The impact of managing a child's Chronic Fatigue Syndrome/ Myalgic Encephalopathy (CFS/ME) on family relationships

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Table of Contents

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Abstract

Although there is a large body of research literature concerning the impact of childhood chronic illness on family relationships, very little of this has specifically focussed on the impact of childhood Chronic Fatigue Syndrome/Myalgic Encephalopathy (CFS/ME). The current study therefore aims to investigate the impact of having a child with CFS/ME on family relationships from the perspective of parents. This study takes a qualitative approach. Semi-structured interviews were used to explore 18 parents' experiences of having a child with CFS/ME and the impact this has had on family relationships. Interview data were transcribed and analysed using thematic analysis. The analysis led to the identification of 5 main themes: “Long and Difficult Journey”, “Uncertainty”, “Isolation and Restriction”, “Focus on the unwell person at the expense of family life” and “Parental roles”. They felt that life had become a constant struggle and often felt exhausted. They felt uncertainty, isolation and restriction. These parents talked about feeling like family life had become focussed on their unwell child, such that some expressed a need to escape from the situation. They also described how, as a consequence of managing their child's CFS/ME, family members could often feel marginalised and the illness could be seen as a destructive force. However, many parents also described working as a team and feeling that family relationships had benefitted in some way. The results of this study are in line with findings of other studies into other chronic illnesses. In addition, this is the first study to focus exclusively on the impact of CFS/ME on family relationships from the perspective of the parents, and is unique in that there is a dual focus on both mothers and fathers. Clinicians should be aware that parents of children with CFS/ME may need additional support, such as information as to what to expect at various stages of the illness and where to access the relevant support.

THESIS OVERVIEW

This thesis reports a study conducted to explore parents' experiences of looking after a child with a diagnosis of chronic fatigue syndrome, and their view on how the condition impacts on family relationships. Chapter One provides an overview of background information regarding Chronic Fatigue Syndrome/Myalgic Encephalopathy (CFS/ME) in childhood, a review of the literature pertaining specifically to the impact of childhood chronic illness on parents and the rationale for the study. Chapter Two addresses the research methodology, providing justification for the choice of the research paradigm, and outlining the sampling method, and the process of data collection and analysis. Issues of ensuring quality in qualitative research and ethical considerations are also discussed. Chapter Three presents the results of the qualitative Thematic Analysis of the data collected from 18 participants interviewed about CFS/ME and family life. The analysis led to the identification of 5 main themes: "Long and Difficult Journey", "Uncertainty", "Isolation and Restriction", "Focus on the unwell person at the expense of family life" and "Parental roles". These themes are explored with reference to various sub-themes. Chapter Four discusses the research findings in relation to the literature identified in Chapter One and additionally provides a discussion of the limitations of the study and implications arising from the study in relation to clinical practice, policy and research.

CHAPTER ONE: INTRODUCTION

CHAPTER OVERVIEW

This chapter will, firstly, provide an overview of the prevalence, presentation and treatment of CFS/ME in childhood. This will incorporate a review of the relevant literature of the impact of childhood CFS/ME on families and because of the paucity of research in this area, the research literature regarding the impact of other childhood chronic illness on families is considered. Finally a rationale for the current study will be presented along with the aims.

1.2 What is Chronic Fatigue Syndrome/ Myalgic Encephalopathy (CFS/ME)?

Chronic Fatigue Syndrome (CFS/ME) is defined as disabling fatigue without another cause (NICE guidelines, 2007; RCPCH guidelines, 2004). There is no generally accepted theory about its cause or causes, and the symptoms can be diverse, with wide variations both between individuals and in each person over time (NICE guidelines, 2007). NICE recommends that diagnosis should be made after other possible diagnoses have been excluded and the symptoms have persisted for 4 months in adults and 3 months in children and young people[1].

1.2.2 Prevalence

There are no published figures on the incidence of CFS/ME in children and young people but several studies have estimated the point or period prevalence. Estimates of the prevalence of paediatric CFS/ME show considerable variation according to survey methodology and diagnostic criteria. A postal survey of 1024 UK general practices (GPs) estimated the prevalence using the Royal College of Paediatrics and Child Health Guidelines to be only 0.06% in children aged 5-19 years (Haines et al. 2005), while population based surveys suggest that the prevalence is between 0.1% and 0.5% (Chalder et al. 2003; Rimes et al. 2007; Jones et al. 2004). According to Crawley et al (2011) these differences could be due to difficulties in diagnosis, non-attendance at medical services or differences in the type of CFS/ME identified by GPs compared with the spectrum of the condition in the childhood population.

1.2.3 Presentation

When young people with CFS/ME are referred to a specialist service there is a clear distinction between days when they will engage in a relatively high level of “high energy” activity (good days) and days when they are unable to manage much “high energy” activity at all (bad days). This pattern of activity makes it difficult to establish any type of routine (Burgess, M. 2005). “High energy” activity is any type of physical, cognitive or emotional

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[1] in this context a “child” an be defined as anyone up to 12 years of age while a “young person” is anyone between 13 to 18

activity that uses up a lot of energy. Children and young people will be engaged in a mix of these types of activity through the day. Initially it is important to explain to the individual and their parents that it is this “boom and bust” pattern of activity that is prolonging the fluctuations in their symptoms. In order to start to feel better individuals need to find their “baseline” of activity; this is essentially the level they can sustain on good days and bad days without a sharp increase in symptoms (payback) (Crawley and Chambers, 2005). “Feeling better” is typically defined as being able to engage in activity previously not managed for some time, typically this will be a return to school, however NICE (2007) recommends that time in education should not be used as a sole marker of progress of CFS/ME, rather that there should be a balance between education and home and social activities.

1.2.4 Treatment

The Royal College of Paediatric and Child Health (RCPCH) recommends that treatment for CFS/ME should be “collaborative, involving parents and family” (RCPCH guidance, 2004, pg 43), while the National Institute for Clinical Excellence (NICE) advise that engagement with the family is particularly important for children and young people with the condition (NICE guidance, 2007).

There is no “cure” for CFS/ME in the traditional sense, therefore “treatment” aims to help the individual and family manage and adapt to the symptoms. A treatment programme is psychosocial and will typically include Cognitive Behavioural Therapy (CBT), Graded Exercise Therapy (GET), sleep management and relaxation training. Ideally the treatment programme will be delivered by a multi-disciplinary team including psychologists (NICE Guidance, 2007).

1.2.5 The contentious nature of CFS/ME

CFS/ME is distinct from other chronic conditions in that it fits into the broader category of Medically Unexplained Symptoms (MUS). Veerhak et al. (2006) state that although the term Chronic Fatigue Syndrome suggests a categorical ‘disease’, there are valid reasons to abstain from such categorizations and to consider the term Medically Unexplained Symptoms as one general condition (Veerhak et al. 2006). Indeed, Wessely et al. (1999) provide the following arguments, after studying case definitions and epidemiological findings concerning 12 different functional somatic syndromes:

- There is a considerable overlap in symptoms, required for a case definition, between several syndromes;
- Patients meeting criteria for one syndrome, often meet criteria for other syndromes as well;
- Patients with different syndromes share non-symptom characteristics;
- Different syndromes respond to the same therapies.

Wessely et al.'s (1999) argument is supported by evidence provided by Robbins et al. (1997) who studied clustering of 23 symptoms, often associated with functional syndromes. These symptoms clustered in five syndromes (many of them loading on more than one syndrome): pain, fatigue, irritable bowel, somatic symptoms of anxiety and somatic symptoms of depression. These five clusters were highly inter-correlated as well, leading to the conclusion of one general latent trait - Medically Unexplained Symptoms. This conclusion is also supported by Stanley et al (2002).

Historically, many within the medical profession have been sceptical of CFS/ME, have not recognised it as a "true" condition, and have lacked agreement on its prevalence (Wallace, 1991; Mounstephen and Sharpe, 1997; Solomon and Reeves, 2004). Contrasting viewpoints have been expressed by different CFS experts. In a letter to the Lancet in 1993, psychiatrists David and Wessely contested the WHO classification of CFS under diseases of the nervous system, arguing that it was a form of neurasthenia[2] to be classified as a psychiatric condition (David and Wessely, 1993). Dutch researchers authored a 1998 study that developed and tested a model where behavioural, cognitive, and affective factors played a role in perpetuating fatigue, and concluded that this was the correct model for CFS (Vercoulen et al. 1998). After an attempted replication of the Dutch model with a population-based study, Song and Jason (2005) stated the model adequately represented chronic fatigue secondary to psychiatric conditions, but not CFS. They reported finding important differences between CFS and psychiatrically explained chronic fatigue which could affect the development of therapy and explanatory models (Song and Jason, 2005).

There has been disagreement over the pathology of CFS/ME, how it should be diagnosed, and how to treat it (Hooge, 1992; Sharpe, 1996; Denz-Penhey and Murdoch, 1993; Greenlee and Rose, 2000; Horton-Salway, 2007). Only 52% of GPs feel confident making

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[2] Neurasthenia is a term that was first used at least as early as 1829 to label a mechanical weakness of the nerves. As a psychopathological term, neurasthenia was used by Beard in 1869 to denote a condition with symptoms of fatigue, anxiety, headache, neuralgia and depressed mood.

a diagnosis of CFS/ME in adults (Bowen et al. 2005). Evidence from qualitative studies in adults with CFS/ME suggest that a diagnosis of CFS/ME brings with it psychosocial challenges perhaps not as frequently encountered when living with more well understood physical illnesses (Fisher & Crawley, 2012). This is exacerbated by ongoing debate over the name (CMO report, 2002). Experiences of stigma and a de-legitimisation of the person’s subjective experience seem common (Arroll & Senior, 2008; Dickson, Knusson, & Flowers, 2007; Ware, 1993). Other studies show the importance participants put on other people understanding CFS/ME as a physical problem. This has been postulated to help avoid feelings of guilt, stigma or blame related to the illegitimacy of the illness (Guise, Widdicombe, & McKinlay, 2007). Hossenbaccus and White (2013) found that 89% of patient organisations thought CFS/ME to be physical, compared with 58% of newspaper articles and 24% of medical authorities. They suggest that this might explain the gulf between patients and doctors, and why many patients are reluctant to engage in behavioural treatments (Hossenbaccus and White, 2013).

1.3 Literature Review

1.3.2 The impact of CFS/ME on family relationships

There is a small amount of research highlighting the impact CFS/ME has on families. Brace et al. (2000) suggested that there are statistically significant differences between children with CFS/ME compared with children with Juvenile Arthritis or healthy controls in terms of psychosocial disability and parental reinforcement of illness behaviour. However, a major limitation was the small sample size (n=10) particularly because this was a quantitative study. Small sample sizes are not so problematic when adopting a qualitative methodology, therefore it would perhaps have been advantageous for Brace et al. to conduct an in-depth qualitative exploration of the views of children and parents. This would have been particularly apt as at the time of their study no research had explored the views of children with CFS/ME and their parents.

Jelbert et al. (2010) explored adolescents' experiences of CFS/ME. Five themes were found from analysing interview data with Interpretative Phenomenological Analysis (IPA). These reflected both the negative and positive aspects of the illness, in particular the impact CFS/ME has on family relationships, specifically a potential increase in stress and strain but also a strengthening of family bonds. However, a limitation to this study is that the adolescents who took part were "recovered" which may have influenced their retrospective accounts and recall of events.

Two studies have focused on the impact of CFS/ME on families from the parents' perspective. Rangel et al (2005) compared 3 groups of parents of children with chronic conditions, Juvenile Rheumatoid Arthritis (JRA) (n=23) CFS/ME (n=25) and Emotional Disorders (ED) (n=21). Differences were found between the JRA parents and the CFS/ME parents in so much as the parents of children with CFS/ME were at almost 50% higher risk of psychopathology, were more "emotionally over-involved" with their child, and there were higher rates of "illness-related family disruption of family interactions". Essentially this meant that the CFS/ME families reported more family arguments, disruption of leisure activities and a feeling of isolation as compared to the JRA families. Rangel et al. report that there were similarities between family characteristics (i.e. parental mental distress, illness-related disruption of family interactions, and possibly emotional involvement) in

CFS/ME and ED. Another retrospective study also established this maternal over-protectiveness in parents of children with CFS/ME (Fisher and Chalder, 2003). This was a small sample of families representing children on the severe end of the illness spectrum, results should therefore be interpreted with caution in generalising to those with less severe CFS/ME.

Most recently Missen (2012) highlighted the financial impact of the CFS/ME on the family. She also found that tensions within the parents' marriage, attributed to having a sick child, were reported by 6 out of 8 mothers, with one relationship ending in separation and one participant reporting a breakdown in the couple's sex life. In addition Missen (2012) describes negative emotions reported by mothers such as frustration, low mood and guilt and physical symptoms such as headaches, cold sores, nausea and weight gain. Other research has also suggested that parents of children with CFS/ME are at high risk of mental distress and physical illness (Rangel et al. 2005, Van de Putte, 2006). In addition it has been suggested that adolescents with CFS/ME and their parents show a reduced internal health control in comparison with healthy families, "their belief in personal control over illness is diminished in favour of a belief in chance or physicians influencing their illness." (van de Putte et al. 2005, pg 1022). However, the small number of studies in this area tend to focus exclusively on mothers and there are no studies that explore the impact of a child's chronic fatigue syndrome on family relationships.

Although the study by Missen et al. (2012) touched on the impact of CFS/ME on family relationships, the focus of the research was the financial impact of the condition. Elsewhere, Morris and Ogden (2012) interviewed 13 mothers of children with "medically unexplained symptoms" (MUS) such as headaches, nausea, dizziness and fatigue, all of which are symptoms which would come under the diagnosis of Chronic Fatigue Syndrome. Morris and Ogden describe the impact of these unexplained symptoms as being similar to the impact of other chronic illnesses. They describe how parents find the experience "hugely stressful" (pg 290) and how there have been "terrible battles and arguments in the family" (pg 291). However, although these experiences are similar to those of the parents managing other chronic conditions, there was no medical confirmation that these children were suffering from chronic fatigue syndrome. Only one parent had a CFS/ME label for their child which they had decided on themselves. Therefore it could be suggested that Morris and Ogden's research reflects the impact of managing an undiagnosed condition, rather than the impact of CFS/ME-like symptoms. Still there is no information on the impact of CFS/ME on fathers: Missen et al. (2012) and Morris and Ogden (2012) both have a sample exclusively of mothers.

1.3.3 Lack of Research into CFS/ME and the family

There is still a paucity of research on the impact of CFS/ME on family relationships. This is consistent with other reviews. For example in a review of 8 studies looking at the impact chronic illness had on parents of children with chronic illness, Fisher (2001) identified three main themes; the need for normality and certainty; the need for information; and the need for partnership. CFS/ME was not one of the illnesses included. Barlow and Ellard (2005) completed a comprehensive review of all reviews (systematic reviews, meta-analyses, literature reviews) of the research literature regarding the psychosocial well-being of children with chronic disease, their parents and siblings. Although the search term “CFS/ME” was included in over 10 online databases, CFS/ME does not feature in any of the final reviews. Coffey (2006) revealed that childhood chronic illness has a significant impact on parents. They carried out a comprehensive metasynthesis of qualitative studies that focussed on experiences of parents caring for a child with chronic illness. The 11 studies included covered 4 different countries, 533 participants and 32 illnesses, but again CFS/ME did not feature. Eggenberger et al (2011) examined the impact on 8 different chronic illnesses on family life with 9 families. The initial 6 families who took part had responded to a newspaper advertisement, while the final 3 families were recruited from “the professional network of the researchers”. It is not clear what this professional network is, and exactly how these final three families were recruited, and again CFS/ME did not feature. More recently Eccleston et al. (2012) completed a systematic review across 4 databases, looking at RCTs of psychological interventions for parents of children with chronic illness. 35 studies were included in the review, covering 14 illnesses. Again, CFS/ME did not feature. The lack of research into CFS/ME may be because of it’s contnetious nature (see section 1.2.5), however, there may also be a disincentive to carry out research into CFS/ME as some leading researchers into have received harassment and even death threats[3].

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[3] BBC News website reference http://www.bbc.co.uk/news/science-environment-14326514

1.3.4 The impact of childhood chronic illness on families

Although there is a paucity of research regarding the impact of childhood CFS/ME on families, there is a larger body of research regarding other chronic childhood conditions and the impact they have on family life. The next section of this literature review concerns the wider area of research looking at childhood chronic illness and it's impact on the family.

In the early 1980's Sergent stated that "chronic illness presents a crisis for the family" (1984). This "crisis" is reflected in many areas of family life such as financial, medical, social, career and family relationships (Martin et al. 1992). Often families will be described as being on an "emotional rollercoaster" when dealing with chronic illness (Morison et al. 2003; Boss and Couden, 2002; Mussatto, 2006). Research into families dealing with childhood chronic illness other than CFS/ME typically reports that parents feel a plethora of negative emotions such as stress, depression, worry, anger, numbness, guilt, tiredness, sorrow and frustration (Britton, 1999; Hobfoll and Spielberger 1992; Nuutila and Salantera, 2006; Tew et al, 2002; Jordan et al. 2007). This "crisis" has been explored in a variety of chronic illnesses, for example Diabetes (Dashiff et al. 2007; Guell, 2011), Cancer (Dolgin et al. 2007), Congenital Heart Disease (Tak and McCubbin, 2002), Cystic Fibrosis (Derouin and Jessee, 1996), Asthma (Svarsdottir et al, 2005), and Juvenile Arthritus (Britton, 1999).

This study is based on the premise from Family Systems Theory that the whole family is affected when a young child is diagnosed with a chronic condition and the assumption that uncertainty surrounding the child's condition is stressful for families. The family is seen as an interlinked system with each member having an impact on each other member. family systems theory avoids labelling behaviours as good or bad but rather focuses on examining the function that a behaviour plays within a system. A critique of family systems theory is that it does not take into account power dynamics in families, and therefore does not hold individuals to account when there are particular problems such as family violence (Murray, 2006). In addition, some family therapists state that systems theory is inadequately able to describe an individual's responsibility (Spronck and Compernolle, 1997) .

A family systems perspective would suggest that if one member of the family system is unwell then the rest of the system, the family, will be effected in some way. This understanding was central to informing the design and interpretation of the current research.

The Family Systems Illness Model provides a useful framework for evaluation, formulation, and intervention with families dealing with chronic illness, enabling us to “think systemically about the interface of any chronic condition and the family, the model casts the disorder itself in systems terms according to its pattern of psychosocial demands over time” (Rolland, 2006, p.243). Rolland suggests that when evaluating general functioning, life-cycle passage, and illness-related family dynamics it is useful to focus on the following areas:

1. The interaction of an illness with individual and family development
2. The multigenerational history of coping with illness, loss and adversity
3. The family’s health/illness belief system and the meaning of the child’s condition
4. family, social, and community resources to manage medical crises and long term care
5. relationships between healthcare institutions, professionals, the patient and family

The Family Systems Illness model emphasising how the impact of chronic illness reverberates throughout the family system, affecting all members and their relationships on physical, psychological and social levels (Rolland and Walsh, 2006; Eggenberger et al. 2011).

1.3.5 Parent’s health

Much of the research literature regarding the impact of childhood chronic illness on parents suggests that they often have impaired physical health. Self reported Health Related Quality of Life (HRQoL) has been found to be lower in parents of children with chronic conditions compared to parents of healthy children (Hatzmann, 2008; Al Robaee and Shahxad, 2010; Morrow et al. 2012; Klassen, 2012) though it is important to note that much of this research focuses on mothers and therefore cannot be generalised to the experience of fathers, in addition a reliance on self report measures raises questions regarding the reliability and validity of results. Kuster (2004) has suggested that HRQoL may be lower in parents of children with chronic conditions because the demands of caregiving can lead to a reduction of health promotion activities such as exercise, however this research was based on a small sample of just 38 mothers and there was no comparison group, therefore it is not possible to verify the amount of participation in health promoting activities by mothers of healthy children. In two studies of families caring for children with chronic illness in China, Martinson et al. (1995 and 1997) report that caregivers identified the most severe problem related to caregiving as its impact on their personal health, though again both these studies focussed exclusively on mothers. Hopia et al. (2005) note that when a child has a serious illness often the whole family is “ill” and the overall health of the family becomes vulnerable. Parents of children with either cancer, diabetes, asthma or cystic fibrosis have been found to suffer insomnia, headaches, stomach aches, a general sense of pain and anxiety (Hopia et al. 2005). Parents of children with epilepsy have been found to experience sleep problems (Cottrell, 2005; Nolan, 2006), and the same has been found for parents of children with eczema (Moore et al. 2006), and cystic fibrosis (Meltzer and Mindell, 2006; Meltzer and Moore, 2008). Jordan et al. (2007) gathered focus group data from parents of children with chronic pain, one such parent is reported as saying: “It’s amazing how exhausting it is...mentally, emotionally and physically, I would never have believed it . . . We’re through that phase now . . . but I remember how incredibly tired I got. I, if I sat down, I would go to sleep” (pg 53). Thyen et al. (1998) found that mothers of children with complex health conditions such as brain injury, congenital malformations, metabolic disease, myopathies and brain tumors reported impaired health status compared to parents of healthy children. Gabor and Farnham (1996) found that mothers reported that they felt more prone to illness, and they directly attributed this to the stress caused by caring for a chronically ill child. This research was however focused on single mothers from deprived backgrounds which makes it impossible to generalise findings to other non-single parent families. In addition deprived single mothers may also face a multitude of other factors that impact on day to day stress levels

In addition to the negative impact on parental physical health there is ample evidence to suggest that having a child with a chronic illness will have a detrimental effect on parental mental health; the experience has been described as an “emotional rollercoaster” by parents (Ganonni and Shute, 2009. pg 44) who feel stress, guilt, depression, fear, sadness, bitterness, and have difficulties coping with everyday life (Martinson et al. 1995; Bystrom, 2012).

There is evidence that although mothers may often provide the majority of the care for the unwell child, they may doubt their ability as parents to cope; In their study of 33 mothers of children with eczema, Faught et al. (2007) found that 46% reported feeling incompetent at managing their child's behaviour. This may be related to the fact that despite parents' best efforts to alleviate their children's suffering, with chronic as opposed to acute illness, children's discomfort is ongoing. This is explored by Maciver et al. (2010) in their research exploring the experience of mothers of children with chronic pain. They report that parents felt an acute sense of helplessness when faced with the nature of their child's pain, primarily at their inability to help, comfort, or soothe their child.

In a study focusing on the impact of a recent diagnosis of childhood cancer or juvenile diabetes/epilepsy on parental quality of life, Goldbeck (2006) reports that parents described considerable restrictions in their emotional stability and their general well-being. A difficulty for parents is that their own emotional experience is just as chronic as their child's condition. Many parents have reported that the ongoing stress can be relentless (e.g. Ware and Raval, 2007). Leonard et al. (1993) suggest that parents' distress levels seem to be effected by increased family responsibility. Parents can be affected to the extent that psychiatric support is needed (Leonard et al. 1993; Bhadada, 2011) and they can be at risk of Post-Traumatic Stress Disorder (Casey, 2012; Gudmunsdottir et al. 2006; Kazak et al. 1997), and depression (Thyen et al. 1998). It is important to note that much of this research evidence focusses on the experience of mothers, and when fathers are the focus (e.g. Ware and Ravel, 2007) the sample sizes are very small. Therefore, while there is evidence to suggest managing a childhood illness has a detrimental effect on parental physical and mental health, it is difficult to draw firm conclusions about wether the impact is the same for fathers as it is for mothers.

1.3.6 Are mothers more vulnerable than fathers?

The evidence suggests that mothers of children with chronic conditions are more psychologically vulnerable than fathers, often scoring higher on depression and anxiety scales (Moore et al. 2006; Mastroyannopoulou et al. 1997; Yildiz et al. 2009), Martinson et al. (1995) report that mothers of children with chronic conditions describe feeling distressed at seeing healthy children. In a pilot study using a self-completed questionnaire Britton (1999) reports that parents of children with Juvenile Rhuematoid Arthritis experience an increase in stress and sorrow after diagnosis, which is particularly true for mothers.However, Britton also reports differences between parental involvement in daily care and between how mothers and father's coped with their situation. Mastroyannopoulou (1997) found that mothers and fathers coped with their child's illness differently. At diagnosis fathers' most commonly used strategy was to cope practically or by emotional withdrawal, whereas mothers were more likely to cope through emotional release. Yildiz suggests mothers are more affected by their children's unwell status than fathers because mothers "are more actively engaged in their children's care than fathers, more often in communication with their children and spend more time with their children" (pg 45).

Although it may seem that mothers may be more vulnerable to the emotional impact of childhood chronic illness than fathers, this should perhaps be interpreted with caution as the majority of studies have a greater female to male ratio, and some studies focus exclusively on mothers (e.g. Van Skiver et al. 1995; Moskowitz et al. 2007; Kashikar-Zuck et al. 2008). There is evidence to suggest that although fathers report fewer psychological symptoms than mothers, fathers and mothers do not differ significantly with regard to physical symptoms (Holm et al. 2008), which suggests that fathers are being impacted by their child's chronic illness, but that the tools being used in the research don't currently reveal the full picture. An alternative explanation that has been posited as to the impact on fathers is that fathers may be more prone to externalise their problems through fighting and drinking (Mastroyannopoulou, 1997)
The overall picture to emerge from the wider research literature is one of fathers showing less outward distress than mothers, fathers having to focus on helping their spouse to cope with the crisis while having to hide their concerns about their child in order to help provide emotional support to their partner. Fathers also strive to maintain a feeling of control through their work outside home (Ware and Ravel, 2007).

1.3.7 The impact of perceived severity of illness

High levels of stress have been reported by mothers of children with congenital heart defects (Davis, 1998), asthma (Vila, 2003), eczema (Faught, 2007), epilepsy (Nolan et al. 2006) and chronic pain (Williams, 2009). However, these studies report on parental samples whose children are at the severe end of their particular illness spectrum, which may itself be a factor related to stress levels. Indeed research suggests that maternal mental health is worse the more severe the child's physical condition (Lustig et al, 1996; Thyen et al, 1998). Lustig et al. (1996) suggest that this may be because mothers experience increased strain as their child's functional status declines. In turn, increased maternal demands might place a burden on both financial and emotional resources needed to meet other family tasks. This perceived burden may lead to mental health problems. Power et al. (2003) found that mothers of children with severe Juvenile Rheumatoid Arthritis were more overprotective than mothers of children with milder arthritis. They suggest that this could be because mothers of the severely affected children are more anxious. Hatzmann et al. (2008) reports that in a sample of 533 parents managing a range of different chronic illnesses the parents of children with metabolic disease are the ones with the lowest health related quality of life. They suggest that this might be explained by the hereditary and progressive nature of these diseases. It may be that parents whose children's conditions are not so severe may experience lower levels of stress. This is supported by Katz (2002) who found that parents of children with life threatening chronic illnesses perceived the impact of the illness to be significantly more catastrophic than parents of children with none life threatening chronic conditions. Zahr et al. (1994) found that the impact on mothers was worse when their child had Leukemia compared to congenital heart disease. They suggest that this is because of Leukemia's chronicity, debilitating effects, social stigma, and poor prognosis. Eddy and Engel (2008) found that parents who had children with "less medically stable" conditions such as neuromuscular disease, spina bifida or cerebral palsy experienced more worry and concern than parents of children with more stable conditions. Although CFS/ME can range from mild to severe, the condition is not life threatening, could it therefore be hypothesised that parents of children with CFS/ME will have lower stress levels that parents managing other more severe conditions? Evidence from similar conditions would suggest not; Lipani and Walker (2006) found that regardless of the severity of their abdominal symptoms, when children viewed their pain condition as serious and evaluated their coping potential as low, mothers reported more worry about their children's health and were more likely to attribute restrictions in family activities and personal time to it. While it is clear that being a parent of a child with a chronic illness is stressful, much of the research into this area is cross-sectional, and therefore only gives a snapshot of the experience at that time. It may be that over time there is a reduction in the experience of stress.

1.3.8 Is there a reduction of "impact" over time?

There is evidence that levels of parental distress can be high at diagnosis and then reduce over time; Gabor and Farnham (1996) found that mothers reported the time of diagnosis as an especially difficult time, with initial emotional responses of denial, confusion, anger, and despair. Notham et al (1996) examined the initial impact and subsequent adjustment of parents to their child's diagnosis of diabetes. They found that immediately after diagnosis, the children and both parents exhibited mild symptoms of psychological distress but these had largely resolved at 12-month follow-up. This is supported by Goldbeck (2006) who found that although parents experienced a high level of stress at the time of diagnosis there was a statistical trend toward increases in maternal family satisfaction and in paternal well-being within three months after onset of disease, independent of diagnosis type. They suggest that this may reflect different coping styles of fathers and mothers as mothers mostly have the role of primary caregiver of the child, they may experience persistent parenting stress with the ill child and may primarily utilise support from their spouse and relatives. Fathers often return to work several weeks after their child's diagnosis, and therefore they experience a greater distance to the medical field and possibly less disease specific parenting stress compared to mothers (Goldbeck, 2006) Tifferet et al. (2007) had similar findings in Israeli mothers of children with neurological conditions. They suggest that a decrease in stress over time may also be attributed to an increase in familiarity and predictability with respect to the child's illness and treatment, since these are known to decrease stress levels.

There is also evidence to suggest that parental levels of distress are related to the age of the child; Hilliard et al. (2011) found that parents of young children with diabetes report more stress than parents of older children with the same condition.

An alternate explanation to a reduction of stress over time could be that gradually parents gain more knowledge over time and move from the initial 'crisis' stage. Therefore the more "unknown" the condition the more stress is experienced.

1.3.9 The experience of uncertainty

Gudmunsdottir et al. (2006) suggest that psychological distress in parents is not a result of the actual life-threat of the disease, but instead a result of the unforeseeable changes the disease brings for the parents and the family. Uncertainty is something that permeates much of the reported parental experience of managing childhood chronic illness. Before a child receives a diagnosis of a specific chronic condition, it is perhaps understandable that parents experience a huge amount of uncertainty and stress (Nolan, 2006). Diagnosis itself can be experienced as a shock and the parents' world is "altered irrevocably" (Ware and Raval, 2007; pg 554).

Uncertainty can also remain after a diagnosis is made (Swallow, 2001). This continuation of uncertainty is reflected in a quote from a father interviewed by Katz (2002): “I am a prisoner of the illness and the terrible uncertainty of the future.” (pg 11). Ongoing uncertainty regarding the prognosis of their child has been found in parents of children with Chronic Pain, Juvenile Rheumatoid Arthritis, Eczema, Heart disease, Diabetes, Epilepsy, Cancer, Downs Syndrome, Cerebral Palsy, Spina Bifida and Cystic Fibrosis (Van Dongen-Melman et al. 1995; Jordan et al. 2007; Dodgeson, 2000; Britton, 1999; Hummelink, 2006; Jessup and Parkinson, 2010; McNeill, 2004). Both McNeil (2004) and Goble (2004) report that fathers worry about what the future holds for their chronically ill children, and whether they are going to be able to continue looking after them into adulthood. Sultana et al. (2007) found that mothers of children with diabetes had concerns about their child’s dependence on medication and their marriage prospects. Fawcett et al. (2005), in their qualitative interview-based study of families caring for children with chronic conditions in Hong-Kong and Scotland, describe how parents experience a huge amount of uncertainty and fear of the future. Similarly, Van Sheppingen et al. (2008) report that parents of children with the rare genetic skin disorder Epidermolysis Bullosa often expressed feelings of uncertainty about the long-term prospects for their child’s illness, for example one mother said: “You never know how bad it’s going to be (. . .) You always carry that fear with you: will she get worse or will it just stay like this?” (pg 550). However, much of this research is based on small, non-representative samples and is cross sectional in nature, making it difficult to generalise to parents of children with other conditions, and to see wether the experience of uncertainty changes over time.

Uncertainty for the future prospects of their children was echoed in research by Liem et al (2011). They report that parents of children with Thalassemia were worried about their children’s ability to find happiness in marriage, have families of their own and achieve independence as adults. Ware and Raval (2007), in a qualitative study focusing exclusively on the experiences of fathers of children with chronic illness note how, for these fathers, the world seemed dangerous and uncertain and they often felt a lack of control and fearful. Jessup and Parkinson (2010) encapsulate the experience of the uncertainty and the unknown in their description of parents managing Cystic Fibrosis in their child: “When the initial tumult abated, families found themselves washed up in parts unknown, cut off from the mundane and familiar, and needing to find their bearings and reconstruct their lives in the context of a new, unanticipated scenario.” (pg 355). Even parents of children who would be classified as “recovered”, in this case from cancer, continue to feel uncertain about the well-being of their children (Van Dongen-Melman, 1995).

Sharkey (1995) suggests that uncertainty evolves from onset of learning about the child's illness. Parents who she interviewed repeatedly said they had no knowledge of the language needed to express themselves during the initial stage of the diagnosis. A common experience was not only dealing with the uncertainty of the chronically ill child's future, but also with the uncertainty of the family's daily lifestyle. However, continual uncertainty in illness may be preferable to negative certainty because it can enable patients and their families to perceive multiple opportunities and increased flexibility when the illness is viewed as chronic and continual (Mishel, 1990, 1999). According to Mishel's (1990) Reconceptualization of Uncertainty in Illness Theory, the reappraisal of uncertainty as promoting a variety of possibilities may evolve over time and uncertainty can become a desirable cognitive state leading to positive psychological changes and personal growth. Indeed, among adult patients and families of children with a chronic illness, researchers have found that continual uncertainty could serve as a catalyst for positive psychological outcomes (Mishel, 1990, 1999; Parry, 2003; Stewart and Mishel, 2000).

There is further evidence for optimism about the future in the research literature. For example Mastroyannopoulou et al. (1997) report that many parents remained "remarkably optimistic" (pg 827) about the future, however this may reflect father's experiences rather than mothers as Mastroyannopoulou also found differences between mothers and fathers reports of impact. Indeed, Ventner (2011) reports that some of the fathers she interviewed felt there were positive aspects to the experience of having a child with chronic illness, with one father saying: "So it didn't start off positive but it's ended up enormously positive because the things that we've learnt on that journey" (pg 10). Mawn (1999) suggests that there can be a "pattern of hope and optimism in the face of uncertainty" (pg 210). Similarly, Britton (1999) found that over half of parents in her study talked about having hope both for the future and that the disease would eventually go away.

1.3.10 Loss

Parents have been described as experiencing grief responses to their child being chronically ill (Mawn , 1999; George et al. 2006; Jordan et al. 2007; Rouf et al. 2012). It is also documented that parents feel loss in terms of their ambitions and aspirations, often having to pursue flexible employment as opposed to the career they may have wanted (George et al. 2008), though there is also contrasting evidence in fathers of children with autism. Gray (2003) reports that most of the fathers interviewed felt that their careers were unaffected; indeed sometimes it was felt that their child's autism actually encouraged a greater commitment to work. However, this may reflect a trend for fathers to "escape" from illness, leaving mothers to cope alone (Gray, 2003).

Theories of grief are useful in understanding the experiences of parents who have "lost" their child to chronic illness. The literature suggests that there are at least two major schools of thought concerning the 'normal' grieving process following bereavement or significant loss. Theories that suggest that the process runs a natural course over time culminating in resolution are termed time bound theories, while an alternative perspective is offered in which a periodic resurgence of grief is proposed as the norm: the theory of chronic sorrow. These two perspectives will now be described.

Time bound theories of grief

Kubler-Ross, in her seminal 1960's work identified five stages of grief; denial, anger, bargaining, depression and acceptance. The belief that the experience of loss can be divided into stages is common to a number of grief theories, some of which suggest that there is a sequential progression through stages of grief, which vary in definition according to different theoretical perspectives (Hayes 1985, Kamm 1985, Clubb 1991, Worden 1995). Such stage or time bound theories about grief have usually been developed through work undertaken with those bereaved, and describe similar end stages such as acceptance (Kubler-Ross 1969) and resolution (Engel 1962). This implies that grief is a common linear process for everyone. However, writers such as Worden (1995) suggest that the experience of grief is unique to the individual in their particular circumstances. Therefore, each person will pass through stages of grief in a unique manner and not necessarily in any predetermined order (Speck 1978, O'Connor 1986, Cook and Phillips 1989, Clubb 1991, Taylor 1995, Worden 1995, Coles 1996). Further, Worden (1995) suggests that the individual does not make the transition between these stages passively, but is an active performer of four basic grief tasks which he terms acceptance of the reality of the loss, working through the pain of grief, adjustment to the environment of the loss, emotional relocation of the deceased (the loss) and moving on with life. Worden considers that if these tasks are not achieved, the grieving process will not be complete. Thus, time bound theories propose that the stages of grief normally culminate in a state of acceptance or resolution. Failure to reach this state is seen as an abnormal response (Teel 1991).

