

**Exploring the Experiences of Two Key Transitions in Cystic Fibrosis: Diagnosis and Transition to Adult Care**

A thesis submitted in partial fulfilment of the requirements for the Doctorate in Clinical Psychology

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# Declaration

This thesis has been submitted for the award of Doctorate in Clinical Psychology at the University of Sheffield. It has not been submitted to any other institution, or for the purpose of obtaining any other qualifications

# Structure and Word Counts

**Literature review**

1. Word count excluding references and tables- 7,998
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**Research report**

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# Lay Summary

**Literature Review:** Health transition is the term given to the process that young people with chronic health conditions experience when leaving children’s health services to enter adult health services. The process has been described as a concern in health, often due to how it is conducted. Therefore, a literature review was conducted to establish how young people and their parents have experienced health transition in Cystic Fibrosis (CF). A total of eight research papers were used for the review. The review identified that participants experienced significant changes in their mental health and personal development during the transition.

Additionally, the transition is experienced as a loss as young people say goodbye to a healthcare team they know very well. Parents also experience a loss as they become more aware of their children growing up and adopt a somewhat different parenting approach. Finally, participants were able to identify what helped the transition process, as the relationships they held with their family and healthcare systems at the time of transition were able to be helpful, providing a positive transition experience. Recommendations were made for how healthcare services could address the findings.

**Empirical Report:** There is guidance that suggests families should be offered support when their new-born baby is diagnosed with cystic fibrosis (CF). However, no research has attempted to capture parents’ experiences receiving the news and its implications for their family and contact with those around them. The study tried to address the lack of understanding in this area. Eight mothers who had received a diagnosis for their new-born baby within the six weeks following birth completed an interview with the lead researcher to describe their experiences. Interviews were analysed, which generated four findings. Participants described receiving the news as a cause of great distress and explained thatthe process, made them feel powerless. Participants felt parenthood was changed forever as their identity and expectations changed. Participants felt guilt they had caused the chronic condition in their children. Additionally, participants felt that the social world did not understand the implications of the CF for them. Finally, participants valued stories of hope, how helpful families are at the time of diagnosis and what they valued in communication with health professionals.

# Acknowledgements

My sincerest thanks goes to the eight parents who took the time to share their experiences of diagnosis with me. Thank you for trusting me to hear about such a personal and emotive time of your life. I hope I have represented your words and experiences accurately and honourably.

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# Section One: Literature Review

The experiences and impact of transition from child to adult healthcare services for young people with cystic fibrosis: a qualitative synthesis

## **Abstract**

**Objectives**

Healthcare transition has been described as a global health concern, often due to the method by which it is conducted. Equally, health transition occurs at a key time in a young persons’ life. This review aimed to investigate the experiences of healthcare transition from the perspectives of young people with cystic fibrosis (CF) and their parents. Previous literature illustrates that health transition has a psychological impact on parents, carers, and young people.

**Design and Method**

Three databases were searched for peer-reviewed published qualitative studies which described young people’s and their parents experience of the transition from paediatric to adult health services.

**Results**

Eight studies met the inclusion criteria and were analysed using thematic synthesis. Synthesis of the data revealed three superordinate themes; ‘*transition comes with a personal cost’*, ‘*nobody can escape the amount of change’,* and ‘*supportive systems are invaluable’.*

**Conclusions**

Participants felt that family and healthcare systems could often work well for them to support transition. Both young people and their parents felt a sense of an ending during transition. The experience of endings were attached to the end of parenthood for parents and for young people the end of relatable, individualised healthcare. Participants also felt a significant personal impact. Participants noticed a change in their mental health prior to the transition and finding a new voice relating to their care.

**Practitioner Points**

* Paediatric and adult healthcare teams with the support of cystic fibrosis (CF) clinical psychologists can support the transition of young people with CF by focusing on fostering positive relationships, developmentally appropriate care and seeing the young person as an individual.
* Parents mark the transition of their child as the end of parenthood and need support in their own transition to a newer role.
* Young people with CF may need support with their mental health as the forthcoming transition causes stress and anxiety. Psychological practitioners are well placed to provide to support this.
* Young people with CF notice finding a new voice to advocate for themselves. Healthcare services can amplify this to promote self-efficacy.

*Keywords*: healthcare transition, cystic fibrosis, young people, parents, qualitative thematic synthesis

# Abbreviations:

# CF – Cystic fibrosis

# CYP – Children and young people

# CASP - Critical Appraisal Skills Programme

## **Introduction**

Chronic health conditions are health problems lasting at least 3 months that require frequent hospital admissions, care at home or other forms of care (Mokkink et al., 2008). As the quality of healthcare provided in health settings for chronic health conditions has improved (Wijlaars et al., 2016), the number of children and young people (CYP) living into adulthood has increased (Nasir et al., 2018; Nelson et al., 2012). Due to these advances, more CYP than ever with chronic conditions are subsequently transitioning to adult healthcare providers when they reach early adulthood.

**Cystic Fibrosis**

It was expected that there would be a 75% increase in young people living with Cystic Fibrosis (CF) transitioning from paediatric to adult healthcare services across Europe in 2015-2025. CF is an inherited, chronic, and progressive condition caused by a defective gene.

Although CF is the most common genetic disease for White people, recent evidence shows that regions of the Middle East, Asia and Latin America, show the condition is present although at lower rates (Scotet et al., 2020; Shteinberg et al., 2021). As the latest prevalence estimates indicate that CF affects 162,428 people worldwide (Guo et al., 2022) remarkable healthcare advances have meant increased life expectancy for these people (Cystic Fibrosis Trust, 2023; Keough et al., 2018). The majority of CYP with CF are now anticipated to transfer their care to adulthood healthcare providers.

CF requires intensive healthcare support across the lifespan from healthcare professionals, family, educational settings, and third-sector organisations. Any disruption to the healthcare provided across these contexts during early childhood and adulthood has been found to have a detrimental impact on later physical and mental health (Dugueperoux, 2008; Quittner et al., 2014).

**Transition**

Unanimously within the literature, transition has been described as “the purposeful and planned movement of adolescents and young adults with chronic physical and medical conditions from child-centred to adult-oriented health-care systems… the optimum goal of transition is to provide health care that is uninterrupted, coordinated, developmentally appropriate, and psychosocially comprehensive” (Blum et al., 1993; p, 570).

Connett and Nagra (2018) suggests that transition should not be viewed as a transactional, single event but instead as an extensive preparatory process planned and expected by the individual transitioning (Sawyer et al., 1997). Typically, transition is assumed to be when the young person must contact and make decisions with healthcare teams more autonomously (Dovey-Pearce & Christie, 2013). For young people with CF, transition can often occur alongside expected deteriorating health associated with the condition, such as worsening lung function (Taylor-Robinson et al., 2018) and reduced body mass index (Marshall et al., 2017). This, coupled with an increase in depressive symptoms found in developing young adults (Ferro et al., 2015), makes the timing and process of transition complex. Alongside potential physical health changes are universal age-related developmental, physical, and social changes such as a change in education setting, living circumstances and employment. Such milestones may impact the ability to self-manage CF and to adhere to substantial treatment regimens (Foster et al., 2001; Tuchman & Schwartz, 2013) as CYP may not prioritise health as highly as other non-health related areas of transition in their lives (Ernst, 2010; Toulany et al., 2022).

Parents and caregivers should be viewed as an integral part of the transition process as they naturally become experts in their child’s care (Balling & McCubbin, 2001). Parents and caregivers are stakeholders in developing the emotional environment necessary to adapt to the difficulties related to young adulthood (Bronfenbrenner, 1979). Studies that have assessed the role of parents and caregivers during the transitional period have found they face novel challenges relating to their mental health, a shifting in the parental role and difficulties related to both their own and their child’s development (Duncan et al., 2014; Williams et al., 2007). Additionally, parents and caregivers are required to adjust their caring roles as the CYP is supported to be autonomous (Reed-Knight et al., 2014; Sasse et al., 2013).

Transition has been found to have significant impacts on CYP and their families and caregivers. Although there are positive findings related to transition programmes across several chronic conditions (Holmes-Walker et al., 2006; McDonagh et al., 2007; Van Walleghem et al., 2008), the healthcare challenges faced by the transition of care have been well documented, too. Across different health conditions such as diabetes, congenital heart defects and intellectual disability, poor transition experience has been found to cause disengagement with healthcare services (Bryden et al., 2003; Kipps et al., 2002), increases in hospitalisation (Nakhla & Daneman, 2012) poor treatment adherence (Annunziato et al., 2007; Watson, 2000) and harmful health outcomes overall (Viner, 2008; Clarke et al., 2011; Shogren & Plotner, 2012). Transition issues are therefore not unique to specific medical conditions or type of care provider.

It has been found that for parents and caregivers who have had a negative transition care experience for their child, subsequently noticed an increase in distress for the safety and well-being of their child (Tomette et al., 2020) and high levels of fear and anxiety (Cheak-Zamora et al., 2015). Heath et al. (2017) noted that for parents who are experiencing high amounts of distress, referral to psychological services may be appropriate.

Although there has been significant guidance and research on transition, there is still no one moment when a young person is ready for transition and no simple tool to identify it (Rosen, 1995).

**Clinical Implications**

As clinical psychologists are seen as key members by the multi-disciplinary teams (MDTs) they work in (McCullough et al., 2018), surveys have shown that their work in MDTs across Europe is highly variable (Abbot et al., 2015). For clinical psychologists who do work in these MDTs, the European CF Society Standards of Care (Conway et al., 2014) highlights that the team psychologist should, for example, assess the person living with diabetes annually, and that this should begin at the age of 12. Importantly, parents and caregivers should be screened annually for their mental health, too. Within the guidance, transition to adult services is seen as a “key transition point” (Castellani et al., 2018; p.170) which can create emotional instability. It is therefore important for clinical psychologists and for their wider MDT colleagues, to understand what the key aspects of transition are so they can offer specific support to young people and the parents and caregivers.

**Justification for Review**

The importance of a well-considered transition being defined as a global health issue (Kerr et al., 2020), a scoping review in 2015 (Hepburn et al., 2015) identified the transition strategies of 9 countries. At that time, only the UK and Australia were making demonstrable efforts to develop and implement transition strategies. More recently, the USA has made transition in healthcare a priority (Healthy People 2030, 2023) and the UK has published a strategy to create a Children and Young People’s Transformation Programme (NHS, 2019). The aim of the strategy is to selectively move towards a ‘0-25 years’ approach that will attempt to improve the quality and continuity of care. The strategy suggests that person-centred and age-appropriate care for physical health needs can improve outcomes, compared to arbitrary transition to adult services based on age not need (NHS, 2019).

Researchers have reported issues that arise throughout transitional care because of generic concerns (Begley, 2013; Sawyer et al., 2007). Although such issues are important, establishing an understanding of transition for young people with CF, based only on research on other genetic and chronic illnesses, risks missing the nuances associated with CF. Much of the guidance that has been published in health transition is disease specific and not evidence based (Pape & Ernst, 2022).

Firstly, research has shown CYP living with CF and their parents are concerned about infection control when attending their new place of care in adult treatment (Al-Yateem, 2012; Boyle, 2001). Secondly, how young people with CF access peer support differs from other chronic conditions. People living with CF have the additional challenge that infection and control guidelines recommend no direct contact with others diagnosed with CF to reduce the likelihood of cross-infection (Jeffrey et al., 2020; Saiman et al., 2014). Peer support within other chronic health conditions is associated with better feelings about transition (Zimmerman, 2022) and impacts feeling reassured, less isolated, and more confident (MacDonald et al., 2019). For young people with CF, there may be an enhanced reliance on the family and caregiver systems which may need to be further understood to maximise a successful transition.

For parents and caregivers who care for CYP with CF, they experience sadness related to the life-limiting characteristic of the condition (Moola & Norman, 2011). It is apparent that parents and caregivers continue to be aware of the reduced life expectancy but hesitate to express their distress and uncertainty with the healthcare team supporting them (Dupuis et al., 2011). Additionally, transition was marked as a moment indicating they are ever closer to losing their child, as transition is an unhelpful reminder of age (Dodgson et al., 2000). Whilst the early death of a child with a chronic condition is not specific to CF, the literature relating to other health transitions does not find that this is in parents’ and caregivers’ consciousness at the time of transition (Kelly et al., 2020; Ludvigsen et al., 2021; Rasalingham et al., 2021).

One existing systematic review (Coyne et al., 2017) aimed to examine the impact of transition on health outcomes and behaviours in CF. Additionally, it aimed to investigate the experiences of healthcare transition from the perspectives of young people, parents, and healthcare providers. It found that structured transition programmes appear to impact experiences positively, despite young people being concerned about leaving behind previous caregivers, experiencing differences in care provision and potential infection risks. A lack of preparation for young people was a consistent, damaging theme.

To the authors' knowledge, Coyne et al. (2017) is the only published systematic review on transition care in CF. However, the review does not use a formal qualitative methodology to synthesise qualitative data. The authors (Coyne et al., 2017) also discussed the need for higher-quality research in CF transition. An initial scoping search indicated that an additional review that includes papers published in the six years since the 2017 review (Coyne et al., 2017) could ultimately strengthen the evidence base (Carroll, 2017; Downe et al., 2019). This could be achieved through qualitative evidence synthesis, which can then be used to inform clinical practice (Carroll & Booth, 2015; Dixon-Woods et al., 2006; Flemming et al., 2019).

The aims of the current review were created following guidance published outlining best practice for transitioning young people to adult healthcare services (Care Quality Commission, 2022; Pape & Ernst, 2022). Additionally, NICE (2016) guidance highlighted recommended areas of research in health transition.

This systematic qualitative review aimed to investigate the experiences of healthcare transition from the perspectives of young people with CF and their parents. Secondly, the review wanted to investigate if transition had an impact on health treatments and what impact transition had on the mental health of young people with CF and their parents and carers.

## **Method**

The protocol for this review was registered on the international register PROSPERO (reference: CRD42022384682).

**Search Strategy**

A SPIDER tool was utilised to develop and refine the search strategy (appendix A; Cooke at al., 2012). The searches were completed in November 2022 across three databases: PsycINFO (via Ovid), Scopus and Medline. Search syntaxes were inputted into Scopus and Medline (table 1); MeSH terms were inputted to PsycINFO (appendix B). Searches focused on titles, abstracts, and keywords. As identifying qualitative literature for systematic reviews has been acknowledged as difficult (Shaw et al., 2004), forward and backward searching of the reference lists and citations of eligible papers were completed.

**Table 1.**

*Search syntaxes*

|  |  |
| --- | --- |
| **Construct** | **Search Term** |
| Health condition | “cystic fibrosis” |
| Population | child\* OR teenager\* OR adolescent\* OR caregiver\* OR parent\* OR adult\* |
| Transition of care | transition OR handoff OR handover OR transfer OR moving |
| Experience/ view | experience OR attitude OR “personal satisfaction\* OR perspective OR perception OR opinion OR understanding OR outcome |
| Qualitative Research | “qualitative methods” OR “thematic analysis” OR interview OR “semi structured interview” OR “focus group” OR ethnography OR “grounded theory” OR “narrative analysis” OR “content analysis” OR ”interpretative phenomenological analysis” OR IPA OR “mixed methods research” |

*Note.* individual search terms for each construct were combined with the Boolean operator ‘OR’, and broad constructs were combined with the Boolean operator ‘AND’.

**Study Selection**

Papers found across all databases were downloaded and imported to the Zotero reference management software programme and duplicates were removed. Papers were then reviewed by the author against the inclusion and exclusion criteria (Table 2). Remaining papers were then subject to a full-text review.

Studies were not included if the sample of participants within papers was made up of varying chronic conditions (i.e., papers that included participants with sickle cell disease, juvenile rheumatoid arthritis, and inflammatory bowel disease) as identifying quotes from patients solely with CF was problematic (Huang et al., 2011; Tuchman et al., 2008).

A second reviewer[[1]](#footnote-1) also screened a random 50% (n=9) of identified papers to ensure consistency and rigour when applying the inclusion criteria, which increased the reliability of the selection process. There were no disagreements relating to the selection of papers alongside inclusion and exclusion criteria.

**Table 2.**

*Inclusion and Exclusion Criteria*

|  |  |
| --- | --- |
| **Inclusion Criteria** | **Exclusion Criteria** |
| Primary empirical research studies exploring experiences relating to the transition of healthcare delivery from paediatric to adult services | Written in languages other than English |
| Experiences of people living with cystic fibrosis and their parents | Utilised only a quantitative research design or were systematic reviews |
| Used qualitative methodology | Papers or studies from the grey literature (e.g., conference proceedings, posters, unpublished theses or dissertations). |
| Published in a peer-reviewed journal | Studies which only explored experiences of professionals working in cystic fibrosis |

**Data Extraction**

The following data was extracted from included articles: author, date of publication, country of study, aims, participant characteristics (age, gender, ethnicity), qualitative data collection method, analysis method, summary of the main themes (Table 3).

**Researcher Reflexivity**

In keeping with quality standards for rigour in qualitative research (Barrett et al., 2020) the researcher considered their view and opinions on transitional care for young people with CF. This is particularly important as identified themes found within the study may have been influenced by those views and opinions.

The researcher identifies as a white British male who does not have a chronic health condition. The researcher has worked in physical health settings with young people and was witness to several transition clinics when working in paediatric diabetes. Upon beginning the research, the researcher believes that young people have ability and resources which have been built up over time (Burnham, 2012). Additionally, they believe that health systems can be overly concerned clinical procedures and processes, which does not consider the impact of developmental and social changes of young people experience.

To further enhance the ethical and methodological rigour (Smith, 1999), the researcher recorded reflective commentary throughout the analysis (appendix C**)**. This consolidated learning from the research process and the feelings that were noticed throughout this process (Thorpe, 2010).

**Data Synthesis**

Data was analysed by the researcher using the three-stage process of thematic synthesis described by Thomas & Harden (2008). The process was supported by NVivo software (QSR International, 2018) due to benefits related to transparency and efficiency in qualitative research (Hoover & Koerber, 2010). Firstly, relevant quotes and findings were coded inductively line by line from the ‘results’ or ‘findings’ sections of each study (appendix D). Secondly, “descriptive themes” were generated to capture the meaning of groups of initial codes. Descriptive themes were created by looking for similarities and differences between the codes to group them. Thirdly, “analytical themes” were then created (appendix E), which went ‘beyond themes’ (Thomas and Harden, 2008) from the primary studies. Thematic synthesis allowed an explicit and transparent process when working with findings of varied brevity, multiple quotes, and differing reporting styles.

