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Differences of the ankle plantar flexor length in typically developing children and children with spastic hemiplegic cerebral palsy

Dae-Yeon Lee*

Faculty of Liberal Arts, Kangnam University, Yongin, Korea

The purpose of this study is to analyze the lengths of the medial and lateral gastrocnemius and soleus muscles in children with spastic hemiplegic cerebral palsy to quantitatively assess the structural differences in skeletal muscles. This study included 10 children with spastic cerebral palsy and 10 children with typically development. To assess the changes in the length of the ankle plantar flexor due to cerebral palsy, we utilized both gait analysis and software for interactive musculoskeletal modeling to model skeletal muscle length. With this model, the differences in the lengths of the medial and lateral gastrocnemius and soleus muscles were assessed at different knee (0°, 45°, and 90°) and ankle (-10°, 0°, 15°, and 30°) angles. Muscle length on the paretic group was shorter than the typically developing and nonparetic group for all

three muscles (medial and lateral gastrocnemius and soleus muscles) for knee and ankle angles. These results were not statistically significant. Normalized muscle lengths in the dynamic/static status revealed a significant difference in the length of the lateral gastrocnemius muscle between the cerebral palsy and typically developing group. I observed muscle shortening on the paretic side of the children with cerebral palsy. This finding suggests that the recovery of plantar flexor length is the most important issue that must be resolved for normal gait and motor function.

Keywords: Cerebral palsy, Muscle length, Musculoskeletal modeling, Triceps surae

INTRODUCTION

Cerebral palsy (CP) is a common disease of the central nervous system arising from abnormal brain development (Barrett and Lichtwark, 2010). It results in severe debilitation of normal gait and motor function. More specifically, spasticity of lower limb muscles or dysfunction in nerve control causes loss of normal muscle activity during gait, leading to secondary changes in the skeletal system and consequent abnormal gait patterns (Barber et al., 2011). Equinus gait is the most commonly observed alteration in the gait pattern seen in patients with CP. This dysfunction is caused mainly by the spasticity or contracture of the gastrocnemius and soleus muscles (Huijing et al., 2013). Patients with this gait pattern exhibit abnormal gait due to unstable stance phase, foot-dragging during swing phase, and hyperextension of the

knee joint. To date, multiple methods are available to evaluate these symptoms, but traditional physical examination and gait analysis are still commonly used (Ko et al., 2014). Muscle length, for example, is often directly measured via ultrasound or magnetic resonance imaging (MRI). Measurement of muscle lengths using these imaging techniques has limitations since these methods cannot provide real-time data for changes in muscle length while gait or from different knee/ankle angles (Kawakami et al., 1998).

Recent developments in musculoskeletal modeling methods allow actual lengths of human muscle—along with gait analysis data—to be measured in real time, and changes in muscle length during different movements can be obtained (Laracca et al., 2014). Thus, analyses can be performed to assess associations between various types of motor dysfunction and muscle function/length during movements. Previous studies have assessed various aspects

*Corresponding author: Dae-Yeon Lee 6 https://orcid.org/0000-0003-0961-4167
Faculty of Liberal Arts, Kangnam University, 40 Gangnam-ro, Giheung-gu,
Yongin 16979, Korea
E-mail: leedy@kangnam.ac.kr
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of these relationships, including normal muscle lengths, and effects of surgery and different exercise therapies for pediatric patients with CP (Malaiya et al., 2007). The plantar flexors of the lower limb include the medial and lateral gastrocnemius muscles. These two-joint muscles are affected both ankle and knee joints (Lieber and Fridén, 2000).

Since effects on gait appear to be different depending on knee and ankle angles, further analysis should consider such dynamic changes. Therefore, we have recorded real-time measurements of changes in the lower plantar flexor length due to CP and performed analysis of muscle length at various knee and ankle angles in parallel.

MATERIALS AND METHODS

Subjects

Ten children with spastic hemiplegic cerebral palsy (SHCP) (age, 83.4 ± 18.2 months; height, 122.8 ± 12.5 cm; weight, 27.5 ± 9.7 kg) and 10 children with typically development (TD) (age, 87.6 ± 9.8 years; height, 123.7 ± 7.9 cm; weight, 25.7 ± 8.2 kg) were included for this study. They voluntarily agreed to participate in the study, and signed an informed consent form in accordance with the ethical standards of the Declaration of Helsinki.

