An exploration of the experiences of cystic fibrosis in family systems

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The candidate confirms that the work submitted is her own and that appropriate credit has been given where reference has been made to the work of others.

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Abstract

Introduction: Whilst Cystic Fibrosis (CF) treatment has recently been transformed with developments in modulator drugs (De Boeck, 2020), it remains a serious genetic condition (Bowen & Hull, 2015). Due to the new CF landscape, it is important to develop an up-to-date understanding of CF experiences. Since working together with whole families is key to improving care (Hisert et al., 2023), an understanding of experiences at a family-level was sought. As family-level research is less common, the methodology was also reviewed.

Methods: Four families with a child with CF were recruited and asked to provide creative items to depict their experiences of CF. Family-level interviews were conducted and the creative items provided by the family were used as stimuli in that family's interview. Interviews were analysed with Interpretative Phenomenological Analysis (IPA).

Results: Results showed that the diagnosis and subsequent lifestyle changes required can feel overwhelming. Families need to negotiate how they live with CF, which can look different for each family. As the child develops or health needs change, renegotiations are needed. Children with CF stand out from their peers. Additionally, fear exists around the child becoming unwell and things becoming overwhelming again.

Discussion: This study supports models of family adaptation to childhood chronic illness but emphasises that the process is not linear. CF clinics should consider how they communicate the initial diagnosis and pacing the information they provide. Holistic support from CF clinics and peer support from older families with CF may help newly diagnosed families. In terms of the methodology, interviewing the family together created a supportive environment but affected what was said. The creative items gave the children power, but this was influenced by pre-existing family dynamics. Future family interviews should consider the location, length and timing of the interview as well as how the creative items are presented.

Table 1. *Acronyms used in the study*

Acronym	Definition	First mentioned
CF	Cystic Fibrosis	Aims
FwCF	Families with Cystic Fibrosis	Introdution
CwCF	Child with Cystic Fibrosis	Introduction
PoCwCF	Parents/Carers of a Child with Cystic Fibrosis	Introduction
SoCwCF	Siblings of a Child with Cystic Fibrosis	Introduction
VEI	Visual elicitation interviews	Introduction
CFTR	CF transmembrane conductance regulator (protein)	Introduction
NBS	Newborn bloodspot screening	Introduction
NHS	National Health Service	Introduction
SES	Socio-economic Status	Introduction
TSC	The transactional stress and coping model	Introduction
RMFAA	The Resiliency model of Family stress, Adjustment and Adaption	Introduction
FAAR	The Family Adjustment and Adaptation Model	Introduction
GET	Group Experiential Themes	Method
AATM	Adjustment Across Time Model	Discussion
ACT	Acceptance and Commitment Therapy	Discussion

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Aims

In this study, we aimed to achieve the following:

Aim 1 – Develop an understanding of experiences of cystic fibrosis (CF) in family systems.

Aim 2 – Reflect on the feasibility of using a novel creative qualitative methodology with the whole family.

Note from Author

Throughout this document, reflexive boxes provide insight into my thinking. To be consistent with what was asked of participants, I will use photos in these boxes to visualise my reflections. Below is a list of where these boxes can be found.

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Chapter One: Introduction

My primary aim was to explore the experiences of Cystic Fibrosis (CF) in families. Thus, after discussing the type of language used throughout this thesis, I will introduce CF before considering medical advancements in its care. I will then describe other external factors that may influence CF experiences, namely socio-economic status and Covid-19. The family system likely also plays a large role within CF experiences. I will consider the definition of a family in detail before discussing models of adjusting and adapting to childhood chronic illness. I will then provide a review of the current literature around how different family members may adapt to CF. I will end this first part of the introduction by introducing how this study hopes to advance current understandings of CF.

The secondary aim of this research was to reflect on the feasibility of using a novel creative qualitative methodology, namely visual elicitation interviews (VEI) with the whole family. I will first introduce the current literature related to VEI and conducting whole family interviews. I will then discuss why it is important to reflect on the feasibility of this methodology.

Language Used

It is important to carefully consider language used in order to respect people and families living with a chronic condition. When referring to someone with a disability or a chronic condition, there is the option of either using identity first (e.g. disabled person) or person first (e.g. person with disabilities) language (Grech et al., 2024). Preference for the type of language used seems to vary across individuals but as a group, those with invisible health conditions (such as cystic fibrosis) seem to prefer person-first language (Grech et al., 2024). Thus, person first language will be used throughout this thesis (e.g. families with cystic fibrosis [FwCF], children with Cystic Fibrosis [CwCF] and parents/carers of children with

Cystic Fibrosis [PoCwCF]). In the literature, siblings without a diagnosis of Cystic Fibrosis are sometimes referred to as 'unaffected siblings' (e.g. Chudleigh et al., 2019). However, this does not make sense from a systemic perspective which sees everyone in a system as being affected by everyone else and everything in the system affecting everyone (Gilliss, 1983). They will therefore be referred to as siblings of a child with Cystic Fibrosis (SoCwCF). When discussing the recruited participants, family members of CwCF will simply be referred to as parent/carer or sibling, for example, as it can be assumed that they are parent/carer(s) or siblings of children with CF. No one will be referred to as a patient because we are exploring daily life, which is greater than simply the times individuals may be seen in clinic or hospital settings.

Families with Cystic Fibrosis, within the regional centre I recruited from, have shared that they prefer the term 'variations' to 'mutations' when referring to changes in the person's genotype. This is because of the negative connotations associated with the term 'mutation'. I will therefore use the term 'variations' in this thesis.

What is Cystic Fibrosis?

Cystic Fibrosis (CF) is an autosomal recessive, life-limiting condition (Bowen & Hull, 2015) caused by variations on the CF transmembrane conductance regulator (CFTR) gene (UK CF Registry 2023). This gene was discovered in 1989 (Bowen & Hull, 2015) and contains the data to make CFTR proteins. Since CFTR proteins help manage the movement of water and salt throughout the body (UK CF Registry 2023), multiple organs are affected when the gene is not working properly (Quinton, 1990, see Table 2). Table 2 highlights that there are a range of challenges which individuals with CF may face (Bowen & Hull, 2015), which can have a large effect on their wellbeing (Gonzalez et al., 2023).

Table 2. *Effect of Malfunctioning CFTR Proteins on the individual*

Effect of malfunctioning CFTR proteins	Consequence for the individual
Reduce the clearance of airway surface pathogens and bacteria may become stuck	Cycles of infection leading to lung damage
Affect the reabsorption of chloride and sodium in sweat glands	Salty sweat. Individuals may lack salt in their body, particularly when they are infants or when exposed to hot weather
Pancreatic enzymes may not reach the gut	Digestion is affected, leading to malnutrition
Changes in gut microbiota and small intestine bacterial overgrowth	Chronic inflammation and reduced immune function in gut
Obstruction of bile ducts	Liver disease

Note. Information adapted from Bowen and Hull (2015)

In terms of the history of CF, European folklore from the middle ages states "woe to the child who tastes salty from a kiss on the brow for he is cursed and soon will die" (Welsh et al., 2001, p. 5121). The first reliable written record of CF dates back to 1595 in which a "bewitched" child is described as having a "swollen hardened gleaming white pancreas" (Littlewood, 2007, p. 3). Since then, there have been many reports of CF and infant deaths related to CF (Littlewood, 2007). However, the life expectancy of people with CF has greatly increased over the years (Elborn, 2016) and recent advances in modulator therapies have transformed life with CF for many (Gifford et al., 2020).

In the past, it was believed that CF largely occurred in populations of European heritage, but evidence now shows that it presents across diverse groups (De Boeck, 2020). However, a lack of newborn screening and high infant mortality reduces the likelihood of a CF diagnosis being made in certain countries and thus the data available on CF is skewed towards populations of European heritage (De Boeck, 2020). Specifically, whilst 162,428 individuals worldwide are thought to be living with CF, only about 65% are thought to have a diagnosis (Guo et al., 2022).

As these numbers are small when considering the world's population, CF is classed as a rare condition (Chen et al., 2021). Thus, CF registry databases play a crucial role in bringing data together from different CF centres to improve the lives of those with CF (Jackson & Goss, 2018). In 2022, 10251 individuals were on the UK CF Registry database, 4226 (41.2%) of whom were under 18 years of age (UK CF Registry 2023). 94.6% of these individuals were white, 3.1% Asian and 0.9% black (UK CF Registry 2023). Whilst these numbers may be slightly affected by minoritised individuals being distrustful of medical systems (Webb Hooper et al., 2019), the register contains data on over 99% of people with CF in the UK and thus can be considered reliable (Keogh et al., 2022). The large proportion of white individuals on the UK CF Register makes sense as 82% of people in England and Wales are white according to the 2021 Census (Office for National Statistics, 2021)

Nearly 2000 variations of the CFTR gene have been reported, however most are uncommon and do not cause CF (Bowen & Hull, 2015). The most common variation in CF is the F508del variation (Kerem et al., 1989; UK CF Registry 2023). Different variations are found more commonly in different ethnic groups, with the most common variations being seen largely in populations of European heritage (Bobadilla et al., 2002). This causes difficulties for screening programmes (Bobadilla et al., 2002) and research into effective treatments for all (McGarry et al., 2022). For example, in America, 95.2% of White patients are able to use modulator therapy due to them having a certain class of variation whereas only 69.7% of Black or African Americans have a class of variation which currently can be treated with modulators (McBennett et al., 2022).

Medical Advancements in CF

I will now consider the medical advancements in CF care that have helped change the landscape for individuals and families with CF. I will discuss newborn screening, symptomatic treatment and the development of modulators in turn.

Newborn Screening

Newborn bloodspot screening (NBS) involves a clinician pricking the baby's heel to collect four drops of blood within the first weeks of life (NHS, 2021). NBS for CF was first introduced in certain areas in England and Northern Ireland in the 1980s (Schlüter et al., 2020). However, it was not until 2007 that it was rolled out across the whole of England (Schlüter et al., 2020).

NBS for CF has improved outcomes as diagnosis is made before significant lung damage has time to occur (Sims et al., 2007). Schlüter et al. (2020) explored longitudinal UK data on the lung functioning and weight for age of children with CF born between 2000 and 2015. They showed that NBS increased weight for age, improved lung function and delayed onset of Pseudomonas aeruginosa infection at one year of age. This is important as Pseudomonas aeruginosa is an indicator of poor survival in CF (Henry et al., 1992). Whilst NBS did not seem to reduce the effect of deprivation on health outcomes early in life, it did seem to have a positive influence on the weight of children living in deprived areas (Schlüter et al., 2020).

Symptomatic Treatment

There are several different daily symptomatic treatments for CF, which can all be demanding. They include chest physiotherapy to help clear the airways, inhaled therapies, pancreatic enzyme replacement therapies (e.g. Creon) and supplements (Boyle et al., 2019). Physical

exercise can also be part of the treatment routine for CF (Palamut, 2023). Wotton (2014) describes in his book how it is exhausting having to do so many different treatments just to stay alive whilst the outside world largely knows nothing about them. Recently, people with CF (n=189) and/ or their carers or friends (n=452) reported to be on a median of 10 therapies for CF taking up to a median of two hours to complete each day (Davies et al., 2020). This highlights the high treatment burden.

Chest physiotherapy can include 'patting' or 'percussion' usually from a relative, which is effective at eliminating excess sputum from the lungs. However, it can be difficult for both the person delivering and the person receiving it (Palamut, 2023). Inhaled therapies use nebulisers that consist of many separate parts, which must be joined correctly together before use. Modern nebulisers are small and easy to use but all the individual parts need to be cleaned before they can be used again and must be transported if the family is going out (Palamut, 2023), requiring sometimes complicated logistics. Further, the administration of Creon requires calculating the required dosage based on how much fat has just been consumed, which is not always clearly stated on food items. Taking too much or too little Creon can lead to stomach pain and thus it can be a difficult but important calculation (Egan et al., 2022).

The Development of Modulators

The treatment landscape of CF has recently been transformed with the development of modulator drugs, which are highly effective (De Boeck, 2020). However, it should be noted that not everyone is able to take these drugs (Bierlaagh et al., 2021). Specifically, whilst the newest modulator drug roll-out is thought to enable 95% of people with CF in the UK to access the drugs due to their specific CFTR variations (NHS England, 2025), some of these individuals are intolerant to the drugs and thus unable to take them (Urbantat et al., 2025).

Additionally, the number of people with CF unable to take modulator drugs due to their specific CFTR variation is greater in other countries such as Turkey (Çobanoğlu et al., 2020) and in many countries, they are not available due to cost.

I will now briefly discuss the development of modulator drugs. Ivacaftor, the first modulator drug developed, was approved for use in 2012 (Burgener & Moss, 2018). It allows chloride to move across the cell membrane (Cystic Fibrosis Foundation, 2024). However, monotherapy (e.g. just Ivacaftor) is ineffective in most CF patients (Purkayastha et al., 2023). Correctors, such as lumacaftor, tezacaftor and elexacaftor were then developed, which improve the shape of CFTR proteins (Cystic Fibrosis Foundation, 2024). Using a corrector alongside Ivacaftor was found to be more beneficial than Ivacaftor alone (Purkayastha et al., 2023). Still triple therapy, in which Ivacaftor is taken with two correctors, is most effective as it enables the targeting of different sites (Purkayastha et al., 2023). Triple therapy, e.g. Kaftrio®, was first approved by NHS England in August 2020 (NHS England, 2020). This was initially for those 12 years and older but is gradually being introduced to children at younger ages (Medicines and Healthcare products Regulatory Agency, 2023). Alyftrek® a new triple therapy drug was just approved as I was finishing this thesis (NHS England, 2025).

People with CF, who are taking modulator drugs, can now expect to live longer, healthier lives (Nichols et al., 2021). The impact of modulators may be even greater in children, who have less lung damage before they start taking them (Nichols et al., 2021). Due to the effectiveness of modulator drugs, some of the more onerous treatments, such as chest physiotherapy may be able to be stopped (Rowbotham & Daniels, 2022). However, as recommended by the National Institute for Health and Care Excellence (NICE, 2024) guidelines, more research is needed into this and thus people with CF, who are taking modulator drugs, are still being advised to complete the demanding treatments. As modulator drugs improve fat absorption, new challenges such as obesity are emerging in the CF

population (Gabel et al., 2022). Thus, as well as improving CF related health outcomes, the drugs may also require lifestyle changes for those taking them to remain healthy (Snowball et al., 2023).

External influences on life with CF

We have now briefly considered the medical world of CF. However, it is not only the medical landscape that influences someone's experiences of CF. In this section, I will explore how socio-economic status and Covid-19 may play a role in CF experiences. The family system may also play a large role and will be considered in detail in the following section.

Socio-Economic Status

Early psychosocial experiences can influence health outcomes in children with chronic diseases (Danese & McEwen, 2012; Taylor et al., 2011). This is no different for CF. For example, studies before the development of modulators showed a relationship between poor socio-economic status (SES) and death (Britton, 1989; O'Connor et al., 2003; Schechter et al., 2001). This relationship was thought to be due to increased exposure to smoke (Rubin, 1990), poorer diets (Schechter et al., 2001) and poorer treatment adherence (Walters et al., 1994), which could be due to the family having less social and financial resources. Exposure to tobacco may continue to have a large effect in the modulator era as it may reduce the effectiveness of modulators (McGarry et al., 2022).

Engaging in physical activity and eating appropriate diets can vary across social classes (Groeniger et al., 2019). This is potentially even more significant for people from the CF community as engaging in physical activity is an important part of CF management (Denford et al., 2020) and will likely remain so in the modulator era (Rowbotham & Daniels, 2022). However, cost has been listed as a barrier to CwCF engaging in physical activity (Denford et

al., 2020). Additionally, parental support can play an important role in adolescents in general exercising and can be affected by social class (Groeniger et al., 2019; Raudsepp, 2006). Parents from higher socio-economic classes may have more favourable opinions towards physical activity themselves (Raudsepp, 2006) but may also simply have the resources to be around more and thus encourage it. Cost is also associated with the high fat, high energy diets that people with CF have traditionally been put on (Collins, 2018). Whilst dietary advice is changing due to modulators improving nutritional status (Wilschanski & Peckham, 2022), it is important to acknowledge that it could have caused much stress in families already experiencing food insecurity. Moreover, not everyone is currently able to take modulator drugs.

Whilst access to treatment is not an issue in the UK due to the National Health Service (NHS), the presence of CF in a family can still present a considerable financial burden. For example, mums of children with CF often do not return to work, likely due to the high treatment demands (Douglas et al., 2016). This could be particularly difficult in the cost-of-living crisis that the UK is currently experiencing (Keith Neal, 2022).

Covid-19

The Covid-19 pandemic was challenging for everyone. However, those with CF were considered to be more vulnerable due to them tending to experience worse viral respiratory tract infections with a greater risk of complications (Colombo et al., 2020). Despite this, data suggests that the presence of CF did not impact on the severity of the Covid-19 virus (Colombo et al., 2020). Nevertheless, the vulnerability status at the time likely led to increased stress for families and individuals alike (Colombo et al., 2020).

Collaço et al. (2021) surveyed 145 PoCwCF and 99 children and young adults with CF to explore the impact of Covid-19 on their lives. Disruption due to having to shield was a key

theme identified. During the pandemic, the participants shared that they worried about never being able to return to normal life and what they would miss out on. PoCwCF shared that they experienced anxiety around their child getting ill, whilst the children and young adults spoke about loneliness as well as anxiety around seeing others. Due to the lack of social distancing by others, FwCF generally felt more restricted to staying at home (Collaço et al., 2021), which likely further worsened the impact of Covid-19 on their wellbeing.

The Family and Childhood Chronic Conditions

In this section, I will consider the definition of a family before thinking about a model of general family wellbeing. I will then discuss models of childhood chronic illness and family adaptation. None of this section discusses Cystic Fibrosis specifically.

Definition of Family

To think about experiences of Cystic Fibrosis within families, it is first important to consider what is meant by the term 'family'. This has been something that scholars have been debating for decades (Miller, 2016). The UK census defines families as a couple living together with or without children or a single parent living with at least one child (Office for National Statistics [ONS], 2023). However, this is limited to a westernised view of a family as it does not consider how some cultures place equal importance on extended family members (Miller, 2016). Moreover, research on 'chosen families' in the 'Lesbian, Gay, Bisexual, Transgender, Queer plus other sexual orientations and gender identities' (LGBTQ+) literature challenges the need for biological or legal ties in families (Miller, 2016). Thus, as there is no universal definition of the term 'family' (Miller, 2016), this study will allow the recruited participants to define their own family. It should be noted that the models introduced below are based on a more structural westernised view.

Model of General Family Wellbeing

Newland (2015) provides a review of the literature on family wellbeing in general. They suggest that family resiliency and self-sufficiency as well as parental physical and mental health are important for family wellbeing. Family resiliency is thought to be built upon positive relationships within the family and a strong social support network within the community (Newland, 2014). Such relationships would suggest that support is available when a crisis is encountered. A bi-directional relationship seems to exist between the family's social network and parental mental health. Specifically, whilst negative parental mental health can reduce the family's social network (Newland, 2015), low social support can also influence parental mental health as they may feel like they must manage alone (Turner & Brown, 2010). Family self-sufficiency refers to how well the family can meet their own needs and thus highlights the impact poverty can have on family wellbeing.

Models of Childhood Chronic Illness

When a chronic illness is first diagnosed, it initially takes on a central role within the family (Frey III, 1984). This fits with the seriousness of the illness and the need for the family to develop new behaviours to manage it. However, it is suggested that in healthy families, the chronic illness only holds this central role temporarily (Frey III, 1984). Several models have been proposed to explore how families may adapt to a chronic illness. I will discuss the Transactional Stress and Coping Model (Thompson, 1985), The Resiliency model of Family Stress, Adjustment, and Adaption (McCubbin & McCubbin, 1993), and the Adjustment Across Time Model (Huang et al., 2022) in turn. I will use the term 'adjustment' to describe changes in the short term and 'adaptation' to describe long-term changes.

The Transactional Stress and Coping Model. The transactional stress and coping model (TSC) was initially proposed by Thompson (1985) and is derived from Bronfenbrenner's ecological systems theory (Bronfenbrenner, 1977). In the model, adaptation is considered in terms of maternal processes, child processes and the relationship between the two. Child processes include self-esteem and how much control they think they have (locus of control) whereas maternal processes include the appraisal of stress, locus of control and efficacy (Thompson, 1985). However, Hocking and Lochman (2005) reviewed the model by looking at research on sickle cell disease and insulin-dependent diabetes mellitus and found that the child's appraisal of stress may also influence adaptation.

The family dynamic is thought to be important within the TSC (Hocking & Lochman, 2005). However, whilst the family dynamic was originally seen to only impact upon maternal adaptation, it has since been shown to also directly affect the child (Hocking & Lochman, 2005; Jaser & Grey, 2010; Szyndler et al., 2005). For example, Jaser and Grey (2010) suggest that child-sensitive parenting is linked to better adaptation and metabolic control in children with type 1 diabetes. Additionally, being in a supportive and expressive family can also help siblings cope (Gold et al., 2008; Gold et al., 2011).

As well as the family context, the severity of the illness and the child's demographic factors (e.g. gender, age, socio-economic status [SES]) are thought to be important in influencing adaptation (Thompson, 1985). SES influences how well the family are able to meet their own needs as also described in Newland (2015) whilst gender and age can affect how the child responds to the illness and it's treatment regimen. Illness severity includes both physiological and functional severity. Physiological severity is the effect of the condition on the body whilst functional severity is the amount of assistance needed or the resources required by the family in caring for the person (Patterson, 2005). Research has found inconsistencies when

looking at the impact of severity, perhaps due to inconsistencies in the type of 'severity' used (Hocking & Lochman, 2005).

The TSC has been criticised for its lack of consideration of others in the system as it focuses on the mum and the child with the chronic illness (Hocking & Lochman, 2005). As people do not exist in isolation nor in isolated dyads, it is important to look at other models. Moreover, the model suggests maternal adaptation plays an important role in child adaptation despite the literature not really supporting this (Hocking & Lochman, 2005). In fact, only studies that have relied solely on maternal reports have found this to be the case (Hocking & Lochman, 2005). This highlights the limitations of relying on certain family members to speak for others.

The Resiliency model of Family Stress, Adjustment, and Adaption.

