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Original Article

Effect of performing daily activities while standing on the quantity and quality of the thigh muscles in adults with severe cerebral palsy: a cross-sectional study

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Abstract. [Purpose] To observe the effect of daily standing, as indicated by gross motor function, on the quantity and quality of the thigh muscles in adults with severe cerebral palsy and to obtain data to determine an appropriate intervention that will improve their quality of life. [Participants and Methods] Thirty-three adults with severe cerebral palsy participated in the study. We assessed the gross motor function using the GMFM-66-IS. We then evaluated the quadriceps muscle thickness and the rectus femoris muscle echo intensity using ultrasonography. We divided the participants into the standing and non-standing groups and then examined the correlations of the GMFM-66-IS score to muscle thickness and echo intensity. We calculated the difference in mean muscle thickness and echo intensity between the two groups using an independent t-test. [Results] Significant positive correlations were found between the GMFM-66-IS score and muscle thickness and echo intensity. In the group-specific analysis, no significant correlation was found between echo intensity and the GMFM-66-IS score in either group. Muscle thickness and echo intensity were greater in the participants of the standing group. [Conclusion] Daily standing, as indicated by gross motor function, affected muscle thickness and echo intensity. Quantitative and qualitative data might need to be evaluated when assessing the muscles of adults with severe cerebral palsy using ultrasonography. Key words: Severe cerebral palsy, Standing in daily activities, Changing muscle quality

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INTRODUCTION

Cerebral palsy (CP) is defined as postural and motor impairment caused by a non-progressive lesion in the brain that develops during the fetal or infantile period and which is often associated with cognitive and communication deficits¹). There is a positive correlation between the gross motor function and muscle strength in children with CP^{2} , but it has been difficult to quantitatively measure muscle strength in children with CP with a low voluntary motor and cognitive function in the clinical setting. Recently, however, ultrasonography has been reported to be effective for estimating the muscle strength in

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such children with CP³⁾. Moreau et al.⁴⁾ showed that the thickness of the vastus lateralis muscle measured by ultrasonography in children with CP with Level I–IV in the Gross Motor Function Classification System (GMFCS) was a highly accurate predictor of knee extension strength. This suggests that the measurement of the muscle thickness using ultrasonography may provide a means of estimating the muscle strength in individuals with CP who have a low voluntary motor and cognitive function. Ohata et al.⁵⁾ reported significant positive correlations between the quadriceps muscle thickness and the Gross Motor Function Measure (GMFM)-66 and the selfcare and mobility items of the pediatric evaluation of disability inventory (PEDI), in children with CP (average age: 12.7 years). This indicates that the measurement of the quadriceps muscle thickness in children with CP can be a useful clinical assessment. The morphological assessment of muscles using ultrasonography is a useful method for estimating muscle strength, especially in children with CP who have severe voluntary motor and communication deficits that make it difficult to measure muscle strength. However, there have been no reports on the relationship between the motor function and quadriceps muscle thickness in adults with severe CP who have very low motor performance. Furthermore, although there have been many reports on children with CP, very few reports have focused on adults with CP. Thus, the first aim of the present study was to investigate the relationship between the quadriceps muscle thickness and the gross motor function, focusing exclusively on adults with severe CP.

