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# Psychological wellbeing and quality-of-life among siblings of paediatric CFS/ME patients: A mixed-methods study

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## Abstract

Chronic fatigue syndrome or myalgic encephalomyelitis (CFS/ME) is a disabling condition known to have a negative impact on all aspects of a child's life. However, little is understood about the impact of CFS/ME on siblings. A total of 34 siblings completed questionnaires measuring depression (Hospital Anxiety and Depression Scale (HADS)), anxiety (HADS and Spence Children's Anxiety Scale (SCAS)) and European Quality-of-life-Youth (EQ-5D-Y). These scores were compared with scores from normative samples. Siblings had higher levels of anxiety on the SCAS than adolescents of the same age recruited from a normative sample; however, depression and quality-of-life were similar. Interviews were undertaken with nine siblings of children with CFS/ME who returned questionnaires. Interview data were analysed using a framework approach to thematic analysis. Siblings identified restrictions on family life, 'not knowing' and lack of communication as negative impacts on their family, and change of role/focus, emotional reactions and social stigma as negative impacts on themselves. They also described positive communication, social support and extra activities as protective factors. Paediatric services should be aware of the impact of CFS/ME on the siblings of children with CFS/ME, understand the importance of assessing paediatric CFS/ME patients within the context of their family and consider providing information for siblings about CFS/ME.

## Keywords

siblings, chronic fatigue syndrome, myalgic encephalomyelitis, CFS/ME, psychological wellbeing, quality-of-life

## Introduction

Chronic fatigue syndrome or myalgic encephalomyelitis (CFS/ME) has a negative impact on a child's schooling (Crawley, Emond, & Sterne, 2011; Crawley & Sterne, 2009; Rangel, Garralda, Levin, &

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Roberts, 2000; Sankey, Hill, Brown, Quinn, & Fletcher, 2006), their social relationships (Bell, Jordan, & Robinson, 2001) and their parents' finances and quality-of-life (Missen, Hollingworth, Eaton, & Crawley, 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). National Institute for Health and Clinical Excellence (NICE, 2007) guidelines recommend that a diagnosis is made after symptoms have been present for 3 months. Paediatric CFS/ME is relatively common with an estimated prevalence of 0.2–2.4% (Chalder, Goodman, Wessely, Hotopf, & Meltzer, 2003; Crawley et al., 2011, 2012).

Children may experience positive and negative consequences if they have a sibling with a chronic health condition (Areemit, Katzman, Pinhas, & Kaufman, 2010; Batte, Watson, & Amess, 2006; Hames & Appleton, 2009; Petalas, Hastings, Nash, Dowey, & Reilly, 2009; Read, Kinali, Muntoni, & Garralda, 2010; Sharpe & Rossiter, 2002; Vermaes, van Susante, & van Bakel, 2012; Williams, 1997). The negative consequences for siblings include both internalising (e.g. depression and anxiety) and externalising problems (e.g. behaviour, social and aggression problems) (Sharpe & Rossiter, 2002; Vermaes et al., 2012). They may experience negative impacts on family life (feelings of exclusion and lack of attention from parents) and their social life (negative public perceptions and impact on peer relationships) (Areemit et al., 2010; Batte et al., 2006; Hames & Appleton, 2009; Petalas et al., 2009; Read et al., 2010). These findings from the sibling studies are consistent with the views of parents of children with CFS/ME who worry that siblings experience anger and frustration, which may be directed at the unwell child (Missen et al., 2011). Benefits of having a sibling with a chronic health condition include increased empathy and personal growth, increased compassion and caring, and increased family cohesion, relationships, maturity and independence (Areemit et al., 2010; Batte et al., 2006; Hames & Appleton, 2009; Petalas et al., 2009; Read et al., 2010; Williams, 1997).

