

Effects of Soft Tissue Surgery on Pelvic and Hip Rotation in Patients with Spastic Diplegia: A Meta-Analysis

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Background: There are several different opinions regarding the improvements seen on the transverse plane after soft tissue surgery alone in independently ambulant patients with cerebral palsy. We performed a meta-analysis using data from previous studies to identify the effects of soft tissue surgery alone on pelvic and hip rotation in children with spastic diplegia.

Methods: We conducted a pilot study to evaluate the improvement in pelvic and hip rotation after muscle-tendon lengthening surgery in children with spastic diplegia. We also searched EMBASE and PubMed and selected 2 previous studies using the same test conditions with kinematic data on the pelvis and hip joints. A meta-analysis of the results of these 3 studies, including this pilot study, was then performed.

Results: The meta-analysis results showed an external rotation decrease ($p = 0.005$) in the mean difference of pelvic rotation of -3.61 (95% confidence interval [CI], -6.13 to -1.09) and a mean difference in hip rotation of 6.60 (95% CI, 3.34 to 9.86), indicating a significant increase in the hip external rotation after surgery ($p < 0.001$).

Conclusions: In independently community-ambulant pediatric patients with spastic diplegia, pelvic retraction and hip internal rotation could be improved after soft tissue surgery.

Keywords: *Cerebral palsy, Pelvis, Hip, Rotation*

Excessive internal hip rotation is a common problem in cerebral palsy and is a major factor contributing to in-toeing. Pelvic retraction, defined as posterior rotation of the pelvis, is also related to in-toeing.^{1,2)} The cause of pelvic retraction differs depending on the type of cerebral palsy. Pelvic retraction in spastic hemiplegia can be caused by shortening of the triceps surae muscle, but that in spastic diplegia is related to the internal rotation of the hip joint.³⁾

Increased femoral anteversion causes increased hip internal rotation during gait, which also leads to compensatory pelvic retraction.^{4,5)} In addition to skeletal deformities such as femoral anteversion, imbalances of the muscles around the hip joint, such as the gluteus maximus, gluteus medius, tensor fascia lata, adductors, hamstrings, and hip flexors (iliacus and rectus femoris), can cause increased internal rotation of the hip joint during gait.^{6,7)}

Some previous studies have demonstrated the influence of muscle imbalances around the hip joint on internal rotation.⁸⁻¹⁰⁾ Chong et al.¹¹⁾ indicated that the medial hamstring plays an important role in in-toeing, and Steinwender et al.¹²⁾ reported that internal rotation improved after hamstring release surgery. Aronson et al.⁸⁾ suggested that in-toeing patterns displayed by children with spastic diplegia could be caused by the strong force exerted by the

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adductor muscle. In some studies, internal rotation decreased after adductor release surgery.^{13,14)}

Although several studies have reported changes in hip internal rotation after soft tissue surgery, the results vary, and it is still unclear how much of the internal rotation of the hip joint could be corrected by soft tissue surgeries alone. In addition, the impact of soft tissue surgery on the hip and pelvic rotation was not clear in patients with normal range of pelvic or hip rotation. This study was conducted to determine the average degree of change in pelvic and hip rotation after soft tissue surgery alone (without osteotomy) in pediatric patients with spastic diplegia. We performed a pilot study to analyze the kinematic changes in the pelvis and hip during gait after surgeries only on the soft tissues in spastic diplegia patients. A meta-analysis was also performed using data from the present study and 2 previous studies conducted under similar conditions.

METHODS

The pilot study and meta-analysis were approved by Institutional Review Board. We collected clinical data for pilot study in spastic diplegia patients with gross motor function classification system level 1 or 2 from March 2010 to March 2013. Patients who underwent only soft tissue surgeries and gait analysis at least 1 year after surgery were included in the study. Patients were excluded for the study if they had hip dislocation or subluxation, had botulinum injection 6 months prior to the study, and had a history of other surgical procedures for cerebral palsy including selective posterior rhizotomy and osteotomies on the femur

or tibia. A total of 12 subjects were selected (10 boys and 2 girls). The mean age of the subjects at the time of operation was 10 years (range, 6 years and 3 months to 15 years and 4 months).