In a study of healthy elderly people, the correlation between quadriceps muscle thickness and knee joint extension strength was moderate⁶⁾, making it difficult to explain muscle strength from muscle thickness values alone. Muscle strength involves not only the muscle mass and muscle morphology, but also neurological factors, such as the number of active motor units and recruitment frequency, and that there are non-contractile tissues within skeletal muscle, such as connective and fat tissue. The muscle thickness obtained by ultrasound included the muscle fibers and the non-contractile tissues inside muscle^{7, 8)}. It is therefore important to examine not only morphological data, such as muscle thickness, but also qualitative data, such as the concentration of non-contractile tissue in the muscle, when evaluating muscles on ultrasound images. The echo intensity of ultrasound images has been used to quantitatively measure the concentration of intramuscular fat⁷) and interstitial fibrous tissue⁸⁾. A previous study demonstrated a strong correlation between the interstitial fibrous tissue content measured in muscle biopsy specimens and the echo intensity⁸⁾. The skeletal muscle of children with CP has been shown to undergo qualitative changes (i.e., increased connective and adipose tissue)^{9, 10}). Pitcher et al.⁹⁾ compared the echo intensity of the medial gastrocnemius muscle in 40 children with CP (GMFCS I-V) with children with typical development and found that the echo intensity of the medial gastrocnemius muscle was higher in children with CP. In addition, Battisti et al.¹⁰ studied the echo intensity of the lower triceps muscle in children with CP (GMFCS I-IV) who were 4-16 years of age and reported that a higher echo intensity tended to be observed with increasing disease severity. These reports suggest that the muscles may change qualitatively in individuals with CP depending on the severity of the disease. Although several reports have examined the relationship between the severity classification of the motor function and the echo intensity in individuals with CP, no reports have examined the relationship between the echo intensity and the detailed motor function, as assessed using the GMFM. Thus, this study also aimed to assess the concentration of non-contractile tissue in the muscles of adults with severe CP using the echo intensity and to examine the relationship between the echo intensity and the gross motor function.

Pitcher et al.⁹⁾ reported that among CPs at the GMFCS I–V level, the echo intensity of the medial gastrocnemius muscle tended to be highest in GMFCS III. Pitcher et al.⁹⁾ hypothesized that, based on the review by Gough et al.¹¹⁾, eccentric loading may promote increased fat and connective tissue in the muscle in children with CP. Consequently, eccentric loading during gait promoted the highest increase in connective tissue in the muscle of children with GMFCS III. The participants in this study had severe CP (GMFCS IV and V). They only have opportunity to undergo gait training during therapy, but some routinely stood with the use of handrails or with assistance during transfers and toileting movements. We hypothesized that whether individuals are standing in their daily activities may affect quantitative (thickness) and qualitative (echo intensity) states of muscles in adults with severe CP (GMFCS IV and V). Thus, the present study aimed to examine the effect of the opportunity to stand in daily activities on the quadriceps muscle thickness and the echo intensity of the rectus femoris.

The first purpose of this study was to investigate the relationships between the gross motor function and the quadriceps muscle thickness (sum of the rectus femoris and vastus intermedius muscle thickness) and the echo intensity of the rectus femoris measured from ultrasound images in adults with severe CP. The second was to examine the effect of the opportunity to stand in daily activities on the quadriceps muscle thickness and the echo intensity of the rectus femoris. The hypotheses to be tested in this study in adults with severe CP are as follows: 1) the greater the quadriceps muscle thickness, the greater the gross motor function; 2) the greater the echo intensity of the rectus femoris, the lower the gross motor function; 3) participants who have the opportunity to stand in daily activities have a greater quadriceps muscle thickness and the echo intensity of their rectus femoris was lower.

PARTICIPANTS AND METHODS

The participants included 33 of 37 adults with CP who were admitted to Kanazawa Disabled Children's Hospital, in whom the severity of CP was classified as IV or V by the GMFCS-expanded and revised version (E&R)¹²⁾. The GMFCS-E&R is a classification of the five levels of gross motor function in individuals with CP. This classification is the clinical gold standard assessment for the functional classification of individuals with CP¹⁰⁾. In general GMFCS E&R¹²⁾, level I indicates that the individual is not limited in mobility in daily activities; level II indicates that the individual is able to walk alone on level

ground but may use a cane or wheelchair for long distances or on inclines; III indicates that the individual is able to walk with a walker and sit with minimal external support; IV indicates that the individual may be able to move with a wheelchair for a limited range but is often transferred by a caregiver; V indicates that the individual requires extensive external support of the head and trunk to maintain posture, and self-mobility is only possible when the individual has mastered the use of a powered wheelchair. In this study, severe CP were defined as GMFCS IV and V; individuals with this classification generally do not have the ability to walk in daily life. The characteristics of the participants are shown in Table 1. The protocol of this study was approved by the Ethics Committee of Kanazawa Disabled Children's Hospital (Approval No. 2019-1). Informed consent was obtained using an opt-out method. Families who did not provide their consent in this way were excluded from the present study. The experimental protocol was conducted in accordance with the Declaration of Helsinki.

