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**What is the impact of CFS/ME and other chronic health conditions on siblings: A
systematic literature review**

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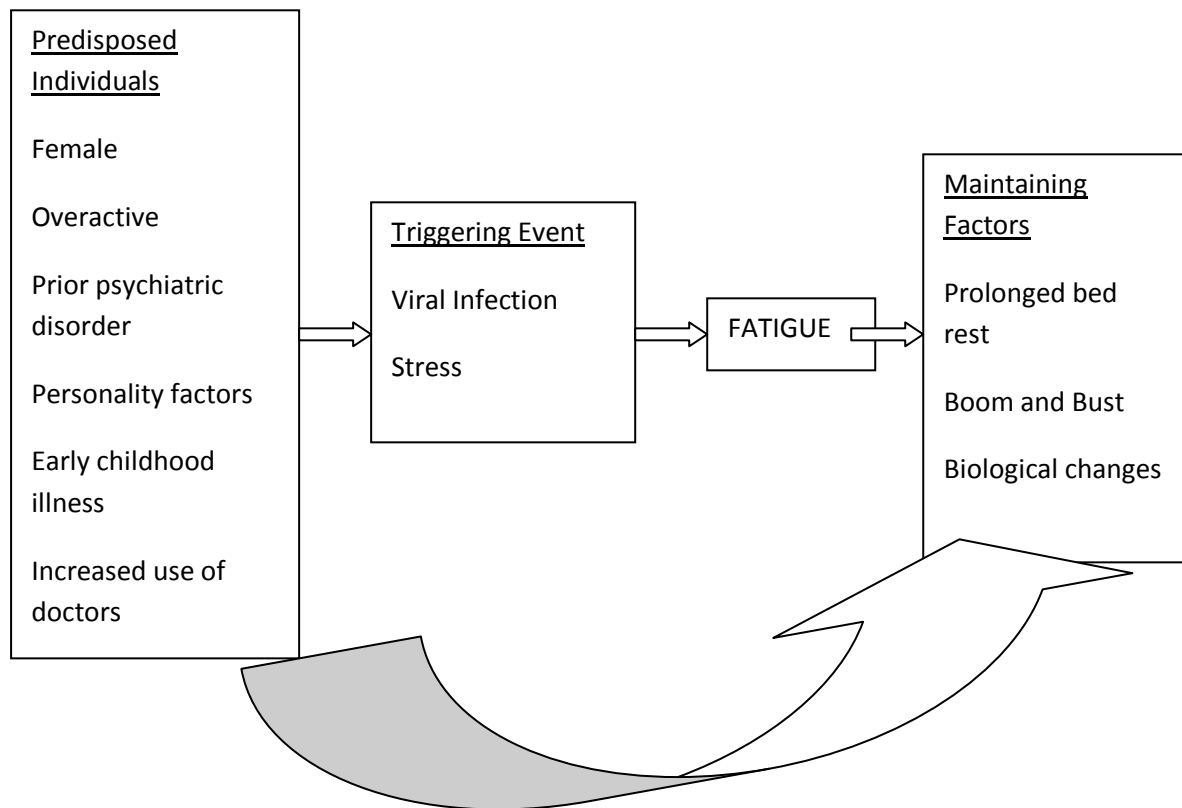
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Introduction

The Royal College of Paediatrics and Child Health in the UK defined chronic fatigue syndrome or myalgic encephalopathy (CFS/ME) as “generalised fatigue, causing disruption of daily life, persisting after routine tests and investigations have failed to identify an obvious underlying ‘cause’” (Royal College of Paediatrics and Child Health, 2004). Harvey & Wessely (2009) suggested a model for understanding the aetiology of CFS/ME in adults (Figure 1). Although no such model has been suggested for children, Patel, Smith, Chalder & Wessely (2003) have suggested that symptoms are similar between children and adults and most commonly include muscle ache, sore throat, headache, abdominal pain, concentration difficulties, increased drowsiness and depression (Feder, Dworkin & Orkin, 1994; Carter, Edwards, Kronenberger et al, 1995; Smith, Mitchell, Corey et al, 1991; Marshall, Gesser, Yamanishi et al, 1991; Krilow, Fisher, Friedman et al, 1998; Bell, 1995). The National Institute for Health and Clinical Excellence (NICE) guidelines recommend children have fatigue for a minimum of 3 months before making a diagnosis (NICE, 2007). The prevalence rates are between 0.19 to 2% of children (Chalder, Goodman, Wessely, Hotopf, & Meltzer, 2003; Jones, Nisenbaum, Solomon, Reyes, & Reeves, 2004; Jordan et al., 2000). CFS/ME has a negative impact on a child’s schooling (Sankey, Hill, Brown, Quinn, & Fletcher, 2006; Crawley & Sterne, 2009; Rangel, Garralda, Levin, & Roberts, 2000), their social relationships (Bell, Jordan, & Robinson, 2001) and their parents (Missen, Hollingworth, Eaton, & Crawley, 2011).

Figure 1: Harvey & Wessely Model for understanding aetiology behind CFS/ME in adults.



Little is known about the impact on siblings' of children with CFS/ME. A number of stressors were identified when talking informally to a parent support worker (Jackson, 1999), including worries about "apparent parental dilution of care or concern" (Jackson, 1999, p31); a change in the sibling relationship; restrictions on family activities; deterioration in peer relationships; and the uncertain or contradictory medical advice given to the family. There are a number of problems with this paper, Jackson (1999) did not describe the method or analysis used, there were no quotes, and terms, such as "parental dilution of care" were not explained. Siblings were recruited whilst visiting the child with CFS/ME in hospital. As few children with

CFS/ME are hospitalised, this is not a representative sample. This is the only study which has explored the impact of CFS/ME on siblings.

The attached major research project explores the quality of life and psychological wellbeing of siblings of children with CFS/ME. The systematic literature review explores the impact of other paediatric chronic health conditions (CHCs) on siblings to increase our knowledge on what the possible impact may be for siblings of children with CFS/ME.

What is the impact of chronic health conditions on healthy siblings?

Literature search

The databases, Web of Knowledge/Science, PsycInfo, Medline (PubMed) CINAHL and PsycARTICLES) were systematically searched using the terms “Impact” AND “Sibling*” and “Sibling*” AND “Chronic illness” were searched between 23rd August 2010 to 17th September 2010, and then updated on 10th January 2012. The term “psycholog*” was initially included, but did not draw any additional papers to “impact”.

In gathering studies for the present review the following inclusion/ exclusion criteria were observed (Table 1):

Table 1

Inclusion and exclusion criteria for literature search

Inclusion Criteria	Exclusion Criteria
Research was written in English	If the ill child had died, as this was felt to have a different impact on the sibling.
Research was published in a peer reviewed journal	If the majority of the siblings were over 18 years of age
Research included the above search terms	If the research described an intervention

in 'keywords'.

Research included conditions that were long-term and had a daily impact, or that were life threatening or life limiting.

If the siblings had not taken part in the research themselves. This is due to findings that parental reports do not accurately reflect sibling reports in childhood chronic illnesses (Sharpe & Rossiter, 2002; Guite, Lobato, Kao, & Plante, 2004).

If the article was published before 1990.

If the articles did not describe original research.

Three thousand, four hundred and twenty five titles were found, and of these 431 abstracts were studied. In total 47 original pieces of research met the inclusion criteria of this review. Citation and reference lists were also studied to ensure no articles were missed.

Results

The 47 research papers included in this study are outlined in Table 2 (Appendix A). Twenty-eight of the studies used quantitative techniques, 15 were qualitative and four used mixed methods. The majority of the qualitative studies used interview data, although a few used diaries or written narrative. Most of the quantitative studies explored the emotional wellbeing and quality of life of the sibling, the sibling relationship or the family cohesion using standardised questionnaires. The chronic conditions described within these studies can be broadly divided into four main categories:

1. Physical Health (29 studies), of which 13 were Cancer, and the rest included conditions such as Traumatic Brain Injury, Muscular Dystrophy, Juvenile Arthritis, Diabetes, Asthma, Cystic Fibrosis and Epilepsy.
2. Developmental Disabilities (12 studies), which included children on the Autism Spectrum, ADHD, Down Syndrome and global developmental delays.
3. Both Physical Health and Developmental Disabilities (five studies).
4. One study included siblings of children with eating disorders (one study).

All the studies were read thoroughly, and were then broadly categorised into four main themes:

1. Impact on emotional wellbeing
2. Impact on the Family
3. Impact on social lives and stigma
4. The positive impact and resilience.

A number of the studies fit into more than one category (Table 3-Appendix B). These themes will now be explored in more detail.

Impact on emotional wellbeing

There were two types of study examining the impact of CHCs on the emotional wellbeing of siblings: questionnaire studies, which specifically tested for levels of depression, anxiety and quality of life; and qualitative studies, which elicited information on the emotional wellbeing of the siblings.

The majority of the questionnaire research suggests that having a child with a chronic disorder within the family does have a negative effect on the siblings' emotional wellbeing

(Alderfer, Labay, & Kazac, 2003; Barrera & Atenafu, 2008; Gallo & Szychliniski, 2003; Hamama, Ronen, & Feigin, 2000; Hollidge, 2001; Houtzager, Grootenhuis, Hoekstra-Weebers, Caron, & Last, 2003; Houtzager, Grootenhuis, Caron, & Last, 2004; Jones, Welsh, Glassmire, & Tavegia, 2006; Lahteenmaki, Sjoblom, Korhonen, & Salmi, 2004; Orsmond & Seltzer, 2009; Silver & Frohlinger-Graham, 2000), and quality of life (Houtzager et al., 2004; Areemit, Katzman, Pinhas, & Kaufman, 2010; Houtzager et al., 2003; Houtzager et al., 2004). The studies used a range of assessment tools to assess for emotional difficulties, such as Revised Children's Manifest Anxiety Scale, PedsQL, Beck Depression Inventory, Self Perception Profile for Children, State Trait Anxiety Inventory for Children (STAIC) and Children's Depression Inventory. However, there are some clear methodological problems with some of these research studies. When siblings of ill children were compared to a sibling group of healthy children, often the healthy sibling group differed from the ill sibling group in terms of income and education (Gallo et al., 2003; Jones et al., 2006; Lahteenmaki et al., 2004). This difference could contribute to the findings of increased anxiety and reduced global self-worth in the sibling (Gallo et al., 2003; Lahteenmaki et al., 2004). One study (Jones et al., 2006) did control for these differences in their analysis, and still found siblings of children with ADHD had higher levels of trait anger than controls.

Although a number of the studies stated that siblings' emotional wellbeing or quality of life was affected, the scores were either still within the normal range for that measure or were not compared to norms (Areemit et al., 2010; Barrera et al., 2008; Hamama et al., 2000; Hollidge, 2001).

However, other questionnaire papers state that siblings' emotional wellbeing (Cuskelly & Gunn, 2006; Houtzager et al., 2004; Houtzager et al., 2004; Macks & Reeve, 2007; Read, Kinali, Muntoni, & Garralda, 2010; Swift et al., 2003; Verte, Roeyers, & Buysse, 2003; Wood, Sherman, Hamiwka, Blackman, & Wirrel, 2008) or their Quality of Life (Barrera et al., 2008)

are comparable to norms. Positive effects on siblings' emotional wellbeing were also found, for example their self worth, self concept, honesty, trust and behaviour (Macks et al., 2007; Verte et al., 2003). All of these studies have comparison control groups, or comparative normative data, and some have made efforts to use normative data, which is matched to the siblings, such as age, gender, ethnic minority and SES (Cuskelly et al., 2006; Macks et al., 2007; Swift et al., 2003; Verte et al., 2003).

Understanding whether there is an impact on siblings is difficult to interpret due to differences in methodology, statistical analyses and the different measures used. One study stated that mean scores on the anxiety, quality of life and behavioural and emotional problems were similar to norms, two years after diagnosis, but significant differences were found in the percentage of adolescents reporting impaired emotional problems, compared to norms (Houtzager et al., 2004)¹². Some studies also reported both positive and negative effects for different outcomes, and some reported changes in the same outcome over time, for example the quality of life would improve over time (Houtzager et al., 2004), or that although they found some significant differences in areas of emotional wellbeing or quality of life, they did not find any significant difference in anxiety (Alderfer et al., 2003; Houtzager et al., 2004; Jones et al., 2006; Silver et al., 2000) depression (Hollidge, 2001; Jones et al., 2006; Lahteenmaki et al., 2004; Silver et al., 2000) or quality of life (Barrera et al., 2008). All studies had small sample sizes (n=10-100).

Some of this discrepancy could be due to the type of illness that the child is experiencing.

However, this is unlikely as all the studies (Alderfer et al., 2003; Barrera et al., 2008; Gallo et al., 2003; Hamama et al., 2000; Hollidge, 2001; Houtzager et al., 2003; Houtzager et al.,

¹ Houtzager described the rationale for studying this as well as mean scores "Rose proposed an alternative method of analysis in which the group is divided in the number of individuals who score below or above the 25th percentile norm (Rose et al., 1999). If this is conducted for age and sex stratified groups, differences in the distributions between the study group and the norm group are accounted for and groups can be compared (Rose et al., 1999). (Houtzager et al, 2004, p504)

² Houtzager et al (2004) used the Youth Self-Report (YSR) to test for behavioural and emotional problems in teenagers. YSR impaired functioning scores correspond with the 16–17% highest scores in the healthy norm group aged 11–18 (Verhulst et al., 1997).

2004; Jones et al., 2006; Lahteenmaki et al., 2004; Orsmond et al., 2009; Silver et al., 2000), (Cuskelly et al., 2006; Houtzager et al., 2004; Macks et al., 2007; Read et al., 2010; Swift et al., 2003; Verte et al., 2003; Wood et al., 2008) included siblings of children with life threatening physical health problems and developmental disabilities.

Most of the qualitative research carried out in this area did not name a specific theme 'emotional wellbeing'. However, many siblings described a number of topics, which related to their emotional wellbeing. The most common adjectives used to describe the emotional impact on the siblings were 'loss', 'sacrifice', 'jealousy', 'loneliness', 'worry' and 'anger' (Areemit et al., 2010; Batte, Watson, & Amess, 2006; Fleitas, 2000; Hames, 2008; Hames & Appleton, 2009; Hollidge, 2001; Hutson & Alter, 2007; Kendall, 1999; Loos & Kelly, 2006; Martinson, Gilliss, Colaizzo, Freeman, & Bossert, 1990; Read, Kinali, Muntoni, Weaver, & Garralda, 2010; Sargent et al., 1995; Wilkins & Woodgate, 2007). The themes, which are described more frequently, are feelings of 'loss' and 'jealousy'. The qualitative studies were varied in terms of sampling, recruitment and analysis methods; however, they all highlighted the impact on siblings' emotional wellbeing.

Summary: It seems that although questionnaire-based studies do not always detect differences in emotional well being between siblings and controls, evidence that their emotional wellbeing is still affected is found when their well-being is explored using interview or focus groups methods.

Impact on family

One of the key themes which emerged is the impact that the chronic conditions have on the family. A large proportion of these studies were qualitative. This section can be divided into two areas: positive impact on the family and negative impact on the family.

Most studies, found a negative impact on the family particularly the disruption on family life and routines (Barr & McLeod, 2010; Batte et al., 2006; Giallo & Gavidia-Payne, 2006;

Hutson et al., 2007; Kendall, 1999; Loos et al., 2006; Martinson et al., 1990; Petalas, Hastings, Nash, Dowey, & Reilly, 2009; Read et al., 2010; Read et al., 2010; Sargent et al., 1995; Sloper, 2000; Williams, 1997). Linked to this, a number of the siblings also spoke about a feeling of responsibility to the family and the child that they now carried (Areemit et al., 2010; Barr et al., 2010; Fleitas, 2000; Hames, 2008; Hollidge, 2001; Loos et al., 2006; Read et al., 2010). The siblings also spoke about feeling rejected by their parents (Read et al., 2010), and the lack of attention they now had, which caused jealous feelings towards their sibling (Barr et al., 2010; Batte et al., 2006; Fleitas, 2000; Hames et al., 2009; Martinson et al., 1990). Lack of communication within the family was cited as a main reason for concern and worry for the siblings (Giallo et al., 2006).

Conversely, some research found that siblings described an improvement in family or sibling cohesion, and enhanced communication due to their ill sibling (Areemit et al., 2010; Hames et al., 2009; Hollidge, 2001; Loos et al., 2006; Read et al., 2010; Sargent et al., 1995), and that this was a protective factor for improved coping with living with an ill child (Giallo et al., 2006). The importance of family and social support as a coping strategies used by siblings has been highlighted (Cox, Marshall, Mandelco, & Olsen, 2003). The majority of these siblings sought this support from family members.

Two of the quantitative studies directly assessed sibling relationships using the Sibling Relationship Questionnaire and the Sibling Relationship Questionnaire Revised. They found similarities in sibling relationships in families who had suffered traumatic brain injury or orthopaedic injury (Swift et al., 2003) and between those siblings in families with juvenile chronic arthritis or healthy families (Weiss, Schiaffino, & Ilowite, 2001).

Summary: Siblings describe a mixed impact on their family life. The detrimental impact includes disruptions to family life and routines, increased responsibility and feelings of rejection. Positive impact includes increased family and sibling cohesion. Communication was cited as a protective and risk factor for impact of chronic conditions.

Social impact/stigma

Peer relationships have been found to be important for siblings of children with chronic conditions (Read et al., 2010; Sloper, 2000). However many of the studies in this review found that having a child with a chronic condition in the family has a negative impact on the siblings' social lives (Areemit et al., 2010; Barr et al., 2010; Fleitas, 2000; Giallo et al., 2006; Hames, 2008; Houtzager, Grootenhuis, Hoekstra-Weebers, & Last, 2005; Hutson et al., 2007; Kendall, 1999; Labay & Walco, 2004; Martinson et al., 1990; Patterson, Millar, & Visser, 2010; Petalas et al., 2009; Read et al., 2010). Siblings of children with disabilities have been found to have more peer problems, and lower pro-social behaviour than UK normative data (Giallo et al., 2006). One of the ways that this impact was felt was through not being able to attend recreational activities with peers (Areemit et al., 2010). The siblings reported a lack of understanding from their peers (Barr et al., 2010; Hames, 2008; Hutson et al., 2007; Petalas et al., 2009), or even teasing or bullying from them about their ill sibling (Barr et al., 2010; Kendall, 1999; Petalas et al., 2009). This meant the well-sibling worried about telling their peers or not about their ill-sibling (Areemit et al., 2010; Hames, 2008). Siblings spoke about feeling different to their peers (Batte et al., 2006), and feeling lonely or embarrassed (Fleitas, 2000; Hutson et al., 2007; Martinson et al., 1990; Petalas et al., 2009).

Some of the possible factors which may alter the negative impact on the sibling are age of the sibling, the birth order of the child with the chronic condition of family communication. Older siblings reported less loneliness than younger siblings (Hamama et al., 2000). This could be due to older siblings feeling less need from parents, or due to older siblings having more peer support than younger siblings. However, if the child with the condition was later in the birth order, the sibling experienced more social problems (Labay et al., 2004). Both of these studies explored the impact of cancer on siblings and only included siblings of children having active treatment. Giallo and Gavidia-Payne (2006) and Gallo and Szychlinksi (2003) noted that there were strong links between pro-social behaviour and high self-perception,

with family communication and problem solving: Better quality family communication meant higher assessment of their pro-social behaviour by parents on the Strengths and Difficulties Questionnaire and higher self-perception on the Self-Perception Questionnaire for Children.

Summary: This theme highlights the social difficulties that can arise from having a child with a chronic health condition as a sibling. The siblings reported a range of feelings, from loneliness to embarrassment, and also experiences of feeling left out and bullied by some peers. Some siblings highlighted the stigma associated with strangers' reactions. However, some studies again reported more positive impacts.

The positive impact and resilience.

Although most studies found that the impact on siblings of having a child with a condition in the family was negative, a number of studies found positive effects, or resilience. The majority of the siblings (65%) used positive coping strategies, which suggests they are independent and proactive in solving problems (Cox et al., 2003). Siblings appeared to be more independent (Fleitas, 2000; Hames, 2008) and more caring and empathic (Areemit et al., 2010; Fleitas, 2000; Hames, 2008; Hollidge, 2001; Sargent et al., 1995; Sloper, 2000). In one qualitative study, a whole theme was extracted, named 'resilience', which included feelings of lessons being learned, taking time for the small things in life, being more altruistic and caring and feeling more grown-up and independent (Fleitas, 2000).

Two studies suggested the impact on the sibling was neither positive nor negative but neutral: with feelings of "acceptance" or feelings of "life goes on" (Petalas et al., 2009; Wilkins et al., 2007).

Discussion

This systematic literature review has explored the impact on siblings of children with any chronic condition, including physical health, developmental disabilities and mental health.

This literature search resulted in 47 papers, which were read and placed into four main

themes: Emotional Wellbeing, Impact on Family, Social Impact/Stigma, and The Positive Impact/Resilience. All but one of these themes also emerged during the CFS/ME paper (Jackson, 1999). Worries about “apparent parental dilution of care or concern”, the loss of sibling relationships and the restriction on family activities could be easily placed within the ‘Impact on Family’ theme. The deterioration in peer relationships could be placed within the ‘Social Impact/Stigma’ theme. The language used within the Jackson (1999) paper, such as ‘loss’, and ‘worry’ could be placed within the ‘Emotional Wellbeing’ theme. This review also found a theme within the qualitative literature on ‘The Positive Impact/Resilience’, which did not emerge in the Jackson (1999) paper. However, the Jackson (1999) paper does have an extra theme relating to the uncertain or contradictory advice given to the family. This is a theme which is not found within these other chronic conditions. It is unclear whether this is due to the Jackson (1999) paper, or due to the intrinsic differences that CFS/ME has compared to the other illnesses described.

Theoretical implications

Prince-Embury (2008) suggests that there are three underlying factors of personal resiliency; Sense of Mastery, Sense of Relatedness and Emotional Reactivity. The interaction between the three of these factors suggest how resilient a young person is. These three areas will be explored, to see whether the findings from this literature review fit with them.

Sense of Mastery, builds on previous definitions of self-efficacy. It has been suggested that self-efficacy allows a child to interact with and have positive cause-and-affect relationships (White, 1959). Prince-Embury (2008) suggests that Sense of Mastery includes optimism about life and their own competence and ability to adapt and problem solve to new situations. One of the themes in this literature review, impact on family, suggested that siblings feel that there is often a negative impact on their family, linked to changes in routines, unpredictability and a heightened sense of responsibility. In addition one of the

studies found that 65% of the siblings used adaptive coping strategies (Cox et al, 2003). This could be due to the ways that the siblings appraise the situation they are in, as well as what level their perceived control is (Abramson, Seligman, & Teasdale, 1978; Lazarus, 19; Salovey, 1991; Rudolph, Kurlakowsky & Conley, 2001). If siblings have low perceptions of control over the situation (Abramson, Seligman, & Teasdale, 1978), this can then lead to stress, anxiety and depression (Rudolph, Kurlakowsky & Conley, 2001). Siblings also spoke about feelings of loss and jealousy, which could also be explained by the lack of 'Sense of Mastery'.

