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Full body gait analysis may improve diagnostic discrimination between hereditary spastic paraplegia and spastic diplegia: A preliminary study

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ABSTRACT

Hereditary spastic paraplegia (HSP) and spastic diplegia (SD) patients share a strong clinical resemblance. Thus, HSP patients are frequently misdiagnosed with a mild form of SD. Clinical gait analysis (CGA) has been highlighted as a possible tool to support the differential diagnosis of HSP and SD. Previous analysis has focused on the lower-body but not the upper-body, where numerous compensations during walking occur. The aim of this study was to compare the full-body movements of HSP and SD groups and, in particular, the movement of the upper limbs. Ten HSP and 12 SD patients were evaluated through a CGA (VICON 460 and Mx3+; ViconPeak®, Oxford, UK) between 2008 and 2012. The kinematic parameters were computed using the ViconPeak® software (Plug-In-Gait). In addition, the mean amplitude of normalised (by the patient's height) arm swing was calculated. All patients were asked to walk at a self-selected speed along a 10-m walkway. The mean kinematic parameters for the two populations were analysed with Mann–Whitney comparison tests, with a significant *P*-value set at 0.05. The results demonstrated that HSP patients used more spine movement to compensate for lower limb movement alterations, whereas SD patients used their arms for compensation. SD patients had increased shoulder movements in the sagittal plane (Flexion/extension angle) and frontal plane (elevation angle) compared to HSP patients. These arm postures are similar to the description of the guard position that toddlers exhibit during the first weeks of walking. To increase speed, SD patients have larger arm swings in the sagittal, frontal and transversal planes. Upper-body kinematics, and more specifically arm movements and spine movements, may support the differential diagnosis of HSP and SD.

1. Introduction

Hereditary spastic paraplegia (HSP) is a rare disorder caused by specific degeneration of the corticospinal tracts. This pathology designates a diverse and heterogeneous group of inherited neurodegenerative disorders (Bien-Willner, Sambuughin, Holley, Bodensteiner, & Sivakumar, 2006). HSP becomes clinically apparent during adolescence or in childhood and progresses slowly throughout the adult years with variable severity of expression (Klebe et al., 2004). HSP is characterised by progressive lower-extremity spasticity and weakness, and it is frequently misdiagnosed as a mild form of spastic diplegia (SD) secondary to cerebral palsy (Fink, 2006). Moreover, the causes and evolution of these two pathologies

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are different. Motor control in patients with SD is associated with a pathological gait pattern in the lower and upper body (Meyns et al., 2011; Romkes et al., 2007) due to a lesion of the central nervous system in the developing brain (Aisen et al., 2011; Dabney, Lipton, & Miller, 1997). Because HSP is progressive and SD is not, the treatments for the two conditions might differ. To obtain an accurate diagnosis and to distinguish patients with HSP from those with SD, magnetic resonance imaging (MRI) of the brain and genetic testing are used (Wren et al., 2011). However, genetic testing cannot always identify HSP reliably (Klebe, Deuschl, & Stolze, 2006). Further, 27 different HSP loci have been discovered so far and different genetic forms of HSP are clinically very similar (Klebe et al., 2006). Consequently, for most HSP patients, a diagnosis of exclusion is used. Even with the emerging availability of laboratory testing for HSP gene mutations, it is still essential that alternative disorders such as SD be excluded by careful history, examination, laboratory studies, neuroimaging and neurophysiologic evaluation.

In this context, a detailed analysis of gait patterns may help to distinguish patients with HSP from those with SD and may help with patient management (Bien-Willner et al., 2006). Clinical gait analysis (CGA), based on three-dimensional movement analyses, objectively quantifies gait patterns. To our knowledge, only three studies have used CGA to compare the gait patterns of HSP and SD patients (Cimolin et al., 2007; Piccinini et al., 2010; Wolf et al., 2011). Their results showed prolonged hip extension during the stance phase of gait and knee hyperextension and ankle plantar flexion during the loading response and stance/swing transition period in HSP compared with SD patients. Similar results were found concerning spatiotemporal and kinematic parameters at proximal joints between the two groups. However, these three studies focused their analyses on the lower limbs and did not include an analysis of the upper extremities (Cimolin et al., 2007; Piccinini et al., 2010; Wolf et al., 2011). Based on the different neuroanatomical levels of impairment for the two pathologies (HSP: corticospinal tracts; SD: brain damage), we hypothesise that upper-body control of movement will be different in each condition.