Chronic sorrow: an alternative perspective

In contrast to time bound theories of grief, some authors who have studied chronic illness suggest that failure to achieve resolution or acceptance is not ‘abnormal’ (Dashiff 1993, Murgatroyd and Woolfe 1993, Hainsworth et al. 1994, Tinlin 1996). It has been proposed that grief may be perpetual, with periods of remission and intensification of grief symptoms (Brown 1985, Jansen 1985, Clubb 1991, Murgatroyd and Woolfe 1993, Tinlin 1996). This has experience been described as “chronic sorrow” (Olshansky, 1962). The theory of Chronic Sorrow has been described as providing a framework for understanding and working with people following a single or ongoing loss (Eakes et al. 1998). Olshansky observed that parents of children with cognitive impairment experienced lifelong, episodic sadness. He proposed that parents never recover from the initial impact, that feelings of guilt are never eliminated and that, although parents adjust and adapt to the situation, these efforts do not represent acceptance. Subsequent research has validated the occurrence of chronic sorrow among parents of mentally or physically disabled young children and expanded the emotions commonly experienced to include not only sadness and sorrow, but also fear, helplessness, anger, frustration, and other feelings characteristic of grief (Burke, 1989; Damrosch and Perry, 1989; Fraley, 1986; Hummel and Eastman, 1991; Phillips, 1991; Seideman and Kleine, 1995; Wikler, Wasow, and Hatfield, 1981). However, there is contrary evidence looking at parents of children with HIV (Mawn, 1999)

1.3.11 Chronic Illness: destructive vs restorative force

Within the research literature there is evidence to suggest that childhood chronic illness can serve as both a destructive and a restorative force, sometimes weakening family relationships and sometimes strengthening them. Increased stress, strain and tension seems to be common for families in this situation (Van Sciver et al. 1995; Thyen et al. 1998; Moore et al. 2013). Van Sheppingen et al. (2008) found that in some cases the child's chronic illness was cited as a major factor contributing to divorce, and even the mothers who were not divorced reported feeling like they were growing apart from their partner (Van Sheppingen et al. 2008). One limitation to this evidence is that the research is cross-sectional. Longitudinal research may be more useful for drawing conclusions regarding the destructive impact of childhood chronic illness. in addition, much of the research is based in self reported questionnaire data, which is subject to recall bias.

Eddy (1999) suggests that perhaps there is a tendency to view child health status as having uniformly negative effect on marriage because the data often come from families involved in some type of clinical services. She asserts that families who seek services are likely to differ from the general population. While this may be true Eddy seems to be suggesting that families who do not seek services may be more self sufficient, however an alternative explanation may be that the families who do not seek services may be experiencing even more problems.

In interviews with 53 parents of children with autism, Gray (2003) found that, in some cases, the child’s problems had led to direct confrontations between the mother and father and threatened their marriage. Gray also reported that some mothers seemed to resent their husbands because they were able to escape from illness regularly by going to work. Similarly, Fawcett (2005) reports that the burden of childhood illness frequently manifests in parental conflict within the marriage. In their interviews, parents talked about arguing more and feeling an “increase in pressure” (pg 13). Parents of children with severe epilepsy described a significant negative impact on their relationship with their spouse (Nolan et al. 2006). Berge et al. (2006) used the Family Adjustment and Adaptation Response (FAAR) model as a theoretical framework to explore marital satisfaction and mental health of couples with children who had chronic health conditions. They found that for mothers, the severity of their child’s condition predicted the level of marital satisfaction and depressive symptoms. Essentially, their marital satisfaction tended to be lower and depressive symptoms were higher when their child was more unwell. They found that although fathers might also report depressive symptoms and low marital satisfaction, this was not influenced by the severity of the child's condition. However, generalizability of the findings is limited because the sample consisted of parents of children aged 12 to 30 months. It could be argued that maternity leave during this time means mothers tend to spend more time with their children than fathers. Mothers may therefore attribute more of what they feel to their caregiving responsibilities. Perhaps this reflects the fact that mothers tend to be the primary caregiver and therefore experience more of the day-to-day struggle of chronic illness.

Although there is evidence to suggest that childhood chronic illness has a negative effect on marriage, there is also evidence to the contrary. For some parents of children with chronic conditions, compared to parents of healthy children, there is no difference in marital quality and stability (Eddy, 1999; Cappelli et al. 1994) or family functioning (Zashikhina and Hagglof, 2009; Rodrigues and Patterson, 2007). Rodrigues and Patterson (2007) suggest that this might be because the presence of a stressor like having a child with a chronic condition challenged these families to develop internal strengths for managing their situation, perhaps leading to stronger family functioning. However, with these studies, especially Rodrigues and Patterson (2007), it is likely that there was a self-selection bias, wherein higher functioning families were more likely to agree to participate in a longitudinal study of this kind. In addition Rodrigues and Patterson did not include single parent families, whereas Zashinka and Hagglof did and found that these families were at risk of family dysfunction. Eddy (1999) conclude that having a child with chronic illness or disability has no uniform impact on marital quality or on perceived marital stability, however, results from this study should be treated with caution as they only asked two questions regarding perceived marital stability and quality. It is surprising that such conclusions were drawn from such a small amount of data, and also it is not clear who asked parents these two questions, this is important as it may have had an impact on what parent's responses were. Although parents may report that the impact on the family is minimal their children may report otherwise; Interestingly Zashikhina and Hagglof (2009) found that there was a significant difference between mothers' and adolescents' ratings in all aspects of family functioning but conflict. Essentially adolescents perceived their families as less competent, less cohesive and less emotionally expressive compared to their mothers.

There is evidence to suggest that childhood chronic illness can make families stronger. Gabor and Farnham (1996) found that despite many ups and downs, low income single mothers of children with chronic illness reported energy to grow and share, which is in contrast to other findings that single parent families are at risk of family dysfunction (Zashikhina and Hagglof, 2009). Interestingly Gabor and Farnham's research was longitudinal, whereas Zashikhina and Hagglof's was cross sectional, suggesting that over time the destruct force can evolve into a restorative force. Chernoff (2001) found that 80% of mothers of children with chronic conditions (diabetes, asthma, sickle cell disease and cystic fibrosis) felt that their family had benefitted in some way from the experience (for example they talked about becoming better parents; of family members looking out for each other; of becoming more aware, sensitive, and tolerant; and of being more confident and emotionally stable), however this finding was based on data from only 3 questions; "learning to manage my child's health condition has made me feel better about myself", "my family is stronger because of my child's condition", and "Some parents say that, despite a lot of challenges, there are also benefits in raising a child with [condition]. For example, some parents say that their child's condition has made them stronger as parents. Do you think you or your family has benefited in any way from [your child's condition]?", in addition there was a very low response rate, 566 families were contacted, though only 190 responded. It could be suggested that the 3 questions asked were quite leading, and also it would be interesting to see what impact the non-responding mothers' felt the illness had had on them. Fawcett et al (2005) reports that the parents in their study identified rewards gained from caring for their child, though it is not reported exactly what these rewards were. Goble (2004) reports a strengthening in committed relationships related to chronic illness, however, this is from the fathers' perspective, and there is evidence to suggest that the mothers' view on this differs considerably. Jessup and Parkinson (2010) observed improved relationships between parents and children with cystic fibrosis, though this was a small sample of 12 parents. Similar findings have been observed in parents of children with chronic pain (Jordan et al. 2007), and fathers of children with life limiting illnesses (Ware and Raval, 2007), though again both of these studies had very small sample sizes. Gannoni and Shute (2010) found that parental confidence increased among parents of chronically ill children. Although most research is with mothers McNeill (2004) describes how fathers of children with juvenile rheumatoid arthritis frequently spoke of their child's condition as a spring- board that helped them to appreciate the priorities in their life and to put them into perspective. In particular, one father is reported as saying: "I think it can strengthen everything, it can strengthen marriages, it can strengthen families . . . because you get a better perspective on what's really important in life. You really do." (pg 537). Similarly, Ware and Raval (2007) found that fathers identified caring for a child with a chronic condition had changed them for the better, reporting a sense of personal growth.

Fiese and Wamboldt (2003) found contrasting evidence that while for some families chronic illness, in this case asthma, could be a strengthening force drawing family members together and fostering feelings of security, for other families it provokes conflict and avoidance of close relationships. Similar experiences were reported by Hopia et al. (2005) who found that some parents felt anger and bitterness and did not want to accept that their child was unwell, while for some parents the child's illness helped to bring family members closer together and made them feel stronger regarding perceived future difficulties. They report one mother saying: "When we got over the shock stage, I got a strong idea that when we survive over this, we will be a thousand times stronger" (pg 191). This is echoed by a mother's account of dealing with her child's cystic fibrosis as reported by Jessup and Parkinson (2010); "It's probably made me a stronger person...the one I am today" (pg 358). Goble (2004) reports that fathers in her study actually felt closer to their wives in some ways but also had to deal with a lack of intimacy. Lack of intimacy was also reported by Missen (2012) in mothers of children with CFS/ME.

Mothers' and fathers' reports differ with regards to the impact of childhood chronic illness; Mastroyannopoulou et al. (1997) report that fathers were more likely to feel that their marriage had remained the same or improved, whereas mothers were more likely to feel that it had become more negative. Gray (2003) found that although fathers of autistic children noted the severe difficulties that their child's autism presented for their families, they usually claimed that their child's condition did not have a significant effect on them personally. In contrast, mothers would be more likely to claim that their child's autism had severely affected their emotional wellbeing. This may reflect the fact that mothers tend to be the main carers for unwell children (Britton, 1999; Gray, 2003; Timmermans and Freidin, 2007), therefore spend more time with the child, and thus may experience more of an impact.

There is clearly contrasting evidence that some families continue to function well when a family member has a chronic illness (Bohachick and Anton, 1990; Donnelly, 1994; Rehm and Catanzaro, 1998; Sawyer, 1992; Youngblut et al. 1994), while other families experience negative outcomes for family functioning (Cornman, 1993; Ferrell et al. 1994; Kopp et al. 1995; Park and Martinson, 1998). Gustafsson et al (2002) suggests that the difference between those families that are weakened by chronic illness and those that are strengthened by it may be that some families are “less cohesive" prior to diagnosis and therefore have less strict patterns of family function, which would mean that the family might show a more "laissez faire attitude" to handling problems. They suggest that "such a lifestyle might be related to fewer psycho-social problems in the family.” (pg 1074). The Resiliency Model of Family Stress, Adaptation and Adjustment (McCubbin and Patterson, 1987) helps to explain why some families succeed in coping while others will move into crisis. Friedman, Bowden and Jones (2003) describe The Resiliency Model as a way to assess the stressors, family coping, and how the crisis has disrupted family functioning. The main emphasis of this model is on resiliency of families and their ability to recover from adverse events, and what strengths influence this process. It consists of two phases, the Adjustment Phase and the Adaptation Phase. The Adjustment Phase is the phase in which families try to maintain the status quo in the face of a stressful event. Multiple stressors during this time make it harder for the family to cope. Pre-existing strains in the family also make it harder to cope with current stressors. Family types and established patterns of functioning are important in the adjustment phase. Parents of children with chronic disease who receive adequate social support generally perceive that they have less stress in their lives (O’Brien, 2007; Sterling, Jones, Johnson and Bowen, 1996) and are important aspects to ongoing adjustment. Outcomes of the adjustment phase are either effective adjustment or ineffective adjustment. If family functioning is not adequate to manage stressful events such as a family illness, the family moves into a crisis situation and the Adaptation Phase of the Resiliency Model of Family Stress (Friedman et al. 2003).

1.3.12 Support

Martinson et al. (1995) suggest that maintenance and use of social relationships can help mothers cope with their child’s chronic illness. Hatzmann et al. (2008) suggested that for parents to cope with chronic illness it is essential to set up an adequate support system to derive emotional support and share child care, though their research is skewed towards mothers of a high educational level. Frankel and Wamboldt (1998) found that mothers who showed high levels of emotional distress and low levels of perceived support report experiencing the child's illness as disruptive and having a major impact on family life. This is supported by findings from Tak and McCubbin (2002) and Wong and Heriot (2007); Tak and Mcubbin (2002) found that, among 92 families with a child under 12 newly diagnosed with congenital heart disease, high levels of perceived social support were related to improved parental coping, while Wong and Heriot's (2007) study of 35 parents of children with Cystic Fibrosis concluded that parents who receive high levels of emotional support are likely to experience lower levels of distress. However, Wong and Heriot's sample were all already members of a support group for parents of children with cystic fibrosis which would suggest that they were already drawing from a cohort who would value emotional support. In addition to this the sample was also mainly mothers, which is true of much of the research evidence, would social support be valued as highly among fathers?

There is evidence to suggest that single mothers have lower levels of family support compared with two parent families (Thyen et al. 1998). It is therefore perhaps unsurprising that the evidence suggests a strong support system is invaluable to single parents (Gabor and Farnham, 1996; Gates, 2012)

Kazak et al (1997) suggest that higher levels of perceived social support is associated with lower levels of posttraumatic symptoms for mothers and fathers, however Gudmunsdottir et al. (2006) found that although lack of social support at the time of diagnosis predicted greater impact of the disease on the family, the lack of social support did not predict parental distress or more trauma symptoms. However, this study is limited by a very low response rate of 18.4% (105 out of 571 parents returned questionnaires).

1.3.13 Isolation

Research suggests that feelings of isolation are common among parents of children with chronic conditions (e.g. Mailick et al. 1994; Martinson et al. 1995). Eddy and Engel (2008) suggest that families who have ongoing concerns about their child's physical or emotional well-being or are unable to participate in normal family activities may feel isolated. Mastroyannopoulou et al. (1997) report that support from relatives and friends is often lost. They suggest that this is because families may be *"regrouping internally"* and not relying on outside support systems. This suggestion makes it seem that parents are choosing to cut themselves off from the outside world, however, there is evidence to the contrary: Hopia et al. (2005) found that parents of chronically ill children feel lonely and desperate for company of other adults, and that they felt "completely alone with the child" (pg 191). This is also highlighted by Timmermans and Freidin (2007) who, in their study of 50 parents of children with asthma, report that there was a feeling amongst the parents of having to cope alone, one parent is reported as saying: “I don’t have anyone who helps me. Who could help me? There’s no one, it’s hard to get help” (pg 1355).

Isolation may also be caused by the perceived stigma of a chronic condition; Chao (1997) found that 83% of parents of children who were chronic bedwetters felt that they would be stigmatised if they openly discussed their child’s problem with others, however, this may also reflect cultural differences; Chao’s research was conducted in Singapore, and there is a suggestion that it is not acceptable to talk openly about these issues in Asian societies. Mawn (1999) explored parents’ experiences of caring for a child with HIV and reported that parents avoid close relationships/friendships for fear of how others will react to their child’s condition. Similarly Ware and Raval (2007) report that fathers of children with chronic conditions often had feelings of alienation and abandonment, highlighted by one particular father: “We found that some people deemed the family as a problem family and it was best to put as much distance between themselves and us as possible, which was quite common actually,” (pg 555). Sometimes parents of chronically ill children can experience insensitive and hurtful comments which makes them feel further isolated (George et al. 2006) though this seems to be reflected more for mothers than fathers. Rouf et al. (2012) suggest that the concern that mothers of children with severe food allergies have about negative social evaluation could potentially increase a sense of social isolation. Rouf states that one mother felt she would be labeled as a “right crazy mother” and “overprotective”. However, it is interesting that this is a specific maternal fear, rather than a paternal one. Rouf comments about the fact that fathers did not seem to participate, and that this is suggestive of gender differences in the experience of parenting younger alergic children. Researchers often make the assertion that parents are isolated, however much of the research evidence suggests that mothers tend to provide the majority of care for unwell children (Britton, 1999; Gray, 2003; Timmermans and Freidin, 2007), therefore perhaps isolation is a problem for mothers but not necessarily for fathers. Some research suggests that fathers are able to escape to the world of work, which may mean that they are not as isolated as mothers. In a study exploring the experiences of 10 families attempting to combine work and care for chronically ill children, Ventner (2011) found two distinct types of families: those who conformed to traditional gender arrangements of the mothers’ role as the primary carer and the fathers working full time, and those with “egalitarian care arrangements” where both parents were involved in caring for their child. Gannoni and Shute (2010) talk about complimentary parental roles and the importance of the father’s role. This has also been supported by other research (Franklyn and Rodger, 2003; Gray, 2003). there is also evidence that fathers sometimes become the primary caregiver to healthy siblings (Goble, 2004). Gavin and Wysocki (2006) have found an association between fathers' involvement in day to day management of illness and healthier functioning mothers, marriages and families. Greater paternal involvement in the medical regimen might enhance marital communication and satisfaction with the partner. Mothers who receive more help may experience less role strain, and may experience less frustration with their partners. Succeeding at such a challenge might have the effect of drawing the parents closer to one another, further enhancing their marital functioning and satisfaction.

Alternatively, it is important to consider that existing levels of marital and family satisfaction may influence spouses' perceptions of paternal involvement. For example, a wife experiencing high marital satisfaction might be more likely to perceive her husband as more involved and helpful. In reality, couples enjoying high marital satisfaction are likely to view each other more positively and be more prepared to collaborate effectively in complex disease management tasks (Gavin and Wysoki, 2006).

Gabor and Farnham (1996) interviewed 5 mothers of children with special health needs[4]. They found that a lack of understanding or acceptance from the extended family could lead to feelings of isolation. One mother is reported as saying: "It kind of freaked my family out. They're used to having healthy, bouncy, annoying kids, you know, and tons of them around them, so to have one that can't communicate, who has some physical and mental disabilities, they just shied away for about the first 18 months." (pg 175). Brewer et al. (2007) report how mothers of children with Huntingtons disease feel isolated because, after a while, other people lose interest in the illness. This is also a parental experience reported by Ware and Raval (2007). Fawcett et al (2005) attributed isolation to friends and family not knowing how to deal with the child's condition. Similar experiences are reported by Gannoni and Shute (2010) in their interview based study of parents of children with cancer, chronic renal failure and type I diabetes; they describe how a lack of understanding and acceptance by others can make families feel isolated. This lack of understanding and acceptance can sometimes stem from the fact that, outwardly, the child may not look unwell (which incidentally is often a particular issue with people with CFS/ME). Ware and Raval (2007) found this when interviewing fathers of chronically ill children; "But people couldn't understand how we had a perfectly healthy 3-year old running around

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[4] Diagnoses included severe bronchopulmonary dysplasia, intracranial hemorrhages, seizure disorders, and cerebral palsy

and we were just beside ourselves" (pg 554). Isolation caused by a lack of understanding and acceptance is also true for parents of children with chronic pain (Jordan et al. 2007) or Thalassaemia (Kumari, 2003; Liem et al. 2011). This is also highlighted by Sharkey (1995) who found that some parents did not even try to get support from friends and family because they felt that they would not understand. One mother is reported as saying: "I couldn't just call a friend and say I am really bugged that Jessie might die or something because they just wouldn't understand what was going on. Talking about her always seemed to make any of my so-called friends uncomfortable" (pg 40).

There is a suggestion that lack of awareness and knowledge of some chronic conditions can lead to parents feeling isolated. In their study of the impact of Thalassemia on Asian families Liem et al. (2011) suggest that the misconceptions held by family and friends can lead to isolation and a general feeling of a lack of support. Parents will then avoid talking to others about their child's condition and do not seek further support. Gannonni and Shute (2009) report interpersonal communication problems about cancer, chronic renal failure and type 1 diabetes with extended family members and school personnel. This lack of awareness and knowledge may be particularly relevant in the case of CFS/ME, as it tends to be a condition that courts controversy (see section about controversy in CFS/ME at the end of the literature review for more information).

1.3.14 Restriction

The literature suggests that having a child with a chronic illness restricts the lives of parents, and that they often feel that their social life becomes limited or non-existent (Thyen et al. 1998; Hunfield, 2001; Goble, 2004; Jordan et al. 2007; Van Sheppingen et al. 2008; Knapp et al. 2010). Goldbeck (2006) reported that parents had restricted time to be with their partner, friends or acquaintances, or to develop their own personal interests, though this was a small sample of parents with recently diagnosed children, perhaps reflecting that this particular time can be quite restrictive Also, Goldbeck uses a self report questionnaire, the findings are therefore dependent on the reliability and validity of self report data. Martinson et al. (1995) found that almost half (41%) of a sample of 75 mothers of children with chronic illnesses[5] stated that they were unable to leave the house because they would feel uneasy if they went out. In Gustafsson et al's (2002) study of 139

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[5] Asthma (33.3%); Renal-kidney problems (28%); Cardiac problems (24.2%); cancer (14.5%)

families with an asthmatic child, 36% thought that contacts with friends and relatives had decreased and 30% often had to make sudden changes in appointments, which negatively affected their relationships with friends and relatives. There is also evidence to suggest that families caring for a child with a chronic illness are unable to go on holiday (Martinson et al. 1995; Gustafsson, 2002; Van Sheppingen, 2008).

1.3.15 Financial burden

There is evidence that childhood chronic illness presents a substantial financial burden to parents (Martinson et al. 1997). One would assume that the financial impact of childhood chronic illness would be related to the heath care available. Health care provision differs depending on the country, some countries offer a form of universal health care whereby a health care system provides health care and financial protection to all it's citizens.

The USA does not have a universal health care system, and health care provision is linked to health insurance provided by employers, or privately purchased. Garwick (2002) reports that even while controlling for family income, both fathers and mothers of preadolescents with uncertain life expectancies reported significantly greater financial burden than parents of preadolescents with normal life expectancies, which is understandable because if a parent has to give up work they would lose some of their health insurance provision and income, and potentially have to pay more for private health insurance.

Although China now has a form of universal health care, this was not the case up until the end of the 1990's. Martinson et al. (1995, 1997) conducted two studies of Chinese parents of chronically ill children. Findings from the first study (1994) suggested that mothers' primary concerns about the child's illness related to obtaining care and bearing the financial impact of the illness. In the second study (1997) it was found that mothers were more likely to experience changes in employment (53%) than fathers (16%). Both studies suggest that financial concerns related to a child's chronic condition can significantly increase parental stress levels, which in turn can make parents "less capable of meeting their child's health needs" (Martinson et al. 1995, pg 374). One limitation to both of Martinson et al's studies is that because both samples are predominantly made up of mothers[6] the evidence provided gives a one sided view of the impact on parents. Research focusing on fathers tend to be quite small scale, for example Goble (2004) found

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[6] 1994 = 100%, 1997 = 77% mothers, 11% fathers, 13% other family member

evidence of financial burdens in a sample of 5 fathers of children with cerebral palsey, autism and osteogenesis Imperfecta in the USA. They found that fathers reported great financial strain particularly because their wives had had to give up work to care for their unwell child. Kumari (2003) reports similar findings in mothers of children with Thalassaemia in Sri Lanka, who lost their jobs because of regularly taking time off to care for their unwell child. One could argue that having to give up work to care for an unwell family member would exert a greater financial strain on the family when there is no universal health care available.

However, the research evidence suggests that there is a financial burden for families regardless of wether the country has universal health care or not. Virtually all of Europe has either publicly sponsored and regulated universal health care or publicly provided universal healthcare. In Sweden, Gustafsson et al. (2002) report a negative impact of childhood asthma of family expenditure, with over half of parents in the sample (52%) attributing this to direct medical expenses. In Germany, Thyen et al. (2003) found that financial strain increased with age of the child. In England Missen et al. (2012) reports the financial burden on families managing a child's Chronic Fatigue Syndrome. Further afield, Australia has a form of universal health care and Gannoni and Schute (2010) found that parents across three illness groups (cancer, chronic renal failure, type I diabetes) noted treatment-related financial concerns such as the cost of special food items, as well as difficulties associated with hospital appointments, including maintaining employment, traveling long distances in some cases, making alternative care arrangements for siblings, and costs of travel. This finding is supported by a more recent study of parents of children diagnosed with cancer in Singapore (also providing universal health care); Aung (2012) found that 61% of families had financial assistance. Limburg (2008) found that 64% of mothers compared to 16% of fathers left their job after their child was diagnosed with cancer. However Limburg et al. also report that although many parents temporarily leave their jobs after their child's diagnosis, the large majority (80%) were able to return to work when they chose to.

1.3.16 Summary

There is very little research into the impact of CFS/ME on family life. Research into other chronic childhood conditions suggest that parental physical and emotional health can be adversely affected. There is evidence to suggest that perceived severity of illness is a factor in the perceived impact on parents. However, much of this research is cross sectional. Longitudinal research suggests that there may be a reduction of the initial adverse affects on parents. Research suggests that mothers and fathers cope with childhood chronic illness differently. Universal experiences of parents managing childhood chronic illness include uncertainty, loss isolation and restriction. The literature reflects that chronic illnesses can serve as both a destructive and a restorative force to families, and in particular that it can be a tremendous financial burden.

1.4 Limitations of previous research

It is often reported that mothers experience a greater negative impact in terms of higher levels of distress (e.g. Yildiz et al. 2009), Yildiz et al. (2009) suggest that this disparity could be due to mothers being more actively engaged in their child's care than fathers, more often in communication with their children and spending much more time with their children. However, one notable issue is that mothers are over represented in the research, and this gives a skewed perspective on the impact on parents. There is evidence to suggest that there are no significant differences in mothers' and fathers' experience of managing childhood chronic illness with regards to the degree of uncertainty in the child's life expectancy and family distress (Dodgeson, 2000). However, Dodgeson (2000) was looking at parents of recently diagnosed children, and although at this point there were no significant differences, over time differences may develop and become more pronounced. An alternative explanation could be that mothers and fathers deal with the impact of childhood illness in different ways; For example, Mastroyannopoulou (1997) found that fathers tended to cope with their child's chronic illness by withdrawing while mothers coped using emotional release. Similarly Gray (2003) found that fathers of children with autism were more emotionally reserved than mothers. This is supported by findings from McNeil (2004) who reports that although fathers of children with juvenile rheumatoid arthritis experience strong emotions, they are often reluctant to express the depth of their feelings openly because they feel that they should be the strong one in the family. In their study of fathers of children with life limiting illnesses Ware and Raval (2007) had similar findings, they report that all 8 of the fathers they interviewed perceived men as not allowing themselves to explore their emotional responses in the same way as women do. They found that this was often attributed to societal expectations of masculinity. One father is reported as saying: “we are always taught from school to be very stoic and all the rest of it, and we don’t talk about issues or problems or anything else, and I think probably that works very much against us and that is a cultural thing.” (pg 557).

There is often a suggestion that mothers should receive more support (e.g. Frankel and Wamboldt, 1998). While there is no doubt that mothers should be supported, research that does include fathers has found that they may also experience a negative impact of childhood chronic illness; Dalheim-Englund et al. (2004) found that fathers of children with asthma younger than 13 had lower quality of life scores than fathers of older children. Garwick (2002) found very little difference between mothers and fathers with regards to the levels of family distress reported. It has been suggested that the reason mothers are over represented in this type of research is because mothers spend more time with the unwell child/children and thus fathers feel hesitant to take part (Goble, 2004). It may also be that if fathers are having to work more to support the family, as suggested by some research (Goble, 2004), then they could be too busy to take part in research, or it may be that research takes place when fathers are at work. A common theme noted in many of the studies reviewed was the need for further research focusing on fathers and how they are affected by their child’s chronic illness.

1.5 Rationale for the current research

The NICE guidelines for treatment of CFS/ME (2007) highlight the importance of engagement with the family when working with children with severe CFS/ME, and the guidelines published by the Royal College of Paediatrics and Child Health (2004) also advise developing a rapport with the family and establishing a cooperative and empathetic relationship. Although these guidelines are evidence based, the available evidence focuses on the treatment of the individual with CFS/ME. Indeed, Rangel et al. (2005) have identified that although clinicians working with childhood CFS/ME have noted disruption to family life, there is hardly any empirical evidence. Although Rangel et al (2005) explored the impact of CFS/ME on family health using both questionnaires and semi-structured interviews, data were analysed quantitively and there was no deeper qualitative analysis.

Outside of CFS/ME research there is a growing body of evidence demonstrating that families can have a beneficial or harmful effect on a family member's health (Campbell, 1986; Campbell, 2003; Campbell and Paterson, 1995; Doherty and Campbell, 1980). Children who experience high family stress have been shown to be at higher risk for infections and hospitalization (Beautrais et al. 1982). Illness can then in turn exacerbate the dysfunctional traits of a family, and effectively create a negative cycle.

The paucity of empirical evidence of the impact of CFS/ME on family life, and the findings of previous research into different chronic illnesses help to build a strong rationale for the current research study. It is vital to gather evidence of the impact of CFS/ME on the wider family unit in order to inform clinical practice. By seeking to understand the wider impact of CFS/ME on parents this research will have implications for support and treatment of the young person, inform future service development and help to expand the current individualistic model of care to possibly include support for parents as well as children. The aims of the current research are therefore to investigate the impact of having a child with CFS/ME on family relationships, from the perspective of parents, and for this information to help to inform future service provision.

1.6 Influence of a Key Paper

The idea for this study was originally conceived having seen the findings of previous research investigating the psychological health of mothers of children with CFS/ME and the financial impact of their child's diagnosis (Missen et al, 2012). This was the first, and to date, only study to demonstrate the financial impact on parents of children with CFS/ME. This research adopted a mixed method approach utilising questionnaires and semi-structured interviews. 40 out of 50 mothers recruited into the study returned questionnaires (the Hospital Anxiety and Depression Scales, the General Health Questionnaire-12, a Loss of Earnings inventory) and 8 out of 50 mothers were interviewed. Through quantitative analysis it was found that mean loss of income for these families was £247 per month and it was suggested that income loss for parents of children with CFS/ME was equivalent to more than 10% of the mean income and expenditure of the general population. Financial concerns have been highlighted by other research in the area (Gustafsson et al, 2002; Gannoni and Shute, 2010). Thematic analysis was used to construct 5 themes: lack of understanding from others; marital tension; child's anxiety and distress; impact on siblings and maternal emotional distress. These represent the negative aspects of family life when dealing with CFS/ME and are consistent with previous research into the impact of childhood chronic illness on families. Of particular interest was the theme of marital tension, which Missen describes as the "broadest and most widely discussed". Mothers felt there was a direct link between their child's CFS/ME and relationship difficulties.

1.7 Aim

The aim of the current study was to explore parents' individual and couple experiences of having a child with CFS/ME, with a particular focus on their perceptions of its impact on family relationships. An additional aim was include fathers' perspectives in addition to mothers'.

CHAPTER 2: METHODOLOGY

CHAPTER OVERVIEW

The following chapter details the design, method and procedure used in this study. First, a detailed consideration of the principles of qualitative methodology and Thematic Analysis are provided alongside a rationale for the chosen methodology. The process of ensuring quality and a consideration of ethical issues are also described.

2.2 Qualitative Methodology

2.2.2 Principles of Qualitative Methodology

Qualitative methodologies are increasingly being utilised in research to understand the complexity of human behaviour (Draper, 2004; Elliott *et al*., 1999). This increase reflects a shift away from the more traditional quantitative method of inquiry. In regards to philosophical basis and research aims, quantitative and qualitative methods differ significantly. Quantitative research is rooted in positivism and aims to test hypotheses and establish cause and effect through the collection and analysis of numerical data. In contrast, qualitative research is broadly rooted in the interpretative tradition and aims to describe and explain social phenomena as they occur in their natural context by collecting and analysing narrative data (Draper, 2004). Qualitative research is concerned with the 'quality and texture of the experience... how people make sense of the world and how they experience events' (Willig, 2008, p. 8).

In quantitative research, hypotheses inform the direction of the study. This is known as the hypothetico-deductive method (Willig, 2008). However, qualitative research is guided by provisional research questions. Such questions identify the phenomenon under investigation and guide the way in which research is conducted without predicting the findings (Willig, 2008). Commonly open ended questions which elicit detailed descriptions of phenomena under investigation, are utilised to explore these research questions (Draper, 2004). Semi-structured interviews appear to be the most widely used data collection tool in qualitative research with other methods, such as observation, focus groups or diaries used to a lesser extent (Willig, 2008).

2.2.3 Rationale for Qualitative Methodology

A qualitative methodology was deemed appropriate for the current study for several reasons. Primarily, this was because, as outlined in Chapter One, there is a limited knowledge base within this field. Thus, a qualitative approach, which seeks to facilitate an exploration into phenomena on which relatively little is known, was considered more appropriate than a quantitative methodology which aims to test out preconceived hypotheses based on the existing literature (Stern, 1980). Furthermore, as the study was concerned with parents' *experiences* it was considered that quantitative methods would not enable the researcher to capture the phenomena easily or explore it at sufficient depth. Qualitative methodology is ideal to make sense of, or interpret, phenomena in terms of the meanings people bring to them (Denzin and Lincoln, 1994). Yardley (2000) points out that a primary reason for adopting a qualitative methodology is a recognition that our knowledge and experience of the world cannot consist of an objective appraisal of some objective reality, but is profoundly shaped by our subjective and cultural perspective, and by our conversations and activities (Yardley, 2000). It seems apt, therefore, when considering how family life is affected by having a child with CFS/ME to adopt a methodology that will enable the researcher to "tap into" the families' lived experience, to understand and represent the experiences and actions of people as they encounter, engage, and live through situations (Elliot et al. 1999). Therefore Thematic Analysis, a "flexible method" allowing a researcher to "focus on meaning across a dataset" (Braun and Clarke, 2012), was considered appropriate. This particular research method is ideal for research that is "experimental and exploratory" (Braun and Clarke, 2012). Thematic analysis was chosen because it is not "wedded to a theoretical framework" (Braun and Clarke, 2012) and therefore does not require the detailed theoretical and technological knowledge which allows for flexibility at the analysis stage. Thematic analysis was chosen as a method of analysis as opposed to IPA or Grounded Theory because although IPA and Grounded Theory seek patterns in the data, they are theoretically bounded. Although Thematic analysis is not "wedded to a theoretical framework" (Braun and Clarke, 2012), it is not "theory-less"; it can be carried out from various theoretical positions. Thematic analysis is perhaps best understood as being on a theoretical spectrum and can be essentialist/realist, or constructionist. Braun and Clarke make clear that although there is flexibility with regards to the theoretical underpinnings of any thematic analysis, a researcher must make it clear to which theoretical position their particular thematic analysis is related to. For the current study the theoretical position taken by the researcher can be described as "critical realism", which acknowledge the ways individuals make meaning of their experience, and, in turn, the ways the broader social context impinges on those meanings, while retaining focus on the material and other limits of 'reality' (Braun and Clarke, 2012). In critical realism, language is understood as constructing our social realities. However, these constructions are theorized as being constrained by the possibilities and limitations inherent in the material world (Sims-Schouten et al. 2007). The basic assumption of critical realism is the existence of a real world independent of our knowledge of it (Bhaskar, 1998). Within the current research, taking a critical realist stance meant that the researcher accepted Chronic Fatigue Syndrome as a “reality”; the assumption was that although all of the families were experiencing this “reality”, the language parents use would reflect differing interpretations of “reality”.

2.3 Thematic Analysis

2.3.2 Principles of Thematic Analysis

Attride-Stirling (2001) has described the process of using Thematic Analysis to seek themes in the text as akin to an Archaeologist seeking to “unearth” hidden treasure. However it is worth pointing out that the process of Thematic Analysis is a much more active process than this archaeological metaphor would suggest; Braun and Clarke (2012) suggest that far from the themes already existing and lying in wait for the thematic analyst to discover them, the analyst is instead like a sculptor, making choices about how to shape the stone (data) into art (analysis).

2.3.3 Thematic Analysis: an explanation

The interviews were analysed using a framework approach to Thematic Analysis (Braun and Clarke, 2006). Thematic analysis is suited to an inductive, data-driven approach. This method of analysis allows for greater flexibility, with both sampling and its allowance of identification of themes at a semantic level (Braun et al., 2006). This is in accordance with the idea that participants’ experiences can be accessed through their verbal accounts. A strength of this approach is that it is “a data, rather than theory-driven process, enabling the researcher to describe and summarize the data in its entirety rather than seeking only parts of the data that were deemed relevant” (Earle and Eiser, 2007,p 284). The interviews were analysed in order to identify issues important to the participants, rather than those thought to be important by the researcher.

