The Experiences of Parents with Cystic Fibrosis:
Staying well to Parent and Parenting to Stay Well

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ABSTRACT

Due to advances in earlier diagnosis and treatment the life expectancy of a person born with Cystic Fibrosis (CF) has increased. Therefore, more people with CF are becoming parents but the psychological understanding of CF has lagged behind advances in medical treatment. There is very limited evidence for parents and professionals to draw from when considering issues of parenting in this context. Therefore, the aim of this research was to explore *how people with CF experience being a parent,* which includes the experiences of both mothers and fathers in relation to *staying well to parent and parenting to stay well.* Nine participants were recruited purposively from the All Wales Adult Cystic Fibrosis Service. Semi-structured interviews were conducted to explore experiences of parenting, CF and the interaction between the two roles. Interpretive Phenomenological Analysis was used to interpret the participants' accounts. Three superordinate themes relating to the experiences of parents with CF were identified: *Being a parent on compressed time, Being a parent on unexpected time and Being a parent on uncertain time.* These themes were considered in relation to the limited evidence base on the psychological experience of living with CF and a systematic review of the qualitative literature relating to parenting with a chronic health condition or disability. The findings have implications for parents with CF, those considering parenting and for health professionals whose guidance needs to be grounded in an evidence-base. Implications for CF services, clinical psychology practice and future research are discussed.

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CHAPTER ONE: INTRODUCTION

1. OVERVIEW OF CHAPTER

This chapter contains an overview of the relevant literature relating to parenting with cystic fibrosis (CF). An introduction to CF, fertility, pregnancy and the relevant psychological issues associated with living with CF is presented. A systematic review of the literature exploring parenting with a chronic illness or disability (CID) is described and critiqued. Finally, there is a description of the rationale and aims for the current study.

The literature was identified using the key words and databases shown in Appendix 1, the systematic review process is described in the relevant section.

1.1 CYSTIC FIBROSIS

CF is a multisystem condition which results from a mutation on chromosome seven (Edenborough, 2002). This causes an abnormal transport of chloride, sodium and water across the epithelial cells in the body (Tzemos, 2011). The consequence is that thick, sticky mucus secretions build up in the lungs and other organs and impair the clearance of microorganisms (Cystic Fibrosis Trust, 2012). This creates an environment for bacterial colonisation which can lead to chronic airway infection, decline in lung function and respiratory failure (Edenborough, 2002; Tzemos, 2011).

CF can also cause obstruction of the pancreatic ducts leading to pancreatic insufficiency. The intestine can also be affected which results in maldigestion and malabsorption of nutrients leading to poor growth, physical weakness and delayed puberty (Edenborough, 2002). People with CF may experience a loss of appetite which is problematic due to the increased energy requirements needed to respond to infections (Cystic Fibrosis Trust, 2013b; Tzemos, 2011). Secondary complications of CF are diabetes, bone diseases such as osteoporosis and osteomalacia and cirrhosis of the liver (Balfour-Lynn & Elborn, 2007; Tzemos, 2011).

In an average CF population, 85% of people have pancreatic insufficiency, 35% of people have some glucose intolerance and 15% of people have some degree of liver impairment (Edenborough, 2002). CF is a life limiting condition and in 2011 the median age of death was 26 (Cystic Fibrosis Trust, 2013d). The most common cause of death in people with CF is respiratory failure, which results from progressive lung damage through chronic inflammation and infection (Balfour-Lynn & Elborn, 2007).

1.1.1 History of Cystic Fibrosis

The understanding, diagnosis, treatment and therefore life expectancy of people with CF has developed significantly over the last century. Many adults with CF today would not have been expected to live beyond childhood or early adulthood: therefore an understanding of this context is important when exploring the experiences of parents with CF.

CF was first recognised as a specific condition in 1938 but the CF gene was not identified until 1989 (Littlewood, 2007). The life expectancy of people with CF has improved from early childhood in the 1940s (Ahmad *et al.*, 2013; Littlewood, 2007) to 41 for children born in 2011 (Cystic Fibrosis Trust, 2013d). Treatment has progressed with the success of antibiotics in the 1940s; nutritional advice and nutritional maintenance in the 1970s; new physiotherapy techniques, enteral feeding, intravenous antibiotics and the first heart and lung transplant in the 1980s (Littlewood, 2007). A greater number of adult CF centres were also introduced in the 1980s as more people were surviving into adulthood and the need for a multi-disciplinary preventative approach was appreciated (Littlewood, 2007).

In the 1990s, it was recognised that Pseudomonas aeruginosa and Burkholderia cepacia infections were a significant problem and so people were more routinely segregated to prevent cross infection (Littlewood, 2007). Due to increasing life expectancy, the needs of people with CF have changed and more recently there has been a greater focus on managing its co-morbidities: diabetes mellitus, osteoporosis, liver disease, pregnancy and infertility (Tzemos, 2011).

1.1.2 Epidemiology

CF is an autosomal recessive genetic disorder (Cystic Fibrosis Trust, 2012). There are over 1,900 identified cystic fibrosis transmembrane conductance regulator (CFTR) mutations which are linked with CF (Ahmad *et al.*, 2013); the most common mutation is at least 50,000 years old (Walters & Mehta, 2007). The distribution of genetic mutations varies across the world and the birth prevalence is higher in some geographically or culturally isolated populations such as Ohio Amish and lower in some populations such as in Norway and Finland (Walters & Mehta, 2007).

In Caucasian populations, the birth prevalence of CF is higher than in other ethnic groups and is also more common in Western Europe than in Eastern Europe (Walters & Mehta, 2007). In the UK, the incidence of CF is thought to be about 1 in 2500 births (Cystic Fibrosis Trust, 2013b). It is difficult to estimate birth prevalence because people with mild cases may not be diagnosed until later in life (Walters & Mehta, 2007). The incidence of CF is similar across genders (Cystic Fibrosis Trust, 2013a).

1.1.3 Diagnosis

From 2007, all babies are screened for CF at birth (Cystic Fibrosis Trust, 2013c). Once babies have been screened, diagnosis is usually confirmed by a sweat test and/or a genetic mutation analysis (Cystic Fibrosis Trust, 2013c). However, children born before 2007, those born abroad or with an unusual genetic variation may not be screened at birth. For these people, clinical signs which indicate CF may prompt further exploration and diagnosis through a sweat test or genetic mutation analysis (Cystic Fibrosis Trust, 2012). Early and accurate diagnosis is considered important because delays in the onset of treatment may have long term implications for physical health and can be emotionally challenging for families (Wallis, 2007).

1.1.4 Symptoms

The symptoms of CF vary in presentation and severity. Some genotypes of CF are associated with a milder clinical phenotype which for these people means fewer symptoms and an improved prognosis. Symptoms normally develop within a child's first year of life but can develop later in adulthood (Cystic Fibrosis Trust, 2013b). However, late onset symptoms do not necessarily indicate a milder clinical phenotype. The most common symptoms in CF are

respiratory (Walters & Mehta, 2007) and may include a persistent cough, wheezing, shortness of breath and chest infections (Cystic Fibrosis Trust, 2013b). Symptoms such as malnutrition, prolonged diarrhoea and low weight can also occur as a result of pancreatic and digestive obstruction (Cystic Fibrosis Trust, 2012). Other symptoms can include sinus infections, nasal polyps and constipation (Cystic Fibrosis Trust, 2013a).

There is a gender difference in the symptoms and the prognosis of CF. Females with CF tend to have a more rapid reduction in lung function, they contract Pseudomonas aeruginosa more frequently and have a shorter life expectancy (Gage, 2012; Cystic Fibrosis Trust, 2013a). The reasons for this are unclear but one hypothesis is that increased oestrogen during menstruation may cause thickening of mucus resulting in increased infections (Gage, 2012). It is also suggested that social pressures for women to be slim may encourage poor nutrition in females (Pfeffer *et al.*, 2003).

1.1.5 Treatment

CF has many physical and psychological consequences; therefore treatment requires an individualised and multi-disciplinary approach (Batitucci, 2012; WHO, 1999). Evidence suggests that physical health in relation to pulmonary disease severity and nutrition is improved if provided by specialist CF centres despite the risk of cross-infection (Mahadeva, 1998). The overall aim of a multi-disciplinary approach is to detect pulmonary infections early, prevent chronic infections, minimise deterioration, maintain independence, optimise quality of life and maximise life expectancy (Agent & Madge, 2007). However, the treatment for CF can be challenging and time consuming (Ashish *et al.,* 2012; Jansen, 2009; Wicks, 2007) and people with CF may need to make difficult decisions about quality as opposed to length of life (Badlan, 2006).

The importance of educating people with CF and their family about the disease and current developments in knowledge has also been recognised (Ribeiro *et al.,* 2002) so that people are able to make informed decisions about their care. Specific treatments can involve: physiotherapy; exercise; antibiotics through oral, intravenous or inhalation routes; nutritional support such as a high calorie diet and enzymes to be taken with food (Cystic Fibrosis Trust, 2012; Tzemos, 2011); and isolation from others with CF, particularly those with certain germs to prevent cross-infection (Ashish *et al.,* 2012). These treatments can help to clear obstructions in the lungs,

manage lung infections and reduce progressive lung damage (Balfour-Lynn & Elborn, 2007; Cystic Fibrosis Trust, 2013a). Lung transplantation may be considered when the condition progresses to the end stage (NICE, 2006; Sands *et al.,* 2011).

1.2 PARENTING AND CYSTIC FIBROSIS

This section contains an overview of the relevant literature relating to parenting with CF, which includes: infertility in males and females; pregnancy; pregnancy decision-making; and parenting with CF.

1.2.1 Infertility in Males

Ninety eight per cent of males with CF are thought to be infertile. This is due to obstructive azoospermia related to absence or atresia of the vas deferens, epididymis and the seminal vesicles (Batitucci, 2012; Götz & Götz, 2001). Assisted conception techniques such as microscopic epididymal sperm injection (MESA) and intracytoplasmic sperm injection (ICSI) are now available (Boyd *et al.*, 2004) and are successful in 62.5% of couples in which the male has CF (Sawyer *et al.*, 2005). However, the number of men with CF opting for infertility treatment remains low; in 2001 one per cent of men (taken from a cohort study of CF registry data) undertook fertility treatment (Boyd *et al.*, 2004).

There are a greater number of studies exploring the knowledge and experience of infertility in males with CF than in females (Gage, 2012) which may be due to the increased incidence of infertility in males. Research in this area has mainly focussed on assessing reproductive knowledge, perceptions of infertility, understanding what information males want about fertility and beliefs about when and how this information should be given (Fair *et al.*, 2000; Havermans *et al.*, 2011; Sawyer *et al.*, 2005). Research suggests that the reproductive knowledge of men is improving and in recent studies almost all men knew the impact of CF on their fertility (Fair *et al.*, 2000; Havermans *et al.*, 2011; Popli *et al.*, 2009; Sawyer *et al.*, 2005). Fair *et al.* (2000) reported that in their sample, two out of three men expressed negative emotions such as shock, bewilderment and anger when learning about likely infertility, which is in contrast to an earlier study showing that men reported little distress (Sawyer *et al.*, 1998).

Research suggests that the emotional and psychological impact of discovering infertility is more challenging for older males (Sawyer *et al.*, 2005) and the significance of this information increases with age and greater lung function (Fair *et al.*, 2000; Sawyer *et al.*, 2005). The evidence indicates that males would like to be told about issues of infertility by CF professionals between the ages of 12 and 16, which is younger than average current practice found in these studies (Fair *et al.*, 2000; Havermans *et al.*, 2011; Sawyer *et al.*, 2005). Most males were told about infertility by CF professionals but found it difficult to broach the subject themselves (Fair *et al.*, 2000).

The knowledge of assisted reproductive technology in males was poorer with many men being unaware of the success rates of these treatments (Havermans *et al.*, 2011; Houser *et al.,* 2008; Popli *et al.*, 2009). Popli *et al.* (2009) suggested that awareness is low because currently few men opt for this treatment and there are also social, personal and economic factors which preclude some men from being motivated to find out about assisted reproduction.

1.2.2 Infertility in Females

Puberty is often delayed in women with CF (Arrigo *et al.,* 2003) but the evidence now suggests that most females do not have infertility problems (Edenborough *et al.*, 2008). However, an accurate picture of fertility is difficult to establish because many women with CF make the decision not to have children (Edenborough *et al.*, 2002). Infertility can occur due to direct changes in the reproductive tract or due to indirect effects of ovulation disturbance or nutritional failure (Ahmad *et al.*, 2013; Götz & Götz, 2001). Depending on the cause of infertility, treatments can be given to assist fertility such as ovulation stimulants, intrauterine insemination, in-vitro fertilisation or intracytoplasmic sperm injection (Ahmad *et al.*, 2013). The number of women with CF pursuing treatment for infertility remains relatively low; a UK study in 2001 showed that 0.5% of women with CF sought infertility treatment (Boyd *et al.*, 2004).

Studies exploring knowledge of fertility in women suggests that understanding of fertility issues was good, but some women were thought to underestimate their fertility (Gage, 2012). Women are more likely than men to initiate discussions of reproduction and infertility with healthcare professionals (Havermans *et al.*, 2011). Fair *et al.* (2000) found that women wanted discussions relating to infertility to take place between the ages of 16 and 19 and that information regarding the health consequences of pregnancy and the long term effects of pregnancy on health to be provided in verbal and written form.

Partners of people with CF who are considering having children can have genetic testing to find out if they are carriers of the most common forms of CF mutations. Couples who are both carriers can opt to have pre-implantation genetic diagnosis of their embryos (Batitucci, 2012) if they are concerned about transmitting CF.

1.2.3. Pregnancy

The first woman with CF to become pregnant was reported in 1960. This woman died six days after giving birth (Götz and Götz, 2001). However, due to an increasing life expectancy and improved treatment for people with CF, an increasing number of people are having the opportunity to become parents (Lomas & Fowler, 2010). Research using data from the United Kingdom Cystic Fibrosis (UKCF) Database found that the total number of pregnancies recorded in the UKCF database in 2001 was 105 (Boyd *et al.,* 2004). Physical health status seems to play a role in successful pregnancies; women with better lung function are more likely to become pregnant (McMullen *et al.,* 2006) and men with a self-reported milder CF (Sawyer *et al.,* 2005) or higher predicted forced expiratory volume (FEV1) are more likely to be fathers (Fair *et al.,* 2000).

Early studies exploring the effects of pregnancy on women with CF suggested a poorer prognosis for those who became pregnant (Edenborough *et al.,* 2000; Edenborough, 2001). Some of the risks in pregnancy can include: the enlarging uterus on lung functioning, relative state of immunosuppression and increased nutritional demands on the mother (Lau *et al.,* 2012). However, recent research in this area is more optimistic (Lau *et al.,* 2012; McArdle, 2011). Studies indicate that women are more likely to receive an increased use of antibiotics, monitoring and hospitalisation during pregnancy (Ahmad *et al.,* 2013; McMullen *et al.,* 2006) but there is no clear evidence suggesting that pregnancy per se is associated with increased mortality or an increased loss of lung function (Burden *et al.,* 2012; Lau *et al.,* 2012; McArdle, 2011; Thorpe-Beeston *et al.,* 2013). There is also no clear 'cut off' for estimated lung function which could predict a poorer prognosis for a woman with CF who becomes pregnant (Lau *et al.,* 2012; McArdle, 2011).

Evidence suggests that women with CF and particularly women with a poorer lung function are more likely to deliver a premature baby (Ahmad *et al.,* 2013; Edenborough *et al.,* 2000). A study exploring the outcome of pregnancies in 41 women showed that women with a FEV1 of less than 60% were more likely to deliver earlier and with caesarean section (Thorpe-Beeston *et al.,* 2013). However, longitudinal studies suggest that babies born to women with milder forms of

the disease and good lung function are not at an increased risk of health problems but there is almost no data following the health outcomes of infants beyond the neonatal period (Edenborough, *et al.,* 2008) or assessing the psychological well-being of these children. It is also speculated that factors such as sleep deprivation may impact on a mother's adherence to treatment and consequently their physical health, but this has not been explored empirically (McArdle, 2011).

1.2.4 Parenting

There is limited research exploring CF and parenting, but some authors have highlighted issues which prospective parents might consider: however this has not been explored through research. It is proposed that women think about the impact of pregnancy on health (Götz & Götz, 2000; Johannesson *et al.,* 1998; Sawyer *et al.*, 1995), the implications of CF for the parenting role (Conners & Ulles, 2005; Popli *et al.*, 2009), genetic risk to the child (Fair *et al.*, 2000; Götz & Götz, 2000; Popli *et al.*, 2009) and managing the demands of treatment with a child (Conners & Ulles, 2005; Götz & Götz, 2000; Sawyer *et al.,* 1995). Johannesson *et al.*, (1998) found that women with CF who were contemplating motherhood thought that they may become 'more motivated to keep in good shape' (p.120).

Simcox *et al.*, (2009) conducted a qualitative study to explore decision-making in pregnancy for women with CF. Four core categories were generated which included: impact, preparation, owning the decision and personal dilemmas. Two categories provide an understanding of the experiences that mothers with CF expected to encounter in their parenting role. 'Impact' included considerations about the child becoming a CF carrier or the child witnessing a deterioration of the parents' health; impact on the mother's own health, anxiety about the effects of pregnancy; and sacrifices in relation to missing out if they decided against pregnancy and missing out by being unable to do things as a family. Personal dilemmas contained issues such as selfishness, questioning motives for having children and the uncertainty about health and longevity. Whilst this information provides a useful framework for initial understanding in this area, this is the first study to explore decision-making in this population therefore further research is needed to assess the generalisability of these findings.

To the researcher's knowledge, the only published study exploring the experience of parenting in people with CF was a study conducted in Sweden and published as a conference abstract. This

was a report of survey outcomes showing that parenting was not seen to negatively impact on the management of CF or as requiring extra support from the CF clinic (Frankl & Hjelte, 2004). However, the research above suggests that this may be an over simplification and future investigation exploring the impact of parenting on the mother's health, the parenting role and the experiences of fathers is indicated (Simcox *et al.*, 2009).

1.3 PSYCHOLOGICAL FACTORS AND CF

There are psychological and emotional challenges associated with living with CF (Nobili *et al.*, 2011) and it is important to gain an understanding of these to appreciate the context in which people with CF become parents. Therefore, this section contains literature relating to the psychological issues associated with CF, psychological well-being, the presence of self-reported feelings of anxiety and depression and the psychological processes underlying living with and adjusting to CF.

CF is an unpredictable and uncertain condition (Badlan, 2006) and requires adjustment to slow deteriorations in health, in addition to managing acute exacerbations and infections (Ashish *et al.,* 2012; Britto *et al., 2002*). As described, CF is a life limiting condition and people may live with the threat of a premature death and an uncertain future (Anderson *et al.*, 2001; Badlan, 2006; Wicks, 2007). Many adults with CF would not have been expected to live until adulthood and so their future is new to them and their families (Badlan, 2006). As Wicks (2007) described 'it is difficult to plan for a time that my parents were told I would not have' (p.1271).

Treatment can be time consuming and may interfere significantly with a person's daily activities (Badlan, 2006; Götz & Götz, 2000) and can become more so as the condition progresses. Transplantation may need to be considered which usually signifies a threatening deterioration in health (NICE, 2006, Sands *et al.,* 2011). For some people, treatment can serve as a constant reminder of the disease and can result in the unwanted disclosure of CF, which is otherwise an invisible condition (Ravert & Crowell, 2008). It is thought that segregation and confinement in hospital can lead to feelings of loneliness, boredom, isolation, abandonment and anger (Duff, 2002). Although, this has been proposed on the basis of research in other populations, therefore research within the CF population is needed before any firm conclusions can be made. In addition, social support from others with CF is often lacking (Badlan, 2006) as Wicks (2007),

expressed 'the road travelled with cystic fibrosis is often deserted, devoid of like-minded companions' (p.1270).

People with CF are more likely to have a shorter stature, low weight, and pubertal delay (Tierney, 2012). Due to cultural expectations and ideals for men and women a low body weight is more likely to result in a poor body image for men but is considered more acceptable and desirable for women (Pfeffer *et al.*, 2003; Tierney, 2012). However, weight is linked to survival in CF so it is important that women are motivated to improve their nutrition through treatment (Tierney, 2012).

Much of the research relating to psychological issues in CF has focused on assessing the presence of symptoms of anxiety and depression using the Hospital Anxiety and Depression Scale (HADS) (e.g. Goldbeck, 2010; Yohannes *et al.*, 2012). The evidence is mixed with some studies showing that adults with CF 'demonstrate a remarkable psychological resilience' (Abbott, 2003, p.45) and suggestions that adults with CF have similar psychological functioning to control groups (Abbott, 2003; Anderson *et al.,* 2001; Casier *et al.*, 2010; Pfeffer & Hodson, 2003). Reliance on the HADS is problematic as it is a self-report questionnaire which cannot 'diagnose' anxiety or depression. Research exploring self-reported feelings of anxiety and depression has not been conducted with parents with CF but it would be important to consider which parents may be more vulnerable to these experiences.

Despite the challenges described, many people with CF are able to lead fulfilling and active lives (Besier & Goldbeck, 2012; Götz & Götz, 2000) and manage the interaction between treatment and other life goals (Besier & Goldbeck, 2012; Casier *et al.*, 2010). This may include meeting developmental milestones such as leaving home, finding employment and having a family (Wicks, 2007). The Cystic Fibrosis Registry (2011) showed that 70 per cent of adults over 16 years reported being in employment or studying (Cystic Fibrosis Trust, 2013d).

1.3.1 Health Related Quality of Life (HRQoL)

There has been an increasing body of research exploring quality of life in people with CF and studies are now more comparable with the development of CF specific HRQoL measures. Some studies have demonstrated an association between objective health status such as pulmonary function (usually measured by FEV1) and HRQoL or life satisfaction, however the association is

usually weak and much of the variance is accounted for by other factors (Besier & Goldbeck, 2012; Gee *et al.*, 2005; Goldbeck *et al.*, 2007). The other factors which contribute to good HRQoL in people with CF remain unclear (Yohannes *et al.*, 2012). However, it is likely to be multi factorial and research suggests correlations between age, male gender, objective health status, body mass index, treatment, hospital readmission, employment, family functioning, social support, coping and hope for the future (Ashish *et al.*, 2012; Besier & Goldbeck, 2012; Goldbeck *et al.*, 2007; Yohannes *et al.,* 2012).

Changes in health such as infection exacerbations and significant decreases in lung function (loss of 10% or more) may have a greater impact on HRQoL than severity of the health condition per se (Britto *et al.*, 2002; Goldbeck *et al.*, 2007). This suggests that it is more challenging to adapt to acute change than slow progression of the disease and it is thought that QoL does not decline until the disease becomes so severe that adaptation is very challenging (Pfeffer *et al.*, 2003). One study explored age related differences in psycho-social development in CF; a lower life satisfaction was found in the older age group compared to the adolescent group, which may be attributable to decline in health and greater treatment burden associated with increasing age (Besier & Goldbeck, 2012). However, as this study was cross-sectional it was not possible to establish what contributed to life satisfaction across the lifespan or how this sample compared to a healthy ageing population (Besier & Goldbeck, 2012). Therefore, longitudinal or prospective research is needed to explore this issue and assess QoL over the course of an individual's life and with changes to health (Besier & Goldbeck, 2012; Goldbeck *et al.*, 2007).

In summary, the evidence suggests that HRQoL is not the consequence of physical health status. Life satisfaction may be more attributable to other psychosocial factors and parenting may be one such factor. This research also indicates for parents with CF, maintaining a good HRQoL may be more challenging when confronted with acute exacerbations in health or increased treatment burden as a consequence of aging with CF.

1.3.2 Acceptance in CF

The concept of acceptance has been explored in relation to cognitive, behavioural and emotional adjustment to CF. Acceptance is considered necessary so that individuals can direct action toward important life goals despite experiencing psychological or physical challenges (Smith & Hayes, 2005; Veehof *et al.*, 2011). It is suggested that acceptance of CF is important to manage

the challenge of pursuing development goals whilst facing the demands of treatment and the uncontrollable and unpredictable aspects of CF (Casier *et al.*, 2010). Acceptance may therefore be an important psychological process for people with CF who hope to become parents and for those fulfilling this role. The limited CF specific research in this area has shown that acceptance is related to fewer depressive thoughts and feelings, lower levels of disability and a better HRQoL (Casier *et al.*, 2008; Casier *et al.*, 2010).

Badlan (2006) proposed that acceptance can occur at a cognitive level but a person may still experience difficult emotions which have an impact on life and identity. However, this hypothesis was derived from one sample; therefore further evidence in this area is needed before any firm conclusions can be made. Acceptance of the experiences associated with CF was not found to be related to disease severity, which suggests that it is the subjective experience of CF which is of greatest importance in understanding the process of acceptance (Casier *et al.*, 2010) and it is the psychosocial as opposed to the biological aspects of CF which need to be investigated. Therefore, further research in this area is needed using qualitative methods to explore the psychological processes underpinning acceptance in CF.

1.3.3 Identity and Cystic Fibrosis

There is a small body of research exploring identity in the context of CF. Understanding identity in this population is important when considering the development and threats to identity as a parent with CF.

Most people are diagnosed with CF as children and have not known a life without it (Badlan, 2006; Williams *et al.*, 2009). A common finding in research with people living with CF is their aim to be as 'normal' as possible (Ravert & Crowell, 2008). Within the framework of Erikson's psychosocial theory of development this would include 'traversing a series of psychosocial stages including exploring self-identity, establishing autonomy, finding a place in society and guiding the next generation' (Ravert & Crowell, 2008, p. 324). Parenting is an example of one important developmental milestone that some people with CF will aim to pursue. People with CF may experience ambiguity as to whether they feel 'normal' or unwell (Badlan, 2006). Many people are in employment, have family and a social life but also need to include treatment in their day. Therefore, 'normal' life may involve pursuing developmental goals in addition to treatment and feeling unwell. Life may only become not 'normal' when a person experiences an exacerbation or

an infection which shifts the person from life as usual with CF (Badlan, 2006). It is also thought that people may only feel 'different' in particular experiences in which reality does not meet anticipated trajectory or biography (Williams *et al.*, 2009). This finding suggests that for parents with CF, changes to health or adjustments to usual parenting roles could pose threats to a 'normal' parenting identity.

Some authors have explored the development of identity based on the assumption of a CF diagnosis in childhood. Williams *et al.* (2009) suggests that younger children are more likely to pursue a self-referential version of normalcy and compare their current self with their past self and find no difference. Families may revise expectations and adapt with CF to create an experience of normality and continuity for the child. These adaptations are also likely to be based on the families beliefs about CF, for example the causes, consequences, cure, controllability and trajectory of the illness (Williams *et al.,* 2009). Adolescents with CF may experience a disruption in their sense of normalcy as they develop their identity. It is suggested that adolescents with CF may engage in behaviours to create a 'normal' identity for themselves and the 'public' (Williams *et al.*, 2009). 'Normality' may be personally derived (this includes personal beliefs about CF) and socially derived from expectations and beliefs from others. Adolescents may aim to create both 'normalities' which may be for themselves directly but often also to maintain social status with peers and wider society (Williams *et al.*, 2009). Threats to normality may involve separating the CF aspect of self from the whole self as a way of preventing CF defining the person. This is in keeping with the finding in young adults which suggests that CF is just one characterisation of the self (Ravert & Crowell, 2008) and that people oscillate between 'sick' and healthy roles (Badlan, 2006). Therefore, people with CF may separate their identity as a parent and a person with CF to maintain a sense of normality as a parent.

In summary, the evidence base exploring psychological factors in living with CF is limited with conclusions based on very few studies, largely using cross-sectional designs, which do not contribute to an understanding of the development of psychological well-being or the processes underlying these mechanisms. Therefore, caution should be applied when relying on this evidence. Further research using qualitative methods is needed to contribute to an understanding of the psychological processes underlying the experiences of living with CF.

However this evidence suggests that psychosocial factors appear to contribute to overall HRQoL. Parenting is one factor which may influence subjective well-being but further research

in this area is needed. Acceptance appears to be a key psychological mechanisms for adjustment to CF and may be important in understanding the psychological experiences of parents with CF. The literature exploring identity, indicates that parents with CF may develop an identity based on life as usual and may only experience feeling 'different' as a parent when CF shifts 'normal' life, for example through a deterioration in health. However, the psychological processes underlying the experiences of parents with CF have not been explored and further investigation in this area is needed.

As described, there is only one study which has sought to understand the experiences of parents with CF. Therefore, the evidence base exploring parents with other chronic conditions or disabilities will be examined to provide an initial understanding and wider context for this study.

1.4 SYSTEMATIC REVIEW: PARENTING WITH A CHRONIC ILLNESS OR DISABILITY

A systematic review of the literature base exploring parenting with a physical health condition was conducted. The systematic review question was defined as: *How do people with a chronic health condition or disability experience parenting?* The review was undertaken to present a structured and critical synthesis of previous research aimed to provide up-to-date knowledge about parenting in this context. For ease of reading, the term chronic illness or disability (CID) will be used. This section contains an outline of the search process, a description of the included studies, a critique of the quality of studies, a narrative synthesis of research findings and implications for future research.

1.4.1 Search Strategy

To identify relevant studies the following electronic bibliographic databases were searched on the 17/01/13: PsychINFO, PsychArticles, Medline, Pubmed, ASSIA, Embase, Scopus, Sociologiocal Abstracts, Web of Knowledge and CINAHL.

1.4.2 Search Terms

Two separate searches were carried out to ensure relevant studies were identified. The search terms 'chronic illness, 'parenting', 'qualitative' and 'identity' (and all relevant variants of these terms, see Appendix 2) were combined using Boolean operators. 'Identity' was used to find studies which focused on the participant's experience of being a parent and how it may have impacted on their experience of themselves as people as opposed to the practical tasks of parenting. There were no date limits on the searches conducted.

1.4.3 Inclusion and Exclusion Criteria

As stated, the aim of the review was to explore the lived experiences of parents with a CID. Therefore qualitative studies were deemed most appropriate to meet this aim and were included in this review. The full exclusion and inclusion criteria are described below.

Inclusion Criteria:

- Qualitative research

- Experience of parenting

- Parent diagnosed with a chronic condition or physical disability

Exclusion Criteria:

- Unpublished studies / abstract only

- Studies not published in English

- Child with a CID

- Focus on understanding the child's well-being

- Parents own childhood is the focus of the study

- Other roles in addition to parenting explored

- Pregnancy / decision-making to become parent with a CID

- Evaluation of therapeutic interventions

- Focus on practical rather than psychological issues of parenting

1.4.4 Search Process

The search generated 4,636 titles which were reviewed according to the exclusion and inclusion criteria (see Appendix 2, for a table showing the outcomes of the two searches). The vast majority of the studies generated by this search explored children with a CID, which is why so many were excluded at the stage of abstract review. Sixty papers were examined in more detail, 47 were excluded on the basis of the specified criteria and 8 were excluded because they were duplicates. Grey literature and references were searched which generated a further four articles (see Figure 1 for a diagrammatic illustration of this process). In total nine studies were retained for inclusion in the review.

Figure 1: Search Process

Inclusion, exclusion criteria & key words identified

↓

Ten electronic databases used to complete two searches
PsychINFO, PsychArticles, Medline, Pubmed, ASSIA, Embase, Scopus, Sociologiocal abstracts, Web of Knowledge and CINAHL.

↓

Search 1= 2,029 hits	Search 2= 2,607 hits	→	4, 576 abstracts discarded as did not meet criteria
33 Full Text Articles Retrieved	27 Full Text Articles Retrieved		

↓

60 full text papers screened against inclusion & exclusion criteria	→	55 full text papers discarded as did not meet criteria
5 articles retained		

↓

Reference lists searched & 8 relevant articles retrieved	→	4 full text papers discarded as did not meet criteria

↓

Nine articles retained & included in the review

1.5 SUMMARY OF INCLUDED STUDIES

An in-depth description of the studies included in the review is shown in Figure 2. A short narrative account of the included studies is provided below to illustrate the design, method and characteristics of the participants in the whole body of literature.

