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Can pacing self-management alter physical behavior and symptom severity in chronic fatigue syndrome? A case series

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Abstract—Given the lack of evidence in support of pacing self-management for patients with chronic fatigue syndrome (CFS), we examined whether physical behavior and health status of patients with CFS would improve in response to a pacing self-management program. We performed an observational study of pacing self-management in seven CFS patients using a single-case study design. Stages A1 and A2 (7-day assessment periods) of the A1-B-A2 design corresponded to the baseline and posttreatment measurements of physical behavior (real-time activity monitoring) and health status (self-reported measures), respectively. Stage B (3 weeks of treatment) consisted of three individual treatment sessions of pacing self-management. When comparing pre- versus posttreatment data, we found that the patients' ability to perform daily activities and the severity of their symptom complexes were improved ($p = 0.043$). Concentration difficulties, mood swings, muscle weakness, and intolerance to bright light improved as well. A statistically significant decrease in the mean time spent doing light activity (<3 metabolic equivalents) was observed, but a change in the way physical activity was spread throughout the day was not. We found that 3 weeks of pacing self-management was accompanied by a modest improvement in symptom severity and daily functioning. The outcome of the present study calls for a randomized controlled clinical trial to examine the effectiveness of pacing self-management for people with CFS.

Key words: activity, activity peak, behavior, CFS, chronic fatigue, pacing, rehabilitation, self-management, syndrome, therapy.

INTRODUCTION

People with chronic fatigue syndrome (CFS) often report a fluctuating pattern to their symptoms, including their physical and cognitive capabilities. A clinical study of CFS patients showed high variability in mental and physical fatigue over a 4-week period [1]. Too vigorous exercise [2–4] or even a 30 percent increase in activity [5] can frequently trigger a relapse, which may consequently explain at least part of the fluctuating symptom pattern commonly seen in CFS. In-line with these results are the findings that (1) the lifestyle of CFS patients is

Abbreviations: CFS = chronic fatigue syndrome; CFS-APQ = Chronic Fatigue Syndrome-Activities and Participation Questionnaire; CIS = Checklist Individual Strength; COPM = Canadian Occupational Performance Measure; MET = metabolic equivalent; PACE = Pacing, Graded Activity, and Cognitive Behavior Therapy: A Randomized Evaluation; SD = standard deviation; SF-36 = Medical Outcomes Study 36-Item Short Form Health Survey; VAS = visual analog scale.

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characterized by activity peaks followed by very long rest periods [6], (2) a premorbid overactive lifestyle may play a predisposing and/or initiating role in CFS [7], and (3) continuing to be active despite increasing fatigue is likely to be a crucial step in the development of CFS [8]. It has been shown that specific activities, which were expected to result in high fatigue levels, were less frequently performed by CFS patients and, furthermore, that high fatigue expectations were related to low activity levels [9]. On the other hand, during short periods, patients with CFS are generally able to perform light to moderate activity or exercise (40% of peak oxygen capacity) without exacerbating their symptoms or affecting their cognitive performance [10–11].

From these findings, including activity management in the comprehensive treatment of people with CFS seems plausible. Activity management is generally included in cognitive behavioral programs for CFS, and evidence exists that supports the effectiveness of cognitive behavioral therapy for patients with CFS [12]. Up to date, no data have been published on the extent to which activity management contributes to the effectiveness of cognitive behavioral therapy. Cognitive behavioral therapy could possibly be equally as effective without the inclusion of activity management. However, cognitive behavioral therapy might rely (in part) on activity management for its effectiveness. In addition, the availability of cognitive behavioral therapy for CFS patients in many countries is limited because of a lack of therapists. Cognitive behavioral therapists often work in specialized fatigue centers, which leads to capacity problems and practical barriers to those patients who live long distances from these centers. Other cognitive behavior therapists work in private practices, which generates financial obstacles for the patients. For all these reasons, studies examining the potential benefits of less time-consuming treatments are warranted.

Graded exercise therapy has been reported to be effective for CFS [13] and is included in many cognitive behavioral therapy programs for CFS. Although no evidence currently exists that graded exercise therapy, on average, causes harm to CFS patients [13], preventing exercise-induced exacerbations in symptoms when applying exercise therapy to patients with CFS remains important, especially in terms of treatment compliance [14]. Initial success of exercise therapy in CFS is most likely due to the patients realizing that exercise can be safely undertaken without the consequence of relapse. This realization assists CFS patients in abandoning any avoidance behaviors to which they may have previously adhered [15]. Therefore, a self-management program that teaches people with CFS to explore all kinds of physical activity appropriate for their individual physical capabilities appears necessary. These types of self-management techniques used together with, or prior to, a graded exercise program appear warranted for people with CFS [14].

Self-management for people with CFS involves encouraging the patients to pace their activities and respect their physical and mental limitations [16–17]. This strategy has been termed “pacing” and involves encouraging the patient to achieve an appropriate balance between activity and rest in order to avoid exacerbating his or her symptoms. It requires the patient to set realistic activity goals on a daily basis [16,18] and to regularly monitor and manipulate activity in terms of intensity, duration, and rest periods in order to avoid possible overexertion, which can worsen symptoms [16,18]. Pacing takes into account the considerable fluctuations in symptom severity [16] and delayed recovery from exercise that typically occur in patients with CFS [19]. The pacing approach is in-line with recent observations regarding the interactions between malfunctioning of the immune system, physical activity, and symptoms in CFS patients [14]. It advocates a symptom-contingent rather than a time-contingent approach. In addition, some patients with CFS are reluctant to undertake psychological treatments, such as cognitive behavioral therapy, for what they believe to be a physical condition. Pacing self-management techniques encourage a behavioral change and at the same time acknowledge the physical aspects of the illness.

Given the lack of evidence in support of pacing self-management as a sole treatment for those with CFS, we undertook an observational study of pacing self-management in seven CFS patients using a single case study design. We examined whether pretreatment physical behavior and health status of patients with CFS differed from posttreatment physical behavior and health status. The outcome of the present single case study will allow us to judge the feasibility of setting up a randomized controlled clinical trial to examine the effectiveness of pacing self-management for people with CFS.