However, the findings may be very different for siblings of children with CFS/ME because it is a long-term stigmatising illness and little is known about it (Beasant, Mills, & Crawley, 2014). One study was previously published exploring the experience of siblings of children with CFS/ME (Jackson, 1999). She found that siblings had worries including 'apparent parental dilution of care or concern', 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. However, this study did not report number of siblings participating in the study, how the siblings were recruited, the methodology used in the study or go into detail about what the themes stated above meant. We used a mixed-methods approach to understand the impact of paediatric CFS/ME on siblings, to investigate whether any factors are psychologically protective and to discover how services should support siblings of young people with CFS/ME.

## Method

### Design

We collected quantitative data on siblings' psychological wellbeing and quality-of-life to enable comparisons with community normative data. We collected qualitative data by interviewing a sample of siblings to gain a greater understanding of individual experiences.

### Participants

Siblings of children with CFS/ME were recruited prospectively between July 2011 and July 2013 from a National Health Service (NHS) specialist paediatric CFS/ME service. This service covers a

region in the south west of England, which has a population of around 400,000 children, aged 5–19 years. In addition, the service offers assessment and treatment to children from outside the region who do not have access to 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 (referred to as the ‘index’ child) was attending a follow-up appointment and was aged between 8 and 18 years, and if the sibling was aged between 12 and 17 years and 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, to permit the researcher to contact the family at home to tell the parents and sibling(s) about the study. The researcher phoned the family and spoke to the parent(s) and the sibling (if interested). If the sibling and parent both agreed, the researcher sent out the questionnaire pack and consent forms with a pre-paid envelope. We used a convenience sample for the qualitative part of the study and interviewed the first nine siblings who consented to participate.

### Measures

The Hospital Anxiety and Depression Scale (HADS) (Zigmond & Snaith, 1983) and the Spence Children’s Anxiety Scale (SCAS) (Spence, 1998) are routinely collected for young people with CFS/ME who attend the Royal National Hospital for Rheumatic Diseases (RNHRD) service. These questionnaires were completed by siblings, in addition to the European Quality-of-life-Youth (EQ-5D-Y) quality-of-life questionnaire (Wille et al., 2010).

**HADS.** The HADS comprises 14 items, each rated on a 4-point Likert scale (‘definitely as much’ to ‘hardly at all’), and has two sub-scales: ‘anxiety’ (e.g. ‘I feel tense and wound up’) and ‘depression’ (e.g. ‘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). In adolescents, the most suitable cut-offs for anxiety are scores of 9–11 indicating possible emotional disorder and >11 indicating probable emotional disorder (White et al., 1999). These cut-offs are higher than the cut-off for anxiety in adults (Zigmond & Snaith., 1983). For the depression sub-scale, scores of 7–9 indicate possible depression and scores of >9 indicate probable depression (White et al., 1999). The HADS normative data used in this study were from a German population as no UK normative data were available (White et al., 1999).

**SCAS.** The SCAS is a 44-item self-report measure that measures the frequency of anxiety symptoms experienced by a child in the following domains: generalised anxiety, separation anxiety, social phobia, panic-agoraphobic, obsessive–compulsive disorder (OCD) and phobia of physical injury. Each item is rated on a point Likert scale (‘never’ to ‘always’). The SCAS has been found to be reliable and valid in adolescents (Spence, Barrett, & Turner, 2003). The normative data used for this study came from an adolescent population from the Netherlands (Muris, Schmidt, & Merckelbach, 2000) as there were no UK normative data available.

**EQ-5D-Y.** EQ-5D-Y is a 5-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 South African and German populations (Ravens-Sieberer et al., 2010). There are no UK normative data for this questionnaire.

### *Statistical analysis*

Data were analysed using Stata (StataCorp, College Station, TX, USA). Characteristics of young people 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 with community norms using Student's *t* tests. Reliability coefficients (Cronbach's alpha) for each instrument used in the sibling sample were as follows: HADS anxiety, .84; HADS depression, .73; SCAS, .94; EQ-5D-Y, .71.

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 with the expected proportion in the population using one-sample binomial tests.