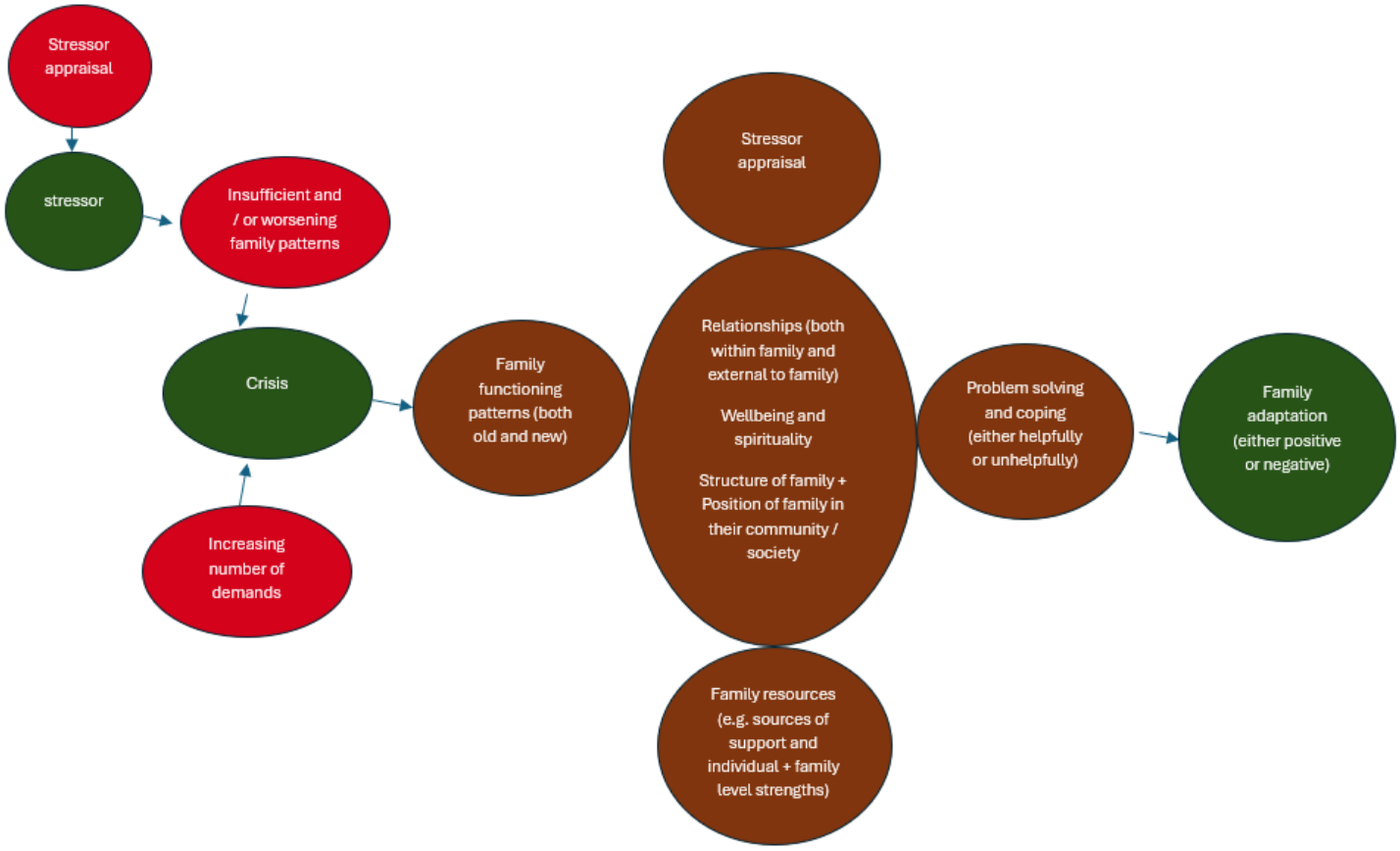
The Resiliency model of Family stress, Adjustment and Adaption (RMFAA, McCubbin & McCubbin, 1993) has evolved through several models within family stress theory (Brown-Baatjies et al., 2008). Hill and Boulding (1949) initially proposed the ABCX Model to describe the adjustment of families during and after the second world war. In this model, the event was thought to interact with the family's resources and how the family define the event to produce the crisis (Brown-Baatjies et al., 2008). However, research during the Vietnam war showed that this model missed factors that influence adaptation over time such as further stressors, new resources, the family re-defining the event and coping strategies tried by the family (Brown-Baatjies et al., 2008). Consequently, the Double ABCX Model of Adjustment and Adaptation (McCubbin & Patterson, 1983) was developed and included all the factors above (Brown-Baatjies et al., 2008). However, longitudinal studies showed that families tend to go through three stages of adaption, namely 'resistance, restructuring and consolidation' (McCubbin & Patterson, 1983), which were still missed

(Brown-Baatjies et al., 2008). Thus, the Family Adjustment and Adaptation Model (FAAR) was proposed (Patterson, 1988).

The FAAR model acknowledges that the family has likely faced previous stressors and suggests that their adaptation to these can influence their current vulnerability (McCubbin & Patterson, 1983). It suggests that when the family face a new stressor, they first enter an adjustment phase, in which they may try to draw on their own resources to reduce disruption (McCubbin & Patterson, 1983; Patterson, 1988). The resources that they draw upon can be at an individual (e.g. self- esteem, sense of mastery), family (e.g. cohesion, communication skills, adaptability) and / or a community level (e.g. health care services and social support) (Patterson, 1988). Additionally, the family may draw upon their 'family coping behaviour', which refers to both their individual and family problem solving skills (Patterson, 1988). Whilst the family may try to reduce disruption, it is likely that disruption occurs anyway because of the crisis. Individual roles of family members may change as well as their typical ways of interacting with each other (McCubbin & Patterson, 1983; Patterson, 1988).

However, the FAAR model still does not fully explain why some families seem to thrive during stressors whilst others cannot cope (Brown-Baatjies et al., 2008). The Typology Model of Family Adjustment and Adaptation (McCubbin & McCubbin, 1988) aimed to make sense of this by including social class and ethnicity as well as a consideration of the family's vulnerability, the family's usual patterns of interaction as well as the family's resources and coping skills (Brown-Baatjies et al., 2008). Although the Typology Model considers ethnicity and social class, it only considered them in comparison to families of European heritage (Brown-Baatjies et al., 2008). Thus, the RMFAA was proposed in an attempt to include diverse families (Brown-Baatjies et al., 2008). Figure 1 shows the RMFAA model.

Figure 1. *The Resiliency model of Family stress, Adjustment and Adaption*



Note. Diagram adapted from Brown-Baatjies et al. (2008, p. 112).

Whilst these models stemmed from research into families facing stressors from war, Patterson (2005) argued based on previous literature that the FAAR model was also helpful in looking at childhood chronic illness as a discrete stressor. Furthermore, the RMFA model has previously been applied to families of premature babies (Gralton, 2017). Whilst war and illness are different, the models are simply looking at family demands and resources and whether one outweighs the other as well as the meanings that families make of situations (Patterson, 2005). The models also allow for families to go through the cycle repeatedly, which will likely be the case with a chronic illness, especially as the family may face new challenges across the family life cycle (Patterson, 2005).

Adjustment Across Time. Huang et al. (2022) completed a qualitative meta-synthesis to explore family adaptation over time in relation to childhood chronic illness. They included 17 studies in the synthesis and suggest three different phases which families may go through. They named these stages: 'Disintegration and Vulnerability', 'Adjustment and Adaptation', 'Recovery and Reconstruction'.

In the 'Disintegration and Vulnerability' stage, emotions are thought to fluctuate as family routine disappears (Long et al., 2015). Interestingly, the synthesis identified differences in the psychological reactions between mums and dads. Specifically, mums were shown to experience more negative emotions, perhaps due to them taking on more responsibility in caring for the child (Douglas et al., 2016; Huang et al., 2022).

As the family get more used to living with the illness, they are thought to move into the 'Adjustment and Adaptation' stage. Across the culturally diverse studies, spirituality was identified as being important in the development of resilience in this stage (Huang et al., 2022). In line with the significance of social support as highlighted in Newland (2015) and the RMFAA model (Brown-Baatjies et al., 2008), families may also seek to expand their social networks when adjusting and adapting (Huang et al., 2022). In this stage, families are thought to find new ways of organising themselves, improve their problem-solving skills and find better ways of appraising the illness and situation (Huang et al., 2022).

Through the processes of adaptation, it is suggested that the family enter the 'Recovery and Reconstruction' stage. During this stage, the family may start to identify positive changes that have occurred due to the diagnosis, such as understanding each other better and valuing life in the present. Conversely, the experience of a chronic illness may highlight to families their differing world views, which could pull them further apart (Patterson, 2005). Additionally, it may be important to consider SES as resources and free time available to the family may

influence whether the family is able to enter this stage. Specifically, the trauma of poverty may affect families' ability to come together as a unit or identify any positive changes. Huang et al. (2022) identify that financial difficulties may play a role in the initial 'Disintegration and Vulnerability' stage. However, their paper discusses how families and friends help with this, a luxury that not all families have.

Whilst the journey through this model may seem linear, this will likely not always be the case. For example, as children grow older they often begin wanting more control over their treatment (Blakemore, 2019). This could change and destabilise the whole family's positioning around the condition. Moreover, national events, such as the Covid-19 pandemic, or individual health events, such as the child picking up an infection, may cause families to go back to an earlier stage of the model. Thus, families may jump around the model depending on stressors they are currently experiencing.

Experiences of Cystic Fibrosis

In this section, I will discuss the current evidence base on how different individuals may experience cystic fibrosis (CF). Due to the development of modulators, the experience of CF in a family has likely changed significantly. Whilst children with CF (CwCF) may be too young to consider their own mortality (Szyndler et al., 2005), modulators may provide parent(s)/carer(s) of a child with CF (PoCwCF) with increased hope. Moreover, as people with CF begin to live longer, the types of stressors they, and their family, encounter are likely to change (Bathgate et al., 2022). Consequently, research from before the wide availability of modulators may tell a different story to how life is for people with CF and their families currently. This needs to be considered when referring to the CF literature and demonstrates a need for new research.

Newborn Screening

It is not just modulators which have changed cystic fibrosis in the last two decades in the UK but also the availability of newborn screening (NBS). NBS changes the time point in which the family start adjusting to life with CF. It means that those who were diagnosed as newborns know no other way of life - living with CF is 'normal' to them (Williams et al., 2009). Younger siblings of a child with CF (SoCwCF) will also know no different than life with CF. Older SoCwCF will have been younger at the point of diagnosis, changing how much they remember about life before a CF diagnosis (Chudleigh et al., 2019).

NBS may also improve PoCwCF experience of the initial CF diagnosis (Oliver et al., 2004). Namely, if the child is diagnosed clinically later, the parent(s)/carer(s) will have likely faced a period of time in which they knew that their child was unwell but were unsure of the reason and therefore the necessary treatment. However, a review into the wellbeing of parents of children with type 1 diabetes showed that diagnosis may be experienced as stressful partly due to the amount of information, for example about treatment, that they were suddenly expected to know (Whittemore et al., 2012). Such stress can exist regardless of the age of diagnosis. In fact, it may be even harder for new PoCwCF suddenly trying to learn how to simply care for a newborn as well as about CF. Feelings of guilt, anxiety and shock can also be heightened in PoCwCF rather than with a different condition as their newborn may show no outwards signs of being unwell (Li et al., 2023) and CF is a genetic condition.

Changes in Family Relationships and Routine

Family relationships may play a critical role in family quality of life (Ridosh, 2014) and these relationships can be affected from the point of diagnosis. For example, PoCwCF may reduce daily activity out of concern for the CwCF's health, affecting how much different family members see of each other (Li et al., 2023). This change in family patterns is represented in both the RMFAA (Brown-Baatjies et al., 2008) and Adjustment Across Time (Huang et al., 2022) models and can reduce family wellbeing generally (Newland, 2015). The loss of the old family routine can be hard for everyone, including SoCwCF (Milo et al., 2021; Tay, 2021), who may feel like their needs are being neglected (Milo et al., 2021). As the family environment can be a protective factor for children adjusting to life with a chronic illness (Hocking & Lochman, 2005; Wallander et al., 1989), these changes can greatly impact upon all children in the family.

Author's Comments and Reflections 1. *Being a mum*

Whilst working on this thesis, I have become a mum to a little boy. I have reflected on how hard it is navigating the newborn and baby phases anyway. I have also reflected on how it feels like a challenge keeping them alive even without the presence of a childhood chronic illness. During the newborn phase, getting out of the house felt like a challenge but in my mock interview, the mum spoke about how she would have so many hospital appointments during the first years with her baby with CF. My little boy loves playing in mud and outdoors. He loves getting into things. I think this would be extremely challenging if I had a reason to be worried about his immune system.

Mental Health

In fact, the presence of anxiety and/or depression in CwCF and their parent(s)/carer(s) has been found to be two to three times higher than in society in general (Quittner et al., 2014). In Blair et al. (1994), mums of children with either CF or anorexia nervosa were found to be more emotionally distressed than mums of typically developing children but this was found not to be the case for dads. More recently, Douglas et al. (2016) reviewed medical records in Australia and showed that the prevalence of mental illness in the included fathers of a child with CF was lower than in the included mothers during the child's first year of life. In support of these studies, Huang et al. (2022) also found mums of children with chronic illnesses to experience more negative emotions than dads ~~of children with chronic illnesses~~. This difference between parents may be due to mums being more likely to take on the role of primary caregiver (Douglas et al., 2016; Unal Yuksekgonul et al., 2020). Taking a lead in ensuring the recommended treatments are completed can be stressful (Andrews et al., 2021). Conversely, it can also be helpful as it provides the parent/carer with a sense of being able to do something for their child (Andrews et al., 2021), which supports the idea that locus of control is important in maternal adaptational processes (Thompson, 1985). It would be helpful for future studies to consider the effect, if any, of other children in the house as parenting more than one child increases the demands on parent(s)/carer(s) regardless of the presence of a chronic condition (McClellan & Cohen, 2007).

As identified in the TSC model (Thompson, 1985), age can play an important role in the wellbeing of the CwCF. For example, as children get older, they often become more socially aware (Eccles, 1999). They may begin to realise that they are different from their peers and want to fit in (Williams et al., 2009). This may lead to worries about their physical appearance as well as how the symptoms of CF may be perceived by others (Williams et al.,

2009). Due to stress around their nutritional intake (Quittner et al., 2014), CwCF may also be more aware of calories at a younger age. This and the fact that they are more likely to have abnormal growth patterns, due to the condition and the impact of modulator drugs (Snowball et al., 2023), may be potential reasons for the increased risk of disordered eating seen in individuals with CF (Bathgate et al., 2022). Modulator drugs can also have a direct impact on mental health, though this is thought to be rarer (Spoletini et al., 2022).

Education

Children and young people spend a lot of time at school. CF can affect school attendance, consequently affecting achievement (Bowdy et al., 2023; Claxton, 2012). Not achieving their potential in school may further affect an individual's sense of self and thus their mental health. Unsurprisingly, improved health has been shown to reduce educational risk (Bowdy et al., 2023) and therefore a shift may be seen in educational outcomes as CwCF start taking modulators earlier. However, it is not just ill health that gets in the way of schooling but also the treatment routine which takes up much time. Gathercole (2019) interviewed fourteen CwCF from a large English regional CF centre alongside their parents. The interviews showed that families try to do treatments before or after school so that the CwCF did not seem different than their peers. Conversely, this led to them having to get up earlier or having less time to study after school. Sleep problems are very common in CwCF (Fauroux et al., 2021) and having to wake up early to complete treatments could compound the problem. Moreover, some of the required treatments affected the CwCF's ability to engage in things like residential trips (Gathercole, 2019), which could affect peer relationships and consequently the child's well-being.

Experiences of Siblings of a Child with Cystic Fibrosis (SoCwCF)

It is also important to consider the wellbeing of SoCwCF. Havermans et al. (2011) asked SoCwCF aged between 10 and 18 years at two CF centres in Belgium to fill in questionnaires about their quality of life and the impact of CF. Surprisingly, the 39 SoCwCF scored higher on all domains of the Child Health Questionnaire than the reference group, which was taken from a Belgium study looking at quality of life in healthy children. This could be because the SoCwCF are grateful for their own health (Havermans et al., 2011). It may also be due to the included families with CF (FwCF) being in the 'Recovery and Reconstruction' phase (Huang et al., 2022) and the CF bringing them closer together (Long et al., 2015) or making them live their lives more in the present (Huang et al., 2022). Siblings in families where the CwCF had been hospitalised rated the impact of CF as higher (Havermans et al., 2011), which fits with the TSC model (Thompson, 1985). It also highlights the need to consider physiological illness severity in research into the effects of chronic conditions on families (McClellan & Cohen, 2007).

Milo et al. (2021) included eight SoCwCF in a six month focus group. The participants were either adolescents or young adults. They spoke about feeling neglected in the family and that CF took up all the family's attention. Research with PoCwCF has supported this, as they report that they tend to give the SoCwCF less attention (Foster et al., 2001; Quittner & Opipari, 1994). SoCwCF in Milo et al. (2021) also discussed how they would try and keep CF hidden from the outside world. This fits with the idea that CwCF also want to appear 'normal' (Williams et al., 2009). It also fits with the idea in McKeever (1983) that PoCwCF give SoCwCF limited information about CF as they want to hide CF from the outside world and SoCwCF may be a means of the information getting out. However, CF at the time of McKeever (1983) was a completely different diagnosis than it is now.

On the other hand, SoCwCF in Larocque (2006) spoke about how they received less sympathy from others due to CF being an 'invisible illness'. This fits with CwCF feeling like their physical difficulties are not understood, and that they receive less support due to their symptoms being invisible (Claxton, 2012). Thus, whilst CwCF and their siblings want to be seen like everyone else, they still at times wish for some acknowledgements of the difficulties that CF causes. In fact, invisible conditions are thought to negatively influence adaptation more than visible conditions due to the uncertainty associated with whether or not to acknowledge the condition in everyday life (Jessop & Stein, 1985).

Daily Treatments

A daily difficulty of living with CF is the demanding treatment regimen (Boyle et al., 2019). Adherence to treatment may depend on both the FwCF's relationship with the health professional and the relationship between the CwCF and their family (Williams et al., 2007). These relationships likely affect how much everyone feels heard and in control (Butow et al., 2010; Stein et al., 2019). For example, effective communication between the clinician and FWCF helps develop trust and creates space to think about how either the treatment can be incorporated into family life or how family life can be altered (Cooley et al., 2018). The more everyone feels like they have some control over the treatment, the more positively CF is likely to be viewed (Huang et al., 2022; Thompson, 1985).

Controllability for the CwCF likely changes in importance and meaning as they develop. Adolescence, in particular, is an important developmental process in which the individual becomes an independent adult (Blakemore, 2019). They may start using their developing sense of independence to say 'no' to treatment routines (Grossoehme et al., 2014), potentially due to them trying to make time for their peers and not be different (Dziuban et al., 2010; Gathercole, 2019). In the adult literature, the importance of shared power in the clinician-

patient relationship is acknowledged and this is likely also needed within the home of the young person (Williams et al., 2007). However, the transition from full parental control and responsibility to the PoCwCF passively supervising is difficult for PoCwCF as they worry about whether or not their young person is doing the treatment well (Williams et al., 2007).

This worry may have some validity as young people with CF in Dziuban et al. (2010) indicated that they believed it was okay to miss some daily treatments. This was especially true for males, who have been shown to engage in more risky behaviours generally (Reniers et al., 2016). Worryingly, if their health was poorer, participants in Dziuban et al. (2010) were more likely to see treatment as limiting their freedom and were more likely to view their clinician as unempathetic (Dziuban et al., 2010). These factors may be related as if the young person is not following what their clinician is telling them to do, they may feel less supported by their clinician and they may notice the effects of treatment less. It is also understandable that if the young person is more ill, they may feel like they have less control over CF and thus that treatment is pointless (Hocking & Lochman, 2005). Another large barrier towards treatment adherence identified in Dziuban et al. (2010) was forgetting or misplacing medications.

The Current Study

Previous research has largely focused on individual family members and their experiences of CF. However, family systems theory argues that all members of a family are connected to each other, so that what happens to one affects all (Segrin & Flora, 2018). Moreover, the family environment is considered to be critically important for healthy development (Prieur et al., 2021; Ridosh, 2014) and people with chronic illnesses likely do better if they live within a healthy family system (Patterson & Garwick, 1994b). Thus, it is important to support families to develop helpful coping strategies and adaptational processes as a unit (Prieur et al., 2021).

Indeed, Hisert et al. (2023) argue that working together with CwCF and their families is key in improving clinical care. Moreover, the world of CF has changed massively in recent years and thus older studies that sought to understand life with CF may be out-of-date. This study therefore aimed to understand the current experiences of CF in family systems by asking family members to bring creative items to the interview and make sense of their experiences of CF together.

The secondary aim was to reflect on the feasibility of using a novel creative qualitative methodology with the whole family. I will introduce this aim in the following section.

Reflecting on the methodology

To introduce the secondary aim, I will first introduce the reasons for interviewing the FwCF as a unit and then discuss visual elicitation interviews (VEI), the creative methodology employed in this study. I will end this section by reflecting on why it is important to evaluate this methodology.

Interviewing the Family as a Unit

It can be argued that we experience things in the world through our relationships with others. Thus, our experiences and the meanings that we make of them are intersubjective (Larkin et al., 2019). Indeed, Heidegger argues that we are embedded in the world (Heidegger, 1962). Therefore, simply studying the individual meanings made by one person can restrict the understanding developed (Larkin et al., 2019).

As we primarily aimed to understand the experiences of CF in family systems, it made sense to consider the family as the unit of analysis. Interviewing members of the family together helps develop an understanding of the shared family narrative (Ummel and Achille, 2016). Specifically, the family can be seen as a complex unit that is “unique from the sum of its

parts" (Gilliss, 1983, p. 51). When the family is making meaning from their experiences, the narrative moves away from the individual and becomes a collective story (Koenig & Trees, 2006). Indeed, people may disclose different things when interviewed individually but joint interviews allow them to add to or challenge each other's reports, providing a different interpretation that is inaccessible otherwise (Taylor & De Vocht, 2011). Thus, whilst many studies have tried to get a multi-perspective understanding by interviewing individuals and then comparing the accounts at a family-level (Larkin et al., 2019), the understanding constructed from this is likely different than the understanding constructed at a family-level.

Moreover, there are inherent ethical difficulties in interviewing families or couples separately as they may recognise each other in the transcripts and thus gain an awareness of what was said (Larkin et al., 2019). In addition, they have no control over what the other member shares. It has also been acknowledged that even purely individual interviews can affect the family, for example through increasing distress within the system (Polfuss et al., 2023). Nevertheless, different ethical concerns also exist in joint interviews, as someone may hold all the power, preventing other's from sharing their narrative (Ummel & Achille, 2016). This worry about power made Cave (2022) cautious about involving parents in the interviews they conducted with children. They thus gave their child participants, aged between 7 and 11 years, the choice. Notably, 70% of the children interviewed chose to be interviewed jointly with their parents. The other 30% chose for their parents to be present in the background rather than somewhere else. Whilst Cave (2022) did not intend on doing family-level interviews, their study suggests that children in this age bracket may want their parents present in research interviews.

Visual Elicitation Interviews

Visual elicitation interviews (VEI) refer to when a visual stimulus is used to aid the interview discussion (Pauwels, 2015). The term has been proposed as an umbrella term to capture all types of elicitation methodology, including photo and object elicitation methods, as there is a lack of agreed upon terminology and it is arguably unhelpful to have different names for each possible type of stimuli (Pauwels, 2015).

VEI can be particularly beneficial as individuals, especially children, are not always used to or comfortable with lengthy verbal exchanges about an abstract topic (Barton, 2015) but may be more used to using visual methods (e.g. photos or drawings) to document their day. Additionally, VEI allow for spontaneous meaning making rather than the participant telling the same verbal story that they are used to telling about their condition (Willig, 2017). Moreover, if the participant is bringing the item themselves, then it increases their power in the interview as they decide what is discussed (Ford et al., 2017). This was something that one of the child involvement partners particularly liked in the mock interview (see Appendix 1). Nevertheless, those who generally experience less power, such as children or individuals with disabilities, may still struggle saying ‘no’ or expressing their real views (Whiting, 2015).

Whilst this type of interview can be enjoyable (Ford et al., 2017), it can also lead to strong emotional responses. For example, images can have greater power at bringing back memories than verbal responses alone (Copes et al., 2018). Moreover, if the participant is bringing the item themselves, they will likely need to engage cognitively when selecting or creating it (Copes et al., 2018), which requires time and may be painful. However, the fact that the participant gains power to direct the interview and share their own story is thought to outweigh the time cost it requires. Additionally, talking about and thinking through painful experiences can have a cathartic effect (Biddle et al., 2013). Further, participants often value

being able to contribute to research and benefit their community (Biddle et al., 2013). It is important that children, as well as adults, are given the opportunity to share their own experiences (Guell, 2007) and VEI can help children engage in the process (Barton, 2015).

Use of a Novel Approach

The current study used a novel method of interviewing families as a whole unit with VEI. As far as we are aware, no other study has interviewed families with CF (FwCF) as a whole unit. Traditionally, it is more common to interview individuals (Eggenberger & Nelms, 2007) and a current literature search mainly finds individual or dyadic interviews when family interviews are searched for.

Eggenberger and Nelms (2007) did interview family members together to understand their experiences of having a critically ill family member. Whilst they did not exclude young children, their youngest participants were 13 years old. They used a semi-structured interview design but did provide pens and paper for the adolescent participants so that they could engage in more creative methods. The paper does not make it clear whether adolescents used the pens and paper provided or found it helpful. Eggenberger and Nelms (2007) found that families were happy to engage in the interviews and tell their stories. Moreover, through being interviewed as a family, individual family members learnt about each other. One family even reported that the family interview was exactly what they needed. This suggests that even at a family crisis point, families enjoy and may even benefit from being interviewed together.

Leshed and Håkansson (2014) included much younger children in their interviews with farming families at home. They also used a type of VEI as they asked the family to show them objects during the interview. However, it is unclear how much this was set up prior to the interview. Moreover, they did not endeavour to interview the family, as a whole, when starting the research. Instead, they planned to conduct individual interviews but very quickly

realised that families preferred being interviewed together. Fitting with their individualistic perspective, there were two interviewers present at each visit so that one could continue talking to someone if someone else in the family started talking about something else.