METHODS

Patients

Fifteen adult women who fulfilled Centers for Disease Control and Prevention diagnostic criteria for CFS [20] were contacted, seven of whom participated in the study. All participants were recruited from a specialized university-based chronic fatigue center, were within the age range of 18 to 65, spoke Dutch as their native language, and provided written informed consent for study participation. Patients who previously received activity management or behavioral therapy were deemed inappropriate for study participation. The study protocol was approved by the local ethical committee.

Design

A single case study with an A1-B-A2 design was used. All study participants had to attend the university hospital on five different occasions at 1-week intervals (**Figure 1** displays the study flow diagram). During their initial visit, participants were asked to read the information leaflet, given the opportunity to ask for additional information, and asked to sign the informed consent form. To account for bias related to cointerventions, we explained to the study participants that they were allowed to continue their ongoing medical treatment but under no circumstances were they to initiate a new medical or conservative treatment. They were then asked to fill in a number of self-report questionnaires: the Chronic Fatigue Syndrome-Activities and Participation Questionnaire (CFS-APQ), CFS Symptom List, Medical Outcomes Study 36-Item Short Form Health Survey (SF-36), and Checklist Individual Strength (CIS). The order of questionnaire administration was randomized to account for test order bias. To prevent administration bias, we had patients fill out the questionnaires personally without input or feedback. Next, we assessed the patient's ability to perform daily activities using the Canadian Occupational Performance Measure (COPM).

After filling out the questionnaires, the patients entered stage A1 of the A1-B-A2 design. Stage A1 corresponded to the baseline measurement of physical behavior prior to treatment (stage B) commencement. During stage A1, participants continuously wore an accelerometer for real-time activity monitoring. Height, weight, and sex were entered before attaching the accelerometer on the nondominant wrist and securing it with an adhesive band. We explained the use of the device and instructed the patient to wear it 24 hours a day until the second appointment 1 week later. Besides wearing the accelerometer, patients were instructed to keep an activity diary; to rate their fatigue severity, pain severity, and concentration difficulties twice a day on a visual analog scale (VAS); and to complete the CFS Symptom List every evening.

At their second appointment (day 8 of the study and termination of stage A1), the participants received their first individual treatment session (initiation of stage B: **Figure 1**). Stage B lasted for 3 consecutive weeks and consisted of one individual treatment session a week for a total of three treatment sessions. The physiotherapists who led the treatment sessions were not involved in any of the measurements (blind assessments and blinded therapists).

The final treatment session (week 4) corresponded to the termination of stage B and the initiation of stage A2. Thus, after their final treatment session, patients were instructed to once again wear an accelerometer 24 hours a day until the final appointment 1 week later. As was the case during stage A1, they were instructed to keep an activity diary; to rate their fatigue severity, pain severity, and concentration difficulties twice a day on a VAS; and to complete the CFS Symptom List every evening. During their final appointment (week 5), participants were asked to complete the same set of questionnaires as during their initial visit. Again, the order of the questionnaires was randomized and patients had to fill out the questionnaires personally without input or feedback from the investigators. Finally, the same investigator assessed the patient's ability to perform daily activities using the COPM.

Questionnaires

The SF-36 assesses functional status and well-being, or quality of life [21]. The SF-36 has been documented to have reliability and validity in a wide variety of patient populations [21–23], and it is the most frequently used measure in CFS research [24].

The CFS Symptom List is a self-report measure for assessing symptom severity in CFS patients. In order to assess the severity of the symptoms included in the CFS Symptom List, we used VASs (100 mm). Psychometric work supporting the use of the CFS Symptom List has been published [25–26].

The CFS-APQ is a self-administered questionnaire aimed at monitoring activity limitations and participation restrictions in patients with CFS. A total score of 1 indicates