Although, the upper body (head, arms and trunk) is often regarded as a single unit (Kubo & Ulrich, 2006), and it is considered as "the passenger part" of the gait (Chung, Park, Lee, Kong, & Lee, 2010; Perry, 2010), the upper body plays an important role during gait. Indeed, the trunk represents 60% of the total body mass and it is optimally situated to support distal limb segment mobility, providing a stable base for walking (Gillet et al., 2003; Massion & Frolov, 2004). An altered gait pattern influences trunk movements by means of compensations to help maintain balance and stability. In SD patients, who have poor balance control (Woollacott & Shumway-Cook, 2002), trunk compensations have been studied and highlighted (Adkin, Bloem, & Allum, 2005; Huxham, Baker, Morris, & Iansek, 2008; Linley, Sled, Culham, & Deluzio, 2010; Romkes et al., 2007; Thummerer, von Kries, Marton, & Beyerlein, 2011). However, there is no study on trunk and other upper body movements in HSP patients. In the literature, some studies have reported that arm movements are not essential to gait (Ford, Wagenaar, & Newell, 2007; Marks, 1997; Umberger, 2008) and that these movements are a consequence of walking that aids in efficiency (Collins, Adamczyk, & Kuo, 2009). However, these studies concern healthy subjects and not patients with deficient equilibrium, such as HSP and SD patients. Indeed, several authors have claimed that arm swing during human locomotion enhances gait stability (Bruijn, Meijer, Beek, & van Dieen, 2010; Li, Wang, Crompton, & Gunther, 2001; Meyns, Desloovere, et al., 2012; Milosevic, McConville, & Masani, 2011; Ortega, Fehlman, & Farley, 2008; Perry, 2010). Therefore, the upper body (head, arms and trunk) should be of interest in distinguishing these two populations because HSP patients mainly display movement alterations of the lower limbs, whereas SD patients can also have alterations of the upper limbs.

Thus, the aim of this study was to observe and compare full-body movements in the SD and HSP pathologies, which are similar in term of functional characteristics, independently of the classification of the multiple functional phenotypes found in these conditions. We have hypothesised that, if differences exist in upper-body movements, they could represent compensatory strategies for the gait that is characteristic of each group.

2. Methods

2.1. Patients

Between 2008 and 2012, 19 HSP patients were seen at the Willy Taillard Laboratory of kinesiology, 10 of whom (5 males and 5 females, age: 16.7 ± 5.8 years) were included in this study. The inclusion–exclusion criteria for this group were the following: clinical diagnosis confirmed by a genetic test, reliability of the gait data, available video documentation, no surgery within 1 year before the CGA, no pharmacological treatment in the 6 months before the CGA and the ability to walk without assistive devices. The gross motor function score (GMFSC) in 3 cases were classified as GMFSC 1 and GMFSC 2 in 7 cases (Table 1). Five patients had past surgery: Achilles tendon, gastrocnemius, hamstring and soleus lengthening, and anterior tibialis tenotomy and anterior tibialis transfer.

In the same period, 77 SD patients were seen in the laboratory. From this group, 12 SD patients were included (6 males and 6 females, age: 12.3 ± 4.5 years) based on MRI results. The inclusion-exclusion criteria for this group were the same than for the HSP group. Seven cases were classified as GMFSC 1 and 5 as GMFSC 2 (Table 1). Two patients had surgery to lengthen the hamstrings and Achilles tendon.

A control group of 17 normally developing subjects (9 males and 8 females, age: 26.2 ± 2.1 years) was added as a reference.

2.2. Protocol

All patients had a CGA assessment performed during routine follow-up between 2008 and 2012. The evaluation was performed with a 12-camera motion analysis system (VICON Mx3+; ViconPeak®, Oxford, UK) set at a sampling frequency of