The 6 phase approach

Braun and Clarke (2012) succinctly describe a 6 phase approach to doing Thematic Analysis: familiarising yourself with the data, generating initial codes, searching for themes, reviewing potential themes, defining and naming themes and producing the report. This 6 phase approach was adopted as a framework for data analysis within this study. An important point to keep in mind is that although this approach is made up of distinct phases the process of analysis is not a linear journey, one may move through phases only to start to develop a new insight into the data, it is therefore necessary to adopt an iterative approach.

1. Familiarising yourself with the data

Braun and Clarke refer to this phase as “immersing” yourself in the data. This was achieved by transcribing the data verbatim and then reading and re-reading the transcripts. Braun and Clarke emphasise that immersion is not a passive exercise, therefore the following questions were used to guide the initial exploration of the data:

How does this participant make sense of their experiences?

What assumptions do they make in interpreting their experience?

What kind of world is revealed through their accounts?

2. Generating initial codes

At this phase the transcripts were all read through again, and sections of text were labelled to describe and interpret what participants are saying, these label are called “codes”. Braun and Clarke describe codes as the “bricks” that will eventually be used to make the theme “walls” which in turn make the overall analysis “house”. Once the initial codes had been generated for the first 4 interviews, all of the codes were reviewed by an additional researcher. This process helped to increase the overall rigour of the resulting analysis , an important aspect of research quality.

3. Searching for themes

This stage began after completing the coding for all of the interviews. All of the codes were listed together, with reference to which participant and page they related to. The codes were then organised into groups of similar codes. At this stage these groups can be described as sub-themes. These sub-themes were given a temporary name to describe them. To coin an analogy used by Braun and Clarke this is the phase when, like a sculptor creating a piece of art from raw material, the analyst starts to shape the data into an interpretation of the participant’s experience.

4. Reviewing potential themes

All of the themes were then reviewed with regards to the volume of data and what was included and excluded. This process involved joining many of the initial sub-themes to create the main themes. All of the sub-themes were also reviewed in relation to the entire dataset. This involved reading back through the interviews to make sure that the sub-themes best reflect the experience of the participants in relation to the research question.

5. Defining and naming themes

The themes were then finalised and named to best describe the parents' experiences. Braun and Clarke suggest that sometimes using direct quotes from the data as theme names "can provide an immediate and vivid sense of what a theme is about, while staying close to participants' language and concepts." (pg 10)

6. Producing the report

Once the themes had been finalised the process of writing up the results began. Braun and Clarke emphasise that the report of the analysis should not just a description of what was found, it needs to be presented as an "argument that answers the research question" (pg 10).

2.4 Ensuring quality in qualitative research

As qualitative methods have become increasingly common in social science research, the debate regarding the legitimacy of these methodologies has intensified (Elliott *et al.*, 1999; Reicher, 2000). As the evaluative criteria applied to quantitative research are disputably not applicable in these contexts, a number of guidelines, specific to qualitative methodologies, have been developed to increase the quality and legitimacy of qualitative research (Elliott *et al.*, 1999). These guidelines recommend that the study should demonstrate the following throughout the research process:

1. Owning one's perspective

   The researcher should specify their theoretical, methodological or personal orientations to enable the reader to interpret the analysis and consider alternative understandings. A summary statement from the researcher is provided in section 2.4.1.

2. Situating the sample

   The researcher should provide information describing the participants and their contexts to assist the reader in evaluating the range of people and situations to which the research findings may be applicable. This information can be found in the results section.

3. Grounding in examples

   The researcher should provide examples from the data to a) demonstrate analytic procedures and b) illustrate their interpretation of meaning. This enables the reader to appraise the fit between the data and the researcher's understanding. Chapter 3 presents the thematic analysis of the data along with direct quotes from the participants.

4. Providing credibility checks

   The researcher should employ methods, such as using an additional analyst, to check the credibility of their findings. Two additional independent analysts who were fully competent with Thematic Analysis provided support in reviewing the data and analysis and offered some elaboration to the initial analysis. In addition to this a consultant Paediatrician considered an expert in the field of Paediatric CFS/ME also provided valuable feedback regarding the initial analysis.

5. Coherence

   The researcher should report understanding in a manner which a) enables the reader to understand the phenomenon and b) preserves the nuances in the data. Chapter 3 provides such a framework; beginning with a diagram (see Figure 3.1) which depicts the relationships between the themes and further provides a comprehensive verbal narrative around the 5 main themes using examples from the data.

6. Accomplishing general versus specific research tasks

   The researcher should provide a clear explanation regarding whether a general or specific understanding of a phenomenon is sought; thus addressing the issues of generalisability. The aims of the study (section 1.9) and clarification of principles of qualitative research (section 2.2.1) provide a clear understanding of the task of the current study.

7. Resonating with readers

   The researcher should present the findings in a manner that enables the reader to connect with the narrative and perceive it to represent an accurate depiction of the researched phenomena. This was firstly achieved through 'member reflections';

inviting the research participants to comment on the initial themes in order to confirm wether the researcher's perspective accurately reflected their experiences. In addition to member reflections draft chapters were regularly submitted to the researcher's supervisors for feedback on the narrative of the experience under investigation.

2.4.2 The Researcher's Perspective

The researcher is a single 33 year old White British male from a middle-class background currently living in the South West of England. The researcher has been employed as a trainee health psychologist by an NHS Specialist Paediatric Chronic Fatigue Syndrome (CFS/ME) team for the last 5 years and manages a large caseload of patients. The researcher's role is to facilitate appropriate changes to a patient's day to day activity and sleep and also provide relaxation training. In addition he sometimes provides cognitive behavioural therapy for the management of CFS/ME. The researcher regularly sees children with their parent/parents though the treatment model is very much focused on the child. This duel role of researcher/clinician put the researcher in a particularly powerful position, and it was therefore important that there was consideration given to the impact this could have. One particular issue was that families may have felt coerced into taking part in the research. Families may have also wanted to present a particularly negative picture in order to elicit further support, or a particularly positive picture to appease the researcher/clinician. Alternatively, being in the unique duel role position the researcher/clinician was already a part of the families' lives, they trusted the researcher and had an ongoing relationship with him, which may have afforded richer data. A negative aspect of the duel role is that it can be difficult "to see the wood for the trees", in that being embedded in families lives can make it difficult to take an objective view of what is going on. For a while, especially during the analysis stage of the research, the researcher/clinician was working with these families and fully immersing himself in the data. However, on the other hand, being embedded as a researcher affords for rich and detailed analysis. Braun and Clarke recommend being immersed in the data. Having recognised that "not seeing the wood for the trees" could be a potential problem the researcher took steps to address this by keeping a reflective journal, having regular supervision meetings and also utilising inter-rater reliability.

Another particular issue is that of gaining consent from the young people with CFS/ME. Although the young people are not directly part of the data collection for the current study, indirectly they are present through the parent's narrative. There may be times when the parents say things that the young person may not be happy with. Although for this study consent was not sought from the young people, this may be worth considering for future research.

The researcher's male gender may have had implications on the way data were elicited and interpreted. Families may react differently depending on gender of the researcher, for example there were differences between what mothers said to a female researcher in Missen's study (2012) and what parents said in the current study, specifically with regards to talking candidly about couple's sex life. Although this was discussed in Missen's study it was not brought up in the current study. Mcneill (2004) noted that in his research fathers were reluctant to express the depth of their feelings openly, whereas in other research with female researchers (Goble, 2004; Ware and Ravel, 2007) intimacy with a partner was discussed openly.

As part of the Professional Doctorate in Health Psychology the researcher has completed a systematic review prior to starting the current research (Appendix viii). The review, "Does family therapy help children to cope with chronic illness?" highlighted that actively engaging with families through a multidisciplinary family therapy approach may increase coping among children and adolescents. This review of 6 studies has informed the current research since it highlighted the impact chronic illness has on families; in addition the review drew attention to the lack of research on the impact of CFS/ME on family relationships.

2.5 Ethical issues

1. Ethical Approval

Prior to the research commencing, the researcher obtained approval from the Local Health Board Research and Development Units, the Local Research Ethics Committee, and the University of the West of England ethics committee

2. Informed Consent

A participant information sheet outlining the study aims and procedure was developed for parents (Appendix i). This informed potential participants of the voluntary nature of participation in research, the right to withdraw from the study at any stage without penalty and the measures in place to ensure anonymity throughout the research process.

Participant's consent, (Appendix ii) regarding the interview being audio-taped and the possibility of anonymised data collected during the study being included in the final report, was obtained several days prior to the interview and confirmed by the researcher on the day of the interview.

3. Confidentiality

The researcher ensured that issues of confidentiality were fully explained in the participant's information sheet (Appendix i). These were re-iterated verbally prior to the commencement of the interview. Specifically, it was highlighted that all information would remain confidential, with the exception of participants' disclosures of risks to themselves, or to others, in which event the researcher would need to share that information with the relevant services in order to provide the participant with the appropriate support.

To further protect participants' anonymity pseudonyms were allocated and specific details referred to during interviews such as holiday destinations and gender of siblings were changed. After the data had been analysed, the transcripts were stored in a locked cabinet (on an NHS site) to remain for five years before being destroyed (Data Protection Act, 1998).

4. Debriefing

At the end of each interview participants were reminded of the research protocols in place (as outlined in the participants' information sheet) to provide additional support in the event of distress arising from, for example, the recounting of past difficult experiences. This additional support was available from the senior clinical psychologist in the team who could talk to parents if they felt this was needed. In addition, participants were also reminded that the research was part of the principal researcher's doctoral studies and that the findings of the study may be published in a peer-reviewed journal.

5. Dissemination of Research Findings

A summary of the research findings was provided to the South West Paediatric CFS/ME service and participants upon completion of the writeup of the study (Appendix iii). It is also intended that the study will be submitted as an article for publication in a peer-reviewed journal publication. The thesis will be held in the University of the West of England library.

2.6 Study design

The study employed a qualitative research design and involved a series of semi-structured interviews with 12 families who were currently involved with the South West Paediatric CFS/ME service. The purpose of the semi-structured interview was to elicit the participants' subjective experience of having a child with CFS/ME; including their expectations about relationships and recovery. The interviews were transcribed and the data analysed using Braun and Clarke's guidelines for Thematic Analysis (Braun and Clarke, 2012).

2.7 Service context of current study

The Specialist Paediatric Chronic Fatigue Syndrome (CFS/ME) service is based at the Royal United Hospital for Rheumatic Diseases (RNHRD) in Bath. Each year the service treats on average 200 children and young people up to the age of 18. The treatment model is based on the guidelines published by the National Institute for Clinical Excellence (NICE) which covers the clinical management of patients given a diagnosis of CFS/ME. Young people with CFS/ME will typically be seen with one or both of their parents by members of the clinical team. The Royal College for Paediatric and Child Health (RCPCH) guidelines recommend that engaging with the family and maintaining a therapeutic alliance is crucial for successful implementation of the management plan, since a greater awareness of the impact that CFS/ME has on families can help facilitate engagement with families through increased empathy.

2.8 Sampling

Purposive sampling was used to recruit parents from consecutive attendees at the paediatric follow up CFS/ME clinics in the RNHRD in Bath, the Gloucester Royal Hospital and Frenchay Hospital in Bristol. Purposive sampling was used in order to achieve a mix of single parents, couples and individual parents; this involved initially asking all CFS/ME clinic attendees (the parents) if they wanted to take part in a research project ensuring that a mix of mothers, fathers and couples were recruited.

In order to address the research question, which aimed to explore the impact of CFS/ME on family relationships, and ensure the safety and well being of participants, a set of inclusion and exclusion criteria were developed. The specific criteria are presented below.

2.8.2 Inclusion Criteria

Parents of children currently receiving treatment from the South West Paediatric CFS/ME service

Parents of children who have been seen regularly by the principal researcher in follow up clinics for at least 3 months since initial assessment.

2.8.3 Exclusion Criteria

Parents of children whose diagnosis of CFS/ME is not confirmed

Parents of children whose symptoms have another potential cause

Parents who cannot speak or read English

2.9 Recruitment of Participants

Participants were recruited from follow up clinics in the South West Paediatric CFS/ME service. Follow up clinics are any appointment that families have after the initial assessment has taken place. Follow up appointments differ from initial assessments in that the diagnosis of CFS/ME has been confirmed, and they tend to be shorter in duration, initial assessments are typically 1 hour 30 minutes while a follow up appointment is 30 minutes to 1 hour. Parents of children with CFS/ME who attended the principal researcher's (AHF) follow up clinics were asked by the principal researcher, at the end of the appointment, if either one or both parents would be interested in taking part in a research study. The research was briefly explained to parents and it was emphasised that access to the clinical service would not be affected whether they took part in the research or not. Those parents interested in taking part were given an information sheet, consent form (including specific consent to audio-taping) and a contact details sheet to complete after the clinic appointment. Only those returning their forms were included and the semi-structured interview was arranged. Parents who did not return their consent forms did not receive any further communication about the study and continued to receive clinical input as normal by the team.

2.10 Data collection

2.10.2 Semi-structured interview

A topic guide for participants (Appendix iv) was developed through an exploration of family-systems literature. The 5 areas recommended by Rolland in the Family Systems Illness model (see section 1.3.3) was particularly influential and helped to focus the topic guide and prompts.

Feedback on the topic guide was given by 2 qualitative researchers at the Centre for Child and Adolescent Health, University of Bristol. Both of the qualitative researchers were experienced in conducting chronic illness research in a family context, one had prior experience of research into childhood CFS/ME.

Once consent was obtained the parent(s) were contacted and an interview was arranged at a place and time that was convenient for them. For all 12 families this was at their own homes. The duration of the interviews was between 30 minutes to 2 hours. Only the parent/parent couple being interviewed was present at the interview, other family members were not present. Parents were reassured of the anonymity and confidentiality of the data prior to commencement of the interview.

Participants were guided through the topic guide in a semi-structured interview. The researcher used open ended questions, alongside expanding and exploratory prompts, to ensure flexibility throughout the course of the interview. This method allowed participants to talk freely about their experiences. In total 18 parents were interviewed representing 12 families. Participants were recruited until no major new issues or themes appeared to emerge during the coding process (data saturation). All the interviews were transcribed and analysed by the principal researcher.

2.10.3 Data analysis

Data were digitally recorded, transcribed verbatim onto a private computer (password-protected) and analysed using thematic analysis (see section 2.3 for further information regarding thematic analysis).

After transcribing all 12 interviews verbatim the principal researcher began familiarising himself with the data and generated initial codes. An additional researcher who was not involved with the clinical service or the data collection also familiarised herself with the interview transcripts and suggested her own initial codes. The principal researcher and the independent researcher discussed both sets of initial codes and found a high level of agreement. Disagreement was resolved through discussion.

CHAPTER THREE: RESULTS

CHAPTER OVERVIEW

This chapter presents the results of the qualitative analysis of the data collected from 18 participants interviewed about their experiences of caring for a child with CFS/ME. The data were analysed using Thematic Analysis and organised into 5 themes and sub-themes.

All identifiers have been removed from the quotations and pseudonyms have been allocated to all participants.

3.2 Response rate

Twenty families were asked about taking part in the research. Of these, 3 families (15%) did not want to participate and 17 agreed, although 5 of these did not return their consent forms.

In total 12 families (60%) agreed to meet with the researcher and participated in the study. The duration of time between providing the research information pack and the interview taking place was approximately three weeks.

Of the 12 families represented in this study 2 were single parent families. Of the remaining 10 families, 4 parents were interviewed alone. Table 1 provides a brief description of the participants. Pseudonyms have been used to ensure anonymity for both young person and parent.

3.4 Table 1: Participant details

Family No.	Pseudonym		
	Child (age)	Parent (Interviewee)	Parent description
1	Karen (13)	Barbara and George	Couple interviewed together
2	Jenny (16)	Sally	Mother of couple interviewed alone
3	Gemma (17)	Carol	Mother, single parent
4	Zoe (12)	Janet	Mother, single parent
5	David (16)	Brian and Sue	Couple interviewed together
6	Rachel (16)	Paul and Anna	Couple interviewed together
7	Hayley (16)	Debbie	Mother of couple interviewed alone
8	Mark(11)	Andy and Claire	Couple interviewed together
9	Julie (14)	Richard and Vicky	Couple interviewed together
10	Emma (15)	Simon	Father of couple interviewed alone
11	Kate (15)	Stephen	Father of couple interviewed alone
12	Tom (17)	Laura and Angus	Couple interviewed together

3.5 Table 2: Themes and sub-themes

This table outlines the 5 main themes that were constructed from what the parents said, included are the various sub-themes that are part of the main themes. All of the themes and sub-themes are explained and explored in section 3.6.

Long and Difficult Journey	**Uncertainty**	**Isolation and Restriction**	**Focus on unwell person at the expense of family life**	**Parental Roles**
Up hills, through tunnels	What are we dealing with?	No one can see this	Life revolves around them	Good cop/ Bad cop
A constant struggle	Who are they and what will they become?	Only we know	Destructive force	It’s a “mother” thing
Exhaustion	There must be something we can do	Our world has shrunk	Resentment	Parents a team
Is the worst behind us?		A need to escape and a feeling of guilt		

3.6 Thematic analysis

What follows is the exploration of 5 themes and their corresponding sub-themes. A full list of all of the quotes used to construct the themes can be found in Appendix vi.

Theme 1:

Long and Difficult journey

One of the mothers, Sue, commented that the experience of living with CFS/ME was "going on too long". This encapsulates a feeling that all of the parents expressed that they were on a long, difficult journey, which is explored within this theme. The sub-theme "up hills, through tunnels" looks at how the narrative of being on a long journey is used by the parents to explain their experience of living with their child's CFS/ME. The sub-theme "a constant struggle" explores how parents find their day to day life difficult because of CFS/ME. "Exhaustion" is a sub-theme focusing on the outcome of the parent's "long journey" and "constant struggle". The final sub-theme "is the worst behind us?" presents an optimistic view that some of the parents had.

Up hills, through tunnels

For parents living with a child with CFS/ME day to day life had become a difficult journey which began from the moment their child became unwell and was not clear as to when it would end: *"It's just taken a long time really, it's a bit of a rough journey" (Anna).* 12 parents (6 mothers, 6 fathers) representing 8 of the families described living with their child with CFS/ME in these sort of terms. The parents seemed uncertain as to the duration and destination of the journey, this is encapsulated by Sally:

"It's a bit like walking towards the top of a hill and you think you're at the peak and then you get to that bit you see there's another bit beyond, and another bit beyond, I think we'll get on top of it eventually but it's the fact, it's a journey of an unknown length that makes it difficult." (Sally)

Sally describes how just as they feel like they are reaching the end of the illness journey there is a lot further to go. Sally's hill metaphor also emphasizes that the experience of managing CFS/ME is very much an uphill struggle. Although this is a long uncertain journey there did seem to be some optimism that what lay ahead would not be quite as difficult to manage, as demonstrated when Sally suggests that the worst part is behind them:

"I think we've been through the most horrible bit and we're back to where, back to where we were." (Sally)

Further optimism is evident in George's positive attitude:

"I'm just hoping that things eventually you know work their way out of this, we'll get her up and running….the best way I can put it is 'so far so good', we haven't had anything to bother us yet, which i think can only be a positive thing I suppose. (pause) and let the thing go its course" (George)

Laura uses a tunnel metaphor to describe her journey, suggesting that once they were being seen by the CFS/ME service the worst was perhaps past:

"You were the light at the end of the tunnel" (Laura)

Laura's use of the "*light at the end of the tunnel"* metaphor suggests that the CFS/ME service served as the first glimmer of hope or the first sign of rescue from a desperate or hopeless set of circumstances. It is as if some parents feel trapped by their circumstances, much like someone might feel trapped in a tunnel or cave walking in complete darkness and uncertainty. This is highlighted by Anna who says:

"*you're in this long, long tunnel of, and a sense that people DO come out of it and DO get better, I think it's really, really important, that hope, keeping hope going" (Anna)*

Spotting a light would be a sign their struggles would soon be over and escape would now be possible. Metaphorically speaking, a light at the end of the tunnel would signal the end of uncertainty or doubt. The significance of the first sign of light is a much-needed suggestion of hope, which often motivates people to keep moving with a renewed sense of purpose, for example Sue felt that there was a glimmer of hope in the sense that her son was finally starting to get better:

“There’s a, a tiny part of us both at the moment I think that actually wonders whether he has turned a corner and whether actually he is a lot better” (Sue)

However, it is important to note that the first light at the end of the tunnel is not necessarily a guarantee of a more positive outcome. The “light” may not be the salvation first thought:

“*because you don’t know how long a time you’re going through you see glimmers of light but then it’s just a little reflection and it’s not the end actually.” (Sally)*

There still could be a number of obstacles to overcome or crucial decisions to make before the source of the light can be reached:

“*From there being a light at the end of the tunnel suddenly there was another rockfall to clear away” (Stephen)*

The supposition is that most tunnels do have an entrance and an exit, but following the path towards those exits can be a long and frustrating experience:

“It’s a drag at the moment, it’s going on too long” (Sue)

There was a sense that some of the parents had grown weary of chronic fatigue and they just wanted their child to be better:

“Just before Christmas I got to the point where I just thought I’ve had enough of this now, you know, ‘why hasn’t it ended?’” (Andy)

A constant struggle

Just as Sally referred to managing CFS/ME as being like always trying to climb a hill , and seeing more hills ahead of you, many of the parents felt that dealing with the CFS/ME was extremely difficult and complicated, and that they were engaged in a constant struggle.

Indeed, husband and wife Brian and Sue referred to how they felt life was “hard enough” without the addition of the “burden” of CFS/ME. This was echoed by the other parents; Vicky mentioned “staggering through” and it being “extremely difficult”, while her husband Richard felt it made day to day life “slightly more complicated”. For Richard and Vicky it seemed that for them the ongoing management of CFS/ME would lead to potential conflict between the two of them:

“I’m not knocking how [my husband] came back [from clinic] and was like ‘oh we’ve got to do this and we’ve got to do this’ and I’m kind of going ‘yes, we’re trying to but actually it’s really complicated” (Vicky)

The potential conflict here arises from Vicky’s husband returning from clinic with a plan of action that, for Vicky, seems difficult to implement. Similarly Sue referred to the constant struggle impacting on her relationship with her husband Brian, though she was unsure as to exactly how that “impact” manifested itself:

“I think it’s had a huge impact on our relationship without us being able to put a finger on exactly what, what it is, umm but I’ve just, I think that I, that I find the family harder work than I think it should be” (Sue)

There was a sense that parents were struggling because they felt that the CFS/ME made life much harder and more challenging than it perhaps needed to be. Everyday tasks are made much harder because of the CFS/ME; This can be seen when Andy reflects on the “challenge” that can be posed by “something like grocery shopping”, while Angus mentions that tasks that may have been taken for granted in the past would now almost feel impossible to tackle:

“You couldn’t do what would normally just be a fairly simple thing” (Angus)

Angus goes on to reflect that overall life is “more complicated” because of the CFS/ME which makes “things less straightforward as a result”.

Sometimes parents talked about their experience as being like an ongoing conflict, Sue refers to family life as a “battleground”, emphasizing the perceived conflict within the family:

“It’s a battleground, almost constantly, we’re not having enough nice time as in the family...There’s a lot of conflict between the children and he’s probably responsible for most of it” (Sue)

For Sue it feels like the conflict is constant, taking it’s toll on family life. Simon also refers to the “battle”, in his case specifically relating to trying to wake his daughter in the morning:

"I tell you, it's a bit of a battle sometimes in the morning to get her up" (Simon)

Like an ongoing battle, living with CFS/ME is a constant struggle for both the unwell individual and the family, as emphasised by both Andy and Stephen:

“I tend to call it the grind, it’s just constantly there isn’t it?” (Andy)

“it’s constantly in the back of your mind, am I supposed to be taking her somewhere, should I have told her to do her homework” (Stephen)

Although the struggle is ongoing it seems to begin prior to diagnosis; for example Sally recounted how after her daughter’s state of ill health was dismissed by the GP she felt that she had to fight for a diagnosis:

“I was outraged, but er, but then I didn’t go away from that appointment leaving it like that, I said ‘well if nothing else I need a formal diagnosis’” (Sally)

11 of the parents gave the impression that they were always busy because of the CFS/ME. Paul talked about there always being a “set of tasks” to carry out that were directly related to the CFS/ME, while Sally felt that, along with her husband, they had to work hard to “push” their daughter through 6th form. Stephen referred to the additional “work” that is created for both himself and his wife because their child is unwell. Similarly, Vicky mentioned that there was “additional work” and “a lot of pressure” and highlights the constant business with the use of a metaphor:

“It does feel like we’re constantly managing things and just keeping, you know, juggling balls in the air, keeping pots boiling” (Vicky)

There was a feeling that part of the struggle was that parents felt they were constantly working "behind the scenes" to keep a semblance of "normal" life for the unwell child.

"[We're] just trying to be as proactive as we can with her and doing everything so she doesn't have to do an awful lot. The fact that, behind that you go in and tidy her bedroom up so it doesn't look untidy, and running around getting her special food... Just the little things to make life normal" (Debbie)

Debbie and Janet both capture this idea well with their use of animal analogies:

"to my mind, the more you can keep everything normal, you know, it's like that duck, the feet are going like mad underneath it, trying to make it normal... So Hayley's life continues to be normal." (Debbie)

For Debbie family life 'on the surface' to the outside observer seems 'normal', however she feels that just under the surface they are having to do everything to stay afloat. This constant state of business and alertness is further highlighted by Janet who, using a metaphor of a hamster on a wheel, suggests that it is not until she is able to stop and take stock of the situation that she realises just how busy she has been:

"It's kind of like on a hamster wheel, you know just going round and round and round and if you get a chance to get off every now and again sometimes I'm thinking 'Gosh I could go out today, but (sigh) I don't know if I really want to really'. So it's, you kind of get that aspect of it as well which is, which is crazy really, but when I do go out, when I make the effort I do go out I do realise how much I've needed it." (Janet)

This highlights an awareness the parents have that life has significantly changed and so they feel that they have to work hard to try and keep everything as "normal" as possible for their child. For example, Barbara stated that "it's important that we keep life the same for [Karen]". Perhaps this is because they feel they can not 'fix' their child in as much as they can not cure them, so they have to work behind the scenes to make life seem "normal". Laura makes clear that this process of normalising life is not about trying to ignore the current difficulties:

“You’re constantly trying to, not gloss over it, but make it seem normal that you’re not all doing it together really, I feel.” (Laura)

Although the parents work behind the scenes to “make things normal” for their unwell child, Sally refers to being painfully aware that this is not the case:

“I think just the painful reminder that my daughter wasn’t able to do that anymore, yeah, and that, if you like, the sense of grieving, yeah, that sort of loss, loss of the child you once had” (Sally)

Sally is often reminded when she sees other young people who are Jenny’s age that Jenny is no longer like them. This is also explored in theme 2.

Exhaustion

The constant struggle that the parents described could lead one to surmise that they must be extremely tired. Indeed, 7 parents talked about this. Laura describes at one point “collapsing in a heap”, and this is particularly indicative of the total exhaustion that many of the parents referred to. In particular, both Sue and Vicky referred to the overwhelming “exhaustion” that they experienced as parents managing their child’s CFS/ME, while from a father’s perspective Angus mentioned that although managing their son’s CFS/ME was “tiring” for both himself and his wife he conceded that this probably impacted his wife more than himself, suggesting a potential imbalance between husband and wife. Sally felt that it had begun to feel like everyone in the family had CFS/ME because everyone felt “very tired”. Anna explains why the experience is particularly tiring for her family:

“You need quite a lot of stamina to keep going and it did remind me of when they were younger, you go from activity to activity and changing your activity levels and it’s tiring” (Anna)

For Anna it is as if she has to go back to an earlier stage of parenting because her unwell child is so dependent on her.

Is the worst is behind us?

Sometimes parents were quite stoic about their experience. Paul commented that “it’s not all bad”; for Brian there was an acceptance that the CFS/ME was “*nowhere near as bad as it could be”.* Anna explained that they always tried to focus on the positive and were proactive:

“We never feel it’s pointless and hopeless and it’s not worth doing, we’re always looking for the way forward” (Anna)

When talking about the CFS/ME there was a sense that parents felt that although it had a massive impact on their lives it could have been a lot worse. For example, Brian refers to the fact that CFS/ME is not a terminal illness, while Simon reflects on how the situation could have been worse:

“I guess there’s always hope that it’s going to get better, so it’s not (pause) it’s not terminal” (Brian)

“She could be disabled, much more disabled, then it might have affected me.” (Simon)

As well as considering that the situation is perhaps not as bad as it could have been, there was also a sense that there were some positives, as explained by Richard:

“It’s sometimes too easy to get caught up in the frustration of what’s not happening to recognise what has happened and I think the journey she’s been through has been quite significant and I think she’s learnt and gained some other skills as part of it as well, which I think are positives” (Richard)

It seems like, to coin a cognitive therapy phrase, Richard is able to cognitively restructure the experience of his daughter being unwell and focus on the potential positives that can come out of the situation. In this case the experience has meant that his daughter is better equipped to deal with illness in the future.

Theme 2:

Uncertainty

A common experience for all of the parents was a feeling of overwhelming uncertainty. Parents felt uncertain about their child's prognosis and a lack of understanding about the etiology of CFS/ME. This is explored in the sub-theme "What are we dealing with?" highlighting a feeling of helpless and powerless. Parents also felt uncertain as to how CFS/ME had changed their child, this is encapsulated in the sub-theme "who are they and what will they become?". Because of the constant uncertainty parents felt that their skills as parents were sometimes called into question. This is part of the sub-theme "there must be something we can do".

What are we dealing with?

Uncertainty was something that all of the parents talked about, particularly with regards to the period before the child is diagnosed with CFS/ME. Vicky reflects that "it takes a while to learn that ME is ME anyway". For Laura it took a year before they knew what was going on:

"It took us nearly a year I'd say to actually get to the point where anyone diagnosed this whole thing as Chronic Fatigue Syndrome" (Laura)

Sue describes this long time up until her son was referred to the CFS/ME service as "a horrible year not knowing". During this period of "not knowing", parents are uncertain as to why their child is not managing what they would normally be able to do, at this stage they are looking for answers. These fathers highlight the feeling that something is wrong but it feels impossible to work out what it is:

"it's quite worrying, I think, when you notice your child is ill and you can't actually put a finger on what's wrong" (Stephen)

“We had an inkling what was wrong with him but until you know for sure you’ve still got, as I say, the sort of huge degree of uncertainty” (Angus)

“we went on holiday and suddenly it was like why hasn’t she got the energy just to do what would be considered a normal walk, you know, what’s wrong?” (Richard)

During this period of uncertainty a high level of fear and helplessness could be experienced, as highlighted by Janet:

“it was quite frightening times, yeah, because I didn’t know really what was happening, she was, you know, getting worse, she was deteriorating, she was (sigh) struggling to eat, um, you know sometimes, you know, speak, and I was thinking ‘I don’t know what to do’” (Janet)

Once the diagnosis of CFS/ME has been made, sometimes the diagnosis can exacerbate rather than reduce feelings of uncertainty and helplessness. This is illustrated by Carol who explained that *“even though you’d told me it was ME I thought there was something else going on”*. George commented that they have *“no idea what’s laying ahead of us”*, which was echoed by Paul who felt the diagnosis *“certainly brings uncertainty”*. Parents also talked about uncertainty caused by the fluctuating nature of the illness itself:

“it seems that everybody seems to suffer with ME differently and umm, so there doesn’t seem to be a, I would like there to be a “it will last for 2 years and you feel this symptom in month one...” but there isn’t any of that and it seems to be a bit up in the air really.” (Sue)

“it can morph and vary from individual to individual quite considerably” (Anna)

“you don’t know how bad it is because everyone is completely different” (Debbie)

Laura expressed an ongoing uncertainty as to the length of time her son would be unwell: *“I don’t know how long it’s going to go on”*. Not knowing how long their child will be unwell for can be a frustrating experience, leading parents to ask questions for which answers may not be readily available: For George and Barbara they wanted to know *“how long is it going to last?” (George)* and *“where has it come from? Why has Karen got it?” (Barbara).*

Brian had similar questions, highlighting that even when you live with CFS/ME it can still remain ambiguous:

“What’s frustrating for ME organisations and for us is ‘what is it?’, are we, is it a physical disease? or is it a disease of the brain?” (Brian)

This feeling of dealing with an ambiguous illness was echoed by other parents. Some said that they did not “understand” the illness (Sue, Andy), while Vicky described CFS/ME as a “strange” illness particularly because of it’s fluctuating nature:

“ME’s strange, you know, it’s like sometimes it has more of an impact than others and there are times with her not at school when she’ll have bags of energy in the evening and it’s kind of like, hang on a minute, if you’ve got this much energy why aren’t you going to school?” (Vicky)

A similar view is expressed by Simon:

"I don't understand it, how can she get so tired without doing a lot to get tired? I cannot understand that" (Simon)

Mother’s Anna and Claire both recount how a lack of understanding can lead to frustration:

“learning that chronic fatigue and ME were the same thing and that post-viral fatigue didn’t really exist in any real terms, it was just really frustrating” (Anna)

“I do find it an incredibly frustrating illness because I think it’s... you just can’t understand it and I have a need to be able to, you know, get it right and do what’s right for him and push him on if I need to, but I don’t understand.” (Claire)

“We’ve got to the point of thinking ‘when is this going to end?’ you know ‘is it?’, well yeah ‘is he ever going to get better? Is he going to be employable? Is he going to have a life?” (Sue)

Sue gives the sense that she and her husband are at some sort of turning point, that perhaps up until recently they had held on to the belief that their son would get better, that they could still envision a “normal” future for him. Sue’s concerns about future employability of her son are echoed by Simon in his interview:

"The bit I worry about is - is she going to get a job?" (Simon)

Who are they and what will they become?

It seemed that parents felt that since their child had become unwell they no longer felt like the same person, in a sense the parents felt like they had lost their child. This is highlighted by Stephen:

“We’ve lost an active, academic child and we’ve got an inactive child that’s struggling with the amount of work she’s going to need to do” (Stephen)

Both Sue and Anna reflected on the fact that it seemed the CFS/ME had had a transformative effect on their children, turning them into people they no longer recognized:

“it’s horrible to see, your child who was a very sociable person turn into somebody who, it doesn’t feel like it’s quite him at the moment” (Sue)

“Her confidence and self-esteem have had an enormous knock - she’s almost like a different person in that respect, since before and afterwards of having it.” (Anna)

Both Sue and Anna’s accounts give the clear impression of two versions of the same person; the person before CFS/ME and the person with CFS/ME. In both cases the person with CFS/ME is a lesser version of the person prior to the illness. For some parents it felt like their child had regressed to an earlier stage of development:

“She isn’t able to do what you would normally expect a little child at her age to be doing” (Vicky)

It is interesting that Vicky refers to her daughter as a “little child” when actually Julie (her daughter) is a teenager. Perhaps this is a reflection of how, because illness has increased their parental dependency, the young people are perceived by their parents as being younger than they really are. This is reflected by comments made by both Anna and Janet:

“it reminded me of having small children when they were young and you’d have to find things for them to do” (Anna)

“She’s like a small child again” (Janet)

The effect of this is that sometimes parents feel that their child is unable to cope and thus the parents find it difficult to relinquish some of the power/support. For example, Anna reflects that it is “hard to let go of [Rachel] and do things on her own”.

