

The management of children with chronic fatigue syndrome-like illness in primary care: a cross-sectional study

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ABSTRACT

Background

Most studies on children with chronic fatigue syndrome (CFS)/myalgic encephalomyelitis (ME) have been undertaken in tertiary care and little is known about their management in primary care.

Aim

To describe the characteristics of patients aged 5–19 years with CFS-like illness in primary care and to examine how GPs investigate and manage patients.

Design of study

Descriptive retrospective questionnaire study.

Setting

Sixty-two UK GP practices in the MRC General Practice Research Framework (GPRF).

Method

One hundred and twenty-two practices were approached; 62 identified 116 patients consulting a GP with severe fatigue lasting over 3 months. Practice nurses and GPs completed questionnaires from medical notes and patients completed postal questionnaires.

Results

Ninety-four patients were considered by a clinical panel, blind to diagnosis, to meet the Oxford CFS criteria with a fatigue duration of 3 months. Seventy-three per cent were girls, 94% white, mean age was 12.9 years and median illness duration 3.3 years. GPs had principal responsibility for 62%. A diagnosis of CFS/ME was made in 55%, 30% of these within 6 months. Fifty per cent had a moderate illness severity. Paediatric referrals were made in 82% and psychiatric referrals in 46% (median time of 2 and 13 months respectively). Advice given included setting activity goals, pacing, rest and graded exercise.

Conclusions

Patient characteristics are comparable to those reported in tertiary care, although fewer are severe cases. GPs have responsibility for the majority of patients, are diagnosing CFS/ME within a short time and applying a range of referral and advice strategies.

Keywords

children; chronic fatigue syndrome; diagnosis; patient management; primary health care.

INTRODUCTION

Prolonged unexplained fatigue including conditions such as chronic fatigue syndrome (CFS) and myalgic encephalomyelitis (ME) are increasingly reported in children.^{1–3} The chronic nature of the condition makes repeated GP visits likely but little is known about the characteristics and management of these patients in primary care. Most previous research on CFS/ME in children has taken place in tertiary care settings.^{4–6} One primary care study⁷ described 36 children in a GP special interest clinic and concluded that they were comparable to those in a tertiary setting, although given the specialist nature of the clinic, it is unclear if patients were representative of the UK population.

The joint Royal Colleges' report on CFS acknowledges that young people with CFS/ME should be managed within primary care with appropriate input from other specialists⁸ and the Chief Medical Officer's report⁹ highlighted the role of the GP in coordinating effective multidisciplinary care and considering the impact on the family. Given these recommendations it is important to know how GPs are reaching a diagnosis of CFS/ME in children and young people, how they are managing their care and whether primary care patients differ from those in other settings. The study reported here describes the characteristics of young patients (aged 5–19 years) suffering from CFS-like illness in primary care and examines how they are investigated and managed by GPs.

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How this fits in

Expert groups advocate the role of the GP in managing children suffering from CFS-like illness, but little is known about patient characteristics in this setting. The majority of young patients with CFS-like illness are being managed by GPs. Fewer patients in this sample were severe cases and from higher social classes compared to those reported in tertiary care settings. GPs gave a diagnosis of CFS and/or ME within 6 months in two-thirds of patients and made paediatric and psychiatric referrals although referral duration was variable.

METHOD

The study was carried out using the Medical Research Council's General Practice Research Framework (MRC GPRF) (www.mrc-gprf.ac.uk), a network of representative UK GP practices. Multicentre ethical approval was obtained.

Practices that had reported potentially eligible patients in a postal survey of the framework practices in 1999¹⁰ and who had indicated an interest in the new study, were contacted by the MRC GPRF in 2001 and invited to participate in this second study. Details of those practices that still had eligible patients and who remained willing to participate were passed to the research team. Nurses in these practices were asked to recruit any patients aged 5–19 years with over 3 months of severe fatigue and a significantly reduced pre-morbid level of activity, who had consulted a GP between 1 January 1999 and 31 July 2001, including those who had subsequently recovered. Non-responding practices were sent two postal reminders followed by a letter from the head of the MRC GPRF. Letters were followed up by at least two reminder telephone calls to the practice nurse leading the study.