Sense of Relatedness suggests that social support and positive relationships can act as mediators of resilience (Werner and Smith, 1982). Prince-Embury (2008) proposes that Sense of Relatedness includes comfort and trust in others, and ability of tolerate differences with others. Within this literature review a theme emerged of 'Social impact/stigma'. This would suggest that many of the siblings in these studies find that their Sense of Relatedness is low, which would increase their vulnerability (Prince-Embury, 2008). This could also fit with Social comparison theory, which would suggest that the siblings are comparing themselves negatively to their peers (upward social comparisons), and then feeling anxious or lonely because of this (Festinger, 1954). This could be increased when the illness is also seen as stigmatizing, and so social support may be diminished. Also linked to this is Rolland & Walsh (2006) family resilience framework, which recognises that crises and persistent challenges, such as an ill child, impacts upon the whole family, and that key family processes can mediate the adaptation of all members and their relationships. These factors include positive communication, connectedness, flexibility and positive outlook.

Finally, Emotional Reactivity, builds on the idea that a child's emotional reactivity and their ability to modulate and regulate this reactivity is related to their resilience (Cicchetti, Ganiban, & Barnett, 1991; Cicchetti & Tucker, 1994; Rothbart & Bates, 1998; Thompson,

1990). Prince-Embury (2008) suggests that Emotional Reactivity consists of sensitivity, threshold and intensity of the reaction, length of time it takes to recover from emotional upset and impairment whilst upset. One of the themes which emerged in this literature review was emotional impact, which suggested that there was a negative impact on the emotions of some siblings. It seemed that some siblings felt strong negative emotions, such as 'loss' 'anger' and 'jealousy', whereas others were more able to accept the situation, and recover from the emotional impact quicker. It has been noted previously that some children seem to be more resilient to negative life events, such as divorce and parental death, than others (Greene, Anderson, Hetherington, Forgatch, & DeGarmo, 2012; Heinzer, 1995).

In many ways the Prince-Embury (2008) theory of resilience in children fits very well with the findings from this literature review. Those children with higher senses of mastery and relatedness, and lower levels of emotional reactivity, tend to be more resilient and have fewer psychological difficulties (Prince-Embury, 2008). However, although many of the studies suggest that factors such as positive family communication, social support or adaptive coping strategies are helpful for siblings, there are been no research using the Prince-Embury (2008) resilience scales to explore the impact on siblings.

Findings from the literature review and implications to CFS/ME

The literature on the impact of chronic conditions on siblings is varied; with the children suffering from a range of conditions and the studies using a range of methodologies, such as different sampling methods, analyses, questionnaires etc. Although all of the results could be easily placed within four main themes, different research found varied, sometimes conflicting results.

The 'Emotional Wellbeing' theme seemed to have the largest amount of discrepancy within it. Although the majority of the quantitative data seemed to suggest a negative impact on

siblings, some suggested that there was no impact on the emotional wellbeing of the siblings, or even that there was a positive impact.

Overall the interview literature does suggest that having a child with a CHC within the family does have a negative emotional impact on the sibling. This is consistent with a recent meta-analysis (Vermaes, van Susante, & van Bakel, 2012), who found that siblings experienced a negative effect, and also had fewer positive self-attributes than normative comparisons.

This review begins to give us an insight into the types of impact CFS/ME may have on siblings. It seems very likely that siblings of children with CFS/ME will be experiencing difficult emotions, such as loss, jealousy and worry. This may be more so than with other chronic conditions, because of the added worry of conflicting medical advice (Jackson, 1999). CFS/ME in children often takes time to be diagnosed, which could add to the sibling's sense of low control, and in turn lead to increased feelings of anger, worry and loss (Abramson, Seligman, & Teasdale, 1978; Sankey et al., 2006), or their low Sense of Mastery (Prince-Embury, 2008).

The literature review also suggests that there will some impact on the family. However, this impact could be either negative or positive, and seems to depend on the mode of communication used within the family. The impact on the family also seems to be linked with feelings of jealousy about the child with the condition, and feelings of rejection from the parents. Again this could be linked to the Sense of Mastery or Sense of Relatedness themes suggested by Prince-Embury (2008).

This literature review suggests CFS/ME may impact on siblings' social life and stigma. Given how severely affected many children with CFS/ME are, it seems likely that siblings will be less able to take part in an active social life. CFS/ME is a stigmatising condition (many people do not know what it is, misunderstand it or underestimate it (Jason, Taylor, Plioplys,

Stepanek, & Shlaes, 2002)). It therefore seems likely that siblings may feel that they experience stigma. What happens to the sibling may depend on parental reaction because of link between appraisal of life events, social support and adaptive behaviour (Jackson & Warren, 2001). They suggest that if a life event is appraised as negative, social support can act as a mediator to adaptive behaviour, and that less social support is needed for positively appraised events (Jackson & Warren, 2001).

There are significant differences between some of these conditions reviewed and CFS/ME. Some studies investigated the siblings of children with life threatening diseases, such as cancer or Cystic Fibrosis. CFS/ME is not life threatening, and so some of the issues which arose for siblings of children with life threatening illnesses, may not arise for siblings for children with CFS/ME. Many of the conditions included in the literature review are well-known and elicit sympathy and understanding, e.g. cancer. However, a number of the included studies involved siblings of children with developmental disabilities, which could also be seen as stigmatizing. Studies investigating the siblings of those with ADHD, all found negative impacts on the sibling, such as increased trait anger, feelings of victimisation, sorrow and loss, and increased conflict in the sibling relationship and increased internalizing and externalizing problems (Jones et al., 2006; Mikami et al., 2008). However, Kendall (1999) and Mikami & Pfiffner (2008) conclude that this could be attributed to the extreme behaviour and disruption cause by the child with ADHD, which is not likely to occur with children with CFS/ME.

In conclusion, there is variation in how siblings experience negative life events, suggesting there are both risk and resilience factors, which either ameliorate or increase the likelihood that having an ill child in the family will have a negative impact on siblings. Research needs to be conducted to explore the impact on siblings of children with CFS/ME.

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Appendix A: Table 2: Description of Included Research Studies

Author (year)	Design	Sample	Aim	Impact Measures	Findings
Alderfer, Labay & Kazac (2003)	Questionnaire Study	78 Adolescent Siblings of Cancer Survivors	Post-Traumatic Stress	RCMAS; IES-R; PTSD-RI; ALTTIQ	49.3% had mild PTSD, 32% had moderate to severe PTSD. Anxiety was in normal range.
Areemit et al (2010)	Questionnaire Study and Qualitative using focus groups.	20 siblings aged 10-18 yrs of adolescents with eating disorders.	Quality of life and general impact.	PedsQL-4.0; Eating Attitude Test-26	80% said QoL was affected by siblings' condition. Key themes were: a desire to understand the ED, acute awareness of ED behaviours and thoughts, challenges in understanding non eating-related obsessive behaviours, increase in family conflict and arguments, compassion and concern for the AED, feelings of loss and sacrifice, overwhelming sense of responsibility for the AED, and a sense of pervasiveness of the ED in all aspects of their lives.
Barr & McLoed (2010)	Qualitative-Sibling Support website analysed	676 contributions to the website. Siblings of children with physical and/or developmental disabilities	General Impact	N/A	Three key themes; Strangers, Peers and Family. Strangers stare and have negative attitudes towards my sibling with a disability; peers don't understand what it's like to be me, use certain words that upset me, say nasty things and tease me about my brother/sister; although my family loves me, they don't have a lot of time for me, our plans are often disrupted, and they give me a lot of responsibility.
Barrera & Atenafu (2008)	Questionnaire Study-comparative of	45 children (3-16 yrs) with Pediatric	Cognitive, educational and	WISC-III; CHQ-PF50; FACES; BDI	Child and sibling IQ scores did not differ significantly.

	siblings and children	Hematopoietic, and their 33 siblings (3-20yrs).	psychosocial impact.		
Batte, Watson & Amess (2006)	Qualitative-semi-structured interviews. Questionnaire.	15 Siblings (8-12 yrs old) of children with chronic renal failure.	General Impact and Anxiety	SCAS	Siblings had significantly more internalizing problems than the children. No Clinical Anxiety in the siblings. Qualitative data indicated that siblings had a variety of concerns concerning their own health and that of their siblings and worried about the effects on family routine and separation from parents. The siblings felt more protective towards their chronically ill sibling and felt that they themselves needed to be more grown up.
Cox et al (2003)	Qualitative-Content Analysis	46 Siblings (6-18 yrs old) of children with developmental disabilities.	Coping responses	Sentence-completion instrument consists of 18 open-ended statements allowing siblings to respond to real or hypothetical stressful situations.	Four types of coping strategies were found: Proactive, interactive, internally reactive and non-active.
Cuskelly & Gunn (2006)	Comparative questionnaire Study	53 siblings (7-14yrs) of children with Down's Syndrome, and matched siblings of typically developing children.	Adjustment, behaviour, self-perception.	CBCL; Self-Perception Profile for Children; Sibling Inventory of Behaviour.	There were no significant differences between the groups on adjustment measures. These included parent perceptions of externalizing and internalizing behaviours, parent perceptions of sibling competence, and sibling perceptions of their own competence and self-worth.
Fay & Barker-Collo (2003)	Comparative Questionnaire Study	10 siblings of children with Traumatic Brain Injury and 10 Siblings of children with Orthopaedic Injury.	Behaviours, emotional wellbeing and sibling relationship.	CBCL; SIQ; SRQ	Well siblings of children with TBI exhibited more internalising behaviours than siblings of children with orthopaedic injury. Well siblings' ratings of the impact of the injury on the family

					a greater in TBI families than Ortho families. and sibling relationship did not differ significantly across groups.
Fleitas (2000)	Qualitative-comments on sibling websites and to Children's nurses.	No attempt to qualify or sort the comments by age were made.	Stressors for siblings.	N/A	Two themes: Stress and Resilience. Stress = (responsibility, loneliness/resentment, fear, jealousy, guilt, sadness, embarrassment, and confusion. Resilience = lessons learned, independence, altruism Results indicated that siblings of children with diabetes were at risk for self-perception problems in the areas of scholastic competence and global self-worth. Male sibling pairs in the diabetes group had lower self-perception scores than male pairs in the asthma group; whereas, female sibling pairs in the diabetes group had lower family functioning scores than female pairs in the asthma or healthy groups. For the siblings in the diabetes group, physical appearance, athletic competence, behavioural conduct, scholastic competence, and global self-worth were significantly associated with family functioning. It was found that parent and family factors were stronger predictors of sibling adjustment
Gallo & Szycliński (2003)	Comparative Questionnaire Study	135 siblings (6-12yrs) -44 of children with Type 1 diabetes, 42 of children with Asthma and 41 of healthy children	Self-perception and family functioning	Family APGAR & Self-Perception Profile for Children.	
Giallo & Gavidia-Payne (2006)	Questionnaire study	Forty-nine siblings (7-16yrs) and parents of children with	Sibling adjustment	The Sibling Daily Hassles and Uplifts Scale; The Self-	

		intellectual, sensory, physical, or developmental disabilities residing at home.		Report Coping Scale; The Strengths and Difficulties Questionnaire – Parent Version; The Perceived Stress Scale; The Parent Behaviour Questionnaire; The Family Hardiness Index; The Family Problem Solving Communication Index; The Family Time and Routines Index;	difficulties than siblings' own experiences of stress and coping. Specifically, socio-economic status, past attendance at a sibling support group, parent stress, family time and routines, family problem-solving and communication, and family hardiness-predicted sibling adjustment difficulties. Finally, siblings' perceived intensity of daily uplifts significantly predicted sibling pro-social behaviour.
Hamama, Ronen & Feigin (2000)	Questionnaire Study	Sixty-two healthy siblings aged 9 to 18 of children with cancer.	Emotional response, anxiety, loneliness and self-control	State-Trait Anxiety Inventory for Children; Loneliness Questionnaire; Children's Self-Control Scale	The findings showed that the stress elicited emotional responses. Anxiety was related to the child's age and duration of the sibling's cancer, and loneliness was related to the child's sex and rank in the family. The outcomes also demonstrated a link between self-control as a coping skill and anxiety and loneliness as emotional distress responses. Healthy siblings' higher self-control rates were associated with their lower anxiety and loneliness reports.
Hames (2008)	Qualitative-semi structured interviews	Interviewed parents and then siblings for 12 years, from when they were pre-verbal, until they were aged 12–14 years, of	Understanding the condition of their sibling.	N/A	Themes included social awareness and describing the impact of the disability.

Hames & Appleton (2009)	Qualitative-semi-structured interviews and a personal account.	children who had developmental disabilities. Fourteen siblings from the 25 families, aged 8–25 years, provided a personal account of what it was like living with a brother or sister with epilepsy.	General Impact	N/A	Themes included the negative impact of having a sibling with epilepsy, e.g. behaviour, feeling lonely, anxious or 'different', impact on family, responsibility. Also positive themes, e.g. how much they loved them and the strength of family.
Havermans, Wuytack, Deboel, Tijtgat, Malfroot, De Boeck & Proesmans (2010)	Questionnaire study	39 siblings (10-18 years) of children with Cystic Fibrosis	Quality of life and impact of illness	CHQ, SPQ (sibling perception questionnaire,	Siblings of children with CF reported a better QoL on several domains than siblings of healthy children. No gender or sibling birth rank effects.
Hollidge (2001)	Mixed methods; qualitative semi-structured interviews and questionnaires	A sample of 28 well siblings between the ages of 8 and 12 of children with diabetes were studied	Psychological Adjustment	Achenbach Child Behaviour Checklist; Piers-Harris Self-concept scale; Reynolds Child Depression Scale; Revised Children's Manifest Anxiety Scale	The findings conclude that well siblings have difficulties negotiating emotions, communications, and activities with their diabetic siblings. The report interprets sibling issues from a psychodynamic orientation, suggesting that many of the healthy psychological functions performed by the sibling relationship are interrupted by the introduction of a chronic illness.
Houtzager, Grootenhuis, Hoeskstra-Weebers, Caron & Last (2003)	Prospective Questionnaire Study	At Month 1, 83 siblings aged 7–18 years and at Month 6, 66 siblings aged 7–18 years, or children with cancer.	Anxiety, social–emotional problems and quality of life (QoL)	Youth Self-Report; Dutch Children's AZL/TNO Quality of Life Questionnaire (DucatQoL); State-Trait Anxiety Inventory for	At 1 month, siblings reported a lower QoL and adolescent girls reported more emotional problems compared with peers. At 6 months, adolescent QoL remained relatively impaired. Over time, adolescent brothers

				Children.	reported fewer emotional and total problems and young girls reported decreased anxiety. No significant amelioration in QoL was found over time. The older the siblings were, the lower their observed QoL at both measurements and in several domains. The occurrence of life events predicted sisters' QoL at 1 month. Changes in sibling functioning were predicted by none of the investigated risk factors. Thus, QoL is impaired shortly after diagnosis. Adolescent siblings risk persisting problems in daily functioning
Houtzager et al (2004a)	Comparative Questionnaire Study	83 siblings (aged 7-19 yrs) of children with cancer.	Anxiety, quality of life, behavioural-emotional problems, and emotional reactions to illness	The State-Trait Anxiety Inventory for Children; The Dutch Children's quality of life questionnaire; Dutch Child behaviour Checklist; Youth Self Report; Situation Specific Emotional Reactions Questionnaire for Siblings; Coping Strategies Scale for Siblings; Dutch version of Family Acceptability and Cohesion Evaluation Scales	Anxiety was comparable at 1 month between siblings and comparative norms, and lower than the norms at 6 months. Quality of life remained lower than norms at all time points until 2 years post diagnosis. Sibling self report behavioural-emotional problems were higher than norms at 1 month (p>.05). Internalizing problems were particularly high but diminished to normal level at F/U.
Houtzager, Grootenhuys, Caron	Prospective and retrospective	Prospective study=2 yrs after diagnosis, 57	Psycho-social problems	State-Trait anxiety Inventory for	No difference in anxiety, internalising or externalising

& Last (2004b)	questionnaire study	siblings aged 9-19 yrs old. Retrospective study=2 yrs after diagnosis, 46 siblings aged 8-18 yrs old.		Children; Youth Self-Report, The Dutch Children's Quality of Life Questionnaire; The TNO AZL Children's Quality of life Questionnaire.	problems between siblings and peers. Siblings aged 7-11 yrs had significantly lower QoL than peers. 42% reported impaired emotional QoL, 34% reported impaired Social QoL.
Houtzager, Grootenhuis, Hoekstra-Weebers & Last (2005)	Questionnaire Study	83 siblings (aged 7-18 yrs) of children with cancer	Quality of life 4-8 weeks after diagnosis.	TNO-AZL Children's Quality of Life Questionnaire; cognitive Coping Strategies Scale for Siblings.	Siblings aged 7-11 yrs old reported lower QoL than norms in motor skills, cognitive skills and positive and negative emotions. Adolescents aged 12-18yrs reported lower QoL than peers in cognitive skills, and positive and negative emotions. Older siblings had more negative emotions. Girls reported more problems on Social QoL.
Hutson & Alter (2007)	Qualitative-semi-structured interviews	9 siblings (11-21 years old) of children with FA (a form of cancer)	General Impact	N/A	Four main themes emerged: Containment, invisibility, worry and despair.
Jones, Welsh, Glassmire & Tavegia (2006)	Comparative Questionnaire Study	45 siblings (aged 9-13 years) of children with ADHD, and a control group (aged 9-13 years).	Psychological Functioning	Children's depression inventory; pediatric Anger Scale	Siblings of children with ADHD had significantly higher levels of trait anger than children in control group.
Kendall (1999)	Qualitative-semi-structured interviews and diaries	13 siblings (mean age 11 years old) of children with ADHD.	Experiences of living with a child with ADHD	N/A	Three main themes emerged: Disruption, Effects of Disruption, e.g. victimizations, caretaking and sorrow and loss, and Managing Strategies, e.g. retaliation, accommodation and avoidance.
Labay & Walco (2004)	Comparative Questionnaire Study	29 Siblings (7-16 years) and 14 children (8-15 years) diagnosed with	Empathy, illness concepts, sibling relationship and psychological	Index of Empathy; Child Behaviour Checklist, Sibling Relationships	Siblings scored significantly below the mean on three social competencies subscales: activities, social relationships

Lahteenmaki, Sjoblom, Korhonen & Salmi (2004)	One year follow up questionnaire study	Cancer 33 siblings of children with cancer, and 357 healthy controls	adjustment. Behavioural and health related problems.	Questionnaire. State-Trait Anxiety Inventory for Children; Children's Depression Inventory.	and school performance. 3 months after diagnosis, state anxiety was greater in siblings than controls. This diminished at F/U. Siblings did not differ significantly on depression from controls.
Lardieri, Blacher & Swanson (2000)	Mixed methods; questionnaires and semi-structured interviews	71 siblings of children with LD and without LD	Psychological Adjustments and perceived impact of their brother or sister.	Child Behaviour Check List; SRQ; SIQ; Youth Self Report;	Siblings of children with LD report higher externalizing behaviour scores than siblings of children without LD. Siblings of children with LD scored higher on the Conflict subscale of SRQ.
Laufersweiler-Plass, Rudnik-Schoneborn, Zerres, Backes, Lehmkuhl & von Gontard (2003)	Comparative Questionnaire study between children with Spinal muscular atrophy (SMA), their siblings and healthy controls	96 adolescents with SMA (6-18 years old), 45 siblings (6-18 years), and 59 healthy controls (6-18 years).	Behavioural adjustment	Child Behaviour check List	Siblings with clinical and/or borderline behaviour scores was 2-3 times higher than the normative population on all three scales. Significantly more siblings had internalizing behaviour symptoms than control, and more siblings had externalizing symptoms than their brothers or sisters with SMA.
Loos & Kelly (2006)	Qualitative-in depth interviews	16 well siblings (6-14 years) of children with diabetes.	General impact and knowledge.	N/A	6 main themes emerged: Reactions to Diagnosis, e.g. sadness, fear of needles, adjustment; Education on Diabetes, e.g. education attendance, knowledge; Care Involvement, e.g. physical, emotional and increased responsibility; Sibling Relationships, e.g. closer or strained; Impact of Diabetes, e.g. anger, feeling left out, family routines; and Fears, e.g.