The GMFM has been widely used for the assessment of activities in children with CP and has been shown to have good reliability and validity¹³⁾. The GMFM-66 is a shortened version of the GMFM, with 66 test items identified by a Rasch analysis (the original GMFM included 88 items arranged in order of item difficulty)¹⁴⁾. The GMFM-66 has been shown to have good validity and reliability, as well as the original GMFM¹⁴⁾. Furthermore, Russel et al.¹⁵⁾ developed the GMFM-66-IS (item sets), which shows high agreement with the GMFM-66 using a screening method. The GMFM-66 only includes 15–39 items, allowing for shorter assessment in comparison to the GMFM-66 items¹⁵⁾. In this study, GMFM-66-IS was used to assess gross motor function. The GMFM-66-IS was measured by a physiotherapist (TS) with 35 years of experience, and who was trained in the GMFM in Japan.

Information on the opportunity to stand in daily activities was obtained by a questionnaire given to facility staff. Participants who routinely stood during activities of daily living such as transferring and toileting with caregivers were classified into the standing (ST) group and those who were unable to stand at all were classified into the non-standing (NST) group. If the participants had difficulty in maintaining standing posture with caregiver in actual daily activities even if they were able to stand shortly as part of training with physical therapists, they were classified into the NST group. The reason for this was that participants receive an average of three to four physical therapy per month, but those who can stand with a care giver will be standing significantly more often than those who cannot. One participant in the NST group was performing standing exercises as a physical therapy intervention. The other participants had not performed standing exercises, including the standing board intervention. Previous standing interventions were not tracked in this study. Those in the ST group had opportunity to stand in daily activities every day. None of the participants in this study were able to hold the standing position on their own, and all of the participants in the ST group required assistance or handrails to maintain the standing position. All of the participants in the ST group had a maximum knee joint extension angle of less than -10° , and none of them were able to achieve full knee joint extension in the standing position.

The muscle thickness of the quadriceps, fat thickness of the anterior thigh and the echo intensity of the rectus femoris (EI) were measured with a linear probe (L64, Hitachi, Tokyo Japan) using a B-mode ultrasound system (Noblus, Hitachi, Tokyo, Japan). In our study, one investigator (SE) performed the ultrasound measurements. During the blinded measurement, care was taken to maintain the participants in a standardized position. They were examined in the supine position with the knees flexed at 60° and feet on the bed. To ensure the reliability of comparing echo intensity between participants, the imaging settings were kept unchanged during all measurements. The probe was placed perpendicular to the long axis of the quadriceps muscle at the midpoint of the line connecting the superior anterior iliac spine and the proximal end of the patella. A sufficient amount contact gel was used to eliminate excessive compression due to probe pressure and resulting image distortions, and the ultrasound images were observed in real time. In accordance with the methods of the study of Young et al.⁷, images were taken three times on each side.

The ultrasound images were digitalized, and the images were created and analyzed with the ImageJ software program (Version 1.52a, National Institutes of Health, Bethesda, MD, USA). The muscle thickness of the quadriceps and the echo

Table 1 Destining to have atomistic

Mean age (SD), years	30.1 (11.3)		
Mean height (SD), cm	147.6 (8.9)		
Mean weight (SD), kg	37.4 (9.7)		
Females: males	10:23		
GMFCS level, n (mean age [SD])			
IV	13 (33.1 [8.1]))		
V	20 (28.1 [11.4])		
Type of cerebral palsy, n			
Spasticity (bilateral)	21		
Dyskinetic	6		
Ataxia	6		

GMFCS: gross motor function classification system.