Consent was obtained from patients and/or parents depending on the patient's age. For consenting patients the GP and the practice nurse completed questionnaires using patients' notes and patients and/or parents completed a postal questionnaire. Parents were sent questionnaires for 5–9 year olds, for 10–15 year olds questionnaires were sent to both parents and patients, and for 16–19 year olds questionnaires were sent to patients only.

The questionnaires asked for patients' sociodemographic characteristics, health in the year

before fatigue onset, pre-consultation fatigue duration, nature of onset, presenting and subsequent symptoms, severity, worst level of functional impairment, coexisting illness, investigations, diagnosis, medication, treatment and referrals.

The study inclusion criteria were deliberately broad as there are no agreed diagnostic criteria for CFS/ME in children and the illness can be variably labelled. In order to identify patients with CFS-like illness, a panel of four paediatricians experienced in the management of children with CFS were paired up to independently review an anonymised data set extracted from the questionnaires for each patient, blind to the GPs' diagnosis against the Oxford criteria for CFS in adults (Box 1).¹¹ A minimum fatigue duration of 6 months has been suggested as too long for children^{8,2} and in cases where this was not met, 3 months was accepted for the purpose of the review.

Cases meeting the criteria were considered to be cases of CFS-like illness. When the reviewers disagreed, the case was reviewed by two further paediatricians (majority verdict prevailed). Remaining discrepancies were resolved at a panel meeting.

RESULTS

Response rates

The research team were sent details of 122 practices with potentially eligible patients who were willing to participate; 62 practices (50%) subsequently took part. Reasons for non-participation were lack of eligible patients in 20 practices (16%) and lack of staffing and time in 25 practices (21%). Fifteen practices (13%) failed to respond at all.

Jarman deprivation scores for responding practices ranged from -28 to 37.61. Responding and non-responding practices did not differ in their Jarman scores ($z = -1.464$; not significant) and total population size ($z = -0.260$; not significant).

The 62 practices identified 151 eligible patients of whom 116 (77%) consented, 13 (8.6%) refused and 22 (14%) did not reply. Responders and non-responders were comparable in terms of sex and ethnicity, but non-responders were older (mean age of 15 years), and more likely to have recovered (50% versus 16%).

Complete data were obtained for 95 patients (82%); 94 were considered to have CFS-like illness, 89 fully met the Oxford CFS criteria and five only partially did but the reviewers considered a CFS-like illness likely in the context of available information. The remaining patient was considered to have depression and asthma (GP diagnosis of tiredness). The following results are based on data from the 94 patients considered to have CFS-like illness.

Patient characteristics

There were 69 girls (73%) and 25 boys (27%). Eighty-

Box 1. Oxford CFS criteria.

- ▶ Fatigue to be the principal symptom
- ▶ Of definite onset
- ▶ Fatigue to be severe, disabling and affecting physical and mental functioning
- ▶ Present for at least 6 months, if not present for at least 3 months
- ▶ Other symptoms to be present, particularly myalgia, mood and sleep disturbance
- ▶ Exclusion of established conditions known to cause chronic fatigue
- ▶ Exclusion of coexisting diagnosis of specified psychiatric disorders

eight (94%) were white British, three (3%) white other and three (3%) of other ethnic origin (one each of Asian Indian, black British and mixed other). Mean age at first consultation with GP about the fatigue was 12.9 years (standard deviation [SD] = 2.87; 95% confidence interval [CI] = 12.3 to 13.5), 12.3 years in boys (SD = 2.96; 95% CI = 11.1 to 13.6) and 13 years in girls (SD = 2.83; 95% CI = 12.5 to 13.8).

GP diagnosis

Ninety-three patients (99%) had been given a diagnosis by the GP; these were CFS ($n = 42$, 45%) post-viral fatigue ($n = 21$, 22%), chronic fatigue ($n = 12$, 13%), ME ($n = 10$, 11%), depression ($n = 4$, 4%) and one case each of an eating disorder, anxiety disorder, allergy and streptococcal infection.

