

RESULTS

Identification of children with fatigue

The project was carried out over three school terms between 3/9/07 and 15/2/08 for school 1 and over two terms between 31/10/07 and 15/2/08 for schools 2 and 3. The school rolls for children in years 7 to 11 at the time of the study were 925; 1055 and 875 children respectively (total 2855).

Figure 1 shows the flow of children through the study. A total of 461 (16.1%) children were reported to have missed 20% or more of school over a 6 week period during one or more terms. Of these, 315 (68.3%) had only a single episode off school, had a known medical illness that could explain the absence (three of these had CFS/ME and were being seen by the specialist CFS/ME service), or were known to have been on holiday or to have been truanting. The remaining 146 children (5.1% of school roll) had missed 20% or more school without an identifiable cause and were invited to a clinical review at school. Of these, 112 (76.7%) attended the school clinic.

Forty eight children (42.9% of those assessed, 2.1% of the school roll) described significant fatigue as a major cause of their absence and were given appointments for more detailed assessments at the specialist service. Of the remaining 64 children assessed, 30 had medical problems that explained their absence. These included recurrent headaches/migraines in 9 children; infections (e.g. tonsillitis, diarrhoea and vomiting or viral upper respiratory tract infections) in 8 children; 24 children were missing school because of social or emotional problems; 8 because they were truanting and 2 had a diagnosis of CFS/ME confirmed by a paediatrician but had not been referred to the specialist service.

Forty one children with fatigue attended further outpatient assessment by the specialist service, while one child was severely affected so was assessed at home. Of these, 23 were diagnosed with CFS/ME that was thought to be the main reason for their reduced school attendance, and were offered rehabilitation. Of the 19 children assessed but not diagnosed with CFS/ME, 14 had fatigue secondary to mood disorders (mostly anxiety and depression). Of these, 11 were referred to local Child and Adolescent Mental Health Services. Five children had other medical problems that were thought to be the cause of their fatigue (sleep apnoea (2), recurrent tonsillitis (1), migraine (1), undiagnosed sleep disorder not CFS/ME (1)).

In summary, a total of 28 children (6.1% of the 461 children who were missing 20% or more of school) had CFS/ME. These included 23 (82.1%) new cases of CFS/ME identified through school clinics, and 2 (7.1%) children identified as having CFS/ME previously diagnosed by a paediatrician but not known to the school or to the specialist service. Only 3 (10.7%) children with CFS/ME were already being seen by the CFS/ME service and so were not offered further assessment. The estimated prevalence of CFS/ME sufficiently severe to cause a child to miss 20% or more of school was $28/2855 = 0.98\%$ (95% CI 0.65% to 1.41%). This is likely to be an underestimate as some of the 39 children who were invited to but did not attend school clinics were likely to have had CFS/ME.

Comparison of children with CFS/ME identified through school with those referred by health services

Table 1 compares the characteristics of the 23 children identified as having CFS/ME through school surveillance and who attended the specialist service with those of 604 children with CFS/ME referred to the specialist service by a health professional.

1 Children identified through surveillance were less fatigued, had less pain, had fewer
2 symptoms and were less physically disabled than those referred to the specialist
3 service. However, the length of time for which children had been unwell (mean 20.2
4 months, SD 17.1) was comparable. There was little evidence that levels of anxiety
5 and depression differed between the two groups.
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14 *Outcomes of treated CFS/ME in children identified through school*

15 Of the 23 children diagnosed as having CFS/ME, 4 were given advice about
16 managing CFS/ME but did not attend follow up appointments or return follow up
17 inventories. Of the remaining 19 children, 12 (63.2%) were attending full time school
18 by 6 months, of whom 6 had made a full recovery and were attending full time school
19 with minimal advice after 6 weeks. One housebound child improved and was
20 attending school part time every day by 3 months. For the remaining 6 children,
21 school attendance did not alter significantly at 6 months.
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DISCUSSION

In school based clinics, 28/2855 (0.98%) of children aged 11 to 16 years were missing 20% or more of school and were diagnosed with CFS/ME. Only 3/28 (10.7%) of these had received a diagnosis and accessed specialist treatment. The duration of symptoms in those identified through clinics was comparable to those referred to via health services, but they were not as severely affected. Outcomes following treatment were encouraging.