Measurements

For gait analysis, 16 reflective markers were attached on anatomical landmark points, and participants were asked to walk on a 9-m path at a specified speed. During gait, a three-dimensional motion analysis system using a Vicon 370 Motion Analysis Sys-

tem (Oxford Metrics Inc., Oxford, UK) and six infrared cameras were used to mark and store trajectories of the markers at 120 Hz. To assess muscle lengths at different joint angles, data obtained via gait analysis were loaded into the software for interactive musculoskeletal modeling (SIMM) (Motion analysis, Santa Rosa, CA, USA). With fully extended knee position (0°) as a standard, muscle lengths were measured at three different angles (0°, 45°, and 90°). Similarly, after assigning minus and plus to angles of dorsiflexion and plantar flexion, respectively, muscle lengths were measured at four different ankle angles (-10°, 0°, 15°, and 30°).

Using this model, we recorded real-time measurements of the changes in the length of the medial and lateral gastrocnemius muscle and soleus muscle while gait. Moreover, we performed comparative analysis of muscle lengths between the paretic (SHCP-P) and nonparetic (SHCP-NP) sides of the patient group and the control group (TD children). To adjust for differences in the height among participants, we normalized the measurements by dividing the muscle lengths measured while gait by muscle length measured while standing (dynamic muscle length/static muscle length).

Statistical analysis

All data collected from the study were analyzed using IBM SPSS ver. 18.0 (IBM Co., Armonk, NY, USA). For the comparison of muscle lengths at different angles for each muscle, one-way repeated measures analysis of variance and Bonferroni correction was performed as a *post boc* test. All statistical significance levels were set as P < 0.05.

Table 1. Muscle length of ankle plantarflexor in typically developing children and spastic