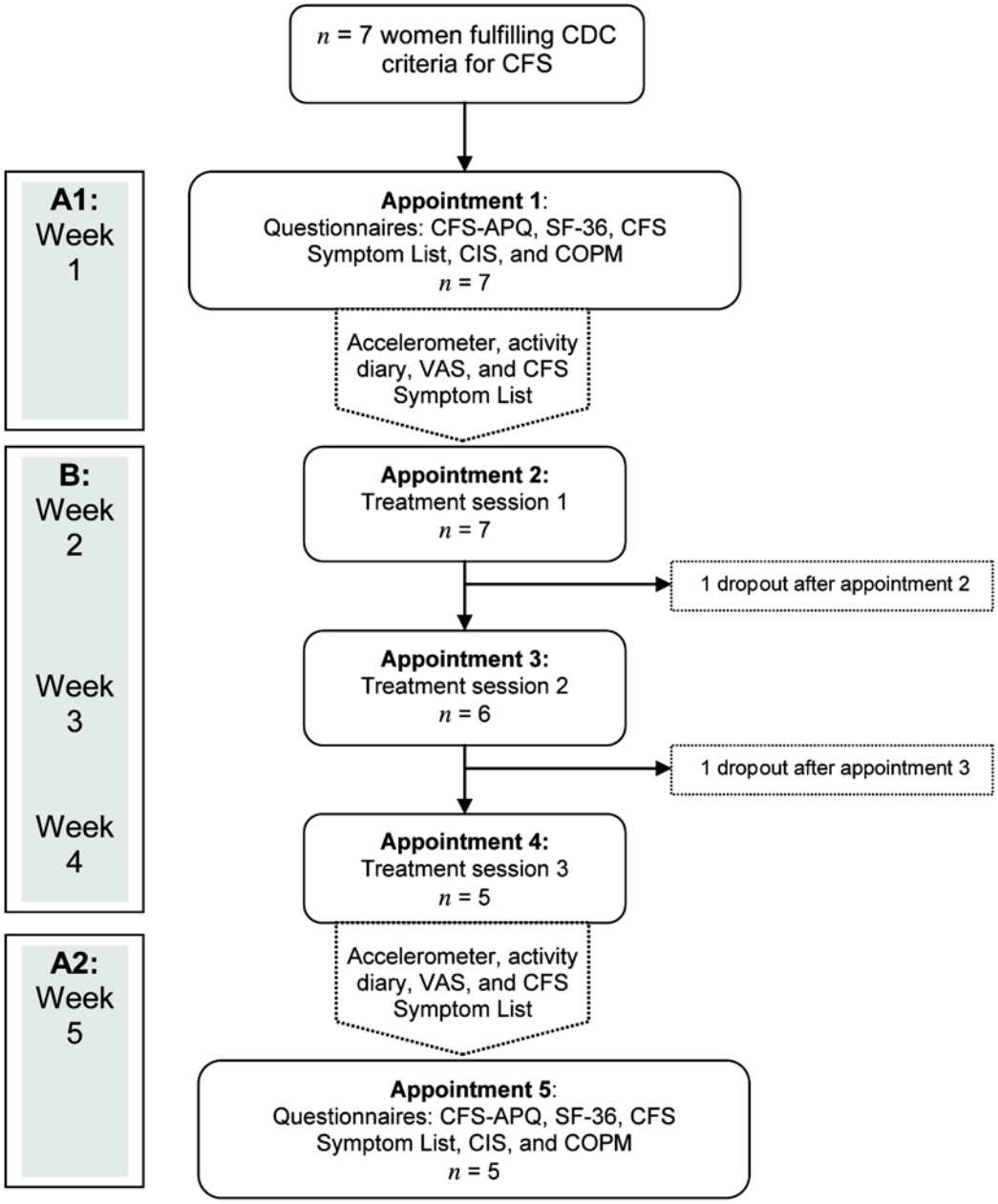

Figure 1.
Study flow diagram. CDC = Centers for Disease Control and Prevention, CFS = chronic fatigue syndrome, CFS-APQ = Chronic Fatigue Syndrome-Activities and Participation Questionnaire, CIS = Checklist Individual Strength, COPM = Canadian Occupational Performance Measure, SF-36 = Medical Outcomes Study 36-Item Short Form Health Survey, VAS = visual analog scale.

no activity limitations or participation restrictions, while 16 represents the maximum score. Data documenting the clinometric properties of the Dutch CFS-APQ are available [27–28].

The CIS aims at assessing subjective fatigue experience, concentration difficulties, motivation, and physical activity [29]. Higher scores on the CIS correspond to severe fatigue, many concentration difficulties, problems with motivation, and a low level of physical activity. Its psychometric properties are well established [29–31].

The COPM uses a semistructured interview for the assessment of the patient's ability to perform activities of

daily living within three domains: self-care, productivity, and leisure time [32]. During the semistructured interview, the assessment is focused on the patient's personal problems in commencing daily activities that are important to the patient's living environment. The COPM generates two subscale scores: the performance score indicates the ability of the patient to perform daily activities and the satisfaction score corresponds to the patient's satisfaction with daily activities. The clinometric properties of the COPM are well established [32–35].

Assessment of Physical Behavior

The Actical (Mini Mitter; Bend, Oregon) accelerometer was used for real-time monitoring of physical behavior. The Actical accelerometer has an omnidirectional sensor and is capable of measuring movement in one plane. The sensor functions via a cantilevered rectangular piezoelectric bimorph plate and seismic mass and is capable of detecting movements in the 0.5 to 3 Hz range. Voltage generated by the sensor is amplified and filtered via analog circuitry. The amplified and filtered voltage is passed into an analog to digital converter, and the process is repeated 32 times per second (32 Hz). The resulting per second value is divided by four and then added to an accumulated activity value (activity counts) for the epoch. The Actical is the smallest accelerometer available (28 × 27 × 10 mm, 17 g) and is water-resistant. Accelerometers are the gold standard for measuring physical behavior during daily activities. The Actical accelerometer has been used in scientific research and has been shown to be valid for the real-time assessment of physical behavior [36]. For the present study, the monitors were initialized to save data in 1-minute intervals (epochs).

Apart from wearing an accelerometer, patients were instructed to report their physical behavior in an activity diary. For each day, the activity diary consisted of a large table with four columns for filling out (1) the type of activity, (2) the initiation and termination time for each activity, (3) any health status changes (e.g., increase of fatigue in response to ironing), and (4) the total activity duration and corresponding metabolic equivalent (MET) values (to be completed by the investigators). Participants were instructed to specify their activities in detail immediately in order to avoid mistakes in recall. The activity diary also contained two sets of VASs (for fatigue severity, pain severity, and concentration difficulties) to be completed in the morning and at noon (immediately after lunch) and a CFS Symptom List (to be completed in the evening). Participants started with the activity diary the morning after the first appointment and were instructed to keep on doing so until the second appointment 1 week later. Apart from the VASs, no information or data generated with the activity dairy were used for further analysis. The activity diary was used by the therapists to coach the patient in the pacing self-management program. More specifically, the activity diary was used to identify activity peaks, to confront the patient, and to discuss solutions for the activities of interest.

Treatment: Pacing Self-Management

The pacing self-management program focused on teaching the patients to estimate their current physical capabilities prior to commencing an activity. In order to appropriately pace activities (daily activities and exercise bouts), CFS patients had to learn to estimate their current physical capabilities prior to commencing an activity, keeping in mind the regular fluctuating nature of their symptoms. Daily activities were defined as those duties typically performed around the home and at work, such as ironing, shopping, housework, gardening, etc. The activity duration used within the program was less than the reported duration to account for typical overestimations made by patients. Each activity block was interspersed with breaks, with the length of the break equating to the duration of the activity. This procedure was followed in order to account for the delayed recovery from exercise commonly demonstrated in CFS patients [19]. "Breaks" were defined as relative rest periods, with the patient just relaxing or performing a different type of light activity (for example, in a break between two ironing sessions, the patient was allowed to perform a light mental activity such as reading).

Employing the pacing principles during a CFS patient's daily life implicates a behavioral change. Thus, care was taken to explain the rationale and potential benefits of the program prior to commencement, while the patient's expectations for care were taken into account and subsequently used to encourage adherence to the program. We provided participants with a booklet that explained the treatment rationale and practical approach. When the patient with CFS is able to manage his or her daily activity (i.e., symptom fluctuation is reduced to a manageable level) (stabilization phase), the therapist can then start to progress activity and exercise levels (grading phase). However, in the present study, only the stabilization

phase was included in the program. An important difference between pacing self-management and cognitive behavioral therapy is that the latter postulates that activity levels can be substantially increased by a time-contingent approach and that a recovery from CFS is possible. The treatment as applied in the present study is described elsewhere [14].

