



Citation for published version:

Loades, M, Rimes, K, Ali, S, Lievesley, K & Chalder, T 2019, 'Does fatigue and distress in a clinical cohort of adolescents with chronic fatigue syndrome correlate with fatigue and distress in their parents?', *Child: Care, Health and Development*, vol. 45, no. 1, pp. 129-137. <https://doi.org/10.1111/cch.12626>

DOI:

[10.1111/cch.12626](https://doi.org/10.1111/cch.12626)

Publication date:

2019

Document Version

Peer reviewed version

[Link to publication](#)

This is the peer reviewed version of the following article, Loades, M. E., 2018. Does fatigue and distress in a clinical cohort of adolescents with chronic fatigue syndrome correlate with fatigue and distress in their parents. *Child: care, health and development* which has been published in final form at <https://doi.org/10.1111/cch.12626>. This article may be used for non-commercial purposes in accordance with Wiley Terms and Conditions for Self-Archiving.

University of Bath

Alternative formats

If you require this document in an alternative format, please contact:
openaccess@bath.ac.uk

General rights

Copyright and moral rights for the publications made accessible in the public portal are retained by the authors and/or other copyright owners and it is a condition of accessing publications that users recognise and abide by the legal requirements associated with these rights.

Take down policy

If you believe that this document breaches copyright please contact us providing details, and we will remove access to the work immediately and investigate your claim.

Abstract

Objectives: Previous studies have found that parents of children with CFS are more fatigued, and mothers are more distressed than healthy controls. Managing the disabling symptoms of CFS can result in disruption and burden for the family. . Most research has focused on mothers. This study sought to further explore the associations between adolescent fatigue and distress, and parental fatigue and distress, as well as family functioning, including both mothers and fathers.

Design: Cross-sectional study of a clinical cohort of consecutive attenders at a specialist chronic fatigue unit.

Methods: Questionnaires were completed by adolescents (N = 115, age 11-18) with a confirmed diagnosis of CFS and their mothers (N = 100) and fathers (N = 65).

Results: Maternal fatigue was significantly correlated with maternal distress, but not with adolescent fatigue, depression, anxiety or functioning. This pattern held true for paternal fatigue. Maternal and paternal anxiety and depression were significantly correlated with family functioning. Paternal and maternal distress were correlated with each other. Mothers and fathers tended to have a consistent view of family functioning. Family functioning, specifically being overwhelmed by difficulties and scoring lower on strengths and adaptability was positively associated with adolescent depression. Unexpectedly, higher levels of adolescent fatigue and poorer physical functioning were associated with better family functioning as rated by the mother.

Conclusions: Parents of adolescents with fatigue scored near to or within normative range for non-clinical samples on distress, fatigue and family functioning. Parental distress may contribute to or result from poorer family functioning. Family functioning, particularly

building strengths and adaptability, may be clinically important in CFS, as well as attending to parental, (particularly paternal) distress in families where adolescents are low in mood.

Keywords

CFS, adolescents, fatigue, distress, parents, family functioning

Key Messages

- Fatigue in a parent was significantly correlated with their own anxiety and depression, and with family functioning, but not with the adolescents' fatigue, anxiety, depression or functioning.
- Within a family, more anxiety and/or depression symptoms in one parent appeared to be related to more anxiety and/or depression symptoms in the other parent.
- Parental distress was also related to poorer family functioning.
- Family functioning, particularly reduced family strength and adaptability, and being overwhelmed by difficulties, reported by the mother, was associated with adolescent depression.
- A family focused approach could address distress in the parent in addition to working with the adolescent.

Chronic Fatigue Syndrome (CFS) is a common illness in adolescence, which impacts significantly on functioning. According to NICE guidelines (NICE, 2007), CFS is diagnosed when an adolescent has experienced persistent and debilitating fatigue for > 3 months in the absence of another explanation (e.g. ongoing exertion, primary medical condition causing secondary fatigue). Prevalence rates range from 0.06 - 0.11% in primary care and in specialist paediatric samples (Haines, Saidi, & Cooke, 2005; Nijhof et al., 2011) and up to 2.4% in population studies which capture those who have chronic disabling fatigue but who may not seek help or receive a diagnosis (Chalder, Goodman, Wessely, Hotopf, & Meltzer, 2003; Crawley et al., 2012). Children and young people (henceforth referred to as 'children') with CFS miss an average of 1 year of school, as a result of their illness (Rangel, Garralda, Levin, & Roberts, 2000) and those attending specialist services have an average school attendance of 40% (Crawley & Sterne, 2009).

