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Original Article

Association of age in motor function outcomes after multilevel myofascial release in children with cerebral palsy

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ABSTRACT

Background: Many recommend deferring orthopedic surgery for cerebral palsy-related disorders in young children. However, age is correlated with musculoskeletal deterioration, and deferral may affect surgical outcomes. We aimed to clarify the relationships among age, degree of musculoskeletal disorder, and postoperative motor function change in children with cerebral palsy.

Methods: We prospectively evaluated children with cerebral palsy and a knee flexion gait disorder who underwent multilevel myofascial release between June 2010 and July 2014. The children were divided into younger (<10 years of age) and older (>10 years of age) groups. Outcome measures included the Gross Motor Function Measure (GMFM), range of motion, spasticity, and physical capacity. Preoperative factors and postoperative changes were compared between the groups using the chi-squared, independent *t*-, and Mann–Whitney tests. Significant factors were plotted by participant age to identify the relationships between age and other variables.

Results: We analyzed 20 patients who underwent multilevel myofascial release (12 and 8 in the younger and older groups, respectively). Whereas most preoperative factors were comparable between the two groups, the older group had a higher range of motion limitation score (44.4 vs. 36.1, $p < 0.05$). The older group also showed less improvement in the GMFM (−0.3 vs. +3.0, $p < 0.05$) and physical capacity (+0 vs. +1, $p < 0.05$) scores after 6 months of postoperative rehabilitation.

Conclusions: Age was positively correlated with the range of motion limitation and negatively correlated with postoperative GMFM improvement. The less favored postoperative rehabilitation course in older children needs to be considered for parents whose children are amenable to surgeries.

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At a glance of commentary

Scientific background on the subject

Orthopedic surgery for gait disorder in cerebral palsy is believed to be safer and better outcomes when performed in older children. However soft tissue contracture and gait disorders often deteriorate with skeletal growth. Optimal age of orthopedic surgery for gait disorders is still controversial.

What this study adds to the field

Age was positively correlated with the range of motion limitation and negatively correlated with postoperative improvement in gross motor function. Less and slower postoperative motor function improvement was noted in children older than 10 years old. Results offer another opinion for parents whose children are amenable to surgeries.

Cerebral palsy (CP) is a non-progressive neurological disease that leads to various musculoskeletal system disorders. The hypertonic muscles cannot gain adequate length during childhood skeletal growth, resulting in a progressively decreased range of motion (ROM) and diminished motor function [1]. In a longitudinal survey, Bell et al. reported that walking function deteriorates after the ROM decreases in ambulatory children with CP [2]. In adolescents with CP, significant correlations have been reported between soft tissue contracture and declining motor capacity [3]. Orthopedic surgery is often indicated when conservative treatments can no longer improve muscle and joint contractures [4–12].

In neurological diseases that cause progressive musculoskeletal system changes, age correlates with the deterioration of musculoskeletal function [2,5] and could be a factor affecting treatment outcomes [13]. Previous studies have reported that joint deformity and muscle contracture recurrence were more common when surgery was performed in children younger than 8 years old [14,15]. Švehlík et al. used logistic regression to retrospectively analyze the gait deviation index in a 10-year longitudinal study of children with CP. They concluded that increased age at the time of surgery was the most important predictor of a satisfactory long-term result [16]. Additionally, they reported that the greatest improvement in gait function was found in 10–12-year-old children with a Gross Motor Function Classification System (GMFCS) level III [17]. These findings suggest that among children with CP, orthopedic surgery is safer and has better outcomes when performed in older children. However, patients who undergo surgery at an older age might have minor underlying neurological disorders and slower disease progression. This potential confounding factor should be controlled to address the controversy about whether surgery should be performed when indicated or deferred until the patient is older. Therefore, we compared surgical outcomes in age-based groups of children with CP with similar neurological involvement, gross motor function, and surgical indications (knee flexion gait disorder). We tested the null hypothesis that older children with CP have the same postoperative motor outcomes as younger children after multilevel myofascial release.

Methods

Study design

We performed a prospective comparative study in our tertiary medical center between June 2010 and July 2014. This study was approved by the Institutional Review Board of the authors' affiliated institutions. We obtained written informed consent for each child's participation from their parent or legal guardian.