		0°			45°			90°	
Ankle position	TD	SHCP-NP	SHCP-P	TD	SHCP-NP	SHCP-P	TD	SHCP-NP	SHCP-P
-10°	31.6±4.8	31.9±4.8	31.2 ± 4.3	30.6 ± 4.3	31.0±5.1	30.3 ± 5.3	29.8 ± 4.1	30.3±3.2	29.6±3.4
0°	30.9 ± 3.4	31.3 ± 4.2	30.7 ± 3.8	30.0 ± 3.3	30.2 ± 4.2	29.8 ± 4.5	29.3 ± 2.2	29.7 ± 4.1	29.0 ± 3.3
15°	29.8 ± 3.9	30.2 ± 4.2	29.6 ± 4.2	29.1 ± 4.8	29.4 ± 3.8	28.7 ± 4.5	28.2 ± 4.2	28.7 ± 4.6	28.0 ± 3.6
30°	28.9 ± 3.1	29.7 ± 4.0	28.8 ± 3.7	27.9 ± 3.9	28.3 ± 3.9	27.7 ± 3.9	27.3 ± 3.3	27.7 ± 4.2	26.9 ± 4.1
-10°	$31.7 \pm 4.6^{a,b}$	32.1 ± 4.1	31.3 ± 4.5	$30.9 \pm 4.3^{a,b}$	31.3 ± 3.8	30.6 ± 2.8	$30.2 \pm 4.0^{a,b}$	30.6 ± 4.8	29.9 ± 3.9
0°	$31.2 \pm 4.5^{a,b}$	31.6 ± 4.4	30.8 ± 4.2	$30.3 \pm 4.7^{a,b}$	30.8 ± 4.3	30.0 ± 3.9	$29.5 \pm 4.3^{a,b}$	30.0 ± 3.2	29.1 ± 4.4
15°	$30.1 \pm 4.1^{a,b}$	30.6 ± 3.8	29.8 ± 3.9	$29.6 \pm 4.5^{a,b}$	29.9 ± 4.6	28.9 ± 3.3	$28.5 \pm 4.1^{a,b}$	29.0 ± 4.4	28.2 ± 3.8
30°	$29.4 \pm 4.4^{a,b}$	29.9 ± 3.1	28.8 ± 4.1	$28.3 \pm 3.9^{a,b}$	28.8 ± 3.6	27.9 ± 3.3	$27.5 \pm 4.2^{a,b}$	27.9 ± 3.8	27.3 ± 4.1
-10°	20.6 ± 3.3	20.8 ± 4.1	20.4 ± 4.4	20.7 ± 3.6	20.9 ± 3.9	20.5 ± 3.2	20.6 ± 4.1	20.9 ± 2.8	20.3 ± 3.1
0°	20.0 ± 3.3	20.3 ± 4.0	19.9 ± 2.9	20.0 ± 3.3	20.2 ± 3.1	19.9 ± 2.9	20.0 ± 3.3	20.3 ± 2.2	19.8 ± 3.1
15°	19.0 ± 2.2	19.2 ± 3.1	18.9 ± 2.9	19.2 ± 4.0	19.2 ± 3.7	18.8 ± 3.6	19.0 ± 3.1	19.2 ± 3.3	18.9 ± 3.6
30°	18.0 ± 4.3	18.8 ± 3.6	17.9 ± 3.7	18.0 ± 3.9	18.2 ± 3.7	17.9 ± 3.3	18.0 ± 3.7	18.2 ± 3.7	18.0 ± 3.5
	-10° 0° 15° 30° -10° 0° 15° 30° -10° 0° 15° 30° -10°	nkle position TD -10° 31.6±4.8 0° 30.9±3.4 15° 29.8±3.9 30° 28.9±3.1 -10° 31.7±4.6 ^{a,b} 0° 31.2±4.5 ^{a,b} 15° 30.1±4.1 ^{a,b} 30° 29.4±4.4 ^{a,b} -10° 20.6±3.3 0° 20.0±3.3 15° 19.0±2.2	nkle position TD SHCP-NP -10° 31.6±4.8 31.9±4.8 0° 30.9±3.4 31.3±4.2 15° 29.8±3.9 30.2±4.2 30° 28.9±3.1 29.7±4.0 -10° 31.7±4.6ab 32.1±4.1 0° 31.2±4.5ab 31.6±4.4 15° 30.1±4.1ab 30.6±3.8 30° 29.4±4.4ab 29.9±3.1 -10° 20.6±3.3 20.8±4.1 0° 20.0±3.3 20.3±4.0 15° 19.0±2.2 19.2±3.1	Inkle position TD SHCP-NP SHCP-P -10° 31.6±4.8 31.9±4.8 31.2±4.3 0° 30.9±3.4 31.3±4.2 30.7±3.8 15° 29.8±3.9 30.2±4.2 29.6±4.2 30° 28.9±3.1 29.7±4.0 28.8±3.7 -10° 31.7±4.6³ab 32.1±4.1 31.3±4.5 0° 31.2±4.5³ab 31.6±4.4 30.8±4.2 15° 30.1±4.1³ab 30.6±3.8 29.8±3.9 30° 29.4±4.4³ab 29.9±3.1 28.8±4.1 -10° 20.6±3.3 20.8±4.1 20.4±4.4 0° 20.0±3.3 20.3±4.0 19.9±2.9 15° 19.0±2.2 19.2±3.1 18.9±2.9	Inkle position TD SHCP-NP SHCP-P TD -10° 31.6 ± 4.8 31.9 ± 4.8 31.2 ± 4.3 30.6 ± 4.3 0° 30.9 ± 3.4 31.3 ± 4.2 30.7 ± 