intensity of the rectus femoris were measured from the same image. The muscle thickness of the quadriceps femoris was defined as the distance between the upper edge of the femur and the lower edge of the ventral fascia of the rectus femoris, and the fat thickness of the anterior thigh was defined as the distance between the lower edge of the skin located just above the rectus femoris and the upper edge of the ventral fascia of the rectus femoris, respectively (Fig. 1). To evaluate the EI value, the contour of the rectus femoris muscle was extracted (Fig. 2), and a grayscale analysis of the extracted area was performed. The EI value was defined as the average value of all pixels in the selected area, quantified in 256 shades of gray from 0 to 255 (black=0, white=255). The mean values of the muscle thickness of the quadriceps, the fat thickness of the anterior thigh and EI (measured from three images) were taken as unilateral representative values. In this study, the average of the bilateral representative values was taken as the representative value for each participant. However, if only one side could be measured due to contracture or joint deformity, the unilateral value was taken as the representative value. It has been reported that EI values vary with subcutaneous fat thickness, even in the same muscle, and it is desirable to correct for this using a correction equation⁷). Thus, in this study, the EI values were corrected using the following equation, which differed according to gender⁷).

Male corrected EI=[0.144 * (40 * fat thickness of the anterior thigh) + uncorrected EI] + 1.126

Female corrected EI = [0.062 * (40 * fat thickness of the anterior thigh) + uncorrected <math>EI + 7.901

SPSS ver. 26 (IBM Japan, Tokyo, Japan) was used for all of the statistical analyses. First, the Shapiro-Wilk test showed normality in all data (p>0.05).

The effects of the opportunity to stand in daily activities and gender on GMFM-66-IS, the muscle thickness of the quadriceps femoris, the fat thickness of the anterior thigh and the corrected EI were examined by an independent t-test. The r value was calculated from the t-value and the degree of freedom (df) as an effect size of the t-test using the following equation. $r=\sqrt{\{t2 / (t2 + df)\}}$

Correlations between age and the GMFM-66-IS, the muscle thickness of the quadriceps, the fat thickness of the anterior thigh and the corrected EI were examined using Pearson's Productive Correlation Coefficient. Similarly, correlations between the GMFM-66-IS and the muscle thickness of the quadriceps, fat thickness of the anterior thigh and corrected EI were examined using Pearson's product correlation coefficients.

If the GMFM-66-IS, the muscle thickness of the quadriceps, the fat thickness of the anterior thigh and the corrected EI differed significantly according to gender or the presence or absence of the opportunity to stand in daily activities, the correlations between the GMFM-66-IS and the muscle thickness of the quadriceps, the fat thickness of the anterior thigh and the corrected EI (by group) were examined using Pearson's productive correlation coefficient. Fisher's exact probability test was also performed to examine the proportion of participants in the ST and NST groups between the genders and between GMFCS levels, respectively. The significance level was set at 5%.







Fig. 2. Extraction of the rectus femoris muscle range. Solid lines indicate the internal margin of the fascia of the rectus femoris muscle, and the echo intensity (grayscale) was measured within the selected range.

RESULTS

The mean values of the measured items in all participants are shown in Table 2. There were significant positive correlations between the GMFM-66-IS and the muscle thickness of the quadriceps (r=0.72, p<0.01) and the corrected EI (r=0.47, p<0.01), but not between the GMFM-66-IS and the fat thickness of the anterior thigh (r=0.26, p=0.14).

The GMFM-66-IS (t=-5.10, p<0.01, r=0.68), the muscle thickness of the quadriceps (t=-4.08, p<0.01, r=0.59) and corrected EI (t=-2.53, p=0.02, r=0.41) were significantly greater in the ST group in comparison to the NST group (Table 2). There was no significant correlation between GMFM-66-IS and the muscle thickness of the quadriceps in the ST group (r=0.56, p=0.95), while there was a significant positive correlation between both values in the NST group (r=0.56, p<0.01). There was no significant correlation between GMFM-66-IS and the corrected EI in either the ST group (r=0.21, p=0.57) or the NST group (r=0.30, p=0.16).