For meta-analysis, we conducted searches of the literature in EMBASE and PubMed published between March 1980 and March 2014 using the keywords of “cerebral palsy,” “rotation,” and “surgery.” The search results returned 314 articles, and 12 studies matched our inclusion criteria (Table 1). Of the 12 searched studies, 10 studies were excluded (2 included femoral osteotomy, 5 included spastic hemiplegia, 1 had insufficient surgical data, 1 presented only maximum and minimum values, and 1 had no pelvic rotation value) and 2 studies were selected for analysis (Fig. 1). We analyzed the results of 3 studies including our pilot study and the 2 studies selected from EMBASE and PubMed. The characteristics of the 3 selected studies are presented in Table 2. The study by Lofterod and Terjesen¹⁵⁾ was counted as 2 independent studies because the study subjects were divided into a group with data within 2 standard deviations (SDs) of the normal mean and the other group with data more than 2 SDs above the normal

Table 1. Inclusion Criteria

Population	Patients with spastic diplegia who are able to walk independently
Intervention	Soft tissue surgery only in lower limbs
Comparison	Before and after surgery
Outcome	Pelvic and hip rotation during walking

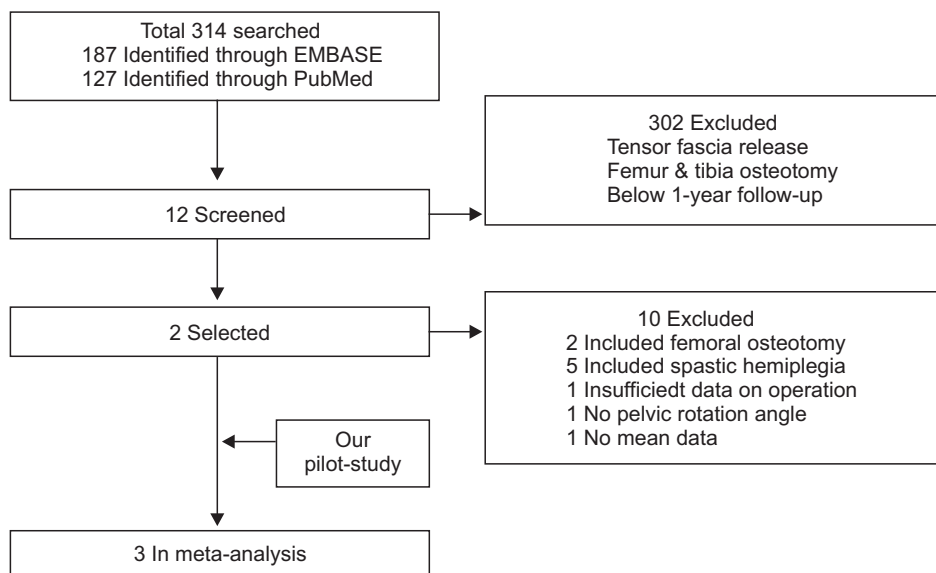


Fig. 1. Systematic search strategy results.

Table 2. Study Characteristics

Study	Study type	Subgroup	Participants	Age (yr)	Follow-up (yr)	Unilateral/bilateral
Steinwender et al. (2000) ¹²⁾	Retrospective	Diplegia	16	10.2	3.4	-
Our pilot study (2014)	Retrospective	Diplegia	12	10.6	1.3	5/7
Lofterod and Terjesen (2010) ¹⁵⁾	Retrospective	Diplegia	28	12.0	1.2	3/25