Sally talks about how seeing other children who are not unwell enjoying themselves can be difficult, because it reminds her of the loss she has suffered:

“it was horrible being with all these normal kids like, like my child used to be. I think just the painful reminder that my daughter wasn’t able to do that anymore, yeah, and that, if you like, the sense of grieving, yeah, that sort of loss, loss of the child you once had” (Sally)

She felt this sense of loss so acutely that she likens the experience to grieving for someone. This is a feeling shared by Stephen:

“I mean she’d been to see her GP a few times but, it’s one of those things, there’s nothing wrong and then she’ll have a spurt when she’s better, you know, down again, so a bit like a, a grieving process.” (Stephen)

Some parents talked about their child losing something about what it means to be a child/teenager:

“At his age it should be normal for him to be going down the youth club on a friday night” (Brian)

“It’s hard to see her not flourishing and not becoming independent, it’s hard to see a young person being quite so restrained” (Anna)

“Hayley’s not a, I say ‘normal ‘ teenager, Hayley I don’t think is a run of the mill teenager” (Debbie)

As well as feeling they had lost who their child once was, 5 parents talked about losing who their child might have been able to become. For Anna her daughter's CFS/ME meant "a loss of her life that she should be having”, this suggests that the CFS/ME has completely disrupted the “normal” course of her daughter’s life, and that this is something that can not be recaptured. In a similar vein Laura describes how her son has "missed a chunk of teenage years". This view is expanded by Sally who said:

“Yeah, so a bit of anger if you like that this thing’s coming along when it has, has robbed her of what should have been a, apart from the exam stress, a particularly exciting fun time of life” (Sally)

Sally expresses a sense of injustice that, like a thief, the CFS/ME has stolen part of their child's life. This perceived loss could make it quite difficult to be optimistic about the future, as seen by Stephen’s comment that the CFS/ME was "restricting her future, it’s made her future very narrow” (Stephen), and by the fear expressed by Simon:

"I worry about her later life, I worry about that tremendously if I'm honest" (Simon)

There must be something we can do

While parents seemed desperate for their children to get better there was also a sense that because they were dealing with a something that they did not fully understand, they were uncertain as to what they could do to help. Frustration was a common emotion that parents experienced when they felt like things couldn’t be fixed:

“at the end of it it’s frustrating, trying to keep us on the straight and narrow is a good thing and valuable but it (pause) what we really want is for it to be fixed” (Brian)

“the worst thing is, the most frustrating thing is that there’s no easy fix and with an illness you really want an easy fix that gets it over and done with” (Debbie)

“that leads to lots of, lots of stress and strain as to, you know, what should we do? Should we be firm? should we try and make her get up, try and make her get into school?” (Sally)

Here Sally seems to be questioning her parenting skills. Some parents expressed that they should be able to make their child better, though this led to a sense of powerlessness when they felt that they were unable to do this:

“I feel helpless because I don’t feel like I have any level of control at all, you know, as a mum I’d just like to take the ME away” (Sue)

“it’s been two years, I should have been able to sort this out by now. I got a bit fed up with it all if I’m honest” (Andy)

Sue’s comment, “as a mum”, suggests an underlying assumption about the role of parents, that a mum should be able to make their child better. This suggests that Sue is putting a lot of pressure on herself. Similarly Andy says that “he should” have sorted things out “by now”, here he is perhaps presenting himself as having failed as a parent.

Richard and Vicky reflect on the feeling of being completely helpless faced with an unwell child:

Richard: “It comes back to that thing about how one feels when you’re just looking at somebody you love suffering essentially and not being able to flourish and...” Vicky: “You can’t help her” (Richard and Vicky)

It almost sounds as if Richard and Vicky are viewing their child through a window into another room, they can see her but they are powerless to do anything to help her.

“at this present point in time we’re struggling about what to do next” (Brian)

“Initially I felt quite relieved and then probably after a while I felt you know, like ‘oh my god, this is the end of the world” you know, especially as she started getting worse” (Janet)

There was a sense that because their child had been unwell for so long, and parents felt powerless to help, they were put in a position where they felt their skills as parents were questioned. Sometimes this questioning was coming from the parents themselves; it was evident that parents were putting huge pressure on themselves as is clear by Sue's comment:

"I just feel guilty that as a parent I should be managing the whole thing a lot better than I am, so it's just constant feelings of guilt all round that I'm not doing the best for David" (Sue)

Here Sue is constantly questioning whether she is "doing the best" for her son, similarly Claire reflects on past decisions and wonders whether she could have managed things in a different way:

"I think it's one of those things in retrospect you perhaps would have done slightly differently" (Claire)

Stephen reflects on worrying that they as parents are somehow responsible when their daughter's health deteriorates:

"She was doing ok for a while and then she started to struggle again, it was really difficult to work out, is it something we've done wrong?" (Stephen)

The guilt that both Stephen and Sue refers to is also touched on by Debbie. Debbie recounted how she had felt guilty in the past as to whether having the HPV cervical cancer vaccine injection had caused her daughter's CFS/ME:

"She had the injection, as all girls did, and then you read that there may have been a link to that, so she had that last year, the year before and you kind of think, ah dear, you know. You worry about all these things ." (Debbie)

There was also evidence that parents felt their parental skills were questioned by others. For Sue there is a perception that other people are critical of her and view her as an "overprotective parent":

“We’re also faced with people saying “isn’t it marvelous that David’s better now!” when they see him on a good day and you just feel like a (sigh) an overprotective parent when you say “oh, but he’s not better” you know, people think you’re crazy!” (Sue)

It is as if Sue feels that the legitimacy of her concerns for her son are being questioned by others, so that instead of being a caring parent she has become “overprotective” and even “crazy”. Stephen makes a similar comment when talking about taking his unwell daughter to the GP:

“You do feel very, almost fraudulent, taking a tired child to the doctor” (Stephen)

Stephen worries that he is “fraudulent” for taking his daughter to the GP when she is just “tired”. This comment highlights the a fear that they will not be believed by others, that although they are certain there is something wrong with their child it isn’t enough to just be tired.

Theme 3:

Isolation and Restriction

There was a strong sense that the interviewees felt isolated. This was felt keenly by single parent Janet who said *“We’re really on our own, we really don’t know what to do”*. However, it wasn't just the single parents who felt isolated. At one point Andy said *"we’re on our own”*, and this was echoed by Laura in her interview saying *"we have felt quite on our own really"*. Isolation could be caused by parents feeling that CFS/ME was an “invisible illness”, this is explored in the sub-theme “no one can see this”. Linked to CFS/ME being an “invisible illness” was a feeling parents had of validity of their child’s unwell status being questioned, this forms the sub-theme “only we know”.

In addition to a strong feeling of isolation this theme also highlights the restriction that parents felt the CFS/ME had imposed on them and their families. The sub-theme “our world has shrunk”, which refers to a specific phrase used by Laura, encapsulates the feeling that the parents’ lives had become so restricted by the CFS/ME that it was as if their social world had become a whole lot smaller, and explores this with specific examples of how the parents felt their lives had become restricted. Finally the sub-theme “a need to escape and a feeling of guilt” explores the consequences of feeling isolated and restricted, essentially that parents felt the need to get away from the CFS/ME and consequently feel guilty about this.

No one can see this

There was a sense that the parents felt that only they were in a position to truly understand what it was like to have a child who has CFS/ME:

“I don’t think anybody understands unless they’ve actually lived with someone who’s had it” (Carol)

This was reflected in what 6 parents said about CFS/ME as an invisible illness in the sense that “people don’t see it” (Barbara). One parent referred to CFS/ME as an “Illness within” (George) in the sense that there are often no visible signs that the person is unwell, compared to other illnesses such as heart disease which Anna refers to as an "open illness", by which she means that there are visible cues which makes it easier for people to see that you are unwell. Richard refers to CFS/ME as "one of those invisible injuries where people know there's something wrong but they can't see there's something wrong so they don't quite know how to react". Both Anna and Debbie highlight the fact that although their child may, to an outsider, look fine, looks can be deceiving.

"She walks out the front door and she's looking quite smart and she's made an effort but half an hour into it she's tired and she wants to come home again" (Debbie)

"It's a hidden disorder, isn't it? it can't be seen, people don't know they just think you look fine and that's it and they don't see beyond that." (Anna)

Managing an "invisible" illness can be incredibly isolating for both the unwell person and the family:

"it is isolating. A lot of people don't really understand about... they see Rachel and they see that she's fine and don't understand that she can't do things" (Anna)

Only we know

In addition to feeling like other people did not understand CFS/ME, all 18 parents talked about others, including other family members, not always believing them:

"She thinks karen's putting a lot of it on [wry laugh] and she thinks she puts it on for us sort of thing, and, but she's always been a little bit jealous of Karen." (Barbara)

"Our oldest child spent the first couple of years telling us that there's nothing wrong with David, that we were imagining it" (Sue)

"her sisters still don't believe she's ill and think she's putting it on" (Carol)

Paul comments that this lack of belief can isolate the family:

“That’s the big issue actually, communicating it outside the family to people that Rachel interacts with. That IS the big issue” (Paul)

Here Paul reflects on the isolation felt. It is almost as if there is an invisible boundary that is almost like the border between two countries where the barriers may be linguistic and cultural. Richard expands on this:

“I find medical terminology something really hard to hold onto because it’s a foreign language” (Richard)

When others do not believe this can be particularly isolating because, as Sue mentions, people don’t want to know about what is going on:

“My father doesn’t, I don’t think believes it, he doesn’t want to hear anything about it” (Sue)

Lack of belief from the medical profession can also create a sense of isolation. Angus refers to the period before they had a diagnosis of CFS/ME, when it felt like no one within the healthcare sector knew what to do with them:

“we kind of dropped through the cracks in the floorboards to be honest” (Angus)

Our world has shrunk

Fifteen parents talked about how their lives were restricted because of the CFS/ME. Having a child with CFS/ME meant that parents may not "go out very much" (Carol), one parent even said that she felt “housebound” a lot of the time (Janet). On the occasions when they did manage to go out for a leisure activity parents found that the activity would be restricted, perhaps not spending "as long as [they] normally would" (Simon). Plans often had to be changed at the last minute, or abandoned completely:

“we’re always having to change plans, if we plan to do something 9 times out of 10 it ends up being cancelled or rearranged, cancelled again and rearranged, so I tend not to plan things now, do things if she’s well we’ll do things.” (Carol)

Barbara referred specifically to important events that had been missed because of the CFS/ME, specifically social, family oriented events that could be seen as “normal” for most families:

“When Karen was first diagnosed everything was Karen, you know, this child just led on that settee sound asleep 24/7, you know, we couldn’t go out, we couldn’t do anything, we missed parties and birthdays and things.” (Barbara)

Barbara’s comment that “everything was Karen” highlights that their world had become focused on their unwell child. There was a sense that once parents started to miss social events the restriction imposed on them could become self perpetuating, as highlighted by both Anna and Laura:

“you get out of the loop of socialising with people and so you don’t get asked places” (Anna)

“it’s that your life just shrinks and your social life shrinks as well inadvertently” (Laura)

Laura’s use of this ‘life shrink’ metaphor evokes images of the family being more and more restricted and perhaps even restrained. This links to another powerful metaphor that Anna uses when she says “it makes you feel like you’re in a prison”. If home is like a prison it is no longer a place of refuge, it is a place that confines you, having been kept there against your will. A central part of being imprisoned is that one’s freedom is taken; this is something that is keenly felt by Vicky:

“It’s a huge stress just managing because there’s no freedom if you’re trying to stick properly to things like that, there’s no freedom” (Vicky)

Anna reflected on how this sense of being confined and restricted by the CFS/ME had impacted on the relationship between herself and her daughter, because of her daughter’s dependence on her:

“her social world diminishes and she’s not so able to be independent and go out so she can’t be, she’s a dependent teenager really so I think it makes a difference on our

relationship... I'm here with her, you know, supporting her in what she needs but it's not really doing what I want to be doing" (Anna)

It seems that Anna might be feeling some resentment to her child encroaching on her space. This feeling of resentment is echoed by other parents when they talk about how having their child constantly with them has restricted what they can now do,

"In the beginning I felt I couldn't do the things I wanted to do" (George)

"So [my husband] gets, well he's like the dog that hasn't been walked because you're going 'but this is MY WEEKEND! I want to enjoy it with my family, but my family can't do it.'" (Vicky)

In some ways it seemed that the parents had lost their independence:

"I didn't like it because it takes away from my independence a little bit" (Sue)

Parents also reflected on how siblings of the unwell children had also been restricted by the CFS/ME:

"[his sister] has felt restricted by, and frustrated by the fact that we're saying we can't do certain things and, whilst we're trying to be careful about why and not to blame it on Mark or, if it is very much due to him, explain the situation, you can get a little bit of resentment" (Andy)

"the other children aren't allowed sleepovers and that's stemmed from David, that actually it's kind of unfair if the others do things and he's not allowed to, it's just had an affect on everybody" (Sue)

"He can't play with his elder brother, so he probably feels a bit left out, probably accounts for his slightly erratic behaviour sometimes, which is probably just attention seeking." (Angus)

There was a sense that the many restrictions on family life along with the perceived isolation had left these parents feeling that life had slowed down and become quite dull.

Indeed, Paul encapsulates this when he says that “we’re living fairly boring existences” and describes life as “stuck”. Debbie describes day to day life as being “stretched”. Vicky and Simon talked about how the CFS/ME had reduced the amount of leisure activities they were now able to engage in:

“It’s actually been really hard to get her outdoors at all. So if the rest of the family are then pent up indoors it’s boring.” (Vicky)

"It's just the leisure side that's affected." (Simon)

Both Paul and Brian talked about the absence of fun in their lives:

“there’s not so much fun full stop” (Paul).

“It consistently takes the fun out of life, takes the fun out of his life and I think it takes the fun out of ours as well. Not all, not all the fun, but it just saps it” (Brian)

Brian suggests that the reduction of fun is a gradual process, with the CFS/ME gradually sapping it from their lives. Sally reflects on how the CFS/ME has made her daughter's life devoid of the sorts of fun experiences she should be having as a teenager:

“All those sort of bits of childhood that should be fun and experimenting and exciting and having loads of energy and being able to do lots of things and all of a sudden that’s gone” (Sally)

A need to escape and a feeling of guilt

Parents felt they had lost a certain amount of their independence and they were somewhat restricted, because of this it is perhaps unsurprising that 8 parents talked about wanting to escape. This sub-theme theme highlights a dichotomy in terms of the parents feeling they wanted to get away from the CFS/ME and also feeling guilty about this. Parents talked about a consequence of the CFS/ME being that their child had to spend a lot of time at home. This led to a feeling of the parents wanting to escape from the situation, and along with this a huge feeling of guilt.

"[my husband and I] are both involved in voluntary youth work and lets you go off and escape" (Sally)

Sally refers to "escaping" into the world of voluntary work, perhaps this helps to divert their attention from focusing on the CFS/ME that can become all encompassing. For Janet there are small amounts of time that she cherishes when she can get away from CFS/ME.

"just to go out, it doesn't happen very often, go out with a friend, have a cup of coffee and talk about their problems and you know, the state of the world instead of ME" (Janet)

The problem was that once parents did "escape" for a while they then felt guilty. Barbara talks about this in terms of doing activities that she enjoys:

"I've got to still do the things I like to do, like [my husband] still needs to go to the gym because he needs to. But then I do feel guilty because Karen would have been coming with me" (Barbara)

Barbara's guilt stems from the fact that Karen is too unwell to join her, and therefore misses out on an enjoyable experience. Barbara is thus "stuck" feeling that on the one hand she should have time for her own pursuits while on the other hand having the constant reminder that she has an unwell child who cannot share the experiences with her. It is perhaps that Barbara feels selfish that she is having time to herself while her daughter suffers. This is something that Laura refers directly to:

"My day to day life changes, I'd just like the house to myself sometimes, I would like some real 'me' time without having to go out of the house in order to get it. Sometimes, I think that's probably one of the things I find really, which sounds selfish sometimes" (Laura)

Laura is not only talking about escaping the illness and the unwell child, she is highlighting a feeling that through the process of caring for the unwell person somehow the "real" you has been lost. She wants to rediscover the "real" her, though feels guilty about this.

Theme 4:

Focus on the unwell person at the expense of family life.

This theme is about how the parents' focus is, perhaps inevitably, often on the unwell child and their needs, which can lead to other family members feeling marginalised, particualrly siblings of the unwell child. The theme contains the sub themes "life revolves around them", "destructive force" and "resentment".

Life revolves around them

"it does become the focal point of arranging everything in your day, because everything's got to be based around it" (Andy)

"life revolves around Mark and his limitations" (Claire)

Parents described day to day life as being tailored to their unwell child. This was described as a full time job by both Barbara and Andy:

"somebody has to be with Karen all the time" (Barbara)

"now everything has to be actually managed around when we're taking him into school or around when someone can be here to look after him or around the length of time he's able to go out and do that thing with you." (Andy)

Andy feels that the restrictions caused by the CFS/ME dictate how a typical day can be managed. Laura reflects on life being organised around her child suggesting that this is vital in order to make their child feel normal:

"It's tempering everything to fit within his limitations is really the normality in order that he feels normal, I suppose. As normal as possible." (Laura)

A consequence of having an unwell child in the family is that they will spend a lot more time at home than when they were healthy. Andy encapsulated this with his comment that his unwell son is *"just always there"*. For some parents there was a sense that because of their child's age it was not normal for them to be spending so much time at home, in particular Brian commented that his son *"should be having more time away from us"* . The feeling that the unwell child was *"always there"* was something that 11 of the parents reflected on, touching on the fact that their relationship with each other is inhibited by the constant presence of their unwell child. This is expanded on by Angus who says that it's like *"a permanent threesome"* , this can feel restrictive for parents, as Anna explains:

"we have less free time together because normally she would be out and she's not so everyone's, we don't get so much time to do stuff do we?" (Anna)

There is a sense that having a chronically unwell child is a drain on family relationships, Anna reflects that they can't do the sorts of things that they would be doing if their daughter was well. Andy also reflects on this saying that the amount of quality time he has with his wife has been reduced:

"I think it's harder for us to have time for us together as a couple and we have to grab our moments to talk when we can" (Andy)

Couple Angus and Laura go further with their explanation, referring to a reduction in the sort of physical contact one may expect from an adult relationship:

"it's definitely inhibited our personal relationship, I'd say, without a doubt." (Laura)

"it doesn't allow you the personal intimacy and contact that you would have had before so it makes you a bit more distant, that's the thing, you're not as close" (Angus)

For Sally her daughter's constant presence has more of a practical impact; Sally felt that she could not get on with chores.

“They’re just there and in the space where otherwise you’d be just chilled out and relaxed or getting on and you know, cleaning or doing some work yourself. So that does make life different, you feel that if somebody’s always there you’ve got no space.” (Sally)

Not only can Sally not complete the housework she also feels that she cannot relax because of the constant presence of her daughter, and her comment that she has “no space” suggests that she has feelings of restriction and confinement. Parallels can perhaps be drawn here with Laura’s comment in the ‘isolation and restriction’ theme that home has begun to feel like a prison.

Another impact caused by the focus being on the unwell person is that sometimes other parts of the family are neglected, this was more apparent in families with multiple children. Angus referred to his other children not getting “their fair share of attention that they perhaps would have had”. Laura agreed with her husband on this point and admitted that she sometimes felt that she just hadn’t spend the time with her other children like she “should have done”. Angus’s comment about the “fair share” of attention and Laura’s feeling that she “should” have spent more time with the other children perhaps suggest that the parents feel guilty. Indeed, Laura goes on to talk about guilt:

“I’ve got the mother’s guilt of stretching myself too thinly with the family” (Laura)

<u>**Destructive force**</u>

A particular impact of the CFS/ME that parents talked about was the destructive affect that it had had on the family life. For Vicky this meant the family had been cut up and divided:

“the way we rejig the family is to cut it up and divide it up and participate or not participate so it’s been very destructive to family” (Vicky)

Similar experiences are reported by both Laura and Carol:

“One of us goes instead of all of us going because I didn’t want to leave Tom or, so we’ve sort of split ourselves up, and so yes it does divide you up and affect you” (Laura)

“I think the biggest impact probably has been that we can’t do things as a family like we used to so it means they go off and do things with their friends because we can’t leave Gemma and go out with them, we can’t go out together, or very rarely, so we tend to split up and do different things” (Carol)

Both Carol and Laura mention that they no longer engage in activities as a whole family because the needs of the unwell child are the priority. As well as whole families being divided up couples are also constrained. Andy said that he and his wife no longer *"go out together anymore"*, similarly Anna mentioned that she and her husband had *"less free time together"*. For Richard this meant there was *"less of an opportunity to relax as a family"*. Because the quality time that families and couples have available to them has effectively been *"squeezed" (Andy)*, the result was often an increase in *"tension" (Richard)*. Married couple Vicky and Angus reported that there was more *"friction"* between them because of the CFS/ME, Carol identified that they had *"had a few arguments"* as a direct result of the CFS/ME. Increased frustration was evident for Vicky who referred to *"tearing our hair out"* and *"often having a row"*. While Stephen felt that his relationship with his wife was *"ok"*, he conceded that because of the *"regimented regime"* that his daughter had to follow there was often *"a bit of strain here and there"* between himself and his wife. Debbie also felt that there was sometimes a *"strain"* between herself and her husband.

Parents also talked about the conflict between each other regarding their individual approaches or roles when managing their unwell child. Sally mentioned that she sometimes had disagreements with her husband about *"how we’re working together and who’s turn it is to be the one who gets up earlier than you would need to do to make sure she wakes up in time."* For married couple Brian and Sue there were clear differences of opinion on how to *"control and discipline"* their unwell son which often lead to *"conflict"*, and Brian felt that it was *"difficult to know what's right"*. Claire recounted an episode regarding when she had been taking her unwell son to the bus stop rather than letting him walk there on his own, to which her husband was reported as saying *"that's rubbish, you don't want to be doing that" (Claire).*

Resentment

“Occasionally, [his brother] has got cross and said, he sort of thinks everything is focused a lot on Mark” (Andy)

“The other kids are missing out. I mean they’re, they’re very good kids, the other kids too, our other children, but they resent it.” (Sue)

Eight parents commented on siblings of the unwell child not getting the attention that they felt they should have, and how these siblings inevitably felt resentment towards the child with CFS/ME:

“I don’t think she should be jealous, and she is, that’s the only word I can say, she’s jealous of Karen” (Barbara)

“[his sister] says we are giving all our attention to David and what about her and she’s been very very jealous hasn’t she?” (Sue)

Both Barbara and Sue commented about sibling jealousy of the unwell child. The resentment can sometimes stem from siblings perceiving that the unwell child gets “special treatment”, as mentioned by Sue, or that the unwell child is exempt from doing certain chores, as Stephen explains:

“she really resents a lot of what David’s going through and the special treatment that he’s had, which is hard” (Sue)

“The youngest daughter is the only family member that is really impacted and I think there is a hint of ‘why are you telling me to do things when Kate gets a rest?’” (Stephen)

Vicky refers to this situation in terms of "sibling rivalry", suggesting that perhaps it is normal for there to be conflict between siblings. However, Vicky seems to be suggesting that despite this it actually is not fair that the unwell child gets special treatment.

“There’s a sibling rivalry thing of ‘it’s not fair, I do all her jobs’ and he’s right, it’s not fair, that’s the bottom line isn’t it?!” (Vicky)

In addition to parents observing sibling resentment there was evidence that the parents themselves sometimes felt resentment too. For example, here Laura refers to having to leave the house just to get some time to herself:

"I feel I have to go out in order not to be around and I resent that sometimes" (Laura)

Theme 5:

Parental roles

This theme focuses on how differences between parents were revealed through their explanations of how they dealt with and managed their child's CFS/ME. This is explored through the following sub-themes: "good cop/bad cop" which highlights the different roles parents took in caring for their child with CFS/ME, and also how these roles could change over time; "it's a mother thing" focusses on how gender is used to explain differences in parental roles; "Parents a team" explores how some of the parents utilised their different roles in order to support each other to look after their unwell child.

Good cop/bad cop

It was apparent that there were sometimes clear differences in parent couples with regards to their approaches to care giving. Indeed, Richard commented that "we both work in very different ways". It seemed that the CFS/ME had the effect of bringing into focus the different attributes that the parents might have:

"Whereas Sue will prefer to tell him what to do, and I think that, that's, we have a different style of approach to it, obviously that's not, my style is not the same as Sue's on this one and so Sue will probably perceive that mine is a wishy washy approach and I will perceive that Sue's approach is leading to, whilst you might succeed in waking David up in that particular half hour you're losing the war." (Brian)

Brian is clear that his wife has a different style of managing their unwell son; the suggestion seems to be that his wife is perhaps firmer than he is, as evidenced by his perceived *"wishy washy"* approach. However, he also comments that while his wife may be able to achieve a small victory of waking their son, ultimately she is *"losing the war"*. This comment again highlights the ongoing conflict that is sometimes felt by families. Differences in parental roles is also evident for Barbara and George*:*

“I do think we are a bit different towards her though aren’t we? It’s like if she says to George ‘I don’t want to do my walk today‘ George is a bit lenient and doesn’t let her do it” (Barbara)

Similar to Brian and Sue, Barbara and George also fit into two contrasting roles; George seems to let Karen do what she wants, allowing her to avoid her daily walk (which is an important part of regular activity management). Barbara, on the other hand, seems to be a bit stricter:

“She says ‘I can’t go any further”, I say ‘no, you can go further karen, you’re going to go that little bit further!;” (Barbara)

Similarly Sally mentions that she felt that she and her husband had *"divided into the firmer and the softer one ever since it happened”*. She expands on this suggesting that her husband has a far more laissez faire attitude to healthcare than she does:

“my husband has never been proactive about trying to find out about what’s best to do, he waits for either the health service or myself to say ‘this is what we should, we could be doing’” (Sally)

A difference in understanding is highlighted by Debbie:

“my husband is probably not so tolerant as I am of Hayley, he has struggled in the first instance, he’s probably struggle more than I have to understand that Hayley isn’t physically capable of doing things” (Debbie)

Interestingly, Debbie was the one who had initially attended clinic appointments with Hayley, so perhaps this had an impact on the perceived difference in understanding between the couple. A difference in understanding may impact on what course of action to take next, as suggested by Andy:

“there’s times we’ve maybe had slightly different view on where we are or what we should do” (Andy)

Not only does the CFS/ME highlight how the parents had different ways of reacting or dealing with the young person, it also can change how they react and this was highlighted by 11 of the parents. Paul felt that he *"used to under-react to things that happened"*, though the CFS/ME had changed him in this respect. For Andy, an increase in his understanding of CFS/ME had changed the way he manages:

"I, from the outset, was pretty much 'lets get on with it, lets push on' and I have had to modify that feeling and understand, or try and understand the situation far more and I think as time's gone by I've got far more understanding." (Andy)

Claire talked about their roles changing so significantly it feels like they have been reversed:

"So I think how I'd say we've handled it is - I am, up until quite recently, I've been the pulling back on it and you've been the pushing forward on it and then, really strangely, we reversed didn't we?" (Claire)

This is not an experience exclusive to couples. Single parent Janet also talked about this.

"In the beginning [my ex-husband] didn't want to accept it, 'no, it's not that, and you've got to push yourself' and 'I can't believe you can't do this and you can't do that, and Zoe instead of just sitting there you should push yourself and do this and do that' and he was quite aggressive, but he's more passive now, actually he's more passive than I am" (Janet)

Janet observes that over time her ex husband has become much more "passive" with regards to managing their daughter. This is very similar to Andy's observation of how he changed over time.

It's a "mother" thing

There was a suggestion among 11 of the parents that the differences in the caregiving roles was partly due to gender. Carol felt that mothers provide more emotional support than fathers:

"I suppose mothers are more emotionally involved than fathers (laughs) anyway I don't know, I think she does feel that she doesn't get any emotional support from her dad" (Carol)

For Claire, this emotional involvement is seen as built into being a mother:

"your maternal instinct kicks in and you just want to look after him and make him better" (Claire)

Sue wonders whether fathers are better than mothers at managing their emotional responses:

"I suppose it's more of a male thing that actually you can just switch off and say 'well, that's that' you know 'he's decided, don't get stressed' I don't think you get stressed as I do" (Sue talking to Brian)

Meanwhile, Sally referred to an unspoken assumption that because she is the mother she should take the main caring role:

"it's almost always been me [who came to clinic with Jenny]. I don't know I think maybe my husband may have come to you occasionally. I don't know whether he's not so comfortable or whether it's just assumed that that's Mum's role" (Sally)

Fathers Paul and Andy also reflected on the fact that it was their wives rather than themselves who took the main caring role, which can have a negative impact i.e. Andy's wife is put under "quite a strain" because of her caring role.

"Anna has always played the mainstay role of caring for everybody's needs" (Paul)

"I think it's put quite a strain on Claire in that she has done the bulk of the caring" (Andy)

Simon's comment about being at work gives a practical explanation as to why the majority of caring falls to the mother:

"I don't notice it as much being at work, [my wife] is the main carer" (Simon)

Parents a team

“ We do have a team approach...working in the same direction really” (Anna)

Nine parents talked about being brought together because of the illness and they were working together as a team to help their child get better. Barbara felt that "really we work well together looking after Karen". Both Claire and Andy emphasised their ability to work together as a team being underpinned by their complimentary roles:

“We do think quite similarly, and I think we’re quite good at counterbalancing each other” (Claire)

“We most likely do tend to play off each other and, if one’s fulfilling one role, the other one of us fulfils another” (Andy)

A similar approach is described by Debbie, Paul and Laura:

“we just get on with it, we take on a different role or someone’ll do something for us to allow us to do something else” (Debbie)

“I think we’re quite good at trying to get what the other person needs and doing a sort of tag team, really, aren’t we, be flexible” (Paul)

“you’re trying to work more as a couple and a unit with the whole.” (Laura)

Sue felt that through dealing with the CFS/ME they had been brought closer together:

“over the last couple of years we’ve been far more united” (Sue)

For Brian, a positive impact of the CFS/ME had been that he and his wife were perhaps now working as more of a team than they had in the past:

“I think there’s been (pause) a strain but to a certain extent we will talk about David in the same way that we talk about all our children, umm, and that’s something, that is an issue

that we very much share (pause) I wouldn't say that is, so that's, the sharing problems is pretty positive for the relationship rather than negative" (Brian)

In addition to working as a team, six parents talked about how their relationship with their partner had improved since dealing with the CFS/ME:

"I think it's probably improved it in a way actually hasn't it?" (Claire)

"I think certainly I've become far better over the period of time of saying to Claire actually how I'm feeling and something so I think I'm a bit more open to that now. And maybe also honest with myself." (Andy)

"We do work well as a team and really in some ways you could argue it's [Anna: "more cohesive] strengthens that because we do get used to 'oh well you do that' and you know... we become better team workers sort of thing" (Paul)

Laura felt that the CFS/ME had given her the opportunity to become closer to her unwell son:

"there have been upsides to it, I suppose, now I think about it, this has probably made us very close I suspect... I'll come out of the experience definitely stronger in that sense" (Laura)

CHAPTER FOUR: DISCUSSION

CHAPTER OVERVIEW

This chapter reviews the results of the study and discusses the main findings in relation to the existing literature. The clinical implications of the study are discussed, followed by a methodological critique and recommendations for future research.

This research highlights the impact that CFS/ME has on families from the perspective of parents. They felt that life had become a constant struggle and often felt exhausted. They felt uncertainty, isolation and restriction. They talked about feeling like family life had become focussed on their unwell child, such that some expressed a need to escape from the situation. They also described how, as a consequence of managing their child's CFS/ME, family members could often feel marginalised and the illness could be seen as a destructive force. However, many parents also described working as a team and feeling that family relationships had benefitted in some way from the experience.

4.2 Unique nature of families with CFS/ME

CFS/ME is perhaps unique among chronic conditions in that there is still a large amount of controversy surrounding the condition, and there remains a lot of misunderstanding and disbelief. Essentially CFS/ME is currently “medically unexplained” whereas other chronic conditions can be “medically explained”. This was reflected by parents in the current study who expressed an ongoing uncertainty that wasn’t addressed by getting a confirmation of the CFS/ME diagnosis. Families with managing CFS/ME are also distinct from families managing other chronic conditions in the UK as there is not universal coverage of support services for families. In addition to this the individual experience of CFS/ME varies hugely, and therefore no one child’s experience will be the same as another’s making it difficult for parents to find common ground with other families, the illness journey can certainly be very lonely, uncertain and isolating, this is highlighted in the current research.

4.3 Synthesis of themes; the “thematic jigsaw”

Although the 5 main themes are distinct and separate, they fit together like a jigsaw to explain the impact of CFS/ME on these families. The first two pieces of the jigsaw that fit together are “long and difficult journey” and “uncertainty”; The “difficult journey” that the families found themselves on was embarked on without the benefit of a map to guide the way, which led to them questioning every aspect of their experience. Parents would often feel uncertain as to what they were dealing with, what the future held for their child, and also as to whether there was anything more they could or should be doing as parents. The next two pieces of the jigsaw fitting together are “isolation and restriction” and “focus on the unwell person at the expense of family life”; CFS/ME isolates and restricts families both physically and emotionally, turning the family in on itself. The fifth theme, or jigsaw piece is, in a way, a product of the other four themes; Roles are highlighted through this process.

4.4 Results in context of previous literature

Previous research exploring the financial impact of CFS/ME on mothers (Missen, 2012) found that mothers experienced a lack of understanding from others; marital tension; concern about their child's distress; concern about the impact on siblings and emotional distress causing physical symptoms. There are similarities between this research and the current study, particularly in the themes "Long and Difficult Journey", "Isolation and Restriction" and "Parental Roles".

This study is consistent with previous studies of childhood chronic illnesses that showed parents experienced loss, uncertainty, isolation and restriction. In addition, consistent with previous studies, parents in the current research also talked about their child's CFS/ME as being both a destructive and strengthening force in the family. However, in contrast to previous research, parents in this study talked about both resenting their child and guilt because they wanted to escape from the illness. What follows is an exploration of where the current research converges with and diverges from previous studies of childhood chronic illness.

4.4.2 Loss

Parents in the current study sometimes talked about their grief of "losing" their child. In some ways it could be argued that the impact of chronic illness is like coping with the death of a loved one. Previous research has certainly found that parents seem to experience a grief-like reaction for the loss of their healthy child (Mawn, 1999; George et al. 2006; Jordan et al. 2007; Rouf et al. 2012). A parent's reaction when faced with a diagnosis of childhood chronic illness has been compared to the stages of grief model developed by Kubler-Ross (Tamlyn and Arklie, 1986). Kubler-Ross theorised a 5 stage model to the adjustment to one's own impending death; denial, anger, bargaining, depression, and acceptance. The model has since been applied to the survivors of a loved one's death. Parents in the current study seemed to display anger, depression and sometimes acceptance, though denial and bargaining were not observed. Worthington (1994) has suggested that a traditional 'stage' based model is "inappropriately applied to all situations of loss and grieving, including when a child is sick or disabled" (pg 1). Because the symptoms of chronic illness are cyclic, it is difficult for families to adapt to the fluctuating absence and presence of the ill person. The alternative model of grieving which is 'chronic sorrow', first described by Olshansky (1962) and adapted by Worthington (1989).   More recently Gordon (2009) has defined chronic sorrow as "a normal grief response associated with an ongoing living loss that is permanent, progressive, recurring, and cyclic in nature" (pg 117). This "living loss" as described by Gordon (2009) makes it difficult for people to cope. When someone loses a loved one because they die they are no longer physically present, however with chronic illness the loved one who is "lost" is still physically there. This was highlighted by some of the parents in the current study who talk about how it feels as if their son or daughter is no longer the child they used to be. This is known within Family Stress theory (Boss, 1999) as "ambiguous loss'. Ambiguous loss refers to a situation where an individual's psychological or physical status is unclear. Boss and Coulden (2002) have suggested that when people are unable to obtain clarity about the status of a family member, they are often "immobilized" (pg 1352), or as Ellenwood (2007) describes it, "families dealing with chronic illness are in a frozen state hyper-focused on the chronic family member's dysfunction" (pg 267). This is certainly true of some of the parents' experiences shared in the current study; Paul expressed this succinctly when he said "life is stuck, everything's just pretty stuck". However, in contrast to this George commented that they were definitely not stuck. It is worth pointing out that Paul's daughter had been unwell for a few years whereas George's daughter had been unwell for only 1 year, perhaps this experience of being "immobilized" is related to the duration of illness. Parents in a later stage might feel stuck while parents in an earlier stage might not feel this way. This is something that could be explored in more detail in future research.

Ambiguous loss can be further complicated by Boundary ambiguity. Boundary ambiguity refers to the difficulty parents can experience when deciding how their child with a chronic illness fits into their family and in turn how they relate to their child. "A state when family members are uncertain in their perception of who is in or out of the family or who is performing what roles and tasks within the family system" (Boss, 1987, pg. 709)

Berge and Holm (2007) suggest that boundary ambiguity is a risk factor for psychological distress in parents of children with chronic health conditions. Boundary ambiguity has been significantly correlated with depressive symptoms in mothers of children with epilepsy (Mu et al. 2005. Mu, Wong et al. 2001). Certainly, parents in the current study reported feeling distressed with regards to their child having changed in some way as a result of being unwell.

4.4.3 Uncertainty

Parents interviewed in this study talked about experiencing a high level of uncertainty with regards to day to day life and the prognosis of their children. This is consistent with previous research. Uncertainty exists when details of situations are ambiguous, complex, unpredictable, or probabilistic (Babrow, Hines, and Kasch, 2000; Brashers, 2001; Jordan et al. 2007). Unlike uncertainty in acute illness, which tends to be localized in issues of diagnosis, treatment, and recovery, uncertainty in chronic illness involves broader aspects of a person's life (e.g. home, work) and influences daily routines and activities (Bayliss et al. 2003; Mishel, 1999; Yorgason et al. 2010). Long term illness can bring "continuous uncertainty" into parents' lives (Nuutila and Salantera, 2006). Cohen (1993) conducted a grounded theory study to explore the concept of uncertainty as experienced by parents following the diagnosis of chronic illness. She eloquently describes how rather than a diagnosis putting an end to uncertainty, it spreads to every aspect of life: "The uncertainty that follows the diagnostic announcement was found to be universal, multidimensional, and noncategorical. It transcends all life-threatening, chronic illnesses." (pg 144). In a synthesis of results from 8 qualitative studies Fisher (2001) found that the diagnosis of a chronic illness marks the end of the previously known world of parents.

