due to the way that physiotherapy restricted their lives compared to their peers (Williams et al., 2007b). Younger children who were receiving percussive physiotherapy from their parents expressed a sense of loss at not being able to stay overnight with friends or go on school trips as they needed parental help with physiotherapy. Older CYP spoke of not being able to stay out as late as friends due to physiotherapy (Williams et al., 2009, 2007b).

Although CYP were aware of the benefits of physical activity, for some CF symptoms meant that even simple everyday activities were exhausting (Moola et al., 2012). In the only study to include CYP’s reflections on an intervention, one participant in Moola and Faulkner's (2014) case series reported feeling less limited by CF and more able to consider attending college following an intervention involving physical activity and counselling.

Positive aspects of difference

CYP in several studies noted positive aspects of difference associated with having CF. CYP who had undergone lung transplant spoke of their surgical scars as representing ‘strength’, ‘survival’ and a ‘signature point in my life’ (Durst et al., 2001).

In Christofides et al.'s (2016) study of decision-making, some CYP described enjoying participating in research, acknowledging opportunities they would not otherwise have had, identifying as professional research participants or attributing an interest in science to this experience.

Discussion

To the author's knowledge, this is the first attempt to synthesise existing qualitative literature relating to empowerment in CYP with CF. This review screened 660 journal articles were screened and 17 qualitative articles relating to empowerment in CYP with CF were selected. Despite a thorough systematic search, no articles explicitly exploring empowerment were identified. However, descriptions of empowerment indicators were found in studies covering a wide range of topics including identity, decision-making, adherence, interpersonal support and communication.

Studies varied in quality. Lack of involvement of CYP in the design and conduct of the research and lack of researcher reflexivity were key weaknesses across studies. Whereas Thomas and Harden (2008) found that studies of higher methodological quality contributed more to their themes, the current study did not find this association. This reflects the findings of Jorgensen et al. (2018), study of facilitators of empowerment in adult patients, a finding the authors attributed to the inclusion of studies that did not have a specific focus on empowerment. Whilst in line with guidance on meta-synthesis, the number of contributing studies did not determine the weight of themes, themes that consisted of only a small number

of poorer quality studies need to be treated with more caution than more robust themes (e.g. ‘positive aspects of difference’ versus ‘feeling heard and respected’).

The key facilitators of empowerment in CYP with CF deduced from this review were having **relational support**, having **information and understanding**, **feeling heard and respected**, and experiencing **mastery and competence**. Barriers to empowerment emerging from the review reflected the opposite of the above-mentioned facilitators, e.g. **prejudice and assumptions** involving bullying or differential treatment could be considered the opposite of **feeling heard and respected**.

This review identified the theme of **navigating being different** that both influences and is influenced by empowerment. Existing theory reflects this dynamic relationship. CYP’s identity and sense of mastery and competence develop through comparison with and feedback from peers and adults (Erikson, 1959). For CYP with CF, comparison with peers may highlight their CF-related differences (coughing, taking medications, fatigue and school absences) (Ernst et al., 2011).

There were resonances between emerging themes and empowerment theories developed within the context of community psychology (Zimmerman, 1995). For example, having **information and understanding** could be thought of as part of an empowering process (a mechanism that increases opportunities for individuals to influence decisions that affect their lives and move closer to their goals). Similarly, experiencing **mastery and competence** could be considered an empowered outcome.

Many of the emerging themes map on to Bravo et al's (2015) theory of patient empowerment which was developed from research with adults with a range of LTCs. This highlighted the importance of 'feeling respected', experiencing 'self-efficacy' and having access to 'knowledge'.

There were also similarities between the current review's findings and those of previous reviews involving studies of adult patients. For example, in a review focusing on facilitators of patient empowerment in cancer patients during follow-up, Jørgensen et al. (2018) identified themes including 'knowledge is power' and 'communication and interaction between patients and health care professionals'. Similarly, in a study of empowerment in adults in the advanced stages of life-limiting illness, Wakefield et al. (2018) identified themes including 'personalised knowledge in theory and practice' and 'negotiating personal and healthcare relationships.'

Key differences between this review and previous work are that the themes identified in this review included a greater emphasis on support from parents and healthy peers; less emphasis on support from patient groups; and greater significance of themes related to identity and difference. The lives of CYP with or without LTCs are highly influenced by their adult caregivers. The findings of the current review suggest that empowerment in CYP with CF may be associated with attachment relationships. For example CYP who are able to reliably depend on parents and other adults including health care professionals and teachers for acceptance, support and information may be more empowered than those with a tendency towards more anxious-avoidant relationship patterns who may attempt to be entirely self-

sufficient or those with an anxious-ambivalent relationship patterns who may struggle to separate from caregivers and thus have fewer opportunities to experience mastery and competence.

Strengths and limitations

Developing the search strategy was a challenge as the concept of patient empowerment overlaps with other concepts such as self-efficacy, and can be understood and defined in many different ways (Bravo et al., 2015). Use of the conceptual definition employed in the Jørgensen et al. (2018) study, afforded a relatively clear and comprehensive definition of empowerment to guide the search. However, whilst in some ways a strength, this definition of empowerment was theoretically based rather than derived from the experiences of patients and so could also be seen as a limitation. As noted by Jørgensen et al. (2018) employing a theoretically driven definition as the basis for the review may to some extent resulted in the review not being able to answer how the patients themselves experienced empowerment. Nonetheless, it was considered important to use a single definition of patient empowerment to enable a rigorous systematic search and synthesis of identified literature.

The number of participants included was moderate. Quality of included studies was variable and sometimes difficult to assess due to the way studies were reported. Most studies were conducted in developed countries, and non-English language articles were excluded, therefore the transferability of the findings is uncertain.

Strengths of this review include the novel review of empowerment in CYP with CF, and that emergent themes were represented across the included studies, supporting the validity of the findings.

Clinical implications

The findings of this review highlight the importance of CYP having accessible information, feeling heard and respected, and experiencing mastery (including in relation to CF self-management) in promoting patient empowerment.

It may therefore be useful for clinicians to continuously review with CYP's understanding of CF and to build a library of accessible resources. It may also be useful for service evaluations to be conducted, for example, using Likert scales to ascertain the extent to which CYP felt heard and respected (etc).

Issues related to CF and other LTCs could be incorporated into the Personal, Social, Health and Economic (PSHE) Education curriculum, potentially with involvement of clinicians. Clinicians might also work with educators to promote acceptance of difference, find ways of managing perceived differential treatment and to support CYP in making choices around managing difference, e.g. by enabling CYP to take medication in private if they wish to.

Strategies that may enhance both feeling heard and respected and mastery and competence include development of CYP expert patient groups or patient educator programmes that could

teach medical students CF-related history and examination, or be involved in service development and in creating CYP-friendly resources.

To support CYP in coming to terms with difference through interventions such as Acceptance and Commitment Therapy (ACT) may be beneficial. The decisions CYP have to make in balancing CF-treatment choices with day to day social and academic activities of childhood are complex, and there may be a role for motivational interviewing or ACT values-based work.

Research implications

None of the studies looked explicitly at empowerment in children with CF. Further qualitative research is required, particularly in relation to developmental and social factors influencing empowerment in CYP with CF. This could further explore how CYP manage competing priorities, and what helps them make complex decisions; how mastery and competence are experienced by CYP with CF at different developmental stages; and the role of adult others around them in facilitating this.

Strict infection control guidelines have been in place since the mid-1990s, and further research might investigate whether peer support occurs in the context of these restriction, as well as the nature and impact of such support.

CYP with CF are not only patients, but studies included in the current review primarily described indicators of empowerment in medical contexts. The term ‘patient empowerment’

has been criticised for being over medicalised (Johnston-Roberts, 1999) – and it would be valuable for future research to explore empowerment across domains.

Conclusion

This synthesis of qualitative studies provides an initial insight into experiences of CYP with CF relating to patient empowerment – a topic which is currently under-researched.

Through the process of thematic synthesis, the review identified a number of key themes relating to the process of empowerment in CYP with CF. Having relational support, information and understanding, feeling heard and respected, and experiencing mastery and competence were identified as factors that may facilitate empowerment. Prejudices and assumptions, particularly bullying or differential treatment were identified as potential barriers to empowerment. The findings of the review also suggested that navigating the impact of difference on self and facing limitations both influence and are influenced by empowerment.

Whilst some of the findings resonated with previous reviews exploring empowerment in relation to adults with a range of LTCs, the review also suggests that the process of empowerment may be influenced by both developmental changes and stages and factors unique to CF.

Findings need to be treated with caution as studies varied in quality. Overall, studies used appropriate research designs and methods for helping children to express their views, but the reporting of data analysis processes varied considerably. Across studies, the lack of

researcher reflexivity was a significant limitation, impacting on the validity of the findings reported.

Nonetheless, there are both clinical implications and a clear need for further qualitative research to more fully understand the contextual and developmental influences on empowerment in CYP with CF.

References

- Acuña Mora, M., Luyckx, K., Sparud-Lundin, C., Peeters, M., van Staa, A. L., Sattoe, J., ... Moons, P. (2018). Patient empowerment in young persons with chronic conditions: Psychometric properties of the Gothenburg Young Persons Empowerment Scale (GYPES). *PLoS ONE*, *13*(7), 1–13. doi:10.1371/journal.pone.0201007
- Acuña Mora, M., Sparud-Lundin, C., Burström, Å., Hanseus, K., Rydberg, A., Moons, P., & Bratt, E. L. (2019). Patient empowerment and its correlates in young persons with congenital heart disease. *European Journal of Cardiovascular Nursing*, *18*(5), 389–398. doi:10.1177/1474515119835434
- Ayers, S., Muller, I., Mahoney, L., & Seddon, P. (2011). Understanding needle-related distress in children with cystic fibrosis. *British Journal of Health Psychology*, *16*(2), 329–343. doi:10.1348/135910710X506895
- Barker, D. H., Driscoll, K. A., Modi, A. C., Light, M. J., & Quittner, A. L. (2012). Supporting cystic fibrosis disease management during adolescence: the role of family and friends. *Child: Care, Health and Development*, *38*(4), 497–504. doi:https://dx.doi.org/10.1111/j.1365-2214.2011.01286.x

- Barroso, J., Gollop, C. J., Sandelowski, M., Meynell, J., Pearce, P. F., & Collins, L. J. (2003). The challenges of searching for and retrieving qualitative studies. *Western Journal of Nursing Research, 25*(2), 153–178. doi:10.1177/0193945902250034
- Birks, M., & Mills, J. (2015). *Grounded Theory: A practical guide* (2nd ed.). London, UK: SAGE.
- Boyatzis, R. (1998). *Transforming qualitative information: Thematic analysis and code development*. London, UK: Sage.
- Braun, V., & Clarke, V. (2006). Using thematic analysis in psychology Using thematic analysis in psychology. *Qualitative Research in Psychology, 3*(2), 77–101. doi:10.1191/1478088706qp063oa
- Bravo, P., Edwards, A., Barr, P. J., Scholl, I., Elwyn, G., & McAllister, M. (2015). Conceptualising patient empowerment: A mixed methods study. *BMC Health Services Research, 15*(1), 1–14. doi:10.1186/s12913-015-0907-z
- Bronfenbrenner, U. (1986). Ecology of the Family as a Context for Human Development. Research Perspectives. *Developmental Psychology, 22*(6), 723–742. doi:10.1037/0012-1649.22.6.723
- Calvillo, J., Román, I., & Roa, L. M. (2015). How technology is empowering patients? A literature review. *Health Expectations, 18*(5), 643–652. doi:10.1111/hex.12089
- Carroll, C., Booth, A., & Lloyd-Jones, M. (2012). Should we exclude inadequately reported studies from qualitative systematic reviews? An evaluation of sensitivity analyses in two case study reviews. *Qualitative Health Research, 22*(10), 1425–1434. doi:10.1177/1049732312452937
- Carter, B., Ford, K., Bray, L., Water, T., Arnott, J., Tichanow, C., & Dickinson, A. (2017).

- “It’s not who I am”: Children’s experiences of growing up with a long-term condition in England, Australia, and New Zealand. *Journal for Specialists in Pediatric Nursing*, 22(4), 1–10. doi:10.1111/jspn.12197
- Cerezo, P. G., Juvé-Udina, M.-E., & Delgado-Hito, P. (2016). Concepts and measures of patient empowerment: a comprehensive review. *Revista Da Escola de Enfermagem Da USP*, 50(4), 667–674. doi:10.1590/S0080-623420160000500018
- Charmaz, K. (2003). Grounded theory in the 21st century. In N. Denzin & Y. Lincoln (Eds.), *The landscape of qualitative research: Theories and issues* (2nd ed., pp. 507–530). London, UK: Sage.
- Charmaz, K. (2006). *Constructing grounded theory: A practical guide through qualitative analysis*. London, UK: Sage Publications Ltd.
- Christian, B. J., & D’Auria, J. P. (1997). The child’s eye: memories of growing up with cystic fibrosis. *Journal of Pediatric Nursing*, 12(1), 3–12. Retrieved from <http://ovidsp.ovid.com/ovidweb.cgi?T=JS&PAGE=reference&D=med4&NEWS=N&AN=9037958>
- Christofides, E., Dobson, J. A., Solomon, M., Waters, V., & O’Doherty, K. C. (2016). Heuristic decision-making about research participation in children with cystic fibrosis. *Social Science & Medicine*, 162, 32–40. doi:10.1016/j.socscimed.2016.06.017
- Cooke, A., Smith, D., & Booth, A. (2012). Beyond PICO: The SPIDER tool for qualitative evidence synthesis. *Qualitative Health Research*, 22(10), 1435–1443. doi:10.1177/1049732312452938
- Corbin, J., & Strauss, A. (1991). A nursing model for chronic illness management based upon the trajectory framework. *Scholarly Inquiry for Nursing Practice: An International*

- Journal*, 5(3), 155–174. Retrieved from <https://www.ncbi.nlm.nih.gov/pubmed/1763239>
- Corbin, J., & Strauss, A. (2015). *Basics of qualitative research: Techniques and procedures for developing grounded theory* (4th ed.). London, UK: Sage Publications Inc.
- Coulthard, K. P. (2018). Cystic fibrosis: novel therapies, remaining challenges. *Journal of Pharmacy Practice and Research*, 48(6), 569–577. doi:10.1002/jppr.1516
- Coyne, I. (2006). Consultation with children in hospital: Children, parents' and nurses' perspectives. *Journal of Clinical Nursing*, 15(1), 61–71. doi:10.1111/j.1365-2702.2005.01247.x
- Cruz, I., Marciel, K. K., Quittner, A. L., & Schechter, M. S. (2009). Anxiety and depression in cystic fibrosis. *Seminars in Respiratory and Critical Care Medicine*, 30(5), 569–578. doi:10.1055/s-0029-1238915
- Cystic Fibrosis Trust. (2019). Cystic fibrosis FAQs. Retrieved September 14, 2019, from [https://www.cysticfibrosis.org.uk/what-is-cystic-fibrosis/faqs#How common is cystic fibrosis?](https://www.cysticfibrosis.org.uk/what-is-cystic-fibrosis/faqs#How%20common%20is%20cystic%20fibrosis?)
- D'Auria, J., Christian, B., & Richardson, L. (1997). Through the looking glass: Children's perceptions of growing up with cystic fibrosis. *Canadian Journal of Nursing Research*, 29(4), 99–112.
- Dashiff, C., Suzuki-Crumly, J., Kracke, B., Britton, L., & Moreland, E. (2013). Cystic fibrosis-related diabetes in older adolescents: parental support and self-management. *Journal for Specialists in Pediatric Nursing : JSPN*, 18(1), 42–53. doi:<https://dx.doi.org/10.1111/jspn.12010>
- Department of Health. (2015). NHS Constitution for England. *The NHS Constitution Establishes the Principles and Values of the NHS in England*, (July), 1. doi:26 March

2013

- Dey, I. (1999). *Grounding Grounded theory: Guidelines for qualitative inquiry*. California, CA: Academic Press.
- Dixon-Woods, M., Bonas, S., Booth, A., Jones, D. R., Miller, T., Sutton, A. J., ... Young, B. (2006). How can systematic reviews incorporate qualitative research? A critical perspective. *Qualitative Research*, 6(1), 27–44. doi:10.1177/1468794106058867
- Durst, C. L., Horn, M. V, MacLaughlin, E. F., Bowman, C. M., Starnes, V. A., & Woo, M. S. (2001). Psychosocial responses of adolescent cystic fibrosis patients to lung transplantation. *Pediatric Transplantation*, 5(1), 27–31. Retrieved from <http://ovidsp.ovid.com/ovidweb.cgi?T=JS&PAGE=reference&D=med4&NEWS=N&AN=11260485>
- Erikson, E. (1959). *Identity and the life cycle*. New York: Norton & Company.
- Ernst, M. M., Johnson, M. C., & Stark, L. J. (2011). Developmental and Psychosocial Issues in Cystic Fibrosis. *Pediatric Clinics of North America*, 58(4), 865–885. doi:10.1016/j.pcl.2011.06.004
- Flemming, K., José Closs, S., Hughes, N. D., & Bennett, M. I. (2016). Using qualitative research to overcome the shortcomings of systematic reviews when designing of a self-management intervention for advanced cancer pain. *International Journal of Qualitative Methods*, 15(1), 1–11. doi:10.1177/1609406916670656
- Foster, C., Eiser, C., Oades, P., Sheldon, C., Tripp, J., Goldman, P., ... Trott, J. (2001). Treatment demands and differential treatment of patients with cystic fibrosis and their siblings: Patient, parent and sibling accounts. *Child: Care, Health and Development*, 27(4), 349–364. doi:10.1046/j.1365-2214.2001.00196.x

- Gathercole, K. (2018). Managing cystic fibrosis alongside children's schooling: Family, nurse and teacher perspectives. *Journal of Child Health Care*.
doi:10.1177/1367493518814930
- Gibson, C. (1991). A concept analysis on the process of empowerment. *Journal of Advanced Nursing, 16*, 354–361. Retrieved from <http://www.ncbi.nlm.nih.gov/pubmed/22104094>
- Glaser, B., & Strauss, A. (1967). *The discovery of grounded theory: Strategies for qualitative research*. New Brunswick: AldineTransaction.
- Goodwin, N., Sonola, L., Thiel, V., & Kodner, D. (2013). *Co-ordinated care for people with complex chronic conditions: Key lessons and markers for success*. The Kings Fund.
doi:/978 1 909029 19 4
- Hegarty, M., MacDonald, J., Watter, P., & Wilson, C. (2009). Quality of life in young people with cystic fibrosis: Effects of hospitalization, age and gender, and differences in parent/child perception. *Child: Care, Health and Development, 35*(4), 462–468.
doi:<http://dx.doi.org/10.1111/j.1365-2214.2008.00900.x>
- Iles, N., & Lowton, K. (2008). Young people with cystic fibrosis' concerns for their future: When and how should concerns be addressed, and by whom? *Journal of Interprofessional Care, 22*(4), 436–438. doi:10.1080/13561820801950325
- Johnston-Roberts, K. (1999). Patient empowerment in the United States: A critical Commentary. *Health Expectations, 2*, 82–92.
- Jørgensen, C. R., Thomsen, T. G., Ross, L., Dietz, S. M., Therkildsen, S., Groenvold, M., ... Johnsen, A. T. (2018). What Facilitates “Patient Empowerment” in Cancer Patients During Follow-Up: A Qualitative Systematic Review of the Literature. *Qualitative Health Research, 28*(2), 292–304. doi:10.1177/1049732317721477

- King, N. (1999). Template analysis. In C. Cassell & G. Symon (Eds.), *Essential guide to qualitative methods in organizational research* (pp. 118–134). London, UK: Sage.
- Kirk, S., Beatty, S., Callery, P., Gellatly, J., Milnes, L., & Pryjmachuk, S. (2013). The effectiveness of self-care support interventions for children and young people with long-term conditions: a systematic review. *Child: Care, Health and Development*, 39(3), 305–324. doi:<http://dx.doi.org/10.1111/j.1365-2214.2012.01395.x>
- Leung, L. (2015). Validity, reliability, and generalizability in qualitative research. *Journal of Family Medicine and Primary Care*, 4(3), 324. doi:10.4103/2249-4863.161306
- Malone, H., Biggar, S., Javadpour, S., Edworthy, Z., Sheaf, G., & Coyne, I. (2017). Interventions for promoting participation in shared decision-making in children and adolescents with cystic fibrosis. *Cochrane Database of Systematic Reviews*, (3), 1–21. doi:10.1002/14651858.CD012578
- Mays, N., & Pope, C. (2000). Qualitative research in health care: Assessing quality in qualitative research. *Bmj*, 320(7226), 50–52. doi:10.1136/bmj.320.7226.50
- Moola, F. J., & Faulkner, G. E. J. (2014). “A tale of two cases:” the health, illness, and physical activity stories of two children living with cystic fibrosis. *Clinical Child Psychology and Psychiatry*, 19(1), 24–42. doi:<https://dx.doi.org/10.1177/1359104512465740>
- Moola, F. J., Faulkner, G. E. J., & Schneiderman, J. E. (2012). “No Time to Play”: Perceptions Toward Physical Activity in Youth With Cystic Fibrosis. *Adapted Physical Activity Quarterly*, 29(1), 44–62. doi:10.1123/apaq.29.1.44
- Nuttall, P., & Nicholes, P. (1992). Cystic Fibrosis: Adolescent and Maternal Concerns About Hospital and Home Care. *Issues in Comprehensive Pediatric Nursing*, 15(3), 199–213.

doi:10.3109/01460869209078252

- Oakley, A. (1994). Women and children first and last: Parallels and differences between children's and women's studies. In B. Mayall (Ed.), *Children's childhoods: Observed and experienced* (pp. 13–33). London: The Falmer Press.
- Oliver, C. (2012). Critical Realist Grounded Theory : A New Approach for Social Work Research, (July 2018), 371–387. doi:10.1093/bjsw/bcr064
- Parliamentary, A. P., & Group on Global Health. (2014). A report by the All Party Parliamentary Groups on Global Health; HIV/AIDs; Population, Development and Reproductive Health; Global Tuberculosis; and Patient and Public Involvement in Health and Social Care. Patient empowerment: for better quality, more su.
- Pizzignacco, T. M. P., & de Lima, R. A. G. (2006). Socialization of children and adolescents with cystic fibrosis: Support for nursing care. *Revista Latino-Americana de Enfermagem*, 14(4), 569–577. Retrieved from <http://ovidsp.ovid.com/ovidweb.cgi?T=JS&PAGE=reference&D=med5&NEWS=N&AN=16967164>
- Potter, J. (1997). Discourse analysis as a way of analysing naturally occurring talk. In D. Silverman (Ed.), *Qualitative research: Theory, method, and practice* (pp. 144–160). London, UK: Sage Publications.
- Prigge, J. K., Dietz, B., Homburg, C., Hoyer, W. D., & Burton, J. L. (2015). Patient empowerment: A cross-disease exploration of antecedents and consequences. *International Journal of Research in Marketing*, 32(4), 375–386. doi:10.1016/j.ijresmar.2015.05.009
- Public Health Resource Unit. (2006). *Critical Appraisal Skills Programme (CASP)*

Qualitative Checklist. Retrieved from https://casp-uk.net/wp-content/uploads/2018/03/CASP-Qualitative-Checklist-2018_fillable_form.pdf

Rappaport, J. (1984). Studies in Empowerment. *Prevention in Human Services*, 3, 1–7.

doi:10.1300/J293v03n02_02

Ritchie, J., & Spencer, L. (1994). Qualitative data analysis for applied policy research. In A. Bryman & R. Burgess (Eds.), *Analysing qualitative data*. London, UK: Routledge.

Rowbotham, N. J., Palser, S. C., Smith, S. J., & Smyth, A. R. (2019). Infection prevention and control in cystic fibrosis: a systematic review of interventions. *Expert Review of Respiratory Medicine*, 13(5), 425–434. doi:10.1080/17476348.2019.1595594

Rowlands, J. (1995). Empowerment Examined. *Development in Practice*, 5(2), 101–107.

Retrieved from

<https://www.jstor.org/stable/pdf/4028929.pdf?refreqid=excelsior%3A6ad6e17fc21fe0ee58ffe8b2a4d10777>

Sandelowski, M. (2000). Whatever happened to qualitative description? *Research in Nursing & Health*, 23(4), 334–340. doi:10.1002/1098-240X(200008)23:4<334::AID-

NUR9>3.0.CO;2-G

Savage, E., & Callery, P. (2007). Clinic consultations with children and parents on the dietary management of cystic fibrosis. *SOCIAL SCIENCE & MEDICINE*, 64(2), 363–374.

doi:10.1016/j.socscimed.2006.09.003

Sawicki, G. S., Heller, K. S., Demars, N., & Robinson, W. M. (2015). Motivating adherence among adolescents with cystic fibrosis: youth and parent perspectives. *Pediatric Pulmonology*, 50(2), 127–136. doi:<https://dx.doi.org/10.1002/ppul.23017>

Sawicki, G. S., Ren, C. L., Konstan, M. W., Millar, S. J., Pasta, D. J., Quittner, A. L., &

- Fibrosis, I. and C. of the E. S. of C. (2013). Treatment complexity in cystic fibrosis: trends over time and associations with site-specific outcomes. *Journal of Cystic Fibrosis : Official Journal of the European Cystic Fibrosis Society*, 12(5), 461–467. doi:<https://dx.doi.org/10.1016/j.jcf.2012.12.009>
- Sawyer, S. M., Drew, S., Yeo, M. S., & Britto, M. T. (2007). Adolescents with a chronic condition: challenges living, challenges treating. *Lancet (London, England)*, 369(9571), 1481–1489. doi:[https://dx.doi.org/10.1016/S0140-6736\(07\)60370-5](https://dx.doi.org/10.1016/S0140-6736(07)60370-5)
- Small, N., Bower, P., Chew-Graham, C., Whalley, D., & Protheroe, J. (2013). Patient empowerment in long-term conditions: Development and preliminary testing of a new measure. *BMC Health Services Research*, 13(1), 263. doi:10.1186/1472-6963-13-263
- Stake, R. (2000). Case studies. In N. Denzin & Y. Lincoln (Eds.), *The handbook of qualitative research*. Thousand Oaks, CA: Sage.
- Stein, R. E. K. (1992). Chronic Physical Disorders. *Pediatrics in Review*, 13(6), 224–229. doi:10.1542/pir.13-6-224
- Stewart, L., Moher, D., & Shekelle, P. (2012). Why prospective registration of systematic reviews makes sense. *Systematic Reviews*, 1(1), 7–10. doi:10.1186/2046-4053-1-7
- Strauss, A., & Corbin, J. M. (1990). *Basics of qualitative research: Grounded theory procedures and techniques*. Newbury Park, CA: Sage.
- Taylor, R. M., Gibson, F., & Franck, L. S. (2008). The experience of living with a chronic illness during adolescence: A critical review of the literature. *Journal of Clinical Nursing*, 17(23), 3083–3091. doi:10.1111/j.1365-2702.2008.02629.x
- The British Psychological Society. (2018). Code of Ethics and Conduct. *The British Psychological Society*, (July), 100–102. Retrieved from www.bps.org.uk.

