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# Muscle paresis and passive stiffness: Key determinants in limiting function in Hereditary and Sporadic Spastic Paraparesis

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ABSTRACT

*Background:* People with Hereditary and Sporadic Spastic Parapresis (SP) walk with a stiff legged gait characterised by a lack of knee flexion.
*Objective:* We investigated the relationship between lower limb strength and stiffness and knee flexion during swing phase while walking in 20 people with SP and 18 matched controls.
*Methods:* Maximal isometric strength was measured using a dynamometer. Passive stiffness and spasticity was assessed during motor-driven slow (5°/s) and fast (60°/s) stretches at the ankle and knee while the subject was relaxed or preactivating the muscle. Walking was assessed using 3D motion analysis.
*Results:* Isometric muscle strength was decreased in people with SP with over a 50% reduction in strength being found in the ankle dorsiflexors. Passive stiffness, assessed during slow stretches, was 35% higher in the plantarflexors in people with SP ($P < 0.05$). Faster stretches induced large stretch evoked muscle activity and over a 110% increase in stiffness at the ankle and knee in people with SP reflecting the presence of spasticity ($P < 0.05$). However, stretch reflex size and stiffness was similar between the groups following identical stretches of the pre-activated muscle ($P > 0.05$). Lower knee flexion during swing phase was associated with reduced knee flexion velocity at the end of stance phase which in turn was associated with reduced plantarflexor strength and increased passive stiffness in the knee extensors.
*Conclusions:* The relative importance of muscle paresis and passive stiffness in limiting walking in SP suggests that these impairments should be the target of future therapies.


## 1. Introduction

Hereditary Spastic Paraparesis (SP) is an heterogeneous degenerative condition. In the type I or uncomplicated presentation people present with predominately lower limb paresis and spasticity with ~40% of people showing an additional reduction in vibration sense. In the type II or complicated forms there may be added signs, for example, myopathy, cerebellar ataxia or dementia [1,2].

Pathological studies in spastic paraparesis reveal a dying back axonal degeneration of the corticospinal tracts, fasciculus cuneatus and spinoceberebellar tracts with additional degeneration of the corpus callosum in people with dementia [3]. Both autosomal, recessive and X linked forms of inheritance have been described with abnormalities in axonal transport being implicated in the pathogenesis of the most common form caused by mutation of the spastin gene [4]. Spastin mutations are also seen in ~13% of people with sporadic onset of spastic paraparesis restricted to the legs [5,6].

Difficulties with walking and standing balance are commonly reported in people with spastic paraparesis. People with Hereditary and Sporadic SP trend to walk with a stiff-legged gait characterised by a reduction in knee flexion during swing phase, often with the addition of increased hip adduction during the swing phase [7,8]. Reduced knee flexion can lead to an increased incidence of trips and falls and to compensatory strategies such as leg circumduction that can greatly increase the effort of walking.

Typically such patterns of walking are felt to be mainly caused by the presence of spasticity [9,10]. Indeed excessive muscle activity of the knee extensors, such as rectus femoris, during preswing and swing phase could limit knee flexion during swing phase. However, studies modelling the contributions of individual muscles during walking suggest that there may be multiple factors contributing to a particular gait pattern. The amplitude of knee flexion in swing phase, for example, is strongly dependent on the velocity of knee flexion at the end of stance phase [11]. The degree

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of knee velocity in turn depends on the activity of the ankle plantarflexors and hip flexors which are responsible for initiating swing phase [12]. Therefore, factors that affect swing phase initiation such as muscle paresis of the hip flexors or plantarflexors can cause a stiff legged gait.

Understanding the underlying cause of a given pattern of walking will allow clinicians to more effectively target therapies. This study examined the relationship between the lack of knee flexion in people with spastic paraparesis and their underlying impairment.

## 2. Methods

### 2.1. Recruitment criteria

Twenty people with Spastic Paraparesis (SP) were recruited from the Hereditary Spastic Paraplegia support group, UK and the Neurogenetics and Spasticity clinics at the National Hospital for Neurology and Neurosurgery, London, UK. People were included if they had a clinical diagnosis of type 1 spastic paraparesis and were able to walk at least 100 m with or without a walking aid. Exclusion criteria included the presence of additional neurological or orthopaedic impairments. None of the participants were on regular anti-spasticity medication or had received botulinum toxin injections within the last 3 months. People matched for age, gender and height with no history of neurological or orthopaedic impairment were recruited from colleagues and spouses/friends of people with SP to act as a control group. People participated with informed written consent and the approval of the local ethics committee in accordance with the Declaration of Helsinki.

### 2.2. Clinical outcome measures

Clinical measures of three functional movements were taken. Maximal and normal walking speed and cadence was measured over 10 m; gait aids were used as required and a 2 min rest was given after each trial. Timed sit to stand ($\times 5$) was assessed as participants stood up and down from a chair (50 cm height) with their arms folded. Balance was assessed using the Berg balance scale.

