SHORT REPORT

A feasibility study comparing two treatment approaches for chronic fatigue syndrome in adolescents

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Chronic fatigue syndrome (CFS) involves severe disabling fatigue that affects physical and mental functioning.¹ Reported prevalence varies between 0.05% and 2% depending on definitions and methodologies.^{2–3} There are significant short and long term effects on young people and their families, including long term school non-attendance.^{4–5} Most reported studies are not randomised, are from a variety of different clinical settings, and show variable outcomes: 5–20% being seriously incapacitated in the longer term, with larger numbers having residual symptoms.^{2–6–9}

There is lack of consensus about the effectiveness of the main treatment approaches. These range from approaches that involve high levels of rest, through “pacing” to more active rehabilitation. More active rehabilitation has developed as a treatment focusing on graded rehabilitation, and more recently cognitive behavioural techniques. They are now more likely to integrate a range of approaches to address physiological, psychological, social, and systemic aspects of the syndrome.

Strong feelings about treatment can be expressed by proponents at either end of the spectrum, sometimes with criticism of alternative approaches. Some advocating active rehabilitation quote research saying that prolonged excessive inactivity has adverse consequences.¹⁰ Conversely advocates of prolonged rest who find support from internet sites,¹¹ some specialist clinics,⁴ and some general practitioners¹² believe active rehabilitation can be damaging. Criticism of overzealous rehabilitation can tend to undermine all rehabilitation, and criticism of rest can tend to undermine all energy management programmes. This polarisation of views conspires against healthy discussion on middle ground treatments. This makes open minded randomised controlled trials essential to provide evidence to guide treatment and restore confidence in treatment centres, particularly as new initiatives to establish centres of excellence unfold.

This study assesses the feasibility of a larger treatment trial comparing the effectiveness of the two current most common treatment approaches. We sought to explore the acceptability of treatments and the numbers needed to show a meaningful difference between treatments.

METHODS

The study was undertaken, with ethical committee approval, at York Hospital, which covers a population of 300 000. A

total of 328 local professionals from health, social services, and education identified young people known to them with CFS or with more than two weeks off school because of physical symptoms but no clear diagnosis. These and any subsequent new cases identified over a 15 month period were assessed by a paediatrician (DWB). Those meeting the Oxford criteria¹ for CFS (using the recommended modification for children of three months’ fatigue) were given further information about the study. Exclusions were other fatiguing medical conditions and pre-existing ongoing treatment for CFS. Informed consent was obtained from participants.

Young people were randomised to one of two treatment groups using remote randomisation at York University. This stratified for age, sex, and mobility. Over one year clinic appointments were weekly for one month, 2 weekly for the next three months, 3 weekly for two months, and 4 weekly for six months. Three clinicians (BW, CW, BA) conducted both treatment options using treatment manuals. The paediatrician (DWB) saw all young people every 12 weeks.

Research assessment interviews were conducted blind to treatment, by a separate researcher (JJ) prior to treatment and at one year after treatment began.

Both treatment arms included:

- A strong emphasis on collaboration with patient and family
- Support and advice to establish a normal eating pattern, a balanced healthy diet, and healthy sleep patterns
- Cooperative work between child mental health professionals and paediatricians.

The two treatment arms were characterised in the following ways:

- “Pacing” is described in a document presented to the Chief Medical Officer’s working group on CFS/ME.¹³ and included: (1) pacing activity to the changing needs and responses of the body by exercising to the point of tolerance, avoiding overexertion; (2) managing energy within an overall limit (“glass ceiling”); (3) resting when necessary, but avoiding total rest; (4) avoiding physically and/or emotionally stressful situations until ready; and (5) tailoring return to school to the needs of the young person,

Table 1 Treatment option by age group, sex, and length of illness

Treatment option (n)	Gender		Age			Median length of illness in months
	Male	Female	0–11	12–14	15–19	
Pacing	2	4	0	4	2	14.5
Stairway	3	4	1	3	3	12.0
p value	1.00*		1.00†			0.248

*Fisher’s exact test.