**Quality Appraisal**

The Critical Appraisal Skills Programme (CASP, 2018) is a tool to evaluate and comment on the credibility of findings in qualitative research and is a commonly used tool in systematic qualitative reviews (Dixon-Woods et al., 2007). Appraisal of quality of papers is assessed using ten questions (appendix F). Appraisal of the studies was undertaken by the lead researcher and then again by an independent secondary reviewer[[2]](#footnote-2).Two disagreements were discussed and resolved. All papers meeting the inclusion criteria were included in the review, regardless of CASP outcome as there is currently no accepted method to do so in qualitative reviews (Carroll et al., 2012; Garside, 2014).

## **Results**

The PRISMA diagram (Figure 1) outlines the study search and selection process (Moher et al., 2009; Page et al., 2021). Following the removal of duplicates, database searches provided 58 papers to screen. Following title and abstract screening, 40 papers were removed as they did not meet the inclusion criteria. 18 papers were then subjected to a full-text review to determine relevance. Following this process, eight papers met inclusion criteria to be included in the final review. No papers were found through citation and forward and backward searching.

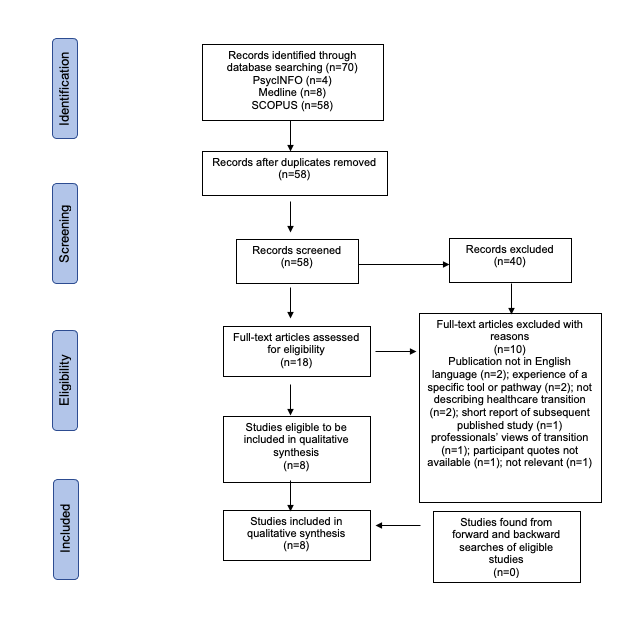
**Study characteristics**

Studies were carried out between 2003 and 2022. Six of the studies used qualitative methodology. Two studies (Coyne et al., 2018; Zack et al., 2003) used mixed methods and collected their quantitative data through self-administered questionnaires.

Within Zack et al., (2003) study the qualitative component of the mixed method design focused solely on young people’s experience of transition. Two papers described parent’s informational needs and role change at the time of and post transition. One paper described the organisational needs prior to transition. Four papers described exclusively the experience young people had throughout the transition process.

**Figure 1.**

*PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analyses; Moher et al., 2009) diagram of study selection process.*



**Table 3.**

*Table of characteristics*

|  |  |  |  |  |
| --- | --- | --- | --- | --- |
| **Author(s), Date, Country** | **Aims** | **Participants’ Characteristics** | **Qualitative Data Collection and Analysis Method** | **Main Themes** |
| Al-Yateem (2012).  Ireland | The transition of young adults with CF moving from child to adult healthcare settings, to identify patients’ needs and the factors that facilitate or hinder this transition | N=15 (no information on gender or age range) | In-depth interviews, data analysis process | Preparing for transition, amorphous service |
| Brumfield & Lansbury (2004).  Australia | Examine and investigate the experiences of adolescents  as they made the transition from paediatric to adult care, the factors contributing to these experiences, and the potential outcomes of these experiences. | N=6 (male, 3; female 3; age range 19-34). | Focused in-depth interviews, thematic analysis | Paediatric care, elements of the transition programme and psychosocial factors |
| Coyne et al. , (2018)  Ireland | Identify parents’ informational needs on the transition process and examine the extent to which these needs are being met by healthcare professionals | N=59 parents (female, 42; male, 17; age range 22-62). N=59 young people with CF (female, 23; male, 36). | Questionnaire, open ended questions, inductive analysis using a coding frame | Positive and negative feelings towards transition discussed |
| Iles & Lowton (2010)  (UK) | To explore the nature of parental support that is perceived to be available at the time of transition | N=50 young people with cystic fibrosis, although focus of the paper was on N=32 (female 17; male, 15) who had experienced transition or accessing adult care. | Semi-structured interviews, thematic analysis | Providing non-clinical practical and emotional support; acting as ‘troubleshooters’ in times of health-related crisis; working in partnership with offspring in ongoing disease management in the home and clinic; Acting as ‘protectors’ of their children |
| South et al., (2022)  (USA) | To explore the perspectives  of a diverse sample of AYAs with CF at one urban academic medical center regarding healthcare  transition | N=12 (female, 3; male 9; age 15-24).  N=3 Black; N=1 Native Hawaiian/Pacific Islander; N=2 Decline to state; N=4 Hispanic/Latina/o Ethnicity; N=6 White[[3]](#footnote-3). | Semi-structured interviews, qualitative descriptive methodology | Independent Care of the Whole Self, preparing for Change and the Unknown, transition Experiences Vary |
| Tierney et al., 2013  (UK) | To explore young people’s experience of transferring. | N=19 (female, 7; male 12; age 17-19). | Semi-structured interviews, systematic thematic analysis “framework” approach | Fracturing, acclimatizing, integrating |
| Vion Genovese et al., (2021)  (France) | To identify the organizational needs both of patients and of parents before the transfer to an adult CF center | N=43 young people with CF (age 16-19). N=41 parents[[4]](#footnote-4). | Semi-structured interviews | Anticipate, accompany, announcement |
| Zack et al., (2003)  (USA) | To investigate the perspectives of adolescents and adults with CF who receive care at a children’s hospital regarding preventive health counselling and transition issues. | N=32 (female, 19; male, 13; age range (16-43).  N=31 White, non-Hispanic; N=1 American Indian | Structured interview, coding techniques | Changes in care experienced with age, best features of care at this hospital, Reasons for maintaining care at this hospital, suggestions for improvement, guidelines for hospital staff |

**Quality Appraisal Results**

A comprehensive review of the quality appraisal is included (appendix G). Critical appraisal suggested that most studies were of satisfactory quality. However, six of the studies failed to fully acknowledge the relationship between the researchers and study participants (CASP qualitative checklist criterion 6). Four papers omitted information relating to rigorous data analysis (criterion 8) and two papers did not report a clear statement of their findings (criterion 9). The other criteria of the CASP checklist were completed by most of the eight studies. Two studies reported all criterion to an adequate standard. All studies fulfilled 7> of the CASP criteria.

**Thematic Synthesis**

The thematic synthesis identified 3 main themes and 6 subthemes (Table 4). Each theme is complimented with a participant quote from the primary research papers to broadly represent the theme. The themes are illustrated with multiple participant quotes (appendix H).

**Table 4.**

*Themes and subthemes*

|  |  |
| --- | --- |
| **Themes** | **Subthemes** |
| Transition Comes With A Personal Cost | Mental Health Impact |
|  | Change of Self |
| Transition Means Everything is Changing | The End of Parenthood?  The End of Stable, Relatable Healthcare |
|  |  |
| Supportive Systems Are Invaluable | Family as Resource |
|  | Equipped Health Services |

***Transition comes with a personal cost***

This theme covers participant’s personal impacts of transition. The personal impact participants felt was experiencing changes in mental health both pre and post transition alongside a significant awareness of personal changes.

**Mental health impact.** Participants acknowledged that the transition process had an impact on their mental health (Brumfield & Lansbury, 2004; Al-Yateem, 2012; Tierney et al., 2013; Coyne et al., 2018; Vion Genovese et al., 2021; South et al., 2022). The resounding feeling participants noticed was “anxiety” as the change of team was approaching (Al-Yateem, 2012; Tierney et al., 2013; Coyne et al., 2018; Vion Genovese et al., 2021; South et al., 2022). Participants struggled to imagine how they would express themselves, how they would be viewed from first impressions and having an inability to imagine the future healthcare. Participants also thought about the wider funding in health relating to health budgets.

*“So he’s known me for a long time. That’s why I was worried about moving here.. he knew a lot of the problems” (Tierney et al., 2013; p. 743).*

Other participants discussed how their confidence would be affected (Iles & Lowton, 2010) and how “on edge” they had felt about the move, meaning the transition was a source of discomfort (Brumfield & Lansbury, 2004). Most of all, participants felt transition had had a negative impact on their wellbeing.

*“It’s very stressful, when we tell you that you’re going to go to an adult center. . . we don’t have a point of reference” (Vion Genovese et al., 2021; p.262).*

**Change of self.** Participants described noticing a profound change that occurs in parallel during the transition. Alongside healthcare transition, there was an ongoing personal transition relating to life changes such as entrance to further education or the workplace. Such changes, on occasions, were met with contention as healthcare transition went ahead alongside important life transition (Tierney et al., 2013; Coyne et al., 2018; Vion Genovese et al., 2021).

“*Allowances should be made for a child to stay in paediatric care until he finishes his leaving certificate*” (Coyne et al., 2018; p. 651).

Participants noticed their autonomy towards treatment changed as they found a “new voice” meaning they could advocate for themselves (Tierney et al., 2013). Furthermore, participants noted a difference in who had responsibility for their care which was ultimately them as young adults (Tierney et al., 2013; South et al., 2022). This was seen as a marker for entering the adult world (Vion Genovese et al., 2021).

*“I’m not really one of those that will say ‘‘I don’t like this, I don’t like that.’’ So it’s more that that’s been my problem, getting used to me having to say ‘‘I don’t really agree with that’’ (Tierney et al., 2013; p. 273)*

***Nobody can escape the amount of change***

This theme comprises of participants’ experience of loss during the transition process whereby there were significant endings of relationships with the health service and within the family. Such experiences have been depicted as endings.

**The end of parenthood?** Parents of young people transitioning to adult health services depicted a sudden change. The nature of loss varied from losing the up-to-date information about their child’s health (Iles & Lowton, 2010; Tierney et al., 2013; Coyne et al., 2018; Vion Genovese et al., 2021; South et al., 2022) to not being present, or on occasions, invited to appointments (Tierney et al., 2013; Coyne et al., 2018; Vion Genovese et al., 2021; South et al., 2022). Consequently, such endings meant losing pre-defined parent-child roles which needed to be re-negotiated in the family (Tierney et al., 2013; Vion Genovese et al., 2021 South et al., 2022).

*‘‘Where will our place be [as parents]?” (*Vion *Genovese et al., 2021; p. 262).*

Adolescents described actively re-aligning the child-parent relationship themselves, as, when support was not required, they asked their parents to take a backwards step in a sense to gain control back (Iles & Lowton, 2010; VionGenovese et al., 2021; South et al., 2022). This was particularly difficult for parents who had described the previous held long-term positive relationships with paediatric CF services (Iles & Lowton, 2010; Vion Genovese et al., 2021; South et al., 2022).

*“Mum always used to sit in on consultations until, until I could get rid of her about three years ago”. (Iles & Lowton, 2010; p. 27).*

Some parents understood that they had to take a backwards step to encourage their young adults to have autonomy (Coyne et al., 2018; VionGenovese et al., 2021). However, this was undoubtedly difficult to do.

*It’s always a step towards the unknown. . . sick child or not, at some time, you have to let them go (Vion Genovese et al., 2021; p. 261).*

**The end of stable, relatable healthcare.** Most participants described strong relationships with the paediatric teams they had received care from prior to transition. This appeared in explicit descriptions of how the paediatric team had connected with the young person across their life (Zack et al., 2003; Brumfield & Lansbury, 2004; Tierney et al., 2013; Coyne et al., 2018; VionGenovese et al., 2021; South et al., 2022). Participants shared that the care received was akin to care you would receive as a member of someone’s family (Zack et al., 2003), based upon a sense of a deep bond of trust (VionGenovese et al., 2021) and that doctors were aware of you as an individual, so much so, that they had abilities to understand you on a deeper level (Brumfield & Lansbury, 2004)

*“Nurses that treat you like you are part of their family, they sit down and talk to you, and they are all concerned about whatever ailment or problem you may have” (Zack et al., 2003; p.380).*

As transitioned occurred, participants consistently found members of healthcare staff they felt unable to connect with. Participants described the new input as “harsh” (Zack et al., 2003; p. 380), as being “separated” from a time of feeling “attached” (Vion Genevose et al., 2021; p. 260) and that new healthcare staff were not deemed trustworthy (Al-Yateem, 2012) and “stand-offish” (Brumfield & Lansbury, 2004; p. 228). Receiving care from the adult care team was, at least initially, a negative experience.

*“The paediatric service was excellent in every aspect of the transition; however, the adult health service was a nightmare” (Coyne, et al., 2018).*

Ultimately, the loss of connection made accessing appointments in the adult services they had transitioned to, difficult. Many participants felt they would no longer be seen as an individual following transition (Zack et al., 2003; Brumfield & Lansbury 2004; Iles & Lowton, 2010; Al-Yateem, 2012; Tierney et al., 2013; Coyne et al., 2018; Vion Genovese et al., 2021; South et al., 2022).

*“I don’t . . . I don’t really trust them, because of the way they treat you, because you are a number” (Brumfield & Lansbury 2004; p. 232)*

As the relational and personal aspects of appointments were felt negatively, participants criticised the treatment they experienced in adult CF services as there was a notable difference from the care they received in paediatric services. Participants felt that the care did not meet their needs (Zack et al., 2003; Al-Yateem, 2012; Coyne et al., 2018; Vion Genovese et al., 2021) and felt that once healthcare services did not meet their needs, this was when disengagement began to occur (Zack et al., 2003; Brumfield & Lansbury, 2004; Al-Yateem, 2012).

“*I was quite compliant to start off with, you know, I was there every three months, then it got less and less to the point whereas if I walked in the door they’d all start clapping… and it was generally after they’d stopped writing me scripts over the phone” (Brumfield & Lansbury, 2004; p. 232)*

***Supportive systems are invaluable***

This theme encompasses the relationship participants had with their family and the healthcare systems around them during the time of transition. Participants felt these available systems and the relationships within them, worked to provide a positive transition experience.

**Family as a resource.** There was a persistent view that parental support during the transition process was sought and appeared “uncontentious, changing relatively little during and after young people’s transition” (Iles & Lowton, 2010; p. 24). Participants saw parental input in their healthcare as a constant throughout their lives, “someone [parents] were always there for me” (Vion Genovese et al., 2021; p.261) and that eliciting support was not age dependent (Tierney et al., 2013; South et al., 2020). When parental support was present, most studies reported it was a positive to have it (Brumfield & Lansbury 2004; Iles & Lowton, 2010; Tierney et al., 2013; Vion Genovese et al., 2021; South et al., 2022).

*“me mum was there and I’m 19 but because me mum was there I was more conﬁdent in asking questions because I knew if I’d said something that had come out a funny way or the wrong way, mum would go well what she actually means is this” (Tierney et al., 2013; p.743).*

Participants described accessing their parental support for ways that were personal to them. Support was linked to what was needed at that time as participants spoke about their parents cooking to improve diet when returning home (Iles & Lowton, 2010), attending appointments to support them (Tierney et al., 2013) and by sharing information parents had researched information independently (Al-Yateem 2012). Within the papers, when parents’ resources were utilised, it was a personal transaction between the young person living with CF and their parent.

*“because I’m home, so mum cooks properly, it’s not me using tins at three in the morning when I get in!” (Iles & Lowton, 2010; p24).*

Participants commented that following transition parents still handled the more complex parts of treatment. Regimes, such as ordering medications, implementing their use and support with emergency care were seen as aspects of treatment parents were still required to support (Iles & Lowton, 2010; Tierney et al., 2013; Vion Genovese et al., 2021; South et al., 2022).

*“For all medical prescriptions and medication management, it’s my parents!” (*Vion *Genovese et al., 2021; p.261)*

Some participants described their transition as starting off with their parents’ support, prior to the healthcare system initiating it (Iles & Lowton, 2010; Al-Yateem, 2012; Vion Genovese et al., 2021; South et al., 2022).

*“It started out, when I was like, I think I was about 13 or 14, my mom would sit next to me, and I would call in my prescriptions to the local pharmacy” (South et al., 2022; p.119).*

**Equipped health services.** This subtheme accounted for participants’ descriptions of a health system that was equipped to support individuals with the transition to adult care services.

One method that participants spoke of was being made aware who would be delivering their care (Brumfield & Lansbury 2004**;** Al-Yateem, 2012;Coyne et al., 2018**;** VionGenovese et al., 2021). The impact of such methods made participants feel positive about the forthcoming transition (*Al-Yateem, 2012;* Brumfield & Lansbury, 2004; South et al., 2022).

*“They are going to take us over to the other hospitals, and talk to the other staﬀ there, that would help” (Al-Yateem, 2012; p. 852).*

Some participants described how age-related care reflected how well the system was equipped to talk to young adults who were either still part of paediatric care or had just left it (Brumfield & Lansbury, 2004; Iles & Lowton, 2010; Tierney et al., 2013; South et al., 2022). Participants shared that the content of discussions, such as re-productive health and relationships were now on the agenda. Prior to this, conversations of this nature were either ignored in paediatric services, or on occasions, not discussed due to parents being present in consultations (Iles & Lowton, 2010; Tierney et al., 2013; South et al., 2022)

Participants stated that how healthcare teams spoke to them, meant they reiterated you were now in an adult healthcare environment which resulted in feeling “very comfortable” (South et al., 2022; p. 121) and “good” (Tierney et al., 2013; p. 743). There was a clear distinction of developmentally and age-related appropriate care both prior and after the transition experience being felt positively.

*“As I grew up, he (paediatric doctors) sort of treated me… as if I was older… he didn’t start treating me like a little kid and stuff, so that was good. (Brumfield & Lansbury, 2004; p 227)”*

## **Discussion**

This objective of the systematic review was to review the qualitative evidence describing the experience of transition for young people and their parents. Three themes were identified; *transition comes with a personal cost; nobody can escape the amount of change, supportive systems are invaluable.*

**Transition comes with a personal cost**

Previous research (Brown et al., 2019) has shown that participants felt transition had a negative impact on their mental health describing anxiety as the change was due to occur. Many participants felt that their worry was future focused relating to how they would share information and how they would be perceived.

