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## Original Article

# Gross motor function change after multilevel soft tissue release in children with cerebral palsy

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## ABSTRACT

**Background:** Improving motor function is a major goal of therapy for children with cerebral palsy (CP). However, changes in motor function after orthopedic surgery for gait disorders are seldom discussed. This study aimed to evaluate the postoperative changes in gross motor function and to investigate the prognostic factors for such changes.

**Methods:** We prospectively studied 25 children with CP (4–12 years) who were gross motor function classification system (GMFCS) level II to IV and underwent bilateral multilevel soft-tissue release for knee flexion gait. Patients were evaluated preoperatively and at 6 weeks and 3 and 6 months postoperatively for Gross Motor Function Measure (GMFM-66), range of motion, spasticity, and selective motor control. The associations between change in GMFM-66 score and possible factors were analyzed.

**Results:** 25 children with gross motor function level II to IV underwent surgery at a mean age of 8.6 years (range, 4–12 years). Mean GMFM-66 score decreased from 55.9 at baseline to 54.3 at 6-weeks postoperatively and increased to 57.5 at 6-months postoperatively ( $p < 0.05$ ). Regression analysis revealed better gross motor function level and greater surgical reduction of spasticity were predictors for decreased GMFM-66 score at 6-weeks postoperatively. Younger age was a predictor for increased GMFM-66 score at 6-months postoperatively.

**Conclusion:** Reduction of contracture and spasticity and improvement of selective motor control were noted after surgery in children with CP. However, a down-and-up course of GMFM-66 score was noted. It is emphasized that deterioration of motor function in children with ambulatory ability and the improvement in young children after orthopedic surgery for gait disorders.

**Level of evidence:** case series, therapeutic study, level 4.

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## At a glance commentary

### Scientific background on the subject

Improving motor function is a fundamental goal in any treatment for children with cerebral palsy. Orthopedic surgeries have been reported to improve gait function, ease of care and to prevent chronic morbidity. However, the surgical effects on gross motor function were variable.

### What this study adds to the field

A down-and-up course of gross motor function was noted after soft tissue release. Children with ambulatory ability had greater risks of deterioration in motor function, and young children had greater potential of improvement in the first post-operative 6 months.

Cerebral palsy (CP) is a complex neurologic disorder caused by nonprogressive encephalopathy occurring in the early years of life [1,2]. This neurologic disorder affects muscle strength, muscle tone, and motor control, leading to a sequence of conditions in the musculoskeletal system [3]. Although the encephalopathy is static, the musculoskeletal problems are progressive and can further affect motor function [4,5]. Bell et al., in their 4.4-year longitudinal survey of ambulatory children with CP, reported deterioration of walking function following decreased range of motion (ROM) in the lower extremities [6]. Preventing the natural course in the musculoskeletal system and deterioration of motor function are the principle goals of therapy for children with CP.

Orthopedic surgery is a common treatment when muscle tightness, joint contracture, and bone deformity have affected motor function [5,7–10]. Orthopedic surgeries have been reported to improve gait function and ease of care and to prevent chronic morbidity [10–16]. However, the surgical effects on gross motor function have been variable. Abel et al. reported that Gross Motor Function Measure (GMFM) score decreased at 3 months postoperatively and increased at 6 months postoperatively after multilevel soft-tissue release [11]. Thomason et al., in their observation of the postoperative course after osteotomy, reported that GMFM score remained at its preoperative status for the first 12 months and improved significantly 24 months after surgery [16]. In another study, Kondo et al. reported that GMFM score decreased in high-function-level children, but there was no change in low-function-level children after surgery [13]. Thus, postoperative changes in motor function have been variable among children with CP with different function levels, musculoskeletal disorders, and surgical procedures. However, the factors affecting the postoperative course remain unknown.