In 90% of cases (84/93) GPs' diagnosis broadly agreed with that of the panel. In the nine cases where these differed, two cases had a secondary diagnosis of 'fatigue' and 'possible CFS'; in one case the GP was delaying making a diagnosis of CFS/ME until adequate evidence was obtained. In six cases GPs had considered a diagnosis of CFS/ME; in three cases this was rejected because the GP felt symptoms were inconsistent; in three cases reasons were not given.

The CFS-like illness

The duration of fatigue before the first GP consultation was a median of 4 weeks ($n = 83$; range = 1–15 months); 65 (78%) consulted their GP when they had been fatigued for less than 3 months, 17 (20%) had been fatigued for 3–5 months and 11 (13%) for 6 months or more.

Seventy-nine (84%) patients were still ill at the time of the study and for these the median illness duration from onset to questionnaire completion was 3.3 years (range = 4.5 months to 14 years).

Seventy-seven patients (82%) reported good health in the 12 months prior to illness onset. Ninety-one patients (96%) reported month of illness onset; 13 (14%) reported onset in spring (March–May), 18 (19%) in summer (June–August), 36 (39%) in autumn (September–November) and 24 (26%) in winter (December–February). χ^2 testing showed a significant difference in seasonal onset ($\chi^2 = 13.149$, 3 degrees of freedom; $P = 0.004$) with peak onset in the autumn. Thirty-five patients (38%) reported gradual onset while 58 (62%) reported a sudden onset. Type of onset was unrelated to season.

Only six patients (9%) had a first-degree relative with severe fatigue; two a parent and four siblings.

Symptoms

Symptoms noted by GPs at all consultations are reported in Table 1. Fatigue was the most frequent symptom, ($n = 100$, 94%), followed by sore throat

Table 1. All symptoms noted by the GP in any consultations with the patient.

Symptom	<i>n</i> (%)
Fatigue	94 (100)
Sore throat	65 (69)
Headaches	61 (65)
Mood disturbance	55 (58)
Sleeping difficulties	54 (57)
Myalgia	41 (44)
Abdominal pain	37 (39)
Nausea/vomiting	35 (37)
Difficulty concentrating	31 (32)
Joint pain	29 (31)
Eye pain/light sensitivity	18 (19)
Lymph node pain	14 (15)
Sensitivity to sound	5 (5)
Other ^a	52 (55)

^a*Dizziness, fainting, fever, speech difficulties, loss of voice, reduced immunity (ulcers, flu, frequent infections, upper respiratory tract infections), increased allergies, excessive sweating, changes in body temperature, changes in appetite, weight changes, skin rashes, irregular heartbeat, palpitations, hyperventilation, shortness of breath, jaundiced appearance, difficulty swallowing, sinusitis, rhinitis, nose bleeds, facial pain, social and psychological stress, loss of interest in schooling, low confidence.*

($n = 65$, 69%) and headaches ($n = 61$, 65%). The GPs assessed the principal presenting symptom to be fatigue in 88% of cases ($n = 83$), headaches in 3% ($n = 3$), abdominal pain and nausea in 3% ($n = 3$), dizziness in 2% ($n = 2$), sore throat in 2% ($n = 2$), (in one patient not stated). The mean number of symptoms was six (SD = 2.85; 95% CI = 5.64 to 6.81).

Illness severity

Eighty-eight patients (93%) had missed some school; median absence in the previous year was 23 weeks ($n = 74$; range = 1–39 weeks); 44 (59%) had missed more than half of the academic year (≥ 20 weeks).

The following criteria were used to assess severity: patient-rated functional impairment using the modified Association of Young People with ME ability scale¹² and GP-rated functional impairment. Ratings referred to the worst illness period. Agreement between scores given by both patient and GP were used to assign patients a severity category of mild, moderate or severe.

In 41 cases (44%) patients and GPs similarly assessed functional impairment; where there was inconsistency the authors independently rated illness severity using the participants' and GP assessments together with time off school, symptom severity and GPs' rating of severity. Inter-rater agreement was examined using the κ statistic (0.55; $P < 0.001$).

