Available online at www.sciencedirect.com

SciVerse ScienceDirect

Neuromuscular Disorders 22 (2012) S221–S225

www.elsevier.com/locate/nmd

Intensive aerobic training improves motor performances and oxidative metabolism efficiency in chronic polymyositis: A case report

S. Dalise, F. Bertolucci, C. Simonella, B. Rossi, C. Chisari *

Unit of Neurorehabilitation, Department of Neuroscience, University Hospital Pisa, Italy

Abstract

We describe the case of a 64-year-old woman affected by chronic polymyositis with gait disturbance, fatty replacement and swelling of thigh muscles. She achieved significant clinical improvement after 5 weeks intensive aerobic training. In particular the patient improved in motor performance tests, showed an improvement in the efficiency of oxidative metabolism and quality of life. Furthermore, analysis of creatinephosphokinase levels showed a reduction of muscle damage susceptibility. In conclusion, a specific intensive exercise program can be safely used with beneficial effects on muscle function in patients with chronic polymyositis.

Keywords: Polymyositis; Aerobic training; Oxidative metabolism; Muscle damage; Myopathies rehabilitation

1. Introduction

Polymyositis (PM) is a chronic, inflammatory and autoimmune skeletal muscle disorder characterized by muscle weakness and fatigue, mainly in proximal muscles such as thigh, shoulder and neck, and infiltration of mononuclear inflammatory cells [1–4]. Despite of initial favorable effects of corticosteroid and aggressive immunosuppressive treatment in most of patients, a majority develops sustained disability characterized by reduced muscle strength and endurance as well as fatigue and myalgia [5].

The pathophysiological mechanism underlying the sustained low muscle endurance and weakness in patients with chronic PM is not completely understood and it seems to vary during different phases of the disease. The inflammation is likely to cause muscle impairment in the early phase while secondary metabolic impairment, such as reduced ATP levels in muscle, could contribute to the disability in the chronic phase of the disease. In the late chronic phase other factors such as steroid myopathy and disuse of muscle due to physical inactivity could also play a role [6–8].

In the past, different approaches were used to study oxidative metabolism in inflammatory myopathies. Cea et al. [9] studied quantitative MRI and phosphorus magnetic resonance spectroscopy (31P-MRS) in a group of patients affected by polymyositis and dermatomyositis (PM/DM) and showed an impaired muscle oxidative metabolism in all subjects; they attributed this reduced aerobic capacity to slow H+ efflux. Wiesinger et al. [10] studied the peak oxygen uptake and described a reduced aerobic capacity in adult PM/DM patients. An abnormal oxygen supply to the muscles seems to be the most likely explanation for the association of oxidative metabolism impairment, the shift towards anaerobic metabolism during exercise and the reduced rate of H+ efflux. This defect could be attributable to some morphologic features that have been found in PM patients such as abnormal regional capillary distribution and microvessel walls thickness [11,12] or reduced number of capillaries per muscle fiber [13].

Physical exercise could be one way to counteract several of the mechanisms that are believed to have a negative effect on muscle function in patients with PM. However, until recently, patients have been advised to refrain from

* Corresponding author. Address: Unit of Neurorehabilitation, Department of Neuroscience, University Hospital Pisa, Via Paradisa 2, 56126 Pisa, Italy. Tel.: +39 (0) 50 996907, Fax: +39 (0) 50 995430.
E-mail address: c.chisari@ao-pisa.toscana.it (C. Chisari).

0960-8966/$ - see front matter © 2012 Elsevier B.V. All rights reserved.
http://dx.doi.org/10.1016/j.nmd.2012.10.015

physical activity because of fear of exacerbation of muscle inflammation and disease progression. This cautious attitude is based on previous studies revealing increased creatinephosphokinase (CPK) levels and signs of inflammation in muscle tissue after excessive exercise in healthy individuals [14,15].

However, recent studies have shown that moderate exercise in combination with immunosuppressive drugs can improve muscle performance without signs of increased muscle inflammation, suggesting that exercise represents a viable therapeutic intervention for PM patients [16,17].

We investigated the effect of a specific aerobic training in a 64-year-old PM patient, at a chronic stage of the disease, suffering from gait disturbance, excessive fatiguability and wasting of thigh muscles.