Improving motor function is a fundamental goal of therapy for children with CP. The postoperative changes in GMFM score, either improving or deteriorating, and the factors associated with such changes are important for rehabilitation planning. The present study enrolled children with CP who

underwent multilevel soft-tissue release for knee flexion gait in order to answer the following questions [1]: what are the short-term postoperative changes in GMFM score in relation to gross motor function level? and [2] what are the significant factors that could predict such postoperative changes.

## Methods

### Study design and subjects

This was a prospective case–control study. Children with spastic diplegic or quadriplegic CP who underwent multilevel soft-tissue release for knee flexion gait at the age of 4–12 years were enrolled. Knee flexion gait was defined when the knee flexion angle remained at more than 20° throughout the stance phase or was more than 30° at terminal swing by clinical observation [17]. The knee flexion gait included apparent equinus gait, jump knee gait, and crouch gait [18]. They shared the similar features of knee motion, and ankle angle could be in plantarflexion, neutral, or dorsiflexion in the stance phase. Patients who underwent concomitant osteotomy were excluded because the postoperative course could be greatly affected by this procedure. Children who were Gross Motor Function Classification System (GMFCS) [19,20] level I or V and had hemiplegia also were excluded to avoid ceiling and floor effects when evaluating changes in GMFM score. GMFCS is a classification of motor function in daily life with emphasis on sitting, transfer, and mobility. Briefly, level I: walks without limitations. Level II: walks with limitations. Level III: walks using a hand-held mobility device. Level IV: self-mobility with limitations; may use powered mobility. Level V: transported in a manual wheelchair. The institutional review board for human studies at the authors' hospital approved this study, and the parents of all the participants provided written informed consent.

### Surgery and physical therapy

Myofascial release of bilateral lower extremities was performed in all patients. Hamstrings release was the general procedure, with or without release at the hip or ankle based on clinical judgment of gait disorder and physical examination [15]. Release at the hip included myotomy of the adductor longus, gracilis, and/or psoas. Release at the ankle included myofascial release of the gastrocnemius and/or tendon lengthening of the tibialis posterior. After surgery, long-leg splints were applied for 2 weeks to facilitate standing training. Short-leg casts were applied for 4 weeks in children who underwent ankle surgery. Nonarticulated or ground reaction force ankle-foot orthoses were applied after the short-leg casts.

Two research therapists (YYC and KKY) conducted the postoperative physical therapy. In the first 2 weeks, therapy included standing and balance training, strengthening of the back and hip muscles, and ROM exercise. After the first 2 weeks, physical therapy focused on strengthening of the knee and hip muscles, ROM exercise, and gait training using a walker. Children underwent postoperative physical therapy at development centers or hospitals near their homes. They returned every 2 weeks for a follow-up examination for the

**Table 1** Baseline data of the 25 study patients.

No.	Sex	Age, y	BMI, kg/m <sup>2</sup>	GMFCS level	Involvement	Surgery	GMFM-66 score	ROM score	Spasticity score	Strength score	SMC score
1	M	7.7	21.2	2	D	HKA	74.2	27	15	4	8
2	M	8.9	15.9	2	Q	HKA	60.1	39	12	3	6
3	M	8.9	16.2	2	D	HK	73.6	28	13	3	2
4	M	9.2	19.7	2	D	HK	65.0	26	16	3	5
5	M	9.3	14.0	2	D	KA	76.8	32	19	3	7
6	M	9.7	15.9	2	D	HKA	63.6	34	21	3	4
7	M	11.6	16.9	2	D	HKA	65.0	32	17	3	1
8	M	11.8	14.9	2	D	HKA	75.3	36	23	3	2
9	M	11.8	14.8	2	D	HK	68.9	51	12	4	4
10	M	5.1	13.6	3	D	HKA	45.3	34	12	0	3
11	M	5.9	18.3	3	Q	HKA	53.9	39	18	1	2
12	M	6.2	17.7	3	Q	HK	55.6	35	17	2	2
13	M	6.9	18.2	3	D	HKA	50.6	42	18	0	0
14	F	8.1	13.6	3	Q	KA	54.6	44	16	3	0
15	M	8.5	13.9	3	Q	HKA	53.1	46	18	0	2
16	M	9	17.7	3	D	HKA	52.1	35	20	4	0
17	M	10.3	19.8	3	Q	HKA	53.1	45	20	1	0
18	F	10.6	15.4	3	Q	HKA	44.2	57	19	0	3
19	F	10.6	15.3	3	Q	HKA	52.9	53	21	4	2
20	F	10.8	17.0	3	D	HK	49.9	33	18	2	0
21	F	12.0	15.2	3	D	HKA	48.7	54	21	2	1
22	M	4.0	15.5	4	Q	HKA	28.7	32	18	0	0
23	M	4.6	14.1	4	Q	HK	44.6	34	18	1	4
24	M	6.2	14.0	4	Q	HKA	45.0	36	22	0	0
25	M	7.4	19.1	4	Q	HK	42.0	59	23	0	0