There were no significant gender differences in the muscle thickness of the quadriceps (t=0.27, p=0.79, r=0.05) and the GMFM-66-IS (t=-0.76, p=0.94, r=0.14); however, there were significant differences in the fat thickness of the anterior thigh (t=-4.79 p<0.01 r=0.65) and the corrected EI (t=-2.92, p<0.01, r=0.46), which were all significantly greater in female (Table 3). In males, there were significant correlations between the GMFM-66-IS and the muscle thickness of the quadriceps (r=0.71, p<0.01), the fat thickness of the anterior thigh (r=0.60, p<0.01), and the corrected EI (r=0.71, p<0.01). In females the GMFM-66-IS only showed a significant correlation with the muscle thickness of the quadriceps (r=0.74, p=0.01); it was not correlated with fat thickness of the anterior thigh (r=-0.30, p=0.927) or corrected EI (r=0.15, p=0.65).

No significant correlations were found between age and the GMFM-66-IS (r=0.16, p=0.38), the muscle thickness of the quadriceps (r=0.24, p=0.18), the fat thickness of the anterior thigh (r=0.30, p=0.09), or the corrected EI (r=0.21, p=0.24).

Fisher's exact probability test showed that there was no significant gender difference in the ratio of the presence or absence of the opportunity to stand in daily activities (φ =0.50, p=1.00); however, there was a significant difference in the ratio for each GMFCS level. In comparison to adults with GMFCS V, a higher proportion of adults with GMFCS IV were classified into the ST group (φ =-0.68, p<0.01).

DISCUSSION

The positive correlation found between GMFM-66-IS and the muscle thickness of the quadriceps was similar to the findings of a previous study⁵). It has been reported that the muscle thickness of the quadriceps reflects the gross motor function, self-care, and mobility in daily life^{5, 16}). The relationship between the muscle thickness of the quadriceps and the gross motor function in children with CP has been widely reported^{5, 17}), but no studies have been limited to adults with severe CP (GMFCS IV and V). It was shown that the muscle thickness of the quadriceps was correlated with the gross motor function even in adults with severe CP who were unable to walk. Choe et al.¹⁷) reported a correlation coefficient of 0.59 between the

 Table 2. Effects of gender on GMFM-66-IS, muscle thickness of the quadriceps, fat thickness of the anterior thigh, and echo intensity of the rectus femoris

(CD)	Tatal	Tratal ST array 10	group, n=10 NST group, n=23 p valu		Mean differ-	95%CI	
Average (SD)	Total	S1 group, n–10		p value	ence	Lower	Upper
GMFM-66-IS	27.62 (10.07)	37.78 (6.19)	23.20 (8.03)	<0.01*	14.58	8.74	20.41
MTQ, cm	2.20 (0.73)	2.86 (0.73)	1.93 (0.54)	< 0.01*	0.92	0.46	1.39
FTA, cm	0.95 (0.61)	1.26 (0.52)	0.81 (0.61)	0.05	0.45	-0.08	0.9
Corrected EI	124.02 (21.64)	137.39 (16.46)	118.21 (21.30)	0.02*	19.18	3.71	34.65

*Significant difference between groups. SD: standard deviation; GMFM: gross motor function measure; MTQ: muscle thickness of the quadriceps; FTA: fat thickness of the anterior thigh; EI: echo intensity of the rectus femoris; CI: confidence interval.