My study is novel as I aimed to include children and their families to understand the experiences of CF in family systems. I aimed to get a family-level understanding as families make meaning together. Moreover, I aimed to do this by fully using VEI with all participating family members. It is important to reflect on family interview methodology as families are different than focus groups (Eggenberger & Nelms, 2007), where individuals may not know each other nor see each other again. Thus, due to the novel nature of this design, and the lack of guidance in the literature, the secondary aim was to reflect on the feasibility of using VEI with the whole family.

Chapter 2: Methodology

This was a qualitative study using adapted respondent-generated visual elicitation interviews (VEI) to understand some of the family-level experiences of cystic fibrosis (CF). I analysed the data using interpretative phenomenological analysis (IPA).

In this section, I will discuss this study's epistemological and ontological positions. I will then discuss the role of the involvement partners in the initial design of the study before discussing the rational for the methodology chosen, providing more information about the VEI approach used here and IPA. I will also describe other approaches that were considered but ultimately decided against. I will then discuss the mock interview and what was learnt from it. To ensure the trustworthiness of the study, I will end this section by reflecting on the perspectives and experiences that I bring.

Qualitative approach

Qualitative approaches allow us to gain insight into the experiences of participants and how they make sense of these experiences (Hollstein, 2011). Little is currently known about family experiences of CF in the current landscape. Therefore, a qualitative approach, looking in depth at family experiences was thought to be most appropriate.

Ontological and Epistemological Position

To understand a study, it is necessary to consider its ontological and epistemological positions. Ontology is what we believe reality is, whilst epistemology is our beliefs about how we access it (Kant, 2014).

Ontology

Realism and idealism sit at opposite poles within ontology. Realism believes that we all observe things exactly the same way (Willig, 2013), whilst idealism argues that physical reality is dependent on our previous experiences (Willig, 2013). Critical Realism sits between realism and idealism. It assumes that whilst there is a physical reality, we see it through what we have already experienced (Danermark et al., 2019). This is the view that I most align with. I think that there is a physical reality, otherwise things seem too abstract to comprehend. However, I think who we are as people, and who we interact with (as well as when we interact with them) affects how we see things. This positioning also fits with IPA, which wants to understand how participants make sense of their experiences (Smith et al., 2021).

Epistemology

Positivism and interpretivism sit at opposite poles in epistemology. Positivism believes that knowledge can be gained objectively through data collection (Alharahsheh & Pius, 2020) and is where the traditional scientific method sits. On the other hand, interpretivism, connected with idealism, believes that reality is dependent on our previous experiences and thus can only be observed on an individual basis (Alharahsheh & Pius, 2020). Contextual Constructionism sits between positivism and interpretivism and states that whilst knowledge exists, we never know everything as we all make sense of it through our unique contexts (Jaeger & Rosnow, 1988). This fits with my own views as I believe that whilst the literature base is important, we need to see everyone as individuals without any preconceived ideas of what life may be like for them. It also fits with IPA, which highlights the double hermeneutic, namely that the researcher is making sense of the participant's sense making based on their own beliefs and experiences (Smith et al., 2021).

Involvement Partner Help in Initial Study Design

To ensure this study was accessible and meaningful for families with CF, involvement partners were recruited to help from design to analysis. Whole FwCF were recruited as I was aiming to recruit whole families as participants. It was initially envisioned that the same involvement partner families would be involved throughout, however this ended up not being possible, perhaps due to a break in the study as I went on maternity leave.

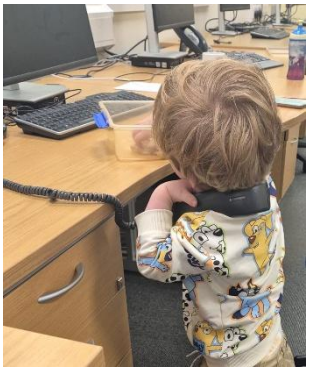
Two FwCF were recruited as involvement partners early in the design of the study. Both families only had one child. It was soon apparent that due to the business of family life, it would not be possible to meet up altogether. Separate meetings were therefore held online. In one family, only the mum attended both meetings. In the second family, the mum attended one meeting and she attended the other with dad and daughter. The recruited involvement partners thought that FwCF would be happy to participate in the study and able to talk about CF. They felt that the information sheets explained the study well. However, they did not agree on the age at which a child would be able to reflect on their experiences of CF. One of the involvement partners gave advise on the visual elicitation task, which will be discussed in the next section.

Visual Elicitation Family Interviews

This study utilised adapted respondent – generated visual elicitation interviews (VEI) to collect the data. Participants were asked to bring ‘creative items’ to the interview to act as stimuli for discussion. This ensured that the interview discussions centred on what was meaningful for the participants (Ford et al., 2017) and provided children with something concrete to anchor discussions onto (Barton, 2015).

The creative items could exist in any medium including photos or pictures as well as stories, comic strips or objects. Whilst bringing written items to the interview is not usually part of VEI, it was felt to be beneficial here as an involvement partner reflected that their CwCF would prefer to write something than submit a picture (see Appendix 1). As the point of the creative items here is purely to help the participants share their story, it was felt that the medium most helpful for each participant should be encouraged. In support of this, Spencer et al. (2023) asked their young involvement partners to select activities which would appeal to participants of a similar age and help them share their experiences. Both written and pictorial activities were chosen to be further developed and used in the study. Moreover, both types of activities were selected by participants, highlighting how different designs appeal to different individuals. Thus, allowing participants to choose the medium that they bring to the interview may help them engage in the task in a way that feels comfortable for them.

Author's Comments and Reflections 2. *Using Visual Elicitation Methodology*



I emailed Olivia Stransky about her experiences of using visual elicitation methodology within the cystic fibrosis population in Stransky et al. (2023). Oliva Stransky's study looked at the experiences of parents with CF. She commented about the need to be flexible as family life makes scheduling a lot harder. She also commented on how the interviews became quite emotional as the participants discussed parenting with cystic fibrosis but that there seemed to be a cathartic effect. She found having a pre-interview meeting helped set up the interview as it allowed her to explain to the participants what was expected of them. She spoke about giving credit to her participants for any photos used in publications. We have spoken to our involvement partners about this and they do not think it is necessary to give credit. As some of the creative items will be done by children, who may change their views as they develop, it is felt that maintaining anonymity in the creative items will be best. Olivia did raise concerns that parents would not speak freely about things like mortality in our study. We agree that different things may be raised than if we were to speak individually with family members. However, as the aim of this study was to gain a family-level understanding, we believe that this is okay.

Interpretative Phenomenological analysis

IPA draws upon three key ideas in the philosophy of knowledge, namely, phenomenology, idiography and hermeneutics (Smith et al., 2021). Phenomenology is embedded in the personal experiences of the individual. It sees the individual as being embodied and situated in their unique context (Smith et al., 2021). Idiography is attending to the particular (Smith et al., 2021). IPA uses small samples to allow the researcher to deeply understand the unique experiences of their participants. Through hermeneutics, IPA recognises that reality is socially constructed through interpretation. Namely, the researcher makes sense of the participant's interpretation of their experiences through the researcher's context (Smith et al., 2021). This is known as the double hermeneutic (Montague et al., 2020). This also fits with the idea that families are making sense of reality together. The hermeneutic circle is concerned with the relationship between the part and the whole, specifically to understand a part one needs to understand the whole and vice versa (Smith et al., 2021).

The use of IPA within focus groups has been criticised for not holding the hermeneutic circle in mind. Specifically, Tomkins and Eatough (2010) highlight that previous IPA research with focus groups has analysed the data at group level, ignoring the individual, but then has presented the data as if it was collected from individuals within the write up. This is problematic as it goes against the principles of IPA. Since focus group participants only come together for the purpose of the research study, their group level meaning making may not exist naturally within the world and thus it could be argued that IPA is best done with individual interviews.

However, family groups are different from typical focus groups as they can be understood as a more meaningful analysis unit (Eggenberger & Nelms, 2007) bound together outside of the research context. The Resiliency model of Family Stress, Adjustment and Adaption

(RMFAA) suggests that family meaning making is important in adaptation (Brown-Baatjies et al., 2008) and therefore it is critical that we consider it.

Koenig and Trees (2006) attempted to understand joint family storytelling processes. They recruited 12 family units, made up of three family members, and analysed how meaning was made in the unit. They suggested that meaning was either made through family-unit sense making, individual sense making or incomplete sense making. In family-unit sense making, a mutual understanding was developed by the family considering all individual perspectives. Families that engaged in individual sense making processes paid less attention to what each other said and thus the individual perspectives were not integrated together. Incomplete sense making was categorised when the researchers could not find any meaning made at either an individual or family-level. This does not fit with IPA's understanding of humans as sense making creatures (Smith et al., 2021). However, whilst Koenig and Trees (2006) say that they could not draw any conclusions from what was said (or not said as a family member was disengaged in the process within this category), the quotes used show the family describing what happened with adjectives showing how they found it. Thus, the researcher's view that no meanings could be drawn may be based on their epistemological position and level of interpretation. Overall, this study shows that families can engage in meaning making together but that the meanings conveyed may not always be within one shared understanding. This highlights the importance of the hermeneutic circle and the need to move between the whole and its parts. As highlighted above, it is reasonable to see the family as a meaning making unit.

Tomkins and Eatough (2010) suggest analysing the data first at a group level and then going through the stages again for each individual to see how their meanings mapped onto the group level meanings. This allows the researcher to move between the whole and the parts. It also enables the researcher to develop an understanding as to whether the family is creating

one coherent story or multiple separate stories as shown in Koenig and Trees (2006). However, care needs to be taken to remember that the individual meaning making is being done in the context of the group, which likely influences the meanings made. Namely, a multiple hermeneutic exists as many individuals, including the researcher, are trying to make sense of what is being said (Tomkins & Eatough, 2010). Thus, the order in which things are said may also be important. Analysing the data at group level and then looking at how the individual members fit or diverge from this understanding within an IPA framework has previously been done in research on couples (Antoine et al., 2013; Wawrziczny et al., 2016) and focus groups (Githaiga & Swartz, 2017).

Author's Comments and Reflections 3. *Family as a unit of analysis in IPA*

I have emailed Prof Jonathon Smith about the use of family as the unit of analysis. He suggested that this was a possibility. He suggested that, depending on the data I get, I could look at the family experience as the unit of analysis and individual experiences within that.

After attending epistemology teaching, I at first felt very lost in how I was going to connect all parts of this study. However, after reviewing the literature, it does seem clearer and I believe it makes sense to do IPA to fulfil the aim of this study.

Alternatives Considered

Purely verbal interviews were considered but it was felt that VEI would help empower all participants to share their stories, especially as young children may be involved. Interviewing individuals or dyads instead of the whole family unit was also considered but it was felt that this would miss family-level stories. This was felt to be limiting as everyone in the family likely influences each other (Segrin & Flora, 2018) and any change from interventions likely takes place within the whole family unit. Only using one type of stimuli, such as purely

using photos, within the VEI was also considered but one of our involvement partners felt that this was limiting as their child would be happier to participate in a written task.

As well as IPA, Discourse Analysis and Grounded Theory were considered for data analysis. Discourse Analysis explores how an experience is understood through language (Willig, 2008). It was considered as the study hoped to develop an understanding of the experiences of families by asking them to discuss their experiences together. However, it was decided that it was not an appropriate methodology to achieve the study's aims as I was not interested in how families construct discourses but instead wanted to focus on the meanings being made of their experiences. Grounded Theory was considered due to it being situated within social constructivism (Charmaz, 2008). However, Grounded Theory generally aims to achieve a large sample size so that a theory can be developed. This was not something that the current study aimed to do.

Eggenberger and Nelms (2007) analysed their whole family interviews with hermeneutic phenomenology. Alsaigh and Coyne (2021) developed guidance on how to do a hermeneutic phenomenology analysis as they realised this was lacking in the literature. Their steps are very similar to the steps provided in IPA. Indeed, IPA has been described as an 'integrative hermeneutic phenomenology' and is a more established methodology within psychology (Tuffour, 2017).

Involvement Partner Help in Mock Interview

A new family was recruited as involvement partners for the mock interview. A family with a sibling of a child with CF (SoCwCF) was deliberately sought as during a research panel held whilst the research was initially being developed, it was raised that SoCwCF may find it particularly hard to talk about CF.

The family recruited consisted of a mum and two children, one of whom had CF. The child with CF was nine years old. Learnings from the Mock Interview are summarised in Table 3. Full details can be found in Appendix 1. The family approached the gathering of the creative items differently than what was originally envisioned in the initial research design. Specifically, it was thought that each family member would come up with at least two of their own creative items independently of each other, but the involvement partners doing the mock interview submitted their creative items as a group. Upon reflection, it was felt unrealistic to expect families not to talk about their creative items and that it is likely that different families will approach the task differently. As a result, the information sheets were adjusted to clearly allow this. A possible negative consequence though of creative items being submitted as a group is that someone's voice may be minimised in the selection of items. Therefore, it was agreed that I would check in the interview if there was anything else that people would like to talk about.

Whilst the SoCwCF seemed more distracted throughout the interview, he was happy to participate and opened up about his experiences of CF. As individual differences exist in terms of how much people talk in interviews, it is felt that too much emphasis should not be placed on his distractibility. The fact that he was happy and able to open up about CF shows that siblings without CF can be included. Overall, the interview set up seemed to enable all participating family members to share their thoughts on a variety of topics picked by them.

The involvement partners who helped with the mock interview also helped with the analysis of the data. This will be discussed later on.

Table 3. *Learnings from the mock interview*

What was learnt	What was done because of the mock interview
Having a pre-interview meeting helps participants understand the task, which can ease anxiety.	Pre-interview meetings were conducted before each interview.
Conversations that are hoped to be had during the interview may start during the pre-interview meeting.	It was agreed that I would stop the family if it seemed like the conversation in the pre-interview meeting was becoming an interview-style discussion. It was agreed that I would tentatively bring up topics raised in the pre-interview meeting when appropriate in the interview. However, I would ensure I gave the family space to say if they did not want to discuss it in the interview.
Participants may be worried about saying the right things.	I stressed to all participants that there are no right or wrong answers.
Families may approach the creative items task differently	I ensured that everyone present was able to discuss what they thought was important. I was curious about how the creative items were created in the interview.
Children below secondary school age can actively take part in the study	The child with CF having to be of secondary school age was removed from the inclusion criteria
The home environment helped the children stay present. However, it meant that the interview was also interrupted by daily tasks such as the post.	Interviews were offered to be done in the family home in the first instance. It was felt that disruptions could happen anywhere and that ensuring participants were at ease was most important.
Rich discussions were had after the interviewer stopped recording.	After consent was obtained, I planned to record all discussions unless the family did not want me to record something specific.
The children were struggling to engage after around 42 minutes	I paid attention to how all family members were doing. The ethics form said that the interviews will take around 60 minutes but it was anticipated that this may be less depending on the family.
The sibling without CF was happy to participate and able to reflect on CF	Well siblings were included
The family enjoyed the mock interview and rich data was collected	The research design was considered to be appropriate for this study

Reflexive Statement

In this statement, I recognise the assumptions, biases and pre-existing beliefs that I brought to the study. Other parts of myself were also triggered as the study progressed (LeVasseur, 2003) and thus are included in reflective boxes at appropriate stages within this thesis.

I am a 32-year-old White British female from a middle-class background. I had no real knowledge of CF before starting this research. However, childhood epilepsy was present in my family, and I experienced how we all made sense of it. Unlike CF, it did not require lengthy treatment schedules, but I was aware of having to buy frozen peas when camping to keep the medication cold and that we were unable to watch things with flashing lights. I also experienced a few of the seizures and the distress and confusion that they caused. As a result, I am aware of how fear can exist in families when a chronic condition is present and how my family found ways of adapting. I did not mind the things we had to do because of childhood epilepsy and instead was focused on protecting and helping the individual. Moreover, I was diagnosed with speech dyspraxia as a child and had to attend speech and language therapy throughout my childhood. This made me feel different from my peers, which I did not like. I have noticed that I can often be pulled to ensuring individuals feel understood and not different from others.

I have completed the PGCert in Systemic Practice and am very interested in how family systems work. I tend to think about cases in terms of their system and systems theory. I have also grown up in a family unit as the oldest of four children. This may mean that I am pulled to understanding sibling dynamics in terms of my own experiences. For example, I feel protective of my younger siblings but am also fully aware of sibling rivalry and wanting to 'win', whatever that means. I am now married and have a son, who at the time of submission was two. I have experienced the huge adjustment that having a baby can have on family life. I

am also aware of the pressure placed on parents and may be drawn to trying to protect parents from feeling judged or not good enough.

Before recruiting participants, I developed knowledge of CF and the theories presented in the introduction whilst I designed the study, applied for ethics and prepared for the Transfer Viva. I tried to remain consciously aware of what may have been coming from my knowledge and what was coming from the participants. I ensured that I continuously reflected on what was happening in contact with participants and when my knowledge of the area may have been leading me to make sense of things in a certain way.

I was mindful of my positioning during all interactions with families including the interview and sought to be curious to each family's unique stories rather than just listening to my own (LeVasseur, 2003). I spent time listening to the audio and reflecting on how the participants' words, tone and stories were affecting me. This helped me become more aware of when my biases were sneaking in (Rodham et al., 2015). I have also tried to share any pulls or emotions I experienced in this report to ensure transparency. Moreover, as I am not part of the CF community, I asked the involvement partners for help with making sense of anonymised sections of the transcripts to enhance the credibility of any interpretations (Smith et al., 2021).

Chapter 3: Method

I have now discussed the design of the study and the principles on which it is based. In this section, I will describe the specific steps taken to fulfil the aims. This section will end with an assessment of the quality of this research.

Sample

Families with a child with CF were recruited for the study. Participants could decide which family members took part as all families are different. Moreover, who is affected by the CF or plays important roles around the CF may vary across families. Parents were offered help to decide on the capability of their children to participate by either the clinician informing them of the study or by myself when arranging the pre-interview meeting. A mock interview was completed with one child who was nine years of age, and they were fully able to participate and share their views (see Appendix 1). However, the ability of children to participate may vary across age ranges as shown by different views within the involvement partner families (see Appendix 1). As this study is looking at the family unit rather than individuals, it is thought that it does not matter if the children included are at different life stages.

Inclusion Criteria:

- At least 2 family members willing and able to participate.
- The child with CF is able and willing to participate.

Exclusion Criteria:

- The child with CF is extremely unwell.
- The child with CF was diagnosed later than four years of age as this will possibly change the experience of CF within the family due to the child likely being quite unwell at point of diagnosis (De Boeck, 2020).

I aimed to recruit 3-5 families to enable detailed analysis with rich, deep personal insights (Hale et al., 2008).

Recruitment

Participants were recruited from one UK Cystic Fibrosis Centre. They were approached by their clinician at their usual clinic visit.

Consent

At the child's usual clinic visit, the clinician explained the study to the family and gave them the study information pack. Initially this pack contained all versions of the information sheets (adult, parent, child and child easy read) as well as information about the creative items and all the consent and assent forms. However, the clinicians felt that this was too much information for the family to be provided with initially. Therefore, just the standard information sheet and consent to contact form were provided. I gave all other forms to the family when arranging the pre-interview meeting.

At the clinic visit, the clinician gained written consent from an adult in the family to pass on the adult's contact details via NHSmail to myself. I then got in touch with the family via the consenting adult to answer any questions and arrange the pre-interview meeting.

The families were able to choose whether the pre-interview meeting happened online or face-to-face. Online meetings were conducted over Zoom. If a face-to-face meeting was preferred, families could choose between meeting at the family home or in a pre-booked university room. All face-to-face meetings ultimately took place at the family home. All participating family members were asked to be at the pre-interview meeting. At the meeting, I verbally explained the study and answered any questions. I talked through the 'creative items' and

gave each participating family member the guidance sheet for the 'creative items' (see Appendix 2). I asked participants to gain verbal consent themselves from anyone present in their creative items before they submitted them. If the pre-interview meeting happened face-to-face, the consent / assent forms were completed at the end of the meeting. If it happened via Zoom, I asked all participating members to sign and email them to me. I verbally checked consent and assent again immediately before the interview.

Data Collection

Interviews were audio-recorded using two encrypted digital Dictaphones that are held by the university. Families could choose between doing the interview in the family home or a pre-booked university room. All families chose to do the interview in the family home. To limit the technology requirements of the interview, participants were asked to email their creative items, or photos of their creative items, to myself to print a week before the interview. To ensure parents were aware of what their under 16-year-olds were bringing to the interview, they were asked to email creative items for any of their children under 16 years of age on their behalf. This was done to protect the child. However, it ultimately meant that parents were the gatekeepers of what was brought to the interview.

It was planned that the interviews would start with the participants sorting their 'creative items' into piles to help them engage with them and ensure that the most important 'creative items' were discussed for each participant (Bugos et al., 2014). However, this felt like a very unnatural step in reality as there was not a large quantity of creative items. The interviews did all start with the families looking through the items, which allowed them to re-familiarise themselves (Bugos et al., 2014) but this was led by the family rather than myself.

I started the interview by asking the family which creative item they wanted to discuss first. In most interviews, this decision ended up being given to the children. I tried to use playfulness to help empower everyone in the discussions that took place around each creative item. Circular questioning (Cecchin, 1987) was used to gain a family perspective. Concrete questions were also used as these can feel safer, especially for children (Bugos et al., 2014). I asked the family for feedback on the interview at the end.

Immediately after the interview, a brief demographic questionnaire was given to an adult in the family to complete. In most cases, all participating family members were still present when it was filled in. The questionnaire asked for the family's ethnicity / ethnicities and the current age, gender and age at diagnosis of the child with CF as well as whether they were currently taking any modulators. It also asked about the ages and genders of immediate family members and if there were any other mental or physical conditions present in the family. It was up to the family who they included as immediate family members. Finally, it asked about parent(s)/carer(s) current occupation(s).

Debrief

At the end of the interview, I debriefed the families. I offered each family a further debrief with the CF clinic within one week of the interview to discuss any difficulties that may have arisen. However, this was not taken up by any of the families as they felt it was not needed.

Reimbursement

Each family received a £30 love2shop voucher as a thank you for their participation.

Transcription

I transcribed all the interviews verbatim. Transcripts did not include anything other than the spoken word unless it was felt to be important to include more detail to help understand the context in which something was spoken (e.g. one child responded in agreement to their parent saying 'anything is possible' but in the room and on tape, their tone sounded like it was something they had heard many times before. A note of the child's tone was therefore added in the transcript).

Analysis

The transcripts were analysed with Interpretative Phonological Analysis (IPA). The analysis steps in Smith et al. (2021) were followed and expanded on to allow for my learning journey and reflections with my supervisors. As IPA is seen as being flexible rather than rule-bound, this was considered appropriate (Smith et al., 2021). The steps taken through the analysis of the research are detailed below. The interviews were analysed in the order in which they were conducted.

1. **Analysis of each transcript at the family-level.** The transcript was read through multiple times before in-depth analysis was started. A map through the transcript was written as a table on Microsoft Word to help me make sense of each interview. It showed how conversations flowed as perhaps rapport developed over the course of the interview (Smith et al., 2021) or participants began to tire. The transcripts were then printed off to enable deep engagement with the data (Smith et al., 2021). The paper copy of the transcript was read through at least once in its entirety. Exploratory notes were subsequently made on the paper copy on a line-by-line basis. These included descriptive and linguistic comments as well as thinking about the meaning

that the individual or family were making and/or the meaning I was making of it. They also included notes on the methodology. The exploratory notes were summarised into experiential statements on the other side of the transcript. Finally, connections between the experiential statements were sought resulting in Personal Experiential Themes (PETs).