Experiences of distress related to transition have been found in other studies identifying transition under a common theme of ‘going into the unknown’ (Kirk, 2008; Stewart et al., 2001). Young people with CF were explicit in discussing their anxiety about the transition, which was different to other qualitative literature on transitions which highlighted parents’ anxiety and young peoples’ “wait-and-see attitude” (Anthony et al., 2009; Moons et al., 2009). There is potential that such needs may be specific to the CF population due to previously mentioned specific transition considerations young people with CF have. For young people with CF transitioning, it may be important for healthcare professionals to acknowledge, assess and treat any emerging mental health concerns to increase transition success. Additionally, healthcare professionals need to be aware of personal change such as employment, education and living circumstances at the time of transition.

Participants noticed that the time of transition coincided with a significant change relating to self-efficacy and finding their voice as a user of a healthcare service. Transition may be a time when there has been a significant amount of mastery experiences (Bandura, 2008) coupled with developmental changes related to ongoing social modelling from parents (Bandura, 2012). Such basic factors may combine at the same time meaning young people feel in a position to take ownership over their healthcare (Zimmerman & Schunk, 2003).

**Nobody can escape the amount of change**

Participants who were parents described the transition process as the end of parenthood in the broader sense. Parents described the loss of up-to-date information, appointment attendance and re-defining child-parent roles enhanced the feeling that their role as a parent had ultimately ended, aligning with previous research (Betz et al., 2015; Ellison et al., 2022). Some parents described how they were aware they had to take a backwards step from the care of their child and reduce parental responsibility. Parents were discussing their own personal transition, which is experienced across a range of transitional healthcare experiences (Heath et al., 2017). Role theory (Eagly et al., 2000) may offer to explain the position of family roles and expectations and behaviours throughout the CF transition. Within role theory, *role negotiation* (Turner, 1956; Graen and Scandura, 1987) emphasises how any change in role, whether this is from a ‘parent carer’ role to solely just ‘parent’, may need to be negotiated between all members who are in contact with the ‘role’. Role theory explains how roles are taken “off” and “on” as and when the context around an individual permits them to be utilised. This is more apparent for parents who support their children with chronic health conditions. For example, the ‘parent carer’ role is switched “on” for the majority of the young person’s first 16 to 18 years of life. Transition may be a time when parents may struggle to “take off” the carer role which is more likely to happen when an individual invests a great deal of time and sacrifice in the acquisition of a role (Turner, 2001).

For parents supporting young people with CF, it is important for healthcare staff to understand parent’s role expectations and previous demands alongside the forthcoming transition. Although the included studies did not identify that parents were impacting on transition progress which has been identified in previous literature (Bashore & Bender, 2016) the importance of supporting families may be a useful area to focus on, as it has been found that parents would like support in redefining their role (Marsh et al., 2011; Morsa et al., 2020).

Participants described the end of personal and relatable healthcare, noticing that strongly built relationships that had been long held with paediatric teams were coming to an end. Participants in this review discussed how newer relationships with the adult teams were different and less person-centred. This replicates previous research in gastroenterology which identified two clear distinct cultures between paediatric and adult health services (Pinzon et al., 2004) and that a key challenge is bridging this gap to improve outcomes (Anthony et al. 2009; Valenzuela et al. 2011).

Participants criticised the adult care teams they were receiving care from, highlighting the differences in relationships that were once present, sharing they were less personal in nature which impacted on the care and chances of attending health appointments independently. At the time of transition, young people are experiencing a developmental transition, too (Snowman & McCown, 2014). Socially, at the time of transition there is a need to belong to a group that provides support and caring relationships. The same may be said for healthcare relationships. Participants were unanimous in noticing how they feared they would no longer be recognised as an individual (Together for Shorter Lives, 2015). Within the literature, however, it is understood that positive relationships are protective factors for distress (Chen & Harris, 2019) which may be utilised in transition to repair ruptures of nature.

Strengths based interventions may be utilised in consultations with young people living with CF as amplifying strengths and resources that the young person, and their family hold, can create positive outcomes and increased self-management (King et al., 2010; Warren-Findlow et al., 2012). Additionally, this can equally allow the young person to be seen and recognised as an individual which is an important factor is to building relationships (Etz et al., 2019).

**Supportive systems are invaluable**

Participants felt that the family and health system could be helpful in managing the transition process. Participants spoke of personal experiences of using their family as a resource to assist with the forthcoming transition. Participants spoke of how the health services were able to facilitate this too through age related care.

The importance of using the family as a resource to provide substantial support in health transition resonated with previous research (Allen et al., 2018; Lowton, 2002; McGuffie et al., 2008). The findings reinforce that young people have a reliance on the support of their family (Lowton & Gabe, 2003) which not only supported the transition process, but the care participants felt throughout and afterwards. There is an understanding that support for the young person only reduces moderately as they reach adulthood (McGuffie et al., 2008) which was detailed in the findings of the current review. The results of the current review confirm that parents are largely, the key facilitators to the process of transition (Colver et al., 2018; Heath et al., 2017).

Guidance published in the UK related to health transition (NICE, 2016) offers direction to increase peer support, to increase independence and maximise transition outcomes. It is known that accessing peer support is difficult given the risk of infection for people living with CF when accessing support of this nature (Al-Yateem, 2012; Boyle, 2001), which might explain why the dominant description of the support that the family offers was so central to young people’s experience. Considering the systemic familial factors when transitioning to adult services (Bronfenbrenner, 1992) may be fundamental as there is an abundance of resources in the mesosystem i.e., family and a lack of support due to infection risks in the exosystem i.e., the peer support community. Identifying needs within these areas may be fundamental in supporting transition.

Participants and their parents felt that when the health service was equipped to care for them, this helped the transition experience. Good care was experienced when clinicians in the health system were aware transition was imminent and an appropriate amount of detailed information was shared about it. This supported previous research across healthcare transition that knowing who will be providing care, where it will take place and who will be providing it, supported good care (Reiss et al., 2005; Shaw et al., 2004). When such information was shared with people living with CF, positive affective attitude was present. As transition is occurring at a time which has been found to be an anxiety provoking experience (Young et al., 2009) such information may enhance young people’s ability to cope with the forthcoming change.

Conversations that were developmentally and socially appropriate, made participants living with CF feel they were within an equipped health system that was talking to them about the right things at the right time, which replicated broader literature (Farre et al., 2016). Participants spoke about experiencing age-appropriate care which permitted reproduction and relationship conversations when in their appointments (Azh et al., 2017; Austrian et al., 2020).

**Critique of Included Studies**

Out of the eight studies included in this review, only two studies commented on the relationship between the participants and researchers. They did not discuss the ways in which biases could have existed between participants and researchers. Ignoring the researcher’s position in qualitative inquiry “is to discount a major component of the research process” (Rae & Green, p 1543). Researcher positionality, particularly when completing research with children, is important due to the potential for power dynamics which, is perhaps, why the lack of reflexivity is more noticeable within these studies. Additionally, although all eight papers documented ethical approval and/or considerations within their study methodology, not including a statement on positionality could be seen as an ethical failure (Morrow & Richards, 1996). It is a weakness that six studies in this review did not acknowledge this and their findings should be treated with caution. Future research in health transition would benefit from the completion of reflexive statements.

Four out of the eight studies did not provide rigorous information relating to how the qualitative data was analysed. Two papers within the review did not detail the steps taken to analyse the data, describing “phrases with similar meaning were grouped into themes” and “results classified by topic”. Indeed, one paper spoke of inductive thematic analysis but not how or by whom in the research team. One paper omitted detailing the analysis altogether. It is acknowledged that findings may be difficult to transfer from one setting to another. Although detailing the aims, methods, participant characteristics and settings of original papers was included by all papers, omitting how data was analysed and by whom means findings should be viewed cautiously.

**Limitations of the Review and Future Directions**

Several limitations of this review should be considered. All the primary papers included in this review have been given equal weight, despite the data provided by each studying varying in breadth and quality. Specifically, Zack et al. (2003) reported limited participant quotes, compared to Vion Genovese et al. (2021) who reported an abundance of participant data to illustrate themes. Therefore, some primary papers may feature more heavily than others which in turn may skew the findings. It is important to note that rigorous descriptions of data analysis in the primary papers were often associated with detailed quotes from participants (South et al., 2022; Tierney et al., 2013; Vion Genovese et al., 2021).

One of the research aims was to understand the experience of parents of children who are experiencing or have experienced the transition to adult services. Only two papers included in the review sought to collect data from parents (Coyne et al., 2018; Vion Genovese et al., 2021). Within the two papers, the total number of parents was limited (n=100) compared to young people who featured in all the primary papers (n=218). The gender of parents was also not reported in one paper (Vion Genovese et al., 2021). As Coyne et al. (2018) reported parents’ gender (n=17 male; n=42 female) the results found in the review relating to parent’s experience are from a small sample, in which male parents may be underrepresented. Although it is not unusual for fathers to be underrepresented in child and family development (Panter-Brick et al., 2014) or health research (Garfield & Isacco, 2012), future research could specifically enquire about fathers’ experiences, particularly as previous research (Balling & McCubbin, 2001), including the findings of this review, has found the importance of wider family input.

As CF does not exclusively affect White people (Farrel, 2008) and has been found to affect Middle Eastern, Asian and Latin American populations (Scotet et al., 2020; Shteinberg et al., 2021), a major criticism resulting from this review is that the primary research studies report solely on Western health systems. Only two of the primary papers report patient ethnicity demographics. Therefore, it is difficult to generalise any findings to other health systems and cultures, and to address the specific needs of ethnic minorities (Oakley et al., 2003). It is widely known that minority ethnic groups are under-represented in health research (Redwood & Gill, 2013). Future research may attempt to understand the needs of young people with CF and their family who are from ethnic minorities, specifically focused on the transition process to inform future practice. Additionally, including studies not in the English language, which this review did not do (Becher et al., 2014), may capture differing cultural experiences of transition in future research.

One limitation is that no review can capture every feeling and emotion expressed across the CYP and their parents transitioning. The main researcher did attempt to fully capture feelings and emotions across the primary papers by second checking identified themes with a secondary researcher. However, the primary coding of papers was completed solely by the lead author of the review. Furthermore, there was limited information from the primary studies related to treatment adherence and health outcomes following transition, hence two aims of the review could not be examined in full.

Post-hoc sensitivity analysis could have been used to strengthen the quality of results (Carroll & Booth, 2015). Grey literature was not included in the review which may have excluded several suitable studies which may be useful. One positive of the review is that secondary reviewers were utilised to apply the inclusion criteria to selected papers and to appraise primary papers using a recognised and endorsed tool (CASP, 2018). Additionally, an enhancing transparency in reporting the synthesis of qualitative research statement (ENTREQ Tong et al., 2012) checklist was completed by independent researcher[[5]](#footnote-5) which found the study met all 21 items for reporting the most common stages of a qualitative synthesis (appendix I).

A further positive of the study is that the author acknowledges their position relative to the participant group, as reflexive comments are published within the research, as are transcripts from the reflective diary (appendix C).

**Clinical Implications**

Healthcare services that support young people with CF through transition can make realistic and tangible changes to their practice that can support the transition process. Within CF team guidance, clinical psychologists are key members of the MDT (Conway et al., 2014). This review highlights that CF teams are the key stakeholders in building positive relationships and can facilitate this with the young people they care for in several ways.

As there is broad support across the literature for young people to be made aware of where post transitional care is going to be and the impact this has on transitioning (Reiss et al., 2005; Shaw et al., 2004), acknowledging and facilitating clinic time that occurs across the two sites of care is a useful foundation in building towards a successful transition. Psychologists in CF teams may want to facilitate changes in transition pathways, particularly as they are skilled in containment, grounding and validating varying levels of distress which are related to health transition (Gray et al., 2015).

There is a substantial challenge in how best to initiate and increase the connection between the transitioning young person and the busy health system. Such a question may require additional exploration beyond the scope of this review. However, consultations relating to future goals and *who* the person transitioning *is* may be useful to further increase the likelihood of reciprocal positive relationships. Indeed, health passports (Wolfstadt et al., 2011) have been found to be useful in communicating needs. However, young people seldom take them to health appointments (Colver et al., 2019)**.**

The traditions associated within solution focused therapy (SFT) (de Shazer, 1985) and its relationship with strengths-based questions may be a viable area to explore. It has been found that healthcare professionals can enhance their consultations by utilising SFT (Blayney et al., 2014; Corcoran & Pillai, 2009) and can learn SFT techniques to a good standard (Medina et al., 2022; Seko et al., 2020). As the essence of SFT is around building resources, such techniques may also be beneficial in amplifying the families’ available resources, too. Informing and guiding health teams to be aware of what the family already does to facilitate their child with CF is important and useful (Barker et al., 2012). Families need to be involved in the therapeutic process to building the relationship with the young person and team, too (Wright & Leahey, 2005) and every opportunity to harness these the benefits of these special relationships needs to be facilitated.

This review highlights that parents and caregivers need support as they experience their own transition. The current review identified that navigating this, albeit expected transition, is difficult (Horn et al., 2013). Acceptance and commitment therapy (Hayes et al., 1999) may be a suitable set of techniques that could offer parents tools to navigate their own transition. Psychologists in CF teams may be well placed to co-facilitate parental support groups within the ACT framework as this has been found helpful in healthcare transition (Kingsnorth et al., 2011).

## **Conclusion**

The results from this synthesis highlight CYP and their parents’ experiences of transition. Participants felt that family and healthcare systems often worked well for them to support transition.

Both young people and their parents felt a sense of endings during transition. The experience of endings were attached to the end of parenthood for parents and for young people the end of relatable, individualised healthcare. Participants also felt a significant personal impact. Participants noticed their mental health prior to the transition changed but also post transition, finding a new voice relating to their care was present.

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## **Appendix A:** SPIDER tool (Cooke et al., 2012).

|  |  |
| --- | --- |
| **SPIDER** |  |
| Sample | Children/adolescents/parents and caregivers experiencing the transition from child to adult services in cystic fibrosis |
| Phenomenon of interest | Studies related to the experience of the transition |
| Design | Gathering qualitative data: interviews, focus groups |
| Evaluation | Studies related to the lived experiences, satisfaction and perceptions of children and their parents of the transition pathway/process |
| Research type | Qualitative or mixed methods |

## **Appendix B**: *MeSH terms*

|  |  |
| --- | --- |
| **Construct** | **MeSH Term** |
| Health condition | exp Cystic fibrosis/ |
| Population | exp Adult/ or Child/ or Parents/ or child\*.mp. or Adolescent/; teenager.mp or adolescent/; exp caregivers/; exp parent/ or parents.mp. |
| Transition of care | transition.mp. or exp patient handoff/ or exp patient transfer/ or transition to adult care/ or transitional care/ |
| Experiences/ view | experience.mp.; attitude.mp. or exp personal satisfaction/ or perspective.mp. or perception.mp. opinion.mp. or understanding.mp. or outcome.mp. |
| Qualitative research | Qualitative Research/ or qualitative method.mp.; exp thematic analysis.mp.; exp interview/ or exp semi-structured interview.mp. or exp focus groups/ or exp grounded theory/ or exp narrative analysis.mp. or exp content analysis.mp. Interpretative Phenomenological Analysis.mp. or IPA.mp. or qualitative research/ mixed methods.mp. |

## **Appendix C:** Examples of reflective notes

1. I think for the first time I am appreciating the parental aspect of the transition. I have worked in supporting young people through transition before, but only young people and I remember their parents staying outside of the room as they had their last clinical psychology session(s). Reading the parental experience and noticing the phrase “where will our place be [as parents]” resonated with me. Unknown. Anguish attached to it? I notice I have thought about my own two children at this point when reading the paper (Vion Genovese et al., 2021) and realise that there might not always be a time when I’m needed. I feel sad and watch out for this interpretation when re-reading the paper.
2. Image of a healthcare team clapping someone into a consultation because they turned up. Am missing the context to this? It is well mannered. Humour can go a long way. My interpretation from the quote – it isn’t. Thoughts are to be critical staff when this might not be the case.
3. Teenagers are being moved from college to uni and from healthcare teams. Thinking of Dr H (GP) who I used to see as a child and always seemed to have time for me during routine illness as a child. Connection. Availability. Knew me and my Mum and Dad, brother. Remember moving house and having a new GP. Didn’t like them when I went for acne medication. Imagine this feeling – but only 1,000 times worse.
4. Enjoy seeing young people talk about their health teams in such positive ways. Reminder I can make a connection that can be useful for peoples’ live
5. Disheartened. CHANGING health systems is clearly hard. This might be changing two health systems to be helpful… if transition was to be better. Think of the family system as well. How do you start? Where do you start? Reminded of a quote from a book I’m reading “when there’s no place for the scalpel, words are the surgeon’s only tool”. Think of other medical professionals changing their words and become hopeful. It can happen.