 Table 3. Effects of standing on GMFM-66-IS, muscle thickness of quadriceps, fat thickness of the anterior thigh, and echo intensity of the rectus femoris

Average (SD)	Female, n=12	Male, n=21	p value	Mean differ-	95%CI	
				ence	Lower	Upper
GMFM-66-IS	27.80 (10.85)	27.52 (9.88)	0.94	0.28	-7.27	7.83
MTQ, cm	2.16 (0.83)	2.24 (0.69)	0.79	-0.72	-0.62	0.48
FTA, cm	1.47 (0.54)	0.65 (0.44)	< 0.01*	0.82	0.47	1.17
Corrected EI	137.13 (15.75)	116.53 (21.23)	< 0.01*	20.6	6.23	34.96

*Significant difference between groups. SD: standard deviation; GMFM: gross motor function measure; MTQ: muscle thickness of the quadriceps; FTA: fat thickness of the anterior thigh; EI: echo intensity of the rectus femoris; CI: confidence interval.

rectus femoris muscle thickness and the GMFM-88 and Ohata et al.⁵⁾ reported a correlation coefficient of 0.52 between the quadriceps muscle thickness and the GMFM-66. The high correlation of 0.72 between the quadriceps muscle thickness and the GMFM-66-IS in the present study may be because the inclusion criteria were strictly limited to adults with severe CP who did not have a gait function. The previous study included individuals with GMFCS I–V and a wide range of participants, and it was likely that there were other factors that affected the muscle thickness.

In the present study, there was no significant correlation between the quadriceps muscle thickness and GMFM-66-IS in the ST group, but there was a significant positive correlation between the two factors in the NST group. This may be because none of the participants in the ST group had a maximum knee joint extension angle of more than -10° , since the knee joint was semi-flexed in the standing position. Thus, flexion was always generated against the knee joint, and—as a consequence—quadriceps muscle activity was required to maintain the standing position. In the standing load might increase the quadriceps muscle thickness. The scoring system of the standing item of the GMFM, which requires an independent standing ability to score, however, the participants in the ST group depended on assistance to stand. Thus, in the ST group, it was possible that quadriceps muscle thickness was not necessarily correlated with the GMFM-66-IS scores. On the other hand, in the NST group, the significant correlation between these factors may have been due to the differentiation in the quadriceps muscle thickness in participants in the NST group, due to gross motor function differentiation other than standing, such as rolling over and crawling.

There was a significant positive correlation between GMFM-66-IS and the corrected EI in all participants as a group, which was contrary to the hypothesis. In other words, in adults with severe CP (GMFCS IV and V), those with a higher gross motor function showed a greater amount of intramuscular noncontractile tissue. Pitcher et al.⁹⁾ suggested that the echo intensity of the medial gastrocnemius muscle in children with CP was highest in GMFCS III and decreased with increasing severity in GMFCS IV and V. The result that participants with severe CP who had lower motor function had lower corrected EI values may have partially supported the report of Pitcher et al⁹. In other hand, the correlation found between the corrected EI and the GMFM-66-IS may be a spurious correlation. In this case, it suggested that the ST group with higher motor function may simply have had a higher EI value. This hypothesis was also supported by the fact that there was no significant correlation between the GMFM-66-IS and the corrected EI in the two groups when the ST and NST groups were examined separately. The results of this study do not allow us to discuss this mechanism in detail. However, there is no doubt that a high load is placed on the rectus femoris muscle due to inadequate extension of the knee joint during standing¹⁸⁾. Hence, it is possible that the overload damaged the muscle fibers, that the damage was not adequately repaired, and that the damaged muscle fibers were replaced by fat or connective tissue. In fact, muscle satellite cells, which play an important role in the process of muscle tissue repair¹⁹), have been shown to be reduced in children with CP²⁰). The effect of standing on the qualitative state of the muscle has been largely unexplored and requires further study. The results of this study suggest that the standing position may well affect the qualitative status of the rectus femoris muscle; thus, it may be necessary to evaluate qualitative data (echo intensity) in addition to quantitative data (muscle thickness) when performing ultrasound imaging assessments of the muscles of individuals with CP.

Maintaining the standing position has been reported to have various benefits in children and adults with CP, including increased bone mineral density of the spine²¹, decreased modified Ashworth scale, improved passive range of motion with dynamic standing posture maintenance using a posture-holding device called the Innowalk²², increased hamstring muscle length, and reduced caregiver assistance²³. In addition to these benefits reported in previous studies, the possibility of muscle qualitative changes caused by standing as suggested in the present study must be taken into account when making decisions on whether or not to provide standing intervention for individuals with severe CP. However, there is a paucity of studies on muscle qualitative changes, and it is not possible to draw any definitive conclusions in this study as to whether the qualitative changes in muscle are negatives or positives, and future research is expected to clarify the meaning of these changes.