### *Qualitative semi-structured interviews*

All siblings who returned their questionnaire were asked whether they wanted to participate, and we interviewed the first nine siblings who provided consent. Siblings were interviewed using a semi-structured interview schedule to explore the impact their siblings' CFS/ME had on them. Interviews were digitally recorded and transcribed. The interviews were analysed using a framework approach to thematic analysis (Braun & Clarke, 2006) to identify issues important to the participants, rather than those thought to be important by the researchers.

The transcripts of the first five interviews were read and reread by two researchers (S.V. and L.B.), who discussed the similarities and differences between the codes before agreeing to a final set of codes. 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*

The study received approval from the local NHS research ethics committee (11/H0102/10, 4 April 2011).

## **Results**

### *Descriptive statistics*

During the recruitment period, 653 young people with CFS/ME<sup>1</sup> were seen for the first time in follow-up. Of these, 162 (24.8%) were eligible. A total of 59 parents were approached, and 48 families consented to be contacted by the researcher. In these 48 families, there were 55 siblings. Of these, 35 (63.6%) returned the questionnaire packs. Only three families had more than one sibling who completed the questionnaires. Out of 35 returned questionnaires, 11 siblings stated that they would like to take part in an interview of whom the first 9 (5 females, 4s male) were subsequently interviewed.

The recruited siblings were aged 12–17 (mean = 14.7, standard deviation (*SD*) = 1.4) years, and 15 (44.1%) were female. To determine whether the 32 'index' young people (patients whose siblings were recruited) were representative of the clinic cohort, we compared their characteristics with the

**Table 1.** Characteristics of children with CFS whose siblings were recruited to this study compared with all other children with CFS/ME assessed by the specialist service.

	Children with CFS/ME whose siblings were recruited ( <i>n</i> = 32 unless otherwise indicated)	All other children with CFS/ME ( <i>n</i> = 692 unless otherwise indicated)	<i>p</i> -value <sup>a</sup>
	Mean (SD)	Mean (SD)	
Age (years)	12.7 (2.7)	13.8 (2.5)	.02
Female	22 (68.8%)	487/685 (71.1%)	.78
	Median (IQR)	Median (IQR)	
Time to assessment (months)	12 (7–18), <i>n</i> = 27	12 (8–24), <i>n</i> = 579	.16
Chalder Fatigue Score (0–33)	25 (23–28), <i>n</i> = 29	25 (21–28), <i>n</i> = 652	.57
SF-36 physical function (0–100)	50 (39–60), <i>n</i> = 30	55 (35–70), <i>n</i> = 640	.40
Anxiety (SCAS) (0–90)	25 (17–37), <i>n</i> = 30	29 (18–44), <i>n</i> = 623	.29
No. of symptoms (0–14)	9 (7–9.5)	9 (7–10), <i>n</i> = 687	.63
Anxiety (HADS) (0–21) <sup>b</sup>	8 (6–10), <i>n</i> = 21	9 (6–12), <i>n</i> = 525	.58
Depression (HADS) (0–21) <sup>b</sup>	7 (4–9), <i>n</i> = 21	7 (5–10), <i>n</i> = 526	.44
Visual analogue pain	54 (24–78), <i>n</i> = 30	52 (23–71), <i>n</i> = 619	.28
School attendance past week	<i>n</i> (cumulative %)	<i>n</i> (cumulative %)	
0–40%	15 (48.4%)	329 (51.0%)	.78
60–100%	16 (51.6%)	316 (49.0%)	
Total	31 (100.0%)	645 (100.0%)	

SD: standard deviation; CFS/ME: chronic fatigue syndrome or myalgic encephalomyelitis; IQR: inter-quartile range; HADS: Hospital Anxiety and Depression Scale; SCAS: Spence Children's Anxiety Scale.

<sup>a</sup>Student's *t* test for comparison of means and Kruskal–Wallis test for comparison of medians, Chi-squared test for comparison of proportions.

<sup>b</sup>HADS was completed only by patients aged  $\geq 12$  years.

characteristics of all young people who attended an initial assessment appointment during the period 1 April 2006 to 31 March 2013 who were also diagnosed with CFS/ME and who were from the same postcode areas (Table 1). The following characteristics were similar between the two groups: percentage of females, time to assessment, fatigue scores, physical functioning, number of symptoms, school attendance anxiety, depression and pain. However, index children were slightly younger (difference = 1.1 (95% confidence interval (CI) = [0.1–2.1]) years). Three index children with CFS/ME had missing data for fatigue and two for the anxiety scales (SCAS), and two index children were missing data for pain.