Macks & Reeve (2007)	Comparative Questionnaire study	51 siblings (7-17 years) of children with autism. And 35 siblings of children with no condition.	Psychosocial and emotional adjustment	Children's Depression Inventory-Short form; Piers-Harris Children's self-concept scale.	catching diabetes, genetic link or death. No significant differences between sibling groups on the Depression inventory. Siblings of children with autism scored significantly higher than comparison group on self-concept total score and behaviour, intellectual and school status. As demographic risk factors increase, so does the unfavourable impact on the siblings.
Martinson, Gilliss, Colaizzo, Freeman & Bossert (1990)	Repeated Qualitative interviews.	At diagnosis, 14 siblings (aged 6-12 years). Five years later, 9 siblings with a living child and 7 siblings with a deceased child.	Emotional reactions	N/A	Some of the themes which emerged were: Hospitalization having impact on the family, feelings of fear an loneliness, lack of peer understanding, feeling left our by parents, feeling jealousy and sadness.
Mikami & Pfiffner (2008)	Comparative questionnaire study	77 siblings (4-18 years) of children with ADHD. 14 siblings of control children (4-18 years)	Sibling relationship and internalizing and externalizing problems	SRQ; Child Depression inventory	Increased conflict in sibling relationships with a child with ADHD than controls. Mothers reported more warmth and closeness than siblings did.
Orsmond & Seltzer (2009)	Questionnaire study	57 siblings (12-18 years) of children with an ASD.	Emotional wellbeing and stress	Centre for Epidemiological Studies-Depression Scale; Revised Children's manifest Anxiety Scale; Life events checklist from NIMH Methods for epidemiology of Child and Adolescent mental Disorders	Over 1/3 (36%) of siblings reported depressive symptoms at or above the cut-off of 16. At the highest cut off of 28, 10% reached this. 8.5% reported clinical relevant anxiety symptoms Sisters reported significantly more depressive symptoms and anxiety than brothers Higher depression in siblings

Patterson, Millar & Visser (2010)	Qualitative semi-structured interviews and focus group	4 siblings (14-17 years) of children with cancer (focus groups). 7 siblings (16-22years) of children with cancer (telephone interview)	Unmet needs for siblings of children with cancer.	N/A	whose child with ASD had behaviour problems. Siblings who reported more life events in previous year, had higher depression ratings. Siblings identified 10 areas of need: peer support (friends), peer support (similar experience), information, sibling relationship, expressing and coping with feelings, respite and recreation, acknowledgement and attention for self, involvement in cancer experience, instrumental support, access to support services.
Petalas, Hastings, Nash, Dowey & Reilly (2009)	Qualitative-semi-structured interviews	8 siblings (9-12 years) with a brother with ASD.	Perceptions and experiences	N/A	Five main themes emerged: Siblings perceptions of the impact of their brother's condition on their lives; Siblings' perceptions of the attitudes of others; siblings' tolerance and acceptance towards their brothers; positive attitudes and experiences; and sibling's views on support for themselves and their brothers.
Petalas, Hastings, Nash, Hall, Joannidi & Dowey (2011)	Questionnaire study	166 Typically Developing (5-17 years) siblings (84 male) of children with an ASD	Psychological adjustment and sibling relationship	SDQ, AQ (Autism Spectrum Quotient), SRQ, FMSS (Five minute speech sample), HADS	Total difficulties on the SDQ of the child with an ASD was a significant independent positive predictor of sibling SDQ total difficulties and conflict and rivalry in the sibling relationship, and also a negative predictor of warmth in the sibling relationship. Critical family climate predicted conflict in the

					sibling relationship. Specifically, siblings older than the child with an ASD and whose brother or sister had been longer diagnosed had less parent-reported conflict in the sibling relationship.
Read, Kinali, Muntoni & Garralda (2010a)	Questionnaire Study	46 siblings (11-18 years) of children with Duchenne Muscular Dystrophy.	General impact and adaption	SDQ; GHQ; HADS; SF-36; Family Assessment Device; Life events Questionnaire	Comparable means on the SDQ between siblings and norms. % of siblings reaching high risk threshold on emotional subscale of SDQ was twice than of norms. Siblings were either comparable or more favourable on GHQ and HADS. Siblings reported the impact on family life, e.g. daily activities and social life. More likely to affect emotional SDQ if closer age gap and less communication between siblings. 6 key frameworks: Knowledge, Caring responsibilities, Activities, Impact, Coping and Support.
Read, Kinali, Muntoni, Weaver & Garralda (2010b)	Qualitative-semi-structured interviews	35 siblings (11-18 years) of children with Duchenne Muscular Dystrophy.	Descriptive accounts of general impact and coping.	N/A	Positive themes included becoming more compassionate and caring (16%), family members become closer (16%), Expanded life experiences (18%). Negative themes included Received less attention (14%), Increased family separations and disruptions (13%), increased negative feeling states (12%)
Sargent, Shaler, Roghmann, Mulhern, Barbarian, Carpenter, Copeland, Dolgin & Zelter (1995)	semi-structured interviews	254 siblings (5-18 years) from 198 families with a child with cancer.	Effects of cancer on self and family	N/A	

Silver & Frohlinger-Graham (2000)	Questionnaire Study	34 siblings (13-19 years) of children with chronic health conditions, including asthma, arthritis, diabetes, cancer, epilepsy, sickle cell disease, thyroid and cardiac disorder.	Psychological Impact	Brief Symptom Inventory; Global Severity Index	23% said there was nothing positive. Ill sibs were significantly higher on anxiety and interpersonal sensitivity subscale of the BSI. Ill-sibs were significantly higher on the global severity index than those well-sibs. Significant combined effect of illness status, birth order and sibling gender for three BSI scores-anxiety, hostility and global severity.
Sloper (2000)	Qualitative Interviews	94 siblings (8-16 years) of children with cancer interviewed at 6 and 18 months after diagnosis. Semi structured interview were conducted.	Psychological Impact	N/A	six months after diagnosis, siblings reported a number of problems: loss of attention and status; loss of their own and their families usual activities and routines; loss of certainty and security; and loss of companionship of the ill child. For many, problems had resolved 18 months after diagnosis, but problems remained or had arisen for some. These were not confined to those whose brothers or sisters had relapsed or continued to have treatment. Supportive relationships were reported to be important resources, providing an opportunity for siblings to express their own feelings and needs, and information about the illness and treatment helped them to understand why family life was disrupted. Positive effects were also apparent: gains

Swift, Taylor, Kaugars, Drotar, Yeates, Wade & Stancin (2003)	Questionnaire study.	103 siblings of 34 children with severe Traumatic Brain Injury (TBI), 30 with moderate Traumatic Brain Injury and 39 with Orthopedic injuries without brain injury (ORTHO).	Long term outcomes of children with TBI, 4 years post-injury	SRQ, Child Behaviour-sibling self-report.	in maturity, understanding and compassion, and closer family relationships No differences between sibling groups of sibling relationships. No differences between sibling groups in internalizing, externalizing or total behaviour problems. More children with TBI behaviour problems, linked to more sibling behaviour problems.
Verte, Roeyers & Buysse (2003)	Comparative Questionnaire study	29 siblings (6-16 years) of children with High Functioning Autism (HFA), 29 siblings without a disorder	Psychological Adjustment	Child behaviour Checklist; Matson Evaluation of Social Skills with Youngsters; Self-description Questionnaire 1 & 11.	Siblings of HFA scored themselves higher on the scale of social skilled behaviour than controls. No differences in SDQ were found. Sisters of HFA have more positive self-concept than control (12-16 years). Siblings scored higher on subscales of honesty-trust and verbal self-concept.
Weiss, Schiaffino & Ilowite (2001)	Comparative Questionnaire study	20 children with Juvenile Chronic Arthritis (7-18 years) and their siblings (7-21 years) compared to 20 comparison sibling pairs.	Sibling relationships	SRQ;	Siblings of JCA did not differ from other's siblings in ratings of sibling relationship.
Wilkins & Woodgate (2007)	Qualitative semi-structured interviews	8 siblings of children Bone Marrow Transplant recipients.	Lived experience	N/A	Interruptions to family life-revolved around hospital visits and ill child. Family trips, birthdays and routines all interrupted. Roles and responsibilities within the family changed. Families better and more cohesive after BMT. Feeling that 'life goes on'.

Williams, Williams, Graff, Hanson et al (2002)	Questionnaire study	252 well children (mean= 11 years) and adult dyads.	Psychosocial variables and health.	Sibling Knowledge about Illness; Sibling Perception Questionnaire-revised; Sibling Perception Questionnaire-Attitude; Self-Perception Profile for Children; Social Support Scale for Children; Child behaviour Inventory	Feelings of anger, worry, fear, sadness, hope and pride. Lack of control over life and comparison to other people. Behaviour of the sibling is directly affected by SES and cohesion in family, age of sibling, support felt by sibling and knowledge about the illness.
Wood, Sherman, Hamiwka, Blackman & Wirrell (2008)	Questionnaire Study	37 siblings (6-18 years) of children with intractable epilepsy and siblings with epilepsy and other chronic conditions,	Anxiety, depression and quality of life	RCMAS; CDI; PedsQL.	None of the siblings were depressed. If siblings with chronic illness were excluded, depression was significantly lower than norm. Siblings did not differ from norm in anxiety. Siblings had non-significant lower scored on QoL than norms Siblings QoL, anxiety and Depression scores were highly correlated. Anxiety score was uniquely predictor of sibling QoL.
Wood Rivers & Stoneman (2008)	Questionnaire Study	50 siblings (7-12 years) of children with autism, Aspergers and PDD.	Sibling temperament, sibling relationship.	Sibling Inventory of Behaviour; Satisfaction with the Sibling Relationship Scale	Siblings higher in persistence were rated by themselves and parents as having a less negative relationship with less unkindness and less avoidance/embarrassment.

RCMAS= Revised Children's Manifest Anxiety Scale; IES-R= Impact of Events Scale-Revised; PTSD-RI= Post Traumatic Stress Disorder-Reaction Index; ALTTIQ= Assessment of Life Threat and Treatment Intensity Questionnaire; Peds-QL=Pediatric Quality of life Inventory ; WISC-III=Weschles Intelligence Scale for Children-third edition;

CHQ-PF50=Child Health Questionnaire; FACES-3= The Family Adaptability and Cohesion Evaluation Scale-3; BDI-Beck Depression Inventory; SCAS= Spence's Children's Anxiety Scale; CBCL= Child Behaviour checklist; SIQ=Sibling Impact Questionnaire; SRQ=Sibling Relationship Questionnaire

Appendix B: Table 3: Themes

Theme	Authors	What the authors found
Emotional Wellbeing	Alderfer, Labay & Kazac (2003)	Post Traumatic Stress Symptoms found. No increased anxiety though.
	Areemit et al (2010)	QoL affected. Feelings of Loss and sacrifice
	Barrera & Atenafu (2008)	Higher levels of internalizing problems than ill child but normal range. QoL did not differ.
	Batte, Watson & Amess (2006)	Jealousy of and worry about ill child
	Cox et al (2003)	13% of siblings coped by being internally reactive, eg. Emotional. 3% coped by showing no physical or emotional response.
	Cuskelly & Gunn (2006)	No difference in sibling perception of their own competence and self-worth.
	Fay & Barker-Collo (2003)	Siblings of TBI had more internalising and externalising problems than siblings of ORTHO.
	Fleitas (2000)	Feelings of loneliness, fear, guilt, sadness, embarrassment and confusion.
	Gallo & Szychlinski (2003)	Lower global self-worth if ill child has diabetes. Higher on social acceptance scale if ill child has asthma.
	Hamama, Ronen & Feigin (2000)	Higher anxiety if the ill child more recently diagnosed. Siblings who were older and older than ill child, and had more self-control, had less loneliness and anxiety.
	Hames (2008)	Feelings of loss and responsibility
	Hames & Appleton (2009)	Worry and anxiety over the child's illness. Fear of being left alone with child. Feelings of love and responsibility.
	Havermans, Wuytack, Deboel, Tijtgat, Malfroot, De Boeck & Proesmans (2010)	QoL in siblings was reduced when the child with CF had been hospitalised and the illness was more severe
	Hollidge (2001)	53.5% of siblings had moderate anxiety, 32.1% had high anxiety. ½ fell below the mean for total self-concept. Siblings not depressed but internalised their problems. Majority of siblings reported 'often' feeling responsible, unhappy, jealous, negative and guilty. Interviews found feelings of anxiety, worry and anger, and a feeling of disavowing their own needs.
Houtzager, Grootenhuis & Last (2001)	75% of siblings had anxiety (8-10 th percentile). Boys of all ages and girls aged 7-12 years were significantly higher on anxiety than norms.	
Houtzager, Grootenhuis, Hoeskstra-Weebers, Caron & Last (2003)	Adolescent girls had more internalising problems and anxiety and peers one month after diagnosis. Child and adolescent had impaired emotional, social and overall QoL at Month 1. 7-11 year olds had impaired physical QoL at Month 1. At 6 months, 35% of adolescent grls reported internalising and externalising problems in clinical borderline range. Adolescent boys improved at 6 months.	
Houtzager et al (2004a)	Similar anxiety to control groups. Siblings had lower QoL than control.	
Houtzager, Grootenhuis,	Lower QoL	

Caron & Last (2004b)	Older siblings had more negative emotions than peers
Houtzager, Grootenhuis, Hoekstra-Weebers & Last (2005)	
Hutson & Alter (2007)	Feelings of trying to constrain or separate aspects of life. Feelings of despair; sadness, jealousy, loneliness and abandonment.
Jones, Welsh, Glassmire & Tavegia (2006)	More trait anger than control
Kendall (1999)	Feelings of loss and sorrow
Lahteenmaki, Sjoblom, Korhonen & Salmi (2004)	Young siblings more likely to have conduct, psychosomatic and behaviour problems at 3 months, but not follow-up. School age children have conduct, learning, psychosomatic, impulsive-hyperactive and other behaviour problems at 3 month and F/U. State and trait anxiety higher at 3 months but same as control at F/U.
Loos & Kelly (2006)	Feelings of sadness, anger and fear. Feelings of being left out and rejected.
Macks & Reeve (2007)	No differences in depression scores or behaviour conduct between siblings of children with autism and siblings of children with no condition . Higher self-concept, behaviour, intellectual and school states.
Martinson, Gilliss, Colaizzo, Freeman & Bossert (1990)	Feelings of fear, loneliness, jealousy and sadness.
Orsmond & Seltzer (2009)	36% reported depressive symptoms over cut off. 8.5% reported clinically relevant anxiety symptoms. Sisters more depressive than brothers.
Patterson, Millar & Visser (2010)	Siblings described some unmet needs: expressing and coping with feelings, acknowledgement and attention for self,
Read, Kinali, Muntoni & Garralda (2010a)	Comparable SDQ between siblings and norms. % reaching high risk threshold o emotional subscale was twice of normative sample. GHQ and HADS were either comparable or more favourable for siblings.
Read, Kinali, Muntoni, Weaver & Garralda (2010b)	Diagnosis caused feelings of shock, anger, sadness, confusion, jealousy, neglect isolation and guilt
Sargent, Shaler, Roghmann, Mulhern, Barbarian, Carpenter, Copeland, Dolgin & Zelter (1995)	Siblings felt they had increased negative feeling states since their sibling became ill.
Silver & Frohlinger-Graham (2000)	Higher on anxiety and interpersonal sensitivity, and higher on global severity Index than well siblings.
Swift, Taylor, Kaugars, Drotar, Yeates, Wade & Stancin (2003)	No difference in siblings on internalising, externalising or total behaviour problems. Child behaviour and sibling behaviour were correlated.
Verte, Roeyers & Buysse	No differences on SDQ between siblings of children with or without autism. Siblings of HFA

(2003)	have higher self-concept. Siblings of HFA are higher on subscales of honesty-trust and verbal self-concept.
Wilkins & Woodgate (2007)	Feelings of anger, worry, sadness, hope and pride. Lack of control over life.
Williams, Williams, Graff, Hanson et al (2002)	Behaviour of siblings is directly affected by SES and cohesion in family, age of sibling, support, and knowledge about the illness.
Wood, Sherman, Hamiwka, Blackman & Wirrell (2008)	No depression in siblings and no difference in anxiety between siblings and norms. Siblings' QoL, depression and anxiety were correlated.
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Family	
Areemit et al (2010)	Themes of family conflict, awareness of their siblings behaviours and thoughts, and feelings of responsibility.
Barr & McLoed (2010)	Themes of disruption of family life, responsibility, worry about their parents and lack of attention from parents.
Batte, Watson & Amess (2006)	Themes of disruption to family routine, worry about their ill sibling (which ½ felt unable to share with parents), and jealousy of their ill sibling.
Fleitas (2000)	Feelings of responsibility, jealousy and resentment
Gallo & Szychlinski (2003)	Siblings of diabetes less satisfied with their family communication than healthy or asthma siblings.
Giallo & Gavidia-Payne (2006)	More likely to have better mental health if more family hardiness and time and routines and problem solving information.
Hames (2008)	Feelings of protectiveness over ill child and taking a caring role. Feelings of responsibility.
Hames & Appleton (2009)	Impact on family and parents, jealousy over lack of parental attention, and feelings of responsibility.
Havermans, Wuytack, Deboel, Tijtgat, Malfroot, De Boeck & Proesmans (2010)	Siblings older than child with CF reported more of an impact than siblings younger than child with CF
Hollidge (2001)	Worry over impact on mother, lack of time/attention from parents, and feelings of responsibility and protectiveness.
Hutson & Alter (2007)	Themes of parents being unwilling to talk, impact on Family life, feeling overlooked and excluded, and worry about parents ad child.
Kendall (1999)	Impact on family life, and feelings of resentment.
Loos & Kelly (2006)	Feelings of responsibility, impact on sibling relationship and family routines, and feelings of rejection and left out.
Martinson, Gilliss, Colaizzo, Freeman & Bossert (1990)	Impact on family, parental rejection ad exclusion, and feelings of jealousy.
Mikami & Pfiffner (2008)	Impact on sibling relationship, and more conflict in sibling relationship.
Patterson, Millar & Visser (2010)	Siblings identified some unmet areas of need: work around sibling relationship,

Petalas, Hastings, Nash, Dowey & Reilly (2009)	Impact on family life and sibling relationship, feelings of lack of attention and worry, and loss of sibling relationship.
Petalas, Hastings, Nash, Hall, Joannidi & Dowey (2011)	More conflict and rivalry in siblings relationship when child with ASD SDQ total difficulties score s higher. Negative sibling relationship when critical expressed emotion in family environment
Read, Kinali, Muntoni & Garralda (2010a)	Impact on family life and daily activities
Read, Kinali, Muntoni, Weaver & Garralda (2010b)	Impact on family activities. Either positive effect on family cohesion, e.g. more home based activities, or negative effect, e.g. loss of relationships, role transfer and less attention.
Sargent, Shaler, Roghmann, Mulhern, Barbarian, Carpenter, Copeland, Dolgin & Zelter (1995)	Family disruptions and separations, lack of parental attention, and positive of family becoming closer.
Sloper (2000)	Loss of attention and status, los of own and families' usual activities and routines, loss of companionship with ill siblings. However, at 18 months F/U, closer family relationships.
Swift, Taylor, Kaugars, Drotar, Yeates, Wade & Stancin (2003)	No difference i sibling relationships.
Weiss, Schiaffino & Ilowite (2001)	No differences found in sibling relationships
Wilkins & Woodgate (2007)	Interruption to family life.
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Social/Stigma	
Areemit et al (2010)	All areas of life effected
Barr & McLoed (2010)	Stigma with strangers, and interactions with peers effected.
Batte, Watson & Amess (2006)	Felt more mature than peers
Cox et al (2003)	Second most common coping strategy (19%) is to seek social support
Fleitas (2000)	Feelings of loneliness and embarrassment
Gallo & Szychlinski (2003)	Siblings of children with asthma were higher on social acceptance scale than siblings of children with diabetes.
Giallo & Gavidia-Payne (2006)	More peer problems and lower pro-social behaviour in siblings compared to UK norms
Hamama, Ronen & Feigin (2000)	Older siblings reported less loneliness.
Hames (2008)	Noticing public reactions. Feelings of lack of peer understanding-worries about disclosure.
Houtzager, Grootenhuis, Hoekstra-Weebers & Last	Girls reported lower social QoL in comparison to norms

(2005) Hutson & Alter (2007) Kendall (1999) Labay & Walco (2004)	Felt socially isolated, and noticed the stigma and felt unable to talk to peers. Feelings of victimisations and impact on peer relationships. Increase in social difficulties, and if the child was late in birth order, the siblings had greater social problems.
Martinson, Gilliss, Colaizzo, Freeman & Bossert (1990) Patterson, Millar & Visser (2010) Petalas, Hastings, Nash, Dowey & Reilly (2009) Read, Kinali, Muntoni & Garralda (2010a) Read, Kinali, Muntoni, Weaver & Garralda (2010b) Sloper (2000) Verte, Roeyers & Buysse (2003)	Feelings of loneliness and lack of peer understanding Siblings identified some unmet needs: peer support (friends), peer support (similar experience). Impact on social relationships, and feelings of lack of understanding and rejection Impact on social life Importance of peer relationships were highlighted. Importance of peer relationships were highlighted Siblings of HFA scored themselves higher on social skilled behaviour than control group.
Resilience	
Areemit et al (2010) Batte, Watson & Amess (2006) Cox et al (2003) Fay & Barker-Collo (2003)	Feelings of compassion and concern Felt more mature than peers and more protective towards their chronically ill sibling 65% of siblings cope by doing something Children whose sibling had experienced a TBI rated the warmth closeness of the relationship higher; while relative status/power, conflict, and rivalry received lower ratings when compared to siblings of children with orthopaedic injury.
Fleitas (2000) Hames (2008) Havermans, Wuytack, Deboel, Tijtgat, Malfroot, De Boeck & Proesmans (2010) Hollidge (2001) Petalas, Hastings, Nash, Dowey & Reilly (2009) Sargent, Shaler, Roghmann, Mulhern, Barbarian, Carpenter, Copeland, Dolgin	Theme of 'resilience' included lessons learned, independence, altruism Feelings of independence and caring Siblings of children with CF reported higher levels of QoL than siblings of healthy children. Feelings of empathy and protectiveness over ill sibling Feelings of acceptance Positive themes included becoming more compassionate and caring (16%), family members become closer (16%), Expanded life experiences (18%).

& Zelter (1995)	
Sloper (2001)	At 18 months siblings felt they had improved in maturity, understanding and compassion.
Wilkins & Woodgate (2007)	Feelings of 'life goes on'.
Wood Rivers & Stoneman (2008)	Siblings higher in persistence were rated as having less negative relationship with siblings and less unkindness and less avoidance and embarrassment.

The psychological wellbeing of siblings of children with CFS/ME: a qualitative study

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Abstract

Chronic Fatigue Syndrome or myalgic encephalopathy (CFS/ME) has a negative impact on a child and their parents. It is not known what the impact is for the siblings of children with CFS/ME. Nine siblings participated in semi-structured interviews. Siblings identified a number of negative impacts to their family and to themselves, as well as describing some protective family factors. These findings have implications for current practice in CFS/ME paediatric services.

Keywords: Sibling, chronic fatigue syndrome, myalgic encephalopathy, psychological wellbeing, quality of life

Introduction

Chronic fatigue syndrome or myalgic encephalopathy (CFS/ME) has a negative impact on a child's schooling (Sankey et al., 2006; Crawley et al., 2009; Rangel et al., 2000; Crawley, Emond, & Sterne, 2011), their social relationships (Bell et al., 2001) and their parents (Missen et al., 2011). CFS/ME is defined as "generalised fatigue, causing disruption of daily life, persisting after routine tests and investigations have failed to identify an obvious underlying 'cause'" (Royal College of Paediatrics and Child Health, 2004). The National Institute of Health and Clinical Excellence (NICE) guidelines (NICE, 2007) recommend children need to have fatigue for a minimum of 3 months before making a diagnosis. CFS/ME is relatively common (the prevalence is 0.19- 2%) (Chalder et al., 2003; Chalder et al., 2003; Jones et al., 2004; Jordan et al., 2000).

Parents of children with CFS/ME have raised concerns about its impact on their healthy child/children (Missen et al., 2011). Parents felt that CFS/ME can cause the ill child to feel angry and irritated, which was often directed at their well siblings, and that well siblings missed out on attention from parents, resulting in them becoming more self-reliant and independent.

Only one study has explored the impact on siblings of children with CFS/ME. Siblings identified a number of stressors when talking informally to a support worker, including worries about "apparent parental dilution of care or concern" (Jackson, 1999, p30); a change in the sibling relationship; restrictions on family activities; deterioration in peer relationships; and the uncertain or contradictory medical advice given to the family. There are a number of problems with this paper (Jackson, 1999); it did not describe either the method or analysis used, no quotes were used, and terms, such as "parental dilution of care" were not described or explained. Siblings were recruited whilst visiting the child with CFS/ME in hospital; as few children with CFS/ME are hospitalised, this is not a representative sample.

There could be both disadvantages and advantages to having a sibling with a chronic illness (Williams, 1997; Sharpe et al., 2002; Vermaes et al., 2012). The negative effects on siblings include an increase in both internalizing (e.g. depression and anxiety) and externalizing problems (e.g. behaviour, social and aggression problems) (Sharpe et al., 2002; Vermaes et al., 2012). Positive effects include increased empathy and personal growth (Williams, 1997).

Interview and diary data describe both a negative impact on the sibling's emotional wellbeing (Areemit et al., 2010; Barr et al., 2010; Batte et al., 2006; Fleitas, 2000; Hames, 2008; Hames et al., 2009; Hollidge, 2001; Hutson et al., 2007; Jackson, 1999; Kendall, 1999; Loos et al., 2006; Martinson et al., 1990; Petalas et al., 2009; Read et al., 2010; Sargent et al., 1995; Sloper, 2000; Wilkins et al., 2007), and a positive impact (Areemit et al., 2010; Barr et al., 2010; Batte et al., 2006; Fleitas, 2000; Hames, 2008; Hames et al., 2009; Loos et al., 2006; Petalas et al., 2009; Read et al., 2010; Sargent et al., 1995; Sloper, 2000; Wilkins et al., 2007). Negative impact includes disruption to family life; feelings of exclusion and lack of attention from parents; and social impact including negative public perceptions and impact on peer relationships. The positive effects include increased compassion and caring, increase in family cohesion and relationships, maturity and independence.