Families managing childhood chronic illness have been described as having to deal with "multiple unknowns" and being faced with "predictable unpredictability" (Eggenberger et al, 2011, pg 287). Britton (1999) describes how parents of children with juvenile arthritis have "vague expectations of the future" and are "unclear about the prognosis of their child" (pg 541), this is certainly reflected in the current study with many parents feeling frustrated that they still did not understand CFS/ME. A good example of this is Brian's comment *"What's frustrating for ME organisations and for us is 'what is it?', are we, is it a physical disease? or is it a disease of the brain?".* Brian's comment also highlight the difficulty of living with medically unexplained symptoms (MUS). The experience of uncertainty is arguably a much more central concern for parents of children with CFS/ME because by it's very nature it is medically unexplained, and there are therefore multiple unknowns.

A feeling of powerlessness was something that many of the parents in the current study talked about experiencing at various points during the illness journey. This is something that is highlighted by Van Sheppingen et al. (2008) who found that parents experienced difficulty in knowing how they can best help their child feel better. Similar findings have been found in studies of parents of children with chronic pain (Jordan et al, 2007; Maciver, Jones and Nichol; 2010). Jordan et al (2007) describe one theme as “mummy couldn’t make it better” highlighting how parents felt powerless to help their child and thus felt they were not fulfilling their parental duty. Maciver et al (2010) report how parents felt they were a failure because they could not alleviate their child’s pain, with one parent describing this as getting a “great big “F” on my report card”. Parents in the current study talked about remarkably similar experiences. A poignant example of this was when David’s mother talked about a constant guilt that she was not doing her best to help her son get better. Similarly Andy felt that it was his responsibility to get his son better and, two years into the illness journey, he had failed to achieve this.

4.4.4 Isolation and restriction

A feeling of isolation was central to parents’ experiences. Part of this was the feeling that CFS/ME is an “invisible illness” meaning that other people were less likely to understand parents’ plight or believe them. This echoes the experiences of adult women in a previous study by Asbring and Narvanen (2002). They report that women with CFS/ME were regularly “challenged by others regarding the veracity of their experience of being ill” (pg 152) and that women felt their illness was called into question because of an absence of evident external symptoms. Other research has had similar findings, for example parents of children with life-limiting illnesses have been described as struggling to manage when outwardly their child may look completely healthy (Ware and Raval, 2007).

Isolation is a common experience for parents of children with a chronic illness. Morrison et al (2003) reported that these parents feel they are not part of “normal society” and that the “rest of the world” moves on without them. Ware and Raval (2007) found that parents of children with life-limiting illnesses felt alienated and abandoned. The feelings of isolation that families have when managing a chronic illness can perhaps, in part, be explained by the stigma that chronic illness creates. Asbring and Narven (2002) found that CFS and fibromyalgia could be particularly stigmatizing because of the lack of understanding and belief among others. This was talked about by the parents in the current study, when describing experiences when others had doubted that their child was really unwell, and how other people outside of the family do not understand what it’s like for them living with CFS/ME on a day to day basis. Other research has highlighted that families with a chronically ill member feel isolated, hopeless, and abandoned by their immediate and extended family members (Gabor and Farnham, 1996; Ware and Ravel, 2007; Timmermans and Friedin, 2007). Brewer et al (2007) reported that parents of children with Juvenile Huntingtons disease found that, as time went by, their child's condition would become "old news" and offers of support would gradually cease. Similar experiences have been reported by Ware and Raval (2007) who report that support from family and friends often lessens over time, with some parents even feeling that some people would actively distance themselves from them because of the child's illness. A lack of understanding and belief was not something that was exclusive to friends and family members of parents in the current study, they also highlighted experiencing lack of belief from health professionals. This is consistent with research into other chronic illnesses, for example Keenan et al (2010) report experiences of parents of children with Autism Spectrum Disorder (ASD) as feeling that that professionals are "against" them.

In addition to feeling isolated, parents in the current study talked about how the CFS/ME had restricted their lives. This seems to be a common experience of parents of children with chronic conditions; Gannoni and Shute (2010) found that in families coping with childhood cancer, chronic renal failure and type 1 diabetes there was a reduction in "spontaneity of lifestyle". Van Sheppingen et al. (2008) report similar experiences of parents of children with the chronic skin condition Epidermolysis Bullosa, with one parent having said "nine times out of ten" appointments would need to be cancelled, something that the parents in the current study could no doubt identify with. Indeed, Carol talked about how having made plans "9 times out of 10 it ends up being cancelled or rearranged". Parallels can again be drawn between the experiences of parents of children with chronic pain. Jordan et al (2007) found that a common experience of these parents was to feel like their lives had been restricted, one participant referring to life being "put on hold" (pg 53), much like Paul's comment in the current study of life being stuck.

4.4.5 Differences between parents

A particularly evocative comment was made by Sally; “I feel like sometimes I’m a single parent”. This perhaps highlights the frustration of a parent when they feel that the other parent is not pulling their weight. Indeed, there have been similar findings in research into other conditions; Van Sheppingen et al (2008) found that mothers of children with a rare skin disorder had a feeling of being on their own despite still being married. However, this may also reflect an actual imbalance of the burden of care between couples. Indeed, one father in the current study felt that his wife provided “the bulk of the caring” (Andy). This imbalance in the burden of care is something that has been documented in previous research. Timmermans and Freidin (2007) report that most mothers of children with asthma estimated the involvement of fathers in care to be less than 10%. One parent they interviewed said that a consequence of the father not being involved in the day to day care of the unwell child meant that the father did not “take it as seriously as he should” (pg 1356). However, Timmermans and Freidin did not interview fathers regarding the perceived seriousness of their child’s condition, or indeed what their opinion was on the caring roles. Ware and Raval (2007) offer a different perspective; they interviewed exclusively fathers of children with life-limiting illnesses, and they found that these fathers “perceived men as not allowing themselves to explore their emotional responses in the same way as women do” (pg 557) and that they attribute this to cultural expectations of what it is meant to be a man. One father is reported as saying:
“We are always taught from school to be very stoic and all the rest of it, and we don’t talk about issues or problems or anything else, and I think probably that works very much against us and that is a cultural thing” (pg 557).

This mirrors findings in the current study, in particular Carol‘s supposition that “mothers are more emotionally involved than fathers”, and Sue’s assertion that “it’s more of a male thing that actually you can just switch off”.

Although the current study seems to support the findings of previous research in the sense that mothers are seen as the primary care giver in the family, parents also talked about having complimentary roles and working as a team.

4.4.6 Parental Stress

All of the parents in the current study referred directly and indirectly to the stress caused by managing CFS/ME. Previous research has shown that parents of children with disabilities report higher levels of distress than those with healthy children. (Dyson, 1997; Kazak and Marvin, 1984; Kazak and Wilcox, 1984; Roach, Orsmond, and Barrat, 1999; Yau and Li-Tsang, 1999). However, there is evidence to suggest that the level of distress that parents face does reduce over time. Siklos and Kerns (2007) found that parents of children with autistic spectrum disorders experience a high level of stress at diagnosis. Similarly Dolgin et al. (2007) found that mothers of children with cancer experienced an increase in stress at diagnosis and consistently observed a steady decline of stress at 3 to 6 month follow up. As the current study was not longitudinal it is not possible to tell whether this is the case with these parents. However, some did mention that “the worst was behind them” (Sally) that they had “been through the worst bit” and that the service seemed to be the “light at the end of the tunnel”. This all suggests that, initially, the experience was extremely stressful, though as time has progressed the stress has perhaps reduced.

4.4.7 Destructive vs Restorative force

Dolgin et al. (2007) found that stress and depression rates were higher in mothers than fathers when managing childhood cancer. Rao et al (2004) reported that mothers were more likely to become neurotic, depressed, introverted, have a sense of lacking in self-confidence and become generally more anxiety-ridden than the fathers of chronically ill children or mothers of healthy controls. This may be related to differences in the roles that parents take when caring for a sick child. Research suggests that there are differences between parental involvement in caring for chronically ill children, Nagy and Ungerer (1990) found that mothers took more responsibility than fathers with regards to care of a child with Cystic Fibrosis. Derouin and Jesse (1995) found that siblings of children with Cystic Fibrosis and asthma reported that mothers were the main caregiver in the family. If mothers often provide the majority of the day to day care it is perhaps unsurprising that the impact on mothers is found to be more pronounced than on fathers. Missen et al. (2012) suggest it is possible that mothers are still viewed by society as the main caregivers or that it could reflect a disengagement on the part of fathers. However, the difficulty with previous studies is that many of the studies had a sample exclusively of mothers, rather than including evidence from the fathers’ perspective.

Many researchers have found marital satisfaction to be lower amongst parents of children with chronic health problems when compared to those with well children (Fisman and Wolf, 1991; Friedrich et al. 1987; Gath, 1972; McAlister et al, 1973; Shek and Tsang, 1993; Singhi et al. 1990; Hauenstein, 1990; Kazak, 1989; Sabbeth and Leventhal, 1984; Zimand and Wood, 1986; Crain et al. 1966; Reynolds et al. 1988; Walker et al. 1996). Quittner and Opipari (1994) found that couples caring for a child with Cystic Fibrosis experienced greater marital role strain than couples caring for a child without an illness. For some families, the increased strain leads to family breakup and divorce (Perrin and MacLean, 1988). There is a suggestion that couples are particularly vulnerable if they had experienced difficulties in the marriage prior to their child becoming unwell: "if pre-existing marital difficulties are present, parents can have problems resolving differences of opinion concerning illness management" (Sergent, 1983; pg 51).

However, much research into marital satisfaction and chronic illness is based on small nonrepresentative samples. Eddy and Walker (1999) suggest that perhaps there is a tendency to view child health status as having a uniformly negative effect on marriage because the data often come from families involved in some type of clinical service, and that these families are likely to differ from the general population.

In contrast to research that suggests marital satisfaction is lower in parents with chronically ill children, there are those that have found evidence to the contrary. Eddy and Walker (1999), for example, found that children with chronic health problems do not necessarily exert a negative effect on marriage, suggesting that "the belief that the stress associated with having a child with a chronic illness leads to marital distress and/or dissolution may be untrue" (pg 25). They argue that while having a child with a chronic illness is undoubtedly stressful, the concept of 'stress' should not be equated with 'distress'. Indeed, in their quantitative study looking at parental quality of life in parents of children with chronic illness, Storch et al. (2008) found that parental stress was higher for the parents managing their child's chronic illness. However they suggest that despite the high stress these families function just as well as families with no chronic illness present. Klinnert et al (1992) go even further and suggest that the stress of managing chronic illness may strengthen relationships: "although the presence of a special-needs child might affect some couples negatively, it might stimulate others to increase their relationship commitment." (pg 340). Clawson (1996) suggests that there is an opportunity for families to grow from their mastery of the chronic illness situation and Eggenberger et al (2011) describe how families are brought closer together during the experience, purposefully spending more time together in order to have more memories of happier times. The idea that chronic illness may strengthen families is further supported by Wray and Maynard (2005) who, in a survey of 209 parents, found that over 90% reported stable or improved family cohesion, and over one-third reported improved relationships between partners. In addition Jordan, Eccleston and Osborn (2007) found that parents of adolescents with chronic pain reported enhancements in their relationship with their adolescent. Ware and Raval (2007) report that fathers of children with life limiting illnesses often identified a sense of personal growth and a change in what, and how, they valued in life.

4.4.8 Role of fathers

This research highlights that there is a significant impact on families when one of the children CFS/ME. One particular issue that was highlighted, and could warrant further exploration in future research, is the role of fathers and the difference in how mothers and fathers cope with childhood CFS/ME.

A chronic illness changes the dynamics of a family, and in the case of CFS/ME, like many chronic conditions, the mother tends to become the primary carer for the child. What impact does this have on the father? previous research has suggested that fathers cope through withdrawal and being practical” (Mastryannopoulou et al. 1997, pg 827). Ware and Ravel (2007) suggest that it may be that fathers exclude themselves from alternative ways of coping with their emotional lives because, at a deeper level, they fear that if they look too closely at their emotional functioning they will be faced with their own vulnerability; and that this vulnerability may be overwhelming and that they will be unable to cope. It may be that there are perceived societal expectations of what fatherhood is that discourage fathers to talk about their families and their own emotional wellbeing (Ventner, 2011; Ware and Ravel, 2007). In different cultures the norms and expectations of the sociocultural environment can vary, for example in israel fathers are actively encouraged to partake in the care of their unwell child (Katz, 2002).

In previous research when fathers are included in the research they tend to be exclusively interviewed as part of a couple (e.g. Britton, 1999), while on the whole mothers are interviewed alone. This has a particular bearing on the data gathered, tending to centre around the mother’s experience. Even when parents are interviewed together the narrative tends to be dominated by the mother; It has been suggested that fathers have less input answering questions when both parents are interviewed together (Holm et al. 2008). This may be because the mothers tend to be the main caregivers and therefore the prevailing view within couples may be that the mother has the greater insight into the impact on the family. However, this does mean that often the father's voice is not being heard. A particular strength of the current research was that some of the fathers were interviewed alone, and therefore the fathers' voices can be "heard" alongside those of the mothers'. Though the current research has only really started to scratch the surface of the issue of how chronic illness changes a father's role in the family.

4.5 Implications of the current study.

The current research highlights that the link between CFS/ME and family relationships is by no means straightforward. There are both positive and negative experiences. In previous research some studies have found evidence that families continue to function well when a family member has a chronic illness (Bohachick and Anton, 1990; Donnelly, 1994; Rehm and Catanzaro, 1998; Sawyer, 1992; Youngblut et al. 1994), in contrast, others have documented negative outcomes for family functioning (Cornman, 1993; Ferrell et al. 1994; Kopp et al. 1995; Park and Martinson, 1998). There is a clear difference between families that reach breaking point and those that grow stronger. This could in part be explained by the resiliency model of Family Stress, Adjustment, and Adaptation by McCubbin and McCubbin (1989). The model suggests that families will vary on their levels of resilience depending on how they have managed adverse experiences in the past. Some of the parents in the current study talked about prior experiences of ill health in the family, in particular Laura referred to having looked after her unwell parents and thus feeling that she was well prepared to look after her unwell son. Mussatto (2006) suggests that families that have successfully coped with challenges in the past will be able to draw from prior experiences, and may find strength in their demonstrated ability to "get over things". In contrast, "a family that has very high expectations for their experience as parents, and little history of coping with stress, may perceive the diagnosis of chronic illness in their child as an insurmountable challenge" (pg 112). Britton (1999) found that 41% of families with a child with juvenile chronic arthritis believed that events that occurred pre-diagnosis strongly affected how they subsequently coped.

In addition to successfully managing prior adverse events McCubbin and McCubbin (1989) suggest that the most important protective factors that sustain families through stressful periods are family celebrations and traditions; family hardiness and family time and routines. It is therefore perhaps unsurprising that the families who took part in the current research seem to have been far more vulnerable to stress because the CFS/ME impacts on many of their established protective factors. For example, Barbara talked about not being able to attend family events, other parents talked about feeling like they had been forgotten or were "out of the loop socially".

Some parents in the current study talked about working very hard "behind the scenes" to keep everything as normal as possible for their unwell child. Some researchers suggest that there is less of a negative impact of chronic illness on some families because of a process of 'normalization' (Knafl, 2010). According to Knafl and Deatrich (1986), a part of the concept of normalization is that the family members define their lives as essentially normal and engage in behaviours to demonstrate their family's normality to others. Normalization seems to be a common coping strategy adopted by parents of chronically unwell children. It involves adapting to the illness in a way that makes life as "normal" as possible for unwell children. This process has been discussed with regards to children with HIV (Mawn, 1999); parents talked about keeping things "typical" for their child , and one mother is reported as saying "I don't think of her as being a kid with HIV, she's just a kid to me" (pg 205).

4.6 Strengths and limitations

This is the first study to look in detail the impact of CFS/ME on family relationships from the perspective of parents. A particular strength of this research is that fathers were recruited as well as mothers, this is unusual as mothers are over represented in the literature and often fathers are not present at all in much of the previous research into the impact of childhood chronic illness.

A further strength of this research regards the methodology. The first four interviews were double coded which helped to ensure rigour, and all themes were checked by an expert in the field which helped to increase the validity of the findings. Overall quality was ensured by following the guidelines recommendations by Elliot et al. (1999).

A limitation to the current study is that participants were recruited from a specialist, tier 3 service, meaning it is likely that they represent the more complex end of the spectrum of

CFS/ME (Garralda et al. 1999; Rangel et al., 2000). This indicates that the findings from this study may not be generalisable to families seen in general practice or community settings. The findings may also not be generalisable to CFS/ME services in other parts of the country. A further limitation is that in 10 of the families the unwell “child” was a teenager, which means that the results may only be generalisable to other families with unwell teenagers. Only 12 out of 20 families returned their consent forms. This may suggest that there is bias in those recruited. There is no evidence to suggest that they had more or less problems, though a comparison of children of the parents interviewed in this study suggests that compared to the overall cohort of children seen by the specialist CFS/ME service with regard to scores on the standard outcome measures for the CFS/ME service there was no significant differences, suggesting that this sample was representative (Appendix v).

Because the interviewer had an ongoing relationship with regards to the care of the participants' unwell children it is likely that this will have had an impact on the recruitment and data collection. Parents may have felt that their child’s care would be adversely affected if they chose not to take part in the study and therefore felt duty-bound to take part, despite steps to reassure potential participants that this would not be the case. Also, if it were the case that parents felt a duty to take part in the research one would assume a higher recruitment rate than 60% would have been achieved. One could argue that because the interviewer was also the main clinician working with the families, parents may have been trying to present an overly negative picture of family life in order to elicit more support or may have wanted to present an overly positive picture to avoid potential criticism. Although the interviewer was aware that this could potentially happen, it was felt that parents were being as open and honest as possible, and many parents voiced an extremely favourable opinion to the research because they felt it would help others to understand about the wider impact of CFS/ME on the family. A potential benefit of the dual role of the interviewer could be that the parents already felt comfortable talking to him because of previous clinic appointments and therefore engendered a more open and honest exchange. A particular issue that parents raised was the difficulty helping people outside of the immediate family to understand CFS/ME. Therefore having an interviewer who the parents knew fully understood CFS/ME may have put them more at ease than if they had been interviewed by someone who may not have fully understood the condition

4.7 Recommendations

This study describes the impact of childhood CFS/ME on families, from a parent's perspective, and their experiences of managing it. It is clear that CFS/ME can have a significant impact; parents can feel a profound sense of loss of the child they felt they once had, they will probably have ongoing feelings of uncertainty, they may also have conflicting feelings of resentment and guilt. Clinicians need to be aware that parents of children with CFS/ME may be experiencing some or all of this at any given time, and therefore parents may need to be offered additional support both individually and possibly in a group setting. From an individual perspective parents could be given more information about CFS/ME and it's prognosis which would help to demystify the condition. This seems particularly important considering that parents in this research felt they did not understand much about CFS/ME. In this way an individual approach would help to provide parents with a personalised "road map" to help guide them on their "journey" through CFS/ME. In a group setting parents may be able to gain support from other parents experiencing similar issues to themselves, which in turn may help to reduce feelings of isolation and normalise their experience.

4.8 Unanswered questions for future research.

Because there were differences between families in the current study with regards to perceived impact there could be two possible hypotheses that could be explored by future research. Firstly it could be hypothesised that the impact of CFS/ME differs depending on type of family; single parent family, "traditional" two parent family, families with extended family support structures. To address this hypothesis future research should include a larger cohort of parents, including other family members such as grandparents and siblings. It would also be useful to include other groups such as non-English speaking parents and same-sex couples. Expanding the research parameters in this way would also help identify whether the current findings are generalisable to a larger cohort of parents. The second hypothesis that could be addressed by future research is that the effect CFS/ME has on the family changes over time. To do this it would be useful to adopt a longitudinal design. This would help to inform practice in that it it would make it easier to tailor the right sort of support package for the most vulnerable families at the right sort of time.

Although fathers were included in the current research it was the mother's voice that dominated much of the "impact talk". This may reflect the imbalance of the burden of care, it may also reflect the findings of Ware and Raval (2007); specifically that culturally fathers feel unable to voice and explore their emotions in the same way as mothers. It would be interesting to hear more from fathers - future research could focus exclusively on fathers and compare their experiences to the findings of research on mothers. If fathers do feel unable to voice their feelings with regards to the stress of managing chronic illness then it would seem appropriate to allow fathers a forum to be able to open up and share how they feel.

It is not clear whether those parents who declined to take part in this research would have had the same issues and experiences as those that did participate. Future research needs to address this; perhaps by increasing the flexibility of data collection techniques, for example parents may feel more comfortable conducting an interview over the phone, or writing down their experiences anonymously. In addition, future studies should investigate whether known strategies to support parents of children with other chronic health conditions such as group and individual support can be helpful to parents of children with CFS/ME.

4.9 Conclusion

In conclusion, this interview based study has explored parents' experiences of having a child with CFS/ME and the impact on the family . It provides a rich pool of findings which expand on the small body of research into the psychosocial impacts of CFS/ME. This research clearly suggests that parents feel there is a significant impact on the family, both in a negative and positive sense, and that over the course of the illness journey the needs of a family will change. With this in mind it seems that in addition to supporting the child/ young person with CFS/ME there is a need to identify what needs parents and families have, and provide extra support and information where needed.

CHAPTER FIVE: REFLECTIVE CHAPTER

CHAPTER OVERVIEW

This chapter is an opportunity to present a critical account of the development of my own professional practice across all the competencies of the D Health programme. The actual evidence relating to these competencies forms the Doctoral Portfolio and the Research Thesis. For this chapter I have chosen to reflect on challenges that i have experienced over the 4 years of the doctorate in the following competency areas: Professional, Research and Teaching and training.

I see the Health Psychology Doctorate as a journey that I embarked on in October 2009. This journey has been a difficult one, during which I have experienced challenges both professionally and personally. On reflection I feel that the challenges I have faced have helped me to develop into the rounded health professional that I feel I am today. To structure my reflections I have chosen to use Rolfe et al's (2001) framework (figure 1) for reflective practice. I have chosen this particular framework because it is often used in healthcare settings and it's simple structure helps to get the most out of the process of reflection.

Figure 1: Rolfe et al's (2001) framework for reflective practice

Descriptive level of reflection	Theory - and knowledge - building level of reflection	Action-orientated (reflexive) level of reflection
What …	**So what …**	**Now what …**
… is the problem/difficulty/ reason for being stuck/reason for feeling bad/reason we don't get on/etc., etc.?	… does this tell me/teach me/ imply/mean about me/my patient/others/our relationship/ my patient's care/the model of care I am using/my attitudes/my patient's attitudes/etc., etc.?	… do I need to do in order to make things better/stop being stuck/improve my patient's care/resolve the situation/feel better/get on better/etc., etc.?
… was my role in the situation?		
… was I trying to achieve?	… was going through my mind as I acted?	… broader issues need to be considered if this action is to be successful?
… actions did I take?	… did I base my actions on?	… might be the consequences of this action?
… was the response of others?	… other knowledge can I bring to the situation? · experiential · personal · scientific	
… were the consequences · for the patient? · for myself? · for others?		
… feelings did it evoke · in the patient? · in myself? · in others?	… could/should I have done to make it better?	
	… is my new understanding of the situation?	
… was good/bad about the experience?	… broader issues arise from the situation?	

Research

I began the health psychology doctorate having completed research both at undergraduate and post graduate level. I had also had a paper published of research into memory problems experienced by children with Chronic Fatigue Syndrome. It was the research element of the doctoral programme that I was most looking forward to.

What...

At the beginning of 2010 the 5 year relationship with my long-term partner began to break down. In March 2010 we ended our relationship, though continued to live together as flatmates because my ex-partner could not find anywhere to live. My ex-partner moved out at the end of July 2010. For much of 2010 I found it extremely difficult to concentrate on my studies because of the trauma and stress that resulted from the breakup. There were periods of time during those months when I found it impossible to work on my doctoral studies due to the emotional turmoil I experienced. I feel that if the relationship with my partner had not broken down I would have been able to devote much more time and effort to my doctoral studies, possibly avoiding the outcome whereby I failed on my first submission of my systematic review. I have since been able to use the experience to my advantage as it has given me a deeper insight into how much emotional turmoil can impact on day to day functioning. In my clinical work I regularly work with children and young people who are extremely distressed because their CFS/ME sets them apart from their peers and often means that they cannot take part in activities that healthy young people may take for granted. In a small way, having experienced my own emotional turmoil and it’s ongoing effects on my work, I have a greater appreciation of how the emotional impact the condition can be just as debilitating as the physical impact.

So What...

The experience also highlighted for me what works with regards to coping strategies, and what can become counter productive. For example, before I submitted my systematic review I had reached a point where I felt that i may not be on the right track with regards to what I was writing, however I had reached a point where I was scared of what sort of feedback I may receive, I worried that I may be told that I had done everything wrong and that my quality of work was substandard. As I ploughed forward with my work these voices just became louder, and when I began to feel stuck and would probably have benefitted from some feedback I blindly pushed forward. What I ended up submitting was, on reflection, more like an extended first draft of my systematic review, and it is not surprising that I failed this first submission. I remember feeling disheartened and upset that I had failed, though this did allow me to get the feedback that I needed, which did not include being told that I was wrong or had produced something ‘substandard’. The feedback was useful and constructive and helped me to build on the first draft to get to a point where the systematic review was at an acceptable standard to pass.

Now what...

It is easy to see now that I should have sought input from my supervisors and tutors at an earlier point, though at the time the level of stress I experienced made it feel impossible to seek guidance. Reflecting on this particular challenge I am reminded of something I regularly talk to patients about; It is often easy to give others advice, but it can sometimes feel impossible to give ourselves similar advice. With this in mind I often suggest that it can be helpful to mentally take a step back and to imagine what you would say to a friend if they came to you with a similar problem as your own. The advice that you would give them can then form the basis of what you should do about your own problem. This is something that I often have to remind myself in situations where I may feel stuck or overwhelmed by the seeming enormity of a task. Since the challenge of completing the systematic review I have tried to seek guidance more regularly when I feel that I am at a point where I am starting to feel stuck, sometimes it feels like I am actively having to fight against the impulse to bury my head in the sand and carry on regardless.

Teaching and training

This has been a particular area of growth for me. I have never felt particularly comfortable with the prospect of having to present to a group of people. At school I would actively avoid situations where I may have to talk infront of a group of people, this was also the case at undergraduate level university. I recall one excruciating experience in the second year of my Psychology BSc when I had to present the case for and against medication use for mental health problems to my psychology cohort. I remember visibly shaking, sweating and feeling decidedly unwell during the whole experience. Thankfully since this experience I have gradually gained confidence through subsequent presentations at university, conferences and through my clinical work.

What...

Although I felt confident in my abilities when it came to the teaching sessions that I conducted as part of the doctorate, I still did not feel completely at ease with the idea of presenting to large groups of people. In 2010 I taught a session on pain to a large group of nursing students at UWE. This was a real challenge as at 100 students this would be the largest group I had ever presented to. I therefore went into the situation with a higher than normal level of anxiety.

So what...

With my initial trepidation I remember trying to reflect on the fact that I am not alone with my feelings of nervousness as my peers on the doctoral programme had also talked about their apprehension with regards to the possibility of teaching a large group. Looking back now I feel that the level of nerves I felt were appropriate and they did not hamper my performance to any great degree, however some of the student feedback suggested that I spoke too fast, this may be a reflection of my nerves. I think that the nerves I felt may also be partly to blame for my apologetic manner where I repeatedly tell the students that I am “not an expert in pain” at the beginning of the teaching session. At the time I think I did this because I wanted to engage with the audience by demonstrating that I recognized that the students, some of whom were mature students with many years more experience than myself, may know more about the subject matter than me. I think that this approach may be appropriate if I was a student giving a presentation, however as I was in the lecturer role the audience has certain expectations that you will know what you are talking about. By admitting that I am not an expert and confused by the subject matter I risk losing the confidence of the audience in my abilities as a lecturer and undermining my own professional status. Dr Morris, who was observing my lecture, also made the point that I need to make sure I don’t let the audience know I might be inexperienced with a subject. I feel that when I am talking about the practical “real life” aspects of pain that I am most confident underpinned by a solid knowledge base. As I work in a clinical setting and regularly have to explain various practical information to patients I feel that this is probably why I sound more confident and at ease when slipping into this role as a lecturer.

Now what...

Overall I felt that the lecture went well, I was only a little nervous to begin with. I forgot to get the group to discuss in pairs the a case study that I had prepared, instead just asked them to read the case study and then asked them about it. I think I was a bit too honest at times – for example I told them that I didn't know what the "neuromatrix" was and that it was confusing for me – maybe shouldn't have spent so much time explaining this. For future lectures I would still be honest if I don't have extensive experience in a certain area, however I will avoid saying things like "I am not an expert in the area" or "this confused me".

I sometimes forget that although there may be those in the audience with a lot of experience in their own line of work I also have a lot of experience in my line of work. I should be mindful of this when conducting any future teaching sessions; It would be better to acknowledge that a certain topic or paper may be complex, but not undermine my own knowledge or experience in the process.

I would say that the teaching sessions I conducted through the doctorate were a challenge for me, and that subsequently my confidence in my own abilities has grown. I still feel an occasional pang of fear when I'm required to present to a room full of people, however I am far from being in a position where I feel paralyzed with fear. Since the teaching and training module in the first year I have continued to provide training support for other professionals through my clinical work. Recently this has involved co-facilitating a conference workshop for teachers. I have also had to opportunity to return to Bath university every year to provide a teaching session to the MSc cohort regarding Health Psychology in practice.

Professional Skills

Over the course of the doctorate my clinical caseload increased, which proved a challenge with regards to managing the ongoing pressure of the doctorate workload. Monthly meetings with my workplace supervisor have been essential in making sure that the balance has stayed in check. Essentially, when either the pressure on the clinical or doctoral side has become too great I have been able to discuss this with my supervisor and we have used the opportunity to put together an action plan in order to manage the increased workload. This has sometimes meant limiting new referals to me, I have also been able to take study leave when I’ve needed additional time to concentrate on my studies.

What...

One particular incident that sticks in my mind was in 2012, it was an uncharacteristically warm for April, almost to “heat wave” levels, I was invited to attend a meeting with the two psychologists in our team and two other senior psychologists at the hospital to discuss outcome measures and the future direction of psychology services in the hospital. Because of the weather I opted to wear shorts and sandals with a short sleeved short and tie. Although this would not be my normal attire if I was seeing patients I decided that as I would not be seeing patients that day It would be fine to look slightly more casual, I would also not feel too uncomfortable in the hot weather. I attended the meeting, and was told by the team manager later in the week that there had been a complaint about my attire.

So what...

I was initially feeling confident that there was no problem with what I was wearing for two main reasons: first of all since it was a non-clinical day for me; second of all I had assumed that the meeting was not going to be formal. As soon as I met with my two collegues before the meeting I doubted my choice, their reaction was that of surprise that I was wearing shorts. However, because they knew that I had never worn shorts to work before they understood that this was just a one off. At the meeting I felt uncomfortable with my choice as one of the other psychologists was wearing a suit, and the meeting had a much more formal atmosphere than I had been expecting. I felt underdressed for the occasion, but felt that despite my appearance I still provided a contribution to the meeting, and I felt excited at the prospect of helping to develop the psychology service. I was mortified when I received the complaint later in the week,though I did feel that as we had all been at the meeting together and were collegues in the sense that we all worked in the same hospital it could have been dealt with in a less formal manner. I felt that the other psychologists could have mentioned to me either at the beginning or end of the meeting that it wasn’t appropriate to wear shorts. I was worried that my faux pas had reflected badly on me as a health professional in that I was seen not to be smart and professional, and that it also reflected badly on our service. I was able to talk about all of this with both my supervisor and my boss, who passed this on to the boss of the other team. This went no further, though I feel I learnt some valuable lessons from the experience.

Now what...

Firstly I feel that this incident highlights that sometimes I have a tendency to under plan for a situation, and rely on assumptions rather than getting all the information I need to make an informed decision. For example, I assumed that the meeting was to be informal, which led to my decision to dress in an informal manner. I also assumed that the other psychologists, who I had not previously met, would react in the same way as my collegues who I see on a regular basis. The fact was that the other psychologists had not met me before and therefore this first impression of me as being informal may well inform their whole view of me. Since this incident I have made sure that regardless of the work situation I should not present myself in a casual manner, there are also staff regulations regarding dress code for therapeutic staff that I have since become familiar with.

The other issue that this incident highlighted was that I sometimes need a bit more preparation when entering into new or unfamiliar situations. I need to make sure that I don’t rely on assumptions that I may have; If I think a meeting is to be casual I should still dress in a professional way. If I have not met other people who are attending the meeting I should not assume that they will know what I am like as a health professional.

Conclusion

By undertaking the Health Psychology doctorate I feel that I have been able to build on my existing skills within research, clinical and teaching settings, further apply my understanding of psychological theory from a health perspective and gain further insight into the role of Health Psychology in today’s society. Moving forward I feel the doctorate will help to broaden my professional horizons in the future, while also giving me extra confidence in my own skill set when working alongside other health professionals. I do not see the completion of the Health Psychology doctorate as the end of my journey; rather I see it as an opportunity to develop into a flexible and dynamic health professional.

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The impact of managing a child's
Chronic Fatigue Syndrome/ Myalgic
Encephalopathy (CFS/ME) on family relationships

Andrew Haig-Ferguson

Appendices

Appendix i

Participant Information Sheet

CFS/NHS/PAEDIATRICS - Specialist help for ME.

A qualitative study investigating the impact on family relationships for families with a child who has Chronic Fatigue Syndrome/ME

INFORMATION LEAFLET

We would like to understand whether having a child with CFS/ME impacts on family relationships and how this is perceived by carers

Before you decide to take part it is important for you to understand *why* the study is being done and *what* it will involve. Please read this leaflet carefully. You can talk about it with your family, friends, doctor, or us if you want to.
Ask us if there is anything you don't understand or if you want more information. Take time to decide whether or not you want to join in.

Thank you for reading this!

Reasons for the study

- In a previous study families identified that they would have liked to be asked about how having a child with CFS/ME impacts on family relationships.
- We want to find out what the impact is on family relationships.
- Understanding the impact on family relationships in families with a child with CFS/ME is an important part of developing appropriate support and treatment for children within the context of their family.
-

What are we asking you to do

We will arrange a time that is convenient for you to talk to you for around 30 minutes about the effects of caring for a child with CFS/ME on family relationships. The interview will be recorded with an audio recorder.

Your privacy

It is very important that all the information you give us is kept as anonymously as possible. We will write down the things that you say from the tape recording and take out any details linking the recording to you and nobody will know that it was you. The audio recordings and memory chips will be destroyed once we have written down the information. The copy of what you said in the interview (the transcript) will be linked to you and your child via an ID code. All personal details or lists that could identify you will be kept secure in locked cabinets in locked offices or on password protected secure NHS computers. If you would like more information on this please ask one of the research team (contact details below).

Consent

We have to be absolutely certain that you are happy to join in this study, so if you say you are, we will ask you to sign our consent form. Even if you do sign it, you will be free to withdraw at any point. Just tell us if this is the case. Whether or not you wish to participate, your child will continue to receive exactly the same service from the clinical team.

Benefits of joining in

There are unlikely to be many benefits for you. Taking part in this study may help other children and young people with CFS/ME and you may find it beneficial to talk about your experiences.

Problems with joining in

You will need to spend 30 minutes talking to us. You may find it difficult thinking about what has happened during your child's illness and the problems you have faced. If you have any problems with this study, please speak to Andy Haig-Ferguson, Dr Esther Crawley or any member of the team that you know. You will be able to complain to the NHS in the usual way if you were not happy with the way the study was carried out.

Who is organising the study?

This research is organised by Andy Haig-Ferguson, a Health Psychologist who is working with Dr Esther Crawley who leads the Paediatric CFS/ME Research team at the Royal National Hospital for Rheumatic Diseases (also known as the RNHRD/Mineral Hospital) in association with the University of Bristol. Andy Haig-Ferguson is undertaking this study on a voluntary basis as part of his doctoral studies in Health Psychology.