1.5.1 Design & Method

As specified in the inclusion criteria, all studies used a qualitative design. Semi-structured interviews were the method of data collection used in all studies. Due to ethical concerns, Wilson (2007) did not ask participants' directly about motherhood in the context of HIV but found that respondents were keen to share their experiences of motherhood; therefore this became the basis of a paper. Thorne (1990) conducted a secondary analysis of qualitative data about motherhood and chronic illness which explored experience of health care relationships in the context of chronic illness.

A variety of methods were used with the single most common being grounded theory which was employed in four studies (Grue & Laerum, 2002; Ingram & Hutchinson, 1999; Thorne, 1990; Wilson, 2007). Other methods included: a phenomenological-constructivist paradigm (Duvdevany *et al.,* 2008), phenomenological method (Nelms, 2005), thematic content analysis (Hebling & Hardy, 2007), discursive approach (Radtke & Mens-Verhulst, 2001) and one study reported no explicit method (Mens-Verhulst *et al.*, 2004).

1.5.2 Sample

Three samples were selected purposively, three were recruited through personal contacts, two through attendance at clinics and one study did not specify a method. The sample size ranged from three to 30, with the mean being 14. One study recruited fathers (Duvdevany *et al.,* 2008), the remaining studies interviewed mothers only. Participants were recruited from a variety of countries including: Israel (Duvdevany *et al.,* 2008), Norway (Grue & Laerum, 2002), Brazil (Hebling & Hardy, 2007), US (Ingram & Hutchinson, 1999; Nelms, 2005), Canada (Mens-Verhulst *et al.*, 2004; Thorne, 1990), Holland (Mens-Verhulst *et al.*, 2004; Radtke & Mens-Verhulst, 2001) and the UK (Wilson, 2007).

Participants were recruited on the basis of the diagnosis of various CIDs which included: spinal cord injury (Duvdevany *et al.,* 2008), multiple sclerosis, neuromuscular diseases, cerebral palsy (Grue & Laerum, 2002), HIV (Hebling & Hardy, 2007; Ingram & Hutchinson, 1999; Nelms, 2005; Wilson, 2007), asthma (Mens-Verhulst *et al.*, 2004; Radtke & Mens-Verhulst, 2001), rheumatoid arthritis and inflammatory bowel disease (Thorne, 1990).

Figure 2: Description of Included Studies

Author	Aim	Method (Design, data collection & analysis)	Participants	Quality Rating *(Derived from Figure 3)*	Findings	Discussion
Duvdevany *et al.,* (2008). Israel	Understanding of the parenting experience of men with a spinal cord injury (SCI), how fathers with a SCI perceive their parenting, their developing relationship with their children and the children's acceptance of the father's disability.	Qualitative, phenomenological-constructivist paradigm. Semi-structured interviews exploring identity, perceptions of social attitudes, interaction with family & processionals and impact on parenting, expectations for change of social attitudes. Four stage inductive analysis.	Twelve Jewish males with a SCI, selected purposively. Six children born before injury, two at the time of injury. Four fathers had three children; seven had two children & one father had one child. Children ages ranged from birth to 21 years.	**17/20**	Grounded in assumptions of social model of disability. Fathers experienced negative attitudes toward parenting such as being viewed as unfortunate. Fear of rejection by children proved false; children's everyday familiarity with fathers' disability was shaped through open discussion & education with children.	Relative neglect of media portrayals of parenting with disability & normative aspects of this role, which undermined positive perceptions. Conflict between how disability viewed by fathers & how understood in society. This was challenged proactively by fathers. The integration of professionals with disabilities in society is needed to change attitudes.
Grue, L. & Laerum, K.T. (2002). Norway	To provide knowledge and to gain insight into the lives of physically disabled women with children. To encourage a general discussion about disability and social integration.	Qualitative, grounded theory. Part of a wider study exploring experiences of growing up with a disability in today's society. Semi-structured interviews exploring experience of being a mother and being a mother in the social relationships in	Thirty women aged 28-49 recruited through the National Birth Registry or personal contacts. Participants had one to three children all born before 1976. Women diagnosed with multiple sclerosis, neuromuscular	**15/20**	Themes included: becoming a mother (changed perception of body and 'lost gender'); a fragile motherhood (legitimising their motherhood); mother's little helper (fear of not being looked upon as a good mother by asking for children's help but	Discourse of disability does not interface easily with discourse of motherhood. Study indicates that motherhood is embedded in a discourse of disability that makes it difficult for them to 'do being ordinary'. Study is indicator of what we

		which mothers are engaged. Data analysed using grounded theory.	diseases, cerebral palsy or spinal cord injury.		importance of socialising children to help); contradictions in keeping children safe).	have achieved so far in granting disabled people equal rights in society. Main challenge is other people's 'disablism', social, maternal framing of motherhood in society.
Hebling, E.M. & Hardy, E. (2007). Brazil	To explore the feelings of HIV-positive women about motherhood, plans for the care of their child in case of death & mechanisms of defence to deal with this.	Qualitative design. Semi-structured interviews. Thematic content analysis using the concept of 'mechanism of defence'.	Twelve women aged 20-39 selected from three sites in Sao Paulo, Brazil. Four women were pregnant when diagnosed as HIV- positive, four had children after diagnosis and four decided not to have more children but had children already.	**14/20**	Women expressed right to be a mother; feelings about motherhood were intense & surpassed all negative feelings about being HIV-positive; preparing for death was difficult to express; women used compensation, denial, rationalisation and projection as mechanisms of defence.	Mothering impacts on the women's identity. Services should offer support to wider family as they were identified as surrogate parents in case of the mother's death. Physical, emotional health of women & their children needs to be recognised in services for people with HIV.
Ingram, D. & Hutchinson, S.A. (1999). US	To provide an explanatory schema that illuminates the experiences of mothers living with HIV infection.	Qualitative, grounded theory. Semi-structured interviews exploring experiences of being an HIV-positive mother. Data analysed using grounded theory.	18 HIV positive mothers aged 18-44 selected purposively. Eight were African American, nine were Caucasian and one was Latina. Aware of diagnosis for an average of three years. Participants HIV status described.	**16/20**	Stigma sets the stage for defensive mothering, three sub processes to defensive mothering: preventing the spread of HIV & stigma; preparing the children for a motherless future; and protecting self through thought control. These processes may occur simultaneously although mothers may focus more on one than another.	Literature on chronic sorrow relevant to this population. Women experience anticipatory grief & disenfranchised grief; due to stigma cannot be publically mourned; research in these areas is needed. Women need support to discuss mothering as stigma poses a problem to discuss openly. Defensive mothering is seen as protective.

Mens-Verhulst *et al*., (2004). Canada & Holland	Mothers with asthma and the influence of their mothering on managing the demands of their chronic illness.	Qualitative design, no specific method stated. Semi-structured interviews (including demographic questions) relating to experiences of being a mother, living with asthma and the interaction between the two. Thematic analysis.	Eight participants, four Dutch and four Canadian, all Caucasian aged 31-65. Selected through personal contacts & systematically through asthma clinics to provide a representative sample. All diagnosed with asthma before children born.	**16/20**	Mothering involves three 'unhealthy spirals' including: anxiety & symptoms; worsening symptoms & depression; and attempting to compensate which results in worsening symptoms.	A difference was noted between the women from Canada & Holland suggesting a need for local research. Appropriate gender specific interventions were indicated. Research needed with fathers with asthma.
Nelms, T.P. (2005). US	In what ways being HIV positive makes mothering different; how focus on mortality operates on their lives; how mothering is characterised & lived experience of being an HIV infected mother.	Qualitative, phenomenological design. Semi-structured interviews exploring experiences of being an HIV infected woman and mother; how HIV affected mothering and what issues they faced including thoughts about a time when they might not be alive. Data analysed using Giorgi method.	16 participants aged 23-54, two Hispanic, two African American, two African, one Jamaican and nine Caucasian, recruited through a service for people with AIDs in North Texas. Children aged between 20 months to early 20s (two diagnosed with HIV).	**14/20**	Mothering with HIV revealed a pattern of burden including: burden of diagnosis & health status; whether or not to reveal to children; unknown future of themselves & children; and relieving the burden through seeking support and finding meaning in HIV diagnosis.	'Spoiled identity' / stigma burdens these women & mothering. Sample unusual as participants were working, not in poverty. Challenges identified & support required for mothers to disclose diagnosis to their children. Family interventions required to help relieve every day stresses to experience less burden.
Radtke, H.L., & Mens-Verhulst, V.J. (2001). Holland	Interested in the responses of women about the experience of being a mother with asthma and whether or not this multiple positioning mattered to them.	Qualitative, discursive approach. Semi-structured interviews exploring: experience as a mother, experience of asthma, if mother role had influenced how lived with asthma and	Three white, Dutch participants aged 31-60 recruited through personal contacts. Two diagnosed as children, one as an adult (after having children). All university educated.	**17/20**	Identity as 'good mothers'; avoided being positioned by others as abnormal or sick; constructed a mothering identity which was very important & all encompassing; orientation to a new 'super mother'	Women subject to constraining discourses within three overlapping worlds of being a mother, citizen and patient. Being a mother is relevant to how women

		vice versa. Discourse analysis.	Children aged 1.5– 31 years.		who successfully combines employment & family.	live with asthma.
Thorne, S. E. (1990). Canada	The experience of raising children when chronically ill.	Secondary analysis of qualitative data originally using grounded theory. Interviews related to experience of health care relationships in the context of chronic illness. Content analysis.	16 participants, no method of recruitment stated. Four diagnosed with rheumatoid arthritis, 4 inflammatory bowel disease & 3 with scleroderma. Women had from 1-4 children. Nine had been diagnosed when children 'were small', the other seven became ill when children were 12 or above.	**9/20**	Themes identified were: performance (women compared themselves to 'normal' mothers & identified what they couldn't do); availability (feeling unreliable, despite obstacles-made self present during important occasions); fear of dependency on child; socialisation of children to illness; & healthcare services which don't accommodate chronic illness and parenting.	These women face complex challenges and perceived themselves to be inadequately supported by healthcare services. Two social constructed roles of mother and illness are incompatible. Conflicting role expectations found in healthcare professionals as well as society at large.
Wilson, S. (2007). UK, Scotland	The inter-relationship between illness and key sources of identity in the context of motherhood and HIV infection.	Qualitative design (some longitudinal data). Semi-structured interviews (when possible on two occasions) at six month intervals. Women asked about their service use in the context of practical issues such as finance and accommodation with some open ended questions. Data analysed using grounded theory.	12 women, aged 26-44, white British or other European origin, selected purposively to reflect different levels of service use. Varying HIV health status. Children aged between seven and 22.	**16/20**	Themes derived were: the importance of establishing and maintaining identities and good mothers in spite of HIV-related stigma, the threat posed to this identity by the potentially fatal nature of their condition; and their attempts to minimise the impact of the diagnosis on children.	Respondents keen to share thoughts about motherhood despite not being asked. Data interpreted as a biological disruption. Threat to identity as a result of the potentially fatal nature of condition as well as stigma. Respondents did not see their illness in individual terms importance of incorporating care giving responsibilities when supporting women.

1.6 QUALITY OF RESEARCH

The studies were critically evaluated to assess quality with the aim of gaining an appreciation of the credibility of the research findings presented. Various checklists have been developed as a bench mark to assess quality in qualitative research (e.g. CASP, 2010; Elliott *et al.,* 1999; Law *et al.,* 1998; Spencer *et al.,* 2003). The CASP criteria were used to assess quality in the current review because it provides a clear checklist which has been tested and is recommended for use in the NHS (Campbell *et al.,* 2011). The CASP (2010) criteria are shown in Appendix 3.

The studies were assessed against CASP (2010) quality indicators and summarised in Figure 3. Studies were rated with a score between zero and two on each quality indicator to give an indication of overall quality. A score of zero indicated no reported adherence, a score of one was given if the indicator was partially fulfilled and two if the study met the criteria, therefore studies were given a rating between zero and 20. Scoring has been suggested as one useful means of comparing and contrasting the articles' quality (Chenail, 2011) and it is proposed that researchers should use their judgement when assessing quality, although some grounding in qualitative methods is useful in the assessor (Spencer *et al.,* 2003). A narrative description of the quality of the whole body of research is presented below.

1.6.1 Narrative of Quality Review

The included studies were given a quality rating of between 9 and 18.

Research Aims, Methodology & Design
All but one study provided a clear statement for the aims of the research; Thorne (1990) did not specify a clear aim as the study was a secondary analysis of data, originally aimed to explore healthcare relationships. A qualitative methodology was deemed appropriate to meet the aims of all studies which broadly sought to understand the subjective experiences of parents in the context of a CID. Seven of the researchers reported some justification for their choice of design.

Recruitment & Data Collection
The method of recruitment was described in all but one study. However, only two researchers explained why the participants they had chosen were most appropriate to meet the aims of the research. Four authors made some comments about the representativeness of their sample but

this was only discussed in depth in two cases, one of which considered their study not to be representative and the other expressed that their sample reflected a range of important demographics found in the population. In three studies, participants were recruited through personal contacts but the position of the researcher in relation to these contacts was only acknowledged in one study. The reporting of data collection methods, process and the type of data collected was transparent in most studies.

Reflexivity

The relationship between the researcher and participants was discussed in one study and this was reflected on in relation to the data generated by participants. The epistemological position of the research method was described in three studies.

Ethical Issues

Issues of consent, confidentiality and anonymity were discussed in almost all studies. However, the reporting of ethical approval was only found in four of the papers.

Data Analysis

A description of the process of data analysis was given in all studies and findings were supported by quotes from participants. Only two studies reported using methods of triangulation during the data analysis process and the researcher's position was only acknowledged in one study.

Findings and Value of the Research

All of the papers described clear findings, which were presented as themes supported by quotes from participants. The majority of the studies demonstrated the value of the research through reporting their findings in relation to theory and clinical practice. One study commented on their findings in relation to current policy. One author acknowledged the limitations of their study in relation to the representativeness of the sample, generalizability of the findings and the position of the researcher in constructing these findings. No studies reported using criteria to ensure the quality of their research.

In the majority of these studies, the authors provided a description of the method of recruitment, data collection and analysis. However, most studies lacked a justification or exploration of the representativeness of their sample and the utility of the method employed. In addition, issues of credibility and rigour were rarely addressed. Therefore, further research is

needed in this area to contribute to the development of an evidence base built on high quality qualitative research. With these limitations in mind, a synthesis of the theoretical and clinical findings is provided below.

Figure 3: Summary of Quality Review

REFERENCE	AIMS Was there a clear statement of the aims of the research?	METHODOLOGY Is a qualitative methodology appropriate?	DESIGN Was research design appropriate to address aims of the research?	RECRUITMENT Was recruitment strategy appropriate to address the aims of the research?	DATA COLLECTION Were the data collected in a way that addressed the research issue?	REFLEXIVITY Has the relationship between researcher and participants been adequately considered?	ETHICS Have ethical issues been taken into account?	DATA ANALYSIS Was the data analysis sufficiently rigorous?	FINDINGS Is there a clear statement of findings?	VALUE OF RESEARCH Does researcher discuss contribution of research?	TOTAL SCORE
Duvdevany et al., (2008). Israel	Clear aim, importance & relevance stated. (2)	Aim of research to explore in-depth understanding of parenting experiences. (2)	Justification & explanation for design given. (2)	Explanation of how participants selected & justified discussion of recruitment process. (2)	Data collection process was transparent & justified. No discussion of setting of interviews. (1)	Epistemological position of research method stated but researchers position not stated. (0)	Ethical issues considered & approval Sought. (2)	Transparent description of data analysis, emergence of themes, & quotes included, triangulation reported. Researcher position not discussed. (1)	Explicit description of findings, discussion of credibility discrepant results explored & discussed in relation to research question. (2)	Research discussed in relation to original research question, theory & practice. (2)	16/20
Grue, L. & Laerum, K.T. (2002). Norway	Aim of research, relevance & importance stated in context of comprehensive literature review. (2)	Aim was to explore experiences of being mother with a disability - appropriate to qualitative methodology. (2)	Justification of design & appropriate in relation to aims of research. (2)	Recruitment method was transparent & reasons for selection stated. Some recruited through personal contacts. (1)	Method and justification of data collection & setting was transparent. (2)	Researcher's position as a personal contact was acknowledged but not explored further. (1)	Ethical issues of consent consider but ethical approval not reported. (1)	Short description of analysis, data presented to support findings with exceptions noted, researcher did not	Findings are explicit, adequate evidence for & against arguments & discussed in relation to research question.	Findings discussed in relation to policy, theory & practice. (2)	15/20

									examine own role in process. (1)	Credibility of findings not reported. (1)		
Hebling, E.M. & Hardy, E. (2007). Brazil	Clear aim with reference to short review of literature. (2)	Feelings of HIV-positive women about motherhood appropriate to qualitative methodology. (2)	Designed reported & appropriate but not justified. (1)	Clear description of recruitment method & discussion of process. (2)	Setting of data collection, process, method and form of data clear. Saturation of data discussed. No justification for the method given. (1)	Relationship between researcher and participants was not discussed. (0)	Ethical approval sought & issues of consent discussed. (2)	Short description of data analysis, supported by data & quotes to illustrate. Contradictory data taken into account. Researcher's position in data analysis not reported. (1)	Explicit findings & discussed in relation to the aims. No discussion of credibility of findings. (1)	Findings discussed in relation to theory & practice. (2)	**14/20**
Ingram, D. & Hutchinson S.A. (1999). US	Clear study aim & importance & relevance stated. (2)	Purpose was to generate a substantive theory about experiences which is appropriate to qualitative methodology. (2)	Explanation & justification of the design. (2)	Recruitment method was reported no discussion of why participants were selected. (1)	Setting of data collection discussed but not specified, method of data collection & form of data collected was made explicit. (1)	Epistemological position of research method stated. Researcher's position was not discussed. (1)	University approval sought, issues of confidentiality & anonymity reported. (2)	Transparent description of data analysis. Findings grounded in quotes. Researcher's own role not examined. (1)	Finds are explicit, Triangulation of data analysis reported to ensure credibility, findings discussed in relation to research question & evidence for and against discussed. (2)	Implications for clinical practice & future research identified. (3)	**16/20**

Mens-Verhulst *et al.*, (2004). Canada & Holland	Clear study aim, relevance & importance based on short literature review. (2)	Qualitative methodology deemed appropriate & justified. (2)	Design was justified by researcher. (2)	Description of recruitment process & reasons for sample selection was justified. Participants recruited via personal contacts. (1)	Setting of data collection described, the method, process & form of data collected was clear. (2)	Researcher's position as a personal contact was acknowledged but not discussed further. (1)	Confidentiality discussed. Ethical approval was not reported. (1)	Short description of data analysis, data is presented to support findings, contradictory data is taken into account. Not clear how researcher's own role was considered in analysis. (1)	Evidence for & against findings is discussed. Credibility managed through 'consensus coding'. Findings discussed in relation to aims. (2)	Theory & practice implications reported. Areas for future research considered. (2)	**16/20**
Nelms, T.P. (2005). US	Clear study aim based on review of literature. (2)	Qualitative method justified as exploring subjective experiences of mothers with HIV. (2)	Clear justification for design which is discussed in relation to aims of research. (2)	Clear description of recruitment processes but no discussion of appropriateness. (1)	Setting for data collection described. Method & form of data collected was reported. (2)	Relationship between researcher & participants not discussed. (0)	Ethical approval sought. Anonymity discussed. (2)	Clear description of data analysis & how categories were formed. Findings grounded in quotes. Researcher's own role not discussed. (1)	Findings are explicit & differences between participant are described. No credibility of findings reported. (1)	Implications for theory & practice identified. Areas for future research not suggested. (1)	**14/20**
Radtke, H.L.., & Mens-Verhulst, V.J. (2001) Holland	Clear study aim based on review of literature. (2)	Qualitative method appropriate & justified. (2)	Design was described & justified. (2)	Description of recruitment method & representativeness of sample. Participants	Setting of interview described. Method, process & form of data	Reflexive account, relationship between participants & researcher	Ethical approval not reported. Anonymity discussed. (1)	In depth description of data analysis. Findings grounded in data.	Findings are explicit, discrepancies are reported & findings are	Implications for theory and practice identified, & applicability to other	**17/20**

Author/Year/Country	Aim	Method	Design	Recruitment	Data collection	Relationship	Ethical issues	Data analysis	Findings/Credibility	Value/Implications	Score
				accessed through personal contacts. (1)	collected was reported. (2)	was discussed. (2)		Contradictory data taken into account. (2)	in relation to research question. Credibility of findings discussed but methods to ensure this e.g. triangulation not reported. (1)	populations is discussed. (2)	
Thorne, S. E. (1990). Canada	Broad study aim which was derived from secondary analysis of data. (1)	Qualitative method appropriate to meet aims of research & justified. (2)	Research design deemed appropriate but little justification of design. (1)	Recruitment method not specified, discussed or justified. (0)	Method of data collection was described & form of data collected was specified. No rationale for method of data collection in relation to research question. (1)	Relationship between researcher & participant was not specified. (0)	Ethical approval was not reporter & ethical issues were not discussed. (0)	Clear description of data analysis process. Data is presented to support findings. Researcher's own role was not explored. (1)	Clear statement of findings with data to support findings. No discussion of credibility of findings. (1)	Implications for theory and practice & future research identified. (2)	9/20
Wilson, S. (2007). UK, Scotland	Clear aim of research. (2)	Qualitative method appropriate & justified. (2)	Design stated & justified. (2)	Description of recruitment process & justification for selection of participants. (2)	Setting of data collection specified. Description of data collection process & form of data collected. (2)	Relationship between researcher & participants was not reported. (0)	Ethical issues discussed in relation to confidentiality and anonymity. Ethical approval reported. (2)	Description of data analysis. Data presented to support findings. Researcher's own role not explored. (1)	Findings are explicit, discrepancies identified. Explored in relation to research question. Credibility of findings not reported. (1)	Discussion of how findings extend understanding of theory and practice & future research. (2)	16/20

1.7 NARRATIVE SYNTHESIS

This section contains a review of the theoretical models discussed in the included studies, followed by a narrative synthesis of the evidence base.

1.7.1 THEORETICAL MODELS

1.7.2 Social Constructionist Models

Chronic illness can be 'understood in the social situations in which they emerge, are managed and have consequences' (Thorne, 1990, p.217). Thorne (1990) proposed that the roles associated with parenting and chronic illness are socially constructed and she explored how women undertaking both roles experience them. Radtke and Mens-Verhulst (2001) also adopted a social constructionist position and employed a discursive approach which conceptualises 'mothers with asthma as both the producers and the products of discourse' (p.381). It was suggested that participants constructed their experiences by drawing on available discourses which were present in the society in which these women lived (Radtke & Mens-Verhulst, 2001).

The two studies exploring physical disability and parenting used the social model of disability to make sense of their findings. This model understands disability at a societal level and suggests that individuals are disabled by societal prejudice and discrimination (Duvdevany *et al.*, 2008). Disability represents a form a social exclusion and not a product of impairment (Grue & Loerum, 2002). This understanding is in contrast to the construction of disability at an individual level which suggests that a person has impairments which need to be supported to improve functioning (Duvdevany *et al.*, 2008). The studies employing this theoretical model focused on understanding experiences that could be interpreted as the parents' reaction to 'disablism' (Duvdevany *et al.*, 2008; Grue & Loerum, 2002).

1.7.3 Biographical Disruption

Bury (1982) proposed that chronic illness could be perceived as a disruptive event which disturbs usual assumptions and behaviours and involves a re-thinking of biography and sense of self. There is little existing literature exploring biographical disruption in the context of

chronic illness and motherhood (Wilson, 2007). The concept of biographical disruption was used to understand how women with a diagnosis of HIV re-negotiated their identity as mothers in the face of threats to this identity resulting from stigma (Wilson, 2007).

1.7.4 Stigma

The majority of the studies exploring motherhood in the context of HIV understood their findings in relation to stigma (Ingram & Hutchinson, 1999; Nelms, 2005; Wilson, 2007). Stigma is 'omnipresent with HIV/AIDS and reflects the interrelated oppressions of heterosexism, sexism, and racism, along with issues of addiction, poverty, and cultural fears of death and dying' (Nelms, 2005, p.4). In these studies, stigma set the context for interpreting the findings in various ways which included: the concept of defensive mothering (Ingram & Hutchinson, 1999); the burden of mothering with HIV (Nelms, 2005); and threat posed to identity as a result of stigma (Wilson, 2007).

In summary, the conceptual frameworks used in the studies above are largely sociological and do not draw heavily on current psychological theory or health psychology research. These models do not provide a comprehensive understanding of the psychological processes which underpin the experiences of parents with a CID.

In understanding the experiences of parents with CF, a social constructionist perspective may offer an insight into identity as a parent and threats to this identity. However, such explanations may be less generalisable to this population as discrimination and prejudice could be less relevant. CF also does not appear to attract the same stigma as HIV therefore the use of this concept in understanding the experiences of people with CF may be limited. In addition, psychological understanding needs to move beyond using the concept of stigma as explanatory. Biographical disruption links to assertions by Williams *et al.* (2009) that people with CF may feel 'different' only in circumstances in which reality does not conform to an anticipated trajectory or biography (Williams *et al.*, 2009). Therefore, these ideas relating to 'disruption' of life with CF may be more applicable in understanding identity in parents with CF. Qualitative studies are needed to understand the nature of the perceived threat to identity when living with a CID such as CF and how people cope with this threat.

1.8 EVIDENCE BASE

Walsh and Downe (2005) suggest an analytical technique for synthesising qualitative research findings which involves tabulating the themes from all studies and comparing and contrasting them to gain an understanding of how they are related. This review followed this method; similarities and differences were identified to produce themes derived from the evidence as a whole. The table of resultant themes is shown in Appendix 4. Five themes were identified and included: *considering the children; identity; being a parent and living with a CID; experience with healthcare professionals and living to mother, mothering to live.* The themes generated and their occurrence in each of the included studies is shown in Figure 4.

To the researcher's knowledge, the only other review of qualitative research exploring the experiences of parents with a CID was conducted by Vallido *et al.* (2010). This review explored mothering disrupted by illness which was defined as 'a women perceiving that her maternal life has become disordered' (Vallido *et al.*, 2010, p.1436). The themes identified in this narrative synthesis were: mechanism of disruption; reframing the mother role; protecting the children; experiencing guilt or shame; problems with healthcare professionals; and living to mother, mothering to live. The review conducted by Vallido *et al.*, (2010) broadly differed from the present review in three ways; the first was the sole inclusion of mothers; second was the inclusion of mothers with mental and physical health problems and third was the focus on disrupted mothering. Despite these differences the themes identified by Vallido *et al.,* (2010) are highly relevant when exploring this body of research and will be compared with the current review for the purposes of triangulation (Walsh and Downe, 2005) and to demonstrate new or contrasting findings.

Figure 4: Table to show the occurrence of the themes across the included studies

Study	Theme				
	Considering the children	Identity	Living with a CID and being a parent	Living to mother, mothering to live	Experiences with healthcare professionals
Duvdevany *et al.,* (2008)	*	*			*
Grue, & Laerum (2002)	*	*			*
Hebling & Hardy (2007).	*	*	*	*	
Ingram & Hutchinson (1999)	*	*	*	*	
Mens-Verhulst *et al.,* (2004)	*	*	*		
Nelms (2005)	*	*	*	*	
Radtke, & Mens-Verhulst, (2001)		*			*
Thorne, (1990).	*				*
Wilson (2007)	*	*		*	*

1.8.1 Considering the children

A theme emerged relating to the potential impact of parenting with a CID on the child(ren). Participants reflected on the way in which they parent in a different way as a result of a CID. This was broadly considered in three ways: shaping the children's perception of disability, managing support from children, preparing children for an uncertain future. Vallido *et al.,* (2010) identified a theme of 'protecting the children' which captures ideas such as shielding children from stigma; concealing the effects of treatment and diagnosis; and preparing the children for a motherless future. This review builds on this work by adding ideas relating to shaping the children's perception of disability and managing support from children.

The fathers in the study conducted by Duvdevany *et al.* (2008) feared the way in which their children would view them as a parent with a disability and proactively worked to shape their children's view of disability. The fathers managed this through strategies aimed at being open with their children; they had direct conversations with their children about the disability; allowed their children's friends to become familiar with the disability; and ensured their children held positive perceptions of disability. Nelms (2005) found that some mothers managed this in a different way by not revealing the diagnosis to their children in an attempt to protect them from an uncertain future and the stigma associated with HIV.

Many participants feared their children supporting them and taking on a caring role (Grue & Laerum, 2002; Nelms, 2005; Thorne, 1990; Wilson, 2007). This required balancing the needs of children to become independent and socialised to help others as part of regular parenting with a fear of children 'being robbed' of their childhood by needing to undertake caring roles or being exposed to illness throughout their childhood (Grue & Laerum, 2002; Nelms, 2005; Thorne, 1990; Wilson, 2007). Grue and Laerum (2002) described how children's help was viewed within a discourse of disability as opposed to a discourse of socialisation.

However, some participants noted the positive impact of parenting with a CID on their children's development. This included children being more tolerant of difference, holding positive attitudes towards people with disabilities, greater compassion and ability to help others (Duvdevany *et al.,* 2008; Grue & Laerum, 2002; Thorne, 1990).

Attempting to prepare children for the parent's death was a theme that emerged in the studies with mothers diagnosed with HIV or Aids (Hebling & Hardy, 2007; Ingram & Hutchinson, 1999; Nelms, 2005; Wilson, 2007). Some parents used formal strategies to plan for their children's future such as making wills, identifying someone to care for their children in the event of their death and leaving tapes of their voices for their children to hear in the future (Ingram & Hutchinson, 1999; Nelms, 2005; Wilson, 2007). Other parents thought that being open about diagnoses would disrupt their children's development, so prepared children through indirect means such as education, developing positive relationships and ensuring positive legacies of their role as mothers (Ingram & Hutchinson, 1999; Nelms, 2005; Wilson, 2007).

1.8.2 Identity

An interpretation of how parents managed the two identities of a being a person with a CID and being healthy was considered a challenge which needed careful management (Mens-Verhulst *et al.*, 2004). Negative perceptions associated with being a parent with a CID was understood as stigma in the participants' diagnosed with HIV (Hebling & Hardy, 2007; Ingram & Hutchinson, 1999; Nelms, 2005; Wilson, 2007) and 'disablism' (understood within the social model of disability) in parents with a physical disability (Duvdevany *et al.*, 2008; Grue & Laerum, 2002). These negative perceptions were considered a threat to the participants' identity as parents; and this threat was managed in various ways.

Some parents normalised their health condition through minimising the impact of their health on the parental role and dealing with parental tasks in a way that allowed them to 'pass as normal' (Grue & Laerum, 2002; Mens-Verhulst *et al.*, 2004, p.82; Radtke & Mens-Verhulst, 2001). The desire for these participants to prove themselves to be a good parent and overcome threats to suggest otherwise was apparent and was described by Grue and Laerum (2002) as a 'fragile motherhood'. Mothers in this study described a need to perform as good mothers beyond the average parent to be perceived as 'good enough'.

Establishing an identity as a good mother was seen as a greater challenge and some parents enhanced their assertion that they were good mothers by emphasising their determination to put the needs of the children first (Wilson, 2007) and by making an 'extreme case' for their investment in the mothering role and their identities as mothers as 'all-encompassing and consuming' (Radtke & Mens-Verhulst, 2001, p. 386, p. 387). The threat to identity as a good mother was considered even greater for women with a life limiting condition which was considered more powerful than the stigma associated with the diagnosis of HIV in itself (Wilson, 2007). Some parents had internalised this stigma and did not view themselves as being good enough to parent (Wilson, 2007). However, others expressed their right to be a parent despite negative beliefs from other people (Hebling & Hardy, 2007).

Vallido *et al.*, (2010) also found that women with mental health problems experienced an incompatibility between an identity as a person with a mental health problem and a good mother. For some women this meant struggling to appear 'normal' to convince others of good mothering skills and resisting seeking help for worsening symptoms if this would

expose them to beliefs that they were not good enough parents (Montgomery *et al.*, 2006 as cited in Vallido *et al.,* 2010).