2. **Discussion with involvement partners.** Anonymised extracts from all transcripts, apart from their own, were taken to an involvement partner family to gain insight into their interpretations as suggested in Smith et al. (2021) because neither myself nor my supervisors have lived experience of CF. This family consist of one mum and two sons. Spencer et al. (2023) have previously shown that children can be involved in the analysis of interview data. No specific analysis training was provided to the family. They were simply told that we wanted their interpretations of the data as they are experts in lived experience of CF. They were also advised that I would take their thoughts and use them to help me with the analysis as they did not have access to the full dataset. I sent some of the quotes to the family ahead of the meeting so that they could start making sense of them. The family looked through these quotes together the night before I came. At the meeting, I laid all the quotes out on the table, and we discussed whichever ones anyone in the family felt to be meaningful. I then asked the family to help sort the quotes into piles based simply on which quotes were thought to be connected. However, by this point the boys had lost interest and so it was primarily the mum who sorted the quotes. This meeting with the family was audio recorded so that I did not miss any important reflections. A summary of the meeting is presented in Appendix 1.
3. **Writing up meeting with the involvement partner family.** I then listened to the recording of my meeting with the involvement partner family and looked at the piles

of quotes created. I wrote this up into a word file which I circulated with my supervisors.

Author's Comments and Reflections 4. *Analysing with the involvement family*

When meeting the involvement partner family for help with the analysis, it became clear that the scales on which people make decisions as to what is okay and what is not okay varies across families. I felt a strong want to defend the participating families. I realised that by providing only isolated quotes to preserve anonymity, the families and the decisions they had made maybe came across differently than they had in the actual interview.

The involvement partner family suggested I read Wotton (2014) as it had given the mum a good insight into how people slightly older than her viewed the condition. Whilst reading the book, I did note many similarities between the things described by Wotton and what was discussed by my participants. I also reflected on how the book was published around the time that the children with CF in my study were being born.

4. **Re-listening of each audio file and preparation of notes to bring to supervisors.**

As different understandings can be achieved from listening to the audio vs reading the transcript (Rodham et al., 2015), I relistened to each audio file as I prepared a document for discussion with my supervisors. The document I prepared included a thorough pen portrait for each family, any reflections I had made at any point relating to the family and the clusters of experiential statements that I had at that moment in time with some quotes. The document also included any scales on which people make decisions that were apparent in each transcript because in previous supervision meetings we had reflected on how this seemed like an important theme across interviews. I also included any quotes that I wanted to discuss further because they either stood out for me or I had a query about how to interpret them. I also included any reflections made by the involvement partner family in step 2.

5. **Discussion of the notes prepared with my supervisors.** This meeting was audio recorded to enable me to engage actively in the discussions and not have to worry about writing everything down.
6. **Transfer of data into Excel spreadsheet.** My paper transcripts felt like they were getting very messy as I repeatedly handled them. I also felt like I did not have space to add further notes. I therefore transferred the transcripts and notes to a Excel workbook designed by Horton (2024). During the transfer, I added further exploratory notes and experiential statements as I noticed or reflected on things whilst reading the transcript again. The workbook was used solely for showing exploratory notes and experiential statements more clearly.
7. **Further Analysis of each transcript at the family-level.** Following the steps described above, the transcripts were further analysed at the family-level with the new reflections and notes in mind. The PETS were updated. At this point, attention was paid to how individuals within each family were relating to each other and whether they were agreeing or disagreeing with what was being said as suggested in Tomkins and Eatough (2010).
8. **Analysis across cases.** Once all transcripts had been analysed at the family-level, patterns of sameness and difference were sought across the families to gain group experiential themes (GETS).
9. **Checking analysis developed with notes from my meeting with the involvement partner family.** I went back to my notes from my meeting with the involvement partner family to check their comments still fitted with the analysis.
10. **Discussion of family-level and group level themes with supervisors.** The PETs and GETS were discussed in supervision. Many quotes were provided to enable us to stay as close to the data as possible. These discussions took place over several meetings. In

between these meetings, I reflected on the points made by myself and my supervisors and the raw data. This resulted in me re-doing steps 7, 8 and 9 between each supervision meeting until agreement was reached with the analysis.

To achieve the secondary aim, I spent time after step 10 thinking about my reflections written in my reflective journal, any relevant experiential statements or Personal Experiential Themes (PETs) as well any other comments made by participants during the research process. I discussed any patterns that I noticed with my supervisors.

Journaling

The keeping of a reflective journal is recommended in Smith et al. (2021) as it produces an audit trail, increases transparency and can help the writer process their thoughts (Vicary et al., 2017). As one of my aims was to explore the use of the methodology, the journal also gave me a way of remembering my thoughts on the process as it unfolded. Thus, as well as improving my own learning and increasing the quality of this study, the journal was a necessary part of meeting the study aims.

The journal was kept on a word document stored on OneDrive and was added to after key moments in the research (e.g. contact with participants) or when a reflection came to mind. As the research progressed, my attitude towards the journal changed. For example, I began to see it as more useful to include the date that reflections were made. I also began having the file open whenever I was doing any work on the thesis and began recording reflections on the theory I was reading as well as on my data and the process. This change happened after reading about the benefits of journalling in Vicary et al. (2017). A few extracts from my journal can be found in Appendix 3.

Assessment of quality

It is important to assess the quality of qualitative research. Many papers have offered guidance on how to do this and fortunately they mainly agree with each other (Cohen & Crabtree, 2008). I assessed the quality of this study using the four principles proposed by Yardley (2000) and Yardley (2008), namely 'sensitivity to context', 'commitment and rigour', 'transparency and coherence' and 'impact and importance' (see Table 4).

Table 4. *How the current study met the quality principles in Yardley (2000, 2008)*

Principle from Yardley (2000, 2008)	How it was met in the current study.
'Sensitivity to context'	An involvement partner family was asked to help with the analysis as neither myself nor my supervisors have lived experience of CF. Any phrases used by participants that seemed unclear were discussed with my supervisors. This included any metaphors used. I have spent time reflecting on my own context and how this may have influenced the interviews and analysis. I have also provided a pen portrait of each family to help set the scene for the interviews but due to concerns around maintaining internal confidentiality, this was kept to a minimum. More detailed pen portraits were used in discussions with my supervisory team.
'Commitment and rigour'	I spent time in the Cystic Fibrosis Centre and have read Wotton (2014) on recommendation by an involvement partner to help develop my understanding of life with CF. I have also spent time learning about IPA. I have continuously referred to Smith et al. (2021) throughout my thesis journey. Moreover, I have been part of an IPA online forum. In addition, I have attended IPA help sessions with others in my university cohort. I have also had email contact with Prof Jonathon Smith about analysing family interviews with IPA. I have considered the data on an individual, family and group level and have paid attention to any similarities and differences present. I have asked an involvement partner family for help with the analysis. I have also spent time with my supervisors thinking in detail about the meanings that are being made by participants.
'Transparency and coherence'	The data collection and analysis process have been laid out in depth. I have included my own reflections during this time. The data has been presented in a way that makes sense and this has been checked by others not knowledgeable about CF.
'Impact and Importance'	The results of this study share the story of some families with CF. This enables their voice to be heard. This may help other families coming to terms with a CF diagnosis. By showing things that can be important for families with CF, this study gives suggestions for clinical practice and future research which can be used to improve the lives of those with CF.

Ethics

NHS ethical approval was gained from Newcastle and North Tyneside 2 Research Ethics Committee. IRAS number: 333858. R and D number: PA24/164497. The approval letter can be found in appendix 4.

Ethical considerations

The psychological impact of discussing CF as a family was considered. New information could be brought to light during the interviews as the participants may not be used to discussing CF as a family. Even if conversations around CF were common in the family, the creative items could evoke powerful emotions (Copes et al., 2018). As a clinical psychology trainee, I have experience and training in managing distress and thus it was felt appropriate for me to conduct the interviews. Moreover, I was well supported by my supervisory team which consisted of a consultant paediatrician within CF, a clinical psychologist with clinical CF experience and a senior research fellow with experience of conducting qualitative interviews on CF. I made sure participants knew that we could stop the interview at any time without them having to give a reason. I also debriefed all participants immediately after the interview. Each family was offered the possibility of being contacted by their CF centre within one week of the interview for further support. However, this offer was not taken up by anyone.

The risk of infection also needed consideration as people with CF can be vulnerable to diseases (Bierlaagh et al., 2021). It was agreed that I would inform the family if I had any signs of illness or had recently been exposed to an infectious disease on the day of the interview and let them decide whether they would prefer to do it online or re-arrange. However, this situation never arose in practice.

Careful consideration was also paid to maintaining confidentiality as the CF community is fairly small due to it being a rare condition (Chen et al., 2021). A dilemma exists in qualitative research of wanting to display rigorous data but also needing to maintain confidentiality (Ummel & Achille, 2016). Thus, a balance is required in terms of how much contextual information is shared. To achieve this balance, I have kept the pen portraits provided for each family brief. My supervisors and I also paid careful attention to the information in the results section and whether it identified participants. Similarly to Ummel and Achille (2016), we deliberately did not use some quotes as they were thought to display too much identifiable information. When group-level quotes are discussed, they are not identified with pseudonyms as this limits the amount of information that is linked to each individual family.

The creative items introduced further complexities around confidentiality. To ensure parents were happy with the creative items selected by under 16-year-olds, it was requested that parents emailed me the items. Whilst this was an attempt to safeguard the family, it did ultimately give parents more power. This is something that will be reflected on later in the Methodological Reflections chapter. Participants were also asked to gain verbal consent from anyone present in the creative items as detailed in Ford et al. (2017). It was felt beneficial to include creative items within dissemination of the results as they help tell the participants' stories. Separate written consent for use of any creative item in dissemination was gained after the interviews from the person who submitted the creative item, and anyone else also present in the item. Everyone involved was shown exactly what the creative item would look like before they gave consent so that they were aware of what others would see. Specifically, the creative items were edited to make them as unidentifiable as possible. It could be argued that it is ethical to credit participants for their creative items as it was their work (Stransky et al., 2023). However, children's views on how their items are used may

change over time as they grow up (Ford et al., 2017). Fully crediting the participants would have made them identifiable, which even if they consent now, may not be something they are happy with in the future. Moreover, an involvement partner was asked about crediting participants for the creative items, and they did not think it was necessary. Therefore, the decision was taken to not credit any participant.

As the involvement partners were from the same CF centre as the participants, maintaining internal confidentiality was also an issue when seeking help with the analysis. I thus only showed them isolated anonymised quotes. However, this did limit how much they were able to make sense of what was said. I also did not show any creative items as consent had not yet been gained to use them in dissemination.

Data Management

Data was stored securely in line with NHS and University of Leeds policies as detailed in the ethics application.

Chapter 4 – Results

This chapter presents the results from my analysis of family experiences of CF. To help understand the context of each family, I will first provide brief pen portraits. I will then present overarching themes to introduce the general findings. I will conclude this section with the individual themes, thinking about how they fit in with and are unique from the overarching themes. In this section, it can be assumed that any mention of parents or siblings are those of a child with CF.

All pictures used, apart from those within my reflection and comment boxes, are those supplied by the participants. Creative items were either photos (either taken specifically for the interview or old family photos), drawings or objects (that were also photographed). Interviews lasted on average 53.5 minutes (range: 42 minutes – 64 minutes).

Demographic information

Four families took part in the study. Each family had one child diagnosed with Cystic Fibrosis. Different constellations of family members took part in the interviews reflecting the different make up of families, the busyness of family life and individual's different willingness to be included. Demographic information is summarized in Table 5. As other mental / physical health conditions present in the family did not seem to play a role in the narratives shared in the interviews and could potentially lead to families being identified, they are not included in the results.

Table 5. *Overview of the sample*

Demographic Area	Participant data
Number of families	4
Number of children with CF	4 (1 in each family)
Number of siblings that took part	1
Number of families that took part with just one parent	2
Number of participating family members in an interview	2 – 3 family members
Ethnicity	All families were White British
Socio-economic status (SES) estimate[1]	2 families had High SES 2 families had Middle SES
How CF was diagnosed	2 x due to meconium ileus[2] at birth 2 x via newborn screening
Age range of children with CF at the time of the interview	7-12 years (M = 9)
Gender of children with CF	3 x female 1 x male
Modulator drugs	3 x taking modulator drugs 1 x not taking modulator drugs

Note. [1]SES estimate based on parent's current occupation according to Office for National Statistics (2010)

[2] Meconium ileus, obstruction in the intestines, is often the first sign of CF and happens in 20% of patients (Sathe & Houwen, 2017).

Pen Portraits

To help understand the individual contexts for each participating family member, I have written brief pen portraits. I initially wrote detailed pen portraits to help myself and my supervisors fully understand each family's context when working on the analysis. However, to maintain anonymity as the CF world is small, only brief pen portraits are presented here. The families are numbered in the order in which they were interviewed. This was also the order in which they were analysed. All the CwCF were born around the time modulators were being developed. However, they will all have only recently trailed or started taking them due to initial age limits set on the drugs.

Family 1

This family was originally recruited as involvement partners for the mock interview. However, they wanted their interview to be included in the study. As nothing changed from the mock interview in terms of the setup of the interview, consent to be included in the study was gained retrospectively. This was done after approval for such consent was obtained from Newcastle and North Tyneside 2 Research Ethics Committee.

The family consisted of a mum, Avery, and two boys, Riley and Harper. Riley is 12 years old and does not have CF. Harper is 9 years old, has CF and is currently taking modulator drugs. Harper was born after the first modular drug was approved and correctors were in development. He was born without any obvious health problems and diagnosed via newborn screening.

Family 2

This family consisted of mum (Aria), dad (Luca) and two children (Charli and Rowan). Only Charli took part in the interview with both her parents. Aria and Luca felt that Rowan was too young to participate. Luca was at another commitment at the start of the interview and only arrived towards the end. Charli is 8 years and 8 months old, female, has CF and is on modulator drugs. Charli has had Pseudomonas aeruginosa twice but was not unwell with it either time. Charli was born after the first modulator drug was approved for use and correctors were being developed and approved. She was born without any obvious health problems and diagnosed via newborn screening.

Family 3

This family consisted of mum (Alex), dad (Cameron) and child (Blake). Blake is 12 years old, female, has CF and is not currently eligible for modulator drugs due to experiencing

adverse side effects when they were trailed. She has been hospitalised several times due to CF. Blake was born the year the first modulator drug was approved for use. She was unwell with meconium ileus as a newborn and spent time on the surgical ward.

Family 4

This family consisted of mum (Eden), dad (Jordan) and three children. Only Eden and their youngest child (Ash) took part in the interview. Ash is seven years old, female, has CF and is on modulator drugs. Apart from being unwell as a baby, Ash has generally been well. She was unwell with meconium ileus as a newborn and spent time on the surgical ward. She was born after the first modulator drug and correctors were approved for use.

Results of the Group Analysis

In this section, I will present the group analysis results. Five group experiential themes (GETS) were developed through the clustering of shared experiences between the individual families. One family gave the metaphor of a sponge, which seemed to make sense for all families. Thus, the GETS are centred on this metaphor and are briefly summarized below.

1. The first GET describes the initial overwhelming experience of the diagnosis, namely 'Initially get the sponge out of the packet and put it in water'.
2. The second GET describes how families find ways of living life with CF, namely 'Learn to squeeze out the sponge through negotiations'. These negotiations happen inside as well as outside of the family and include negotiations around treatment.
3. The third GET centres around how families must find new ways of coping as the child develops or health needs change, namely 'Renegotiating through changes'. This involves increasing the child's awareness, parents letting go and medication changes.
4. The fourth GET describes the fear and dread of things becoming too much again, namely 'A sponge only absorbs so much water before it starts coming out'.

5. The last GET is ‘Being different from others’.

Note. Picture of a sponge

A simple sponge carries much meaning for families with CF anyway as they are told at diagnosis “if you’ve got sponges get rid of them because … they absorb water [which] harbours dangerous bacteria”. In the metaphor used within this analysis, the sponge can be seen as being the family and the water that it soaks up can be seen as being all the information required to manage CF as well as the stress associated with it. Unsurprisingly, this can become too much for the family to contain just like the sponge can “only absorb so much water before it starts coming out”. Thus, families must find ways of squeezing out the water. This is not a single solution because as the family grows up or health needs change, new ways must be found. Moreover, there is always the risk of too much water being poured over the sponge again, for example if the child with CF becomes unwell. Additionally, just like a bright yellow sponge stands out on the drainer, families with CF can stand out from others.

Table 6 displays how each GET relates to the individual family narratives. Whilst the interview with family 1 did not contain as many of the GETs, the family shared more experiences relating to each one whilst helping with the analysis. This suggests that the GETs do fit with their experiences. Each GET will be discussed in detail. To limit the amount of information shared about each family, standalone quotes are presented in the group level results without indicating who they are from. Quotes are presented with quotation marks. If an extract from the interviews contains multiple speakers, each new speaker is presented on a new line. Pronouns and connected words are changed in the quotes to ‘they / them’ to further reduce the amount of information shared about each family. A longer list of relevant quotes can be found Appendix 5.

Table 6. How the group experiential themes relate to the individual family narratives

	Family Number			
Theme	**1**	**2**	**3**	**4**
'Initially get the sponge out of the packet and put it in water' – the overwhelming experience of the diagnosis and initial information provided	-	x	x	x
Learn to squeeze out the sponge through negotiations – families finding ways of living life with CF	x	x	x	x
Renegotiating through changes – families finding new ways of coping as the child develops or health needs change	x	x	x	x
"A sponge only absorbs so much water before it starts coming out" – fear and dread around things becoming too much again	-	x	x	x
Being different from others	x	x	-	x

Initially Get the Sponge Out of the Packet and Put It in Water

The sponge metaphor, proposed by one of our participating families, suggests that the diagnosis of CF is like a lot of water suddenly being poured over the sponge. In all the families, the diagnosis happened in early infancy. In two families, the diagnosis happened after the child had been hospitalised as a newborn.

"well [they were] born and … had Meconium Ileus so … was transferred to … surgical ward … and eventually they sent us home but obviously they were still waiting for the heel prick test to come back ... and then it was like when they said alright we've got the test results back can you come in, we will tell you what it is and obviously the instructions we were given were to go to the door at the side … and it says cystic fibrosis unit on the door"

…. "which was a bit of a shock really"

Note. A Picture of all the medication needed to be taken by the young child

It was not just the shock of the diagnosis that caused the sponges to fill up. The diagnosis led to them being given "a lot of information" about how to care for their new infant and keep them safe. They had to get used to "the regime, the medicines and all the treatments" that were needed. Some families spoke about how their sponges overfilled due to all the new information.

Learn to Squeeze Out the Sponge Through Negotiations

Over time, the families learnt to adapt to life with CF. They engaged in negotiations about how to live 'typical' daily life. This can be seen as them learning ways of squeezing out the sponge so that it does not overfill. Sometimes the negotiations happened naturally, and they only noticed afterwards that things had changed. For example, one parent spoke about how she initially worried that her child's need for Creon, a pancreatic replacement therapy, would affect her child's social life but quickly the family got into the routine of the child taking Creon tablets with her when going out with others.

"I think when [they] first started school and going to parties I felt quite sad about it because I thought oh [they] can't just go to a party, [they] can't just go for tea, but actually, I think it's like with everything when we first had [them], ... when its new it's quite scary and then when you realize , you are getting so, like now [they go] for tea, and the parents [they go] to tea with, they know [they are] going to have Creon"

Note. Picture of a pet rabbit

Other times, the process was a more conscious decision. For example, “it was a big decision to let [them] have a rabbit when we got her”. This was because whilst the child was desperate for a pet rabbit and the family wanted them to be able to have this experience, the “bedding is not good for [their health]”. The family negotiated this by agreeing that the mum would clean out the hutch and the child would feed and play with the rabbit. However, the child wished they could do more to care for their rabbit.

The above examples display how negotiations happen both within and out with the family unit. Negotiations are also needed around the treatment regimen. These different types of negotiations will now be discussed in more detail.

Negotiating as a Family. Each family had to work out for themselves what they felt happy with. They had to negotiate boundaries to keep everyone safe but also allow for life to continue, such as whether they could get a pet rabbit or if the child could play in autumnal leaves. This is something that always happens within families, but CF requires an “extreme” version.

Note. Picture of a young child playing in leaves.

“… [they’d] just started walking, straight over to the leaves and wants to start playing in the leaves, children with CF aren’t meant to play in the leaves… how do you let a small child have those experiences that every small child is having ... or do you stop [them]? What do you do? We ended up walking around with a bottle of hand gel really, washing [their] hands all the time”

Different families negotiated these boundaries differently. For example, another family spoke about how they tried to avoid all leaves and “muddy puddles”. They spoke about how seeing

Peppa Pig, a children's cartoon character, jump in puddles made this extra hard, something that the child still remembered. This highlights that having CF does not seem to fit with the media's portrayal of children's outdoor play. FwCF must therefore be creative in terms of how they live their lives.

The differences between where families put boundaries suggest that there is no black and white to living with CF. This lack of certainty was a cause of anxiety for parents as they worried that they "were inadvertently making [their child] very ill" or that their child was missing out on experiences or getting a special treatment that was not needed.

Parents would sometimes engage in negotiations around what was acceptable without their child knowing so that their child was allowed to just be "happy".

> "… [child] can't use a face flannel"
>
> "I can, I can use a flannel!"
>
> "You have to have it dried in between"
>
> "and we do dry it … so I am allowed flannels mummy"
>
> "yea but it was something that daddy and I had to think about"

Another common negotiation is how to handle the different needs of siblings. Whilst life with CF can be difficult, there are things such as being able to eat lots of treat food and salt that may make other children in the family jealous. For young children, parents may be able to hide these differences, through for example, pretending to give salt. However, it is likely that the children will eventually become aware of it. Conversely, children having different needs is not an unusual concept in a family. Open communication around everyone's individual needs may aid understanding.

Negotiating Treatment. The treatment regimen is something that needs negotiating in all FwCF The family needs to work out how it fits with their daily life. Exercise was often incorporated into fun activities such as family walks, dance and gymnastics. One family bought a trampoline to continue to help their child with CF exercise in a fun way. However, the whole treatment regimen, including physio exercises, can be difficult to fit into busy family life. In some ways, the covid-19 pandemic helped families have time to complete all treatments. One family said that "we were stuck inside so we had the time to do all the treatments really well, we weren't rushing anything". On the other hand, the pandemic also reduced physical exercise opportunities and increased anxieties around maintaining health, thus making other forms of physio more necessary. For example, one parent explained, "at one point it felt like we had to be quite rigid with it, especially around covid because we were quite worried".

Note. A picture of a family completing physio activities together during the covid-19 pandemic

Negotiating with Others. Negotiations also must happen outside of the family unit, especially as the CwCF reaches school age and starts needing and wanting to do more without their parents. This can be stressful for parents as they need to trust others, and/or their child, in keeping the agreed boundaries. For example, one parent said, "I would try to make more of a big deal of it as I want them to know how important it is". Parents must also judge whether someone else is able to manage the child's care in different situations.

"… so like if somebody said oh, we will babysit them, but we are going out, no then we will just not go out because that is going to be too much for you to be able to manage"

Negotiations outside of the family unit, for example with school, may be limited in terms of the amount of flexibility that is possible. It was sometimes down to the child to initially highlight any problems.

" ...first aid is like here and my classroom is all the way down here, … I had to walk down to the classroom to get my water bottle, bring it back, and then filled up and then by the time I had finished my Creon it was the end of break because I had to take, to take my pack and my water bottle back"

Note. A picture of Creon

… "but they are the things you need to speak about aren't they, because we didn't know about that for a long time until you mentioned that and then that frustrated you quite a bit, didn't it? … now [they have] got [their] own cup that's labelled so [they] can take the Creon, so [they are] not having to walk backwards and forwards to drop [their] water bottle off"

Part of the negotiations with school involved parents advocating for their child. For example, by suggesting that their child make a "cystic fibrosis healthy plate" in health education rather than a typical healthy plate that is not healthy for them. One family decided to send their children to private school to increase engagement in physical activity. This decision was a result of trying to manage CF and was supported financially by the wider family.

Renegotiating Through Changes

All negotiations within and out-with the family are naturally time dependent. As the family grows up or health needs change, re-negotiations around how to live with CF are needed. This section will discuss how families coped with this.

Increasing the Child's Awareness. Parents were aware of the need for their CwCF to become independent, including with the management of CF. In the interviews,

parents demonstrated this awareness by helping their child think through CF care and what their bodies needed. For example, the following conversation was had between a parent and child with CF: “you tend to do like practicing gymnastics moves and things. Why do you think it’s important to do exercise and things?” “get me some health” “what do you think it does?”

Parents Letting Go. By helping their CwCF become aware of CF health needs, they were able to “let go a bit”, allowing the children to live their own lives, including going on residential trips.