Participants

Children with spastic diplegic or quadriplegic CP who were scheduled to undergo bilateral multilevel soft tissue release for a knee flexion gait disorder were included in the study. Children who underwent osteotomy were excluded because the postoperative course of recovery after that procedure is

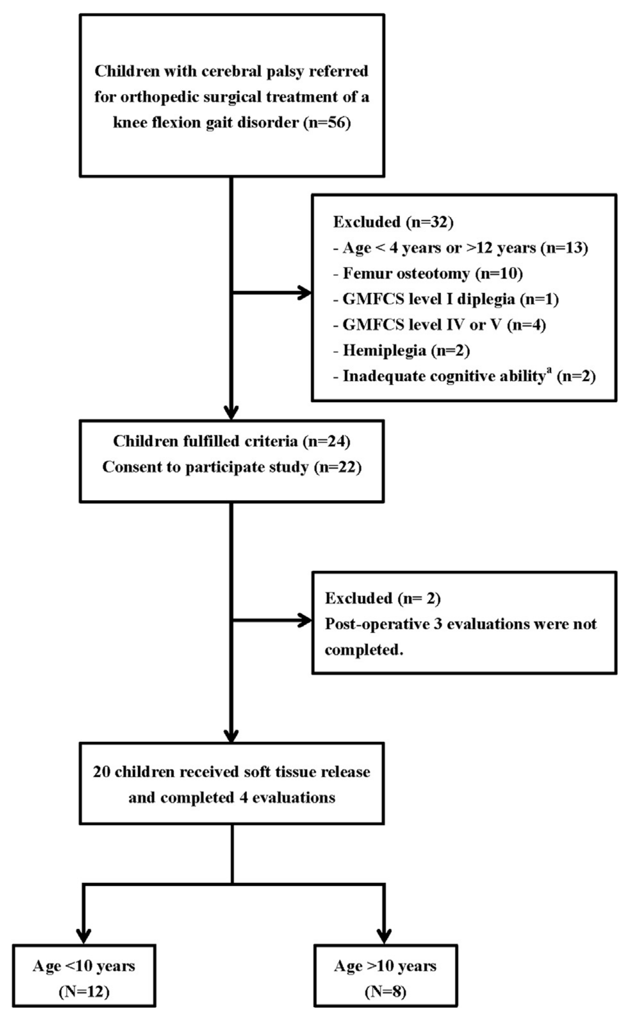

Fig. 1 Patient enrollment flowchart.

different from that following soft tissue release. Children with a GMFCS level of I, IV, or V and hemiplegia were excluded to prevent from ceiling and floor effects in the evaluation of motor function changes. Other exclusion criteria are listed in Fig. 1. We defined knee flexion gait as a gait with a knee flexion angle of 20° or higher throughout the stance phase or 30° or higher at the terminal swing [18,19].

Children were divided in a younger (<10 years of age) and older (≥10 years of age) groups. The age 10-year division was chosen in accordance with the study by Švehlík et al. which reported the greatest improvement in gait function in 10–12-year-old children [17]. We intended to compare the short-term postoperative rehabilitation improvement in children younger and older than 10 years old.

Surgery and postoperative therapy

Single-event multilevel soft tissue releases of both lower extremities were performed in all patients. The muscles that required release were identified by preoperative clinical evaluation of gait characteristics [18] and physical examination. The surgeries varied among participants including procedures performed around the hips (tenotomy of the adductor longus, gracilis, psoas, or combined), knees (myofascial release of the semimembranosus and semitendinosus muscles), and ankles (myofascial release of the gastrocnemius, tibialis posterior, or both).

Postoperatively, long leg splints were applied for 2 weeks to maintain the knee flexion contracture correction and facilitate standing training. Non-articulated or ground reaction force ankle-foot orthoses were applied for gait training. Physical therapy was conducted by two certified pediatric physical therapists. In the first 2 weeks, therapy included standing, balance training, and strengthening of the back and hip muscles. Thereafter, physical therapy focused on strengthening the knee and hip muscles and gait training using a walker. The children returned every 2 weeks for a checkup by research physical therapists for the first 6 weeks and then every 6 weeks until 6 months after surgery.

Each patient underwent four assessments as follows: the week before surgery and postoperatively at 6 weeks, 3 months, and 6 months to evaluate the changes following surgery and after rehabilitation. The outcome measures included gross motor function, ROM limitation, spasticity, selective motor control, and physical capacity. These evaluations were performed by the same physical therapists who had provided postoperative physical therapy.