- The British Psychological Society (BPS). (2014). *Code of Human Research Ethics*. The British Psychological Society. Retrieved from http://www.bps.org.uk/sites/default/files/documents/code_of_human_research_ethics.pdf
- Thomas, J., & Harden, A. (2008). Methods for the thematic synthesis of qualitative research in systematic reviews. *BMC Medical Research Methodology*, 8, 1–10.
doi:10.1186/1471-2288-8-45
- Thomas, J., Sutcliffe, K., Harden, A., Oakley, A., Oliver, S., Rees, R., ... Kavanagh, J. (2003). *Children and healthy eating: A systematic review of barriers and facilitators*. London, UK.
- Tong, A., Flemming, K., McInnes, E., Oliver, S., & Craig, J. (2012). Enhancing transparency in reporting the synthesis of qualitative research: ENTREQ. *BMC Medical Research Methodology*, 12(Figure 1), 1–8. doi:10.1186/1471-2288-12-181
- UK Cystic Fibrosis Registry. (2013). *Cystic Fibrosis: Our focus*. London. Retrieved from <https://www.cysticfibrosis.org.uk/the-work-we-do/uk-cf-registry>
- Vines, E. E., Fisher, P., Conniff, H., & Young, J. (2018). Adolescents' experiences of isolation in cystic fibrosis. *Clinical Practice in Pediatric Psychology*, 6(4), 375–385.
doi:10.1037/cpp0000256
- Wakefield, D., Bayly, J., Selman, L. E., Firth, A. M., Higginson, I. J., & Murtagh, F. E. M. (2018). Patient empowerment, what does it mean for adults in the advanced stages of a life-limiting illness: A systematic review using critical interpretive synthesis. *Palliative Medicine*, 32(8), 1288–1304. doi:10.1177/0269216318783919
- Williams, B., Corlett, J., Dowell, J. S., Coyle, J., & Mukhopadhyay, S. (2009). I've never not

had it so I don't really know what it's like not to: nondifference and biographical disruption among children and young people with cystic fibrosis. *Qualitative Health Research*, 19(10), 1443–1455. doi:<https://dx.doi.org/10.1177/1049732309348363>

Williams, B., Mukhopadhyay, S., Dowell, J., & Coyle, J. (2007a). From child to adult: an exploration of shifting family roles and responsibilities in managing physiotherapy for cystic fibrosis. *Social Science & Medicine* (1982), 65(10), 2135–2146. Retrieved from <http://ovidsp.ovid.com/ovidweb.cgi?T=JS&PAGE=reference&D=med5&NEWS=N&AN=17719160>

Williams, B., Mukhopadhyay, S., Dowell, J., & Coyle, J. (2007b). Problems and solutions: accounts by parents and children of adhering to chest physiotherapy for cystic fibrosis. *Disability and Rehabilitation*, 29(14), 1097–1105. Retrieved from <http://ovidsp.ovid.com/ovidweb.cgi?T=JS&PAGE=reference&D=med5&NEWS=N&AN=17612996>

Williams, R., & Barker, H. (2010). Cystic Fibrosis. *InnovAiT: Education and Inspiration for General Practice*, 3(12), 743–752. doi:10.1093/innovait/inq072

World Health Organization Regional Office for Europe. (2013). *Health 2020: A European policy framework and strategy for the 21st century*. Copenhagen, Denmark: World Health Organization Regional Office for Europe.

Yeh, M. Y., Wu, S. C., & Tung, T. H. (2018). The relation between patient education, patient empowerment and patient satisfaction: A cross-sectional-comparison study. *Applied Nursing Research*, 39(45), 11–17. doi:10.1016/j.apnr.2017.10.008

Zimmerman, M. A. (1995). Psychological empowerment: Issues and illustrations. *American Journal of Community Psychology*, 23(5), 581–599. doi:10.1007/BF02506983

Part B

Abstract

Whilst the importance of patient empowerment is increasingly being recognised, little is known about empowerment in children and young people (CYP) with long term conditions. Empowerment may be particularly important in CYP with cystic fibrosis (CF) due to high treatment burden and limited opportunities for peer support.

This study employed a Grounded Theory Method to develop a preliminary theory of empowerment in CYP with CF. Seven CYP with CF, five parents and four professionals were interviewed.

The emerging model suggests that ‘thriving alongside CF’ may be supported by interactions between ‘having a team’ and ‘taking charge and having a voice’, leading to ‘being able to just be a child/getting on with life’, that ‘concealing self’ may get in the way of ‘thriving alongside CF’ and that these processes occur within wider medical and developmental contexts. Study limitations, clinical and research implications are discussed.

Key words: Cystic Fibrosis, Empowerment, Adolescents, Children

Introduction

Patient empowerment

Supporting increasing numbers of people living with long-term conditions (LTCs) is challenging for the NHS (Goodwin, Sonola, Thiel, & Kodner, 2013). Since the 1990s, policy has shifted towards collaborative healthcare models, with policies now referring to patient empowerment to enable patients with LTCs to better manage their health and achieve better outcomes (Small, Bower, Chew-Graham, Whalley, & Protheroe, 2013). Empowering patients is consistent with NHS values, e.g. 'improving lives' and 'respect and dignity' (Department of Health, 2015). However, clear definitions of patient empowerment are lacking, and the concept has been found to overlap with other concepts including self-efficacy, self-determination, self-management and autonomy (Cerezo et al., 2016).

Most patient empowerment literature has been written from a nursing perspective (Coyne, 2006; Gibson, 1991). Psychological theories of empowerment come largely from community psychology, e.g. Rappaport (1984) and Zimmerman (1995). There are several models of patient empowerment, though arguably, the most relevant to patients with LTCs is that of Bravo et al. (2015). This was based on interviews with key stakeholders: adults with various LTCs, health managers and researchers. Self-efficacy, knowledge and skills, perceived control, a sense of meaning, and feeling respected were identified as indicators of empowerment. Bravo et al. (2015) suggested empowered adult patients participate in shared decision-making, self-manage, and utilise support groups.

Much of what is known about patient empowerment focuses on adults. Although there is growing interest in patient empowerment in adolescents and a scale of empowerment has recently been developed for children and young people (CYP) with LTCs, this has been based on an adult patient empowerment model (Acuña Mora et al., 2018). Patient empowerment in relation to CYP is complex due to constant developmental changes, the relatively greater impact of systemic influences such as parents, teachers, and peers (Kirk et al., 2013) and the influences of the wider social contexts that impact CYP indirectly including cultural patterns and values, dominant beliefs and ideas, as well as political and economic systems (Bronfenbrenner, 1986). Empowering CYP with LTCs is essential for future engagement and has potential to impact significantly on long-term health outcomes. This is particularly important in cystic fibrosis (CF) due to the significant treatment burden and barriers to face-to-face peer support associated with risks of cross-infection (Cystic Fibrosis Trust, 2019).

Cystic fibrosis

CF is a multisystemic condition primarily affecting the lungs and digestive tract (Cystic Fibrosis Trust, 2019). CF is the most common life-limiting genetic condition affecting white populations: one in every 2,500 babies born in the UK have the condition (Cystic Fibrosis Trust, 2019; R. Williams & Barker, 2010). Fifty years ago it would have been unlikely for a child with CF to live to their 10th birthday (Coulthard, 2018). With universal screening, earlier diagnosis, new understandings and treatments there have been dramatic increases in life expectancy with over half of people with CF living beyond the age of 47 (Cystic Fibrosis Trust, 2019).

CF has a high treatment burden: daily therapies typically include chest physiotherapy, nebulised medication, pancreatic enzymes, nutritional supplements and antibiotics (Cystic Fibrosis Trust, 2019). Whilst there is variation in disease severity and progression, many individuals require frequent inpatient treatment throughout their lives.

Dramatic increases in life expectancy have opened up both new possibilities and new problems for CYP growing up with CF. Whilst people with CF live longer, they are often affected by significant morbidity (Coulthard, 2018). Improved survival means that they and their families must cope with the demands of living with the condition for longer, managing increasingly complex regimens (Sawicki et al., 2013).

Although there is some disagreement within the literature, it appears that the additional psychosocial challenges associated with having CF increase the risk of experiencing psychological distress (Cruz et al., 2009).

Developmental perspectives

According to Erikson's model of psychosocial development (1959), identity versus role confusion is the key conflict to be resolved during adolescence. For CYP with LTCs, adolescence is characterised by multiple physical, cognitive, social and emotional changes that must be negotiated alongside the medical condition (Sawyer, Drew, Yeo, & Britto, 2007). This often includes learning to accommodate the condition and accepting more responsibility for managing self-care into adulthood (Stein, 1992).

Little is known about the influences on and impact of empowerment in CYP with CF during adolescence, a period associated with life transitions such as changing schools, becoming more independent at home, developing friendships and romantic relationships, forming an independent sense of identity and planning for the future (Erikson, 1959). For adolescents with CF this period of rapid developmental change often coincides with worsening CF symptoms, e.g. cough and fatigue, increasingly frequent pulmonary exacerbations (episodes of intense disease activity) and associated increases in treatment burden (Hegarty et al., 2009). At a time when CYP would otherwise be becoming more independent and spending time with friends, deteriorating health could make them become isolated and more dependent on family, needing to stay at home or being admitted to hospital (Iles & Lowton, 2008).

Historically, CYP with CF benefited from peer support through residential social events. This helped to reduce isolation, protected against stigmatisation and increased quality of life through the sharing of adaptive coping strategies. In the mid-1990s, the dangers of cross-infection were recognised, resulting in strict infection control policies, including guidance that patients should never meet in person (Rowbotham, Palser, Smith, & Smyth, 2019).

Rationale

In summary, empowering patients is consistent with the aims and values of the NHS. Much of what is known about patient empowerment relates to the experiences of adult patients. Due to medical advances there have been dramatic increases in longevity amongst patients with CF, a condition with a high treatment burden, typically diagnosed at birth. To the author's

knowledge, there are no existing studies of empowerment in CYP with CF. Given that current theory is insufficient, it was felt helpful to use grounded theory (Corbin & Strauss, 2015) to develop new theory rather than attempt to force data into existing theories of patient empowerment that have largely been developed from experiences of adult patients with other LTCs.

Aim

This study aimed to develop a grounded theory of perceived facilitators of, barriers to, and consequences of empowerment in CYP with CF during key life transitions.

Research questions:

- 1) How do CYP with CF experience empowerment during key life transitions?
- 2) What are the perceived facilitators of empowerment during these transitions?
- 3) What are the perceived barriers to empowerment during these transitions?
- 4) What are the perceived consequences of empowerment during these transitions?

Method

Design overview

A qualitative approach was adopted to enable a detailed exploration of participants' views and experiences. Grounded Theory Birks and Mills (2015) was selected as a methodology that can be particularly useful where little is known about the study area, where the

phenomena of interest involves a process, and where developing new theory with explanatory power is a desired outcome.

Interviews were conducted using semi-structured interview schedules, giving focus to the interviews while allowing participants freedom to describe their subjective experiences in their own words. This method, along with line-by-line analysis of the data, aimed to give a voice to CYP with CF and the adults that support them (Charmaz, 2006).

Epistemological position

Grounded theory can be conducted from a range of philosophical and methodological positions (Birks & Mills, 2015). This study employed a critical realist approach, assuming that whilst an objective reality exists, it can only be made sense of and described through the lenses of language and social context (Oliver, 2012). This approach suited the researcher's philosophical standpoint and is likely to have more utility than a social constructionist perspective in medical settings, which tend to be predominantly realist.

Patient and public involvement

Feedback was sought from the Cystic Fibrosis Trust (UK based charitable organisation) patient and public involvement (PPI) lead, suggesting that research into empowerment in CYP with CF, would be of interest to its members. CYP from a youth advisory panel (based at a children's hospital not involved in the research) commented on the CYP information sheets, assent/consent forms and draft interview schedules. Feedback was incorporated into

final versions of these documents. Attempts were made to gain similar parental input, however this was hampered by the local PPI group disbanding.

Sampling

Following principles of grounded theory, sample size was not prospectively determined. Data collection and analysis was conducted concurrently, later participants were recruited according to the likelihood that they would be able to answer questions raised by earlier interviews and fill gaps in the emerging theory including possible exception (theoretical sampling). Participants were continuously recruited on the basis of theoretical sampling until no new concepts arose during analysis of successive interviews. See Appendix E for a flow chart detailing theoretical need for participant selection and the order in which this occurred).

Strauss and Corbin (1990) used the term ‘theoretical saturation’ to describe the point where no new codes are identified in later rounds of data collection, and where categories are developed to the point that properties (characteristics) and dimensions (variations) of all categories are well defined and integrated within the model. Dey (1999) highlighted the subjective nature of ‘theoretical saturation’, arguing that data collection and analysis could go on indefinitely and researchers should instead be aiming for ‘theoretical sufficiency’.

Theoretical sufficiency is the point at which the emerging theory is considered to have good explanatory power.

Following joint coding of extracts from several transcripts with trainee research peers, asking questions such as ‘what processes are occurring here?’ and ‘when, why and how does the process change’ (Charmaz, 2006, p. 51) and discussion about developing categories in

supervision, theoretical sufficiency was considered to be attained at 14 interviews; two subsequent interviews yielded no new concepts.

Participants

Participants were recruited from two NHS paediatric CF services in England. One service was based within a children's hospital where patients typically transferred to adult CF services aged 17-19; the other was based within a large regional hospital where patients typically transferred at 16.

The researcher attended multi-disciplinary team meetings to introduce the research. CF teams were emailed information (Appendix F), including information sheets (Appendices G-J), and consent/assent forms (Appendices K-N). Eligible participants recruited via the NHS were identified by CF clinicians using clinic databases. Inclusion/exclusion criteria are shown in Table 1.

As recruitment was initially slower than expected, further ethical approvals were obtained to expand recruitment to CF charities and social media, though this did not recruit further participants. The main barriers to participation were thought to be pre-existing pressure on CYP and their families due to the high treatment burden of CF, and competing requests for biomedical research with greater personal benefit (e.g. inclusion in novel drug trials).

Table 1

Inclusion and exclusion criteria

<u>Inclusion</u>	<u>Exclusion</u>
Children and young people (CYP) aged 11-19 years with a diagnosis of cystic fibrosis (CF)	Insufficient command of English language or cognitive difficulties that would affect the interview process.
Parents/guardians of eligible CYP	CYP person has significant non-CF related co-morbidities
Professionals working with CYP with CF	Families considered by clinical team to be currently too vulnerable (for whatever reason) to participate in research.
Under the care of (CYP with CF) or known to (professionals and parents/guardians) an NHS paediatric CF team in the UK	Families associated with significant risks identified by the clinical team (e.g. risk of aggressive behaviour).
	Professional has under six months' experience of working with CYP with CF.
	CYP who have already transferred to adult CF services.
	Professionals working solely in adult CF services

Sixteen participants were included in total; participant demographic and health information are shown in Tables 2-4 (data amalgamated to protect confidentiality).

Table 2

Young person demographic and health information

Age	Range: 12-18 Mean: 14.7
Gender	Male: 3 Female:4

Ethnicity	White British: 7
Age at CF diagnosis	Newborn: 4 ≤ 2 years: 2 ? ≤ 5 years: 2
FEV-1 ^a	Range: 79 – 122% Mean: 100.5%
CF related comorbidities & procedures	CF related diabetes; intestinal obstruction; gastrostomy; ACE procedure; ileostomy; lobectomy; microcytic anaemia
Number of episodes of IVs in previous year	Range: 0 – 4 Mean: 1.5
Number medications	Range: 5 – 15 Mean: 11

^a FEV₁ (forced expiratory volume in 1 second) is a measure of CF disease severity. An FEV₁ of 100% means that lung function is as would be expected for a person of the same age and height who doesn't have CF. FEV₁ ≥ 85% is considered normal; 70-84% indicates mild lung disease, 41-69% indicates moderate lung disease; ≤ 40% suggests severe lung disease (UK Cystic Fibrosis Registry, 2013).

Table 3 <i>Parent/carer demographic information</i>	
Relationship to child with CF	Father: 1 Mother: 4
Gender	Male: 1 Female: 4
Ethnicity	White British: 5
Age of children	Range: 11-18 Mean: 13.8

Table 4 <i>Professional demographic information</i>	
Profession	Dietician Physiotherapist Specialist nurse Pharmacist
Years of experience	1-5 years: 1 6-10 years: 1 11+ years: 2

Interviews

A flexible interview schedule was developed in accordance with the research questions. Interviews took place on NHS premises, familiar to participants or via telephone. Duration was from 17-56 minutes. Participant comfort was prioritised; CYP were first asked about their general interests to build rapport (Charmaz, 2006). Similarly, to ease adult participants into the interview process, initial questions were more general and related to CYP's interests. Initial interview schedules for each interview group are included as Appendices O-Q. Topics included: times when CYP feel most confident, accessing information, handling change, managing CF treatment, hopes for the future and messages for people wanting to help CYP feel more empowered.

To avoid social desirability and accommodate varying cognitive and developmental levels, concrete questions were initially used, progressively addressing more abstract concepts. Intermediate questions were designed to be unobtrusive and flexible; questions were guided by participants' responses and appropriate prompts used throughout. This flexible approach enabled detailed exploration of experiences and helped prevent the researcher imposing preconceived ideas (Charmaz, 2006). Final questions moved away from more sensitive experiences which was deemed particularly important for younger participants. Throughout the interviews, reflection and paraphrasing were used to check for inconsistencies within the data and clarify meaning.

All interviews were audio-recorded and professionally transcribed (see Appendix R for confidentiality agreement).

Ethical considerations

Ethical practice was guided by the BPS code of human research ethics (The British Psychological Society (BPS), 2014) and the BPS code of ethics and conduct (BPS, 2018).

Ethical approval was granted by a local NHS ethics committee (Appendices S-T) and the Health Research Authority [HRA] (Appendix U). Research and development (R&D) approval was granted by two NHS Trusts (Appendices V-W).

Informed consent/assent

Families and professionals were introduced to the researcher by a member of the CF team. The researcher explained the study and answered any questions. This included details about confidentiality, rights to withdraw from the study and storage and use of data. Information was also provided in written format (Appendices G-J).

Potential participants were given a minimum of 24 hours to decide whether to participate. Children under 16 whose parent/guardian had given consent were asked for their assent, particular care was taken to avoid children feeling undue pressure to participate. Over 16s consented for themselves.

Managing potential harm to participants

As talking about empowerment/disempowerment in the context of growing up with CF can be sensitive, the study was designed such that participants were only exposed to experiences that were already known to them (their own experiences or experiences that they had witnessed or been told about relating to their child or a child that they had cared for professionally). No participant was required to discuss anything they were uncomfortable with.

Local CF specific infection control policies were complied with at all times, e.g. participants with CF were not seen back-to-back in the same room, nor asked to wait in the same waiting area as others with CF.

To increase convenience and reduce infection risk, all participants were given the opportunity to provide written consent electronically and to participate in interviews via videoconferencing or telephone if preferred.

Special considerations – working with children

Given the specific ethical and methodological issues raised with conducting research with and for CYP, care was taken to explain the research in an accessible way, to avoid ‘pseudo-friendships’ and to acknowledge and minimise power differentials between researchers and young participants (Oakley, 1994).

Data analysis

The data were analysed using grounded theory as described by Corbin and Strauss (2015).

This approach was in line with the epistemological stance of the researcher and has previously been used in research exploring the experiences and perspectives of individuals with physical health conditions (Corbin & Strauss, 1991). This method utilises three stages of coding: open, axial and selective. This process is outlined in Table 5; a more detailed coding table and example transcript are included in Appendices X-Y.

Codes were continuously reviewed throughout data analysis, particular efforts were made to find comments appearing to contradict the developing theory, consider alternative interpretations, and adjust codes accordingly. The NVivo 12 software package was used to facilitate data analysis.

Table 5	
<i>Description of the analytic process</i>	
Initial 'open' coding	<p>Audio files were checked against verbatim transcripts and coded line-by-line enabling the researcher to become thoroughly immersed in the data.</p> <p>This initial coding was used to break the data down and allow segments to be compared with other segments, between and across transcripts (Glaser & Strauss, 1967).</p> <p>During open coding the researcher stuck closely to the data and was particularly careful to reflect on their own assumptions when making early analytical decisions (Corbin & Strauss, 2015).</p>

Concurrent data collection and analysis	<p>Data was analysed after each interview.</p> <p>In each case the researcher asked ‘what is the data saying and where are the gaps?’ (Glaser & Strauss, 1967).</p> <p>On the basis of this, interview questions were refined and ‘theoretical sampling’ was used to identify which other potential participants to interview.</p>
Theoretical sampling	<p>Additional participants were recruited according to likelihood that they would be able to answer some of the questions raised by other interviews (Birks & Mills, 2015).</p>
Intermediate ‘axial’ coding	<p>Axial coding was used to reassemble the data that was fractured during open coding at a more conceptually abstract level (Corbin & Strauss, 2015).</p> <p>First categories were developed by linking subcategories and considering their properties and dimensions. Categories were then linked together (Birks & Mills, 2015).</p> <p>The main question here was what are the relationships between categories (who, what, when, and how) (Corbin & Strauss, 2015).</p>
Selective coding and theoretical integration	<p>Core category identified and systematically related to other categories (Corbin & Strauss, 2015).</p> <p>Storyline technique used as a mechanism for theoretical integration (Corbin & Strauss, 2015).</p>
Theoretical sensitivity	<p>Idea that all researchers have their own way of seeing the world, influenced by their own experiences and things they’ve read.</p> <p>As data analysis progresses, they will become more sensitive to emerging concepts that resonate with their own previous experience</p>
Memo writing and diagrams	<p>Diagrams and memos were used to note emerging ideas and track theory development, acting as a bridge between data collection and theoretical analysis.</p> <p>Memos are used throughout the process to record initial reflections, and as analysis continued links between increasingly abstract concepts (Birks & Mills, 2015).</p>
Constant comparison	<p>Throughout the process data was constantly compared within and between all three levels of coding and across memos.</p>

Quality assurance

There is a lack of consensus around criteria and processes for evaluating qualitative research (Thomas & Harden, 2008). This report was guided by a tool for appraising qualitative research developed by the Critical Appraisal Skills Programme [CASP] (Public Health Resource Unit, 2006), guidance provided by Mays and Pope (2000), and the principles of good practice for conducting research with children described in Thomas et al. (2003).

In keeping with the critical realist stance, researcher reflexivity was prioritised. Reflexivity refers to the consideration of the ways that the researcher's prior experiences, biases and assumptions may impact on the research process at all levels from initial proposal to dissemination (Mays & Pope, 2000). The researcher was mindful of how her own preconceptions might influence the research. She mitigated the projection of her own understandings through the use of a research diary (Appendix Z), bracketing interviews (Appendix AA), active listening, adhering closely to the data and making constant comparisons within and between transcripts. While the researcher did not discuss her own health with participants, her own visible disability may have affected participants' disclosures during interviews and allowed greater sensitivity to some of the socially contextualised concerns participants raised.

Triangulation between interviews with CYP, parents and professionals, and the use of diagrams (Appendix BB) and theoretical memos (Appendix CC) also added comprehensiveness and supported researcher reflexivity (Mays & Pope, 2000)

Feedback to stakeholders

Findings were shared with relevant stakeholders including: NHS ethics committee, HRA, relevant R&D departments (Appendix DD); all participants (Appendix EE); and services in which the research was conducted.

Results

Due to space constraint, some subcategories will be described in more detail than others. The focus will be on categories that address the research questions most directly, the aspects that are not explained in previous literature, and those that are most relevant for clinical practice, future research and policy.

Table 6: Selective and axial codes	
<u>Selective code</u>	<u>Axial codes</u>
Having a team	Team membership Provides a sense of acceptance/ 'normalising' Share burden of CF Makes room for mistakes/ learning from experience Trusting relationships
Taking charge and 'having a voice'	Information and understanding Active participation in decision-making Explaining to others 'Finding ways round and through'/ practical problem solving

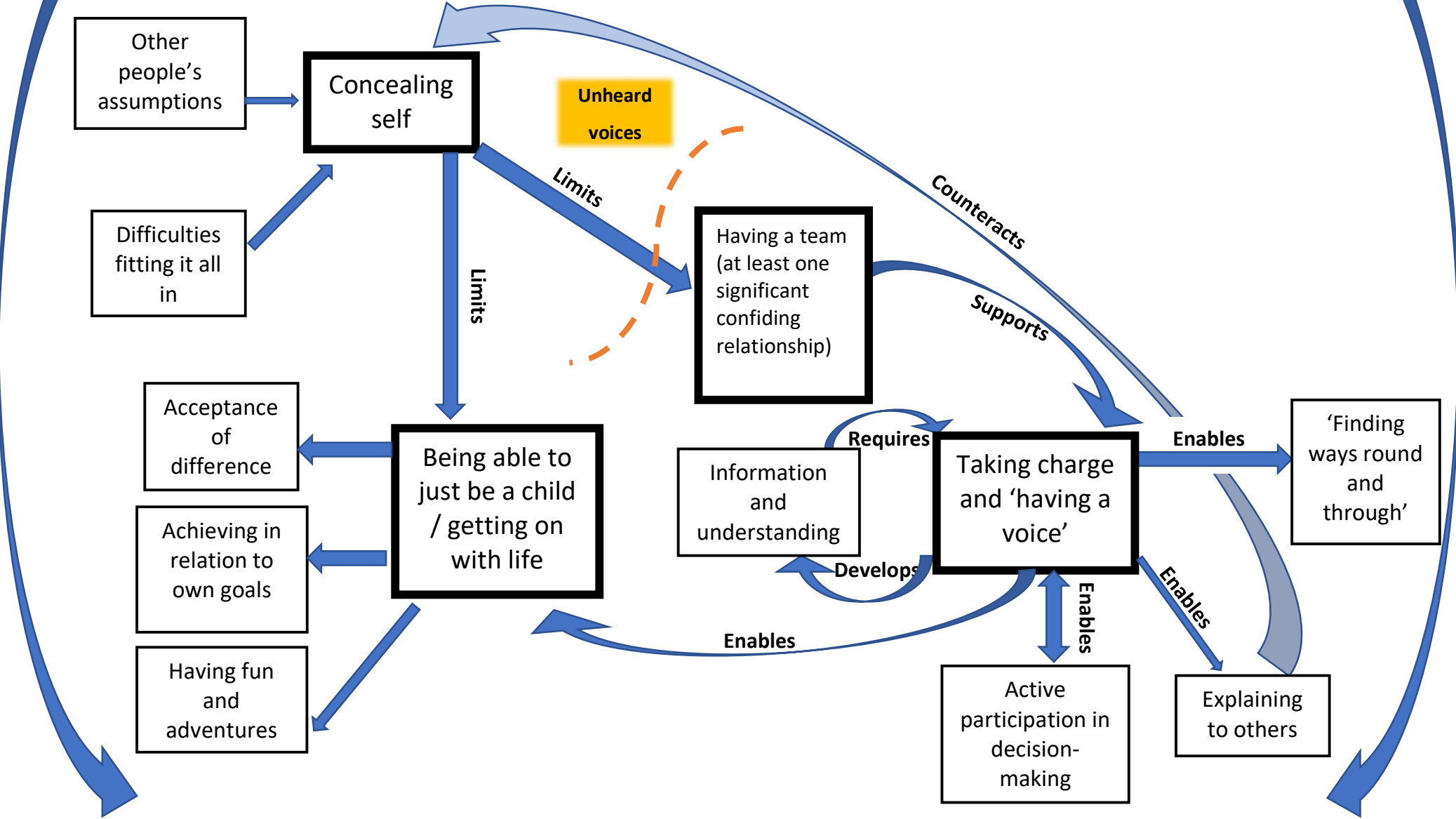
Being able to just be a child/getting on with life	Self-acceptance/dealing with difference Achieving in relation to own goals Having fun and adventures
Concealing self	Other people's assumptions Difficulties fitting it all in Concealing self Unheard voices
Wider context	Unique individuals Interaction with developmental changes and stages Constantly changing landscape of CF

Overview of the model

The model presented in Figure 1 illustrates the phenomenon of **'thriving alongside CF'**.

