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The Effects of Functional Electrical Stimulation on Walking in Hereditary and Spontaneous Spastic Paraparesis

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Objectives: To investigate in people with spastic paraparesis (SP): 1) the factors contributing to foot drop and reduced toe clearance while walking; 2) short-term effects of bilateral functional electrical stimulation (FES) of the common peroneal nerve.

Materials and Methods: Long term (>0.5 years) users of FES with SP were compared to matched controls ($N = 11$ per group). Ankle strength and plantarflexor stiffness and walking kinematics were objectively recorded. The effects of FES on: 1) perceived efficacy; 2) muscle torque and ankle motion; 3) clinical outcome measures and walking kinematics were assessed. Results were compared using an analysis of covariance.

Results: Ankle weakness and stiffness is higher among people with SP. Higher plantarflexor stiffness is associated with reduced swing phase dorsiflexion; higher toe clearance while walking is associated with increased hip flexion. FES increases dorsiflexor torque, improves toe clearance and dorsiflexion in swing phase, and significantly improves walking speed ($p < 0.05$).

Conclusions: There are multiple causes of tripping in people with SP; FES reduces foot drop and improves walking speed.

Keywords: Functional electrical stimulation, hereditary spastic paraparesis, spasticity, stiffness, walking, weakness

Conflict of Interest: In April 2006 the clinical and commercial activities of Department of Medical Physics and Biomedical Engineering were "spun off" from Salisbury NHS Foundation Trust to form a company Odstock Medical Limited (OML). OML is the manufacturer of the ODFS device used in this project. The majority share holding in the company remains with Salisbury NHS Foundation Trust. Members of the FES group at Salisbury District Hospital, including the authors of this manuscript (IS and PT) have been allocated "token shares" that may in the future have monetary value. A proportion of the 4th and 5th author's time is seconded to Odstock Medical Limited to provide clinical FES treatment and general support. However, they remain an employee of Salisbury NHS Foundation Trust, which receives remuneration for the seconded time. Paul Taylor has two patents in his name relating to the ODFS. These are assigned to his employer Salisbury NHS Foundation Trust, which in turn licenses the intellectual property to Odstock Medical Limited.

INTRODUCTION

Hereditary and spontaneous spastic paraparesis (SP) is a heterogeneous degenerative condition that is associated with a dying back axonal degeneration affecting the corticospinal tracts, dorsal columns, and spinocerebellar tracts (1). This can lead to multiple impairments such as weakness, spasticity, and stiffness. The symptoms mainly affect both legs and result in difficulties with standing balance and walking, especially over uneven ground. People with SP (pwSP) often trip due to poor clearance of the foot in swing phase. This, in part, may be related to impaired ankle dorsiflexion in swing phase, although other factors, such as reduced knee and hip flexion, may also affect foot clearance (2). A reduction in dorsiflexion could, in turn, be caused by a number of factors such as dorsiflexion weakness; increases in passive stiffness in the antagonist plantarflexor muscle, or increased spasticity in the plantarflexors (3,4). Understanding the cause of foot drop and reduced foot clearance will help in appropriately targeting focal pharmacological and physical interventions.

Functional electrical stimulation (FES) to the common peroneal nerve can improve dorsiflexion and toe clearance during swing phase and contribute to a reduction in the incidence of tripping following an upper motor neuron (UMN) lesion (5). FES is associated with improvements in walking speed and efficiency in people with hemiplegia as a result of a cerebrovascular accident (CVA) (4–9),

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multiple sclerosis (9), cerebral palsy (10) and spinal cord injury (11). In recent years FES, usually of the common peroneal nerve, has been used clinically in people with SP to improve ankle dorsiflexion and foot clearance during the swing phase of walking.

This study aimed to: 1) assess the factors affecting the amplitude of ankle dorsiflexion and the degree of foot clearance in swing phase in pwSP; 2) describe the short term effect of functional electrical stimulation on: (i) torque generation at the ankle; (ii) lower limb kinematics; and (iii) clinical outcome measures.

METHODS AND MATERIALS

Through the National FES Centre (Salisbury District Hospital, Wiltshire, UK) 11 long-term (>0.5 years) users of FES with hereditary or spontaneous spastic paraparesis were recruited. Participants were able to walk at least 10 m with or without a walking aid. They had no other neurological or orthopedic conditions that could affect their walking. Participants with SP were compared to 11 healthy controls. This study was performed with the approval of the local ethics committee and following informed written consent of the participants.

