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Review

Respiratory rehabilitation in multiple sclerosis: A narrative review of rehabilitation techniques

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ABSTRACT

Background: Respiratory disorders in multiple sclerosis (MS) are an important issue. They can occur early during the course of the disease, are associated with the neurological impairment, and can lead to pneumonia and respiratory failure, which are the main causes of death in advanced MS. Prevailing impaired expiratory muscles and cough abilities has been demonstrated in this population and might constitute a specific target for rehabilitation interventions. However, international guidelines lack recommendations regarding respiratory rehabilitation in MS. Here we performed a systematic review of the published literature related to respiratory rehabilitation in MS.
Methods: We searched the databases MEDLINE via PubMed, PEDro and Cochrane Library for English or French reports of clinical trials and well-designed cohorts published up to December 2016 with no restriction on start date by using the search terms "multiple sclerosis", "respiratory rehabilitation", "respiratory muscle training", "lung volume recruitment", "cough assistance", and "mechanical in-exsufflation". Literature reviews, case reports and physiological studies were excluded. The Maastricht criteria were used to assess the quality of clinical trials. We followed the Oxford Centre for Evidence-Based Medicine guidelines to determine level of evidence and grade of recommendations.
Results: Among the 21 reports of studies initially selected, 11 were retained for review. Seven studies were randomized controlled trials (RCTs), 2 were non-RCTs, and 2 were observational studies. Respiratory muscle training (inspiratory and/or expiratory) by use of a portable resistive mouthpiece was the most frequently evaluated technique, with 2 level-1 RCTs. Another level-1 RCT evaluated deep-breathing exercises. All reviewed studies evaluated home-based rehabilitation programs and focused on spirometric outcomes. The disparities in outcome measures among published studies did not allow for a meta-analysis and cough assistance devices were not evaluated in this population.
Conclusion: Although respiratory muscle training can improve maximal respiratory pressure in MS and lung volume recruitment can slow the decline in vital capacity, evidence is lacking to recommend specific respiratory rehabilitation programs adapted to the level of disability induced by the disease.

1. Introduction

Multiple sclerosis (MS) is a chronic demyelinating disease of the central nervous system. Because of the random localization of MS lesions along the neuraxis, patients can experience a wide range of symptoms, among which respiratory disorders are important [1]. The clinical expression of such disorders varies widely among individuals with the disease, and even if their onset can occur early in the disease evolution [2–4], they are associated with the overall neurological impairment assessed by the Expanded Disability Status Scale (EDSS) [1,3].

Respiratory disorders can be chronic or acute. Chronic respiratory failure involves bulbar dysfunction with swallowing disorders; altered central respiratory drive; motor and neuromuscular disorders following lesions disrupting the corticospinal tract; or sleep-disordered breathing. Acute conditions mainly involve spinal or bulbar relapse with extensive plaques, neurogenic pulmonary edema or acute respiratory failure, often following sepsis [1].

Infections and respiratory complications are the main causes of mortality in this population, with 11.7-fold more risk of death from

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respiratory issues than in the overall population [5]. Therefore, respiratory management and prevention of infections, especially aspiration pneumonia, are a major concern for physicians and caregivers [6,7]. For milder disease, although still debated, respiratory involvement is thought to be associated with fatigue [4,8], decreased motor performance and psychological and/or behavioral impairments [9].

Current practical guidelines and recommendations for MS care and rehabilitation acknowledge the lack of evidence for practical recommendations on respiratory rehabilitation [10,11]. The prevailing impaired expiratory muscles and cough abilities in this population has been demonstrated [11–14] and may be specific targets for rehabilitation interventions. Two recently published literature reviews assessed the benefits of respiratory muscle training (RMT) in MS [15,16], suggesting that RMT may have a significant impact on maximal inspiratory and expiratory pressures (MIP, MEP). However, the reviews did not address other techniques, combined protocols, or functional issues (fatigue, activity, quality of life, etc.).

Therefore, we performed a systematic review of the literature to assess the evidence for the various respiratory rehabilitation techniques used for MS. We aimed to establish a basis for evidence-based recommendations and future protocols.