## **Appendix D:** Descriptive Themes and Code Development

Graphical user interface, text, application

Description automatically generated

Graphical user interface, text

Description automatically generated

Table

Description automatically generated

## **Appendix E:** Descriptive Themes into *Analytical Theme Development*

Graphical user interface, application

Description automatically generated

## **Appendix F:** CASP Appraisal Tool

*Graphical user interface, text, application, email

Description automatically generated*

Graphical user interface

Description automatically generated with low confidence

A picture containing text

Description automatically generated

A picture containing text

Description automatically generated

A picture containing graphical user interface

Description automatically generated

Graphical user interface, text, application

Description automatically generated

## **Appendix G:** Quality Assessment Results

|  |  |  |  |  |  |  |  |  |  |  |
| --- | --- | --- | --- | --- | --- | --- | --- | --- | --- | --- |
| **Author(s) (year)** | **1. Clear statement**  **of aims** | **2. Qualitative methodology** | **3.**  **Research design** | **4.Recruitment strategy** | **5. Data collection** | **6. Relationship between**  **researcher and participant** | **7.**  **Ethical issues** | **8. Rigorous data**  **analysis** | **9. Clear statement of findings** | **10. How valuable is the research?** |
| Al-Yateem (2012) | Yes | Yes | Yes – although author could have elaborated on what the methodology was as opposed to only including a reference to it | Yes | Can’t tell –  doesn’t talk about the interview types, lengths, how this was recorded etc. Saturation is not discussed. | Can’t tell  Reflexivity and objectivity not discussed beyond being independent from the CF unit. Not discussed the impact of self on questions used within interviews. | Yes  Discussed that letters were sent to potential participants describing the study and asking to participate. | No  Not done in this paper. Referenced to somewhere else, but not discussed in any detail within this paper. | Yes | This research gives a voice to young adults in CF services in Ireland and is recommended to take this approach within other services to support young people in transition.  Says that things need to happen in transition but falls somewhat short of saying exactly what would be recommended. |
| Brumfield & Lansbury (2004) | Yes  Although not explicitly stated, the description of what the study wanted to explore conveyed this. | Yes  Used a less intensive methodology but still valid | Yes | Yes | Yes | Can’t tell.  Mentioned interviewer’s age as similar, but beyond that not much discussion about this or impacts at all | Yes | Yes  Discussed the process of second coding and simple thematic analysis | Yes | This research demonstrates a small sample with at least some diversity.  Future recommendations for practice are put forwards for this.  Little is discussed in terms of limitations of the research which would have been useful to see the authors reflect on to inform their recommendations. |
| Coyne et al., 2018 | Yes | Yes  Brief inductive analysis conducted as a part of a wider quantitative piece of work. | Yes  This was justified for their aims, however, the stated aims don’t highlight much qualitatively in terms of themes. | Yes | Yes | No | Yes | No  Mentioned that inductive thematic analysis was conducted, not how or by whom. Most data analyzed using quantitative methods. | No  Mention findings but just gives quotes with minimal context. No comprehensive themes due to nature of analysis | The overall paper is helpful and some of the qualitative data is welcome, however, this is very limited in scope due to the methods and analysis undertaken. |
| **Author(s) (year)** | **1. Clear statement**  **of aims** | **2. Qualitative methodology** | **3.**  **Research design** | **4.Recruitment strategy** | **5. Data collection** | **6. Relationship between**  **researcher and participant** | **7.**  **Ethical issues** | **8. Rigorous data**  **analysis** | **9. Clear statement of findings** | **10. How valuable is the research?** |
| Iiles & Lawton (2010) | Yes | Yes | Yes | Yes | Yes  Although no mention of saturation | Can’t tell  Mentioned that they had no clinical relationship but doesn’t mention bias or reflexivity. | Yes | Yes | Yes | The research provides insight into patient and professionals views of the parental role in transition and blends this together to discuss the potential conflicts / supports that this can bring. |
| South et al., (2022) | Yes | Yes | Yes | Yes | Yes | Yes | Yes | Yes | Yes | A very thorough accounting of young people’s experiences during transition using a specific transition protocol. |
| Tierney et al., (2013) | Yes | Yes | Yes | Yes | Yes | Yes | Yes | Yes | Yes | A detailed paper showing the experiences of young people transitioning which details concrete clinical activities that may be implemented to support |
| Vion Genovese et al., (2021) | Yes | Yes | Yes | Yes | Yes | Can’t tell  Mentioned that collecting data were not employees of the CF centre but no additional information relating to data analysis | Yes | No  No formal analysis method conducted; researchers spoke of “results classified by topic” | No  No formal table of themes or statement | Vast amounts of qualitative data provide a thorough account of the pre-transfer needs of young people and their parents. The findings are limited in scope due to the methodology utilised. |
| Zack et al., 2003 | Yes | Yes | Yes | Yes | Yes | Can’t tell  Mentioned that during recruitment researchers were not part of the CF transition programme - no details about the three researchers who grouped themes | Yes | No  Responses transcribed from notes taken during interviews, then phrases with similar meaning were grouped into themes | Yes | The use of participant quotes is welcome but how the data has been analysed limits any findings.  The research does discuss the need for clinical and guideline changes and details limitations of the research. |

## **Appendix H:** Examples of additional quotes for each theme

|  |  |  |
| --- | --- | --- |
| **Super-ordinate**  **theme** | **Subtheme** | **Quotes** |
| The Personal Impact | Mental health impact | *“Like … like about the diabetes, is it going to get worse” (Al-Yateem, 2012; p.852)*  *“big step … on edge … rocky uncomfortable time … I’d feel uncomfortable even more …” (Brumfield & Lansbury, 2004; p.228)*  *“it's deﬁnitely going to be a bit nerve wracking. I think I'm pretty much okay with it. I mean what's different about it I'm not sure” (South et al., 2022; p.)*  *“So he’s known me for a long time. That’s why I was worried about moving here.. he [paediatric Dr] knew a lot of the problems that I’ve had…” (Tierney et al., 2013; p. )*  *“It’s very stressful, when we tell you that you’re going to go to an adult center. . . and we don’t have a point of reference” (Vion Genovese et al., 2021; p262).* |
|  | Change of self | *“Since I went over to the adults’ [clinic] it’s been me more involved and she’s just sat back and she’ll take me if I want her to and she’ll sit there and she won’t say anything unless I ask her” (Iles & Lowton, 2010; p25).*  *“I started to listen a lot more and do more research and start to learn more and learn about the reason ofwhy I have to do the treatments and what the treatments could do to me and the medications that I consume. I just started to learn a lot more” (South et al., 2022; p.120)*  *“I manage my drugs myself, I get my drugs alone in a drugstore, they give me prescriptions, I do nebulizer cleaning, I manage quite well anyway” (Vion Genovese et al., 2021; p261).*  *“You’re treated more as an adult. You realize you can make your own decisions without needing parental consent” (Zack et al., 2003; p.379)*  *“I started to listen a lot more and do more research and start to learn more and learn about the reason of why I have to do the treatments and what the treatments could do to me and the medications that I consume. I just started to learn a lot more” (South et al., 2022; p. 120).* |
| What helped: supportive systems | Family as a resource | *“but Mum has always told me that you’ve got to keep your foot in the door in case we need them (the clinic)” (Brumfield & Lansbury, 2004; p.232 )*  *“She knows to make sure I start to slow down, grab a bucket, get in the car, shoot to the nearest hospital; let her deal with it” (Iles & Lowton, 2010; p24).*  *“When I was younger obviously my mum did them (set up nebulisers), and then obviously she just, I started to do it and then when I ﬁrst started doing it, she, my mum, just used to make sure that I didn’t overdose myself or anything” (Iles & Lowton, 2010; p25).*  *“She [Mum] tried to start falling back once I hit 18, but she was still there” (South et al., 2022; p. 25).*  *“I know it sounds dead daft, me mum was there and I’m 19 but because me mum was there I was more conﬁdent in asking questions because I knew if I’d said something that had come out a funny way or the wrong way, mum would go well what she actually means is this” (Tierney et al., 2013; p.743 )*  *“For all medical prescriptions and medication management, it’s my parents! Yeah, later I can start looking at some prescriptions, after treatments: I’ll try, physiotherapist, I’ll try too… it’s just for prescriptions...” (Vion Genovese et al., 2021; p261).*  *“there are things I don’t want to talk about and she says it for me” (Vion Genovese et al., 2021; p262).*  *“I prefer that there is always someone [parent] with me. Just in case I forget something, then, someone says it for me*” (Vion Genovese et al., 2021; p.261).  *“If I start coughing up blood again, she immediately knows what to do. She knows to make sure I start to slow down, grab a bucket, get in the car, shoot to the nearest hospital; let her deal with it” (Iles & Lowton, 2010; p. 24)* |
|  | Equipped health services | *“People need to be looked at and listened to … and then you make your diagnosis from there” (Al-Yateem, 2012; p.853)*  *“They had a special room aside for some of us adults who . . . knew each other from past get togethers, to sort of . . . make ourselves feel a bit more at ease” (Brumfield & Lansbury, 2004; p. 229)*  *“It was very smooth, it was within the one hospital and many of the health professionals were the same people” (Coyne et al., 2018; p.650)*  *“But that’s the good thing about the adult clinic, is the parents don’t have to be there” (Iles & Lowton, 2010; p26).*  *“I think that they [paediatric services] should start patients at 16, 17, even the parents just doing...letting the patient do things on their own so they could be ready and there's no surprises to them once they hit the adult clinic” (South et al., 2022; p.120)*  *‘‘as soon as the doctor started talking to me I was like, oh you’re talking to me rather than me mum, which was good” (Tierney et al., 2013; p.743 )*  *No, my doctor treats me as an adult. When I’m in the hospital, all the nursing staff treat me as an adult. I get the treatment I want’ (Zack et al., 2003; p.380)*  *“they were giving us information about the clinic and how it was run and stuff like that which I thought was pretty good”. (Brumfield & Lansbury, 2004; p.229).* |
| Endings | The end of parenthood | *“we had to take a back seat immediately” (Coyne et al., 2018; p.650)*  *“Mum obviously gets upset if you mention stuff like dying. So you have to be really careful” (Iles & Lowton, 2010; p27).*  *“I don't need my parents or my grandparents anymore, and you feel a little bit more conﬁdent doing It”.. (South et al., 2022; p.119)*  *“Because he doesn’t necessarily think about telling us things that seem essential, maybe he transcribed everything, but we are never sure” (Vion Genovese et al., 2021; p261).*  *“I think we really need to prepare parents beforehand, so that they can really start making their way to the idea that they have to let go of their child.”. (Vion Genovese et al., 2021; p. 261).* |
|  | The end of stable, relatable healthcare | *“I don’t really know, it might be either X hospital or Y hospital” (Al-Yateem, 2012; p.852)*  *“I don’t know we have to go and see … I don’t know how they will be like” (Al-Yateem, 2012; p.852)*  *“Situations like medications . . . they’d, they’d lecture to you and say oh, you know ‘if you don’t take these medications, you’re going to end up real sick’, and I just thought, oh . . . mate, I know” (Brumfield & Lansbury, 2004; p.228)*  *“it would have been good if someone explained how it would work and what we could expect” (Coyne et al., 2018; p.650)*  *“Sometimes it was like he (paediatric doctor) could read your mind…” (Brumfield & Lansbury, 2004; p.227).*  *“I was just clicking. I was like, this is the answer and you're telling me it's not right. Even though I know damn well it's right.” (South et al., 2022; p120.)*  *“didn’t seem to show interest in how you felt about moving over. It was more like we’ve sent your notes over to that side so we’re just waiting for them to reply”. (Tierney et al., 2013; p. )“Oh, when I went to… (the ﬁrst adult hospital) the Doctors were, sort of, a bit stand-ofﬁsh… said something when they were asked, not… they didn’t come out” (Brumfield & Lansbury, 2004; p.228).*  *“I don’t really like to come here… like they don’t do much for me… (Al-Yateem et al., 2012; p. 853)* |

## **Appendix I:** Enhancing Transparency in Reporting the Synthesis of Qualitative Research (ENTREQ) Statement (Tong et al., 2012)

|  |  |  |  |
| --- | --- | --- | --- |
| **No.** | **Item** | **Guide and Description** | **Page of Current Review** |
| 1 | Aim | State the research question the synthesis addresses | pp. 10 |
| 2 | Synthesis methodology | Identify the synthesis methodology or theoretical framework which underpins the synthesis, and describe the rationale for choice of methodology (e.g. meta-ethnography, thematic synthesis, critical interpretive synthesis, grounded theory synthesis, realist synthesis, meta-aggregation, meta-study, framework synthesis). | pp. 14 |
| 3 | Approach to searching | Indicate whether the search was pre-planned (comprehensive search strategies to seek all available studies) or iterative (to seek all available concepts until they theoretical saturation is achieved). | pp. 10 |
| 4 | Inclusion criteria | Specify the inclusion/exclusion criteria (e.g. in terms of population, language, year limits, type of publication, study type). | pp. 12 |
| 5 | Data sources | Describe the information sources used (e.g. electronic databases (MEDLINE, EMBASE, CINAHL, psycINFO, Econlit), grey literature databases (digital thesis, policy reports), relevant organisational websites, experts, information specialists, generic web searches (Google Scholar) hand searching, reference lists) and when the searches conducted; provide the rationale for using the data sources. | pp. 10 |
| 6 | Electronic search strategy | Describe the literature search (e.g. provide electronic search strategies with population terms, clinical or health topic terms, experiential or social phenomena related terms, filters for qualitative research, and search limits). | pp. 11 |
| 7 | Study screening methods | Describe the process of study screening and sifting (e.g. title, abstract and full text review, number of independent reviewers who screened studies). | pp. 11-12 |
| 8 | Study characteristics | Present the characteristics of the included studies (e.g. year of publication, country, population, number of participants, data collection, methodology, analysis, research questions). | pp. 17-19 |
| 9 | Study selection results | Identify the number of studies screened and provide reasons for study exclusion (e,g, for comprehensive searching, provide numbers of studies screened and reasons for exclusion indicated in a figure/flowchart; for iterative searching describe reasons for study exclusion and inclusion based on modifications t the research question and/or contribution to theory development). | pp. 16 |
| 10 | Rationale for appraisal | Describe the rationale and approach used to appraise the included studies or selected findings (e.g. assessment of conduct (validity and robustness), assessment of reporting (transparency), assessment of content and utility of the findings). | pp. 14 |
| 11 | Appraisal items | State the tools, frameworks and criteria used to appraise the studies or selected findings (e.g. Existing tools: CASP, QARI, COREQ, Mays and Pope [25]; reviewer developed tools; describe the domains assessed: research team, study design, data analysis and interpretations, reporting). | pp. 28 |
| 12 | Appraisal process | Indicate whether the appraisal was conducted independently by more than one reviewer and if consensus was required. | pp. 14 |
| 13 | Appraisal results | Present results of the quality assessment and indicate which articles, if any, were weighted/excluded based on the assessment and give the rationale. | pp. 42 |
| 14 | Data extraction | Indicate which sections of the primary studies were analysed and how were the data extracted from the primary studies? (e.g. all text under the headings “results /conclusions” were extracted electronically and entered into a computer software). | pp. 13 |
| 15 | Software | State the computer software used, if any. | pp. 13 |
| 16 | Number of reviewers | Identify who was involved in coding and analysis. | pp. 13 |
| 17 | Coding | Describe the process for coding of data (e.g. line by line coding to search for concepts) | pp. 13-14 |
| 18 | Study comparison | Describe how were comparisons made within and across studies (e.g. subsequent studies were coded into pre-existing concepts, and new concepts were created when deemed necessary). | pp. 13-14 |
| 19 | Derivation of themes | Explain whether the process of deriving the themes or constructs was inductive or deductive. | pp. 13 |
| 20 | Quotations | Provide quotations from the primary studies to illustrate themes/constructs, and identify whether the quotations were participant quotations of the author’s interpretation | pp. 28 |
| 21 | Synthesis output | Present rich, compelling and useful results that go beyond a summary of the primary studies (e.g. new interpretation, models of evidence, conceptual models, analytical framework, development of a new theory or construct). | pp. 29-36 |
| **No.** | **Item** | **Guide and Description** | **Page of Current Review** |
| 1 | Aim | State the research question the synthesis addresses | pp. |
| 2 | Synthesis methodology | Identify the synthesis methodology or theoretical framework which underpins the synthesis, and describe the rationale for choice of methodology (e.g. meta-ethnography, thematic synthesis, critical interpretive synthesis, grounded theory synthesis, realist synthesis, meta-aggregation, meta-study, framework synthesis). | pp. |
| 3 | Approach to searching | Indicate whether the search was pre-planned (comprehensive search strategies to seek all available studies) or iterative (to seek all available concepts until they theoretical saturation is achieved). | pp. |
| 4 | Inclusion criteria | Specify the inclusion/exclusion criteria (e.g. in terms of population, language, year limits, type of publication, study type). | pp. |
| 5 | Data sources | Describe the information sources used (e.g. electronic databases (MEDLINE, EMBASE, CINAHL, psycINFO, Econlit), grey literature databases (digital thesis, policy reports), relevant organisational websites, experts, information specialists, generic web searches (Google Scholar) hand searching, reference lists) and when the searches conducted; provide the rationale for using the data sources. | pp. |
| 6 | Electronic search strategy | Describe the literature search (e.g. provide electronic search strategies with population terms, clinical or health topic terms, experiential or social phenomena related terms, filters for qualitative research, and search limits). | pp. |
| 7 | Study screening methods | Describe the process of study screening and sifting (e.g. title, abstract and full text review, number of independent reviewers who screened studies). | pp. |
| 8 | Study characteristics | Present the characteristics of the included studies (e.g. year of publication, country, population, number of participants, data collection, methodology, analysis, research questions). | pp. |
| 9 | Study selection results | Identify the number of studies screened and provide reasons for study exclusion (e,g, for comprehensive searching, provide numbers of studies screened and reasons for exclusion indicated in a figure/flowchart; for iterative searching describe reasons for study exclusion and inclusion based on modifications t the research question and/or contribution to theory development). | pp. |
| 10 | Rationale for appraisal | Describe the rationale and approach used to appraise the included studies or selected findings (e.g. assessment of conduct (validity and robustness), assessment of reporting (transparency), assessment of content and utility of the findings). | pp. |
| 11 | Appraisal items | State the tools, frameworks and criteria used to appraise the studies or selected findings (e.g. Existing tools: CASP, QARI, COREQ, Mays and Pope [25]; reviewer developed tools; describe the domains assessed: research team, study design, data analysis and interpretations, reporting). | pp. |
| 12 | Appraisal process | Indicate whether the appraisal was conducted independently by more than one reviewer and if consensus was required. | pp. |
| 13 | Appraisal results | Present results of the quality assessment and indicate which articles, if any, were weighted/excluded based on the assessment and give the rationale. | pp. |
| 14 | Data extraction | Indicate which sections of the primary studies were analysed and how were the data extracted from the primary studies? (e.g. all text under the headings “results /conclusions” were extracted electronically and entered into a computer software). | pp. |
| 15 | Software | State the computer software used, if any. | pp. |
| 16 | Number of reviewers | Identify who was involved in coding and analysis. | pp. |
| 17 | Coding | Describe the process for coding of data (e.g. line by line coding to search for concepts) | pp. |
| 18 | Study comparison | Describe how were comparisons made within and across studies (e.g. subsequent studies were coded into pre-existing concepts, and new concepts were created when deemed necessary). | pp. |
| 19 | Derivation of themes | Explain whether the process of deriving the themes or constructs was inductive or deductive. | pp. |
| 20 | Quotations | Provide quotations from the primary studies to illustrate themes/constructs, and identify whether the quotations were participant quotations of the author’s interpretation | pp. |
| 21 | Synthesis output | Present rich, compelling and useful results that go beyond a summary of the primary studies (e.g. new interpretation, models of evidence, conceptual models, analytical framework, development of a new theory or construct). | pp. |

# Section Two: Research Project

Exploring how parents experience the diagnosis of cystic fibrosis for their baby in the post-natal period: An interpretative phenomenological study

## **Abstract**

**Objectives**

Despite guidance suggesting specialist psychological support is offered to families at the time of diagnosis of Cystic Fibrosis (CF) for their new-born, to date no study has sought to understand the experiences of parents at this time. This study aimed to explore the experiences of parents who have children who received a diagnosis of CF in the post-natal period.