Investigating the Factors Affecting Dorsiflexion and Toe Clearance in SP

Measuring Impairment

Ankle strength and stiffness were measured using a dynamometer (Biodex Systems 3, IPRS Mediquipe, Little Blakenham, Suffolk, UK). The foot was supported in a manipulandum and the ankle axis was aligned to the axis of the motor. The starting position was plantigrade with the knee extended. The shank, thigh, and trunk were constrained with straps and manual resistance.

Ankle isometric strength: The maximum voluntary contraction (MVC) of the ankle dorsi- and plantar-flexors was assessed. Two contractions were performed in each case with verbal encouragement and visual feedback of the applied torque. The maximal increase in torque above the pre-contraction resting baseline level was determined.

Stiffness of the ankle plantarflexors: This was measured using ramp and hold stretches with the participant resting (5° amplitude; repeated 6× with a 3 sec inter-stretch period). Two speeds were investigated, 5°/sec and 60°/sec. Muscle activity in the ankle dorsi- and plantar-flexors was measured using surface electromyography (EMG; MT8 Telemetry, MIE, Leeds, UK). The torque, position, velocity, and EMG were digitized at 2 kHz (Power 1401, Spike 2, Version 5, CED Electronics, Cambridge, UK) and stored for offline analysis. Position and torque were measured over two periods of 300 msec; immediately prior to stretch onset or immediately after stretch offset (from 200–300 msec for the 60°/sec stretch and 1400–1500 for the 5°/sec stretch).

Stiffness was defined as: Stiffness = change in Torque/change in Position.

Measuring Lower Limb Kinematics

Lower limb kinematics were measured using 3D motion analysis (Codamotion, Charnwood Dynamics, Leicester, UK). Markers were placed on standardized bony landmarks and wands attached to the leg and aligned with the coronal axis of the leg. Foot contact and foot lift off were defined from the acceleration trace of the heel and foot markers. Gait cycles were normalized to 100% duration and the average of four cycles were calculated. The following variables were determined:

1. Foot clearance defined as the lowest vertical height of a marker placed at the level of the 5th metatarsal head during mid-swing phase (the period from 5% of the gait cycle after the onset of swing phase to 5% prior to the onset of foot contact; approximately 65–95% of the gait cycle).
2. Maximum dorsiflexion in swing phase.
3. Maximum knee and hip flexion, hip abduction, and pelvic elevation in swing phase.

Investigating the Short-Term Effects of FES

The exact pattern of stimulation was determined clinically for each individual. The perceived efficacy and any discomfort experienced during walking with the stimulator were assessed using a 0–10 visual analogue scale. For efficacy, 0 was defined as "not at all effective" and 10 defined as "allows the user to walk normally." For discomfort, 0 was defined as "no discomfort" and 10 as "extremely painful."

Participants' walking was assessed either without stimulation (NOSTIM); using bilateral stimulation of the common peroneal nerve (BICP) or using their preferred pattern of stimulation (PREF), as determined by the clinical team. BICP was routinely used by eight people. Other configurations targeted the hip abductors and lumbar extensor muscles ($N = 1$) and the flexor withdrawal reflex with stimulating electrodes placed in the lateral border of the popliteal fossa ($N = 2$).

All participants used an Odstock two channel stimulator (O2CHSII, Odstock Medical Limited, Salisbury, Wiltshire, UK). Stimulation was elicited via foot switches that were placed within the participants' usual footwear and sited either under the heel or more distally depending on their degree of equinus while walking.

For BICP the electrodes were sited such that they elicited maximum dorsiflexion with the ankle aligned midway between inversion and eversion or in slight (~5°) eversion. The clinically prescribed electrode positions were used for the preferred patterns of stimulation. The participants' usual stimulation settings (stimulation frequency; pulse width and ramp up and down times) were noted and used throughout. The order of testing of the stimulus conditions was randomized between participants and rest (>15 min) was provided between the three conditions (NOSTIM, PREF, and BICP). For each condition, the following measures were taken:

1. Stimulus-elicited dorsiflexor torque at the participants' usual stimulus intensity. In 7/11 participants the isometric dorsiflexor torque elicited as current intensity was increased up to a comfortable level was additionally recorded using a dynamometer.
2. Range of movement of dorsiflexion and degree of toe clearance while walking
3. Walking speed as measured over a 10 m walkway. The average of three trials per condition was calculated.
4. Physiological cost index (PCI). Heart rate was recorded when rested after 5 min of sitting and immediately on cessation of walking with the participant still standing. PCI was defined as:

$$\text{PCI} = (\text{Heart rate after walking} - \text{resting heart rate}) / \text{walking speed (beats per meter)}$$

Analysis

Differences in ankle muscle strength, stiffness, and walking speed between the SP and control groups were analyzed using unpaired *t*-tests. Significant differences were seen in walking speed between the control and SP groups (see further discussion). Therefore, differences in the amplitude of kinematic variables between the control and SP groups were analyzed using an analysis of covariance with walking speed as a covariate.

The relationship between ankle muscle strength and stiffness and the amplitude of ankle dorsiflexion and the relationship between lower limb kinematics and toe clearance were assessed using a Pearson correlation. Results were said to be significant if $p < 0.05$. Mean +/– standard deviation is indicated throughout.

RESULTS

Eleven people with SP were assessed (nine male, age 57 years +/– 14.2; mass 78.7 kg +/– 12.8). The mean reported duration of symptoms was 20.4 years (18.6 years); six participants had a family history of SP. Participants with SP were compared to 11 healthy controls (nine males, age 56.4 years (+/–8.0); weight 78.1 kg (+/–7.3).

Investigating the Factors Affecting Dorsiflexion and Toe Clearance in SP

Differences in Ankle Strength and Stiffness

The MVC of the ankle plantar- and dorsi-flexors was significantly reduced in the SP group (Table 1). Ankle plantarflexor stiffness when measured following slow and fast stretches was significantly higher in people with SP. The slow (5°/sec) stretch did not elicit any increase in muscle activity and so measured the passive component of muscle stiffness. The faster stretch in contrast elicited a stretch reflex in all the pwSP (Table 1). The difference in stiffness between the slow and fast stretch-induced stiffness, a marker of stretch reflex excitability, was also significantly higher in the SP group.

Differences in Walking Pattern (NOSTIM vs. Controls)

Five pwSP used walking aids; walking speed was significantly reduced in the SP group (Table 2). Mean differences in the degree of toe clearance of 24 mm were observed between the SP and control group. In pwSP, lower walking speeds were associated with lower toe clearance ($R^2 = 0.43$ $p < 0.5$). The difference in toe clearance between the SP and control groups was not statistically significant when co-variation due to differences in walking speed were considered. In contrast, hip flexion was significantly higher in the SP group, even accounting for differences in walking speed ($p < 0.05$).

Table 1. Strength and Stiffness Characteristics.

Parameter	SP	Control
Plantarflexor strength (Nm/kg)	0.74 (±0.1)	1.49 (±0.6)*
Dorsiflexor strength (Nm/kg)	0.12 (±0.1)	0.58 (±0.5)*
Stiffness (5°/sec) Nm/rad	74.3 (±29.4)	57.4 (±14.0)*
Stiffness (60°/sec) Nm/rad	120.4 (±53.4)	67.5 (±19.6)*

*Indicates significant difference between the SP and control group.
Mean +/– standard deviation is indicated.

Factors Affecting Ankle Dorsiflexion and Toe Clearance in pwSP

The maximal dorsiflexion amplitude during swing phase in pwSP was correlated to the degree of stiffness observed after the fast stretch ($R^2 = 0.4$, $p < 0.05$), with greater stiffness being associated with lower dorsiflexion amplitude. The size of the correlation decreased when either the slow stretch stiffness or the difference between the fast–slow stretch difference were compared.

Toe clearance was not related to the degree of ankle stiffness or weakness, the range of ankle dorsiflexion, and knee flexion during swing phase or with the degree of hip abduction or pelvic elevation. Greater toe clearance was seen in pwSP who had greater hip flexion in swing phase ($R^2 = 0.34$ $p < 0.05$).

Investigating the Short Term Effects of FES

Usage and Acceptability of FES

Participants had used FES for an average of 2.6 years (+/–1.6). People rated the effectiveness of FES 6.6/10 (+/–1.8) and their discomfort with stimulation as 1.3/10 (+/–2.5). Subjectively, pwSP reported that FES was most helpful when walking longer distances and over rough and uneven terrain and that it provided increased confidence that the foot would not drag.