Table 3. Available Values of Included Studies

Study	Variable	Mean \pm SD ($^{\circ}$)			<i>p</i> -value (preoperative vs. postoperative)	
		Normal	Preoperative	Postoperative		
Steinwender et al. (2000) ¹²⁾	Pelvic rotation	Mean in 1st double leg support	5.7 \pm 2.0	4.9 \pm 7.2	5.0 \pm 5.5	0.74
		Mean in single leg support	0.1 \pm 1.8	-0.1 \pm 6.3	-1.2 \pm 4.6	1.36
		Mean in 2nd double leg support	-5.0 \pm 2.1	-5.8 \pm 6.0	-5.2 \pm 6.0	0.70
	Hip rotation	Mean in 1st double leg support	-9.8 \pm 6.3	7.8 \pm 13.5	-6.2 \pm 10.1	0.00
		Mean in single leg support	2.5 \pm 6.0	9.7 \pm 10.4	-2.8 \pm 9.8	0.00
		Mean in 2nd double leg support	3.3 \pm 7.0	13.6 \pm 10.7	2.0 \pm 9.9	0.00
Our pilot study (2014)	Pelvic rotation	Mean over stance	1.46 \pm 1.11	-0.9 \pm 5.0	1.9 \pm 4.6	0.04
	Hip rotation		3.27 \pm 1.48	-2.8 \pm 10.2	-8.4 \pm 9.1	0.85
Lofterod and Terjesen A (2010) ¹⁵⁾	Pelvic rotation	Mean over stance (< 2 SD)	0 \pm 4	-6	-2	0.07
	Hip rotation		1 \pm 7	11	13	0.60
Lofterod and Terjesen B (2010) ¹⁵⁾	Pelvic rotation	Mean over stance (> 2 SD)	0 \pm 4	-13	-6	0.02
	Hip rotation		1 \pm 7	21	18	0.10

prior to the surgery. Therefore, the study by Lofterod and Terjesen¹⁵⁾ was denoted as either “A” or “B” (Table 3). The study of Steinwender et al.,¹²⁾ which included both sides, was not suitable for the statistical analysis of pelvic rotation, so we discarded these data in the meta-analysis of the pelvis.

In the pilot study, if a patient showed asymmetric pelvic rotation, we selected the leg with external pelvic rotation during stance phase. We included both legs if pelvic rotation was similar on both sides. When describing the direction of rotation in transverse plane of kinematics, “minus” represents external rotation of the joints. The average values of pelvic and hip rotation during the stance phase were compared between the preoperative gait analysis and final follow-up. Statistical analyses were performed using the SAS ver. 9.1 (SAS Institute, Cary, NC, USA). The distribution of continuous data was assessed using the Shapiro-Wilk test. Paired *t*-test was performed to compare

preoperative and postoperative kinematic variables. The significance level was set at *p* < 0.05.

In the meta-analysis, the mean pelvic and hip rotations during stance phase were evaluated. The effect size was calculated using the standardized mean change difference derived from the preoperative and postoperative test results.¹⁶⁾ R Project 3.1.0 (R Foundation for Statistical Computing, Vienna, Austria) was used to analyze the effect size and homogeneity of the selected studies. The significance level was set at *p* < 0.05.

RESULTS

In our pilot study, pelvic retraction was changed from $-0.9^{\circ} \pm 5.0^{\circ}$ to $1.9^{\circ} \pm 4.6^{\circ}$ (*p* = 0.04) (Fig. 2A and B). Hip internal rotation was changed from $-2.8^{\circ} \pm 10.2^{\circ}$ to $-8.4^{\circ} \pm 9.1^{\circ}$, although this change was not statistically significant (*p* = 0.85) (Fig. 2C and D). Furthermore, the changes were