Outcome measures

The Gross Motor Function Measure (GMFM) is a standardized instrument used to quantify changes in gross motor ability in children with CP [20,21]. We used the GMFM-66 that includes 66 items in 5 domains: lying & rolling, sitting, crawling & kneeling, standing, and walking, running & jumping skills. The GMFM-66 was modified from previous version of 88 items to improve the interpretability of changes following interventions [21]. The maximal range of scores for children with GMFCS level II at age of 5–12 years was 44.8–92.2 and that for GMFCS level III was 36.1–65.4 [22]. Higher scores indicated better gross motor function.

We measured ROM limitation using the Spinal Alignment and Range of Motion Measure [23]. The bilateral hip (12 items), knee (4 items), and ankle (4 items) scores were summed (0–80) to represent the overall lower extremity ROM limitation; higher scores indicated more severe ROM limitations.

Spasticity was measured using the 5-point Modified Ashworth Scale [24] which is scored as follows: 1, normal tone; 2, mild spasticity with catching in limb movement or minimal resistance throughout the remainder (<50%) of the ROM; 3, moderate spasticity with increased tone throughout most of the ROM; 4, severe spasticity with difficulty in passive motion; and 5, extreme spasticity with rigidity in flexion and extension. The scores for bilateral hamstring and gastrocnemius muscles were summed to indicate the global spasticity in key muscles responsible for knee flexion gait. The spasticity score ranged from 020, and the scores increased with severity.

Physical capacity was evaluated on the basis of the number of times the participant could stand up from a squatting position in 30 s [25]. Light support was allowed for participants who might lose their balance while standing up. Physical capacity was graded as follows: 0, could not stand up even with support; 1, stood up once or twice with support; 2, stood up 3–8 times with support; 3, stood up > 8 times with support; and 4, stood up > 8 times without support; higher scores indicated better physical capacity.

We performed measurements of test-retest reliability at 2-week intervals and assessed the interrater reliability for each of the four tests used to evaluate surgical outcomes.

Statistical analysis

We compared preoperative baseline measurements and postoperative changes between the two age groups. Baseline measurements included body mass index (BMI); GMFCS level; and GMFM-66, ROM limitation, spasticity, and physical capacity scores. Surgical outcomes were measured by the changes in GMFM-66, ROM limitation, spasticity, and physical capacity scores. Chi-squared, independent *t*-, and Mann–Whitney tests were used for categorical (GMFCS level), continuous (BMI, GMFM-66, ROM limitation), and ordinal (spasticity and physical capacity scales) variables, respectively. Following the comparison between age groups, significant factors were plotted by participant age to identify the relationships between age and variables. Correlation between postoperative GMFM change and preoperative baseline measurements was analyzed using Pearson' correlation. The pre-operative factors that were significantly correlated to GMFM change were entered multiple regression test. We used SPSS Statistics for Windows, Version 20.0 (IBM Corp., Armonk, NY, US) for all analyses, and the level of significance was set at a p-value <0.05.

Results

Participants

Fig. 1 shows the study flow diagram. Of 22 consenting participants, two in the older group failed to attend all scheduled

Table 1 Baseline data in 20 children with cerebral palsy.

No	Age	Sex	BMI	GMFCS	GMFM	ROM	Spasticity	Capacity	Surgery
1	5.1	M	13.6	III	45.3	34	12	0	HKA
2	5.9	M	18.3	III	53.9	39	18	1	HKA
3	6.2	M	17.7	III	55.6	35	17	2	HK
4	6.9	M	18.2	III	50.6	42	18	0	HKA
5	7.7	M	21.2	II	74.2	27	15	4	HKA
6	8.1	F	13.6	III	54.6	44	16	3	KA
7	8.5	M	13.9	III	53.1	46	18	0	HKA
8	8.9	M	15.9	II	60.1	39	12	3	HKA
9	9.0	M	17.7	III	52.1	35	20	4	HKA
10	9.2	M	19.7	II	65	26	16	3	HK
11	9.3	M	14	II	76.8	32	19	3	KA
12	9.7	M	15.9	II	63.6	34	21	3	HKA
13	10.3	M	19.8	III	53.1	45	20	1	HKA
14	10.6	F	15.4	III	44.2	57	19	0	HKA
15	10.6	F	15.3	III	52.9	53	21	4	HKA
16	10.8	F	17	III	49.9	33	18	2	HK
17	11.6	M	16.9	II	65	32	17	3	HKA
18	11.8	M	14.9	II	75.3	36	23	3	HKA
19	11.8	M	14.8	II	68.9	51	12	4	HK
20	12.1	F	15.2	III	48.7	54	21	2	HKA