Paediatric CFS has a considerable impact on parents. The journey to having their child diagnosed with CFS and being referred to specialist CFS services for treatment can be difficult; mothers of children with CFS may have needed to be proactive and persistent to achieve this (Beasant, Mills, & Crawley, 2014; Sankey, Hill, Brown, Quinn, & Fletcher, 2006). As compared to parents of children with juvenile arthritis and children with emotional disorders, parents of children with CFS report higher levels of family disruption and increased family burden, including higher levels of arguments, changed relationships, family atmosphere, isolation and reduced family visits (Rangel, Garralda, Jeffs, & Rose, 2005). They also felt the financial impact of the illness, including a loss of monthly income and increased monthly expenditure (Missen, Hollingworth, Eaton, & Crawley, 2012).

It is not surprising then that research has shown that parents of children with CFS have elevated levels of fatigue and more distress (Rangel et al., 2005; van de Putte et al., 2006). In one study, the presence of fatigue in the mother corresponded with a 5.3 times higher likelihood of fatigue in their adolescent offspring (van de Putte et al., 2006). However, fathers did not report higher levels of fatigue in a small sample of 32 fathers and 34 mothers (van de Putte et al., 2005), which also specifically highlighted the associations between paternal locus of control in relation to health, and that of their offspring. Two studies have reported higher levels of distress, as measured by the general health questionnaire (GHQ-12) in parents (mostly maternal respondents, and only 1 parent per family) of children with CFS. This was found in comparison to children with juvenile arthritis and children with emotional disorder (Rangel et al., 2005) as well as when compared to healthy controls (Missen, Hollingworth, Eaton, & Crawley, 2012). In the latter study, 72% were above the clinical cut-off versus 20% in the healthy control group. More specifically, self-reported maternal depression, anxiety and somatization was found to correspond to a 5.6 times higher odds ratio for the presence of CFS in the adolescent (van de Putte et al., 2006). In a qualitative study using in-depth interviews, 8 mothers attributed their distress to a lack of understanding by others, marital strain, concern about their child's distress and the impact on their other children, as well as their own feelings of frustration (Missen et al., 2012).

There are a number of possible explanations for associations between fatigue in the mother and the child. It may reflect a biological vulnerability, shared environmental stressors or a behavioural modelling process (Chalder, Deary, Husain, & Walwyn, 2010; Nijhof et al., 2013). Regardless of cause, maternal fatigue severity is important as it may be associated with treatment outcome for CFS in children. At the long term follow-up of a randomised

control trial of CBT in adolescents with CFS, maternal fatigue, present in 28% of mothers at baseline, was found to be a significant predictor of the child's fatigue after treatment (Knoop, Stulemeijer, de Jong, Fiselier, & Bleijenberg, 2008). However, in a trial of internet based CBT for CFS in adolescents, there was no evidence to confirm an association between higher maternal fatigue severity and inferior treatment outcome (Nijhof et al., 2013), although increased maternal focus on bodily symptoms was associated with reduced odds of the child's long-term recovery.

In summary, previous research has highlighted the associations between maternal fatigue and distress in mothers of children with CFS, and there is mixed evidence about the importance of maternal fatigue for treatment outcome. Few studies have considered fathers as well as mothers, and in the small scale studies that exist, the focus was paternal fatigue and specifically on health beliefs, rather than on broader constructs of distress or family functioning, which may also be important. Given the findings from previous research, this study aims to examine the associations between child fatigue, distress and functioning and maternal and paternal fatigue and distress. In addition, the association between maternal- and paternal-reported family functioning will be examined. The research questions are: 1) is parental fatigue associated with child fatigue? 2) is parental distress associated with child distress? 3) is parental distress associated with child fatigue and functioning? 4) are views of family functioning correlated between mothers and fathers, and associated with child symptomatology?

Method

Participants

All adolescents (11-18 years old) who attended an initial assessment at a specialist CFS Unit from August 2010 to January 2017 were invited to participate. During this period, 192 adolescents attended an initial assessment. Of these, 125 had a confirmed diagnosis of CFS, and 115 (92%) of these completed the measures for this study.

Measures

Basic demographics and self-reported percentage school attendance were gathered as part of routine clinical practice.

Adolescent Measures:

Fatigue – The Chalder Fatigue Questionnaire, CFQ (Chalder et al., 1993) is an 11-item scale which assesses the severity of mental and physical fatigue. Each item is rated as ‘less than usual’, ‘no more than usual’, ‘more than usual’ and ‘much more than usual’ with a recall period of 1 month. It has good reliability and validity (Cella & Chalder, 2010).

Physical Functioning –The Short Form 36 (SF-36) physical functioning scale (Ware Jr, Kosinski, & Keller, 1996) is a 10-item physical function scale which lists activities of daily living such as ‘bathing or dressing yourself’ and asks respondents to rate, on a 3 point scale, how much their health limits them in these activities. Higher scores indicate better functioning. The SF-36 has been psychometrically examined in adolescent chronic illness samples e.g. cystic fibrosis (Gee, Abbott, Conway, Etherington, & Webb, 2002).