Some studies found that parents developed a new meaning to their sense of self as a person with a CID when they fulfilled this role. Grue and Laerum (2002) identified that parenting enabled the women in their sample to be 'included in a community not rooted in their disability, but rooted in their role as a mother' (p. 676). Hebling and Hardy (2007) discovered that the participants' feelings about motherhood were intense and outweighed the negative challenges associated with having a diagnosis of HIV.

1.8.3 Living with a CID and being a parent

The psychological processes underlying living as a parent with a CID were considered in a limited way by the majority of these studies. For parents with HIV this was considered in the context of living with a life limiting condition but for the other parents the focus was primarily on managing treatment and the physical effects of the condition. This theme was not explored in the review by Vallido *et al.*, (2010).

It was suggested that women with HIV or aids may employ cognitive strategies to manage and protect themselves from the implications and uncertainty of the future. These strategies were rationalisation, denial, positive reframing and gaining control of negative thoughts. Many of these participants also described living life to the full and finding meaning in their lives as mothers with HIV (Hebling & Hardy, 2007; Ingram & Hutchinson, 1999; Nelms, 2005).

Some parents sought practical and emotional support from their children, partners, wider family and God (Hebling & Hardy, 2007; Mens-Verhulst *et al.*, 2004). Mens-Verhulst *et al.* (2004) described 'non-medical prevention' techniques to manage the challenges of living with asthma and being a parent, for example making alterations to the environment such as bringing the baby's bed downstairs to avoid climbing stairs to minimise risks to health.

1.8.4 Living to mother, mothering to live

This theme was replicated from Vallido *et al.*, (2010) who developed the concept 'living to mother, mothering to live' to describe how mothers expressed a need to live for the sake of their children but also an uncertainty about their existence if they were no longer required to fulfil their mothering role (Wilson, 2007). Two of the studies included in the review by Vallido *et al.*, (2010) were also included in the current review so overlap of the evidence base is expected. However, this theme was also found in additional studies. Women identified children as a motivator to stay well and expressed a wish to stay alive to see their child reach a certain age which signified that they had reached adulthood (Hebling & Hardy, 2007; Ingram & Hutchinson, 1999).

1.8.5 Experiences with healthcare professionals

Experiences with healthcare professionals were considered in two ways; the first was the negative perception that some professionals held of parents with a CID; and the second was how services supported the participants' dual identities of patient and parent. Duvdevany *et al.* (2008) described how fathers experienced some professional interactions which were based on negative social attitudes towards parents with a disability. Wilson (2007) found that mothers in her sample encountered insensitive and disapproving treatment. Vallido *et al.* (2010) also identified this theme with women diagnosed with cancer and mental health problems.

A central theme in the study by Thorne (1990) was the experience of healthcare services. This highlighted the challenge that services faced when needing to support a person with a chronic illness who is also a parent. It was found that the priorities of the two roles were at odds with each other, for example suggestions were made to make lifestyle changes to support health which were incompatible with a role as a parent. However, it is noted that this study was published in 1990 therefore further progression in this area may have been made.

Radtke and Mens-Verhulst (2001) found that the women in their sample 'told few stories of medical encounters where their positioning as mothers was of interest' (p.383). However, although this gap was noted it was not necessarily a problem for these women. It was

hypothesised that (as captured in the *identity* theme) these participants worked hard to ensure that they were perceived as good mothers so a limited connection between physical health and their parenting was constructed (Radtke & Mens-Verhulst, 2001). In support of this hypothesis, Grue and Laerum (2002) identified that women feared being reported to the authorities and worked hard to convince others of their suitability as a mother; acknowledging the impact of a CID on mothering may have undermined this position.

1.8.6 Implications for future research

A large proportion of the research in this area is medically orientated or focused on the implications of a health condition for the children's well-being or the families' functioning. As Duvdevanny *et al.*, (2008) expresses 'the parents' voice is almost completely absent from parenting research' (p.1021). In studies which do explore this role there is often a greater focus on the practical and social barriers to parenting as opposed to the psychological or emotional issues related to being a parent with a CID. It is suggested that this focus reflects public health priorities as opposed to the concerns of parents. Therefore, further research is needed to explore parenting experiences for people diagnosed with different health conditions so that comparisons can be made between samples within specific populations.

The most notable absence of literature in this evidence base is fathers' experiences of parenting with a CID. With the exception of the study by Duvdevanny *et al.* (2008) all studies included mothers only. Research is needed to identify how fathers negotiate these two roles; which may be different given the differing societal expectations and assumptions relating to fathering roles. This narrative review also only included one study conducted in the UK (Wilson, 2007). There are many factors which make comparisons between research generated in different countries problematic, such as differing experiences of poverty, health care systems and cultural expectations of parents and people with a CID. Further research is needed to generate an understanding of the experiences and needs of parents which is reflective of parenting and healthcare culture in the UK.

The evidence base is largely understood from a sociological perspective with only small references to individual psychological experiences such as the cognitive processes some parents adopt to manage the uncertainty of a life limiting condition. These studies do not provide an understanding of the psychological processes which enable parents with a CID to

continue functioning and draw benefit from their parenting roles. This review indicates that a person with CF may experience various psychological challenges as a parent, which could include: deteriorations in CF; threats to identity as a parent and person with CF; perceived time limitations; and the impact of the demands of parenting on psychological well-being and adjustment. Therefore, alternative frameworks are needed which examine how parents appraise and manage these threats and the tension between being 'good enough' as a parent and a person with CF. This understanding is needed for people with CF who are parenting or considering parenting but also for health professionals who need to base their interventions in evidence-based and researched positions.

There is a small evidence base exploring coping and coping styles in people with CF which could be applied as an alternative framework in understanding parenting in this population. In addition, the literature in the area of parenting with a CID suggests that parenting with a life limiting condition may be the greatest threat to identity as a 'good parent'. Given, the time consuming nature of CF treatment and the dominant position treatment adherence is given in the psychological literature in CF, a framework which captures the challenges of CF as a time limiting condition is also needed. Therefore, two alternative frameworks: coping and time perception (Zimbardo & Boyd, 1999) are explored in more detail below.

1.9 ALTERNATIVE THEORETICAL MODELS

1.9.1 Coping

Coping refers to the behaviours, thoughts and feelings that are used to avoid being harmed by life stressors (Abbott, 2003). A small evidence base suggests that the principle approach to coping in adults with CF is an optimistic style (Abbott, 2003) and is associated with a better HRQoL (Abbott *et al.,* 2008). Distraction is associated with a poorer HRQoL (Abbott *et al.*, 2008). However, people with CF are likely to use more than one method of coping to meet the short and long term demands of a changing condition (Abbott, 2003: Abbott *et al.,* 2008) and approaches such as avoidance may be beneficial in enabling functioning in the face of a challenging condition (Abbott, 2003). Therefore coping strategies for parents with CF are likely to be adaptive for different individuals at various points in time. However, there are limitations when relying on this evidence base, for example there are no consistent

ways of measuring coping or psychological functioning, the sources of information from which coping is established varies from self-report to other-raters and it is not possible to establish a cause and effect relationship between coping and psychological functioning from these studies (Abbott, 2003). Therefore longitudinal studies are needed to establish the long term implications of different approaches to coping in this population and research needs to develop as the treatment for CF progresses (Oxley & Webb, 2005).

As described earlier, acceptance may be an important construct when understanding adaptation to CF. Therefore, a model of coping which includes acceptance may enhance the understanding of parents with CF. Secondary control is one such model which incorporates acceptance and adjustment. In contrast to primary control in which people choose to influence their environments (as described above) secondary control is defined as an *adjustment* to some aspect of the self and an *acceptance* of circumstances as they are (Morling & Evered, 2006). More specifically, secondary control consists of adjusting, adapting or changing the self and a process of accepting the existing environment. It is proposed that secondary control facilitates a sense of control through the ability to accept or adjust to existing realities (Thompson, *et al.,* 1996). This approach could be of benefit in the context of CF when circumstances relating to health cannot be changed.

One of the proposed beneficial outcomes of secondary control is a lack of helplessness and despair as it fosters wellness even though it does not enhance efficacy (Morling & Evered, 2006). People optimise their functioning across the lifespan by using a combination of primary and secondary control (Skinner, 2007). However, these assertions are limited in their evidence, particularly when applied to a population of people with a CID. There is an on-going debate as to whether secondary control follows primary control, if secondary control fosters perceived primary control and if it should be considered a means of coping or 'passive' acceptance (Morling & Evered, 2006; Skinner, 2007). There is also limited evidence to suggest which approach to coping is more adaptive in particular populations. Contrary to expectations, Thompson *et al.* (1996) found that secondary control resulted in increased distress in men with HIV in a prison population who had very low control over their environment. Therefore more research with people with a CID is needed. Secondary control has not been explored in the CF population but may provide a useful means of understanding how parents accept CF whilst making adjustments to enable them to fulfil parental roles.

1.9.2 Time Perception

Zimbardo and Boyd's (1999) theory of time perception is an extension of Lewin's Life Space Model which considered the influence of past and future on present behaviour. It is proposed that to create a sense of order and coherence experiences are assigned to time categories. Time perceptions are used to reconstruct past experiences and imagine the future, which influences present decision making (Zimbardo & Boyd, 1999; Zimbardo & Boyd, 2008). Successful temporal functioning is an ability to balance the integration of past experiences and future hopes into current decision making and experiences which requires an adaptation process involving internal (cognitive and emotional) and external (environmental) adjustments (Livneh, 2013). Therefore, a close link between coping and temporal adaptation is hypothesised but to date research in this area is limited and inconclusive (Livneh, 2013).

Personal experiences such as a CID are thought to influence the perception of time (Livneh & Martz, 2007). Previous research suggests that the onset of a CID disrupts the continuity of developmental processes and therefore the experience of time (Livneh, 2013) although the processes involved in this are not clearly evidenced. This concept could usefully be applied to the CF population. A time perspective scale was developed to assess the time perceptions of: past negative, present hedonistic, future, past positive and present fatalistic (Zimbardo & Boyd, 1999; Zimbardo & Boyd, 2008). Research with people with disabilities or chronic conditions has primarily focused on exploring future time orientations as it suggested that 'the stronger the light [future time orientation] the farther you see—the more objectives you discover—the brighter and clearer they appear…and the more able you are to structure and plan for future actions' (Livneh & Martz, 2007, p. 456) which is particularly important in the on-going management of chronic illness. Parenting with CF could be understood as an investment in a future orientation which may enable parents to direct action towards fulfilling a goal as a parent.

Research has demonstrated that future orientation is linked to psychosocial adaptation in participants' diagnosed with diabetes (Livneh & Martz, 2007) and that pain, distressed awareness of death, shock and depression are related to a foreshortened future orientation in people with a spinal cord injury (Martz & Livneh, 2003; Martz, 2004). However, 'empirical data on time orientation and perspective, and psychosocial adaptation to physical CID are

scarce at best' (Livneh, 2013, p.76). Further exploration is needed to understand the processes involved in time perception such as the development of a time perspective, changing perspectives and what particular perspectives might predict for a person with a CID. There is no research exploring time perception in people with CF. This could be an important area for investigation given the time consuming nature of CF treatment which is set in the context of a life limiting condition, changing medical technologies and treatments for CF which has caused people to re-evaluate their perception of having a foreshortened future.

As described, a close link between coping and temporal adaptation is hypothesised. Therefore, to better understand the psychological experiences of parents with CF, the theoretical model of secondary control and its relationship with time perspective could usefully be explored.

1.10 CONCLUSION

In conclusion, there is a small body of research exploring the experiences of parents with a CID. The systematic review included nine qualitative studies which met the inclusion criteria and were explored in more detail. A summary of these studies, a review of the quality, and narrative synthesis of the findings were presented. The theoretical models used in this evidence base are limited in their application to understanding the psychological mechanisms underpinning the experiences of parents with CF. Therefore, two theoretical models were described which sought to this fill this gap. Given the lack of research in the UK, specifically exploring individual parents' experiences, with the inclusion of fathers, it is recommended that further research in this area is conducted.

1.11 CURRENT STUDY

1.11.1 STUDY RATIONALE

The rationale for this current study is a reflection of the lack of research exploring parenting with a CID generally and parenting with CF more specifically. The broader evidence base

exploring parenting with a CID indicates a need for further research to understand how individual parents experience this role, particularly fatherhood.

Parenting is now becoming a reality for people with CF but there is very limited research in this subject area. The majority of the evidence relates to pregnancy outcomes for people with CF and whilst this is important information; there is a gap in knowledge relating to how people with CF experience being a parent and the implications for healthcare professionals working with this population. The application of the broader evidence base is a useful starting point for exploration but specific research within the CF population is needed as CF presents unique challenges and experiences.

People with CF have expressed difficulty in obtaining appropriate and useful information regarding motherhood (Johannesson *et al.*, 1998) and have stated a desire for information relating to the experiences of other people with CF to be shared and the emotional impact of pregnancy and fertility to be recognised in clinics (Fair *et al.,* 2000). Due to the lack of available evidence, there is a limited understanding regarding this subject area. It is therefore argued that further qualitative studies are needed which are able to explore the experiences of parents with CF from their perspective.

1.11.2 STUDY AIMS

The aim of this research was to explore *how people with CF experience being a parent,* which includes the experiences of both mothers and fathers. The theme living to mother, mothering to live, was captured from the broader evidence base relating to chronic illness and parenting and is used as a starting point for exploration of parenthood in this population. However, to better reflect the CF population and include fathers; this was re-named *'staying well to parent and parenting to stay well'*. In the current study, this is understood as an exploration of the participants' experiences of looking after themselves to be a parent and being motivated to remain well as a consequence of being a parent. This focus was used to generate a psychological understanding of how parents manage the tensions between being a parent and a person with CF and threats to this identity.

Therefore the aim of this study was to explore the participants' hopes, beliefs and relationships (including with health professionals) in the past, present and future and how

they live as a parent and a person with CF. A qualitative methodology was deemed the most appropriate approach to gain access to the experiences of individual participants. The lack of available research in this area indicates that this approach would be a useful starting point to begin exploration. The aim of this study was to identify emergent themes from within the data, using Interpretive Phenomenological Analysis.

The theoretical models described in the wider evidence base are limited in their applicability to understanding CF and offer a limited contribution to the psychological understanding of parents with a CID. Therefore, a further aim of this research is to identify other theoretical models which provide a useful understanding of the experiences of parents with CF. Time perspective (past, present and future) is important when exploring CF as it is 'life limiting' and because advances in medicine have forced those with CF and the health professionals who work with them to re-evaluate the meaning of 'life-limiting' and its implications. Therefore, the psychological mechanisms underlying parenting in this context will also be considered.

As a result of the rapidly evolving understanding and treatment of CF which has now made parenting possible, it is expected that this research will provide evidence with implications for the practice of professionals working with this population. Participants were recruited from the All Wales Adult Cystic Fibrosis Service, which offers a specialist service to adults with CF across Wales. The service offers inpatient and community services delivered by a multi-disciplinary team which has two Clinical Psychologists employed on a part time basis. The aim of this service is to ensure all patients receive appropriate optimal therapy and achieve the best possible quality of life. Given this aim, it is important that the needs of parents with CF are understood to ensure resources are allocated appropriately and people with CF in Wales receive optimal psychological sup

CHAPTER TWO: METHODOLOGY

2. OVERVIEW

A qualitative approach using semi-structured interviews was considered the most appropriate method to fulfil the aims of the research. Interviews were conducted with nine parents with CF who were recruited from an Adult CF Service. Data were analysed using Interpretative Phenomenological Analysis (IPA) to explore how people with CF experience being a parent. This chapter includes a rationale for the use of the methodology, the background and philosophy of IPA and a description of the design and procedure of the research process. Ethical issues and the process of data analysis are also considered.

2.1 QUALITATIVE METHODOLOGY

2.1.1 Philosophical Underpinning

The aim of qualitative research is to explore how people understand and make sense of their experience of particular situations (Elliott *et al.,* 1999; Willig, 2001). Unlike some quantitative methodology which it could be argued, aims to identify cause and effect, qualitative research is concerned with examining richness of experience and the meanings that people attribute to events (Willig, 2001). Therefore, the aim of qualitative research is to describe and understand experiences but not to predict them. There are various qualitative approaches that exist within this methodology and take different positions in relation to epistemology, reflexivity and language (Willig, 2001).

The philosophy of epistemology is concerned with knowledge; how, what and can be known (Willig, 2001). Epistemological positions can be seen on a continuum with positivism at one end and relativism at the other. A pure positivist approach would propose that there is a direct relationship between objects in the world and our perceptions, so an objective truth can be discovered unbiased by the researcher (Willig, 2001). Relativism, for instance as exemplified by social constructionism, argues that knowledge is not formed through direct

perception of reality but that reality is constructed between people and is emergent from the culture or society in which we live (Burr, 1995). Therefore, in this approach language is not seen as a means of expressing ourselves but a construction of reality (Burr, 1995). Findings of qualitative research are acknowledged to be co-constructed by the researcher and the participant through language. Knowledge derived through this research method is relativist and socially constructed. Extreme relativism takes this position further in rejecting the concept of 'truth' and 'knowledge' completely (Willig, 2001).

2.1.2 Rationale for Using a Qualitative Design

The objective of this research was to explore how participants make sense of and experience being a parent with CF. The aim was to allow the development of an understanding of experience rather than test existing theory (Elliott *et al.,* 1999; Willig, 2008). With this aim, it was important to choose a methodology which allowed participants to reflect on their experiences with minimal imposition of pre-defined categories in which to share their experiences within. This allowed the emergence of themes and ideas which may not have been considered by the researcher or predicted by existing theory and allowed participants to express their personal meaning of this experience. To fulfil these objectives a qualitative methodology was deemed an appropriate choice.

2.2 INTERPRETATIVE PHENOMENOLOGICAL ANALYSIS

IPA is considered an approach rather than an explicit method (Larkin *et al.*, 2006). It holds an epistemological position (described below); it contains guidelines for conducting research and describes a body of research (Smith, 2004). As described, IPA is concerned with exploring how participants make sense of situations and the meanings they ascribe to these experiences (Smith, 2003). This process is interpretative, that is, it is shaped by the researcher's own values, beliefs and ideas in making sense of the participants' experiences (Smith, 1996) and reporting on them. Therefore, IPA seeks to understand the first person perspective from the third person position (Larkin, 2012). IPA is derived from phenomenological, interpretive and idiographic perspectives (Smith *et al.*, 2009). These are described in more detail below.

2.2.1 Phenomenology

Phenomenology is a philosophical approach to studying experiences (Smith *et al.*, 2009) and is concerned with exploring the meaning of experiences for participants (Landridge, 2007). IPA is phenomenological as it attempts to explore individuals' perceptions of events rather than seek to find an objective truth about the world (Smith, 2003). Phenomenology takes a position between realism and relativism (Larkin, 2012). It is agreed that direct access to experience is impossible as researchers use their own thoughts and feelings to make sense of a participant's experience (Willig, 2008). However, IPA makes the assumption that participants' accounts provide access to thoughts and feelings which are derived from experiences; this assumption could be considered a realist approach to generating knowledge (Willig, 2008). The types of experience that IPA is normally concerned with are moments which take on particular significance in people's lives. These are events which people reflect on and become important as the person tries to make sense of them (Smith *et al.*, 2009).

Experience can be a first order activity or a second order reaction to the experience, in which a person engages in remembering, regretting or hoping in relation to the experience. It is not possible to understand or 'get to' an experience purely as it is reflected upon after an event (Smith *et al.*, 2009). Therefore IPA attempts to understand the meaning a participant attributes to an experience and the aim is to do research which is 'experience close' (Smith *et al.*, 2009, p.33). An understanding of experience is a result of the researcher's own ideas and relationships to objects and people in the world (Smith, *et al.*, 2009). This is discussed further below.

2.2.2 Interpretation

Analysing data is an interpretative process; it is a two stage procedure in which the participant makes sense of their experiences and the researcher attempts to understand the participant making sense of their experiences (Smith, 2003). Interpretation is made through the lens of the researcher who reflects upon their own prior concerns, experiences and thoughts when understanding the experiences of participants. A cyclical approach is taken in which the researcher reflects on changes in ideas and assumptions after meeting with participants (Smith *et al.*, 2009).

Interpretation during analysis can be at different levels, a combination of an empathic and questioning interpretation is usually taken in IPA (Smith, 2003; Smith *et al.*, 2009). An empathic interpretation is used to try to understand the person's experience from their point of view. The aim of this is to provide an interpretation of the participant's experience, which is as close to their words as possible (Larkin *et al.*, 2006; Smith *et al.*, 2009). As identified, this can only be partial because an understanding of the participants' world involves the researcher as they are part of the world that they are trying to understand (Larkin *et al.*, 2006).

Questioning the text when constructing themes from the data, involves interpreting what the person might be trying to communicate and moves beyond describing the experiences of participants and attempts to interpret them in relation to social, cultural and theoretical constructs (Larkin *et al.*, 2006). This is an interpretation by the researcher to understand why participants may have had particular feelings, concerns, and experiences in particular situations. Questioning interpretations can be at different levels of meaning and depth (Smith, 2003) but should still remain as close as possible to the participants' own words (Willig, 2008).

2.2.3 Idiographic Approach

IPA is an idiographic approach; the aim is to explore particular cases rather than make generalisations within the population. IPA is concerned with understanding detail and depth of experience in particular contexts for particular people (Smith *et al.*, 2009). It is proposed that this method produces research which is compatible with the complexities of human behaviour (Smith *et al.*, 2009).

Analysis starts with understanding meaning from one participant's account and then similarities and differences are drawn between their data and other participants' accounts. As more qualitative studies are conducted within a specific population, gradually more ideas can be generated about a phenomenon, process or experience based on the analyses of individual cases (Smith, 2003) and new insights can be offered to existing nomothetic research (Smith *et al.*, 2009).

2.2.4 Limitations of IPA

There are several limitations acknowledged with the use of IPA. This method relies on the quality of people's accounts of their experiences. As IPA is concerned with experiences and the meanings attached to these, it is dependent on participants being able to reflect on their thoughts, feelings, relationships, emotional and physical lives rather than simply to offer their opinions about a phenomenon (Willig, 2008). Therefore, the IPA method may be easier for the researcher to apply with some participants than others; this may limit the breadth of participants who can usefully be recruited using this method (Willig, 2008) and potentially skew the sample towards more articulate and educated participants.

IPA is also concerned with describing and interpreting experience. Research using this method seeks to understand experience rather than explain it; it is not concerned with why such experiences take place or why there are differences between people (Willig, 2008). It is suggested that in order to explain phenomena researchers would need to understand the context behind experiences. It could be argued that without explanation, understanding is limited (Willig, 2008).

Furthermore, IPA relies on language as a valid means of representing experiences. Critics adopting a social constructionist position would argue that language constructs reality rather than describes it (Burr,1995; Willig, 2008). Therefore, it is argued that IPA needs to attend more to the constructionist role of language (Willig, 2008). Interview transcripts are the usual data sets used in IPA; this also limits the richness of an interview as all non-verbal elements of communication are lost in the transcribed document.

2.2.5 Rationale for Using IPA

IPA is often used when experience takes on a particular significance for people, usually due to important changes in a person's life (Smith *et al.*, 2009). The aim of this research was to capture the meaning of parenting for these participants, which is a personal, significant, complex and changing process. IPA enabled an understanding of the meaning of this experience at a specific point in the participant's parenting role, in the chronicity of their illness and in the developmental stage of their child(ren). This approach allowed the subjective nature of this experience to be interpreted and facilitated an understanding of the

ways in which the same 'objective' event of being a parent with CF could be experienced (Willig, 2008).

IPA was also used because it enabled an understanding of this experience within its context. It is acknowledged that IPA does not place context in the central position that some other approaches suggest such as discourse analysis (Willig, 2008). However, IPA is an interpretive approach and therefore the context in which the research took place is understood when interpreting the data.

It is important that parenting with CF is understood within the complex context in which it sits. Medical advances in CF have moved relatively quickly within the participants' lifetime and parenting with CF is set against a background of cultural and scientific prioritisation that directs resources to medical procedures such as assisted fertility, which have made this experience possible for some people with CF. Employing IPA allowed an understanding of the evolution of parenting with CF through interpreting the reflections of participants who have lived its relatively recent history in South Wales.

IPA has also frequently been employed in health psychology contexts as it offers a way of understanding how individuals might make sense of their health condition (Smith, 1996). This is because IPA attempts to understand the relationship between an object (this could be our body) and an individuals' perception of it (Smith, 1996), which was considered useful in understanding how people make sense of CF when also fulfilling a parenting role.

In addition, there is very little existing research exploring parenting with CF. Reid *et al.* (2005) suggested that 'IPA is particularly suited to researching unexplored territory, where a theoretical pretext may be lacking' (p.23) because it allows an exploration of the quality and nature of experience to describe and interpret rather than predict experience (Larkin *et al.*, 2012). IPA was therefore also considered appropriate to employ in this study as a starting point for new research in this area.

2.3 ENSURING QUALITY IN IPA

Establishing criteria for quality in qualitative research is made challenging by the number of different epistemological positions and methodological differences that exist within the

broad spectrum of qualitative approaches (Meyrick, 2006; Willig, 2008; Yardley, 2000). It is recognised that for this reason it is not possible to have rigid rules or exact criteria which fit all qualitative methodologies (Yardley, 2000). However, to ensure quality in qualitative research some guidelines and criteria (which can be fulfilled to a greater or lesser extent according to the approach employed) have been proposed (e.g. Critical Appraisals Skills Programme (CASP), 2010; Elliott *et al.*, 1999; Meyrick, 2006; Yardley, 2000).

Yardley (2000) suggested that quality can be assessed against the dimensions of: sensitivity to context; commitment and rigour; transparency and coherence; and impact and importance. Meyrick (2006) developed a framework which was based on agreed principles drawn from pre-existing views on rigour in qualitative research. These were transparency (Yardley, 2000) and systematicity. This framework describes how researchers can ensure rigour at every stage of the research process and reflects the diversity of ways to do this according to methodological approach taken (Meyrick, 2006). Elliott *et al.* (1999) published a set of seven guidelines for the purpose of monitoring qualitative research to ensure better quality control across the spectrum of qualitative methodologies.

Willig (2008) described different priorities for judging quality in qualitative research based on the epistemological position of the qualitative approach taken. It is suggested that the epistemological approach used in IPA implies that researchers need to pay particular attention to the role of the researcher's position in ensuring quality in these studies (Willig, 2008).

For clarity, the quality of this research is assessed in relation to the seven standards proposed by Elliott *et al.* (1999) but the guidelines suggested by other authors will also be incorporated within these seven standards to ensure breath of perspectives and greater assurance of quality.

2.3.1 Owning one's Perspective

In qualitative research, it is suggested that the researcher should take a position of reflexivity (Elliott *et al.*, 1999; Willig 2008). This should be an acknowledgement of how the researcher influences and shapes the research process as a researcher, both professionally and personally (Willig, 2008). The researcher is required to outline their motivations and

assumptions about the research topic in advance and also as they develop throughout the research (Elliott *et al.*, 1999; Yardley, 2000). An acknowledgment of the social context of the relationship between the researcher and the participants should also be made to allow an understanding of the interaction between the researcher and participants (Yardley, 2000). Reflexivity goes beyond acknowledging 'biases' and serves to consider how the researcher makes possible certain insights and understandings within the research process (Willig, 2008) as the researcher acts as a channel or filter through which experiences are interpreted (Larkin, 2012).

In IPA, the role of the researcher's perspective is recognised but the way in which this perspective is incorporated into the research process is not made explicit (Willig, 2008). In this study, the researcher's perspective was acknowledged through a description of the researcher's personal and professional position in relation to the research. The development of the researcher's position during the research process was also captured through the researcher's reflective diary (appendix 5). The researcher also undertook a 'bracketing interview', which involved exploring changing beliefs and assumptions in relation to this area to increase self-awareness (Rolls & Relf, 2006). This was conducted with a group of trainee clinical psychologists, all with an interest in qualitative methods. Interviews took place during the data analysis process and discussions focused on assumptions, expectations and values prior to data collection and appreciating change throughout the research process. This enabled the researcher to notice what she expected participants to share during data collection and attend to unexpected or difficult information. For example, contrary to the researcher's pre-existing assumptions, participants described many positive aspects of CF and some explained that they would not choose a life without CF. These changing assumptions also contributed to the researcher's reflective diary.

Researcher's Position

The researcher is a 30 year old, white, middle class, English female. She is younger than eight of the nine participants. The researcher was born in London and has lived in South Wales for the last eleven years. She would still consider herself an observer of the culture and values which shape families and communities in South Wales. This is in contrast to all of the participants who are from South Wales.

She was brought up with two parents who both played active parenting roles. In her clinical work, the researcher is aware of noticing and paying attention to the role of fathers when they become absent from conversations. The researcher is a twin and is interested and influenced by her own experiences in thinking about how this relationship impacts on a family. The researcher is not a parent and is conducting this research through the perspective of a person who does not have a physical health condition and had no personal or professional experience of cystic fibrosis until this research commenced.

The researcher is in the final year of her clinical psychology training and is currently working within a physical health context in critical care and oncology. She has very occasional contact with people with cystic fibrosis in her work but often considers the psychological issues affecting parents and the children of these parents with physical health conditions. The researcher has also worked with parents with mental health problems and learning disabilities and has observed the motivations and challenges children can bring to these parents. She has also witnessed the discrimination that some parents experience within services. The researcher is interested in systemic approaches to her work and believes that experiences should be understood in context. Research findings are viewed from the same perspective and therefore the impact of the researchers and the participant's position in understanding the data is considered important by the researcher.

The research topic was developed in collaboration with the researcher's clinical and academic supervisors. The research topic was of interest to her for two reasons; the first was after meeting children with life limiting conditions in her child and family placement and wondering how these children may go on to develop roles and identities outside the medically dominated context in which they inhabited from their early years. The second was thinking from the parent's perspective as a result of previous clinical work and being intrigued as to how these parents coped and experienced challenging life events while maintaining their parenting roles.

Prior to conducting this research the researcher understood a physical health condition such as CF, to be something which would challenge a parenting role. She believed that the parent would need to be more resilient and in need of support to fulfil this role. As the research has progressed, the researcher began to understand this as too simplistic and appreciated the

multiple ways in which people negotiate and experience CF, which changes over time and in different contexts.

2.3.2 Situating the Sample

A description of research participants and their life circumstances should be made to allow the reader to judge the range of participants, the similarities and differences between participants (Smith *et al.,* 2009) and their contexts in relation to the research findings (Elliott *et al.*, 1999). This should be sufficient so that the research can be evaluated by assessing the extent to which observations have been grounded in the contexts in which they occurred (Willig, 2008). Demographic information and information about the participant's family is described. The participant's most recent lung function test results are also included to provide a measure of the severity of one aspect of CF.

2.3.3 Grounding in Examples

Examples of data to illustrate the analytic procedure and the understandings developed should be included to allow others to make sense of this, generate new ideas (Elliott *et al.*, 1999) and demonstrate transparency of the research process (Meyrick, 2006; Yardley, 2000). Therefore, multiple examples were used to illustrate each theme in the presentation of the results and extracts from transcripts are included in appendix 6.

2.3.4 Providing Credibility Checks

A number of methods to ensure the credibility of the themes generated and the rigour of the data analysis are suggested by Elliott *et al.*, (1999) and Yardley (2000). These include checking with the original informants, using more than one qualitative method and employing multiple qualitative analysts. Themes generated in this research were reviewed and explored with a group of trainee clinical psychologists interested in qualitative methods. The analytical process and the development of the themes were also discussed with the researcher's academic and clinical supervisors. This allowed multiple perspectives on the development of themes to ensure credibility.

2.3.5 Coherence

The data should be presented in a way that achieves coherence and integration but retains detail (Elliott *et al.*, 1999) the results should form a coherent argument which is meaningful to the audience in which the research was intended (Yardley, 2000). The data was discussed with the academic and clinical supervisors throughout the process of data analysis to ensure a useful and productive account of the data. Tables to show how themes are related and a narrative description of themes are presented in chapters three and four to ensure coherence.