**Design and Method**

Purposeful sampling was utilised to find parents who had received a diagnosis of CF in the post-natal period in the last three years. Eight participants took part in a semi-structured interview. Data was analysed using interpretative phenomenological analysis (IPA).

**Results**

Four group experiential themes were found - *“The diagnosis of CF changed everything”, “the unimaginable start to parenthood”, “following diagnosis, I’m alone*” and “*coping with diagnosis is impossible without support*”. Participants described the emotional impact the diagnosis had and how they felt their identity as a parent had changed as a result. Participants shared that ‘hope’ was an integral part of support, as were having support from family and a CF team that communicated *with* them.

**Conclusion**

Participants experienced the diagnosis as a moment that changed everything. The clinical implications of how to support parents with the changes associated with new-borns diagnosed with CF is discussed.

**Practitioner Points**

* The diagnosis of CF for parents of babies born in the post-natal period creates a significant emotional response.
* The gap between the initial telephone call indicating diagnosis and subsequent visit from the CF centre was a notable negative aspect of the diagnosis experience.
* Parents should be offered an assessment to identify any significant changes in parental wellbeing. In particular, the feeling of guilt that parents have appears to be significant.
* The social world is difficult to navigate as a parent following diagnosis.
* Parents benefit from hearing hopeful stories relating to CF prognosis and a CF team that communicates effectively. Furthermore, family is viewed as a helpful resource.

*Keywords:* Cystic fibrosis, diagnosis, parental experience, interpretative phenomenological analysis, qualitative.

## **Introduction**

Cystic Fibrosis (CF) is the most common genetic disease for white people in the United Kingdom (UK) (Schwarz et al., 2009). CF is a multisystem condition that predominantly affects the respiratory and gastrointestinal systems (Bishay & Sawicki, 2016). In the UK, the diagnosis of CF is commonly identified through the heel prick test screening (Connet, 2009) usually on a new-borns 5th day of life (Public Health England, 2018).

Following diagnosis in the UK, specialist centres facilitate most of the treatment to people living with CF as such care has been associated with better care outcomes, compared to locally provided, but non-specialist CF care (Johnson et al., 2003; Mahadeva et al., 1998; Quittner et al., 2014). According to Conway et al. (2014) specialist CF teams’ care should be provided by a multidisciplinary team (MDT) as this is essential to delivering good care. Indeed, the vast improvement in outcomes facilitated by specialist CF teams is no more apparent than when reviewing life expectancy figures (Keogh et al., 2020) as half of babies born today with CF in the UK are expected to survive beyond the age of 47 (Cystic Fibrosis Trust, 2017; Keogh et al., 2018) compared to the age of 31 between 1993-1997 (Dodge et al., 2007).

As CF is usually diagnosed in the first 5 days of the 6-week post-natal period (World Health Organisation, 2018) there are significant changes that occur to parents. Notably, the transition to parenthood, developing, and nurturing secure attachments for the new-born and finding one’s new identity within family and community life (Finlayson et al., 2020) are significant processes. Giving birth to a chronically unwell new-born can make these processes highly demanding (Carpenter et al., 2018; van den Tweel et al., 2008) as there are emotional (Halterman et al., 2004), financial (Zan et al., 2015), and psychosocial (DeRigne 2012; Stabile & Allin, 2012) changes occurring, parallel to the diagnosis. Coutinho et al. (2020; p.2) identified that the process of diagnosis is a “potentially traumatic experience for children and their parents”.

Ernst et al. (2010) stated that the widespread nature of CF diagnosis means it is a “family diagnosis”. The diagnosis of CF impacts not only the new-born but also the parents and wider family. For CF, the immediacy of the delivery of treatment (Cystic Fibrosis Trust, 2023) may increase the burden of responsibility on parents (Hodgkinson & Lester, 2002) as uncertainty around the condition and changes progress.

Research specifically relating to parental experience of a chronic health diagnosis at the time of birth have found parents face a range of difficulties. A recent systematic review (Hill et al., 2019) found that diagnosis of osteogenesis imperfecta (OI) in the post-natal period resulted in feelings of anxiety and distress. Additionally, it was found parents had to seek information about a condition they knew little about and had to do so, on their own. Similarly, research in post-natal diagnosis of complex congenital heart disease (CCHD) and sickle cell disease (Owoo & Tadros, 2021; Abu Ali et al., 2017), found diagnosis was a critical time for parents (Brosig et al., 2007).

Much of the existing literature is dominated by investigating the impact of diagnosis on care. However, how parents experience the social world around them following diagnosis of CF for their new-born may require further exploration due to the well-established link between looking after children with chronic illness and their quality of life (Golics et al., 2013). The social changes experienced with family and friends is stark as parents report they feel different to their parental peers and that conversation from them is seen as ‘depressive’ (Davis et al., 2009). Equally, family relationships become strained as family members seek to understand how best to support each other in the presence of the illness (Lewis, 1990). Within the literature, it appears that mothers of chronically unwell children are subsequently at a disadvantage through lower employment rates, thus reducing their quality of life compared to their spouses (Artazcoz et al., 2004). Conversely, there is literature to suggest that family relationships can grow stronger in the presence of chronic illness (Kim et al., 2007) although this has been seldom researched in families coping with diagnosis of CF with their new-born child.

Three studies on the experience parents have when their children are diagnosed with CF have been completed. All studies completed grounded theory analysis. Tluczek et al., (2006) identified parent’s experiences of the counselling support received at the time of their child’s positive screening test result. Chudleigh et al. (2017) sought to establish the impact on the family of a positive new-born screening test result. Grob (2008) elicited first-person accounts of how new-born screening programs areexperienced by parents whose children screen positive for genetic disease. Although all studies provided useful insights, the grounded theory design was utilised to produce theory from the data set, rather than focussing on the lived experience of parents (Barker & Pistrang, 2021). Barker (2015; p.71) considered that “well-conducted qualitative research exploring patients’ experience, their knowledge and experiences can be utilised for the benefit of others and the impact of the patient voice truly heard when designing and commissioning healthcare services”.

As the current literature on parental experience of diagnosis in CF is sparse, it has not yet included a qualitative approach that sets out to gain a more in-depth understanding of the parental lived experience of receiving a diagnosis of CF for their new-born child.

**Clinical Implications**

There are nuanced, complex and life limiting aspects of CF (Conway et al., 2014). Supporting bodies addressing the caring needs of people living with CF and their families have highlighted the importance of research to identify effective ways of supporting aspects of CF from the early stages of diagnosis (CF Foundation, 2023; NICE, 2017). Although parents and caregivers of babies diagnosed with CF naturally become experts in the child’s care (Balling & McCubbin, 2001), what experiences shape the development of such expertise has been seldom researched.

Clinical psychologists may be well placed to provide more tailored support to parents as they are viewed as one of key members of the CF MDT by professional guidelines (Conway et al., 2014) and colleagues (Mccullough et al., 2018). However, as the CF Foundation (CF Foundation, 2009) stated although the psychosocial impact of the diagnosis of CF on the family must be ‘carefully addressed’ at the initial visit, there has been limited research identifying where the focus should be, despite NICE guidance (2017) and the National Health Service (NHS) Long Term Plan (2019) advocating for specialist psychological support at the point of diagnosis. This research will attempt to identify information through the experiences of parents to guide these developments.

**Research Aims**

To use Interpretative Phenomenological Analysis (IPA) to explore the experiences of parents who have a child who received a diagnosis of cystic fibrosis in the post-natal period.

It particularly seeks to explore how the diagnosis was experienced, how it affected parenthood, and the development of a relationship with the new-born, and how interacting with others was subsequently experienced (e.g., physical health services, family, peer support and friends).

Primary Questions

* How do parents experience a diagnosis of CF for their new-born baby in the post-natal period?
* How do parents experience the process of becoming a new parent of a child with CF?
* How do parents of children with CF experience personal and professional relationships following diagnosis?

## **Method**

**Design**

The study utilised a qualitative design and adopted IPA as Smith et al. (2009) outlined. A qualitative approach was chosen because it enables an in-depth understanding of complex social and psychological experiences, including individual beliefs and interpretations of events (Barker & Pistrang, 2016), which aligned with the aim of the study.

**Reflexivity**

The researcher identifies as a White British male. They work as a Trainee Clinical Psychologist and are a parent. They do not access physical healthcare with their children, other than general visits to primary care when required. The researcher tended to reflexivity in further depth throughout the study in two ways. Firstly, they reflected on their personal position and the differences between themselves and the participant group (appendix A). Secondly, the researcher documented their beliefs, assumptions, thoughts, and emotional responses throughout the data collection and analysis stages in a reflective diary (Bradbury-Jones, 2007; appendix B). The reflections that will arise in this process were discussed with the research supervisor, enabling some level of “bracketing off” of personal experience (Yardley, 2008).

**Patient and Public Involvement (PPI)**

The Health Research Authority (2023) state that research should be done ‘with’ and ‘by’ members of the public rather than to them.

The researcher discussed the aims, interview schedule and methodology of the study with three parents of children who have been diagnosed with CF. One parent consulted was suitable for the study but was unable to be contacted. All parents consulted had received the diagnosis of CF for their children under five years ago and their children were receiving care from the specialist CF centre where the study was taking place. The aim of each discussion was to enhance the study’s methodology and to make sure the prepared research materials met the research aims. One change that was made related to the interview schedule brevity, as parents felt there was too many questions. One further change was made to the demographic information questions as parents shared that relationship status would be important to collect as it would give single parents a chance to be heard.

**Ethics**

Ethical approval was obtained via the Integrated Research Application System (IRAS, 316477) following full Research Ethics Committee review and Health and Research Authority Approval. All ethics approval documentation is contained in (appendix C).

**Recruitment and Participants**

According to Smith et al., (2009; p.48) “samples are selected purposively (rather than through probability methods) because they can offer a research project insight into a particular experience”. Additionally, Smith et al. (2009; p.3) emphasized that “IPA studies are conducted on relatively small sample sizes, and the aim is to find a reasonably homogeneous sample, so that, within the sample, we can examine convergence and divergence in some detail”.

To help ensure homogeneity, purposeful sampling was used to recruit a sample to examine parental experience of the diagnosis of CF for their new-born in the peri-natal period. Participants were identified in one specialist paediatric CF centre in the North of England, according to the study’s inclusion criteria, shown in Table 1.

**Table 1.**

*Inclusion and exclusion criteria*

|  |  |
| --- | --- |
| **Inclusion Criteria** | **Exclusion Criteria** |
| Adults (Age 18-65 years) | Received the diagnosis out of the post-natal period (e.g., at aged 6 months old) |
| Any gender | Any parents of children who had multiple diagnoses in the post-natal period |
| Biological parent of a child who has been diagnosed with CF | Individuals who were unable to provide informed consent. |
| Diagnosis was made in the post-natal period and no more than three years ago at the time of interview | Parents’ initial description of thinking and talking about the diagnosis of their child is still experienced as traumatic |
| Spoke English |  |

Potential participants were identified alongside the inclusion criteria by two clinical psychologists who worked in the MDT at the specialist CF site and through information posters placed on the walls of the CF specialist centre (appendix D). Participants who were deemed suitable (n=35) were approached either during their clinic appointment or through a telephone call. Initially, potential participants were provided a brief overview of the study and consent for the researcher to contact them with more information. Consenting participants were then contacted by the researcher and informed of the study’s aims and scope. If participants were interested in the study, they were then booked in to complete an interview at a time convenient for them. Following the opportunity to ask further questions, a participant information sheet was provided (appendix E) and informed consent was gained and recorded (appendix F).

Eight participants took part in the study. Smith et al (2009) recommend between four and ten interviews for doctoral-level work, whilst Smith & Eatough (2009) revised this number to be between 8-10 interviews. Eight interviews were deemed appropriate for the research aims and to provide higher information power (Malterud et al., 2016) as all participants were deemed suitable to generate rich data and regarded to be well informed about the diagnosis process. Participant demographic details are displayed in Table 3 (appendix G).

A total number of 22 people were invited to take part in the study. The participation rate was approximately 36.6%. Potential participants that did not consent often cited that they did not have enough time.

**Data Collection**

Data were collected via single face-to-face interview with each interview lasting between 50-86 minutes, an average of 66 minutes. Participants could complete the interview in person (n=1) or via video call (n=7). Prior to the interview starting, participants were reminded of the aims of the study and time for any questions was provided.

A semi-structured interview was used (Smith et al., 2022). An interview schedule (appendix H) of open-ended questions was developed following consultation with cystic fibrosis parents, a paediatric clinical psychologist working with the study population, a clinical psychologist who has two children with CF and an experienced qualitative researcher. Participants were given a £10 voucher for their participation in line with following research guidance by Gelinas et al. (2018).

Interviews were audio-recorded using an encrypted digital recorder and were subsequently transcribed. Audio-recordings were transcribed by a professional transcriber (n=6) and the researcher (n=2).

**Quality and Rigour**

Elliot et al., (1999) presented guidelines for reviewing qualitative research. The researcher adhered to the guidelines as presented in Table 2.

**Table 2.**

*Quality control*

|  |  |
| --- | --- |
| Guideline | Application to current study |
| Owning one’s perspective | The reflexive statement (appendix A) and reflexive diary entries (appendix B) have ensured that the researcher has considered their own thoughts, biases, assumptions and position. |
| Situating the sample | The researcher collected demographic information to allow the reader to understand the context in which the data was collected. |
| Grounding in examples | To evidence the themes, quotes from the interview are included in the results section. |
| Credibility checks | 20% of two transcripts were co-analysed by the researcher’s supervisor. To generate a ‘verification step’ a meeting was facilitated to discuss any similar codes, themes, or inconsistencies.  The author also discussed the final themes with an independent colleague.  Participants did not complete credibility checks which is an acknowledged weakness of the study. |
| Coherence | Within the results section, there is a succinct and clear summary of the findings. Additionally, the analysis methods are documented as how codes were created and how they link to themes. |
| Accomplishing general vs. specific research tasks | The findings of the current study provide a snapshot perspective of those who participated in the research. The researcher did not generalise the findings to all parents with children living with CF within the U.K. The researcher highlights generalisability as a weakness of the research. |
| Resonating with readers | The researcher described the findings in a coherent, clear way that corresponds closely to participants experiences and views. The findings may inform how specialist CF MDT teams support parents who experience the diagnosis of CF in their new-born babies |

**Analysis**

The analysis followed the integrative principles of Smith et al. (2009). Firstly, each interview was read line by line, and notes made aside from each of the transcripts. While the raw data was being reviewed, the researcher recorded thoughts and feelings that appeared within this process to tend to the reflexivity discussed (Smith, 2011).

Next, the researcher examined the transcripts and created exploratory notes, which aimed to collect comments as close to the participants explicit meaning as possible (Smith et al., 2009). Any parts of the interviews that become frequent and recurrent points in the data were noted. These initial interpretative comments were centred around participants’ descriptions of their experiences as a highly emotive, upsetting experience.  Next, the researcher read through the transcript and created experiential statements. The researcher then looked for connections and clustered similar themes from the experiential statements to create personal experiential themes (PETs) that went beyond what was stated explicitly (Ng & Barlas, 2023). The researcher then searched for connections across the PETs. This process was repeated for each transcript. Finally, grouping the PETs together to complete group experiential themes (GETs) was completed. Examples of each stage of the analysis are presented (appendix I & J).

## **Results**

Data analysis revealed four GETs, with several group level subthemes (Table 4). The themes are illustrated with multiple participant quotes (Appendix K). Participants’ contributions to each theme are presented in Table 5.

**Table 3.**

*Participant demographic information*

|  |  |  |  |  |  |  |  |
| --- | --- | --- | --- | --- | --- | --- | --- |
| Participant | Gender of participant | Gender of child | Year of diagnosis | Ethnicity | Relationship  status | Employment Status | Additional health needs of child |
| 1 | Female | Male | 2020 | White British | Married | Self employed | None |
| 2 | Female | Male & Female (Twins) | 2022 | White British | Married | Unemployed | None |
| 3 | Female | Male | 2022 | White British | Co-habiting | Employed full time | None |
| 4 | Female | Female | 2022 | White British | Married | Full time | None |
| 5 | Female | Male | 2022 | White British | Single | Unemployed | None |
| 6 | Female | Male | 2021 | White British | Co-habiting | Unemployed | None |
| 7 | Female | Female | 2020 | White British | Co-habiting | Employed part time | None |
| 8 | Female | Male | 2020 | White British | Married | Employed full time | None |

**Table 4.**

*Group experiential themes and group level subthemes*

|  |  |
| --- | --- |
| **Group Experiential Theme** | **Group Level Subtheme** |
| The diagnosis of CF changes everything | Immeasurable distress |
|  | I’m powerless |
| The unimaginable start to parenthood | The everchanging identity of Mum |
|  | This isn’t what I expected |
|  | It’s my fault |
| Following diagnosis, I’m alone | Is there anybody who understands? |
| Coping with diagnosis is impossible without support | Stories of hope |
|  | Family is so important |
|  | I’m communicated with |

**The diagnosis of CF changes everything**

This theme encapsulates the emotional response that participants experienced when the initial diagnosis conversation was occurring. The emotional response was strong as all participants recollected this as a critical moment that changed their lives.

***Immeasurable distress***

Many of the participants described the period of receiving a phone call from the specialist CF site to arrange a home visit or to attend a hospital appointment as a “heart sink” (P4), being hit by a “double decker [bus]” (P2) and that the phone call was “an out of body experience” (P6).

