

# Progression of Hip Displacement during Radiographic Surveillance in Patients with Cerebral Palsy

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## INTRODUCTION

Cerebral palsy (CP) is a group of permanent developmental disorders caused by nonprogressive disturbances occurring in the brain of a developing fetus or infant (1). The estimated prevalence in the general population is 2 to 4 per 1,000 live births (2,3).

Patients with CP experience difficulties with movement and posture, leading to activity limitations and musculoskeletal problems. A common musculoskeletal problem in this population is hip displacement (e.g., subluxation or dislocation) (4). Hip displacement occurs in approximately 21% of patients with CP (5) and 50% of patients with quadriplegic CP (6). In patients with CP, hip displacement can cause pain and severe contractures, resulting in problems with positioning, sitting, standing, and walking (4,7). However, hip displacement can be detected

Progression of hip displacement is common in patients with cerebral palsy (CP). We aimed to investigate the rate of progression of hip displacement in patients with CP by assessing changes in radiographic indices according to Gross Motor Function Classification System (GMFCS) level during hip surveillance. We analyzed the medical records of patients with CP aged < 20 years who underwent at least 6 months interval of serial hip radiographs before any surgical hip intervention, including reconstructive surgery. After panel consensus and reliability testing, radiographic measurements of migration percentage (MP), neck-shaft angle (NSA), acetabular index (AI), and pelvic obliquity (PO) were obtained during hip surveillance. For each GMFCS level, annual changes in radiographic indices were analyzed and adjusted for affecting factors, such as sex, laterality, and type of CP. A total of 197 patients were included in this study, and 1,097 radiographs were evaluated. GMFCS classifications were as follows: 100 patients were level I-III, 48 were level IV, and 49 were level V. MP increased significantly over the duration of hip surveillance in patients with GMFCS levels I-III, IV, and V by 0.3%/year ( $P < 0.001$ ), 1.9%/year ( $P < 0.001$ ), and 6.2%/year ( $P < 0.001$ ), respectively. In patients with GMFCS level IV, NSA increased significantly by 3.4°/year ( $P < 0.001$ ). Our results suggest that periodic monitoring and radiographic hip surveillance is warranted for patients with CP, especially those with GMFCS level IV or V. Furthermore, physicians can predict and inform parents or caregivers regarding the progression of hip displacement in patients with CP.

**Keywords:** Cerebral Palsy; Progression; Hip Displacement; Hip Surveillance

with routine hip surveillance (8,9), which is now a part of CP management. Therefore, physicians need to know the characteristics and risk factors associated with hip displacement.

Many studies have discussed the risk factors for hip displacement in patients with CP. One such relationship is between gross motor disability and Gross Motor Function Classification System (GMFCS) level. GMFCS level is an important factor in determining the risk of hip displacement in patients with CP; the risk is increased with increased gross motor disability, with the highest risk among children classified as GMFCS level V (7,10). In addition to GMFCS level, type of CP is also associated with hip displacement. For example, hip displacement is more frequent in quadriplegic CP than in hemiplegic CP (11,12).

The present study aimed to measure the rate of progression of hip displacement by assessing changes in radiographic indices during hip surveillance according to GMFCS level, and to

investigate factors that affect the progression of hip displacement in patients with CP.

## MATERIALS AND METHODS

### Subjects

Consecutive patients with CP aged < 20 years who underwent at least 6 months of serial hip radiographs before any surgical hip intervention, including reconstructive surgery, were included in this study. Exclusion criteria were as follows: 1) presence of hip deformity caused by trauma, infection, tumor, etc.; 2) presence of neuromuscular disease other than CP; 3) initial migration percentage (MP) > 100%; and 4) inadequately taken hip radiographs (Fig. 1).

Baseline demographic data, such as age and sex, type of CP (unilateral vs. bilateral), laterality (right or left), and GMFCS level were obtained from medical record review. Hip radiographs were obtained in the supine position with the hips rotated internally by approximately 30° (13). For patients with hip flexion contractures, the technicians manipulated the patients to fit onto the cassette by gently pushing them into hip and knee extension according to our clinical protocol. Radiographs were

obtained using a UT 2,000 unit (Philips, Eindhoven, the Netherlands) under the following conditions: source-to-image distance, 100 cm; 60 kVp; and 10 mAs. All conventional radiographic images were digitally acquired using a picture archiving and communication system (PACS; Infinitt, Seoul, Korea). All radiographic measurements were subsequently obtained using PACS software.

### Consensus building and reliability testing

A consensus-building session to select and define the radiographic measurements was held by five surgeons who had orthopedic experience of 26, 15, 13, 9, and 8 years, respectively. Previous studies were reviewed (14-19), and four radiographic measurement parameters were selected by panel consensus. The following measurements on serial hip radiographs were considered appropriate in determining the risk of developing hip displacement in patients with CP: MP (16), neck-shaft angle (NSA) (14,15), acetabular index (AI) (19), and pelvic obliquity (PO) (17) (Fig. 2).