Data Analysis

Data were analyzed with SPSS, version 16.0 (SPSS Inc; Chicago, Illinois). Given the small number of study participants, nonparametric and appropriate descriptive statistics were used. The accelerometer data were subdivided into four activity levels: sedentary activity (≤1 MET), light activity (<3 METs), moderate activity (3–6 METs), and vigorous activity (>6 METs). The METs were computed by the Actical software based on activity counts and demographic data of the participant. Besides the METs, the pure, untransformed data generated by the accelerometer were used: the mean ± standard deviation (SD) amount of daily physical activity, the amount of physical activity (i.e., activity counts) during the peak activity hour for each day (identified as the hour with the highest number of activity counts), and the peak ratio for each day (counted as the amount of physical activity during the peak activity hour/mean amount of physical activity on that day). A high peak ratio indicated a stronger concentration of physical activity, while a low peak ratio suggested that the participant spread her physical activity throughout the day more equally.

The Wilcoxon signed rank test was used to compare pre- versus posttreatment data (variability): questionnaires completed during week 1 versus week 5, mean and SD symptom severity score for symptom fluctuations (both CFS Symptom List) during week 1 versus week 5, and mean activity value/SD of the accelerometer data (activity counts and METs) during week 1 versus 5. In order to account for missing data, we used the "last observation carried forward method" for intention-to-treat analysis. The level of significance was set at 0.05. Clinically meaningful improvements of the SF-36 subscale scores were counted and interpreted according to the method described by Vitorino et al. [37]. Effect sizes were calculated as Cohen's *d*, with *d* defined as the difference between the two means divided by the pooled SD for those means. A *d*-value of 0.20 is described as small, 0.50 as medium (moderate), and 0.80 as large.

RESULTS

At study entry, the seven female participants had a median age of 43 ± 13 years (range: 23–59), median weight of 69 ± 19 kg, median body length of 170 ± 6 cm, and a median illness duration of 96 ± 44 months (range: 60–180). Two patients ended their cooperation during the program. One dropped out after the second appointment because of overlap between the treatment content of previous treatments (i.e., a multidisciplinary treatment program including activity management), and a second patient dropped out after the third appointment because she could not see how another approach besides magnesium intake could benefit her.

The comparison of the pre- versus posttreatment questionnaire data (appointment 1 vs appointment 5) is presented in **Table 1**. The total score on the CFS Symptom List, reflecting the severity of the entire symptom complex experienced by the patient, was improved (p = 0.043; mean improvement = 7 ± 8 mm; effect size = 0.4). The CIS revealed a trend toward improved concentration difficulties (p = 0.066; effect size = 0.6), an observation strengthened by the decrease in VAS score for concentration difficulties (p = 0.043; mean improvement = 12 ± 10 mm). Furthermore, the severity of mood swings (mean improvement of 18 ± 19 mm), muscle weakness (mean improvement = 19 ± 18 mm), and intolerance to bright light (mean improvement = 22 ± 24 mm) of the CFS Symptom List decreased from appointment 1 to appointment 5 ($p < 0.05$).

At the posttreatment assessment, patients' ability to perform daily activities improved (both subscale scores of the COPM; p = 0.043 with large effect sizes). Five of seven patients improved on both subscale scores, with a mean improvement (all seven patients) of 34 ± 31 percent for the performance subscale and 57 ± 60 percent for the satisfaction subscale. However, no change was found in activity limitations and participation restrictions (CFS-APQ) or quality of life (SF-36) ($p > 0.05$). Clinically meaningful improvements in the SF-36 subscale scores were observed rarely.

The previous paragraphs present the comparison of the first with the final assessment day, and those data might have been biased by the fluctuating nature of CFS. If, by coincidence, all patients had a bad day during the first appointment and a good day during their final appointment (unrelated to the treatment response), then the comparison of the first with the final assessment day

Table 1.
Intention-to-treat analysis of pre- (n = 7) versus posttreatment (n = 7) symptom and health status scores.

Variable	Appointment 1: Pretreatment (mean ± SD)	Appointment 5: Posttreatment (mean ± SD)	p-Value*	Effect Size: Cohen's d
CFS-APQ	8.2 ± 1.7	7.7 ± 1.8	0.35	0.3
CIS Fatigue	47.3 ± 8.1	41.9 ± 9.9	0.14	0.6
CIS Concentration Difficulties	26.0 ± 5.5	22.3 ± 6.7	0.07	0.6
CIS Motivation	15.1 ± 6.6	15.1 ± 6.6	0.89	0
CIS Physical Activity	13.7 ± 4.8	10.6 ± 3.6	0.10	0.7
COPM Performance	3.7 ± 0.8	4.8 ± 1.3	0.043†	0.9
COPM Satisfaction	3.1 ± 0.9	4.7 ± 1.7	0.043†	1.2
CFS Symptom List Total Score	51.3 ± 17.5	44.1 ± 21.7	0.043†	0.4
VAS Fatigue	68.9 ± 19.7	62.4 ± 29.2	0.30	0.3
VAS Pain	50.1 ± 31.5	50.6 ± 32.9	0.67	0.0
VAS Concentration Difficulties	63.4 ± 28.3	51.9 ± 34.5	0.043†	0.4
VAS Muscle Weakness	61.4 ± 18.1	42.7 ± 31.2	0.043†	0.7
VAS Mood Swings	42.3 ± 15.4	25.0 ± 22.5	0.042†	0.9
VAS Intolerance to Bright Light	52.7 ± 27.4	30.3 ± 36.1	0.043†	0.7
SF-36 Bodily Pain	44.9 ± 18.8	49.0 ± 15.9	0.89	0.2
SF-36 Physical Functioning	42.1 ± 20.8	46.4 ± 16.8	0.22	0.2
SF-36 Role Limitations: Physical Functioning	39.3 ± 31.8	28.6 ± 30.4	0.08	0.3
SF-36 Role Limitations: Emotional Problems	57.1 ± 46.0	71.4 ± 40.5	0.32	0.3
SF-36 Social Functioning	48.2 ± 18.3	46.4 ± 21.3	0.32	0.1
SF-36 Mental Health	56.6 ± 12.5	60.0 ± 14.2	0.28	0.2
SF-36 Vitality	41.4 ± 13.8	41.4 ± 16.5	0.99	0
SF-36 General Health Perception	34.6 ± 12.6	29.5 ± 7.7	0.41	0.5