**Comparison of sibling mood and quality of life with control data from community samples.** We compared sibling scores for the HADS and SCAS with data from healthy community samples (Muris et al., 2000; White et al., 1999) (Tables 2 and 3). The healthy community sample for the SCAS had a similar age range (13–19 years), approximately 50% were girls and there was similar ethnic origin (95% White Caucasian). The healthy community sample for the HADS also had similar age range (12–16 years), and 44% were female. There was no information about ethnic origin (White et al., 1999). Table 2 shows that the mean HADS depression and anxiety scores were similar in the siblings and normative sample.

**Table 2.** Comparison of HADS anxiety and depression scores in German school population ( $N=248$ ; White, Leach, Sims, Atkinson, & Cottrell, 1999) with scores in siblings of children with CFS/ME ( $N=32$ ).

	Siblings Mean (SD)	Normative sample Mean (SD)	Proportion [95% CI] exceeding cut-off in sibling sample <sup>a</sup>	Proportion [95% CI] exceeding cut-off in normative sample <sup>a</sup>	<i>p</i> -value <sup>b</sup>
HADS anxiety <sup>a</sup>	6.75 (4.19), $n=32$	7.2, $n=248$	2/32=6.3% [0.8–20.8%]	35/249=14.1% [10.0–19.0%]	.52
HADS depression <sup>a</sup>	3.09 (2.74), $n=32$	3.4, $n=249$	1/32=3.1% [0.1–16.2%]	5/248=2.0% [0.1–4.6%]	.28

HADS: Hospital Anxiety and Depression Scale; CFS/ME: chronic fatigue syndrome or myalgic encephalomyelitis; SD: standard deviation; CI: confidence interval.

<sup>a</sup>HADS anxiety cut-off > 11, HADS depression cut-off > 9 (White et al., 1999).

<sup>b</sup>Fisher's exact test for proportions comparing sibling sample with normative sample.

**Table 3.** Comparison of anxiety scores according to age and gender with cut-off scores described by Muris, Schmidt, and Merckelbach (2000) defining the 90th centile (top 10%) of the normal population.

	N	Mean (SD)	Comparison with normal population			<i>p</i> -value <sup>a</sup>
			Cut-off score	Number >90th centile	% [95% CI] exceeding 90th centile	
<b>Total anxiety score</b>						
Boys	18	17.9 (11.6)	25	5	28% [10–53%]	.03
Girls	15	30.1 (18.5)	36	3	20% [4–48%]	.18
<b>Generalised anxiety</b>						
Boys	19	4.5 (3.0)	6	5	26% [9–51%]	.04
Girls	15	6.7 (3.7)	9	2	13% [2–40%]	.45
<b>Separation anxiety</b>						
Boys	19	2.1 (1.5)	4	0	0%	1.00
Girls	15	2.9 (2.6)	5	5	33% [12–62%]	.01
<b>Social phobia</b>						
Boys	19	5.3 (3.4)	6	8	42% [20–67%]	<.001
Girls	15	8.1 (3.9)	8	6	40% [16–68%]	.002
<b>Panic-agoraphobic</b>						
Boys	19	2.2 (2.0)	4	2	11% [1–33%]	.58
Girls	15	4.9 (4.8)	6	3	20% [4–48%]	.18
<b>Obsessive-compulsive</b>						
Boys	19	2.7 (2.6)	5	4	21% [6–46%]	.11
Girls	15	4.4 (3.6)	6	3	20% [4–48%]	.18
<b>Physical injury fears</b>						
Boys	19	1.9 (1.8)	5	0	0%	1.00
Girls	15	4.0 (2.8)	6	4	27% [8–55%]	.06

SD: standard deviation; CI: confidence interval.