It may be useful to think of the impact of CFS/ME on siblings within a resilience model. Key risk and protective factors are known to impact on children's development, including family relationship and family situation factors (see Gilligan (2004) and Velleman (2009) for review). Resilience is described as "a dynamic process encompassing positive adaptation within the context of significant adversity." (Luther, Cicchetti, & Becker, 2000). Some children seem to be more resilient to negative life events, such as divorce and parental death, than others (Greene, Anderson, Hetherington, Forgatch, & DeGarmo, 2012; Heinzer, 1995). Prince-Embury (2008) suggests that there are three key factors which impact on a child's resilience; sense of mastery (e.g. self-efficacy), sense of relatedness (e.g. ability to relate effectively with others) and emotional reactivity (e.g. sensitivity, threshold and intensity of the reaction,

and length of time it takes to recover from emotional upset and impairment whilst upset). It has been suggested that there is no single characteristic that is protective across all life experiences, so it is important to assess these possible characteristics within each negative life event (Glantz & Sloboda, 1999).

It may be that the differences in sibling experience could be due to the way in which the sibling appraises the situation. Previous literature has suggested a link between appraisal of life events, social support and adaptive behaviour (Jackson & Warren, 2001). They suggest that if a life event is appraised as negative, social support can act as a mediator to adaptive behaviour, and that if a life event is appraised positively, then less social support is needed for an adaptive behaviour outcome (Jackson & Warren, 2001). However, if a life event is appraised negatively, and there is little social support, there could be a greater emotional impact on the sibling.

The impact described by siblings of children with CFS/ME (Jackson, 1999) and the siblings of children with other chronic health conditions (CHCs) are very similar. However, siblings of children with CFS/ME also described the uncertain or contradictory medical advice given to the family, as difficult. No positive effects were described in the Jackson (1999) paper. It is unclear whether these differences are due to the particular group of children interviewed (Jackson, 1999), or due to the intrinsic differences that CFS/ME has compared to the other illnesses described.

Aims of the study

The aims of this study were to gain understanding into the psychological wellbeing of siblings of children with CFS/ME. I had three research questions:

1. Is there a psychological, physical, social or quality of life impact on siblings of children with CFS/ME?

2. Are there any psychological or social factors, which may be protective for siblings of children with CFS/ME?
3. Should/could services be psychologically supporting siblings of children with CFS/ME?

Method section

Design

A qualitative, interview approach³ was used in order to gain a greater understanding of the individual experiences of siblings.

Participants

Siblings of children with CFS/ME were recruited between July 2011 to April 2012 from a Specialist Paediatrics CFS/ME Service. This service covers a region in the South West of England, with a population of around 400,000 children aged 5 to 19 years. In addition, the service offers assessment and treatment to children from out of the region who cannot access more local specialist services. Children are assessed and offered treatment in outpatient clinics, unless they are too severely affected to attend clinic, in which case they are seen at home. Siblings were eligible for this study if the child with CFS/ME (later referred to as 'index' child) was attending a follow-up appointment, if the index child was between 8-18 years old and if the sibling was aged 12 to 17 years and who lived with the index child full-time.

Procedure

During follow-up appointments parents were told about the study by the clinician and asked to sign a consent-to-contact form (Appendix B), to allow the researcher to contact them to

³ Originally a mixed method approach was used but due to small sample size, only the qualitative part of the study will be reported here. See Appendix N for more detail about the quantitative part of the study

tell them (and the sibling) more about the study. At this point they were also given a participant information sheet (Appendix C). The researcher phoned the family, spoke to the parent, and if the sibling was interested, the sibling. If the sibling and parent both agreed, the researcher immediately sent out the questionnaire pack (Appendix D) and consent forms (Appendix E), with a pre-paid envelope. Every participant was given the opportunity to take part in the interview (if they were interested, they could tick the 'yes' box on the questionnaire). If the questionnaire was not returned within two weeks, a reminder letter was sent (Appendix F). It was hoped that purposive sampling could be used, in order to ensure that interviews included a range of informants, in terms of age, sex, sibling order for the siblings as well as illness severity and length of illness for the index child with CFS/ME. However, due to slow recruitment, the first nine participants were contacted in order to arrange a suitable time and location to be interviewed.

Qualitative Semi Structured Interviews

All siblings who returned their questionnaire were given the opportunity to take part in an interview. Out of 18 returned questionnaires (nine female), 11 siblings (six female) asked to take part in an interview. Siblings were asked whether they would prefer to be interviewed at home, at their school or at their local CFS/ME clinic. Siblings and parents completed a consent form prior to the interview⁴. Siblings were interviewed using the semi-structured interview schedule as the basis for a qualitative, exploration of the psychological, social and quality of life impact their siblings' CFS/ME had on them (Table 1). Previous research had highlighted the impact that chronic health conditions can have on all aspects of a siblings' life; school life, home life, friends, family, relationships and mood. Due to this, the interviews were focussed on asking about all aspects of a young person's life, in order to ascertain

⁴ See Appendix E and G for consent forms

whether any of them was affected by the CFS/ME. All interviews were digitally recorded and transcribed⁵.

Table 1: Description of questions asked in qualitative interviews

Thoughts about the study

Thoughts/feelings/expectations when told about the study

Life at Home

Who do you live with? Typical day? Typical weekend? Arguments? Family days? Weekends at home? If you could change one thing at home what would it be? Home life before and after CFS/ME?

Life at School

Who do you spend time with? Best and worst days at school? Favourite/worst lessons/plans for the future? Importance of school?

Friends

Friends from school? Best friends/group? Link between friends and home? Importance of friends?

Physical Health

Time off school/work? What kind of illnesses do you get? How is illness treated within the family?

Relationship with Sibling

Age between you? Share a room, friends, life? Argue? Get on? Difference between siblings with CFS/ME and those without? Change before/after diagnosis?

CFS/ME

What does it mean to you? How did you hear about it? What do other people think about it? Does it have an impact on you? What's the worst thing about having a sibling with CFS/ME?

⁵ See Appendix H for example transcript

The interviews were analysed using a framework approach to Thematic Analysis (Braun & 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 & 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 researchers⁶.

The transcripts of the first five interviews were read and reread by two researchers (SV and LB⁷), who highlighted any words or segments which related to the siblings' experiences of CFS/ME. If these words or segments were repeated or seemed to be important to the sibling, they were then coded by the individual researcher, and then discussed together to check for similarities and differences between the codes. Any differences between the codes were checked, discussed and agreed. From this initial coding, two additional questions were added to the interview; in what way do you think having a sibling with CFS/ME has changed you or your family? What, if anything, do you think our service should offer for siblings of children with CFS/ME?

Ethics

There were two main ethical issues that were thought about, discussed and brought to the local NHS ethics committee. Firstly, there were concerns about whether there would be an impact on the child with CFS/ME. Due to this, the study was discussed with the parents and the sibling, but not the child with CFS/ME. This was to stop any additional burden on the

⁶ Reflexivity is discussed in Appendix M

⁷ LB is a qualitative researcher at the Centre for Child and Adolescent Health

child with CFS/ME, and to try and move the focus of the interview away from the child with CFS/ME. Secondly, there were concerns that the interviews may upset the siblings. To prevent this from happening, the siblings were told they could stop the interview at any time, or could choose not to answer particular questions. The interviews were conducted by a trainee clinical psychologist, who was trained in risk assessment, and was aware of the local services available to children. There was also a team consultant clinical psychologist, who could be available if needed. The study received full approval from both the local NHS ethics committee and the Doctorate student's University (Appendix J & K).

Results

Descriptive Statistics. Siblings were recruited between July 2011 to April 2012. During this period, 302 new follow-up children⁸ were seen. Of this number, 91 were eligible, and 34 parents were approached during a follow-up appointment by the clinician. Ten did not return the consent to contact forms, and twenty-four signed consent-to-contact forms were returned to the researcher. Of these, twenty-three agreed to be sent the questionnaire pack (11 female), and of those eighteen questionnaires were returned (9 female). Eleven of the siblings, who returned their questionnaire, stated that they would like to take part in the interview (6 females). Nine of those siblings were subsequently interviewed (5 females). The interviewer (first author) was a 28 year old, white, female, psychologist in training. Seven of the siblings opted for the interviews to take place in their home, one opted for it to take place at the CFS/ME service, and one opted for it to take place at her local GP surgery.

To determine whether those recruited were representative of the clinic cohort, a comparison of the index children with CFS/ME with all the children attending an initial assessment appointment, aged between 8-18 years (Table 2) within the last 6 years⁹ was made. The size of the qualitative sample was insufficient to make meaningful statistical comparisons with the group of unselected participant, so the full 18 index children were explored. The siblings recruited to this study had an index child who was similar to the cohort apart from being slightly younger (mean age index child 12.8 years (SD=3.1), mean age cohort 14.2 years (SD=2.1)).

⁸ Most of these children were seen on multiple occasions, but each follow-up child was counted only once.

⁹ The index children from this study had attended an initial assessment appointment since 2006.

Table 2

Characteristics of Index Children¹⁰ and general CFS/ME population at clinical assessment

	N	Index Children	General CFS/ME Bath population (N=267)	P-value*
		Mean (SD)	Mean (SD)	
Age (years)	17	12.8 (3.1)	14.2 (2.1)	0.01
Female	17	13 (76.5%)	199 (74.8%)	0.88
		Median (IQR)	Median (IQR)	
Time to assessment (months)	17	11 (7.5 – 18)	12 (7 – 24)	0.76
Chalder Fatigue Score (0 – 33)	15	26 (22 – 29)	25 (22 – 29)	0.94
SF-36 physical function (0 – 100)	17	19 (16 – 21)	21 (16 – 24)	0.24
Anxiety (SCAS) (0 – 90)	15	25 (20 – 43)	32 (18 – 47)	0.57
No. of Symptoms (0 – 14)	17	9 (9 – 10)	8 (7 – 10)	0.68
Anxiety (HADS) (0 – 21) ^{***}	17	7 (6 – 10)	9 (5 – 13)	0.44
Depression (HADS) (0 – 21) ^{***}	17	7 (3 – 9)	8 (5 – 11)	0.10
Visual Analogue Pain	16	52 (27.5 – 76.5)	53 (18 – 73)	0.49
School attendance past week	17	n (cumulative %)	n (cumulative %)	

¹⁰ Index children are the children with CFS/ME whose siblings were recruited to the study.

None	4 (23.5%)	59 (23.8%)	0.30
10%	3 (41.2%)	18 (31.1%)	
20%	2 (52.9%)	20 (39.1%)	
40%	0 (52.9%)	32 (52.0%)	
60%	6 (88.2%)	49 (71.8%)	
80%	2 (100.0%)	49 (91.5%)	
100%	0 (100.0%)	17 (98.4%)	
Not applicable	0 (100.0%)	4 (100.0%)	

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

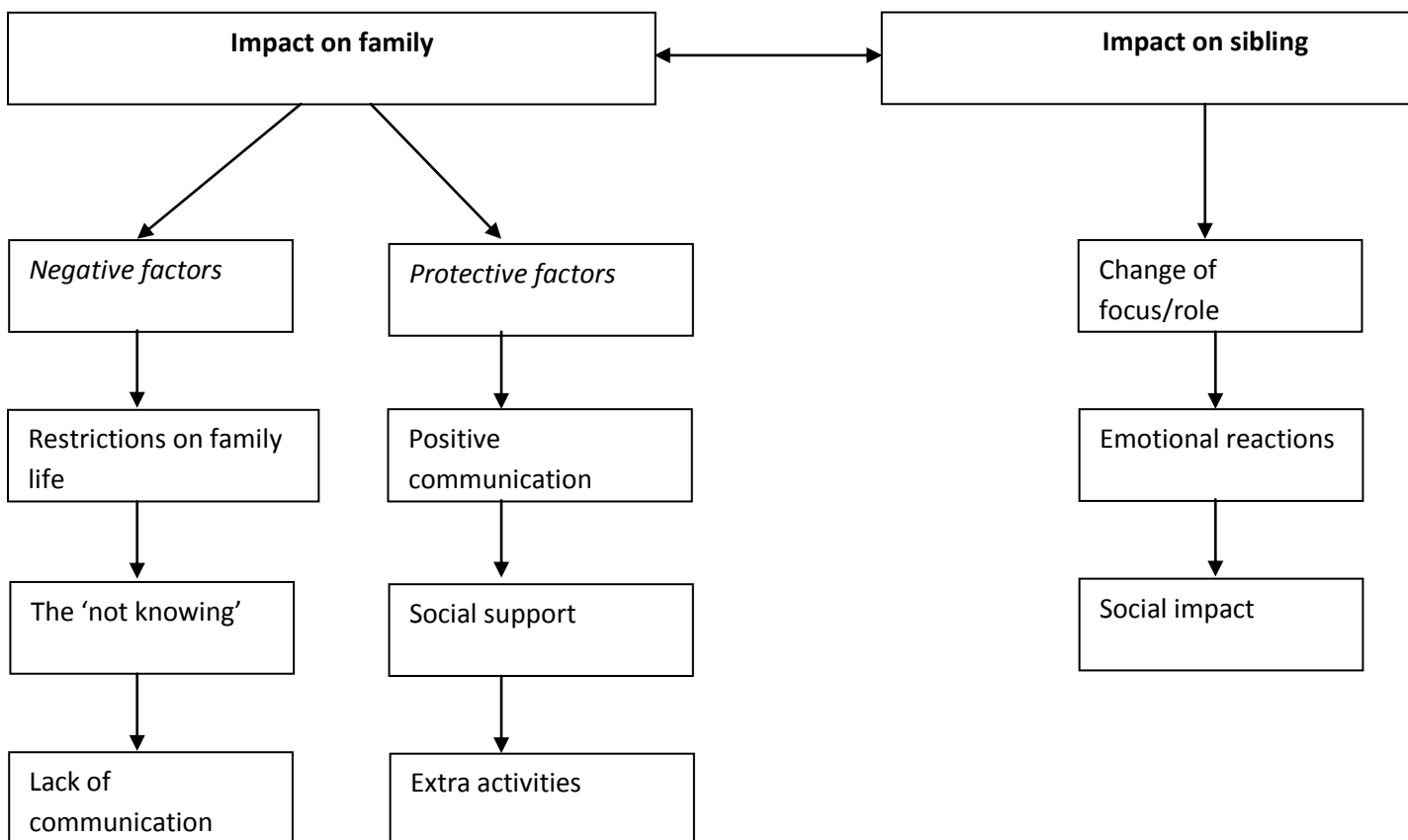
*** Only completed by patients age \geq 12 years (11 whose siblings were recruited)

Qualitative Results

Recruited siblings who wanted to participate in the interviews were similar to those who did not want to have an interview in terms of age and gender of sibling and age and gender of index child ($p>0.05$).

Two overarching themes emerged from the data: Impact on Family and Impact on Siblings with a number of sub- themes (Table 5). These overarching themes will be explored, with sibling accounts given for each theme. ¹¹

Table 5: Qualitative themes



¹¹ For a full description of all the quotes for each theme, see Appendix K

Impact on family.

All of the siblings interviewed talked about the impact that CFS/ME had on their family. When asked how things were at home, they made links between how things were at home, with how well or ill the child with CFS/ME was currently, for example “they’re really good at the moment because ****’s doing really well” (P7, page 1). Some siblings described the impact as negative (“negative factors”) and some as positive (“protective factors”).

Negative factors.

Three main ‘negative impact on the family’ related themes emerged: Restrictions on Family Life, ‘Not Knowing’ and ‘Lack of Communication’.

Restrictions on family life All of the siblings talked about some level of restriction that having a child with CFS/ME in the family caused, for example, limiting going out as a family, limiting family activities, such as holidays, and limiting activities inside the house, such as TV and friends coming over:

“That’s another thing that’s changed because of ****’s illness, is that before we were big campers, big walkers, big cyclers and now we just can’t do that.” (P4, page 7)

‘Not knowing’. All of the siblings had difficulties in understanding what CFS/ME is, how it affects their siblings, and whether the behaviours their siblings exhibit could be explained by the diagnosis:

“I find it quite difficult really cause I don’t really fully understand what it is” (P7, page 5)

Three siblings also talked about how the unpredictability of CFS/ME; the symptoms and the impact they have on the sibling and the family, changes:

“We can’t plan too far ahead because we don’t know if she will be OK” (P8, page 3)

Two siblings described that their attitudes to CFS/ME changed over time, which they linked to an increase in knowledge and understanding

“I sort of thought ‘oh well she’s putting it on’ at first [and then] I just thought well yeah this is serious” (P2, page 3)

Three of the nine participants believed that either their siblings did not have CFS/ME, or that the diagnosis allowed the sibling to behave in ways that they would not be able to do if they had not been diagnosed:

“Well I didn’t feel like sorry for him because he’s always been lazy, and I didn’t know if it was genuine or not” (P1, page 6)

The siblings seemed cautious when describing their disbelief, for example by wondering whether the ill brother or sister could have pushed themselves a bit more, or the fact that sometimes the ill child seems to have more energy, and other times they have less.

“There was definitely a difference between when she didn’t know what she had and when she knew what she had” (P6, page 12)

Lack of Communication. Seven of the nine siblings talked about a range of ways in which lack of communication, or negative communication impacted on them and their family, for example feelings of being left out, changes in how the family have fun together, more arguments or more silence.

Three of those siblings talked about the difficulties they had around communication about CFS/ME. These siblings also highlighted feelings of disbelief around the diagnosis. The difficulty in communication seemed to occur in two ways, either in feeling that CFS/ME was not communicated well to them;

“I was never properly told about it” (P5, page 6)

Or that they found it difficult to explain or describe to others what CFS/ME was;

“I don’t really explain it cause, cause I don’t even know exactly what it means” (P1,
page 12)

Five of the seven siblings talked about negative, or lack of, communication within the family, e.g. a change in the way the family communicated since their sibling became ill and a feeling of being unable to talk to their parents or siblings about their feelings:

“We used to have debates, the kind of, just jokey debates round the table; it’s hard to remember that far back” (P6, page 3-4)

Six siblings talked about difficulties in talking to people outside of their family about the CFS/ME. All of them mentioned how hard it was to talk to their friends about it:

“They don’t understand [other people]” (P4, page 10)

Two of those siblings talked specifically about the lack of understanding, and how difficult it was to communicate to services, such as schools:

“Yeah you kind of feel like there’s just a big brick wall and um you can’t talk to the school and really explain it to them.” (P9, page 2)

Protective Factors.

Eight of the siblings described factors which they described as helping them cope with the impact on the family. These emerged in three themes: ‘Positive communication’, ‘Social Support’ and ‘Extra Activities’

Positive Communication. Seven of the nine siblings spoke about the way in which positive communication within the family was helpful to them. Six of the siblings talked about how the openness in their family seemed to help them to understand more about CFS/ME and how it may impact their family:

“They [parents] helped me to understand and stuff. Cause after that I understood why they had to help her out” (P7, page 9)

Three of those siblings talked about how communication within the family was facilitated by things like family meals or days out:

“I come back from school and then I usually go talk to *** for a bit, me and Mum” (P2, page 2)

Five of the siblings specifically stated that if they had a problem or worry they would feel most comfortable talking to either their Mum or Dad:

“If anyone I would speak to my mum, or even my dad” (P9, page 5)

One of the siblings in particular highlighted how important they think positive communication is in a family affected by CFS/ME:

“If a family isn’t very functional to start with I think a family could get completely ripped apart by it” (P4, page 11)

Social Support. Eight of the nine siblings talked about the importance of social support. Six spoke about the close, supportive relationship they had with their brother or sister:

“We’ve always got on really well” (P8, page 5)

Five of the siblings mentioned the close relationship they had with their parents:

“If it was a family problem, I’d go to family” (P4, page 5)

Six also spoke about the close relationships they have with people outside of the family, such as friends:

“Obviously they understand...that it is harder, but they don’t mind” (P3, page 5)

Extra Activities. Two siblings also seemed to be able to cope more with the negative impact of CFS/ME if they had extra activities outside of the family, for example hobbies, or friendships away from the family:

“If I have a bad day either I go out on my road bike, I go mountain biking or I go for a run” (P4, page 5)

Impact on Sibling

All of the siblings talked about the impact that the CFS/ME had directly on them. The ways in which the siblings described the impact were divided into three main themes: Change of Focus/Role, Emotional Reaction and Social Impact.

Change of Focus/Role. Eight of the nine siblings talked about how they felt the focus from the parents had shifted from being a shared focus, to being more focussed on the sibling with CFS/ME:

“So, family life changed quite a bit. It’s become more focussed on ****” (P4, page 3)

Two of the siblings spoke about personality or emotional changes in the child with CFS/ME, since the diagnosis.

“She has a tendency to strike out at things she thinks is wrong or doesn’t fit with her idea of good. She never used to do that” (P6, page 5)

Some talked about how the child with CFS/ME was treated differently with different rules, for example, routines surrounding bed times, going to school or not and tolerance to bad behaviour.

“whatever time I go to bed, I still have to wake up between 6 and half 6, but he, he...sort of had that choice as well of whether to wake up or not”. (P1, page 6)

They also talked about their role having changed within the family. Some took on the caring role, or feeling they had to be more protective of their brother or sister.

“I do feel a little more protective of her” (P3, page 3)

However, others seemed to take on a more grown-up role than their age would suggest:

“My sister can reduce my Mum to tears....and she [Mum] obviously has to talk to someone and Dad’s at work” (P6, page 3)

In contrast, one participant felt that his brother’s illness had allowed to him to develop more:

“Because in a way I’ve found my own voice I suppose” (P4, page 11)

Although some of the siblings saw this shift in role and focus as an understandable and acceptable occurrence:

“It’s fine because she’s my sister” (P8, page 7)

Others found this shift very hard to bear:

“it was almost like unfair that, because I go to school that I decided that it was time for me to go to bed and then he just, he just stayed up...but that’s because he could just sleep it off for as long as he wants the next day” (P1, page 15)

Emotional Reactions. Eight of the nine siblings talked about some level of emotional impact. Four spoke about the change in their relationship, and how this felt like a loss,

“I suppose the worst part would be...not being able to go out with her anymore” (P2, page 7)

Some of the four felt this loss almost like a bereavement:

“I’ve lost my brother... we used to do everything together and now we just don’t” (P4, page 11)

One of the siblings spoke about feeling guilty about the things that they could still do, but that their ill brother could no longer do:

“So it makes you kind of feel guilty, you know, every now and again. Like when I learnt to surf this summer, it was one of those things that we were going to do together”. (P4, page 2)

Five of the siblings talked about feelings of hopelessness, upset and worry about their brother or sister, for example feelings that there is nothing they can do to ease the illness, worry about how or whether their sibling will improve and upset in seeing them ill and in pain:

“As it moved on and time went by and she was ill, it got more and more...upsetting in a way” (P5, page 9)

Four of the siblings talked about their feelings of anger, stress and frustration, either at their brothers or sisters for being ill, or at the illness for taking away their brother or sister.

“I could get quite frustrated and like ‘why can’t you just do this’” (P3, page 8)

Two of the siblings spoke in more positive terms about increasing hope and acceptance:

“She’s going to get better. It doesn’t really matter how long it takes” (P2, page 6)

Social Impact. Finally, eight of the siblings talked about the social impact of having a brother or sister with a diagnosis such as CFS/ME. Five talked about the decision they had made not to tell certain people about the CFS/ME, mostly because they felt that some of their friends may not understand

“Not all my friends know” (P8, page 4)

Two of the siblings also worried that the information about the CFS/ME may be used by people in their school in a negative way:

“And someone else would hear and someone else would use it” (P6, page 9)

A third of the siblings talked about a lack of understanding or a lack of interest in CFS/ME from those around them.