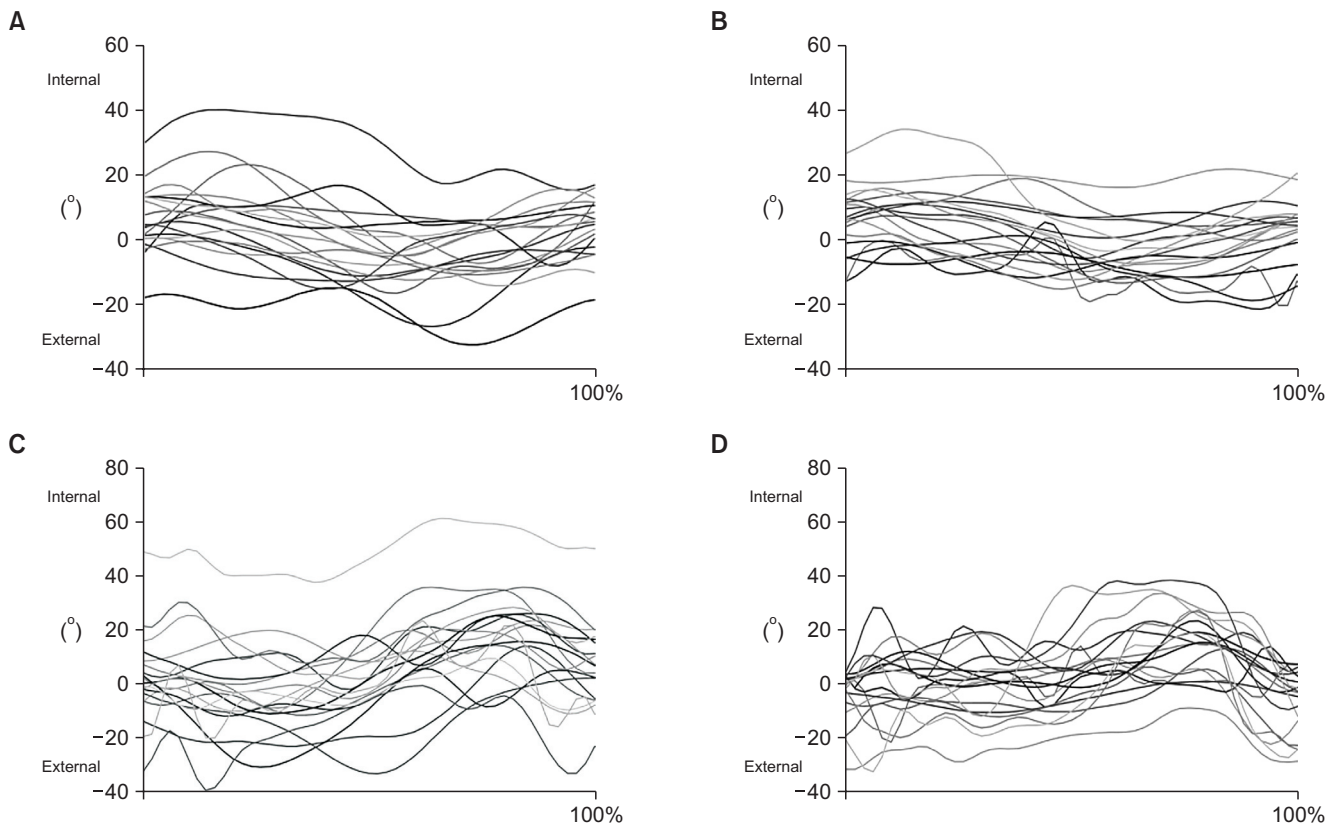


Fig. 2. Pelvic and hip motion in transverse plane during gait in our pilot study. (A) Preoperative pelvic rotation, (B) postoperative pelvic rotation, (C) preoperative hip rotation, and (D) postoperative hip rotation.

Table 4. Type of Surgical Procedures

Study	Patients (limbs)	Psoas recession	Adductor release	Medial hamstring lengthening	Lateral hamstring lengthening	Rectus femoris transfer	Triceps surae lengthening	Foot operation	Total
Steinwender et al. (2000) ¹²⁾	16 (29)	24	0	29	5	29	4	0	91
Our pilot study (2014)	12 (19)	0	1	15	0	1	25	4	46
Lofterod and Terjesen (2010) ¹⁵⁾	28 (53)	38	17	49	0	44	18	0	166

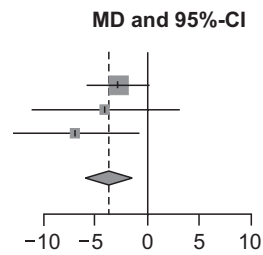
small and the SD was larger than the change itself.