<sup>a</sup>From comparison with expected proportion (10%) using one-sample binomial test.

**Table 4.** Comparison of sibling group with community German (Ravens-Sieberer et al., 2010) and South African (Ravens-Sieberer et al., 2010) samples.

	Siblings n (%)	Germany n (%)	p-values (siblings vs Germany) <sup>a</sup>	South Africa n (%)	p-values (siblings vs South Africa) <sup>a</sup>
Mobility			$p = .002$		$p = .81$
No	31 (91.2%)	695 (91.9%)		220 (85.3)	
Some of the time	3 (8.8%)	57 (7.5%)		31 (12.0)	
A lot of the time	0 (0.0%)	1 (0.1%)		1 (0.4)	
Looking after myself			$p = 1.00$		$p = 1.00$
No	34 (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			$p = .74$		$p = .12$
No	33 (97.1%)	705 (93.3)		213 (82.6)	
Some of the time	1 (2.9%)	47 (6.2%)		38 (14.7)	
A lot of the time	0 (0%)	2 (0.3%)		2 (0.8)	
Pain or discomfort			$p = .02$		$p = .001$
No	28 (82.4%)	469 (62.0%)		133 (51.6)	
Some of the time	5 (14.7%)	272 (36.0%)		118 (45.7)	
A lot of the time	1 (2.9%)	10 (1.3%)		2 (0.8)	
Feeling sad, worried or unhappy			$p = .95$		$p = .89$
Not	20 (58.8%)	452 (59.8%)		155 (60.1)	
A bit	13 (38.2%)	271 (35.8%)		88 (34.1)	
Very	1 (2.9%)	29 (3.8%)		10 (3.9)	

<sup>a</sup>p-values from Fisher's exact test.

The mean SCAS total anxiety score for siblings was 6.9 (95% CI=[2.8–11.0]) points higher than in the normative sample ( $p_{t \text{ test}} = .001$ ), and 24% (8/25) of siblings exceeded the 10% cut-offs for the SCAS total anxiety score ( $p = .01$ ). More male siblings scored above the threshold for total anxiety (28% [10–53%]) compared to female siblings (20% [4–48%]) (Table 3). The anxiety domain that was most likely to be a problem was social phobia with 42% [20–67%] of male siblings and 40% [16–68%] of female siblings scoring above the threshold. Separation anxiety appeared to be a problem for female siblings.

The siblings' EQ-5D-Y scores were similar to those from a community German school sample (Table 4). A total of 28 (82.4%) siblings reported 'no pain or discomfort' in comparison with 133 (51.6%) of the South African school sample ( $p = .03$ ). In all, 20 (58.8%) siblings were sad, worried or unhappy compared with 98 (35%,  $p = .37$ ) of the South African school sample.

### Findings from qualitative data

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 ( $p > .05$ ). Two overarching themes emerged from the data: Impact on Family and Impact on Siblings with a number of sub-themes.



## *Impact on family*

All the siblings 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 John's doing really well'.

**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 Sam's illness, is that before we were big campers, big walkers, big cyclers and now we just can't do that'.

**'Not knowing'.** Those interviewed had difficulties in understanding what CFS/ME is, how it affected their siblings and whether their behaviour could be explained by the diagnosis: 'I find it quite difficult really 'cause I don't really fully understand what it is'.

One sibling in particular talked about not wanting to have more information about what CFS/ME is: 'I kind of didn't really, didn't really want to know, really know much about it'.

The unpredictability of CFS/ME was mentioned by some of the siblings, including the symptoms, the impact they have on the sibling and the impact on the family: 'we can't plan too far ahead because we don't know if she will be OK'.

A few of the 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'.

Some siblings believed that either their siblings did not have CFS/ME or the diagnosis allowed the sibling to behave in ways that they would not be able to do if they had not been diagnosed with CFS/ME: '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'.

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'.

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

Difficulties around communication about CFS/ME were discussed. These siblings also talked about 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') 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').

Over half the siblings talked about negative, or lack of, communication within the family, for example, 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'.

Almost all of the 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]'.

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'.