Overall 16 (17%) patients were rated as mildly ill (able to carry on with most previous activities), 47 (50%) as moderately ill (given up most previous activities) and 31 (33%) were severely ill (housebound, given up all previous activities).

Table 2. Tests ordered by the GP.

Test	n (%)	Number of patients with abnormal test results (%)
Haemoglobin	80 (85)	4 (5)
White cell count	75 (78)	6 (8)
Thyroid function	56 (60)	4 (7)
Kidney function	58 (62)	4 (7)
Liver function	57 (61)	8 (14)
Monospot	63 (67)	6 (10)
ESR/CRP	51 (54)	6 (12)
Urine test	30 (32)	3 (10)
Bone profile	23 (24)	0
Blood glucose	18 (19)	1 (5)
Immunoglobulins	16 (17)	1 (6)
Creatine kinase	12 (13)	0

Table 3. Health professional referrals.

	No of patients n (%)	Median time before referral in months n (%) [range]
Health professional		
Paediatrician	71 (82)	2 (68) [0–42]
Psychiatrist/psychologist	42 (48)	13 (37) [0–69]
Neurologist	9 (10)	14 (9) [0–40]
ENT	8 (9)	21 (8) [0–70]
Immunologist	7 (8)	13 (7) [0–33]
Rheumatologist	4 (5)	24 (4) [5–60]
Other hospital referrals ^a	19 (22)	22 (19) [0–72]
Referral to allied health professional		
Physiotherapist	20 (23)	13 (18) [0–52]
Homeopath	10 (11)	16 (8) [9–68]
Dietician	8 (9)	6 (7) [2–40]
CFS/ME specialist	4 (4)	24 (4) [9–66]
Occupational therapy	2 (2)	48 (2) [33–63] ^b
Other ^c	21 (24)	18 (19) [0–104]

^aGastroenterologist, endocrinologist, physician and unspecified. ^bThe value of 48 refers to the mean duration, as only two patients were referred to occupational therapy. ^cSpeech therapy, counsellor, osteopath, self-help group.

GP management

The GP had primary clinical responsibility for 58 patients (62%); 22 (24%) were managed by a paediatrician, six (6%) by a psychiatrist, two (2%) by an adult physician and one (1%) by a complementary practitioner. Care was shared between the GP and a paediatrician in four patients (4%) and between a GP and an adult physician in one patient (1%). There was no relationship between illness severity and the health professional managing patients.

Number of GP consultations ranged from 1 to 116 (median = 10). Girls consulted their GP more frequently, a median number of 12 in girls (interquartile range [IQR] = 7–28) compared to seven in boys (IQR = 3–15) ($z = -2.844$; $P = 0.004$). In patients ill for up to 2 years, the median number of consultations was seven, rising to 10 in those ill for 4 years and to 24 in those ill for 6 years or longer.

In 86 patients (91%) the GP had ordered diagnostic tests with a mean of 7 per patient (SD 4; 95% CI = 6.9 to 8.8) (Table 2). The majority of test results were normal, although liver function and ESR/CRP tests

gave the highest percentage of abnormal results (14% and 12%, respectively).

Eighty-seven patients (91%) had been referred to another health professional; 71 (82%) to a paediatrician (median of 2 months after presentation in 67 cases with available information); 33 (49%) were referred within 2 months; 42 (48%) to a psychiatry/psychology service (median time of 13 months); 20 (23%) to a physiotherapist, and two patients (2%) to an occupational therapist within an average of 4 years. The median number of referrals per patient was two (range = 0–7).

Patients were more likely to be referred to a psychiatrist if they had been ill longer (median of 52 weeks (IQR = 39–76) for those referred, compared to 29 weeks (IQR = 21–42) for non-referred patients. ($z = -3.57$; $P < 0.001$). Severity was not related to choice of referrals (Table 3).