In the meta-analysis, 56 children with spastic diplegic cerebral palsy (6–19 years old) who underwent only soft tissue surgery were included. Twenty-nine limbs of 16 patients were included from the study of Steinwender et al.,¹²⁾ and 19 limbs of 12 patients were included from our pilot study. The number of limbs included from the study of Lofterod and Terjesen¹⁵⁾ varied. A total of 303 operative procedures were performed (Table 4). The most frequently performed surgery was medial hamstring lengthening, followed by rectus femoris transfer. In our series, 4 foot procedures (calcaneal lengthening osteotomy) were included.

Before calculating the effect size to compare the preoperative and postoperative results of pelvic and hip rotation in pediatric patients with spastic diplegia, the homogeneity of these target studies was evaluated using a fixed-effects model. In the meta-analysis, the mean difference between preoperative and postoperative pelvic rotation was -3.61 (95% confidence interval [CI], -6.13 to -1.09), indicating a statistically significant decrease in the pelvic external rotation ($p = 0.005$) (Fig. 3A). The mean difference between preoperative and postoperative hip rotation was 6.6 (95% CI, 3.34 to 9.86), indicating a statistically significant change toward hip external rotation ($p < 0.001$).

A

Study	MD	95%-CI	Weight (%)
Our pilot study 2014	-2.80	[-5.78; 0.18]	71.6
Lofterod_A 2010	-4.00	[-11.07; 3.07]	12.7
Lofterod_B 2010	-7.00	[-13.36; -0.64]	15.7
Fixed effect model	-3.61	[-6.13; -1.09]	100
<i>Heterogeneity: I-squared = 0%, tau-squared = 0, p = 0.499</i>			
<i>Test for overall effect: Z = -2.809 (p = 0.005)</i>			



B

Study	MD	95%-CI	Weight (%)
Steinwender 2000	12.70	[7.07; 18.33]	33.52
Our pilot study 2014	5.60	[-0.39; 11.59]	29.61
Lofterod_A 2010	-2.00	[-13.22; 9.22]	8.44
Lofterod_B 2010	3.00	[-3.11; 9.11]	28.43
Fixed effect model	6.60	[3.34; 9.86]	100
<i>Heterogeneity: I-squared = 63.4%, tau-squared = 20.22, p = 0.042</i>			
<i>Test for overall effect: Z = -3.968 (p < 0.001)</i>			

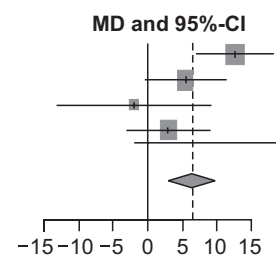


Fig. 3. Meta-analysis of the effect of soft tissue surgery on pelvic rotation (A) and hip rotation (B) in pediatric patients with spastic diplegia. MD: mean difference, CI: confidence interval.

(Fig. 3B).

DISCUSSION

At the initial contact of normal gait, the ipsilateral pelvis is oriented at maximum internal rotation. The pelvis then starts to rotate externally, as the contralateral limb advances forward, and the pelvis is oriented at maximum external rotation at the preswing phase.¹⁷⁾ The hip remains in the neutral position at initial contact and then shows gradual internal rotation during the loading-response and single-limb support stages.¹⁷⁾ Pediatric patients with spastic diplegia display excessive hip internal rotation due to skeletal deformities or muscle imbalances, including muscle weakness and spasticity.²⁾ If a patient shows increased hip internal rotation and femoral anteversion compared to typically developing children, osteotomy is the treatment of choice. However, it is difficult to calculate separately the change in hip rotation achieved by osteotomy and by musculotendon lengthening, if the two procedures are performed simultaneously. In addition, it has not been determined whether a hamstring or adductor lengthening would result in changes in the hip and pelvic rotation in patients with normal hip or pelvic rotation. In this study, we attempted to evaluate the average change in the hip and pelvic rotation during stance phase after soft tissue surgery in spastic diplegic patients.

