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## FUNCTIONAL OUTCOMES OF ANAEROBIC REHABILITATION IN A PATIENT WITH CHRONIC FATIGUE SYNDROME: CASE REPORT WITH 1-YEAR FOLLOW-UP

Staci R. Stevens, MA<sup>1</sup>; Todd E. Davenport, PT, DPT, OCS<sup>2</sup>

<sup>1</sup> Executive Director, Pacific Fatigue Laboratory, Department of Sport Sciences, University of the Pacific, Stockton, CA, USA

<sup>2</sup> Assistant Professor, Department of Physical Therapy, Thomas J. Long School of Pharmacy and Health Sciences, University of the Pacific, Stockton, CA, USA

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### ABSTRACT

This case study aimed to document the effect of pacing self-management and short-duration exercise on physiological functioning and disability in an individual with chronic fatigue syndrome/myalgic encephalomyelitis (CFS/ME). A 28-year-old woman with CFS/ME received a rehabilitation program that involved: (1) pacing self-management using a heart rate monitor to maintain heart rate below ventilatory threshold and (2) a pragmatic approach to restorative strengthening and flexibility exercises conducted at an intensity below anaerobic threshold, which were completed 3 times weekly. Cardiopulmonary exercise testing (CPET) with analysis of expired gases and functional self-report were obtained before and 1 year after initiation of intervention. At 1-year follow-up, the patient reported she was able to complete daily activities without reproducing symptoms and 75% improvement in time to recover from CPET. Prior to intervention, patient demonstrated impaired cardiovascular and pulmonary responses at peak and anaerobic threshold, which improved at 1-year follow-up. Pacing self-management combined with short-duration restorative exercise resulted in a favorable clinical outcome in this patient with CFS/ME. Additional research is necessary to determine the efficacy and mechanism of action for this treatment.

**KEYWORDS:** Chronic fatigue syndrome, myalgic encephalomyelitis, rehabilitation, exercise, intervention.

## **BACKGROUND**

Chronic fatigue syndrome/myalgic encephalomyelitis (CFS/ME) affects 1-4 million adults in the United States (1). The most common definition of CFS/ME involves persistent or relapsing fatigue for greater than 6 months (2). To meet case definition criteria for the diagnosis of CFS/ME, an individual concurrently must also demonstrate at least 4 additional symptoms, including post-exertional malaise (PEM) for at least 24 hours following exercise, impaired memory or concentration, unrefreshing sleep, muscle pain, multiple joint pain without signs of inflammation, headaches of new type or severity, sore throat, and tender cervical or axillary lymph nodes (2).

Among all fatiguing conditions, PEM appears uniquely characteristic of CFS/ME and it is a prominent source of functional disability in individuals with CFS/ME. PEM refers to intermediate-term discomfort or unease following even minimal physical activity, which can last up to 2 weeks or more (3). One possible explanation for PEM involves a spiraling cycle of decline in aerobic energy system function and autonomic regulation (4). Genetic, cellular, and systemic metabolic deficits are thought to cause aerobic system impairment, perhaps secondary to either active or latent viral activation (5). Clinically-relevant cognitive and behavioral factors also are associated with PEM, involving centrally-mediated disturbances in attentional, perception, and affect (6). PEM is characterized in part by impaired effort perception, which may lead to decreased maximal exertion. However, behavior responses to post-exertional symptoms in patients with CFS/ME can range from maintenance of a sedentary lifestyle to abrupt increases in activity during periods of symptom remission that serve to exacerbate symptoms.

The importance of pain and fatigue in CFS/ME symptomatology places a premium on developing appropriate rehabilitation strategies for individuals with this condition. To date, much of the literature involving physical rehabilitation for patients with CFS/ME has examined the effectiveness of graded aerobic exercise to improve physiological characteristics and functional outcomes. Training effects have been observed across studies (7, 8), but results involving functional improvement have been more mixed. Indeed, self-reported functional improvement in these studies also may be an artifact of performing an exercise program, rather than reflecting personally meaningful change in a patient's status (9). Many studies involving graded exercise also demonstrate a high drop out rate, which suggests this intervention may be ineffective for a substantial portion of patients with CFS/ME.

We recently proposed a promising rehabilitation model that aims to promote meaningful functional improvement using interventions that avoid excessive activation of the impaired aerobic system (10). These include pacing self-management strategies combined with short-duration restorative exercise. To date, no conceptual work has been completed that applies the model in practice. The purpose of this case report is to describe and apply a rehabilitation model that involves pacing self-management and short-term exercise in the clinical management of a patient with CFS/ME.

## CASE DESCRIPTION

The patient was a 28-year-old female who presented with a diagnosis of CFS/ME and orthostatic intolerance. Her chief functional concern was avoidance of all daily activities, except for walking and household chores. Her main symptoms included whole-body fatigue and aching pain after activities. Her reported activity coping style involved cyclic periods of substantially increased activity during periods of low symptoms, followed by periods of decreased activity associated with increased symptoms of PEM that precluded activity (ie, 'push-crash' cycle). She noted her periods of symptoms and corresponding decreased activity were becoming progressively longer and more frequent.

Single cardiopulmonary exercise tests (CPET) were used to evaluate aerobic and anaerobic capacity before and after intervention, as well as to provide a standardized physiological stressor aimed at provoking symptoms of PEM. The pre-test involved treadmill CPET with 2.0mph ramping protocol and the post-test included bicycle CPET with a 10 watt/minute ramping protocol (11). Electrocardiography was monitored continuously for signs of cardiac arrhythmia or ischemia, and pulse oximetry was monitored to ensure safe levels of arterial oxygenation. The patient was encouraged to continue on each test as long as possible, and testing was terminated when objective criteria for maximal effort were met (11). The patient was then monitored for 2-5 minutes following each test.