Categories and subcategories are highlighted in bold. Quotations were selected to best represent the categories while demonstrating the range of views expressed, and are italicised in the text.

Wider context: - Unique individuals;
- Interaction with developmental changes;
- Constantly changing landscape of CF



The core category that emerged from the data was **‘thriving alongside CF’**. Five higher level categories (selective codes) appeared to influence **‘thriving alongside CF’**: **having a team, taking charge and having a voice, being able to just be a child/getting on with life, concealing self and the wider context.**

Two higher level categories appeared to support **‘thriving alongside CF’**: **having a team, and taking charge and having a voice.** These categories seemed to interlink with each other and enable **being able to just be a child/getting on with life.**

Concealing self appeared to obstruct **‘thriving alongside CF’** because having to conceal oneself limits **having a team** and **being able to just be a child/getting on with life.**

‘Thriving alongside CF’ also appeared to be influenced by **wider contextual factors.**

‘Thriving alongside CF’ occurs in the context of constant developmental changes, changes in the condition and treatments at the individual level, and constant developments in biomedical understandings and CF treatment.

Core category

The core category which links all the data together is **‘thriving alongside CF’**. This refers to CYP being able to live without their diagnosis defining them or preventing them from making choices that impact on how they live.

'they're happy... they live their life, and CF is a part of it, but CF isn't their whole life' (Professional)

Professionals noted that there is a group of CYP with CF who aren't currently **'thriving alongside CF'**. Due to the nature of this research, these voices remain silent and are referred to as **'unheard voices'**.

Having a team

All participants referred to the importance of CYP with CF having a support network. CYP, parents and professionals all used 'team language' for example *'we're very much a team'* giving rise to the in vivo code (codes that place the emphasis on the words spoken by participants) 'having a team'.

Team membership

Family, friends, the medical team, and school were key members of the team for many CYP. For some, a connection with God and a faith community were important, for others, sports clubs or pets.

Trusting relationships

All participants referred to the importance of trusting relationships. CYP and their families valued enduring friendships and consistency of medical team members.

'it's a good feeling when there's someone there who's been with you since you were born' (YP 17, M)

Having a team of trusted people facilitated open and honest conversations. Whilst having friends in general was beneficial having **'at least one good friend'** seemed particularly important and links to the subcategory (axial code) of **'explaining to others'**.

'Get a really close friend... one person you can really trust and tell them' (YP 15, F)

CYP varied in how open they were about CF. Some chose to tell one or two carefully selected friends or teachers on a strictly need-to-know basis.

'I mean, we've got some children who... have... a couple of best friends who know... they don't want it blazoned across... for all in sundry to hear about it.' (Professional)

Others spoke proudly of the confidence they had gained through standing up in front of large groups and raising awareness of CF, for example in assemblies at school

'Once it was off my chest I felt so much better, and they know what I'm going through and why I'm missing school so much' (YP, 13 F)

Having **'at least one good friend'** protects against **'having to keep secrets'/concealing self.**

'we definitely see a difference in the children who, for example... haven't told their friends at school and have to take their Creon in secret and they find that it's... always stressful' (Professional)

Sharing the burden of CF

CF was described by CYP and parents as too much for any one person to manage.

'It's so important that it's not just down to us parents, it's not just down to [child], it's a team effort, work with the CF team as well, you've got their support, their backing, their help, their guidance.' (Parent)

The CYP's team has several roles which may be fulfilled in different ways by different members at different times. Several participants described how having a team helped share the burden of CF through practical and emotional support.

'If I'm down... the dumps, my family and my friends always help me out. And my little pet lizard always keeps me calm' (YP, 12, M)

Several CYP explained that although they completed all aspects of their CF regime, doing all of the CF-related tasks completely independently was not their priority. Sharing the burden of CF with help from family, friends and the medical team to complete tasks provided CYP with the time and energy to do other things that were important to them.

'I can do my IVs myself, but I choose not to... My mum will do them if I'm at home. But if I'm in hospital, the nurses will.' (YP, 18, F)

For some CYP the CF community formed an important part of their team

'It's nice to know that people have the same thing and that can relate to... what we're going through' (YP, 14 M)

Others expressed a yearning for connection with other CYP with CF.

'I know absolutely no-one whatsoever... I would love to know someone my age and talk to them... – not necessarily about CF – but talk to someone daily, how you would talk to anyone else that's in the same position as you. Because you can have really

supportive people around you, really amazing people, but they're not in the same position as you' (YP, 14 F)

Providing a sense of acceptance/'normalising'

One of the team's roles was providing a sense of acceptance. Many CYP spoke of the importance of feeling 'normal'. Feeling accepted at home, with friends and at school was experienced as 'normalising' by many CYP.

'I'd class my friends as normal, when I'm out with them not worrying about my health, like I'm not doing my medication then, I'm not in hospital then, I'm more classed as normal, and that's quite nice.' (YP 18, F)

Making room for mistakes and learning from experience

Another theme that emerged, primarily from interviews with parents and professionals, was the team's role in making room for mistakes and allowing CYP to learn from experience. This was the case both in relation to the lessons CYP needed to learn for themselves (as opposed to being told) in directly managing CF, as well as generic learning experiences which would have an added layer of complication for CYP with CF.

'I've certainly had a couple of chats about alcohol with our older teenagers. It's generally quite positive. They're teenagers, they're going to experiment.... So I try really hard to show that I'm not judging them and I'm not going to tell them off.'

(Professional)

Some of the professionals reflected on how taking on the role of a coach facilitated the CYP taking charge.

'team members to act like coaches, and to allow the children, as they get older... to feel like they can ask really simple questions, and... can tell us and be honest about their compliance with treatments... and be honest about when they're really struggling.' (Professional)

Many of the adult participants described how allowing CYP to learn from experience both required and built trusting relationships.

'Whatever's going on, we can support you and if you want to make a change or get back on track, whatever direction you want to take following whatever's happened, we're here to support you' (Professional)

Several parents and the three oldest CYP with CF reflected on the role that their families and medical team had in enabling learning from experience.

'he knows if he doesn't take his medication he will get poorly... cos he's tried it once before, he's hidden his tablets when he was younger' (Parent)

'stop badgering...Remind me a few times, but it's on me if I don't do it' (YP, 17 M)

Taking charge and having a voice

‘Having a team’ supports **‘taking charge & having a voice.’** All participants referred to taking charge and developing a voice, both directly in relation to the medical context and more broadly. This was something that interacted with developmental changes and evolved over time. Taking charge and ‘having a voice’ appeared to both require and develop information and understanding.

‘I think if they take charge of it themselves... be more independent... understand it and do more treatments for themselves, I think that’s a big help for children’ (Parent)

Professionals spoke of a wide range in levels of understanding about CF amongst the CYP that they work with.

‘there’s a huge range of between what the level of detail they know about their condition’ (Professional)

Professionals and parents noted the role of parents in passing on information about CF to CYP, with professionals in particular reflecting on how the informational needs of CYP change over time.

'it's quite up to the parents to give the information' (Parent)

'Some of our parents are very open with their children about the condition and complications... Others are very secretive about it and they don't want to talk about certain things in front of their children... they don't want to worry their child about things that might happen' (Professional)

All professionals described strategies that they had developed to encourage CYP to take ownership of their own understanding of CF/CF treatments – for example developing tailored information sheets and encouraging CYP to ask questions in clinic.

A number of CYP made references to the difference between getting information from professionals or information sheets and getting information from others with lived experience of CF.

'someone who has the experience to know... instead of... people who have either known people with CF or have just found things out from reading things. It would have been nice to have had a first-hand experience' (CYP 14, M)

Approximately half of the CYP interviewed were connected to other CYP with CF through internet based support groups which acted as a source of information and social interaction. For those who did not have any contact with others with CF, this was experienced as a loss

with CYP feeling they were missing out on an important source of information and connection.

'Because I'm not allowed near any other person with it... it's a bit concerning, and upsetting... then I can't talk to people who understand what I'm going through' (CYP 12, M)

A range of views were expressed by parents and professionals about information from the internet, including from support groups. Some had had their own positive experiences of connecting with others in similar circumstances; others feared that support groups could be the source of frightening or inaccurate information, or encourage non-compliance.

'she's quite well informed but I don't think she knows everything, all the different paths that the future could lead to.... if you connect with other people you don't know what they know... she's still very young, in terms of having to sort out if some people are rebelling against regimes... I'm just wondering if that's unhelpful as well as helpful.' (Parent)

Having information and understanding seemed to enable taking charge and having a voice which in turn seemed to enable active participation in decision-making.

'I think the first step is understanding what it is... and from there, you can make choices' (CYP 15, F)

Many participants stressed the importance of having information so that the CYP could make decisions based on their individual preferences and circumstances. This included both treatment-related decisions such as medication regime timings and broader life decisions such as career choices.

‘Taking control of their CF and being able to have a voice in the management and direction that they take...being able to manage it alongside all the rest of their life... to make the decisions that help them direct their journey through it’ (Professional)

Taking charge and ‘having a voice’ appeared to enable ‘finding ways round & through’/practical problem solving. All of the CYP and parents gave examples of finding ways of doing the things that are important to them despite the significant challenges that CF and associated treatment regimens can present. For many, having a routine facilitated this. Others described emotional coping strategies and creative solutions to practical problems.

‘We have done things like administering IV antibiotics in the car, before she went in to a party’ (Parent)

Taking charge and ‘having a voice’ seemed to enable explaining to others which appeared to help counteract other people’s assumptions.

'I basically said, "If I cough, it's not contagious." And as the weeks went on, they found out a little bit more (YP 18, F)

Taking charge and 'having a voice' appeared to enable 'being able to just be a child/getting on with life'.

Being able to just be a child/getting on with life

'Being able to just be a child/getting on with life' seemed to be a consequence of 'thriving alongside CF'. This was something that many of the CYP and their families strived for.

'our attitude is, that if you've done all your treatments then we can do anything else... let's get everything done, out the way, forget about it, and now crack on with normal stuff' (Parent)

'he needs to live a full life and be like any other child' (Parent)

'Being able to just be a child/getting on with life' seems to require 'acceptance of difference'.

'if I never had CF, I would never be me' (YP, 13 F)

Acceptance of difference appeared to enable 'achieving in relation to own goals' and 'having adventures and fun' both of which were described as examples of 'being able to just be a

child/getting on with life'. For some CYP, 'achieving in relation to own goals' meant spending time with friends, others accomplished this through sport or academic achievements, or through raising awareness about CF.

'recently where she is starting to feel more comfortable about herself, her body, how it looks, how she's feeling... she actually did a talk in one of her lessons' (Parent)

'to be able to do all the things they want to do in life... relationship building or ...a particular career pathway' (Professional)

Concealing self

'Other people's assumptions' and/or 'difficulties fitting it all in' can put CYP in the position of having to making difficult choices and may lead to 'having to keep secrets' or concealing aspects of themselves. 'Other people's assumptions' described by participants included misunderstandings about what CF is (e.g. contagious), low expectations (e.g. in relation to sport), and judgements (e.g. teachers and peers making comments about CYP with CF eating 'unhealthy' foods, when in fact, a high fat / high calorie diet is medically necessary).

All participants referred to the 'difficulties fitting it all in', and many CYP described arriving home exhausted and having to make a choice between completing homework or their full CF regimen.

'it is hard to fit homework in as well as medicines' (YP 13, F)

Some referred to difficulties in having honest conversations with the medical team when it isn't possible to fit everything in. This seemed to relate to fear of offending or disappointing the medical professionals that CYP and families valued so much as part of their team.

'I think she is quite conscious that everyone's there to help her so she's worried about offending them if she appears ungrateful... she hates so many of her treatment... I think she finds it hard to talk honestly about that without offending them' (Parent)

All of the professionals and some parents reflected on some CYP 'having to keep secrets.' This was described as stressful for the CYP and appeared to get in the way of 'thriving alongside CF'

'they feel embarrassed about some of the things they have to do, and so they don't tell people. I think that... just increases the stress' (Professional)

'Having to keep secrets/concealing self' appeared to get in the way of developing 'trusting relationships' and seemed to limit 'having a team' as CYP may be unable to test out whether they will be accepted.

‘she felt quite embarrassed about that [side effect of medication] and so we were thinking about ways you could open up and own that a bit without feeling mortified. I don’t know if... you can ever... be so bold and say “... it’s my illness, it happens from time to time! Let’s all get over it and have a laugh and just move on”’ (Parent)

The accounts of professionals and some parents suggested that ‘having to keep secrets’ may limit ‘active participation in decision-making’

‘She will just nod and say yes and be polite. I don’t know how much she really feels able to really tell them [CF team] truthfully’ (Parent)

Whilst some of the CYP reflected on times when they had felt that they needed to conceal aspects of themselves and keep secrets, the CYP interviewed spoke of this as something they had done in the past. A sense of the CYP currently feeling they have to conceal aspects of themselves (as opposed to those making an active choice to keep some information private) came through the professionals and parents of CYP who opted not to participate. This gave rise to the axial code ‘unheard voices.’

‘I guess it’s a bit of a shame... but it is the young people that do quite like to talk that have got involved [in the research] ... it would be nice to hear their [those that didn’t take part] voice and for them to say, “Actually, I’m just completely pissed off with having CF and I don’t want to talk about it”.’ (Professional)

Thriving alongside CF was described by participants as occurring in the wider context of all CYP with CF being unique individuals, with unique constellations of experiences, hopes, fears, challenges, preferences and goals.

'It's one of those things that people have their own... definition of. Like, what is empowering to one person won't be to the other' (YP 18, F)

There also seemed to be an interaction with developmental stages and changes – CYP growing up with CF are constantly changing in terms of levels of cognitive, emotional and physical maturity. All participants spoke of the additional struggles faced by CYP with CF as they navigate the typical challenges of adolescence. Professionals in particular, highlighted the impact of uncertainty and the decline in health that many older adolescents face just when peers without CF will be in their physical prime.

'Puberty, changing peer groups, family dynamics, all those [things that]... children without CF, have to deal with... then I guess the changes in health that they may or may not suffer with' (Professional)

In addition to the constant changes in the condition and treatments at the individual level, 'thriving alongside CF' also occurs in the context of constant developments in biomedical understandings of and treatments for CF. Although this appeared important in maintaining a sense of optimism for the future for some participants, many CYP spoke of acceptance and

living more in the moment. Some spoke of difficulties in keeping up with constant medical developments

'It's a bit confusing at first because when you're used to the old stuff, then they give you new stuff, I kind of get a bit worried that it's gonna be hurting or... it's not gonna work because it's brand new, and they've not tested it properly yet' (YP, M, 12)

Discussion

Research has focused on parents and families of children with chronic conditions – few studies have focused on the children's own perspectives (Carter et al., 2017). Five main themes emerged from the stories told by the CYP, parents and professionals about the process of **'thriving alongside CF': having a team, taking charge and having a voice, being able to just be a child/getting on with life, concealing self and the wider context.** The model 'thriving alongside CF' resonated both with broad psychological theories such as empowerment theory and social ecological theory, and with previous findings in CF-specific literature.

Empowerment theory

The model is consistent with Rappaport's (1984) description of empowerment. Both Rappaport's empowerment theory and the 'thriving alongside CF' model describe a process by which people gain mastery over their lives. Rappaport asked the question 'under what conditions do we find people reporting a sense of control over their lives?' The model provides some answers to this question in relation to CYP with CF. Similarly to Rappaport,

the model suggested that there are often multiple possible solutions to challenges, that different solutions suit different individuals and that professional input may open up more possibilities.

The model is also consistent with Zimmerman's (1995) conceptualisation of psychological empowerment, in that 'thriving alongside CF' was found to take different forms in different CYP, can take different forms in different contexts (for example at home, at school and in clinic) and fluctuates over time.

Zimmerman (1995) made the distinction between empowering processes (in which individuals are given opportunities to influence decisions that affect their lives and access to resources that enable them to move closer to their goals) and empowered outcomes (mastery and control, resource mobilisation and participation) both empowering processes and empowered outcomes are reflected in the model of 'thriving alongside CF'.

Other models of patient empowerment

There was some overlap between the current model and models of patient empowerment based on research with adult patients (e.g. Bravo et al., 2015; Small et al., 2013), particularly in relation to having information and decision-making.

There were also some significant differences. Some of these relate to developmental stages – for example feeling or being perceived to be 'different' and confronting other people's assumptions are both likely to be more of a challenge for CYP than for adults, and children

are more likely to be dependent on adults for reliable sources of information (Ernst et al., 2011).

Other differences were specific to CF. For example the impact of infection control guidelines and the limited opportunities for some CYP to connect with others with CF (Vines, Fisher, Conniff, & Young, 2018) meant that the role patient peer support was more complex than is often reflected in models of empowerment based on other patient groups.

Links to the CF literature

The model is also consistent with previous research involving CYP with CF. For example, Barker, Driscoll, Modi, Light, and Quittner (2012) described the importance of family and friends in successfully managing CF during adolescence. Parents in the Sawicki, Heller, Demars, and Robinson (2015) study made reference to the role of allowing adolescents to make and learn from mistakes in promoting adherence. Adolescents and their parents in Dashiff, Suzuki-Crumly, Kracke, Britton, and Moreland, (2013) acknowledged the importance of understanding the medical aspects of CF when making decisions, and highlighted a need to make information more accessible to children and their families. Children participating in D'Auria, Christian, and Richardson (1997) study spoke of the process of deciding if and who to share information about CF with. However, in contrast to the current study, the D'Auria et al (1997) participants generally agreed that it was better to keep CF a secret. One explanation for this difference is that participants in the current study were older, indeed some reflected on having been afraid to tell their peers when they were younger.

There are numerous references in the literature to limited time and the burden of CF and CF treatment within the context of short lives (e.g. Barker et al., 2012; Foster et al., 2001).

Whilst this clearly featured in the current study, there was much more of a sense of optimism and expectation of surviving and thriving into adulthood.

In contrast to previous studies focusing on adherence in CF for example: (Sawicki et al., 2015) the current study suggested that CYP taking full responsibility for CF care may be neither achievable nor desirable.

What this study adds

Increasing longevity associated with recent medical advances is affording many CYP living with CF opportunities not previously considered possible. All participants made references to hopes for the futures of the CYP with CF. This included hopes for careers, getting married and having families of their own – all of which would have been unthinkable until relatively recently and are therefore not accounted for in much of the previous literature.

Given the prominence of patient peer support in previous models of patient empowerment, it is particularly important to highlight the impact of infection control guidelines limiting physical contact between CF. These guidelines are still relatively recent and whilst other research on the impact of isolation is beginning to emerge (Vines et al., 2018) the current study adds an additional perspective.

To the author's knowledge, the impacts of developmental stages and changes, the progressive nature of CF and the particularly rapid nature of current advances in CF care have not been accounted for in previous models of patient empowerment.

Strengths and limitations

Strengths of the current study included actively involving stakeholders (CYP and clinicians) in the design and implementation of the study, and the recruitment of participants from different groups (CYP, parents and professionals) across two hospitals.

Whilst the recruitment of three separate participant groups enabled triangulation (Mays & Pope, 2000), and is likely to have encouraged greater reflexivity in data analysis, resulting in a more comprehensive understanding of 'thriving alongside CF', the limited scope of the project meant that it was only possible to recruit relatively small numbers of each category of participant. Consequently, although each category identified in the data was deemed to reach theoretical sufficiency, the theoretical model's validity could be enhanced with a larger sample size.

Data analysis was primarily conducted by a single researcher. Whilst analytic decisions were discussed in supervision and with research peers, and bracketing techniques were used, a second coder would have added to the validity of findings. Similarly, the validity of the model could have been enhanced through use of respondent validation (Mays & Pope, 2000).

Although participants were recruited from two NHS sites and attempts were made to also recruit via CF charities and social media, there remained a lack of diversity. All participants were white British, to a certain extent this is consistent with the genetic predisposition of CF (Williams & Barker, 2010). Due to the complexities of measuring socio-economic status in CYP, this was not recorded, though it could be postulated to have an impact on CYP's experiences of empowerment.

The nature of the research is likely to have meant that CYP who are not currently 'thriving alongside CF' would be less likely to have the time, energy, confidence or inclination to participate. This group was alluded to by several participating professionals, and captured in the model under the category of 'unheard voices'. This will be an important group to consider in future research.

Implications for research

It would be valuable to explore the extent to which the findings of the current study are transferable to other settings. This could be done by replicating the current study using larger samples.

Previous work (Acuña Mora et al., 2018) has developed a scale of empowerment for children by applying a model that was originally developed based on research with adult patients. An advantage of the current model is that it is grounded in the views of CYP and the adults most closely involved in supporting them. A possible next step could be to develop a scale of

empowerment specifically for children with CF using a mixed method approach similar to those used by Acuña Mora et al. (2018).

The CYP who are not currently ‘thriving alongside CF’, whose voices are rarely heard in clinic and whom professionals most struggle to support provide a clear focus for future qualitative research.

There is likely to be a need for creativity in considering how best to access this population, for example use of peer researchers as in Gathercole (2018) or use of anonymous questionnaires may go some way to mitigating power differentials between researcher and participants.

Implications for clinical practice

Given the importance ascribed by participants to ‘having a team’ it may be valuable for medical professionals to routinely ask CYP questions about who is in their support network and how CYP would like them to be involved. This could include questions about whether CYP have access to support from other CYP with CF. Access to peer support could be facilitated through the development of internet-based group interventions.

The current study adds weight to recommendations previously made by Sawicki et al. (2015) for example developing communication tools for use by CYP and professionals that incorporate a mutual understanding of competing priorities and time pressures, and developing problem-solving skills using techniques such as motivational interviewing. This

could involve parents and professionals (including teachers), allowing choices, noting initiative and encouraging opinions and problem-solving.

Clinicians may find it helpful to continuously review with CYP's understanding of CF and to develop a stock of accessible resources. Service evaluations could be conducted to understand the extent to which CYP feel they have the opportunity for 'taking charge' / 'having a voice', e.g. using Likert scales.

To support CYP with 'being able to just be a child/getting on with life' Acceptance and Commitment Therapy values-based work or motivational interviewing may help CYP make choices around CF-treatment and balance this with their social and educational activities.

Conclusion

This study drew on the experiences of CYP growing up with CF, their parents and professionals to develop a preliminary theory of empowerment during the key life transitions of adolescence. The emerging model suggests that 'thriving alongside CF' was supported by interactions between 'having a team' and 'taking charge and having a voice', leading to 'being able to just be a child/getting on with life'. 'Concealing self' gets in the way of 'thriving alongside CF'. 'Thriving alongside CF' itself occurs within a wider context.

The model resonates strongly with models of empowerment developed within the context of community psychology (Rappaport, 1984; Zimmerman, 1995) but moves beyond existing

models of patient empowerment to incorporate both a developmental perspective and factors unique to CF.

Given the relatively small-scale of the study it is important to treat findings with caution. However, there are important clinical and research implications. It may be possible to develop interventions to support CYP to have a voice and enable CYP with CF to develop their support network by connecting with each other virtually.

Further qualitative and quantitative research should be undertaken to refine aspects of the model and test hypotheses, particularly in relation to the unheard voices of CYP not ‘thriving alongside CF’.

References

- Acuña Mora, M., Luyckx, K., Sparud-Lundin, C., Peeters, M., van Staa, A. L., Sattoe, J., ... Moons, P. (2018). Patient empowerment in young persons with chronic conditions: Psychometric properties of the Gothenburg Young Persons Empowerment Scale (GYPES). *PLoS ONE*, *13*(7), 1–13. doi:10.1371/journal.pone.0201007
- Acuña Mora, M., Sparud-Lundin, C., Burström, Å., Hanseus, K., Rydberg, A., Moons, P., & Bratt, E. L. (2019). Patient empowerment and its correlates in young persons with congenital heart disease. *European Journal of Cardiovascular Nursing*, *18*(5), 389–398. doi:10.1177/1474515119835434
- Ayers, S., Muller, I., Mahoney, L., & Seddon, P. (2011). Understanding needle-related distress in children with cystic fibrosis. *British Journal of Health Psychology*, *16*(2), 329–343. doi:10.1348/135910710X506895

- Barker, D. H., Driscoll, K. A., Modi, A. C., Light, M. J., & Quittner, A. L. (2012). Supporting cystic fibrosis disease management during adolescence: the role of family and friends. *Child: Care, Health and Development*, 38(4), 497–504. doi:<https://dx.doi.org/10.1111/j.1365-2214.2011.01286.x>
- Barroso, J., Gollop, C. J., Sandelowski, M., Meynell, J., Pearce, P. F., & Collins, L. J. (2003). The challenges of searching for and retrieving qualitative studies. *Western Journal of Nursing Research*, 25(2), 153–178. doi:10.1177/0193945902250034
- Birks, M., & Mills, J. (2015). *Grounded Theory: A practical guide* (2nd ed.). London, UK: SAGE.
- Boyatzis, R. (1998). *Transforming qualitative information: Thematic analysis and code development*. London, UK: Sage.
- Braun, V., & Clarke, V. (2006). Using thematic analysis in psychology Using thematic analysis in psychology. *Qualitative Research in Psychology*, 3(2), 77–101. doi:10.1191/1478088706qp063oa
- Bravo, P., Edwards, A., Barr, P. J., Scholl, I., Elwyn, G., & McAllister, M. (2015). Conceptualising patient empowerment: A mixed methods study. *BMC Health Services Research*, 15(1), 1–14. doi:10.1186/s12913-015-0907-z
- Bronfenbrenner, U. (1986). Ecology of the Family as a Context for Human Development. Research Perspectives. *Developmental Psychology*, 22(6), 723–742. doi:10.1037/0012-1649.22.6.723
- Calvillo, J., Román, I., & Roa, L. M. (2015). How technology is empowering patients? A literature review. *Health Expectations*, 18(5), 643–652. doi:10.1111/hex.12089
- Carroll, C., Booth, A., & Lloyd-Jones, M. (2012). Should we exclude inadequately reported

studies from qualitative systematic reviews? An evaluation of sensitivity analyses in two case study reviews. *Qualitative Health Research*, 22(10), 1425–1434.

doi:10.1177/1049732312452937

Carter, B., Ford, K., Bray, L., Water, T., Arnott, J., Tichanow, C., & Dickinson, A. (2017).

“It’s not who I am”: Children’s experiences of growing up with a long-term condition in England, Australia, and New Zealand. *Journal for Specialists in Pediatric Nursing*, 22(4), 1–10. doi:10.1111/jspn.12197

Cerezo, P. G., Juvé-Udina, M.-E., & Delgado-Hito, P. (2016). Concepts and measures of

patient empowerment: a comprehensive review. *Revista Da Escola de Enfermagem Da USP*, 50(4), 667–674. doi:10.1590/S0080-623420160000500018

Charmaz, K. (2003). Grounded theory in the 21st century. In N. Denzin & Y. Lincoln (Eds.),

The landscape of qualitative research: Theories and issues (2nd ed., pp. 507–530).

London, UK: Sage.