2.3.6 Accomplishing General vs. Specific Research Tasks

It is important to be clear about whether a general understanding of a phenomenon is intended or a specific instance of an event is to be understood. The limitations of either goal should be addressed (Elliott *et al.*, 1999). The current study contains a sample of parents with cystic fibrosis who live in South Wales and the findings are not intended to be generalised to any other sample. Details of the participants are provided so that the reader can assess similarities and differences to other samples in this population and assess the extent to which the reader can relate to these results when understanding another sample (Meyrick, 2006). An acknowledgement of the limitations of this research method is examined in chapter four.

2.3.7 Resonating with Readers

The presentation of the research should allow readers to judge that it accurately represents the subject matter and expands their understanding of it (Elliott *et al.*, 1999). The research should have a useful impact on the reader which is consistent with the aims of the research (Yardley, 2000). It is also important that the researcher develops a good awareness of the subject matter and literature to allow them to draw upon pre-existing theories and ideas to develop further reaching interpretations of the data which are more useful to the reader (Yardley, 2000).

In order to ensure that the research resonated with the reader, an overview of relevant clinical and theoretical issues in relation to the research is outlined within the literature review in chapter one. These ideas were drawn upon in the data analysis. The researcher also

discussed and sought feedback from academic and clinical supervisors to gain multiple perspectives on the presentation of the research and literature review to ensure that it was useful to readers.

2.4 ETHICAL CONSIDERATIONS

2.4.1 Ethical Approval

Approval for this study was sought from the Research and Development Department (R&D) in Cardiff and Vale University Health Board (see appendix 7). Ethical approval was granted by the National Research Ethics Service (NRES) Committee South West – Central Bristol (see appendix 7).

2.4.2 Informed Consent

In accordance with the British Psychological Society (BPS) (1995, 2009) and the Health and Care Professions Council (HCPC) (2012) guidance, informed consent to participate in the research was sought from each participant. In line with the approved proposal to LREC, the clinical supervisor sent potential participants an invitation to participate in the research and information about the research (see appendix 8). This included a request for permission to access the participant's last lung function test result from the CF database, should they decide to participate in the research. Only potential participants deemed to meet the inclusion criteria were sent this information. Potential participants expressed their interest in participating in the research by contacting the researcher directly.

On meeting the potential participant, the researcher read the participant information sheet with them and invited them to ask questions. If they were satisfied with the information and indicated that they felt able to make an informed decision about participating they were also asked if they would consent to the interviews being tape-recorded for the purposes of transcription. It was explained that the recorded interview would be destroyed after transcription and that transcripts would be anonymised to maintain confidentiality. It was explained to participants that the interview transcripts would be stored electronically and would be password protected until being destroyed on completion of the study.

Participants were asked if they would give permission for the researcher to access their last lung function test result from the database held by the CF service. If the participant remained in agreement, informed consent was provided in writing and this was also signed by the researcher. It was explained to participants that they could withdraw from the study at any time without implications for their care within the CF service.

2.4.3 Confidentiality and Anonymity

Professional Practice Guidelines (BPS, 2008) and standards of conduct (HPC, 2007) were followed in assuring the maintenance of participants' confidentiality and anonymity. The participants were informed via the information sheet that confidentiality would only be broken if they disclosed that they posed a serious risk to themselves or another person; for example, their child. This was reiterated when the researcher met with participants. Each participant was assigned a number and their personal details were recorded using this number so that data generated by participants did not need to contain any identifying information. This coding system allowed participants to be traced by the researcher if necessary. The coding system was saved electronically and password protected to ensure confidentiality.

In accordance with Local Research Ethics Committees (LREC) and R&D approval, the interviews were transcribed by the researcher. The audio recordings of the interviews were deleted electronically when they had been transcribed and anonymised. The transcripts and the completed questionnaires were then saved electronically and the folder containing this information was password protected so that only the researcher had access. On completion of the study these were destroyed. The consent forms were stored in a locked filing cabinet in the CF centre base.

To ensure anonymity, any person or place that participants' identified in the course of the interview was given a pseudonym. This was used when transcripts were prepared and quotes were included in the results. Because of the potential to identify participants amongst the small CF cohort in Wales, some demographic details (for example employment, education, marital status) were not matched with the participant's pseudonym in the presentation of the results. This was designed to minimise the breadth of information that was presented and ensured that all quotes remained anonymous. Therefore, only information which would not

expose the participant's identity and was considered important in interpreting the data was presented with the participant's pseudonym. This included: age of participants, age of children, lung function (all presented as a range) gender, and questionnaire responses.

2.5 PARTICIPANTS AND RECRUITMENT

2.5.1 Sample

In accordance with IPA methodology, the sample was selected purposively. This was with the aim of gathering detailed information from a specific group on the particular topic of interest (Landridge, 2007). A sample which was as homogenous as possible given the constraints of population size was recruited in order to address the research question meaningfully (Smith *et al.*, 2009). The selection of participants (the inclusion and exclusion criteria) was based on what was considered to be the most important similarities between participants. This was their parenting role (in terms of involvement) and their experience of CF.

It is suggested that there is no correct sample size; the size should be judged on the basis of depth of analysis, the richness of individual cases and pragmatic constraints (Smith *et al.*, 2009). Ten participants are considered to be at the higher end of sample size (Smith *et al.,* 1999 as cited in Reid, Flowers & Larkin, 2005). Smith *et al.,* (2009) suggests between four and ten participants for professional doctorates. Therefore, the aim was to recruit ten participants.

2.5.2 Inclusion and Exclusion Criteria

The inclusion criteria for participation in the research were parents (mothers or fathers) who had a diagnosis of CF and received services from the Adult CF service in South Wales or Bristol. Potential participants needed to be primary care givers which was defined as living with their child(ren) on a full time basis. This was based on the assumption that these people would have shared in or provided all parental roles and had consistent contact with their children. This definition was adopted to increase the probability of recruiting a

homogeneous sample. However, it is acknowledged that this may be limiting the diversity of parenting roles that are present in modern families, which is captured within this research.

The numbers and ages of the children were not used as exclusion criteria because the challenges of recruitment from this small population were recognised and because experience of parenting roles and responsibilities was the focus of the study. Participants also needed to have sufficient command of spoken and written English to read the information sheet and take part in semi-structured interviews; resources for the research did not extend to funding for translators or interpreters.

The Exclusion Criteria Included:
- Parents with CF who have had a lung transplant.
- Parents with CF who would not be able to participate in an interview of 60-90 minutes (although this could be split over more than one session).
- Parents who are vulnerable due to unstable mental health problems.
- Individuals who might pose a risk under the lone worker policy for the relevant LHB/ NHS trust.

Participants were excluded from the research if they have had a transplant because a transplant will alter symptoms and treatment and therefore these people's experiences may differ from those parents who have not had a transplant.

2.5.3 Recruitment Process

Potential participants were identified by the clinical supervisor from the All Wales Adult Cystic Fibrosis Centre on the basis of the inclusion and exclusion criteria. A database of potential participants was created and sorted alphabetically. Participants were selected randomly from this list by inviting every third person and recruiting in waves of ten potential participants. The first ten participants selected were either sent a letter or were given a letter if they were attending a clinic appointment that week. This included an information sheet, a response sheet and an invitation to take part in the study (see appendix 8). A reminder letter was sent to participants after two weeks (see appendix 8). The same procedure was repeated until a sufficient sample size was obtained for the purposes of the study. Participants were not recruited from the Adult Cystic Fibrosis Service within University Hospitals Bristol NHS Foundation Trust because a sufficient sample size was obtained in Wales.

Participants who wished to take part were asked to return the reply sheet to the researcher and indicate their willingness to be considered for the study. Upon receiving the reply slip, the researcher contacted potential participants and their eligibility for the study as specified in the inclusion and exclusion criteria were checked. If appropriate, an interview was arranged with participants at a time and place that was convenient to them.

2.5.4 Response Rates

Twenty eight invitation letters and 11 reminder letters were sent or given to potential participants between the 6th August 2012 and the 29th of November 2012. Four potential participants declined to take part and 14 did not respond. Ten participants responded and one participant was excluded because he did not meet the definition of a primary care giver.

2.5.5 Participant Group Demographics

Four participants were male and five were female. The participants were all white Welsh parents with a mean age of 37. Participants lived in South Wales and received a service from the Adult Cystic Fibrosis Service. Participants were asked to report the highest level of education that they held. Three participants had GCSEs, four participants had A/AS – Levels, one had a degree and another had a different higher education qualification. Two participants defined themselves as a full time homemaker and the remaining seven participants were in either part or full time employment.

Six participants conceived through assisted fertility. Five participants had two children (two with twins) and four participants had one child. Two parents were diagnosed with CF as adults and the remaining participants were diagnosed as children. The participants' lung function ranged from FEV1 40-50 – 91-100% predicted with the mean being 70.2%.

As described, to ensure anonymity participants were given a pseudonym. Only information which was deemed important for interpreting the participants' quotes is presented with the participant's pseudonym in Figure 5 to maintain anonymity. For the same reason, estimated lung function and age are presented as a range. The children's age ranges were constructed to represent different periods of development. Each age bracket has been given a name for ease

of reference when presenting the results in chapters three and four. The pseudonym reflects the participant's gender.

Figure 5: Participant Demographics

Participant Pseudonym	Age Range	Age Range of Child	Lung Function (FEV1 % predicted) (shown as a range)
Adam	41-50 (older)	5-12 years (mid)	61-70
Anna	41-50 (older)	18 + (older)	51-60
Ben	21-30 (younger)	0-4 years (younger)	61-70
Beth	21-30 (younger)	0-4 years (younger)	91-100
Catrin	21-30 (younger)	0-4 years (younger)	51-60
Lucy	31-40 (mid)	0-4 years (younger)	91-100
Luke	41-50 (older)	5-12 years (mid)	91-100
Nina	31-40 (mid)	13-18 years (older)	51-60
Tom	31-40 (mid)	0-4 years (younger)	40-50

2.6 PROCEDURE

2.6.1 Semi-structured Interview Schedules

Research using IPA, requires in-depth, detailed and first person accounts of experiences. In-depth interviews are considered one useful means of collecting this data (Smith *et al.*, 2009). In-depth interviews allow participants to tell their stories, reflect on experiences and explore concerns at length to generate rich data. Interviews should also allow the participant the opportunity to express their ideas about the topics under consideration which may mean the interview extends into unanticipated areas and perspectives (Smith *et al.*, 2009).

This research used semi-structured interviews which were based on an interview schedule and prompts. Questions were designed to facilitate the discussion of relevant experiences with the aim of exploring the research question – *how do people with CF experience being a parent?* Questions focused on exploring their experience of themselves as parents, a person with CF,

the interaction between the two, and their ideas about other people's perception of them as parents with CF.

The interview schedule was developed with the researcher's supervisors and was based on five questions and prompts that were expected to generate an interview lasting between 45-60 minutes (see appendix 9). The interview schedule was discussed with a CF Clinical Care Patient Adviser for Wales and revised on the basis of her comments. Although all questions were applied during the interviews, the questions and prompts were used flexibly to allow the participant to take the interview in their own direction (Reid *et al.*, 2005).

2.6.2 Measures

Participants were asked to complete three questionnaires. These were used to situate the sample and included:

- The Brief Illness Perception Questionnaire (Brief IPQ) (see appendix 10)
- The Hospital Anxiety and Depression Scale (HADS) (see appendix 11)
- Cystic Fibrosis Questionnaire UK (CFQ-UK) (see appendix 12)

Participants were given the questionnaires on completion of the interview so that their thinking during the interview was not biased by the questions asked in the questionnaires. A description of these questionnaires and the rationale for their use is described below.

The Brief Illness Perception Questionnaire (Brief IPQ)
The Brief IPQ (Broadbent *et al.*, 2006) is a nine item scale designed to rapidly assess cognitive and emotional representations of illness. Illness representations have been linked to a range of psychological factors such as coping, mood, adaptation to illness and adherence (Moss-Morris *et al.*, 2002). This measure was used in this research to gain a context of illness experience for the participants, which as highlighted in the research may predict other psychological factors associated with experience of CF.

The Brief IPQ includes five dimensions which are designed to assess cognitive illness representations, which are: consequences, timeline, personal control, treatment control and identity. Two items assess emotional illness representations and include: concern and

emotions and one item assesses illness comprehensibility. The final item is an open ended question to assess causal representation; participants are asked to rate the three most important causal factors in their illness (Broadbent, *et al.*, 2006). This item was not included in this research because the causal factor of CF is well established and not multi-factorial.

Each dimension is measured by a single item scored on an 11 point likert scale. Higher scores on each item represent a greater experience of the dimension assessed. A composite Brief IPQ score was derived which reflects the degree to which the illness is perceived as threatening or benign (shown in chapter three). This assesses the strength of illness perceptions rather than specific beliefs or emotions. The BIPQ was derived from the longer Illness Perception Questionnaire which was revised for brevity, speed of completion and ease of interpreting scores (Broadbent *et al.*, 2006). Some people with CF experience fatigue, so a shorter assessment was deemed more appropriate in this research, therefore the Brief IPQ was used to minimise interview time. Research suggests that the Brief IPQ has good test-retest reliability and there are moderate to good associations between the Brief IPQ and the IPQ-R (Broadbent *et al.*, 2006).

Hospital Anxiety and Depression Scale (HADS)
The participant's mood was assessed using the HADS (Zimond & Snaith, 1983). The HADS specifically screens for feelings of anxiety and depression. Mood was assessed because psychological distress would have an impact on the participant's experiences of being a parent, which was considered important to know when interpreting the data. The HADS was developed with a population of people from non-psychiatric based clinics, so that physical symptoms such as fatigue and dizziness which may be the result of a CID are not assessed as symptoms of depression or anxiety (Bjelland *et al.*, 2002). Therefore, this measure was considered an appropriate mood screening tool for this population.

The HADS consists of seven items assessing 'depression' and seven items measuring 'anxiety'. Each item has a four point response category ranging from zero to three, so possible scores on each measure of 'anxiety' and 'depression' range from zero to 21 (Snaith, 2003). A score of zero to seven is thought to be in the 'normal' range and above eight is considered to be an appropriate 'cut off' score as a case finder for 'anxiety' and 'depression' (Bjelland *et al.*, 2002). An updated literature review exploring the reliability and validity of the HADS using 71 studies suggests that the measure has sufficient internal consistency;

sensitivity and specificity with a threshold of above eight; and *good* to *very good* concurrent validity (Bjelland *et al.,* 2002).

A total score for each participant's symptoms of anxiety and depression was calculated and presented in chapter three.

Cystic Fibrosis Questionnaire Revised (CFQ-UK)

This measure was developed as a tool for assessing HRQoL in people with CF. It was developed as an outcome tool for clinical trials, to assess disease progression and for monitoring individual patients in a clinical context (Quittner *et al.,* 2005). It was considered important to gain an understanding of each participant's subjective experience of CF and the implications of CF on their perceived quality of life in order to understand the context in which they were reflecting on the parenting role.

The CFQ-UK was developed in France (Henry *et al.,* 2003) and has been translated for use in the UK. The CFQ-UK assesses experience on 12 dimensions which are: emotion, treatment burden, health perception, social, body image, role, vitality, physical functioning, eating, weight, respiratory, and digest. These are assessed within the categories of: demographics; quality of life; school, work or daily activities and symptom difficulties. Each item has a four point response category. Raw scores are standardised so for each scale, a participant's score can range from zero to 100. A higher score indicates the perception of a better quality of life. The CFQ-UK was shown to have internal reliability above the cut off of 70 in seven of the scales tested and re-test reliability was also demonstrated in seven of the domains (Quittner *et al.,* 2005). Construct validity was tested through demonstrating an inverse relationship between CF, age and disease severity (Quittner *et al.,* 2005).

2.6.3 Interview Process

The researcher met with each participant individually, on one occasion, for a maximum of one and a half hours. Four interviews took place in the CF service and five were undertaken in the participants' homes. Interviews ranged from 40 to 70 minutes. At the meeting the researcher reiterated the purpose of the interview, outlined the role of the participant and offered them an opportunity to ask questions. Issues of confidentiality and consent were discussed in detail to enable the participants to seek reassurance.

A semi-structured interview of open-ended pre-formulated questions based on stem questions and prompts which were expected to last approximately 45–60 minutes was conducted. Key topics were raised, each with additional questions to expand discussion if necessary. The interview was tape recorded and transcribed. Following the interview, participants were also asked to complete the three questionnaires referred to above, which were self-administered and took approximately 10-20 minutes to complete.

2.6.4 Data Analysis

All interviews were transcribed verbatim. During transcription, the interviews were anonymised using pseudonyms to replace any names used. The researcher kept a record of the process and reflections on changing ideas and assumptions in the researcher's diary throughout the data collection and analysis (appendix 5). There is no single accepted method for analysing data when using IPA but several authors have proposed practical steps to aid researchers in this process (e.g. Landridge, 2007; Smith *et al.*, 2009; Willig, 2008). This research used the method devised by Smith *et al.* (2009) as a guideline for analysis. The steps used are outlined below.

1. Re-reading

The data analysis first consisted of the researcher listening to the audio recordings and reading through the transcripts a number of times. Immersion in the data is considered important so that the researcher is able to 'enter into the participant's world' and also gain an understanding of the interview as a whole (Smith *et al.*, 2009, p.82).

2. Initial Noting

This stage involved writing initial ideas on the margin of the transcript. The initial notes were considered at three levels. The first was a description of the key things which seemed important to the participant (e.g. relationships). The second focused on the language used and initial ideas were noted about how the participant presented meaning through language. The last stage was a more interpretative process in which the researcher considered the transcript at a more conceptual level. This stage required the researcher to draw upon experience and knowledge, for example of the relevant literature, to facilitate this process.

3. Developing Emergent Themes

The analysis moved from a focus on the transcript to a deeper exploration of the initial notes. Themes were identified by mapping the interrelations, connections and patterns in the notes. These were grounded in text but were at a more abstract level. Themes that emerged were illustrated with a quote from the participant to ensure that they represented the participant's meaning (Elliott, *et al.*, 1999).

4. Connections across Themes

Master themes were identified and developed through the clustering of emergent themes. This was carried out visually by exploring all the emergent themes which were written on pieces of paper. Themes which were oppositional were also noted and formed master themes.

5. The Next Case

The same process was carried out for the other eight transcripts. The researcher reflected on the ideas developed from the cases analysed previously and ideas were as far as possible bracketed so that new ideas could be noted on subsequent analyses.

6. Patterns across Cases

The connections between participants were explored visually by looking at patterns across cases. The frequency with which a theme was supported across cases was used as one method of the establishing the importance of a theme. A table to show the frequency of master themes across the transcripts is shown in appendix 13. Three super ordinate themes were developed with master and emergent themes subsumed within these. Themes were organised into a table which consisted of super-ordinate, master and emergent themes. Master and emergent themes were labelled with the participant's own words when possible. Following the data analysis, a literature review was conducted to consider how existing literature related to these findings.

2.6.5 Dissemination of Results

All participants agreed to receive an accessible summary of the results, which was sent to them on completion of the study.

An analysis of the data collected using the methodology described is presented in chapter three.

CHAPTER THREE: RESULTS

3. OVERVIEW OF RESULTS CHAPTER

This chapter contains an analysis of data collected from nine interviews exploring the experiences of parents with CF. IPA was used to identify three superordinate themes along with nine master themes and two to four emergent themes within each master theme. A diagrammatic summary of the themes is presented below. Each master theme is illustrated with quotes from participants. To situate the sample, firstly the results of the three questionnaires completed by participants are presented.

3.1 QUESTIONNAIRE RESPONSES

Participants completed three measures which included: the Hospital Anxiety and Depression Scale (HADS), Brief Illness Perception Questionnaire (BIPQ) and the Cystic Fibrosis Questionnaire Revised UK (CFQ-UK). All nine participants completed the measures. Scores are presented for each measure as well as mean scores for the sample, along with the participants' predicted lung function.

3.1.1 Hospital Anxiety and Depression Scale (HADS)

In this sample, the mean level of self-reported feelings of anxiety was 6.9, which is similar to the mean level for the general population (6.14) (Crawford *et al.*, 2001). The mean level of depressive feelings was 5.4 which is higher than the mean level for the general population (3.68) (Crawford *et al.*, 2001). Within the CF population, HADS scores have been categorised as: 'none' (0-7), 'mild' (8-10), 'moderate' (11-14) and 'severe' (15+) symptoms of anxiety or depression (Quittner, 2012). The largest international epidemiological study to measure self-reported anxiety and depression using the HADS (with this system of categorisation) showed the prevalence of anxiety was: 'none' - 66%, 'mild' - 20%, 'moderate' - 12%, 'severe' - 2% and for depression: 'none' - 87%, 'mild' - 9%, 'moderate' - 3% and 'severe' - 1% (Quittner, 2012). In this sample six people reported symptoms which were classified as

'none' for anxiety, one person reported 'mild' feelings of anxiety and two reported 'severe' anxiety. Self-reported depression was lower, with seven people reporting levels of depression categorised as 'none', one as 'mild' and one as 'moderate'.

Figure 6: Summary of the Participants' Scores on Measures of Anxiety and Depression using the HADS.

Participant's pseudonym	Anxiety	Depression	Lung Function (FEV1 % predicted)
Adam	4 (none)	2 (none)	61-70
Anna	1 (none)	1 (none)	51-60
Ben	7 (none)	7 (none)	61-70
Beth	1 (none)	2 (none)	91-100
Catrin	7 (none)	6 (none)	51-60
Lucy	9 (mild)	5 (none)	91-100
Luke	15 (severe)	11 (moderate)	91-100
Nina	15 (severe)	8 (mild)	51-60
Tom	3 (none)	7 (none)	40-50
Mean	6.9	5.4	70.2
Range	1-15	1-11	

3.1.2 Brief Illness Perception Questionnaire (Brief IPQ)

Figure seven shows participants' composite scores on the Brief IPQ; a comprehensive summary of participants' scores is shown in appendix 14. A higher composite score on the Brief IPQ reflects a more threatening appraisal of the illness. For this sample, the mean composite score was 38.67 (range 23-63). On each domain, participants chose a response from 0-10. The highest perception of threat reflected illness perceptions of time (mean = 10), illness identity (mean = 6.9) and concern (mean = 5.2). Coherence (mean = 0.7) and treatment control (mean = 1.7) reflected the lowest perceptions of illness threat. These results also showed that a lower estimated lung function was not necessarily associated with a more threatening appraisal of the illness.

Figure 7: Summary of Participants' Scores on the Brief IPQ

Participant's pseudonym	Composite Score	Lung Function (FEV1 % predicted)
Adam	25	61-70
Anna	26	51-60
Ben	47	61-70
Beth	31	91-100
Catrin	45	51-60
Lucy	23	91-100
Luke	54	91-100
Nina	63	51-60
Tom	34	40-50
Mean	**38.67**	**70.2**
Range	**23-63**	

3.1.3 Cystic Fibrosis Questionnaire Revised (CFQ-UK)

A summary of the participants' scores on the CFQ-UK is shown in Figure 8 and 9. Figure 8 shows the scales measuring physical quality of life (QoL) and Figure 9 is a summary of the scales reflecting psychosocial QoL. Scores were standardised using the method suggested by the authors and ranged from 0-100 (CFQ-R, 2008). A higher score indicates a better perceived quality of life. The highest QoL on the physical domains were 'eating' and 'weight' with the lowest being on 'physical health' and 'respiratory'. Measures assessing 'body image' and 'emotions' reflected the highest QoL on psychosocial domains and 'vitality' and 'health perception' reflected the lowest perceived QoL on psychosocial domains.

Figure 8: Summary of Participants' Scores on the Dimensions Measuring Physical QoL

Participant	Lung Function (FEV1 % predicted)	Physical	Eating	Weight	Respiratory	Digest
Adam	61-70	33.3	66.7	100	38.9	77.8
Anna	51-60	58.3	88.9	100	77.8	100
Ben	61-70	20.8	77.8	0	16.7	77.8
Beth	91-100	8.3	100	100	11.1	33.3
Catrin	51-60	70.8	100	100	61.1	88.9
Lucy	91-100	91.7	100	100	83.3	77.8
Luke	91-100	29.2	77.8	100	55.6	55.6
Nina	51-60	4.2	77.8	66.7	5.6	55.6
Tom	40-50	37.5	77.8	66.7	61.1	100
Mean	**70.2**	**39.3**	**85.2**	**81.5**	**45.7**	**74.1**
Range		**4.2-91.7**	**66.7-100**	**0-100**	**5.6-83.3**	**33.3-100**

Figure 9: Summary of participants' scores on the scales measuring psychosocial aspects of QoL

Participant	Lung Function (FEV1 % predicted)	Emotion	Treatment Burden	Health Perception	Social	Body Image	Role	Vitality
Adam	61-70	80.0	44.4	55.6	83.3	100	75	33.3
Anna	51-60	100	77.8	55.6	83.3	100	91.7	83.3
Ben	61-70	46.7	22.2	0	44.4	44.4	16.7	25.0
Beth	91-100	93.3	55.6	55.6	61.1	88.9	50	41.7
Catrin	51-60	73.3	33.3	44.4	66.7	100	100	41.7
Lucy	91-100	73.3	100	77.8	72.2	100	100	50
Luke	91-100	46.7	33.3	22.2	33.3	77.8	66.7	41.7
Nina	51-60	46.7	44.4	22.2	22.2	55.6	33.3	8.3
Tom	40-50	73.3	33.3	55.6	83.3	88.9	41.7	33.3
Mean	**70.2**	**70.4**	**49.4**	**43.2**	**61.1**	**84**	**63.9**	**39.8**
Range		**46.7- 100**	**22.2- 100**	**0-77.8**	**22.2-83.3**	**44.4-100**	**33.3-100**	**8.3-83.3**

In conclusion, all measures reflected a wide variety of scores showing diversity of subjective experience and heterogeneity of the sample. As identified in previous studies, self-reported HRQoL (Besier & Goldbeck, 2012; Gee *et al.*, 2005; Goldbeck *et al.*, 2007) and illness perceptions do not appear to be dependent on physical health status which suggests that there other psycho-social factors underpinning experience of CF; this will be explored in the qualitative analysis.

3.2 QUALITATIVE ANALYSIS

All participants had an understanding of CF as being 'forever' which was rated as 10 out of 10 by all participants on the perception of the timeline of CF on the Brief IPQ. One of the most salient aspects of being a parent with CF was the concept of time. Three superordinate themes which capture this context of parenting were derived from the data, these include: *'Being a Parent on Compressed Time', 'Being a parent with Unexpected Time, 'Being a parent with Uncertain Time'.* These themes will be discussed in relation to the aims of the research which were to explore the experiences of parents with CF and more specifically, the experiences of these participants in *staying well to parent and parenting to stay well.*

Quotations are used to illustrate the themes and are marked with the participant's pseudonym, page number and line number from the transcripts. Information which appears in square brackets [] has been added by the researcher and when text has been edited to shorten the quotation three full stops (…) indicate this.

Figure 10: Summary Table of Superordinate, Master and Emergent Themes

Superordinate Themes	Master themes	Emergent themes
Being a parent on compressed time	I'll have them while I'm young	Anxiety Fearing the future
	It's sharpened my focus	New perspectives New meaning to CF
	I've got to prioritise	Putting self second Sacrificing self Missing out Feeling different
Being a parent with unexpected time	It was such euphoria!	Exceeding expectations Elation
	I had accomplished something	Defying all odds Being a success Changing sense of self
	I never think that I've got CF	Normality of CF Being the lucky one with CF Avoiding CF Hoping, wishing
Being a parent with uncertain time	I want to see my children's children	Guilt Bargaining for time
	I look at myself and into the future	Coping with uncertainty & fear The future in focus New perspectives
	Parenting with uncertainty	Noticing the impact on children Parenting differently Making the most of time

3. 3. BEING A PARENT ON COMPRESSED TIME

As expressed by Seymour (2002 as cited in Livneh, 2013) 'living with a disability is living a life dominated by time, but disability is also a time-consuming lifestyle' (p.139). This superordinate theme reflects the challenge that many parents with CF confront when they are parenting with a limited life trajectory but also with a demanding day to day consumption of time through adherence to complex treatment regimes and because illness may disrupt planning and goal attainment. The three master themes captured within '*Being a parent on compressed time'* include both elements of this concept; long term life trajectory and a time consuming lifestyle.

3.3.1 I'll have them while I'm young

The master theme *'I'll have them while I'm young'* emerged from participants' reflections about becoming a parent. All participants spontaneously disclosed that when they reached a decision to have children they then had a sense of urgency to conceive. This was compounded for some by the necessity to conceive through assisted fertility treatment. Most participants reflected on their anxiety to have children quickly once they had made the decision to have them. This did not seem to reflect an urgency to have children when younger and more fertile; the anxiety seemed to be in relation to factors associated with CF and their physical health.

For some participants this was related to fearing that a decline in health could preclude them from having children in the future:

My maternal instinct kicked in and I was thirty…I said look we weren't even married and that but I said we haven't got time for that, I'm thirty. I'm lucky if I can have one. Lucy, p.11, 350-353.

Beth and Nina wanted to ensure that they could spend as much time as possible with their children whilst they were well, this was related to quantity and quality of time.

I felt though at the time was a massive sense of pressure and stress because I thought that if I don't have children while I am very well, I'm going to have either less time with them…it's not going to be

that quality time and it's not going to be the time that I have imagined in my head where you can take your kids out to you know, on day trips and all the rest of it. Beth, p.7, 229-235.

I thought if I'm going to have a child I'll have him when I'm young and even though I always said that I wouldn't have a child until I'm about 30 … I thought well if I'm going to have him then I'll have him when I can play and mess about with him and at least have a couple of years with him. Nina, p. 5, 163-166.

This feeling of pressure or anxiety was exacerbated when participants had to wait for assisted fertility treatment.

The waiting time for my operation [infertility related surgery] alone was four to six years and the answer we gave to that was again, as I said it seemed a bit extreme, but was we said well I could be dead in four to six years. Tom, p.12, 339-341.

First time was unsuccessful [IVF] and that was very hurtful. I remember thinking at that time and I'm sure [wife] did as well, how much time have I got on this planet having CF, how much time is actually left for me to [pause] and you start working it out. Ben, p.3, 94-96.

Ben described it as a timeline and compared it to other people's experiences of 'fitting things in' in their life.

It's a timeline and you're thinking to yourself …if I have them now, I'll have to be that old before. I've always said if they are 18, 20 and if I die… Ben, p.3, 99-101.

I suppose women do it in the opposite by working out well I want to get married by and I want to have kids by this age. Ben, p. 3, 96-98.

Tom explained that his wife felt concerned about time and although he agreed, in the context of his understanding of a 'normal' life trajectory children came too soon. He talked about finding it difficult to adjust the responsibility and the permanence of a child.

I don't think I really thought along the same lines as my wife did where you know come on we need to get it done now just in case you know. Tom, p. 12, 367-369.

Between 6 months to a year we were talking about having children whereas I think normal couples or couples where there isn't sort of CF or anything like that affecting the relationship …I think they wait a bit longer than six months to start talking about sort of having children. Tom, p.13, 371-374.

Coming back to that sort of responsibility it sometimes feels that it can be, oh you can't switch off from this. Tom, p. 7, 197-199.

Lucy also reflected on the timing of having her children and thought that she had left it too late. This difference was a result of needing to negotiate timing with her partner.

I said to him it's your fault I should have been 25 having my children, not old, I said my health you know I shouldn't be having kids at 30 I should be having them a bit younger. Lucy, p. 16, 527-529.

This sense of urgency and the resulting anxiety experienced when the participants could not conceive children as they had hoped, had a significant impact on their current parenting experience and provided some of the context to the master theme *'It was such euphoria!'* Some parents demonstrated a remarkable ability to frame very demanding experiences positively perhaps consistent with the priority they assigned to their wish to have a child whilst relatively younger and therefore potentially healthier.

3.3.2 It sharpened my focus

Since having children, the majority of participants expressed a new perspective on keeping well and a resulting motivation to adhere to difficult and time consuming treatment routines in the hope that they could lengthen their life and remain as well as possible to be a parent. One participant explained that she noticed this change when she was pregnant:

When you're pregnant all of a sudden if you are ever lax with doing medication or anything like that you suddenly realise the importance of it because you want your children to be as healthy as possible. Beth, p.3, 94-96.