†Mann-Whitney U test.

taking careful heed of symptoms, the child, and the family.

- *The STAIRway to Health* programme involved a Structured Tailored Incremental Rehabilitation programme. This incorporated some aspects of previously researched approaches.^{14 15} In particular time was spent:
 - providing a holistic understanding of CFS that moved away from an exclusively physical or an exclusively psychological understanding of the illness
 - explaining vicious cycles that exacerbate illness, including those of nutrition, sleep patterns, physical deconditioning, social isolation, educational estrangement, and emotional cycles (including loss of self-esteem and confidence)
 - bolstering adaptive coping strategies and re-evaluating negative attributions about the illness and the future.

Collaboratively agreed targets were set around six areas: nutrition, activity, sleep, social activity, emotional factors, and school reintegration. The young person monitored these using a diary. Young people were not expected to do any activities they had not agreed.

As the treatment progressed, young people were encouraged to discuss constructively how their lifestyles,

temperaments, and approaches to life may impact on illness or recovery. A tailored gradual return to school was planned where possible, as was a gradual return to normal social activity.

Outcome measures

The following five main outcome measures were used.

- Global health
 - rated from the Child Health Questionnaire¹⁶ by the child
 - rated by the clinician at the end of treatment
- Activity
 - rated from the Child Health Questionnaire¹⁶ by the child
 - rated by the paediatrician and child using the Young Person Functional Ability Scale¹⁷
- School attendance (percentage of possible days attended in six month periods)
- Fatigue on a self report measure (using the 14 item version)¹⁸
- Emotional symptoms on self report measures
 - depression¹⁹
 - anxiety.²⁰

Table 2 Mean scores for outcome measures before and after treatment

	Pacing				Stairway to health			
	Mean (SD)	25th centile	Median	75th centile	Mean (SD)	25th centile	Median	75th centile
Global health (1–5; higher score worse health)								
Global health rating before treatment	4.20 (0.837)	3.50	4.00	5.00	4.00 (1.095)	3.50	4.00	5.00
Global health score after treatment	4.20 (0.837)	3.50	4.00	5.00	2.20 (0.447)	2.00	2.00	2.50
Child activity ratings (1–4; higher score fewer difficulties, 4 = no problems)								
Difficulty doing highly exertional activities (before) child rated	1.80 (0.837)	1.00	2.00	2.50	1.33 (0.516)	1.00	1.00	2.00
Difficulty doing highly exertional things (after)	2.00 (1.414)	1.00	1.00	3.50	3.20 (0.827)	2.50	3.00	4.00
Difficulty doing moderately exertional activities such as swimming (before)	2.80 (1.304)	1.50	3.00	4.00	1.83 (1.169)	1.00	1.50	2.50
Difficulty doing moderately exertional activities such as swimming (after)	2.60 (1.517)	1.00	3.00	4.00	4.00 (0)	4.00	4.00	4.00
Difficulty walking and climbing several flights of stairs (before)	3.00 (0.707)	2.50	3.00	3.50	2.67 (1.366)	1.00	3.00	4.00
Difficulty walking and climbing several flights of stairs (after)	3.20 (0.837)	2.50	3.00	4.00	4.00 (0)	4.00	4.00	4.00
Difficulty climbing one flight of stairs (before)	3.60 (0.894)	3.00	4.00	4.00	3.17 (1.169)	2.50	3.50	4.00
Difficulty climbing one flight of stairs (after)	3.40 (0.894)	2.50	4.00	4.00	4.00 (0)	4.00	4.00	4.00
Difficulty getting in and out of bed (before)	3.40 (0.894)	2.50	4.00	4.00	2.83 (1.329)	1.75	3.00	4.00
Difficulty Getting in and out of bed (after)	3.80 (0.447)	3.50	4.00	4.00	4.00 (0)	4.00	4.00	4.00
Paediatrician activity ratings (YPAS¹⁷) (0–100%; 100% is fully healthy and active)								
Baseline Physical Activity Score (YPAS)	67.00 (18.23)	52.5	75	77.5	52.50 (22.75)	31.25	55	75
Exit Physical Activity Score	68.50 (27.02)	40	80	91.25	81.25 (17.59)	60	87.5	95.63
School attendance (percentage of possible half days in six month period)								
School attendance in the 6 months before treatment as a percentage	45.74 (29.92)	16.75	53.00	71.10	45.25 (40.90)	0.00	45.75	86.00
School attendance at the end of treatment	28.12 (31.82)	0.00	16.20	62.20	73.00 (37.53)	49.50	89.50	95.75
School attendance in the six months after treatment concluded	28.70 (36.24)	0.00	14.50	64.50	84.60 (34.8)	76.90	98.00	100.00
Fatigue scale¹⁸ (0–42; higher score worse fatigue)								
Fatigue score before treatment	23.20 (9.23)	13.50	26.00	31.50	32.17 (6.113)	26.00	34.00	36.25
Fatigue score at the end of treatment	18.00 (6.519)	12.00	18.00	24.00	14.00 (9.582)	6.25	14.00	21.75
Birleson Depression Scale¹⁹ (0–36; higher score lower mood)								
Depression score before treatment	14.00 (5.657)	9.00	15.00	18.50	15.83 (5.529)	10.00	15.50	21.00
Depression score at the end of treatment	12.60 (6.580)	7.50	15.00	16.50	10.67 (4.844)	7.50	11.00	13.50
Anxiety score²⁰ (0–21; higher score more symptoms)								
Anxiety score before treatment	6.80 (3.56)	3.00	9.00	9.50	10.17 (3.71)	6.75	9.50	12.50
Anxiety score at the end of treatment	6.60 (4.73)	2.00	7.00	11.00	6.00 (3.63)	3.00	6.50	9.25