There are several possible reasons why children missing significant amounts of school with CFS/ME are not identified. Those with mild or moderate CFS/ME may not see their GP, or may not be recognised as having CFS/ME if they are seen.

Alternatively, GPs and paediatricians may not be aware of specialist CFS/ME services or may feel that the child's CFS/ME is not sufficiently serious to warrant a referral. Children referred to the Bath Specialist CFS/ME service are more severely affected than those described by other specialist services¹². Differences in characteristics at referral did not appear to be explained by the length of time from symptom onset or co-morbid mood problems.

Strengths and Weaknesses

Children were offered assessment regardless of whether their absence had been classified as "authorised" or "unauthorised". This is because fatigue that is causing a child to miss school may have been ascribed to social or educational problems, leading her absence to be classified as unauthorised. All children given a diagnosis of CFS/ME had been screened for other medical and emotional causes of fatigue, were prospectively characterised and followed up with well validated inventories.

Our clinics were conducted in three schools in one city in the South West of England in which there is already a well established specialist CFS/ME service. The prevalence of CFS/ME, and reasons for school absence, may be different in regions

1 where there is no specialist service and may vary according to the prevalence of
2 socio-economic factors that impact on school attendance. One of the schools was a
3 girls only school. A cross sectional study of British children found little evidence that
4 female gender was a risk factor for CFS/ME diagnosed using the CDC criteria or
5 parental report³ although adult CFS/ME is more common in women than in men¹⁸. If
6 the prevalence of CFS/ME were higher in girls than boys aged 11-16 then we would
7 have overestimated the overall prevalence. We were not able to obtain information on
8 children who did not attend the specialist service for assessment.
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10 *Results in context with previous literature*

11 The prevalence of diagnosed CFS/ME in this study (3/2855 = 0.11%) is similar to the
12 prevalence of 0.07% reported in two other school surveys¹⁹ and the prevalence of
13 CFS/ME diagnosed in primary care (0.06%)². We identified more children with
14 CFS/ME than a recent British study which identified chronic fatigue syndrome in only
15 7 of 8839 children enrolled in 10 schools²⁰. However that study screened children
16 who missed 20% of school and whose absence was recorded as “medical absence”
17 whereas we assessed children who missed school for any reason, including those
18 whose absence was classified as unauthorised. Our prevalence estimate was also
19 higher than estimates of between 0.1% and 0.5% from population based studies³⁻⁵,
20 perhaps because these used the adult definition of CFS/ME, which requires 6 months
21 of fatigue and four additional symptoms. Our prevalence estimate is lower than that of
22 2.3% for CFS/ME with three months’ duration, from a study in twins²¹.
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52 *Implications*