Ethical Approval

The study has been approved by the South West 4 Research Ethics Committee. It has also been checked and approved by the RNHRD research committee.

Contact / Further Information:

If you have any questions or problems then please speak to:

- *Andy Haig-Ferguson – Health Psychologist*
- *Dr Esther Crawley* - Paediatric Consultant and Head of the Paediatric CFS/ME Service.

Both Andy Haig-Ferguson and Dr Crawley can be contacted at the Specialist CFS/ME Service for Children and Young People, Royal National Hospital for Rheumatic Diseases NHS Foundation Trust, Upper Borough Walls, Bath, BA1 1RL
Tel: 01225 465941

Appendix ii

Participant Consent Form

A qualitative study investigating the impact on family relationships for families with a child who has Chronic Fatigue Syndrome/ME

Please complete the two copies of this consent form provided, return one to a member of the research team.

Please only tick ONE of these boxes:

I/We **agree** to join in the study ☐ OR I/We **do not** agree to join in the study ☐

I/We agree that, if I am/we are interviewed, the interview will be tape-recorded

If you agree to take part, please fill in the information below:

Please tick box if "yes"

I/We have read the leaflet about the study. I/we understand what the study is about and have had the chance to ask questions. ☐

I/We understand that it is my/our choice whether or not I/we agree to take part in the study and that it is ok for me/us to withdraw from the study at any time. ☐

If you agree to take part, please fill in the information below:

Your name

…………………………………………………...

Your signature

…………………………………………………..

Today's date

……../………/20……….

Your address: ....

....

....

Post code: ....

Email:..

Telephone:…………………………………

This consent form will be kept in a locked filing cabinet in a locked office in the University of Bristol.

An encrypted password protected database will be created to store your details. This will be kept on a secure NHS server in the Royal National Hospital for Rheumatic Diseases.

All interview transcripts will be linked to you and your child via an ID code on separate lists. The list linking the code to you will be kept in the University of Bristol with the consent forms and the list linking the code to your child will be kept in a locked cabinet in a locked office in the RNHRD.

Thank you!

Appendix iii

Summary of Research Findings

The impact of managing a child's Chronic Fatigue Syndrome/ Myalgic Encephalopathy (CFS/ME) on family relationships

Research study by Andrew Haig-Ferguson

Completed as part of the degree of professional doctorate in Health Psychology.

Research summary

Although there is a large body of research literature concerning the impact of childhood chronic illness on family relationships, very little of this has specifically focussed on the impact of childhood Chronic Fatigue Syndrome/Myalgic Encephalopathy (CFS/ME). The study aimed to investigate the impact of having a child with CFS/ME on family relationships from the perspective of parents. This study took a qualitative approach using Semi-structured interviews to explore 18 parents' experiences of having a child with CFS/ME and the impact this has had on family relationships. Interview data was transcribed and analysed using thematic analysis. The analysis led to the identification of 5 main themes: "Long and Difficult Journey", "Uncertainty", "Isolation and Restriction", "Focus on the unwell person at the expense of family life" and "Parental roles". They felt that life had become a constant struggle and often felt exhausted. They felt uncertainty, isolation and restriction. These parents talked about feeling like family life had become focussed on their unwell child, such that some expressed a need to escape from the situation. They also described how, as a consequence of managing their child's CFS/ME, family members could often feel marginalised and the illness could be seen as a destructive force. However, many parents also described working as a team and feeling that family relationships had benefitted in some way. The results of this study are in line with findings of other studies into other chronic illnesses. In addition, this is the first study to focus exclusively on the impact of CFS/ME on family relationships from the perspective of the parents, and is unique in that there is a dual focus on both mothers and fathers. Clinicians should be aware that parents of children with CFS/ME may need additional support, such as information as to what to expect at various stages of the illness and where to access the relevant support.

Appendix iv

Topic Guide

Families with a child who has CFS/ME – Topic Guide

Welcome, thanks, check consent form, check consent to audio-recording. Explain confidentiality. Define ground rules if interviewing a couple (allow each to have a point of view, respect each others opinion, explain need to speak one at a time otherwise the audio recording will not pick up).

Topic: The effect of CFS/ME on family relationships

Prompts: Has there been an effect? What effect has there been?

Topic: The effect of CFS/ME on the parent's relationship

Prompts: Has CFS/ME changed your relationship with each other? Is the relationship better/worse? Why do you think it changed/didn't change?

Topic: Dealing with CFS/ME

Prompts: How do you think you both handle the illness? Philosophy/experience of illness/ underlying roles in families. Have you had any support from professionals? Could we have done anything to support you?

Topic: Things that made the experience harder

Prompts: Where there particular things that made things harder/more difficult? Time at home/money/lack of money/who went to the appointment/different understandings of the illness

Topic: Things that made the experience easier

Prompts: Have you found any strategies that have helped?

Thank for taking part, answer any questions

Appendix v

Comparison Table

Characteristics of children in current study compared with all other children with CFS assessed by the specialist service during the same period (2008-2012) who were in the same age range (11-17 years)

	Study children (n=12 unless otherwise indicated)	**All other children with CFS (n=445 unless otherwise indicated)**	**P-value***
	Mean (SD)	**Mean (SD)**	
Age (years)	13.5 (1.6)	14.3 (1.7)	0.13
Female	9 (81.8%)	325 (73.0%)	0.52
	Median (IQR)	**Median (IQR)**	
Time to assessment (months)	9 (6 – 18)	12 (8 – 24), n=382	0.06
Chalder Fatigue Score (0 – 33)	26.5 (25 – 30), n=10	26 (22 – 28), n=423	0.10
SF-36 physical function (0 – 100)	50 (30 – 70)	55 (35 – 70), n=416	0.75
Anxiety (SCAS) (0 – 90)	27 (20 – 51)	27 (17 – 41), n=408	0.32
No. of Symptoms (0 – 14)	9 (7 – 10)	9 (7 – 10), n=444	0.86
Anxiety (HADS) (0 – 21)**	10.5 (8 – 13), n=10	8 (5 – 12), n=392	0.18
Depression (HADS) (0 – 21)**	10.5 (5 – 16), n=10	7 (5 – 10), n=393	0.16
Visual Analogue Pain	70 (24 – 76)	55 (24 – 73), n=402	0.47
School attendance past week	**n (cumulative %)**	**n (cumulative %)**	
None	3 (30.0%)	109 (25.7%)	0.68
10%	1 (10.0%)	37 (8.7%)	
20%	2 (20.0%)	28 (6.6%)	
40%	0 (0.0%)	65 (15.3%)	
60%	2 (20.0%)	80 (18.8%)	
80%	1 (10.0%)	73 (17.2%)	
100%	1 (10.0%)	27 (6.4%)	
Not applicable	0 (0.0%)	6 (1.4%)	

* Student's t test for comparison of means, Kruskal-Wallis test for comparison of medians, Chi-squared test for comparison of proportions

** Only completed by patients age ≥ 12 years

Appendix vi

Theme list with corresponding quotes

Appendix vi: Theme list with corresponding quotes

Long, difficult journey	
Up hills, through tunnels	Parent, page
It does take time doesn’t it really. It does take time. It’s a slow process	George, 10
I actually realised the other day that we’re coming up to her being a year being poorly	Barbara, 10
It’s a frustrating experience because you think you’re getting somewhere and you take a step back and you think ok more gently, more gently, and you don’t know how long a time you’re going through	Sally, 8
because you don’t know how long a time you’re going through you see glimmers of light but then it’s just a little reflection and it’s not the end actually.	Sally, 8
It’s a bit like walking towards the top of a hill and you think you’re at the peak and then you get to that bit you see there’s another bit beyond, and another bit beyond, I think we’ll get on top of it eventually but it’s the fact, it’s a journey of an unknown length that makes it difficult.	Sally, 8
But we’re still in this situation and we didn’t expect ever to be in the situation still	Sue, 17
it’s going on so long	Sue, 18
it’s a drag at the moment, it’s going on too long	Sue, 18
I think maybe an appointment with the parents without the child after about a year to sort of say listen guys, you mm, this would only be appropriate if it was actually true, to sit them down and say look guys, you know, you might be here for the long haul,...	Brian,18
it’s a bit of a rough journey	Anna, 11
You’re in this long, long tunnel	Paul, 16
I’ve had enough of this, why hasn’t this ended?	Andy, 8
It helps to have someone to guide you through	Claire, 18
From there being a light at the end of the tunnel suddenly there was another rockfall to clear away	Stephen, 1
It takes a long time to appreciate that it’s not just a cold or a virus	Stephen, 2
[my husband] is fully on board with us now for the long haul	Debbie, 3
it’s how long the journey is	Debbie, 5
I give her a few little rewards or a few little milestones to get through to the next point	Debbie, 12
I can understand where she was but it’s nice to know that’s where she is now	Debbie, 12
You were a bit of a proverbial light at the end of the tunnel	Angus, 16
It’s insidious, it sort of creeps up on you really and then you realise, you know, all these things have passed by, it’s years suddenly isn’t it? Weeks become months, months become years and you realise that lots of things, landmark things slip by really.	Angus, 17
A constant struggle	
Bringing up children is hard enough without, without this added, added kind of burden	Sue, 6
We’re often in conflict if we try and enforce on David what we think is best for him that will not always lead to a situation where i think is a long term win because he gets upset	Brian, 1
Life’s hard enough without having this on top of it	Brian, 9
the battle’s in the mind now	Brian, 18
I think it’s had a huge impact on our relationship without us being able to put a finger on exactly what, what it is, umm but I’ve just, I think that I, that I find the family harder work than I think it should be	Sue, 9
Implementing your advice, you can’t do that for us, we have to do that, and that’s, that’s the bit that’s really difficult	Sue, 17

Appendix vi: Theme list with corresponding quotes

🟧	I suppose it’s just that... it’s difficult isn’t it?	Paul, 7
🟧	It can be quite tiring and the less energy hours they have the more tiring it is, because of that reason of just trying to find, trying to fill the time with things. It’s really hard sometimes	Anna, 6
🟧	trying to arrange the structure of every day to get the right amount of red and green and whatever time and she can be quite challenging on some of that sometimes	Claire, 3
🟧	Even something like grocery shopping becomes a bit more of a challenge doesn’t it?	Andy, 16
🟧	It’s been a challenge	Andy, 11
🟧	we’ve staggered through but it’s been extremely hard	Vicky, 5
🟧	it just makes everything slightly more complicated	Richard, 6
🟧	It has added a certain something which is, you know, maybe harder to process	Richard, 5
🟧	I’m not knocking how [my husband] came back [from clinic] and was like ‘oh we’ve got to do this and we’ve got to do this’ and i’m kind of going ‘yes, we’re trying to but actually it’s really complicated	Vicky, 5
🟧	It’s then the logistics, how do we fit in, how do we get it all to work and fit together?	Vicky, 9
🟧	If we’d both been working it would have been... I don’t know what we could have done	Stephen, 2
🟧	You couldn’t do what would normally just be a fairly simple thing	Angus, 4
🟧	It’s made life more complicated, you know, that’s the thing, where it’s just, as I say, things are less straightforward as a result	Angus, 8
🟧	it’s hard to manage family activities, it’s also hard to manage her activities	Vicky, 4
🟧	there’s less time to be able to find to relax	Richard, 5
🟧	the logistical balance and that time-keeping and things it becomes extra stressful as a family and quite often very difficult for us	Vicky, 5
🟧	how it’s going to be managed is quite a challenge	Vicky, 5
🟧	I say to her if you don’t do things then Karl will stop coming round, it’s the only weapon I’ve got, is the boyfriend really	Debbie, 7
🟧	It’s a battleground, almost constantly, we’re not having enough nice time as in the family	Sue, 10
🟧	You can see that he’s really struggling in his own mind about what he can and can’t do	Sue, 3
🟧	I tell you, it's a bit of a battle sometimes in the morning to get her up	Simon, 2
🟧	It’s all quite a struggle	Sue, 10
🟧	I think that what’s stopping him at the moment is himself, rather than, rather than possibly the ME	Sue, 20
🟧	I tend to call it the grind, it’s just constantly there isn’t it?	Andy, 4
🟧	we had to do a lot of phoning around to get the right people to come and see her	Stephen, 5
🟧	it’s constantly in the back of your mind, am I supposed to be taking her somewhere, should I have told her to do her homework	Stephen, 6
🟧	it’s always on the back of your mind	Stephen, 1
🟧	you do have to push for every bit of information you need	Stephen, 3
🟧	I was outraged, but er, but then I didn’t go away from that appointment leaving i like that, I said ‘well if nothing else I need a formal diagnosis’	Sally, 5
🟧	It’s quite a big subject isn’t it and we do spend a lot of time talking about it’s you know, very important to both of us to get it right	Claire, 7
🟧	getting her to do things takes time	Barbara, 10
🟧	it’s made more work for both of us [parents]	Stephen, 2
🟧	it just feels relentless that actually you can’t, can’t switch off all the time, you want to do the right thing, and as [my husband] said there’s a lot of conflict between us quite often	Sue, 2

Appendix vi: Theme list with corresponding quotes

there’s always a set of tasks relating to something	Paul, 3
It’s one extra thing you have to think about, so, everything is just a tad more complicated	Brian, 7
It ends up creating a lot of additional work and a lot of pressure	Vicky, 10
theres lots of organisation every day with the schools to do and communications with various different parties in order to try and make her life as ordered as possible, and that takes up quite a lot of time really	Anna, 3
we’re really trying to push her through sixth form	Sally, 7
everything has to be considered, if you do anything you’ve got to consider how long’s it going to take	Anna, 5
[We’re] just trying to be as proactive as we can with her and doing everything so she doesn’t have to do an awful lot. The fact that, behind that you go in and tidy her bedroom up so it doesn’t look untidy, and running around getting her special food... Just the little things to make life normal	Debbie, 1
you’ve got many different things to bear in mind as you’re doing things and sometimes you forget things	Anna, 8
It does feel like we’re constantly managing things and just keeping, you know, juggling balls in the air, keeping pots boiling	Vicky, 7
there’s lots of scheduling every day	Paul, 9
I think the big thing is that i cant actually force hayley into it, she has to grasp it for herself	Debbie, 7
to my mind, the more you can keep everything normal, you know, it’s like that duck, the feet are going like mad underneath it, trying to make it normal... So Hayley’s life continues to be normal.	Debbie, 9
It’s kind of like on a hamster wheel, you know just going round and round and round and if you get a chance to get off every now and again sometimes I’m thinking Gosh I could go out today, but (sigh) I don’t know if I really want to really. So it’s, you kind of get that aspect of it as well which is, which is crazy really, but when I do go out, when I make the effort I do go out I do realise how much i’ve needed it.	Janet, 5
[my other son] had got better and so when she became ill it was like God now we’re going to have to deal with this!	Vicky, 19
We’re going to have to build her up again	Vicky, 19
It’s important that we keep life the same for her	Barbara, 7
Everyone’s, I think, pulling for her to recover and get back to somewhere near normal again	Debbie, 10
You’re constantly trying to, not gloss over it, but make it seem normal that you’re not all doing it together really, I feel.	Laura, 11
Exhaustion	
We were just tired, tired, really struggling	Sue, 13
I think i’d also like to say it is absolutely exhausting for us as parents	Sue, 1
We can just never relax	Sue, 2
It makes everyone in the family very tired I think. You begin to think everyone’s got ME!	Sally, 3
I am exhausted. I think we both are exhausted from the whole thing aren’t we?	Sue, 2
You need quite a lot of stamina to keep going and it did remind me of when they were younger, you go from activity to activity and changing your activity levels and it’s tiring	Anna, 7
it can be quite tiring	Anna, 7
I’m just worn out by it I think	Sue, 3
I think the impact on us is, frankly, exhaustion	Vicky, 12
probably over the period of time it’s quite tiring on us, particularly for you	Angus, 11
I definitely just collapse in a heap	Laura, 12

Appendix vi: Theme list with corresponding quotes

Is the worst behind us?	
I'm just hoping that things eventually you know work their way out of this, we'll get her up and running	George, 6
the best way I can put it is 'so far so good', we haven't had anything to bother us yet, which i think can only be a positive thing I suppose. (pause) and let the thing go its course	George, 7
I think we've been trough the most horrible bit and we're back to where, back to where we were.	Sally, 5
There's a, a tiny part of us both at the moment I think that actually wonders whether he has turned a corner and wether actually he is a lot better	Sue, 6
You were the light at the end of the tunnel	Laura, 17
there are people with far worse situations to deal with, so we're lucky because, you know, fundamentally he's ok	Andy, 8
you've just got to keep going, really and we say to her just keep chugging, just got to keep on doing it and that's all we know to do and make the best of what you can do, so just keep going, don't panic, but just keep focusing on what you can do	Anna, 24
you're in this long, long tunnel of, and a sense that people DO come out of it and DO get better, i think it's really, really important, that hope, keeping hope going	Paul, 16
it's not all bad	Paul, 2
it's important to have positive things, small things, you can live on those, like that's holidays for you isn't it?	Paul, 23
I guess there's always hope that it's going to get better, so it's not (pause) it's not terminal	Brian, 24
obviously it's not like a child who's, you know, terribly ill with cancer or something but it does have an impact	Laura, 2
You could be disabled, much more disabled, then it might have affected me.	Simon, 2
it's nowhere near as bad as it could be	Brian, 24
sometimes it's too easy to get caught up in the frustration of what's not happening to recognise what has happened and I think the journey she's been through has been quite significant and I think she's learnt and gained some other skills as part of it as well, which i think are positives	Richard, 21
Uncertainty	
What are we dealing with?	
where has it come from? Why has Karen got it?	Barbara, 3
I said 'well if nothing else I need a formal diagnosis' and then I got the referral to the specialist.	Sally, 8
I still don't understand the disease either, and that's hard	Sue, 10
What's frustrating for ME organisations and for us is 'what is it?', are we, is it a physical disease? or is it a disease of the brain?	Brian, 22
it all seems to be a bit up in the air really	Sue, 22
It's really difficult and you never know really if it's that or if it's something else or what if it is that brings that sort of flare around and it makes it hard and frustrating really	Anna, 7
learning that chronic fatigue and ME were the same thing and that post-viral fatigue didn't really exist in any real terms, it was just really frustrating	Anna, 14
She had the injection, as all girls did, and then you read that there may have been a link to that, so she had that last year, the year before and you kind of think, ah dear, you know. You worry about all these things .	Debbie, 12
to be honest I don't think now I understand the illness	Andy, 8

Appendix vi: Theme list with corresponding quotes	
I think it’s the sort of illness I feel, personally, you just think you’re getting to grips with it, you just think you’re understanding it, and then something else happens and you think - no, I haven’t got a clue.	Claire, 8
it isn’t something you can explain in one sentence to somebody. Because there isn’t, whilst there is a much better understanding now than there used to be it’s still massively, not surprisingly misunderstood because we don’t even get it do we?	Claire, 8
ME’s strange, you know?	Vicky, 4
suddenly it was like why hasn’t she got the energy to just do what would be considered a normal walk, you know, what’s wrong?	Richard, 4
ME is quite a weird illness, I don’t even know if it is an illness? it’s a syndrome, whatever but the treatment for it is quite odd	Vicky, 5
I don't understand it, how can she get so tired without doing a lot to get tired? I cannot understand that	Simon, 3
it’s that whole, as I say... worse case scenario, sort of thing, could it be something?	Angus, 2
Who are they and what will they become?	
it was horrible being with all these normal kids like, like my child used to be seeing them getting on with life, enjoying themselves ,and thinking ‘my child can’t do this anymore’	Sally, 2
I think just the painful reminder that my daughter wasn’t able to do that anymore, yeah, and that, if you like, the sense of grieving, yeah, that sort of loss, loss of the child you once had	Sally, 2
it made it hurt when you saw that hits at a time when they’re taking public exams so she’ll be very much changing her expectations of what she’s going to do it terms of university	Sally, 6
it’s robbed her of what should have been	Sally, 6
bits of childhood that should be fun and experimenting and exciting and having loads of energy and being able to do lots of things and all of a sudden that’s gone	Sally, 6
because they’re coming out of school earlier you’re losing opportunities you might have had to work longer	Sally, 6
it’s horrible to see, your child who was a very sociable person turn into somebody who, it doesn’t feel like it’s quite him at the moment	Sue, 5
it reminded me of having small children when they were young and you’d have to find things for them to do	Anna, 2
Her confidence and self-esteem have had an enormous knock - she’s almost like a different person in that respect, since before and afterwards of having it.	Anna, 8
it makes it hard to let go of her and do things on her own, which can’t really be right	Anna, 7
I mean she’d been to see her GP a few times but, it’s one of those things, there’s nothing wrong and then she’ll have a spurt when she’s better, you know, down again, so a bit like a, a grieving process.	Stephen, 2
You can restrict your aspirations	Stephen, 3
She’s lost her future	Stephen, 4
It’s restricting her future	Stephen, 4
She’s a totally different person	Barbara, 8
She’s changed, she takes everything seriously now	Barbara, 10
we still have glimpses of the old karen	Barbara, 12
she looks like the old karen and everything	Barbara, 13
we want karen back	Barbara, 15

Appendix vi: Theme list with corresponding quotes

I know they're still underneath, because you get flashes of the old person, you know, you get bits of the old person	Sally, 6
She's like a small child again	Janet, 7
At his age it should be normal for him to be going down the youth club on a friday night	Brian, 3
It doesn't feel like it's quite him at the moment	Sue, 2
I think that he's changed in the last year or so	Sue, 7
he was lovely to begin with but now he's just fed up with it all	Sue, 9
He really is underneath, he's a really nice guy, and so everything that we see is just so out of character	Sue, 12
If he has a day off school it's ME, it's never anything else, so maybe we're slightly blind to it	Brian, 16
It's hard to see her not flourishing and not becoming independent, it's hard to see a young person being quite so restrained	Anna, 8
you need quite a lot of stamina to keep doing that and it did remind me of when they were younger	Anna, 7
we're getting there and, as I said the other day, there are some glimpses now of the old hayley	Debbie, 13
Hayley's not a, I say 'normal ' teenager, Hayley I don't think is a run of the mill teenager	Debbie, 11
She isn't able to do what you would normally expect a little child at her age to be doing	Vicky, 3
we grieve over her wellbeing in that she was reasonably athletic, she swam and she did gym and she just can't do that anymore.	Stephen, 4
We've lost an active, academic child and we've got an inactive child that's struggling with the amount of work she's going to need to do	Stephen, 4
What once was a girl that was active and would have a good, what you'd call a good set of all round... a good all round education, sufficient to go to university, you know, without question, now we're talking about a girl that's so unfit she's restricted	Stephen, 4
Before he's be able to go into school at half past seven, and now he can't do a normal school run	Angus, 2
It's a loss of her life that she should be having	Anna, 3
he has missed a chunk of teenage years	Laura, 10
it's restricting her future, it's made her future very narrow	Stephen, 7
Hopefully she'll either grow out of it or control it so she doesn't get any relapses, but I don't know, I don't know	Simon, 6
Yeah, so a bit of anger if you like that this thing's coming along when it has, has robbed her of what should have been a, apart from the exam stress, a particularly exciting fun time of life	Sally, 9
There must be something we can do	
that leads to lots of, lots of stress and strain as to, you know, what should we do? Should we be firm? should we try and make her get up, try and make her get into school?	Sally, 1
Initially I felt quite relieved and then probably after a while I felt you know, like 'oh my god, this is the end of the world you know, especially as she started getting worse	Janet, 2
I feel helpless because I don't feel like I have any level of control at all, you know, as a mum I'd just like to take the ME away	Sue, 10
at this present point in time we're struggling about what to do next	Brian, 11

	Appendix vi: Theme list with corresponding quotes	
🟨	at the end of it it’s frustrating, trying to keep us on the straight and narrow is a good thing and valuable but it (pause) what we really want is for it to be fixed	Brian, 17
🟨	it’s about the fact that it’s going on so long and actually, I suppose it’d be nice ‘ok well there’s been new evidence that actually, we’ll give you this tablet and he’ll be better in the morning’	Brian, 18
🟨	the magic wand says we want him better	Brian, 23
🟨	not being able to give a tablet for it just to make it go away again is actually kind of quite scary	Debbie, 6
🟨	the worst thing is, the most frustrating thing is that there’s no easy fix and with an illness you really want an easy fix that gets it over and done with	Debbie, 12
🟨	it’s been two years, I should have been able to sort this out by now. I got a bit fed up with it all if I’m honest	Andy, 8
🟨	Richard: It comes back to that thing about how one feels when you’re just looking at somebody you love suffering essentially and not being able to flourish and... Vicky: You can’t help her	Richard and Vicky, 10
🟨	Just getting the impression from the GP that there’s nothing wrong with her, yeah, she’s just naughty, ‘you’re wasting my time’ sort of thing.	Sally, 4
🟨	We’re also face with people saying isn’t it marvelous that David’s better now! when they see him on a good day and you just feel like a (sigh) an overprotective parent when you say oh, but he’s not better you know, people think you’re crazy!	Sue, 23
🟨	I just feel guilty that as a parent I should be managing the whole thing a lot better than I am, so it’s just constant feelings of guilt all round that I’m not doing the best for David	Sue, 20
🟨	I think it’s one of those things in retrospect you perhaps would have done slightly differently	Claire, 18
🟨	sometimes I realise I haven’t sat down and read with them, like I should have done	Laura, 7
🟨	I’m probably a bit more on edge because than there are times when [my husband] is not here then, obviously, i’m dealing with all of the children	Laura, 7
🟨	She was doing ok for a while and then she started to struggle again, it was really difficult to work out, is it something we’ve done wrong?	Stephen, 4
🟨	You do feel very, almost fraudulent, taking a tired child to the doctor	Stephen, 6
🟨	it’s just like ‘what are we supposed to do here?’	Sally, 1
🟨	I really don’t know if it’s the right thing or not but he entered for a race at the weekend	Brian, 12
🟨	We tried various things, we tried changing his diet	Brian, 17
🟨	The fact that he’s sociable with us most of the time, you know, makes us feel good, and that gives us, I think some confidence that we’re getting it right most of the time.	Brian, 24
🟨	[the therapist] gave us clear implementation methods, so there was no further decoding or, there was nothing further required.	Paul, 15
🟨	sometimes you lose your way a little bit or you forget about things, it’s really good to come back and [the therapist] refocus again and off you go again and it’s really helpful to have that	Anna, 16
🟨	it’s hard to know when to turn back and say stop	Stephen, 3
🟨	you don’t really know the answer and you’re afraid to go to far along the line	Debbie, 1
🟨	it is a lot about trial and error isn’t it?	Debbie, 10
🟨	I think in some ways it was a learning curve with her	Richard, 13
🟨	managing it is a different language	Vicky, 17
🟩	Isolation and Restriction	
🟩	**No one can see this**	

Appendix vi: Theme list with corresponding quotes

people don’t see this	Barbara, 12
I would put it it’s like an ‘illness within’, you don’t see it but it’s in the person and that makes some days very hard to understand you	George, 13
my mother had heart disease...but that’s what you might call a, an ‘open illness’, people can see you’ve got it	Sally, 3
it is isolating. A lot of people don’t really understand about... they see Rachel and they see that she’s fine and don’t understand that she can’t do things	Anna, 4
It’s a hidden disorder, isn’t it? it can’t be seen, people don’t know they just think you look fine and that’s it and they don’t see beyond that.	Anna, 10
She looks fine, there doesn’t look anything wrong with her particularly	Debbie, 2
She walks out the front door and she’s looking quite smart and she’s made an effort but half an hour into it she’s tired and she wants to come home again	Debbie, 8
it’s one of those invisible injuries where people know there’s something wrong but they can’t see there’s something wrong so they don’t quite know how to react	Richard, 18
Only we know	
the extended family, you get the feeling they just don’t believe it	Sally, 1
it took so long to get a diagnosis because I felt that I was being treated as a munchausen’s by proxy parent, yeah, so i didn’t dare suggest anything.	Sally, 4
[her sister] thinks karen’s putting a lot of it on [wry laugh] and she thinks she puts it on for us sort of thing, and, but she’s always been a little bit jealous of Karen.	Barbara, 1
trying to explain to other people’s very hard isn’t it?	Barbara, 12
[the GP] thought she was lying, she was putting it on	Janet, 5
her sisters still don’t believe she’s ill and think she’s putting it on	Carol, 1
it took [my ex husband] quite a long time to work out what the illness meant	Carol, 1
Our other children, the oldest of whom spent the first couple of years, telling us that there’s nothing wring with David, that we were imagining it	Sue, 2
close family members also don’t believe, and that’s hard	Sue, 23
My father doesn’t, I don’t think believes it, he doesn’t want to hear anything about it	Sue, 23
it’s difficult isn’t it? because a lot of other people don’t believe that it is a real disease and so we’re faced with that, that we’re always sort of explaining away when we shouldn’t feel should need to	Sue, 23
You go to your GP and you say my child’s not right and they go well, what’s wrong? and you go well, she’s not right, she hasn’t been to school today or yesterday or the day before, why not?, Well, she’s too tired. And it’s hard to keep taking a child back under those circumstances because the doctor goes Well, there’s nothing wrong, you’re just tired	Stephen, 7
I suspect they don’t all believe in it, all of them, they don’t think they’re really dealing with something tangible	Laura, 15
it can’t be very nice to have somebody who doesn’t really believe that you’re as ill as you are	Carol, 7
I don’t think anybody understands unless they’ve actually lived with someone who’s had it	Carol, 8
That’s the big issue actually, communicating it outside the family to people that Rachel interacts with. That IS the big issue	Paul, 18

Appendix vi: Theme list with corresponding quotes

🟩	I find medical terminology something really hard to hold onto because it’s a foreign language	Richard, 12
🟩	I think generally it’s unhelpful when people don’t have an understanding and they think you, you know, that’s difficult, even with the friends and that comes back to the various simple information and thing where it just says, like, you know, they’re not conning you, they’re not, you know.	Paul, 18
🟩	we kind of dropped through the cracks in the floorboards to be honest	Angus, 11
🟩	people see them on the good days and say why don’t they go to work? yeah, and they don’t see them on the next day in the wheelchair. it’s felt like that sometimes	Sally, 4
🟩	I found it hard to understand because to me any weakness is not a good thing, if i’m fit i’m feeling good.	George, 4
🟩	I don’t think anybody understands unless they’ve actually lived with somebody that’s had it, I don’t think they do.	Carol, 3
🟩	it’s very difficult for the other kids to understand	Sue, 2
🟩	Dad: I’m not sure I understand it. Mum: No, I don’t understand it either	Sue & Brian, 19
🟩	I don’t think [the school] understand it, I don’t think they’ve got time for it	Sue, 19
🟩	sometimes I think, school and I think I find it difficult to understand why, if you’ve got these important lessons, why are you going to be doing climbing or something like that because obviously doesn’t seem to make any sense at all	Anna, 9
🟩	when this person makes a choice that’s a really good thing they make a choice, and they choose something they enjoy, that’s a really positive thing towards their recovery, they are not shirking	Paul, 10
🟩	but it’s a challenge, isn’t it, to be constantly educating, especially as they move on	Paul, 10
🟩	How many times have I heard you look fine. Yeah, because I wouldn’t be here would I? If I didn’t feel alright?	Paul, 10
🟩	[the GP] wasn’t giving us any advice, we were in limbo and it feels like no-one knows anything and he wasn’t doing anything and it got so frustrating	Anna, 13
🟩	when I got it, had it myself, I knew exactly what that is	Anna, 20
🟩	a lot of people have an opinion on it and think they know what it’s all about and I do find that quite a challenge at times to deal with that situation	Andy, 13
🟩	They just don’t know, it’s one of those things, until you actually deal with it day to day	Andy, 13
🟩	not surprisingly misunderstood because we don’t even get it do we?	Claire, 15
🟩	after a period of time it falls into a bit of a chore and becomes a bit more of a struggle to quite understand what’s what	Andy, 19
🟩	it’s difficult to explain to other people as well, why she can’t do things so it is a little bit, I find that a little bit frustrating	Debbie, 13
🟩	someone came up to me and said I’m sorry to hear he’s got diabetes and I said What?! you know, and they hear something and they take it further, so then you need to sit down and explain to them	Laura, 2
🟩	**Our world has shrunk**	
🟩	we don’t go out so much because of course Rachel’s got chronic fatigue	Anna, 14
🟩	I don’t go out very much	Carol, 5

	Appendix vi: Theme list with corresponding quotes	
🟩	When Karen was first diagnosed everything was Karen, you know, this child just led on that settee sound asleep 24/7, you know, we couldn't go out, we couldn't do anything, we missed parties and birthdays and things.	Barbara, 3
🟩	you get out of the loop of socialising with people and so you don't get asked places	Anna, 2
🟩	Her social world diminishes	Anna, 1
🟩	you can't go further afield, you can't spend a day out or go to the, you know, seaside for a day.	Anna, 2
🟩	I think she feels like home can be a bit like a prison sometimes	Anna, 2
🟩	her life over the last few years has been constrained	Paul, 7
🟩	it makes you feel like you're in a prison	Anna, 8
🟩	sticking to the same thing is sometimes difficult and sometimes having those constraints can make you feel a whole lot worse	Anna, 8
🟩	it's that your life just shrinks and your social life shrinks as well inadvertently	Laura, 18
🟩	I suppose our world shrunk when he was really bad	Laura, 1
🟩	I think one's life shrinks, is the thing. You don't have time to do, you don't have time to do anything	Angus, 1
🟩	I end up going to bed earlier	Laura, 6
🟩	everything shrinks again	Laura, 19
🟩	they keep their distance and they fall by the wayside don't they, maybe they feel awkward, I don't know but you do feel that one or two people, your social life does shrink a bit, no doubt about it.	Laura, 19
🟩	life is stuck, everything's just pretty stuck	Paul, 10
🟩	We do MUCH less	Vicky, 12
🟩	you just want to get on with things and see it as a constraint	Vicky, 13
🟩	we're on our own	Andy, 5
🟩	I feel like I'm a single parent at the moment	Sally, 3
🟩	I don't go out very much	Carol, 1
🟩	I think it's made it harder because there's only one of me, and so if there yeah, if there were two of us obviously it would be a lot easier because we wouldn't always be the same parent staying with Gemma, umm one parent could go out with the others and, I have to try and spread myself around	Carol, 7
🟩	when he went to secondary school he was on his own, he didn't know anybody, he didn't know anybody, he only vaguely knows people and i think he's friendly with people but he doesn't feel confident enough to invite them back	Sue, 3
🟩	you have to deal with the upset of having a teenager not being a teenager	Sue, 8
🟩	We're really on our own, we really don't know what to do	Janet, 3
🟩	I think relationships with the extended family, of which we have quite a lot, are inevitably just curtailed	Paul, 2
🟩	It's isolating for everybody	Anna, 5
🟩	communicating it outside the family to the people that she interacts with, that is a big issue.	Paul, 10
🟩	you can feel so alone with it	Paul, 22
🟩	he doesn't heave the level of contact with his friends as he used to have	Andy, 16
🟩	it is isolating	Anna, 4
🟩	one of the regrets we have, I suppose, that we weren't able to find this information out or, certainly, find it out in a way that we could implement	Stephen, 6
🟩	no-one can give you any information	Debbie, 1