Table 3 Analyses of covariance for each outcome

	Difference (95% CI) All differences show more improvement in STAIRway arm than pacing arm	F (df = 1,8)	p value	n for 80% power
School attendance comparing six months prior to study to last six months of treatment (percentage)	45.1 (−1.8 to 92.0)	4.9	0.057	18
School attendance comparing six months prior to study to six months post study (percentage)	56.1 (6.3 to 105.7)	6.8	0.032	14
Difficulty doing highly exertional activities (child rated)*	1.46 (−0.33 to 3.25)	3.7	0.095	36
Difficulty doing moderately exertional activities such as swimming*	1.56 (−0.20 to 2.33)	4.4	0.075	20
Difficulty walking and climbing several flights of stairs*	0.93 (0.02 to 1.84)	5.8	0.046	16
Difficulty climbing one flight of stairs*	0.71 (−0.18 to 1.61)	3.5	0.10	26
Difficulty getting in and out of bed*	0.31 (−0.17 to 0.78)	2.4	0.17	40
Young Person Functional Ability Scale ¹⁷ (percentage score rated by paediatrician)	17.0 (−17.0 to 51.0)	1.3	0.28	56
Anxiety score ²⁰ (0–21 child rated anxiety subscale, Hospital Anxiety and Depression Scale)	−1.60 (−8.31 to 5.10)	0.30	0.60	260
Depression score ¹⁹ (0–36 Birmleson Depression Rating Scale)	−2.99 (−10.0 to 4.06)	1.0	0.36	86
Fatigue score ¹⁸ (0–42 14 item version)	−5.2 (−19.8 to 9.49)	0.67	0.44	120
Global health rated by child (Child Health Questionnaire), 1 = excellent, 5 = poor	−1.8 (−0.94 to −2.74)	23.4	0.002	10†

Analysis uses the "before" score as covariate, and the treatment group as independent variable. The total sample size was 11, because of two dropouts. The 95% confidence intervals (CI) describe the difference with pacing subtracted from stairway. These show greater improvements across measures for the STAIRway arm. The effects presented are for the effect of treatment, controlling for baseline score. In the n for power column, our data are used to estimate future study sample sizes required for 80% power (alpha=0.05), assuming that the effect size was consistent with this study.

*Scale 0–4 with 4 being best score (fully healthy). For the first eight items positive scores represent greater improvements in activity and occupation for the STAIRway arm. For the last four items negative scores represent greater reduction in negative symptoms for the STAIRway arm.