School and social adjustment –The Work and Social Adjustment Scale, WSAS (Mundt, Marks, Shear, & Greist, 2002) is a 5-item scale which instructs respondents to rate, on a 0-8 scale, their functioning in work, domestic, social and leisure activities and close relationships. It has been shown to be valid and reliable in CFS patients (Cella, Sharpe, & Chalder, 2011). In the current study, the word ‘work’ in the first item was replaced by the words ‘school/college’, and the scale will therefore be referred to as the School and Social Adjustment Scale. Despite this amendment, the reliability of this scale was good (Cronbach’s alpha = .807).

Anxiety – The State Trait Anxiety Inventory, STAI (Speilberger, Gorsuch, & Lushene, 1970) is a 40 item scale assessing the intensity of felt anxiety, capturing both state anxiety (experienced in particular situations) and trait anxiety (threat sensitivity). Each item is rated on a 1-4 scale. It is considered to be valid and reliable (Speilberger et al., 1970).

Depression - Children’s Depression Inventory, CDI (Kovacs, 1992) is a 27-item self-report scale, which asks about the presence of depressive symptoms including negative mood, ineffectiveness, anhedonia, low self-esteem and interpersonal problems, with a recall period of 2 weeks. Responses are rated on a 0-2 scale. It is reliable and validated for use in 7 to 17 year olds (Kovacs, 1992).

Parent measures:

Parents were asked about their own psychiatric history and current mental health. They also completed the following measures:

Parental fatigue - Parents completed the CFQ, reporting on their own fatigue.

Parental anxiety/depression - Hospital Anxiety and Depression Scale (HADS;(Zigmond & Snaith, 1983) is a 14-item scale, with 7 items pertaining to anxiety and 7 items to depression. Each item is rated on a 0-3 scale. In addition to scoring each subscale separately to generate an anxiety and a depression score, the total HADS score (anxiety + depression) can be used as an index of general distress (Norton, Cosco, Doyle, Done, & Sacker, 2013).

Family Functioning – The Systemic CORE, SCORE-15 (Stratton, Bland, Janes, & Lask, 2010) is a 15-item questionnaire, which presents statements about family life and relationships (e.g. hostility, communication, atmosphere, flexibility, roles and rules) which respondents are required to rate on a 6-point scale. It is composed of 3 subscales (Strengths and Adaptability, Overwhelmed by Difficulties, and Disrupted Communication) and is validated for use with adolescents and adults (Stratton et al., 2010). Higher scores on each of the subscales indicate poorer functioning, and lower scores indicate better functioning. For clarity, ‘strengths and adaptability’ will henceforth be referred to as ‘reduced strengths and adaptability’.

Procedure

All patients invited to attend an initial assessment at a specialist CFS unit were sent questionnaires to complete. Included with the questionnaires was an invitation letter explaining the use of this data for audit and potential research purposes. During the

appointment, patients and their parents were given information sheets and an explanation of the study, and consent procedures were completed.

Ethical Approval

Ethical approval was granted by an NHS research ethics committee (REF omitted for blinding), the Research and Development departments at the (omitted for blinding). The collection of routine outcomes was approved by the clinical audit committee of (omitted for blinding).

Data Analysis Plan

The data was analysed using SPSS 23.0. Descriptive statistics were conducted. Bivariate correlations (Pearson's, 2 tailed, missing cases excluded pairwise) were utilised to explore the associations between parent and adolescent variables of interest.

Results

The sample consisted of 115 adolescents, age 11 to 18, who met the NICE criteria for CFS (NICE, 2007) based on clinician judgement. Characteristics of the participants and their parents are shown in table 1. Of the 65 fathers, 64 were biological fathers and 1 was a step-father. In most families, both parents were the main carers (57.4%), with the mother being the main carer in 38.7% of the families, and the father being the main carer in 2 of the families (1.8%).

[insert table 1 about here]

Around two thirds of mothers (63%) classified themselves as employed, and 69% were married. Ten percent of mothers reported a past history of CFS, 22% a past history of depression, and 11% reported anxiety in the past. Eleven percent of mothers currently had self-reported CFS, and 22% scored > 18 on the CFQ (reflecting severe fatigue). Furthermore, 15% self-reported current depression and 9% anxiety. In 7% of the sample, mothers had co-morbid self-reported anxiety and depression. Fifteen percent of mothers scored > 10 on the depression subscale of the HADS, and 3% scored >10 on the anxiety subscale of the HADS.

The vast majority of fathers (88.9%) classified themselves as employed, and 88.9% were married. One father had a past history of CFS, 12 (19%) had a past history of depression, and 6 (9.5%) had previously experienced anxiety. No fathers currently had self-reported CFS/ME, and 4 (6.2%) scored > 18 on the CFQ. Furthermore, 7 (11.1%) reported current depression, and 5 (7.9%) anxiety. In 3% of the sample, fathers had co-morbid self-reported anxiety and depression. Twenty percent of fathers scored > 10 on the depression subscale of the HADS, and 7.8% scored > 10 on the anxiety subscale of the HADS.