Conversely, many 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'.

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'.

*Life outside the family.* Extra-curricular activities were mentioned as a tool to coping with the negative impact of CFS/ME, for example, hobbies and friendships: 'If I have a bad day either I go out on my road bike, I go mountain biking or I go for a run'.

### **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.* Most 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 Sally'.

A few 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'.

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'.

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'.

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'.

In contrast, one participant felt that his brother's illness had allowed him to develop more: 'Because in a way I've found my own voice I suppose'.

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'), 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.

*Emotional reactions.* Almost all talked about some level of emotional impact. Four spoke about the change in their sibling relationship and how this felt like a loss: 'I suppose the worst part would be . . . not being able to go out with her anymore'.

Some of them felt this loss almost like a bereavement: 'I've lost my brother . . . we used to do everything together and now we just don't'.

Feelings of guilt were mentioned due to being able to still do things 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'.

Some described 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'.

Some 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"'.

A few 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'.

*Social emotional support/impact.* Most of the siblings talked about social/emotional support as well as the negative impact.

They talked about the negative social impact of having a brother or sister with a diagnosis such as CFS/ME. Around half 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'.

A few 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'.

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'.

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'.

The importance of social support, through friends or family, was mentioned. Six spoke about the close, supportive relationship they had with their brother or sister: 'I come back from school and then I usually go talk to Nell for a bit, me and Mum'.

However, one of the siblings did say that the CFS/ME had negatively impacted the close relationship he previously had with his brother: 'We, you know, just don't really have anything, anything that we share anymore because of the illness, so . . . it's horrible'.

Many siblings mentioned the close relationship they had with their parents: 'If it was a family problem, I'd go to family' and with the people outside of the family, such as friends: 'Obviously they understand . . . that it is harder, but they don't mind'.

*Advice to CFS/ME specialist services or to families.* 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'.

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'.

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'.

## Discussion

The siblings of young people with CFS/ME have high levels of anxiety with a quarter of siblings scoring above the 90th centile for anxiety suggesting they may benefit from treatment. All those interviewed described a change in family function and/or a reduction in social support which they relate directly to their sibling's illness. Some described feelings of loss and bereavement, anger, frustration and worry about their sibling, but few felt they had been given explanations about the illness or treatment. Many siblings did not believe their brother/sister had a real illness which may be related to poor communication.

### *Results in context of previous literature*

Our study is consistent with a previous study from this patient cohort (Missen et al., 2011) where parents described that children with CFS/ME were angry and irritated, which was often directed at their well siblings. Our results of change in role/focus, restrictions on family life/activities and the negative social impact were consistent with the Jackson study's themes of change in sibling relationship, restriction on family activities and deterioration in peer relationships. However, there are also some differences. Unlike the Jackson study, we have shown that siblings are concerned about the impact on their family as a whole, as well as the impact on themselves. Additionally, we describe how positive communication was protective and how a lack of or negative communication could affect understanding/knowledge and therefore sibling acceptance of the diagnosis. Our finding of disbelief in the siblings is a new finding.

Our finding that such a high percentage of siblings were very anxious is novel for CFS/ME but is consistent with previous studies that have shown that having a child with a chronic disorder within the family does have a negative effect on the siblings' emotional wellbeing (Alderfer et al., 2010; Areemit et al., 2010; Batte et al., 2006; Hamama, Ronen, & Feigin, 2000; Houtzager, Grootenhuis, Hoekstra-Weebers, Caron, & Last, 2003). This suggests that many of the issues are similar across chronic diseases.

We were also able to explore the types of anxiety presenting in the male and female siblings. Female siblings were more likely to experience separation and social anxiety, whereas boys were more likely to experience generalised and social anxiety. The heightened social anxiety in both groups could highlight the social stigma that is still attached to CFS/ME. On the other hand, since this pattern is also seen in children with CFS/ME (Crawley, Hunt, & Stallard, 2009), it could indicate a familiar trait. The interview data supported this finding, with most of the siblings describing worries around telling friends about their sibling's ill health. The differences in the ways that the male and female siblings seem to show their anxiety (generalised vs separation) could highlight the different ways in which the young people cope with the situation they are in. Both types could illustrate the unsettling natures and the shift in the family pattern described by the adolescents throughout the interviews.