Abbreviations: BMI: body mass index; GMFCS: Gross Motor Function Classification System; GMFM: Gross Motor Function Measure; ROM: range of motion; SMC: selective motor control; M: male; F: female; Q: quadriplegia; D: diplegia; HK: hip and knee; HKA: hip, knee, and ankle; KA: knee and ankle.

first 6 weeks, and then every 6 weeks until 6 months after surgery. After the first 6 months, regular physical therapy including ROM exercise, muscle strengthening, and gait training were conducted in developmental.

### Measurements

Each patient underwent 4 assessments: the week before surgery and at 6 weeks, 3 months, and 6 months after surgery. The assessments included GMFM score, ROM, spasticity, muscle strength, and selective motor control (SMC), which were performed by 2 research therapists who were trained by careful review of written instructions and repeated practice before the study.

GMFM score was the main measure of therapeutic effect in this study. GMFM score is a standard measure of motor function and can quantify changes in gross motor ability in children with CP [21–23]. It consists of 88 items designed to measure activities such as lying, rolling, walking, running, and jumping. Each item is graded on a 4-point scale: 0, unable to initiate the task; 1, initiates the task; 2, partially completes the task; and 3, completes the task. GMFM-66 is the modified scale from the 88 items, which has been proved to show improved interpretability of total score and change scores [23]. Higher GMFM-66 score indicates better gross motor function. In reliability testing, the test–retest performed 2 weeks apart (intraclass coefficient [ICC], 0.997;  $p < 0.001$ ) and interrater reliability (ICC, 0.998;  $p < 0.001$ ) were excellent.

Limitation in ROM was measured using the Spinal Alignment and Range of Motion Measure [24]. Limitation in ROM at each joint was characterized as follows: 0, no limitation; 1, dynamic limitation; 2, mild structural limitation; 3, moderate structural limitation; and 4, severe structural limitation. ROM scores were used in this study because all surgical releases were performed in bilateral lower extremities. ROM score was defined as the sum of the scores in bilateral hips (12 items), knees (4 items), and ankles (4 items). A higher ROM score indicates more severe limitation in ROM. The test–retest (ICC, 0.95–0.97;  $p < 0.001$ ) and inter-rater reliability between 2 testers (ICC, 0.89;  $p < 0.001$ ) were good.

Spasticity was measured using the modified Ashworth scale [25]. This 5-point scale was scored as follows: 1, normal tone; 2, mild spasticity with catching in limb movement or minimal resistance throughout less than 50% of ROM; 3, moderate spasticity with increased tone throughout most of ROM; 4, severe spasticity with difficulty in passive motion; and 5, extreme spasticity with rigidity in flexion and extension. Scores for bilateral quadriceps, hamstrings, and gastrocnemius were summed to indicate the extent and magnitude of spasticity. Total spasticity score ranged from 0 to 20, with higher score indicating more severe spasticity. The test–retest (ICC, 0.73–0.79;  $p < 0.001$ ) and inter-rater reliability (ICC, 0.86;  $p < 0.001$ ) were good to moderate.