3.8 30.0 ± 3.3 15° 29.8 ± 3.9 30.2 ± 4.2 29.6 ± 4.2 29.1 ± 4.8 30° 28.9 ± 3.1 29.7 ± 4.0 28.8 ± 3.7 27.9 ± 3.9 -10° $31.7 \pm 4.6^{a.b}$ 32.1 ± 4.1 31.3 ± 4.5 $30.9 \pm 4.3^{a.b}$ 0° $31.2 \pm 4.5^{a.b}$ 31.6 ± 4.4 30.8 ± 4.2 $30.3 \pm 4.7^{a.b}$ 15° $30.1 \pm 4.1^{a.b}$ 30.6 ± 3.8 29.8 ± 3.9 $29.6 \pm 4.5^{a.b}$ 30° $29.4 \pm 4.4^{a.b}$ 29.9 ± 3.1 28.8 ± 4.1 $28.3 \pm 3.3^{a.b}$ -10° 20.6 ± 3.3 20.8 ± 4.1 20.4 ± 4.4 20.7 ± 3.6 0° 20.0 ± 3.3 20.3 ± 4.0 19.9 ± 2.9 20.0 ± 3.3 15° 19.0 ± 2.2 19.2 ± 3.1 18.9 ± 2.9 19.2 ± 4.0	Inkle position TD SHCP-NP SHCP-P TD SHCP-NP -10° 31.6±4.8 31.9±4.8 31.2±4.3 30.6±4.3 31.0±5.1 0° 30.9±3.4 31.3±4.2 30.7±3.8 30.0±3.3 30.2±4.2 15° 29.8±3.9 30.2±4.2 29.6±4.2 29.1±4.8 29.4±3.8 30° 28.9±3.1 29.7±4.0 28.8±3.7 27.9±3.9 28.3±3.9 -10° 31.7±4.6ab 32.1±4.1 31.3±4.5 30.9±4.3ab 31.3±3.8 0° 31.2±4.5ab 31.6±4.4 30.8±4.2 30.3±4.7ab 30.8±4.3 15° 30.1±4.1ab 30.6±3.8 29.8±3.9 29.6±4.5ab 29.9±4.6 30° 29.4±4.4ab 29.9±3.1 28.8±4.1 28.3±3.9ab 28.8±3.6 -10° 20.6±3.3 20.8±4.1 20.4±4.4 20.7±3.6 20.9±3.9 0° 20.0±3.3 20.3±4.0 19.9±2.9 20.0±3.3 20.2±3.1 15° 19.0±2.2 19.2±3.1 18.9±2.9 19.2±4.0 19.2±3.7	Inkle position TD SHCP-NP SHCP-P TD SHCP-NP SHCP-P -10° 31.6±4.8 31.9±4.8 31.2±4.3 30.6±4.3 31.0±5.1 30.3±5.3 0° 30.9±3.4 31.3±4.2 30.7±3.8 30.0±3.3 30.2±4.2 29.8±4.5 15° 29.8±3.9 30.2±4.2 29.6±4.2 29.1±4.8 29.4±3.8 28.7±4.5 30° 28.9±3.1 29.7±4.0 28.8±3.7 27.9±3.9 28.3±3.9 27.7±3.9 -10° 31.7±4.6ab 32.1±4.1 31.3±4.5 30.9±4.3ab 31.3±3.8 30.6±2.8 0° 31.2±4.5ab 31.6±4.4 30.8±4.2 30.3±4.7ab 30.8±4.3 30.0±3.9 15° 30.1±4.1ab 30.6±3.8 29.8±3.9 29.6±4.5ab 29.9±4.6 28.9±3.3 30° 29.4±4.4ab 29.9±3.1 28.8±4.1 28.3±3.9ab 28.8±3.6 27.9±3.3 -10° 20.6±3.3 20.8±4.1 20.4±4.4 20.7±3.6 20.9±3.9 20.5±3.2 0° 20.0±3.3 20.3±4.	Inkle position TD SHCP-NP SHCP-P TD SHCP-NP SHCP-P TD -10° 31.6±4.8 31.9±4.8 31.2±4.3 30.6±4.3 31.0±5.1 30.3±5.3 29.8±4.1 0° 30.9±3.4 31.3±4.2 30.7±3.8 30.0±3.3 30.2±4.2 29.8±4.5 29.3±2.2 15° 29.8±3.9 30.2±4.2 29.6±4.2 29.1±4.8 29.4±3.8 28.7±4.5 28.2±4.2 30° 28.9±3.1 29.7±4.0 28.8±3.7 27.9±3.9 28.3±3.9 27.7±3.9 27.3±3.3 -10° 31.7±4.6ab 32.1±4.1 31.3±4.5 30.9±4.3ab 31.3±3.8 30.6±2.8 30.2±4.0ab 0° 31.2±4.5ab 31.6±4.4 30.8±4.2 30.3±4.7ab 30.8±4.3 30.0±3.9 29.5±4.3ab 15° 30.1±4.1ab 30.6±3.8 29.8±3.9 29.6±4.5ab 29.9±4.6 28.9±3.3 28.5±4.1abb 30° 29.4±4.4ab 29.9±3.1 28.8±4.1 28.3±3.9ab 28.8±3.6 27.9±3.3 27.5±4.2abb -10°	Inkle position TD SHCP-NP SHCP-P TD SHCP-NP SHCP-P TD SHCP-NP SHCP-P TD SHCP-NP TD SHCP-NP </td