**Table 1.** Pre-intervention and post-intervention cardiopulmonary exercise measurements

	<b>Baseline</b>	<b>1 Year Follow Up</b>	<b>% Change</b>
<b>VO<sub>2</sub><sub>max</sub></b>	25.5	26.1	▲2%
<b>VE</b>	73	90	▲19%
<b>RR</b>	38	54	▲42%
<b>HR</b>	177	189	▲7%
<b>BP</b>	112/72	170/98	▲51% / ▲25%
<b>VT HR</b>	103	121	▲17%
<b>Recovery</b>	1 month	1 week	▼75%

**Abbreviations:** VO<sub>2</sub><sub>max</sub> – peak oxygen uptake (ml/kg/min), VE – minute ventilation (L/min), RR – respiratory rate (breaths/min), HR – heart rate (beats/min), VT HR – ventilatory threshold heart rate (beats/min), recovery – time to full symptomatic recovery from a test-retest maximal exercise challenge

The pre-intervention CPET met objective criteria for a maximal test. Pre-intervention CPET measurement and recovery questionnaire revealed abnormal exercise responses. Although peak volume of oxygen consumption was within the low end of normal range (25.5 ml/kg/min), peak exercise systolic blood pressure (112/72), respiratory rate (38 breaths/min), and ventilation (73 L/min) were blunted (Table 1). Blood pressure dropped upon standing and failed to rise during exercise to pre-test supine resting levels. However, the patient remained asymptomatic of orthostatic hypotension throughout the pre-intervention CPET procedure. The patient experienced exercise intolerance with poor recovery. Following the CPET, the patient's exercise recovery questionnaire indicated she felt "great [and] energetic for the first 6 hours"; "weak and tired but the 2nd and 3rd days were worse;" and she estimated it took at least one month to fully recover.

The patient completed the intervention program at home 3 times per week for 52 weeks. Intervention involved pacing self-management and restorative exercise conducted at an intensity below anaerobic threshold (10). Pacing self-management involved activity biofeedback by way of heart rate monitoring. This was intended to provide the patient with an objective measurement of workload during daily tasks. An alarm was set on the monitor so it would inform the patient when she was exerting at workloads greater than anaerobic threshold. The patient was encouraged to function at workloads that would not prompt the alarm, and to rest in a seated or supine position when the alarm sounded. Restorative exercises involved general flexibility, resistance, and short-term endurance exercises conducted at intensities below anaerobic threshold. Specific interventions were selected considering the symptoms that were identified as most concerning to the patient. For example, diaphragmatic breathing and gentle upper extremity stretching were used to facilitate active pain management. Also, supine upper and lower extremity resistance band exercises were used to minimize the potentially aggravating influence of gravity on the patient's apparent circulatory impairment, even though she was asymptomatic during testing.

## **OUTCOMES**

At 1 year follow-up, the patient demonstrated good self-reported adherence to the treatment program according to weekly exercise logs. Functional improvements were seen in exercise dosage parameters, and she reported increased tolerance to daily activities without reproducing symptoms of PEM. Follow-up CPET conducted at that time recorded associated improvements in cardiopulmonary measures. At post-intervention follow-up, the patient also met objective criteria for a maximal test. Peak VO<sub>2</sub> (26.1 ml/kg/min), ventilation (90 L/min), respiratory rate (54 breaths/min), heart rate (189 beats/min), and systolic blood pressure (170/98) all increased. Heart rate at the ventilatory threshold increased to 121 beats/min (64% of peak value). All measurements represented improvements over baseline testing. The patient's self-reported recovery time also was reduced 75% from the initial CPET.

## **DISCUSSION**

This case study is the first to document physiological and functional responses to pacing self-management combined with short duration exercise in an individual with CFS/ME. At 1 year follow-up, the patient continued assertive disease self-management in combination with a program of short duration exercises in order to manage her chronic fatigue symptoms. In this manner, the patient was able to increase her tolerance for usual daily activities without exacerbation of PEM symptoms.

Although causation is impossible to infer from a single case, the patient's improvement in functional activities appeared to occur in parallel with selected improvements in submaximal cardiovascular and pulmonary responses to exercise. Elevation in ventilatory threshold heart rate suggests a potential increase in the reserve for submaximal physiological functioning that is associated with usual daily activities. Although estimates for peak VO<sub>2</sub> and HR are expected to be slightly higher during treadmill CPET than CPET conducted using bicycle ergometry, VO<sub>2</sub> and HR at ventilatory threshold are generally considered to remain stable between the 2 testing modes. Improved submaximal metabolic functioning may partially explain the patient's improved tolerance to functional activities, even though no substantial increase in peak VO<sub>2</sub> was observed. Indeed, this trend is predicted from the specificity principle of exercise, given that activity prescription in this case study focused on short-duration tasks that should not be expected to promote improvements in peak metabolic performance.

It should be noted that since the pre-intervention CPET is known to result in higher estimations of metabolic capacity than the post-intervention CPET, it is possible a larger treatment effect would have been observed if similar testing modalities had been used in this case study. Nevertheless, observed improvements in peak systolic and diastolic function, peak heart rate, and peak respiratory rate suggest the possibility of effects at the level of cardiovascular and pulmonary impairments that may contribute to PEM in patients with CFS/ME. These observations regarding potential improvements in cardiovascular and pulmonary system impairments require additional replication in the context of controlled intervention studies.

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