MP, which ranges from 0% to 100%, was calculated as the width of the femoral head lateral to Perkin's line divided by the total width of the femoral head (16,20). NSA was measured as

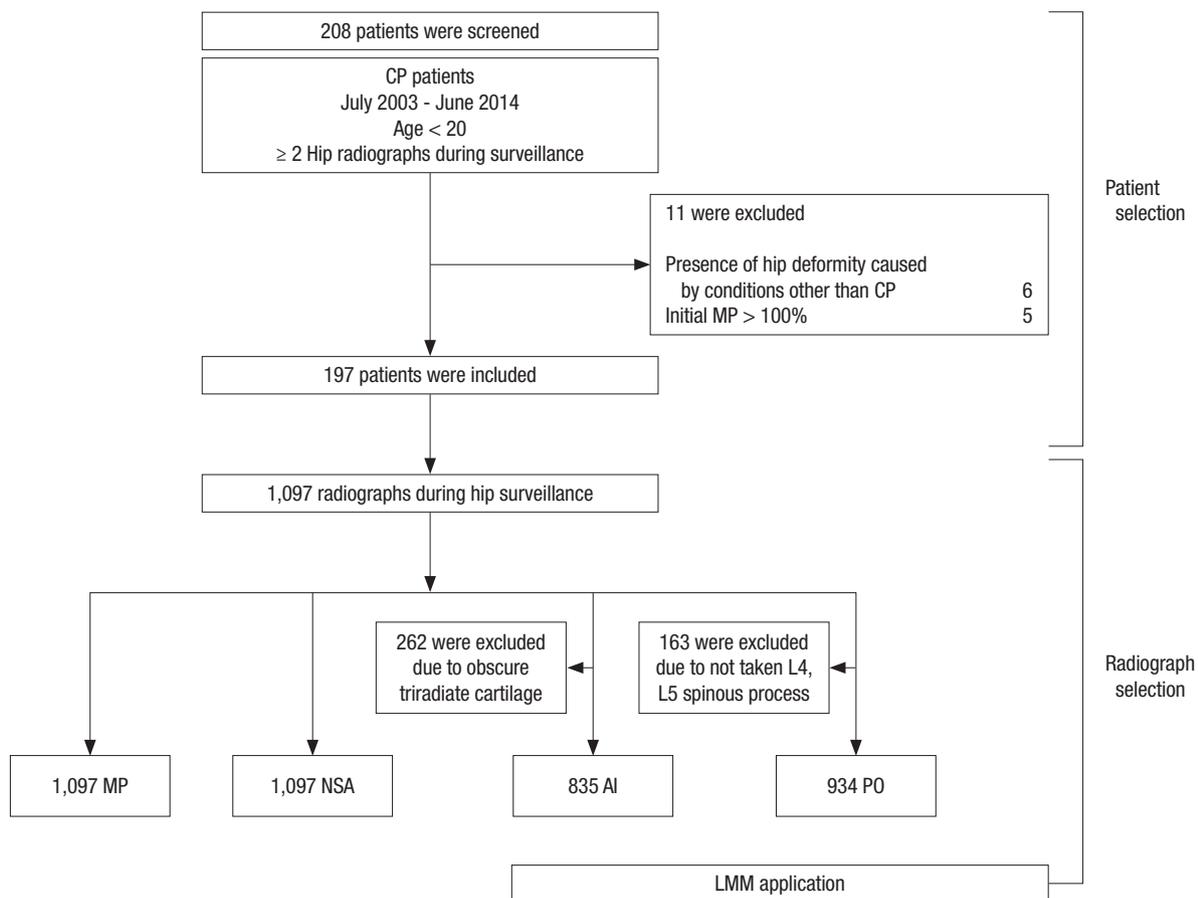
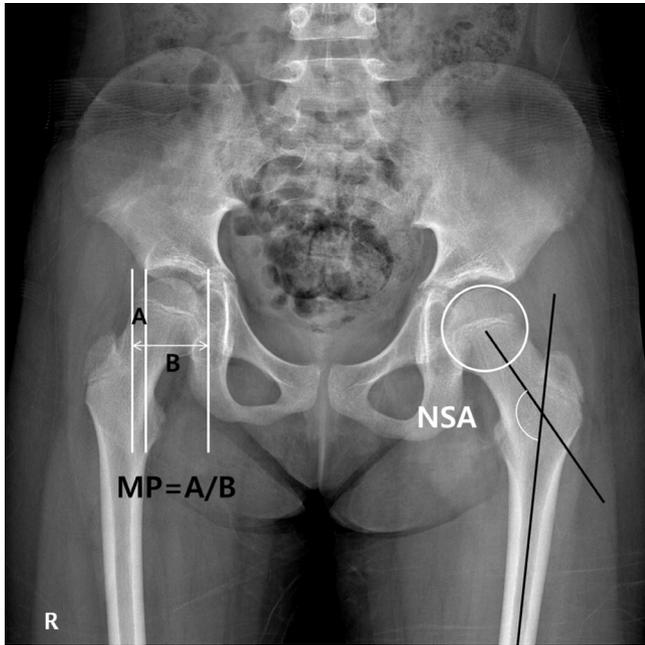


Fig. 1. Inclusion and exclusion criteria.

