Gait & Posture 42 (2015) 7–15

Contents lists available at ScienceDirect

Gait & Posture

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

Winner of the 2014 combined ESMAC/SIAMOC meeting's best clinical and best methodological paper award

# The validity and reliability of modelled neural and tissue properties of the ankle muscles in children with cerebral palsy

Lizeth H. Sloot [a,*], Marjolein M. van der Krogt [a], Karin L. de Gooijer-van de Groep [b], Stijn van Eesbeek [c], Jurriaan de Groot [b], Annemieke I. Buizer [a], Carel Meskers [a], Jules G. Becher [a], Erwin de Vlugt [c], Jaap Harlaar [a]

[a] Department of Rehabilitation Medicine, MOVE Research Institute Amsterdam, VU University Medical Center, Amsterdam, The Netherlands
[b] Department of Rehabilitation Medicine, Leiden University Medical Center, Leiden, The Netherlands
[c] Faculty of Mechanical Engineering, Delft University of Technology, Delft, The Netherlands

ARTICLE INFO

*Keywords:*
Muscle stiffness
Muscle spasticity
Cerebral palsy
Biomechanics
Rehabilitation

ABSTRACT

Spastic cerebral palsy (CP) is characterized by increased joint resistance, caused by a mix of increased tissue stiffness, as well as involuntary reflex and background muscle activity. These properties can be quantified using a neuromechanical model of the musculoskeletal complex and instrumented assessment. The construct validity of the neuromechanical parameters was examined (i.e. the internal model validity, effect of knee angle, speed and age, sensitivity to patients versus controls, spasticity severity and treatment), together with the repeatability. We included 38 children with CP and 35 controls. A motor driven footplate applied two slow (15°/s) and two fast (100°/s) rotations around the ankle joint, at two different knee angles. Ankle angle, torque and EMG of the gastrocnemius (GA), soleus (SO) and tibialis anterior (TA) muscle were used to optimize a nonlinear neuromuscular model. Outcome measures were tissue stiffness, reflex and background activity for GA, SO and TA. The internal model validity showed medium to high parameter confidence and good model fits. All parameter could discriminate between patients with CP and controls according to CP pathology. Other measures of external model validity (effect of test position, speed and age) showed behaviour along the lines of current knowledge of physiology. GA/SO background activity was sensitive to spasticity severity, but reflex activity was not. Preliminary data indicated that reflex activity was reduced after spasticity treatment. The between-trial and -day repeatability was moderate to good. The large variance between patients in the ratio of stiffness and neural resistance indicates that the method could potentially contribute to patient-specific treatment selection.



## 1. Introduction

Spastic cerebral palsy (CP) is characterized by increased joint resistance to motion, which is caused by neural as well as tissue impairments [1]. Among the neural impairments are increased reflex activity (i.e. a velocity dependent increase in muscle tone or spasticity [2]), co-contraction, and non-stretch related contractions (i.e. background activation) [3,4]. The tissue impairments comprise altered visco-elastic properties of connective tissues, muscle or tendon [4]. The discrimination between neural and tissue impairments largely guides the selection of treatments that aim to reduce joint resistance. In case of suspected neural origin, muscle activation can be reduced by botulinum toxin (BTX-A) or selective dorsal rhizotomy (SDR) [5,6], while suspected tissue impairments can be treated by corrective casting or splinting [7]. Thus, objective quantification of neuromechanical joint parameters could contribute to patient specific treatment in CP.

Current clinical assessment of joint resistance is based on manual testing of the resistance to slow and/or fast movements, such as the Ashworth and Tardieu-like tests. These clinical tests do not allow for objective quantification, because they are found to be subjective, of low resolution, and limited in discriminating neural and tissue components [8–10]. Instrumented manual measurements have been shown to result in an increased precision [11–13]. Nevertheless, these manual tests still lack standardization of exerted force and movement velocity, while spasticity is known to be force- and velocity dependent [13].

* Corresponding author.
*E-mail address:* l.sloot@vumc.nl (L.H. Sloot).

http://dx.doi.org/10.1016/j.gaitpost.2015.04.006
0966-6362/

Neuromuscular modelling combined with motorized assessment has been demonstrated to quantitatively distinguish between reflex activity and tissue viscoelasticity around the ankle in stroke patients and patients with CP [14,15]. However, previous models only tested in the dorsiflexion direction and did not include the tissue properties of the major antagonist (tibialis anterior), even though its field of activity could overlap with those of a shortened triceps surae muscle. In addition, they modelled the lumped properties of the triceps surae and were thus unable to distinguish between the contribution of the soleus and gastrocnemius. These models did not take any non-stretch related background activity into account [3,4]. Finally, the validity of the model has not yet been established in young children with CP, but only in adolescents, while the impairments of these children worsen during growth and are thus often assessed and treated before the end of their growth.

Therefore, the aim of the study was to present an extended motorized assessment protocol (to separate between soleus and gastrocnemius) combined with an extended neuromuscular ankle model (including antagonists and estimation of background activity); and to assess the construct validity and repeatability of derived neural and tissue parameters. For construct validity, i.e. the extent to which the results correspond with current knowledge of the neuromuscular ankle system in CP by a lack of a true gold standard [16,17], we examined the internal model validity (parameter confidence and model fit), and external model validity (effect of two different test positions, movement speed, and age; patients versus controls; spasticity severity, and treatment).