Charmaz, K. (2006). *Constructing grounded theory: A practical guide through qualitative*

analysis. London, UK: Sage Publications Ltd.

Christian, B. J., & D’Auria, J. P. (1997). The child’s eye: memories of growing up with cystic

fibrosis. *Journal of Pediatric Nursing*, 12(1), 3–12. Retrieved from

<http://ovidsp.ovid.com/ovidweb.cgi?T=JS&PAGE=reference&D=med4&NEWS=N&AN=9037958>

Christofides, E., Dobson, J. A., Solomon, M., Waters, V., & O’Doherty, K. C. (2016).

Heuristic decision-making about research participation in children with cystic fibrosis.

Social Science & Medicine, 162, 32–40. doi:10.1016/j.socscimed.2016.06.017

Cooke, A., Smith, D., & Booth, A. (2012). Beyond PICO: The SPIDER tool for qualitative

evidence synthesis. *Qualitative Health Research*, 22(10), 1435–1443.

doi:10.1177/1049732312452938

Corbin, J., & Strauss, A. (1991). A nursing model for chronic illness management based upon the trajectory framework. *Scholarly Inquiry for Nursing Practice: An International Journal*, 5(3), 155–174. Retrieved from <https://www.ncbi.nlm.nih.gov/pubmed/1763239>

Corbin, J., & Strauss, A. (2015). *Basics of qualitative research: Techniques and procedures for developing grounded theory* (4th ed.). London, UK: Sage Publications Inc.

Coulthard, K. P. (2018). Cystic fibrosis: novel therapies, remaining challenges. *Journal of Pharmacy Practice and Research*, 48(6), 569–577. doi:10.1002/jppr.1516

Coyne, I. (2006). Consultation with children in hospital: Children, parents' and nurses' perspectives. *Journal of Clinical Nursing*, 15(1), 61–71. doi:10.1111/j.1365-2702.2005.01247.x

Cruz, I., Marciel, K. K., Quittner, A. L., & Schechter, M. S. (2009). Anxiety and depression in cystic fibrosis. *Seminars in Respiratory and Critical Care Medicine*, 30(5), 569–578. doi:10.1055/s-0029-1238915

Cystic Fibrosis Trust. (2019). Cystic fibrosis FAQs. Retrieved September 14, 2019, from [https://www.cysticfibrosis.org.uk/what-is-cystic-fibrosis/faqs#How common is cystic fibrosis?](https://www.cysticfibrosis.org.uk/what-is-cystic-fibrosis/faqs#How%20common%20is%20cystic%20fibrosis%3F)

D'Auria, J., Christian, B., & Richardson, L. (1997). Through the looking glass: Children's perceptions of growing up with cystic fibrosis. *Canadian Journal of Nursing Research*, 29(4), 99–112.

Dashiff, C., Suzuki-Crumly, J., Kracke, B., Britton, L., & Moreland, E. (2013). Cystic fibrosis-related diabetes in older adolescents: parental support and self-management.

Journal for Specialists in Pediatric Nursing : JSPN, 18(1), 42–53.

doi:<https://dx.doi.org/10.1111/jspn.12010>

Department of Health. (2015). NHS Constitution for England. *The NHS Constitution Establishes the Principles and Values of the NHS in England*, (July), 1. doi:26 March 2013

Dey, I. (1999). *Grounding Grounded theory: Guidelines for qualitative inquiry*. California, CA: Academic Press.

Dixon-Woods, M., Bonas, S., Booth, A., Jones, D. R., Miller, T., Sutton, A. J., ... Young, B. (2006). How can systematic reviews incorporate qualitative research? A critical perspective. *Qualitative Research*, 6(1), 27–44. doi:10.1177/1468794106058867

Durst, C. L., Horn, M. V, MacLaughlin, E. F., Bowman, C. M., Starnes, V. A., & Woo, M. S. (2001). Psychosocial responses of adolescent cystic fibrosis patients to lung transplantation. *Pediatric Transplantation*, 5(1), 27–31. Retrieved from <http://ovidsp.ovid.com/ovidweb.cgi?T=JS&PAGE=reference&D=med4&NEWS=N&AN=11260485>

Erikson, E. (1959). *Identity and the life cycle*. New York: Norton & Company.

Ernst, M. M., Johnson, M. C., & Stark, L. J. (2011). Developmental and Psychosocial Issues in Cystic Fibrosis. *Pediatric Clinics of North America*, 58(4), 865–885. doi:10.1016/j.pcl.2011.06.004

Flemming, K., José Closs, S., Hughes, N. D., & Bennett, M. I. (2016). Using qualitative research to overcome the shortcomings of systematic reviews when designing of a self-management intervention for advanced cancer pain. *International Journal of Qualitative Methods*, 15(1), 1–11. doi:10.1177/1609406916670656

- Foster, C., Eiser, C., Oades, P., Sheldon, C., Tripp, J., Goldman, P., ... Trott, J. (2001). Treatment demands and differential treatment of patients with cystic fibrosis and their siblings: Patient, parent and sibling accounts. *Child: Care, Health and Development*, 27(4), 349–364. doi:10.1046/j.1365-2214.2001.00196.x
- Gathercole, K. (2018). Managing cystic fibrosis alongside children's schooling: Family, nurse and teacher perspectives. *Journal of Child Health Care*. doi:10.1177/1367493518814930
- Gibson, C. (1991). A concept analysis on the process of empowerment. *Journal of Advanced Nursing*, 16, 354–361. Retrieved from <http://www.ncbi.nlm.nih.gov/pubmed/22104094>
- Glaser, B., & Strauss, A. (1967). *The discovery of grounded theory: Strategies for qualitative research*. New Brunswick: AldineTransaction.
- Goodwin, N., Sonola, L., Thiel, V., & Kodner, D. (2013). *Co-ordinated care for people with complex chronic conditions: Key lessons and markers for success*. The Kings Fund. doi:978 1 909029 19 4
- Hegarty, M., MacDonald, J., Watter, P., & Wilson, C. (2009). Quality of life in young people with cystic fibrosis: Effects of hospitalization, age and gender, and differences in parent/child perception. *Child: Care, Health and Development*, 35(4), 462–468. doi:<http://dx.doi.org/10.1111/j.1365-2214.2008.00900.x>
- Iles, N., & Lowton, K. (2008). Young people with cystic fibrosis' concerns for their future: When and how should concerns be addressed, and by whom? *Journal of Interprofessional Care*, 22(4), 436–438. doi:10.1080/13561820801950325
- Johnston-Roberts, K. (1999). Patient empowerment in the United States: A critical Commentary. *Health Expectations*, 2, 82–92.

Jørgensen, C. R., Thomsen, T. G., Ross, L., Dietz, S. M., Therkildsen, S., Groenvold, M., ...

Johnsen, A. T. (2018). What Facilitates “Patient Empowerment” in Cancer Patients During Follow-Up: A Qualitative Systematic Review of the Literature. *Qualitative Health Research*, 28(2), 292–304. doi:10.1177/1049732317721477

King, N. (1999). Template analysis. In C. Cassell & G. Symon (Eds.), *Essential guide to qualitative methods in organizational research* (pp. 118–134). London, UK: Sage.

Kirk, S., Beatty, S., Callery, P., Gellatly, J., Milnes, L., & Prymachuk, S. (2013). The effectiveness of self-care support interventions for children and young people with long-term conditions: a systematic review. *Child: Care, Health and Development*, 39(3), 305–324. doi:http://dx.doi.org/10.1111/j.1365-2214.2012.01395.x

Leung, L. (2015). Validity, reliability, and generalizability in qualitative research. *Journal of Family Medicine and Primary Care*, 4(3), 324. doi:10.4103/2249-4863.161306

Malone, H., Biggar, S., Javadpour, S., Edworthy, Z., Sheaf, G., & Coyne, I. (2017). Interventions for promoting participation in shared decision-making in children and adolescents with cystic fibrosis. *Cochrane Database of Systematic Reviews*, (3), 1–21. doi:10.1002/14651858.CD012578

Mays, N., & Pope, C. (2000). Qualitative research in health care: Assessing quality in qualitative research. *Bmj*, 320(7226), 50–52. doi:10.1136/bmj.320.7226.50

Moola, F. J., & Faulkner, G. E. J. (2014). “A tale of two cases:” the health, illness, and physical activity stories of two children living with cystic fibrosis. *Clinical Child Psychology and Psychiatry*, 19(1), 24–42. doi:https://dx.doi.org/10.1177/1359104512465740

Moola, F. J., Faulkner, G. E. J., & Schneiderman, J. E. (2012). “No Time to Play”:

- Perceptions Toward Physical Activity in Youth With Cystic Fibrosis. *Adapted Physical Activity Quarterly*, 29(1), 44–62. doi:10.1123/apaq.29.1.44
- Nuttall, P., & Nicholes, P. (1992). Cystic Fibrosis: Adolescent and Maternal Concerns About Hospital and Home Care. *Issues in Comprehensive Pediatric Nursing*, 15(3), 199–213. doi:10.3109/01460869209078252
- Oakley, A. (1994). Women and children first and last: Parallels and differences between children's and women's studies. In B. Mayall (Ed.), *Children's childhoods: Observed and experienced* (pp. 13–33). London: The Falmer Press.
- Oliver, C. (2012). Critical Realist Grounded Theory : A New Approach for Social Work Research, (July 2018), 371–387. doi:10.1093/bjsw/bcr064
- Parliamentary, A. P., & Group on Global Health. (2014). A report by the All Party Parliamentary Groups on Global Health; HIV/AIDs; Population, Development and Reproductive Health; Global Tuberculosis; and Patient and Public Involvement in Health and Social Care. Patient empowerment: for better quality, more su.
- Pizzignacco, T. M. P., & de Lima, R. A. G. (2006). Socialization of children and adolescents with cystic fibrosis: Support for nursing care. *Revista Latino-Americana de Enfermagem*, 14(4), 569–577. Retrieved from <http://ovidsp.ovid.com/ovidweb.cgi?T=JS&PAGE=reference&D=med5&NEWS=N&AN=16967164>
- Potter, J. (1997). Discourse analysis as a way of analysing naturally occurring talk. In D. Silverman (Ed.), *Qualitative research: Theory, method, and practice* (pp. 144–160). London, UK: Sage Publications.
- Prigge, J. K., Dietz, B., Homburg, C., Hoyer, W. D., & Burton, J. L. (2015). Patient

empowerment: A cross-disease exploration of antecedents and consequences.

International Journal of Research in Marketing, 32(4), 375–386.

doi:10.1016/j.ijresmar.2015.05.009

Public Health Resource Unit. (2006). *Critical Appraisal Skills Programme (CASP)*

Qualitative Checklist. Retrieved from [https://casp-uk.net/wp-](https://casp-uk.net/wp-content/uploads/2018/03/CASP-Qualitative-Checklist-2018_fillable_form.pdf)

[content/uploads/2018/03/CASP-Qualitative-Checklist-2018_fillable_form.pdf](https://casp-uk.net/wp-content/uploads/2018/03/CASP-Qualitative-Checklist-2018_fillable_form.pdf)

Rappaport, J. (1984). Studies in Empowerment. *Prevention in Human Services*, 3, 1–7.

doi:10.1300/J293v03n02_02

Ritchie, J., & Spencer, L. (1994). Qualitative data analysis for applied policy research. In A.

Bryman & R. Burgess (Eds.), *Analysing qualitative data*. London, UK: Routledge.

Rowbotham, N. J., Palser, S. C., Smith, S. J., & Smyth, A. R. (2019). Infection prevention

and control in cystic fibrosis: a systematic review of interventions. *Expert Review of*

Respiratory Medicine, 13(5), 425–434. doi:10.1080/17476348.2019.1595594

Rowlands, J. (1995). Empowerment Examined. *Development in Practice*, 5(2), 101–107.

Retrieved from

<https://www.jstor.org/stable/pdf/4028929.pdf?refreqid=excelsior%3Abad6e17fc21fe0ee58ffe8b2a4d10777>

Sandelowski, M. (2000). Whatever happened to qualitative description? *Research in Nursing*

& Health, 23(4), 334–340. doi:10.1002/1098-240X(200008)23:4<334::AID-

NUR9>3.0.CO;2-G

Savage, E., & Callery, P. (2007). Clinic consultations with children and parents on the dietary

management of cystic fibrosis. *SOCIAL SCIENCE & MEDICINE*, 64(2), 363–374.

doi:10.1016/j.socscimed.2006.09.003

- Sawicki, G. S., Heller, K. S., Demars, N., & Robinson, W. M. (2015). Motivating adherence among adolescents with cystic fibrosis: youth and parent perspectives. *Pediatric Pulmonology*, *50*(2), 127–136. doi:<https://dx.doi.org/10.1002/ppul.23017>
- Sawicki, G. S., Ren, C. L., Konstan, M. W., Millar, S. J., Pasta, D. J., Quittner, A. L., & Fibrosis, I. and C. of the E. S. of C. (2013). Treatment complexity in cystic fibrosis: trends over time and associations with site-specific outcomes. *Journal of Cystic Fibrosis : Official Journal of the European Cystic Fibrosis Society*, *12*(5), 461–467. doi:<https://dx.doi.org/10.1016/j.jcf.2012.12.009>
- Sawyer, S. M., Drew, S., Yeo, M. S., & Britto, M. T. (2007). Adolescents with a chronic condition: challenges living, challenges treating. *Lancet (London, England)*, *369*(9571), 1481–1489. doi:[https://dx.doi.org/10.1016/S0140-6736\(07\)60370-5](https://dx.doi.org/10.1016/S0140-6736(07)60370-5)
- Small, N., Bower, P., Chew-Graham, C., Whalley, D., & Protheroe, J. (2013). Patient empowerment in long-term conditions: Development and preliminary testing of a new measure. *BMC Health Services Research*, *13*(1), 263. doi:10.1186/1472-6963-13-263
- Stake, R. (2000). Case studies. In N. Denzin & Y. Lincoln (Eds.), *The handbook of qualitative research*. Thousand Oaks, CA: Sage.
- Stein, R. E. K. (1992). Chronic Physical Disorders. *Pediatrics in Review*, *13*(6), 224–229. doi:10.1542/pir.13-6-224
- Stewart, L., Moher, D., & Shekelle, P. (2012). Why prospective registration of systematic reviews makes sense. *Systematic Reviews*, *1*(1), 7–10. doi:10.1186/2046-4053-1-7
- Strauss, A., & Corbin, J. M. (1990). *Basics of qualitative research: Grounded theory procedures and techniques*. Newbury Park, CA: Sage.
- Taylor, R. M., Gibson, F., & Franck, L. S. (2008). The experience of living with a chronic

illness during adolescence: A critical review of the literature. *Journal of Clinical Nursing*, 17(23), 3083–3091. doi:10.1111/j.1365-2702.2008.02629.x

The British Psychological Society. (2018). Code of Ethics and Conduct. *The British Psychological Society*, (July), 100–102. Retrieved from www.bps.org.uk.

The British Psychological Society (BPS). (2014). *Code of Human Research Ethics*. The British Psychological Society. Retrieved from http://www.bps.org.uk/sites/default/files/documents/code_of_human_research_ethics.pdf

Thomas, J., & Harden, A. (2008). Methods for the thematic synthesis of qualitative research in systematic reviews. *BMC Medical Research Methodology*, 8, 1–10.
doi:10.1186/1471-2288-8-45

Thomas, J., Sutcliffe, K., Harden, A., Oakley, A., Oliver, S., Rees, R., ... Kavanagh, J. (2003). *Children and healthy eating: A systematic review of barriers and facilitators*. London, UK.

Tong, A., Flemming, K., McInnes, E., Oliver, S., & Craig, J. (2012). Enhancing transparency in reporting the synthesis of qualitative research: ENTREQ. *BMC Medical Research Methodology*, 12(Figure 1), 1–8. doi:10.1186/1471-2288-12-181

UK Cystic Fibrosis Registry. (2013). *Cystic Fibrosis: Our focus*. London. Retrieved from <https://www.cysticfibrosis.org.uk/the-work-we-do/uk-cf-registry>

Vines, E. E., Fisher, P., Conniff, H., & Young, J. (2018). Adolescents' experiences of isolation in cystic fibrosis. *Clinical Practice in Pediatric Psychology*, 6(4), 375–385.
doi:10.1037/cpp0000256

Wakefield, D., Bayly, J., Selman, L. E., Firth, A. M., Higginson, I. J., & Murtagh, F. E. M.

- (2018). Patient empowerment, what does it mean for adults in the advanced stages of a life-limiting illness: A systematic review using critical interpretive synthesis. *Palliative Medicine*, 32(8), 1288–1304. doi:10.1177/0269216318783919
- Williams, B., Corlett, J., Dowell, J. S., Coyle, J., & Mukhopadhyay, S. (2009). I've never not had it so I don't really know what it's like not to: nondifference and biographical disruption among children and young people with cystic fibrosis. *Qualitative Health Research*, 19(10), 1443–1455. doi:https://dx.doi.org/10.1177/1049732309348363
- Williams, B., Mukhopadhyay, S., Dowell, J., & Coyle, J. (2007a). From child to adult: an exploration of shifting family roles and responsibilities in managing physiotherapy for cystic fibrosis. *Social Science & Medicine* (1982), 65(10), 2135–2146. Retrieved from <http://ovidsp.ovid.com/ovidweb.cgi?T=JS&PAGE=reference&D=med5&NEWS=N&AN=17719160>
- Williams, B., Mukhopadhyay, S., Dowell, J., & Coyle, J. (2007b). Problems and solutions: accounts by parents and children of adhering to chest physiotherapy for cystic fibrosis. *Disability and Rehabilitation*, 29(14), 1097–1105. Retrieved from <http://ovidsp.ovid.com/ovidweb.cgi?T=JS&PAGE=reference&D=med5&NEWS=N&AN=17612996>
- Williams, R., & Barker, H. (2010). Cystic Fibrosis. *InnovAiT: Education and Inspiration for General Practice*, 3(12), 743–752. doi:10.1093/innovait/inq072
- World Health Organization Regional Office for Europe. (2013). *Health 2020: A European policy framework and strategy for the 21st century*. Copenhagen, Denmark: World Health Organization Regional Office for Europe.
- Yeh, M. Y., Wu, S. C., & Tung, T. H. (2018). The relation between patient education, patient empowerment and patient satisfaction: A cross-sectional-comparison study. *Applied*

Nursing Research, 39(45), 11–17. doi:10.1016/j.apnr.2017.10.008




Zimmerman, M. A. (1995). Psychological empowerment: Issues and illustrations. *American*

Journal of Community Psychology, 23(5), 581–599. doi:10.1007/BF02506983

Part C Appendices A-LL

CASP Checklist: 10 questions to help you make sense of a **Qualitative** research

How to use this appraisal tool: Three broad issues need to be considered when appraising a qualitative study:

-  Are the results of the study valid? (Section A)
-  What are the results? (Section B)
-  Will the results help locally? (Section C)

The 10 questions on the following pages are designed to help you think about these issues systematically. The first two questions are screening questions and can be answered quickly. If the answer to both is “yes”, it is worth proceeding with the remaining questions. There is some degree of overlap between the questions, you are asked to record a “yes”, “no” or “can’t tell” to most of the questions. A number of italicised prompts are given after each question. These are designed to remind you why the question is important. Record your reasons for your answers in the spaces provided.

About: These checklists were designed to be used as educational pedagogic tools, as part of a workshop setting, therefore we do not suggest a scoring system. The core CASP checklists (randomised controlled trial & systematic review) were based on JAMA 'Users' guides to the medical literature 1994 (adapted from Guyatt GH, Sackett DL, and Cook DJ), and piloted with health care practitioners.

For each new checklist, a group of experts were assembled to develop and pilot the checklist and the workshop format with which it would be used. Over the years overall adjustments have been made to the format, but a recent survey of checklist users reiterated that the basic format continues to be useful and appropriate.

Referencing: we recommend using the Harvard style citation, i.e.: *Critical Appraisal Skills Programme (2018). CASP (insert name of checklist i.e. Qualitative) Checklist. [online] Available at: URL. Accessed: Date Accessed.*

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Paper for appraisal and reference:

Section A: Are the results valid?

1. Was there a clear statement of the aims of the research?

Yes	<input type="checkbox"/>
Can't Tell	<input type="checkbox"/>
No	<input type="checkbox"/>

- HINT: Consider
- what was the goal of the research
 - why it was thought important
 - its relevance

Comments:

2. Is a qualitative methodology appropriate?

Yes	<input type="checkbox"/>
Can't Tell	<input type="checkbox"/>
No	<input type="checkbox"/>

- HINT: Consider
- If the research seeks to interpret or illuminate the actions and/or subjective experiences of research participants
 - Is qualitative research the right methodology for addressing the research goal

Comments:

Is it worth continuing?

3. Was the research design appropriate to address the aims of the research?

Yes	<input type="checkbox"/>
Can't Tell	<input type="checkbox"/>
No	<input type="checkbox"/>

- HINT: Consider
- if the researcher has justified the research design (e.g. have they discussed how they decided which method to use)

Comments:

4. Was the recruitment strategy appropriate to the aims of the research?

Yes	<input type="checkbox"/>
Can't Tell	<input type="checkbox"/>
No	<input type="checkbox"/>

HINT: Consider

- If the researcher has explained how the participants were selected
- If they explained why the participants they selected were the most appropriate to provide access to the type of knowledge sought by the study
 - If there are any discussions around recruitment (e.g. why some people chose not to take part)

Comments:

5. Was the data collected in a way that addressed the research issue?

Yes	<input type="checkbox"/>
Can't Tell	<input type="checkbox"/>
No	<input type="checkbox"/>

HINT: Consider

- If the setting for the data collection was justified
- If it is clear how data were collected (e.g. focus group, semi-structured interview etc.)
- If the researcher has justified the methods chosen
 - If the researcher has made the methods explicit (e.g. for interview method, is there an indication of how interviews are conducted, or did they use a topic guide)
 - If methods were modified during the study. If so, has the researcher explained how and why
 - If the form of data is clear (e.g. tape recordings, video material, notes etc.)
 - If the researcher has discussed saturation of data

Comments:

6. Has the relationship between researcher and participants been adequately considered?

Yes	<input type="checkbox"/>
Can't Tell	<input type="checkbox"/>
No	<input type="checkbox"/>

HINT: Consider

- If the researcher critically examined their own role, potential bias and influence during (a) formulation of the research questions (b) data collection, including sample recruitment and choice of location
- How the researcher responded to events during the study and whether they considered the implications of any changes in the research design

Comments:

Section B: What are the results?

7. Have ethical issues been taken into consideration?

Yes	<input type="checkbox"/>
Can't Tell	<input type="checkbox"/>
No	<input type="checkbox"/>

HINT: Consider

- If there are sufficient details of how the research was explained to participants for the reader to assess whether ethical standards were maintained
- If the researcher has discussed issues raised by the study (e.g. issues around informed consent or confidentiality or how they have handled the effects of the study on the participants during and after the study)
- If approval has been sought from the ethics committee

Comments:

8. Was the data analysis sufficiently rigorous?

Yes	<input type="checkbox"/>
Can't Tell	<input type="checkbox"/>
No	<input type="checkbox"/>

HINT: Consider

- If there is an in-depth description of the analysis process
- If thematic analysis is used. If so, is it clear how the categories/themes were derived from the data
- Whether the researcher explains how the data presented were selected from the original sample to demonstrate the analysis process
- If sufficient data are presented to support the findings
 - To what extent contradictory data are taken into account
- Whether the researcher critically examined their own role, potential bias and influence during analysis and selection of data for presentation

Comments:

9. Is there a clear statement of findings?

Yes	<input type="checkbox"/>
Can't Tell	<input type="checkbox"/>
No	<input type="checkbox"/>

HINT: Consider whether

- If the findings are explicit
- If there is adequate discussion of the evidence both for and against the researcher's arguments
- If the researcher has discussed the credibility of their findings (e.g. triangulation, respondent validation, more than one analyst)
- If the findings are discussed in relation to the original research question

Comments:

Section C: Will the results help locally?

10. How valuable is the research?

HINT: Consider

- If the researcher discusses the contribution the study makes to existing knowledge or understanding (e.g. do they consider the findings in relation to current practice or policy, or relevant research-based literature
- If they identify new areas where research is necessary
- If the researchers have discussed whether or how the findings can be transferred to other populations or considered other ways the research may be used

Comments:

Appendix B. Criteria for assessing research conducted with children used by (Thomas et al., 2003)

1. Did the study use appropriate data collection methods for helping children to express their views?
2. Did the study use appropriate methods for ensuring the data analysis was grounded in the views of children?
3. Did the study actively involved children in the design and conduct of the study?