Participants reported there was a significant time delay between the initial phone call and then receiving formal CF information relating to the condition, its causes, treatments, and lifestyle changes. Some participants commented this was half an hour, whilst some participants said the wait was overnight. Most participants had limited knowledge of CF which they shared created concern. As participants were informed of the potential of a CF diagnosis via telephone and were offered a home visit, without further information, they experienced being placed into a position of not knowing. This gave way for what participants described was a disbelief, a frantic search for hope and a substantial emotional response.

*“Me and NAME-(partner) were just like pacing round the house, pacing what are were going to do, holding each other, crying, erm so yeah it was just terrible, terrible, terrible experience and then NAME –(CF Nurse) came” (P6).*

Due to how damaging the news was, some parents commented that they would have preferred not to be told over the phone, however, parents understood that there may not be another method.

*“but I don't know, just some kind of indication rather than just a phone call when we thought it was fine, like and “yes, he's got CF”. But she didn't say it like that you know, but, you know, just out of nowhere that word has been introduced to our world when we thought he was completely fine” (P3).*

One participant highlighted that they had to wait for half an hour for a member of the CF team to visit which had helped them cope with the initial distress which they described as “worry” and “fright” (P7).

*“it wasn’t prolonged to Google things or ring around… it stopped the anxiety building and I found it quite helpful”. (P7)*

***I’m powerless***

Several participants described that as they waited for more information relating to CF, they felt “powerless” (P6) alongside CF professionals who knew a great deal about the diagnosis and its implications. Indeed, as a lack of information was available to parents at this initial point of the diagnosis process, this echoed the lack of power they felt as a result. Participants spoke of how they had to request more information, sometimes repeatedly for it to be answered. Participants reported feeling the need to push healthcare professionals for more information, which additionally increased distress. The information provided in this context made participants “scared” (P3) as the answers could have huge implications for their baby.

*“Yeah, if she would have withheld that, that would have really frustrated me that, that would have really frustrated, erm and it would have felt like, I guess it would have felt like a power thing as well” (P6).*

Some participants were only aware of the condition as they had family friends who had been diagnosed or had memories from school. The lack of knowledge about CF understandably required participants to ask for information. As information was provided to parents about the treatments available, there was a disconnect between healthcare professionals and participants in what was deemed “exciting” in CF.

*“cos she would be like' it's an exciting time for CF right now'. I'm like I'm sure it's exciting for you, it's not really exciting for me” (P2)*

One parent was aware that they may have been high risk of having a CF baby. With prior knowledge of a chance of CF, they experienced the diagnosis as a damaging experience as she perceived healthcare professionals did not listen to their concerns. For them this meant they felt powerless in the situation.

*“watching all these nurses basically hold them down and they didn’t explain what it was they were [doing] (P3)*

*Participants described how s*ome healthcare professionals used their power to good effect to offer valuable, reassuring information which helped participants come to terms with what their life supporting their new-born with CF would look like.

*“he [Consultant] said on day one; “we’re not asking you to bubble wrap her and we don’t want you to, she’s got to”, TEARFUL sorry its getting me upset, you know “she’s got to experience life and go to nursery, and do normal things” (P4)*

**The unimaginable start to parenthood**

This theme covers participants narratives of their experiences of becoming a new parent. Participants felt that CF had changed the expectations they had for motherhood and how they were viewed as a new parent.

***The everchanging identity of Mum***

Participants reflected on how their identity as a Mum is everchanging. Identity was formed internally by new mothers and by the family and social structures around them. For some, participants described how they were now “overprotective” (P.1; P2), that they were an “educator” (P5) to family and friends and that the parenting as they did with other siblings was no longer compatible with a child with CF. One participant described not being a “normal parent” (P2).

*“I seem like the most overprotective mum in the world … I'm like sanitise your hands before you touch him” (P3)*

Some parents reported that being a Mum and looking after your child is in “the manual” (P2) and that you must be a Mum that cares “whether you feel good or not” (P1). Whilst these accounts of being a caring Mum when thinking about the early stages of CF were apparent across all the participants accounts, participants described how there was a fine line between the perception of a ‘good’ and ‘bad’ Mum. Participants described themselves on a pendulum between the two positions due to the complexities of managing the newly diagnosed condition and the related infection control risks.

*“you are constantly beating yourself up about if you’re doing a good job or a bad job, you’re constantly gonna have that voice in your head that’s saying ‘oh but really you shouldn’t be doing that’, you have just got to learn to manage it” (P1)*

Unanimously, participants shared that their identity as a Mum is as a “decision maker” as the complexities of the infection risk grew. As such, participants shared that they felt responsibility is on them to care for their new-born. Participants felt that this came at a cost as some described it as “tiring” (P4) and an extra layer on top of an already difficult task of being a parent.

*“I feel like it adds to the level of anxiety (LAUGH) to parenthood, erm I mean I think I feel like I’ve got that extra alarm bell in my head all the time about is this situation alright for NAME-(daughter” (P4)*

***This isn’t what I expected***

Participants spoke of how their expectations of parenthood had changed once CF had been diagnosed. Participants described varying emotions towards their new-born in the post-natal period such as “sadness and guilt” (P4; P6; P7) and feeling “gutted” for them (P3). For parents who had experienced a period of not knowing their baby had CF and had taken them home for several weeks, they noticed a shift in expectations.

*“the new-born bubble burst” (P7)*

Participants felt that due to CF diagnosis, they were required to re-negotiate internally with some of their family traditions, values and hopes. The expectations of the future, within these moments needed to be adjusted. These unexpected adjustments were experienced as losses for participants.

*“we were so excited, we are going to take our baby to as many places as we possibly can he will be the most travelled baby and then it's kind of that realisation there are certain things that he's not going to be able to or we kind of had planned or he might not be able to do” (P3)*

***It’s my fault***

The majority of participants described a change in feeling following diagnosis about the relationship they had with the diagnoses and the causes of it. Participants blamed themselves for the diagnosis. The primary feelings associated with this were feeling “guilt” (P4; P6; P7; P8), “selfishness” (P2) and “hurt” (P4; P8). Some participants attempted to create meaning as to why CF was in their lives describing how they must have “attracted it into my life” (P3) and that there was “something wrong with you [me]” (P4). One participant shared they made these perceptions of themselves emerged from previous life experiences “of course this was gonna happen to me, it was never gonna be like life was going too good” (P6).

*“like ‘god I did that, that’s my fault, I was being selfish wanting to have children’, and that’s hard” (P2)*

**Following diagnosis, I’m alone**

Participants unanimously spoke of accessing the social world following diagnosis with caution. Participants spoke of how the social world - defined as employers, family, friends, and strangers - did not understand the enormity of infection control and the treatment implications. To participants, keeping a safe distance meant they were doing right by their child by keeping away from uncertain, potentially infectious environments.

***Following diagnosis, I’m alone***

The significance of accessing the social world and being cautious in doing so was evident across all participant’s narratives. Some participants conveyed the view that accessing the social world is no different, as, if precautions are taken and in line with care guidance provided by the CF team, their baby in the initial stages of life and CF can stay safe. Participants felt pressure as participants shared their awareness of knowing that if the CF community was to see such activities on social media, there would be a “judgement” (P5). Not accessing social media altogether was explicitly described as not being a loss for one participant for this reason (P8). Additionally, *who* was present in social environments was perceived as a danger. Participants often felt that in the social environment, they were required to disclose CF into conversations to prompt people to maintain a safe distance.

*“I was like “oh hello, we are just waiting to be let in because we are going up to the Cystic Fibrosis Unit”, oh and stepped back, “he has CF?”, I was like yes, I was like how quickly could I get CF into the conversation so you back off (LAUGH)” (P2)*

Participants spoke of occasions they would need to create a story, to enable strangers to keep a safe distance. Although people were responsive to feedback to keep a safe distance, participants felt that this behaviour was unwarranted and that people with CF should be “given more protection” (P6).

*“Yeah, I’ve had a ticket person on the tram go to touch him, and I was like no, like lunge to forward, almost slapped his hand away, I was like “please don't”. But he was like “ok I won't touch your baby”, I was like “\_\_”. You kind of like have to make something up, like I'm just a very cautious parent (P4)*

One parent spoke of friendship groups changing as a result of the diagnosis as asking friends to understand what caring needs they had was difficult.

*“I lost a big friendship circle, they couldn’t understand that my care needs had changed, not just to a little girl, but to a girl that has special, extra needs… trying to educate was hard” (P7)*

Some participants had attempted to access support from other CF parents. The type of support discussed by participants was mostly through social media which detailed other families’ journeys of supporting a loved one with CF. Support through social media was described as a “minefield” which can interfere with your pre-set judgement as a Mum (P4). Peer support accessed in this way often created negative emotions as one participant felt that peer support caused them to feel bad (P1), experience “judgement from others” (P5) and to be “freaked out” (P3).

*“she's going through a bad patch where her little one has caught a bug or anything, her little one is about a year older than NAME-(son), she will post a lot and then I'll get freaked out, I’m like ‘oh my gosh he catches stuff here, here and here and I find for a couple of days after I'm like frantically cleaning” (P3)*

One participant was keen to share that peer support within the acute hospital setting following diagnosis worked for her. The participant noted how helpful her approach to coping with the initial process of diagnosis was able to be shared with others.

*“Yeah, and there’s no right or wrong or, you know, because NAME-(baby’s mum) next to me said “I’ve got two dogs and I’ve not been home and seen my dogs, and I said “just go, go and see they like, NAME-(baby) will be fine”. I can remember her saying “you’re the best thing that’s happened since I’ve been on this ward because I didn’t feel I could leave but I’ve seen you and you’ve left” (P1)*

**Coping with diagnosis is impossible without support**

This theme covers people’s understanding of what helped them get through the initial days of diagnosis. Participants actively wanted stories of hope, particularly in the presence of damaging conversations about CF being a life limiting disease. Participants were unanimous in describing the support they had received from the CF team was experienced positively.

***Stories of hope***

Participants shared that the feeling of hope came from familiar places such as family and friends and also the CF team. Such stories of hope were able to change participants’ pre-conceived ideas of what CF meant for their new-born. During the early stages of diagnosis many participants were seeking information about life expectancy. This usually occurred in an online context, often providing participants with significantly out-of-date information.

*“that initial Google search brings up a lot of either out dated information, like one thing that for me personally, I get really, really hung up on is the life expectancy” (P3).*

Participants described that hearing stories first hand, particularly from or about middle-aged people living with CF, allowed participants to see that their children could live fulfilling lives.

*“she told me about her son, he’s like fifty eight now and he competes in marathons and at the time when he was diagnosed he was told he wouldn’t live til ten and he didn’t get diagnosed until he was two… all these things were just like ‘yeah he can have a normal life” (P6)*

Participants felt that when the CF team were able to share such stories, they felt a change in feeling able to start “cracking on with it” (P5), having a renewed “focus” (P3) and that conversations at the stage of diagnosis were slightly more positive as result.

*“I can’t remember exactly what they said but I can always remember it being really hopeful and really positive” (P1)*

Participants felt hopeful that there would be role models for their children to follow when they are older as, by chance, coming across a famous person who was successful with CF provided hope that their child could one day do the same.

“*my boys could grow up… he could be a role model for them if they got social media and followed him” (P8)*

***Family is so important***

Participants expressed that the support they had from family was significant. Participants felt family were able to provide emotional and practical support either as the diagnosis news was in its infancy or in the following days and weeks.

*“actually, absolutely overwhelming amount of support from family” (P4)*

Family members were able to offer practical support such as keeping diaries of hospital events at the time of diagnosis (P2), attending the hospital when requested (P1) and being present at the time of the initial CF team visit (P6).

*“We are phoning up, “NAMEfather-in law, you are going to have to come and pick NAME-(daughter) up, like something is not going right to plan, we need to go down to Special Care” (P2)*

***I’m communicated with***

Participants were keen to share their recollections of how they were communicated to at the time of diagnosis. Participants spoke of the diagnosis being shared in a way that was provided with “no fluff” and that was “frank and honest” in content (P1). Participants felt they responded well to care that was done in a “considerate” way (P4) which was delivered “softly” (P8) and by healthcare professionals who could “listen” (P2; P6; P8). Participants shared that consistency helped and a ”caring” (P7) approach helped relieve the early difficulties associated with diagnosis.

*“Because they are dead good at listening too (LAUGH), and they would often, they would often just sit and listen while they were doing their physio, like say ‘what’s going on today’” (P2)*

As the content of conversations following diagnosis began to focus on how to live life with CF, there were distinct individual differences in what participants valued. One participant particularly valued the ‘*doing with*’ than ‘*doing to*’ approach.

“I was like “wow”, are they actually asking what I want to happen rather than just like ‘this is happening” (P2)

Another participant was keen to share how they enjoyed the “detail” in information sharing (P1; P7) whilst another wanted the “brutal truth” at all times to facilitate their processing of the news (P6). Participants shared they wanted to know the “risks and benefits” (P4) as this was the best way for them to get on board with any treatment plan.

## **Discussion**

The study aimed to explore the experience of mothers who had received a diagnosis of CF for their new-born baby in the peri-natal period. Four themes were identified: “*the moment that changed everything”, “parenthood changed forever”, “the social world”* and *“impossible without support”.*

**The diagnosis of CF changed everything**

The distress experienced by participants when notification that genetic markers indicated CF diagnosis, replicates previous research showing parents who receive a diagnosis of a genetic condition for their new-borns, have a significant emotional reaction (Hill et al., 2019; Salm et al., 2012; Tluczek et al., 2006). As with other research, participants’ pre-existing knowledge of CF was found to be limited (Chudleigh et al., 2016), which in turn for participants contributed to the distress felt, aligning with previous research which has shown uncertainty can negatively affect parental wellbeing (Folkman & Greer, 2000; Madeo et al., 2012).

Qualitative research has found that parents would like healthcare professionals to be aware of the impact of extended waiting for bad news related to health diagnosis (Brouwer et al., 2021). For participants, waiting was reported by them to be one of most distressing parts. Noticeably, all participants were able to recall these events easily during interviews aligning with other recollections in research when parents were first told about their child’s genetic condition (Forrest et al., 2008).

The waiting period between a telephone call identifying a diagnosis and a subsequent visit is a recognised problem following a positive result on the new-born screening test (Ulph et al., 2017), despite UK guidance recommending the call (Southern, 2019). As participants waited for information, and informational needs were not met, this triggered what Ulph et al., (2015) identified as the reason anxiety and catastrophic rumination occur. Johnson et al. (2019) described “information gathering action” occurs during the wait to lessen the undesirable anxiety state. Indeed, many participants sought information at this time which was unhelpful to them.

Mothers made sense of the experience of waiting for information through a perceived lack of power which resonates with previous research discussing power imbalances in healthcare (Johnson et al., 2019). Participants felt they were not in equal communication with healthcare professionals as professionals held knowledge that participants ultimately wanted quick access to. This may have reflected the ‘classic’ paternalistic model of the healthcare professional-carer relationship as parents often want to be seen as an equal partner when bad news is given (Gabe et al., 2004). In addition to an initial power imbalance, participants highlighted a memorable emotional disconnect, too. For parents who receive the diagnosis of CF for their new-born, one sentence such as “this is an exciting time for CF” can influence families’ narratives of diagnosis (Tobler et al., 2014). For parents of children who have been diagnosed with CF in the post-natal period, managing the non-conversational aspects of diagnosis such as the wait to be seen, a lack of clarity about future treatment and by feeling prepared for the conversation, compared to a sudden telephone call (Brouwer et al., 2021) may be important in supporting mothers who receive a diagnosis of CF for their new-born baby.

**The unimaginable start to parenthood**

Changes in how you perceive yourself as a parent following diagnosis are apparent in health research (Larun & Malterud, 2007). Changes in identity related specifically to parents whose children have been diagnosed with CF was termed in previous literature as “a whole new world of CF” by Boardman & Clark (2022) due to the immediacy of the health threat. Other participants reported they felt their identity change as they were required to become an “educator”, noticing they were “overprotective” and “not a normal parent”. Changes in identity were demonstrated in previous research with parents of children with CF (Iles & Lowton, 2010) with the findings also replicating research in CF that identified that parents see themselves as “decision makers” within the role of their new-born’s care (Buchanan et al., 2022).

Participants felt that they were on a pendulum between being a ‘good’ and ‘bad’ parent which differed from previous research identifying mothers of children with CF as confident in their abilities (Cammidge et al., 2016; Frankl & Hjelte, 2004). As participants were reflecting on the start of their caring journey in supporting their new-born children with CF, competence in your own abilities as a parent may develop with time, as found in previous research (McDonald et al., 2013). Additionally, post-traumatic growth (Tedeschi & Calhoun, 2004), identified as occurring in parents with children of CF (Byra et al., 2021) and the positive adaptations made in the context of highly challenging life circumstances, specifically related to competence, may have not been noticed or experienced in the early stages of diagnosis (Irie et al., 2019).

The findings echo previous research which shows that feelings of guilt and fear occur as the adaptation in supporting your baby begins (Tluczek et al., 2005). The expectations that mothers held for their child suddenly changed which has been identified in other research relating to genetic disorders (Leff & Walizer, 1992; Suza et al., 2020).

A prominent feeling associated with the diagnosis was “guilt” as mothers blamed themselves for giving their child a genetic condition. The feeling of guilt is unanimously present in other research identifying parents’ experiences of diagnosis for their children (Berge et al., 2004; Clark et al., 2020; George et al., 2006; James et al., 2006) and equally in parents of children with CF (Barker et al., 2017; Cammidge et al., 2016).

**Following diagnosis, I’m alone**

Participants described that the social world they accessed, at times, did not ‘fit’ with the CF diagnosis as social activities were required to be thought about and negotiated, as infection risk increased and changes with friendships occurred. Participants noted that they either created stories to enable people to keep their distance or openly shared the diagnosis. Changes in the social world following diagnosis are evidenced across health literature (Davis et al., 2010) as care needs change (Bowen et al., 2010). Research proposes (Golics et al., 2013) that due to these occurrences, considering the effect of treatment has on patients’ close social group is therefore important.