Results of changes in hip rotation after soft tissue surgery vary among studies. Lovejoy et al.⁹⁾ reported a decrease of approximately 5° in internal rotation of the hip after soft tissue surgery alone. However, this study was not included in our meta-analysis because it represented a

comparison of range values rather than mean values of internal rotation. Steinwender et al.¹²⁾ reported a significant improvement (12°) in internal hip rotation after hamstring release and distal rectus femoris procedures, indicating positive effects of multilevel soft tissue surgery, especially the medial hamstring release. Even though our pilot study and the study of Lofterod and Terjesen¹⁵⁾ did not show any significant changes, the rotation of the hip joint changed toward external rotation in the meta-analysis. Therefore, preoperative internal hip rotation could be expected to improve slightly after soft tissue surgery alone based on the meta-analysis. However, changes in the preoperatively normal or externally rotated hip are not clear. In our pilot study, the preoperatively externally rotated hips rotated more externally after operation, and the amount of internal rotation of the hip in Lofterod A group (< 2 SD) was increased, although the changes were not significant in both studies. Further studies will be needed in these cases.

The average pelvic rotation after surgery was changed toward internal rotation, and results of the meta-analysis suggest that it improved significantly considering preoperative external rotation in each study. In the study by Lofterod and Terjesen,¹⁵⁾ soft tissue surgery did not result in significant changes in pelvic rotation in subjects whose preoperative external pelvic rotation was within the normal range, whereas subjects whose preoperative average pelvic retraction was above 2 SD than the control group had an average of 7° improvement. In children with spastic diplegia, the pelvis might be retracted (externally rotated) to compensate the internally rotated hip.^{18,19)} Accordingly, we expected pelvic retraction could be improved

with the correction of the hip internal rotation. However, there was no notable improvement in hip internal rotation in Lofterod B (> 2 SD) and pelvic retraction was improved regardless of hip external rotation in our pilot study. In a study by Kay et al.²⁰⁾ that included femoral osteotomy, the improvement in pelvic retraction after soft tissue surgery varied from 3.3° to 21° . Therefore, it seems that pelvic retraction is a compensatory movement that could be influenced by gait improvement as well as hip internal rotation.²¹⁾

Our study has several limitations. In three-dimensional (3D) gait analysis, hip rotation is measured along the longitudinal axis of the femur, which is defined as a line connecting the knee and hip joint center relative to the pelvis. Hence, measurement errors can occur depending on the places of markers attached by each clinician.²²⁾ Few studies have been conducted on the effects of soft tissue surgery alone (without femoral osteotomy) in patients with spastic diplegic cerebral palsy; therefore, few studies were available for the meta-analysis. Furthermore, the standard deviation was very wide in our pilot study and the study of Steinwender et al.¹²⁾ Although we chose the

previous studies carefully, there was no grouping between Achilles tendon lengthening and gastrosoleus recession. In addition, we did not evaluate the foot progression angle. The pilot study did not involve psoas recession surgery but did include 4 foot procedures, so it may be difficult to compare with other cases. We selected studies with at least 1-year follow-up because the effects of surgery on children with cerebral palsy change as they grow, and thus long-term follow-up studies are essential in this population.

Nevertheless, this study is the first systematic review to investigate changes in the transverse plane in ambulant pediatric patients with diplegia who underwent soft tissue surgery alone. In this meta-analysis, during overall stance phase, pelvic retraction was improved and hip rotation was changed toward external rotation after surgery, although the improvement was not remarkable.

CONFLICT OF INTEREST

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

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