Appendix C. Key strengths and limitations of studies

<i>Key strengths and limitations of studies</i>		
<u>Study</u>	<u>Strengths</u>	<u>Limitations</u>
Ayers et al. (2011)	<p>Acknowledged those choosing not to participate</p> <p>Interview procedure appropriate for helping children express their views</p> <p>Researcher not associated with clinical team</p> <p>Clear consideration of ethics</p> <p>Data analysed by more than one researcher</p> <p>Clearly described process of thematic analysis and theme deduction</p> <p>Used appropriate methods for ensuring data analysis grounded in the views of children</p> <p>Quotes used to support findings</p> <p>Clear clinical and research implications</p>	<p>No evidence of having involved children in design or conduct of the study</p> <p>Data saturation not discussed</p> <p>No evidence of researcher reflexivity</p>
Barker et al. (2011)	<p>Clear consideration of ethics</p> <p>Used appropriate methods for helping children express their views and ensuring data analysis grounded in the views of children</p> <p>Clear description of template analysis process</p> <p>Data analysed by more than one researcher, inter-rater reliability reported</p> <p>Quotes used to support findings</p> <p>Clear clinical and research implications</p>	<p>No evidence of having involved children in design or conduct of the study</p> <p>Unclear how participants were selected by medical team</p> <p>Data saturation not discussed</p> <p>No evidence of researcher reflexivity</p>
Christian & D'Auria (1997)	<p>Clear description of grounded theory process</p> <p>Used appropriate methods for helping children express their views and ensuring data analysis grounded in the views of children</p> <p>Discussed data saturation</p>	<p>No evidence of having involved children in design or conduct of the study</p> <p>No evidence of researcher reflexivity</p> <p>No reference to ethical approval</p>

Appendix C. Key strengths and limitations of studies

	Quotes used to support findings Clear clinical and research implications	
Christofides et al. (2016)	<p>Reflected on why some potential participants declined to take part</p> <p>Used appropriate methods for helping children express their views and ensuring data analysis grounded in the views of children</p> <p>Clear description of thematic analysis process</p> <p>Data analysed by more than one researcher</p> <p>Quotes used to support findings</p> <p>Clear discussion of implications</p>	<p>No evidence of having involved children in design or conduct of the study</p> <p>Data saturation not discussed</p> <p>No evidence of researcher reflexivity</p>
Dashiff et al. (2013)	<p>Sample clearly described</p> <p>Used appropriate methods for helping children express their views</p> <p>Some use of quotes to support findings</p> <p>Data analysed by more than one researcher</p>	<p>No evidence of having involved children in design or conduct of the study</p> <p>Description of 'qualitative description' limited and unclear</p> <p>Unclear whether sufficient data to support findings</p> <p>Data saturation not discussed</p> <p>No evidence of researcher reflexivity</p>
D'Auria et al. (1997)	<p>Researcher not associated with clinical team</p> <p>Clear description of grounded theory process</p> <p>Used appropriate methods for helping children express their views and ensuring data analysis grounded in the views of children</p> <p>Discussed data saturation</p> <p>Data analysed by more than one researcher</p> <p>Quotes used to support findings</p>	<p>No evidence of having involved children in design or conduct of the study</p> <p>No evidence of researcher reflexivity</p> <p>No reference to ethical approval</p>

Appendix C. Key strengths and limitations of studies

	Clear discussion of implications	
Durst et al. (2001)	<p>Sample clearly described</p> <p>Used appropriate methods for helping children express their views</p> <p>Some use of quotes to support findings</p>	<p>No evidence of having involved children in design or conduct of the study</p> <p>Study design and method unclear, mentions ethnography in abstract but this is not elaborated on in the methods section, in main body study described as 'descriptive'</p> <p>Unclear how the data from interviews was collected - no mention of audio recording</p> <p>No evidence of researcher reflexivity</p> <p>No reference to ethical approval</p>
Foster et al. (2001)	<p>Researcher not associated with clinical team</p> <p>Clear description of grounded theory process</p> <p>Used appropriate methods for helping children express their views and ensuring data analysis grounded in the views of children</p> <p>Data analysed by more than one researcher</p> <p>Quotes used to support findings</p> <p>Discussion of implications</p>	<p>No evidence of having involved children in design or conduct of the study</p> <p>No evidence of researcher reflexivity</p> <p>Data saturation not discussed</p>
Moola & Faulkner (2014)	<p>Some use of quotations to support findings</p> <p>Implications discussed</p>	<p>No evidence of having involved children in design or conduct of the study</p> <p>Sampling strategy unclear (case series described 2 of the 6 participants)</p> <p>Data analysis process unclear - thematic analysis mentioned but unclear how themes were derived</p> <p>Unclear if appropriate methods were used to ensure data</p>

Appendix C. Key strengths and limitations of studies

		analysis was grounded in the views of children No evidence of researcher reflexivity
Moola et al. (2012)	<p>Researcher not associated with clinical team</p> <p>Clear description of grounded theory process</p> <p>Used appropriate methods for helping children express their views and ensuring data analysis grounded in the views of children</p> <p>Evidence of researcher reflexivity</p> <p>Discussed data saturation</p> <p>Quotes used to support findings</p> <p>Clear discussion of implications</p>	No evidence of having involved children in design or conduct of the study
Nuttall & Nicholes (1992)	<p>Clear description of content analysis process</p> <p>Used appropriate methods for helping children express their views and ensuring data analysis grounded in the views of children</p> <p>Inter-rater reliability reported</p> <p>Quotes used to support findings</p> <p>Clear discussion of implications</p>	<p>No evidence of having involved children in design or conduct of the study</p> <p>No evidence of researcher reflexivity</p>
Pizzignacco & de Lima (2006)	<p>Used appropriate methods for helping children express their views</p> <p>Quotes used to support findings</p> <p>Clear discussion of implications</p>	<p>No evidence of having involved children in design or conduct of the study</p> <p>Data analysis process unclear - thematic analysis mentioned but unclear how themes were derived</p> <p>No evidence of researcher reflexivity</p>
Savage & Callery (2007)	<p>Clear description of discourse analysis process</p>	No evidence of having involved children in design or conduct of the study

Appendix C. Key strengths and limitations of studies

	<p>Used appropriate methods for helping children express their views and ensuring data analysis grounded in the views of children</p> <p>Triangulation of data (interviews & observations of consultations)</p> <p>Quotes used to support findings</p> <p>Clear discussion of implications</p>	<p>No evidence of researcher reflexivity</p>
<p>Sawicki et al. (2015)</p>	<p>Used appropriate methods for helping children express their views</p> <p>Some evidence of researcher reflexivity</p> <p>Discussed inter-rater reliability</p> <p>Quotes used to support findings</p> <p>Clear discussion of implications</p>	<p>No evidence of having involved children in design or conduct of the study</p> <p>Unclear which qualitative data analysis method was used</p>
<p>Williams et al. (2007a)</p>	<p>Evidence of having involved children in design/conduct of the study</p> <p>Clear description of framework analysis process</p> <p>Used appropriate methods for helping children express their views and ensuring data analysis grounded in the views of children</p> <p>Data analysed by more than one researcher</p> <p>Quotes used to support findings</p> <p>Clear discussion of implications</p>	<p>Limited evidence of researcher reflexivity</p>
<p>Williams et al. (2007b)</p>	<p>Evidence of having involved children in design/conduct of the study</p> <p>Clear description of framework analysis process</p> <p>Used appropriate methods for helping children express their views and ensuring data</p>	<p>Limited evidence of researcher reflexivity</p>

Appendix C. Key strengths and limitations of studies

	<p>analysis grounded in the views of children Data analysed by more than one researcher Quotes used to support findings Clear discussion of implications</p>
<p>Williams et al. (2009) UK</p>	<p>Evidence of having involved children in design/conduct of the study Clear description of framework analysis process Evidence of researcher reflexivity Used appropriate methods for helping children express their views and ensuring data analysis grounded in the views of children Data analysed by more than one researcher Quotes used to support findings Clear discussion of implications</p>

Appendix D. Overview of quality assessment ratings according to CASP and Thomas et al. (2003) criteria

Authors, Year	Title	1	2	3	4	5	6	7	8	9	10	11	12	13
Ayers et al., 2011	Understanding needle-related distress in children with cystic fibrosis.	Y	Y	Y	Y	Y	?	Y	Y	Y	Y	Y	Y	N
Barker et al., 2012	Supporting cystic fibrosis disease management during adolescence: the role of family and friends.	Y	Y	Y	?	Y	?	Y	Y	Y	Y	Y	Y	N
Christian & D'Auria 1997	The Child's Eye: Memories of growing up with cystic fibrosis	Y	Y	Y	Y	Y	?	?	Y	Y	Y	Y	Y	N
Christofides et al., 2016	Heuristic decision-making about research participation in children with cystic fibrosis.	Y	Y	Y	Y	Y	?	Y	Y	Y	Y	Y	Y	N

Dashiff et al, 2013	Cystic fibrosis-related diabetes in older adolescents: Parental support and self-management.	Y	Y	Y	Y	Y	?	Y	?	Y	Y	Y	?	N
D'Auria et al., 1997	Through the looking glass: children's perceptions of growing up with cystic fibrosis.	Y	Y	Y	Y	Y	?	Y	Y	Y	Y	Y	Y	N
Durst et al., 2001	Psychosocial responses of adolescent cystic fibrosis patients to lung transplantation.	Y	Y	?	Y	?	?	?	?	?	Y	Y	?	N
Foster et al., 2001	Treatment demands and differential treatment of patients with cystic fibrosis and their siblings: Patient, parent and sibling accounts.	Y	Y	Y	Y	Y	?	Y	Y	Y	Y	Y	Y	N
Moola & Faulkner 2014	'A tale of two cases:' the health, illness, and physical activity stories of	Y	Y	Y	N	Y	?	?	?	?	Y	Y	?	N

	two children living with cystic fibrosis.													
Moola et al., 2012	No time to play: Perceptions toward physical activity in youth with cystic fibrosis.	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y	N
Nuttall & Nicholes 1992	Cystic fibrosis: Adolescent and maternal concerns about hospital and home care.	Y	Y	Y	Y	Y	?	Y	Y	Y	Y	Y	Y	N
Pizzignacco & de Lima 2006	Socialization of children and adolescents with cystic fibrosis: Support for nursing care.	Y	Y	Y	Y	Y	?	Y	?	Y	Y	Y	Y	N
Savage & Callery 2007	Clinic consultations with children and parents on the dietary management of cystic fibrosis.	Y	Y	Y	Y	Y	?	Y	Y	Y	Y	Y	Y	N

CASP Qualitative checklist

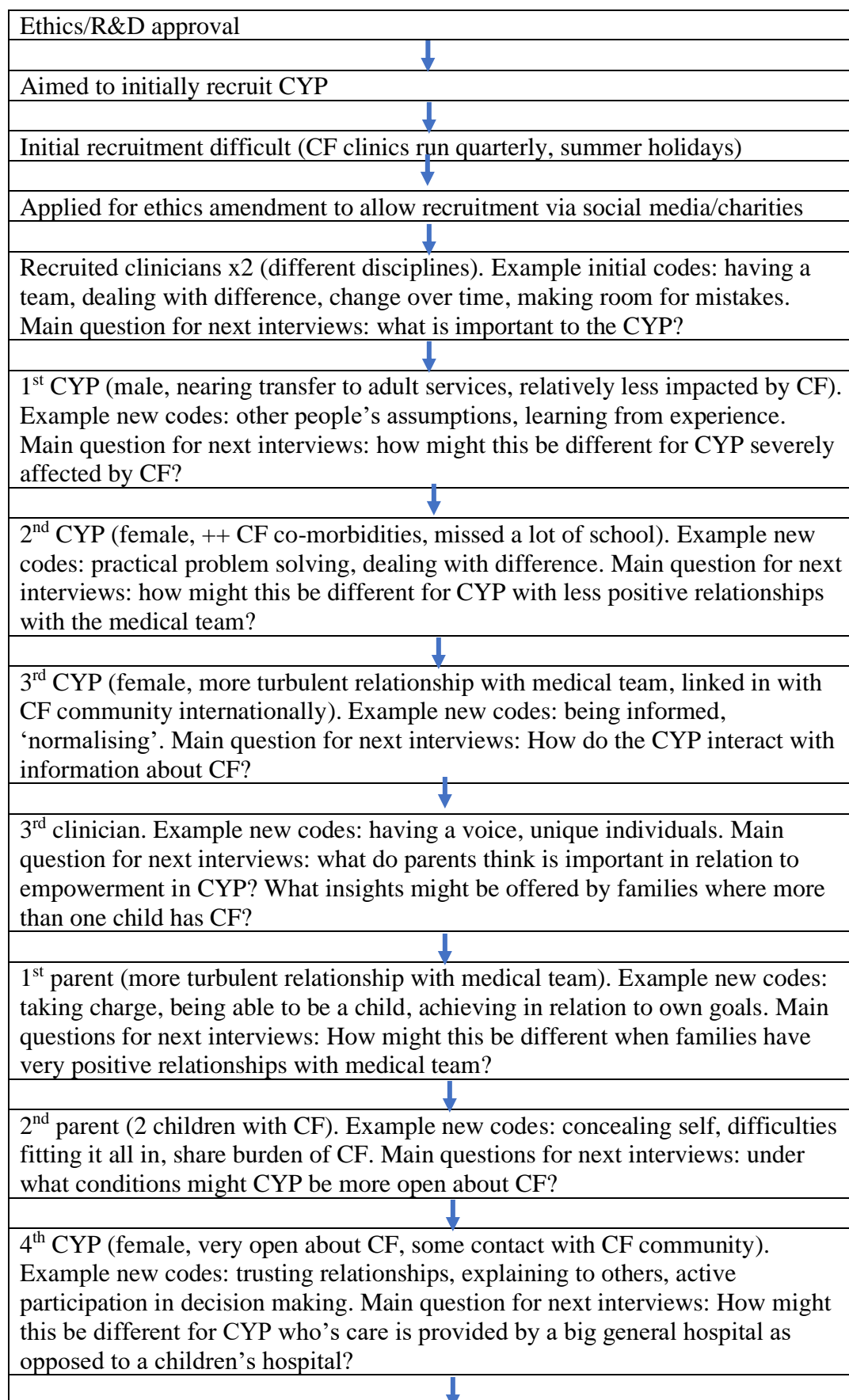
- 1. Clear statement of aims?**
- 2. Qualitative methodology appropriate?**
- 2. 3. Was the research design appropriate to address the aims of the research?**
- 3. 4. Was the recruitment strategy appropriate?**
- 4. 5. Was data collected in a way that addressed the research issue?**
- 5. 6. Has the relationship between researcher & participants been adequately considered?**
- 6. 7. Have ethics been considered?**
- 7. 8. Was data analysis sufficiently rigorous?**
- 8. 9. Is there a clear statement of findings?**
- 9. 10. How valuable is the research? Thomas et al. 2003**
- 10. 11. Used appropriate data collection methods for helping children to express their views?**
- 11. 12. Used appropriate methods for ensuring the data analysis was grounded in the views of children?**
- 12. 13. Actively involved children in the design and conduct of the study?**

Y = Yes

N = No

? = Don't know

Appendix E. Theoretical sampling flow chart



3rd parent (parent of younger child, recruited from 2nd hospital site). Example new codes: none. Main question for next interviews: how might younger male CYP experience empowerment?



5th CYP (male, younger, recruited from 2nd hospital site). Example new codes: none. Main question for next interviews: what insights could dads offer in relation to empowerment in CYP



4th parent (dad, family historically chose to keep CF relatively private). Example new codes: having adventures, achieving in relation to own goals. Main question for next interviews: What insights could parents of children who are not currently 'thriving alongside CF'?



5th parent (parent of younger child who is not 'thriving with CF', child chose not to be interviewed themselves): Example new codes: concealing self, unheard voices. Main question for next interviews: Can CYP 'thrive alongside CF' whilst keeping CF private in some contexts?



6th CYP (recruited from 2nd hospital site, chooses to keep CF relatively private at school, would like to find a way of having contact with other CYP with CF). Example new codes: none. Main questions for next interview: what insights might a clinician with many years' experience offer?



4th clinician (≥ 20 years' experience). Example new codes: none. Main question for next interview:



7th CYP (male, middle of age range, open about CF): none

Appendix F. Email to CF teams

My name is Naomi Fairweather and I am a trainee clinical psychologist at Canterbury Christ Church University.

I am currently working on my doctoral research project, exploring empowerment in children and young people with CF during times of change. The findings of this research will inform clinical practice.

I am interested in talking to children and young people aged 11-19 years living with CF, their parents, and clinical staff. Interviews will last up to one hour and will take place at [Children's Hospital] or via Skype/phone if preferred. I'm aiming to recruit a total of 3-5 members of staff (ideally from a range of professional backgrounds) from [Trust] and [Trust]. I can be flexible around days and times for interviews which will be taking place between now and December.

If you are interested in taking part or have any questions about the research please get in touch: n.h.fairweather323@canterbury.ac.uk

Research Information Leaflet: Children and Young People

Empowerment of children and young people living with cystic fibrosis during times of change

Hello. My name is Naomi Fairweather and I am a trainee clinical psychologist at Canterbury Christ Church University. I would like to invite you to take part in a research study. Before you decide it is important to understand why the research is being done and what it involves for you. If you decide to take part please keep this information sheet so that you can look at it again if you want to.



What is this study for?



We know that many children and young people with medical conditions want to feel confident and in control of their lives, but can face barriers. Empowering (giving power to) people with medical conditions might mean they can stay happier and healthier. I want to know more about the empowerment of children and young people with cystic fibrosis (CF).

Why have I been invited?

I am interested in talking to children and young people aged 11-19 years living with CF during times of change like moving schools, becoming more independent at home, making new friends and having relationships, and moving on to the adult CF team.

I also want to speak with parents and medical staff. You have been invited to take part because you are within an age range where lots of important life changes happen and are under the care of a CF team in the UK.

Do I have to take part?

It is up to you to decide whether or not to join the study. You don't have to take part – even if your parents tell you they want you to. If you agree to take part, I will ask you to sign a consent form. If you are under 16 years, we will need your parents to agree and sign a form first. You are free to change your mind at any time, without giving a reason. This would not affect the care you receive in any way.

What will happen to me if I take part?

Some information about your CF-related health will be collected from a database. You will be invited to take part in an interview so that I can ask you some questions about CF and feeling confident in different situations like home, school, and the hospital. Interviews can take place at _____ Hospital, by phone or using Skype, whichever you prefer. The interview will last under an hour. You don't have to answer all the questions.

If you choose to have the interview by phone or Skype, I won't be able to make sure that no one can hear what you are saying where you are, but I will talk to your parents/guardians about how important it is that your interview is private. I will make sure no one can overhear our conversation where I am.

Some people might be invited to a second interview and asked extra questions – it will be up to you whether you want to or not.



Expenses and payments

I can give you or your parents up to £10 towards travel costs.

What are the possible benefits of taking part?

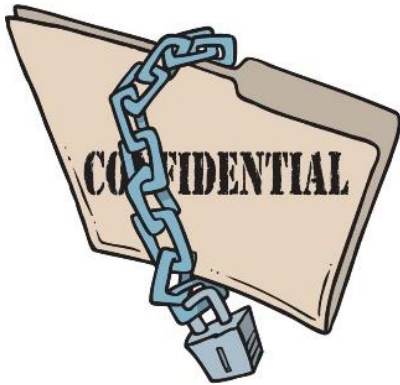
Many people like to know that they have helped with research which might help children and young people with CF in the future. We hope this research will help us to better support children and young people with CF.

What are the possible disadvantages and risks of taking part?

Sometimes talking about your experiences can be upsetting. I am trained to talk to both children and adults and to help if someone becomes upset. I can also tell you about other people that might be able to help, for example the Cystic Fibrosis Trust. You can contact them via their website www.cysticfibrosis.org.uk or call their helpline on 0300 373 1000 or 020 3795 2184.

What will happen to the information given by me?

All information about you will be kept confidential (private) within the research team. There are two situations where I would not be able to keep information private.



- 1) If I thought you were at risk of harm
- 2) If I thought someone else was at risk of harm

If this is the case I would always try to talk to you first.

The interview will be audio-recorded. and the recording will be stored safely so that no-one else can listen. I write a report based on yours and other people's answers - no-one will know who you are.

I will tell your medical team that you are taking part but I won't let them know what you have said. You can talk to anyone you want to

about the research.

What if there is a problem?

I don't want anything about the research to be upsetting. If you are unhappy about anything to do with the research you can talk to me or your CF team. You can also choose to drop out of the research at any time and you don't have to so why. I can also tell you about other people who might be able to help if you are upset. If you do get upset, I will tell your CF team so that they can support you.



If you have a complaint I will do my best to put it right (there is a phone number and email address at the end of this information sheet) or you can talk to the CF team. If you are still unhappy or don't want to talk to me about it, you can talk to Professor Paul Camic, who is in charge of research at my university – paul.camic@canterbury.ac.uk, tel: _____. There is also an NHS Complaints Procedure, you can ask anyone in the CF clinic about this.



What will happen to the results of the research study?

At the end of the research a short summary will be available for anybody who wants it who has taken part. I will write a report for my university course and will let other people know about the findings. No-one will be able to tell who the people who took part in the research are.

Who is organising and funding the research?

This project is funded by Canterbury Christ Church University. In the research team, as well as me, are Dr Fergal Jones, Dr Sally Clarke and Dr Martha Deiros Collado. They are all clinical psychologists who are helping me to do the study. I may also ask someone to type out what you have said to me. They will sign a form promising to keep your information private.

Who has reviewed the study?

All research in the NHS is looked at by independent group of people, called a Research Ethics Committee, to make sure it is safe and fair. This study has been looked at by _____ Research Ethics Committee.

What happens now?

If you would like to help us by taking part in this research, you or your parents can contact me directly (phone number and email address at the end of this information sheet), or you can let me know when I contact you in a couple of weeks' time. We will need to arrange a time to meet at _____ Hospital and will try and make it before or after a clinic appointment so you don't have to travel especially to see me. Alternatively we could do the interview on the phone or Skype. I will explain the study and answer any questions you might have.



I will ask everyone who wants to take part to sign a form to say that they understand what the research is about and are willing to be involved. If you are under 16, legally I also need your parent/guardian to sign a form to say they agree to you taking part.

Further information and contact details

If...

- you would like to help us by taking part in this research
- you would like information about the research
- you would like to know more about this study

please contact:

Naomi Fairweather
Trainee Clinical Psychologist
Salomons Centre for Applied Psychology,
Canterbury Christ Church University

Email: N.H.Fairweather323@canterbury.ac.uk

Telephone: You can leave a message for me on a 24-hour voicemail phone line at 03330117070. Please say that the message is for Naomi Fairweather and leave a contact number so that I can get back to you.

Thank you for reading about my study. I will contact you by phone within two weeks if I have not heard from you, to see if you want to take part.

Research Information Leaflet: Young people

Empowerment of children and young people living with cystic fibrosis during times of change

Hello. My name is Naomi Fairweather and I am a trainee clinical psychologist at Canterbury Christ Church University. I would like to invite you to take part in a research study. Before you decide it is important that you understand why the research is being done and what it would involve for you. If you decide to take part please keep this information sheet so that you can look at it again if you want to.



What is this study for?

We know that many children and young people with medical conditions want to feel confident and in control of their lives, but can face barriers. Empowering people with medical conditions might mean they can stay happier and healthier. I want to know more about the empowerment of children and young people with cystic fibrosis (CF).

Why have I been invited?

I am interested in talking to children and young people aged 11-19 years living with CF during times of change like going to college, becoming more independent at home, making new friends and having relationships, and moving on to the adult CF team.

I also want to speak with parents and medical staff. You have been invited to take part because you are within an age range where lots of important life changes happen and are under the care of a CF team in the UK.

Do I have to take part?

It is up to you to decide whether or not to join the study. If you agree to take part, I will then ask you to sign a consent form. If you are under 16 years, we will need your parents to agree and sign a form first. You are free to change your mind at any time, without giving a reason. This would not affect the care you receive in any way.

What will happen to me if I take part?

Some information about your CF-related health will be collected from a database. You will be invited to take part in an interview so that I can ask you some questions about CF and feeling confident in different situations like home, school or college, and the hospital. Interviews can take place at _____ Hospital, by phone or using Skype, whichever you prefer. The interview will last under an hour. You don't have to answer all the questions.

If you choose to have the interview by phone or Skype, I won't be able to make sure that no one can hear what you are saying where you are, but I can talk to your parents/guardians about how important it is that your interview is private if you want me to. I will make sure no one can overhear our conversations where I am.

Some people might be invited to a second interview and asked extra questions – it will be up to you whether you want to or not.

Expenses and payments

I can give people who take part up to £10 towards travel costs.

What are the possible benefits of taking part?

Many people like to know that they have helped with research which might help children and young people with CF in the future. We hope this research will help us to better support children and young people with CF.

What are the possible disadvantages and risks of taking part?

Sometimes talking about your experiences can be upsetting. I am trained to talk to children, young people and adults, and to help if someone becomes upset. I can also tell you about other people that might be able to help, for example the Cystic Fibrosis Trust. You can contact them via their website www.cysticfibrosis.org.uk or call their helpline on 0300 373 1000 or 020 3795 2184.

What will happen to the information given by me?

All information which is collected from you during the research will be kept strictly confidential, and any information which leaves _____ hospital will have names removed so that you cannot be recognised. There are only two occasions when I might need to break confidentiality, this would be if you were to say something that made me worry that you or someone else may be at risk. I would normally talk to you first before breaking confidentiality in this situation unless I thought this would lead to further risk to you.

Interviews will be audio recorded and transcribed, either by the researcher or by a professional transcriber who has signed a confidentiality agreement. Once transcribed, the audio recordings will be deleted. The interviews will be analysed by the researcher Naomi Fairweather. The only other people who will have access to your information are the other members of the research team, clinical psychologists Dr Fergal Jones, Dr Sally Clarke and Dr Martha Deiros Collado.

All of the information collected in this study will be stored securely, and all names and personal information will be removed to ensure the information is anonymous. Personal details that have been collected for the purposes of making contact with you and arranging an appointment to meet will be kept separately and securely, and will be destroyed at the end of the study. All information collected in interview will be kept in a password protected file at Canterbury Christ Church University, at _____ hospital and with me (Naomi Fairweather) for 5 years.

Your medical consultant will be informed that you are participating in the research but will not be given access to any audio recordings or transcripts. You are welcome to discuss this research with whoever you want, including friends, family, the doctor or clinical nurse specialist.

What if there is a problem?

I don't want anything about the research to be upsetting. If you are unhappy about anything to do with the research you can talk to me or a doctor or a nurse. You can also choose to drop out of the research at any time and you don't have to do so why. I can also tell you about other people who

might be able to help if you are upset. If you do get upset, I will tell your CF team so that they can support you.

If you have a complaint you should ask to speak to me and I will do my best to put it right (there is a phone number and email address at the end of this information sheet). If you are still unhappy or don't want to talk to me about it, you can talk to Professor Paul Camic, who is in charge of research at my university – paul.camic@canterbury.ac.uk, tel: _____ . There is also an NHS Complaints Procedure, you can ask anyone in the CF clinic about this.

What will happen to the results of the research study?

At the end of the research a short outline of the findings will be available for participants on request. Findings will be written up as part of a doctorate research project and it is hoped that results will be published in a professional journal. In the initial write up and any subsequent publications anonymised quotes from interviews will be used but care will be taken to make sure that participants are not identifiable.

Who is organising and funding the research?

This project is funded by Canterbury Christ Church University.

Who has reviewed the study?

All research in the NHS is looked at by independent group of people, called a Research Ethics Committee, to make sure it is safe and fair. This study has been looked at by _____ Research Ethics Committee.

What happens now?

If you would like to help us by taking part in this research, you can contact me directly or you can let me know when I contact you in a couple of weeks' time. We will need to arrange a time to meet at _____ hospital and will try to combine this with your usual clinic appointment. Alternatively we can arrange for the interview to be conducted via phone or videoconferencing app such as Skype. I will explain the study and answer any questions you might have.

Further information and contact details

If.....

- You would like to help us by taking part in this research
- You would like information about the research or like to know more about this study

Please contact:

Naomi Fairweather
Trainee Clinical Psychologist,
Salomons, Canterbury Christ Church University
Email: N.H.Fairweather323@canterbury.ac.uk

Telephone: You can leave a message for me on a 24-hour voicemail phone line at 03330117070. Please say that the message is for me, Naomi Fairweather and leave a contact number so that I can get back to you.

Thank you for reading about my study. I will contact you by phone within two weeks if I have not heard from you, to see if you want to take part.

Research Information Leaflet: Parents

Empowerment of children and young people living with cystic fibrosis during times of change

Hello. My name is Naomi Fairweather and I am a trainee clinical psychologist at Canterbury Christ Church University. I would like to invite you and/or your child to take part in a research study. Before you decide it is important that you understand why the research is being done and what it would involve. If you decide to take part please keep this information sheet for reference.



What is the purpose of the study?

We know that many children and young people with medical conditions want to feel confident and in control of their lives, but can face barriers. Empowering people with medical conditions has the potential to impact significantly on long term health and social outcomes. Much of what is known about patient empowerment focuses on adults. To the author's knowledge there is no research (with adults or children) looking specifically at cystic fibrosis (CF) and empowerment.

Why have I been invited?

I am interested in talking to children and young people aged 11-19 years living with CF, their parents, and staff. I want to ask some questions about living with CF during time of change, like starting a new school or college, becoming more independent at home, making new friends and having relationships, and moving to the adult CF team.

You and your child have been invited to take part because your child falls within an age range where lots of important changes happen and is under the care of a CF team in the UK. There is a separate information sheet for children and young people, but they may wish to read this one too.

We don't need all members of a family to take part. Some children might not want to even if their parent(s) chooses to. Some parents might not want to even if their child chooses to.

This information sheet applies if you are thinking about participating yourself, or if you are deciding whether or not to give consent for your child to participate. If your child is under 16 years old you will need to sign a consent form to say that you agree to them taking part. Even if you consent to their participation, they will only be eligible to participate if they want to.

Do I have to take part?

It is up to you (and your child) to decide whether or not to join the study. If you agree to take part, I will then ask you to sign a consent form. You are free to withdraw at any time, without giving a reason. This would not affect the care you or your child receive in any way.