“The second I mention chronic fatigue they sort of switch off” (P9, page 8)

One of the siblings talked about how the CFS/ME diagnosis felt like a private thing that should be kept within the family and not shared.

“I don’t think it’s fair on my sister if everyone knows” (P8, page 4)

One of the siblings also talked about how the CFS/ME had impacted on the close relationship he had previously had with his brother:

“We, you know, just don’t really have anything, anything that we share anymore because if the illness, so...it’s horrible” (P4, page 11)

After the first five interviews, the next four siblings were asked at the end of the interview if they had any advice to siblings whose brother or sister has just been diagnosed, or any advice to CFS/ME services. Two suggested more information should be given to siblings:

“Maybe just basic information, because I’ve only heard from word of mouth. Maybe just some facts” (P6, page 14)

Two of the siblings promoted better communication within the family:

“Probably tell them to talk to their family to help them understand more about it and stuff like that” (P7, page 9)

The final sibling promoted a more understanding and caring attitude to the ill brother or sister:

“You’ve just got to look out for them really, and care a bit more and make them feel great...you’ve got to be prepared to make sacrifices” (P9, page9)

Discussion

This paper highlights the psychological impact that CFS/ME has on siblings’ lives. All of the siblings described some negative impact on their life; restrictions on their family life, a lack of communication or negative communication occurring within the family or a feeling of lack of understanding or ‘not knowing’ about CFS/ME. However, many siblings also described protective factors occurring within the family, such as positive communication, positive social supports and extra activities, reducing the illness impact.

Strengths and limitations

This is the first paper that has qualitatively explored the impact on siblings of children with CFS/ME in a methodologically sound way.

The index children of the recruited siblings were on average one year younger than those children with CFS/ME whose siblings were not recruited to this study. This could be due to the fact that older index children had siblings who were too old for this study (i.e. over 18 years of age). The siblings were recruited from a specialist, tier 3 service, meaning it is likely that their brothers and sisters represent the more complex end of the spectrum of young people with CFS/ME (Garraida, Rangel, Levin, Roberts, & Ukoumunne, 1999; Rangel et al., 2000). This indicates that the findings from this study may not be generalisable to the siblings of patients with CFS/ME seen in general practice or community settings.

Only 33 out of a possible 91 siblings were approached by clinicians about this study. There are a number of possible reasons for this low recruitment rate. It is likely that some clinicians occasionally forgot to recruit participants. Some clinicians may not know, or ask about siblings living at home with the child with CFS/ME. Consequently, siblings who are struggling may not be noticed by the team, unless a family brings it up themselves. This has implications for how the service assesses families. Finally, some clinicians may worry about recruiting from families who were already struggling, if they saw this project as an additional burden. This could mean that although the sample is representative of the children attending the local CFS/ME service, it may not reflect the breadth of families. Only 18 participants completed and returned their questionnaires and of those only 11 asked to take part in the interviews. Although no differences were found between those who took part in the interviews and those who did not, it is not known why some opted to not take part in the interviews.

Results in context of previous literature

The previous study exploring the impact of CFS/ME on siblings found that siblings had worries including “apparent parental dilution of care or concern” (Jackson, 1999, p31); a change in the sibling relationship, including change of role and care needs; restrictions on family activities; deterioration in peer relationships; and the uncertain or contradictory medical advice given to the family. There are similarities between this previous study and the current study. The themes, which seem to overlap from this current study to the previous study are the change in role/focus, restrictions on family life/activities and social impact. However, there are some differences. The current study found that siblings spoke about both the impact on their family as a whole, as well as the impact on themselves. Additionally, they talked about protective factors, which seemed to affect the impact that the CFS/ME had on them. A strong emphasis on the importance of positive communication, and how lack of communication or negative communication could impact on the lack of

understanding/knowledge and acceptance of the diagnosis, was also found. As in the previous paper, siblings described a change in role and relationship with the ill child. However, when the siblings in this current study described how this change in role and relationship affected them, it seemed to impact on their emotional wellbeing, and their sense of what their role or focus was within the family.

In addition the siblings in this current study describe a problem with lack of knowledge ('not knowing' theme). Many siblings described feeling unsure about what CFS/ME actually was, and what the impact was on their brother or sister. This could be due to a general lack of knowledge within our society and the health system around what CFS/ME is (Bowen, Pheby, Charlett, & McNulty, 2005). Or it could be due to the lack of communication between the service and siblings, or within the family around the issue of what CFS/ME is, what the impact may be on the ill-child, and so what the impact may be on the rest of the family, including siblings.

Linked to this, a third of the siblings interviewed did not believe the index child had a "real" illness. They could not tell which behaviours their ill brothers and sisters exhibited were because they were unwell or because the diagnosis of CFS/ME gave them permission to act in that way (i.e. lazy). This could be linked to attribution theory (i.e. if the siblings make negative attributions about their brother or sisters' illness, this allows them to feel angry at them rather than sorry for them) (Heider, 1958). In this current study, this disbelief seemed to be linked to how much the family had communicated to the sibling about what CFS/ME is and how it impacts on a child. It may be that lack of belief stems from lack of communication within the family. However, some of the siblings explained that they had not wanted to hear about what CFS/ME was, and so had not asked. Siblings were hesitant in describing the disbelief, which could be due to feelings of guilt; worry about seeming callous; or wanting to protect and look after their ill sibling. This is not consistent with previous literature suggesting that the feelings disbelief may be unique to CFS/ME.

As well as the negative impact on siblings, this current study describes some protective factors, which have been found in previous literature for siblings of children with chronic illnesses, such as good communication, social support and extra activities (Areemit et al., 2010; Batte et al., 2006; Cox et al., 2003; Fay et al., 2003; Fleitas, 2000; Hames et al., 2009; Havermans et al., 2011; Hollidge, 2001; Petalas et al., 2009; Sargent et al., 1995; Sloper, 2000; Wilkins et al., 2007; Wood Rivers & Stoneman, 2008). These are important to note when thinking of how to promote resilience in these young people. It could be argued that those siblings whose emotional wellbeing does not seem to be affected by their brother or sister's ill health could be more resilient than other siblings; some children seem to be more resilient to negative life events, such as divorce and parental death, than others (Greene et al, 2012; Heinzer, 1995).

Meaning of the study and possible mechanisms

It is possible to frame this current study within the resilience framework suggested by Prince-Embury (2008), which suggests that resilience is made up of three factors; heightened sense of mastery and sense of relatedness and lowered emotional reactivity. One factor which siblings described as difficult was lack of communication or difficult communication within the family. This had not been highlighted as a risk in previous sibling literature, although good communication was found as a protective factor (Cohen et al., 1994; Daniels, Miller, Billings, & Moos, 1986; Daniels, Moos, Billings, & Miller, 1987). Poor communication could be seen as a reduced sense of relatedness (Prince-Embury, 2008), which could cause greater family vulnerability (Rolland & Walsh, 2006) and possibly greater misunderstandings and heightened feelings of anger and jealousy for siblings. Rolland & Walsh (2006) recognises that crises and persistent challenges, such as an ill child, impacts upon the whole family, and that key family processes can mediate the adaptation of all members and their relationships. These factors include positive communication, connectedness, flexibility and positive outlook.

Siblings with less knowledge and understanding about CFS/ME in this study seemed to struggle more with feelings of anger and frustration directed at the ill child. This could be linked to low perceptions of control over the situation (Abramson, Seligman, & Teasdale, 1978), which can then lead to stress, anxiety and depression (Rudolph, Kurlakowsky & Conley, 2001). One could also understand it using Prince-Embury's (2008) sense of mastery factor, which promotes the importance of self-efficacy in resilience.

Family dynamics seem to play a particularly important part in how well siblings adjust to having a brother or sister with CFS/ME. Those siblings who described a good relationship with their siblings prior to the diagnosis, seemed to be more protective, understanding and knowledgeable when talking about their brother or sister now. Most of the siblings said that they would go to their Mum and Dad if they were having a problems, but some did not feel able to do this. Families where there was open communication and support around the diagnosis of CFS/ME and the impact of that on the family seemed to be described as more supportive by the siblings, than those families where very little discussion and support occurred. This supports previous resilience research, which suggests positive communication, positive family relationships and good communication with parents create an environment that supports successful youth adjustment, and is linked to positive feelings of self-worth in siblings as well as increased satisfaction with their family with less conflict (Jackson, Bijstra, Oostra, & Bosma, 1998; Steinberg, 2001; Gilligan, 2004; Prince-Embury, 2008). A link has also been found between appraisal of life events, social support and adaptive behaviour (Jackson & Warren, 2001). They suggest that if a life event is appraised as negative, social support can act as a mediator to adaptive behaviour, and that if a life event is appraised positively, then less social support is needed for an adaptive behaviour outcome (Jackson & Warren, 2001). However, if a life event is appraised negatively, and there is little social support, there could be a greater emotional impact on the sibling.

Illness characteristics, which have previously been found to have more of a negative impact on siblings of other CHCs, include whether the illness affects the day-to-day functioning of the child (Sharpe et al., 2002), or if the illness has higher mortality rates (Vermaes et al., 2012). The 'index' children in this study are known to be very ill, so daily impact would be expected (Crawley et al., 2011), although mortality is not. The variability in the way that CFS/ME impacts on children may explain some of the siblings' disbelief; if their brothers and sisters are sometimes able to do things and sometimes not able, this may cause the sibling to question how unwell they really are. Prince-Embury (2008) suggests that heightened emotional reactivity is linked to heightened vulnerability and reduced resilience. Disbelief, jealousy, anger and loss (all emotional reactions described by the siblings in this study) could be thought of as heightened emotional reactions, which could mean that they are more vulnerable.

Impact on clinical practice

This research has highlighted the need to identify those siblings who may be at risk from mental health difficulties. Rolland & Walsh (2006) promote the use of a family systems approach, which "aim to identify and fortify key interactional processes that enable families to withstand and rebound from disruptive life challenges" (Walsh, 2006) (p 3). As part of this acceptance that crises and persistent challenges, such as an ill child, impacts upon the whole family, clinicians should ensure they ask about siblings when first meeting with a family; to allow the clinician a better insight into the family as a whole.

One of the main risks for siblings being more distressed was lack of or negative communication within the family as well as a lack of knowledge about CFS/ME. Siblings should be provided with specific, tailored information about CFS/ME and what their sibling with CFS/ME is coping with. In particular this should include information about the variability of the illness, and the impact on the index child's emotional wellbeing. Clinicians can promote positive communication within the family ensuring that parents are aware of the

possible impact this may have on the sibling, and suggesting more communication, 'family time' and positive, family experiences.

Further research, exploring effectiveness of interventions is important. In order to fully explore the sibling relationship, a further study with both the index child and the sibling should be conducted, to explore whether the sibling relationship impacts on the index child's illness trajectory. It may also be useful to compare the siblings of children with CFS/ME with siblings of other CHCs, such as chronic pain. This may allow us further insights into how CFS/ME impacts on siblings differently to other CHCs.

Conclusion

In conclusion this qualitative study has explored the impact on siblings of children with CFS/ME, and found that siblings described a negative impact on both themselves and their family. In particular they noted how communication (either negative or positive), and also their knowledge of belief about CFS/ME impacted on their own adjustment and acceptance of their siblings' illness. Siblings also described some protective factors, such as communication, social support and extra activities outside of the home. This research tentatively suggests that there is an impact on siblings, and that services should provide extra support and information for siblings of children with CFS/ME.

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Appendixes

Appendix A: Dissemination Statement

The following steps will be taken to inform interested parties of the research findings.

Participants:

All participants taking part in the interviews have been offered a copy of the research paper once it is published. A summary of the research findings will also be included with copy of the paper.

Services involved in the study:

The local CFS/ME service from which participants were recruited will be offered a presentation of the research project, describing the findings and suggesting changes to current clinical practice.

The R&D department for the host trust, and the local NHS ethics committee will be sent a copy of the final write up for their records.

The University of Exeter Doctorate in Clinical Psychology:

The write up will be submitted in partial completion of this programme.

The wider clinical and academic community:

Both the research manuscript and the literature review will be submitted to the peer reviewed journal to Clinical Child Psychology and Psychiatry.

I am also intending to submit an abstract for consideration for presentation at British Association for Community Child Health Conference in October 2012.

Parental consent to contact form: Sibling Study

The psychological impact of CFS/ME on adolescent siblings

Please initial boxes if “yes”

I have read the leaflet about the study (Version 6, dated 12.03.11). I understand what the study is about and have had the chance to ask questions.	<input type="checkbox"/>
I understand that it is mine and my child’s choice about whether or not to take part in the study and that it is ok for my child to withdraw from the study at any time.	<input type="checkbox"/>
I agree that a researcher may contact me at home to discuss this study further.	<input type="checkbox"/>
I agree that a researcher may visit my child at a place that we choose and interview them for 30 minutes <i>(only complete this if you child is under 16 years old)</i>	<input type="checkbox"/>

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

Your name: Signature: Your e mail:	Your address: Your phone number: Today’s date:/...../20..... Your child’s name:
--	--

We will give you a copy of this consent form. A copy 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 personal 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 via an ID code on separate lists. The list linking the code will be kept in the University of Bristol with the consent forms.

CFS/NHS/PAEDIATRICS - Specialist help for ME.

**The impact of Chronic Fatigue Syndrome/Myalgic Encephalopathy (CFS/ME)
on adolescent brothers/sisters.**

INFORMATION LEAFLET FOR YOUNGER ADOLESCENTS

We would like to invite you to take part in a research study. This will help us learn about what happens to the brothers/sisters of young people with CFS/ME. Before you decide to take part it is important for you to understand *why* the study is being done and *what* you need to do.

Please read this leaflet carefully.

You can talk about it with your family, friends, or us if you want to. The leaflet is split into two parts. Part 1 tells you about the study and what will happen to you if you take part. Part 2 gives details about how the study will be run.

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 take part.

Thank you for reading this!

Part 1

Why are we doing this study?

We want to find out about what happens to the brothers/sisters of young people with CFS/ME.

Why have I been asked to take part?

You have been asked if you want to take part because:

- you are between 12 and 17 years of age
- you have a brother/sister who you live with fulltime
- Your brother/sister has CFS/ME and has attended an assessment at the Bath Specialist CFS/ME service.

Do I have to take part?

You do not have to take part in this study.

If you decide to take part but change your mind later, we will destroy the information we collected.

We hope that up to 40 young people will take part in this study but it is up to you to decide whether or not to take part. If you decide not to take part or decide to withdraw at any time, this will not affect the medical care that your brother/sister will be given.

Who should not take part in this study?

We do not think you should take part if you:

are under the age of 12

you do not live full time with your brother/sister who has CFS/ME

if you cannot read or write English.

What are we asking you to do?

If you and your parent agree to take part in this study, you will be sent a questionnaire pack. This pack will ask you about your mood, your family and your health. These questionnaires will take you up to 40 minutes to complete. A reminder letter will be sent to you if we have not received the questionnaire after two weeks. A reminder phone call will be made if we have not received the questionnaire back after another two weeks.

At the bottom of the questionnaire you can tick a box if you want to be interviewed. Not all the young people who tick that box will be interviewed, as only 10 interviews are taking place. If you want to be interviewed, and if the study still needs interviewees, a researcher may call you. They will arrange a time to interview you in the next two weeks at a place and time of your choice. The interview will ask about your home, school and friends and your thoughts and feelings about CFS/ME. The interview will be audio-recorded, with your permission, and will last for around 30 minutes.

Are there any problems with taking part in this study?

By filling out the questionnaires, you may be reminded of things that upset you. The questionnaires will take up to 40 minutes of your time. If you take part in the interviews, you may need to spend time talking to a researcher for about 30 minutes.

Good things about joining in

There are no specific good things for you in taking part in this study. However, if we understand more about the wellbeing of brothers and sisters of those with CFS/ME, we may be able to offer help to them in the future. It also may help you to explore your thoughts and feelings around having a brother/sister with CFS/ME.

What happens when the research study stops?

After the study stops, your brother/sister will continue to access specialist medical care if they still need it. You will not be contacted again for this study.

What if there is a problem?

We will try and deal with any problem you have during this study. . If you feel upset after taking part in the study, you can speak more with a Clinical Psychologist. To do this, you can ask the researcher, or ring Dr Esther Crawley (01225 465941. esther.crawley@bristol.ac.uk) or any member of the clinical team that you know. If the researcher is worried about anything you have told us during the interview, they may need to speak to someone else about this. They will always tell you if this is going to happen. Detailed information is given in part 2.

Will my details be kept private?

Yes. Your privacy is important to us and all your details will be handled in confidence. The details are included in part 2.

If the information in Part 1 has interested you and you are considering taking part in this study, please read the additional information in Part 2 before making any decision.

What will happen if I don't want to carry on with the study?

You can leave the study at any point and this will not affect the care that we give your brother/sister. We will keep the information that we have collected up to the time you leave the study but this is completely unidentifiable (nobody will know it is you).

What should I do if I have a problem with this study?

If you have any problems with this study, please speak to Dr Esther Crawley (01225 465941. esther.crawley@bristol.ac.uk) or any member of the clinical team that you know. You would be able to complain to the NHS in the usual way if you were not happy with the study through the Patient Advice and Liaison services (PALS) 01225 473424.

Your privacy

It is very important that all the information you give us is completely private. We will write down the things that you say from the audio-recording and take out any details linking the recording to you so that nobody will know that it was you. We may use small bits of what you say when we report the study, but the quotes will not have your name on so nobody will know it was you. The recording will be encrypted and password protected (so no-one else can listen to it) before it is stored on a secure university server. The written copy of what you said in the interview will be linked to you and your parents via a code. All personal details that could identify you will be kept secure in locked cabinets in locked offices or password protected on secure NHS computers.

All questionnaires that you fill out will not have your name on. We will give you a 13 digit identification code that will be on the top of the questionnaires. A list of names and corresponding identification numbers are kept separately and securely on a password protected NHS server.

Data protection

All data is completely anonymised and is kept on secure encrypted password protected University Servers.

Consent

We have to be absolutely certain that you are happy to take part this study, so if you say you are, we will ask you to sign our consent form. We will ask you to sign another consent form to interview you. We will also ask your parents to sign a consent form. Even if you do sign the forms, you will be free to leave at any point.

Just tell us if this is the case. Whether or not you wish to participate, you will continue to receive the same care from the clinical team.

As we said in above, everything you tell us is private. However, if you tell us something which means we feel worried about your, or someone else's safety, the interviewer will need to tell someone else about this. You will be reminded of this at the beginning of the interview. You will always be told if the interviewer feels they need to tell someone else.

What will happen to the results of the study?

This study will give us information about the wellbeing of brothers/sisters of young people with CFS/ME. We aim to publish these results in journals to help other people seeing teenagers with CFS/ME.

Who is organising and funding the study?

This research is organised by Sophie Velleman, who is a Trainee Clinical Psychologist at the University of Exeter, and Dr Esther Crawley who is the Clinical Lead for the Bath specialist CFS/ME service at the RNHRD and leads the Paediatric CFS/ME Research team at the University of Bristol.

Will I be paid to take part in this study?

No, there is no payment for taking part in this study.

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:

Dr Esther Crawley - Paediatric Consultant and Clinical Lead of the Paediatric CFS/ME Research Team. Oakfield House, Oakfield Grove, Clifton, Bristol, BS8 2BN.
Tel: (0117) 33 14099 esther.crawley@bristol.ac.uk

Sophie Velleman - Trainee Clinical Psychologist at Psychology Office, Washington Singer Building, University of Exeter, Exeter, EX4 4SB. sv248@exeter.ac.uk

THANK YOU for taking the time to read this leaflet

The impact of Chronic Fatigue Syndrome/Myalgic Encephalopathy (CFS/ME) on adolescent brothers/sisters.

INFORMATION LEAFLET FOR OLDER ADOLESCENTS AND PARENTS

We would like to invite you to take part in a research study which will help us understand what happens to the brothers/sisters of young people with CFS/ME.

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, or us if you want to. The leaflet is divided into two parts. Part 1 tells you about the study and what will happen to you if you take part.

Part 2 gives details about how the study will be run.

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 take part.

Thank you for reading this!

Part 1

Why are we doing this study?

We want to find out about what happens to the brothers/sisters of young people with CFS/ME.

Why have I been asked to take part?

You have been asked if you want to take part because you are between 12 and 17 years of age, and you have a brother/sister who you live with fulltime, who has CFS/ME and who has attended an assessment at the Bath Specialist CFS/ME service.

Do I have to take part?

You do not have to take part in this study.

If you decide to take part but change your mind later, we will destroy the information we collected.

We hope that up to 40 young people will take part in this study but it is up to you to decide whether or not to take part. If you decide not to take part or decide to withdraw at any time, this will not affect the medical care that your brother/sister will receive.

Who should not take part in this study?

We do not think you should take part if you are under the age of 12, or do not live full time with your brother/sister who has CFS/ME, or if you are unable to read or write English.

What are we asking you to do?

If you agree to take part in this study, you will be sent a questionnaire pack, which will ask you about your mood, your family and your health. These questionnaires will take you up to 40 minutes to complete. A reminder letter will be sent if we have not received the questionnaire after two weeks. A reminder phone call will be made if we have not received the questionnaire back after another 2 weeks. At the bottom of the questionnaire there is a box you can tick, which will say if you want to be interviewed or not. Not all the young people who tick that box will be interviewed, as only 10 interviews are taking place. If you want to be interviewed, and if the study still needs interviewees, a researcher may arrange a time to interview you in the next two weeks at a place and time that is convenient for you. The interview will ask about your home, school and friends and your thoughts and feelings about CFS/ME. The interview will be audio-recorded with your permission and will last for around 30 minutes.

Are there any disadvantages of taking part in this study?

By filling out the questionnaires, you may be reminded of things that upset you. The questionnaires will take up to 40 minutes of your time. If you take part in the interviews, you may need to spend time talking to a researcher for about 30 minutes.

Benefits of joining in

There are no specific benefits for you in taking part in this study. However, if we understand more about the wellbeing of brothers and sisters of those with CFS/ME, we may be able to offer help to them in the future. It also may help you to explore your thoughts and feelings around having a brother/sister with CFS/ME.

What happens when the research study stops?

After the study stops, your brother/sister will continue to access specialist medical care if they still need it. You will not be contacted again for this study.

What if there is a problem?

We will try and deal with any problem you have during this study. . If you feel upset after taking part in the study, you can speak more with a Clinical Psychologist. . To do this, you can ask the researcher, or ring Dr Esther Crawley (01225 465941. esther.crawley@bristol.ac.uk) or any member of the clinical team that you know. If the researcher is worried about anything you have told us during the interview, they

may need to speak to someone else about this. They will always tell you if this is going to happen. Detailed information is given in part 2.

Will my details be kept private?

Yes. Your privacy is important to us and all your details will be handled in confidence. The details are included in part 2.

If the information in Part 1 has interested you and you are considering taking part in this study, please read the additional information in Part 2 before making any decision.

What will happen if I don't want to carry on with the study?

You can withdraw from the study at any point and this will not affect the care that we give your brother/sister. We will keep the information that we have collected up to the time you leave the study but this is completely unidentifiable (nobody will know it is you).

What should I do if I have a problem with this study?

If you have any problems with this study, please speak to Dr Esther Crawley (01225 465941. esther.crawley@bristol.ac.uk) or any member of the clinical team that you know. You would be able to complain to the NHS in the usual way if you were not happy with the study through the Patient Advice and Liaison services (PALS) 01225 473424.

