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Plantar flexor muscle weakness and fatigue in spastic cerebral palsy patients

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ABSTRACT

Background: Patients with cerebral palsy develop an important muscle weakness which might affect the aetiology and extent of exercise-induced neuromuscular fatigue.
Aim: This study evaluated the aetiology and extent of plantar flexor neuromuscular fatigue in patients with cerebral palsy.
Methods: Ten patients with cerebral palsy and 10 age- and sex-matched healthy individuals (~20 years old, 6 females) performed four 30-s maximal isometric plantar flexions interspaced by a resting period of 2–3 s to elicit a resting twitch. Maximal voluntary contraction force, voluntary activation level and peak twitch were quantified before and immediately after the fatiguing task.
Results: Before fatigue, patients with cerebral palsy were weaker than healthy individuals (341 ± 134 N vs. 858 ± 151 N, $p < 0.05$) and presented lower voluntary activation ($73 \pm 19\%$ vs. $90 \pm 9\%$, $p < 0.05$) and peak twitch (100 ± 28 N vs. 199 ± 33 N, $p < 0.05$). Maximal voluntary contraction force was not significantly reduced in patients with cerebral palsy following the fatiguing task ($-10 \pm 23\%$, $p > 0.05$), whereas it decreased by $30 \pm 12\%$ ($p < 0.05$) in healthy individuals.
Conclusions: Plantar flexor muscles of patients with cerebral palsy were weaker than their healthy peers but showed greater fatigue resistance.
What this paper adds: Cerebral palsy is a widely defined pathology that is known to result in muscle weakness. The extent and origin of muscle weakness were the topic of several previous investigations; however some discrepant results were reported in the literature regarding how it might affect the development of exercise-induced neuromuscular fatigue. Importantly, most of the studies interested in the assessment of fatigue in patients with cerebral palsy did so with general questionnaires and reported increased levels of fatigue. Yet, exercise-induced neuromuscular fatigue was quantified in just a few studies and it was found that young patients with cerebral palsy might be more fatigue resistant that their peers. Thus, it appears that (i) conflicting results exist regarding objectively-evaluated fatigue in patients with cerebral palsy and (ii) the mechanisms underlying this muscle fatigue – in comparison to those of healthy peers – remain poorly understood. The present

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study adds important knowledge to the field as it shows that when young adults with cerebral palsy perform sustained maximal isometric plantar flexions, they appear less fatigable than healthy peers. This difference can be ascribed to a better preservation of the neural drive to the muscle. We suggest that the inability to drive their muscles maximally accounts for the lower extent of exercise-induced neuromuscular fatigue in patients with cerebral palsy.