†Current sample size provided 98% power for significant effect.

Analysis

The data were analysed with SPSS using parametric and non-parametric bivariate statistics. For each parametric outcome measure, analysis of covariance was used, with the baseline score as a covariate and the treatment option being used as a predictor. As this was a pilot study, we were interested in finding a recommended future sample size that would be required to detect a difference equal to that found in this study. This was calculated for 80% power at 5% significance based on the non-centrality parameter from the current study. This assumes that the effect size in this study is an unbiased estimator of the effect size in the population.²¹

RESULTS

Fourteen young people had newly diagnosed CFS; one declined involvement in the study. Thirteen (age range 8.9–16.9 years) gave informed consent. They were all in mainstream schools and were incapacitated by CFS to the point of not being able to attend. Six had been unwell for less than 1 year, five for between 1 and 2 years, and two for longer than 2 years. All were markedly restricted in their ability to walk from the house, but none were permanently bed or wheelchair bound. Randomisation resulted in young people being distributed as in table 1. The results are shown in tables 2 and 3.

At the end of the study 100% of children in the STAIRway to Health arm showed completely healthy scores for all activity up to and including moderate exertional activities. No child had any problems with bike riding, swimming, and similar activities. In the pacing arm, 60% had problems in all three of these activities and still had some difficulties getting around school, compared with no problems in the STAIRway arm. Forty per cent in the pacing group still had problems with bending and lifting.

DISCUSSION

Statistical significances found should be treated with caution given the small sample sizes. The pacing programme used showed little improvement in activity scores rated by child or clinician and a deterioration in school attendance, whereas

activity and school attendance were improved markedly in the STAIRway to health arm. The differences were clinically large. Global health improved in both arms although more in the STAIRway arm than the pacing arm whether measured by child or clinician.

This study suggests that contrary to the fears of some, active rehabilitation does not appear to make young people worse if managed correctly, although a larger study would be needed to explore whether such problems may affect a minority.

The apparent acceptability of these treatments, with low drop out rates is encouraging. Only one child did not consent to the study. Two dropped out, one from each arm of the study. This represents 11 out of 14 (79%) of those consecutively diagnosed and approached. The centre concerned was highly motivated to engage and work positively with families. Whether this could be transferred to clinic settings remains to be seen. One boy in the STAIRway arm received an antidepressant, a relatively low rate given reported use of up to 94% in one series.⁹ The study has been useful in the development of treatment manuals.

There are few RCTs looking at children under 18,²² although work is underway in London and Holland. This study is too small to be conclusive, but shows that a larger study is feasible and that these treatments are acceptable to patients and families. We would strongly recommend a well designed and larger multicentre trial.

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IMAGES IN PAEDIATRICS

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Toys and potentially lethal games

A 15 year old was brought to our emergency department with mild headache after having been shot at contact range with an air pistol. Examination revealed a circular entry wound of 1 cm on the forehead without a corresponding exit wound. Neurologically there were no focal signs. Skull x ray examination showed a radio-opaque foreign body in the frontal lobe. Computed tomography scan showed pellet fragments in the left frontal lobe with surrounding haemorrhage. The child was referred to the regional neurosurgeons for further management.

Air pistols are known to inflict potentially lethal injuries such as extensive brain damage, traumatic aneurysms, and penetrating injuries of the abdomen.¹ There have been reported incidents of suicides and non-accidental injuries with air pistols.² Shanon *et al* noted that 85% of the injuries, where the mode of injury was known, were self inflicted while playing with air guns; 12.7% of their patients had long term disabilities.³

Air pistols, despite their capacity to inflict hazardous injuries, are still used as toys and continue to be mishandled by children. Increased public awareness of the hazardous nature of air pistols is

required to prevent their mishandling. Educating the public in appropriate handling and supervision of children while playing with these deceptively harmless but lethal toys combined with a stricter legislation could reduce the incidence of these injuries. Physicians should be aware that some of these patients may appear clinically stable with minimal external injuries and still have significant trauma to the internal organs.

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