The clinical characteristics of people who were prescribed bilateral common peroneal stimulation ($N = 8$) or an alternative configuration of stimulation ($N = 3$) is compared in Table 3.

1. Stimulus-elicited torque.
   Stimulation of the common peroneal nerve at the participant's usual level of stimulation resulted in a 30% (+/–22) increase in dorsiflexor torque compared to their MVC. There was a sigmoidal relationship between stimulus intensity ($N = 7$; Fig. 1).
2. Ankle dorsiflexion and toe clearence
   BICP improved toe clearance and maximal range of dorsiflexion during swing phase (Table 2). All other comparisons were non-significant when co-variation due to speed was considered.
3. Clinical outcome measures
   FES resulted in a small but statistically significant increase in walking speed (ANOVA $p < 0.05$; effect size 0.35). The increase in walking speed compared to the NOSTIM condition was slightly larger for PREF (+2.1 m/min ± 0.9) compared to BICP (1.6 m/min ± 0.8). There was no change in the PCI with either type of stimulation [NOSTIM = 1.64 beats/m (± 1.41) BICP = 1.46 beats/m (± 1.02) PREF = 1.36 beats/m (± 0.93)].
4. Effect of alternative stimulation configurations
   In one participant, the left lumbar extensors and right gluteus

Table 2. Walking Characteristics.

Parameter	Control ($N = 11$)	SP (NOSTIM $N = 11$)	SP (BICP $N = 11$)
Walking speed (m/min)	89.4 (±13.6)*	23.7 (12.1)	25.3 (±12.0)†
Toe clearance (mm)	105.6 (±6.6)	81.3 (±5.9)	98.5 (±6.5)†
Maximal dorsiflexion (degrees)	0.13 (±1.0)	3.9 (±2.4)	10.8 (±2.2)†
Maximal knee flexion (degrees)	60.0 (±2.3)	53.8 (±4.0)	50.8 (±3.2)
Maximal hip flexion (degrees)	25.0 (±2.0)*	35.8 (4.5)	37.0 (±2.9)

*Indicates significant difference between control and NOSTIM.
†Indicates significant difference between NOSTIM and BICP conditions.
Mean +/– standard deviation is indicated.

Table 3. Clinical Characteristics of People Who Were Prescribed BICP ($N=8$) and Those Who Were Prescribed Alternative Configurations ($N=3$ see Text for Details).

Parameter	Alternative Stimulation Group ($N=3$)	BICP ($N=8$)
Age (years)	57.7 (12.9)	56.8 (14.5)
Symptom onset (years)	12.5 (17.7)	41.9 (14.2)
Length of FES use (years)	4 (0)	2.1 (1.0)
Stiffness (5°/sec) Nm/rad	70.7 (22.9)	75.7 (32.8)
Stiffness (60°/sec) Nm/rad	143.2 (87.6)	111.9 (40.1)
Plantarflexor strength (Nm/Kg)	0.82 (0.1)	0.69 (0.3)
Dorsiflexor strength (Nm/Kg)	0.16 (0.05)	0.12 (0.07)
Walking speed (m/min) (no stimulation)	27.7 (20.7)	22.1 (8.7)
PCI (beats/m) (no stimulation)	1.59 (1.68)	1.66 (1.43)

Mean +/– standard deviation is indicated.

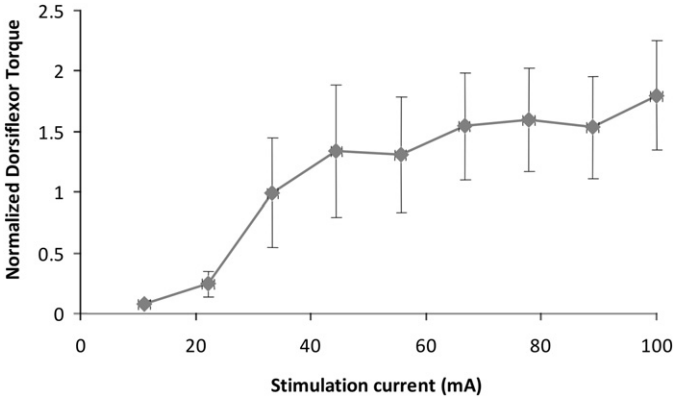

Figure 1. Effect of stimulus intensity on isometric dorsiflexion torque ($N=7$). The torque is shown normalized to the participant's MVC so that 1 indicates people's maximal volitional contraction.

medius/maximus were stimulated on right heel strike with the aim of increasing pelvic elevation and foot clearance on the left side during left swing phase. This stimulation configuration produced an increase of 11.1° in left-sided pelvic elevation and a 1.0 W/kg increase in right hip extensor power generation at the start of stance phase compared to the NOSTIM condition. This was accompanied by an 8 mm increase in left foot clearance, a 3.9 m/min improvement in walking speed and a 0.06 beats/m reduction in PCI.