Abbreviations: A: surgeries around the ankles; BMI: body mass index; F: female; GMFCS: gross motor function classification system level; GMFM: gross motor function measure score; H: surgeries around the hips; K: surgeries around the knees; M: male; ROM: lower extremity range of motion limitation score.

clinic appointments and were excluded. Therefore, we analyzed 20 patients (mean age, 9.2 years; age range, 5.1–12.1 years). In these patients, the mean GMFM-66 score was 58.1 (range, 44.2–76.8), and all had spastic gait with excessive knee flexion. Observational gait pattern analysis showed that 16 and 4 children had equinus and crouch gait, respectively. Children with crouch gait received multilevel soft tissue release, sparing the ankle (cases 3, 10, 16, and 19; Table 1).

After grouping the children by age, there were 12 and 8 in the younger and older groups, respectively. Preoperative BMIs; GMFCS levels; and GMFM, spasticity, and physical capacity scores were comparable between the two groups. Children in the older group had higher ROM limitation scores than did those in the younger group (44.4 vs. 36.1, respectively; *t*-test, $p = 0.028$, Table 2). The scatter plot of data from all subjects revealed a positive correlation between age and preoperative ROM limitation scores [Fig. 2], indicating that age >10 years was associated with more severe soft tissue contractures.

Postoperatively, the young patients experienced a decrease of GMFM scores at 6 weeks, recovery to pre-operative condition at 3 months, and improvement at 6 months. A common condition in the older patients was decreasing GMFM scores at post-operative 6 weeks and 3 months. Some recovered to pre-operative condition after 6 months, and the others did not [Table 3].

Compared with the younger group, the older group showed less improvement in GMFM scores (change from preoperative

Table 2 Preoperative measurements in children with cerebral palsy grouped by age.

	<10 years old (n = 12)	>10 years old (n = 8)	*p*-value
Age	7.9 (1.5)	11.2 (0.7)	
GMFCS (level II:III) [c]	5:7	3:5	0.852
BMI[a]	16.6 (2.6)	16.2 (1.7)	0.648
GMFM66 scores[a]	58.7 (9.5)	57.2 (11.1)	0.750
ROM limitation scores[a]	36.1 (6.2)	44.4 (9.3)	0.028*
Spasticity scores[b]	17.5 (15.25–18.75)	19.5 (17.25–21)	0.111
Capacity scores[b]	3 (0.25–3)	2.5 (1.25–3.75)	0.811

Continuous variables are reported as means (standard deviation), and ordinal variables, as medians (interquartile ranges).
*$p < 0.05$.
[a] Independent *t*-test.
[b] Mann-Whitney test.
[c] Chi-squared test.

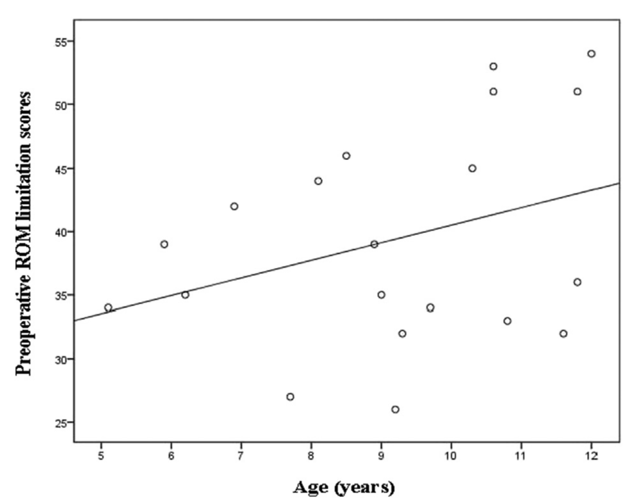

Fig. 2 Scatter plot of age-related preoperative range of motion limitation (ROM) scores in children with cerebral palsy. A positive correlation suggests that the range of motion decreases with age in growing children.