Maternal fatigue was significantly correlated with maternal anxiety and maternal depression (see table 2). Maternal fatigue was not significantly correlated with child fatigue (CFQ), child depression (CDI), child state anxiety (STAI-state), child physical functioning (SF-36 PFS) or child school and social adjustment (SSAS). Maternal fatigue was not significantly correlated with family functioning.

Paternal fatigue was significantly correlated with paternal anxiety and paternal depression (see table 2). Paternal fatigue was not significantly correlated with their child's fatigue, physical (SF-36 PFS) or school and social adjustment (SSAS), depression (CDI) or anxiety (STAI-state). Paternal fatigue was positively correlated with their score on the Overwhelmed by Difficulties subscale of family functioning (SCORE).

Maternal anxiety was significantly correlated with mothers' own depression and fatigue, and with paternal anxiety and depression, but not with any of the child variables (fatigue, school/social and physical functioning, depression and anxiety). Maternal depression was significantly correlated with maternal anxiety, but not with any of the child variables. Using the HADS total score (anxiety + depression) as an index of general distress (Norton et al., 2013), maternal general distress was correlated with own fatigue ($r = .458^*$, $p < .000$) and paternal' general distress ($r = .279^*$, $p = .034$). Higher levels of both maternal depression and maternal anxiety were significantly correlated with poorer family functioning, specifically being overwhelmed by difficulties and having disrupted communication (see table 2).

Paternal anxiety was significantly correlated with their own depression and fatigue, and with maternal anxiety, but not with the child variables. Paternal depression was significantly correlated with paternal fatigue and anxiety, maternal anxiety, and with the child's depression. Paternal general distress (HADS total) was correlated with own fatigue, child CDI score and maternal general distress. Higher levels of paternal depression and anxiety were significantly correlated with poorer family functioning (SCORE) across all 3 subscales.

Mothers' and fathers' scores of family functioning (SCORE) on each of the 3 subscales were correlated with one another, and with each other's scores, indicating that parents tended to have a fairly consistent view of family functioning (see table 3). The family functioning subscales were not associated with adolescent anxiety, but 2 subscales, the reduced Strengths and Adaptability Scale (maternal report but not paternal report) and the Overwhelmed by Difficulties Scale (maternal report but not paternal report), were associated with the adolescent depression score on the CDI (see table 4). Adolescent physical functioning (SF-36 PFS), school and social functioning (SSAS) and fatigue (CFQ) were also associated with the SCORE reduced scores on the Strengths and Adaptability scale (maternal report only, apart from SF-36 PFS, which showed an association with both parents' report).

[insert tables 2, 3 and 4 about here]

Discussion

In this cohort of adolescents with CFS, presenting to a specialist CFS service, maternal fatigue was significantly correlated with maternal anxiety and depression, and with family functioning, but not with the adolescents' fatigue, anxiety, depression or functioning. Similarly, paternal fatigue was significantly correlated with paternal anxiety and depression, and with family functioning, and but not with the adolescent variables, with the exception of paternal depression which was positively associated with adolescent depression. Raised levels of distress (anxiety and depression) in one parent in a family appeared to be related to higher levels of distress in the other parent, and raised levels of parental distress were also related to poorer parent-reported family functioning. Family functioning, particularly

reduced strength and adaptability, and being overwhelmed by difficulties, reported by the mother, was associated with adolescent depression. Unexpectedly, reduced family strength and adaptability was related to higher levels of adolescent physical functioning and lower levels of fatigue, but to poorer social and school functioning.

In this sample, parental fatigue was lower than in some other studies (van de Putte et al., 2005; van de Putte et al., 2006) but comparable to others (Rangel et al., 2005); see table 5). Previously, it has been suggested that intergenerational fatigue patterns may be accounted for by environmental (van de Putte et al., 2006) and/or by genetic (Hickie, Kirk, & Martin, 1999) factors. However, it is interesting that the mean CFQ scores for the parents in the current study were comparable to those reported by a community sample of adults (Cella & Chalder, 2010).

Furthermore, this sample of parents reported lower rates of distress than has been found in other studies of parents whose children have CFS (see table 5). Compared to normative data on the HADS in non-clinical populations, parents scores on the HADS were slightly raised but not dramatically so, and the mean score of this study sample falls within the 50th to 65th percentile when compared to normative data (Crawford, Henry, Crombie, & Taylor, 2001). Fathers who were depressed tended to have children who were depressed, although the same was not true for mothers. This merits further exploration in future research.