In general, the concentration of noncontractile tissue in the muscle tends to increase with age⁶⁾. However, the results of the present study did not show a significant correlation between age and EI. It has been reported that there was no significant correlation between age and the EI levels in children with CP⁹⁾, and the results of the present study in adults with CP were similar to those in children. For individuals with CP, the EI levels seem to be unrelated to age; however, certain daily activities, such as the opportunity to stand with caregivers in daily activities, can influence the muscle EI, especially in adults with severe CP.

The present study showed that among adults with severe cerebral palsy, the subcutaneous fat thickness of the anterior thigh and corrected EI were significantly greater in females than in males. Thus, the effect of gender on EI in adults with CP was clear and similar to the differences found in healthy participants²⁴). The gender ratio did not differ between the ST and NST groups according to the results to Fisher's exact probability test. Thus, the difference in EI between the ST and NST groups was not influenced by gender.

In this study, the participants were divided into two groups, the ST and NST groups; however, the sample size after the division was small and the statistical power was low. Secondly, since the participants in this study were limited to those with severe CP (GMFCS IV and V), the relationship between motor function and muscle thickness and the echo intensity in individuals with less severe CP (GMFCS I–III). In addition, in this study, conclusions were limited to the rectus femoris and vastus medialis muscles, in order to reduce the time required to complete the assessments. Other muscles should be investigated in the future.

This study suggested the following: 1) in adults with severe CP, those with a greater quadriceps muscle thickness had a higher gross motor function. 2) In adults with severe CP, higher GMFM-66-IS scores were associated with a higher echo intensity of the rectus femoris; however, these relationships may have been influenced by the the opportunity to stand with caregivers in daily activities. Although there are still few studies on the muscle echo intensity in adults with CP, it is desirable to perform both quantitative and qualitative evaluations of the muscles using ultrasound imaging.

Conflict of interest

The authors declare no conflicts of interest in association with the present study.