What will happen to me if I take part?

If you decide to take part, I will meet with you (and/or your child) for an interview to ask some questions about CF and empowerment. Some information about your child's CF-related health will be collected from a database. I will be asking questions to children, parents, and staff in separate interviews. Interviews will take place at _____ Hospital or via telephone or Skype if preferred, at a time which is convenient for you. All interviews will last under an hour. I will be asking some questions about children and young people with CF and confidence in different situations like at home, school or college, and the hospital. No one will have to talk about anything they feel uncomfortable talking about.

If you or your child choose to be interviewed by phone or Skype, it is important to note that the researcher will not be able to guarantee that you/your child will not be overheard where you are. It is important that participants have privacy for their interview. If your child is being interviewed by phone or Skype, please try to ensure that no one can overhear them.

Some people might be invited to a second interview and asked extra questions – it will be up to you whether you want to or not.

Expenses and payments

I can reimburse travel costs of up to £10 per participant.

What are the possible benefits of taking part?

Many people like to know that they have helped with research which might help children and young people with CF in the future. We hope this research will help us to better support children and young people with CF.

What are the possible disadvantages and risks of taking part?

Sometimes talking about your experiences can be upsetting. I am trained to talk to children, young people and adults, and to help if someone becomes upset. I can also tell you about other people that might be able to help, for example the Cystic Fibrosis Trust. You can contact them via their website www.cysticfibrosis.org.uk or call their helpline on 0300 373 1000 or 020 3795 2184.

What will happen to the information given by me and/or my child?

All information which is collected from you or your child during the research will be kept strictly confidential, and any information which leaves _____ hospital will have names removed so that you cannot be recognised. There are only two occasions when I might need to break confidentiality, this would be if you or your child were to say something that made me worry that you or someone else may be at risk. I would normally talk to you and/or your child first before breaking confidentiality in this situation unless I thought this would lead to further risk to you/your child.

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Interviews will be audio recorded and transcribed, either by the researcher or by a professional transcriber who has signed a confidentiality agreement. Once transcribed, the audio recordings will be deleted. The interviews will be analysed by the researcher, Naomi Fairweather. All participants will be given the opportunity to check the accuracy of data held about them and correct any errors. The only other people who will have access to your information/your child's information are the other members of the research team, clinical psychologists Dr Fergal Jones, Dr Sally Clarke and Dr Martha Deiros Collado.

All of the information collected in this study will be stored securely, and all names and personal information will be removed to insure the information is anonymous. Personal details that have been collected for the purposes of making contact with you and arranging an appointment to meet will be kept separately and securely, and will be destroyed at the end of the study. All information collected in interview will be kept in a password protected file at Canterbury Christ Church University, at _____ hospital and with me (Naomi Fairweather) for 5 years.

Your child's medical consultant will be informed that you and/or your child is participating in the research but will not be given access to any audio recordings or transcripts. You and your child are welcome to discuss this research with whoever you want, including friends, family, the doctor or clinical nurse specialist.

What if there is a problem?

The questions are not intended to cause distress and we do not expect that your child will have any problems taking part. However, if your child is unhappy about anything relating to the study they can talk to their doctor or nurse. They also have the choice to withdraw from the study at any time of they want to.

Any complaint about the way that you or your child have been dealt with during the study or any concerns about possible harm they might suffer will be addressed. If you have a concern about any aspect of this study, you should ask to speak to me and I will do my best to address your concerns (see contact details at the end of this information sheet). If you remain unhappy and wish to complain formally, you can do this by contacting Professor Paul Camic, Research Director, Salomons Centre for Applied Psychology – paul.camic@canterbury.ac.uk, tel: _____. If you remain unhappy and wish to complain formally, you can do this through the NHS Complaints Procedure. Details can be obtained from the CF clinic

What will happen to the results of the research study?

At the end of the research a short outline of the findings will be available for participants on request. Findings will be written up as part of a doctorate research project and it is hoped that results will be published in a professional journal. In the initial write up and any subsequent publications anonymised quotes from interviews will be used but care will be taken to make sure that participants are not identifiable.

Who is organising and funding the research?

This project is funded by Canterbury Christ Church University.

Who has reviewed the study?

All research in the NHS is looked at by independent group of people, called a Research Ethics Committee, to protect your interests and those of your child. This study has been reviewed and given favourable opinion by _____ Research Ethics Committee.

What happens now?

If you and/or your child would like to help us by taking part in this research, they, or you, can contact me directly or you can let me know when I contact you in a couple of weeks' time. We can arrange a time to meet at _____ hospital and will try to combine this with your child's usual clinic appointment. Alternatively we can arrange for the interview(s) to be conducted via phone or Skype. I will explain the study and answer any questions you or your child might have.

I will ask all parents taking part to sign a consent form for themselves. If your child is 16 years old or over they are legally able to consent to participate and I will ask them to sign a consent form. If they are under the age of 16 years we will require your agreement for them to participate and I will ask you to sign a consent form on their behalf, and ask your child to sign an assent form.

Further information and contact details

If.....

- You and/or your child would like to help us by taking part in this research
- You would like information about the research or like to know more about this study

Please contact:

Naomi Fairweather
Trainee Clinical Psychologist,
Salomons, Canterbury Christ Church University

Email: N.H.Fairweather323@canterbury.ac.uk

Telephone: You can leave a message for me on a 24-hour voicemail phone line at 03330117070. Please say that the message is for me Naomi Fairweather and leave a contact number so that I can get back to you.

Thank you for reading about my study. I will contact you by phone within two weeks if I have not heard from you, to see if you and/or your child want to take part.

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Research Information Leaflet: Staff

Empowerment of children and young people living with cystic fibrosis during times of change

My name is Naomi Fairweather and I am a trainee clinical psychologist at Canterbury Christ Church University. I am contacting you to invite you to participate in my doctoral research study. Please retain this information sheet for your reference.



What is the purpose of the study?

Previous research has shown that many children and young people with medical conditions want to feel confident and in control of their lives, but can face barriers. Empowering people with medical conditions has the potential to impact significantly on long term health and social outcomes. Much of what is known about patient empowerment focuses on adults. To the author's knowledge there is no research (with adults or children) looking specifically at cystic fibrosis (CF) and empowerment.

Why have I been invited?

I am interested in talking to children and young people aged 11-19 years living with CF, their parents, and clinicians. You have been invited to take part because you work within a CF team in the UK. There are separate information sheets for children and young people, and parents.

Do I have to take part?

It is up to you to decide whether or not to join the study. If you agree to take part, I will then ask you to sign a consent form. You are free to withdraw at any time, without giving a reason.

What will happen to me if I take part?

If you decide to take part, I will meet with you for an interview to ask some questions about CF and empowerment. I will be asking questions to children, parents, and staff in separate interviews. Interviews will take place at _____ Hospital or via telephone or Skype if preferred, at a time which is convenient for you. All interviews will last under an hour. I will be asking some questions about children and young people with CF and confidence in different situations like at home, school or college, and the hospital. No one will have to talk about anything they feel uncomfortable talking about.

If you choose to be interviewed remotely, e.g. via telephone, the researcher cannot guarantee privacy at your location, but will ensure the interview cannot be overheard at the researcher's location.

You may be invited to a second interview. Consent to participating in this additional interview will be sought following the initial interview if applicable, and will not be assumed. You can withdraw from the study at any time.

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Expenses and payments

I can reimburse travel costs of up to £10 per participant.

What are the possible benefits of taking part?

There are no direct personal benefits to taking part in this study, but participants may help to make a contribution to the research evidence in this area.

What are the possible disadvantages and risks of taking part?

Discussing your clinical experience of working with children and young people with CF and their families may be distressing. I am trained to talk to children, young people and adults, and to help if someone becomes distressed. I have experience of conducting research involving healthcare professionals. I can also signpost to other sources of support.

What will happen to the information given by?

All information which is collected from you during the research will be kept strictly confidential, and any information which leaves _____ hospital will have names removed so that you cannot be recognised. There are only two occasions when I might need to break confidentiality, this would be if you were to say something that made me worry that you or someone else may be at risk. I would normally talk to you first before breaking confidentiality in this situation unless I thought this would lead to further risk to you/other people.

Interviews will be audio recorded and transcribed, either by the researcher or by a professional transcriber who has signed a confidentiality agreement. Once transcribed, the audio recordings will be deleted. The interviews will be analysed by the researcher, Naomi Fairweather. Other members of the research team who will have access to the data are clinical psychologists Dr Fergal Jones, Dr Sally Clarke and Dr Martha Deiros Collado.

All of the information collected in this study will be stored securely, and all names and personal information will be removed to ensure the information is anonymous. Personal details that have been collected for the purposes of making contact with you and arranging an appointment to meet will be kept separately and securely, and will be destroyed at the end of the study. All information collected in interview will be kept in a password protected file at Canterbury Christ Church University, at _____ hospital and with me (Naomi Fairweather) for 5 years.

You are welcome to discuss this research with whoever you want, including friends, family and colleagues.

What if there is a problem?

The questions are not intended to cause distress and we do not expect that you will have any problems taking part. However, if you are unhappy about anything relating to the study you can raise this with me or my university.

Any complaint about the way that you have been dealt with during the study or any concerns about possible harm anyone taking part might suffer will be addressed. If you have a concern about any aspect of this study, you should ask to speak to me and I will do my best to address

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your concerns (see contact details at the end of this information sheet). If you remain unhappy and wish to complain formally, you can do this by contacting Professor Paul Camic, Research Director, Salomons Centre for Applied Psychology – paul.camic@canterbury.ac.uk, tel:_____. If you remain unhappy and wish to complain formally, you can do this through the NHS Complaints Procedure.

What will happen to the results of the research study?

At the end of the research a short outline of the findings will be available for participants on request. Findings will be written up as part of a doctorate research project and it is hoped that results will be published in a professional journal. In the initial write up and any subsequent publications anonymised quotes from interviews will be used but care will be taken to make sure that participants are not identifiable.

Who is organising and funding the research?

This project is funded by Canterbury Christ Church University.

Who has reviewed the study?

All research in the NHS is looked at by independent group of people, called a Research Ethics Committee, to protect your interests and those of your child. This study has been reviewed and given favourable opinion by _____ Research Ethics Committee.

What happens now?

If you would like to help us by taking part in this research you, can contact me directly or you can let me know when I contact you in a couple of weeks' time. We will need to arrange a time to meet at _____ hospital. Alternatively we can arrange for the interview to be conducted via phone or Skype. I will explain the study and answer any questions you might have. I will ask everyone taking part to sign a consent form.

Further information and contact details

If.....

- You would like to help us by taking part in this research
- You would like information about the research or like to know more about this study

Please contact:

Naomi Fairweather
Trainee Clinical Psychologist,
Salomons, Canterbury Christ Church University
Email: N.H.Fairweather323@canterbury.ac.uk

Telephone: You can leave a message for me on a 24-hour voicemail phone line at 03330117070. Please say that the message is for me (Naomi Fairweather) and leave a contact number so that I can get back to you.

Thank you for reading about my study. I will contact you via phone if I have not heard from you within two weeks.

Form to be on headed paper)

Centre Number:




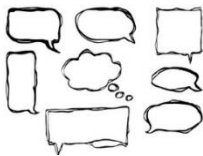
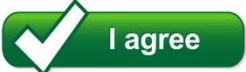
Participant Identification Number for this study:

ASSENT FORM

Title of Project: **Empowerment of children and young people living with cystic fibrosis during times of change**

Name of Researcher: Naomi Fairweather

If you agree, please put your initials in the boxes

<p>1. Yes, I have read the information sheet and understand this.</p>		
<p>2. Naomi gave me time to think about what I read and ask questions about it. I am happy with her answers and understand these.</p>		
<p>3. I understand that I am choosing to take part in this interview, and I can change my mind at any time and don't need to tell Naomi why. Choosing to take part doesn't affect my treatment or support from the service in any way. I might be asked to do a second interview but I don't have to say yes.</p>	<p>IT'S OKAY TO CHANGE YOUR MIND</p>	
<p>4. I agree that Naomi can record my interview. I am happy for her to use bits of what I say in her work as long as nobody could tell that it was me who was interviewed.</p>		
<p>5. I agree that Naomi can use quotes from my interview in any published papers as long as I can't be recognised.</p>		
<p>6. I agree to take part in the above study.</p>		

Name of Participant _____ Date _____

Signature _____

Name of Researcher taking Assent _____ Date _____

Signature _____

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Form to be on headed paper)

Centre Number:

Study Number:

Participant Identification Number for this study:

CONSENT FORM

Title of Project: **Empowerment of children and young people living with cystic fibrosis during times of change**

Name of Researcher: Naomi Fairweather

Please initial box

1. I confirm I have read and understand the information sheet dated [date] (version 2.0) for the above study.
2. I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily.
3. I understand that my participation is voluntary and that I am free to withdraw at any time without giving any reason, and without my care being affected in any way.
4. I understand that I may be invited to a second interview but can decide not to take part.
5. I agree to my interview being audio-recorded and anonymous quotes appearing in academic work.
6. I agree that anonymous quotes from the interview may be used in published reports of the study findings.
7. I agree to take part in the above study.

Name of Participant _____ Date _____

Signature _____

Name of Researcher taking Consent _____ Date _____

Signature _____

Parent consent form

[Trust logo] Version 2.0 14.04.18 IRAS Ref Number: 234289

Form to be on headed paper)

Centre Number:

Study Number:

Participant Identification Number for this study:

CONSENT FORM

Title of Project: **Empowerment of children and young people living with cystic fibrosis during times of change**

Name of Researcher: Naomi Fairweather

Please initial box

1. I confirm I have read and understand the information sheet dated [date] (version 2.0) for the above study.
2. I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily.
3. I understand that my participation is voluntary and that I am free to withdraw at any time without giving any reason, and without my child's care being affected in any way.
4. I understand that I may be invited to a second interview but can decide not to take part.
5. I agree to my interview being audio-recorded and anonymous quotes appearing in academic work.
6. I agree that anonymous quotes from the interview may be used in published reports of the study findings.
7. I agree to take part in the above study.

Name of Participant _____

Signature _____

Name of Researcher taking Consent _____ Date _____

Signature _____

[Trust logo] Staff consent form

Version 2.0 14.04.18 IRAS Ref Number: 234289

Form to be on headed paper)

Centre Number:

Study Number:

Participant Identification Number for this study:

CONSENT FORM

Title of Project: **Empowerment of children and young people living with cystic fibrosis during times of change**

Name of Researcher: Naomi Fairweather

Please initial box

1. I confirm I have read and understand the information sheet dated [date] (version 2.0) for the above study.
2. I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily.
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4. I understand that I may be invited to a second interview but can decide not to take part.
5. I agree to my interview being audio-recorded and anonymous quotes appearing in academic work.
6. I agree that anonymous quotes from the interview may be used in published reports of the study findings.
7. I agree to take part in the above study.

Name of Participant _____

Signature _____

Name of Researcher taking Consent _____ Date _____

Signature _____

Child and young people interview schedule

Thank you for taking part in my research. I am interested in finding out how children and young people living with CF experience empowerment in different situations (like home, school and the clinic/hospital). It is difficult to say exactly what empowerment is, some people think that empowerment involves having enough information and understanding it, having choices and being in control, and being able to cope with difficult situations and feelings – I am interested in any other ideas you might have about empowerment. I want to know what helps children and young people feel empowered, what gets in the way and what the consequences of empowerment are. I want to ask you some questions today about yourself, about CF and about your experiences of empowerment at home, at school and at the hospital. If there are any questions you do not want to answer that is OK, just let me know.

- Tell me a little bit about yourself [what sorts of things are you interested in? what sorts of things do you like doing?]
- When do you feel most confident?
- What is it like to be a [age] year old with CF? [at home, at school, with friends, at the hospital]
- How do you handle change? [what important changes have there been in your life so far? What changes do you think will come up in the future?]
- How do you take care of yourself? (What challenges have you had? Can you give me an example of a time you've handled things really well? What helped you do this?)
- What helps you manage CF? [what helps you manage difficult things about CF or CF worries?]
- What do you think could be done to help children feel more in charge of CF?
- What do you think about transitioning to the adult CF team? Do you have any hopes or fears?
- What hopes or wishes do you have for your life?
- If you had to give a message to hospital staff wanting to learn about how to help children with CF what would it be? [what about friends, teachers, parents?]

We have now reached the end of the interview – do you have any questions for me or anything else you would like to say before we finish? Thank you for taking part in my study.

Parent interview schedule

Thank you for taking part in my research. I am interested in finding out how children and young people living with CF experience empowerment during key life transitions. I want to find out what helps children and young people feel empowered, what gets in the way and what the consequences of empowerment are. I want to ask you some questions today about your child, about CF and about your child's experiences at school, with friends, at home and at the hospital. If there are any questions you do not want to answer that is OK, just let me know.

- In what situations is your child most confident? [what is it about those situations that means they feel more confident?]
- How much do you think your child knows about CF? (where did they get the information? How has this changed over time?)
- How does your child handle change? [what important changes have there been in their life so far? What changes do you think will come up in the future? What do you think helps them handle change? What gets in the way?]
- How does your child take care of her/himself (What challenges has your child had? Can you give me an example of a time when your child handled things really well? What helped them do this?)
- What bits of your child's CF care are they doing? What bits are you/other people doing? How has this changed as they have got older?
- What helps your child manage CF related worries?
- What do you think could be done to help children feel more in charge of CF?
- What are your thoughts about your child transitioning to adult services? What are your fears? What are your hopes?
- What hopes do you have for your child's life?
- If you had to give a message to hospital staff wanting to learn about how to empower children with CF what would it be? [what about your child's friends, teachers, other parents of children with CF]

We have now reached the end of the interview – do you have any questions for me or anything else you would like to say before we finish? Thank you for taking part in my study.

Clinician interview schedule

Thank you for taking part in my research. I am interested in finding out how children and young people living with CF experience empowerment in different settings. I want to find out what helps children and young people feel empowered, what gets in the way and what the consequences of empowerment are. I want to ask you some questions today about the children and young people you work with, about CF and about your perceptions of children and young peoples' experiences of empowerment at home, at school and in the hospital. If there are any questions you do not want to answer that is OK, Just let me know.

- When do you think the children and young people you work with feel most confident? [what do you think it is about those situations that helps them feel confident?]
- How much do you think the children and young people you work with (aged 11-19) know about CF? (where did they get the information? How has this changed over time?)
- How do you think the children and young people you work with handle change? [what important changes do they face? What do you think helps them handle change? What gets in the way?]
- Let's talk about children and young people with CF taking care of themselves (What challenges do you think the children and young people you work with have? [without giving any identifying details] Can you give me an example of a time a child handled things really well? What helped them do this?)
- What bits of CF care do you think children and young people aged 11-19 are they doing themselves? What bits are you/other people doing? How does this changed as they have got older?
- What helps children and young people aged 11-19 manage CF related worries?
- What do you think could be done to help children and young people aged 11-19 feel more in charge of CF? [at home, with friends, at school, at the hospital]
- What hopes do you have for the lives of the children and young people aged 11-19 that you work with?

- If you had to give one message to other hospital staff wanting to learn about how to empower children and young people with CF what would it be? What message would you give to parents about how to empower children and young people with CF?

We have now reached the end of the interview – do you have any questions for me or anything else you would like to say before we finish? Thank you for taking part in my study.

Confidentiality Statement for Persons Undertaking Transcription of Research Project Interviews

Project title: Empowerment of children and young people with cystic fibrosis during times of change

Researcher's name: Naomi Fairweather

The tape/s or recording/s you are transcribing have been created as part of a research project. Tapes may contain information of a very personal nature, which should be kept confidential and not disclosed to others. Maintaining this confidentiality is of utmost importance to the University. Signing this form means you agree not to disclose any information you may hear on the recording to others, and not to reveal any identifying names, place-names or other information on the recording to any person other than the researcher/s named above. You agree to keep the recording in a secure place where it cannot be accessed or heard by other people, and to show your transcription only to the relevant individual/s who is involved in the research project, i.e. the researcher/s named above.

You will also follow any instructions given to you by the researcher about how to disguise the names of people and places talked about on any recordings as you transcribe them, so that the written transcript will not contain such names of people and places.

Following completion of the transcription work you will not retain any recordings or transcript material, in any form. You will pass all tapes back to the researcher and erase any material remaining on your computer hard drive or other electronic medium on which it has been held.

You agree that if you find that anyone speaking on a tape is known to you, you will stop transcription work on that tape immediately and pass it back to the researcher.

Declaration

I agree that:

1. I will discuss the content of the recording/s only with the researcher/s named on the previous page.
2. I will keep all recordings in a secure place where they cannot be found or heard by others.
3. I will treat the transcripts of the recordings as confidential information.
4. I will agree with the researcher how to disguise names of people and places on the recordings.
5. I will not retain any material following completion of transcription.
6. If the person being interviewed on a recording is known to me I will undertake no further transcription work on the recording and will return it to the researcher as soon as is possible.

I agree to act according to the above constraints

Your name _____

Signature _____

Date _____

Occasionally, the conversations on recordings can be distressing to hear. If you should find it upsetting, please speak to the researcher.

Appendices S-W have been removed from the electronic copy

Appendix X. Coding table

As hundreds of open codes were produced, the following table contains examples of from each category rather than an exhaustive list.

Selective code	Axial codes	Open codes
Having a team	Team membership	<ul style="list-style-type: none"> • Ref to having a team • YP ref to role of family • Parent ref to role of family • Prof ref to role of family • Ref to at least one good friend • YP ref to role of friends • Parent ref to role of friends • Prof ref to role of friends • Ref to role of medical team • Ref to role of school • Ref to CF community
	Provides a sense of acceptance/ 'normalising'	<ul style="list-style-type: none"> • Parent ref to belonging • Prof ref to belonging • YP ref to being accepted • YP ref to fitting in
	Share burden of CF	<ul style="list-style-type: none"> • Ref to sharing responsibility • Ref to practical support • Ref to emotional support • Ref to multiple supportive roles
	Makes room for mistakes/ learning from experience	<ul style="list-style-type: none"> • Prof describing role as coaching • Parent talking about YP learning from experience • Prof reflecting on role of mistakes
	Trusting relationships	<ul style="list-style-type: none"> • YP reflecting on trusting relationships • Parent ref to trusting relationships • Prof ref to trusting relationships
Taking charge and 'having a voice'	Information and understanding	<ul style="list-style-type: none"> • YP ref to interacting with information about CF • Parent ref to information and understanding • Prof ref to information and understanding • Ref to technology

	Active participation in decision-making	<ul style="list-style-type: none"> • YP reflecting on making decisions • Parent reflecting on making decisions • YP reflecting on having a voice • Parent highlighting importance of YP having a voice • Parent ref to YP confidence • Prof reflecting on making decisions
	Explaining to others	<ul style="list-style-type: none"> • YP reflecting on explaining to others • YP reflecting on sharing personal information • YP ref to raising awareness • Parent reflecting on YP explaining to others • Prof ref to YP explaining to others
	'Finding ways round and through'/ practical problem solving	<ul style="list-style-type: none"> • YP reflecting on asking for help • YP describing coping strategies • YP describing ways around limitations • Parent describing YP's coping strategies • Parent ref to YP facing challenges • YP making the most of things • YP ref to routine • YP talking about the importance of hope • Ref to preparing for future
Thriving alongside CF	Being able to just be a child/getting on with life	<ul style="list-style-type: none"> • YP talking about who they are • YP ref to non-CF challenges • Parent ref to child's life being more than CF • Prof hopes for the future of YP they support • Ref to priorities
	Self-acceptance/dealing with difference	<ul style="list-style-type: none"> • Ref to dealing with difference • YP expressing acceptance • YP reflecting on developing confidence • YP sharing stories of survival • YP showing wisdom

	Achieving in relation to own goals	<ul style="list-style-type: none"> • YP describing achieving • YP showing determination • YP sharing hopes for the future • Parent hopes for their child's future • Prof ref to YP goals • Prof ref to YP being determined • Parent ref to YP helping others
	Having adventures and fun	<ul style="list-style-type: none"> • YP ref to enjoyment • YP describing having adventures • Prof ref to YP having adventures
Concealing self	Other people's assumptions	<ul style="list-style-type: none"> • Reflecting on other people's assumptions • Parent ref to responses of other's • Ref to lack of flexibility
	Time/difficulties fitting it all in	<ul style="list-style-type: none"> • YP ref to time • YP describing the challenge of CF • Prof ref to logistical challenges
	Having to keep secrets/concealing self	<ul style="list-style-type: none"> • Struggling with being different • Having to keep secrets
	Unheard voices	<ul style="list-style-type: none"> • Unheard voices
Wider context	Unique individuals	<ul style="list-style-type: none"> • YP explaining it's all about the individual • Ref to individual differences
	Interaction with developmental stuff: changing emotional, physical, cognitive (maturation)	<ul style="list-style-type: none"> • Ref change over time – responsibility • Ref to change – getting older • Prof reflection on interaction with developmental norms
	Constantly changing landscape of CF	<ul style="list-style-type: none"> • YP reflecting on change over time in own CF • Prof reflecting on change over time in disease • Ref to changing landscape of CF

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Appendix Z. Abridged Research Diary

March 2017

Visited the children's hospital cystic fibrosis (CF) team. They were really encouraging and I'm feeling all fired up and excited about the project. They highlighted transitions (e.g. between primary and secondary schools) as a particularly challenging time for children and young people (CYP) with CF, their families and the medical team. I'll amend my interview schedule accordingly.

June 2017

I've been reflecting on my choice of methodology. I chose to do a qualitative project despite my research background being very strongly quantitative because I find qualitative research interesting to read and useful clinically, and I knew if I didn't do qualitative research during training I never would. Trying to get into the qualitative mindset is proving a real challenge – it's such a different way of approaching things. I've been trying to get my head around an introductory grounded theory text and I'm beginning to wonder if my choice of research design was brave or daft!

August 2017

I've been reflecting on my choice of topic and client group. I felt a lot of pressure to choose a topic very early on in training during a time when I was pre-occupied with getting to grips with clinical work and navigating the academic demands of training. I think I fell into choosing a topic that was familiar both professionally (I'd worked as an assistant psychologist in paediatrics prior to training) and personally (I have my own experiences of empowerment/disempowerment having grown up with long term health conditions). I'm passionate about the topic but it turns out that empowerment is a really complex concept and

sometimes I wonder if the research process would be easier if I'd chosen a topic less close to home.

March 2018

I attended the REC meeting today, it's taken a lot longer than I'd hoped to get to this point and I got a real grilling from the committee (15 panel members!). I don't think they really understood qualitative research or CF as they asked me lots of questions about how I was going to achieve a representative sample and suggested that I run in person focus groups with children with CF (big infection control no no!)

June 2018

My project has finally got ethics approval. Now to get R&D approvals for my two Trusts.

July 2018

I've been trying to find a question for my literature review for what feels like months! I feel conflicted about doing a literature review prior to my grounded theory study as this is not in keeping with the method.

August 2018

I've finally heard back from one of the R&D departments. They want me to do some training that is only really appropriate for large drug trials! I'm getting frustrated (and a bit panicked) about all the hoops!

October 2018

Recruitment is so slow, I'm really worried. Have submitted an amendment to enable recruitment from CF charities and social media.

Early December 2018

Recruitment has continued to be really slow and I've found approaching potential participants much harder than I expected. I have my first interviews booked in for later this month

Late December 2018

Disaster!! I had 3 interviews booked in but ended up ill in hospital myself. I really hope that the participants will be understanding and that we can re-book.