Family functioning in this study was more similar to that of non-clinical families than of families receiving family therapy (Stratton et al., 2010). Importantly, parents tended to agree with one another about family functioning. Often one parent's report is used as a

proxy for family functioning, therefore it is reassuring to find in this study that mother's and father's views were reasonably consistent. Family functioning was related to parent distress and lower levels of family strengths and adaptability (as rated by the mother) were associated with higher levels of adolescent depression symptoms and worse school and social functioning. However, the converse was true for fatigue and physical functioning; that is, reduced family strengths and adaptability was associated with less fatigue and better physical functioning. It may be protective in terms of adolescent mood and school and social functioning to be in a family where family members talk to each other about things that matter and are good at finding new ways of dealing with difficult things, look after each other at times of distress and listen to and trust each other. The family being overwhelmed by difficulties (as rated by the mother) were associated with higher levels of adolescent depression symptoms. When children are depressed they tend to withdraw and to talk less, so this could explain why more severe depression is associated with mothers rating family functioning as worse. The direction of these associations cannot be determined from this study and it is possible that there are bi-directional influences.

Strengths and Limitations

The addition of fathers as well as mothers in this study is important and previously understudied; this has provided insight into fathers, as well as into parent dyads. Including parental reports of family functioning in addition to fatigue and distress has shed light on the associations between these variables. Consecutive attenders in routine clinical practice were invited to participate, reducing the likelihood of selection bias. Despite this, the sample was limited to those attending specialist services, and therefore it is unclear to what extent these findings represent those with CFS who do not attend specialist services. This

study included fathers as well as mothers. However, data was available for a larger number of mothers than fathers, and it is possible that the lack of significant findings with regard to paternal report on the family functioning measures may be the result of a lack of statistical power. Furthermore, the cross-sectional design of this study means that cause and effect cannot be established. This study was also limited by the lack of a control group.

Clinical implications

Assessment of adolescents with CFS should include parental distress (both maternal and paternal) as well as family functioning. A family-focused treatment approach could particularly build up family strengths and adaptability, and may be particularly relevant where parents are distressed and in families where adolescents are low in mood. Supporting parents with their own distress and particularly attending to fathers' distress, where adolescents are low in mood may be important. Within a family-focused approach, distress could be addressed in parents as well as children (Chalder et al., 2010). This may be particularly appropriate if the child's fatigue and/or mood is a significant contributory factor to the parent's own distress. However, if addressing parental distress within a family intervention is deemed inappropriate, it may help to refer the distressed parent to an adult mental health service.

Conclusions

In this large clinical cohort of adolescents presenting to specialist services, and their parents, levels of parental fatigue, distress and family functioning were comparable to population norms of non-clinical samples. Parental fatigue and distress did not seem to be strongly associated with adolescent fatigue and distress. However, adolescent mood may be related

to reduced family strengths and adaptability, and parental distress may be related to poorer family functioning. These are important findings as they are different to prior research which was limited by small sample sizes and the relative absence of data from fathers.

Table 1. Characteristics of the participants

Gender	Male	35 (30.4%)
	Female	78 (67.8%)
Ethnic Origin	White Caucasian	88 (80%)
	Black	4 (3.6%)
	Mixed race	3 (2.7%)
	White other	5 (4.5%)
	British (unspecified)	10 (9.0%)

	Range in the current sample	Mean (S.D)
Age (years)	11-18	15.07 (1.69)
Fatigue (Chalder Fatigue Scale)	5-33	23.08 (5.82)
Percentage school attendance	0-100	40.55 (35.96)
Occupational Functioning (SSAS Total Score)	4-39	23.96 (7.94)
Physical functioning (SF-36PFS)	0-100	50.35 (25.42)
STAI State Anxiety	21-78	44.93 (12.22)
STAI Trait Anxiety	26-69	43.31 (10.89)
Depression (CDI)	4-40	15.84 (8.28)

	Mothers		Fathers	
Ethnic Origin	White Caucasian	93 (93%)	59 (90.8%)	
	Black	3 (3%)		
	Asian	1 (1%)	4 (6.2%)	
	Other	3 (3%)	2 (3%)	

	Range	Mean (S.D)	Range	Mean (S.D)
CFQ Fatigue	4-33	14.51 (6.22)	0-29	12.18 (4.44)
HADS Anxiety	0-20	7.01 (3.92)	0-17	6.59 (4.44)
HADS Depression	0-20	4.38 (3.58)	0-14	3.77 (3.53)
HADS Total Score	1-40	11.40 (6.65)	0-31	10.35 (7.44)
Family Functioning (SCORE) – Lack of Strengths and Adaptability*‡	5-22	10.42 (4.14)	5-29	12.15 (5.45)
Family Functioning (SCORE) – Overwhelmed by Difficulties*	5-25	10.04 (4.60)	5-21	10.66 (4.54)
Family Functioning (SCORE) – Disrupted Communication*	5-19	8.99 (2.87)	5-23	10.57 (4.29)

CDI – Children's Depression Inventory, CFQ – Chalder Fatigue Questionnaire, HADS = Hospital Anxiety and Depression Scale, SCORE = Systemic CORE Family Functioning Measure, SF-36 – Short Form 36, STAI – State-Trait Anxiety Inventory, SSAS – School and Social Adjustment Scale