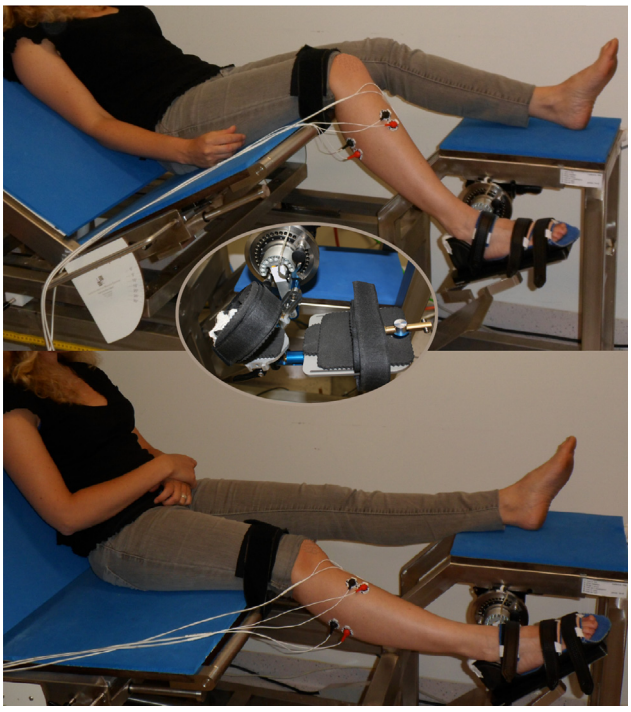

**Fig. 1.** Measurement set-up with 70° (A) and 20° (B) knee flexion. The rigid footplate is shown in both subfigures, the adjustable footplate is shown in the inset. The broad seating area enabled measurement of either the right or the left foot.

## 2. Methods

### 2.1. Subjects

We included a convenience sample of 38 children with CP (10.5 ± 2.9 years; 16 GMFCS-I, 16 GMFCS-II, 6 GMFCS-III; 18 male) and 35 typically developing children (10.1 ± 2.7 years, 15 male). Patients were included if they were between 6 and 18 years old, had a clinical diagnosis of spastic uni- or bilateral CP and excluded if they were not able to extent their knee to 20° knee flexion, had additional medical problems interfering with joint neuromechanical characteristics, or severe cognitive deficits interfering with participation in the study. Informed consent was provided and the study was approved by the local medical ethics committee.

### 2.2. Procedures

All children were measured at the ankle dynamometer. In a random subset of 29 patients (10.3 ± 3.3 years), the spasticity score (SPAT) was collected and subdivided into a low (SPAT 0 and 1, $n$ = 16) and high (SPAT 2+, $n$ = 13) spasticity severity group [18]. Four patients (12.3 ± 1.5 year, GMFCS I–III) were measured before and after treatment (two received BTX-A in the gastrocnemius muscles and two underwent SDR). To assess repeatability, 12 patients (9.8 ± 3.1 years; GMFCS I–II) were measured on two occasions, 9.0 ± 7.3 days apart, without any treatment in between.

### 2.3. Instrumentation

Subjects were seated in an adjustable chair, with a fixed 120° hip angle and the knee adjusted to 20° and 70° flexion (Fig. 1). In patients, the foot of the most affected leg was fixed in an adjustable footplate that allowed for talus repositioning, i.e. correction of ab/adduction and pro/supination of the forefoot with respect to the talus and calcaneus for an optimal fixation (Fig. 1) [19]. In controls, the right foot was fixed in a rigid footplate. The footplates were motor driven and applied rotations around the ankle joint in the sagittal plane (MOOG, Nieuw Vennep, The Netherlands). The axes of the ankle (talo crural joint) and the motor were visually aligned by minimizing knee translation during rotation. EMG electrodes (Ø 15 mm, 24 mm inter-electrode distance) were placed on the tibialis anterior (TA), soleus (SO), gastrocnemius medialis and lateralis (together GA) muscles according to SENIAM guidelines [20]. Angular displacement, foot reaction torque and muscle activity (Porti7, TMSi B.V., The Netherlands) were measured at 1024 Hz. EMG was high-pass filtered (bi-directional 3rd order Butterworth at 20 Hz), rectified and low-pass filtered (uni-directional 4th order Bessel filter at 20 Hz) to obtain the envelope. The angle and torque data were identically low-pass filtered and all data were resampled to 128 Hz.

### 2.4. Measurement protocol

The ankle angle offset was calibrated by measuring the position of the footplate corresponding to 30° ankle plantar flexion, which was determined by goniometry. The passive ankle range of motion (ROM) was determined by imposing an age- and disorder-dependent maximal flexion and extension torque (Appendix A). Next, two repetitions of ramp-and-hold rotations were imposed at 15 and at 100°/s, starting at a random time instant to prevent anticipation and with at least 20 s rest between measurements. Subjects were instructed to remain relaxed and not resist any motion during the measurements. The measurements were performed at both 20° and 70° knee flexion, conform clinical testing [18]. The outcomes at these angles are from here on referred to as SO and GA/SO, because the soleus is expected to be the dominant muscle at 70° knee flexion, since the gastronemii are too short to generate force [21]. TA properties were included for both angles, but examined at 20° knee flexion.

### 2.5. Neuromuscular model

Ankle angle and EMG signals were cropped from start to stop of the dorsal and plantar flexion movement and used as an input to