Appendix vi: Theme list with corresponding quotes

🟩	you feel quite isolated	Debbie, 4
🟩	we kind of dropped through the cracks in the floorboards	Angus, 3
🟩	we all just were beginning to feel a bit hopeless that nobody seemed to know anything	Laura, 3
🟩	we felt quite on our own really	Laura, 12
🟩	there’s not so much fun, full stop	Paul, 12
🟩	not being able to flourish and not just being able to enjoy the now in quite the same way as you would otherwise and live a life driving round at 20 miles an hour.	Paul, 13
🟩	he’s fed up, he is really i’d say quite depressed and fed up	Sue, 7
🟩	maybe we get to a place where we don’t even discuss doing things that we might otherwise do	Brian, 8
🟩	It’s taken, taking away some fun that ought to be there	Brian, 9
🟩	it’s taken a bit of fun out of life	Brian, 9
🟩	he’s getting a bit boring and he’s very bored	Sue, 21
🟩	what David’s got really takes the fun out of life	Brian, 23
🟩	it consistently takes the fun out of life, takes the fun out of his life and I think it takes the fun out of ours as well, not all, not all the fun, but it just saps it	Brian, 23
🟩	there’s not much fun, full stop	Paul, 3
🟩	if she didn’t have chronic fatigue I think it would just be more fun	Paul, 4
🟩	we’re all living fairly boring existences	Paul, 5
🟩	life is missing the fun	Paul, 5
🟩	it is just dull and boring	Anna, 5
🟩	day to day life has been a bit stretched	Debbie, 6
🟩	It’s actually been really hard to get her outdoors at all. So if the rest of the family are then pent up indoors it’s boring.	Vicky, 7
🟩	It's just the leisure side that's affected.	Simon, 2
🟩	the family activity side’s also stopped	Vicky, 2
🟩	it’s much harder to think of activities which cater for everyone’s needs	Vicky, 2
🟩	suffering from boredom is part of the problem	Vicky, 13
🟩	Restriction	
🟩	[my husband] had plans to go to the gym regularly which now he can’t do as often because somebody has to be here with Karen all the time	Barbara, 1
🟩	I’ll have to wait until she gets better before I think of anything else	George, 5
🟩	I don’t feel right doing things with my other daughter that the three of us would have done together	Barbara, 13
🟩	she used to go and stay with her father at weekends but obviously she can’t do that anymore	Janet, 1
🟩	If she overdoes it on the saturday then on the sunday if we were planning on doing anything it just wouldn’t happen	Janet, 2
🟩	we changed our holiday plans in the summer because we didn’t feel that we would be up to it	Brian, 8
🟩	the children aren’t allowed sleep overs	Sue, 8
🟩	we don’t tend to do nearly as much as we used to on weekends or on holidays, even down to booking a holiday	Paul, 3
🟩	it’s changed the things we do	Andy, 5
🟩	we change the way we do activities, we have to think about it	Claire, 16
🟩	we used to go out for walks and things altogether as a family. Suddenly all those sort of things we didn’t do at the weekends anymore	Laura, 1
🟩	we haven’t done any major holidays or anything and things like that	Laura, 4
🟩	In the beginning I felt I couldn’t do things that I wanted to do	George, 13

Appendix vi: Theme list with corresponding quotes

🟩	When we're out for a day we're not out for as long as we normally would have been out	Simon, 3
🟩	We’re pretty much housebound	Janet, 1
🟩	it makes you feel like you’re in a prison	Anna 5
🟩	I can’t see a way out of it	Sue, 21
🟩	her social world diminishes and she’s not so able to be independent and go out so she can’t be, she’s a dependent teenager really so I think it makes a difference on our relationship	Anna, 12
🟩	All those sort of bits of childhood that should be fun and experimenting and exciting and having loads of energy and being able to do lots of things and all of a sudden that’s gone	Sally, 6
🟩	It’s a huge stress just managing because there’s no freedom if you’re trying to stick properly to things like that, there’s no freedom	Vicky, 5
🟩	In the beginning I felt I couldn’t do the things I wanted to do	George, 3
🟩	He can’t play with his elder brother, so he probably feels a bit left out, probably accounts for his slightly erratic behaviour sometimes, which is probably just attention seeking.	Angus, 13
🟩	She has felt restricted by, and frustrated by the fact that we’re saying we can’t do certain things and, whilst we’re trying to be careful about why and not to blame it on A or, if it is very much due to him, explain the situation, you can get a little bit of resentment	Andy, 3
🟩	There’s jobs that I haven’t gone for because of it, because I haven’t been able to put the effort into it.	Sally, 6
🟩	the other children aren’t allowed sleepovers and that’s stemmed from David, that actually it’s kind of unfair if the others do things and he’s not allowed to, it’s just had an affect on everybody	Sue, 8
🟩	If we had a half term or whatever we’d have something planned for every day to go and do, which we don’t now.	Claire, 5
🟩	I think maybe we get to a place where we don’t even discuss doing things that we might otherwise do	Brian, 17
🟩	we don’t go out so much for all those reasons in the evening	Anna, 4
🟩	sometimes the best way to face this is to take it day by day	George, 2
🟩	You just take it as it goes along, there’s no other way to, as i’m concerned, you can look at it	George, 6
🟩	you’ve got to be positive, there’s no other way	George, 15
🟩	knowing what the problem is then dealing with it facing it	George, 15
🟩	you get up and you get on with it every morning don’t you, and you do what needs to be done because she needs, she needs looking after	Janet, 2
🟩	You get in this habit that you’re just, what’s today’s set of tasks? you know, there’s always a set of tasks relating to something	Paul, 3
🟩	I don’t tend to blame people for things or get hooked up on that sort of thing. it’s just, well, it’s happened now	Andy, 9
🟩	last week you couldn’t do this and you couldn’t do this and you couldn’t do that and you were unable to do this, I don’t like talking to him in those terms all the time	Andy, 12
🟩	Whole family effected	
🟩	for the first month we were quite positive and then when we didn’t get the response that we thought we were going to get we kind of all dipped	Debbie, 1
🟩	you find you have less time for the other children and less time for ourselves	Angus, 8
🟩	his brother probably feels a bit left out, probably accounts for his slightly erratic behaviour sometimes, which is probably just attention seeking	Angus, 8
🟩	I think the family activity side’s also stopped	Richard, 2
🟩	Slow down	
🟩	lots of activities just were curtailed, stopped.	Angus, 1

	Appendix vi: Theme list with corresponding quotes	
🟩	I end up spending half the week it seems, in bristol sort of killing hours	Laura, 2
🟩	day to day life has been a bit stretched	Debbie, 1
🟩	life is stuck, everything's just pretty stuck	Paul, 7
🟩	not being able to flourish and not being able to enjoy the now in quite the same way as you would otherwise and live a life driving at 20 miles an hour	Paul, 8
🟩	**A need to escape and a feeling of guilt**	
🟩	there needs to be someone in the house at all times now	Barbara, 1
🟩	they're just there and in the space where otherwise you'd be just chilled out and relaxed or getting on and you know, cleaning or doing some work yourself. So that does make life different, you feel that if somebody's always there you've got no space	Sally, 6
🟩	he should be having more time away from us	Brian, 3
🟩	we have less free time together because normally she would be out and she's not so everyone's, we don't get so much time to do stuff do we?	Anna, 11
🟩	he's always there isn't he?	Claire, 6
🟩	He is just always there	Andy, 6
🟩	our time together has been squeezed and I miss that actually, I miss that we don't have that sort of together time so much any more	Andy, 6
🟩	having your son at home for so long has an impact on your personal relationship with each other	Laura, 1
🟩	It's almost like being in a permanent threesome with him isn't it, because he's always there, you know, just always around	Angus, 6
🟩	sometimes when she's been really bad I wished there was somewhere she could go and they could try, try and, there were experienced people that could try and work with her and get her going	Janet, 9
🟩	he uses his illness sometimes as an excuse	Sue, 2
🟩	when David is tired he is far more argumentative and it just raises the temperature for the whole family	Brian, 1
🟩	It's frustrating, she doesn't do her chores	Vicky, 11
🟩	I didn't like it because it takes away from my independence a little bit	Sue, 12
🟩	She becomes dependent and we become less independent	Anna , 7
🟩	I'm here with her, you know, supporting her in what she needs but it's not really doing what I want to be doing	Anna, 7
🟩	So [my husband] gets, well he's like the dog that hasn't been walked because you're going 'but this is MY WEEKEND! I want to enjoy it with my family, but my family can't do it.'	Vicky, 10
🟩	the gym in many ways helped me, you know I could just wake up and go there, come back, and deal with it in my own way	George, 1
🟩	my husband reacted to it by going and doing more and escaping	Sally, 1
🟩	[my husband and I] are both involved in [activities] and lets you go off and escape	Sally, 1
🟩	i think it's something that [my husband] doesn't want to know too much about	Sally, 7
🟩	to have a little bit of normality sometimes just when her dad comes over to be able to go out and not talk about ME. Go out, it doesn't happen very often, go out with a friend, have a cup of coffee and talk about their problems and you know, the state of the world instead of ME.	Janet, 3
🟩	just to go out, it doesn't happen very often, go out with a friend, have a cup of coffee and talk about their problems and you know, the state of the world instead of ME	Janet, 5
🟩	from my point of view there's not a lot I can do in that time and certainly it wouldn't be something I would choose to have gone and done within that time	Claire, 4
🟩	I went away for a touch of normality	Laura, 9

	Appendix vi: Theme list with corresponding quotes	
🟩	sometimes you just, you want the real normal, or what you think is the real normal. You want to step outside that world where you are concentrating on the sick child. You just want to just be yourself	Laura, 18
🟩	My day to day life changes, I'd just like the house to myself sometimes, I would like some real 'me' time without having to go out of the house in order to get it. Sometimes, I think that's probably one of the things I find really, which sounds selfish sometimes	Laura, 19
🟩	I feel I have to go out in order not to be around and I resent that sometimes	Laura, 19
🟩	I've got to still do things that I like to do, but sometimes I do feel guilty because Karen would have been coming with me	Barbara, 13
🟩	you want to do the right thing	Sue, 2
🟩	I've got to still do the things I like to do, like [my husband] still needs to go to the gym because he needs to. But then I do feel guilty because Karen would have been coming with me	
🟩	it's quite difficult that makes in my own personal sense feel quite guilty and sometimes [my wife] will pass a comment that I'm not taking my parenting duties seriously	Brian, 9
🟩	we have permanently got the stress of worrying that we're doing the right thing	Sue, 9
🟩	I've got the mother's guilt of stretching myself too thinly with the family	Laura, 18
🟩	I just feel that as a parent I should be managing the whole thing a lot better than I am, so it's just constant feelings of guilt all round that I'm not doing the best for David and I'm not doing the best, the best I can for the other children	Sue, 11
🟩	I don't know what's right, what to do for the best	Sue, 15
🟩	I sometimes feel guilty about trying to persuade him to do things, I feel guilty sometimes that I'm not helping because I'm not being positive	Brian, 16
🟩	he sort of said that was a miserable birthday, really, which I felt guilty about	Laura, 4
🟩	I have the mother's guilt of stretching myself too thinly with the others	Laura, 18
🟦	**Focus on unwell person at the expense of family life.**	
🟦	**Life revolves around them**	
🟦	it does become the focal point of arranging everything in your day, because everything's got to be based around it	Andy, 18
🟦	Occasionally, [his brother] has got cross and said, he sort of thinks everything is focussed a lot on A	Andy, 17
🟦	somebody has to be with Karen all the time	Barbara, 3
🟦	we've had to manage our life and tailor our life to how we can manage it	Anna, 11
🟦	now everything has to be actually managed around when we're taking him into school or around when someone can be here to look after him or around the length of time he's able to go out and do that thing with you.	Andy, 18
🟦	she might feel the whole family has been reorganised around them	Vicky, 16
🟦	life revolves around A and his limitations	Claire, 18
🟦	It's tempering everything to fit within his limitations is really the normality in order that he feels normal, I suppose. As normal as possible.	Laura, 15
🟦	We used to go out for walks and things altogether as a family. Suddenly all those sort of things we didn't do at weekends anymore	Laura, 13
🟦	With a smaller family I don't know but with a bigger one you end up, as I say, doing things as smaller groups as opposed to, that's the noticeable thing, or just not doing things.	Angus, 5
🟦	I think maybe we get to a place where we don't even discuss doing things that we might otherwise do	Brian, 13
🟦	We can't go out with family, other family members anymore, erm, because Zoe can't or is too unwell to go out	Janet, 2

Appendix vi: Theme list with corresponding quotes	
We've planned to do things and in the day we've gotten up and she hasn't been well enough to do it	Simon, 3
we're always having to change plans, if we plan to do something 9 times out of 10 it ends up being cancelled or rearranged, cancelled again and rearranged, so I tend not to plan things now, do things if she's well we'll do things.	Carol, 1
You find you have less time for the other children and less time for ourselves	Angus, 4
The other kids are missing out. I mean they're, they're very good kids, the other kids too, our other children, but they resent it.	Sue, 7
The children aren't allowed sleepovers	Sue, 7
the other children don't get probably their fair share of attention that they perhaps would have had	Angus, 13
Sometimes I realise I haven't sat down and read with them like I should have done	Laura, 14
you're stretched thinly, you are with the others, because you do end up spending so much time on the child who's unwell	Laura, 2
[his brother] is probably the one who's missed out the most	Angus, 8
Destructive force	
If I'm keeping her company here I can't go out and be independent i'm, you know, I'm here with her and that's her, you know, supporting her in what she needs but it's not really doing what I want to be doing	Anna, 5
it's sometimes a struggle and it's got you down and I think it's fair to say I don't naturally see that until we get to the point where you've got upset basically, or it becomes very obvious.	Andy, 4
we don't go out really together anymore, not that often	Andy, 5
One of us goes instead of all of us going because I didn't want to leave Tom or, so we've sort of split ourselves up, and so yes it does divide you up and affect you	Laura, 2
when you go out it will tend to be now with your friends because I'm here, rather than the two of us going out together, which perhaps it would have been before	Andy, 5
the way we rejig the family is to cut it up and divide it up and participate or not participate so it's been very destructive to family	Vicky, 12
the other boys have probably missed having both of us with them all the time	Laura, 4
The opportunity to relax as a family has gone and I think that tension has ended up having an impact	
just that he's there, just sort of inhibits, you know	Angus, 6
I think the biggest impact probably has been that we can't do things as a family like we used to so it means they go off and do things with their friends because we can't leave Gemma and go out with them, we can't go out together, or very rarely, so we tend to split up and do different things	Carol
it ends up creating a lot of additional work and a lot of pressure which comes with that	Richard, 5
I feel like sometimes i'm a single parent	Sally, 2
it becomes extra stressful as a family and quite often very difficult for us	Vicky, 5
We don't go out together really anymore	
there's this terrible split of, well, it feels really wrong to constantly leave her at home on her own	Vicky, 12
It sometimes makes a strain on myself and my husband	Debbie
we have less free time together because, again, normally she would be out and she's not so everyone's, we don't get so much time to do stuff do we?	Anna, 3
the time we can have together has been squeezed and I miss that actually, if i'm honest	Andy, 6
I think it's harder for us to have time for us together as a couple and we have to grab our moments to talk when we can	Andy, 6

Appendix vi: Theme list with corresponding quotes	
it causes us to have friction between us	Laura, 8
It did cause a lot of friction between the two of us	Vicky, 16
it’s definitely inhibited our personal relationship, i’d say, without a doubt.	Laura, 13
We don’t have that time together so there’s not as much intimate ‘us’ time basically, to put it in a nutshell!	Angus, 14
it’s not made a barrier, but it just inhibits, it’s inhibiting.	Angus, 14
it doesn’t allow you the personal intimacy and contact that you would have had before so it makes you a bit more distant, that’s the thing, you’re not as close	Angus, 6
there’s probably been more friction	Angus, 15
we end up tearing our hair out and quite often having a row don’t we?	Vicky, 11
it was quite a trial, disagreeing, and also working out who was going to be the carer	Sally, 1
we still disagree sometimes about how we’re working together	Sally, 2
Initially I think I’ve got a fairly stubborn resistant teenager	Sally, 7
we used to go to london and Catherine said well why can’t we go? and I said well it’s not fair on karen if we go	Barbara, 5
we’ve had a few arguments	Carol, 2
you know, we just had to say look we might have to, there is a limit, you can, you might have to leave home!	Brian, 1
we seem to have conflict in the family the whole time	Sue, 2
there’s a lot of conflict between [my husband and I] quite often	Sue, 2
there’s a lot of conflict between david and his sister	Brian, 2
we’re not being quite united all the time on it and it’s hard	Sue, 6
we fight about it, we argue about it and I hate that added, that added stress in the relationship	Brian, 6
it puts pressure on the relationship	Paul, 1
if she wasn’t unwell we’d have more time to do things on our own	Anna, 2
se gets frustrated and gets cross with us as a backlash	Anna, 3
it winds the other two up and they’ll argue	Andy, 2
I was taking him to the bus stop wasn’t I? and you were saying, that’s rubbish, you don’t want to be doing that	Claire, 10
relationships with me and my wife are ok, though there is an added strain	Stephen, 1
you’re tired and you’re getting information in and it’s not gone 100% to plan, that you can sort of go ‘why not?’ but in general, [my wife] accepts we’re doing our best	Stephen, 4
it makes a strain on myself and my husband	Debbie, 1
we’ve had a few arguments	Debbie, 7
we’ve had arguments and, you know, tensions and things	Laura, 1
sometimes [my husband’s] been tactless with him and things like that and I sort of then go and try and make the peace	Laura, 7
I get cross and wound up	Laura, 8
I was furious that you hadn’t managed his activity, we ended up having a massive row	Laura, 9
the opportunity to relax as a family has gone and I think that tension has ended up having an impact	Richard, 2
quite often we end up having a row	Vicky, 3
I think the impact on the relationship it’s had most is exhaustion leads to fractiousness and lack of care	Vicky, 8
we had a little battle this morning	Barbara, 16
we’ve had a few arguments	Carol, 5

Appendix vi: Theme list with corresponding quotes

Relationships with me and my wife are ok, there is added strain in that, because Kate's got quite a regimented regime that ensuring that's followed as best as possible is, can cause a bit of strain here and there.	Stephen, 8
it puts a strain on myself and my husband	Debbie, 10
how we control and discipline David in those situations can lead to conflict between Sue and I because it's difficult to know what's right	Brian, 13
I was taking him to the bus stop, wasn't I, and you were saying 'that's rubbish, you don't want to be doing that	Claire, 10
We still disagree sometimes about how we're working together and who's turn it is to be the one who gets up earlier than you would need to do to make sure she wakes up in time.	Sally, 5
[the relationship] It's not better, that's probably for sure!	Angus, 11
Resentment	
I don't think she should be jealous, and she is, that's the only word I can say, she's jealous of Karen	Barbara, 14
she's always been a little bit jealous of Karen	Barbara, 1
[his sister] says we are giving all our attention to David and what about her and she's been very very jealous hasn't she?	Sue, 2
the other kids are missing out, I mean they're very good kids, but sometimes they resent it	Sue, 8
she really resents a lot of what David's going through and the special treatment that he's had, which is hard	Sue, 14
There's a lot of conflict between the children and he's probably responsible for most of it	Sue, 14
[his brother] thought it had changed us a little bit, how we react and he thinks it's made him different, which he wouldn't say in a good way	Claire, 2
[his sister] has felt restricted and frustrated by the fact that we're saying we can't do certain things	Andy, 3
it's really impacted her younger sister, i think there's a hint of why are you telling me to do this when she gets to rest.	Stephen, 1
I think she's argued a bit more with her sister because of it	Simon, 3
they get resentful and then they see a problem	Laura, 5
The youngest daughter is the only family member that is really impacted and I think there is a hint of 'why are you telling me to do things when J gets a rest?'	Stephen, 9
There's a sibling rivalry thing of 'it's not fair, I do all her jobs' and he's right, it's not fair, that's the bottom line isn't it?!	Vicky, 6
Parental Roles	
Good cop/ Bad cop	
[my husband] is probably not so tolerant as I am of Hayley, he has struggled in the first instance, he's probably struggle more than I have to understand that Hayley isn't physically capable of doing things	Debbie, 2
because he hasn't come to some of the meetings and listened to some of the things I think he finds it difficult to accept	Debbie, 2
He tends to worry quite a lot whereas I will just get on with it	Debbie, 7
we just take on a different role or someone'll do something for us to allow us to do something else	Debbie, 9
I used to under-react to things that happened	Paul, 5

Appendix vi: Theme list with corresponding quotes	
Whereas [my wife] will prefer to tell him what to do, and i think that, that's, we have a different style of approach to it, obviously that's not, my style is not the same as [my wife's] on this one and so [my wife] will probably perceive that mine is a wishy washy approach and i will perceive that [my wife's] approach is leading to, whilst you might succeed in waking David up in that particular half hour you're losing the war.	Brian, 11
there's times we've maybe had slightly different view on where we are or what we should do	Andy, 5
we both work in very different ways	Richard, 7
[my husband] has never been proactive about trying to find out about what's best to do, he waits for either the health service or myself to say 'this is what we should, we could be doing'	Sally, 8
we've divided into the firmer and the softer one ever since it happened	Sally, 3
I do think we are a bit different towards her though aren't we? It's like if she says to [my husband] 'I don't want to do my walk today' [my husband] is a bit lenient and doesn't let her do it	Barbara, 5
we take it in turns to look after her	Barbara, 2
He was always at work and I was at home, and that's changed now	Barbara, 7
He was quite aggressive, pushing her to do activity, but he's, he's more passive now actually infact he's more passive than I am, he's almost taken a back seat in that he's not pushing her to do really very much at all	Janet, 3
I gave up my job completely after the diagnosis	Sue, 9
we'll back off because of her condition	Paul, 3
i used to under-react to things that happened	Paul, 12
he says I tend to over react, he under reacts and somewhere in between we meet and come up with a solution	Anna, 13
everyone in the family has their individual bit	Paul, 21
he's ended up acting a bit like a parent to [his siblings]	Andy, 2
we'll talk about the big things going on, and if there's meetings with consultants etc, I've been to all bar one, so we'll discuss those big things but perhaps I've missed the day to day aspects	Andy, 3
I think how we were to start with has now changed, we're different and I am far more conscious that I don't want him to overdo it	Claire, 8
you're coping with trying to understand and trying to interpret and it's not that we both interpret in the same way so what I might see as a positive solution might not be [my wife's]	Richard, 5
I tend to do more management during the day because my wife is out at work so that's obviously the way it is.	Stephen, 4
I'm the one, it has to be said, who gets up early and I'm the first one up so I get him up	Laura, 6
I think m[my wife] takes on mist of the caring role	Angus, 12
I suppose you just find out how people react differently to things	Laura, 17
it's almost always been me who's come to the clinic appointments, I don't know maybe my husband may have come to you occasionally. I don't know wether he's not so comfortable or whether it's just assumed that that's mum's role	Sally, 7
I do think we are different towards her though aren't we?	Barbara, 5
sometimes [my wife] and I will react differently to him	Brian, 1
we've both got slightly different opinions about how to go about managing it	Sue, 2
it's probably a difference between us that's always existed, I mean it's not an effect of the ME, it's just our personalities	Stephen, 3
[my husband] probably isn't as tolerant as I am, he has struggled in the first instance	Debbie, 2
I have little or no sympathy for anyone who's ill, especially men.	Debbie, 7

	Appendix vi: Theme list with corresponding quotes	
	I, from the outset, was pretty much ‘lets get on with it, lets push on’ and I have had to modify that feeling and understand, or try and understand the situation far more and I think as time’s gone by I’ve got far more understanding.	Andy, 10
	I think you have to work more or less as a unit, I think if you don’t it’s going to pull you apart	Laura, 19
	So I think how i’d say we’ve handled it is - I am, up until quite recently, I’ve been the pulling back on it and you’ve been the pushing forward on it and then, really strangely, we reversed didn’t we?	Claire, 4
	we both work in very different ways	Richard, 5
	In the beginning [my ex-husband] didn’t want to accept it, ‘no, it’s not that, and you’ve got to push yourself’ and ‘I can’t believe you can’t do this and you can’t do that, and Zoe instead of just sitting there you should push yourself and do this and do that’ and he was quite aggressive, but he’s more passive now, actually he’s more passive than I am	Janet, 3
	It’s a “mother” thing	
	I’ve always been the one that looks after everybody when they’ve been ill	Barbara, 7
	it’s almost always been me [who came to clinic with Jenny]. I don’t know I think maybe my husband may have come to you occasionally. I don’t know whether he’s not so comfortable or whether it’s just assumed that that’s Mum’s role	Sally, 8
	I suppose mums are more emotionally involved than fathers (laughs) anyway I don’t know, I think she does feel that she doesn’t get any emotional support from her dad	Carol, 3
	I suppose it’s more of a male thing that actually you can just switch off and say ‘well, that’s that’ you know ‘he’s decided, don’t get stressed’ I don’t think you get stressed as I do (Sue talking to Brian)	Sue and Brian, 12
	[my wife] has always played the mainstay role of caring for everybody’s needs	Paul, 8
	I think it’s put quite a strain on [my wife] in that she has done the bulk of the caring	Andy, 1
	your maternal instinct kicks in and you just want to look after him and make him better	Claire, 11
	I’d deal with any illness at all	Claire, 10
	I’m obviously the main carer	Laura, 1
	I’m trying to keep the peace between everyone really	Laura, 7
	I can’t let him out of my sight	Laura, 9
	I suppose it’s a mother thing, I’m not going to have him written off, you know, that he can’t achieve what he wants to achieve and do those sorts of things	Laura, 10
	I don’t notice it as much being at work, [my wife] is the main carer	Simon, 2
	My father was never ill, well, he never talked about being ill, he just got on with it and I, I’m like that too	Simon, 7
	my father died from a coronary thrombosis when I was 21 but up until then he hadn't been ill, he just dropped dead.	Angus, 16
	Parents a team	
	I think really we work well together looking after Karen really	Barbara, 2
	we take it in turns to look after her	Barbara, 2
	we both took it on the shoulders and face the same, and that’s a good thing in many ways	George, 9
	I suppose you just group together and try to get through it and get on with it really. You just sort of bunker down	Laura, 19
	We do think quite similarly, and I think we’re quite good at counterbalancing each other	Claire, 6
	undoubtedly when the two of us are together, dealing with the situation, it’s better for the both of us	Andy, 7

	Appendix vi: Theme list with corresponding quotes	
	We most likely do tend to play off each other and, if one’s fulfilling one role, the other one of us fulfills another	Andy, 9
	we share doing things, so it doesn’t become one person constantly having to do it	Paul, 5
	[my husband] is fully on board now with anything and everything that we try and do so he’s been to the doctor’s with Hayley this evening, he’s got her round the chemist to get her prescriptions and he’s good as gold with that	Debbie, 3
	it’s enabled us to to talk through how we’re both feeling about something and come to an understanding or agreement about what the best way to go is	Andy, 3
	we just get on with it, we take on a different role or someone’ll do something for us to allow us to do something else	Debbie, 3
	We’re quite complimentary in the roles we take aren’t we?	Paul, 13
	I think we’re quite good at trying to get what the other person needs and doing a sort of tag team, really, aren’t we, be flexible	Paul, 6
	I suppose because we’re complimentary in our natures that [my daughter] gets different things from us	Paul, 6
	we do have a team approach...working in the same direction really	Anna, 9
	I think we’ve both been reasonably good at listening to what the other person’s had to say and trying to take that into account	Andy, 3
	you’re trying to work more as a couple and a unit with the whole.	Laura, 8
	The majority of the time we’re actually getting it right	Brian, 25
	I think [my husband] and I are more united	Sue, 10
	I think we’ve got the same opinion so we don’t particularly disagree about when the child should have time off	Sue, 15
	over the last couple of years we’ve been far more united	Sue, 7
	it’s all working together isn’t it really?	Debbie, 8
	I think there’s been (pause) a strain but to a certain extent we will talk about David in the same way that we talk about all our children, umm, and that’s something, that is an issue that we very much share (pause) I wouldn’t say that is, so that’s, the sharing problems is pretty positive for the relationship rather than negative	Brian, 6
	suddenly we were getting, you know, a bit more understanding and sympathy and it’s only when you suddenly mention the word that other people will come out and say ‘actually I’ve got a relative who has this’ or ‘I’ve got somebody who had that’, yeah, people start to share with you their experiences of people who’ve come out the far side which makes life all a bit better	Sally, 3
	It’s quite nice to have somebody with the same, very similar age child who’s been through it very recently and they’ve been really good	Claire, 17
	I mean it hasn’t confined us [to the house], we’re not feeling stuck	George, 3
	it’s not changed how we are with each other, I mean we’ve always been fine	Simon, 9
	it hasn’t caused arguments between us or anything like that so we still get on I think	Andy, 5
	it most likely hasn’t been dramatic but undoubtedly there has been some impact on us i’d say	Andy, 1
	It’s actually had a huge impact and one that we’re still only just realising really	Vicky, 8
	I don’t think it’s fundamentally changed how we are with each other or how we react to each other. - there are the basics to the relationship underneath that are the same.	Andy, 5
	this has probably made [myself and my son] very close I suspect	Laura,17
	hopefully she’ll be getting back on track	Sally, 8
	I think it’s very fortunate the way the cards have been dealt in our favour	George, 17
	we haven’t really argued	Andy, 3

Appendix vi: Theme list with corresponding quotes	
I’m a big believer in thinking positively about situations and thinking about what you can do rather than what you can’t do and I do talk about that sort of stuff quite a lot	Andy, 8
We never feel it’s pointless and hopeless and it’s not worth doing, we’re always looking for the way forward	Anna, 7
but we’re not fighting all the time	Laura, 8
I try to look on it that I’m having extra time with him	Laura, 19
Relationship improved? -	
I think it’s probably improved it in a way actually hasn’t it?	Claire, 7
I think certainly I’ve become far better over the period of time of saying to [my wife] actually how i’m feeling and something so I think I’m a bit more open to that now. And maybe also honest with myself.	Andy, 9
I’m always a glass is half full guy so that the positive, if you press on the positive all of the time then hopefully that’s going to help us get through it so it’s, if there are some less positive areas maybe we don’t need to know about them.	Debbie, 5
We do work well as a team and really in some ways you could argue it’s [Mum: more cohesive] strengthens that because we do get used to ‘oh well you do that’ and you know... we become better team workers sort of thing	Paul, 4
this has probably made us very close I suspect	Laura, 19
I’ll come out of the experience definitely stronger in that sense	Laura, 19
there have been upsides to it, I suppose, now I think about it	Laura, 20
I think actually, the way it’s been redistributed between us has been really good	Vicky, 16
i had a chronically sick mother...I’m probably well equipped to do it, doesn’t mean I want to	Laura, 11
there’s a certain amount of sailing along that comes with knowing the systems	Vicky, 8
working now with her is no different from what we’ve always done with karen you see	Barbara, 4
because you’re used to it, it’s a tolerance thing.	Vicky, 9
we got used to illness with [our other child] so dealing with her, you’re more used to it and more tolerant	Vicky, 13
if you haven’t been exposed to illness it’s harder to be understanding and tolerant really	Richard, 13
it hasn’t confined us to the house, that’s the main thing	George, 16
it doesn’t really drive us apart	Anna, 4
Has it changed our relationship? No, not at all.	Debbie, 2

Appendix vii

Systematic Review

“Does family therapy help children to cope with chronic illness?”

Systematic Review for the Professional Doctorate in Health Psychology

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Table of Contents

ABSTRACT

INTRODUCTION

Chronic illness presents children and families with a tremendous coping burden. There is a growing body of evidence demonstrating that families can have a beneficial or harmful effect on a family member's health. There is a paucity of systematic reviews looking at family therapy and childhood chronic illness

OBJECTIVES

To carry out a systematic review to assess the effectiveness of family therapy interventions in improving coping among children and adolescents with a chronic illness

METHODS

The review strategy was adapted from the Cochrane Collaboration Handbook for Systematic Reviews of Health Promotion and Public Health Interventions. A method of Narrative synthesis as described by Poppay et al. (2006) was used.

RESULTS

Following database searches, 892 references were located. Titles and abstracts were examined and 30 articles were acquired. After searching reference lists of acquired articles, 6 were examined.

CONCLUSIONS

Six studies evaluating the effectiveness of family therapy interventions met the inclusion criteria for this review. Four out of the six studies found improvements in coping. Studies highlight the importance of any sort of family intervention taking place within the context of a multidisciplinary treatment regime. For any sort of intervention to work there needs to be a good level of communication, both outside the family with regards to health professionals and services and also within the family between members

Note: The Systematic Review is presented in a format corresponding to the journal Archives of Diseases in Childhood.

INTRODUCTION

Traditionally treatment of illness tends to focus on the unwell individual. However, with any illness it is not only the individual that is affected, those close to the ill person will also be affected. Chronic illness presents children and families with a tremendous coping burden[34]. As well as the physical restrictions chronic illness exerts, the impact of chronic illness on children and adolescents can involve psychological and social repercussions [40], for example chronic illnesses has been associated with substantive emotional distress and suicide ideation, especially in female adolescents[43]; Strauss (1975) describes the way individuals strategically withdraw from the field of social interaction under the impact of chronic illness[42].

There is a growing body of evidence demonstrating that families can have a beneficial or harmful effect on a family member's health ([5,7,15]). Children who experience high family stress have been shown to be at higher risk for infections and hospitalization (Beautrais, A.L., Fergusson, D.M., & Shannon, F.T. (1982). Life events and childhood morbidity: A prospective study. Paediatrics, 70, 935-940.)Illness can then in turn exacerbate the dysfunctional traits of a family, and in effect a negative cycle is created. There is an assumption that addressing the dysfunction within the family through a family centred therapeutic approach can help to facilitate increased coping and positive adjustment in both the child and the family[21], which in turn can break the negative cycle. Essentially an intervention that addresses the system can have outcomes that directly benefit the individual. This is why a family therapy approach is suggested to provide a greater benefit than medical or psychosocial interventions focused solely on the patient in terms of improvements in both individuals' health and well-being[29].

Family therapy is a therapeutic approach to deal with individual's problems within the context of the family. Family therapists may work with the whole family, or with individual family members, but always with the overall view of the family in mind. There is a paucity of evidence looking at family therapy and childhood chronic illness. Only one systematic review was found that looked directly at family therapy and childhood chronic illness. Yorke and Shuldham (2005) aimed to assess the effects of family therapy as an adjunct to medication for the treatment of asthma in children[50]. Although they reported that there was some indication of the usefulness of family therapy they recognised that the findings were limited due to small study sizes and lack of standardisation in the choice of outcome measures.

A thorough review of the literature regarding family therapy for chronic illness in children will be conducted. The focus will be on the children’s ability to cope with having a chronic illness. Coping is defined by the Oxford English dictionary as to “deal effectively with something difficult”. To deal effectively with chronic illness children may need to adopt treatment regimens such as using asthma inhalers or injecting insulin, they may also need to psychologically adjust to their new way of life. In contrast to the previous review this review will include other chronic illnesses in addition to asthma.

METHODS

The review strategy was adapted from the Cochrane Collaboration Handbook for Systematic Reviews of Health Promotion and Public Health Interventions.

Search strategy for identification of studies

Individualized searches were run for each separate database, search results were initially screened for suitability by a single reviewer. The following databases were searched from their inception toFebruary 2010:Embase, Medline, Psychinfo, Pubmed, Sciencedirect, CINAL

Published and unpublished studies were considered for inclusion in the review. Grey literature was searched using SIGLE, ETHOS and Conference Proceedings Citation Index- Science (CPCI-S) via the Web of Science. The full search strategies for each database are included in Appendix i. Table 1 details the references found by searching each database and the abstracts retained by database.

In addition to the electronic searches, reference lists of included studies were searched to identify further published and unpublished research. Experts in the field were contacted regarding any published or unpublished research they would consider inclusion within the current review.

Search terms

Search terms pertaining to relevant participants, populations and interventions such as “children”, “adolescents” and “family therapy” were used to identify potentially relevant research. (see Appendix i for full list of search terms)

Inclusion and exclusion criteria

Published and unpublished studies were considered for inclusion in the review. Inclusion criteria were as follows:

1. A focus on children or adolescents (up to 18 years old) with chronic illness
2. A therapeutic approach that explicitly involves the family
3. An outcome measure used to assess coping

Studies were not excluded due to date of publication, study design or quality of methodology. Due to resource limitations the decision was made to exclude studies which were not published in the English language. As the review was focused on children and adolescents with chronic illness studies of adults with chronic illness were also rejected. Results were excluded by title if they were primarily mental health related (e.g. schizophrenia, eating disorder, bipolar), focused on the elderly or end of life care, or about drug or alcohol dependency and sexual abuse. Results were also excluded if they dealt with children's terminal illness' such as cancer. This was because it was felt that a family therapy in these cases would have quite different objectives from family therapy dealing with non-life threatening chronic illness. Similarly results focusing on chronic pain were excluded because it was felt that chronic pain presented a different set of treatment challenges than other chronic conditions, not least that there does not seem to be one "standard" treatment for chronic pain. Only primary research studies were included, books and reviews were therefore excluded.