The three mothers with younger children said that a shift had taken place cognitively and they were aware of wanting to adhere to treatment but in practice this had not happened and they explained that this was a result of time constraints. This theme is linked to the master

theme *'I've got to prioritise'* in which this experience is explored in more depth. This seemed to cause all three mothers with younger children feelings of anxiety.

I'd like to say that having children gives you that massive motivation to stay as well as possible and it absolutely does because if I'm unwell because I haven't done medications or anything like that then obviously they are going to suffer as well but I don't know how that is going to go in the next few months. Beth, p.4-5,133-137.

If you don't exercise you're taking ten years off. And I'm, and I'm consciously, I'm always thinking of that now, you know I've got kids I want to see my grandkids. You know, so it gets me down a little bit thinking, I got to exercise, I got to exercise but I think when? Lucy, p.8, 263-266.

The fathers of young children had a slightly different perspective and described a new sense of responsibility as a motivator to keep well for their whole family.

I think yeah you should pull your finger out and do it for your family and your children yeah. Tom, p.4, 120-121.

You are very mindful of something that you're nurturing…and the family that is so and you want to stay you got this emphasis that you want to stay around the family as long as you can. Luke, p.9, 301-304.

Because of my drugs and routine that we have to do urrm there is no point in me seeing my kids if I am going to wreck my routine because unless I'm here for the majority of their life or a large proportion of it there is no point. Ben, p.1, 10-12.

The mothers with older children also described a strong motivation to keep well for their children so that they could continue to see their children grow up and they were able to implement this in practice. They explained how their children are involved in helping to keep them well by reminding, encouraging and helping practically with treatment.

I honestly do not miss nothing, whereas years ago if I didn't have [child] and I had all this treatment years ago I think I probably would have missed some if I'm honest. He keeps me focused to stay alive

basically to stay healthy as I can to see him grow up and not get into any trouble. Nina, p.4, 121-124.

So he would be making me, it wouldn't be my husband or my family making me, it would be my son saying mum you haven't done your nebs yet, mum you haven't done your drugs yet. Nina, p.1, 31-33.

He'll [son] do my physio for me and that sort of thing, you know. Anna, p.5, 163-164.

This change in perspective is linked to the superordinate theme *'Being a parent with uncertain time'.* Parents described an uncertain future which had come into sharper focus when they took on the responsibility of becoming parents and this uncertainty in combination with responsibility seemed to serve as a motivator to *stay well to parent* for many participants.

3.3.3 I've got to prioritise

This master theme reflects the parents' experiences of managing the two roles of being a person with CF and being a parent. Both roles are time consuming and both are invested with powerful internal and external expectations about the importance of undertaking them successfully. For these participants, this required balancing and juggling the two roles which had implications for their emotional and physical wellbeing. When one role was performed at the expense of another, some participants felt guilty, anxious, sad, and frustrated and a feeling of loss was evident. This master theme contains accounts relating to past and present experiences and ideas for the future.

As captured in the theme '*It sharpened my focus'* the experience of many participants was a new perspective on keeping well but in practice this was enacted very differently among participants. The majority of participants expressed that caring for their child(ren) took priority.

I come second now; I don't look after myself like I should. But you know my kids come first and unfortunately I don't get a chance to look after myself as well. Lucy, p.2, 55-56.

When you have them it does seem a strange thing, you got less time and you're not so conscious about yourself. Beth, p.3, 97-99.

It's not about you anymore and it's never about you, it's about your children. Ben, p.11, 162.

Nothing or no one comes before him. Nina, p.13, 408-409.

If I had to weigh up do a bottle or do my nebulisers, it had to be the bottle. Catrin, p.3, 71-72.

Managing both roles was achieved by many parents through adjusting CF routines to fit around the child(ren).

If I got up in the morning I sometimes used to get up about five just to do my chest clearance so before, so when he woke up I could give him his bottle without coughing and spluttering all over him. Nina, p.3, 93-95.

I like my schedule and I like to do my physio or do this at certain parts of the day, so I've really had to sort of try and bend that around you know helping my wife with [child(ren)]. Tom, p. 5, 126-128.

All participants explained that they sometimes had to de-prioritise their own needs to attend to being the child's primary caregiver. However, the majority of the mothers reported always putting their children's needs first and endured feeling physically unwell to ensure that they fulfilled their ideals as a mother and remained being the primary caregiver.

I'm thinking oh my God I could cough loads of blood up and think oh my God this blood could kill me but I know my son's playing football and I'll put that aside… I'm thinking in my head I just want to go home and I want to cry and I'm thinking oh my God, I could die. Nina, p.8, 264-268.

The first thing it does is make me tired and runs me right down. If I got a chest infection I just want to go to bed but I know that I can't, so I just get on with it. Lucy, p. 10, 321-323.

I feel quite annoyed that I have to go on IVs and frustrated because I know I'm going to be on, you know I'm going to be tired, it's going to be that extra workload, while I'm trying to look after these. Beth, p.6, 186-187.

Some mothers expressed how this 'endurance' was impacting on them now and how they feared the future.

I am constantly on the go… it doesn't help my mental health because I am exhausted, Lucy, p.6, 199-200.

Even though I love him and he's my motivation sometimes the bad days can weigh out the good days especially now that my health has deteriorated and I think oh God. Nina, p.5, 157-159.

Some mothers also described parenting experiences which appeared at times to be isolated and lonely. They reported little help from their own families.

We had no family support or anything, nothing no. Anna, p.3, 86-87.

It's just me the unlucky one to have parents who just move to the back seat more than coming down to help me. They'd help my sisters before they came to help me so that was probably the most difficult thing for me than anything else. Nina, p.4, 102-105.

Lucy explained that she did not want to be supported differently because of the CF.

I don't want you to give me extra help because I got CF I want you to give me the extra help or the help because you want to do it because it's your grandchild, not because I need it. Lucy, p.21, 706-708.

The fathers also expressed a need to adjust treatment routines to fit around the needs of their children but adhered to treatment at the cost of spending less time with their family in the short term with a view to spending longer time with their family in the long term. These experiences were reflected in the emergent theme of '*Missing out*' and '*Feeling different*'. The fathers expressed frustration, sadness and described the strain that this places on their partners.

The only thing that I hate is actually coming in here [hospital] leaving them all behind that's, that's really hard, Adam, p.6, 199-200.

Not only does it take you away from your family and it's not the normal thing, you know it's not normal you know people; families without cystic fibrosis don't have to contend with that… it can get on your nerves to say it politely. Tom, p.2, 43-47.

The first thing I do is bang straight on my neb and it is hard to sit there and watch my kids but I know that within 15-20 minutes I can do that…for the partner I think that is a lot harder for them. Ben, p.10, 333-335.

I think it is particularly difficult the week in hospital on the person who is left at home looking after the child... And it's obviously you know difficult on me as well because I am obviously away. Tom, p.1, 11-13.

This is in contrast to two of the mothers who describe their conflicting feelings associated with the possibility of needing to go into hospital and suggested that they would set different criteria for deciding on taking time away from their families to adhere to treatment demands.

A couple of times the doctor would say to me 'If you don't come in you could die' and I'd be really scared thinking I really want to go in and I'd really want to go in and I'd really want to have a rest and this and that and I'd look at my son and he'd be standing there about that high and I'd think oh god, I can't put him through that. Nina, p.2-3, 66-69.

If I did have to go in it would have to be childcare that would have to come first. Catrin, p.10, 320-321.

Anna was an exception to this theme; she described no struggle in prioritising both roles. This may be linked with the master theme *'It was such euphoria!'* as she had waited a considerable amount of time to have a child and explained:

It's still elation the fact that we actually had a son so I've never found it a struggle. Anna, p.2, 66.

Anna also seemed to cope by accepting a need to co-parent which implied drawing on her husband's abilities to compensate for what she could not do and enabled her to feel that she was not missing out on fulfilling her role as a mother.

I can't honestly think of anything that I haven't been able to do with him because I got cystic fibrosis you know so I urrrm. I couldn't go running around the fields with him but his father could do that. Anna, p.6, 172-174.

In relation to the powerful internal and external expectations that participants felt were impacting on their priority setting, some participants described a 'trade off'. They explained that they did not feel as responsible or felt that they could not be 'blamed' if their health declined since having children because it would be understood that combining CF treatment and parenting is challenging. This seemed to reflect a sense of responsibility to maintain their health for others around them but a sense that others should make allowances now that they had become parents. However there was an underlying feeling of failure and resignation in their disclosures.

I'm sure they do understand the deterioration from pregnancy and having a child and that is taken into account and you're not really expected to keep up your lung functions as much. Catrin, p.10, 328-330.

They do look at the whole situation now that I do have a child and probably understand that you know you are up in the nights and it's quite, you are going to be doing more etc so if I do get ill possibly more often, I suppose they know the reason why really. Tom, p.9, 250-253.

By contrast, Luke believed that others should see him first as a unique individual and that to allow treatment priorities to dominate how he lived would mean that he was 'giving up'. Failure appeared to be associated with adherence to prescribed treatment regimens for this participant.

And also on the other side respect yourself and realise that you got to be you know err kind of errm kind of careful with yourself almost and you know a bit of self-respect I think and I think respect it but don't let it dominate. Luke, p.10, 327-329.

I probably am non-compliant but you know there is a bit of psychology in that as well, the minute that I feel that I have to that I need it, I've lost myself, I've lost my determination. Luke, p.3, 97-99.

3.4 BEING A PARENT WITH UNEXPECTED TIME

The second superordinate theme *'Being a parent with unexpected time'* also refers to the context in which the participants are fulfilling their parental role. The majority of participants were not expected to live into adulthood or become parents. In addition, many of the participants did not expect to have children due to CF related infertility and many went on to experience a period of uncertainty when accessing assisted fertility treatment and trying to conceive. This theme reflects the participants' feelings of joy and elation in exceeding past expectations in the master theme *'It was such euphoria!'* and the participants' changing sense of self as they surpassed their expected potential is captured in the master themes *'I had accomplished something'* and *'Never think that I've got CF'*. This superordinate theme reflects the concept of *parenting to stay well*.

3.4.1. It was such euphoria!

The master theme *'It was such euphoria!''* emerged from the majority of the participants describing what they expected to achieve in life and how they experienced life after they judged themselves to have achieved more than this by becoming parents. Expectations were internalised from parents, family and the medical community and reflected the understanding of CF when the participants were children. Some participants also experienced dissuasion from their own parents to have children. Expectations differed in relation to the age of the parent with older parents experiencing fewer hopes for meeting developmental milestones such as having children and perhaps a greater sense of achievement when these were exceeded.

Some participants explicitly described expectations from the past and connected this to how they felt when planning or having children in the present day.

I was told, told don't go too high, don't you know, exceed yourself and then obviously I didn't tell them I was trying for a baby because I knew they would have literally have said no. Nina, p.9, 289-291.

When I was diagnosed growing up they used to expand my life expectancy every couple of years, I mean when I was diagnosed when I was two I think they gave my parents until I was five, until I was eight and you

know when I became when I was about 12, it was like possibly 17 or 18 and even then I think it was-I can't die yet I want a baby. Anna, p.10, 323-328.

They always said when I was younger – oh she won't reach five years old, oh she won't reach 18 and then all of a sudden I think I was [age] having him, [age] and I got a little boy, I defied all the odds…people do they, you're having a baby, you got a couple of years we are starting making your cardboard box now, that's what it's like. Lucy, p.18, 580-585.

They tell people that they didn't really think that I was going to ever of had children or that I would manage as I do. Catrin, p.7, 221-223.

However, Beth who is one of the younger participants described how the expectations from her family and professionals differed. She explained that through having her children she had discovered these discrepant views and this had caused her to think about herself with CF in a different way.

My mum has always assumed I was going to have kids and she is going to be a grandmother. Beth, p.9, 228-229.

There were a few doctors there that had never seen a mother with CF, let alone a [age] year old with CF that was as well as I am…one of the doctors the main doctors…referred to me on several occasions as a medical miracle. Beth, p.5, 149-152.

Makes you think am I really that lucky or am I, you know it kind of brings it home to you that actually I do have something wrong with me. Beth, p.6, 175-176.

The majority of the participants conceived children through assisted fertility. Adam described how he felt about infertility and how it changed how he felt about being a parent.

The hardest part was actually getting them, how long it actually took and when you see people on the street…having absolutely loads of children and not looking after them…you think why, why you know it's not fair…it's made a difference on how I look at children. Adam, p.8, 240- 267.

I've gone from being told over the phone by a GP that I can't have children to you know at that point breaking down. Luke, p.10,315-316.

Participants described a resulting feeling of joy and elation when they became parents; this was still evident on reflection some years after their children were born.

Having two beautiful children that are mine you know they are not from the sperm bank or anything like that they are mine so massive euphoric. Luke, p.10, 320-321.

It was just, just, wild, it was funny, after waiting so long for children it was nice. Adam, p.11, 349-350.

We had to have treatment and I think having them gives you that massive sense of pride…you've got them after all the hard work of you know trying to get them or trying to get pregnant. Beth, p.7, 222-224

I mean we wanted a family for so long that you know it's still elation the fact that we actually had a son. Anna, p.2, 65-66.

I had this baby and I was like, he's amazing and I felt so, oh I was on cloud nine for the first year. Nina p.9, 303-304.

Proud very kind of okay I never thought this would happen no one thought that this would happen…I didn't think I could have kids. Ben, p.2, 61-63.

Anna also went further to explain that she thinks that feeling of elation has helped her to stay well; she made a link between her physical and mental health and described how like the weather, her emotions can also affect her physically.

My health tends to deteriorate with the weather, big time, affects me very much so…and any sort of emotional effect then, tends to affect me. Anna, p.4, 116-120.

I think in some ways it has helped my health…we sort of went through a lot of trauma trying to have children…the elation of actually having that child I'm sure within myself has helped, you know my wellbeing. Anna, p.3, 97-101.

3.4.2 I had accomplished something

Some parents also described how exceeding expectations had changed other peoples' perception of them and their sense of themselves as being more valued as people with CF. This was in contrast to feeling viewed as selfish for having children with a life limiting condition which was described in the theme *'I want to see my children's children'*. Both perceptions were experienced by some participants and seemed to vary with the context.

This probably sounds absolutely ridiculous but I think you gain a little bit more, I don't know,…respect [as a result of having children]. Tom, p.8, 226-227.

I think I liked them to think, that I did actually accomplish something. Nina, p. 13, 420-421.

It's just a look, it's not that they treat you or speak to you any differently, they look they give you of kind of potential, not a potential that's the wrong word, totally is the wrong word. You've achieved, or they've achieved [medical professionals]. Ben, p.16, 539- 542.

For Ben, he felt he was judged negatively by some friends and family but as a success in the eyes of CF professionals. He thought that they should also be praised for their part in helping him to be a success by keeping him well enough to have children. This quote illustrates Ben's empathy for CF professionals and his desire to be a 'success' not only for himself but for others too. This sense of shared accomplishment seemed to contribute to Ben's feelings of anticipatory guilt if his health was to deteriorate and he died younger than expected. He feared their joint accomplishment would be diminished if his health declined.

I think it is very hard for them to see people who they have spent a lot of time with medically and then they pass away. I've been in the service for a long time and they've spent a lot of time on me. Perhaps it's a fist in the air moment when those kids were born. Ben, p.18, 578-581.

Some participants explained that being a parent had changed their perception of themselves. This was mostly a positive change and some parents had a feeling of being an achievement as a result of fulfilling an important goal in their lives:

I definitely look at myself as an achievement, Ben, p.17, 560.

I was just self-satisfied that I had achieved what I wanted to achieve out of life… I have certainly changed totally in character and in personality. Anna, p.10, 312-314.

I think they thought that I would never get a husband, I know that because they probably wouldn't want to be with me…I thought to myself you know what I've brought my house, I've paid off my mortgage, I've had a job on and off and I had a son. Nina, p.10, 320-325.

Beth and Nina also described how their friends who viewed them through the eyes of parents without CF, perceived them in a positive light because of the way that they manage the two roles of being a parent and having CF.

Lots of my friends, especially my best friend she thinks that I am some kind of like ah some kind of like super hero or something because I have managed to do it with CF. Beth, p.8, 246-248.

I think they are stronger than what they thought I was, I think I know my friend says to me I can't believe how you cope you are amazing. Nina, p.11, 368-369.

3.4.3 Never think that I've got CF

Exceeding expectations was a contrast to a master theme *'Never think that I got CF'*. Some of the same parents explained that they did not think that having CF played a large part in their decision to have children or said that they did not frequently consider CF in their parenting role. However, they also still described many of the ideas contained in all three of the superordinate themes which suggested a significant impact of CF on their identity as parents and on their anticipated parenting roles in the future. This is reflected in the theme *'I look at myself and into the future'*.

For some parents this may be because CF and its treatment is 'normal to them' (Williams *et al.*, 2009) so largely their identity as a parent was not linked to CF.

I don't tend to think of anything I just think, I'm having kids and the only change for me is I'll be looking after kids. I didn't think - oh how is my CF going to come into that. Lucy, p.13, 435-436.

I never think of myself as having anything wrong; just being normal. Beth, p.12, 387.

I say I've got practically as much energy as a dad without CF to be honest so yeah. Tom, p.13, 389.

I've never had a child without having cystic fibrosis. Anna, p.6,171.

Linked with the master theme *'I had accomplished something'* many participants described being lucky with CF and contained within this theme were ideas of hoping, wishing, denying and trying to forget that CF was a part of their life as a parent because it hadn't yet 'caught up with them' and they were the 'lucky parents with CF'.

I tend not to talk about it anyway because I don't think; I don't look at it as an illness. Adam, p.16, 520-521.

I know that the life expectancy has always been around 30 but I could never really see that for myself because I have always been so well. Catrin, p.6, 195-197.

Catrin and Nina explained how they viewed doing treatment:

I just blank off and it's just a mother who does the washing, I just treat it as that, it's just another chore, Nina, p.4-5, 135-136.

I don't think about it, if I'm honest it's at the back of mind, it's a chore, it's something that I have to do every day otherwise I will struggle to breathe. Catrin, p.4, 127-128.

However, for some participants CF seemed to take on a different significance in their identity as a parent if they experienced an exacerbation or deterioration in their health.

I am like a normal mother apart from obviously when I am ill then obviously I am ill. But luckily day to day I am you know I go to baby groups and all that nobody would ever know that there was anything wrong with me. Lucy, p.13, 442-444.

Nina reflected on how this perception had changed over time as her health had worsened.

I just never thought CF would catch up with me, so because you're healthy you don't think of what, you read about what the future could hold. You don't actually think it's going to happen to you. So not really I didn't think it [being a parent] would have an impact. Nina, p.5, 151-154.

3.5 BEING A PARENT WITH UNCERTAIN TIME

The third superordinate theme *'Being a parent with uncertain time'* refers to how parents experience managing the uncertainty of CF and the time ahead of them. Contained within this theme are the participants' feelings, thoughts and strategies for managing this uncertainty for themselves as parents and for their children.

3.5.1 I want to see my children's children

Many of the parents managed the uncertainty of time through creating a target age that they would like to reach before they died which was based on living to see their children reach a certain age. The master theme *'I want to see my children's children'* is a reflection of the guilt and sadness that some parents experienced as a result of having children with an uncertain lifespan and this seemed to be one way of managing it. Ben and Anna described how they have experienced being judged or being made to feel selfish for having children. Ben described his fear of being blamed for his children 'failing' after his death.

I think in a selfish way they look at me and think how could you be so selfish to have kids knowing you are not going to live long. And I've seen a few people look at me like that. Ben, p.15, 500-502.

I want them to succeed but I don't want them to fail because of me. So I don't want to be blamed for that. Ben, p.4, 129-130.

I still used to think very selfishly probably even if I knew I was gonna die young I still wanted, that was what I wanted out of life, I wanted a baby. Anna, p.10, 333-335.

Anna described this in the past tense and seems to be struggling less with feelings of guilt or anxiety about the future in comparison to other participants and this was reflected in her

very low scores on the HADs (See Figure 6). This may be a reflection of a feeling that she had already achiev*ed*:

I was just self satisfied that I had achieved what I wanted to achieve out of life. Anna, p.10, 312.

Some of the participants based their target age on concrete factors such as the level of education they would like their child to reach or a developmental milestone such as their children having their own children. This seemed to reflect a desire to have the time to experience joy and pleasure from parenting their children, as well as to fulfil for their children's needs as they matured.

I know the importance of keeping myself well now so I can hopefully grow up and see them get married themselves, have children themselves. Tom, p.6, 166-167.

Because now I got CF and I got children, I want to see my children's children. Lucy, p.14, 467.

I'm a bit selfish I'd obviously want to be there to see them smiling happy and joyful. So I've always said 18, 20 years I'm happy to, it's not a massive lifespan but for CF it's huge. I'd be 50, 52 then. Ben, p.4, 111-113.

Some participants explained that they would be able to 'cope' if they were able to support their children to reach a certain age. Commonly this age seemed to signify their child had reached adulthood and no longer needed them in the same way. Ben described how he thought it would be traumatic for children to lose a parent but it would be preferable if this could happen after the child had gained a level of education which would 'set them up' for the future.

I've always said if they are 18, 20 and if I die, or pass away sorry err I can handle that because I think they would have done all their GCSES all their A 'levels or whatever they have got to do and they are going to uni…they have got that underway and you can always, you can never take that away from them, Ben, p.3-4, 101-105.

I got really anxious when I used to see blood I got anxious that maybe I wouldn't, I always had in my head that if I can get my son to 16 and then anything happened then I could cope. Nina, p.3, 76-78.

This master theme reflects the concept of *staying well to parent* and is linked to the theme *'It sharpened my focus'* as this strategy gave participants a concrete goal and a new motivation to reach their child(ren)'s target age. However, as explored in the theme *'I got to prioritise'* being able to look after themselves in the context of managing two roles was not always perceived to be possible. This seemed to lead to a heightened anxiety and a greater fear about the future, a theme which is explored in the next section *'I look at myself and into the future'.*

3.5.2 I look at myself and into the future

The master theme '*I look at myself and into the future*' reflects a new perspective that participants said they had experienced since becoming a parent. Many of the participants explained that they had not considered their future in the same way until they became parents and since then they had started thinking and worrying about the future and their mortality. This perspective also shifted as changes occurred in their health. The participants described how they manage this new perspective through different styles of coping.

I never ever thought of my future before. Never thought oh when will I live to or anything like that but now you got kids, now I got kids I do think all the time about you know will I. Lucy, p.15, 484-486.

It didn't used to, it didn't used to bother me at all. Lucy, p.15, 506.

I think you are very mindful of err something that your nurturing that you have a big part to play in developing it and the family…you want to stay, you got this emphasis that you want to stay around the family as long as you can you know and that's there all the time. Luke, p.9, 301-304.

It is you know more of a concern you know that I do have a child that you know will need looking after in the future, I never really thought about it before. Catrin, p.6,192-195.

Yes. Yes before I never really cared. Ben, p.5, 136.

I've been really lucky but all of a sudden you think am I, is it just luck you know, am I really going to get that bad or do you know you think of the negative things. Beth, p.5, 162-163.

Ben explained that watching his children develop had made him more aware of the deterioration in his health and his life trajectory in comparison to how his children's lives were progressing. This increased his anxiety about the future:

I was thinking well hang on they are progressing [children] and I'm not I'm getting worse if anything so how does that bode for the future. Ben, p.8, 248-249.

Participants reflected on how they could cope with this new perspective on their mortality. Ben devised a problem focused way of coping through making concrete changes such as planning to go on holiday sooner than expected.

So I've always said to her let's bring it forward to maybe five or six [years]and its coming down now [lung function] and that's why I'm starting to question how long have I got left. Ben, p.5, 165-166.

A more fatalistic way of coping was adopted when efforts to control CF were not effective or because they felt that they were already 'doing' what they could to keep themselves well, so the rest was down to luck.

For now, I just got to get on day to day. I don't think of it. Lucy, p.15, 503-504.

I try not to think about it too much because there is nothing that I can do about it until the time comes and I get ill. Catrin, p.6,193-194.

I think if you can increase life then brilliant bla bla but I always thought if it's my time to go, it's my time to go. Ben, p.5, 138-140.

Ben moved between these two positions which may reflect the need to cope in multiple ways when managing such uncertainty.

This is in the sense of money can't help me, nothing can help me. Ben, p.8, 237-238.

The only thing that can help me is myself and if I keep on exercising and do my medication then I can hold off CF I'm not saying you can't beat CF you can only hold it off for so long and the longer I hold it off the better. Ben, p.8, 240-242.

Luke and Lucy describe how they felt emotionally when they thought about their mortality and Beth described her fears for the future.

Sometimes you do dwell on it but I used to sort of beat myself up but I don't anymore you know when you think well sometimes that it's going to happen and you snap out of it, Luke, p.2, 62-64.

Yeah, I mean some days I don't think about it at all and other days then especially when I am not feeling well, I get all depressed. Lucy, p.16, 540-541.

A lot of my friends with children that are a lot more unwell than me to put it that way erm get very down about it and get very frustrated. So I hope I don't get like that one day. Beth, p.6, 191-193.

Some participants feared what might happen to their children if they became unwell or died.

Now I have I think to myself if I weren't around what on earth would happen to them two. Lucy, p.16, 510-511.

I'm very conscious that the minute I get ill or the minute that I'm not 100% it's going to affect my children…I'm not going to be able to take such good care of them as I might… that I would if I was 100% well and in good health. Beth, p.13, 409-416.

Furthermore, when Ben was unable to improve his health despite trying, this led to a feeling of being out of control and a greater feeling of anxiety.

I didn't expect to become an athlete I expected a slight increase of my lung function and it didn't happen and I kind of thought oh Jesus, Ben, p.5, 159-160.

It's coming down now [lung function] and that's why I'm starting to question how long have I got left. Ben, p.5, 165-166.

3.5.3 Parenting with Uncertainty

This master theme reflects participants' experiences of noticing the impact of the uncertainty of CF on their children, the way in which they managed parenting in this context and the feelings associated with these experiences.

Some of the parents described the ways in which their children reacted to the uncertainty and separation from them. This was only described by the participants with middle or older children.

He was very very nervous, worried about me leaving him, worried about me dying, he thought I could go to bed one night and not wake up…I'd wake up in the middle of the night sometimes and a hand would be there and he would be checking if he could feel my breath. Nina, p.15, 484-490.

I'll find myself saying I won't be around forever and what you need to do is, all those kinds of things you know, and the kids don't like that, no they don't like it they get upset by that…and then you do feel guilty. Luke, p.6, 200-203.

It's a bit sad sometimes every time I go home after about a week he says yeah are you home for good and I'm like nah and he is like oh okay so he does, he is missing me. Adam, p.18, 583-585.

The way in which the participants managed parenting as a result of this uncertainty was discussed by some parents. Luke and Ben described how they believed that they were more protective as parents with CF:

The children thing, I am very very very protective over my children you know, I think, I think the CF obviously has its part to play in that you know in terms of how I bring them up you know in all kinds of ways really, Luke, p.5, 140-143.

So my parenting is going to be, I think I've become more protective or very kind of wrap my arms around my kids and we didn't let people in for the first few weeks…we were like no you might have a cough, cold, we are not even taking chances. Ben, p.13, 408-412.

Luke and Adam described trying to manage CF by hiding it or avoiding talking about it with their children.

I don't want to pass on anxiety, so for me I tend to err hide a lot of things and clam up a little bit that way, you know I don't divulge anything. I certainly don't make a play of it if I can help it you know. Luke, p. 8, 550-552.

She never talks to me about it [daughter]. I would talk to her about it but I don't really want to. Adam, p.18, 616.

Anna and Nina manage this in a different way; they told their children about CF and encouraged them to participate in their treatment. However, they both acknowledged limits to this and contemplated the effects of being open about CF.

Sometimes [husband] says that I tell him too much but I've always been very open with him about everything…they accept it better don't they, if it's not suddenly thrown at them as a shock at the end of it. Anna, p.9, 282-285.

He likes to know but I don't tell him everything obviously but he likes to know that everything is okay. Nina, p.2, 54-55.

Luke and Nina described how they did practical things to prepare their children for an uncertain future and compensate for the effects of having a parent with CF.

I got to look after them or and you know try and teach them…there is a lot of philosophy that is passed on I think, you know that I do to help them later because I am always thinking later later on. Luke, p.1, 27-30.

Exactly and none of his friends mothers have been ill at all they are all working…I just feel I should be the same as them so I try to overcompensate then my making sure that he has everything that he wants. Nina, p.7, 205-207.

Luke also did practical things to ensure that his children would know he cared for them after his death.

What I do is they are not aware but I write all notes on their little letters … Luke, p.2, 46-47, ….You know, sometimes it's hard like but…if it's my son playing rugby I write the same thing on there…so they can at least you know at some point or another you know they can see that and they can think oh I did actually care. Luke, p.2, 49-52.

Parenting with uncertain time and perhaps linked to the elation described in the master theme '*It was such euphoria!'* led some parents to suggest that they were fulfilling their role as a parent by treasuring time and immersing themselves fully in the experience.

I think when you have got maybe an imaginary gun pointing at you… it makes you feel more appreciative of things definitely. So you know I can pass on to my kids, I want them to appreciate everything. Luke, p.13-14, 440-443.

I want my life to revolve around them; I want every moment with them. Ben, p.6,172-173.
You just want to put everything into that child, you know to fulfil your life, Anna, p.8, 241.

3.6 Conclusion

These findings suggest that being a parent with CF results in a different perspective, experience and awareness of time. *Being a parent on compressed time* links directly to the experiences of participants in *staying well to parent. Being a parent on unexpected time* refers to experiences relating to *parenting to stay well.* These results will be discussed in more detail and in relation to existing literature and theory in chapter four.

CHAPTER FOUR: DISCUSSION

4. OVERVIEW OF CHAPTER

This chapter contains a summary of findings and a discussion of these in relation to relevant literature and theory. The clinical and service implications of these results will be outlined, followed by a discussion of the methodological strengths and limitations of this study. Finally, ideas for future research will be described. When themes are cited, italics will be used.

4.1 RESEARCH FINDINGS & EXISTING LITERATURE

The main aim of this research was to explore how people with CF experience being a parent. To the author's knowledge this was the first study to investigate the experiences of mothers and fathers with CF. Three superordinate themes emerged from the data, which include: *Being a parent on compressed time; Being a parent with unexpected time;* and *Being a parent with uncertain time.* These superordinate themes, and the master themes contained within them, will be discussed in relation to existing literature and theory. However, as described the evidence base relating to parenting with a CID is limited in its application to understanding the psychological processes underpinning this experience and more specifically in its relevance to making sense of the unique challenges associated with CF.

As described in chapter one, personal experiences such as a CID are thought to influence the perception of time (Livneh & Martz, 2007; Livneh, 2013). Successful temporal functioning requires an ability to balance the integration of past experiences and future hopes and to apply this to current decision making and experience; this requires adaptation involving internal and external adjustments (Livneh, 2013). Therefore, a strong relationship between coping and temporal adaptation is hypothesised. However, to date research in this area is limited and inconclusive (Livneh, 2013). In the current study, parenting with CF is understood to influence the passage and experience of time; CF is a time limiting and time consuming condition and parenting represents a transitional event in life trajectory (Rolland, 1994).

Temporal adaptation in this context is explored with reference to the coping processes adopted to integrate past experiences and future hopes into current experiences of being a parent. As previously described, secondary control is made up of two processes; the first being an adjustment to some aspect of the self and the second an acceptance of circumstances as they are (Morling & Evered, 2006). An engagement in both processes is understood as secondary control although some researchers have also explored the benefits of engaging in either process in isolation (Morling & Evered, 2006). The theory of secondary control will be used to understand these findings as both processes of acceptance and adjustment were emergent in the participants' accounts.

Therefore these findings will be discussed with reference to the relationship between secondary control (Morling & Evered, 2006) and temporal functioning (Livneh, 2013) to understand the experiences of parents in *staying well to parent and parenting to stay well*. A judgement or an assessment of 'successful' or 'unsuccessful' temporal functioning will not be made, rather the participants' adaptation and adjustment to their context as a parent with CF will be considered. Findings will be compared to the evidence base on parenting experiences with a CID. A detailed account of each superordinate theme will be discussed with exceptions and discrepancies between participants noted to provide a rich account of the findings.