Table 3 Change at post-operative 6 months in the 20 children.

No	GMFM	ROM	Spasticity	Capacity
1	2.6	−3	0	2
2	9.7	−6	−2	2
3	0.5	−6	0	1
4	1.3	−12	−6	3
5	7.7	−7	−4	0
6	1.6	−13	−4	0
7	0.8	−11	−8	3
8	−0.8	−2	2	0
9	2.8	−14	−4	0
10	2.4	7	5	0
11	3.2	13	−2	1
12	−0.9	−7	−9	1
13	0.8	−18	−4	1
14	−3.8	−10	−3	0
15	2.3	−19	−5	0
16	−1.2	−7	−6	−1
17	3.1	−16	−7	0
18	−2.7	2	−9	0
19	1.1	−7	−1	0
20	−2.4	9	−5	0

Abbreviations: GMFM: Gross motor function measure score; ROM: lower extremity range of motion limitation score.

measurement, −0.3 vs. +2.6 in the older vs. the younger group; *t*-test, $p = 0.045$). After 6 months of rehabilitation, the older group showed lesser improvement in physical capacity than the younger group (change from preoperative measurement, 0 vs. +1 in the older vs. younger group; Mann–Whitney test, $p = 0.024$; Table 4). The scatter plot of data from all subjects revealed an inverse relationship between age and post-operative improvement in GMFM scores [Fig. 3].

The Pearson's correlation showed the postoperative GMFM change was significantly correlated to age ($r = -0.535$, $p < 0.001$), BMI ($r = 0.430$, $p < 0.05$), and pre-Op ROM limitation scores ($r = -0.386$, $p < 0.05$). The multiple regression showed age was the only significant factor for the post-operative GMFM change (GMFM change $= -0.45 \times$ age $+ 1.86$, $p = 0.042$).

Test-retest and interrater reliabilities

The GMFM test-retest and interrater reliabilities were excellent (intraclass correlation coefficients [ICCs], 0.997 and 0.998, respectively). The Spinal Alignment and Range of Motion Measure test-retest and interrater reliabilities were good (ICC, 0.95–0.97 and 0.89, respectively). The Modified Ashworth Scale test-retest and interrater reliabilities (ICC, 0.73–0.79 and 0.86, respectively) were moderate to good. The physical capacity evaluation test-retest (ICC, 1.0) and interrater reliabilities (ICC, 1.0) were good.

Table 4 Changes 6 months after bilateral multilevel lower extremity soft tissue releases.

	<10 years old (n = 12)	>10 years old (n = 8)	*p*-value
GMFM66 scores[a]	2.6 (3.2)	−0.3 (2.5)	0.045*
ROM limitation scores[a]	−5.1 (8.1)	−7.5 (9.9)	0.557
Spasticity scale[b]	−3 (−5.5–0.25)	−5 (−6.75–3.25)	0.162
Capacity scale[b]	1 (0–2)	0 (0–0)	0.024*

Continuous variables are reported as means (standard deviation), and ordinal variables, as medians (interquartile ranges).
*$p < 0.05$.
[a] Independent *t*-test.
[b] Mann-Whitney test.

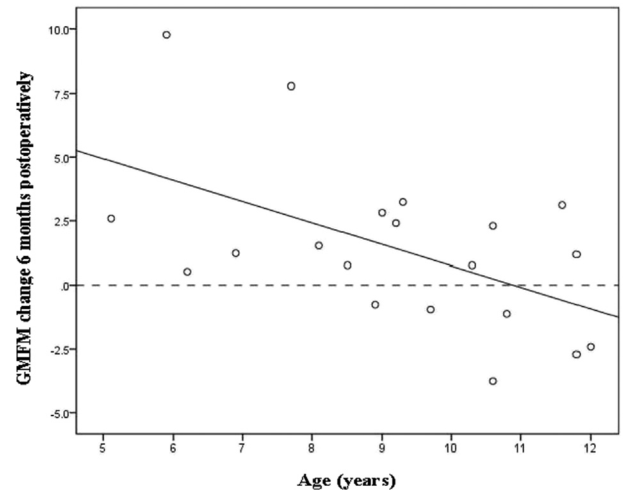

Fig. 3 Scatter plot of age-related gross motor function measure (GMFM) changes in children with cerebral palsy 6 months postoperatively. A negative correlation suggests that the benefits of surgery decrease with age.