Your privacy

It is very important that all the information you give us is completely private. We will write down the things that you say from the audio-recording and take out any details linking the recording to you so that nobody will know that it was you. We may use small bits of what you say when we report the study, but the quotes will not have your name on so nobody will know it was you. The recording will be encrypted and password protected (so no-one else can listen to it) before it is stored on a secure university server. The written copy of what you said in the interview will be linked to you and your parents via a code. All personal details that could identify you will be kept secure in locked cabinets in locked offices or password protected on secure NHS computers.

All questionnaires that you fill out will not have your name on. We will give you a 13 digit identification code that will be on the top of the questionnaires. A list of names and corresponding identification numbers are kept separately and securely on a password protected NHS server.

Data protection

All data is completely anonymised and is kept on secure encrypted password protected University Servers.

Consent

We have to be absolutely certain that you are happy to take part this study, so if you say you are, we will ask you to sign our consent form. We will ask you to sign another consent form to interview you. We will also ask your parents to sign a consent form. Even if you do sign the forms, you will be free to withdraw at any point. Just tell us if this is the case. Whether or not you wish to participate, you will continue to receive the same care from the clinical team.

As we said in Part 1, everything you tell us is private. However, if you tell us something which means we feel concerned about your, or someone else's safety, the interviewer will need to tell someone else about this. You will be reminded of this at the beginning of the interview, if you decide to take part. You will always be told if this is going to happen.

What will happen to the results of the study?

This study will give us information about the wellbeing of siblings of young people with CFS/ME. We aim to publish these results in journals to help other people seeing young people with CFS/ME.

Who is organising and funding the study?

This research is organised by Sophie Velleman, who is a Trainee Clinical Psychologist at the University of Exeter, and Dr Esther Crawley who is the Clinical Lead for the Bath specialist CFS/ME service at the RNHRD and leads the Paediatric CFS/ME Research team at the University of Bristol.

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Contact / Further Information:

Dr Esther Crawley - Paediatric Consultant and Clinical Lead of the Paediatric CFS/ME Research Team. Oakfield House, Oakfield Grove, Clifton, Bristol, BS8 2BN. Tel: (0117) 3314099
esther.crawley@bristol.ac.uk

Sophie Velleman - Trainee Clinical Psychologist at Psychology office, Washington Singer Building, University of Exeter, Exeter, EX4 4SB. sv248@exeter.ac.uk

Demographic Questionnaire

The psychological impact of CFS/ME on adolescent siblings

Demographics

Your Age:

Your Gender: Male Female

Age of your sibling with CFS/ME:

Gender of your Sibling with CFS/ME: Male Female

Who lives in your house?

Do you share your bedroom? Yes No
If 'yes', who do you share it with?

How many bedrooms do you have in your house?

Would you like to be contacted again to talk about taking part in an interview? (please circle one)
YES NO

ID # _____

GROUP _____

Sibling Relationship Questionnaire - Revised (Child) 3/90

My name is _____ (completed by)

The phrase "this sibling" refers to _____ (completed about)

1. Some siblings do nice things for each other a lot, while other siblings do nice things for each other a little. How much do both you and this sibling do nice things for each other?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> Extremely much
2. Who usually gets treated better by your mother, you or this sibling?	<input type="checkbox"/> My sibling almost always gets treated better <input type="checkbox"/> My sibling often gets treated better <input type="checkbox"/> We get treated about the same <input type="checkbox"/> I often get treated better <input type="checkbox"/> I almost always get treated better
3. How much do you show this sibling how to do things he or she doesn't know how to do?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> Extremely much
4. How much does this sibling show you how to do things you don't know how to do?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> Extremely much
5. How much do you tell this sibling what to do?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> Extremely much
6. How much does this sibling tell you what to do?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> Extremely much

<p>7. Who usually gets treated better by your father, you or this sibling?</p>	<p><input type="checkbox"/> My sibling almost always gets treated better <input type="checkbox"/> My sibling often gets treated better <input type="checkbox"/> We get treated about the same <input type="checkbox"/> I often get treated better <input type="checkbox"/> I almost always get treated better</p>
<p>8. Some siblings care about each other a lot while other siblings don't care about each other that much. How much do you and this sibling care about each other?</p>	<p><input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> Extremely much</p>
<p>9. How much do you and this sibling go places and do things together?</p>	<p><input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> Extremely much</p>
<p>10. How much do you and this sibling insult and call each other names?</p>	<p><input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> Extremely much</p>
<p>11. How much do you and this sibling like the same things?</p>	<p><input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> Extremely much</p>
<p>12. How much do you and this sibling tell each other everything?</p>	<p><input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> Extremely much</p>
<p>13. Some siblings try to out-do or beat each other at things a lot, while other siblings try to out-do each other a little. How much do you and this sibling try to out-do each other at things?</p>	<p><input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> Extremely much</p>
<p>14. How much do you admire and respect this sibling?</p>	<p><input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> Extremely much</p>

15. How much does this sibling admire and respect you?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> Extremely much
16. How much do you and this sibling disagree and quarrel with each other?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> Extremely much
17. Some siblings cooperate a lot, while other siblings cooperate a little. How much do you and this sibling cooperate with other?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> Extremely much
18. Who gets more attention from your mother, you or this sibling?	<input type="checkbox"/> My sibling almost always gets more attention <input type="checkbox"/> My sibling often gets more attention <input type="checkbox"/> We get about the same amount of attention <input type="checkbox"/> I often get more attention <input type="checkbox"/> I almost always get more attention
19. How much do you help this sibling with things he or she can't do by him or herself?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> Extremely much
20. How much does this sibling help you with things you can't do by yourself?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> Extremely much
21. How much do you make this sibling do things?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> Extremely much
22. How much does this sibling make you do things?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> Extremely much

<p>23. Who gets more attention from your father, you or this sibling?</p>	<p><input type="checkbox"/> My sibling almost always gets more attention <input type="checkbox"/> My sibling often gets more attention <input type="checkbox"/> We get about the same amount of attention <input type="checkbox"/> I often get more attention <input type="checkbox"/> I almost always get more attention</p>
<p>24. How much do you and this sibling love each other?</p>	<p><input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> Extremely much</p>
<p>25. Some siblings play around and have fun with each other a lot, while other siblings play around and have fun with each other a little. How much do you and this sibling play around and have fun with each other?</p>	<p><input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> Extremely much</p>
<p>26. How much are you and this sibling mean to each other?</p>	<p><input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> Extremely much</p>
<p>27. How much do you and this sibling have in common?</p>	<p><input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> Extremely much</p>
<p>28. How much do you and this sibling share secrets and private feelings?</p>	<p><input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> Extremely much</p>
<p>29. How much do you and this sibling compete with each other?</p>	<p><input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> Extremely much</p>
<p>30. How much do you look up to and feel proud of this sibling?</p>	<p><input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> Extremely much</p>

31. How much does this sibling look up to and feel proud of you?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> Extremely much
32. How much do you and this sibling get mad at and get in arguments with each other?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> Extremely much
33. How much do both you and your sibling share with each other?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> Extremely much
34. Who does your mother usually favor, you or this sibling?	<input type="checkbox"/> My sibling almost always is favored <input type="checkbox"/> My sibling is often favored <input type="checkbox"/> Neither of us is favored <input type="checkbox"/> I am often favored <input type="checkbox"/> I am almost always favored
35. How much do you teach this sibling things that he or she doesn't know?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> Extremely much
36. How much does this sibling teach you things that you don't know?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> Extremely much
37. How much do you order this sibling around?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> Extremely much
38. How much does this sibling order you around?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> Extremely much
39. Who does your father usually favor, you or this sibling?	<input type="checkbox"/> My sibling almost always is favored <input type="checkbox"/> My sibling is often favored <input type="checkbox"/> Neither of us is favored <input type="checkbox"/> I am often favored <input type="checkbox"/> I am almost always favored
40. How much is there a strong feeling of affection (love)	<input type="checkbox"/> Hardly at all

between you and this sibling?	<input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> Extremely much
41. Some kids spend lots of time with their siblings, while others don't spend so much. How much free time do you and this sibling spend together?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> Extremely much
42. How much do you and this sibling bug and pick on each other in mean ways?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> Extremely much
43. How much are you and this sibling alike?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> Extremely much
44. How much do you and this sibling tell each other things you don't want other people to know?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> Extremely much
45. How much do you and this sibling try to do things better than each other?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> Extremely much
46. How much do you think highly of this sibling?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> Extremely much
47. How much does this sibling think highly of you?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> Extremely much
48. How much do you and this sibling argue with each other?	<input type="checkbox"/> Hardly at all <input type="checkbox"/> Not too much <input type="checkbox"/> Somewhat <input type="checkbox"/> Very much <input type="checkbox"/> Extremely much

ID Code: Flag

1 - Pre	5 - 24 mth
2 - 6 wk	6 - Misc
3 - 6 mth	7 - D/C
4 - 12 mth	

CFS/NHS/**- Specialist help for ME****HADS**

This questionnaire is designed to help describe how you feel. Please read each item and then place a cross in the box next to the reply that comes closest to how you have been feeling in the past week. Try to give your first reaction. This will probably be more accurate than spending a long time thinking about an answer.

Please cross only one box for each question

1.1 I feel tense / wound up: A	1.8 I feel as if I am slowed down: D
Most of the time 3 <input type="checkbox"/>	Nearly all of the time 3 <input type="checkbox"/>
A lot of the time 2 <input type="checkbox"/>	Very often 2 <input type="checkbox"/>
Occasionally 1 <input type="checkbox"/>	Sometimes 1 <input type="checkbox"/>
Not at all 0 <input type="checkbox"/>	Not at all 0 <input type="checkbox"/>
1.2 I still enjoy things I used to: D	1.9 I get a frightened feeling like 'butterflies' in my stomach: A
Definitely as much 0 <input type="checkbox"/>	Not at all 0 <input type="checkbox"/>
Not quite as much 1 <input type="checkbox"/>	Occasionally 1 <input type="checkbox"/>
Only a little 2 <input type="checkbox"/>	Quite often 2 <input type="checkbox"/>
Hardly at all 3 <input type="checkbox"/>	Very often 3 <input type="checkbox"/>
1.3 I get a sort of frightened feeling as if something awful is about to happen: A	1.10 I have lost interest in my appearance: D
Very definitely and quite badly 3 <input type="checkbox"/>	Definitely 3 <input type="checkbox"/>
Not too badly 2 <input type="checkbox"/>	I don't take as much care as I should 2 <input type="checkbox"/>
Little doesn't worry me 1 <input type="checkbox"/>	I may not take quite as much care 1 <input type="checkbox"/>
Not at all 0 <input type="checkbox"/>	I take just as much care as ever 0 <input type="checkbox"/>
1.4 I can laugh and see the funny side of things: D	1.11 I feel restless as if I have to be on the move: A
As much as I ever could 0 <input type="checkbox"/>	Very much indeed 3 <input type="checkbox"/>
Not quite as much now 1 <input type="checkbox"/>	Quite a lot 2 <input type="checkbox"/>
Definitely not so much 2 <input type="checkbox"/>	Not very much 1 <input type="checkbox"/>
Not at all 3 <input type="checkbox"/>	Not at all 0 <input type="checkbox"/>
1.5 Worrying thoughts go through my mind: A	1.12 I look forward with enjoyment to things: D
A great deal of the time 3 <input type="checkbox"/>	As much as I ever did 0 <input type="checkbox"/>
A lot of the time 2 <input type="checkbox"/>	Rather less than I used to 1 <input type="checkbox"/>
From time to time 1 <input type="checkbox"/>	Definitely less than I used to 2 <input type="checkbox"/>
Only occasionally 0 <input type="checkbox"/>	Hardly at all 3 <input type="checkbox"/>
1.6 I feel cheerful D	1.13 I get sudden feelings of panic: A
Not at all 3 <input type="checkbox"/>	Very often indeed 3 <input type="checkbox"/>
Not often 2 <input type="checkbox"/>	Quite often 2 <input type="checkbox"/>
Sometimes 1 <input type="checkbox"/>	Not very often 1 <input type="checkbox"/>
Most of the time 0 <input type="checkbox"/>	Not at all 0 <input type="checkbox"/>
1.7 I can sit at ease and feel relaxed: A	1.14 I can enjoy a good book, radio or TV programme: D
Definitely 0 <input type="checkbox"/>	Often 0 <input type="checkbox"/>
Usually 1 <input type="checkbox"/>	Sometimes 1 <input type="checkbox"/>
Not often 2 <input type="checkbox"/>	Not often 2 <input type="checkbox"/>
Not at all 3 <input type="checkbox"/>	Very seldom 3 <input type="checkbox"/>

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Acta Psychiatrica Scandinavica (1983) (67: 361.370)

ID Code:

Flag

1 - Pre	5 - 24 mth
2 - 6 wk	6 - Misc
3 - 6 mth	7 - D/C
4 - 12 mth	

CFS/NHS/PAEDIATRICS

- Specialist help for ME



EQ-5D™

Describing your health today

Under the heading, mark the ONE box that best describes

1.1 Mobility (walking about)	
I have <u>no</u> problems walking about	<input type="checkbox"/> 1
I have <u>some</u> problems walking about	<input type="checkbox"/> 2
I have <u>a lot</u> of problems walking about	<input type="checkbox"/> 3
1.2 Looking after myself	
I have <u>no</u> problems with washing or dressing myself	<input type="checkbox"/> 1
I have <u>some</u> problems with washing or dressing myself	<input type="checkbox"/> 2
I have <u>a lot</u> of problems with washing or dressing myself	<input type="checkbox"/> 3
1.3 Doing Usual Activities (for example, going to school, hobbies, sport, playing, doing things with family or friends)	
I have <u>no</u> problems doing my usual activities	<input type="checkbox"/> 1
I have <u>some</u> problems doing my usual activities	<input type="checkbox"/> 2
I have <u>a lot</u> of problems doing my usual activities	<input type="checkbox"/> 3
1.4 Having pain or discomfort	
I have <u>no</u> pain or discomfort	<input type="checkbox"/> 1
I have <u>some</u> pain or discomfort	<input type="checkbox"/> 2
I have <u>a lot</u> of pain or discomfort	<input type="checkbox"/> 3
1.5 Feeling worried, sad or unhappy	
I am <u>not</u> worried, sad or unhappy	<input type="checkbox"/> 1
I am <u>a bit</u> worried, sad or unhappy	<input type="checkbox"/> 2
I am <u>very</u> worried, sad or unhappy	<input type="checkbox"/> 3

SPENCE CHILDREN'S ANXIETY SCALE

Your Name:

Date:

PLEASE PUT A CIRCLE AROUND THE WORD THAT SHOWS HOW OFTEN EACH OF THESE THINGS HAPPEN TO YOU. THERE ARE NO RIGHT OR WRONG ANSWERS.

1.	I worry about things.....	Never	Sometimes	Often	Always
2.	I am scared of the dark.....	Never	Sometimes	Often	Always
3.	When I have a problem, I get a funny feeling in my stomach.....	Never	Sometimes	Often	Always
4.	I feel afraid.....	Never	Sometimes	Often	Always
5.	I would feel afraid of being on my own at home.....	Never	Sometimes	Often	Always
6.	I feel scared when I have to take a test.....	Never	Sometimes	Often	Always
7.	I feel afraid if I have to use public toilets or bathrooms.....	Never	Sometimes	Often	Always
8.	I worry about being away from my parents.....	Never	Sometimes	Often	Always
9.	I feel afraid that I will make a fool of myself in front of people.....	Never	Sometimes	Often	Always
10.	I worry that I will do badly at my school work.....	Never	Sometimes	Often	Always
11.	I am popular amongst other kids my own age.....	Never	Sometimes	Often	Always
12.	I worry that something awful will happen to someone in my family.....	Never	Sometimes	Often	Always
13.	I suddenly feel as if I can't breathe when there is no reason for this.....	Never	Sometimes	Often	Always
14.	I have to keep checking that I have done things right (like the switch is off, or the door is locked).....	Never	Sometimes	Often	Always
15.	I feel scared if I have to sleep on my own.....	Never	Sometimes	Often	Always
16.	I have trouble going to school in the mornings because I feel nervous or afraid.....	Never	Sometimes	Often	Always
17.	I am good at sports.....	Never	Sometimes	Often	Always
18.	I am scared of dogs.....	Never	Sometimes	Often	Always
19.	I can't seem to get bad or silly thoughts out of my head.....	Never	Sometimes	Often	Always
20.	When I have a problem, my heart beats really fast.....	Never	Sometimes	Often	Always
21.	I suddenly start to tremble or shake when there is no reason for this...	Never	Sometimes	Often	Always
22.	I worry that something bad will happen to me.....	Never	Sometimes	Often	Always
23.	I am scared of going to the doctors or dentists.....	Never	Sometimes	Often	Always
24.	When I have a problem, I feel shaky.....	Never	Sometimes	Often	Always
25.	I am scared of being in high places or lifts (elevators).....	Never	Sometimes	Often	Always

26.	I am a good person.....	Never	Sometimes	Often	Always
27.	I have to think of special thoughts to stop bad things from happening (like numbers or words).....	Never	Sometimes	Often	Always
28.	I feel scared if I have to travel in the car, or on a Bus or a train.....	Never	Sometimes	Often	Always
29.	I worry what other people think of me.....	Never	Sometimes	Often	Always
30.	I am afraid of being in crowded places (like shopping centres, the movies, buses, busy playgrounds).....	Never	Sometimes	Often	Always
31.	I feel happy.....	Never	Sometimes	Often	Always
32.	All of a sudden I feel really scared for no reason at all.....	Never	Sometimes	Often	Always
33.	I am scared of insects or spiders.....	Never	Sometimes	Often	Always
34.	I suddenly become dizzy or faint when there is no reason for this.....	Never	Sometimes	Often	Always
35.	I feel afraid if I have to talk in front of my class.....	Never	Sometimes	Often	Always
36.	My heart suddenly starts to beat too quickly for no reason.....	Never	Sometimes	Often	Always
37.	I worry that I will suddenly get a scared feeling when there is nothing to be afraid of.....	Never	Sometimes	Often	Always
38.	I like myself.....	Never	Sometimes	Often	Always
39.	I am afraid of being in small closed places, like tunnels or small rooms.	Never	Sometimes	Often	Always
40.	I have to do some things over and over again (like washing my hands, cleaning or putting things in a certain order).....	Never	Sometimes	Often	Always
41.	I get bothered by bad or silly thoughts or pictures in my mind.....	Never	Sometimes	Often	Always
42.	I have to do some things in just the right way to stop bad things happening.....	Never	Sometimes	Often	Always
43.	I am proud of my school work.....	Never	Sometimes	Often	Always
44.	I would feel scared if I had to stay away from home overnight.....	Never	Sometimes	Often	Always
45.	Is there something else that you are really afraid of?.....	YES	NO		
	Please write down what it is _____				

	How often are you afraid of this thing?.....	Never	Sometimes	Often	Always

CFS/NHS/PAEDIATRICS - Specialist help for ME.

Assent form for those under 16 years

The psychological impact of CFS/ME on adolescent siblings

Please complete this form if you are under 16 years old. Please initial boxes if “yes”

I have read the leaflet about the study (Version 2, dated 08.03.11).. I understand what the study is about and have had the chance to ask questions.	<input type="checkbox"/>
I understand that it is mine and my parent’s/guardian’s choice about whether or not to take part in the study and that it is ok for me to withdraw from the study at any time.	<input type="checkbox"/>
I understand that relevant sections of data collected during the study may be looked at by regulatory authorities or from the NHS trust, where it is relevant to my taking part in this research. I give permission for these individuals to have access to these data.	<input type="checkbox"/>

If you agree to take part, please sign below:

Your name: Signature:
Today’s date:/...../20..... Today’s date:/...../20.....
Researcher’s name: Signature: Today’s date:/...../20.....

We will give you a copy of this consent form. 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 personal 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 via an ID code on separate lists. The list linking the code will be kept in the University of Bristol with the consent forms.

Older Adolescents consent to study

The psychological impact of CFS/ME on adolescent siblings

Please fill this form if you are between 16 and 17 years old. Please initial boxes if “yes”

I have read the leaflet about the study (version 6, dated 08.03.11). I understand what the study is about and have had the chance to ask questions.	<input type="checkbox"/>
I understand that it is my choice about whether or not to take part in the study and that it is ok for me to withdraw from the study at any time.	<input type="checkbox"/>
I understand that relevant sections of data collected during the study may be looked at by regulatory authorities or from the NHS trust, where it is relevant to my taking part in this research. I give permission for these individuals to have access to these data.	<input type="checkbox"/>

If you agree to take part, please sign below:

Your name:	Signature:
Today's date:/...../20.....	Today's date:/...../20.....
Researcher's name:	
Signature:	Today's date:/...../20.....

We will give you a copy of this consent form. 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 personal 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 via an ID code on separate lists. The list linking the code will be kept in the University of Bristol with the consent forms.

Parental consent form: Sibling Study

The psychological impact of CFS/ME on adolescent siblings

Please initial boxes if “yes”

I have read the leaflet about the study(version 6, dated 08.03.11). I understand what the study is about and have had the chance to ask questions.	<input type="checkbox"/>
I understand that it is mine and my child’s choice about whether or not to take part in the study and that it is ok for my child to withdraw from the study at any time.	<input type="checkbox"/>
I agree that a researcher may visit my child at a place that we choose and interview them for 30 minutes (<i>only complete this if you child is under 16 years old</i>)	<input type="checkbox"/>
I understand that relevant sections of data collected during the study may be looked at by regulatory authorities or from the NHS trust, where it is relevant to my child taking part in this research. I give permission for these individuals to have access to these data.	<input type="checkbox"/>

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

Your name:	Your address:
Signature:
Your e mail:	Your phone number:
Today’s date:/...../20.....	Your child’s name:

We will give you a copy of this consent form. A copy 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 personal 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 via an ID code on separate lists. The list linking the code will be kept in the University of Bristol with the consent forms.

Appendix F: Reminder Letter



Centre for Child & Adolescent Health

School of Social and Community Medicine

University of Bristol

Oakfield House, Oakfield Grove

Bristol

BS8 2BN

Dr Esther Crawley

Email: esther.crawley@bristol.ac.uk

12th March 2011

Dear _____,

The Impact of CFS/ME on adolescent siblings

Thank you for agreeing to take part in this study. As we have not received your filled in questionnaire, we have included a new one just in case you cannot find yours.

Please could you try to fill in the questionnaire and send it back to us as soon as possible? If you have any questions about the questionnaire, or about the study, please ring Dr Esther Crawley or me on 01225 465941. Or email me on sv248@exeter.ac.uk.

We look forward to receiving your questionnaire soon. We are sorry if this letter arrives after you have sent back your questionnaire. You do not need to fill it out again if this has happened.

Best wishes,

Sophie Velleman

Teenage (12-17) consent/assent to interview: Sibling Study

The psychological impact of CFS/ME on adolescent siblings

Please initial boxes if “yes”

I confirm that I consent to being interviewed about my mood, schooling, home life, friends and experience of CFS/ME.	<input type="checkbox"/>
I understand that the interview will be audio-recorded but that I can switch off the recorder or stop the interview without having to give an explanation.	<input type="checkbox"/>
I understand that small parts of what I say may be quoted anonymously when the results of this part of the research are reported.	<input type="checkbox"/>
I confirm that I have had the opportunity to ask any questions about this interview.	<input type="checkbox"/>

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

Your name:	Interviewer’s name:
Signature:	Signature:
Your Child’s name:	
Today’s date:/...../20.....	Today’s date:/...../20.....