*p-values based on Wilcoxon signed rank test.
†$p < 0.05$.
CFS = chronic fatigue syndrome, CFS-APQ = CFS-Activities and Participation Questionnaire, CIS = Checklist Individual Strength, COPM = Canadian Occupational Performance Measure, SD = standard deviation, SF-36 = Medical Outcomes Study 36-Item Short Form Health Survey, VAS = visual analog scale.

would have been biased. However, study participants completed the CFS Symptom List not only during the baseline and final assessment day but also every evening during the week prior to and immediately following treatment. For each day, the mean symptom severity score of all participants together was counted and plotted (**Figure 2**), revealing a positive change. **Table 2** presents the change in mean symptom severity between week 1 (A1: pretreatment) and week 5 (A2: posttreatment). The mean total score on the CFS Symptom List and the mean pain severity improved from week 1 to week 5. When analyzing the SD (as a measurement of fluctuation) of the scores obtained with the CFS Symptom List, we found no statistically significant change between week 1 and week 5 (data not shown).

Comparing the data generated by the accelerometer between week 1 and 5, we found a statistically significant decrease in the mean time spent doing light activity (<3 METs) but no change in time spent doing sedentary (≤1 MET), moderate (3–6 METs), or vigorous activity (>6 METs) (**Table 3**). Furthermore, no changes in the mean amount of physical activity (i.e., activity counts) during the peak activity hour for each day, the total mean time spent active, or the way physical activity was spread throughout the day (i.e., the mean peak ratio) were found. To further analyze the fluctuating nature of the physical activity pattern, we compared the SD of the accelerometer data between week 1 and 5. No statistically significant changes were observed (data not shown).

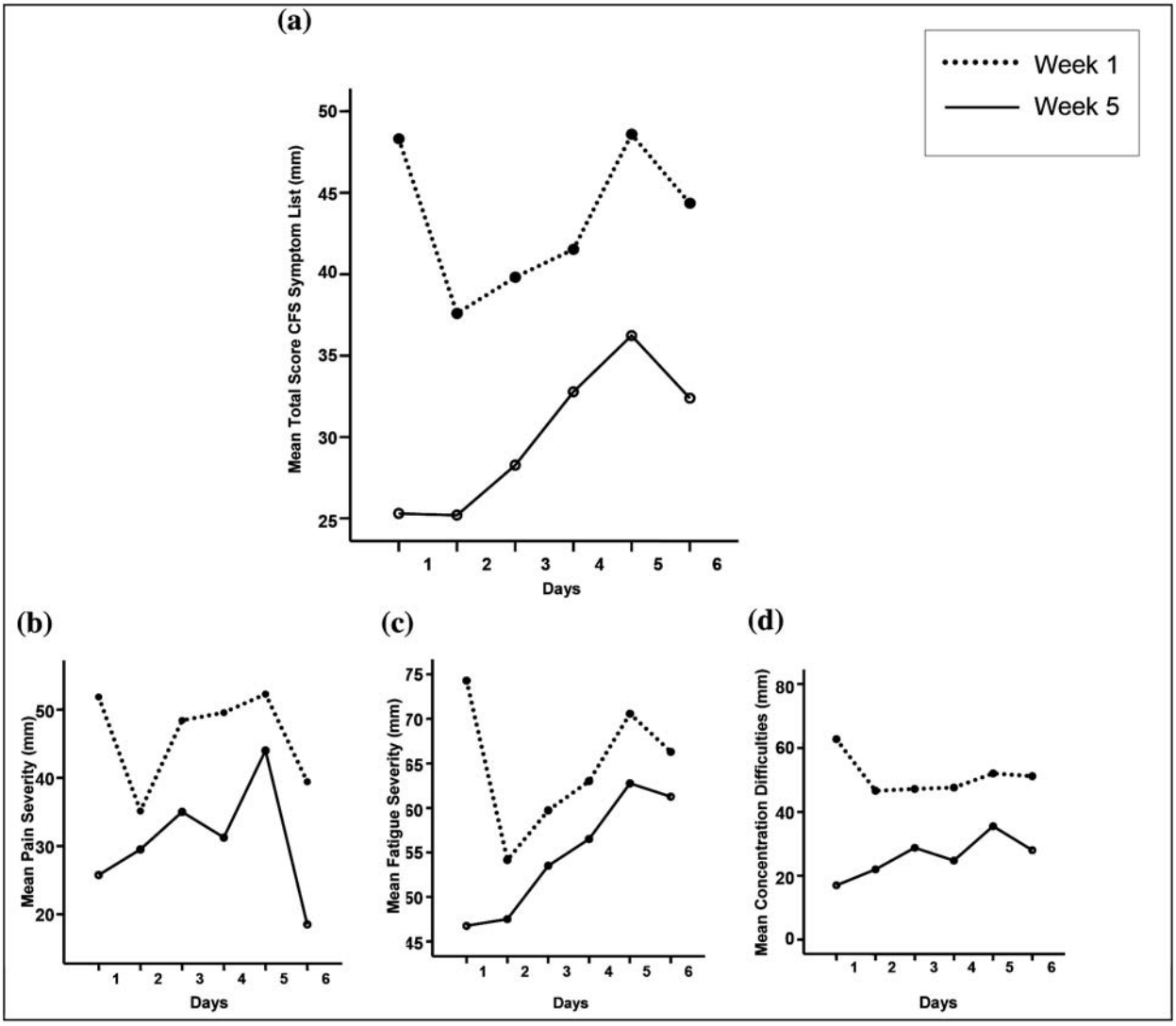

Figure 2.
Change in mean symptom severity between week 1 (A1: pretreatment) and week 5 (A2: posttreatment). **(a)** Change in mean total scores of the CFS Symptom List between week 1 and week 5. **(b)**, **(c)**, and **(d)** Change in mean pain severity, mean fatigue severity, and mean concentration difficulties, respectively. CFS = chronic fatigue syndrome.