Time taken from first consultation to a diagnosis known in 91 patients was a median of 2.8 months (range = 1 month to 6 years 5 months). Among patients with a diagnosis of CFS/ME ($n = 52$), 33 were given a diagnosis within 6 months (63%), in 13 patients (25%) this took over 6 months and in six patients (11%) over 2 years.

Antidepressants were prescribed most frequently ($n = 36$, 38%), followed by analgesics ($n = 12$, 13%) and antibiotics in 15 (16%).

For the 52 patients diagnosed with CFS/ME by their GP, the most frequently reported advice given to patients and families was pacing out activities ($n = 31$, 60%), followed by remaining active (such as continuing with school and leisure activities) ($n = 26$, 50%), identifying and setting goals ($n = 25$, 48%), a programme of graded exercise ($n = 19$, 37%), rest ($n = 12$, 23%) and reassurance only ($n = 17$, 32%).

DISCUSSION

Summary of main findings

This is the first and largest UK study of children with CFS-like illnesses, investigating the characteristics, diagnosis and management of children in primary care. The majority of children with CFS-like illness, with all ranges of illness severity presenting in primary care are being managed by their GPs. GPs are diagnosing CFS/ME relatively quickly with two-thirds given a diagnosis within 6 months of their initial consultation; in a significant minority however, the diagnosis is considerably delayed. Multidisciplinary referrals were common and the majority of patients were referred to a paediatrician, half within 2 months although there were longer delays before referral to other specialties. GPs are using a range of diagnostic labels to describe what is likely to be the same condition although their management strategy is unrelated to the diagnostic label used.

Comparison with existing literature

In terms of age, sex ratio, and ethnicity, participants were similar to those described in primary, secondary and tertiary care and had a similar range of symptoms, illness duration, high consultation rates and inconclusive laboratory test results^{2,4,5,7,13} although the only other primary care study of childhood CFS is set in a specialist clinic⁷ and may not be representative of the UK primary care population. A peak autumn onset is consistent with previous research⁴ and may be related to school year onset. Over half of our sample had missed more than 20 weeks' schooling, however fewer patients in our sample were severely housebound compared to tertiary care samples, 33% compared to 62% in tertiary settings with severity scored as a measure of self-rated global wellbeing and school absence¹⁴ and 57% in a sample of children classified as bedridden during their worst illness period.⁴

Strengths and limitations of the study

The study benefits from high patient response rates, providing us with information on a large sample of children in a primary care setting in the UK.

Although our invitation letter called for GP practices with cases of severe fatigue, we may have had a more positive response from those practices where CFS/ME is a recognised condition. Data were collected retrospectively relying on the accuracy of patients' notes, GPs' recall and subjective reporting by patients and the cross-sectional nature of the study means that patients were captured at different stages of their illness. Although retrospective assessment concluded that 99% of patients had a CFS-like condition, we were unable to establish the reasoning behind the alternative GP diagnoses. The lack of formal standardised fatigue and severity measures limit comparison between studies.

Implications for future research and clinical practice

The study suggests that the majority of children with CFS-like illness are managed by their GP. With a mean illness duration of 3 years, high consultation rates and a high level of functional impairment in up to a third of patients, the management of children with CFS-like illness in primary care will often require long-term GP support. A substantial number were not given a confirmed diagnosis and the use of established diagnostic criteria would improve recognition of CFS/ME by GPs.

The management of CFS/ME requires input from multidisciplinary teams.^{8,9,15} Our finding that 92% of patients had been referred to other health professionals suggests that GPs are aware of this need. However, the study data do not discriminate between referrals for ongoing multidisciplinary care initiated by the GP, or

made in an attempt to establish a diagnosis, or at patients' request, although in practice GPs may not make this distinction. In a minority of patients delays preceded referrals to both paediatric and psychiatry services and the time taken to establish a diagnosis was over 2 years. Future research is needed to identify the reasons behind such delays. Use of standardised criteria to establish severity is imperative if conclusions are to be drawn between studies.

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Ethical approval

The Northern and Yorkshire Ethics Committee (MREC/0/3/29)

Competing interests

The authors have declared that there are none

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