SMC was measured by asking a participant to extend his/her knee and dorsiflex the ankle separately without support of the foot while in a seated position [26]. Possible scores

**Table 2** The effects from soft tissue release on study variables.

	Pre-OP (A)	Post-OP 6 weeks (B)	Post-OP 3 months (C)	Post-OP 6 months (D)	Repeat ANOVA <i>p</i> value	Post hoc test
Mean (SD)						
GMFM-66 (0–100)	55.9 (12)	54.3 (10.5)	56.6 (12.3)	57.5 (12.1)	0.005	A vs. B (0.036) A vs. D (0.024) B vs. C (0.012) B vs. D (0.001)
ROM scores (0–80)	39.1 (9.1)	31.6 (7.6)	31.5 (8.2)	32.8 (10.3)	0.001	A vs. B (0.001) A vs. C (0.001) A vs. D (0.001)
Median (IRQ)						
Spasticity (0–30)	18 (16–21)	12 (12–16)	14 (12–17)	14 (12–16)	<0.001	A vs. B (0.001) A vs. C (0.001) A vs. D (0.001) B vs. C (0.022)
Muscle strength (0–4)	2 (0–3)	3 (0–3)	3 (0–4)	3 (2–4)	0.021	A vs. D (0.020) B vs. D (0.013)
Selective motor control (0–8)	2 (0–4)	4 (0–5)	2 (2–6)	3 (0–4)	0.038	A vs. B (0.045) A vs. C (0.003)

Abbreviations: GMFM: Gross Motor Function Measure; SD: standard deviation; IRQ: interquartile range; ROM: range of motion.

included 0 (no SMC, only synergistic movement), 1 (first movement shows SMC, but becomes synergistic during later movement), and 2 (SMC for full ROM). Scores from bilateral knees and ankles produced a total score between 0 and 8, with higher score indicating better SMC. The test–retest (ICC, 0.56–0.92;  $p < 0.001$ ) and inter-rater reliability (ICC, 0.79;  $p < 0.001$ ) were good to moderate.

General muscle strength was evaluated according to the number of times that a participant could stand up from a squatting position in 30 s [27]. Light support by one hand placed on a stable table was allowed for participants who might lose balance while standing up. This test is an evaluation of general muscle strength rather than maximal isometric strength of each muscle [27]. Muscle strength was graded as follows: 0, cannot stand up even with support; 1, stands up 1 or 2 times with support; 2, stands up 3 to 8 times with support; 3, stands up more than 8 times with support; and 4, stands up more than 8 times without support. A higher score indicates better general muscle strength. The test–retest (ICC, 1.0;  $p < 0.001$ ) and inter-rater reliability (ICC, 1.0;  $p < 0.001$ ) were good.

### Data analysis

For our first question, collected data were analyzed using descriptive analysis for the changes in the first 6 months after surgery. Changes between the preoperative and 3 post-operative values were compared, respectively, using the repeat ANOVA test and post hoc test by least significant difference.

For the second question, postoperative change in the GMFM-66 score was used as the dependent variable. Factors that were assumed to be associated with changes in GMFM included baseline conditions and surgical effects. Baseline conditions included age, sex, body mass index, GMFCS level and GMFM-66, ROM, spasticity, muscle strength, and SMC scores. Surgical effects included ankle surgery or not and the changes in the first 6 weeks after surgery, such as changes in ROM, spasticity, muscle strength, and SMC scores. The

conditions at 6 weeks were closely related to surgery, and children were free from wound pain and splinting, which could interfere with the evaluations. Age, body mass index, baseline GMFM-66 score, and ROM score were regarded as continuous variables, whereas GMFCS level and muscle strength, spasticity, and SMC scores were regarded as ordinal variables. The associations between changes in GMFM-66 score and the other factors were tested using the Pearson correlation analysis for continuous variables and the Spearman rank correlation analysis for ordinal variables. The statistical software was IBM SPSS for Windows, version 20.0 and level of significance was set at a  $p$  value of less than 0.05.