DISCUSSION

The present observational study showed that 3 weeks of pacing self-management in a small group of CFS patients was accompanied by a modest improvement in symptom severity, an improved ability to perform daily activities, and a change in physical behavior (i.e., a decrease in the mean time spent doing light physical activity). No group change in self-reported quality of life or activity limitations and participation restrictions was observed, but large effect sizes were observed for the patients' ability to perform activities of daily living. Although the study design precludes causal interpretations of the results, these findings might point to short-term benefits of pacing self-management for people diagnosed with the severe illness, CFS. The outcome of the present single-case study design calls for a randomized controlled clinical trial to examine the effectiveness of pacing self-management for people with CFS.

The main study findings were unlikely to be biased by the fluctuating nature of the health status of CFS patients. Indeed, the outcome of the comparison of the

Table 2.
Change in mean symptom severity between week 1 (pretreatment) and week 5 (posttreatment).

Variable	Week 1: Pretreatment (mean ± SD)	Week 5: Posttreatment (mean ± SD)	*p*-Value*
CFS Symptom List Total Score	43.4 ± 19.1	33.9 ± 16.7	0.043†
VAS Fatigue	63.7 ± 23.6	57.1 ± 23.9	0.14
VAS Pain	44.9 ± 24.4	37.1 ± 22.2	0.03†
VAS Concentration Difficulties	49.9 ± 22.6	39.2 ± 22.5	0.08

*p-values based on Wilcoxon signed rank test.
†$p < 0.05$.
CFS = chronic fatigue syndrome, SD = standard deviation, VAS = visual analog scale.

Table 3.
Change in physical behavior assessed with accelerometer between week 1 (pretreatment) and week 5 (posttreatment).

Variable	Week 1: Pretreatment (mean ± SD)	Week 5: Posttreatment (mean ± SD)	*p*-Value*
Total Mean Time Active (min)	235.2 ± 47.6	221.6 ± 27.9	0.89
Mean Time Sedentary Activity: ≤1 MET (min)	746.6 ± 61.8	736.0 ± 48.1	0.35
Mean Time Light Activity: <3 METs (min)	567.5 ± 38.1	592.5 ± 38.8	0.043†
Mean Time Moderate Activity: 3–6 METs (min)	125.8 ± 36.7	111.5 ± 21.5	0.69
Mean Time Vigorous Activity: >6 METs (min)	0.1 ± 0.2	0.1 ± 0.2	0.32
Mean Amount of Physical Activity (i.e., activity counts) During Peak Activity Hour for Each Day	829.9 ± 144.0	781.3 ± 179.5	0.67
Mean Peak Ratio (activity counts/activity counts)	3.6 ± 0.2	3.5 ± 0.5	0.67

*p-values are based on the Wilcoxon signed rank test.
†$p < 0.05$.
MET = metabolic equivalent, SD = standard deviation.

self-reported data generated during the first and final appointment (**Table 1**) was confirmed by the analysis (**Table 2**) and plotting (**Figure 2**) of the mean symptom severity score of all participants together throughout week 1 versus week 5. Thus, symptom severity improved from week 1 to week 5 for the CFS patients studied here and the effect sizes were moderate to small.

The change in physical behavior observed here is difficult to interpret; it might even be a negative finding. Apart from a decrease in the mean time spent doing light activity (<3 METs), no changes in physical behavior were observed. The pacing self-management program specifically targets behavioral change by attempting to spread the amount of physical activity throughout the day and stabilize the fluctuating nature of the physical activity pattern throughout the week. However, no change in the way physical activity was spread throughout the day (i.e., the mean peak ratio) or the fluctuating nature of the physical activity pattern was observed. This might be due to the inability of the program to alter these behavioral parameters, the small sample size, or the wrong choice of outcome parameters. The peak ratio for each day was counted as the amount of physical activity during the peak activity hour divided by the mean amount of physical activity on that day. Whether this represents a valid outcome parameter remains unclear.

A high dropout rate (2/7 or 28.6%) was observed. However, the results presented were based on intention-to-treat analysis. We conclude that despite the high dropout rate, improvements in symptom severity, daily activity performance, and physical behavior accompanied the pacing self-management program. Given the lack of evidence

in support of pacing self-management as a sole treatment or treatment component for those with CFS, comparison with other studies is impossible. The ongoing, large Pacing, Graded Activity, and Cognitive Behavior Therapy: A Randomized Evaluation (PACE) trial [38] will provide more insight into the potential benefits of pacing self-management for those with CFS.

A follow-up period was not included in the present study, which means that we have no idea about the patients' long-term response. However, three sessions of pacing self-management are not intended for long-term improvements. Rather, they serve as the first stage (stabilization phase) of a rehabilitation program designed specifically for those with CFS [14]. This study focused on the initial response of people with CFS to the stabilization phase of pacing self-management. Additional treatment sessions would be required to provide a comprehensive rehabilitation program and, hence, generate effect sizes similar to the ones reported in response to cognitive behavioral therapy [39]. On the other hand, not all patients with CFS can be motivated for the subsequent grading phase and thus will remain in a long-term stabilization phase. For those patients, it would have been of interest to monitor the long-term responses.

As explained in detail elsewhere [14], a comprehensive rehabilitation program for people with CFS comprises the initial stabilization phase studied here followed by a grading phase. During the second phase, the same pacing techniques are applied to grade both daily activities as well as exercise levels. When an appropriate exercise level is being determined, a formal, regulated exercise regime that is gentle, graded, flexible, and manageable according to each individual's capabilities is required. Support for this type of grading for people with CFS comes from an observational study [40] and a well-designed randomized controlled clinical trial, which reported that paced and individually tailored graded exercise was superior to relaxation and flexibility training in patients with CFS [41]. Our view of the initial stabilization and subsequent grading phase seems to contradict the adaptive pacing therapy as advocated in the ongoing PACE trial, in which graded exercise therapy is compared with rather than preceded by the initial stabilization phase of our pacing program.