DISCUSSION

Factors Affecting Ankle Dorsiflexion in SP

People with SP had reduced strength in the ankle dorsiflexors and an increase in stiffness of the ankle plantarflexors compared to matched controls. The ankle plantarflexors were stiffer when measured at either slow or fast speeds of stretching. Slow stretches were not associated with any change in muscle activity; this stretch therefore measured passive stiffness due to changes in the viscoelastic properties of the muscle and surrounding connective tissue. The fast stretch (60°/sec) elicited large short-latency stretch reflexes in all pwSP, indicating the presence of spasticity. The stiffness recorded following a fast stretch is a combination of passive stiffness and stretch-reflex mediated stiffness. The difference in stiffness between the fast and slow stretch was higher in the SP group; this has previously been shown to correlate with the size of the stretch-elicited muscle activity (12).

The range of dorsiflexion in pwSP was related to the degree of ankle stiffness; higher stiffness measured after a fast stretch was associated with a reduction in the range of dorsiflexion. One possibility for this association is that increased stiffness in the plantarflexors (mediated by both stretch reflexes and passive stiffness) limits the range of dorsiflexion. When the variation in walking speed was considered there was no difference in the range of dorsiflexion between the patient and control group. It has been proposed that reductions in walking speed may be a compensatory strategy to reduce the speed of joint angle change and thus decrease stretch-induced resistance from antagonist muscles (13).

Factors Affecting Toe Clearance in SP

Decreased toe clearance during swing phase was seen in pwSP. The causes of reduced toe clearance are potentially complex, with changes in motion of the pelvis, hips, knees, and ankles all potentially contributing (2). In the current sample, reduction in toe clearance was not associated with reductions in the peak ankle dorsiflexion during swing phase but with reductions in more proximal knee and, especially, hip flexion. The overall increase in hip flexion in swing phase seen in the SP group compared to the controls may therefore be a compensatory strategy to reduce the incidence of tripping. With progression of SP over time, the development of hip flexor weakness may prevent people from adopting this strategy. This would result in people with less hip flexion showing lower toe clearance. Overall, this suggests that interventions for tripping and reduced toe clearance should target both proximal and distal joints.

Use and Effect of FES in SP

FES may aid walking by improving toe clearance and ankle dorsiflexion. Most people felt the FES was effective and produced little discomfort. FES resulted in an increase in dorsiflexor torque over and above the level that they can produce volitionally. This suggests that it is mainly a reduction in central drive rather than muscle atrophy that limits active dorsiflexion. The FES further produced an increase in ankle dorsiflexion and toe clearance. Long-term use of FES, as seen in the patient group studied, may well have additional benefits. A reduction in both passive stiffness and spasticity, for example, have been described following long-term (>16 months) use in people with spinal cord injury (14). In 27% of people with SP, alternative stimulation configurations were prescribed that aimed

to either elicit a flexor withdrawal reflex at the knee or to stabilize the pelvis while walking. This group had earlier onset of symptoms and had used FES for longer (Table 3). This suggests that alternative patterns of stimulation may be beneficial as the disease progresses.

CONCLUSION

Significant improvements in walking speed with FES were observed the effect size (0.37) was similar to other groups who have explored the immediate and long term effects of FES post stroke (4–8). Future work should evaluate the effectiveness of FES with a naive population of pwSP who have not used FES in the past. It should include a control group and use outcome measures that directly or indirectly assess the impact on community ambulation and confidence in balance and walking ability.

Authorship Statements

All authors contributed significantly to the design, management, or writing of this paper. Jonathan Marsden: Study design, data collection, analysis. Valerie Stevenson: Patient recruitment, manuscript preparation. Carol McFadden: Patient recruitment, manuscript preparation. Ian Swain: Study design, patient recruitment, manuscript preparation. Paul Taylor: Study design, patient recruitment, manuscript preparation.

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