“[child] went on [their] first residential this, last year to London … you had to sort of work out your own Creon when you were there”

“it went very well”

This growing independence required the boundaries of what was okay to be re-negotiated. This was welcomed by the families, even if it felt forced at the time.

Note. A picture of medals from the gym

“when…gym started back up with the social distancing, parent’s couldn’t go in so the instructor would come to the door and take the kids off you and take them and do it and bring them out again ...so that was good from the point of view that it forced me to let go a bit so [they] could go off with the teacher and do it, I think it were good for [them] to have a bit of independence”

Medication Changes. Re-negotiations were also needed because of medication changes. For example, as the children with CF started taking modulator drugs, their health was observed to improve. This included how well their bodies were able to break

down fats, proteins and carbohydrates meaning that these nutrients became more available for use in the body. Thus, the way the family approached food needed re-negotiating as the CwCF was now unable to just eat anything without putting on weight.

Note. A picture of the treat food that used to be enjoyed unlimited

"but now that [they have] got [their] new medication of the Kalydeco and the Kaftrio…[they] maybe needs to cut down on the amount of treat food…. I feel a bit mean, I feel a bit like since [they were] born, I've been like just eat more, eat more and giving [them] more food … but I now feel that I have to be a bit like okay no you can't have more"

"A Sponge Only Absorbs so Much Water Before It Starts Coming Out"

However, whilst physical health had improved for all families at the time of the interviews, fear and "dread" of things becoming too much again was a common experience. In terms of the sponge metaphor, the families feared the sponge being put in far too much water again. This fear was mostly around the child getting unwell and needing more treatments or hospital stays. For most of the included families, the idea of hospital stays was an unknown that they had not yet experienced.

"we tend to find around January February time when things are going round the school and [they start] to pick up a cough and if that doesn't shift you do kind of think oh no, is this going to be something that puts [them] in hospital"

Even if a hospital stay was not needed or was something the family was used to, the extra work needed to manage any signs of illness was dreaded. For example, if Pseudomonas aeruginosa is detected on a cough swab, then the daily treatment routine is intensified.

"obviously [they are] on a lot of treatments anyway just as a standard thing just to try to keep on top of it … when [they get] something additional, then its more tablets, more treatments, more ... and it's like, we seem to be able to cope with a certain amount and then once we have a few more … last year just tipped us over the edge"

Being Different from Others

The necessary treatments and concerns about their physical health make children with CF different from their peers. However, how the children felt about being different varied. One child seemed happy to be the reason for a change in their school in terms of allowing packed lunches, sharing that they were "the start of a revolution!". On the other hand, having to take Creon was experienced as embarrassing for a different child. They did not like the reactions that they got from others, particularly the shock or impressed reactions that they received from grownups when they took tablets independently. To cope with this, they tried to reduce their requirement for Creon when in public. They did this by trying to only eat food types that did not require Creon.

Note. A drawing of a young person having to take Creon at a party and others looking shocked

"I wanted to dance and do something ... except eat the food because I don't want my Creon with it"

…"yea I think sometimes [they don't] eat as much because [they know they have] got to take Creon so [other parents] will say oh [they] just had a few bits of cucumber"

Being different from others also opens the possibility of bullying. In the interviews, bullying was not raised by the children with CF themselves. However, those around them, either parents or siblings tried to protect them from it.

"so I don't really like it when people call [them] chubby, it makes me quite angry because I don't think they understand what kind of medical condition [they have]…I usually say [they are] either bloated or big boned … [they have] a medical condition and I don't think you understand that or I just walk away and say you wish, if you want to see chubby you should look in the mirror."

Themes Specific to Individual Families

The individual family narratives will now be presented. Attention will be paid to where they fit with the GETs as well as any differences between individual family members. Quotes are presented with quotation marks after the pseudonym of the person who said it. To help preserve anonymity, it was decided to not provide further quotes for each family in the appendix.

Family 1

Family 1 spoke about how they found it difficult discussing CF with others due to the condition largely being unknown in general society. This fits with the GET of being different from peers. Harper said, "I try to explain it to some people who ask me about it but in the end they…are not listening and ask questions that I have already stated about". The family felt that a more common condition such as diabetes would be easier to talk about.

The management of CF also requires devices and medications with complex medical names. Whilst these words become part of the family's typical vocabulary, others may find them hard to pronounce. This was particularly frustrating for Harper (child with CF), potentially

because it means so much to him. Specifically, he said, “another thing that was finding annoying is that … people kept calling my Creon crayon”

It is, however, not just the names of medications which are complicated but also how to take them. Specifically, Creon requires you to work out how much fat is in your food and then take the appropriate number of tablets based on this. Thus, maths as well as awareness of what you are eating is required. This can be difficult, especially for people who are not used to it. The family have learnt that not everyone is able to manage the administration of Creon despite it being a typical part of their daily life. Avery said, “….one teacher you had one year we actually ended up having to move classes because she … had such bad anxiety about it”. This experience fits with the general subtheme of having to engage with negotiations out with the family.

The family also spoke about negotiations that they had engaged in within the family unit around CF. Riley (sibling) spoke about how he had decided to take charge of packing Creon, drinks and snacks when going out as a family after experiencing a time when Creon was forgotten.

Riley: “if we don’t have the Creon then he can’t really eat and that kind of makes me sad as well because then I feel bad if I get something to eat and he doesn’t get anything”

Note. The family out walking a llama, one of the physical activities that they do together

The above quote also highlights how close knit and caring the family are. Another example of this is that Avery (mum) plans physical activities that they can all enjoy together without singling anyone out - something that both children appreciated.

Avery: “and I have tried to create it so that all three of us are doing it together...”

Harper: “and its really fun”

Avery: “...so that it’s not like right [Harper] you are now doing your physical activity on your own and it’s, like its singling you out”

However, the focus on physical activity also made the children different to their peers who preferred spending their free time gaming. This again highlights the distinction between the family and others. Avery reflected on how having to engage in physical activity due to CF resulted in them all being healthier. However, there was also an individual difference between the boys evident in the interview. Specifically, whilst Riley had grown up preferring physical activity to gaming, Harper would be happy to spend the day gaming with friends if he could.

In the interview, the family were in slight disagreement in terms of the language used. Riley and Avery spoke about how Harper’s diet was needing to become “normal” due to the modulator drugs. In their choice of language, they were suggesting that in having to be careful about how much food you eat, Harper was going to become ‘normal’. In response, Harper suggests that he is already “normal”. Instead of being “normal” by becoming more like the world, Harper is suggesting that “normal” is living with CF and perhaps being different from the world, because that is all he has known. In other words, “normal” is being the bright yellow sponge standing out on the counter.

Family 2

Note. Luca patting Charli as part of her CF care.

Family 2 spoke about how CF care felt unnatural, which they found difficult. For example, patting, to help the child clear their lungs, can feel very unnatural, especially when the child is still a little baby and seems fragile.

Luca: “the patting particularly was the one that probably, when you have a newborn baby that’s your first child, you cuddle, and you’re close and you do all those kind of things and actually what you’ve got to do as a CF parent is actually you’ve got to, beat your child, you’ve got to carry out that task”

Patting must be done a lot harder than people would generally expect and most people “wouldn’t do it hard enough”. Now that Charli (child with CF) is older, she is given a choice over who does the patting. Charli recognises that Luca (dad) does it harder but still will choose Luca to do it. Charli choosing something that she says hurts to be done by the person who does it harder also seems unnatural. However, patting does have to be done at a certain pressure to be helpful.

Charli: “and daddy does it really hard ….”

Aria: “we give you, we tend to give you the choice, we tend to give her the choice if she’s having patting of who does it, and you do your mask yourself now as well, don’t you?”

Charli: “and most of the time, I say daddy”

Note. A tub of salt in the kitchen cupboard

Another unnatural aspect of CF care is the need for extra salt in the diet. This can go against common dietary advice to limit salt intake. Such advice can seem even more judgemental when you are parenting a small child as there seems to be a greater sense of judgement around what infants and young children are eating, potentially because it is down to the parent(s)/carer(s) to manage this.

Aria: "we had to wean her early , so she was 4 months or 16 weeks we had to wean her, to get extra calories into her, so you had people looking at you because, then she was small … you were giving her food 'oh you are not meant to wean her until she is 6 months old' ….and then on top of that we had to put salt on the food…. and people would be looking at me and I'd be sat there like why don't you ask, instead of looking at me and casting assumptions".

Author's Comments and Reflections 5. *Feeling judged for parenting decisions*

I understand the worry about being judged by others just for parenting decisions. For example, always wondering what people are thinking of your interactions with your child and the random comments by strangers about whether they are wearing socks. Parenting a child with CF sounds like it could be open to so much more judgement from others as it requires an approach to feeding, for example, different than the approach preached on social media. Not related to CF, but I have read on social media how parents have reacted to other parents who have maybe started on solid food earlier or have given their child a food item that others do not perceive as acceptable. We started solid food earlier than 6 months, the age given, as we felt like our son was very keen to start eating. However, I did always feel like I needed to justify this to others.

Having to do unnatural things opens oneself up to perceived, or actual judgement from others. This can make going out in public emotionally difficult and leaves parents feeling

vulnerable about the decisions they must make to keep their child healthy. Luca spoke about how "if you dared to have a day out then you would definitely get the eyes of what are you doing". The family spoke about how the CF unit encouraged them to do things in public so that others, who they had close connections with, were aware of the family's struggles. Additionally, the family decided to make a video about CF for school to help Charli's peers understand CF.

The presence of CF can also make family life different to the life that parents had hoped for. For example, Aria spoke about how Rowan's birth was particularly hard as Charli was diagnosed with Pseudomonas aeruginosa at this time and thus was unable to meet Rowan in the way the whole family had likely dreamt of. This was made even more difficult due to all the additional care that Charli needed, alongside the care of a newborn.

Fear of Charli getting ill was expressed by both Aria and Charli. This relates to the general theme around fearing the sponge soaking up too much water again. Interestingly this fear was around different things. Aria was worried about germs around the house or when they are out together whereas Charli was worried about the school environment as another person with CF also attends the school. Specifically, people with CF are not meant to be too close to each other due to the potential spread of infection. The school environment is more Charli's own space whereas Aria (mum) has control, and responsibility, over other spaces. Charli perhaps feels safer in environments where she knows her parents are keeping her safe. Whilst Aria expressed a lot of concern about things that could harm Charli, she shared that she was not worried about school and instead was reassured that the school already had some experience of CF.

Aria: "no, but we don't, we generally don't tend to have any qualms about [them] being in school…"

Charli: “no, I, I think it worries that I walk, that I have to walk past [them] sometimes…”

The family attempted to live life despite fear. Charli still went to school and Luca and Aria continuously problem solved how Charli could have typical life experiences as safely as possible. Living life, however, did come with challenges; for example, a lot of guesswork is involved when going out to eat and having to work out the amount of Creon required. It also comes with risks and Aria feared she had got it wrong when Charli did get Pseudomonas aeruginosa. Aria said, “you try to do everything to keep them safe but like its everywhere and like she could have got it from anywhere … but because we felt like we failed her”.

Family 3

The family described being suddenly thrown into the world with CF and the sponge soaking up far too much water. They had to give up the life they were planning, e.g. literally fill in the pond they had just dug out for this new life where everything seemed threatening. Alex and Cameron (mum and dad) worked hard to negotiate their lives to completely protect Blake. Alex spoke about how she would “try to control every situation just to try and avoid all these dangers”. Blake added “stay in the same spot”.

The family recognized that how they were living meant they were missing out and not moving forwards. They also realised that they were doing everything they could and yet Blake was still getting ill. This led them to the realisation that “there’s only certain things you can control”. They, therefore, decided to challenge themselves to live life despite CF.

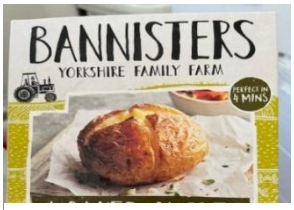
Alex: “so we, I don’t know why we had a discussion, but we were like we need to start doing something and it’s going to have to be something big so that it isn’t an easy decision to cancel it, so we booked a trip [abroad]”

Note. A holiday picture from the holiday that changed the family's outlook on CF

There were many unknowns with this holiday as it involved travelling to a different country, with different standards and a different healthcare system. It was also unknown whether they would be able to go in the first place as Blake was in hospital right before. However, the family rose to the challenge of accepting these unknowns and controlling what they could.

The holiday was described as "the best thing we have ever done". It made them realise that they could cope. They learnt that CF advice was "about balance" rather than simply black and white. This allowed them to bring things that they liked into their lives, resulting in them renegotiating how they lived with CF. For example, Alex said, "I think like bottled water [Blake] would have never had bottled water before …. soap definitely got introduced after*"*

This family also had much experience of being "stuck in hospital". This was an experience that the other families did not have. They spoke about how hospital food was extremely monotonous. Blake described how "always when I was in hospital, there would always be an option to have a jacket potato for food, always…[menu] has not changed.... do you guys have anything else apart from potatoes?"



Note. A picture of baked potatoes, something that was always on the hospital menu

Author's Comments and Reflections 6. *Hospital food.*

I remember being stuck in hospital with the hospital food. At first, I was excited to be able to order baked potatoes as I loved them. However, I did very quickly also become bored of them. I used to go off the ward to go and get strawberry milkshakes.

The lack of good, varied food in hospital meant that anything offered which was different, such as tins of tomato soup seemed so much better than it would normally. However, ensuring adequate nutrition with CF is just as important in the hospital as at home. The family negotiated this by sourcing outside food, including "absolute legendary" pizza. Whilst this is spoken positively about by Blake, there is a financial cost involved. This adds further stress to hospital stays for both Alex and Cameron.

Another part of the hospital that largely stays the same is the staff. This consistency was welcomed by the family. They were able to build up relationships with the staff and this helped them feel safe despite the potentially threatening environment. Alex said, "the bulk of the team have been the same since [Blake] was born which is nice because obviously when she does go in, it's better to have familiarity". Blake spoke about how she remembered how the nurses cared for her and how this care, however brief, resulted in her calling them "my friends".

Family 4

Note. Ash before CF was diagnosed. Eden sees the picture now and wonders how much pain Ash was in.

Ash (child with CF) and Eden (mum) seemed to have different feelings around CF. This may be due to Eden having now lived many years with the awareness and knowledge of CF. On the other hand, Ash is only slowly becoming aware of CF's effect on her life. Specifically, Eden has adapted to the medicine regime and blood tests and is witnessing Ash "developing and becoming … the crazy person you are" despite CF. She has learnt how to squeeze out the sponge.

Eden: "… I think at that time when she was a baby, it was quite difficult for us… it felt like we were sort of thrown into this kind of like pit

of despair really but then … we often say, we wish that we, I could see where she is now and go back and tell that person all those years ago that it was going to be alright”

On the other hand, Ash is only just becoming aware of how she is different from others. She is noticing the yellow colour of the sponge, and this is causing it to soak up too much water.

Note. A drawing of a blood test depicting it as a very stressful event.

Whilst Eden is now able to make downward comparisons, thinking about how CF is better than other conditions, Ash is making upward comparisons, realising how CF affects her life. For example, she realises how due to CF, she needs blood tests and more medication than others.

Ash: “I said in a thought I wish I never had CF, so I never have a blood test ever in my whole entire life”

Chapter 5– Methodological Reflections

The second aim of this study was to reflect on the feasibility of using a novel creative qualitative methodology, namely visual elicitation interviews (VEI) with the whole family. My reflections regarding this will now be presented. Quotes are presented with quotation marks. To help maintain confidentiality, the speaker is not identified. For ease, any quotes including me as the interviewer, are labelled with 'interviewer'. A more detailed list of quotes can be found in Appendix 5. Quotes are only provided from the actual recorded interview. It can be assumed that all mentions of parents and siblings refer to parents / siblings of a child with CF. A summary of my reflections on the methodology will be discussed at the end of this section.

Initial Consent

Children in this study seemed to have the power to say if they wanted to participate as also reported in a different methodological paper on VEI (Ford et al., 2017). Most of the participating families commented that their child had been excited by the creative items when they were initially approached and that this was the reason for them taking part. Similarly, a reason given for some families not taking part was that they had been through the information sheet with their child, who was not interested. Moreover, I was told by one of the participating parents that their other children did not want to participate. Thus, as stated in Ford et al. (2017), creative methods only appeal to some children.

However, it was often the parents who were the gatekeepers on whether their children could participate. It was the parents reporting in the earlier stages whether their child was interested rather than me communicating directly with the child. Moreover, in one family, the mum decided the younger sibling was too young to participate. This was a decision we had given

parents to make on an individual basis due to uncertainty from the involvement partners about the age at which a child could reflect on CF. However, previous research using creative methods has been conducted with much younger children e.g. Gray and Winter (2011). Furthermore, the family did speak about how they would discuss CF openly as a family, suggesting that we were not having completely different discussions than they were used to. The sibling was also present for the last bit of the interview but not involved. Thus, the parents were gatekeepers in terms of whose voice was heard. In retrospect, it may have been helpful asking all siblings, if they themselves were willing, to be involved.

It is also interesting that parents commented on whether their child wanted to take part rather than talking about their own views of the study. This may have been simply because parents, who were not interested, did not complete the consent to contact form and therefore, I did not have any contact with these families. It may also be due to the study being about CF and being advertised through the child's clinic, making the child with CF seem key. It could be argued that because children typically have less power within families, it is good that their voice was placed in the centre of research into a condition that they have been diagnosed with. Namely, whilst one way of viewing a condition is by looking at its impact upon a whole system through a family systems perspective (Patterson & Garwick, 1994a), the traditional concept of the medical model would situate the condition within one specific person. As the medical model is more dominant in Western society (Haegele & Hodge, 2016), it is likely that the child is used to seeing CF as a difficulty situated within them. Therefore, an interview about CF could affect them the most and consequently listening to their voice regarding participation is important.

Not all family members were present when initial information about the study was distributed because information was given to potential participants at the child's clinic visit. It can be difficult for parents to get time off work to attend their child's outpatient appointments

(Phoenix et al., 2020) and consequently it is common for just one parent to attend. School-aged siblings are also unlikely to attend during term time. Whoever was at the clinic visit therefore had the initial power in terms of whether they conveyed the study to other family members or even consented to be contacted (Lewis, 2009). Indeed, one family spoke about how they came back from the clinic and discussed it with the family member who was not present at the clinic, "when you come home from the clinic appointment and say …what you think of this". Such open communication likely depends on the style of communication already present in the family. Thus, it is unclear how much everyone knew about the interview before the pre-interview meeting took place, especially as I only communicated with the parent who completed the consent to contact form to arrange the pre-interview meeting. At the pre-interview meeting, all participants did seem to have some sort of understanding of the study. However, I did not assess this.

Unfortunately, not all family members, as identified by the family, were present in the interviews. It may have been better asking more explicitly for all family members to be involved, unless the individuals themselves did not consent. Specifically, whilst two older siblings did not want to participate and it is important that their choice was respected, the dad who did not participate due to being busy and the younger sibling who was excluded due to his age by his parents may have participated if we had directly asked them to. For example, dads are often excluded from childcare discussions and research, thus the family may have felt like his involvement was less important especially as we did not specifically ask for it. Keeping the recruitment as open as possible in order to not impose our own biases on who could participate, may have also influenced decisions around participation made by families from the global majority or of low socio-economic status. Namely, by not setting clear guidelines on who we were recruiting, they may have also assumed it was for those most

represented in research. However, despite these limitations, it is still felt that we achieved a window into family life for certain families with cystic fibrosis.

The Pre-interview Meeting

Unlike in Lewis (2009), all participants attended the pre-interview meeting. This may have been because it was set up as an essential part of the study whereas Lewis (2009) simply offered it as a possibility. Similarly to Lewis (2009), it was seen as a way of getting consent from everyone before an interview date was set up. One family decided not to participate after the pre-interview meeting as they did not want to be recorded. It may have been harder for them to say 'no', if I had sought consent on the day of the interview. The pre-interview meeting was also a place where I could fully explain the creative item task. The family who completed the mock interview suggested that this helped them understand the task which eased the anxiety present around it. This suggests that taking the time to go through the creative items in the pre-interview meeting was helpful in setting up the interview.

The pre-interview meetings were either online or in-person. This choice was given to the families. Of the participating families (n=4), two choose to do it online. I preferred doing the pre-interview meeting in-person as I found it difficult to establish rapport and get a sense of the different family members online. For example, one child was very quiet during their online pre-interview meeting, and I struggled to get a sense of what they thought of the study. I thus made time to speak with them in-person before the interview to check out their assent. Whilst it is unknown if having the pre-interview meeting in-person would have made any difference to the child on that day, I did get a feeling that the screen presented an additional barrier between us.

Creative Items

In line with previous studies, the use of creative items seemed to empower each participant as the items easily directed the interview discussion onto things important to them (Ford et al., 2017; Shaw, 2021; Terton et al., 2022). One child participant knew straight away that they wanted to take a picture of their rabbit, and the start of the interview was centred on this picture and the emotions behind it. It is unlikely that I would have gained the same level of detail, especially in terms of the emotions involved, if I had simply used a traditional interview schedule. Furthermore, the sponge metaphor would also likely not have been developed from a traditional interview structure. I made sure I also included the picture of their rabbit in this report as it was clear how important it was to them and their story.

However, parents were the gatekeepers in terms of which creative items were included. This was because we asked parents to email me the creative items for any under 16-year-olds to safeguard the family. Nothing came out during the interview that would suggest that the parents abused this power in any way. Moreover, it is likely that all items would have been sent by one person anyway as the adults in each family also did not send individual emails with their creative items. During the interviews themselves, the children would bring up other topics on their own accord, often using different objects to help them discuss these topics. Nevertheless, how much children were truly able to participate will be further discussed in the child participation section below.

In the interviews, all the creative items were laid out in the centre of the room. This was to enable everyone to be able to fully engage with them. However, this also presented challenges as both child and adult participants would get distracted by other creative items during the interview. This meant that as the interviewer, I had to regularly refocus the family on what we were talking about if I felt like there was still more to discuss. Conversely, this

was then me using my power to direct the family. It may have helped actually having the whole family sort the items at the start into an agreed upon order as done with the individual participants in Bugos et al. (2014) and then only present one item at a time. However, this would have removed some of the participants' control during the interview and felt very unnatural in practice.

The use of creative items here was different than is typical in VEI studies as they were discussed as a group. This may have resulted in discussions straying further from the items themselves than what is typically observed in VEI studies. Specifically, the family was the unit of analysis here and the creative items were a way into understanding family experiences. The family members could each reflect on each other's creative items, and this allowed different meanings to be developed from the same items within a family dynamic.

Thus, the creative items were purely stimuli from which families were able to make meaning of their experiences of CF. The items themselves were not analysed in depth in the interview, which was not our aim and was consistent with participants being able to provide creative items in any medium. Namely, the differences in type and time, from which the creative items stemmed, would have presented challenges to achieving a coherent in-depth analysis of the items.

Having a visual task to prompt discussions ensured everyone in the family could bring items that were important to them in a medium that they were used to. This was advantageous over a topic guide, even one shown to the family in advance, as it ensured we were talking about things meaningful to the family. However, participants brought varying quantities of creative items to the interview. This perhaps meant that we spent more time discussing things that are important to specific individuals. It may have been better specifying a number of items each family member had to bring.

Playfulness

Play is how children express themselves (Landreth & Homeyer, 2021) and thus to understand children it is helpful to engage in play (Koch, 2021). It can also help reduce power imbalances in interviews (Koch, 2021). The methodology allowed for playfulness through the use of creative items (Blaisdell et al., 2019). I also tried to maintain an element of playfulness throughout. This included responding to the children's jokes or imaginary statements and interacting with any toys that they showed me.

Child: "[rabbit name] want some salt?" [Laughter] Interviewer: "she prefers her dandelions" Chid: "sometimes if you get a tiny bit of salt and put it in a dandelion, it actually tastes better" Interviewer: "oh does it?"

However, parents were sometimes less on board with playfulness in the interviews. This may have been because they felt like there was a way in which their children should be behaving within research.

New Ways of Interacting with Memories

It has been proposed that VEI enable things to come to light in a new way for participants (Banks, 2001). One of the participants reflected on this.