CONCLUSIONS

In conclusion, the outcome of the present single case study design suggests that 3 weeks of pacing self-management improves symptom severity and performance of daily activities, but a large randomized controlled evaluation is required to confirm these preliminary observations.

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REFERENCES

1. Wallman KE, Morton AR, Goodman C, Grove R. Reliability of physiological, psychological, and cognitive variables in chronic fatigue syndrome. Res Sports Med. 2005;13(3): 231–41. [PMID: 16392538] DOI:10.1080/15438620500222562
2. Jammes Y, Steinberg JG, Mambrini O, Brégeon F, Delliaux S. Chronic fatigue syndrome: Assessment of increased oxidative stress and altered muscle excitability in response to incremental exercise. J Intern Med. 2005;257(3):299–310. [PMID: 15715687] DOI:10.1111/j.1365-2796.2005.01452.x
3. Sorensen B, Streib JE, Strand M, Make B, Giclas PC, Fleshner M, Jones JF. Complement activation in a model of chronic fatigue syndrome. J Allergy Clin Immunol. 2003; 112(2):397–403. [PMID: 12897748] DOI:10.1067/mai.2003.1615
4. Lapp CW. Exercise limits in chronic fatigue syndrome. Am J Med. 1997;103(1):83–84. [PMID: 9236491]

5. Black CD, O'Connor PJ, McCully KK. Increased daily physical activity and fatigue symptoms in chronic fatigue syndrome. Dyn Med. 2005;4(1):3. [PMID: 15745455] DOI:10.1186/1476-5918-4-3
6. Van der Werf SP, Prins JB, Vercoulen JH, Van der Meer JW, Bleijenberg G. Identifying physical activity patterns in chronic fatigue syndrome using actigraphic assessment. J Psychosom Res. 2000;49(5):373–79. [PMID: 11164063] DOI:10.1016/S0022-3999(00)00197-5
7. Van Houdenhove B, Neerinckx E, Onghena P, Lysens R, Vertommen H. Premorbid "overactive" lifestyle in chronic fatigue syndrome and fibromyalgia. An etiological factor or proof of good citizenship? J Psychosom Res. 2001;51(4): 571–76. [PMID: 11595245] DOI:10.1016/S0022-3999(01)00247-1
8. Harvey SB, Wadsworth M, Wessely S, Hotopf M. Etiology of chronic fatigue syndrome: Testing popular hypotheses using a national birth cohort study. Psychosom Med. 2008;70(4):488–95. [PMID: 18378866] DOI:10.1097/PSY.0b013e31816a8dbc
9. Vercoulen JH, Bazelmans E, Swanink CM, Fennis JF, Galama JM, Jongen PJ, Hommes O, Van der Meer JW, Bleijenberg G. Physical activity in chronic fatigue syndrome: Assessment and its role in fatigue. J Psychiatr Res. 1997;31(6):661–73. [PMID: 9447571] DOI:10.1016/S0022-3956(97)00039-3
10. Clapp LL, Richardson MT, Smith JF, Wang M, Clapp AJ, Pieroni RE. Acute effects of thirty minutes of light-intensity, intermittent exercise on patients with chronic fatigue syndrome. Phys Ther. 1999;79(8):749–56. [PMID: 10440661]
11. Cook DB, Nagelkirk PR, Peckerman A, Poluri A, Mores J, Natelson BH. Exercise and cognitive performance in chronic fatigue syndrome. Med Sci Sports Exerc. 2005;37(9): 1460–67. [PMID: 16177595] DOI:10.1249/01.mss.0000179921.48404.ef
12. Price JR, Mitchell E, Tidy E, Hunot V. Cognitive behaviour therapy for chronic fatigue syndrome in adults. Cochrane Database Syst Rev. 2008;3:CD001027. [PMID: 18646067]
13. Edmonds M, McGuire H, Price J. Exercise therapy for chronic fatigue syndrome. Cochrane Database Syst Rev. 2004;3:CD003200. [PMID: 15266475]
14. Nijs J, Paul L, Wallman K. Chronic fatigue syndrome: An approach combining self-management with graded exercise to avoid exacerbations. J Rehabil Med. 2008;40(4):241–47. [PMID: 18382818] DOI:10.2340/16501977-0185
15. Deale A, Chalder T, Wessely S. Illness beliefs and treatment outcome in chronic fatigue syndrome. J Psychosom Res. 1998;45(1 Spec No.):77–83. [PMID: 9720857]
16. Shephard C. Pacing and exercise in chronic fatigue syndrome. Physiotherapy. 2001;87(8):395–96. DOI:10.1016/S0031-9406(05)65457-0
17. Pardaens K, Haagdorens L, Van Wambeke P, Van den Broeck A, Van Houdenhove B. How relevant are exercise capacity measures for evaluating treatment effects in chronic fatigue syndrome? Results from a prospective, multidisciplinary outcome study. Clin Rehabil. 2006;20(1): 56–66. [PMID: 16502751] DOI:10.1191/0269215506cr914oa
18. CFS/ME Working Group. Report to the Chief Medical Officer of an independent working group [Internet]. London (UK): Department of Health; 2002 Jan 11. 82 p. Available from: http://www.dh.gov.uk/en/Publicationsandstatistics/Publications/PublicationsPolicyAndGuidance/DH_4064840/.
19. Paul L, Wood L, Behan WM, Maclaren WM. Demonstration of delayed recovery from fatiguing exercise in chronic fatigue syndrome. Eur J Neurol. 1999;6(1):63–69. [PMID: 10209352] DOI:10.1046/j.1468-1331.1999.610063.x
20. Fukuda K, Straus SE, Hickie I, Sharpe MC, Dobbins JG, Komaroff A. The chronic fatigue syndrome: A comprehensive approach to its definition and study. International Chronic Fatigue Syndrome Study Group. Ann Intern Med. 1994;121(12):953–59. [PMID: 7978722]
21. Ware JE, Snow KK, Kosinski M, Gandek B; New England Medical Center Hospital. SF-36 Health Survey: Manual and interpretation guide. Boston (MA): The Health Institute; 1993.
22. Wells KB, Stewart A, Hays RD, Burnam MA, Rogers W, Daniels M, Berry S, Greenfield S, Ware J. The functioning and well-being of depressed patients. Results from the Medical Outcomes Study. JAMA. 1989;262(7):914–19. [PMID: 2754791] DOI:10.1001/jama.262.7.914
23. McHorney CA, Ware JE Jr, Lu JF, Sherbourne CD. The MOS 36-Item Short Form Health Survey (SF-36): III. Tests of data quality, scaling assumptions, and reliability across diverse patients groups. Med Care. 1994;32(1):40–66. [PMID: 8277801] DOI:10.1097/00005650-199401000-00004
24. Nijs J, Vaes P, Van Hoof E, De Becker P, McGregor N, De Meirleir K. Activity limitations and participation restrictions in patients with chronic fatigue syndrome—Construction of a disease specific questionnaire. J Chronic Fatigue Syndr. 2002;10(3–4):3–23. DOI:10.1300/J092v10n03_02
25. Nijs J, Aerts A, De Meirleir K. Generalized joint hypermobility is more common in chronic fatigue syndrome than in healthy control subjects. J Manipulative Physiol Ther. 2006; 29(1):32–39. [PMID: 16396727] DOI:10.1016/j.jmpt.2005.11.004

