Clinical Biomechanics 48 (2017) 63–72

Contents lists available at ScienceDirect

Clinical Biomechanics

journal homepage: www.elsevier.com/locate/clinbiomech

Increased lower limb muscle coactivation reduces gait performance and increases metabolic cost in patients with hereditary spastic paraparesis

Martina Rinaldi[a,b,*], Alberto Ranavolo[c], Silvia Conforto[a], Giovanni Martino[d,e], Francesco Draicchio[c], Carmela Conte[f], Tiwana Varrecchia[a,b], Fabiano Bini[g], Carlo Casali[h], Francesco Pierelli[h], Mariano Serrao[b,h]

[a] Department of Engineering, Roma TRE University, Via Ostiense 159, 00154 Rome, Italy
[b] Rehabilitation Centre, Policlinico Italia, Piazza del Campidano 6, 00162 Rome, Italy
[c] Department of Occupational and Environmental Medicine, Epidemiology and Hygiene, INAIL, Via Fontana Candida 1, 00078 Monte Porzio Catone, Rome, Italy
[d] Centre of Space Bio-Medicine, University of Rome Tor Vergata, Via Orazio Raimondo 18, 00173 Rome, Italy
[e] Laboratory of Neuromotor Physiology, Istituto Di Ricovero e Cura a Carattere Scientifico Santa Lucia Foundation, Via Ardeatina 306, 00179 Rome, Italy
[f] Fondazione Don Gnocchi, Piazzale Morandi 6, 20121 Milan, Italy
[g] Department of Mechanical and Aerospace Engineering, Mechanical & Thermal Measurement Lab, University of Rome Sapienza, Via Eudossiana 18, 00184 Rome, Italy
[h] Department of Medico-Surgical Sciences and Biotechnologies, University of Rome Sapienza, Via Faggiana 34, 04100 Latina, Italy

ARTICLE INFO

Keywords:
Muscle co-activation
Hereditary spastic paraparesis
Gait analysis
Energy consumption
Energy recovery

ABSTRACT

Background: The aim of this study was to investigate the lower limb muscle coactivation and its relationship with muscles spasticity, gait performance, and metabolic cost in patients with hereditary spastic paraparesis.
Methods: Kinematic, kinetic, electromyographic and energetic parameters of 23 patients and 23 controls were evaluated by computerized gait analysis system. We computed ankle and knee antagonist muscle coactivation indexes throughout the gait cycle and during the subphases of gait. Energy consumption and energy recovery were measured as well. In addition to the correlation analysis between coactivation indexes and clinical variables, correlations between coactivation indexes and time–distance, kinematic, kinetic, and energetic parameters were estimated.
Findings: Increased coactivity indexes of both knee and ankle muscles throughout the gait cycle and during the subphases of gait were observed in patients compared with controls. Energetic parameters were significantly higher in patients than in controls. Both knee and ankle muscle coactivation indexes were positively correlated with knee and ankle spasticity (Ashworth score), respectively. Knee and ankle muscle coactivation indexes were both positively correlated with energy consumption and both negatively correlated with energy recovery.
Interpretation: Positive correlations between the Ashworth score and lower limb muscle coactivation suggest that abnormal lower limb muscle coactivation in patients with hereditary spastic paraparesis reflects a primary deficit linked to lower limb spasticity. Furthermore, these abnormalities influence the energetic mechanisms during walking. Identifying excessive muscle coactivation may be helpful in individuating the rehabilitative treatments and designing specific orthosis to restrain spasticity.

1. Introduction

Hereditary spastic paraparesis (HSP) is a heterogeneous group of inherited neurodegenerative disorders characterized by retrograde degeneration of the corticospinal axonal fibers (Lo Giudice et al., 2014). Lower limb spasticity, which predominates on muscle weakness, is the key clinical feature in patients with HSP (Faber et al., 2014). The presence of spasticity in these patients greatly impairs their walking ability and thus their autonomy and quality of life (Klimpe et al., 2012; Orsucci et al., 2014). Although the paraparetic gait has been widely described by an observational point of view (Adams et al., 2015; Bertolucci et al., 2015; Faber et al., 2014; Fink, 2006; Heetla et al., 2015; Lo Giudice et al., 2014), only few recent studies have provided a detailed quantitative analysis of walking strategies (Klebe et al., 2004; Marsden et al., 2012; Piccinini et al., 2011; Serrao et al., 2016). Very recently, we reported the presence of three different abnormal walking patterns in patients with HSP (Serrao et al., 2016) according to the reduction of the lower limb joint range of motion (RoM) induced by spasticity.

* Corresponding author at: Department of Engineering, Roma TRE University, Via Ostiense 159, 00154 Rome, Italy.
E-mail addresses: martina.rinaldi@uniroma3.it (M. Rinaldi), silvia.conforto@uniroma3.it (S. Conforto), g.martino@hsantalucia.it (G. Martino), fabiano.bini@uniroma1.it (F. Bini), carlo.casali@uniroma1.it (C. Casali), francesco.pierelli@uniroma1.it (F. Pierelli), mariano.serrao@uniroma1.it (M. Serrao).

http://dx.doi.org/10.1016/j.clinbiomech.2017.07.013
Received 2 March 2017; Accepted 27 July 2017

Table 1
Patients' characteristics.