MG, medial gastrocnemius; LG, lateral gastrocnemius; Sol, soleus; TD, typically developing children; SHCP, spastic hemiplegic cerebral palsy; NP, nonparetic side; P, paretic side. a.b. Significant difference in normalized muscle length ratio between TD vs. SHCP-NPa¹, TD vs. SHCP-Pb¹ groups (P<0.05).



RESULTS

Muscle length of the SHCP-P was decreased than the SHCP-NP and TD for all muscles under all conditions (0°, 45°, 90° and -10°, 0°, 15°, and 30° knee joint and ankle joint angles, respectively). The differences in muscle lengths were not statistically significant under any of these conditions. The proportion of normalized muscle length was lowest in the TD group under all conditions, and highest in SHCP-NP. For the lateral gastrocnemius muscle, significant differences were observed between the TD and the SHCP-P and SHCP-NP respectively (P < 0.05) (Table 1).

DISCUSSION

The length of the gastrocnemius muscle during gait in normal humans is associated with the kinematics of ankle and knee joints, while the length of the soleus muscle is closely associated with the kinematics of the ankle joint (Matsunaga et al., 2018). Spasticity and contracture of muscles due to cerebral lesions can affect structural changes in the bone. These three factors—bone, joint, and muscle—are necessary components of normal movement and interact with one another to produce quality movements (Mohagheghi et al., 2008). Therefore, analysis of muscle shortening, adequate muscle length, intersecting joints, and the forces applied to joints is essential for understanding smooth movement and gait.

Several previous studies have reported the application of various methods to analyze the muscles of patients with CP (Mohagheghi et al., 2007). Recent developments in medical imaging techniques and musculoskeletal modeling software allow for more diverse approaches. SIMM is a program developed based on MRI data from normal adults. The software utilizes specified gait analysis data to create a model of the musculoskeletal system of a patient and displays the data in the form of a graph or an animation. With the SIMM model, measurement of changes in muscle strength during gait allows for concomitant measurement of muscle length change not limited to the standing position. Moreover, the model reflects changes in each joint position and skeletal structure. This model, thus, provides the most accurate measurement of the muscle length currently available (Shortland et al., 2002).

The gastrocnemius and soleus muscle are essential lower limb muscles, providing support against body weight while gait and adjusting ankle joint angles at both stance and swing phases. Consequently, contracture of the gastrocnemius and soleus muscles is an important factor of muscle dysfunction and inability to perform daily activities (Moreau et al., 2009).

In this study, we measured the changes in the muscle length while gait, confirmed—as known from the previous studies—that both the gastrocnemius and soleus muscles were shortened in patients during equinus gait, and provided quantitative measurements of the severity of this condition. Our findings are consistent with those of previous studies, suggesting that the decreased plantar flexor lengths lead to abnormal gait patterns. Moreover, decreased plantar flexor length also affects structural changes in ankle and knee joints, causing abnormal equinus gait. Novel approaches (i.e., surgical treatment and rehabilitation) to recover normal function in these muscles are actively pursued.

Several previous studies that assessed shortening and spasticity of muscles associated with CP report shortening of various muscles (Wren et al., 2004). However, a study claims that the length of the hamstring muscle is not lesser than normal and that only the ability to change speed is lesser than normal. This difference was likely due to assessment of equinus gait from the aspect of knee joint flexion in the sagittal plane (Tisha et al., 2019).

In this study, we confirmed that changes in the muscle length while gait were closely associated with kinematic data. More specifically, the gastrocnemius muscle is a biarticular muscle closely associated with maximal knee joint angle during the swing phase.

Additionally, we observed that the muscle length of the SHCP-NP was more than the muscle length of TD, although this difference was not statistically significant. These findings are consistent with those of a previous study suggesting that the hemiplegic CP exhibit imbalance due to a relatively overused non-paretic limb, leading to reduced muscle strength or activity in the paretic limb (van der Krogt et al., 2007).

Further, the musculoskeletal model used in this study does not differentiate muscle and tendon but defines them as a single unit. It is not possible to assess whether muscles or tendons cause muscle shortening. Analysis made in parallel using ultrasound or MRI can be combined with data from modeling to allow a more objective assessment.

CONFLICT OF INTEREST

No potential conflict of interest relevant to this article was reported.

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