### 2.3. Measurement of walking

Three-dimensional joint kinematics and kinetics were measured via markers placed on standardised bony landmarks and wands (Codamotion, Charnwood dynamics, UK) while the person walked on a customised walkway containing two embedded force plates (9286AA Kistler, Instruments Ltd., Hampshire, UK). People used a gait aid as required (1 stick $n = 6$; 2 sticks/crutches $n = 2$) but without the use of any external electrical stimulation or use of any orthotic. A total of 3 steps with either leg landing on a single force plate were recorded. Control participants walked at a matched speed and cadence. Lines at the start of the walkway indicated the desired step length and auditory cues about the required step frequency were provided via a metronome, practice trials were provided prior to recording the data. Data was AD converted (1 kHz for EMG and 200 Hz for force plate and motion analysis data) for off line analysis.

### 2.4. Measures of impairment

Measures of lower limb impairment were taken after the assessment of walking to avoid muscle fatigue associated with the tests impacting on the pattern of walking. A 20 min rest was provided between the walking test and impairment measures. In all cases the right leg was measured.

*Isometric strength* was measured using a dynamometer (Biodex Systems 3, IPRS Mediquipe, UK). Agonist–antagonist pairs at the hip, knee and ankle were measured in standardised positions (see supplementary material). The axis of the motor was aligned with the axis of the joint and the proximal segment fixed. The maximal voluntary contraction (MVC) was recorded twice and the applied torque was recorded (2 kHz AD sampling rate).

*Limb stiffness* was measured by applying ramp and hold stretches to the ankle plantarflexors and the knee extensors (Biodex Systems 3, IPRS Mediquipe, UK). Stretches had a 5° amplitude with a peak velocity of either 5 or 60°/s with a return velocity of 5°/s. Six stretches per velocity condition were recorded with a 6.5 s inter-stretch interval. The order of the conditions was randomised between participants. Stretches were either delivered with the participant resting or pre-activating the muscle of interest to achieve a torque of 10 Nm. This torque level corresponded to approximately 10% of the maximal voluntary contraction (MVC) achieved by the people with SP. Additionally, control subjects pre-activated their muscle to the same percentage of their MVC that was achieved in their matched participant with SP. Surface electromyography (EMG, MT8 Telemetry, MIE, Leeds, UK) was recorded from the medial head of gastrocnemius, tibialis anterior and rectus femoris and medial hamstrings at mid thigh level with an inter-electrode distance was 2.5 cm. During ankle stretches the participant was supine with the knee extended and the ankle in plantigrade. During knee extensor/flexor stretches the participant was supine with the hip extended and the knee flexed by 90°. The contralateral leg was supported in extension. The torque, position, velocity and surface EMG were AD converted at 2 kHz (Power 1401, Spike 2, Version 5, CED Electronics, Cambridge, UK) and stored for off-line analysis.

## 3. Analysis

### 3.1. Walking

Three dimensional joint angles, internal joint moments and power normalised to body weight were calculated using inverse dynamics (CODAmotion, Leister, UK). One step cycle including ipsilateral and contralateral foot on and foot off was defined from the vertical ground reaction force and the horizontal and vertical acceleration of the toe and heel markers. Each gait cycle was normalised to 100% and 3 cycles for each leg were averaged. Preswing was defined as the period of double stance between contralateral foot down and ipsilateral foot off.

The peak knee flexion and extension amplitude in swing phase and the peak knee flexion velocity in preswing were determined. Peak ankle and hip power generation and knee extensor torque during pre-swing was assessed.

*Isometric strength*: The MVC was defined as the peak difference between maximal and baseline torque and was normalised to the body weight.

*Limb stiffness*: Imposed stretches were aligned to the onset of the stretch; the first stretch was omitted to allow for the effects of thixotropy and the final 5 stretches were averaged. The average torque and position was calculated over a 100 ms period prior to the onset of the stretch and immediately following the cessation of the stretch. Stiffness was defined as:

$$\text{Stiffness} = \frac{\Delta\,\text{Torque}}{\Delta\,\text{Position}}$$

Stiffness was normalised to the body weight. Surface EMG was filtered (30 Hz low pass filter) and rectified. A stretch-evoked response occurred if the EMG signal moved above a level of the baseline mean + 4 standard deviations within a 25–125 ms post stretch window. The mean amplitude between the onset and offset of activity, when the EMG fell rose and below this level, was calculated.

### 3.2. Statistical analysis

Differences in strength and stiffness between the SP and control group were compared using an unpaired two-tailed *t*-test. Although walking speed during the walking test was not significantly different between groups the control group did tend to walk faster despite auditory and visual cues about step length and cadence. Therefore measures of walking were analysed using an analysis of covariance with walking speed as a covariate. A Bonferroni correction was applied to account for multiple comparisons during the assessment of walking ($n = 6$), muscle strength ($n = 8$) and stiffness ($n = 4$ per muscle). The relationship between impairment and gait-related variables and peak to peak swing phase knee amplitude and peak knee flexion velocity in preswing phase were assessed using a Pearson correlation. Data was felt to be significant if $P < 0.05$.

## 4. Results

Twenty people with spastic paraparesis were compared to 18 healthy participants matched for gender, age, height and weight (Table 1). A family history was present in 15 of the people with SP with five of these having a genetic diagnosis (SPG4 $n = 4$ and X-