"when you think about it, when somebody says Moana, that was it, my mind goes straight back to that because [they] did nothing but watch Moana…so some things that aren't physical things you know like creative things like a movie or whatever, it does remind you of things because you've watched it like that many times when you were there… [the interview] made me think of more things that remind of CF"

Another participant from a different family also reflected after the interview that they were not expecting to talk so much. Having pictures to prompt conversation may have enabled them to go deeper into their experiences than they would have expected to have done with a stranger. Indeed the creative items may have allowed participants to express themselves in ways that words alone cannot (Church & Quilter, 2021). On the other hand, it may not have just been the VEI that helped individuals reflect and arrive at new insights, as whole family interviews can also help families better understand themselves, their family and their experiences (Eggenberger & Nelms, 2007). Thus, the current methodology may have been even more powerful by combining family interviews with creative methodology.

Power Imbalances

Whilst the creative items may have enabled the children to engage with the topic, power still naturally existed in the room. This may be partly due to how communication and power is managed in the individual families. For example, there were some negotiations around whose voice is heard. In one interview a child started answering for their sibling, "I think it makes [them] feel responsible about stuff but…" and the mum interjected with "let [them] answer".

Parents were also still parenting during the interviews and were the gatekeepers to how their children behaved. They decided on how much playfulness to allow and whether their child could leave the room during the interview. I did not feel like it was my place to step in during these situations.

Child: "[they] made me a bracelet, hold on can I get it, I really like it?" Mum: "no" Child: "please? But that ties in with the other, it ties in" … Mum: "no because you will spend ages looking for it"

It should be noted that whilst the child was not able to go and get the bracelet and use it as a physical item in the conversation, we did discuss it and the meaning that it held for the child.

To sum, pre-existing family dynamics may have played out in the family interview and affected what was said or brought. However, in the recruited families, the dynamics did not seem to silence anyone. All participants across the families seemed able to bring up areas for discussion and whilst parents gatekept some of the things that were brought to the interview, verbal conversation seemed less affected. Nevertheless, if children or other individuals in the family are used to not being able to talk freely, they may have been more constrained in what they shared. How well the children were able to fully participate will be considered further in the next section.

Child Participation

Models of child participation show how the presence of children in an activity does not necessarily equate to them having been actively involved (Hart, 1992). This must be considered in terms of the validity of the current methodology as a family level understanding will not have been truly reached if children were unable to voice their own opinions.

I will consider child participation in this study using Hart's ladder of participation (Hart, 1992). The lowest level of Hart's ladder of participation (Hart, 1992) is when children are manipulated into doing something without any idea of the reason or possible outcomes. 'Tokenism' is slightly higher and refers to when children seem to have a voice but in reality, they have no choice. 'Consulted and informed' refers to situations in which adults have the power and are in control of the project, but the children understand what is happening and their opinions are seriously considered. The top three levels in order are: 'Adult-initiated,

Shared Decisions with Children', 'Child-initiated and Directed' and 'Child-initiated, Shared Decisions with Adults' (Hart, 1992).

In terms of this thesis, I designed the project alongside my supervisory team, who are all adults. Whilst I tried to recruit whole families to help in the design of the study initially, it was mainly adults who engaged in this. Care was taken to ensure that the children who were involvement partners, were fully aware of the project and the aims of their participation. The children's reflections on how the mock interview went were also seriously listened to. Thus, this likely fits in the 'Consulted and Informed' level of the ladder.

During the data collection phase, parents/carers often reported that it was their child's initial interest or not in the project which influenced the family's decision to participate. Thus, according to reports from adults, the children were initiating their family's involvement in the study. I ensured all children were fully aware of the project, it's aims and what their participation would involve when I met with them for the pre-interview meeting. Whilst it was assent from parents/carers that was legally required, I ensured all participating children were also given the formal opportunity to consent or not.

Families went about the creative task differently. Whilst children were unable to directly send me their items, they had clearly selected at least one item which was meaningful to them in most families. However, one child claimed to have not selected any as their family approached it as a group task. This child was asked if there was anything else they wanted to bring to the interview during the interview itself. Conversations had by the families suggested that the adults did manage and ensure the completion of the creative task. Additionally, adults made decisions around their child(ren)'s behaviour in the interview, including around additional items that their child(ren) wanted to bring into the room. Whilst children in most families were allowed to decide on the order in which topics were discussed, parents took

charge of this in one interview. Thus, the interview and creative item parts of this study likely fall under 'Adult-initiated, Shared decisions with Children' overall but the level of participation varied across families and was likely dependent on pre-existing family dynamics.

In terms of the data, a lot of the quotes presented in this thesis are from adults simply because they are more concise. However, many of the creative items presented are from the children. The photo of the rabbit was particularly important and so I ensured the rabbit was featured in this thesis rather than using my adult power to remove it. Moreover, within three out of the four interviews, the interview itself seemed to be directed by the children, especially in terms of the order in which items were discussed. In fact, I was worried at points in the interviews that it was mainly the children sharing their views rather than the adults due to the parents/carers privileging their child's voice. This likely depended on pre-existing dynamics within families.

An involvement partner family then helped with the analysis. Their help with this was agreed with the mum, though I did check with the children if they were happy to help when I arrived at their house. Me already being there and conversations they may have already had as a family, however, likely made it difficult for them to say 'no' at this time. The children also only participated for half of this activity. Thus, this was also clearly adult led, though the children's opinions were taken seriously and valued.

To sum, the current study moved between some of the levels of the ladder. It is limited that children were not more actively involved in the study design as previous research has shown that this is possible (e.g. Spencer et al., 2023). Whilst adult's quotes are perhaps more concise, children were supported to share their narratives and what was important to them. The children's contributions were greatly valued both within the interview context and then

during analysis. Specifically, all individual narratives, including those from the children, were checked to see if they fitted with the PETs and any that did not were explicitly discussed in the results.

Families as a Place of Support and Tension

Both benefits and difficulties of interviewing the family together were observable during the interviews. These will be discussed in turn.

A Place of Support

Despite my best efforts to make everything accessible, the questions I asked were sometimes difficult for the children to engage with. When this happened, parents were able to step in to help. This help was either asked for by the child or spontaneously offered by the parent. For example, when I tried asking a child about how they felt, the child responded with "err help me" directed towards the mum. The mum rephrased the question, and the child was able to give an emotion and elaborate on it.

Parents also helped their children stay focused on the interview.

> Child: "[child's name] did it so so so so so so so so good, [they] nailed it twice"
>
> Mum: "wow that's amazing, why do you think I picked that picture?"
>
> Child: "twice amazing"
>
> Mum: "why do you think I put that picture in there to talk about?"
>
> Child: "what picture?"

However, the support offered was not just from a parent to their child. Children would also help parents with parts of the story and parents would help each other. For example, one mum asked, “so we walk this, is a llama?” and a child responded with “alpaca”.

A Source of Tension

However, interviewing the family together also sometimes proved to be challenging. For example, one child seemed to find it hard to acknowledge that parts of their care had been traumatic for their parents.

Mum: “probably more traumatic for me and daddy than it was for you” [talking about patting when the child was a baby]

Child: “uh huh huh huh, I nearly cried all the time”

Mum: “you didn’t”

Child: “I did”

Mum: “you didn’t cry during patting at all…”

Child: “no you didn’t”

Mum: “yes I did, you weren’t even aware”

Moreover, another parent pointed out that their child was unaware of things because they were “just happy”. This parent seemed to be very aware that their child, who had gone to get something, was listening at the door, potentially not wanting them to hear certain things.

Mum: “…[they lead] a normal life so … that makes life easier for us as parents I think and [they are] crazy like, of on [their] own thing”

Interviewer: “does it all feel a lot more manageable and better now whereas in the past it felt scary?”

Mum: “yea, have you got them [child’s name]? Do you want to bring them in?”

These ideas align with current ideas around family coping. Indeed, despite the criticism around it, the TSC model suggests that maternal adjustment is an important part of coping with childhood chronic illness (Hocking & Lochman, 2005; Thompson, 1985) and parents may be aware of this on some level. Further, the RMFAA model suggests that families cope in crisis situations partly by challenging negative perceptions (Brown-Baatjies et al., 2008). Thus, parents may be less willing to discuss negative emotions in front of their children and their children may be less used to hearing this side of narratives around CF. However, having open discussions as a family can also have a therapeutic effect (Eggenberger & Nelms, 2007). When gaining consent for whole family interviews, both the positives and potential challenges should be discussed and weighed up with the family.

The Hermeneutic Circle

Interviewing the family together enabled family members to support each other but also made it challenging for individuals to fully engage at times. It also presented a challenge in the analysis as both the whole and parts needed consideration (Tomkins & Eatough, 2010). Indeed, as IPA is focused on the particular (Smith et al., 2021), it is usually conducted with individual interviews. However, as argued earlier, it makes sense to also consider the family as a unit of analysis.

During analysis, I was aware of the need to hold both the whole and parts in mind. Whilst we aimed to gather a family-level understanding, I also paid attention to what individual family members said and whether there were any disparities between them as suggested in Tomkins and Eatough (2010). I tried my best to display this in the write-up of the results. For example, the mum and daughter in family 4 seemed to be viewing CF from different lenses and family

1 had different views on the word ‘normal’. It is important to keep space for these individual differences as otherwise key information in the family may be lost. However, most of the time the families in this study were engaged in family-unit sense making as described in Koenig and Trees (2006). This potentially made it easier to conduct an IPA analysis than if they had been engaging in more individual sense making or incomplete sense making (Koenig & Trees, 2006). Therefore, considerations regarding the hermeneutic circle may be more difficult in different family interviews.

Relationship between IPA and Participant-Led Data Collection

In IPA, the researcher is paying attention to each individual’s lived experience and interpreting what was said based on their own understanding (Smith et al., 2021). Namely, the researcher uses their previous knowledge and experiences to make sense of the participants’ sense making. This study also wanted to prioritise the voices of families with CF (FwCF). It has been proposed that since IPA is focusing on lived experiences, these approaches complement each other well (MacLeod, 2019). However, it is important to reflect on how, and if this was achieved within this thesis.

Since I completed much analysis away from the families, I was interpreting what was said independently. I tried to stay close to the experiences of FwCF by asking a family to act as involvement partners and help with the analysis. However, their input was limited due to concerns around confidentiality as CF is a rare condition. As discussed in Humphrey and Lewis (2008), I tried to reach a “emphatic interpretation” (p.29) with my participants. Namely, I consciously paid attention to the raw data and my interpretations, ensuring that they did not stray too far apart from each other.

An interesting dilemma is also created in the write up of the study. Specifically, I wanted to remain as true to the participant data and the meanings created by individual families as possible, but as part of IPA also had to take a wider perspective. A lot of thought was therefore paid to how the results were presented. It was decided that rather than trying to fit the data into a structure, I would present the results in a way that reflected the families' own narratives. Verbatim quotes and the creative items are presented to share the participant's perspectives directly. The creative items help share some of the children's perspectives better than words alone could have done as well as adding more context generally to the quotes. Before and after the quotes, I used my own knowledge to interpret what was said on a more abstract basis.

Practicalities of the Interview

I will now discuss some practicalities (e.g. timing, location and length) that likely need consideration when planning family interviews.

Timing

When organizing the interviews, it was helpful to think about what would fit into each individual family life rather than being strict on a research timetable as also found in Leshed and Håkansson (2014). School holidays seemed to be a quieter time for the recruited families but also meant some families were away. One family signed the consent to contact form during term time but was too busy to set a pre-interview date at that time. Being flexible by offering to contact them again during school holidays enabled them to participate. Having an extended recruitment period helped with this.

Although I tried to arrange the interview at the pre-interview meeting when everyone was present, this was not always the case. For example, in one pre-interview meeting at a family

home, I ended up speaking with all participating family members separately as they were busy doing other things. As a result, I arranged the interview solely with the mum, which ended up being when the dad was busy at football. I was told that he would be back soon and was unsure as to whether to start the interview or wait till he was back. I felt like the child was keen to discuss the pictures and so decided to start the interview whilst their attention was present. This ended up being a good decision as the dad arrived much later than I had expected. He was able to join in with the interview when he arrived. This also highlights the business of family life, which poses a challenge when conducting interviews with families.

Location

As the interview took place in the family home as in Leshed and Håkansson (2014), it felt a lot more intimate than maybe it would have done elsewhere. Eggenberger and Nelms (2007) conducted their family interviews in the hospital but reflected that families may have spoken more freely away from the medical setting. Having the interviews at home, in the family's natural environment did seem to help everyone feel at ease and perhaps have time for the interview within their busy lives. The children were able to wander about and engage with other things in their house, such as eating and playing, whilst taking part in the interview. The adults also engaged in family life around the interview and would sometimes have to talk with non-participating children during the interview.

Being at home also meant that things were able to be spontaneously brought to the interview. This included toys, videos and medication equipment. For example, one child asked, "can I show my Stitch teddy?" and went upstairs to get it.

Length

The children were only able to stay engaged in the interview for short amounts of time. They would get distracted and change topic often. As the children became frequently distracted, the

adults were perhaps unable to share their narratives as they would have liked to. Leshed and Håkansson (2014) suggested having more than one interviewer present so that one interviewer could take over if a child was getting distracted with the other. It may have been easier in my study if more than one researcher was present in the interviews and may have helped everyone feel heard. However, that would have also set up a situation in which multiple conversations were happening at once, which takes away from the family narrative that we hoped to hear.

Previous research suggests that creative items are less tiring for children than traditional interviews (Shaw, 2021). However, there is still likely a limit and one of the child participants voiced that they had found the interview "very tiring". The current methodology did not just involve the children talking about their creative items, but they also had to listen to their parent(s)', sometimes lengthy, narratives which were maybe more in the style of traditional interviews. This may have been more tiring for them than if we had just spoken with the children separately. A couple of the families initially suggested that I could meet them after their clinic visit to save me travelling time. However, everyone who suggested this reflected after the interview that combining the clinic visit and interview on the same day would have been too tiring for the children.

Summary

This study has shown that whole family interviews using VEI are feasible and allow for rich, meaningful insights to be made. These insights may even be new to the individual and family themselves. Children seem to be able to say whether they want to participate in this methodology, and some enjoy the opportunity of bringing their own creative items to the interview. When using VEI with a whole family, the creative items likely purely act as stimuli for discussion as family members make sense of their experiences together rather than

being led by the researcher. Creative items inherently allow for playfulness within the interview, which can help children engage. However, pre-existing dynamics around power and communication within the family will likely persist within the interview and affect the level of child participation. Additionally, whilst interviewing family members together can provide a supportive environment, it can also change what is shared and the details in which it is shared. It can be hard for families to hear about each other's pain. Additionally, some families may find it more difficult to discuss things together and support each other than our participants did. This needs to be discussed when initially recruiting families and attended to when interpreting the results. If families struggle to engage in family-unit sense making (Koenig & Trees, 2006), then IPA analysis may be harder. Future studies using this methodology should consider how the creative items are displayed as well as the timing, location and length of the interview. It may also be helpful to state explicitly that it is hoped that all immediate family members, as defined by the family, participate if they themselves are willing. Additionally, it may be helpful for future research to provide clearer guidelines around who the research hopes to recruit as if the recruitment is too open, people may feel like it is not for them.

Chapter 6 –Discussion

This study aimed to achieve the following:

1) Develop an understanding of experiences of cystic fibrosis (CF) in family systems.
2) Reflect on the feasibility of using a novel creative qualitative methodology with the whole family.

The previous chapter has focused on the second aim and ended with a summary of my reflections. I will therefore first discuss the findings for the first aim here in terms of relevant literature. I will then discuss the strengths and limitations of the whole study before giving clinical implications and directions for future research.

Summary of Findings in Relation to the Literature

This study has identified a journey, starting from when cystic fibrosis (CF) is first diagnosed, that families with CF (FwCF) may embark on. The interviews suggest that this journey may start with fear and information overload but, in their own way, FwCF learn ways of adapting to life with CF. However, the journey is not linear due to the need for renegotiations around how the family live with CF, particularly as the child(ren) with CF (CwCF) develop(s) or their health needs change. Moreover, whilst FwCF may make sense of the condition together, individuals may experience points of the journey asynchronously. For example, the participating CwCF were not aware of the diagnosis when it was made but became aware of it as they grew older whilst their parents were thrown, with full awareness, into the CF world at diagnosis. Other findings are that CF is not well understood by the general public, potentially because it is a rare condition, and can make children stand out from their peers, which can be challenging for FwCF to cope with.

The current findings offer some support for the models of adapting to childhood chronic illness presented in the introduction. However, not all parts of the models are present in our findings. This is to be expected as we were asking FwCF to talk about what was important to them and analysing the transcripts based on their narratives rather than trying to test a theory or make the results fit into a model. Just because something was not brought up by our participants does not mean it is not an important factor for other FwCF and thus the absence of a point here cannot be used to critique existing models. I will use the three stages identified in the Adjustment Across Time Model (AATM, Huang et al., 2022), namely 'Disintegration and Vulnerability', 'Adjustment and Adaptation' and 'Recovery and Reconstruction', to structure this section. This is because it offers a simple journey that somewhat aligns with the journey we identified. However, it should be remembered that this journey is not linear.

'Disintegration and Vulnerability'

The 'Disintegration and Vulnerability' phase aligns with the idea of the sponge initially filling up. Parents of a child with CF (PoCwCF) in this study found the initial diagnosis stressful due to the amount of information given and the changes to lifestyle required, which supports previous literature on the diagnosis of CF specifically (Jedlicka-Köhler et al., 1996; Jessup et al., 2016) and other chronic conditions generally (Whittemore et al., 2012). It also supports the idea in the Resiliency model of Family stress, Adjustment and Adaptation (RMFAA, McCubbin & McCubbin, 1993) that increasing demands leads to a crisis (Brown-Baatjies et al., 2008).

However, Havermans et al. (2015) found that PoCwCF between one- and five-years of age in Belgium reported to receive 'sufficient information'. Moreover, Seddon et al. (2021) who surveyed PoCwCF who had gone through the diagnostic process at a London CF centre found that most (90%) believed that the right amount of information was given at diagnosis and 7%

even thought that too little was given. This is not necessarily in conflict with the current or abovementioned studies as the amount of information given may be both necessary and overwhelming. The current FwCF, for example, spoke about being given lots of information but none of them said that it was more than necessary. Instead, there was an idea of wanting more guidance to remove the element of guesswork and confusion as to how to manage situations such as their CwCF wanting to play in muddy puddles and leaves. How much information is seen as being sufficient may also vary between individual PoCwCF as some may want to know everything at once whilst others may prefer to pace themselves more (Grob, 2008). Thus, the health professional giving the diagnosis and initial information likely needs to be skilled in determining what the FwCF in front of them needs (Koller et al., 2024).

'Disintegration and vulnerability' can also occur at other points during the CF journey, highlighting that adapting to childhood chronic illness is not always linear. For example, if the child experiences ill health or must complete extra treatments due to the presence of Pseudomonas aeruginosa then a crisis may occur. The extra treatments themselves can be burdensome but also simply having to manage increased healthcare appointments can be difficult (Rowbotham & Daniels, 2022) and increase the demands placed on the family.

'Adjustment and Adaptation'

The 'Adjustment and Adaptation' stage in the AATM (Huang et al., 2022) links here with FwCF learning to squeeze out the sponge through negotiations. This also supports the idea in the RMFAA that families engage in family-level problem solving to cope (Brown-Baatjies et al., 2008). As can be expected, family-level problem solving looked different for each of the participating FwCF. In general, some FwCF may be able to quickly start thinking about how to live life with CF, whereas others may feel stuck, terrified of the child's health deteriorating and getting things right. This difference may be due to factors that have been identified as

being important in adaptation, such as illness severity, stressor appraisal, locus of control, access to resources, forming of new family routines, social support and family dynamics (Brown-Baatjies et al., 2008; Cave & Milnes, 2020; Crespo et al., 2013; Hocking & Lochman, 2005; Huang et al., 2022; Jaser & Grey, 2010; Szyndler et al., 2005; Thompson, 1985). It should be noted that not all FwCF may successfully learn ways of adapting to CF. Specifically, whilst all of our participants had negotiated ways of living life well with CF, other FwCF may remain in struggling or floundering states for extended periods of time (Knafl et al., 1996). I will now discuss some of the factors that may influence adaptation in turn, starting with locus of control.

Whilst the participating PoCwCF tried to control what they could, they all learnt in their own way that they could not actually control everything that their CwCF was exposed to. This was both freeing and scary. It gave them permission to relax and enjoy things like holidays but also came with awareness that their CwCF could get ill at any time. The idea behind 'locus of control' in the literature is that the more control people believe that they have over their environment, the less stress they experience (Rotter, 1966). However, our findings also suggest that trying to control everything negatively affects wellbeing, which supports ideas around perfectionism and how it can lead to distress (Flett et al., 2022). Fear of failure, which is common in perfectionism (Flett et al., 2022) may be heightened in families with a childhood chronic illness as the failure could mean the child becoming unwell and the parent potentially being judged as a 'bad parent'. However, any kind of perfectionism is unattainable (Flett et al., 2022) and it is impossible to avoid all germs. Additionally, there is more to life and child development than simply staying well. Accepting some uncontrollability and learning ways of coping with the subsequent stress may be an essential part of family wellbeing for FwCF.

CwCF may also try to control what they can. For example, one of the participating CwCF attempted to control their food intake when out in public to reduce their need for Creon. This was an attempt to avoid unwanted attention and can be seen as a negative coping strategy. Acceptance and Commitment therapy (ACT) has been proposed as a helpful intervention in improving mood and ability to stay in the present moment as well as reducing self-reported treatment barriers in people with CF (O'Hayer et al., 2024). Thus, it may be an appropriate therapy modality for CF clinics to offer, especially as attempts at reducing the need for Creon may be common for young people with cystic fibrosis. For example, some adult participants in Cave and Milnes (2020) also spoke about how they would choose to eat food which required little or no Creon when at school. ACT may also be helpful for whole FwCF as they negotiate treatment demands.

However, all participants in Cave and Milnes (2020) reported that they ultimately learnt to appreciate the positives of Creon and found ways of talking with their peers about it instead of hiding it. Similarly, most of the current participants did not actively hide CF, partly because they realised that they needed to tell people. Telling people may also be a way of maintaining some control as it means others can help or at least appreciate what you must do. However, telling people did not mean they understood, potentially because CF is so far removed from society's general awareness. As a result, FwCF may still feel separate from others.

All the participating FwCF had access to resources to help them cope. For example, they were able to choose their CwCF's school and supplement unappealing hospital diets. These things seemed to help our participants adapt to life with CF, which supports the current literature base (Brown-Baatjies et al., 2008; Thompson, 1985). Families with access to less resources, such as those experiencing poverty, may turn to spirituality and religion more as

they may feel like they have less control themselves (Banthia et al., 2007). Spirituality did not come up in our interviews and thus cannot be discussed here.

Being able to engage in everyday 'normal' activities may also be important for adaptation as it helps create a sense of normalcy and allows the family and young person to continue doing activities that they enjoy (e.g. Crespo et al., 2013; Davies et al., 2025; Snelgrove, 2015). In terms of CF specifically, Davies et al. (2025) interviewed parents of two- to five-year-olds and found a desire for 'normalcy' including grandparents being able to babysit. However, engaging in everyday activities sometimes also brought anxiety for our participants as they worried about making the right decision in terms of balancing the CwCF's health with having 'typical' experiences.

The Covid-19 lockdowns reduced everyone's ability to engage in everyday activities, which meant that FwCF had time to complete all health treatments. It maybe meant that FwCF did not feel so different from everyone else as no-one could engage in 'typical' activities anyway and so were happy to dedicate much time to the treatments. This and the fact that the Covid-19 virus was ultimately shown not to affect people with CF more than others (Colombo et al., 2020) may have meant that the CwCF's physical health was not affected. However, like in Collaço et al. (2021) anxiety was present for our participants at the time of the lockdowns and may have also been a reason for much time being spent doing the treatments well. Thus, whilst physical health may not have been affected per se, there may still have been an impact on family wellbeing. Some FwCF may also have re-entered the 'Disintegration and Vulnerability' stage because of anxieties related to the pandemic.