### Results

We collected series of data from 25 consecutive CP children who received multilevel soft tissue release for knee flexion gait from May 2010 to July 2012. They were 20 boys and 5 girls. Thirteen children had spastic diplegia, whereas the other 12 had spastic quadriplegia. The GMFCS level was II in 9 children, III in 12, and IV in 4. All the patients underwent multilevel soft-tissue release at a mean age of 8.6 years [Table 1].

The direct surgical results were decreased ROM and spasticity scores. SMC also improved significantly by surgical reduction of contracture and muscle tone. General muscle strength increased after surgery and reached statistical significance at 6 months postoperatively. Mean GMFM-66 score was 55.9 at baseline, which decreased to 54.3 at 6-weeks postoperatively, improved to 56.6 at 3-months postoperatively, and increased to 57.5 at 6-months postoperatively. Compared with the preoperative baseline GMFM-66 score, the decrease at 6 weeks and the increase at 6 months were significant. Therefore, changes in GMFM-66 score at 6-weeks and 6-months after surgery served as the dependent variable in subsequent analyses [Table 2].

The decrease in GMFM-66 score at 6-weeks postoperatively was significantly associated with lower GMFCS level ( $r$ , 0.58),



greater baseline GMFM-66 score ( $r, -0.56$ ), older age ( $r, -0.55$ ), and greater decrease in postoperative muscle strength ( $r, 0.46$ ). Regression analysis revealed that change in GMFM-66 score at 6-weeks postoperatively =  $2.9 \times \text{GMFCS level} + 0.4 \times \text{change in spasticity} - 8.1$  ( $R^2, 0.49$ ;  $p = 0.001$ ). GMFCS level was the strongest factor that could replace other collinear factors, such as age, baseline GMFM-66 score, and change in muscle strength. Because spasticity was decreased by surgery, a positive parameter suggested that greater reduction of spasticity led to greater decrease in GMFM-66 score at 6-weeks postoperative.

At 6-months after surgery, the increase in GMFM-66 score was significantly associated with younger age ( $r, -0.53$ ) and less reduction of postoperative spasticity ( $r, 0.50$ ). Regression analysis revealed that change in GMFM-66 score at 6-months postoperatively =  $-0.75 \times \text{age} + 8.1$  ( $R^2, 0.285$ ;  $p = 0.006$ ). Younger age patients had a greater possibility of increased GMFM-66 score at 6-months after surgery.

When changes in GMFM-66 scores were analyzed separately for GMFCS levels II, III, and IV, a continuous trend of increasing GMFM-66 score was noted in level IV children after surgery. However, in higher function levels II and III children, surgery resulted in decreasing GMFM-66 scores at 6-weeks postoperatively. Then the scores gradually recovered and were above the baseline scores at postoperative 6 months. The post-operative decrease of GMFM-66 score was even significant in level II children [Fig. 1].

## Discussion

Knee flexion gait is a common energy-consumption gait disorder in children with CP [17]. This specific gait disorder was used in the present study as a model to evaluate the short-term changes after multilevel soft-tissue release. The direct surgical effects were improved ROM, spasticity, and SMC. However, GMFM-66 scores decreased at 6 weeks postoperatively and then increased at 6 months postoperatively. This postoperative down-and-up course of motor function was most apparent in older children with GMFCS level II.