4.2 BEING A PARENT ON COMPRESSED TIME

The superordinate theme *Being a parent on compressed time* captures the participants' experiences of parenting with a limited life trajectory but also with a demanding day to day consumption of time (Seymour, 2002 as cited in Livneh, 2013) through adherence to complex treatment regimes and disruptions associated with illness. This theme could be understood as the participants' adaptation towards a future time orientation (Livneh, 2013; Zimbardo & Boyd, 1999) which is utilised to make current parenting decisions based on the perception of compressed time. The master themes: *I'll have them while I'm young; It's sharpened my focus; and I've got to prioritise* may reflect adjustments to aspects of the self, whilst accepting circumstances relating to time as they are (Morling & Evered, 2006). A future time perspective is associated with good decision making and greater goal attainment (Livneh, 2013) and in this context may serve to guide participants in *staying well to parent* by making

choices with a view to the future. The cognitive and emotional challenges of coping with this perception of time are explored below.

4.2.1 I'll have the while I'm young

Some participants disclosed that when they reached a decision to have children they experienced a sense of urgency to conceive and described the anxiety that they had felt when this took longer than hoped. In accordance with the findings of Simcox *et al.* (2009) participants considered the timing of having children in the context of their health and life expectancy. This decision was based on their own needs as a parent to fulfil the role in the way in which they hoped for as long as possible and also based on the perceived needs of the child to be cared for in the way that the parent wanted. Decision-making about pregnancy was considered with reference to mothers with HIV, who identified fears of transmitting HIV, passing on the stigma associated with HIV, leaving young children without a parent or being unable to care for children as expected (Hebling & Hardy, 2007; Ingram & Hutchinson, 1999; Nelms, 2005; Wilson, 2007). Most parents in this study explained that they ensured that their partners were not CF carriers before deciding to have children, therefore transmitting CF was not discussed in depth by these participants but had been an important consideration.

Again, consistent with Simcox *et al.* (2009), it was acknowledged by two participants that they broached decision-making about parenting with their partners earlier than they anticipated in the context of completing predictable developmental tasks associated with life trajectory (Rolland, 1994). This experience could be understood as an acceptance of the life limiting nature of CF which enabled participants to make cognitive adjustments (Morling & Evered, 2006) to their beliefs regarding their developmental trajectory in comparison to their healthy peers. This enabled participants to fulfil their goal of becoming parents, but having taken this step, acceptance varied between participants and could be considered a process; participants were at different stages of this process.

4.2.2 It sharpened my focus

The master theme *It sharpened my focus* is in accordance with previous research exploring the experiences of mothers with a CID which suggested that some parents express a motivation

to keep well which was derived from their desire to live for the sake of their children (Hebling & Hardy, 2007; Ingram & Hutchinson, 1999, Wilson, 2007; Vallido *et al.,* 2010). In the current study, this concept was captured as a new perspective on keeping well, which was enacted differently between participants and is explored in more detail within the master theme *I've got to prioritise.*

An interesting finding was related to the different perspective held by fathers. Some fathers expressed a motivation to keep well for the whole family, which included their children and partners. This was in contrast to the mothers in this study who constructed their motives in relation to their relationship between themselves and their child. This discrepancy could be understood from a social constructionist perspective; fathers may be drawing upon the dominant narratives in society which may locate fathers as 'providers' for the whole family. In addition, although fathers in this study were fully participant in child care, they did not appear to regard themselves as the principal care giver for their child(ren) which may have allowed them to occupy a space in their family as supporter and provider for their partner who has taken on the role as main care giver. The mothers positioning will be explored below.

4.2.3 I've got to prioritise

The master theme *I've got to prioritise* is a reflection of the parents' experiences of living with the two roles of being a person with CF and a parent. This theme refers directly to concept of *staying well to parent*. In summary, mothers with younger children expressed a motivation to stay well but described more experiences of prioritising their children's needs first which for some parents resulted in powerful feelings of anxiety and fear. These participants may have been acting in accordance with the dominant discourse surrounding motherhood which is to fulfil the role with complete availability, responsibility and without selfishness (Thorne, 1990). This narrative may be even more powerful and conflictual when a mother is living with a threat to this identity (Vallido *et al.*, 2010) through a CID such as CF. In addition, threat to identity as a 'good mother' is thought to be even more prevailing when living with a life limiting condition (Wilson, 2007) and was shown in the study (by Radtke and Mens-Verhulst, 2001) to result in making an extreme case for investing in the mothering role and behaving in accordance with this. In addition, these mothers may have invested heavily in becoming parents and for some participants this choice was not always supported by wider

social networks. Therefore, the desire to be and be seen as a 'good mother' may be heavily prioritised. This could be one factor which contributes to the gender differences identified in the prognosis of people with CF (Cystic Fibrosis Trust, 2013a; Gage, 2012).

However, this explanation does not account for the finding that mothers with older children described many more experiences in which they were able to prioritise their health needs. This difference may be due to the needs of children at different developmental stages. Older children are less dependent, they may have needs which are more predictable and adolescents are in a developmental position to think about the needs of another person (Herbert, 1998). The older children in this study seemed to be encouraging and supporting their mothers in putting their own needs first. This difference seemed to allow these mothers to maintain their identity as a 'good mother' whilst also prioritising CF treatment. However, this is not to suggest that this was an adjustment without challenges.

It could also be hypothesised that the mothers with older children are a subsample that are healthier as they have survived pregnancy and parenting younger children. Being healthier could have made it easier for these parents to prioritise their health needs. However, this hypothesis is not supported in this sample as participants with older children did not have a better lung function indicating that it is factors relating the child's needs which is important in understanding the theme *I've got to prioritise*. However, it needs to be acknowledged that no direct comparisons can be made between the health status of these participants when they had young children and those currently raising young children, to enable any firm conclusions to be made.

There is a lack of available evidence concerning fathers with CF or a CID. Fathers in this study disclosed that they prioritised adherence to CF treatment and suggested that this resulted in them sacrificing spending time with, or being separated from, their child(ren). Some fathers acknowledged that this placed a burden on their partners and that they felt guilty as a result. As described in the theme *It sharpened my focus* fathers were not positioning themselves as the principal care giver, therefore for these participants their identity as 'good fathers' may not have entailed always putting their children's needs first. However, it appeared that adherence to CF treatment may have disrupted their role as a supporter and provider for the family. These fathers expressed feelings of sadness and anger at not being a 'normal' father. This finding is in keeping with evidence that people with CF might feel

'different' only in specific conditions when their lived experience does not meet an anticipated trajectory or biography (Williams *et al.,* 2009).

Crudely these findings suggest an interaction between time, gender and CF on parenting roles. That is, participants were accepting some difference in their experience of time as a person with CF; and as a result males were making cognitive and emotional adjustments in their role as a father and females were doing the same in their position as a person with CF.

4.3 BEING A PARENT WITH UNEXPECTED TIME

The superordinate theme *being a parent with unexpected time* may be understood as the participants' experience of projecting themselves into the past and using this information to make sense of themselves in the 'present' (Livneh, 2013) as parents with CF. Three master themes are contained within this superordinate theme and include: *It was such euphoria!; I had accomplished something;* and *I never think that I've got CF*. The participants' 'past' perspective (Zimbardo & Boyd, 1999) may be comprised of internalised expectations relating to a shortened life expectancy, hopes for meeting developmental milestones (such as having children) and concerns regarding fertility. Thus, these participants may be living in a time that they were not expected to have, and with children that they had not thought were possible. This master theme could be understood as the participants' experience of *parenting to stay well*. These parents may be making adjustments to their sense of self (Morling & Evered, 2006) as a person with CF as they assimilate who they are now with who they had been expected to be. In the majority of interviews this was infused with positive emotions.

4.3.1 It was such euphoria!

It was such euphoria! was a theme emergent in the accounts of many participants whose expectations of their lives had been exceeded by becoming parents. This resulted in a feeling of joy described by many as a sense of euphoria. This finding was consistent with those of a study of mothers with HIV who expressed an overwhelming sense of elation in their role as mothers, beyond that suggested to be experienced by mothers without a diagnosis of HIV (Hebling & Hardy, 2007). It is, however, acknowledged that a sense of euphoria is difficult to quantify and therefore it is hard to compare this feeling amongst different people.

Euphoria was also a result of being able to conceive children after assisted fertility or for some, after a long and difficult period of waiting for this treatment. Conversely, the feelings of euphoria emphasised the distress that some parents felt when discovering they needed to conceive children via assisted fertility treatment and the challenges they endured in undergoing this. In the current study, two fathers were diagnosed with CF as a result of fertility issues and therefore simultaneously had to assimilate the diagnosis of CF and infertility. Perhaps unsurprisingly, these participants expressed particular distress at not being able to conceive naturally. However, distress permeated the accounts of all participants who sought infertility treatment. This supports the findings of Fair *et al.* (2000) who identified that males with CF reported feelings of shock, anger and bewilderment when learning of issues of infertility. This highlights the priority that people with CF may ascribe to issues of infertility in the context of the many other challenges that CF might present.

4.3.2 I had accomplished something

The master themes *It was such euphoria!* and *I had accomplished something* is a reflection of the discrepancy between the evolution of CF management, which has moved quickly in these participants' lives, and the expectations that these parents may have internalised and transmitted since their childhood. It highlights a challenge that these parents faced in living with a condition which has increasingly offered greater hope for meeting developmental milestones, whilst also managing the reality of CF as a life limiting condition. Some participants expressed guilt and the feeling of being perceived as selfish for having children with CF. Anna and Nina explained that they had been dissuaded from having children or barriers were put in the way of assisted fertility treatment:

We are going to meet a young lady in a moment, she has got some fanciful idea that she wants to have children [conversation between Doctor and medical students overheard by Anna]…I just said but wait a minute this is the reason I am coming here is to ask your advice, I'm not some fanciful young lady…so that was, as I say, the start of the hurdles basically. Anna, p.1, 18-26.

I was never going to accomplish anything honestly, I didn't even my Paediatrician was you know don't get too much of a high expectations because I wanted to be a writer I wanted to be an English teacher, I was told, told don't go to high, don't you know exceed yourself and then obviously I didn't tell them I was trying for a baby because I knew they would have literally have said no. Nina, p.9, 287-291.

These findings may be understood with reference to the social model of disability as highlighted in the experiences of parents with a physical disability (Duvdevany *et al.,* 2008; Grue & Laerum, 2002) which might indicate that people with CF are subject to discrimination, making it more difficult for them to become parents. However, it may also be a reflection of the medical knowledge at that time which highlighted the risks of pregnancy for women with CF and the very limited life expectancy of people born in the 1960s and 1970s (Littlewood, 2012). This understanding is consistent with the finding that these experiences was more apparent in the accounts of the 'older' participants' and may reflect how the development of knowledge about CF management has changed. It is noted that in comparison to the daily experiences of discrimination and stigma that were described by parents with physical disabilities (Duvdevany *et al.,* 2008; Grue & Laerum, 2002) or HIV (Ingram & Hutchinson, 1999; Nelms, 2005; Wilson, 2007) this was less apparent for people with CF. This may be because unlike CF, the transmission of HIV is characterised by stigma and because CF is largely an invisible condition (Ravert & Crowell, 2008).

4.3.3 I never think that I've got CF

In contrast to the stark reality of CF that is present in many of the participants' experiences, is the master theme: *I never think that I've got CF*. This may be understood within the literature relating to the development of identity in people with CF (Badlan, 2006; Williams *et al.,* 2009) and to theories of temporal adaptation (Livneh, 2013). As described by Williams *et al.* (2009) people who are diagnosed with CF as children may develop a self-referential version of normalcy in which their past experiences with CF are reflected upon in relation to the present day and no differences are found. As expressed by some participants, a life without CF was not known and similarly being a parent without CF was not known. This is in keeping with the finding suggesting that CF is just one characterisation of the self (Ravert & Crowell, 2008) and threats to 'normality' may result in separating the CF aspect of the self from the whole self as a way of preventing CF being the defining characteristic of the person (Williams *et al.,* 2009) or parent.

This may reflect a gradual adjustment to living and being a person with CF. Similarly, when the participants' health remains usual to them (Badlan, 2006) they may experience a feeling of *Being the lucky one with CF* as they are able to fulfil their parental role with treatment as usual and may not experience a threat to their sense of 'normality' as a parent (Williams *et al.,*

2009). This experience of CF as usual could also be understood within the concept of secondary control (Morling & Evered, 2006); participants may accept the differences that CF brings and make cognitive adjustments to their sense of self, which enable them to experience life as usual. However, this is more difficult when a person experiences an acute deterioration in their health (Britto *et al.*, 2002; Goldbeck *et al.*, 2007) and adjustment becomes more challenging (Pfeffer *et al.*, 2003) which in the current study, was an experience reflected in some accounts.

Acceptance and adjustment in the context of parenting with CF appeared to be more challenging for participants who were diagnosed as adults, regardless of health status (estimated by predicted lung function) in comparison to other participants in the sample. These participants knew a life without CF and have not had the opportunity to adjust to incremental deteriorations in health and the limitations that this might bring over their lifespan. These participants appeared to engage more frequently in avoiding, hoping and wishing that CF was not part of their life as a parent. It is suggested that avoidance of the realities of CF may at times be adaptive (Abbott, 2003) as also demonstrated with other conditions such as cancer (Brenan, 2004). As reflected in these participants' experiences, different approaches to coping may be used throughout a person's lifespan which may include both primary and secondary control (Morling & Evered, 2006).

4.4 BEING A PARENT WITH UNCERTAIN TIME

The superordinate theme *Being a parent with uncertain time* refers to how participants manage the uncertainty of CF and the time ahead, which may be understood as a 'thrust' towards a 'future time' perspective (Livneh, 2013) and which has emerged as a consequence of becoming a parent. Three master themes were captured in this superordinate theme and included: *I want to see my children's children; I look at myself and into the future;* and *Parenting with uncertainty*. In contrast to previous research suggesting the development of a truncated future orientation to protect against death anxiety in people with a SCI (Martz, 2004) this superordinate theme reflected participants' orientation towards the future. This difference may be the result of existing research which has focused on a CID as something which is acquired during adulthood (Livneh, 2013); for the majority of the participants in the current study, CF was diagnosed in childhood. As discussed, this may have enabled participants to

make adjustments in accordance with the life limiting consequences of CF over their life span and development to date.

However, maintaining a future perspective may be more manageable when CF has not 'caught up' with the person and health remains stable; for these participants they were able to make cognitive and emotional adjustments (Morling & Evered, 2006) to maintain a future perspective that enabled greater opportunities for planning future actions (Zimbardo & Boyd, 2008). As discussed previously, adjustment was more challenging when participants experienced a significant deterioration or change in health. However, for all participants holding a future perspective was not without emotional challenges and the master themes reflect feelings of anxiety, guilt, sadness and despair.

4.4.1 I want to see my children's children

In accordance with previous literature on parenting with a life limiting condition, participants' described the wish to see their child reach a certain age such as 18, which signified that they had reached adulthood (Hebling & Hardy, 2007; Ingram & Hutchinson, 1999). This was reflected in the theme *I want to see my children's children.* The target age that participants hoped to reach was based on concrete factors such as the level of education they would like their child to obtain or a developmental milestone such as their child having their own children. Although these parents were not able to exert complete control over their health, creating a target age seemed to give participants some sense of perceived control to manage feelings of anxiety and guilt regarding the possibility of leaving their child without one parent.

The current study did not support the finding by Wilson (2007) that parents experienced an uncertainty about their existence once their child had reached this age. Two participants had seen their child reach their target age; and they continued to express a strong desire to keep well in the hope of more time with their child(ren). In contrast to the findings by Wilson (2007) one participant also expressed a feeling of satisfaction that she had achieved in life by reaching this target age. This participant seemed to enjoy a greater sense of self-esteem and pride as a result of keeping herself well and seeing her child reach this target age.

The discrepancy between this study and the research by Wilson (2007) may be the result of the stigma experienced by the women diagnosed with HIV, which as identified, is less evident in the accounts of people with CF. Some mothers diagnosed with HIV and with physical disabilities were thought to have re-gained a sense of self-worth after becoming parents (Grue & Laerum, 2002; Wilson, 2007). Although this was evident in the accounts provided by parents with CF, these parents were also invested in other roles such as in employment, in relationships with partners or wider family; and seemed to have a greater sense of self-worth built on engagement in other roles, as well as the parental one. Therefore, the parents in the current study were able to identify reasons to keep well for themselves and their children beyond seeing their children reach a target age such as 18.

4.4.2 I look at myself and into the future

The master theme *I look at myself and into the future* suggests that for these participants becoming a parent had given them a different perspective on their future (Livneh & Martz, 2007) which resulted in a different understanding of the life limiting nature of CF. The way in which participants managed this perspective seemed to depend upon the perceived controllability of CF. When a situation was considered uncontrollable, such as when participants described efforts to manage CF as no longer having the same effect, they disclosed fewer thoughts indicative of cognitive or emotional adjustment and more of accepting the situation as being down to fate or luck. Acceptance of this future perspective is described in the emergent theme *Making the most of time* which is explored below. In situations which were deemed more controllable, participants engaged in adjustments to accommodate changes to health. Emergent in the participants' accounts were examples of moving between these strategies for coping. Such oscillation might be expected throughout a person's lifespan (Livneh, 2013) and may be important when managing uncertainty (Abbott, 2003).

4.4.3 Parenting with uncertainty

The third master theme reflected participants' experiences of noticing the impact of the uncertainty of CF on their children, the way in which they managed parenting in this context and the feelings associated with these experiences. This was a theme found in the literature exploring parenting with a CID (Hebling & Hardy, 2007; Ingram & Hutchinson, 1999; Nelms, 2005; Thorne, 1990; Vallido *et al.,* 2010; Wilson, 2007). The emergent theme *Parenting*

differently resonates with the challenges identified in parents diagnosed with a CID who feared 'passing on' anxiety to their children or damaging their childhood through exposure to illness or disability (Grue & Laerum, 2002; Nelms, 2005; Thorne, 1990; Wilson, 2007). However, unlike the findings of parents with a CID (Grue & Laerum, 2002) and consistent with Simcox *et al.* (2009) parents with CF did not express fear in asking their children for help. Most participants did not describe experiences in which this occurred and for those that did, this was viewed positively. This may be a reflection of the differences in attitudes relating to people with CF, so contrary to the findings of Grue and Laerum (2002); help was viewed within a discourse of socialisation as opposed to chronic illness.

The emergent theme *Making the most of time* was a reflection of the belief that due to parenting with uncertainty, these parents treasured and immersed themselves in the parental role. This was also described in mothers with HIV (Ingram & Hutchinson, 1999) and is illustrated by the following quote: 'HIV helped me put things in a positive form, a forgiving way. I learned to love by having this disease. I'm still learning from it. In that way, I'm a better mom and better for my kids' (p.253). This understanding is influenced by models of acceptance (Smith & Hayes, 2005; Harris, 2009) in which participants may have been willing to accept circumstances and adjust their perception of parental experiences (Morling & Evered, 2006) in accordance with these values. Described another way, participants appeared willing to increase action in the pursuit of living life towards a valued goal (Smith & Hayes, 2005) which encompassed investment in the parental role.

In summary, the findings suggest that parents engage in *staying well to parent and parenting to stay well.* Themes emerged which highlighted the significance of time in parenting with CF which resulted in the emergence of three superordinate themes based on the concept of time. Therefore, parenting with CF was conceptualised using the theory of time perspective proposed by Zimbardo & Boyd (1999) and developed by Livneh (2013). Temporal adaptation was explored in relation to the theory of secondary control (Morling & Evered, 2006). Factors which influenced secondary control for these participants were identified. These included: exacerbations in health; age and gender of the parent; and age of the child.

As described, the evidence base relating to parenting with a CID had limited applicability when understanding the psychological experiences of parents with CF. Therefore, alternative theoretical frameworks were utilised. To the researcher's knowledge this was the first study

to apply the theory of time perspective (Livneh, 2013; Zimbardo & Boyd, 1999) in understanding the experiences of people with CF. It extended the application of this model to a population in which a diagnosis is usually received in childhood. It also allowed connections to be made between coping and time perception, and generated a number of hypotheses which might be the subject of further research. Findings in the current study were most closely related to the parenting experiences of mothers with HIV, due to the life limiting nature of the condition. However, the concept of stigma which characterises many of these studies was not considered as relevant in the experiences of parents with CF.

A number of findings that might have been anticipated were not emergent in this research. These included interactions with healthcare professionals; the impact of partners and wider family on the parental role; and interactions between parenting and other roles such as employment. Whilst some interviews contained allusion to these issues, the current findings were constructed to provide the most meaningful representation of all of the participants' experiences. The inclusion and interpretation of findings was also motivated by clinical applicability and relevance.

4.5 CLINICAL & SERVICE IMPLICATIONS

The principal clinical implication of this research is to increase understanding of the needs and experiences of parents with CF. Such understanding may inform service delivery and, consequently, improve the quality of life of people with CF. Although the role of clinical psychology is recognised in adult CF services (Cystic Fibrosis Trust, 2011), it is hoped that this research will raise awareness and therefore the priority given to the role of psychological factors when developing services for people with CF and more specifically for those who are also parents.

In simple terms, these results suggest that parenting with CF is influenced by past expectations and future hopes and fears. The findings indicate that for these participants, parenting serves as a motivator to keep well and the parental role provides cognitive and emotional benefits when living with CF. Exceptions and factors influencing this experience will be described in more detail below. This research did not employ a methodology which would claim to produce findings that are representative of the CF population, so conclusions

about clinical and service implications need to be advanced with caution. However, at present there is no evidence base from which to draw when working with parents or prospective parents and no evidence-based information for service users to utilise when making the decision to become parents so this research provides a useful starting point for understanding in this area.

4.5.1 Clinical Implications for CF Services

One of the central clinical aims of this research was to gain insight into the challenges faced by parents who may be experiencing difficulties managing parental responsibilities and coping with the symptoms and treatment associated with CF. These findings indicate that there are particular challenging points in the parents' illness trajectory and the child's development which could impact on ability to *stay well to parent* and on the opportunity to experience the rewards of parenting. This knowledge could be used to target scarce resources systematically at appropriate points in the life of a parent with CF.

As identified by Fair *et al.* (2000) issues of infertility were distressing for the participants in the current study. In this sample, discovering infertility was most challenging for those people diagnosed with CF as adults and highlighted the challenge of accepting infertility in the context of also assimilating a diagnosis of CF. As shown in research with the general population, experiencing assisted infertility can be distressing (Chachamovich, *et al.,* 2007). However in the context of a life limiting condition when time may be perceived differently, this could exacerbate the potential for distress.

Previous findings suggest that conversations regarding issues of fertility should take place earlier than in current practice (Fair *et al.*, 2000; Havermans *et al.*, 2011; Sawyer *et al.*, 2005). This is supported by these findings; people with CF may want to think about having children earlier than anticipated in the context of a lifespan without CF. However, some people may find it difficult to initiate these discussions; previous research suggests women with CF were more likely than men to broach issues of fertility and reproduction (Havermans *et al.*, 2011) but some people with CF may avoid consulting with professionals through fear of being advised against pregnancy (Conway *et al.,* 1994). Given the distress reported by these participants who had children, it could also be hypothesised that people with CF who are

unsuccessful in their attempt to conceive may be a particularly vulnerable group, although this requires further exploration through research.

This study suggests that becoming a parent may result in a new perspective on life trajectory and a changing sense of self as a person with CF. This new perspective may serve as a motivator for parents to keep themselves well but also potentially increased anxiety and fear about the future. Parents who have made the decision to have children earlier than previously anticipated may be more vulnerable and may require space to adjust to this new role. An acute deterioration in health may also mark a particularly difficult transition point for parents who are now considering their life trajectory in the context of their responsibilities as a parent. Additionally, parents with younger children are more likely to come into contact with infections from their children. Such information, and advice on how to protect themselves, could be useful for prospective parents when anticipating this role and when parenting infants and young children.

Parents with children in the middle age group may face dilemmas such as how to talk about CF and more specifically about its consequences and treatment implications. The results of this study indicated that parents with adolescent children may experience challenges associated with feeling different as a parent and may benefit, for instance, from discussion about managing treatment in the presence of children's peers. In addition, these parents may be faced with the dilemma of what to share with children about CF and how much to involve them with their care. There is a lack of evidence relating to the experience of children with parents with CF but the current findings suggest that some children experienced anxiety and fear, particularly associated with the life limiting and uncertain nature of their parent's condition. In addition, in the event of parental death, evidence suggests that children of all ages will experience a bereavement reaction with some children going on to experience longer term psychological and emotional difficulties (Dowdney, 2000). Therefore, systemic interventions targeted at the needs of children may be beneficial to families.

The differences in the experiences described by fathers and mothers were a feature of these findings; fathers may more frequently put their own health needs first and construct their experiences in relation to the whole family. Mothers with young children in this sample described more isolated care giving experiences, in which their needs often came last when

faced with a situation that involved separation from their child and/or time consuming treatment. In keeping with previous research suggesting that mothers with mental health problems may hide worsening symptoms if they consider that they will be asked to go into hospital (Davis & Allen, 2007), some mothers with younger children in this sample explained that they may avoid appointments if they believe that this might result in a hospital admission. For these mothers, this choice was not based on a lack of knowledge about treatment and resulted in powerful feelings of distress and anxiety. Therefore, some mothers may benefit from the option of treatment at home as an alternative to hospital admission. However, these findings indicate that mothers may feel able to put their own health needs first as their children get older.

The experiences of fathers in this study indicates that they may benefit from psychological interventions targeted at exploring the loss, sadness and adjustment to thoughts of feeling different as a father with CF. Almost all fathers will have conceived children through assisted fertility and interventions might usefully be targeted during or following this process. In addition, some fathers indicated that their partners might benefit from support as they were likely to have undergone fertility treatment, to have taken on a greater caring role and to be preparing, psychological and practically, for being a single parent in the event of their partner's death. The importance of supporting family and carers has been recognised when planning services for people with long term conditions (Fellow-Smith, 2012).

A proactive approach to service delivery which anticipated the transitions outlined would give parents the opportunity to make informed choices and to begin making practical and psychological adjustments when physically healthier. This may also allow professionals to work collaboratively through these transitions with people with CF, whilst maintaining emotional resilience and rapport. However, it should be acknowledged that parents may find it difficult to initiate conversations with professionals regarding parenting experiences, particularly if challenges are encountered. As indicated by these findings, parents and mothers in particular may feel they that they need to work hard to protect their identity as a good parent and may fear creating suspicion that they are not able to manage parental responsibilities or that they will be advised against having further children. Discussing issues relating to mortality may also be challenging for parents to initiate with professionals who have invested in their care over a number of years. However this research suggests that some parents may want to express their fears of declining health or death in the context of being a

parent and gain an understanding of how best to make plans for their children having recognised their own uncertain future.

This research also indicates that the experience of parents and prospective parents needs to be understood in the context of the medical understanding of CF during the parent's childhood and development. Information relating to CF which was internalised in childhood may have a significant impact on confidence and self-esteem. CF treatment and medical management strategies to promote fertility and support pregnancy and parenting may have also have evolved so that their knowledge of current treatments is now outdated. A coping strategy used to manage CF is denial and avoidance (Abbott, 2003) and some parents in this study actively sought to avoid learning about developments in CF to protect themselves from becoming hopeful about accessing treatments which were not available to, or suitable for them. Therefore, prospective parents may be making decisions based on out-dated expectations and knowledge and may be fearful of engaging health professionals in pro-actively updating their understanding.

It is hoped that the findings and the implications described above will improve the support offered to parents with CF from clinical psychologists and the wider health care team. People with CF have expressed a desire for information relating to the experiences of others with regard to infertility and reproduction (Fair *et al.,* 2000); therefore it is also important that these findings are disseminated to people with CF who might benefit from this information.

4.5.2 Implications for Clinical Psychologists

Clinical psychologists could take a lead role in training other professionals in the psychological implications of parenting with CF. This is important given that many people with CF will not be routinely reviewed by a clinical psychologist. The factors highlighted above could usefully be considered when developing psychological formulations. One approach to sharing psychological knowledge in teams is through the development of shared formulations (British Psychological Society, 2011; Jackman, 2013) which could be used by clinical psychologists in CF multi-disciplinary teams.

There is little available evidence for the application of psychological theory when working with adults with CF (Oxley & Webb, 2005). Some research has explored the use of cognitive

and behavioural (CBT) interventions for working with this population, but this has produced mixed findings (Glasscoe & Quittner, 2009; Hains *et al.,* 2001; Oxley & Webb, 2005). However, one study was based on a very small sample size (Hains *et al.,* 2001) and another was restricted to children with CF (Glasscoe & Quittner, 2009). Therefore, further research is needed before any conclusions can be drawn. CBT is recommended for adults with chronic health problems experiencing depression (NICE, 2009) and may be useful approach to enable parents with CF to manage past negative feelings or cognitions and move toward a future perspective (Livneh, 2013). This model also enables physical factors associated with CF to be incorporated into the formulation and intervention plan.

Acceptance and commitment therapy (ACT) is an alternative approach which could be applied when working with parents with CF. There is no research exploring the use of ACT within the CF population but this approach has been shown to be effective for people living with other physical health conditions such as low mood in chronic pain (Veehof *et al.,* 2011) and managing the distress of end stage cancer (Branstetter *et al.,* 2004). It is suggested that in situations in which an alternative evidence base to ACT does not exist or there is a good reason not to use existing evidence-based practice, ACT could be applied to any problem that fits the underlying model (Hayes, 2008). This approach could be applied to increase acceptance of difficult thoughts and feelings related to being a parent with CF and to enable parents to in live accordance with important values in the context of managing the demands of CF and parenting.

4.6 STRENGTHS AND LIMITATIONS OF THE STUDY

The aim of this research was to explore the lived experiences of parents with CF in relation to *staying well to parent and parenting to stay well.* A review of the relevant literature, presented in chapter one, highlighted the lack of research in this area and in parenting with a CID more broadly. Therefore, the current study sought to fill this gap by pursuing a qualitative analysis of the experiences of a sample of parents with CF. As discussed in chapters one and two there are various ways in which qualitative methodology can be assessed for quality. The criteria proposed by Elliott *et al.* (1999) was outlined in chapter two and considered throughout the research process. Therefore, these guidelines (Elliott *et al.,* 1999) and the

CASP (2010) criteria will be considered when assessing the methodological strengths and limitations of the current study.

4.6.1 Methodology & Design

A qualitative approach and more specifically the use of IPA was a method deemed compatible with the objective of capturing the complexities of parenting with CF. The use of IPA was considered an appropriate method to begin exploration in a new area and enabled the emergence of themes which had not been considered in existing literature or considered previously by the researcher. This method allowed the researcher to remain close to the participants' own words and to construct themes and illustrate them using rich, nuanced quotes. Providing quotes is considered important to allow others to make sense of the data, generate new ideas (Elliott *et al.*, 1999) and to demonstrate transparency of the research process (Meyrick, 2006; Yardley, 2000). However, the interpretive element of IPA also enabled the participants' disclosures to be analysed for their meaning in relation to relevant psychological theory and clinical practice.

A limitation acknowledged with the use of IPA is a reliance on the participant's ability to reflect on their thoughts, feelings, relationships, emotional and physical lives rather than simply offer their opinions about a phenomenon (Willig, 2008) which could skew the sample towards more articulate and educated participants. In this study, participants offered in-depth reflections regarding their experiences as a parent with CF, which enabled the development of broad, overarching superordinate themes but also sufficient detail to identify discrepancies and subtleties between participants. This fulfilled the aim outlined in chapter two of accomplishing the specific research task (Elliott *et al.,* 1999) of gaining an in-depth exploration of the experiences of one sample of participants from this population. However, it is acknowledged that there may be a bias in the people who opted to participate. This is explored in more detail below.