REFERENCES

- Rosenbaum P, Paneth N, Leviton A, et al.: A report: the definition and classification of cerebral palsy April 2006. Dev Med Child Neurol Suppl, 2007, 109: 8–14. [Medline]
- Ross SA, Engsberg JR: Relationships between spasticity, strength, gait, and the GMFM-66 in persons with spastic diplegia cerebral palsy. Arch Phys Med Rehabil, 2007, 88: 1114–1120. [Medline] [CrossRef]
- Barrett RS, Lichtwark GA: Gross muscle morphology and structure in spastic cerebral palsy: a systematic review. Dev Med Child Neurol, 2010, 52: 794–804. [Medline] [CrossRef]
- Moreau NG, Simpson KN, Teefey SA, et al.: Muscle architecture predicts maximum strength and is related to activity levels in cerebral palsy. Phys Ther, 2010, 90: 1619–1630. [Medline] [CrossRef]
- 5) Ohata K, Tsuboyama T, Haruta T, et al.: Relation between muscle thickness, spasticity, and activity limitations in children and adolescents with cerebral palsy. Dev Med Child Neurol, 2008, 50: 152–156. [Medline] [CrossRef]
- 6) Fukumoto Y, Ikezoe T, Yamada Y, et al.: Skeletal muscle quality assessed from echo intensity is associated with muscle strength of middle-aged and elderly persons. Eur J Appl Physiol, 2012, 112: 1519–1525. [Medline] [CrossRef]
- 7) Young HJ, Jenkins NT, Zhao Q, et al.: Measurement of intramuscular fat by muscle echo intensity. Muscle Nerve, 2015, 52: 963–971. [Medline] [CrossRef]
- Pillen S, Tak RO, Zwarts MJ, et al.: Skeletal muscle ultrasound: correlation between fibrous tissue and echo intensity. Ultrasound Med Biol, 2009, 35: 443–446.
 [Medline] [CrossRef]
- Pitcher CA, Elliott CM, Panizzolo FA, et al.: Ultrasound characterization of medial gastrocnemius tissue composition in children with spastic cerebral palsy. Muscle Nerve, 2015, 52: 397–403. [Medline] [CrossRef]
- Battisti N, Milletti D, Miceli M, et al.: Usefulness of a qualitative ultrasound evaluation of the gastrocnemius-soleus complex with the Heckmatt scale for clinical practice in cerebral palsy. Ultrasound Med Biol, 2018, 44: 2548–2555. [Medline] [CrossRef]
- Gough M, Shortland AP: Could muscle deformity in children with spastic cerebral palsy be related to an impairment of muscle growth and altered adaptation? Dev Med Child Neurol, 2012, 54: 495–499. [Medline] [CrossRef]
- 12) Palisano R, Rosembaum P, Bartlett D, et al.: Gross motor function classification system expanded & revised (GMFCS-E&R). CanChild 2007; https://www.canchild.ca/en/resources/42-gross-motor-function-classification-system-expanded-revised-gmfcs-e-r.
- Russell DJ, Rosenbaum PL, Cadman DT, et al.: The gross motor function measure: a means to evaluate the effects of physical therapy. Dev Med Child Neurol, 1989, 31: 341–352. [Medline] [CrossRef]
- Russell DJ, Avery LM, Rosenbaum PL, et al.: Improved scaling of the gross motor function measure for children with cerebral palsy: evidence of reliability and validity. Phys Ther, 2000, 80: 873–885. [Medline] [CrossRef]
- Russell DJ, Avery LM, Walter SD, et al.: Development and validation of item sets to improve efficiency of administration of the 66-item Gross Motor Function Measure in children with cerebral palsy. Dev Med Child Neurol, 2010, 52: e48–e54. [Medline] [CrossRef]
- 16) Ohata K, Tsuboyama T, Ichihashi N, et al.: Measurement of muscle thickness as quantitative muscle evaluation for adults with severe cerebral palsy. Phys Ther, 2006, 86: 1231–1239. [Medline] [CrossRef]
- Choe YR, Kim JS, Kim KH, et al.: Relationship between functional level and muscle thickness in young children with cerebral palsy. Ann Rehabil Med, 2018, 42: 286–295. [Medline] [CrossRef]
- Hsu AT, Perry J, Gronley JK, et al.: Quadriceps force and myoelectric activity during flexed knee stance. Clin Orthop Relat Res, 1993, (288): 254–262. [Medline]
- 19) Seale P, Rudnicki MA: A new look at the origin, function, and "stem-cell" status of muscle satellite cells. Dev Biol, 2000, 218: 115–124. [Medline] [CrossRef]
- 20) Smith LR, Chambers HG, Lieber RL: Reduced satellite cell population may lead to contractures in children with cerebral palsy. Dev Med Child Neurol, 2013, 55: 264–270. [Medline] [CrossRef]
- Caulton JM, Ward KA, Alsop CW, et al.: A randomised controlled trial of standing programme on bone mineral density in non-ambulant children with cerebral palsy. Arch Dis Child, 2004, 89: 131–135. [Medline] [CrossRef]
- 22) Tornberg ÅB, Lauruschkus K: Non-ambulatory children with cerebral palsy: effects of four months of static and dynamic standing exercise on passive range of motion and spasticity in the hip. PeerJ, 2020, 8: e8561. [Medline] [CrossRef]
- 23) Gibson SK, Sprod JA, Maher CA: The use of standing frames for contracture management for nonmobile children with cerebral palsy. Int J Rehabil Res, 2009, 32: 316–323. [Medline] [CrossRef]
- 24) Caresio C, Molinari F, Emanuel G, et al.: Muscle echo intensity: reliability and conditioning factors. Clin Physiol Funct Imaging, 2015, 35: 393-403 [Cross-Ref]. [Medline]