53 Surveillance is particularly important for diseases, such as CFS/ME, where there may
54 be poor parental recognition of symptoms and presentation for assessment²². This
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1 project suggests that undiagnosed CFS/ME can be identified through school based
2 clinics, and may be an important and under-appreciated cause of school absence in
3 children aged 11-16 years. We also identified potential medical or psychological
4 causes of fatigue in children who were not diagnosed with CFS/ME. Although we do
5 not have outcome data for these children, the identification of a potential cause is
6 likely to be of benefit in this group. Reduced school attendance is associated with
7 worse educational attainment, and may increase the risk of unemployment²³.
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10 Surveillance can be justified when improved outcomes in those identified and treated
11 outweigh both harms and costs. The NICE guidelines¹³ recommend that even those
12 children who have mild CFS/ME should be referred to a specialist CFS/ME service
13 after 6 months. There is some evidence that particular treatments (graded exercise
14 therapy (GET) and cognitive behaviour therapy (CBT) are moderately effective in
15 children⁸⁻¹¹, while the recently reported PACE trial provided strong evidence that
16 these treatments are moderately effective in adults²⁴. Large scale randomised
17 controlled trials are needed in children, however our finding that more than half
18 (12/23) of children with school-clinic-diagnosed CFS/ME returned to full time school
19 within six months of the start of treatment compares well with results from other follow
20 up studies²⁵, suggesting that offering specialist services to children with CFS/ME
21 identified through surveillance may reduce school absence. Nonetheless, it would be
22 of interest to evaluate whether school nurses, rather than doctors, can undertake the
23 initial assessments in school clinics, and to conduct randomised controlled trials
24 evaluating the effectiveness and cost-effectiveness of school based surveillance
25 programmes for CFS/ME. Further studies are needed to investigate the potential for
26 harm in offering surveillance for fatigue to children who do not have a medical
27 diagnosis to explain their absence from school.
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4 In conclusion, school based clinics are feasible and have the potential to identify
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6 children with CFS/ME, which may reduce school absence and its harmful
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8 consequences. Together with referral to specialist services, school based clinics have
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10 the potential to improve overall school attendance.
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Table 1 Characteristics of children with CFS/ME identified through school project (school CFS/ME) compared to those with CFS/ME referred to a specialist CFS/ME service (service CFS/ME)

	<i>School CFS/ME</i>		<i>Service CFS/ME</i>		Mean Difference (95% CI)	P value
	N		N			
Age yrs: mean (SD)	23	14.6(1.5)	604	14.3 (2.8)	-0.38 (-1.5, 0.8)	0.53
Female N (%)	23	17 (74)	607	422 (69.5)		0.20
Fatigue score: mean (SD)	22	20 (5.7)	541	24.4 (5.1)	4.4 (2.2, 6.6)	<0.001
Pain score: mean (SD)	16	37.4 (29.4)	503	46.9 (29.7)	9.5 (-5.3, 24.3)	0.21
SF36: mean (SD)	22	25.5 (3.9)	526	19.8 (5.2)	-5.7 (-7.9, -3.5)	<0.001
Number of symptoms: mean (SD)	22	6.6 (2.2)	582	8.5 (2.5)	1.86 (0.80, 2.93)	<0.001
Depression (HADS): mean (SD)	23	6.7 (2.2)	399	7.3 (3.7)	0.6 (-0.9, 2.16)	0.43
Anxiety(HADS): mean (SD)	23	9.4 (4.0)	400	8.4 (4.3)	-1.0(-2.8, 0.78)	0.27
Anxiety(SCAS): mean (SD)	22	35.2 (20.1)	471	29.3 (17.8)	-6.0 (-13.6, 1.7)	0.13
% school attendance: mean(SD)	23	65.9 (28.7)	569	38.6 (36.1)	-27.3 (-42.6, -12.01)	<0.001
Time to assessment (months): mean (SD)	21	22.5 (18.3)	590	25.3 (25.3)	2.7 (-8.2, 13.7))	0.62

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Competing Interests

The authors declare that (1) EC, AE and JS did not receive any financial support from companies for the submitted work (2) EC, AE and JS have no relationships with any companies that might have an interest in the submitted work in the previous 3 years; (3) their spouses, partners, or children have no financial relationships that may be relevant to the submitted work; and (4) AE and JS have no non-financial interest that may be relevant to the submitted work. EC is a medical advisor for the Association for Young people with ME (AYME).

Authors contributions

EC conceived the idea for this study; conducted the school clinics; analysed the data with support from JS and wrote the first draft of the paper. AE contributed to writing, and the interpretation of the data helping to critically revise the paper. JS helped with the analyses and contributed to writing the paper. All authors read and approved the final manuscript. All authors had full access to all of the data in the study. Dr Crawley will act as Guarantor for this paper.

Data sharing: no additional data available.