The role of language and more specifically how participants presented meaning through language was considered in the analysis of data. However, as an alternative a discursive approach may have allowed further exploration of how participants' constructed their experience in that particular moment in time and for the purposes of the interviews. This

would have enabled a greater incorporation of the context in understanding participants' meanings which is privileged less in IPA.

4.6.2 Recruitment & Sample

Participants were selected purposively and the responses rates were recorded. However, to minimise any pressure to participate, the reasons for non-participation were not sought. The recruitment process ensured that participants were invited to take part by random selection; however those who came forward and volunteered may have represented a biased group. Participants choosing to take part may have held particular positions in relation to the topic which motivated them to participate, possibly reflecting a bias in the sample. One such bias may be towards parents who consider they are coping well with this role or who are experiencing distress and seeking emotional containment. However, this can only be surmised.

A particular strength of this study was the inclusion of fathers with CF which are an under-researched group in the literature relating to parenting with a CID. The balance between the heterogeneity and homogeneity of the sample was a further strength of this study. The sample was considered sufficiently homogeneous to enable meaningful comparisons to be made between participants. However, the diversity of the sample allowed an understanding of the differences in needs and priorities of parents at different developmental stages and with different degrees of severity of CF. Given this diversity, it could be argued that the data reflected the rich experiences of parents with CF.

The sample size was also considered sufficient to enable an in-depth exploration of this area whilst maintaining a manageable data set (Smith *et al.,* 1999) but one which displayed the richness of the individual cases. However, a limitation regarding the sample was the exclusion of parents who were not primary care givers. This is not representative of the diverse family structures in the UK. However, only one potential participant was excluded on this basis.

Furthermore, Elliott *et al.* (1999) highlights the importance of situating the sample, this was carried out through a variety of means. However, a limitation of this study was the restricted information provided about each participant in relation to their quotes. This may have

limited an understanding of the context of the participants' quotes. However, anonymity needed to be prioritised.

4.6.3 Data Collection & Analysis

The use of semi-structured interviews allowed participants to provide an in-depth description of their experiences which generated rich data. The diversity in participants' accounts was a reflection of the flexibility that the interview questions gave participants to take their responses in their own direction (Smith *et al.,* 2009). The interviews took place either in the participants' homes or the CF clinic. Perhaps unsurprisingly the participants' accounts seemed to vary with context and the content of interviews leaned towards the priorities of the particular context in which they were held perhaps because of the prompts in that environment. Neither location was deemed more appropriate but this inconsistency may be considered a weakness of this study.

A limitation regarding the use of interview transcripts was the loss of non-verbal communication and the subtleties in expression which were not included in the interview transcripts. Perhaps due to the emotional and challenging nature of some of the participants' reflections, incongruence between presentation of content and the words transcribed was often noted by the researcher when listening and recalling the interviews. However, an in-depth analysis of the meaning of participants' words would have gone some way to bridging the gap between what was spoken and an interpretation of what was meant.

The questionnaire measures used in this study provided a useful means of situating the sample and provided interesting insights into the subjective experiences of participants in relation to their cognitive, emotional and physical experiences of CF. However, it may have been useful to include an assessment of time perception, given the significance of this concept in interpreting the results. The Zimbardo Time Perspective Inventory (Zimbardo and Boyd, 1999) is one such measure that could have been used.

4.6.4 Ensuring Credibility

Credibility of the research findings was considered throughout this study. This was ensured by owning ones perspective (Elliott *et al.,* 1999) by adopting a position of reflexivity during

the research process (Willig 2008). The researcher's position was considered in four ways. The first was the statement of the researcher's position in chapter two; the second was the use of the researcher's reflective diary (Elliott *et al.*, 1999; Yardley, 2000), the third was reflection with the researcher's academic and clinical supervisors throughout the process. These processes assisted the researcher in appreciating changing assumptions and beliefs relating to this topic and enabled the reader to consider the lens from which this research was being interpreted. The fourth approach was the bracketing interview (Rolls & Relf, 2006) which supported the researcher's aim to get 'experience close' during the data analysis by reflecting on own biases and assumptions during this process.

Triangulation of the data analysis with the researcher's clinical supervisors and a qualitative special interest group was adopted to ensure credibility of the themes generated (Elliott *et al.*, 1999) by obtaining multiple perspectives on the analysis. However, research findings were not discussed with the participants who took part in the study or other parents with CF which could be considered a methodological weakness in relation to credibility. This was not carried out due to ethical constraints regarding approaching participants on more than one occasion and due to concerns about the sensitive nature of the data and the challenges of discussing the themes with potential participants who did not take part and have no prior rapport or relationship with the researcher.

4.7 IMPLICATIONS FOR FUTURE RESEARCH

This study is the first to offer insights into the experiences of parents with CF, therefore a number of recommendations for further research in this area are highlighted. This research was conducted with a sample of parents from South Wales and interpreted from the perspective of a female without CF or parenting experience. This study could therefore be replicated with other samples and a different researcher to develop new insights and gain an understanding of the applicability of these findings to other people with CF. The themes generated in this research could be used as a basis for further exploration.

Although it was not intended, all participants recruited in this study planned their pregnancies. Obviously not all pregnancies are planned (Simcox *et al.*, 2009) and it would perhaps be beneficial to develop an understanding of the experiences of parents who did not

plan to become parents and to explore how these parents adjust. As indicated, an understanding of people with CF who could not conceive children is also suggested given the distress experienced by those participants who undertook successful fertility treatment. A third group who were not considered in this study were parents who had had a lung transplant. A transplant may provide an opportunity for parenting but is also considered a 'high risk pregnancy' (Edenborough *et al.,* 2008). Therefore, parenting in this context may result in different parenting experiences and call for different adjustment strategies because although a transplant may be transformative it also signals that the condition has entered its end stage and implies limited life expectancy (NICE, 2006, Sands *et al.,* 2011).

This research focused on understanding the perspectives of parents. As indicated by this research, children, partners and CF professionals take different positions in relation this topic. An examination of their perspectives would enhance understanding of the family and professional system around the parent with CF and potentially enable the development of services which could meet the needs of the whole system.

As described in chapter one, the evidence base relating to parents with a CID is limited when attempting to understand the psychological processes which enable parents with a CID to continuing functioning and draw benefit from their parenting roles. The qualitative evidence base rests upon a small number of studies of variable quality which are largely understood from a sociological perspective. Only small references are made to the individual psychological experiences such as the cognitive processes some parents adopt to manage the uncertainty of a life-limiting condition. In addition, the systematic review only contained one study which was conducted in the UK (Wilson, 2007). There are many factors which make comparisons between research generated in different countries problematic, such as differing experiences of poverty, health care systems and cultural expectations of parents and people with a CID. Therefore, further research in this area, using a sample of parents living in the UK in needed.

This evidence base also had limited application to understanding the unique challenges associated with parenting with CF which include factors such as an early onset, unpredictable trajectory, infertility, limited life expectancy, time consuming treatment and rapidly changing medical advances. These psychological challenges pose different threats to identity as a parent and person with CF and present unique experiences when living as a parent with CF.

Therefore, alternative theoretical frameworks were used to interpret these findings which have not been previously explored in a CF population. Further research could usefully extend the application of these theoretical models to understanding the psychological experiences of parents with CF and the wider CF population.

As described, empirical data investigating time perspective (Zimbardo & Boyd, 1999) and psychosocial adaptation to a CID is limited (Livneh, 2013). The psychological processes underpinning the development of a particular psychological time perspective in the context of a CID is not well evidenced, further longitudinal research could usefully fill this gap. The association between time perspective and particular emotional experiences such as anxiety and low mood is more widely researched (Livneh, 2013). As an example, the evidence indicates that feelings of hopelessness associated with low mood points to a truncated or blocked future orientation and therefore a neglect of future goals (Livneh, 2013).

Therefore, particular challenges associated with CF such as infertility or an exacerbation in health could result in low mood and a truncated future perspective which might limit future goal planning and obtainment. Quantitative research could usefully be employed to examine the relationship between time perspectives and psychological challenges associated with CF. As identified, the evidence base for psychological interventions with people with CF is limited (Oxley & Webb, 2005). An exploration of psychological models such as CBT or ACT which may enable parents with CF to manage past negative feelings, cognitions and move toward a more adaptive future perspective may be of benefit (Livneh, 2013).

As described, the theory of secondary control (Morling & Evered, 2006) is under researched, particularly when applied to people with a CID. Further research is needed within the CF population to understand when secondary may be adaptive and if it fosters perceived control or has other benefits such as reduced helplessness and despair as described by Morling and Evered (2006). It could be hypothesised that particular challenges such as managing an exacerbation to health or a particular developmental stage in a child's life may warrant particular mechanisms of coping, which could be explored through further research. As identified, an association between coping and time perspective is hypothesised but under researched (Livneh, 2013). Quantitative research to investigate this association could be applied within the CF population to better understand the interaction between living with a

health condition which influences the perception of time and fulfilling the dual roles of parenting and living with CF.

4.8 CONCULSION

Due to improvements in treatment and understanding of CF, increasing numbers of people are becoming parents. Authors have speculated that parenting with CF may involve balancing the roles of being a person with CF with those of being a parent and devising strategies to optimise functioning in each role. Despite this, no research has explored this area to date. These findings suggest that being a parent with CF results in a different perspective, experience and awareness of time. As such, three superordinate themes were identified which are based on the experience of time and include: *Being a parent on compressed time; Being a parent on unexpected time; and Being a parent with uncertain time.* The first two superordinate themes refer to the experiences of parents in *staying well to parent and parenting to stay well.*

These findings were understood in relation to an interaction between Time Perspective (Zimbardo & Boyd, 1999) and coping, which was explored in relation to secondary control (Morling & Evered, 2006). Factors thought to influence secondary control were identified, which included: exacerbations in health; age and gender of the parent; and age of the child. The experiences of parents with CF were compared to parents diagnosed with a CID by exploring the evidence base, generated by a systematic review. The evidence base was limited in its application to a psychological understanding of these experiences and to the unique challenges experienced by people with CF. This research broadened this evidence base to include fathers and the unique experiences of people with CF.

It is anticipated that an increased understanding of the needs and experiences of parents with CF will be used to inform service delivery. An awareness of the needs of this group may increase the priority given to the role of psychological factors when developing services for people with CF and more specifically people with CF who become parents. The research indicated various transition points in the participants' experience of parenting and chronicity of CF which highlight potential challenges for these parents and therefore targets for intervention. There is little available evidence for the application of psychological therapy

when working with adults with CF, therefore further research is needed but the use of CBT and ACT was discussed. Further research is needed to explore the experiences of parents who did not plan pregnancy, parents who have had a transplant and the perspectives of others in the wider system in which parents with CF are located.

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Key Words & Databases for Literature Review on Parenting with Cystic Fibrosis

<u>Keywords:</u>

Cystic Fibrosis

AND

History; epidemiology; diagnosis; symptoms; treatment; gender; adult services; infertility; knowledge AND infertility; pregnancy; parent*; mother*; father*; psychological; psychosocial; health related quality of life; anxiety; depression; body image; experiences; well-being; acceptance; coping; identity; identity AND development; mental health problems.

<u>Databases:</u>

PsychINFO; PsychArticles; Medline; Pubmed

Search Terms & Outcome of Systematic Review Search

Search 1 - Key Words:

Chronic illness

AND

Parent* OR mother* OR father*

AND

Identity OR Self OR Self-identity OR Self concept OR Self image OR Personal identity OR Personal characteristics OR Self reference OR Self perception OR Self evaluation OR Self talk OR Character OR Selfhood OR Personality OR Individuality

Table to Show Outcome of Search 1:

Database	Number of hits	Number of articles reviewed as full text	Excluded after reading full text	Reason for Exclusion	Number of duplicates	Total Included
ASSIA	76	1	1	-Practical parenting tasks	0	0
PsychINFO & PsychArticles	1108	9	7	- Parents with mental health problems - Evaluation of therapeutic intervention - Focus on family experience x2 - Mixed method x3	0	2 (Radtke & Mens-Verhulst, 2001; Wilson, 2007)
Pubmed	258	2	1	-Unpublished	0	1 (Mens-Verhulst *et al.*, 2004)
Medline	220	5	3	- Child's well-being - Pregnancy - Mixed method	2	0
Embase	197	7	4	- Mixed method x2 - Not parenting focus - Unpublished	2	1 (Thorne, 1990)
Scopus	5	0	0		0	0
Sociological abstracts	77	4	3	- Child's well being - Unpublished - Parenting experience not the focus	1	0
Web of knowledge (Searched 'title')	6	0	0		0	0
CINAHL	82	5	5	-Mixed method x3 - Not in English - Roles other than parenting	0	0
TOTAL	**2,029**	**33**	**24**		**5**	**4**

Search 2 – Key Words:

Chronic illness OR Chronic condition OR Physical Disorders OR Asthma OR Disability OR Kidney failure OR MS OR HIV OR AIDS OR COPD OR Sickle cell OR Diabetes OR SCI OR Cerebrovascular disease OR Arthritis OR Cystic Fibrosis OR Respiratory failure OR Digestive system disorders OR Lung disorders OR Metabolism disorders OR Congenital Disorders OR lung cancer OR brain injury OR head injury OR renal failure OR kidney disease OR dialysis OR cardiovascular disorders OR chronic pain OR osteoporosis OR epilepsy OR chronic hepatitis

AND

Parent* OR mother* OR father NOT Child* NOT Adolsecen*

AND

Qualitative OR IPA OR grounded theory

Table to Show Outcome of Search 2:

Database	Number of hits	Number of included articles reviewed as full text	Excluded after reading full text	Reason for Exclusion	Number of duplicates	Total included
ASSIA	88	0	0	-	0	0
PsychINFO	291	6	6	- Not parenting experience - Focus on pregnancy - Not parenting experience as the focus x3 - Unpublished	0	0
Pubmed	507	4	3	-Not parenting experience x2 -Focus on pregnancy & motherhood	1	0
Medline	264	4	3	-Focus on pregnancy & motherhood -Not parenting experiencex2	1	0
Embase	491	3	3	-Not parenting experience x2 - Sample of women who were pregnant	0	0
Scopus	38	0	0	-	0	0
Sociological abstracts	131	3	3	- Mixed sample - Unpublished - Parenting not the focus	0	0
Web of knowledge Searched 'title'	53	5	3	-Child with CID - Not parenting experience - Unpublished	1	1 Hebling & Hardy, 2007
PsychArticles	612	0	0	-	0	0
CINAHL	132	2	2	-Unpublished - Parenting not the focus	0	0
TOTAL	**2, 607**	**27**	**23**		**3**	**1**

Appendix 3

CRITICAL APPRAISAL SKILLS PROGRAMME
Making sense of evidence about clinical effectiveness

find → appraise → act

10 questions to help you make sense of qualitative research

These questions consider the following:

Are the results of the review valid?

What are the results?

Will the results help locally?

A number of italicised prompts are given after each question. These are designed to remind you why the question is important. There will not be time in the small groups to answer them all in detail!

Screening Questions

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

Consider:
- *What the goal of the research was*
- *Why is it important*
- *Its relevance*

2. Is a qualitative methodology appropriate?

Consider:
- *If the research seeks to interpret or illuminate the actions and/or subjective experiences of research participants*

Detailed questions

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

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

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

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

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

Consider:

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

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

Consider:

- *If the researcher critically examined their own role, potential bias and influence during:*
  - *Formulation of the research questions*
  - *Data collection, including sample recruitment and choice of location*
- *How the researcher responded to events during the study and whether they considered the implications of any changes in the research design*

7. Have ethical issues been taken into consideration?

Consider:

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

8. Was the data analysis sufficiently rigorous?

Consider:

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

9. Is there a clear statement of findings?

Consider:

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

10. How valuable is the research?

Consider:

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

Narrative Synthesis: List of themes from Included Studies

Duvdevany *et al.,* (2008).	• Confronting negative social attitudes toward parenting • Fear of rejection by his children proven false • Children's everyday familiarity with their father's disability
Grue & Laerum, (2002).	• Becoming a mother • A fragile motherhood? • Mothers little helper • Contradictions in keeping children safe
Hebling & Hardy (2007).	• The right to be a mother • Feelings about motherhood • Care of children in case of death • Not being able to breast feed • Mechanism of defence
Ingram & Hutchinson (1999)	• Defensive mothering • Preventing the spread of HIV & stigma • Preparing the children for a motherless future • Protecting self through thought control
Mens-Verhulst *et al.,* (2004)	• Non-medical prevention • Normalising • Mobilising support
Radtke & Mens-Verhulst (2001)	• Medical discourse • Being healthy / being sick • Invested in mothering • Orientating to the norm for modern women combining mothering & paid employment
Nelms (2005)	• Burden of diagnosis & health status • Burden of whether or not to reveal to children • The burden of unknown future of themselves & children • Relieving the burden of the diagnosis.
Thorne (1990)	• Performance issues • Availability issues • Dependency issues (on child) • Socialisation issues (of child) • Health care issues
Wilson (2007)	• Establishing and maintaining an identity as a good mother in the context of a stigmatised condition • Preserving an identity as a good mother in the context of a potentially fatal illness • Shielding children from the implications of a potentially fatal illness

Extracts from Reflective Diary

20/04/11
I feel really excited that there is an opportunity to do some research in the cystic fibrosis service, I had decided before today that I wanted to carry out a project in a physical health setting which would be related to child or family work. So this seems ideal, although my knowledge about CF extends to a documentary I watched recently about a teenage boy with CF.

27/01/12
I met with an expert patient advisor today to ask for her feedback on my interview questions. I was really aware of not wanting to ask questions which might extend her thinking about CF beyond what she was comfortable with, but I also needed to think in enough depth to make this meeting helpful. Some feedback that she gave me wasn't possible within the boundaries of IPA. I didn't want this meeting to be tokenistic but I also need to work within the methodology. I have decided that I need to take something different from this meeting; an opportunity to find out what CF is about from the point of view of someone living and working in it. It seems that motivation is a big factor. I am surprised at just how much treatment people need to do before leaving the house in the morning. CF all seemed very matter of fact too, just a part of everyday life.

29/06/12
I attended the ethics review meeting today. The panel wanted reassurance that I wouldn't ask participants about their life expectancy. I wasn't planning to, but it struck me just how much anxiety sounds the issue of death and the assumption that talking about would cause harm.

25/10/12
I conducted my first interview today. He didn't say what I thought he was going to say. CF didn't seem to play a big part in this life. I'm not sure if it is his reflections which didn't match my initial assumptions or if my questions aren't good enough. He laughed all the way through it but the things he said, didn't match up with this laughter.

19/11/12
The interviews are progressing. It feels very sad to listen to people my age talk about their fears of death; it's difficult to believe that they probably won't see their children grow up. I've also spent the week in critical care and the teenage cancer ward and I'm struck by the resilience in people but also how difficult it is to listen to all this distress and physical suffering. I sensed that participants initially wanted to protect me from listening to their fears of death; I wonder if this is how they feel in many conversations.

I'm also finding myself being drawn into feeling angry with the male partners and families of the people I am interviewing. Some families do seem to be supporting the participants to do treatment. I need to be aware of my position as a female and how this is influencing the stories that participants are telling me and also what I pay attention to.

12/12/ 12
I have conducted a few interviews in the CF centre now. I was surprised at the close relationships between participants and professionals in the team, which seemed to be strengthened by having children as it is a common ground between people. I have noticed my position as an outsider but also with close links to the team. I am also aware of being organised by 'treatment adherence'. I am attempting to stand back from this and try to understand how participants make sense of CF treatment.

05/12/13
I have begun to become influenced by the social model of disability; I am surprised at just how much my thinking was shaped around an individual understanding of disability. I have become more aware of my assumptions about disability entailing support, resilience and defining the person. Now I am starting to realise just how little CF can feature in a person's sense of themselves even when in terms of time, it occupies a big space in the day.

03/01/13
I am analysing the data and finding it difficult to know how much to go beyond the person's words, for example can I really label what she told me as denial? Some concepts seem to be more value laden. I don't want to misrepresent what participants have said with my interpretations but I need to think more deeply about the meaning within the participants' accounts.

15/03/13
I have gone through so many different ways of interpreting these results now. I met with the qualitative special interest group and asked them to look at my emergent themes. It was reassuring that they arrived at some similar ideas but they also gave me some different ways of thinking about the normality of CF and life trajectory.

24/04/13
These are themes from the bracketing interview we conducted today. We discussed my changing ideas about CF and this research which included:

- I'm not a parent – I have questioned throughout this process, how do I know what is about CF and what is about being a parent? Does this matter, if I'm paying attention to the participants' words?
- I didn't expect people to be positive about being diagnosed with CF – which may have initially influenced how I heard these stories.
- I have developed a lot of respect and admiration for the way people live with what I consider to be challenging circumstances but some people don't see it this way, it is 'life as usual'.

about it he is not allowed to by my husband he's likes no he doesn't want to talk about he just cuts him dead.

[handwritten: feeling def not shared as a family]

Right he changes the subject.

Yeah yeah.

How did you think of yourself before you became a parent?

I think I grew up straight away. Prior to being pregnant I was more self involved not CF wise I was more concerned about not so much even though I didn't do much of my treatment I did know how to keep myself healthy by doing exercise and things like that and obviously when my son came along even though I knew I still had to keep myself healthy he took preference so I, then I went out to keep myself healthy to keep my son healthy it was that would be a thing but yeah I think I was very erm if my friends had colds and this and that I would have thought twice and said right, I'm not coming down today you got a cold I'll see you in a weeks' time it didn't bother me if we were all going out clubbing and I didn't feel well I'm not coming. I didn't care what they thought now I'm more, I'm more worried about what everybody else thinks than what I think.

[left margin handwritten: Cognitive dissonance]
[left margin handwritten: Changing importance of self / putting self second.]
[left margin handwritten: Changing sense of self]

Why do you think that has changed?

I thinks it's because urmm I don't know, I think my son I suppose really because he you're not thinking of yourself so much are you and I'm always thinking, I put my sons feelings before my own and whereas before even if my son before had football or something and I was really really ill I would have turned round and said before oh you know what I'm not coming, and now I'm thinking oh my god I could cough loads of blood up and think oh my god this blood could kill me but I know my sons playing football and I'll put that aside and stand in the freezing cold and the belting down rain just to watch him play football and then I'm thinking in my head I just want to go home and I want to cry and I'm thinking oh my god I could die, I do think that but then I think no I got to watch my son play football and as soon as I get home I jump in the bath and then I think oh I got to have IVs in the next couple of days and I end up doing it then. But usually before that I would have thought no chance I would have thought I don't

[left margin handwritten: Putting self second.]
[left margin handwritten: Sacrificing self.]

[right margin handwritten notes: Need to do what she can if she isn't in control of CF / Second class citizen.]

I love it!

Do you think they show more concern for you having *?

changing views of host

trying to withhold

They did when * was younger, I wouldn't say now that * has got older. when * was younger yes because I think they wanted me in hospital quite a lot but obviously I wouldn't come in because of *, I mean * she was the one that was around was I younger, she would know more and she'd be like oh will you come in and I'd be like oh I can't because I can't leave * and they all knew what * was like he was very errm very nervous, worried about me leaving him, worried about me dying, he thought I could go to bed one night and not wake up.

Projecting son

Needing to prove to her son that she wouldn't leave

That was his fear and some point he was, I think he used to sleep with me until I think he was about 6 and I'd wake up in the middle of the night sometimes and a hand would be there and he would be checking if he could feel my breath and I'd be like oh * what are you doing and he'd be like oh nothing, nothing but I knew what he was doing he was checking the breath out of my nose or out of mouth if I'm still alive but yeah they were but now I wouldn't think say they are more over cautious because of being a mum now, no.

*Noticing impact of * on child.*

Relationship - son - anxious + clingy

question interpreted as a negative way of being by a clinic

I think obviously if was having * now I think they would be but he is an adult now, he is out of the way he is not really going to have an impact on my health now as much as when he was younger.

Child as an obstacle to health.

need to bracket himself - focus on self?

child seen as an obstacle to service.

Would you like them to do things differently / expect things differently?

The only thing that I found was when Dr first started here, he as I said * was about 13, so he was still didn't want me to go into hospital I didn't want to leave him and Dr was like oh you got to be there for your son so you should come in, he didn't think what my son was going through as well, it was just natural his first concern was my health and in his eyes your son , he'll have you when you come home. He said to me if you don't come you could die and then your son could have no one. He was quite abrupt like that and I was thinking I knew what he was saying but I just could not bring myself to. I mean I had come in once or twice because I'd had to come in to have some operation or

Not being treated as a mother.

Not feeling understood. Feeling threatened.

15

NHS

Health Research Authority

NRES Committee South West - Central Bristol

Whitefriars
Level 3, Block B
Lewin's Mead
Bristol BS1 2NT
Email: ubh-tr.SouthWest3@nhs.net

Telephone: 0117 342 1335
Facsimile: 0117 342 0445

03 July 2012

Miss Hazel Barker
Trainee Clinical Psychologist
Cardiff and Vale University Health Board
South Wales Training Programme in Clinical Psychology
1st Floor Archway House
Llanishen,
Cardiff   CF14 5DX

Dear Miss Barker

Study title:	Staying well to parent and parenting to stay well. The experiences of parents with cystic fibrosis.
REC reference:	12/SW/0207
Protocol number:	N/A

The Research Ethics Committee reviewed the above application at the meeting held on 29 June 2012. Thank you for attending with Dr. Moses to discuss the study.

The Committee thought this was a well written study.

Issues discussed:

1. The participant information sheet should:
   - Request permission to access participant's lung function test results
   - Include relevant wording regarding child protection issues
   - Provide a list of organisations that could provide support should a parent become distressed
   - Mention the interview much earlier

2. Upon querying you confirmed you would not ask participants about their possible mortality.

3. Where would interviews be conducted?
   This would be at Llandough Hospital. The Committee wanted to know whether it would be possible to reimburse travel expenses for those who agreed to come to the hospital. You advised that this would be difficult, given the limited funds available.   The Committee thought reimbursement might help with recruitment.

A Research Ethics Committee established by the Health Research Authority

4. Was it considered feasible to obtain ten participants?
    You confirmed that parents were very interested in talking about their condition, and ten was considered a realistic number.

5. When asked, you confirmed you would transcribe the interviews yourself.

Ethical opinion

The members of the Committee present gave a favourable ethical opinion of the above research on the basis described in the application form, protocol and supporting documentation, subject to the conditions specified below.

Ethical review of research sites

NHS Sites

The favourable opinion applies to all NHS sites taking part in the study, subject to management permission being obtained from the NHS/HSC R&D office prior to the start of the study (see "Conditions of the favourable opinion" below).

Conditions of the favourable opinion

The favourable opinion is subject to the following conditions being met prior to the start of the study.

Management permission or approval must be obtained from each host organisation prior to the start of the study at the site concerned.

Management permission ("R&D approval") should be sought from all NHS organisations involved in the study in accordance with NHS research governance arrangements.

Guidance on applying for NHS permission for research is available in the Integrated Research Application System or at http://www.rdforum.nhs.uk.

Where a NHS organisation's role in the study is limited to identifying and referring potential participants to research sites ("participant identification centre"), guidance should be sought from the R&D office on the information it requires to give permission for this activity.

For non-NHS sites, site management permission should be obtained in accordance with the procedures of the relevant host organisation.

Sponsors are not required to notify the Committee of approvals from host organisations

Additional conditions specified by the REC:

1. The consent form
    *   should include boxes large enough to accommodate participant's initials.
    *   should quote the date and version number of the PIS
    *   should include the name and signature of the witness (usually researcher) and date

2. The participant information sheet should:
   - state much earlier that interviews will form part of the study
   - bear the name of our committee as "NRES Committee South West - Central Bristol"
   - include a list of organisations that might be able to help
   - include the name **and** contact details of the person/s to contact in case of problems/complaints
   - request permission to access results of the participant's lung function test
   - include appropriate wording regarding possible breach of confidentiality should any safety issues come to light.

   Link for PIS/CF
   http://www.nres.nhs.uk/applications/guidance/consent-guidance-and-forms/

3. In the IRAS application form (A13) it is stated that a reminder will be sent to participants after two weeks. However, the PIS states that if the participant did not wish to take part, they need do nothing. Please confirm whether the latter is correct, and if so, in the letter of invitation, the last sentence needs to be checked and corrected. The Reminder letter should be withdrawn.

Revised copies of study documentation should be lodged with the Ethics Office.

It is responsibility of the sponsor to ensure that all the conditions are complied with before the start of the study or its initiation at a particular site (as applicable).

You must notify the REC in writing once all conditions have been met (except for site approvals from host organisations) and provide copies of any revised documentation with updated version numbers. The REC will acknowledge receipt and provide a final list of the approved documentation for the study, which can be made available to host organisations to facilitate their permission for the study. Failure to provide the final versions to the REC may cause delay in obtaining permissions.

Approved documents

The documents reviewed and approved at the meeting were:

Document	Version	Date
Interview Schedules/Topic Guides	2	19 April 2012
Investigator CV		
Letter of invitation to participant	2	19 April 2012
Other: Reply Slip	2	19 April 2012
Other: Reminder letter**	2	19 April 2012
Other: CV for Catherine O'Leary		
Other: CV for Dr J Moses		
Participant Consent Form**	2	19 April 2012
Participant Information Sheet**	2	19 April 2012
Protocol	2	19 April 2012
Questionnaire: CFQ-UK Adolescents & Adults	1	
Questionnaire: The Brief Illness Perception Questionnaire		
Questionnaire: HADS		

Membership of the Committee

The members of the Ethics Committee who were present at the meeting are listed on the attached sheet.

Statement of compliance

The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.

After ethical review

Reporting requirements

The attached document "After ethical review – guidance for researchers" gives detailed guidance on reporting requirements for studies with a favourable opinion, including:

- Notifying substantial amendments
- Adding new sites and investigators
- Notification of serious breaches of the protocol
- Progress and safety reports
- Notifying the end of the study

The NRES website also provides guidance on these topics, which is updated in the light of changes in reporting requirements or procedures.

Feedback

You are invited to give your view of the service that you have received from the National Research Ethics Service and the application procedure. If you wish to make your views known please use the feedback form available on the website.

Further information is available at National Research Ethics Service website > After Review

12/SW/0207	Please quote this number on all correspondence

With the Committee's best wishes for the success of this project

Yours sincerely
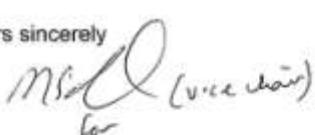
(signature) (vice chair)
for

Dr Pamela Cairns
Chair

Email: naaz.nathoo@UHBristol.nhs.uk

....

NRES Committee South West - Central Bristol

Attendance at Committee meeting on 29 June 2012

Committee Members:

Name	Profession	Present	Notes
Dr Kay Barnard	Ex-research scientist	Yes	
Mr Trevor Beswick	Director - SW Medicines, Information & Training	Yes	
Dr Pamela Cairns	Consultant Neonatologist	Yes	
Mrs Angela Clarke	(Ex-social worker)	Yes	
Dr Simon Croxson	Consultant Physician	Yes	
Dr Ian Davies	Consultant in Cardiac Anaesthesia & Intensive Care	No	
Dr Michael Halliwell	Medical Physicist (retired)	No	
Ms Patsy Hudson	Coach/Mentor	Yes	
Mr Geoffrey Jones	Retired Solicitor	Yes	
Mr John Judges	Lay Member (Insurance/audit)	Yes	
Mr Paul Lewis	Patient Involvement Coordinator	Yes	
Miss Sylvia Pearson	Senior Clinical Trials Coordinator	Yes	
Dr Margrid Schindler	Consultant Senior Lecturer	Yes	

Also in attendance:

Name	Position (or reason for attending)
Ms Angela Chen	Pharm.D. Candidate (Observer)
Professor Julie Kent	Professor of Sociology/Chair - UWE REC (Observer)
Mrs Naazneen Nathoo	Coordinator
Ms Leigh Taylor	Research Manager, UWE (Observer)

Ysbyty Athrofaol Cymru
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Minicom 029 2074 3632

Eich cyf/Your ref
Ein cyf/Our ref
Welsh Health Telephone Network 1872
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Fax: 029 20745311
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From: Professor JI Bisson
R&D Director
R&D Office, 2nd Floor TB2
University Hospital of Wales
Cardiff, CF14 4XW

26 July 2012

Ms Hazel Barker
South Wales Training Programme in Clinical Psychology
1st Floor
Archway House
Llanishen
Cardiff
CF14 5DX

Dear Ms Barker

Cardiff and Vale UHB Ref : 12/MEH/5350 : Staying Well To Parent And Parenting To Stay Well. The Experiences of Parents With Cystic Fibrosis
NISCHR PCU Ref: 84722

The above project was forwarded to Cardiff and Vale University Health Board R&D Office by the NISCHR Permissions Coordinating Unit. A Governance Review has now been completed on the project.