1. Introduction

Spastic cerebral palsy (CP) is caused by a non-progressive perinatal brain lesion, resulting in abnormal development of the spared corticospinal neurons (Cheney, 1997) and in muscle morphological changes (Barrett & Lichtwark, 2010). Important muscle weakness often ensues and can be quantified as considerably lower-than-normal maximal voluntary contraction (MVC) force compared to age-matched healthy individuals (Brouwer, Wheeldon, Stradiotto-Parker, & Allum, 1998; Elder et al., 2003; Engsberg, Ross, Olree, & Park, 2000; Hussain, Onambele, Williams, & Morse, 2014; Leunkeu, Keefer, Imed, & Ahmaidi, 2010; Moreau, Li, Geaghan, & Damiano, 2008; Rose & McGill, 2005; Stackhouse, Binder-Macleod, & Lee, 2005; Tammik, Matlep, Ereline, Gapeyeva, & Pääsuke, 2007; Wiley & Damiano, 1998). The altered neuronal development may result in impaired connections at the spinal cord level and might subsequently lead to lower pre-synaptic inhibition at Ia-afferent – motoneuron synapses in patients with CP in comparison to healthy individuals (Cheney, 1997). This possible lower pre-synaptic inhibition might explain the greater antagonist coactivation (Brouwer et al., 1998; Engsberg et al., 2000; Hussain et al., 2014; Stackhouse et al., 2005) and the lower voluntary activation (Hussain et al., 2014; Leunkeu et al., 2010; Stackhouse et al., 2005) commonly observed in patients with CP compared to healthy individuals. In addition, it was suggested that patients with CP might present impaired motor unit recruitment and/or firing rate modulation (Marciniak, Li, & Zhou, 2015; Mockford & Caulton, 2010). Together, these alterations might be responsible for the important muscle weakness observed in this pathology. However, even if the primary lesion is located within the central nervous system, neural alterations might not account completely for the typical muscle weakness observed in patients with CP. Indeed, alterations in muscle contractile properties, as assessed with transcutaneous electrical stimulation, were reported for the plantar flexors (Stackhouse et al., 2005; Tammik et al., 2007). Accordingly, a large number of alterations in muscle morphology and structure have been reported. These alterations include reductions in muscle volume and muscle belly length (Barrett & Lichtwark, 2010; Mockford & Caulton, 2010), reductions in cross-sectional area (Barrett & Lichtwark, 2010; Johnson, Miller, Subramanian, & Modlesky, 2009; Mockford & Caulton, 2010), shorter fascicle length (Mohagheghi et al., 2008), greater *in vivo* sarcomere length and stiffness (Smith, Lee, Ward, Chambers, & Lieber, 2011), aberrant acetylcholine receptor localization (Theroux et al., 2002), increases in pennation angles (Mohagheghi et al., 2008), disorganization and disorientation of myofibrils (Marbini et al., 2002), mitochondrial aggregation (Marbini et al., 2002) and increased intermuscular adipose tissue (Johnson et al., 2009). In addition to these morphological and structural alterations, impaired lever arms caused by bone deformities might also contribute to difficulty to use force during daily life activity such as walking (Gage & Novacheck, 2001). Any of these alterations might represent potential causes of weakness.

As highlighted in the review of Brunton and Rice (2012), the substantial muscle weakness observed in this pathology associated with an inefficient gait (Vitiello et al., 2016) might result in a greater percentage of force being required for daily activities and as such lead to greater energy expenditure (Johnston, Moore, Quinn, & Smith, 2004) and reported fatigue in daily life (as assessed with questionnaires/interviews) in patients with CP compared to healthy individuals (Opheim, Jahnsen, Olsson, & Stanghelle, 2009; Van Der Slot et al., 2012). For instance, fatigue can seriously limit locomotion capacity in patients with CP (Brunton & Rice, 2012). However, in the few studies which tried to explore neuromuscular fatigue (defined as a transient reduction in the capacity to generate maximal force after exercise (Gandevia, 2001)), signs of greater resistance to fatigue were reported in patients with CP with respect to healthy individuals (Moreau et al., 2008; Stackhouse et al., 2005), although this is not a universal finding (Leunkeu et al., 2010; Stackhouse et al., 2005). These discrepant findings between the extents of self-reported fatigue and objectively-measured neuromuscular fatigue could probably be attributed to the different nature of the task causing fatigue. Studies assessing reported fatigue by means of questionnaires/interviews commonly evaluate fatigue induced by daily activities, whereas neuromuscular fatigue is objectively measured in response to laboratory-based fatiguing tasks (Brunton & Rice, 2012). While daily activities generally involve multiple-joint or whole-body exercises, laboratory testing mostly consists of single-joint tasks. Laboratory-based testing allows to qualitatively and quantitatively assess neuromuscular adjustments to exercise. Indeed, getting better knowledge of the extent and aetiology of neuromuscular fatigue experienced by patients with CP is primordial for developing optimal rehabilitation programs, which are needed to preserve their force capacity and in turn their locomotor capabilities. If discrepant results exist regarding the extent of neuromuscular fatigue experienced by patients with CP, the aetiology of this neuromuscular fatigue also lacks consensual explanation. Although abnormal motoneuronal projections (Brouwer & Ashby, 1991; Brunton & Rice, 2012; Cheney, 1997; Theroux et al., 2005) might result in altered central fatigue development in response to exercise, alterations occurring within the muscle might also affect neuromuscular fatigue development (Leunkeu et al., 2010; Moreau et al., 2008; Stackhouse et al., 2005).