We will give you a copy of this consent form. A copy 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 personal 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 via an ID code on separate lists. The list linking the code will be kept in the University of Bristol with the consent forms.

Parental consent to sibling interview

The psychological impact of CFS/ME on adolescent siblings

Please complete this if your child is under 16 years old. Please initial boxes if “yes”

I confirm that I consent to my child being interviewed about their mood, schooling, home life, friends and sibling with CFS/ME.	<input type="checkbox"/>
I understand that the interview will be audio-recorded but that they can switch off the recorder or stop the interview without having to give an explanation.	<input type="checkbox"/>
I understand that small parts of what they say may be quoted anonymously when the results of this part of the research are reported.	<input type="checkbox"/>
I confirm that I and they have had the opportunity to ask any questions about this interview.	<input type="checkbox"/>

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

Your name:	Interviewer’s name:
Signature:	Signature:
Your Child’s name:	
Today’s date:/...../20.....	Today’s date:/...../20.....

We will give you a copy of this consent form. A copy 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 personal 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 via an ID code on separate lists. The list linking the code will be kept in the University of Bristol with the consent forms.

Appendix H: Example Transcript (Participant 4, pages 1-3)

Int: and how are things at home?

P4: [sigh] it's always a little bit difficult because obviously my brother, he's got ME, so quite a lot of life is based around that. But with, we're a very active family; like me and dad go out cycling, we're always just out and about doing stuff, so it's a great family life, I really love it, but it's obviously restricted by what **** can do and, sorry am I not allowed to say his name or...

Int: No, its fine, I'll block it out.

P4: um, you know, it's difficult because he has restriction that he just can't go past, which has limited what we can do as a family, but we get around it.

Int: What kind of things does it limit?

P4: Um, to give an example, we went to Spain, I don't know five years ago and we went for a planned four day walk in the Pyrenees, and obviously there's just no way we could do that now. Yeah it just, [sighs] it's not even, it doesn't even come into the planning because there's no way he can actually cope with it, uuh, we can't go on a family bike ride, family walks, every holiday we go on is based around exercise because we're a very active family. We can't just, you know, sit there a do nothing. We can't just sunbathe on the beach and, it, we've had to kind of change what we do, what we plan to do around ****'s limitations. So, you know, it's difficult.

Int: Mmm, quite a lifestyle change.

P4: mmm, and also just, you know, we still do active things but it's, obviously feel guilty if I let, because we always used to do everything together, me and him, and now we can't. Because I'm a very active person, I do cycling, I do running, I used to play tennis, and we just can't do that together any more. So it makes you kind of feel guilty, you know, every now and again. Like when I learnt to surf this summer, it was one of those things that we were going to do together.

Int: what's the age difference between you two?

P4: Um, its 18 months, so technically, he's two school years ahead of me, so he's second year of sixth form and I'm year 11, so. I'm 15, he's 17, no sorry, I'm 16, he's 17. But he will be 18 soon.

Int: Ok, and what are the kind of, what's the best weekend that you can remember as a family?

P4: what post or?

Int: Post.

P4: Um, don't really know really, they all kind of merge into one. I can't think of a, an individual one. Probably just one where on holiday where we all, because its, it's very difficult because **** has to rest all the time, um, (sighs), I can't really think of an individual one. Oh, my Mum's birthday, that was nice. Um, we went to the Brecon's, and obviously **** couldn't do walking but we hired out a nice um B&B and we did some walks, really short walks as a family, just you know, 100 metres, 200

meters, just to this nice waterfall, and we had nice meals. That was really good, but obviously we had to leave him out when we went off on a day walk, but you know, he managed to get some work done, which was all right.

Int: yeah, A Level year, there must be a lot to do. OK, so who do you live with? Who's at home at the moment?

P4: Um, Dad, Mum, sister, Brother and me. So happy five, I'm the middle child. Um, Dad is a contractor for IT stuff, so he works from home some of the time and then goes in. Mum's a teacher assistant up at the local school. ****'s [sister] in year 8 at, at***** [name of school] over there, and ****'s [brother] obviously in 6th form, doing just a few A-levels.

Int: And is he in the same school as you?

P4: uh no, no he went uh to ***** for, up until year 11, until he got ME, and then took a year off recovering and then passed his GCSEs at home schooling and then went to***** college, whereas I'm at ***** back over at [names local town].

Int: And what, what's a weekend at home like, just a normal weekend at the moment?

P4: ell I do a lot of running, I'm a competitive runner so Saturday morning I train, uh, at the moment we watch rugby on world cup. Uh ****[brother] does work. Um, if he has enough strength we might go up and play tennis together for 15 minutes. Um, I hopefully go out biking. Uh, **** [sister] might, [laughs] I don't know what she does, she meets friends, you know just does, just general homework. It's kind of just resting and we kind of do our own things, but you know manage to eat as a family. We're big eaters, not that you can probably tell but, we cook, we cook all of our own meals, you know. We all sit down on Saturdays and Sundays and eat together in the evenings and that's nice.

Appendix I: NHS Ethics Approval Letter

04 April 2011

Miss S Velleman

Trainee Clinical Psychologist

Taunton and Somerset NHS Trust

DClin Psychology Office

Washington Singer Building

University of Exeter, Exeter

EX4 4QG

Dear Miss Velleman

Study title: Psychological and physical impact of Chronic Fatigue Syndrome/Myalgic encephalopathy on adolescent siblings

REC reference: 11/H0102/10

Thank you responding to the Committee's request for further information on the above research and submitting revised documentation.

The further information was considered in correspondence by a sub-committee of the REC. A list of the sub-committee members is attached.

Confirmation of ethical opinion

On behalf of the Committee, I am pleased to confirm a favourable ethical opinion for the above research on the basis described in the application form, protocol and supporting documentation, subject to the conditions specified below.

Ethical review of research sites

NHS sites

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

Conditions of the favourable opinion

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

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

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

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

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

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

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

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

Approved documents

The final list of documents reviewed and approved by the Committee is as follows:

Document	Version	Date
Covering Letter		
Covering Letter		21 March 2011
Evidence of insurance or indemnity		02 August 2010
Interview Schedules/Topic Guides	4	08 March 2011
Investigator CV		
Letter from Sponsor		11 January 2011
Other: Academic Supervisor CV		
Other: Insurance Certificate		27 July 2010
Other: Reminder Letter	1	12 March 2011
Participant Consent Form: Teenage (12-17) consent to interview: sibling study	3	11 November 2010
Participant Consent Form: Parental Consent to Contact: Sibling Study	4	08 March 2011
Participant Consent Form: Assent Form for under 16s	2	08 March 2011
Participant Consent Form: Parental Consent Form: Sibling Study	3	11 March 2011
Participant Consent Form: Older Adolescents Consent to Study	4	08 March 2007
Participant Consent Form: Parental Consent to Sibling Interview	4	08 March 2011
Participant Consent Form: Teenage (12-17) Consent/Assent to Interview: Sibling Study	3	11 November 2010
Participant Information Sheet	4	11 November 2010
Participant Information Sheet: Younger Adolescents	2	08 March 2011
Participant Information Sheet: Older Adolescents & Parents	6	12 March 2011
Protocol	3	18 March 2011
Questionnaire: Sibling Relationship Questionnaire Revised (child) 3/90	1	10 January 2011
Questionnaire: Spence Children's anxiety scale		
Questionnaire: HADS 1 11 January 2011		
Questionnaire: Demographic Questionnaire	3	11 November 2010

Questionnaire: Sibling Relationship Questionnaire (SRQ)		
REC application	3	11 January 2011
Referees or other scientific critique report		25 October 2010

Statement of compliance

The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees (July 2001) and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.

After ethical review

Now that you have completed the application process please visit the National Research Ethics Service website > After Review

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

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

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

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

We would also like to inform you that we consult regularly with stakeholders to improve our service. If you would like to join our Reference Group please email referencegroup@nres.npsa.nhs.uk.

11/H0102/10

Please quote this number on all correspondence

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

Yours sincerely

Dr David Evans

Chair

Appendix J: University Ethics Approval Letter



SCHOOL of PSYCHOLOGY

ETHICS COMMITTEE

Washington Singer Laboratories

Perry Road

Exeter

EX4 4QG

Telephone +44 (0)1392 264626

Fax +44 (0)1392 264623

Email Marilyn.evans@exeter.ac.uk

To: Sophie Velleman
From: Cris Burgess
CC: Dr Esther Crawley
Re: Application 2010/230 to Ethics Committee
Date: 03 September 2012

The School of Psychology Ethics Committee met recently and your NHS Local Research Ethics Committee application and approval were reviewed. In line with our procedures, your project is now de facto approved.

The agreement of the Committee is subject to your compliance with the British Psychological Society Code of Conduct and the University of Exeter procedures for data protection (<http://www.ex.ac.uk/admin/academic/datapro/>). In any correspondence with the Ethics Committee about this application, please quote the reference number above.

I wish you every success with your research.

Yours sincerely,

A handwritten signature in black ink, appearing to read 'Cris Burgess', with a horizontal line underneath.

Cris Burgess

Chair of School Ethics Committee

Appendix K: Table of interview quotes

Quotes	Participant, page, line
<i>Impact on the Family, Negative Factors, Lack of Communication</i>	
She always says to me, like...she knows what, what I feel like. But I don't like it when she says that cause like, it's a bit different.	P1, 4, 3-4
You'll tell him [Dad] something and he'll, he won't want to talk about it	P1, 5, 4
I don't really explain it cause, cause I don't even know exactly what it means.	P1, 12, 26-27
It kind of makes me quite angry so when I talk about it, it like makes me frustrated, so I don't really talk about it that much, cause.	P1, 12, 29-30
I can talk to my friends about most things, but I don't, I don't really, cause at home like, cause my family and the situation is so confusing, I don't really feel like I, like explaining all of that to them	P1, 12, 5-7
They [parents] didn't really tell me much [about ME]	P1, 12, 36
I kind of didn't really, didn't really want to know, really know much about it...	P1, 13, 24-25
I didn't ask about it	P3, 8, 16
I could get quite frustrated and be like 'why can't you just do this'	P3, 8, 32
I think people do react like that to people that do have it, like, cause they don't understand it	P3, 8, 34-35
Most of his classmates thought he had died, because he wasn't, he didn't go to school for a year	P4, 7, 35-36
It's very difficult to explain it to anybody, because most doctors can't really explain in	P4, 8, 14-15
They don't understand [other people]	P4, 10, 24
They [other people] don't really ask about it	P4, 10, 33
I was never properly told about it	P5, 4, 12-13
She[child with CFS/ME] pushes everyone away	P6, 2, 31
We used to have debates, the kind of, just jokey debates round the table, it's hard to remember that far back	P6, 3-4, 32-1
The meals kind of separated as well	P6, 4, 16-17
So yeah, there are more arguments and, purely because she is more on edge	P6, 4, 33
I still would want to um talk to anyone really, of our age because once you know something is up with someone you kind of want to make sure they are alright and I don't think I'd want that	P6, 9, 4-5
Well some people don't understand what it is really	P7, 7, 3
Near the beginning they were more trying to help out **** so, yeah I sort of felt sort of left out	P7, 8, 9-10
Yeah you kind of feel like there's just a big brick wall and um you can't talk to the school and really explain it to them.	P9, 2, 7-8
They don't really believe it they don't see how serious the condition is.	P9, 2, 22
Sometimes I can't really talk to her [Mum] because it's so, she's had such a hard day	P9, 8-9, 35-1
Well they kind of know but I don't talk to them too much about it, I	P9, 5, 11-13

mean because I don't think they really, I mean, it's not that they don't need to know but they, you know I don't think they would sort of remember it.

The second I mention chronic fatigue they sort of switch off P9, 8, 19

Impact on the Family, Negative Factors, The 'Not Knowing'

With the ME things, she like lets him, she lets him do less and he's older than me P1, 4, 23-24

Like he would say like I've got this and so I can't do this, and he would be telling himself that he can't do this, but really he probably could do a lot of things that he said he couldn't do P1, 4, 26-28

He's just so lazy. Like he'll be on, like on his computer a lot of the time and I think that, I mean...I remember going on my computer once and, and it doesn't help you sleep and it messes up your sleep patterns P1, 6, 3-6

I don't think he's putting it on or anything but I think he was exaggerating a lot of the time P1, 6, 24-25

Whatever time I go to bed, I'll still have to wake up between 6 and half 6, but he, he, and he sort of like had that choice as well of whether to wake up or not, P1, 6, 31-33

Well I didn't feel like sorry for him because he's always been lazy, and I didn't know if it was genuine or not. P1, 6, 49-50

And it's kind of like my brother is sitting on the side, like, not really doing anything. P1, 10, 32-33

It kind of makes me quite angry so when I talk about it, it like makes me frustrated, so I don't really talk about it that much, cause. P1, 12, 29-30

I kind of didn't really, didn't really want to know, really know much about it... P1, 13, 24-25

so it was almost like unfair that, because I go to school that I decided that it was time for me to go to bed and then he just, he just stayed up thinking that, almost like he was kind of cool, just not going to bed, but that's because he could just sleep it off for as long as he wants the next day P1, 15, 43-45

I sort of thought 'oh well she's putting it on' at first [and then] I just thought well yeah this is serious P2, 2-3, 34-3

When I first heard about it, I didn't really know what it was and I wasn't that interested, so I didn't ask about it. P3, 8, 15-16

This is so long term P4, 1, 10

There's no real treatment, proper treatment, to it if you see what I mean P4, 1, 11

Life just decided to throw a rock on him P4, 7, 18

Most of his classmates thought he had died, because he wasn't, he didn't go to school for a year P4, 7, 35-36

I don't think anyone wants to have ME, but I think it's more physical than psychological because my brother so wants not to be ill P4, 8, 23-24

It's very difficult to explain unless you're experiencing it, and seeing as I haven't experienced it, it's taken me a long time to actually understand how it effects you P4, 10, 7-9

Unless you have firsthand experience of what it does to a person, P4, 10, 24-26

and I mean it's different for different people, but unless you have a sibling or a close friend, or, you know a relative, you, you can't really understand	
It just doesn't go away; it stays and stays and you can't do much cause it's like draining...it feels like you have no energy	P5, 4, 14-15
When she was ill...her personality was completely different to how it is now	P5, 4, 24-26
Cause she obviously shouted a lot and I just had, I couldn't react or anything because obviously it was part of the illness	P5, 5, 1-2
When I first heard about it I found it surprising because...it had not come up before and I'd never heard about it	P5, 9, 4-5
I wasn't sure whether I was like, cause it could have been something more serious where she hit her head...because it was ME I wasn't sure if I was like, I hate to say it but relieved	P5, 9, 6-8
I don't know what it is but it just feels like I don't fully understand what it is, obviously because I don't have it I wouldn't understand what it feels like	P5, 10, 3-4
So there are things that we know she can do, but she...knows she can't do them even though she can	P6, 2, 13-14
Because she can't go out, either she can and she doesn't want to, we don't know	P6, 3, 22-23
She has a tendency to strike out at things she thinks is wrong or doesn't fit with her idea of good. She never used to do that	P6, 5, 1-2
She can get along with me fine one day and then it will be hell the next day	P6, 11, 17-18
So the majority of the time she will be grumpy and annoyed easily and...then the odd occasion she's happy, we don't know why	P6, 11, 22-23
There was definitely a difference between when she didn't know what she had and when she knew what she had	P6, 12, 2-3
As soon as she knew what she had it was "leave me alone I've got no energy"	P6, 12, 3-4
She tends to use it as an excuse, and that's the way it looks	P6, 12, 9-10
It looked like she used it to get her way to, to relax or to get some time off	P6, 12, 14-15
She seems to use it as a reason to do things, and she uses it to push people away.	P6, 12, 21-22
And then what I heard from my sister is 'it means I'm always tired, it means I can't do things, it means I'm not allowed to do thing, I have to relax more, I have to rest.'	P6, 12, 32-34
I think the family opinion is very different	P6, 13, 1
Because I don't know much about it and I know she doesn't want anything to change because she likes how it is at the moment. She would probably like to have chronic fatigue for the rest of her life by the looks of it, because she's got everything very easy and doesn't have to work for anything.	P6, 13, 5-8
I tell them to leave her alone, to leave her alone because I'm pretty sure she would like the attention if they went up and asked her how she was. So I try and keep it on the lowdown as I don't want her ego to grow-that sounds so selfish.	P6, 13, 25-27
I find it quite difficult [to explain] cause I don't really fully understand it	P7, 5, 28
Cause Mum couldn't really cope with it. She [sister] was just so	P7, 6, 12-13

upset all the time and she didn't want to do anything.	
We can't plan too far ahead because we don't know if she will be OK	P8, 3, 2-3
Yeah cause it's so change, it changes so often you don't actually know	P8, 7, 5
It's like different all the time	P8, 7, 7
when you describe it [CFS/ME], it doesn't sound as bad as it is	P9, 2, 23
He doesn't really know that, like properly, what chronic fatigue syndrome is	P9, 7, 27
I think if I was him I wouldn't kind of see properly what, how she would be feeling	P9, 7, 28
A lot of people, it takes a lot of explaining to do, and they think you're just tired and lazy	P9, 8, 10-11
Like I said my brother, he, he roughly knows what it is but he doesn't quite understand it as much as I do	P9, 9, 10-11

Impact on the Family, Negative Factors, Restrictions on Family Life

There's been about 3 times or something that maybe he [brother] hasn't gone [to Dads house]. And I, I actually preferred it because those weekends we, we did more,	P1, 8, 2-3
I can't really spend that much time with Mum cause obviously *** can't be left for over an hour and half and that sort of things.	P2, 1, 16-18
, if I have anyone to stay over we tend to stay down here so it's not noisy for Kim, and stuff.	P2, 4, 5-6
My little sister can't do a lot of red activity, um, like it's not busy if you see what I mean	P3, 1, 24-25
Yeah really slow paced [home life]	P3, 1, 27
When I have been home it's been like quite slow	P3, 2, 8
When they go out the more like slow paced things sort of thing, cause obviously my...sister, cant like walk around a lot	P3, 2, 22-23
She goes to bed at 7 and then I can't be in the room cause obviously it wouldn't work	P3, 3, 3
It's become slower, not necessarily more relaxed, because like obviously now there's strict guidelines of what she's allowed to do and what she's not	P3, 3, 10-11
It's harder for them to stay over, like cause obviously because of ****	P3, 5, 17
I think it can be quite restricted	P3, 9, 8-9
If we were like to do something, we have to think before, would we all be able to keep up with that, like have to plan in advance for everything	P3, 9, 9-11
You can't just decide to do things like other people would, cause with her you need to be like 'will she be OK with this'.	P3, 9, 12-13
Obviously my brother, he's got ME, so quite a lot of life is based around that	P4, 1, 25-26
It's a great family life, I really love it, but it's obviously restricted by what **** can do	P4, 1, 27-28
He has restriction that he just can't go past, which has limited what we can do as a family	P4, 1, 30-31
Obviously there's just no way we could do that now	P4, 2, 2
We've had to kind of change what we do, what we plan to do around *****'s limitations. So you know, it's difficult	P4, 2, 6-7

Before we were big campers, big walkers, big cyclers and now we just can't do that	P4, 7, 25-26
If a family isn't very functional to start with I think a family could get completely ripped apart by it.	P4, 11, 27-28
And you can't do anything or go outside because ****'s ill	P5, 3, 19-20
Me, just coming down, wanting to change the channels, she would go absolutely mad	P5, 4, 33-34
We didn't go on holiday, I don't think, when she was properly ill	P5, 10, 18
Whenever I get a break [from revision] I can't really go on the TV because my sister's in there and I can't sit down on the Xbox because my sisters on there, so I have to find something else to do	P6, 5, 8-10
And no one gets out of the house really either	P6, 6, 31
Last weekend...I came into the house on Friday and I didn't leave till Monday morning	P6, 6, 33-34
Everything, everything in the house kind of relates to her somehow	P6, 9, 13-14
She is always on the tv so if someone wants to relax they kind of cant	P6, 1, 23
She is now in there[main sitting room] from when I get home to when I go to bed...so the room is quite often occupied by her	P6, 2, 22-24
She gets annoyed very easily so if I walk in the room while she is watching, or walk through the room and back out again, you can't go back in again or it will annoy her	P6, 2, 26-28
We haven't really been able to do anything as a family anymore because she can't go out	P6, 3, 21-22
That's when we stopped doing things as a family [18 months]...so it's been, it's been a while	P6, 3, 27-29
Things changed, someone has got to be in the house with her	P6, 4, 2-3
Sometimes we had to stay behind because she didn't want to do things	P7, 2, 18-19
When my sister not ill we go out, but when she's ill we cant	P8, 1, 27
If ***** is not well we just stay at home	P8, 2, 20
We can't plan too far ahead because we don't know if she will be OK	P8, 3, 2-3
We haven't been able to do as much. We don't go out as much.	P9, 3, 19
We would walk the dog a lot or go out but **** can't always do that and it might be too much for her, and so we're definitely restricted to what we can do, but we seem to manage	P9, 3, 20-22
Everything's much slower, and stuff like that.	P9, 3, 27-28
She is so limited to what she can do	P9, 8, 21

Impact on the Family, Protective Factors, Positive Communication

I come back from school and then I usually go talk to *** for a bit, me and Mum will sit up there and talk to her	P2, 2, 3-4
We all sit down on Saturdays and Sundays and eat together in the evenings and that's nice	P4, 3, 10-11
If it was a family problem, I'd go to family	P4, 5, 29
Family problems we always sort out as a family	P4, 5, 30-31
He has to be in his mid resting, which means he needs to be in a secure situations, nothing going on around him, in his bed, I-Pod in	P4, 7, 27-29
Probably the most important thing is understanding what situation they are in, talking to them about it, talking with your family, as a	P4, 11, 32-35

whole family with everyone there, saying this is what is happening, let's deal with it together. Because if you're dealing with it on your own, it's a bad plan	
I'd either go to talk to Mum or Dad really, cause they both understood	P5, 5, 10
Dad knew I wouldn't want to be in the house with them [sister and mum] and so he would probably try and get me out the house more	P5, 10, 30-31
Certainly with my Mum [feeling closer] because she talked to us about it.	P6, 3, 7
We usually go shopping on a Sat morning and then on Sundays we normally go out to the woods or something.	P7, 2, 9-10
when she's happy with how much she's doing, they'll ask her to stay 20 minutes longer	P7, 3, 8-9
I would talk to my parents or like another close friend	P7, 5, 8
Just sort of hang out and talk about random stuff	P7, 7, 4
They helped me to understand and stuff, cause after that I understood why they had to help her out	P7, 9, 4-5
Probably talk to my Mum and Dad	P8, 8, 22
[about family] we just had fun, had a laugh	P8, 2, 17
We don't mind talking about it but we don't talk about it loads cause we try and treat her the same	P8, 8, 9-10
It's almost sometimes good to argue because we can sort of settle things and we definitely find that we're all friends at the end.	P9, 3, 11-12
If anyone I would speak to my mum, or even my dad	P9, 5, 17
We've just sort of communicated a bit more and we've sort of let **** feel more relaxed	P9, 9, 28-29