GMFCS level and age were predictors for postoperative changes. For knee flexion gait requiring soft-tissue release, children with GMFCS level IV reach surgical criteria at a younger age. In this study, level IV children were significantly younger (mean, 5.6 years) at the time of surgery compared

with level II (mean, 9.9 years) and III (mean, 8.7 years) children. Knee flexion gait in level II children rarely requires surgery at a young age, whereas in level IV children, osteotomy is often required at an older age. These 2 factors were entangled because of the study design, which recruited subjects according to surgical event for the same musculoskeletal disorder. Future studies recruiting more subjects in a single GMFCS level are required to define the influence of age.

Regression analysis revealed that the decrease in GMFM-66 score at 6-weeks after surgery was significantly associated with GMFCS level. Following GMFCS level, change in spasticity had a positive correlation with change in GMFM-66 score. The results indicated that greater surgical reduction of spasticity resulted in greater decrease in GMFM-66 score at 6-weeks postoperatively. This relationship can be explained by muscle weakening accompanied by excessive reduction of muscle tone by soft-tissue release. Spasticity is an over-excitability state of lower motor neuron in response to muscle stretching. Soft tissue release increases the muscle length and achieves greater excursion of muscle stretching before onset of reflex. Besides, ankle surgery and following cast immobilization could result in calf muscle weakness and post-operative deterioration of GMFM scores. However, the study results did not support ankle surgery as a significant factor.

Age was the only predictor for change in GMFM-66 score at 6-months postoperatively. Children who underwent surgery at an age of 4–7 years showed an increase of 3.9 from the baseline GMFM-66 score of 52.7. Children who underwent surgery at an age of 8–12 years showed an increase of 0.4 from the baseline score of 60.2. Between the 2 age groups, baseline GMFM-66 scores were comparable, but younger children showed a greater increase in GMFM-66 score after surgery (3.9 vs. 0.4;  $p = 0.01$ ). For children who had recovered from surgery 6 months later, younger patients showed additional improvement in the natural potential of motor development, which was no longer limited by musculoskeletal disorders. On the other hand, older children had reached a plateau of motor development, and improvement was only because of eliminating musculoskeletal disorders.

Limitations of this study include variations in disease characteristics, which confounded outcome analysis, as well as the small number of cases, which prevented further analysis of the age effect in each GMFCS level. The surgical indication was knee flexion gait, but our study did not include gait analysis to prove the improvement in knee angle during walking. In addition, the postoperative physical therapy was not strictly controlled, and the changes in motor function could have been confounded. This study was to survey the short-term post-operative outcome that was closely related to surgery and rehabilitation. The GMFM change after the first 6 months requires further study. Finally, disease severity moderated the timing of surgical intervention for knee flexion gait. Therefore, GMFCS level and age at surgery were entangled when analyzing the outcome predictors.

This study used a model of knee flexion gait to analyze the surgical outcomes of soft-tissue release and the predictors for postoperative changes in gross motor function. Surgical reduction of contracture and spasticity led to improvement in SMC. However, a deteriorating and then improving course of gross motor function was noted. Deterioration at 6 weeks after

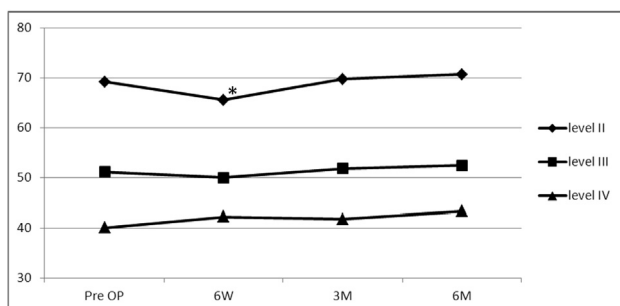


Fig. 1 Postoperative changes in GMFM-66 scores in children with different GMFCS levels.

surgery was noted in children with ambulatory function, whereas greater improvement at 6 months after surgery was noted in younger children. This knowledge is valuable for parents and medical professionals to have a realistic expectation of and proper preparation for orthopedic soft-tissue release in children with CP.

### Conflicts of interest

The authors have no conflicts of interest to declare.

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