The Covid-19 pandemic may also have led to people in general feeling less supported socially (Sommerlad et al., 2022). Social support has been argued to be important for adaptation to childhood chronic conditions as well as family wellbeing in general (Brown-

Baatjies et al., 2008; Huang et al., 2022; Newland, 2015). Our findings further highlight the importance of social networks. For example, positive relationships with NHS staff and being able to trust others with the care of the CwCF seemed to help FwCF adapt to the condition. On the other hand, feeling judged by others made it harder for families to engage in the world. If a FwCF isolate themselves for fear of judgement, their wellbeing could reduce as others will be less available to offer support (Newland, 2015).

A supportive environment within the family system is also important (Brown-Baatjies et al., 2008; Cave & Milnes, 2020; Hocking & Lochman, 2005; Jaser & Grey, 2010; Szyndler et al., 2005; Thompson, 1985). Cave and Milnes (2020) define a supportive family as one in which CF is managed together, information is shared freely and the child with CF is supported to become independent. In my study, this kind of supportive family environment was seen across the interviews. Specifically, the FwCF spoke about how they were used to having conversations about CF with each other, parents would try and increase their CwCF's understanding of CF within the interview and CF was being managed together with no one being singled out. As no other type of family participated in this study, we cannot comment on the effect of having a supportive family here. However, the children in this study did seem to appreciate their parents' approach to managing CF as a family.

'Recovery and Reconstruction'

The final stage in the AATM is the 'Recovery and Reconstruction' stage, in which families are thought to start appreciating the positive consequences of the diagnosis (Huang et al., 2022). Family 1 realising that due to CF, they were engaging in more physical activity as a family, which is beneficial for everyone, may be an example of this stage. However, other clear examples of the whole family being in this stage did not seem to be present in the interviews. This may have been due to what the families chose to talk about. It may also be

the case that not all the families were in or had ever reached ‘Recovery and Reconstruction’. Further, different family members may move through the model asynchronously, making it harder to identify if the family had reached this stage. For example, in family 4 the mum may have been in ‘Recovery and Reconstruction’ at the time of the interview, but the child was just learning to adapt as she was noticing that she was different than her peers. Moreover, PoCwCF and potentially other family members may move between ‘Recovery and Reconstruction’ and chronic sorrow as either the benefits or drawbacks of CF become highlighted by daily life or simply conversations, such as the interview, about daily life. Chronic sorrow is a concept used in the literature to describe times when parent(s)/carer(s) go through periods of grief for their child with a chronic illness or disability (Coughlin & Sethares, 2017). It can occur when parent(s)/carer(s) see that their child is unable to do something that their peers are doing (Northington, 2000), such as playing in muddy puddles, or when the family must engage in clinic visits and hospital stays (Coughlin & Sethares, 2017), which may highlight the difference between them and other families. It may also be the case that FwCF do not need to reach ‘Recovery and Reconstruction’ at all. Specifically, the current FwCF were all able to engage in life around CF and reflect on its impact together without clearly showing they were in this stage.

Strengths

The current study had multiple strengths which will be discussed in turn. Firstly, through interviewing multiple family members together, we were able to gain a perspective that is largely missing in the literature. This is a strength because chronic illnesses, such as CF, do not just affect isolated individuals but instead exist within family systems (Prieur et al., 2021). The methodology also enabled individual family members to support each other in the telling of their experiences.

Another strength is the creative methodology employed. Through using adapted respondent – generated visual elicitation interviews (VEI), participants were able to bring what was important to them to the interview (Ford et al., 2017). Moreover, the use of the creative items seemed to empower the child's voice (Ford et al., 2017; Shaw, 2021; Terton et al., 2022) and enabled them to engage. Being able to use the creative items in the write up of the study, further helped participants' quotes and narratives come to life. Additionally, having the time to be flexible in terms of when the interviews were conducted enabled different families to take part. Doing the interviews at home allowed families to participate despite busy family lives.

Furthermore, attention has been paid to ensure that the current research is of high quality. To do this, four quality markers identified by Yardley (2000) and Yardley (2008) have been considered, including transparency. I have laid out exactly what was done in the study and my role in the double hermeneutic (Montague et al., 2020). The reflexive boxes throughout the thesis have helped with this and allowed me to experience what finding creative items may have been like for my participants. I have also reflected on both the strengths and limitations of the methodology employed.

Involvement partners were included in both designing the study and helping with the analysis. This helped ensure this study was meaningful and understandable for FwCF. Gaining their input on the analysis also greatly helped me make sense of the data, especially as I am not part of the CF community myself. This study could have been further improved by checking my interpretations of each family's interview with that family themselves. However, this was not done as the families had already given up a lot of their time for the study and we did not want to increase the study burden.

Limitations

As is true for all studies, my study also has limitations which must be considered. For example, we only recruited from one regional CF centre in the UK. This means that whilst participants are from different places within the region, only one area of the UK and one CF clinic is represented. Very different experiences may be had by FwCF from different parts of the UK under different CF teams. Additionally, as the study required a lot of input from families, it is likely that only a specific type of family was approached and consented to be included. Specifically, families who perhaps were not coping as well may not have wanted to sit down as a family and discuss it. The included families are also all white British with either high or middle approximated socio-economic status (SES). One family from the global majority did complete the consent to contact form and engaged in the pre-interview meeting. However, they were concerned about being recorded and ultimately decided to not take part. Ideas around how to enable families from the global majority are presented in the future research section of the discussion. IPA does not aim to be generalisable, but the demographics of the included families should be noted when making sense of the results.

Another possible limitation is that whilst this study sought to gain a family-level perspective, only two interviews included the whole family, as identified by participants. This was because one child was considered too young to take part in one family and the siblings and dad in another family either did not want to participate or did not have the time. This meant that despite my efforts to gain a family perspective, some voices were still missing and thus some experiences of CF in the participating families may remain unknown. However, it is still felt that the results present a window into family life. Moreover, a family level understanding was sought and the individuals, who did not participate, were still often held in

mind by the families during the interview. Ideas around how to encourage more family members to participate will be discussed in the future research section of the discussion.

Additionally, individual interviews were not conducted because the study focused on gaining a family-level perspective. Parents may have adjusted what they spoke about as they were aware that their children were listening. Different things may have also been discussed than what would have been shared in individual interviews, where solely what the individual and myself deemed to be important would have been explored. Both individual and family interviews could have been conducted, but this would have been even more burdensome for the participants and would have likely led to the duplication of information. Moreover, the two different types of interviews would have influenced each other, and thus still different information may have been discussed.

Further, participants were aware that I was being supervised by a consultant at their CF centre and knew that I was a trainee psychologist. These factors may have affected what the participants shared. No family that I contacted at any stage expressed any concern about my job role. However, my being a trainee psychologist may have meant families spoke about what they thought I'd be interested in. Some families were interested in asking me about my training after the interview, showing that they were very much aware of it. The participants were not asked about their views on the consultant being involved in the research and again no concerns were raised. However, effort was taken to distance the study from the CF centre. This was done by not offering any clinic rooms for either the pre-interview meeting or interviews. Instead, for face-to-face meetings, participants were either given the choice of their house or a room at the University.

Additionally, limitations existed around power, participation and gatekeeping. Specifically, children were not equal to their parents. Parents were asked to send me any of their children's

creative items to help safeguard the family. Additionally, parents maintained the role of parenting throughout the interviews and thus likely had ultimate power over the stories that were expanded upon and the items that were brought. Power also existed between me and the families as whilst I tried to reflect their narratives, I added my interpretations onto the data. Further, I was the one that decided which data to present in the results. This would have been improved upon by having a FwCF, including the children, fully on the research team.

In terms of the analysis, Rodham et al. (2015) identified the benefits of the whole research team engaging with the data in both written and audio format due to the influence of the researcher(s) on the findings. Due to the nature of the thesis and the time available, I was the only one that listened to the audio recordings. Whilst my supervisors had access to the transcripts, they mainly focused on the information that I pulled out for discussion. Moreover, I only brought extracts from the transcripts to the involvement partners who helped with the analysis due to the need to preserve anonymity of the participants. This influenced how the involvement partners understood the quotes, which needed to be considered when completing the analysis. I also did not provide them with any training on analysis. This perhaps made it harder for them to engage with the quotes and maintained power imbalances between me and them. Future research could maybe consider working alongside involvement partners throughout the whole analysis process and providing them with appropriate training to enable them to be able to partake in the process as equals. However, a potential drawback of providing training is that it could then influence how the family approach the analysis.

Clinical Implications

I sought to explore in depth the experiences of a few families with CF. To be able to understand the experiences in depth, breadth was sacrificed. Thus, the findings are not generalisable, but this was never the aim. This means that the clinical implications are based

on what some families may experience, and further research will likely be required to determine how universal they are in practice. The findings are likely transferable across families with a CwCF as the experiences described in this thesis resonate with previous research. The findings also potentially have some relevance for families living with other childhood chronic conditions as previous research has highlighted that experiences across conditions are not too dissimilar from each other (Hocking & Lockman, 2005).

The current study and previous research (Huang et al., 2022; Nielsen et al., 2022; Sheriffs, 2010) have shown that having positive relationships with medical staff can be important for families. For example, the CF clinic helped one of my families realise the importance of sharing the realities of CF with those around them. This seemed to help them go out into the community as a family, which is likely important for family wellbeing (Newland, 2015). On the other hand, a different family felt stuck due to fear around CF which prevented them from living their lives for a while. Thus, it may be helpful for CF staff to be aware of the difficulties that families may face, not just from a physical health perspective but also socially. Whilst poverty was not present in the present-day lives of the current participants, family self-sufficiency is also likely an important factor for staff to consider. With this awareness, CF staff could help individual families think about what would be most helpful for them. They could help families risk-assess activities from a balanced perspective and signpost to services that could help resource-wise. ACT may also be a helpful therapy modality for CF clinics to offer both families and individuals (O'Hayer et al., 2024).

Learning from others with CF may also be helpful but needs careful consideration due to the segregation that is in place to prevent cross-infection. For example, the participants in Cave and Milnes (2020) spoke about how their peers with CF were important to their learning about the condition. However, they grew up before segregation was common practice. Specifically, segregation in CF care only came in around 2007. When segregation was being

introduced, Russo (2007) interviewed young people with CF to explore their views on segregation and found that those who were used to nothing different found it easier to adapt to than more experienced patients. However, the value of peers with CF was also noted in their study. The participants in my study, expressed no desire to physically meet others with CF, likely because segregation is all they have known. Indeed, it caused anxiety for one child participant that they were at the same school as someone with CF. Nevertheless, one of the participating parents identified that they wished they could go back in time to tell themselves it would all be okay. Thus, in the time of segregation within CF care, it may be helpful for CF teams to try and fill this gap by expressing what the future may look like for the family at different stages and how families can still live family lives with case examples.

This could be done using a social media platform on which personal experiences are shared (Dale et al., 2016). Allowing comments on posts would allow families with CF to interact with each other and share further expertise, although, bullying could be a worry (Fitch et al., 2015). Risks may also be heightened if children or young people were to access the platform. Nevertheless, the benefits may outweigh any well-managed risks, as it could help reduce the sense of isolation suggested in my interviews and help families adapt to life with CF. Moreover, the National Institute for Health and Care Excellence (NICE, 2024) guidelines recommend connecting people with CF safely with each other through online platforms. Face-to-face meetings may also be beneficial for parents of children with CF (Edwards et al., 2018). For example, parents of older children could be paired with parents of infants or young children. The more experienced CF parents could then offer support, guidance and hope to the newly diagnosed family.

Additionally, it may be important for CF clinics to encourage young people with CF to become increasingly independent. In the participating families, parents naturally worked on building up their child(ren)'s knowledge of CF and its treatment within the interview.

Research suggests that this helps create a supportive family dynamic (Cave & Milnes, 2020). Additionally, being able to manage their own treatment may mean that young people with CF can take part in residential school trips, which are important in peer relationships (Gathercole, 2019) and can help create a sense of normalcy. Current literature suggests that there exists a spectrum on which PoCwCF sit in terms of how much they help and encourage their young person to become independent (Williams et al., 2007). Williams et al. (2007) also found that PoCwCF tend to take back more control if the child's health deteriorates. Thus, there may be a role for CF clinics to support these conversations and transitions within families.

Future Research

This study has shown the value of using VEI with families. As models of adjustment and adaptation to childhood chronic illnesses involve the family system (Huang et al., 2022; McCubbin & McCubbin, 1989; Thompson, 1985), it will likely be helpful for future paediatric research in general to consider using this research methodology. When doing so, clear consideration should be paid to the number of creative items each family member brings, how the creative items are presented in the interview as well as the timing, length and location of the interview. Effort should also be made to ensure the whole family, as identified by the participants, is able to participate. The benefits and potential drawbacks for the participants should be carefully discussed before any interviews are conducted. It may also be helpful to recruit a whole family to be fully part of the research team, though ethical issues around confidentiality would need consideration.

In terms of CF, it is likely important to think about how families find out about the CF diagnosis as this can remain a core memory (Havermans et al., 2015). One family in the current study spoke about how they realised the diagnosis due to reading it on the door they had been told to go through when given directions for their appointment over the phone. This

was experienced as shocking for them. However, sharing the diagnosis over the phone, a possible alternative, was considered to be insensitive by parents in Edwards et al. (2018) and Seddon et al. (2021). A hospital in London gets round this by the initial diagnosis being shared by a nurse on a home visit (Seddon et al., 2021). As regional CF centres, like the one we recruited from, cover a large geographical area, this would require a lot of travel time and thus may be impractical within the constraints of the NHS. Another possibility would be sharing the diagnosis in a general paediatric setting before inviting the family to be part of the CF clinic. However, this may not be feasible in terms of how clinics are set up. Thus, further research is needed to consider how best to share the initial diagnosis. Such research could also seek to understand how best to present and pace the initial information about CF to help it seem less overwhelming. For example, some parents in Nielsen et al. (2022) felt that they should have been admitted to hospital to help them understand the information. However, as this would be very resource heavy and disruptive for families, other ways should be considered.

Future research should also consider how best to help families negotiate how they live their lives with CF. Whilst this may look different for each family, and likely always has an aspect of uncertainty, it does not always happen quickly or naturally. Clinical research could design protocols for CF teams to have these discussions with families and balanced resources for families to take away with them. However, it is likely important that the family is empowered to make decisions about what would work for them (Huang et al., 2022; Thompson, 1985).

Living life with CF also involves navigating the necessary treatments. Whilst research is currently being conducted on how we can reduce treatment requirements, it may also be helpful designing and trailing interventions, such as ACT, aimed at changing how families perceive and respond to necessary treatment routines (Rowbotham & Daniels, 2022). This

could help reduce the chances of the sponge overfilling again. Such research would still be important in the modulator era as not everyone is able to benefit from modulators (Bierlaagh et al., 2021) and non-adherence remains an issue (Hansen et al., 2024).

Research is also needed to explore the experiences of CF in families from the global majority within the UK. It may be helpful to consider how best to help these families feel safe participating in research. From my experience, this may involve not recording the interviews but instead having two researchers present with one acting as a note taker whilst the other interviews the family. As CF is a rare condition and does not affect many families from the global majority living in the UK, it will likely be important for future research to recruit from multiple CF centres. It may also be helpful for recruitment information to be more explicit about who the study hopes to recruit. This may encourage people from more diverse backgrounds to take part. Specifically, a lack of detail could lead to families assuming the research is for those who are already often represented within the literature. For similar reasons, research with whole families should be more explicit that all family members should take part unless they do not consent themselves.

Conclusion

The CF landscape has greatly changed in recent years due to the availability of modulators as well as the widespread use of newborn screening within the UK. These changes have affected the course of CF (Nichols et al., 2021; Sims et al., 2007) and thus new research on experiences of CF is needed. Furthermore, previous research has mainly focused on individual experiences of CF, however, it is likely helpful if support around CF is provided on a family-level (Hisert et al., 2023). This study aimed to help fill in these gaps by conducting in-depth interviews using creative items with families. Due to the novel nature of the approach, we also wanted to reflect on the methodology to inform future research.

One of the participating families gave the metaphor of a sponge to represent the journey of CF. This metaphor fit with the other families' narratives and thus was used to describe the group experiential themes (GETs). Specifically, when you initially put a sponge in water, it soaks up the water until it is unable to hold any more. The initial diagnosis and subsequent change in lifestyle, which can be seen as the water, were experienced as being overwhelming by the participating families, who can be seen as the sponge. However, families learnt how to squeeze out the sponge through negotiations both within and outside of the family. These negotiations involved working out how to balance living their lives with managing CF. Re-negotiations were needed as the child with CF developed and became more independent as well as when their health needs changed, for example due to the commencement of modulator drugs. Fear remained about the sponge overfilling again. Specifically, there was fear around the child with CF becoming unwell and needing increased treatments. CF also made the child with CF and their family different from their peers. How this difference was perceived varied across and between families.

The study showed that creative qualitative interviews with the whole family are a feasible way of getting a family perspective. It shows that creative items help reduce power imbalances, but family dynamics may still influence power within the interview and the level of child participation. It also shows that how the creative items are displayed as well as the timing, length and location of the interview need consideration when utilising such a methodology. The potential positives and challenges of being interviewed as a family should also be discussed carefully with the family before consent is given. Conducting interviews at the family's home may reduce the time burden on families and allow them to engage in family life around the interview.

Clinically, this study suggests that it may be helpful to support families with CF to link up safely with each other. This could involve, for example, introducing families with older

children with CF to newly diagnosed families as those with experience could help new families anticipate what the future may look like and show them how they can still live their lives. It may also be helpful for CF centres to have social media platforms in which they share case examples (Dale et al., 2016).

Another implication is for CF clinics to support conversations and transitions in families associated with the young person becoming independent with their CF care. Additionally, the clinical team should consider the family's social landscape and wellbeing as well as the child's physical health and offer support accordingly. One family found it helpful receiving support from the CF clinic to talk with those around them about CF. This helped them reengage with their community. A different family struggled for some time trying to find the balance between protecting their child with CF from germs and living life. More research is needed to determine how best CF staff can help families work out this balance.

There is a need for further research to explore ways in which the initial diagnosis is made. Specifically, if the family is directed to a CF dedicated building for the diagnosis, then they may become aware of the diagnosis by the name on the building, which is not ideal. However, giving the information over the phone initially or at a general paediatric setting may also be limiting. Future research could also consider what information is initially given at diagnosis and whether it could be presented or paced in ways that are less overwhelming. Additionally, as this study has shown that creative qualitative interviews are feasible with families, more general paediatric research should be conducted using this methodology to increase our understanding of family experiences of childhood illness in general.

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Appendixes

Appendix 1 – PPI involvement

Mock Interview

The mock interview was done with three family members - Avery (mum) Harper (9) and Riley (12). Names have been changed to maintain confidentiality. Harper has cystic fibrosis (CF) and was diagnosed at birth. The family entered the pre-interview meeting with as much information as any participant would enter into the study. The pre-interview meeting took place at their home and started with some general chat and rapport building. I then explained the study to the family and the fact that they were hoping to do a mock interview. As part of the explanation of the study, I went through the creative items guidance sheet, and they began thinking together about the effect of CF on their lives. I stopped this conversation as it mirrored the kind of discussions that I hoped to have in the interview. One question the family had was whether their results could be used in the final study as they wanted to help other families with CF. At this point, the role they were playing in the mock interview was explained to them and how this would help inform the study. All three family members were happy to proceed, and a date was arranged for the actual interview.

The family told me that they discussed together what they wanted to talk about in the interview. They decided on food, sport and outdoor activities and the treatment plan. Avery and Harper then chose the photos whilst Riley did not see them until I brought them for the interview. I did ask Riley in the interview if there was anything else he wanted to discuss. Avery emailed me the photos in each of the categories they had decided they wanted to discuss. She wrote in her email that she was not sure if they were right and said she found it very helpful that I emailed back saying they were perfect. Avery felt that whilst her initial reaction was that it would be easier to have a direct list of

possible things to take photos of, she realizes that no list is probably better as it opens space for more data from different families.

The interview took place at the family home. It was decided to first conduct the interview and then have Domino's pizza, ordered by myself, whilst reflecting on the process. The pizza served the dual role of separating the interview from the reflections and of thanking the family for their participation. The children were very excited by the pizza and as a result pizza kept being mentioned in the interview discussion.

Before starting the interview, I took out the Dictaphones to set them up. I used two in case one failed. This initially caused some anxiety for Riley, but he said he was quickly able to pretend they were not there when the interview started. Both Riley and Harper had some fun talking into the Dictaphone before we started the interview properly. This likely also helped them relax.

As the family had already grouped their creative items, I did not start by asking them to sort the individual items. However, I did ask them which one they would like to talk about first. As some different discussions had happened during the pre-interview meeting, I referred to them in the interview where appropriate so that they were also recorded. I introduced these discussion points by asking the family if it was something they wanted to talk about.

Unfortunately, pizza arrived before the treatment plan was discussed. Whilst Avery did very much want to talk about it, the children were both tired before the pizza arrived and did not want to go back to the interview. Once the children had left, Avery spoke about life when Harper was newborn. Unfortunately, this was not recorded at the time as it was not a discussion with the family. However, upon reflection, it would be good to record all discussions that take place during the interview that help show the effect of CF.

Interestingly, Avery and Harper were more engaged in the interview than Riley seemed to be. Specifically, Riley seemed more distracted throughout. This may be because I was there due to the CF and Riley may have experience of conversations about CF happening around them. However, Riley was happy to participate and able to open up about both positive and negative effects of CF. Moreover, all families will be diverse with different people taking up different amounts of space in the interview. Thus, it is felt that too much emphasis should not be put on Alexander's distractibility. The fact that he was happy and able to open up about CF shows that siblings without CF can be included.

The family all agreed that they liked the interview taking place at their home as this helped them to feel at ease and made it easier to talk. Both children moved around the kitchen, where the interview was taking place, at different times. Being able to move around likely helped them stay engaged in the interview and this is another benefit of the interview taking place in the home environment.

After the interview, the family shared how they found the process. Both children reported that they felt at ease and comfortable during the interview. Harper also spoke about how being able to choose the photos in advance was helpful as he was able to avoid talking about certain things. Avery shared that it was a very good experience and that the communication had been good. Overall, the family agreed that they enjoyed the mock interview.

Thus, the mock interview showed that the methodology works and is acceptable. In the initial research design, it was planned to only recruit families whose child with CF is of secondary school age. This was initially decided to ensure the families were at similar life stages and to ensure the child with CF could actively take part. However, Harper was only nine years old and was able to be fully involved. Moreover, 8–14-year-olds are more

likely going to want to take part than older teenagers (especially as taking part would take away time from studying for exams). Additionally, upon reflection, it seems less important having all children with CF at the same life stage as the families will be organised differently anyway and the study is looking at the effect CF has on the family. Thus, after the mock interview, it's felt that it is best not to put restrictions on the ages of the children but to make individual decisions about whether each child is able to participate.

The interview ended up being 42 minutes long, which seemed to be a good length of time for the children to be engaged for. Whilst it was the arrival of pizza that ended the discussions rather than anything else, the children were reaching their limit and would likely not have managed much longer. Moreover, in the 42 minutes, a lot of rich data was collected.

Analysis with the Involvement Partners

Below is a summary of my meeting with some involvement partners (one family unit). This involvement partner family was the same family as in the mock interview. I also gained consent retrospectively to include them in the main study. The analysis discussion involved discussing discrete quotes from the interviews, apart from their own. I sent some of the quotes prior to my visit to the family so that they could start thinking about them. The family had looked through these quotes together the night before I came. I laid all the quotes out on the table, and we discussed whichever ones the family felt to be meaningful. This discussion lasted for about an hour when the pizza that I had ordered arrived. After pizza, I asked the family to help sort the quotes into piles. However, by this point the boys had lost interest and so it was primarily the mum who sorted the quotes.

Below are the reflections that the family made during the discussion.