In addition to the negative impact on siblings, this study describes some protective factors, which have been found in previous literature for siblings of children with chronic illnesses, such as good communication, social/emotional support and extra activities (Areemit et al., 2010; Batte et al., 2006; Hames & Appleton, 2009; Havermans et al., 2011).

### *Meaning of the study and possible mechanisms*

The siblings in the study showed an increase in anxiety but not depression. Many of the siblings describe a lot of changes experienced since their brother or sister had become unwell, for example,

change of role and focus, changes to the ways in which the family spend time together and the unpredictability of the CFS/ME. Many of them also describe a lack of understanding or a difficulty of communication within the family. All of these situations may be causing the increase in anxiety in these adolescents. However, we do not know whether or not these children have only become anxious since their brother or sister became ill or whether they have always been anxious.

Many siblings interviewed described a lack of knowledge about CFS/ME and the impact on their brother or sister. This could be due to a general lack of knowledge within our society and the health system (Bowen, Pheby, Charlett, & McNulty, 2005). Or it could be due to the lack of communication reported within the family. Lack of knowledge appeared to contribute to disbelief about whether the index child had a 'real' illness. Siblings 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'). Although the lack of belief may be because of lack of communication within the family, some siblings explained that they had not wanted to know what CFS/ME was and had not asked. Siblings were at times 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. 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 (Gilligan, 2004; Jackson, Bijstra, Oostra, & Bosma, 1998; Steinberg, 2001).

Illness characteristics, which have previously been found to have more of a negative impact on siblings of other chronic health conditions, include whether the illness affects the day-to-day functioning of the child (Sharpe & Rossiter., 2002) or whether 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 children may explain some of the sibling's 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.

### *Strengths and limitations*

We used the qualitative results to interpret the quantitative data. We were able to compare whether the index young people were similar to the clinic population using routinely collected data. The index children of the recruited siblings were on average 1 year younger than those children with CFS/ME whose siblings were not recruited to this study. This could be because older index children had siblings who were too old for this study (i.e. over 18 years of age). In addition, index children were less likely than the general clinic population to be attending no school suggesting that those with the more severely affected siblings were less likely to take part in the study which may affect the generalisability of the study.

Only 59 out of a possible 162 siblings were approached by clinicians about this study. There are a number of possible reasons for this low recruitment rate. Although it is likely that some clinicians forgot to recruit participants, 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 those interviewed do not represent children with significant family problems.

The use of normative data, rather than data sampled from the same population, is one limitation of our study. Also, the normative data were from Germany, South Africa and the Netherlands

because there were no UK-based normative data. These normative data were chosen because the Netherlands group for the SCAS, the German and South African groups for the EQ-5D-Y and the German group for the HADS covered the matching age range (13–19, 10–18, 13–19 and 12–16 years, respectively), and the Netherlands group was a predominantly White population, which is similar to the sibling population. The German and South African groups did not report the ethnic mix in their studies. The German and South African samples for the EQ-5D-Y also had an even distribution of males and females, which is similar to the sibling sample. Our statistical comparisons were limited by the small size of the sibling sample.

Siblings showed an increase in anxiety only on the SCAS, rather than on the HADS. This could be due to the SCAS being a lengthier, and so possibly more sensitive and in-depth, measure of anxiety than the HADS. The SCAS was able to pick up the differences between the different types of anxiety, which the HADS is not sensitive enough to do.

### ***Impact on clinical practice***

This research has highlighted the need to identify those siblings who may be at risk from mental health difficulties. 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, and also about whether the sibling may need separate support.

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. For some families, where the risks have been identified and communication and knowledge are particularly poor, family therapy may be a useful tool.

This study has shown that siblings of children with CFS/ME are more anxious than the normative samples with similar depression and quality of life. In interviews, 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 their own adjustment and acceptance. 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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### **Note**

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



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