Patients	Gender	Height (m)	Body wt. (kg)	Age (yr)	Diagnosis	Onset (yr)	Duration (yr)	SPRS		
								Ashworth ankle	Ashworth knee	Total
P1	F	1.56	66	57	SPG5	36	21	2	2	20
P2	M	1.60	57	34	SPG4	1–2	33	4	3	25
P3	M	1.64	76	67	_AR	45	22	2	3	21
P4	M	1.70	73	58	SPG4	45	13	3	2	27
P5	M	1.77	104	24	SPG4	14	10	1	2	11
P6	M	1.70	88	48	_AR	10	38	2	2	13
P7	M	1.80	85	25	_AD	13	12	1	0	3
P8	M	1.82	109	49	SPG4	37	12	2	2	21
P9	F	1.58	69	72	SPG4	40	32	4	3	31
P10	F	1.62	58	43	SPG4	5	38	1	2	7
P11	F	1.42	56	78	SPG4	45	33	3	3	28
P12	F	1.59	73	56	_AR	35	21	2	3	20
P13	F	1.58	61	64	SPG31	15	49	1	0	12
P14	M	1.57	87	59	_AR	30	29	3	2	28
P15	M	1.64	76	32	_AR	14	18	3	4	26
P16	M	1.70	104	39	_AD	36	3	1	2	12
P17	M	1.81	81	28	SPG4	13	15	1	2	12
P18	M	1.61	78	58	SPG4	43	15	2	3	17
P19	M	1.77	103	70	SPG4	60	10	2	2	23
P20	M	1.65	69	28	_AD	20	8	2	3	16
P21	M	1.86	136	39	SPG3A	20	19	2	2	27
P22	M	1.61	84	62	SPG4	40	22	1	1	5
P23	M	1.83	78	38	SPG4	30	8	3	4	27

AD = autosomal dominant; AR = autosomal recessive; F = female; M = male; SPRS = spastic paraplegia rating scale; _ = molecular diagnosis still not available. The table lists the SPRS scores; higher scores indicate higher disease's severity.

Furthermore, in these patients, we observed an impaired motor drive for lower limb ankle muscles resulting in an increased muscle coactivation. It is known that muscle coactivation is an important component of motor control (Mari et al., 2014; Patten and Kamen, 2000). Indeed, simultaneous coactivation of antagonist muscles during gait stiffens the joints and ensures lower limb stability (Boudarham et al., 2016; Hirokawa et al., 1991; Peterson and Martin, 2010; Simmons and Richardson, 1998). Conversely, inappropriate coactivation (excessive and/or prolonged) impairs the functional gait performance by increasing the metabolic cost (Dierick et al., 2002; Falconer and Winter, 1985; Macaluso et al., 2002; Peterson and Martin, 2010) and compressive loading across the joint, which may lead to cartilage loss (Childs et al., 2004; Collins et al., 2011; Griffin and Guilak, 2005; Lewek et al., 2004).

High levels of muscle coactivation in knee and in ankle joints during gait have been reported in elderly people (Peterson and Martin, 2010), individuals who have undergone knee arthroplasty (Fallah-Yakhdani et al., 2012), patients with several central nervous system lesions including Parkinson's disease (Dietz et al., 1995), cerebellar ataxia (Mari et al., 2014), and multiple sclerosis (Boudarham et al., 2016). Increased muscle coactivation in these neurological disorders seems to reflect different abnormalities of the motor control. On the one hand, muscle coactivation may be the result of balance-related adaptive compensatory mechanism aimed at reducing instability in the lower limbs such as in cerebellar ataxia (Mari et al., 2014; Martino et al., 2014). On the other hand, it is an expression of primary deficits reflecting either abnormal descending motor commands or lack of reciprocal inhibition of the neural circuits of the spinal cord such as in Parkinson's disease (Meunier et al., 2000).

Up to now, the causal relationship between spasticity and increased coactivation is unknown. Coactivation may be somehow linked to spasticity. For instance, a rearrangement of the interneuronal circuits may be the common neural mechanism at the bases of both features. Otherwise, coactivation may reflect the lack of selectivity by descending drive in tuning the motoneurons of agonist/antagonist muscles. So far, no studies have investigated the relationship between lower limb muscle coactivation and joint kinematic, kinetic, and energetic parameters in patients with HSP.

In order to clarify the role of muscle coactivation on the gait performance of HSP patients, we analyzed a sample of 23 HSP patients from a kinematic, kinetic, electromyographic, and energetic point of view. Identifying the level of muscle coactivation may be helpful in individuating the rehabilitative treatments and designing specific orthosis that may provide a greater joint stability, thus restraining spasticity.

The first aim of this study was to determine the level of coactivation of agonist-antagonist muscles at the knee and ankle joints during gait in individuals with varying severity of HSP compared to control participants. The second aim was to evaluate the relationship between muscle coactivation during gait and limb spasticity, energy consumption, and gait performance in patients compared to control subjects.

We hypothesized increased lower limb muscle coactivation in patients with hereditary spastic paraparesis, compared to controls, that may reflect a primary deficit linked to lower limb spasticity and influence the energetic cost during walking.

2. Methods

2.1. Subjects

Twenty-three patients with HSP were recruited (6 women and 17 men, age: mean 49.04 (SD 16.31) years, height: mean 1.67 (SD 0.11) m, weight: mean 80.07 (SD 21.70) kg). All patients included in the study were able to walk without assistance or walking aids on a level surface. None of the patients showed any involvement of neurological systems other than the pyramidal one (e.g., cerebellar or sensory deficits). The severity of the disease was rated using the spastic paraplegia rating scale (SPRS). The spasticity of ankle and knee joint muscles was scored by the modified Ashworth scale included in the SPRS as a spasticity-related subscale (Schüle et al., 2006). Table 1 summarizes the patients' clinical features and genotypes. Five of the twenty-three patients were taking oral antispastic drugs (baclofen or tizanidine) for 4 to 6 years. Their clinical assessment (SPRS) did not change over the last six months prior to the study. At the time of the evaluation, all patients were