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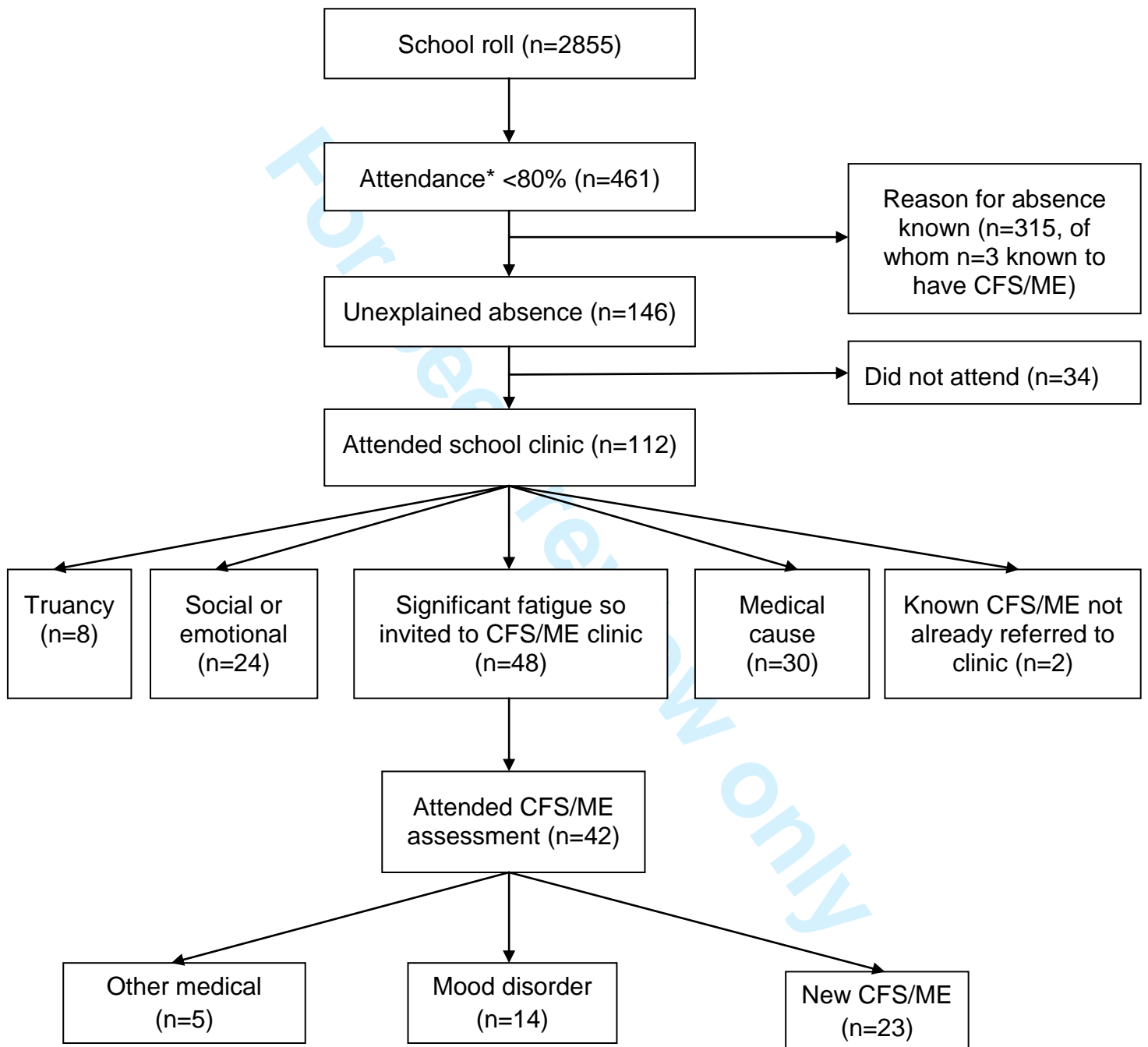
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Figure 1: Flow of children through study



*attendance <80% for at least 6 weeks in any term over 3 terms

STROBE STATEMENT: **Unidentified Chronic Fatigue Syndrome (CFS/ME) is a major cause of school absence:
school based surveillance study**