Following the initial search, a three-phase screening strategy was used to identify relevant articles. Initially, two investigators independently screened potentially relevant studies based on a review of their titles and abstracts against the inclusion and exclusion criteria and agreed the removal of articles which were not relevant to the review question. Secondly, papers were obtained for examination independently by the two investigators against the same inclusion and exclusion criteria and agreement reached on articles which did not meet the selection criteria. Finally, both investigators independently reviewed the full text of remaining articles against the selection criteria and consensus was reached for their final inclusion in the review (figure 1). Data was extracted from the final papers used for the review using the full data extraction form (Appendix ii)

Figure 1. Data screening process:

Assessment of methodological quality

Quality assessments of the included studies were undertaken in order to ascertain the reliability of the available evidence. The quality assessments were carried out by two independent investigators and disagreements were resolved by discussion. The checklist for measuring study quality developed by Downs & Black (year) was used; this is a validated checklist that is recommended to be used to assess quality of randomised and non-randomised studies of health care interventions. The checklist is made up of 26 items relating to various aspects of a study's quality (Appendix iii). The items are split into 4 sub-groups (as described by Downs & Black):

Reporting (10 items, total score of 11) – to assess whether the information provided in the paper was sufficient to allow a reader to make an unbiased assessment of the findings of the study. External validity (3 items, total score of 3) – to address the extent to which the findings from the study could be generalised to the population from which the study subjects were derived.Bias (7 items, total score of 7) – to address biases in the measurement of the intervention and the outcome. Confounding (6 items, total score of 6) – to address bias in the selection of the study subjects.
Scores for each subgroup are combined to give an overall total score out of 27. A total score of 1 to 9 suggests low quality; a total score of 10 to 18 suggests medium quality; a total score of 19 to 27 suggests high quality. Quality scores are reported in the results section.

Data Synthesis

The six studies in the final review were similar in that they all assessed the effect of a family based intervention on children with a chronic illness, however the studies were a mix of RCTs and pre-post intervention and differed in three key areas: the family based intervention, the outcome measures used and the population (table 1). There was no “standard” family therapy “package” that was used across the studies, some used an individual therapeutic approach dealing with 1 family at a time, whereas some studies used a multifamily group approach; studies had a mix of outcomes looking at both practical aspects of dealing with the particular chronic illness (clinical outcomes) and psychological adjustment to having a chronic illness (relational outcomes); most of the studies used a clinical sample, however one study (Creedy et al. 2004) used a non-clinical sample. This heterogeneity was very high, in particular due to the above it was decided that a narrative synthesis approach would be appropriate for synthesis rather than a statistical synthesis of the data.

RESULTS

Study selection

Following database searches, 892 references were located. Titles and abstracts were examined and 30 articles were acquired. After searching reference lists of acquired articles, 6 were examined and included in the final review.

Methodological quality

Overall quality was assessed using the checklist for measuring study quality developed by Downs & Black (year?). Two studies achieved a high quality score; three studies achieved a medium quality score; one study was given a low quality score (Table 1). There were no disagreements between the two independent investigators.

Table 1. Study quality ratings

	Reporting	External validity	Internal validitiy (bias)	Internal validity (confounding)	Total	Overall quality
Onnis	10	1	6	5	22/27	High
Wamboldt	3	2	2	2	9/27	Low
Creedy	9	0	4	1	14/27	Medium
Satin	9	1	5	5	20/27	High
Lask	7	2	3	4	16/27	Medium
Gustafsson	6	1	5	5	17/27	Medium

Data synthesis

Narrative synthesis as described by Popay et al. (2006) is a textual approach to the process of synthesis to ‘tell the story’ of the findings from the included studies. This approach was used to synthesise the findings of this review.

Developing a preliminary synthesis

Table 2. Data extracted from papers

Author and year	Location and setting	Target population	Sample	Study design	Intervention details	Outcome measures	Main findings

Onnis, L. et al. (2001)	Italy. Family therapy service, Department of psychiatry and psychological medicine, “La Sapienza” University of Rome	Children with Astma treated at the Allergy Centre of the Paediatric Clinic	20 children with the most sever chronic bronchial asthma	RCT	Family psychotherapy, divided into 3 phases. Cycle of 10 to 15 fortnightly sessions for 5 to 7 months. Delivered by family therapists. Follow up6 months, 1 year and 2 years after end of therapy.	**Clinical** • Frequency of attacks per year • Frequency of hospitalization and emergency treatment per year • Absence of rapid response to therapy with bronchodilators (within 24-48 hours) without oral corticosteroids • Symptoms between attacks • Interference of symptoms with everyday activities • Differences in chest x-rays between attacks • Respiratory function testing (level of alteration between attacks • Asthma triggered by physical exercise. **Relational** • Enmeshment • Overprotectiveness type 1 (demand for overprotectiveness) • Overprotectiveness type 2 (offer of overprotectiveness) • Conflict avoidance type A (total conflict avoidance • Conflict avoidance type B (Early suppression of conflict) • Conflict avoidance type C (conflict	Change in the family’s dysfunctional interaction, and improvement in patient coping with asthma symptoms. Improvements remained stable after 2 years.

Wamboldt, M.Z. Levin, L. (1995)	USA. The National Jewish Centre for Immunology and Respiratory Medicine, Denver, Collorado.	272 families with children who have asthma treated at the Immunology and Respiratory Medicine	164 people representing 72 families	Pre-post intervention	Multifamily group intervention. 5 hour programme over 2 consecutive days. Delivered by clinical nurse specialists, psychotherapists and an art therapist Follow up consisted of an attitude survey immediately after the group.	**Relational** • The 6 item visual analogue scale survey	Families participating in a brief MFG perceive that people are more helpful and understanding after the group than before it. Families also place more importance on the value of sharing feelings regarding the illness with other family members after MFG than before.
Creedy, D. Lulow, T. Collis, D. & Cosgrove, S. (2004)	Australia. Gold Coast health services district.	Children within the Gold Coast health services district with chronic medical conditions.	12 children. diabetes (2), friedrich's ataxia (2), cystic fibrosis (4), arthrogryposis/ visual impairment (1), coeliac (1), asthma (1), lymphodema (1)	Pre-Post intervention	The Children and Parent Support (CAPS) programme. 8 week intensive programme Delivered by nurses, OTs and psychologists. Followup 3 months after end of programme.	**Relational** Parents completed: • A 5 point likert scale to assess parents' perceptions of the impact of the chronic illness on themselves and their child. • the Child Behaviour Checklist and • the Family Environment Scale. Children completed: • the Coopersmith Self Esteem Inventory • The Revised Child Manifest Anxiety Scale • the Child Depression Inventory.	All children reported improved levels of self-esteem after assessment, suggestive of improved coping. No significant changes in any other measures.

Satin, W. La Grca, A.M. Zigo, M.A. & Skyler, J.S. (1988)	USA. Diabetes/ metabolic unit, university of Miami Hospitals and clinics.	Children with insulin-dependent diabetes mellitus (IDDM) recieving treatment at the Diabetes unit in Miami	32 adolescents with IDDM	RCT	Multifamily group intervention. 6 weekly session, 90 minutes each. Delivered by 2 group leaders (roles not defined). Further guidance and support offered from a psychologic social worker and a nurse practitioner Follow up 3 and 6 months after end of group	**Clinical** • Metabolic control was measured at the beginning of treatment and 3 and 6 months later. **Relational** • Family and adolescent psychological evaluation at the beginning and 6 weeks after treatment. • Attitudes and perceptions were measured using semantic differentials. • Family Environment Scale pre and post treatment.	Data provides support for effectiveness of multifamily groups coupled with a parent simulation exercise for improving metabolic control among adolescents with IDDM
Lask, B. & Matthew, D. (1979)	UK. Asthma clinic at the Hospital for Sick Children, London.	Children with asthma attending clinic at the hospital for sick children, London.	29 children with grade C or D asthma (severe)	RCT	Family psychotherapy. 6 one hour sessions over 4 months. Delivered by a family therapist. Followup comprised of 1 six week assessment period one year after end of therapy	**Clinical** • Diary cards to record day-wheeze and activity limitation on a 0-3 scale. • Peak flow before am and pm medication. • 1 measure of forced expiratory volume • 1 measure of thoracic gas volume • Family and child psychological assessment before treatment.	Family Psychotherapy alleviated psychopathology within the family, and thus contributed to increased coping with asthma

Gustafsson, P.A. Kjellman, N-I, M. & Cederblad, M. (1985)	Sweden. Paediatric Outpatient Department of the University Hospital, Linkoping.	600 Children with asthma treated at the Paediatric Outpatient Department of the University Hospital, Linkoping.	17 of the most severe, chronic children	RCT	Family therapy. 2-21 sessions (mean=8.8) over 8 months Delivered by 2 family therapists Followup 8 months after end of therapy	**Clinical** • General Paediatric Assessment. (assessment of child's total situation) • Clinical grading (5 grade function scale) • Peak flow expiratory flow • Patient compliance • Emergency in-patient days and casualty visits • Daily functional ability diary completed by the parents • No. of doses of Beta-2-agonists and no. of nights beta-2-agonists had to be inhaled • Prescription of steroids for periods longer than 2 months	Family therapy appeared to improve children's ability to cope with severe asthma.

Narrative synthesis

Essentially family therapy is a therapeutic approach that treats the whole family (Gustaffson et al, 1985). This is true for all the studies included in this review, however the way this therapy was delivered, by whom and the outcomes measured differed across the studies. Therefore, as a framework to explore the findings in this review an approach recommended by Popay et al. arranging studies into groups was used. It is possible that studies can be grouped in various different ways depending on what aspect one focuses on, one can then explore the notion of coping with regards to these groupings. The studies included in the current review have been arranged into the following groups: Type of Intervention; Population studied; Type of study (Table 3).

Table 3. Grouping

Grouping according to:	
(1) Delivery	**(2) Outcome measures**
Individual family sessions with family therapists: Onnis et al. (2001); Lask et al. (1979); Gustafsson et al. (1985)	Clinically relevant outcomes with regards to coping with symptoms of the chronic illness: Lask et al (1979); Gustafsson et al (1985).
Group sessions with multidisciplinary teams: Wamboldt and (1995); Satin et al. (1988); Creedy et al (2004).	Relational outcomes with regards to emotional adjustment to the chronic illness: Wamboldt et al (1995); Creedy et al (2004).
	Clinical and relational outcomes: Onnis et al (2001); Satin et al. (1988)

Delivery; Individual vs. Group

The type of intervention used in the studies can be categorised as either Individual or Group. Individual therapy was delivered either weekly or fortnightly by trained family therapists to one family with no standard amount of sessions, the minimum and maximum amount are reported by Gustaffson et al: 2 to 21 sessions over 8 months with two therapists. Onnis et al report 10 to 15 fortnightly sessions for 5 to 7 months with more than 1 therapist while Lask et al report 6 one hour sessions of family psychotherapy over 4 months with 1 therapist. Clearly some families do not require as much therapy contact as other families, and an individual approach can be flexible enough to deal with this. However, this makes it difficult to assess what would be the optimal duration of family therapy.

Although there is no standard model described for the type of therapy used in these studies Gustafsson et al. define family therapy as “treating the whole family, or parts of it, with psychological and pedagogical methods aimed at changing interpersonal relations”, Two therapists delivered the therapy, highlighting and helping to change dysfunctional patterns of interaction, explored the role of the asthma in the family, reveal conflicts, strengthen boundaries and enhance communication about the emotional impact of asthma on the family. Onnis et al describe the therapy in terms of three phases The initial phase which serves as an information gathering phase, the intermediate phase concerned with exploring and uncovering how the family unit works, the closing phase deals with children and parents separately to help strengthen and clarify relationships. All three studies assessing individual family therapy found that this approach could enhance coping in children with chronic illness.

Group therapy involved more than one family taking part in what was described as either a “multifamily group intervention” (Satin et al. 1988, Wamboldt & Levin 1995) or a “Child and Parent Support programme” (Creedy, 2004). This type of group approach involved a multidisciplinary team, rather than just family therapists in the individual approach. Wamboldt & Levin studied groups delivered by clinical nurse specialists, psychotherapists and an art therapist. In the Creedy study nurses, OTs and psychologists were involved in the delivery of groups. Satin et al describe their groups as being delivered by 2 group leaders, though they do not state their roles. Further guidance and support offered from a psychologic social worker and a nurse practitioner. Compared to individual family therapy the impact group therapy has on children’s coping is less clear. Satin concluded that the adolescents in his study coped more with their diabetes after the family intervention that before. However, In the Wamboldt study it is unclear whether there were positive effects on the child as the researchers only looked at parental attitude change. Creedy concluded that the children in her study did not cope any better than they did before the intervention, however she suggested that the range of coping strategies they used was larger post intervention.

Outcome measures used; Clinical, relational or both

Outcome measures used in the studies were either clinical, relational or both. Clinically relevant outcome measures were for example reduction in use of medication to control the illness, or reduction of symptoms. Relational outcomes were those that assessed the impact of the illness on the relationships of parents with their children or the emotional impact the illness had on either the parents or children. Though different both types of outcomes are directly related to the extent to which the child copes with the illness.

Clinical:

The studies that just looked at clinical outcomes observed improvements in coping and concluded that family therapy assisted this improvement. Gustafsson et al (1985) suggested that their findings supported the idea that family therapy improves asthma in children. Improvements were found to be stable over three and a half years after therapy. Lask et al. (1979) also conclude that family therapy can improve children's ability to cope with asthma. They found that peak flow significantly improved for the experimental group at followup and they observed significant change for the experimental group in day-wheeze score. They explain this improvement in terms of family treatment helping to "alleviate psychopathology within the family" and therefore contributing to an alleviation of asthma. However, neither studies measure "family psychopathology", it is therefore difficult to draw firm conclusions regarding this. To explore this link more fully studies use relational outcomes.

Relational:

Creedy et al. (2004) completed all outcome measures before their programme over the phone, and 3 months after completion of the programme. They found that children's self esteem scores significantly increased from pre to post group assessment. No significant difference was found in mean scores for FRI, RCMAS, CDI and CCSC from pre to post assessment. Parental perceptions of impact reduced significantly pre to post group assessment. The authors conclude that the study suggests that psychosocial support groups enable children to understand and manage the demands of having a chronic condition by receiving peer support and learning balanced coping strategies.

Wamboldt & Levin (1995) administered a survey before and after every 2 hour group session. They conclude that families participating in a brief MFG perceive that people are more helpful and understanding after the group than before it. They suggest that families also place more importance on the value of sharing feelings regarding the illness with other family members after MFG than before.   It is not clear within the Wamboldt study whether the 72 families out of the 272 families who attended the multifamily groups completed the attitude survey because they were the families who completed the groups or whether the other families also completed the groups but didn't complete a survey. Wambolt also makes no reference to the validity or reliability of the attitude survey they used as an outcome measure.

When looking at either clinical or relational outcomes in isolation one effectively is only getting half the picture; children may be coping with their treatment regime but what about their emotional coping? Similarly if it is suggested that a child is coping in an emotional sense does this also reflect a greater ability to cope with symptom management? Therefore to get a fuller picture of coping it seems sensible to use both clinical and relational outcomes.

Clinical and relational

Both the Onnis et al. (2001) and Satin et al. (1988) studies included both clinical and relational outcomes. Onnis et al. reported an improvement in both the clinical and relational parameters in the experimental group. The improvements were still evident two years after the intervention. However, they only used the relational measure with the experimental group and not the control group. It is therefore difficult to draw firm conclusions from their findings.

In the Satin et al (1988) study both groups that received multifamily group intervention showed improvements in metabolic functioning (statistically and clinically) as compared to control group. An improvement was also observed in Attitudes and perceptions for the experimental groups. No significant changes in FES were observed. Improvement sustained 6 months after intervention. They conclude that the data provide support for effectiveness of multifamily groups coupled with parent simulation exercise for improving metabolic control in adolescents with diabetes. The authors also suggest that a more positive self image was gained from taking part.

DISCUSSION

Six studies evaluating the effectiveness of family therapy interventions met the inclusion criteria for this review. Four out of the Six studies found improvements in coping. Satin concluded that enhanced coping with diabetes was apparent in adolescents who had participated in the family intervention. In their three studies Gusstafsson, Lask and Onnis found improvement in family functioning post family therapy and concluded that this had a direct affect on the child's coping with asthma symptoms. All 6 studies represent two approaches to family therapy, group based and individual. Both an individual and group approach highlight the importance of communication with regards to positive outcomes. On one level communication is seen as important between healthcare services or healthcare professionals, the studies recommend multidisciplinary approaches, this is perhaps best encapsulated by Onnis with the statement: "this interdisciplinary approach must be a dialectical expression of various viewpoints, an expression and integration based on a shared methodology that can be a useful systemic methodology".

Good communication between families and treatment teams is also seen as important, Wamboldt describes the setting up of "therapeutic alliances" between families and health care staff as enabling better care, while communication between families in the form of peer support and allowing for normalization. Creedy espouses the way a parent and child support group can help children to gain a shared understanding of their experiences, normalise responses and reduce social isolation: Both Satin and Wamboldt highlight the opportunity though group work for families to share their experiences of living and coping with chronic illness.

A lack of communication is related to dysfunction and enhanced communication is related to an increase in coping. Gustafsson describes lack of communication embodied in parent's permissiveness in the face of illness as a maintaining factor in chronic illness, and a breakdown in communication as shown by conflict within the family can fuel illness.

Communication can also be seen as a metaphor for the interplay between the mind and the body. Onnis states that "soma and psyche are not two separate, non-communicating realities but are integrated and act together". It is suggested that a splitting of the two whether this be at the level of treatment (whereby treatments are all completely separate), or within the expectations and knowledge of the family or individual, has detrimental effects on the patient's prognosis.

As well as communication it seems that the studies also highlight the idea of "acceptance" as allowing for a greater feeling of control over an illness. This idea of acceptance can be seen in the sense of families having to accept that traditional models of health may not be useful when considering chronic illness. Onnis makes the point that the traditional dichotomous mind-body view may hamper recovery because it leaves a large group of patients with the impression that there is nothing that can be done to help them to recover. It is therefore much better to integrate the concepts of mind and body by making sure that services and health professionals work together in a multidisciplinary setting. In addition the process of family therapy seems to allow for families to accept the "reality" of illness, this occurs through a process of comparison and reflection. Comparison is more apparent in the multifamily group format when families compare themselves to other families, while reflection seems to be more apparent in an individual family therapy setting.

Strength of the evidence

It is unclear whether the studies used representative samples, if the group being studied is not representative of the wider population this effectively weakens the evidence. Participants in the Onnis study were not representative of the entire population from which they were recruited; they were the most serious cases of chronic bronchial asthma seen in clinic. This was also the case with the Gustaffson study and the Lask study. In the Lask study participants were recruited from all those who attended the clinic, however there is no description of this population other than the fact that they may have been referred for a 2nd or 3rd opinion, suggesting that the diagnosis was probably not clear, possibly because the symptoms were not particularly severe among this population. Satin et al. (1988) state that the characteristics of families who did not elect to participate in the study are unknown, therefore it is impossible to assess whether the sample used was representative.

Subjects in the Creedy et al study (2004) were not representative of the entire population because the researchers essentially "advertised" to the general public through newspaper and radio. There are two main problems with this method of recruitment, firstly it relies on families to value research and be willing to take part, secondly it requires families to recognise and accept that they may have a problem with coping and be willing to accept support for this. These problems are reflected in the slow recruitment rate and low uptake of the study. Wamboldt et al. highlight a potential problem that often groups that are most at risk are also those that may be least likely to follow through on a suggestion from their primary physician that the family seek mental health care. Wamboldt state that 272 families attended the multifamily group intervention over the study period, however they report that only 72 families completed their outcome measure during this same period. There is no information reported as to whether this smaller group of families were representative of the whole population completing the intervention.

There were methodological issues that weaken the strength of the evidence presented in the studies. Onnis et al. (2001), a "high quality" study, had a major limitation to their methodology whereby a main outcome measure was only used with the experimental group, thereby severely weakening the potential evidence of effectiveness. This also suggests that in this case the quality measure was inadequate for the type of studies included in this review. In the Satin study the recruitment method fuels the suspicion that the sample was most likely not representative, the researchers used a notice in the hospital unit that patients were being treated. As with the Creedy study (2004) the researchers recognise that it was likely that the selection procedures favored families who were receptive toward behavioural/psychosocial interventions and who were concerned about their youngster's health. Often families taking part in the studies where those who were already being treated, and had therefore already sought medical assistance, these may be families who are already willing to adhere to medical advice and change their behaviour accordingly. Satin et al reflect on the possibility that their selection criteria may well have favoured families receptive toward behavioural/psychological interventions and who were concerned about their youngsters' metabolic functioning. Wamboldt et al. suggest that families perceive that people are more helpful and understanding after participating in multifamily group therapy and also recognize the importance of a shared communication within the family. However, the families who completed the questionnaires may have already been more willing to accept that there needed to changes in the way they dealt with their child's chronic illness. The families who did not complete the questionnaire may have been unhappy with the group intervention. However, this is supposition as those who did not complete the attitude survey are not described.

Limitations of the review process

Although this review has been conducted in accordance with the Cochrane Collaboration guidelines for systematic reviews a number of factors may limit the findings. It is possible that unpublished studies, studies from non-indexed journals and relevant studies from lesser known databases may have been missed. The possibility of publication bias cannot be excluded. Only six relevant studies were identified in this review thereby limiting the conclusions that could be drawn.

The studies included in the final review were all English language. There were a number of studies that were excluded because they were not in English. It is recognised that this is a limitation of the current review which, with more time and resources, should be rectified should the review be repeated.

The process of locating evidence was as rigorous as it could be with the limited resources and time available to the reviewer. Through the process of locating the evidence the main internet databases were used, grey literature was searched and hand searches of journals were conducted, along with locating further useful material from key paper reference lists. Two attempts were made to contact experts in the field by email, however no response was forthcoming.

All studies included in the final review were quality assessed using the checklist formulated by Downs and Black (1998). Item 8 of the checklist "Have all the important adverse events that may be a consequence of the intervention been reported" was problematic to assess. This was not directly addressed in any of the studies included in the review. This may be because there was the assumption that a family approach was the best to use. The authors all have a multidisciplinary clinical background; it is therefore unsurprising that they would support an integrative systemic approach.

One possible adverse effect from an intervention treating the family rather than the individual the danger is that family members in some way feel to blame for the slow recovery of illness. A family approach can uncover uncomfortable issues that should be identified and worked through. Satin et al. report that after multifamily group sessions fathers of adolescents with diabetes tend to view themselves more negatively over time. They suggest that this may reflect the father's greater awareness of difficulties in diabetes management. Wamboldt et al (1995) describe how through the process of sharing experiences in a multifamily group setting many parents are very emotional after listening to the children speak.

Narrative synthesis is sometimes seen as a "poor cousin" of meta analysis, the Cochrane guidance argues that:

'a possibility that systematic reviews adopting a narrative approach to synthesis will be prone to bias, and may generate unsound conclusions leading to harmful decisions'

With an awareness of this the reviewer took steps to make sure that the current synthesis was conducted in the best way possible, following the published guidance by Popay et al.

The reviewer works in the area of chronic illness with families, therefore has prior knowledge and experience of the impact of chronic illness on families. It is likely that this affected the judgements made through the review process, in particular during the thematic analysis. A reviewer with no prior experience of working with families with and chronic illness may have reached different conclusions. However, it could also be suggested that with prior experience in the area the reviewer was able to provide a greater understanding of the relevant issues that a reviewer without prior experience may well lack. From this point of view it could be suggested that this is a strength rather than a limitation.

Only one study explicitly referred to a theoretical basis to the intervention. Creedy et al. (2004) assess the Children and Parent Support (CAPS) programme based on 4 principles derived by Dunst et al. (2002), these are as follows: Firstly identifying client needs, second focus on strengths, third built and strengthen natural support systems, and forth create opportunities to display strengths.

All studies observed sustained positive effects at follow-up. However there was no standard follow-up point across studies. For family level interventions to be sustainable the research suggests that there needs to be continued communication between healthcare providers and families.

CONCLUSIONS

The objective of this review was to assess the efficacy of family therapy in improving coping among children and adolescents. Overall findings suggest that family therapy may be a potentially effective treatment in increasing coping among children and adolescents. There are fundamental systems that are described within the studies; these are the family, the individual (made up of psyche and soma) and healthcare. These systems all interact with each other. Stress affects both the family system and the individual system. Healthcare can be psychological, medical or a mix of both (multidisciplinary). A multidisciplinary approach is preferable as it incorporates both the medical and the psychological. An issue with splitting the psychological and the medical is that the message is sent out that the body and the mind (soma and psyche) and completely separate, this in turn can cause stress for the family and individual.

Family therapy tends to be seen as a “psychological” treatment either a in a group or individual setting. The outcomes of family therapy are explicitly relational, in that interactions and relationships are changed and therefore the child’s coping will be improved, however there can also be implicit clinical changes whereby families and children will manage their illness more affectively. For any sort of intervention to work there needs to be a good level of communication, both outside the family with regards to health professionals and services and also within the family between members. If there is a breakdown of communication the intervention simply won’t have the desired effect. There also needs to be awareness that a family therapy approach may uncover uncomfortable truths for family members and they need to be supported through this process.

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Abstract I. *Full Search Strategies*

CINAL

1. AB Family or families or kinship network or stepfamily or stepfamilies or relatives or extended family
2. AB male or female or teen or youth or adolescent
3. AB boy or girl
4. family/
5. minors/
6. child/
7. adolescent/
8. AB chronic disease or chronic illness or chronically ill
9. chronic disease/
10. AB group psychotherapy or group therapy
11. (group psychotherapy or group therapy).tw.
12. group psychotherapy/
13. AB group psychotherapy/
14. AB exp psychotherapy, group/
15. AB family therapy
16. family therapy.tw.
17. family therapy$.tw.
18. family therap$.tw.
19. family therapy/

Embase
1. exp adolescence/
2. adolescent/
3. child/
4. juvenile/
5. minor.mp.
6. youth.mp.
7. child$.mp.
8. teen$.mp.
9. adolescen$.mp.
10. family.mp. or exp family/ or exp extended family/
11. exp family centered care/ or exp family assessment/ or exp family systems theory/ or exp family therapy/ or exp family counseling/
12. group therapy/
13. group therapy.mp.
14. 1 or 2 or 3 or 4 or 5 or 6 or 7 or 8 or 9 or 10
15. chronic disease/
16. chronic disease.mp.
17. chronic illness.mp.
18. 15 or 16 or 17
19. 14 and 18
20. 11 or 12 or 13
21. 19 and 20

MEDLINE

1Child$/
2Adolescen$/
3Teen$/
4Minors/
5(child$ or adolescen$ or teen$ or minors).tw.
6Young person.tw.
7Young people.tw.
8Girl$.tw.
9Boy$.tw.
10Teenager$.tw.
11Student$.tw.
12(young person or young people or girl$ or boy$ or teenager$ or student$).tw.
13or/1-12

14(chronic adj illness)/
15Fatigue.tw.
16Asthma.tw.
17Chronic/

18Cystic fibrosis.tw.
19Headache.tw.
20Migraine.tw.
21or/14-20

22Psych$/
23Intervention$/
24(psych$ or intervention$).tw.
25(Evidence-based psychological intervention$).tw.
26Psychopathology.tw.
27(Self adj help)/
28(Guided adj CBT).tw.
29(Minimal adj contact)/
30(Minimal adj contact).tw.
31Or/22-30

32No Psychological Intervention.tw.
33therapist adj (led or intervention).tw.
34Group intervention/
35Family based intervention/
36Systemic/
37Didactic/
38CBT/
39(group intervention or family based intervention or systemic or didactic).tw.
40Cognitive/
41Behaviour$/
42Therap$/
43(cognitive or behaviour or therap$).tw.
44Cognitive–behavioral treatment.tw.
45Group treatment.tw.
46Or/32-45

47RCT/
48Qualitative/
49Quantitative/
50cohort study/
51Comparison group.tw.
52Random allocation.tw.
53Outcome study/
54Or/47-53

5513 and 21 and 31 – children and chronic illness and psychological self help
5613 and 30 and 46 – children and chronic illness and therapist led psychological intervention

Psychinfo

1. child.mp.
2. adolescent.mp.
3. exp Only Children/
4. exp Family/ or exp Adolescent Attitudes/
5. teen.mp.
6. youth.mp.
7. teen$.mp.
8. childhood.mp.
9. kidnapping/ or exp kindergarten students/
10. exp kindergarten students/
11. exp Chronic Illness/
12. chronic illness.mp.
13. (male$ or female$ or teen$ or youth$ or adolescen$).mp.
14. exp Family Crises/ or exp Family/ or exp Extended Family/ or exp Family Relations/ or exp Family Structure/ or exp Family Intervention/ or exp Biological Family/ or exp Family Therapy/ or exp Family Systems Theory/
15. family.mp.
16. exp Interracial Family/ or exp Nuclear Family/ or exp Biological Family/ or exp Family/ or exp Interethnic Family/ or exp Extended Family/
17. exp Family Intervention/ or exp Family Therapy/ or exp Family Systems Theory/
18. group psychotherapy/
19. child$.mp.
20. adolescen$.mp.
21. 4 or 10 or 13 or 15 or 16 or 19 or 20
22. 11 or 12
23. 21 and 22
24. family therapy.mp.
25. 17 or 18 or 24
26. 23 and 25

PUBMED

1. "child"[MeSH Terms] OR "child"[All Fields] OR "children"[All Fields]
2. "adolescent"[MeSH Terms] OR "adolescent"[All Fields]
3. "adolescent"[MeSH Terms] OR "adolescent"[All Fields] OR "teenager"[All Fields]
4. "adolescent"[MeSH Terms] OR "adolescent"[All Fields] OR "youth"[All Fields]
5. ("adolescent"[MeSH Terms] OR "adolescent"[All Fields] OR "youth"[All Fields]) OR ("adolescent"[MeSH Terms] OR "adolescent"[All Fields]) OR ("child"[MeSH Terms] OR "child"[All Fields]) OR ("adolescent"[MeSH Terms] OR "adolescent"[All Fields] OR "teenager"[All Fields])
6. "family"[MeSH Terms] OR "family"[All Fields]
7. "chronic disease"[MeSH Terms] OR ("chronic"[All Fields] AND "disease"[All Fields]) OR "chronic disease"[All Fields] OR ("chronic"[All Fields] AND "illness"[All Fields]) OR "chronic illness"[All Fields]
8. "family therapy"[MeSH Terms] OR ("family"[All Fields] AND "therapy"[All Fields]) OR "family therapy"[All Fields]

9. (("child"[MeSH Terms] OR "child"[All Fields]) AND ("adolescent"[MeSH Terms] OR "adolescent"[All Fields]) AND ("adolescent"[MeSH Terms] OR "adolescent"[All Fields] OR "teen"[All Fields]) AND ("adolescent"[MeSH Terms] OR "adolescent"[All Fields] OR "youth"[All Fields]) AND ("family"[MeSH Terms] OR "family"[All Fields])) AND "family therapy"[All Fields] AND "chronic illness"[All Fields]

Sciencedirect

1. (((Chronic PRE/0 illness) OR (Chronic illness OR Chronic disease)) AND ((family) AND (child OR adolescent OR teen OR young person))) AND (TITLE-ABSTR-KEY(family PRE/0 therapy)) AND EXCLUDE(topics, "schizophrenia,mental health,eating disorder,schizophrenia bulletin,sexual abuse") AND EXCLUDE(topics, "hiv infection")
2. (((Chronic PRE/0 illness) OR (Chronic illness OR Chronic disease)) AND ((family) AND (child OR adolescent OR teen OR young person))) AND (TITLE-ABSTR-KEY(family PRE/0 therapy)) AND EXCLUDE(topics, "schizophrenia,mental health,eating disorder,schizophrenia bulletin,sexual abuse")
3. (((Chronic PRE/0 illness) OR (Chronic illness OR Chronic disease)) AND ((family) AND (child OR adolescent OR teen OR young person))) AND (TITLE-ABSTR-KEY(family PRE/0 therapy))
4. ((Chronic PRE/0 illness) OR (Chronic illness OR Chronic disease)) AND ((family) AND (child OR adolescent OR teen OR young person))
5. (Chronic PRE/0 illness) OR (Chronic illness OR Chronic disease)
6. (family) AND (child OR adolescent OR teen OR young person)
7. TITLE-ABSTR-KEY(family PRE/0 therapy)
8. Chronic PRE/0 illness
9. Chronic illness OR Chronic disease
10. TITLE-ABSTR-KEY(family therapy)
11. family
12. child OR adolescent OR teen OR young person
13. TITLE-ABSTR-KEY(chronic PRE/0 disease)
14. (TITLE-ABSTR-KEY(family)) AND (TITLE-ABSTR-KEY(adolescent OR teen OR child))
15. ((TITLE-ABSTR-KEY(family)) AND (TITLE-ABSTR-KEY(adolescent OR teen OR child))) AND ((TITLE-ABSTR-KEY(chronic PRE/0 disease)) OR (TITLE-ABSTR-KEY(chronic PRE/0 illness))) AND (TITLE-ABSTR-KEY(family PRE/0 therapy))
16. (TITLE-ABSTR-KEY(chronic PRE/0 disease)) OR (TITLE-ABSTR-KEY(chronic PRE/0 illness))
17. child OR adolescent OR teen OR young person
18. TITLE-ABSTR-KEY(chronic PRE/0 illness)
19. TITLE-ABSTR-KEY(family PRE/0 therapy)
20. TITLE-ABSTR-KEY(family)

Appendix II. *Data Extraction Form*

“Does family therapy help children to cope with and adjust to chronic illness?”

Data Extraction Form

Paper number:

Year of Paper:	**Reviewer (initials):**
1st Author:	**Journal:**

Include the paper (tick one)?: Yes No

Please tick reasons for inclusion:

The focus is on children with chronic illness

It involves some sort of therapeutic approach that explicitly involves the family

It is clear that some sort of measure of coping or adjusting was used to assess outcome such as a self-efficacy scale

Tick the following boxes if these are clear from the paper, if not leave blank:

Design of study	observational	intervention
Focus of study	outcome	onset
Sample size	n < 50	n = > 50

What is the Chronic illness? ....

Please complete the following details where possible:

Intro	
Hypothesis and aims?	Page:
Methods	
Sample size	Page:
Population characteristics	Page:
Intervention details	Page:
Theoretical framework	Page:
Provider	Page:
Setting	Page:
Target group	Page:
Study details (date, follow-up)	Page:
Process measures – adherence, exposure, training, etc	
Results	
Missing data, loss of patients	Page:
Response rate reported?	Page:

Drop out reported?	Page:
Sources of bias	Page:
Confounding	Page:
Significance – clinical Vs statistical	Page:
Statistical analysis used?	Page:
What was the outcome and how was it measured?	Page:
Briefly, what were the author's conclusions?	Page:
Discussion	
Interpreted correctly (taken into account study hypothesis, sources of potential bias or imprecision and the dangers associated with multiplicity of analyses and outcomes.	
Limitations?	Page:
Consistent with previous studies?	Page:
Notes:	

Appendix III. *Quality Assessment Form*

	Score		
Reporting	Yes (1)	No (0)	Unable to determine (0)
1. Is the hypothesis/aim/objective of the study clearly described?			
2. Are the main outcomes to be measured clearly described in the Introduction or Methods section?			
3. Are the characteristics of the patients included in the study clearly described ?			
4. Are the interventions of interest clearly described?			
5. Are the distributions of principal confounders in each group of subjects to be compared clearly described?			
6. Are the main findings of the study clearly described?			
7. Does the study provide estimates of the random variability in the data for the main outcomes?			
8. Have all important adverse events that may be a consequence of the intervention been reported?			
9. Have the characteristics of patients lost to follow-up been described?			
10. Have actual probability values been reported(e.g. 0.035 rather than <0.05) for the main outcomes except where the probability value is less than 0.001?			
External validity			
11. Were the subjects asked to participate in the study representative of the entire population from which they were recruited?			
12. Were those subjects who were prepared to participate representative of the entire population from which they were recruited?			
13. Were the staff, places, and facilities where the patients were treated, representative of the treatment the majority of patients receive?			
Internal validity - bias			
14. Was an attempt made to blind study subjects to the intervention they have received ?			
15. Was an attempt made to blind those measuring the main outcomes of the intervention?			
16. If any of the results of the study were based on “data dredging”, was this made clear?			
17. In trials and cohort studies, do the analyses adjust for different lengths of follow-up of patients, or in case-control studies, is the time period between the intervention and outcome the same for cases and controls ?			
18. Were the statistical tests used to assess the main outcomes appropriate?			
19. Was compliance with the intervention/s reliable?			
20. Were the main outcome measures used accurate (valid and reliable)?			
Internal validity - confounding (selection bias)			
21. Were the patients in different intervention groups (trials and cohort studies) or were the cases and controls (case-control studies) recruited from the same population?			
22. Were study subjects in different intervention groups (trials and cohort studies) or were the cases and controls (case-control studies) recruited over the same period of time?			
23. Were study subjects randomised to intervention groups?			

24. Was the randomised intervention assignment concealed from both patients and health care staff until recruitment was complete and irrevocable?			
25. Was there adequate adjustment for confounding in the analyses from which the main findings were drawn?			
26. Were losses of patients to follow-up taken into account?			