**Coping with diagnosis is impossible without support**

Participants were keen to share how hope played an important role following diagnosis. Hopeful conversations are celebrated by parents of children with CF (Jessup et al., 2018). There is unanimous agreement within the paediatric cancer literature, too, that hopeful conversations are a key asset to the care provided as the impact on parents is significant in improving outcomes (Graetz et al., 2022) and psychological adjustment (Truitt et al., 2012). Snyder et al. (1991) offered a theory that hope results from an individuals perceived ability to develop numerous and flexible pathways towards their goals. The goal of mothers, as shared in their experiences was to keep their new-born safe and well. To work towards this goal, information related to fulfilling lives, may have initiated participants to identify barriers and strategies to move towards that goal.

Participants’ families offered them support with the diagnosis process. Support from family in the process of CF diagnosis is an established link (Barker et al., 2017; Cammidge et al., 2016; Jessup et al., 2018) and recognised as a ‘basic need’ (Maslow, 1943). As the link between early CF treatment and outcomes is evidenced (Quittner et al., 2014) support from family too, and at this early stage, is seen to contribute to successful CF management (Barker et al., 2012).

Finally, mothers, valued a communication style grounded in consideration, which was “soft” in style and containing “no fluff”. This resonated with previous literature detailing how to be able to give support to a family at a sensitive time of life (Jansson et al., 2001; Tluczek et al., 2022).

**Strengths, Limitations, and Future Directions**

The research study addressed a gap in the literature of mothers experiences of the diagnosis of CF in the post-natal period for their new-born child. It is the first study attempting to gain a more in-depth understanding of the parental lived experience of receiving a diagnosis of CF for their child in the post-natal period.

Due to who volunteered for the study, unintentionally, the research offered mothers the platform for their experiences to be heard. As such, this is a valid weakness of the study too as although Fathers (n=2) were invited to take part in the research, they were unable to do so. This reflected a wider issue of there not being adequate number of Fathers involved in child health and development research (Davison et al., 2017). Capturing Father’s experiences of the CF diagnosis in the post-natal period may be an essential part of future research.

As 22 participants were contacted to participate, those who chose not to take part may have felt unable to share their story. As a result, the experiences of those who felt unable to take part may have been missed.

The diversity within the sample of the study is a weakness. Although CF is the most common genetic disease for White British people, recent evidence (UK CF Registry Annual Data Report, 2021) shows that 3% of those on the Registry are Asian, 0.2% are Black or Black Caribbean, 1% identified as ‘Mixed’ ethnicity and a further 3% identify as ‘Other’ or do not state their ethnicity. The study’s sample comprised of eight White British mothers’ experiences only which is a weakness of the study. Future research would benefit from seeking to understand the lived experiences of people affected by CF from ethnic backgrounds, as racial and ethnic disparities exist and have been found to exist in a broad amount of literature (Oates & Schechter, 2021). As such, the ability to generalise to other contexts and demographic groups (Myers, 2000) is a concern.

The researcher acknowledged the reflexive position in which they conducted the research. As the researcher was a White male, it should be recognised that the researcher was fully responsible in examining and interpreting the experiences of the eight female participants. Alongside the researchers’ gender, and due to working in clinical psychology, a different gendered researcher with a different professional background may have yielded different results. Although this was acknowledged throughout the analysis, the researcher may still have had limited awareness of specific considerations.

Finally, one strength is the initial involvement of PPI as three mothers of children diagnosed with CF from the CF centre were consulted on the study’s methodology and related research materials.

**Implications**

Within CF team guidance, clinical psychologists are key members of the MDT (Conway et al., 2014). Such guidance suggests that their work should focus on supporting children, parents and caregivers and the wider MDT to support the recently diagnosed new-born. The clinical psychology profession, due to their training in therapy and consultancy (BPS, 2023**),** may therefore be well placed to facilitate the findings of the research into clinical practice.

Participants described the emotional response they had to the diagnosis. Their narratives of the distress it caused were profound and echoed the understandable emotive responses of receiving a health diagnosis like CF on behalf of your new-born baby (Brosig et al., 2007). The time between the initial telephone call and visit from the team were experienced as damaging. There is potential that as the initial telephone call is made, it is only done so when a member of the CF team can attend their home address as soon as possible after the call. It may be of the interest to teams and families that psychologists facilitate and co-create a new initial diagnosis pathway with such measures in mind. Equally, supporting teams who deliver the news can be beneficial, too. Afterall, as Brouwer et al., (2021; p. 280) identified “for the time being, the most pragmatic approach may be the one put forward by the parents in the study—"Ask parents how they wish to be informed”.

As mothers struggled with the everchanging identity of themselves as a parent, it may be of importance to psychologists supporting mothers in the early stages to initially notice and subsequently explore and amplify mothers’ values in conversations. Acceptance Commitment Therapy (ACT; Hayes et al., 1999) tends to the notion that life includes painful events and attempts to avoid pain tend to magnify the pain rather than eliminate it. Within the early stages of diagnosis, mothers experienced the diagnosis as changing how they saw themselves; even as ‘bad’ mothers. ACT has been found to be helpful for parents when their children are faced with life threatening diseases (Burke et al., 2014). It particularly allows individuals to focus on what is important to them and ‘in service’ of personally chosen values.

The feeling of guilt has been associated with hopelessness, effective parenting, and depression alongside genetic illness for parents’ children (Nixon & Singer, 1993). Through routine assessment, psychologists should be alert towards hearing the feelings that parents are experiencing. Wong and Heriot (2008) found that parents who blame themselves for their child’s illness, reduce pursuing goals for their child and have high vicarious despair. It may be important for healthcare teams working with newly diagnosed parents to offer formal, brief support, to be alert to changes within these domains and to offer subsequent treatment if required as high levels of emotional support and encouragement to have hope are likely to experience lower levels of distress (Wong & Heriot, 2008). Although support groups in person or online have been found to help parents with feelings of guilt, such interventions within CF care should be approached with caution as some participants shared that their experiences of peer support had not been helpful (Nixon & Singer, 1993).

Mothers wanted conversations that contained “no fluff” and that was “frank and honest”. Clearly, there is some advantage, when timed right, that stories from adults with CF that are thriving are helpful. It would be encouraged to incorporate such stories of hope in answers to newly diagnosed parents. Equally, traditions from positive psychology (Barakat et al., 2009; Seligman et al., 2005) and solution focused brief therapy (SFBT; deShazer, 1985) can allow healthcare professionals to enquire what hopes parents have for their children and what strengths are available to meet short and long-term goals. Questions from SFBT practice and its link to systemic therapy can initially view the family in relation to the contexts in which they operate i.e., a family experiencing a significant emotional reaction to bad news in order to unlock family resources which can give wider health teams a chance to amplify them. As SFBT approaches have been found to be easily trained to and delivered by wider MDT members, in particular nurses (Bowles et al., 2001), psychologists are well placed to train and model such approaches to ultimately support families.

## **Conclusion**

Mothers of children with CF shared that their experience of the diagnosis of CF for their new-born was a cause of significant distress which had implications on their identity as a parent. Mothers noticed unexpected feelings once the diagnosis was given that they would not usually associate with a new-born. Additionally, the social world added significant challenges. Participants noticed how stories of hope, family support and being communicated *with* were all vital to coping.

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## **Appendix A:** Reflexive Statement

The researcher identifies as a white British male, in his thirties. His family were all born in the UK. He grew up in a middle-class family, within and around British norms. He considers himself a person who values family and is a parent themselves. The researcher does not access extensive health systems with their two children, occasionally visiting primary care for routine care. The researcher’s parents both worked in public service which they believe ultimately forms many of the values they hold today. From conversations about who people are, what their vulnerabilities are and how the systemic system around people forms many of their experiences, values of honesty, integrity and being ethical are held. The researcher believes that those in positions of power should use their power to benefit those who are less able. Additionally, the researcher believes that healthcare, free at the point of access, should always be the healthcare model of the UK.

Professionally, the researcher the researcher is a trainee clinical psychologist with experience of working in paediatric physical health teams in the NHS. The researcher has always been struck by how emotive events in the healthcare system, such as diagnosis and transition, are often handled. The researcher has observed that such events are an anxiety provoking time for both families and young people. Such work has enabled the researcher to want to understand how best families can be supported at these times.

The researchers default position is how best healthcare teams can be pragmatic and at the same time take a backward step. Although the researcher highly values the NHS and the staff that work within it, he seldom believes that the NHS always has the right approach to support people. The researcher much prefers a ‘helpful visitor’ stance when working in the NHS and with children and families, preferring to rely on the families’ pre-existing resources.

## **Appendix B:** Example excerpt from the researcher’s reflexive diary log

I enjoyed all of this interview but found aspects of it hard. Personally, I felt was fully engaged with the conversation we were having about the diagnosis. I enjoyed asking for more information and the relaxed tone I brought to the interview.

I noticed for a short period I was able to really hold in mind their experiences, and was allowed to be myself, and ask questions to develop their narrative about experiences. I’m taken aback slightly at how emotive the conversation was as the process was not straightforward for them. This was the longest interview and wonder whether it went on for as long as it did due to my interest in their experiences or because they wanted to keep sharing their story with me. I believe I connected with the person because they share the characteristics I enjoy in people, such as being warm and friendly and having a few similar interests and outlook on life.

The difficult parts of the interview were the aspects that completely contrasted with my experiences of the first 6 weeks post birth of my two children. My first child and the participants child were born 3 days apart. The difference in our experiences were vast. I can recall the first week of my son’s life as this was the week the UK went into lockdown. I can remember the Prime Minister locking the country down on the Monday night and the weeks ahead from that point very well. During the interview, the participant took me through their own experiences that week and each key event of lockdown sounded so similar and so different to mine. The contrast was that the patient was on their way to hospital whilst I was sat at home without any worry and only happiness. It makes me think how lucky I am and I notice this happening in the interview. I feel guilty that the participant asked if I was a parent and I replied yes which, although I don’t know, felt that the participant was talking to someone who ‘got it’ and what it is like to be a parent.

I was really struck with how well the staff responded to the patient and her needs. I am struck with the care she has for other people as her experiences involve supporting strangers around her who were going through something similar.

## **Appendix C:** Health Research Authority HRA Ethical Approval Form

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## **Appendix D:** Information poster about the research project

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## **Appendix E:** Participant information sheet

Mark Guyers,   
Trainee Clinical Psychologist,   
The University of Sheffield.



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**PARTICIPANT INFORMATION SHEET**

**STUDY TITLE: A qualitative study exploring parent’s experience of the diagnosis of cystic fibrosis for their new-born baby.**

You are being invited to take part in a research study. Before you decide, it is essential to understand why this research is being done and what it will involve. Please read the following information carefully and contact me with any questions that you may have.

**Why have I been invited?**

You have been identified during one of your routine clinic appointments by a clinician or through the research advert you saw in the cystic fibrosis unit. You have agreed to seek further information to participate in this research which was asking for parents of children who have been diagnosed with cystic fibrosis in the first 6 weeks of their life to take part in this research project.

You have also been identified as an adult living in the UK who is fluent in English, can meet in person or use the internet for virtual video calls.

**What are the aims of the study?**

To use Interpretative Phenomenological Analysis (IPA) to explore the experiences of parents who have children who received a diagnosis of cystic fibrosis in the post-natal period.

It particularly seeks to explore how the diagnosis was experienced, affected parenthood, developing a relationship with the new-born, and interacting with others (e.g., physical health services, family, peer support and friends).

*Primary Questions:*

How do parents experience a diagnosis of cystic fibrosisfor their new-born baby in the post-natal period?

How do parents experience the process of becoming a new parent of a child with cystic fibrosis?

How do parents of children with cystic fibrosisexperience personal and professional relationships following diagnosis?

**Do I have to take part?**

No. Your participation in this study is voluntary. It is up to you whether you would like to take part. If you decide to take part, you can keep this information sheet and will be asked to sign a research consent form. **You can withdraw from this research at any time without giving a reason**.

**What will happen if I take part?**

The lead researcher will contact you via telephone, email, or post (please indicate which is your preferred method) within two weeks. You will be asked to take part in an interview which could last up to 90 minutes. In this interview, you will be asked some questions about your experience of receiving the diagnosis of cystic fibrosis for your child and the subsequent time following this.

The interview will be either in person (in the cystic fibrosis unit, Sheffield Children’s Hospital) or via a virtual video platform (Google Meet/Microsoft Teams). If you decide to complete the interview online, a link will be emailed or posted to you, and you will need to click the link at the allocated time given to you. You are welcome to have someone help to support to set this up too. However, they are unable to participate in the interview with you.

Your interview will be recorded and then transcribed using a 3rd party, University of Sheffield approved transcriber. The transcriber will receive instructions on how the store the data that they receive and will be asked to sign a confidentiality agreement. Following transcription, it will then be analysed using Interpretative Phenomenological Analysis. If you have any questions about this type of analysis, you are welcome to ask about these.

**What are the benefits of taking part?**

You have an opportunity to share your experiences of receiving a diagnosis for your child. A written report of the findings will be can be sent to you, and we hope this will be useful for parents receiving a diagnosis in the early weeks of their child’s life and NHS teams, like the one at Sheffield Children’s Hospital, who provide care.

**What if there is a problem?**

If you feel there is a problem at any time, you can let the researcher know. You can let the researcher know by telephone via the research support officer on 0114 222 6650.

You can also let the researcher know by email or by speaking to the researcher during any contact in the study. This may be a topic that is difficult to talk about or could feel distressing at times. If required, the researcher can discuss what further support may be of help (e.g., contacting your GP, talking to the cystic fibrosis team).

**Will all the information be kept confidential?**

Yes. **All your information will be kept strictly confidential**. You will not be identifiable in any reports or publications.

The only exception to this would be if during the interview the researcher held some concern about a risk of harm to yourself (e.g., thoughts of hurting yourself), or someone else (e.g., a child or another adult) that you talk about (e.g., risk of neglect of physical harm). In such a situation, the researcher will discuss the need to breach confidentiality. This is to best support you, others and ensure safety. This may involve contacting relevant services (for example, your GP) to let them know about the situation, provide help and the proper support.

**Will I receive any reimbursement of expenses for taking part in the research?**

Yes. There will be a reimbursement of relevant travel expenses incurred by participating in this research (subject to providing receipts of travel).

You will also receive a £10 voucher of your choice for participating in the study.

**What will happen to the results of the study?**

The results will be submitted as part of the researcher’s doctoral thesis in May 2023 and then prepared for publication in the summer of 2023. You can let the researcher know at the start of the study if you would like a copy of this, and it will be sent to you.

The University of Sheffield is organising and funding this research. This project has been ethically approved by the Cambridge & Hertfordshire Research Ethics Committee (IRAS No. 316477)

**What if I wish to complain about the way the study has been carried out?**

Should there be any complaints about this study, you can contact the lead researcher, Mark Guyers, or, alternatively, you can contact the supervisors involved in this project

* **Mark Guyers**, at mguyers1@sheffield.ac.uk
* **Dr Vyv huddy** Lecturer in Clinical Psychology at Sheffield University at v.huddy@sheffield.ac.uk
* Dr Jaime Delgadillo, Director of Research at the University of Sheffield on j.delgadillo@sheffield.ac.uk

**If you feel that your complaint has not been handled to your satisfaction following this, you can contact:**

Professor Gillian Hardy, Head of Department, on g.hardy@sheffield.ac.uk

**Contact information**

Mark Guyers, Trainee Clinical Psychologist, is conducting this research. This research will be used to write a thesis that fulfils part of their doctoral training. If you have any questions about the study, you can contact the primary researcher by email at [mguyers1@sheffield.ac.uk](mailto:mguyers1@sheffield.ac.uk). Alternatively, you can leave a telephone message with the Research Support Officer on 0114 222 6650, and he will ask Mark Guyers to contact you.

**How your information will be used**

In this research study we will use information from you. We will only use information that we need for the research study. We will let very few people know your name or contact details, and only if they really need it for this study.

Everyone involved in this study will keep your data safe and secure. We will also follow all privacy rules.

At the end of the study we will save some of the data in case we need to check it.  
We will make sure no-one can work out who you are from the reports we write.

The information pack tells you more about this.

**How will we use information about you?**

We will need to use information from you from the semi structured interviews we complete for this research project.

This information will include your initials and contact details. People will use this information to do the research or to check your records to make sure that the research is being done properly.

People who do not need to know who you are will not be able to see your name or contact details. Your data will have a code number instead.

We will keep all information about you safe and secure.

Once we have finished the study, we will keep some of the data so we can check the results. We will write our reports in a way that no-one can work out that you took part in the study.

**What are your choices about how your information is used?**

* You can stop being part of the study at any time, without giving a reason, but we will keep information about you that we already have.
* We need to manage your records in specific ways for the research to be reliable. This means that we won’t be able to let you see or change the data we hold about you.
* Where can you find out more about how your information is used?

You can find out more about how we use your information

* at [www.hra.nhs.uk/information-about-patients/](https://www.hra.nhs.uk/information-about-patients/)
* by asking one of the research team
* by sending an email to Luke Thompson (Head of Data Protection and Legal Services) on [dataprotection@sheffield.ac.uk](mailto:dataprotection@sheffield.ac.uk) https://www.sheffield.ac.uk/govern/data-protection/contact
* by ringing us on 0144 222 6650

**Helpful Telephone Numbers**

If you feel you need some support following completion of the interview, you can seek immediate help:

* Contact your GP for an emergency appointment or the out-of-hours teams
* Call NHS 111 (Non-emergency medical help and advice for people living in England)
* Contact the Samaritans (24/7) samaritans.org116 123 (freephone) jo@samaritans.org

Samaritans - samaritans.org

Mental Health Foundation - mentalhealth.org.uk

MIND - mind.org.uk

NHS 111 - 111.nhs.uk

Cruse bereavement care - cruse.org.uk

Maytree (suicidal support telephone number) - maytree.org.uk 020-7263-7070

NHS Drug addiction - nhs.uk/live-well/healthy-body/drug-addiction-getting-help

Alcoholics anonymous - alcoholics-anonymous.org.uk

CALM - thecalmzone.net

National suicide prevention alliance - nspa.org.uk

SHOUT - crisistextline.uk

## **Appendix F:** Informed consent form

Mark Guyers,   
Trainee Clinical Psychologist,   
The University of Sheffield.