January 2019

I've just done my first interview and I'm buzzing – the YP I spoke to was so reflective and insightful, lots of really rich data. I feel re-invigorated after the gruelling ethics process.

January 2019

I'm to figure out how to analyse the data. I've read at least 4 books about grounded theory and there seem to be lots of ways of doing it and no way of knowing if you're doing it 'right'! Getting really bogged down in fine details.

Early February 2019

Recruitment really picked up, I've been a bit inundated! I'm really enjoying the interviews – I've heard some amazing stories. I feel a lot of pressure with the data analysis – time is precious for CYP with CF and their families and clinicians, I want to do a good job for all the people who have taken the time to talk with me.

March 2019

I have all my data now and am slowly getting there with the initial coding but don't think I can get this done before the deadline.

April 2019

I've decided to defer – this felt like a really difficult decision but I need to concentrate on my clinical work.

Late September 2019

I'm now focusing on research full time. I've completed the line by line coding and have developed axial codes. I've been spending a lot of time thinking about what the data is saying and how my own experiences might impact on the data analysis – feeling very grateful for other trainees and friends that know nothing about empowerment in CF, they have been excellent sounding boards!

October 2019

I've been working on the relationships between my axial categories – the model is beginning to take shape. I'm feeling excited about part B again.

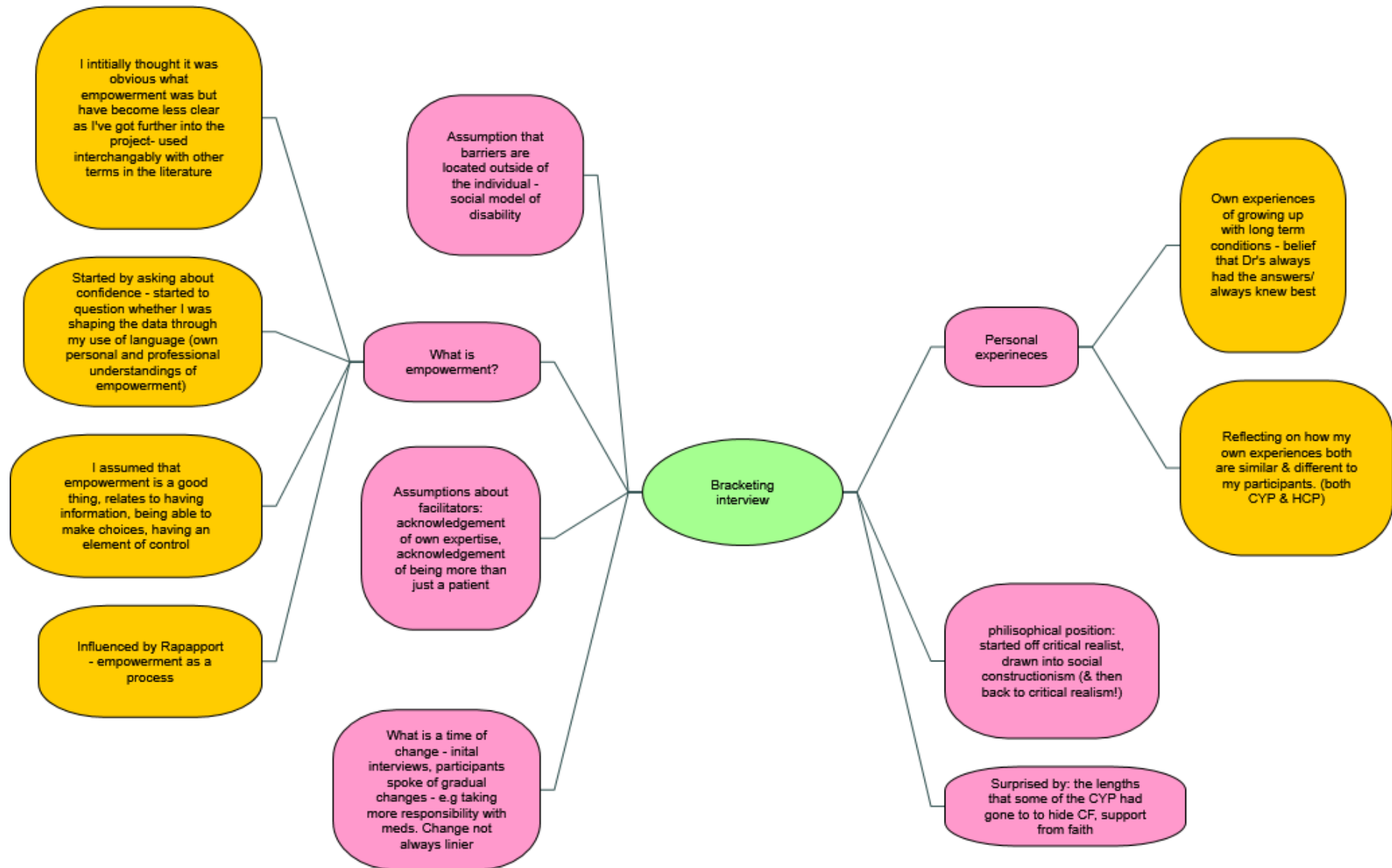
November 2019

I think this qualitative literature review is probably one of the most academically challenging things I've ever done. I think the structure of traditional systematic reviews suits me much better!

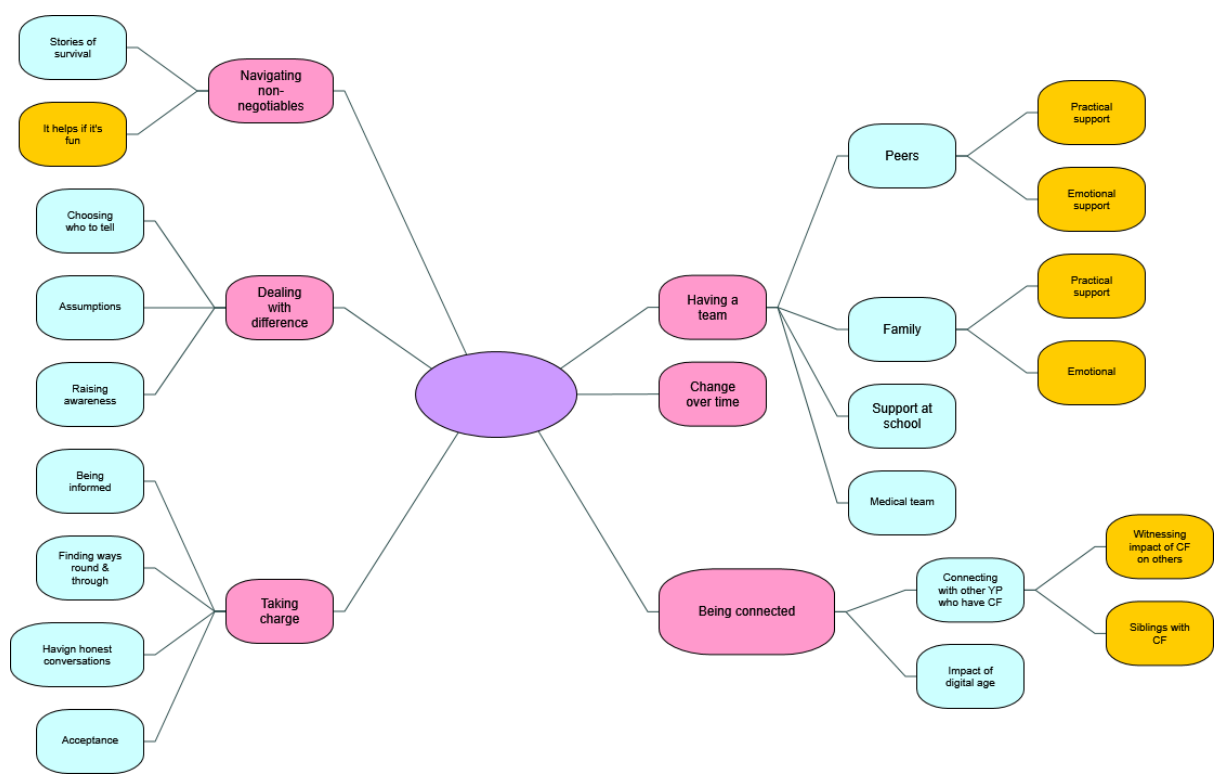
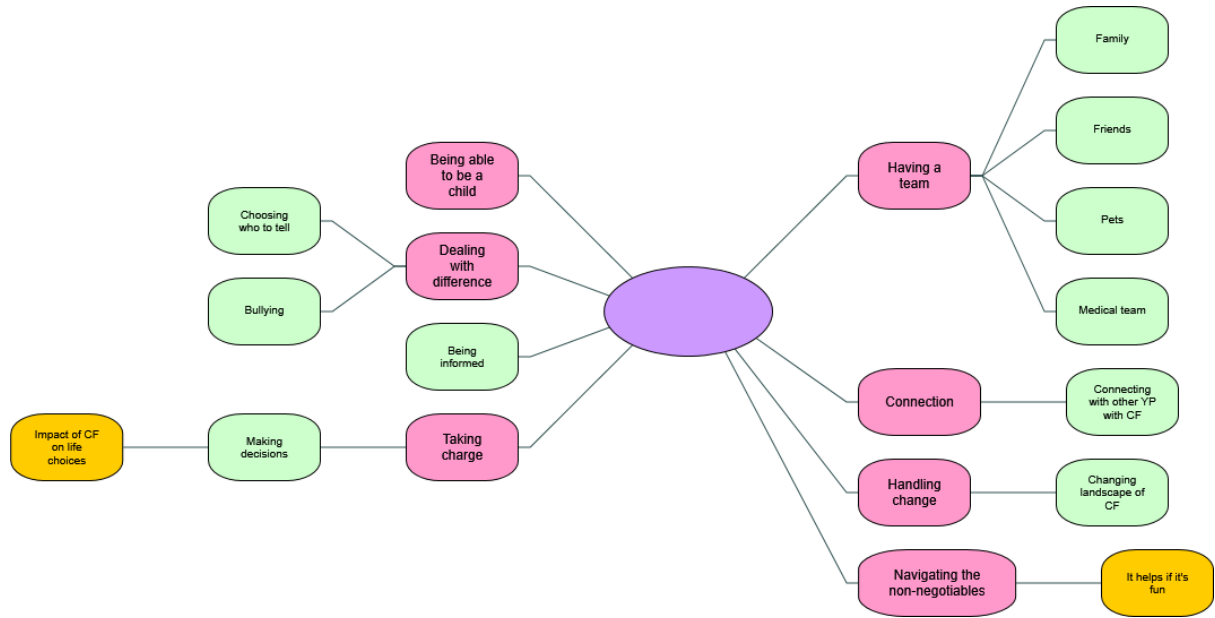
December 2019

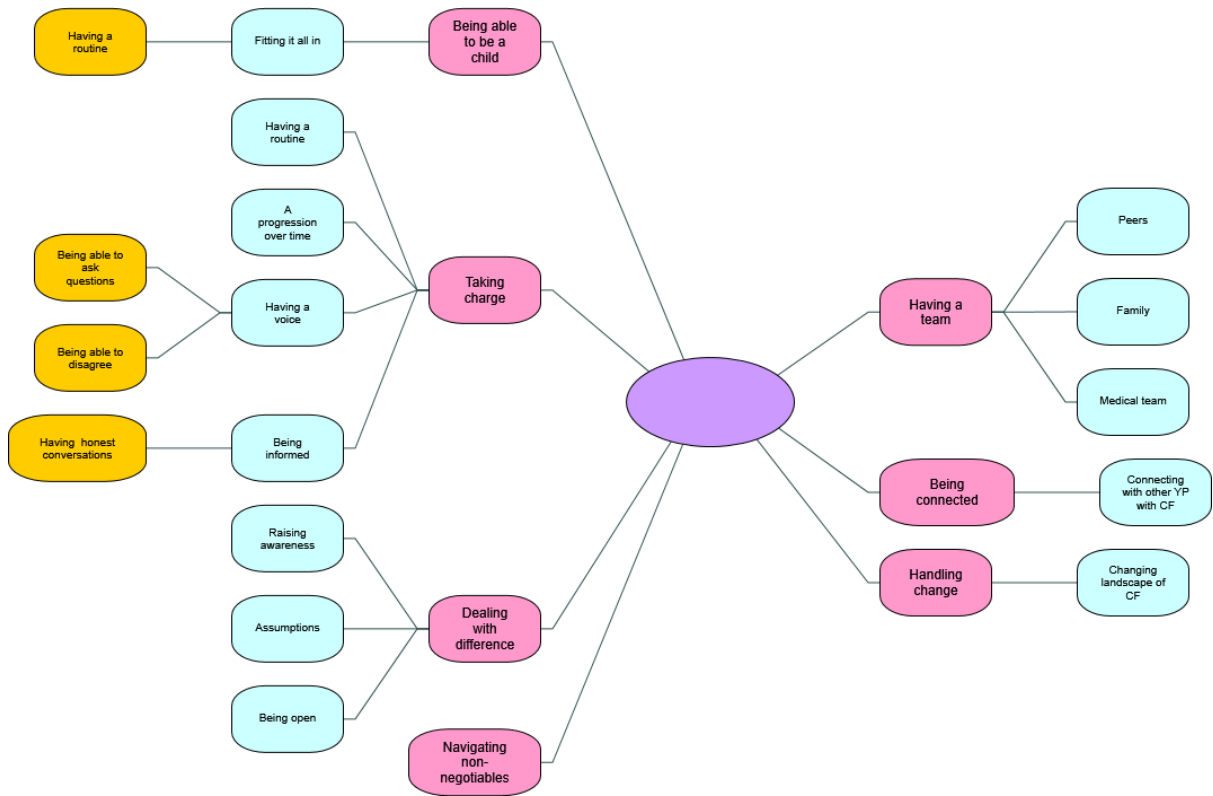
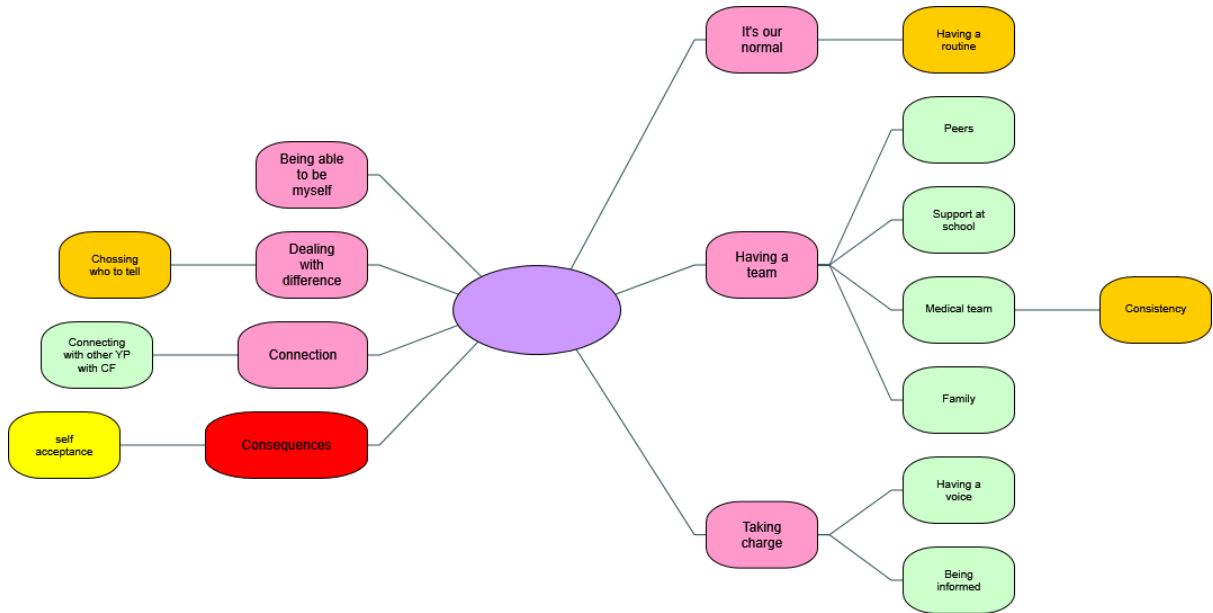
Final drive for completion. Exhausted but glad to be handing it in!

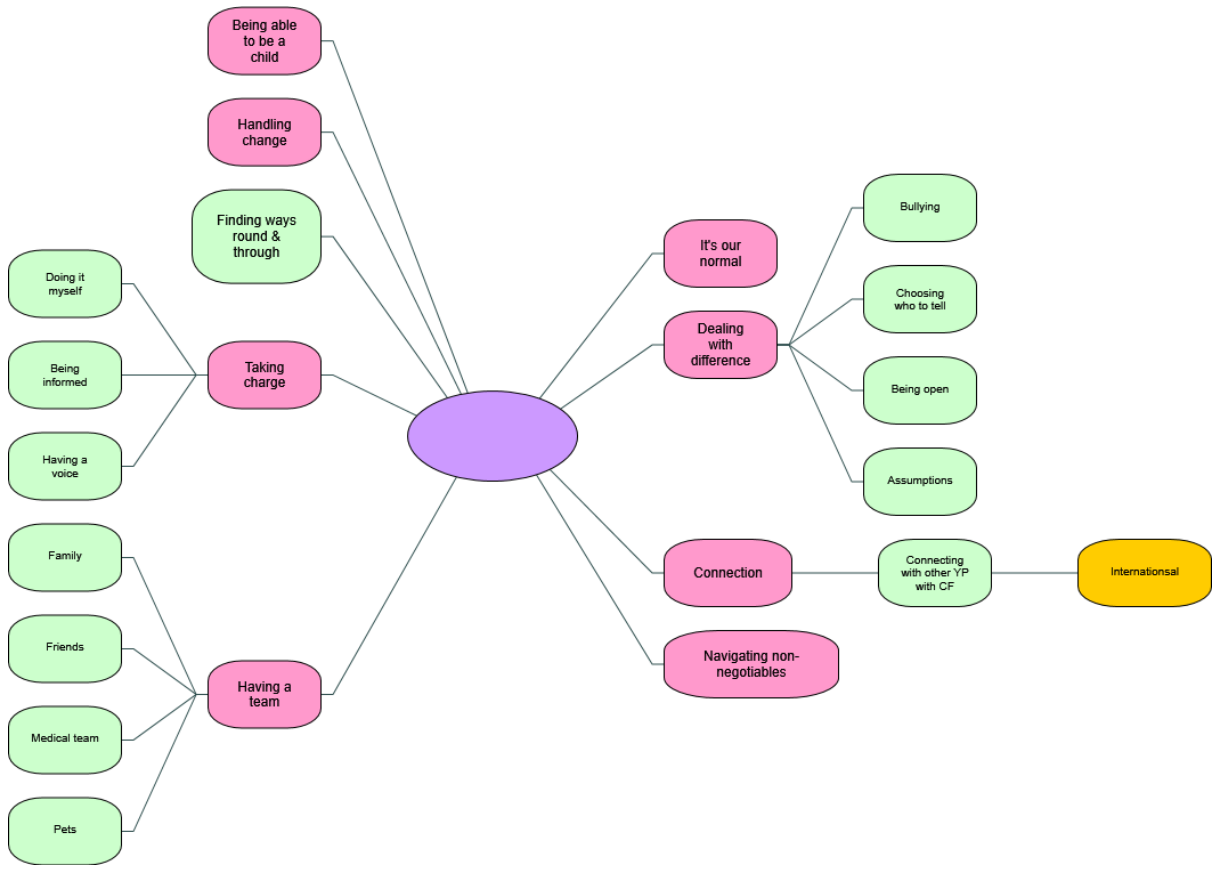
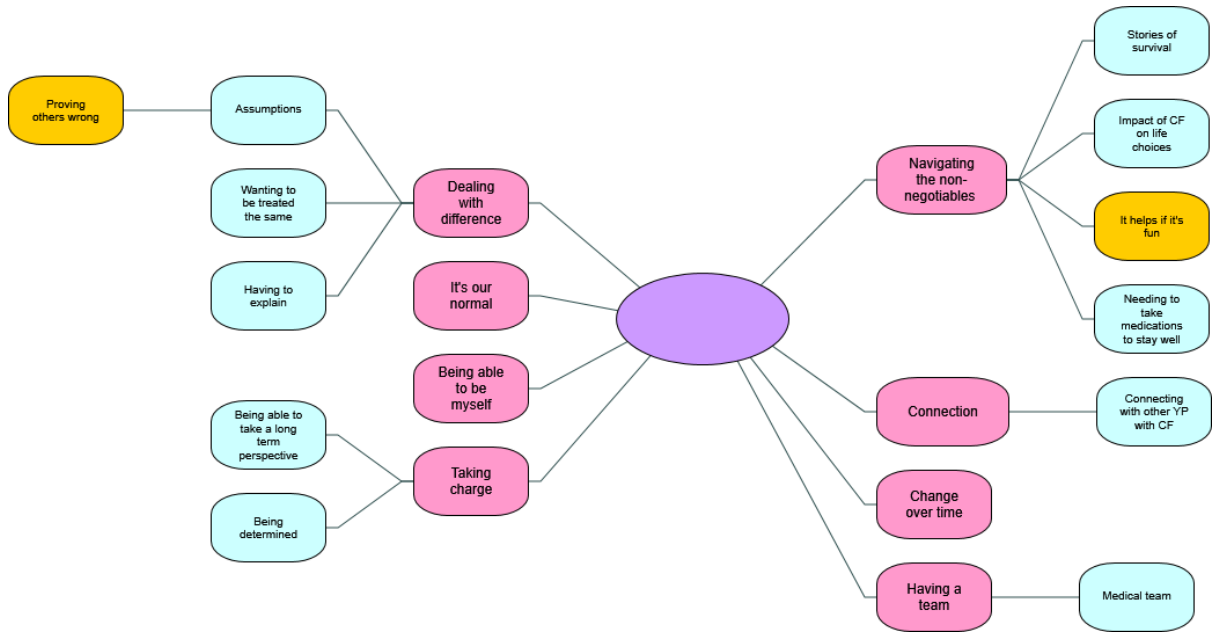
Appendix AA. Bracketing interviews mind map

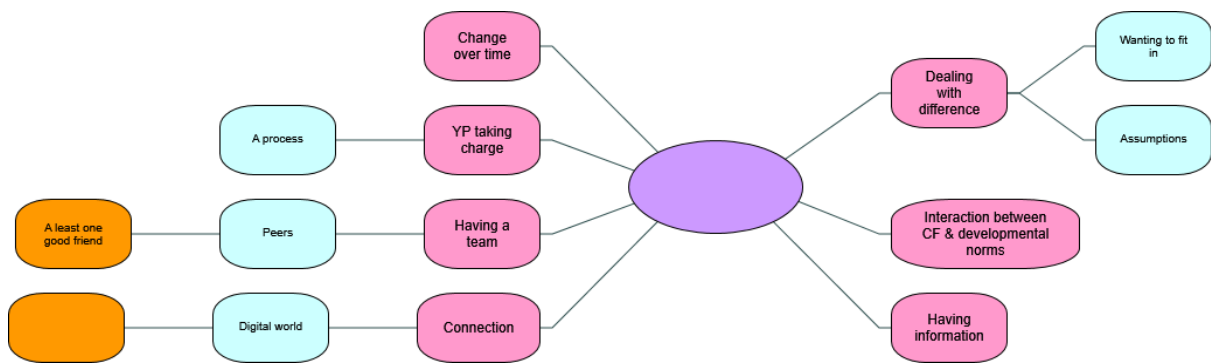
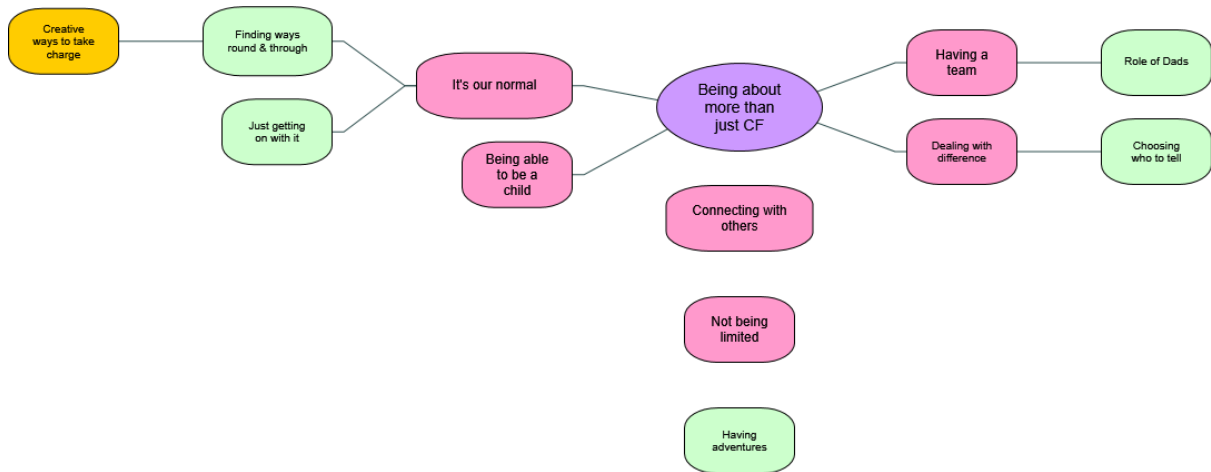


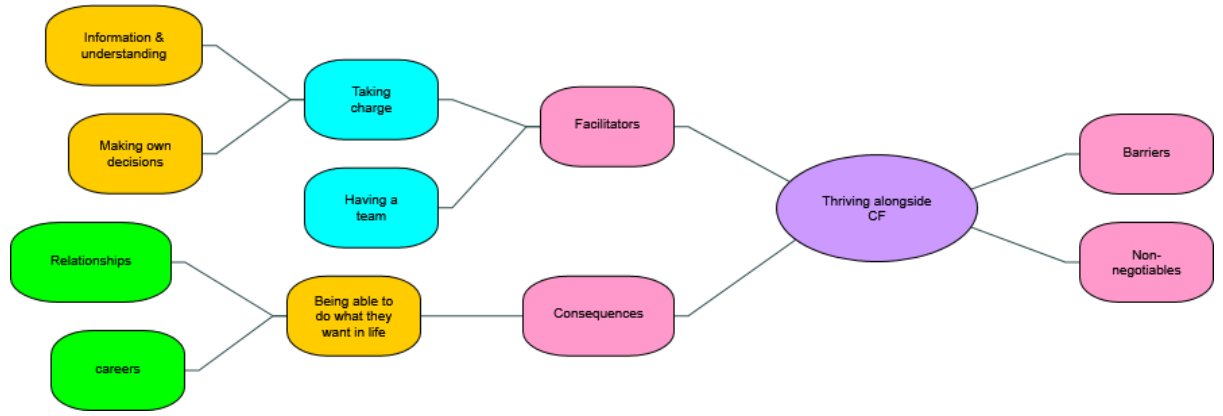
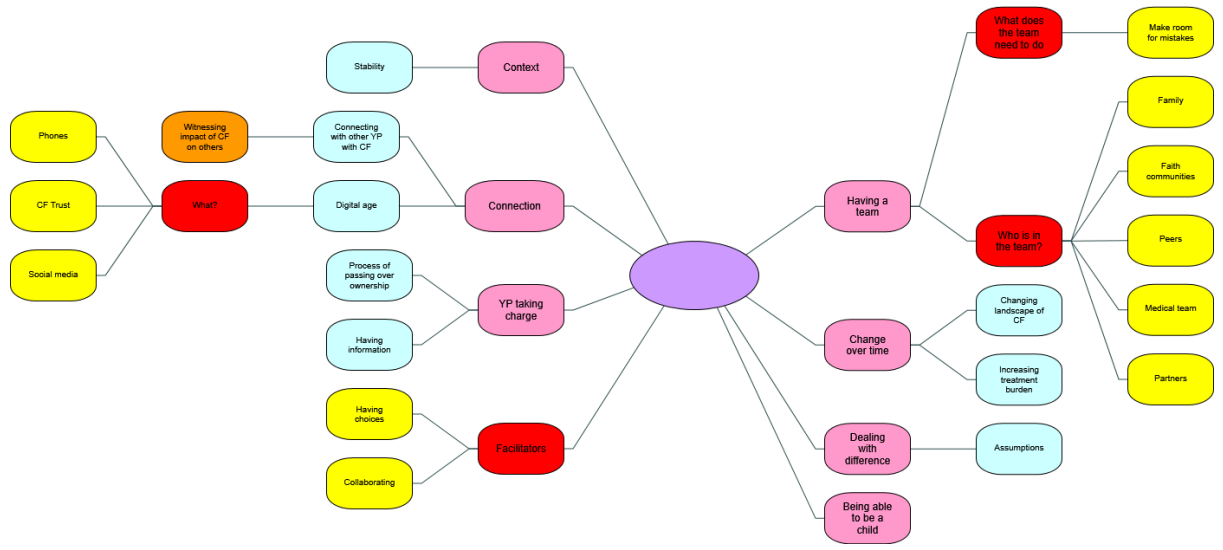
Appendix BB. Theoretical diagrams