- You have to be cruel to be kind (e.g. not let the child play in leaves). You have to think of solutions to the problem so that the children still get that same experience but in a safe way (e.g. cut up paper)
- You have to work out how to do daily life whilst balancing the risks. You need to adapt what you do. CF requires extreme parenting – you are just doing things that everyone does a little bit more, for example most people who have a family would not want them to be eating a load of dirt and would want their children to exercise but it just requires weighing it up a little bit more than what someone without CF would do
- Each family has to make situation-specific decisions around 'what is the limit of your fear here?'
- There may be gender difference between girls and boys. It may be more socially acceptable for boys to huff / spit in public. Age may also come into it as in older brother's class, girls are more self-conscious than the boys are. Mum would also be worried about girls being more prone to eating disorders and if they did not take the Creon, then the fat would just come out of their body, like when people with diabetes do not take their insulin to try to manage their body image.
- On differences between the needs of siblings - Sometimes it just has to be 'we are doing what is best for you and we are also doing what is absolutely best for you', so both are treated the same as both are getting their needs met but their needs may be quite different. This is quite hard for people to come to terms with as they think everyone has to be treated the same but it is not good to treat everyone just exactly the same. If one child can't eat dairy – it's not fair if the other child can't eat diary either. However, in CF, it is often harder for the sibling without CF because the sibling with CF usually gets the upper hand (e.g. eating salt / squirting you with water gun but you

can’t squirt them). Overall, later on in life, you are going to have different needs than others. If you learn this from very young age, you are going to be better off rather than creating a culture of that's not fair. Instead need to think, this is my body and this is what I want to do to look after my body. We are all born differently and need to approach things differently (e.g. schoolwork / food) differently.

- Having CF in the family, makes the children more compassionate towards other children with conditions as they realise that everyone is different.
- The CF diagnosis is hard for parents – e.g. if you are suddenly diagnosed with something yourself later in life, you have to come to terms with that yourself, that is what it's like but it's your child that has that diagnosis and you have to now shift your mindset like ‘right this is now what I have got to deal with’. It is so much harder to change how you are expecting or used to live. Parents have to suddenly follow a medical regime for somebody else – similar to anyone who gets diagnosed with something later on in life.
- When you have a child under 5, strangers think it is their responsibility to come and give you unsolicited advice. This happens to all parents. It is harder for children CF because maybe it feels like more comments or more people making comments. Also comments like ‘he sounds like he is going to cough a lung up’ or ‘he has a good pair on lungs on him’ hit differently when child has CF. It is the underlying feeling of ‘are people thinking I’m a bad parent?’
- It is very natural to feel like you have failed you children. There is no black / white with CF but you do just want a definitive answer about what you can or can’t do. Possible dangers are everywhere but you also need to a bit of a health and safety inspector. But you also need to manage that so that you do not spend your whole life feeling like a failure or worrying about things. You never know what may happen to

anyone so there is no point in being overly worried. You do as much as you can but then enjoy your life.

- Hard to know how much information around CF care that you need to share with other parents when your child goes round to their house. Hard judging their level of knowledge around things.
- As children get older, you don't worry about these things as much (e.g. Creon) as you see how capable your child becomes
- There are definite themes in the quotes e.g., things surrounding medication, things surrounding eating, things surrounding lifestyle and environment (e.g. things that you go to)
- A lot of the quotes are to do with people's feelings, namely their sadness or their guilt or them feeling like they are restricting their child's experiences. It is surprising that there are no mentions around the difficulties of getting medications.
- Mum thought it would have been good if there was an older child with CF included in the research. Mum assumed that all the participants were younger than her child with CF based on the quotes shared. This was not the case.

The following themes were developed during the meeting based on the quotes provided.

- Creon and food
- Treatment
- Missing out
- Worries and fears
- Things that deal with (e.g. schools / hospital / other parents)
- Environment

Appendix 2 – Guidance Sheet for the Creative Items

leeds children's hospital

UNIVERSITY OF LEEDS

Creative Items Guidance

This is a help sheet for the creation / collection of creative items for the interview. You do not need to look at or follow this sheet if you already have ideas of what you want to do. There are no right or wrong answers. We are interested in what you want to bring.

The interview aims to find out how Cystic Fibrosis impacts upon you as an individual and then we can explore this together with the rest of the family. The creative items will be used as a starting point for our conversations.

We would like each family member to bring at least 2 creative items. You can bring as many as you want but remember there will be limited time in the interview and so we may not be able to discuss everything. If you want, you can also decide on, create and bring your creative items as a family.

We have called them creative items because we want you to be able to use whatever you are most comfortable with. So this could be: Photos, pictures that you have found on the internet / in books, drawings, creative writing pieces, objects ….

If you are stuck on what to bring, the following questions may help you think of ideas:

- How does Cystic Fibrosis impact upon you as an individual? How does Cystic Fibrosis affect your daily life?

  _____

  _____

- What would you like to talk about in the interview? What do you think is important for us to know about the impact CF has on your life?

  _____

  _____

For example, participants in one study looking at falls and activity after a spinal cord injury (Musselman et al., 2018) took photos of:

- Cracks and gaps between paths
- Steps at cafes
- Electrical cords lying around the house
- Shoes worn before the injury and shoes worn now
- Bikes tied up as can't cycle at the moment but wishes they could

These are just ideas from a completely different study.

Remember there is no right or wrong answer. We are asking about what is important to you.

The quality of the creative item also does not matter. It is just to help prompt conversations.

If you have any questions or would like more support, please email me on Rosalind.hatton@nhs.net .

Please email all creative items that you wish to discuss to my email – Rosalind.hatton@nhs.net at least 2 days prior to your interview date. If the item cannot be emailed, please take a photo of it and email the photo. Please bring the original item to the interview if possible.

References

Musselman, K. E., Arnold, C., Pujol, C., Lynd, K., & Oosman, S. (2018). Falls, mobility, and physical activity after spinal cord injury: an exploratory study using photo-elicitation interviewing. *Spinal cord series and cases*, *4*(1), 1-10.

Appendix 3 – Extracts from Reflective Journal

Below are a few extracts from my reflective journal.

CF clinic visit 26/04/2022

Lots of meds, stress for parents, fear of coming into clinic, being different? Being physically affected by treatment (ports / steroids), parents worried about siblings as carriers and how they'd be able to cope with a CF child, control story of own illness?

Becoming a parent

I think there is a strong realisation of how hard it must be to be a parent of cf as we are constantly trying to stop my child from eating mud and dirty things but with less urgency and also just making sure they are okay / worrying when they have a cough must be a million times harder when have cf . hard enough keeping child alive as it is (or that feeling)

Reflections after a pre-interview meeting.

Difficulties of doing it online. Hard to establish rapport. Hard to really get a sense of what people think. Daughter was very quite…Also the getting consent / assent forms harder – get daughter to complete assent form at start of interview when in-person as easier to gather / understand if she is happy.

Reflections on one interview in supervision

need for parental advocacy e.g. CF healthy living plate (not all parents may feel able to do this) , dilemnas family face (normal dilemnas plus CF specific dilemnas) and how families negotiate this , families may use humour to cope with situations , whether families talk regularly with each other about CF or don't , Discussed the importance of capturing anything interesting in note

05/08/2024

Thinking about ontology and epistemology led me onto the following two papers.

(Vicary et al., 2017) made me think about this log that I am keeping. Made me realise how helpful it can be – in terms of providing an audit trail and helping people understand my thoughts and what I did. I think I need to work on making it clearer. Make sure I date it as well as including a time frame in which I accessed it and always have it open and ready when doing work on the thesis.

(Rodham et al., 2015) made me think about how I have agreed to go through the analysis with my supervisors but don't know how that will look. I was already going to raise it in supervision in terms of how it will look. This paper made me really think about how they will be able to get their understanding of the data – need to listen to the audio files?

Appendix 4 – Ethical Approval

Ymchwil Iechyd a Gofal Cymru
Health and Care Research Wales

NHS
Health Research Authority

Mrs Rosalind Hatton
Clinical Psychology Training Programme
Leeds Institute of Health Sciences, University of Leeds
Clarendon Way
LS2 9NL

Email: approvals@hra.nhs.uk
HCRW.approvals@wales.nhs.uk

13 February 2024

Dear Mrs Hatton

HRA and Health and Care Research Wales (HCRW) Approval Letter

Study title:	**What are the experiences of families in managing the dynamics around cystic fibrosis? - A qualitative photo elicitation study**
IRAS project ID:	333858
Protocol number:	N/A
REC reference:	24/NE/0016
Sponsor	University of Leeds

I am pleased to confirm that HRA and Health and Care Research Wales (HCRW) Approval has been given for the above referenced study, on the basis described in the application form, protocol, supporting documentation and any clarifications received. You should not expect to receive anything further relating to this application.

Please now work with participating NHS organisations to confirm capacity and capability, in line with the instructions provided in the "Information to support study set up" section towards the end of this letter.

How should I work with participating NHS/HSC organisations in Northern Ireland and Scotland?
HRA and HCRW Approval does not apply to NHS/HSC organisations within Northern Ireland and Scotland.

If you indicated in your IRAS form that you do have participating organisations in either of these devolved administrations, the final document set and the study wide governance report (including this letter) have been sent to the coordinating centre of each participating nation. The relevant national coordinating function/s will contact you as appropriate.

Please see IRAS Help for information on working with NHS/HSC organisations in Northern Ireland and Scotland.

How should I work with participating non-NHS organisations?
HRA and HCRW Approval does not apply to non-NHS organisations. You should work with your non-NHS organisations to obtain local agreement in accordance with their procedures.

What are my notification responsibilities during the study?

The standard conditions document "*After Ethical Review – guidance for sponsors and investigators*", issued with your REC favourable opinion, gives detailed guidance on reporting expectations for studies, including:

- Registration of research
- Notifying amendments
- Notifying the end of the study

The HRA website also provides guidance on these topics, and is updated in the light of changes in reporting expectations or procedures.

Who should I contact for further information?
Please do not hesitate to contact me for assistance with this application. My contact details are below.

Your IRAS project ID is 333858. Please quote this on all correspondence.

Yours sincerely,
Kevin Ahmed

Approvals Manager

Email: approvals@hra.nhs.uk

Copy to: *Jean Uniacke*

Appendix 5 – Quotes

Some details have been omitted to maintain confidentiality.

Family experiences

Initially get it out of packet and put it in water	"a sponge only absorbs so much water before it starts coming out ... and it's like, we seem to be able to cope with a certain amount and then once we have a few more that that last year just tipped us over the edge"
	"well [they were] born and … [were] transferred to X surgical ward ... and then we spent sort of 2 and a half, 3 weeks there …and eventually they sent us home but obviously they were still waiting for the heel prick test to come back ... and then it was like when they said alright we've got the test results back can you come in we will tell you what it is and obviously the instructions we were given were to go to the door at the side of [hospital] wing main entrance ... and it says cystic fibrosis unit on the door, that's why er are here then so" "It's like right" ..."which was a bit of a shock really"
	"and when we went it was the entrance to the CF clinic at [hospital] so we went in and obviously we got a lot of information and one of the things that always, it just always reminds me every time I look at a sponge is that it were like if you've got sponges get rid of them because obviously they absorb water"
	"… I had a caesarean, so I was still... So we had to go , we went in an ambulance, their lights were going and everything, we went to the surgical baby ward, ... you were in a box, in a little clear box, looked like you were going in a rocket to the moon and you had a drip in your arm and you had some tubes, you had a tube down you.. ."
	"we were sort of thrown into this kind of like pit of despair really"
	"we had a massive pond that I'd dug out and put this pond liner round so I had to start ripping all that out and filling that in and we had a tropical fish tank" "yea, a big tank there [points in the room]" "so we had to get rid of that"
Learn to squeeze through negotiations	"It' amazing though how quickly, because you do just think when they are tiny, how will they ever take these medicines, how will they ever swallow these tablets"
	"I find it quite, I think when [they] first started school and going to parties I felt quite sad about it because i thought oh [they] can't just go to a party, [they] can't just go for tea, but actually, I think it's like with everything when we first had [them], that the first things, when its new it's quite scary and then when you realize , you are getting so, like now [they go] for tea, and the parents …they know [they are] going to have Creon, they know that they are going to have to give it to [them] … so it is a lot easier now, so I relax a lot more"
	"so we'd been to clinic, [child] had just started walking, we went to X after clinic for a walk about , … straight over to the leaves and wants to start playing in the leaves, children with CF aren't meant to play in the leaves, there's that thing of , how do you let a small child have those experiences that every small child is having ... or do you stop [them]? What do you do? We ended up walking around with a bottle of hand gel really, washing [their] hands all the time... but yea because then they automatically want to put things in their mouths and all sorts, because there is another one of [them] holding two leaves walking along and it was between that one and other one but even he was walking behind us and he was like 'should we let [them] …, do we need to keep [them] away from those leaves"
	"[their] auntie has already cleaned out the jets in the whirlpool bath because they love the whirlpool bath …" "got to make sure that it's not stagnant"
	"it's always been a big thing pets, because couldn't have fish, you can't have a fish tank...anything like that, and then other people's pets ... it was a big decision to let [them] have a rabbit when we got her..."

	“all kids do but then when you get told, no don’t let her jump in puddles and play with leaves and stuff like this then sure enough on tv, most popular tv programmes are a pig playing” “in muddle puddles *oink*” “how did yous negotiate that as a family then, what…” “distraction” “di di di di di” “or not go where there were puddles”
	“because [they are] much more active, they’ve sort of, ... we do keep up the acapella and stuff like that but there is less pressure”
	“something that we always worried about and things like, can’t use a face flannel, can’t use sponges” “I can, I can use a flannel!” “you have to have it dried in between” “and we do dry it” “yea so that why...” “so I am allowed flannels mummy” “yea but it was something that daddy and I had to think about”
	“so it was a bit of a do we send [them] in to the [gym] and take that risk of [them] mixing with kids and [they] may pick up covid or do we not send [them] and then [they are] missing out on the exercise that is benefitting [them] in other ways so there's again there was that balance in what do we do”
	“I think [other sibling has] been a little bit resentful at times when [child with CF has] been able to have all these crisps”
	“and just stuff like, so it's like today, being out with [other sibling], [other sibling] can eat what [they want], and you are not having to think, right how many Creons [do they] need, have we got Creon with us? Have we got everything we need? And that's why it was always more difficult with [child with CF]”
	“does your sibling want to eat a handful of salt?” “mm-hm” “[sibling] would quite like to sometimes” “yea, does that cause difficulties in the house” “no, not really because don’t tell [them], just pretend”
	“[school] then tried to like make [them their] own menu ...” “it was absolutely terrible” “it was terrible, so they like, [school] would make [them] like an omelette but [school] would make it in the morning so it was then a bit rubbery by the time ...[school] then would give [them] a pot of cheese so every day, [they were] maybe just eating this pot of grated cheese and bread” “...it tasted okay, it tasted like” “and then the nurse that was giving the Creon” “the cheese was the only thing that tasted okay” “yea but [the nurse] felt a bit bad” [so got to take lunch box]
	“and then there was the time at school last year when you told your teacher that you didn’t need any Creon because you wanted to go out to play with your friends, because your friend wanted you to go out and play , so you didn’t and you would have had to walk across the school to go and get your Creon” “yes because , first aid is like here and my classroom is all the way down here, I had to run, I had to walk down to the classroom to get my water bottle, bring it back, and then filled up and then by the time I had finished my Creon it was the end of break because I had to take , to take my pack and my water bottle back” “I know [child], I remember you telling me, but they are the things you need to speak about aren’t they , because we didn’t know about that for a long time until you mentioned that and then that frustrated you quite a bit, didn’t it ? … so effectively the problem was that you are not allowed to share a cup with anyone else, are you? So, [school] used to make [them] walk down to get [their] drink and take the Creon where it were and then come back, now [they have] got [their] own cup that's labelled so [they] can take the Creon , so [they are] not having to walk backwards and forwards to drop her water bottle off”
	“Sometimes I’m allowed to do my healthy plate” “yea, that's what we asked for wasn't it” “mmhum” “we asked school about if [they] could do one for somebody with CF”
	“but the thing is though, it's really difficult when [they’ve] been in hospital for weeks at a time because obviously this menu doesn’t change that much .... they’d say oh your next IV is not due for 2 hours, it's like right get your stuff we are allowed to go, quick get, go to [pizzeria], right get back, get hooked up again coz [they’d] have them hooked” “and then while I'm on these IVs I'm like, pizza, I just get the table”

	"so I don't know if that will help people that when they are going to look around primary schools that they maybe ask for a teacher that is maybe not , emotional which sounds like, you obviously want someone who is going to be emotional to care about them … but then in another way" "but not like that stressed" "like actually somebody who is a little bit like let's just" "chop chop" "decompartmentalises things"
Renegotiating through changes	"Kaftrio because that's changed everything"
	"but now that [they have] got [their] new medication of the Kalydeco and the Kaftrio…[they] maybe need to cut down on the amount of treat food….yea, because you used to be very sort of slimmer and then as soon as [they have] got this new medication [they have] kind of like" "hey!" "increased your weight" "Hey! Hey!"
	'it is a bit because I feel a bit mean, I feel a bit like since [they were] born, I've been like just eat more, eat more and giving [them] more food and [they'd] be like…can I have some grapes or some cucumber and I'd be like oooo but like, no, I know, but I now feel that i have to be a bit like okay no you can't have more or you can't ...'
	"so [they] went on to do the capsules,...[they] found the transition quite difficult, I suppose all kids do because they are quite big,… to take the capsules but , so we still used, we used to do the Creon on , in like scoops and then we put puree on top like baby puree so when we first gave [them] capsules we put them on a spoon and then put puree on so they felt a bit similar"
	"you do know why its salt" "why what's salt?" "why you can eat salt" "cystic fibrosis" "yea, what it do?...stops you.... stops your cells...stops your cells processing salt and that's what makes you mucusy and that's why"
	"you tend to do like practicing gymnastics moves and things. Why do you think it's important to do exercise and things?" "get me some health" "what do you think it does?"
	"so who packs the Creon when you go out now?" [child points at mum] "your mum?" "and me!" .... "it's your responsibility, isn't it?" "we try to make it [child's] responsibility as much as possible really"
	"so you've then got to like divide it by the number of grams that's in what the packet is so its, it not only, its dividing it and then dividing it again by 6" "that's very fun isn't it" "do you do it all in your head?" "err, most maths I do in my head" ... "[they do] and then I check it with my phone"
	"so [they] went on a residential, … last year to London so that was then [they] had,… you had to sort of work out your own Creon when you were there" "it went very well"
	"and we always took charge really, it was only us two doing everything and you don't really want to relinquish that to anyone else when they are small, but then when we, when [sibling] went into hospital, … that was the first time anyone else had ever done [child's with CF] physio,…" "yea and [child with CF was] three …" "that was something, for me it weren't so much about relinquishing it, it was more the fact that it is a big responsibility for somebody else to take on , it's a massive part of our everyday life like"
	"and its therefore knowing so like if somebody said oh, we will babysit them, but we are going out, no then we will just not go out because that is going to be too much for you to be able to manage, you don't know what the expectations are"
	"it was even good during, post the initial covid issues when obviously we were locked away , when [gym] started back up with the social distancing, parent's couldn't go in so the instructor would come to the door and take the kids off you and take them and do it and bring them out again ...so that was good from the point of view that it forced me to let go a bit … I think it were good for [them] to have a bit of independence to actually have to go off through the building and you know"

	"I think it was much easier to control stuff as well when [they were] little before [they] went to school, you know when you look at it, when you look at how many infections, [they] had between being born and going to school...it was a lot less than the first years after going to school where all of a sudden there's hundreds of kids that [they are] mixing with and [they are] picking up things left right and centre so..."
	"… it wasn't as terrifying as I think it would have been if [they'd] have been our first baby going to school, but yea it was just handing over that responsibility, I hadn't realised how much control I had of it until we had to send [them] to school"
	"but actually I think, like now I have let go of some of that responsibility and I am happy for other people, like at parties, I don't feel like I have got to hover and be there, so it has got better like that and I know it's important for [them] to become more independent with things like her Creon and all those sort of things"
	"I think in the, in the past I tried to make more of a big deal of it as I want them to know how important it is"
Fear of the sponge soaking up again	**"...**[they have] got Pseudomonas on her cough swab, so when [they] grew that, that was a year of treatment because [they] had to go onto the growers clinic , you had to go to clinic on a different day and then we had to learn how to put together this 13 piece nebulizer that had to be, we had to prepare a liquid to go in it and we had to wash it in warm soapy water and dry it in between each one"
	"and then if they grow it again in that year afterwards, then they are hospitalized, so you spend the whole year going to clinic ... having the cough swabs and just dreading, like I know what the number is whenever they ring me"
	"were it last year?...when [they] got a few bugs that [the clinic] picked up on the cough swab that [they do] every 6 weeks in clinic...and obviously everything that [they get], obviously [they are] on a lot of treatments anyway just as a standard thing just to try to keep on top of it and ...obviously when [they get] something additional, then its more tablets, more treatments, more ... and it's like, we seem to be able to cope with a certain amount and then once we have a few more that that last year just tipped us over the edge"
	"because [they are] much more active, ...we do keep up the acapella and stuff like that but there is less pressure whereas at one point it felt like we had to be quite rigid with it, especially around covid because we were quite worried"
	"we tend to find around January February time when things are going round the school and [they start] to pick up a cough and if that doesn't shift you do kind of think oh no, is this going to be something that puts [them] in hospital , it never has so, which is great but then there's always something worrying that it's going to, one day it's going to happen and it's going to really take us back"
Bright yellow sponge standing out on drainer	"and then there was the time at school last year when you told your teacher that you didn't need any Creon because you wanted to go out to play with your friends, because your friend wanted you to go out and play , so you didn't and you would have had to walk across the school to go and get your Creon"
	"so at school everybody always has school dinner, no one is allowed a pack lunch at the school …, never in the history of time given a child…" "me" "a pack lunch no matter what allergies or conditions they have" "I am the start of a revolution"
	"most parents wouldn't have battered an eye lid about it, covid is covid, if they get it, they'll be unwell but then they will be fine but we still didn't know what was going to happen with kids with CF"
	"and we didn't have any puree with us so [they] took it with a drink and all of the grownups were like ahhhh, weren't they?" "I was embarrassed"
	"at that party, I don't know, I wasn't and I think the reason why everyone was so shocked was because we had to leave the Creon with [friend's mum], so you had a bit of

	an audience didn't you because … [friend's mum] said you should take it with a drink of water, and so she gave you the tablet didn't she, and everybody watched you, so it wasn't just the little ones...I think it was grownups who were really, they said I can't believe [child with CF] took them when I picked [child] up, I can't believe that [child] takes those without any fuss …"
	"the worry for me is that someone at school will say something horrible to you"
	"so I don't really like it when people call [them] chubby, it makes me quite angry because I don't think they understand what kind of medical condition [they have]…I usually say [they are] either bloated or big boned …[they] has a medical condition and i don't think you understand that or I just walk away and say you wish, if you want to see chubby you should look in the mirror"
	"but now that we are in the middle of it all, we see, the amount of research that is happening and all of the treatment, that we really do feel lucky, what you know, if your child is going to be diagnosed with something, its something that has got plenty of treatment options for it because we knew other kids that were in the intensive care and things at the time when we were staying in [hospital] that were that had really complicated issues but they didn't really necessarily have an understanding of so we do count ourselves to be very fortunate"

Methodological Reflections

In this section, quotes are only presented if there were several competing to be used in the main report.

Use of creative items	Child: "right, so I'm doing that one last. {one as a baby] … and I'm doing this one first"
	Parent: "I think with these photos, I tried to pick like different activities from all year round"
	Child: "this one is really funny [talking about another picture]" Interviewer: "this one? Should we talk about the creon party…"
	Parent: "I'm just thinking that second one as well, its going to be about the leaves isn't it?"
	Parent: "its connected in a similar way to [other parent's] photo" Child: "I've just realized the tiara is on wonky" Parent: "shh, so that's from that same first…"
Power imbalance within families	Child: "[friend] made me a bracelet, hold on can I get it, I really like it?" Parent: "no" …Child: "please? But that ties in with the other, it ties in" …. Child: "it does" Parent: "no because you will spend ages looking for it" Child: "no I know where it is, I know exactly where it is"
	Child: "I want to show you on my trampoline, can I?" Parent: "well not right now because we are talking with Rosie but you can go on your trampoline later definitely"
Families as a place of support	Interviewer: "how do you feel about …." Child: "err help me" Parent: "how do you feel about…" Child: "sad , especially when…"
	Interviewer: "do you want to talk about another picture?" Child: "what?" Interviewer: "do you want to talk about another picture? What's the other picture you've got?" Parent: "let's have a look...should we move on from that one?"
	Parent: "so we walk this, is he a llama?" Child: "alpaca"
	Parent: "[were they] seven?" Child: "I don't even know, I had those twilight sparkle bangs" Parent: "five going six were it?" Parent: "five going six" Child: "I had twilight sparkle bangs" Parent: "it was May 2017, so ..."