Email: mguyers@sheffield.ac.uk

Department of Psychology.  
The University of Sheffield,  
Floor F, Cathedral Court,   
1 Vicar Lane,   
Sheffield, S1 2LT   
UK

**Title of research project: A qualitative study exploring how parent’s experience the diagnosis of cystic fibrosis for their baby.**

IRAS No. 316477

**Name of researcher**: Mark Guyers

**Participant identification number for this study**: \_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_

*Please read the following form.*

*If you answer ‘No’ to any question below, please do not proceed with this consent form. If you would like to have further clarification on any question below, please ask the researcher.*

*For you to be able to take part in this research, all questions below need to be answered ‘yes’. You are free at any time to withdraw if you do not agree to any of the below statements. You do not have to provide a reason to the researcher if this arises.*

|  |  |  |
| --- | --- | --- |
| **Please tick the appropriate boxes** | **Yes** | **No** |
| 1. I have read and understood the project information sheet of the study has been fully explained to me. *N.B. If you answer ‘No’ to this question, please do not proceed with this consent form until you are fully aware of what your participation in the study will mean* |  |  |
| 1. I have been given the opportunity to ask questions about this study. |  |  |
| 1. I agree to take part in the study. I understand that taking part in the study will include participant in an interview that will be audio recorded. |  |  |
| 1. I understand that my participation is voluntary and that I am free to withdraw at any time without giving any reason and without there being any negative consequences. In addition, should I not wish to answer any question or questions, I am free to decline. |  |  |
| 1. I understand that my responses will be kept confidential meaning that I will not be identified or identifiable in the report or reports that result from the research. |  |  |
| 1. I understand and agree that my words may be quoted in publications, reports, web pages, and other research outputs. I understand that I will not be named in these outputs unless I specifically request this. |  |  |
| 1. I understand and agree that other authorised researchers may use my data in publications, reports, web pages, and other research outputs, only if they agree to preserve the confidentiality of the information as requested in this form. |  |  |
| 1. I agree for the data collected from me to be stored anonymously and potentially used in future research. |  |  |
| 1. I agree to assign the copyright I hold in any materials generated as part of this study to The University of Sheffield. |  |  |

\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_ \_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_  
Name of participant Date Signature

\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_ \_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_\_  
Lead researcher Date Signature

*Ask for participant to give contact details if they wish to receive further information and make sure you have a process for giving them the voucher!*

***To be signed and dated in presence of the participant***

*Copies:*

*Once this document has been signed by all parties the participant should receive a copy of the signed and dated participant consent form and the information sheet. A copy of the signed and dated consent form should be placed in the study’s main record. (e.g., a site file). This must be kept in a secure location.*

## **Appendix G:** Demographic questionnaire

Thank you for attending today.

As you know, after reading the information sheet and consent form, you are taking part in a study hoping to understand how parents experience the diagnosis of cystic fibrosis for their new-born baby.

Before we start, I would like to just go over a few things (revisit confidentiality, consent/information sheets, taping of interviews, storage).

It is important that you know that you don’t have to answer questions if you don’t want. I also want you to know that these questions are not a test and there are no right or wrong answers, I just want to know about *your* experience. Please feel free to give as much detail and as many examples as you like. At the end if there are bits of the conversation you don’t want to take forward into the study, you can let me know, it’s no problem.

Do you have any questions before we start?

**Background information:**

If it is okay, I would now just like to ask some questions to get some background information

|  |
| --- |
| What gender is your child? |
|  |
| How old are they? (month of birth and year **only**) |
|  |
| What date did they get diagnosed? |
|  |
| What name or terminology do you prefer to use to refer to the diagnosis (i.e. CF) |
|  |
| How would you describe your ethnicity? |
|  |
| What is your marital or relationship status? |
|  |
| What is your employment status? |
|  |
| Does your child have any other diagnoses from healthcare professionals? |
|  |

## **Appendix H:** Interview schedule

**1. Experiences of diagnosis:**

*First, I would like to ask you some questions about receiving the diagnosis*

1. ***Prior to the diagnosis of cystic fibrosis what was your knowledge of it?***
2. ***Can you give me an idea of what the process/stages/journey of diagnosis was like?***
3. ***How did you feel in the days after diagnosis?***
4. ***Is there anything you would have changed about the diagnosis process?***

**Prompts:**

* *How would you describe how you felt at that time (Happy? Sad? Worried? Confused? Scared?)*
* *Who else was around you?*
* *Tell me more about that*
* *How was it different?*
* *Can you tell me more about that?*

***Prompt for break if required***

2. **Experiences of becoming a new parent of a child with *cystic fibrosis*:**

1. **What was it like for you when you became a parent of a child with a genetic condition such as cystic fibrosis?**

**Prompts:**

* What thoughts, feelings/emotions did you have?
* Do you think the presence of *cystic fibrosis*affected you being a parent?
* How well did you manage being a new parent?
* What did you find easy/difficult? What were the ups and downs of being a parent to a child with a diagnosis of *cystic fibrosis*?
* Was I different from what you had expected?

1. **Can you tell me about your initial relationship with your child?**

**Prompts:**

* *What was it like communicating and interacting with your child initially?*
* *What thoughts, feelings, emotions did you have?*
* *How connected (or not) did you feel towards your child initially?*
* *How well (or otherwise) did they bond with you?*
* *What was easy or difficult about building an initial relationship? How was this different from what you’d expected? Do you think having a child diagnosed with cystic fibrosis**affected your initial relationship (positively, negatively, or both)?*

***Prompt for break if required***

**3. Experiences of the social world and connection following diagnosis:**

**7. How have your family communicated with you in relation to parenting your baby?**

**8. How have other people been towards you? Friends, other medical professionals?**

**Prompts:**

* How have they reacted, treated you, perceived you?
* Have they been helpful/supportive or difficult/interfering or both?
* Did anyone make assumptions about how things were?
* Have you had peer support
* Do you think cystic fibrosishas impacted the role others have had in your relationship with your child?

***Prompt for break if required***

**Final Question**

***Is there anything else about your experiences of attending that I have not asked about that you would like to tell me?***

**Ending:**

* ***Thank participant for their time & give list of helpful services & copies of consent forms.***
* ***Ask if they would like to be notified when the final thesis is available* *and if they would like a summary of the findings.***

***Further useful examples of General Prompt and Probe Questions:***

* *What is that like?*
* *What does that mean for you?*
* *What do you think about that?*
* *Can you tell me more about that?*
* *What do you mean by \_\_\_\_ ?*
* *How does it make you feel?*

**End of interview.**

## **Appendix I:** Analysis Evidence/Audit

Development of PETS (colours indicate different participants)

**A picture containing screenshot, rectangle, parallel, pattern

Description automatically generated**

A picture containing rectangle, pattern, parallel, design

Description automatically generated

*Development of GETS*

**A desk with papers on it

Description automatically generated with low confidence**

## **Appendix J:** Table of Personal Experiential Themes (PETS) for each participant

|  |  |
| --- | --- |
| **Participant 1** | **Participant 2** |
| Nursing style  Personal resources  Symptoms  Identity  Family support  CF emergency  Feelings  Good care experience  CF impact  Timely care  Retaining information  Positive communication  Identity preservation  Loss of contact  Relationships  Information sharing  Self-care  Social world helps  Peer support  Critical social world  Hope within difficult information  Disconnect with family  Role of hope  Life expectancy  Decision fatigue  Reassuring care  Judgment from peer support  Identity changes  Constant decisions  Life disruptive  Treating children differently  Bad Mum  Social world | Impact of diagnosis  Disconnected  Physical responses  Minimising diagnosis  Physical responses  Parental changes  Not normal parenting  Alone  Avoidance  Social world doubtful  Family support  Expectations of parenthood  Not part of the care  Bad care experience  Seeing children differently  Family helping  Compassionate healthcare  Building healthcare relationships  Diagnosis conversations  Wrong timing  Providing hope  Choice  Expertise developed  No voice  Responsive care  Clash with cultural norm  Self blaming  Bonding impacted  Normality disrupted  Social world not understanding  Scared of how others view you  On guard |
| **Participant 3** | **Participant 4** |
| CF is dangerous  Care experience  Uncertainty  Searching for information  False hope  Outdated information  Professionals hold power  Absence of knowledge  Complex compassion  Hope  Direct and constant care  No control  Benefits of hope  Relationships are formed  Family tradition  Identity as a Mum  Social world perception  Damaging visuals  No control  Altruism  Significant change  Different version of motherhood  CF is a threat  Finding the balance  All on Mum  Judgement  Sadness  Lost futures  Bonding  Future focus of plans  Not feeling worthy  Comfort can’t help  Indentity  Bad experiences  Positive stories equal hope  Hope reduced  Behavioural changes | Development of CF skills  Lack of up to date knowledge  Hope within information  Personal resources  Surprise of CF  Significant emotions  CF news  Shock of visit  Good care  Asking for more information  Social world support  Searching online for information  Identity as a Mum  Self-blame  Compassion from CF team  Bad news  CF care trusted  Limited knowledge early on  Impact of diagnosis  Unhelpful behaviours  Loneliness  Difficult care  Seeking hope  Uncertainty  Complex decisions made  Untimely care  Peer support  Social world giving information  Never ending care  Keeping values close  Sharing with others  Mental health consequences  Guilt  Enormity of condition  Social world not seeing reality  Family traditions  Safety outside  Negotiation to go outside  Not in control  Decisions  Family support  Peer support  Fear of judgment  Trying your best  Social world judges  Expectation of parenthood  Identity |

|  |  |
| --- | --- |
| **Participant 5** | **Participant 6** |
| CF is serious  Expectations change  Poor peer support  Impersonal communication  Daily life affects  Feeling alone with news  Blunt direct language  Desperate for information  Power in conversation  Not knowing information  Carrying on  Identity as a Mum  Personal resources  Sparse support  Sharing feelings  Connecting with professionals  Information sharing needs  Bonding  Treatment as an indicator  Connection  Losing connections  Social world  Others judgement  Advantages to CF  Social opportunity  Different instincts  Role as a Mum  Social world unsafe  Role as educator  Parental changes with other siblings  Hopeful stories  Peer support avoided  Aware of death  I’m alone with caring  Power in healthcare | Significant emotion  Unable to describe  Lack of care  Physical responses  Not believing  Emotion  Social support  Blunt care  Family support  Why us  Sadness  Unexpected feelings  Self-blame  Anger  Personal resources  Changeable emotional states  Social world connection  Stories of hope invited  Questioning everything  Loss  Seeking control  Requesting more information  Healthcare style  Uncertainty  Good care experience  Power  Different with other children  Different parenthood  Rejecting self  Emotions towards child  Nobody understands  Wider world problems  Lack of listening  Advocacy |
| **Participant 7**  Emotions with news  Personal resources  Worry  Unknown element of CF  No control  Family support  Friendly care  Expectations changed  Not hearing information  Compassionate care  Guilt  No logic  Family support  Identity  Normal child  Others not understanding  Lockdown impact  Lost friendships  Decision maker  Educating others  Responsive care  Others judgement  Always learning  Relationships different  Lack of control  Bonding  Positivity  Consistent care  Experience of psychology  Peer support  Fearful of negative judgement | **Participant 8**  Limited knowledge of CF  Compassionate care  Information sharing  Upset  Reassurance  Learning  Power imbalance  Worry  Judgement  Guilt  Unable to access care  Family support  Infection control  Mental health  Social media  Peer support  Social world mixed  Stories of hope  Intuition |

## **Appendix K:** Examples of additional themes to illustrate themes

|  |  |  |
| --- | --- | --- |
| Group Experiential Theme | Group Level Subtheme | Additional examples of quotes to illustrate them |
| The moment that changed everything | Immeasurable distress | *“Between that call and her arriving, just frantically Googling everything. Which is probably one of the worst things we could have done” (P3)* |
|  |  | *“So I just, if I could change anything it would be that because I don’t think she should have told me over the phone. Erm I was on my own…” (P.5)*  *yeah you are clutching at straws and you are trying to find one little bit of silver lining to soften the blow (LAUGH) that things might not be as, you know, bleak as you first worry that they will be, so I think yeah, I think that’s how I spent that day. (P4)* |
|  |  | *“Erm, a lot really, so anger was a big one, why is this happening to me, you know that sort of title” (P6)* |
|  |  |
|  | I’m powerless | *“That was the worst moment, just like that split second, because she answered me straight away, but it was that split second of waiting for an answer and if she’d go, or like pull an expression that might suggest yes, cos obviously I didn’t know anything, erm but yeah” (P3)* |
|  |  | *“I do still feel powerless to like faits or some higher power, I don’t know, it just didn’t feel fair and I did feel powerless still” (P6)*  *“That was the worst moment, just like that split second, because she answered me straight away but it was that split second of waiting for an answer and if she’d go, or like pull an expression that might suggest yes, cos obviously I didn’t know anything, erm but yeah, I had no power in what she was about to say” (P3)* |
|  |  | *“Erm I can remember NAME and NAME – (staff) literally being like this is the most exciting year we have ever head, and they literally used the word exciting, like “exciting, there is nothing exciting about this”, like her daughter died in her 20’s” (P1)* |
| Parenthood changed forever | The everchanging identity of ‘Mum’ | *“I would never have batted an eyelid with NAME”-(other sibling) (P1).* |
|  |  | *“I’m worrying as well, what do those other mums think of me in the way that I’m trying to do things and I’m trying to keep her best interests at heart but being an over thinker” (P4).* |
|  |  | *“whereas I’m constantly being knocked off my horse being like ‘oh god no, oh crap mum, I was about to take you swimming” (P2)* |
|  |  | *“I've become obsessive with certain things, erm cleaning and anything to do with stagnant water and soil and dirt, like water in particularly, really freaks me out, but I don't think I've learnt of that until a little while after when I was talking to NAME-(CF staff)” (P3)* |
|  | This isn’t what I expected | *“You see that was horrendous because you look at your four-week-old baby and you flick back to what you remember and you just think they don’t go together, the pictures don’t match” (P5)*  *“I think I thought when we bought this ball pit that we felt like we were depriving her of soft play (LAUGH), and the balls can go in the dishwasher and the mat can go in the washing machine, so it’s as hygienic I think as soft play can be at the moment” (P4).*  *“I’d just fallen in love with this amazing human being and I knew what that was like because I’d already had NAME-(daughter), so I can imagine it’s a little bit worse for new parents but I’d already dealt with those emotions before but just such a disappointing feeling, just such a distress, not disappointment, what’s the word? I guess it is disappointment” (P6)*  *“I feel most sorry for cos she were really happy as an only child (LAUGH), not only did she get a brother but she got a brother with extra complications, which must be really, really hard to succumb” (P1)* |
|  | It’s my fault | *“we were carrying this and give her this and made her have it, I was very upset” (P7)*  *“and then I had the sort of feelings of guilt, well its my fault for either being pessimistic and attracting this into my life” (P6)*  *“I get terrible guilt if she does, and then you’ve got to live with that” (P4)*  *something wrong with you if you like, even though being a carrier wouldn’t have any health implications for us personally, I felt a little bit almost hurt to hear that I had a faulty gene of some sort (P4)*  *“felt I was to blame, and all of those feelings, I remember saying it” (P8)* |
| The social world | Is there anybody who understands? | *“now I’ve got to decide where I’m gonna stand, am I gonna be the person that disinfects the swing before I put her in it (LAUGH), or you know and then face judgement by people in the park looking on thinking what on earth is she doing” (P4)*  *“I wont take him to one of my friends houses because I don’t think she cleans it very well, so he doesn’t go, I make up every excuse in book, or we go somewhere or she comes to mine” (P1)*  *“ they don’t seem to understand what we go through on a daily basis, the care, everything, and they don’t seem to take it as serious as.. like there are things that you have to avoid” (P6)* |
| Impossible without support | Stories of hope | *“but yeah I think because they covered the fact that CF kids are not actually lining up for lung transplants anymore, they’re not sitting in wheelchairs, they are not, do you know, skinny and sickly, the medication’s come on so well, because I had those answers I just said “right, this is what we’ve got to do, lets crack on with it then” (P5)*  *““I think we presumed initially that that would be the case for NAME-(daughter) [bad outcomes with life expectancy] but fingers crossed it sounds like it might be a brighter picture” (P4)*  *“I know that hearing about others has helped” (P7)*  *“Obviously he’s got CF, but he doesn’t look like he’s got CF, he’s managed to have his kids, his twins naturally, he’s got a high-powered job, he does his exercise now and he’s younger than me. He’s not dropping off the edge of the earth, you know, he’s not on deaths door” (P5)*  *“It was the life expectancy figure… as soon as I knew more, it was better, I felt more hopeful whereas I had felt… I think…. I don’t know… like nothing was ever going to be better again (P8)* |
|  | Family is so important | *I think he was saying really supportive things like “it will be fine, we will work through this”, you know, “it will all be okay” (P4)*  *“because me and my nan, like growing up, we’ve always said everyone should aim for 100, life’s too short” (P3)*  *“we were talking to Mum and Dad and they kept a diary of events that I don’t want to see yet” (P2).* |
|  | I’m communicated with | *““she was absolutely fantastic, she was really, really kind and considerate and very, very warm gentle nature in the way that she was speaking to us and she gave us plenty of time, I mean she did it perfectly” (P4)*  *“they have been so clear with us, the information from that point has been fantastic” (P8)*  *“And he was, he was really good bless him, he answered all our questions and he wasn’t, you know he didn’t like, “yes he will definitely be old” but kind of gave a bit of hope but I think there was a good balance between them all to be fair, I do think I don’t reflect on that negatively” (P3)*  *“They talk you through everything in detail” (P1)*  *“finally, people who get it” (P7)*  *“The Consultant was very direct but then we saw a couple of the nurses and the Dietician and they were less formal, and that works for me, I like to speak to people like I’ve known them for years because then everybody’s comfortable” (P5)* |

1. Trainee Clinical Psychologist with experience of qualitative research [↑](#footnote-ref-1)
2. Clinical Psychologist with experience in qualitative research [↑](#footnote-ref-2)
3. Email contact with South et al. (2022) was made to discuss the discrepancy related to participant sample (N=12) and the reported number of ethnicities (N=16) in the paper. South et al., (2022) reported race and ethnicity as separate categories. For example, a participant could report both of White race and Hispanic/ Latina/o ethnicity. The number of participants in the study is N=12.   [↑](#footnote-ref-3)
4. Email contact with Vion Genovese et al. (2021) was made to establish the breakdown of parents’ gender. Unable to contact the author due to the email address listed on the paper no longer being in use. [↑](#footnote-ref-4)
5. Trainee Clinical Psychologist with experience in qualitative methods [↑](#footnote-ref-5)