Discussion

We compared the postoperative results after multilevel lower extremity soft tissue releases in age-based groups of children with CP and found that both age and muscle contracture affect surgical outcomes in this population. The short-term outcome was not comparable to the long-term outcomes in the study by Švehlík [16,17]. However, the less favored rehabilitation course after surgery in older children needs to be considered when discussing surgical interventions with parents.

The relationships among age, motor function, and ROM limitation are important in children with CP. Previous studies have revealed that GMFM scores increase with age through the first few years of life [22,26]. In adolescents, gross motor function may decline, and ROM limitation is the most significant factor leading to this decline [3]. In our analysis, we found an inverse relationship between age and ROM. Older age and substantial ROM limitation were associated with worse surgical outcomes. The relationships among increasing age, diminishing ROM, declining motor function, and less optimal surgical outcomes should be considered when a child is amenable to surgical intervention.

This study had different results from those of the studies by Švehlík et al. [16,17]. Švehlík et al. retrospectively analyzed 10-year longitudinal data of the gait deviation index and concluded that the greatest improvement in gait function was found in 10–12-year-old children. This study showed that children older than 10 years old had less postoperative improvement in GMFM and physical capacity at 6 months postoperatively than did children who had surgery when younger than 10 years old. The different timing of assessment, 6 months postoperatively vs. 10 years postoperatively, and the different outcome measures, motor function/capacity vs. gait deviation index, might be responsible for the different conclusions in this study and the study by Švehlík et al.

Younger children have greater motor development potential than older children, and this could be an underlying factor for the better postoperative outcomes we found in the younger group. Performance of motor function could be suppressed by muscle contracture and spasticity in the preoperative assessment. Motor development potential had the chance to express properly after removing the suppression. Data from this study suggested that young children can reach their expected motor potential after surgical treatment, while in older children, they only plateaued at their existed preoperative motor function. Evidence from a randomized controlled trial indicated that compared with physical therapy alone, surgery provided greater improvement in motor function in children with CP-related disorders who were amenable to surgery [12].

Another factor that could have affected postoperative improvement in our patients was compliance with physical therapy. We found that younger patients participated in postoperative physical therapy more regularly than older children did. The children in the older age group spent more time participating in school activities, and two older group patients did not complete the study. The improvement in the GMFM (activity/participation) shown in this study was related to spastic diplegic CP (health condition), ROM limitation (body structure), age (personal factor), and school life (environmental factor), in accordance with the model of the International Classification of Functioning, Disability, and Health.

This study had several limitations. First, our postoperative follow-up was for only 6 months. With a longer follow-up, physical capacity and GMFM in the older group may have improved. Nonetheless, the most intensive rehabilitation was administered in the first 6 months, and most of the postoperative improvement occurred during this period. Second, the number of participants in each group was small after applying our exclusion factors. The responses to myofascial release were variable among children with cerebral palsy. Variable responses in a small number of study subjects can lead to an unstable statistical result. The association between GMFM change and age became insignificant when the two cases (Nos. 2 and 5) with a greater improvement in the younger age group were regarded as outliers. Therefore, greater sample size is needed. Third, the Ashworth Scale for spasticity classifies ordinal variables. This study summed the bilateral knee and ankle scores to characterize the condition of the entire lower extremity and effects of multilevel soft tissue releases. Fourth, physical capacity was assessed by squat-to-stand in this study. Walking speed, endurance, and other spatial gait parameters were not performed. Fifth, the surgical indication was a knee flexion gait disorder. However, we did not include a gait analysis to document the postoperative improvement in knee flexion angle. Further studies using the Gillette gait index or gait deviation index to measure the change in gait pathology are desirable [27]. GMFM is a universal and basic measurement of gross motor function and has been used to record the post-operative change in children with different GMFCS levels [9]. In this study we focused on the effects from age on short-term change in GMFM.

In conclusion, our findings disprove the hypothesis that multilevel surgery at an older age has the same result as that at a younger age in children with CP from a viewpoint of postoperative recovery and rehabilitation. Age was positively associated with decreased ROM and negatively correlated with postoperative motor function improvement. This study offers another opinion for parents whose children are amenable to surgeries.

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Conflicts of interest

The authors declare no conflicts of interest.

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