2. Methods

2.1. Data collection

We performed a systematic review of the literature by searching for articles in English or French in the databases MEDLINE via PubMed, Physiotherapy Evidence Database (PEDro) and the Cochrane Library published up to December 2016 with no restriction on start date. The following search terms were used: "Multiple Sclerosis"[Mesh] and "respiratory rehabilitation"; "Multiple Sclerosis"[Mesh] and "breathing", and "rehabilitation"; "Multiple Sclerosis"[Mesh] and "respiratory muscle training"; "Multiple Sclerosis"[Mesh] and "respiratory training"; "Multiple Sclerosis"[Mesh] and "expiratory muscle training"; "Multiple Sclerosis"[Mesh] and "inspiratory muscle training"; "Multiple Sclerosis"[Mesh] and "lung volume recruitment"; "Multiple Sclerosis"[Mesh] and "cough assistance"; "Multiple Sclerosis"[Mesh] and "mechanical in exsufflation". The term "Multiple Sclerosis" was used as a MeSH term or as "all fields" without any difference in results obtained. We also checked for ongoing or terminated trial protocols registered at ClinicalTrials.gov. We excluded literature reviews, single case reports and physiological studies after careful examination of citations by a single reader (JL). Single abstracts were kept if they provided enough material.

2.2. Data processing and analysis

A reading grid was designed by 2 of the authors (JL, HP) to summarize the trial design, protocols and main outcomes for each study (Table 1). We used the Maastricht criteria [17,18] to assess the quality and level of evidence for interventional studies in rehabilitation (Table 2). We assigned a score of 1 to Maastricht items verified within the article's full text. After the study selection process, 2 authors (JL and HP) separately assessed the articles for quality. When the reviewed article did not fulfill a quality criterion or the design of the study was not appropriate, the corresponding item was given a score of 0. Ratings were comparable between both readers. For levels of evidence and grades of recommendation, we followed the guidelines of the Oxford Centre for Evidence-Based Medicine (http://www.cebm.net). To consider that a study had level 1 evidence, we arbitrarily chose a cutoff score of 10/15 by the Maastricht criteria, with the item A (adequate method of randomization) being mandatory.

3. Results

The PEDro and Cochrane Library databases did not reveal published reports of trials and studies in addition to those from MEDLINE. After screening titles and excluding duplicates, reports of 21 studies were selected; 10 were excluded and 11 reports of clinical trials were investigated (Fig. 1). The studies are described in Table 1 according to the rehabilitation method investigated.

3.1. Spirometric and respiratory muscle assessment

Although they referred to different recommendations for assessing MIP, MEP and other spirometric parameters, all reports complied with the following quality criteria for measuring lung volume and pressures: MIP was recorded from residual volume and MEP from total lung capacity; participants were asked to repeat each maneuver at least 3 times; variations between values had to be $< 10\%$; and the best of 3 values was retained. The methodology differed mainly in the use of different standard values to express spirometric results [19–21], which affected the pertinence of a meta-analysis.

3.2. Respiratory muscle training (RMT)

Two studies [22,23] evaluated the efficacy of respiratory muscle training (both expiratory and inspiratory). Both protocols used a threshold mouth-resistive device with a unidirectional valve that was set for inspiration or expiration and adjusted to airflow for progressive resistance. Although the Olgiati et al. study was not controlled, after a 4-week program, respiratory muscle strength significantly improved, as assessed by MIP and MEP [22]. Ray et al. compared a 5-week program of assisted RMT to no intervention in an MS population with moderate neurological impairment [23]. MIP and MEP were significantly improved as compared with baseline in the intervention group and as compared with the control group after 5 weeks. The intervention also had a positive effect on fatigue: the global modified fatigue impact scale (MFIS) score and physical and cognitive fatigue sub-scores were all significantly improved ($P = 0.029$, 0.024 and 0.021, respectively). Although health-related quality of life did not significantly improve in the treatment group, the difference between groups was significant at 5 weeks because of a decrease in quality-of-life scores among controls [23].

In keeping with the predominant expiratory muscle dysfunction, 3 studies addressed the efficacy of expiratory muscle training [12,24,25]. The same kind of RMT device, set for expiration load, was used in all 3 studies. The first 2 studies evaluated the same protocol: 3 sets of 15 repetitions repeated twice daily for 3 months. MEP significantly improved in less neurologically impaired patients [12], whereas a homogenous population of wheelchair-dependent participants and bedridden patients did not differ in muscle strength measurements (MIP or MEP) [24]. The third study was a case–control study, and patients were compared to healthy volunteers [25]. Performing daily sets of exercise with progressive resistance for 8 weeks significantly improved MEP and peak expiratory flow (used as a cough efficacy indicator) [25]. However, this study investigated only mild forms of MS. In all 3 studies, baseline MEP was less than MIP. None of the 3 studies used functional outcomes such as fatigue, health-related quality of life or a walking test. For expiratory muscle training, Gosselin et al. evaluated tolerance and did not report any major side effects [24].

Inspiration muscle training was evaluated in 3 other RCTs using similar 10-week protocols [26–28]. The 2 studies by Fry's group

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