2. Case report

Here we describe the case of a 64-year-old woman affected by polymyositis who underwent a 5 week rehabilitative treatment to improve her motor-functional efficiency.

In 2003 was diagnosed a pharyngo-esophageal dyskinesia with erosive esophagitis. In 2004 high levels of CPK were occasionally noticed. In 2009 she developed fatigue and dysphagia. Electromyography showed a myopathic pattern and typical irritative signs of myositis such as fibrillation potentials and positive sharp waves. A muscle biopsy revealed nonspecific alterations consisting of scattered necrotic and regenerating fibers, abnormal variation in fiber size and varying degrees of endomysial and perimysial fibrosis and more specific features of polymyositis such as the occurrence of inflammatory cells at perivascular, perimysial and endomysial site. The endomysial inflammatory aggregates contained a high percentage of T cells, particularly activated CD8+ cells, very few natural killer cells and few or no B cells. Polymyositis with esophageus involvement was diagnosed. The patient was treated with Azathioprine and Immunoglobulins. In December 2010 the dysphagia and the asthenia worsened. A magnetic resonance showed marked atrophy in both quadriceps, fatty replacement of muscle bellies with swelling of residual muscle fibers and slight swelling of back thigh muscles (Fig. 1).

In February 2011 the patient was hospitalized to our Neurorehabilitation Unit because of a clinical exacerbation of the disease characterized by widespread and marked iposthenia, slowdown in postural changes, ambulation fatigue and inability to manage a flight of steps.

3. Methods

The rehabilitative treatment had a duration of 5 weeks with a frequency of 5 days/week. Prior to and after it the following evaluations were performed:

3.1. Oxidative metabolism

Oxidative metabolism was indirectly evaluated by means of dosage of blood lactate levels before and after a submaximal incremental exercise on a treadmill. The exercise protocol consisted of 11 steps of 2-min duration each steps at a constant speed of 3 km/h. The slope was 0% at the start level and increased of 2, 5% at each step. Heart rate was monitored during the exercise: if the patient reached 75% of the maximum theoretical heart rate (220-patient's age), the test was stopped, so keeping the exercise predominantly aerobic. Venous blood samples were executed at rest and at 1, 5, 10 and 30 min after the end of exercise [18].

3.2. The susceptibility to myofibral damage

The susceptibility to myofibral damage was evaluated through the dosage of CPK serum level before and after a walking test. The test consisted of 15 min walk on treadmill, at constant velocity, which was equivalent to the maximum speed tolerated by the patient. During exercise, heart rate was monitored in order to not to exceed the theoretical maximum value. Venous blood samples were performed at rest and at 1, 3, 6 and 24 h after the end of the exercise [19].

3.3. Isometric strength (MVC)

Isometric strength (MVC) was quantified by means of an isokinetic dynamometer (PrimusRS BTE). The patient was positioned on a high chair 30° tilted, with 90° hip

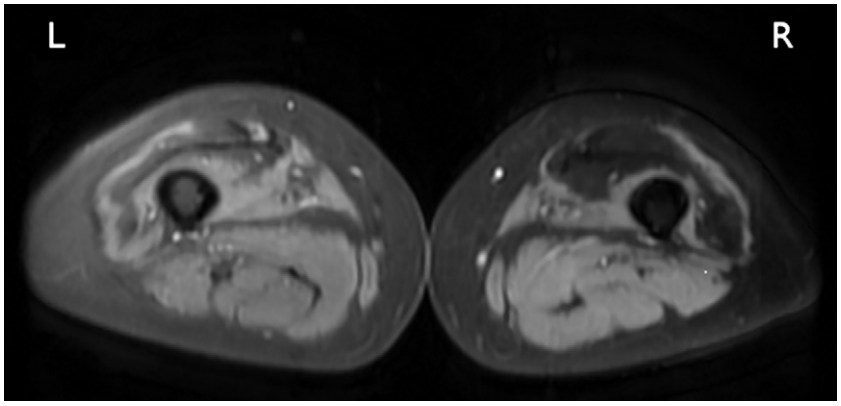

Fig. 1. T1 weighted MRI of thigh muscles performed 2 months before the training program.