*higher scores are indicative of poorer functioning

‡ higher scores indicate reduced strengths and adaptability

Table 2. Correlations between parental distress and parental fatigue and the adolescent variables and family functioning - data shown as r (p)

	HADS Anxiety (Mother)	HADS Depression (Mother)	Fatigue - CFQ (Mother)	HADS Anxiety (Father)	HADS Depression (Father)	Fatigue - CFQ (Father)
HADS Depression (Mother)	.563* (<.000)					
Fatigue - CFQ (Mother)	.220* (.030)	.600* (<.000)				
HADS Anxiety (Father)	.272* (.037)	.138 (.302)	-.068 (.614)			
HADS Depression (Father)	.324* (.012)	.229 (.084)	-.079 (.554)	.742* (<.000)		
Fatigue -CFQ (Father)	.013 (.919)	.025 (.849)	.035 (.792)	.516* (<.000)	.555* (<.000)	
Fatigue - CFQ (Adolescent)	-.147 (.152)	-.133 (.199)	.115 (.264)	.101 (.433)	-.082 (.525)	.104 (.412)
SF-36 PFS (Adolescent)	.030 (.781)	.020 (.854)	-.198 (.058)	-.050 (.708)	-.50 (.706)	-.061 (.645)
CDI (Adolescent)	.176 (.111)	.004 (.973)	.023 (.837)	.235 (.091)	.290* (.035)	.159 (.252)
STAI-State (Adolescent)	.126 (.252)	-.052 (.640)	-.008 (.939)	.160 (.235)	.145 (.283)	.012 (.931)
SCORE Family Functioning Reduced Strengths and Adaptability[‡] (Mother)	.367* (<.000)	.196 (.060)	-.036 (.728)	.333* (.013)	.472* (<.000)	.048 (.727)
SCORE Family Functioning Overwhelmed by Difficulties (Mother)	.555* (<.000)	.459* (<.000)	.197 (.052)	.332* (.013)	.396* (.003)	.048 (.724)
SCORE Family Functioning Disrupted Communication (Mother)	.269* (.008)	.147 (.156)	.025 (.806)	.275* (.040)	.341* (.010)	-.061 (.653)
SCORE Family Functioning Reduced Strengths and Adaptability[‡] (Father)	.206 (.124)	.047 (.730)	-.181 (.183)	.316* (.013)	.422* (.001)	.040 (.757)
SCORE Family Functioning Overwhelmed by Difficulties (Father)	.458* (<.000)	.338* (.011)	-.045 (.742)	.564* (<.000)	.574* (<.000)	.363* (.004)
SCORE Family Functioning Disrupted	.324* (.015)	.326* (.015)	-.053 (.699)	.320* (.013)	.444* (<.000)	.122 (.347)

Communication

(Father)

‡ higher scores indicate reduced strengths and adaptability

Table 3. Correlations between mothers' and fathers' scores on the SCORE 15 family functioning subscales - data shown as r (p)

	Family Functioning Overwhelmed by Difficulties (Mother)	Family Functioning Disrupted Communication (Mother)	Family Functioning Lack of Strengths and Adaptability[‡] (Father)	Family Functioning Overwhelmed by Difficulties (Father)	Family Functioning Disrupted Communication (Father)
Family Functioning Lack of Strengths and Adaptability[‡] (Mother)	.483* (<.000)	.497* (.000)	.461* (<.000)	.307* (.022)	.273* (.046)
Family Functioning Overwhelmed by Difficulties (Mother)		.601* (<.000)	.313* (.019)	.421* (.001)	.369* (.006)
Family Functioning Disrupted Communication (Mother)			.457* (<.000)	.395* (.003)	.339* (.011)
Family Functioning Lack of Strengths and Adaptability[‡] (Father)				.541* (<.000)	.589* (<.000)
Family Functioning Overwhelmed by Difficulties (Father)					.694* (<.000)

[‡] higher scores indicate reduced strengths and adaptability

Table 4. Correlations between adolescent variables and SCORE 15 family functioning as rated by mothers and fathers – Data shown as r (p)

	Family Functioning Reduced Strengths and Adaptability[‡] (Mother)	Family Functioning Overwhelmed by Difficulties (Mother)	Family Functioning Disrupted Communication (Mother)	Family Functioning Reduced Strengths and Adaptability[‡] (Father)	Family Functioning Overwhelmed by Difficulties (Father)	Family Functioning Disrupted Communication (Father)
Adolescent anxiety (STAI-State)	.165 (.142)	.183 (.095)	.109 (.330)	.067 (.631)	.163 (.239)	.093 (.506)
Adolescent depression (CDI)	.240* (.034)	.230* (.040)	.111 (.324)	.213 (.133)	.254 (.072)	.185 (.198)
Adolescent physical functioning (SF-36)	.230* (.032)	.005 (.964)	.121 (.263)	.286* (.031)	.143 (.290)	.223 (.098)
Adolescent school and social adjustment (SSAS)	-.206* (.049)	.063 (.544)	-.035 (.733)	-.204 (.125)	-.122 (.361)	-.046 (.733)
Adolescent fatigue (CFQ)	-.261* (.012)	-.102 (.327)	-.125 (.230)	-.176 (.176)	.099 (.449)	-.076 (.566)