	Item No	Recommendation	
Title and abstract	1	(a) Indicate the study's design with a commonly used term in the title or the abstract	√
		(b) Provide in the abstract an informative and balanced summary of what was done and what was found	√
Introduction			
Background/rationale	2	Explain the scientific background and rationale for the investigation being reported	√
Objectives	3	State specific objectives, including any prespecified hypotheses	√
Methods			
Study design	4	Present key elements of study design early in the paper	√
Setting	5	Describe the setting, locations, and relevant dates, including periods of recruitment, exposure, follow-up, and data collection	√
Participants	6	(a) <i>Cohort study</i> —Give the eligibility criteria, and the sources and methods of selection of participants. Describe methods of follow-up	√
		<i>Case-control study</i> —Give the eligibility criteria, and the sources and methods of case ascertainment and control selection. Give the rationale for the choice of cases and controls	
		<i>Cross-sectional study</i> —Give the eligibility criteria, and the sources and methods of selection of participants	
Variables	7	(b) <i>Cohort study</i> —For matched studies, give matching criteria and number of exposed and unexposed	NA
		<i>Case-control study</i> —For matched studies, give matching criteria and the number of controls per case	
Variables	7	Clearly define all outcomes, exposures, predictors, potential confounders, and effect modifiers. Give diagnostic criteria, if applicable	√
Data sources/ measurement	8*	For each variable of interest, give sources of data and details of methods of assessment (measurement). Describe comparability of assessment methods if there is more than one group	√
Bias	9	Describe any efforts to address potential sources of bias	√
Study size	10	Explain how the study size was arrived at	
Quantitative variables	11	Explain how quantitative variables were handled in the analyses. If applicable, describe which groupings were chosen and why	
Statistical methods	12	(a) Describe all statistical methods, including those used to control for confounding	√
		(b) Describe any methods used to examine subgroups and interactions	

		(c) Explain how missing data were addressed	
		(d) <i>Cohort study</i> —If applicable, explain how loss to follow-up was addressed	
		<i>Case-control study</i> —If applicable, explain how matching of cases and controls was addressed	
		<i>Cross-sectional study</i> —If applicable, describe analytical methods taking account of sampling strategy	
		(e) Describe any sensitivity analyses	
Results			
Participants	13*	(a) Report numbers of individuals at each stage of study—eg numbers potentially eligible, examined for eligibility, confirmed eligible, included in the study, completing follow-up, and analysed	√
		(b) Give reasons for non-participation at each stage	√
		(c) Consider use of a flow diagram	√
Descriptive data	14*	(a) Give characteristics of study participants (eg demographic, clinical, social) and information on exposures and potential confounders	√
		(b) Indicate number of participants with missing data for each variable of interest	√
		(c) <i>Cohort study</i> —Summarise follow-up time (eg, average and total amount)	
Outcome data	15*	<i>Cohort study</i> —Report numbers of outcome events or summary measures over time	√
		<i>Case-control study</i> —Report numbers in each exposure category, or summary measures of exposure	
		<i>Cross-sectional study</i> —Report numbers of outcome events or summary measures	
Main results	16	(a) Give unadjusted estimates and, if applicable, confounder-adjusted estimates and their precision (eg, 95% confidence interval). Make clear which confounders were adjusted for and why they were included	
		(b) Report category boundaries when continuous variables were categorized	
		(c) If relevant, consider translating estimates of relative risk into absolute risk for a meaningful time period	
Other analyses	17	Report other analyses done—eg analyses of subgroups and interactions, and sensitivity analyses	
Discussion			
Key results	18	Summarise key results with reference to study objectives	√
Limitations	19	Discuss limitations of the study, taking into account sources of potential bias or imprecision. Discuss both direction and magnitude of any potential bias	√
Interpretation	20	Give a cautious overall interpretation of results considering objectives, limitations, multiplicity of analyses, results from similar studies, and other relevant evidence	√
Generalisability	21	Discuss the generalisability (external validity) of the study results	√
Other information			
Funding	22	Give the source of funding and the role of the funders for the present study and, if applicable, for the original study on which the present article is based	√

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