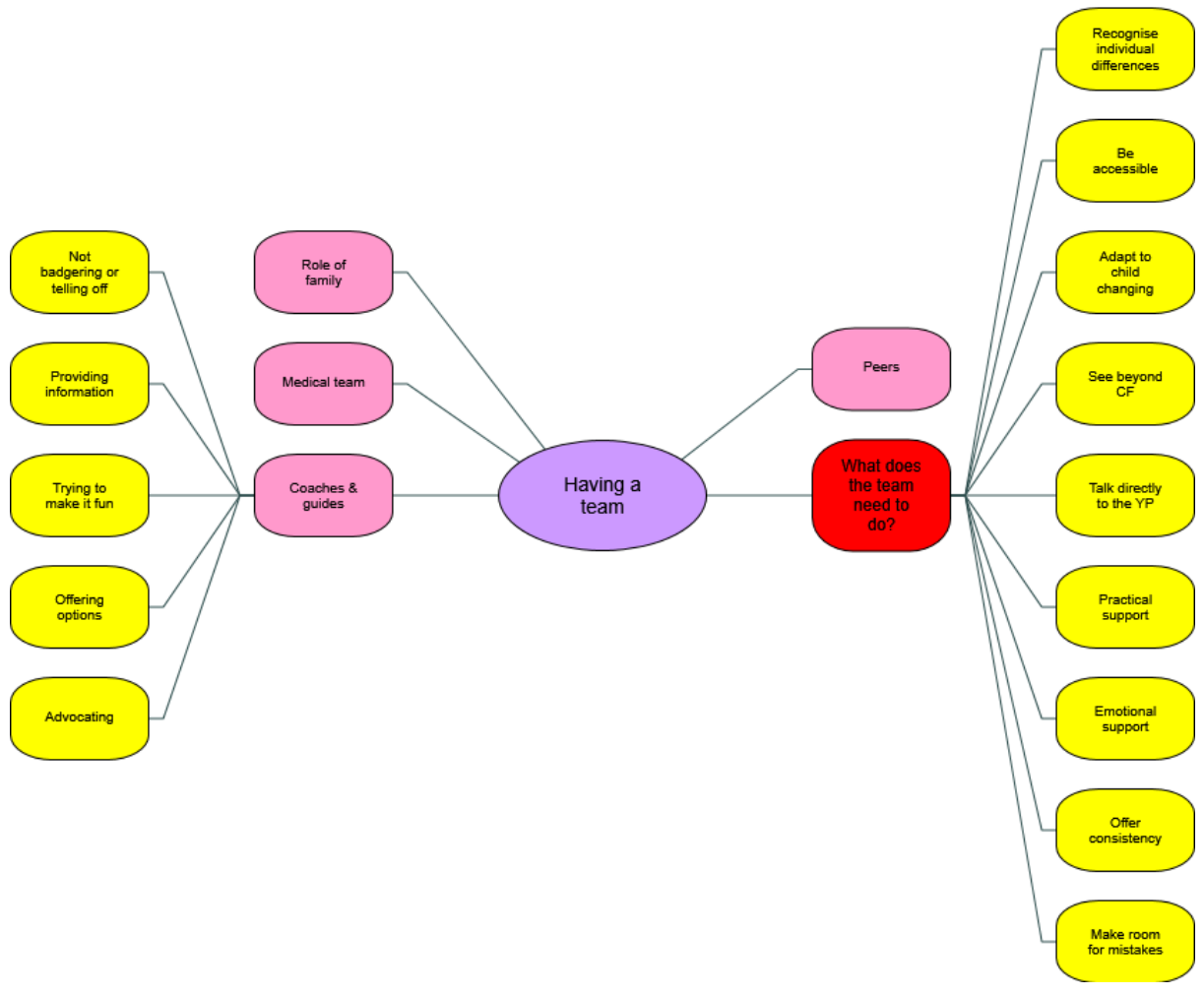


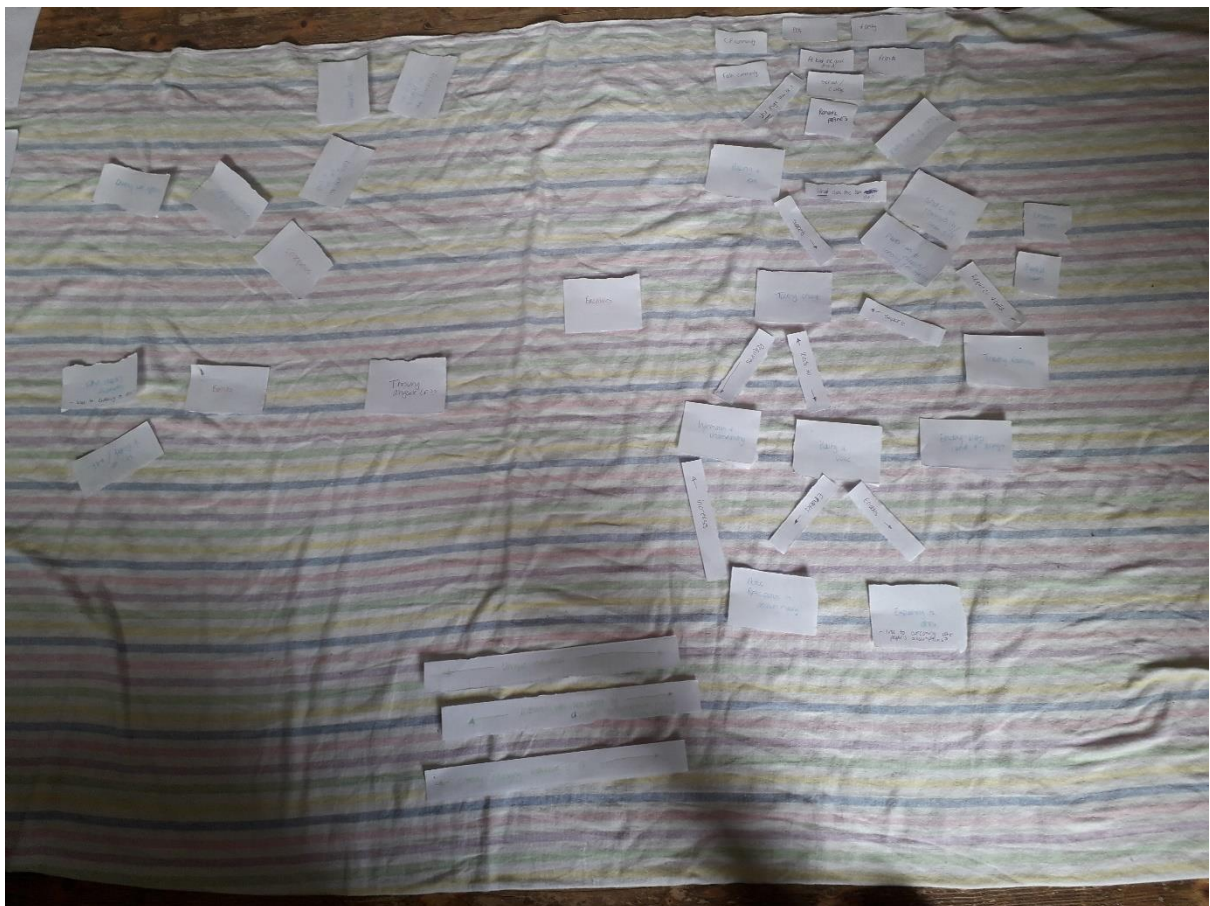
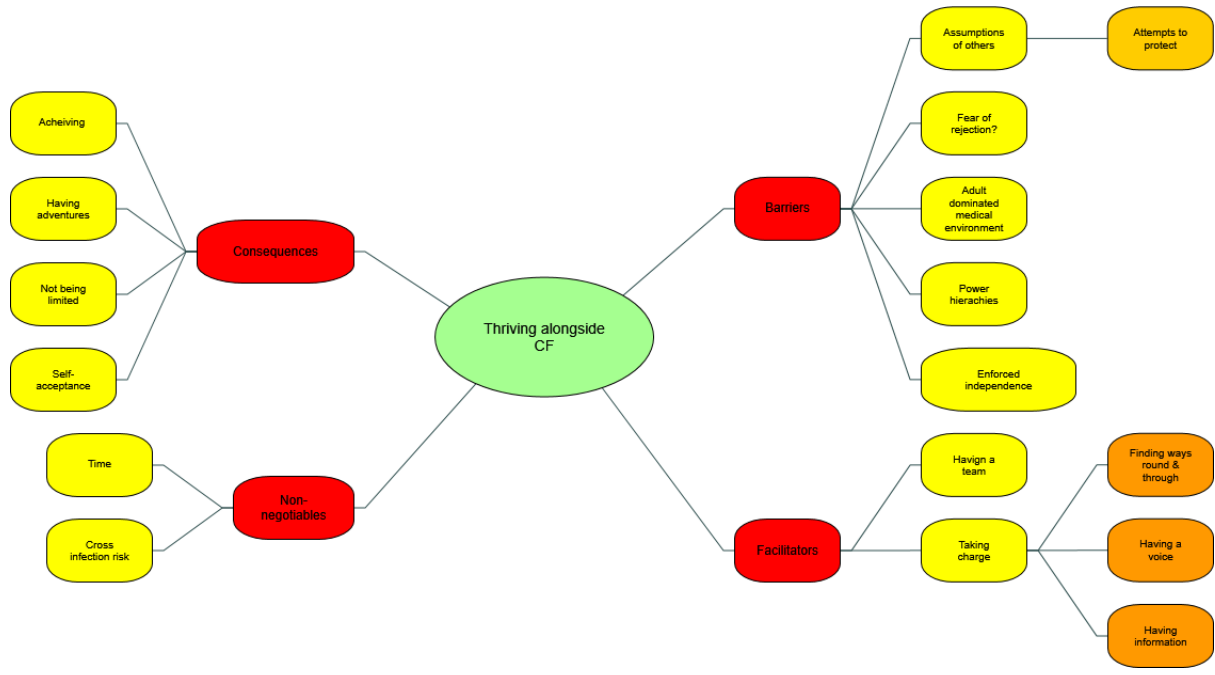


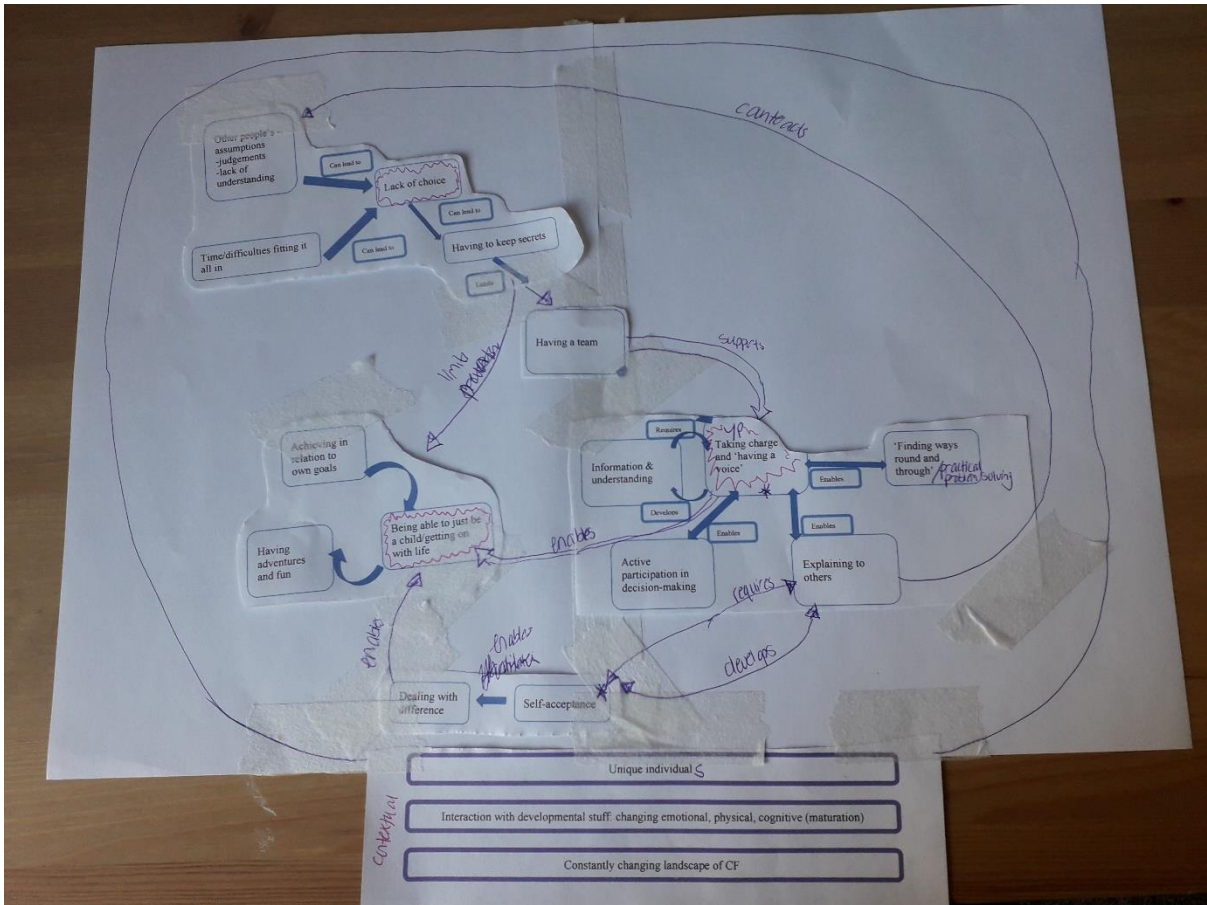












Appendix CC. Example theoretic memos

Barriers:

Is keeping secrets a barrier? or could it be a choice? maybe having to keep secrets is a barrier? choosing to keep some things private could be part of taking charge.

Are lack of choice and lack of flexibility the same concept? All participants indicated that having choice and input into decisions was important. Some also referred to lack of societal flexibility e.g around allowing children with CF to eat what they need to eat in the context of 'healthy schools'

Not sure how to code participant suggestions for improving medical and/or school systems - could this be coded under barriers as it helps explain what some of the sub-category mean? For e.g. coding suggestions about making clinical environments more child friendly under 'adult dominated systems'? I suppose they could also be coded as 'being able to be a child'

Being able to be a child:

Being able to be a child or being able to be myself? Depends who's perspective this is from - some of the young people taking part wouldn't necessarily identify as children even if their parents would describe them as such. Does one code need to fit all instances? Need to think about the language used.

Is 'being able to be a child/just getting on with life' the same as 'thriving alongside CF'? I'm feeling more confident that 'thriving alongside CF' is the central phenomena as it does seem to be the thing that links all of the data. I wonder if 'being able to be a child/just getting on with life' is a part or consequence of 'thriving alongside CF' - for example Mum of 11yr old F seemed to suggest that her daughter was able to just be a child through escapism (although could that in itself be a way of taking charge?!).

Taking charge:

Are being in charge and taking charge different concepts? The YP often refer to situations where they have actively taken charge rather than someone else putting them in charge.

Adults (professionals and parents?) more likely to refer to external circumstances (e.g. starting secondary school) that lead to the YP being in charge.

Is 'holding it all together' what happens when a YP has taken charge? or is that a separate code?

How does taking charge relate to changes in responsibility? The professionals tend to see taking charge as having complete responsibility but interviews with YP suggest that sharing responsibility can be a way of taking charge in a way that enables them to fit in their non-CF priorities.

I initial thought of preparing for the future as part of taking charge but now think it might be more of a consequence of thriving alongside CF.

Appendix DD. Feedback form for ethics panel and R&D departments

Study feedback summary

Name: Naomi Fairweather

Study: 'Thriving alongside CF': Developing A Grounded Theory of Empowerment in Children & Young People with Cystic Fibrosis During Key Life Transitions

REC reference: 18/LO/0450

Whilst the importance of patient empowerment is increasingly being recognised, little is known about empowerment in children and young people (CYP) with long term conditions. Empowerment may be particularly important in CYP with cystic fibrosis (CF) due to high treatment burden and limited opportunities for peer support.

This study employed a Grounded Theory Method to develop a preliminary theory of empowerment in CYP with CF.

Ethical practice was guided by the BPS code of human research ethics (The British Psychological Society [BPS], 2014) and the BPS code of ethics and conduct (BPS, 2018). Approvals were granted by the REC, HRA and relevant R&D departments.

Participants were recruited from two NHS Trusts. Informed consent was obtained for all participants, with those under 16 years providing assent. Seven young people with CF, four parents and four professionals were interviewed. All participants were fully debriefed, no participants reported being distressed by the research.

Interviews were audio recorded and professionally transcribed (a confidentiality agreement was signed), data were analysed using Grounded Theory.

The emerging model suggests that 'thriving alongside CF' may be supported by interactions between 'having a team' and 'taking charge and having a voice', leading to 'being able to just be a child/getting on with life', that 'concealing self' may get in the way of 'thriving alongside CF' and that these processes occur within wider medical and developmental contexts.

Comparisons were made with existing models of patient empowerment which were based on research with adults living with a range of long term conditions. Some similarities were noted, particularly in relation to facilitators of empowerment. For e.g. having information featured as a key facilitator of empowerment in previous models of empowerment and was found to enable 'taking charge and having a voice' in the current study. The current study moved beyond previous models of patient empowerment by incorporating developmental perspectives and factors specific to CF such as the impact of rapid medical advances and complexities of peer support in the context of strict infection control guidelines.

Limitations of the current study included relatively small sample size, and lack of respondent validation. Clinical and research implications were discussed including, a need for research with CYP who are not currently 'thriving with CF' and whose voices were not heard in the current study, increased focus on CYP's support networks in clinic consultations and the potential for developing virtual peer support interventions.

Findings of the study are currently being disseminated to all participants and relevant stakeholders.

‘Thriving alongside CF’ - Empowerment in Children & Young People with Cystic Fibrosis (CF) During Times of Change

Thank you for participating in my research and helping me to understand what might help children and young people living with CF to feel confident and in control of their lives. I am going to briefly outline the research, explain how I analysed the interview data and summarise the findings.

Overview of the research

Previous research had shown that many children and young people with medical conditions want to feel confident and in control of their lives, but often face barriers. Empowering people with medical conditions might mean they can stay happier and healthier. I wanted to know more about the empowerment in children and young people with CF.

I interviewed 7 young people with CF, 5 parents and 4 CF professionals to find out their views. I spent a long time going through the interviews in detail and comparing them to each other. I was able to work out some key themes from doing this.

What research show?

I found that there are things that make it easier for young people to ‘thrive alongside CF’ these were:

- Having a team
- Taking charge and having a voice

I also found that there were some barriers to ‘thrive alongside CF’ the main one was having to keep secrets or hide CF from others.

It was clear that all young people with CF are unique and things that make it easier or harder to thrive change over time.

Do you want more information about the study?

Please contact me via email: n.h.fairweather323@canterbury.ac.uk or leave a message for me on a 24-hour voicemail phone line at 01227 927070. **If you are under 16 please ask your parent/guardian to do this for you.**



Naomi Fairweather
Trainee Clinical Psychologist
Salomons Centre for Applied Psychology
Canterbury Christ Church University
1 Meadow Road
Tunbridge Wells
TN1 2YG



Participant ID:

Demographics

You do not need to answer any of these questions if you do not want to.

Gender

Female

Male

Other

Ethnicity

How do you define your ethnic identity? Tick all that apply

White (British, Irish, Traveller, European, other)

Black (British, African, Caribbean, other)

Asian (British, Indian, Pakistani, Bangladeshi, Chinese, other)

Mixed ethnicity

Other (please specify) _____

Disability

Do you consider yourself to be disabled? Tick all that apply

Yes – physical

Yes – mental

Yes – sensory

Yes – other

No

Participant ID:

Education

Are you currently in education? Tick all that apply

- Yes – at school
- Yes – educated otherwise
- Yes – at college
- Yes – at university
- No
- Full time
- Part time

Who do you live with?

- Relatives
- Friends
- Alone
- Other (please specify) _____

Staff

Profession

Professional experience with CF in years

IRAS Ref Number: 234289 Participant ID:

Health Data

Age of CF diagnosis, if known

Most Recent Lung Function (FEV₁, %) Score

Most Recent BMI

Pancreatic insufficiency

Yes

No

CF-related diabetes

Yes

No

PEG

Yes

No

Central venous catheter (Port or Hickman)

Yes

No

Chronic infections

Episodes of IV antibiotics in previous 12 months

Episodes of oral antibiotics in previous 12 months

On transplant list?

Yes

No

Parent/guardian

Relationship to child/young person

Current age of child/young person

Research Participants Needed

Empowerment of children & young people with cystic fibrosis during times of change

We know that many children and young people with CF want to feel confident and in control of their lives, but can face barriers. Empowering (giving power to) people with CF might mean they can stay happier and healthier. This project aims to find out more about the empowerment of children and young people with CF during times of change.



Who?

- Children & young people with CF aged 11-19 years
- Parents of eligible children & young people
- Clinicians with children & young people with CF

What?

Participation would involve taking part in a one-hour interview about CF and confidence during times of change like moving schools, getting more independent at home, making new friends and moving on to adult CF care.

Where?

At hospital, via phone or Skype.



For more information please speak to the CF clinical team

[Recruitment post/flyer Version 2.0 14.04.2018 IRAS
Ref Number: 234289]

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Manuscript Submission Guidelines: *Qualitative Health Research*

This Journal is a member of the [Committee on Publication Ethics](#)

This Journal recommends that authors follow the [Recommendations for the Conduct, Reporting, Editing, and Publication of Scholarly Work in Medical Journals](#) formulated by the International Committee of Medical Journal Editors (ICMJE).

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Only manuscripts of sufficient quality that meet the aims and scope of *Qualitative Health Research* will be reviewed.

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1. What do we publish?

1.1 Aims & Scope

Before submitting your manuscript to *Qualitative Health Research*, please ensure you have read the [Aims & Scope](#).

1.2 Article types

Each issue of *Qualitative Health Research* provides readers with a wealth of information —, commentaries on conceptual, theoretical, methodological and ethical issues pertaining to qualitative inquiry as well as articles covering research, theory and methods.

1.2.1 What types of articles will QHR accept?

QHR asks authors to make their own decision regarding the fit of their article to the journal. Do not send query letters regarding article fit.

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- Note the sections: General articles, critical reviews, articles addressing qualitative methods, commentaries on conceptual, theoretical, methodological, and ethical issues pertaining to qualitative inquiry.
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- *QHR* does NOT publish pilot studies.

Look Out for These Regular Special Features

Pearls, Pith and Provocation: This section fosters debate about significant issues, enhances communication of methodological advances and encourages the discussion of provocative ideas.

Mixed Methods: This section includes qualitatively-driven mixed-methods research, and qualitative contributions to quantitative research.

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Evidence of Practice: Theoretical or empirical articles addressing research integration and the translation of qualitatively derived insights into clinical decision-making and health service policy planning.

Ethics: Quandaries or issues that are particular to qualitative inquiry are discussed.

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1.3 Writing your paper

The SAGE Author Gateway has some general advice and on [how to get published](#), plus links to further resources.

1.3.1 Make your article discoverable

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2. Editorial policies

2.1 Peer review policy

Qualitative Health Research strongly endorses the value and importance of peer review in scholarly journals publishing. All papers submitted to the journal will be subject to comment and external review. All manuscripts are initially reviewed by the Editors and only those papers that meet the scientific and editorial standards of the journal, and fit within the aims and scope of the journal, will be sent for outside review.

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As part of the submission process you may provide the names of peers who could be called upon to review your manuscript. Recommended reviewers should be experts in their fields and should be able to provide an objective assessment of the manuscript. Please be aware of any conflicts of interest when recommending reviewers. Examples of conflicts of interest include (but are not limited to) the below:

- The reviewer should have no prior knowledge of your submission
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- Reviewer nominees from the same institution as any of the authors are not permitted

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The list of authors should include all those who can legitimately claim authorship. This is all those who:

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It is the policy of *Qualitative Health Research* to require a declaration of conflicting interests from all authors enabling a statement to be carried within the paginated pages of all published articles.

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2.6 Research ethics and patient consent

Medical research involving human subjects must be conducted according to the [World Medical Association Declaration of Helsinki](#)

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- All papers reporting animal and/or human studies **must state in the methods section that the relevant Ethics Committee or Institutional Review Board provided (or waived) approval**. Please ensure that you blinded the name and institution of the review committee until such time as your

article has been accepted. The Editor will request authors to replace the name and add the approval number once the article review has been completed

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Information on informed consent to report individual cases or case series should be included in the manuscript text. A statement is required regarding whether written informed consent for patient information and images to be published was provided by the patient(s) or a legally authorized representative. Please do not submit the patient's actual written informed consent with your article, as this in itself breaches the patient's confidentiality. The Journal requests that you confirm to us, in writing, that you have obtained written informed consent but the written consent itself should be held by the authors/investigators themselves, for example in a patient's hospital record.

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The relevant [EQUATOR Network](#) reporting guidelines should be followed depending on the type of study. For example, all randomized controlled trials submitted for publication should include a completed [CONSORT](#) flow chart as a cited figure and the completed CONSORT checklist should be uploaded with your submission as a supplementary file. Systematic reviews and meta-analyses should include the completed [PRISMA](#) flow chart as a cited figure and the completed PRISMA checklist should be uploaded with your submission as a supplementary file. The [EQUATOR wizard](#) can help you identify the appropriate guideline.

Other resources can be found at [NLM's Research Reporting Guidelines and Initiatives](#)

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3. Publishing Policies

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- Include figures, charts, and tables created in MS Word in the main text rather than at the end of the document.
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