26. Nijs J, Thielemans A. Kinesiophobia and symptomatology in chronic fatigue syndrome: A psychometric study of two questionnaires. Psychol Psychother. 2008;81(3):273–83. [PMID: 18644213] DOI:10.1348/147608308X306888
27. Nijs J, Vaes P, McGregor N, Van Hoof E, De Meirleir K. Psychometric properties of the Dutch Chronic Fatigue Syndrome-Activities and Participation Questionnaire (CFS-APQ). Phys Ther. 2003;83(5):444–54. [PMID: 12718710]
28. Nijs J, Cloostermans B, McGregor N, Vaes P, De Meirleir K. Construct validity and internal consistency of the Chronic Fatigue Syndrome-Activities and Participation Questionnaire (CFS-APQ). Physiother Theory Pract. 2004; 20:31–40. DOI:10.1080/09593980490424950
29. Vercoulen JH, Swanink CM, Fennis JF, Galama JM, Van der Meer JW, Bleijenberg G. Dimensional assessment of chronic fatigue syndrome. J Psychosom Res. 1994;38(5): 383–92. [PMID: 7965927] DOI:10.1016/0022-3999(94)90099-X
30. Vercoulen JH, Hommes OR, Swanink CM, Jongen PJ, Fennis JF, Galam JM, Van der Meer JW, Bleijenberg G. The measurement of fatigue in patients with multiple sclerosis: A multidimensional comparison with patients with chronic fatigue syndrome and healthy subjects. Arch Neurol. 1996;53(7):642–49. [PMID: 8929171]
31. Vercoulen JH, Alberts M, Bleijenberg G. The Checklist Individual Strength (CIS). Gedragstherapie. 1999;32:31–36.
32. Law M, Darrah J, Pollock N, King G, Rosenbaum P, Russell D, Palisano R, Harris S, Armstrong R, Watt J. Family-centred functional therapy for children with cerebral palsy: An emerging practice model. Phys Occup Ther Pediatr. 1998;18(1):83–102. DOI:10.1300/J006v18n01_06
33. McColl MA, Paterson M, Davies D, Doubt L, Law M. Validity and community utility of the Canadian Occupational Performance Measure. Can J Occup Ther. 2000;67(1):22–30. [PMID: 10695166]
34. Chan CCH, Lee TMC. Validity of the Canadian Occupational Performance Measure. Occup Ther Int. 1997;4(3): 231–249. DOI:10.1002/oti.58
35. Cup E, Thijssen M, Scholte op Reimer W. [Usefulness of the Canadian Occupational Performance Measure in stroke patients.] Nederlands Tijdschrift voor Ergotherapie. 2003; 31:157–160. Dutch.
36. Finn KJ, Specker B. Comparison of Actiwatch activity monitor and Children's Activity Rating Scale in children. Med Sci Sports Exerc. 2000;32(10):1794–97. [PMID: 11039655] DOI:10.1097/00005768-200010000-00021
37. Vitorino DF, Carvalho LB, Prado GF. Hydrotherapy and conventional physiotherapy improve total sleep time and quality of life of fibromyalgia patients: Randomized clinical trial. Sleep Med. 2006;7(3):293–96. [PMID: 16564209] DOI:10.1016/j.sleep.2005.09.002
38. White PD, Sharpe MC, Chalder T, DeCesare JC, Walwyn R; PACE trial group. Protocol for the PACE trial: A randomised controlled trial of adaptive pacing, cognitive behaviour therapy, and graded exercise, as supplements to standardised specialist medical care versus standardised specialist medical care alone for patients with the chronic fatigue syndrome/myalgic encephalomyelitis or encephalopthy. BMC Neurol. 2007;7:6. [PMID: 17397525] DOI:10.1186/1471-2377-7-6
39. Scheeres K, Wensing M, Knoop H, Bleijenberg G. Implementing cognitive behavioral therapy for chronic fatigue syndrome in a mental health center: A benchmarking evaluation. J Consult Clin Psychol. 2008;76(1):163–71. [PMID: 18229994] DOI:10.1037/0022-006X.76.1.163
40. Coutts R, Weatherby R, Davie A. The use of a symptom "self-report" inventory to evaluate the acceptability and efficacy of a walking program for patients suffering with chronic fatigue syndrome. J Psychosom Res. 2001;51(2): 425–29. [PMID: 11516764] DOI:10.1016/S0022-3999(00)00220-8
41. Wallman KE, Morton AR, Goodman C, Grove R, Guilfoyle AM. Randomised controlled trial of graded exercise in chronic fatigue syndrome. Med J Aust. 2004;180(9): 444–48. [PMID: 15115421]

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