CDI – Children’s Depression Inventory, CFQ – Chalder Fatigue Questionnaire, SF-36 – Short Form 36, STAI – State-Trait Anxiety Inventory, SSAS – School and Social Adjustment Scale

[‡] higher scores indicate Reduced strengths and adaptability

Table 5. Table showing rates of parental fatigue and distress in comparison to previous studies

	Maternal fatigue	Paternal fatigue	Maternal Distress	Paternal Distress
Rangel et al. (2005)	Parental informant: 22% CFS-like diagnosis, 9% fatigue is markedly impairing		Parental informant: 52% caseness on the General Health Questionnaire (score of > 3)	
van de Putte et al. (2006)	50% fatigue present 2.4% CDC criteria present	43.1% fatigue present 1.1% CDC criteria present		
van de Putte et al. (2005)	56% self-reported fatigue	13% self-reported fatigue		
Missen et al. (2012)			72% caseness on the General Health Questionnaire (score of > 3). 41% anxiety, 13% depression on the HADS	
Current study	11% current CFS (self-report) 22% scored > cut-off of 18 on CFQ	No current CFS (self-report) 6.2% scored > cut-off > 18 on CFQ	15% self-reported current depression, and 15% scored > cut-off of 10 on the HADS-D. 9% self-reported current anxiety and 3% scored > cut-off of 10 on HADS-A.	11.1% self-reported current depression, and 20.3% scored > cut-off of 10 on HADS-D. 7.9% self-reported current anxiety and 7.8% scored > cut-off of 10 on HADS-A.

REFERENCES

- Beasant, L., Mills, N., & Crawley, E. (2014). Adolescents and mothers value referral to a specialist service for chronic fatigue syndrome or myalgic encephalopathy (CFS/ME). *Prim Health Care Res Dev*, *15*(2), 134-142. doi:10.1017/S1463423613000121
- Cella, M., & Chalder, T. (2010). Measuring fatigue in clinical and community settings. *J Psychosom Res*, *69*(1), 17-22.
- Cella, M., Sharpe, M., & Chalder, T. (2011). Measuring disability in patients with chronic fatigue syndrome: reliability and validity of the Work and Social Adjustment Scale. *J Psychosom Res*, *71*(3), 124-128.
- Chalder, T., Berelowitz, G., Pawlikowska, T., Watts, L., Wessely, S., Wright, D., & Wallace, E. (1993). Development of a fatigue scale. *J Psychosom Res*, *37*(2), 147-153.
- Chalder, T., Deary, V., Husain, K., & Walwyn, R. (2010). Family-focused cognitive behaviour therapy versus psycho-education for chronic fatigue syndrome in 11- to 18-year-olds: a randomized controlled treatment trial. *Psychol Med*, *40*(08), 1269-1279. doi:doi:10.1017/S003329170999153X
- Chalder, T., Goodman, R., Wessely, S., Hotopf, M., & Meltzer, H. (2003). Epidemiology of chronic fatigue syndrome and self reported myalgic encephalomyelitis in 5-15 year olds: Cross sectional study. *British Medical Journal*, *327*(7416), 654-655.
- Collin, S. M., Tilling, K., Joinson, C., Rimes, K. A., Pearson, R. M., Hughes, R. A., . . . Crawley, E. (2015). Maternal and childhood psychological factors predict chronic disabling fatigue at age 13 years. *Journal of Adolescent Health*, *56*(2), 181-187. doi:<http://dx.doi.org/10.1016/j.jadohealth.2014.09.002>
- Crawford, J., Henry, J., Crombie, C., & Taylor, E. (2001). Normative data for the HADS from a large non-clinical sample. *British Journal of Clinical Psychology*, *40*(4), 429-434.
- Crawley, E., Hughes, R., Northstone, K., Tilling, K., Emond, A., & Sterne, J. A. (2012). Chronic disabling fatigue at age 13 and association with family adversity. *Pediatrics*, *130*(1), e71-79. doi:10.1542/peds.2011-2587
- Crawley, E., & Sterne, J. A. (2009). Association between school absence and physical function in paediatric chronic fatigue syndrome/myalgic encephalopathy. *Arch Dis Child*, *94*(10), 752-756. doi:10.1136/adc.2008.143537
- Gee, L., Abbott, J., Conway, S., Etherington, C., & Webb, A. (2002). Validation of the SF-36 for the assessment of quality of life in adolescents and adults with cystic fibrosis. *Journal of Cystic Fibrosis*, *1*(3), 137-145.
- Haines, L., Saidi, G., & Cooke, R. (2005). Prevalence of severe fatigue in primary care. *Arch Dis Child*, *90*(4), 367-368.
- Harvey, S. B., Wadsworth, M., Wessely, S., & Hotopf, M. (2008). The relationship between prior psychiatric disorder and chronic fatigue: evidence from a national birth cohort study. *Psychol Med*, *38*(7), 933-940.
- Hickie, I., Kirk, K., & Martin, N. (1999). Unique genetic and environmental determinants of prolonged fatigue: a twin study. *Psychol Med*, *29*(02), 259-268.
- Knoop, H., Stulemeijer, M., de Jong, L. W., Fiselier, T. J., & Bleijenberg, G. (2008). Efficacy of cognitive behavioral therapy for adolescents with chronic fatigue syndrome: long-term follow-up of a randomized, controlled trial. *Pediatrics*, *121*(3), e619-625. doi:10.1542/peds.2007-1488
- Kovacs, M. (1992). *Children's depression inventory*: Multi-Health System North Tonawanda, NY.
- Missen, A., Hollingworth, W., Eaton, N., & Crawley, E. (2012). The financial and psychological impacts on mothers of children with chronic fatigue syndrome (CFS/ME). *Child Care Health Dev*, *38*(4), 505-512. doi:10.1111/j.1365-2214.2011.01298.x
- Mundt, J. C., Marks, I. M., Shear, M. K., & Greist, J. M. (2002). The Work and Social Adjustment Scale: a simple measure of impairment in functioning. *The British Journal of Psychiatry*, *180*(5), 461-464.