Impact on the Family, Protective Factors, Social Support

When she got ill we actually became a lot closer because she didn't have the energy to argue with me	P2, 5, 2
Obviously they [friends] understand, that it is harder, but they don't mind	P3, 5, 25-26
I tell them [friends] pretty much everything, like we, yeah, we do talk about everything	P3, 5, 28
We get on [sibling], we've always gotten on	P3, 6, 33
I try to be like, really understanding with her and that sort of thing	P3, 7, 10
We all sit down on Saturdays and Sundays and eat together in the evenings, and that's nice	P4, 3, 10-11
If I had a serious problem, it would be Mum and Dad [that I'd go to]	P4, 5, 24-25
Me and my Mum, yeah we have a good relationship	P4, 6, 1-2
I tried to spend time with her as much as I could	P5, 5, 4-5
I'd either go to talk to Mum or Dad really, cause they both understood	P5, 5, 10
I can talk to him [friend] pretty much about anything and he can talk to me about pretty much anything so yeah, it's good	P5, 8, 2-3
Obviously they [friends] have sympathy for her because they know she's ill, and she's been ill for a long time	P5, 8, 13-14
[relationship with sister] like if she wants to she'll come upstairs and play on my playstation with me, or if it's a nice day we'll go outside	P5, 8, 21-22
Yeah I can if I want to [talk to friends] but I don't, not really	P6, 8, 32

I do have a kind of best friend...she knows all about it but she's going to keep it quiet, she always would	P6, 9, 9-10
Sometimes I'll stay with her and then Mum and Dad will go out. But I don't have to stay with her or anything, just sort of keep her company	P7, 2, 20-21
I would talk to my parents or like another close friend	P7, 5, 8
We're quite close [sibling]	P7, 5, 13
My friends were really helpful with that, really understanding	P7, 5, 23-24
We can have a laugh and stuff, and sort of, she's there, if I can't hang out with my friends I'll hang out with her	P7, 7, 20-21
I think it's quite helpful for her, cause obviously she's missed a lot of school so sometimes she doesn't understand things, I'll try and help her	P7, 8, 2-3
We watch TV together and things like that [family]	P8, 2, 26
Well the one who comes round here quite a lot, she knows quite a bit cause she normally comes round when she's ill	P8, 4, 5-6
We get on really well [sister]...we've always got on really well	P8, 5, 24-26
My friends argue a lot more than me and my sister do, they can't believe that we get on as well as we actually do	P8, 6, 9-10
We're definitely quite a strong, we all have quite a strong family bond, and we do quite a lot together.	P9, 3, 7-8
They often come round and we all, I play with my brother and sister, stuff like that, we all sort of have a good time together.	P9, 5, 4-5
One of my friends is sort of, is quite a good family friend and knows about ****'s condition.	P9, 5, 9
I guess it's really my only chance in the week to spend time with **** so that's nice.	P9, 6, 18-19
Yeah we've always been really close.	P9, 6, 33
We're good friends and we don't argue as much and when we do we sort it out and it seems to be OK.	P9, 7, 3-4

Impact on the Family, Protective Factors, Extra Activities

I'm not really home a lot on the weekend cause I go out quite a bit	P3, 7, 2
I usually have my own plans to do different things	P3, 2, 21-22
If I have a bad day either I go out on my road bike, I go mountain biking or I go for a run	P4, 5, 22-23

Impact on the Sibling, Change of Focus/Role

She'll [mum] sometimes get angry at me a bit more than she gets angry at ****	P1, 4, 19-20
Sometimes I feel like I don't have as strong relationship with my Mum as my brother does	P1, 4, 21-22
With the ME things, she like lets him, she lets him do less and he's older than me	P1, 4, 23-24
If he cries, and he actually cries quite a lot for a 16 year old, and if he cried, then without me even getting to explain what happened, I'll be in trouble because it's like, what he says is the truth and I won't be able to say anything.	P1, 5, 32-35
Whatever time I go to bed, I'll still have to wake up between 6 and half 6, but he, he, and he sort of like had that choice as well of	P1, 6, 31-33

whether to wake up or not, I've had to do a lot of things and like grow up quite fast because of how, how he acts.	P1, 13, 26-27
so it was almost like unfair that, because I go to school that I decided that it was time for me to go to bed and then he just, he just stayed up thinking that, almost like he was kind of cool, just not going to bed, but that's because he could just sleep it off for as long as he wants the next day	P1, 15, 43-45
I do feel a little bit more protective of her	P3, 3, 14
I try to be like, really understanding with her and that sort of thing	P3, 7, 10
My sister's like quite dependent on my parents and like, like quite close to my parents and stuff	P3, 7, 22-23
It's become more focussed on **** at home	P4, 3, 29-30
**** needs more attention because of his situation, whereas I'm fine	P4, 3, 38
The focus has changed slightly but not undeservedly	P4, 4, 2
Because, in a way I've found my own voice I suppose	P4, 11, 15
Cause she obviously shouted a lot and I just had, I couldn't react or anything because obviously it was part of the illness	P5, 5, 1-2
I wouldn't say unfair but obviously cause she was ill but yeah it was quite difficult because obviously she's at home and I'm at school	P5, 5, 21-22
Obviously she's in her childhood and the best time for like playing and stuff and she's stuck inside on the sofa	P5, 9, 13-14
I probably did more stuff with Dad cause obviously Mum was looking after her	P5, 10, 25
My sister can reduce my Mum to tears....and she obviously has to talk to someone and Dad's at work	P6, 3, 7-9
They've been more stressed, they've had less time to do things	P6, 4, 6-7.
It will often be when my brother has done something wrong and she, she does not have the energy to deal with it so I have to step in and stop him from doing something he should not be	P6, 11, 12-14
By the time we've usually gone [to school] she's getting up for school	P7, 3, 2
Near the beginning...I sort of felt left out a bit...obviously she was upset and tired and so they were focussing most of their attention on her	P7, 8, 9-11
I have to do a lot more for my sister than I did before	P8, 7, 19
I have to cook for her, make her stuff if she wants anything, stuff like that	P8, 7, 21
Its fine because she's my sister	P8, 7, 23
I realised how much I care for my sister and how much I look out for her and you know make sure she's ok, and the same she does for me.	P9, 1, 11-12
my parents are a little bit stressed out with things like school	P9, 1, 27
But it's incredibly stressful for my parents to have to go through this and it definitely cuts away. I mean they want to, they definitely want to care for **** it's just that, you know, it's a lot, a lot of effort, and its tiring for them and its tiring for ****.	P9, 2, 33-35
I guess I'm much more caring for her than I was. It was always a much more playful sort of brother and sister thing but I, you know, I really look out for her because she's got something that not very nice.	P9, 4, 6-8
She doesn't believe that she's great at art and I'm always telling	P9, 4, 31-32

her she is.	
I would be worried that she wouldn't be able to catch up with stuff like that	P9, 8, 26-27
I want to look out for her really	P9, 8, 31

Impact on the Sibling, Emotional Reaction

It kind of makes me quite angry so when I talk about it, it like makes me frustrated, so I don't really talk about it that much, cause.	P1, 12, 29-30
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He makes me really angry and upset all the time	P1, 5, 31-32
She's going to get better. It doesn't really matter how long it takes	P2, 6, 15-16
Not being able to go out with her any more	P2, 7, 27
It can be quite stressful but at the same time, like, it's not too bad	P3, 1, 23
I could get quite frustrated, or like, not frustrated, but like I wasn't always completely understanding	P3, 8, 29-30
I think now I like do understand a lot more	P3, 9, 23
It's one of those things that you get used to	P3, 1, 16
So it makes you kind of feel guilty, you know, every now and again	P4, 2, 9
We always used to do everything together, me and him, and now we cant	P4, 2, 10
You can't, can't do anything to help them, you can't do anything to fix what their problem is	P4, 3, 24-25
Life's not fair	P4, 11, 10
I would give it all back [positive changes] to have him there again, to be honest	P4, 11, 15-16
We just don't really have anything in common, because the things that we had in common was active, active, being active.	P4, 11, 22-23
As it moved on and time went by and she was ill, it got more and more...upsetting in a way	P5, 9, 12-13
But she was ill, so it sort of limited me as well because, obviously going out by yourself and trying to do things, it's not as fun as when you have two people	P5, 10, 12-14
It doesn't seem easy to have a sibling who has something not quite right with them	P6, 1, 7-8
Parents tend to be a lot more agitated	P6, 1, 25
If she's upset it kind of, the house kind of goes into this upset kind of thing	P6, 2, 4-5
Everything is stressful really	P6, 5, 11
Things are stressful everywhere	P6, 6, 6
There's a lot of tension in the morning	P6, 8, 8
I worry more than I should	P6, 10, 14
I don't like it when she's in pain or like have to go to hospital and stuff like that	P8, 6, 19
It's definitely not as great as it was a few years ago	P9, 3, 22
, and for me it's just sort of been worrying	P9, 3, 25
You know, knowing that she's not very well and its, ME is a horrible thing to go through. It's really; it's just like a slow and painful thing.	P9, 3, 25-27
, it's quite hard, it's quite hard because she's so ill and I haven't been able to do as much things with her, but um	P9, 4, 4-5
When she first got diagnosed it wasn't, I wasn't really that happy	P9, 7, 33-34
I knew what I had been through and to see her having to go	P9, 7, 34-35

through all of that

Impact on the Sibling, Social Impact

I can talk to my friends about most things, but I don't, I don't really, cause at home like, cause my family and the situation is so confusing, I don't really feel like I, like explaining all of that to them	P1, 12, 5-7
It's harder for them to stay over, like cause obviously because of ****	P3, 5, 17
I think lots of times people don't take ME seriously	P3, 8, 8-9
I think people do react like that to people that do have it, like, cause they don't understand it	P3, 8, 34-35
I guess they couldn't imagine what having ME's like	P3, 8, 37-38
We, you know, just don't really have anything, anything that we share anymore because of the illness, so...it's horrible.	P4, 11, 23-24
I'm not sure too many of them know, know about her	P5, 8, 5
I still would want to um talk to anyone really, of our age because once you know something is up with someone you kind of want to make sure they are alright and I don't think I'd want that	P6, 9, 4-5
I don't think I would want a regular conversation about how things are going at home	P6, 9, 5-6
And someone else would hear and someone else would use it	P6, 9, 6-7
I tell them to leave her alone, to leave her alone because I'm pretty sure she would like the attention if they went up and asked her how she was. So I try and keep it on the lowdown as I don't want her ego to grow-that sounds so selfish.	P6, 13, 25-27
Well some people don't understand what it is really	P7, 7, 3
But not all my friends know	P8, 4, 6
I don't think it's fair on my sister is everyone knows	P8, 4, 8
Well they kind of know but I don't talk to them too much about it, I mean because I don't think they really, I mean, it's not that they don't need to know but they, you know I don't think they would sort of remember it.	P9, 5, 11-13
I try to keep it in the family because I just don't want to, sort of, get blackmailed or anything	P9, 5, 21
The second I mention chronic fatigue they sort of switch off	P9, 8, 19

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Appendix N: Reflexivity

When reflecting on my experiences whilst completing this research, three main areas come to mind: Qualitative vs quantitative; sibling relationships; positivity.

Quantitative vs. Qualitative

I decided that I wanted to conduct a mixed-methods research project because I feel that both disciplines are important areas for research, and allow us to gain a fuller picture of what is occurring. However, I am aware that although I know the quantitative information gives us valid and important information, it is with the qualitative interviews that I believe that one really begins to understand more about the individual experience of the siblings. This is the area where you can explore the true impact of CFS/ME on siblings, as although the quantitative information can tell us that siblings are generally more anxious, but no higher percentage of those exceeding the cut-off, it is the qualitative interviews that can explore the question of *why*.

Sibling relationships

I was also aware of my relationship with my sister; a relationship that used to be very acrimonious, but which has calmed down over the past 2 years. I now have a very close relationship with my sister. I noticed a number of times in the interviews that comments said by the participants reminded me of my relationship with my sister, and that difficult sibling relationships reminded me of how our relationship used to be. This may mean the extracts regarding 'siblings' relationships' were more apparent to me.

Positivity

I was aware that I was hopeful that there might be some positive impacts or resilience found in these siblings. Due to this, I was very careful throughout the interviews to ensure I asked

balanced questions. We also had a second qualitative researcher read through and code the transcripts to ensure my views did not seep into the research findings.

Appendix N

Inventories for quantitative analysis

Hospital Anxiety and Depression Scale (HADS), (Zigmond & Snaith, 1983). The HADS was used to assess anxiety and depression symptoms in the siblings. The HADS consists of 14 items, rated on a 4-point Likert scale (“definitely as much” to “hardly at all”), and has two subscales: “anxiety” (“I feel tense and wound up”), and “depression” (“I feel as if I am slowed down”). This questionnaire is valid and reliable as a screening tool in adolescents (White, Leach, Sims, Atkinson, & Cottrell, 1999). The cut off scores differ slightly from adolescent use to adult use. In adolescents the most suitable cut off for anxiety was scores 9-11 indicating possible emotional disorder and above 11 indicating probable emotional disorder (White et al., 1999), which is higher than the cut off for anxiety in adults (Zigmond et al., 1983). For the depression subscale, scores of 7-9 indicate possible depression and scores above 9 indicate probable depression (White et al., 1999), which are lower than the cut offs for adults (Zigmond et al., 1983).

Sibling Relationship Questionnaire (SRQ) (Furman & Buhrmester, 1985). The SRQ is a 48 item questionnaire, measuring 16 dimensions of sibling relationship contributing to four scales: warmth/closeness (e.g., “How much do you and this sibling tell each other everything”), conflict (e.g., “How much do you and this sibling insult and call each other names?”), relative status/power (e.g., “How much do you tell this sibling what to do?”), and rivalry (“Who usually gets treated better by your father, you or this sibling?”). Potential ranges for the factor scores are 21-105, 9-45, -24-24 and 0-24 respectively. Higher scores on warmth and closeness and conflict scales indicates higher perceptions on these characteristics. A positive Status and Power suggests the child helps, teaches and controls the sibling more than she/he is helped, taught and controlled by the sibling. Higher scores on

Rivalry indicate more favouritism. Internal consistency was high for all factors (Cronbach's alpha $>.70$), and test-retest reliability was also high (mean $r = .78$) (furman et al., 1985).

Spence Children's Anxiety Scale (SCAS) (Spence, 1998). The SCAS is a 44 item self-report measure that measures the frequency of anxiety symptoms experience by a child in the following domains: generalised anxiety, separation anxiety, social phobia, panic-agoraphobic, obsessive-compulsive (OCD) and phobia of physical injury. Each item is rated on a 4 point Likert scale ("never" to "always"). The SCAS was originally designed for children younger than 13 years of age (Spence, 1998), but has more recently found to be reliable and valid with adolescents (Spence, Barrett, & Turner, 2003).

European Quality of life- Youth (EQ-5D-Y) (Wille et al., 2010). EQ-5D-Y is a five item standardised instrument for use as a measure of health outcome. It is applicable to a wide range of health conditions, and provides a simple descriptive profile and single index value for health status. The EQ-5D-Y has been found to be feasible, reliable and valid in child and adolescent populations (Ravens-Sieberer et al., 2010). This questionnaire is also currently being used on all patients within the local CFS/ME service.

Statistical Analysis

Data were analysed using Stata v12 (StataCorp, College Station, TX, USA). Characteristics of children with CFS/ME whose siblings were or were not recruited for this study were compared using Student's t test for comparison of means, Mann-Whitney test for comparison of medians and Chi-squared test for comparison of proportions. Siblings' mean scores on HADS and SCAS were compared to community norms using t tests. The

percentage of siblings whose total anxiety score on SCAS and total anxiety and depression score on HADS were in the upper 10th percentile were compared to the expected proportion in the population using one-sample binomial tests. Pearson correlation coefficients were calculated for sibling SRQ, HADS, SCAS and EQ5D scores to look for relationships between these factors.

A power calculation was completed on the maximum sample size permitted by the time and resources available, N=40. This sample size gives 80% power (two-sided P=0.05) to detect differences between the two groups of: 10 points on the SCAS (scored 0 - 88), 2 points each on the HADS depression and anxiety sub-scales (each scored 0 - 21).

Results

Quantitative Results

Descriptive Statistics. Siblings were recruited between July 2011 to April 2012. During this period, 302 new follow-up children¹² were seen. Of this number, 91 were eligible, and 34 parents were approached during a follow-up appointment by the clinician. Ten did not return the consent to contact forms, and twenty-four signed consent-to-contact forms were returned to the researcher. Of these, twenty-three agreed to be sent the questionnaire pack (11 female), and of those eighteen questionnaires were returned (9 female). Two of the siblings came from the same family. Eleven of the siblings, who returned their questionnaire, stated that they would like to take part in the interview (6 females). Nine of those siblings were subsequently interviewed (5 females). The interviewer (first author) was a 28 year old, white, female, psychologist in training.

Two index children with CFS/ME had missing data for fatigue and SCAS, and one index child was missing data for pain. To determine whether those recruited were representative of

¹² Most of these children were seen on multiple occasions, but each follow-up child was counted only once.

the clinic cohort, we compared the index children with CFS/ME, to all the children attending an initial assessment appointment, aged between 8-18 years (Table 1) within the last 6 years¹³. The siblings recruited to this study had an index child who was similar to the cohort apart from being slightly younger (mean age index child 12.8 years (SD=3.1), mean age cohort 14.2 years (SD=2.1)).

The sibling (n=18, female=9) mean scores for the HADS and SCAS were compared to community norm samples (n=248, female=110, (White et al., 1999) and (n=1011, female=487) (Muris, Schmidt, & Merckelbach, 2000) respectively (Table 2 and 3). The siblings' mean scores on the SCAS were significantly higher than the normative sample's mean score ($p < 0.001$). However the percentage of children who exceeded the cut-off for anxiety on the SCAS did not differ significantly between the sibling and the normative sample group. The mean HADS anxiety and depression scores were similar in the siblings and community sample. The siblings did not significantly differ from the normative sample in the percentage of children who exceeded the cut off for depression and anxiety on the HADS. The percentage of siblings who scored above the cut off on the HADs depression scale was the same as the normative sample (13%). However, the percentage of siblings who scored above the cut off on the HADS anxiety scale was 50% compared to 33% in the normative sample ($p = 0.18$).

¹³ The index children from this study had attended and initial assessment appointment since 2006.

Table 2

Comparison of depression with German school population (n=248) (White et al., 1999) and anxiety with school population from The Netherlands (n=2559) (Muris et al., 2000) and German school population (N = 248) (White et al., 1999) with siblings of children with CFS/ME.

	Siblings (N=18)		Normative Sample	Number > Cut- off*	% of(95% CI) exceeding cut off in sibling sample	% of(95% CI) exceeding cut off in normative sample	P -values
	N	Mean (SD)	Mean (SD)				
HADS Anxiety *	16	7.5 (5.1)	7.2	8	50% (25 – 75%)	33% (28-40%)	0.18
HADS Depression *	16	3.1 (3.0)	3.4	2	13% (2 – 38%)	13% (9-18%)	1.00
SCAS **	16	27.1 (19.1)	16.56 (11.68)***	6	37.5% (15-65%)	31% (22-40%)	0.72

*HADS anxiety cut-off was >8, HADS depression cut-off was >6 (White et al., 1999)

**SCAS cut off was >25 for males and > 36 for females (Muris et al., 2000)

*** Significant difference in means (p<0.001)

The siblings' EQ5D scores were similar to those from a community German school sample. However, more siblings reported 'no pain or discomfort' in comparison to a community South African school sample ($p < 0.05$). A larger proportion of the sibling sample stated that they were 'a bit' sad worried or unhappy (50%) compared to the German (35.8%) or South Africans (34.1%) samples ($p = 0.41$ and $p = 0.37$ respectively).

Table 3

Comparison of Sibling group with a community German sample, and the sibling group with a community South African sample (Ravens-Sieberer et al., 2010)

	Siblings N (%)	Germany N (%)	P values (Siblings vs. Germany)	South Africa N (%)	P Values (Siblings vs. South Africa)
Mobility			P= 0.34		P= 0.84
No	15 (83.3%)	695 (91.9%)		220 (85.3)	
Some of the time	3 (16.7%)	57 (7.5%)		31 (12.0)	
A lot of the time	0 (0.0%)	1 (0.1%)		1 (0.4)	
Looking after myself			P=0.84		P=0.75
No	18 (100%)	740 (97.9%)		245 (95.0)	
Some of the time	0 (0%)	12 (1.6%)		7 (2.7)	
A lot of the time	0 (0%)	2 (0.3%)		1 (0.4)	
Doing usual activities			P=0.97		P=0.49
No	17 (94.4%)	705 (93.3)		213 (82.6)	
Some of the time	1 (5.6%)	47 (6.2%)		38 (14.7)	
A lot of the time	0 (0%)	2 (0.3%)		2 (0.8)	
Pain or discomfort			P=0.20		P=0.032
No	13 (72.2%)	469 (62.0%)		133 (51.6)	
Some of the time	4 (22.2%)	272 (36.0%)		118 (45.7)	
A lot of the time	1 (5.56%)	10 (1.3%)		2 (0.8)	
Feeling sad, worried or unhappy			P=0.41		P=0.37
Not	8 (44.4%)	452 (59.8%)		155 (60.1)	
A bit	9 (50.0%)	271 (35.8%)		88 (34.1)	
Very	1 (5.56%)	29 (3.8%)		10 (3.9)	

Pearson correlations were calculated for the SRQ factor scores, the HADS anxiety, HADS depression, SCAS and EQ5D scores (Table 4). Unsurprisingly the EQ5D was significantly negatively correlated to the HADS depression and anxiety scores and the SCAS scores ($p < 0.01$), indicating a negative relationship between siblings Quality of Life and their depression and anxiety. The SCAS was also significantly positively correlated to the HADS anxiety and depression score ($p < 0.01$). The Conflict scale of the SRQ was significantly negatively correlated to the Warmth and Closeness scale ($p < 0.05$). The Status/Power scale also had a negative relationship with the Warmth and Closeness scale, which was nearing significance ($p = 0.056$). The Rivalry Scale was also significantly positively related to the HADS depression scores ($p < 0.01$) and to the SCAS ($p < 0.05$).

Table 4

Pearson Correlations of SRQ factor scores, HADS anxiety, HADS depression, Spence's Anxiety Scale and EQ5D in siblings

	Factor Warmth/ closeness	Factor status/ power	Factor Conflict	Factor rivalry	HADS anxiety	HADS depression
Factor Warmth/ Closeness	1.00					
Factor status/power	-0.458 P=0.056	1.00				
Factor Conflict	-0.551 P=0.017*	-0.020 P=0.94	1.00			
Factor rivalry	-0.017 P=0.947	0.050 P=0.843	-0.404 P=0.096	1.00		
HADS anxiety	-0.304 P=0.252	0.183 P=0.498	0.449 P=0.081	0.343 P=0.193	1.00	
HADS Depression	-0.106 P=0.696	-0.042 P=0.876	0.492 P=0.053	0.831 p=0.000**	0.679 P=0.004**	1.00
Spence's Anxiety Scale	-0.052 P=0.843	0.139 P=0.596	0.277 P=0.281	0.499 p=0.041*	0.8309 P=0.000**	0.763 P=0.000
eq5d	0.089 P=0.724	-0.088 P=0.728	-0.354 P=0.149	-0.271 P=0.277	-0.717 P=0.002**	-0.886 P=0.000

*p<0.05 ** P<0.01