Documents approved for use in this study are:

Document	Version	Date
Protocol	2	19 April 2012
Participant information sheet	3	5 July 2012
Reply Slip	3	5 July 2012
Invitation letter	3	5 July 2012
Reminder Invitation letter	3	5 July 2012
Questionnaire: Brief illness perception		
Questionnaire: Adolescents and adults		
Questionnaire: HADS		
Interview questions	2	19 April 2012
Participant consent form	3	5 July 2012
List of Organisations	1	5 July 2012

I am pleased to inform you that the UHB has no objection to your proposal.

Version 1.0. 05 03 11

Please accept this letter as confirmation of sponsorship by Cardiff and Vale University Local Health Board under the Research Governance Framework for Health and Social Care, and permission for the project to begin within this UHB.

May I take this opportunity to wish you success with the project and remind you that as Principal Investigator you are required to:

- Inform NISCHR PCU and the UHB R&D Office if any external or additional funding is awarded for this project in the future
- Submit any substantial amendments relating to the study to NISCHR PCU in order that they can be reviewed and approved prior to implementation
- Ensure NISCHR PCU is notified of the study's closure
- Ensure that the study is conducted in accordance with all relevant policies procedures and legislation
- Provide information on the project to the UHB R&D Office as requested from time to time, to include participant recruitment figures

Yours sincerely

Professor Jonathan I Bisson
R&D Director

CC   R&D Lead   Prof Nick Craddock
      Dr Jennifer Moses
      Dr Catherine O'Leary

<u>Appendix 8</u>

Information about the Research

Version 3

The Experiences of Parents with Cystic Fibrosis

You are invited to take part in a research study which is being carried out by Hazel Barker, Trainee Clinical Psychologist, under the supervision of Dr Catherine O'Leary (Clinical Psychologist, Cardiff and Vale University Health Board) and Dr Jenny Moses (Consultant Clinical Psychologist, Cardiff and Vale University Health Board).

This research will involve taking part in an interview and completing questionnaires. The results of the research will be written and submitted towards a doctorate in clinical psychology. It may also be published as a journal article. No participants will be identified in either published work.

Before you decide whether you would like to take part, please read this information sheet which explains the purpose of the research and what your role in the research would be. Please feel free to discuss this with others or contact the researcher (details below) to ask any questions if there is anything you are not sure about or if you would like more information.

What is the purpose of the study?

Being a parent can be a demanding, challenging, yet rewarding job. Being a parent with a health condition such as cystic fibrosis might impact on parenting in different ways. There has been some research exploring some of the factors people with cystic fibrosis have considered when making the decision to become a parent. However, there is very limited research exploring the experiences of parenting with a diagnosis of cystic fibrosis.

The purpose of this study is to develop an understanding of the needs of parents, to contribute to the development of services for people with cystic fibrosis and to better inform health professionals' understanding of being a parent with cystic fibrosis.

Why have I been invited to take part?

You have been invited to take part in this research because it has been identified that you are a parent and you have a diagnosis of cystic fibrosis. Ten people will be needed for this research.

1

Do I have to take part?

It is up to you to decide whether or not you would like to take part in this research, it is entirely voluntary. Your decision whether or not to take part, will not affect any of the services that you currently receive from the cystic fibrosis service. If you take part you are free to withdraw at any time without giving a reason.

If you decide to take part, please contact the researcher by returning the reply slip enclosed with this document or by telephoning the researcher (details below).

What will happen if I do agree to take part?

If you decide to take part you will be invited to participate in an interview with the researcher, Hazel Barker. It is anticipated that the interview will last for 45-60 minutes. The interview will take place during the working day and will be arranged at a time and place which is convenient to you (either your home or the cystic fibrosis clinic, Llandough Hospital or Bristol Royal Infirmary). The interview will involve being asked about your experiences of being a parent and about how this role might have impacted on you.

You will then be asked to fill in three questionnaires which should take approximately 10-20 minutes to complete. The questionnaires will be made up of questions that you can complete yourself regarding your mood and well-being. It is anticipated that the whole meeting should not take more than 1 ½ hours of your time and you will only meet me on that one occasion.

In order to remember what you say, the interviews will be audio-recorded so that I can transcribe the information and analyse it.

I will also request your permission to obtain your last lung function test by looking at the database of these test results, held by the Cystic Fibrosis Service.

What are the possible benefits in taking part?

The aim of this research is that your contribution will help to develop healthcare staff's understanding of being a parent with cystic fibrosis and it will also provide you with the opportunity to share your experiences with other parents.

What are the possible disadvantages in taking part?

This study is a psychological study. There are no known risks to taking part. However, discussing your experiences could be difficult and if at any point during the interview you would like to withdraw from the

2

study you will be free to do so, without giving a reason and it will not impact on the care that you usually receive.

If you feel concerned by any of the issues raised in the interview you are invited to contact Dr Catherine O'Leary (Clinical Psychologist, Cystic Fibrosis Service) / Dr Samantha Phillips (Clinical Psychologist, Cystic Fibrosis Service) to discuss them with her.

Will my participation in this study be confidential?

Your participation in the research, what you say in the interview and the questionnaires you complete will be kept confidential; this information will only be available to the researcher. Confidentiality would be broken if you disclosed that you posed a serious risk to yourself or another person. If concerns were raised about the welfare of a child, confidentiality would be broken according to the Children's Act (Acts of Parliament, 1989) and I would contact the relevant child protection department.

The questionnaires will be kept in a locked filing cabinet.

The information from the interviews will be transcribed and some quotes from the interviews will be used. However, all names will be changed so that you cannot be identified.

What will happen to the results of this study?

The results of the research will be written up as a thesis and submitted as part of the researchers Doctorate in Clinical Psychology and may also be published as a research article. You will be sent a summary of the research results and if you would like, I will send a copy of the research on completion of the study.

What if I have a problem with the study or want to make a complaint?

If you have any questions or you are unhappy with this study, please contact the researcher. If you remain unhappy or wish to complain formally you can do this via the Cardiff and Vale University Health Board complaints procedure. Contact details: Mr Adam Cairns, Chief Executive, Cardiff and Vale University Health Board Headquarters, Whitchurch Hospital, Park Road, Whitchurch, Cardiff, CF14 7XB. Alternatively, you can contact a member of the Complaints Department on: 029 2074 4095.

Who has reviewed the study?

All research is looked at by a Research Ethics Committee in order to protect your safety, rights, wellbeing and dignity. This study has been reviewed and approved by the NRES Committee South West – Central Bristol.

Further information

If you have any further questions about taking part in this research please do not hesitate to contact the researcher (Hazel Barker) on:

07577 044576 or return the contact slip to the address below and you will be contacted as soon as possible.

If you do not wish to take part you do not need to do anything. In two weeks time I will send a reminder letter to all participants. If you have already replied or do not wish to participate please disregard this letter.

Thank you for taking the time to read this information sheet.

Hazel Barker
Trainee Clinical Psychologist

Cardiff University
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Park Place
Cardiff CF10 3AT
Wales, UK
www.cardiff.ac.uk/psych

Prifysgol Caerdydd
Adeilad y Tŵr
Plas y Parc
Caerdydd CF10 3AT
Cymru, Y Deyrnas Unedig

Version 3

Invitation to Participate in Research

The experiences of parents with cystic fibrosis

My name is Hazel Barker and I am a Trainee Clinical Psychologist. I am carrying out research exploring the experiences of parents with cystic fibrosis. This information has been sent to you by the Cystic Fibrosis Service on my behalf.

You are invited to take part in this research study and have been sent this information because you attend the Adult Cystic Fibrosis Service and you are a parent. I have enclosed information about the research and a reply slip. If after reading the information you decide that you would like to take part in the research, please complete the reply slip and return it to me in the envelope provided or telephone me on **07577 044576.**

If you do not wish to participate, then you do not need to do anything. In two weeks time I will send a reminder letter to all participants. If you have already replied or do not wish to participate please disregard this letter.

Thank you for your time,

Hazel Barker

Information sent and research supervised by: Dr Catherine O'Leary, Clinical Psychologist

Scientific and Governance Approval by Cardiff and Vale University Board Ref: 12/MEH/5350. Ethical Approval: NRES Committee South West – Central Bristol.

Tel/Ffon: 029 208 70582 Email/Ebost: CAV_Psychology.Training@wales.nhs.uk

Version 3

Reminder Invitation to Participate in Research

The experiences of parents with cystic fibrosis

My name is Hazel Barker and I am a Trainee Clinical Psychologist. I am carrying out research exploring the experiences of parents with cystic fibrosis.

A few weeks ago you were invited to take part in this research study and you were sent an information pack. You were sent this information because you attend the Adult Cystic Fibrosis Service and you are a parent.

If after reading the information you decide that you would like to take part in the research, please complete the enclosed reply slip and return it to me in the envelope provided or telephone me on **02920 870587.**

If you have already replied or do not wish to participate please disregard this letter.

If you do not wish to participate, then you do not need to do anything.

Thank you for your time,

Hazel Barker

School of Psychology
Ysgol Seicoleg

South Wales Doctoral Programme in Clinical Psychology
De Cymru Rhaglen Doethuriaeth mewn Seicoleg Glinigol

CARDIFF
UNIVERSITY

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Reply Slip

Version 3

The experiences of parents with cystic fibrosis

Please tick:

☐ I am interested in taking part in the research.
☐ I would like further information before I decide whether or not to take part.

The following information is to enable me to contact you; it will not be used in the study.

Name: _____

Address:
_____
_____
_____

Email address: _____

Telephone Number: _____

Can a message be left at this telephone number? (please tick)

☐ Yes
☐ No

How old is the child / children that you live with?

_____

Please send this reply slip to Hazel Barker, Trainee Clinical Psychologist in the stamped address envelope provided or telephone **07577 044576.**

Thank you for your time,

Hazel Barker

School of Psychology
Ysgol Seicoleg

South Wales Doctoral Programme in Clinical Psychology
De Cymru Rhaglen Doethuriaeth mewn Seicoleg Glinigol

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Consent Form

Version 3 (05/07/12)

The experiences of parents with cystic fibrosis

Researcher: Hazel Barker, Trainee Clinical Psychologist

Please put your initials in the boxes to indicate your agreement with the following statements:

☐ I confirm that I have read and understood the information sheet for the above study (version 3, dated: 05/07/12). I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily.

☐ I understand that my participation in this study is entirely voluntary and that I can with draw at any time without giving a reason.

☐ I understand that my participation in this study will involve being interviewed by the researcher regarding my experiences of being a parent which will take approximately 45-60 minutes and then completing 3 questionnaires.

☐ I understand that the interview will be audio-recorded and that I can request that it is turned off at any time.

☐ I understand that the information provided by me will be held confidentially, until the point of transcription where all identifying information will be made anonymous and transcripts destroyed. Only the researcher can trace this information back to me individually. The information will be retained for up to 5 years when it will be destroyed. I understand that I can ask for the information to be destroyed at any time and I can have access to the information at any time.

☐ I give my permission for extracts from the transcripts to be used in reports of the research on the understanding that my own and my family member (s) anonymity will be maintained.

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Ysgol Seicoleg

South Wales Doctoral Programme in Clinical Psychology
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Plas y Parc
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Cymru, Y Deyrnas Unedig

Interview Questions   Version 2

1. **Parenting is often seen as a fulfilling but demanding job; looking back across the last year how have you coped with its demands?**
   - What sources of help and support do you have?
   - Which are the most helpful?
   - Sometimes sources of help and support turn out to be unhelpful or of limited use? Have you experienced this?
   - How do you think you would manage if you didn't have these supports?'

2. **Do you think being a parent has impacted on the way you look after yourself?**
   - Did you expect that being a parent would affect the way you manage the above?
   - Are there changes that you hoped that you would make, but haven't been able to?

3. **How has being a parent changed the way you think about yourself as a person with CF?**
   - Has it impacted on the way that you feel about CF?
   - Some people say that becoming a parent can create a feeling of achievement or pride, have you noticed this?
   - Did you hope that being a parent would bring about these changes?
   - Were there any changes that you hoped would happen but haven't?

4. **How might being a parent have changed the way other people view you?**
   - How do you feel about these changes?
   - Did you anticipate these changes?
   - Other than what you have described above, would you like people to view you differently/ expect different things from you?

5. **How might being a parent have changed the way professionals in the CF clinic respond to you?**
   - How might being a parent have changed the way professionals in the CF clinic show concern for you?
   - How do you think being a parent has changed what professionals in the CF clinic want to do for you?
   - How do you feel about these changes?
   - Other than what you have described above, would you like people to respond to you differently/ expect different things from you?

Appendix 10

The Brief Illness Perception Questionnaire

For the following questions, please circle the number that best corresponds to your views:

How much does your illness affect your life?
0   1   2   3   4   5   6   7   8   9   10   no affect at all   severely affects my life

How long do you think your illness will continue?
0   1   2   3   4   5   6   7   8   9   10   a very short time   forever

How much control do you feel you have over your illness?
0   1   2   3   4   5   6   7   8   9   10   absolutely no control   extreme amount of control

How much do you think your treatment can help your illness?
0   1   2   3   4   5   6   7   8   9   10   not at all   extremely helpful

How much do you experience symptoms from your illness?
0   1   2   3   4   5   6   7   8   9   10   no symptoms at all   many severe symptoms

How concerned are you about your illness?
0   1   2   3   4   5   6   7   8   9   10   not at all concerned   extremely concerned

How well do you feel you understand your illness?
0   1   2   3   4   5   6   7   8   9   10   don't understand at all   understand very clearly

How much does your illness affect you emotionally? (e.g. does it make you angry, scared, upset or depressed?
0   1   2   3   4   5   6   7   8   9   10   not at all affected emotionally   extremely affected emotionally

Please list in rank-order the three most important factors that you believe caused your illness. *The most important causes for me:-*

1. _____

2. _____

3. _____

Appendix 11

The Hospital Anxiety and Depression Scale (HADS)

Directions for use

Description
The Hospital Anxiety and Depression Scale (HADS) is a 14-item scale developed by Zigmond and Snaith (1983) to provide a brief state measure of both anxiety (seven items) and depression (seven items). It was designed for use in medical out-patient clinics to detect clinical cases of anxiety and depression and to assess the severity of anxiety and depression, without contamination of scores by reports of physical symptomatology.

Administration
The scale is self-administered with instructions on the printed form and takes about 10 minutes to complete.

Scoring
Each item is scored from 0 to 3 and so the total scores range from 0 to 21 for the anxiety subscale and also for the depression subscale.

Interpretation
Higher scores indicate greater anxiety or depression. Based on Zigmond and Snaith's study of 100 medical out-patients, scores from 8 to 10 on each scale have been taken to indicate possible clinical disorder and from 11 to 21 to indicate probable clinical disorder, as these scores resulted in fewest false positives and false negatives when compared with psychiatric assessment (see Table 2 on page 9).

In addition, the HADS can be repeated at intervals to gauge progress. The HADS Chart, which follows the HADS in this unit, may be used to record these serial scores; the grey band indicates the borderline area.

In a clinical population of 573 people with cancer at the time of initial diagnosis or first recurrence, Moorey et al. (1991) found mean anxiety and depression subscale scores of 5.44 (SD 4.07, range 0–19) and 3.02 (SD 2.98, range 0–15) respectively. The percentage of people scoring above the cut-off (see glossary) (8) for possible clinical disorder was 27 per cent for anxiety and 8.7 per cent for depression.

Later experience with the HADS has established that it may be used as a measure of severity of the states. The four score ranges can be classified 'normal' (0–7), 'mild' (8–10), 'moderate' (11–14) and 'severe' (15–21) (authors' unpublished study).

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Table 2: Number of people in each psychiatric assessment category within each score band on the relevant HADS subscales

| | Psychiatric Assessment | | | | | |
| | Depression | | | Anxiety | | |
HADS scores	Non-cases	Doubtful	Cases	Non-cases	Doubtful	Cases
0–7	57	11	1	41	4	1
8–10	8	7	3	10	9	1
11–21	1	4	8	5	15	14

Evaluation and psychometric status

The internal consistency (*see glossary*) of the two subscales as assessed by Cronbach's alpha was 0.93 for anxiety and 0.90 for depression (Moorey *et al.*, 1991). The HADS has good face validity (*see glossary*) and respondents find it easy and acceptable. Concurrent validity (*see glossary*) was assessed by comparison with 5-point psychiatric rating scales of anxiety and depression for 100 medical out-patients (Zigmond and Snaith, 1983). The HADS subscales correlated significantly with these ratings (anxiety, $r = 0.54$; depression, $r = 0.79$). The construct validity (*see glossary*) of the scale as a measure of two factors was confirmed in a factor analysis of the responses of 568 cancer patients, also by Moorey *et al* (1991). Two independent factors were found accounting for 53 per cent of the variance. These factors were replicated on subsamples. In each case items loaded as expected with the exception of item 7 which loaded on both factors. The correlation between the two factors was 0.37 for men and 0.55 for women. While these data indicate good psychometric properties, they are based on specific populations (medical out-patients and people with cancer) and data are not yet available for other populations.

Comparison

There are many other well-validated scales which measure either anxiety or depression or both and which have good psychometric properties. However, no other measure combines the virtues of being short, measuring both anxiety and depression, giving an index of state and giving a cut-off for probable clinical levels.

References

MOOREY, S., GREER, S., WATSON, M., GORMAN, C., ROWDEN, L., TUNMORE, R., ROBERTSON, B. and BLISS, J. (1991). 'The factor structure and factor stability of the Hospital Anxiety and Depression Scale in patients with cancer', *British Journal of Psychiatry*, 158, 255–9.

ZIGMOND, A. S. and SNAITH, R. P. (1983). 'The Hospital Anxiety and Depression Scale', *Acta Psychiatrica Scandinavica*, 67, 361–70. Test materials now available from NFER–NELSON, Windsor.

Hospital Anxiety and Depression Scale

Name .... Date ....

Clinicians are aware that emotions play an important part in most illnesses. If your clinician knows about these feelings she or he will be able to help you more.

This questionnaire is designed to help your clinician to know how you feel. Ignore the numbers printed on the left of the questionnaire. Read each item and **underline** the reply which comes closest to how you have been feeling in the past week.

Don't take too long over your replies; your immediate reaction to each item will probably be more accurate than a long thought-out response.

fold along dashed line

A

I feel tense or 'wound up':

3   Most of the time

2   A lot of the time

1   From time to time, occasionally

0   Not at all

D

I still enjoy the things I used to enjoy:

0   Definitely as much

1   Not quite so much

2   Only a little

3   Hardly at all

A

I get a sort of frightened feeling as if something awful is about to happen:

3   Very definitely and quite badly

2   Yes, but not too badly

1   A little, but it doesn't worry me

0   Not at all

HOSPITAL ANXIETY AND DEPRESSION SCALE

D		
0		
1		
2		
3		

I can laugh and see the funny side of things:

As much as I always could

Not quite so much now

Definitely not so much now

Not at all

A		
3		
2		
1		
0		

Worrying thoughts go through my mind:

A great deal of the time

A lot of the time

From time to time but not too often

Only occasionally

D		
3		
2		
1		
0		

I feel cheerful:

Not at all

Not often

Sometimes

Most of the time

A		
0		
1		
2		
3		

I can sit at ease and feel relaxed:

Definitely

Usually

Not often

Not at all

D		
3		
2		
1		
0		

I feel as if I am slowed down:

Nearly all the time

Very often

Sometimes

Not at all

A		
0		
1		
2		
3		

I get a sort of frightened feeling like 'butterflies' in the stomach:

Not at all

Occasionally

Quite often

Very often

fold along dashed line

HOSPITAL ANXIETY AND DEPRESSION SCALE

I have lost interest in my appearance:

Definitely

I don't take as much care as I should

I may not take quite as much care

I take just as much care as ever

I feel restless as if I have to be on the move:

Very much indeed

Quite a lot

Not very much

Not at all

I look forward with enjoyment to things:

As much as ever I did

Rather less than I used to

Definitely less than I used to

Hardly at all

I get sudden feelings of panic:

Very often indeed

Quite often

Not very often

Not at all

I can enjoy a good book or radio or TV programme:

Often

Sometimes

Not often

Very seldom

Now check that you have answered all the questions

For office use only:

D : ☐ Borderline 8–10

A : ☐ Borderline 8–10

(left margin scoring column, top to bottom)

D 3 2 1 0

A 3 2 1 0

D 0 1 2 3

A 3 2 1 0

D 0 1 2 3

D   A

fold along dashed line

Hospital Anxiety and Depression Scale Chart

for the monitoring of repeated scores

Name: .... Age: .... Sex: ....

Anxiety

20 18 16 14 12 10 8 6 4 2

Depression

20 18 16 14 12 10 8 6 4 2

Treatment

Date

Appendix 12

Understanding the impact of your illness and treatments on your everyday life can help your healthcare team keep track of your health and adjust your treatments. For this reason, this questionnaire was specifically developed for people who have cystic fibrosis. Thank you for your willingness to complete this form.

Instructions: The following questions are about the current state of your health, as you perceive it. This information will allow us to better understand how you feel in your everyday life.

Please answer all the questions. There are **no** right or wrong answers! If you are not sure how to answer, choose the response that seems closest to your situation.

Section I. Demographics

Please fill-in the information or tick the box indicating your answer.

A. What is your date of birth?

Date [ ] [ ] [ ] [ ] [ ] [ ]
Day   Month   Year

B. What is your gender?
☐ Male   ☐ Female

C. During the **past two weeks**, have you been on holiday or out of school or work for reasons **NOT** related to your health?

☐ Yes   ☐ No

D. What is your current marital status?
☐ Single/never married
☐ Married
☐ Widowed
☐ Divorced
☐ Separated
☐ Remarried
☐ With a partner

E. Which of the following best describes your racial background?
☐ White - UK
☐ White - other
☐ Indian/ Pakistani
☐ Chinese/ Asian
☐ African
☐ Caribbean
☐ Other [not represented above or people whose predominant origin cannot be determined/ mixed race]
☐ Prefer not to answer this question

F. What is the highest level of education you have completed?
☐ Some secondary school or less
☐ GCSEs/ O-levels
☐ A/AS-levels
☐ Other higher education
☐ University degree
☐ Professional qualification or post-graduate study

G. Which of the following best describes your current work or school status?
☐ Attending school outside the home
☐ Taking educational courses at home
☐ Seeking work
☐ Working full or part time (either outside the home or at a home-based business)
☐ Full time homemaker
☐ Not attending school or working due to my health
☐ Not working for other reasons

Section II. Quality of Life

Please tick the box indicating your answer.

*During the past **two weeks**, to what extent have you had difficulty:*

	A lot of difficulty	Some difficulty	A little difficulty	No difficulty
1. Performing vigorous activities such as running or playing sports.........	☐	☐	☐	☐
2. Walking as fast as others	☐	☐	☐	☐
3. Carrying or lifting heavy things such as books, shopping, or school bags.........	☐	☐	☐	☐
4. Climbing one flight of stairs	☐	☐	☐	☐
5. Climbing stairs as fast as others.........	☐	☐	☐	☐

*During the past **two weeks**, indicate how often:*

	Always	Often	Sometimes	Never
6. You felt well	☐	☐	☐	☐
7. You felt worried.........	☐	☐	☐	☐
8. You felt useless.........	☐	☐	☐	☐
9. You felt tired.........	☐	☐	☐	☐
10. You felt full of energy.........	☐	☐	☐	☐
11. You felt exhausted	☐	☐	☐	☐
12. You felt sad.........	☐	☐	☐	☐

Please circle the number indicating your answer. Please choose only one answer for each question.

*Thinking about the state of your health over the last **two weeks**:*

13. To what extent do you have difficulty walking?
    1. You can walk a long time without getting tired
    2. You can walk a long time but you get tired
    3. You cannot walk a long time because you get tired quickly
    4. You avoid walking whenever possible because it's too tiring for you

14. How do you feel about eating?
    1. Just thinking about food makes you feel sick
    2. You never enjoy eating
    3. You are sometimes able to enjoy eating
    4. You are always able to enjoy eating

15. To what extent do your treatments make your daily life more difficult?
    1. Not at all
    2. A little
    3. Moderately
    4. A lot

16. How much time do you currently spend each day on your treatments?
    1. A lot
    2. Some
    3. A little
    4. Not very much

17. How difficult is it for you to do your treatments (including medications) each day?
    1. Not at all
    2. A little
    3. Moderately
    4. Very

18. How do you think your health is now?
    1. Excellent
    2. Good
    3. Fair
    4. Poor

Please select a box indicating your answer.

*Thinking about your health during the past **two weeks**, indicate the extent to which each sentence is true or false for you.*

	Very true	Somewhat true	Somewhat false	Very false
19. I have trouble recovering after physical effort	☐	☐	☐	☐
20. I have to limit vigorous activities such as running or playing sports	☐	☐	☐	☐
21. I have to force myself to eat	☐	☐	☐	☐
22. I have to stay at home more than I want to	☐	☐	☐	☐
23. I feel comfortable discussing my illness with others	☐	☐	☐	☐
24. I think I am too thin	☐	☐	☐	☐
25. I think I look different from others my age	☐	☐	☐	☐
26. I feel bad about my physical appearance	☐	☐	☐	☐
27. People are afraid that I may be contagious	☐	☐	☐	☐
28. I get together with my friends a lot	☐	☐	☐	☐
29. I think my coughing bothers others	☐	☐	☐	☐
30. I feel comfortable going out at night	☐	☐	☐	☐
31. I often feel lonely	☐	☐	☐	☐
32. I feel healthy	☐	☐	☐	☐
33. It is difficult to make plans for the future (for example, going to college, getting married, getting promoted at work, etc.)	☐	☐	☐	☐
34. I lead a normal life	☐	☐	☐	☐

Section III. School, Work, or Daily Activities

Questions 35 to 38 are about school, work, or other daily tasks.

35. To what extent did you have trouble keeping up with your schoolwork, professional work, or other daily activities during the past **two weeks**?
    1. You have had no trouble keeping up
    2. You have managed to keep up but it's been difficult
    3. You have been behind
    4. You have not been able to do these activities at all

36. How often were you absent from school, work, or unable to complete daily activities during the last two weeks because of your illness or treatments?
    ☐ Always   ☐ Often   ☐ Sometimes   ☐ Never

37. How often does CF get in the way of meeting your school, work, or personal goals?
    ☐ Always   ☐ Often   ☐ Sometimes   ☐ Never

38. How often does CF interfere with getting out of the house to run errands such as shopping or going to the bank?
    ☐ Always   ☐ Often   ☐ Sometimes   ☐ Never

Section IV. Symptom Difficulties

Please select a box indicating your answer.

*Indicate how you have been feeling during the past **two weeks**.*

	A great deal	Somewhat	A little	Not at all
39. Have you had trouble gaining weight?	☐	☐	☐	☐
40. Have you been congested?	☐	☐	☐	☐
41. Have you been coughing during the day?	☐	☐	☐	☐
42. Have you had to cough up mucus?	☐	☐	☐	☐ Go to Question 44

43. Has your mucus been mostly: ☐ Clear ☐ Clear to yellow ☐ Yellowish-green ☐ Green with traces of blood ☐ Don't know

*How often during the past **two weeks**:*

	Always	Often	Sometimes	Never
44. Have you been wheezing?	☐	☐	☐	☐
45. Have you had trouble breathing?	☐	☐	☐	☐
46. Have you woken up during the night because you were coughing?	☐	☐	☐	☐
47. Have you had problems with wind?	☐	☐	☐	☐
48. Have you had diarrhoea?	☐	☐	☐	☐
49. Have you had abdominal pain?	☐	☐	☐	☐
50. Have you had eating problems?	☐	☐	☐	☐

Please make sure you have answered all the questions.

THANK YOU FOR YOUR COOPERATION!

Appendix: 13   Frequency of Master Themes within the Transcripts

	Anna	Adam	Ben	Beth	Catrin	Lucy	Luke	Nina	Tom
I'll have them while I'm young	Page: 1, 5	N/A Diagnosed after children	Page: 3, 7, 15	Page: 7	Page: 5, 6	Page: 11, 16, 17	N/A Diagnosed after children	Page: 5	Page: 7, 12, 13
Its sharpened my focus	Page:2, 3, 5	Page: -	Page:1, 4, 5, 14	Page:3, 4, 5,	Page:2, 3, 6	Page: 7, 8, 16	Page: 1,2, 8,9	Page:1, 4, 8, 12, 13	Page: 4, 6, 7, 9, 11
I've got to prioritise	Page:5, 6, 14	Page:4, 6	Page:1, 2, 4, 10, 11	Page:2, 3, 6	Page:1, 2, 3, 10	Page: 2, 6, 7, 10	Page:3, 5, 6, 10, 16,	Page:1, 2, 3, 4, 5,6, 8, 9 , 15, 16, 17	Page: 1, 2, 3, 5, 7, 9, 13
It was such euphoria!	Page:2, 3, 4, 8, 10, 12, 16, 17	Page: 3, 8, 11	Page:2, 6	Page: 5, 6, 7, 9	Page:7, 10, 11	Page: 4, 10, 11, 12, 18	Page:10,	Page:3, 9 10, 11, 13, 14, 16	Page: -
I had accomplished something	Page:1, 10, 12, 14, 16, 17	Page:6, 7	Page:15, 1617, 18	Page:5, 7, 8, 11	Page:7, 8	Page: 6, 18, 20, 21	Page:10	Page:3, 9, 10, 11, 12, 1314,	Page:6, 8
I never think that I've got CF	Page:3, 4, 6, 13,	Page:3, 4, 5, 6, 16	Page: 8, 9, 10, 13	Page:1, 2, 5, 9, 12	Page:4, 5, 6, 7, 10, 11	Page: 13, 14, 20, 22, 11, 5	Page:14, 1, 4, 8, 14,	Page:2, 4, 5, 6, 7	Page: 6
I want to see my children's children	Page:10	Page:4	Page:3, 4, 5, 12, 15, 16	Page: 6	Page:6, 2, 4	Page: 11, 16, 14, 23,	Page: -	Page:2,3, 7, 16	Page:6, 12, 13
I look at myself and into the future	Page: 10,	Page:7	Page:3, 5, 7, 8, 11	Page:5, 6, 13	Page: 4,6, 7, 10,	Pages: 6, 12, 15, 16, 22	Page: 1, 8, 9, 14	Page:2, 3, 16	Page:12, 13
Parenting with uncertainty	Page:8, 9	Page:4, 5, 18	Page:4, 5, 6, 9, 12, 13, 15	Page:13,	Page:10, 11, 8	Page: 14, 23, 11, 16,	Page:1, 2, 3, 5, 6, 8, 7, 9, 10, 13, 14	Page:1, 15, 16,12, 13, 2, 3, 14, 15	Page: 1

Brief Illness Perception Questionnaire: Summary of Participants' Scores on the BIPQ

Participant Number	Composite Score	Consequences	Timeline	Personal Control	Treatment Control	Identity	Concern	Coherence	Emotional Response
Adam	25	1	10	8	0	2	2	1	1
Lucy	23	3	10	1	0	3	3	0	3
Anna	26	3	10	1	0	8	2	0	2
Beth	31	5	10	2	0	9	3	0	2
Tom	34	5	10	3	0	6	4	0	6
Nina	63	9	10	7	8	10	9	2	8
Ben	47	8	10	7	3	9	8	0	2
Luke	54	6	10	10	4	7	8	1	8
Catrin	45	6	10	6	0	8	8	2	5
Total	348	44	90	45	15	62	47	6	37
Mean	**38.67**	**4.9**	**10**	**5**	**1.7**	**6.9**	**5.2**	**0.7**	**4.1**