- NICE. (2007). *Chronic fatigue syndrome/myalgic encephalomyelitis (or encephalopathy): Diagnosis and management of CFS/ME in adults and children.*
- Nijhof, S. L., Maijer, K., Bleijenberg, G., Uiterwaal, C. S., Kimpen, J. L., & van de Putte, E. M. (2011). Adolescent chronic fatigue syndrome: prevalence, incidence, and morbidity. *Pediatrics*, *127*(5), e1169-e1175.
- Nijhof, S. L., Priesterbach, L. P., Uiterwaal, C. S., Bleijenberg, G., Kimpen, J. L., & van de Putte, E. M. (2013). Internet-based therapy for adolescents with chronic fatigue syndrome: long-term follow-up. *Pediatrics*, *131*(6), e1788-e1795.
- Norton, S., Cosco, T., Doyle, F., Done, J., & Sacker, A. (2013). The Hospital Anxiety and Depression Scale: a meta confirmatory factor analysis. *J Psychosom Res*, *74*(1), 74-81.
- Rangel, L., Garralda, M., Levin, M., & Roberts, H. (2000). The course of severe chronic fatigue syndrome in childhood. *Journal of the Royal Society of Medicine*, *93*(3), 129-134.
- Rangel, L., Garralda, M. E., Jeffs, J., & Rose, G. (2005). Family health and characteristics in chronic fatigue syndrome, juvenile rheumatoid arthritis, and emotional disorders of childhood. *Journal of the American Academy of Child & Adolescent Psychiatry*, *44*(2), 150-158.
- Sankey, A., Hill, C. M., Brown, J., Quinn, L., & Fletcher, A. (2006). A follow-up study of chronic fatigue syndrome in children and adolescents: symptom persistence and school absenteeism. *Clin Child Psychol Psychiatry*, *11*(1), 126-138.
- Speilberger, C., Gorsuch, R., & Lushene, R. (1970). The state trait anxiety inventory manual. *Palo Alto, Cal.: Consulting Psychologists.*
- Stratton, P., Bland, J., Janes, E., & Lask, J. (2010). Developing an indicator of family function and a practicable outcome measure for systemic family and couple therapy: the SCORE. *Journal of Family Therapy*, *32*(3), 232-258.
- van de Putte, E. M., Engelbert, R. H., Kuis, W., Sinnema, G., Kimpen, J. L., & Uiterwaal, C. S. (2005). Chronic fatigue syndrome and health control in adolescents and parents. *Arch Dis Child*, *90*(10), 1020-1024.
- van de Putte, E. M., van Doornen, L. J., Engelbert, R. H., Kuis, W., Kimpen, J. L., & Uiterwaal, C. S. (2006). Mirrored symptoms in mother and child with chronic fatigue syndrome. *Pediatrics*, *117*(6), 2074-2079.
- Viner, R., & Hotopf, M. (2004). Childhood predictors of self reported chronic fatigue syndrome/myalgic encephalomyelitis in adults: national birth cohort study. *BMJ*, *329*(7472), 941.
- Ware Jr, J. E., Kosinski, M., & Keller, S. D. (1996). A 12-Item Short-Form Health Survey: construction of scales and preliminary tests of reliability and validity. *Medical care*, *34*(3), 220-233.
- Zigmond, A. S., & Snaith, R. P. (1983). The hospital anxiety and depression scale. *Acta Psychiatr Scand*, *67*(6), 361-370.