CP, cerebral palsy; MP, migration percentage; NSA, neck-shaft angle; AI, acetabular index; PO, pelvic obliquity; LMM, linear mixed model.



**Fig. 2.** Hip internal rotation view. For the right hip, migration percentage (MP) was calculated by dividing the width of the femoral head lateral to Perkin's line (A) by the total width of the femoral head (B). For the left hip, neck-shaft angle (NSA) was defined as the angle between a line passing through the center of the femoral shaft and another line connecting the femoral head center and the midpoint of the femoral neck. Femoral head center was the center of the largest best-fitting circle inside the femoral head.

the angle between a line passing through the midpoint of the femoral shaft and another line connecting the femoral head center and the midpoint of the femoral neck. Femoral head center was defined as the center of the largest best-fitting circle inside the femoral head (14,15). AI was measured as the slope of the acetabular roof, which is the angle between the acetabular roof and Hilgenreiner's line (19). PO was measured as the angle between the spinous process line and the iliac crest line (17). The spinous process line was drawn through the spinous processes of the L4 and L5, whereas the iliac crest line was drawn perpendicular to the spinous process line that ran across the top of the iliac crests.  $PO > 3^\circ$  was recorded as positive (+),  $\leq 3^\circ$  as negative (-), and in the case of inappropriate radiographs, "uncheckable".

Interobserver reliability testing was conducted before obtaining the main measurements. Interobserver reliability was determined among three surgeons who had orthopedic experience of 13, 9, and 8 years respectively, using intraclass correlation coefficients (ICCs). These surgeons measured radiographic indices independently, and were blinded to the patients' clinical information and the other surgeons' measurements. One surgeon repeated the radiographic measurements to assess intraobserver reliability 3 weeks after interobserver reliability testing. Reliability tests were performed using 36 patients. The sample size of 36 patients was calculated with a target ICC of 0.8 and

a 95% confidence interval of 0.2 with the setting of a single measurement and absolute agreement (21).

### Main measurement and linear mixed model application

After reliability testing, one of the authors who had orthopedic experience of 8 years assessed all the radiographs. The patients' sex, age, laterality, and date of the radiographs were included in the measurement data. A linear mixed model (LMM) was used to analyze annual changes in radiographic indices of hip displacement during hip surveillance. A research assistant who did not otherwise participate in the study collected all the measurements.

An LMM is a parametric linear model for longitudinal data; this model quantifies the relationships between a continuous dependent variable and various predictor variables, providing a simple and effective way to incorporate within- and between-subject variations and the correlation structure of longitudinal data. An LMM is a statistical model consisting of both fixed and random effects. Fixed effects represent categorical levels that are measurable and not random, such as sex. Random effects are factors that can be specified for individuals within a population that account for variation within individuals. Therefore, estimation of annual changes in radiographic indices of hip displacement using an LMM may provide more practical information to clinicians.

For each GMFCS level, MP, NSA, AI, and PO were adjusted for affecting factors using an LMM, with sex and type of CP as fixed effects and duration of hip surveillance, laterality (left or right) (22), and each subject as random effects. The covariance structure was assumed to be the variance components. The estimation method used restricted maximum likelihood estimation to produce unbiased estimators. The LMM was used to estimate the rate of lateral migration of the femoral head by incorporating the linear duration of hip surveillance and sex as covariates. Examination of the individual pattern of the rate of hip displacement along with follow-up time suggested a model with a random slope and random intercept. The linear effects of duration of hip surveillance, sex, and age were integrated to evaluate the estimates of the four measurements.

### Statistical analysis

In the present study, reliability was assessed using ICCs and a two-way random-effect model, assuming a single measurement and absolute agreement (23,24), with a target ICC of 0.8 and Bonett's approximation to set the width of 95% confidence intervals to 0.2 (21). Descriptive statistics were used to summarize patient demographics and radiographic measurements.

This study included bilateral cases. To consider data dependency within subjects, we adopted an LMM, as proposed in a previous study (22). Furthermore, the LMM was used to model the measurements (MP, NSA, AI, and PO) for assessing the co-

variate effect and for examining factors significantly contributing to the four measurements. Statistical analyses were conducted using R version 2.15.2 (R Foundation for Statistical Computing, Vienna, Austria) with the NLME package. All statistics were two-tailed, and *P* values < 0.05 were considered significant.

**Ethics statement**

The institutional review board of the Seoul National University Bundang Hospital reviewed and approved the protocol of this study (IRB approval number; B-1307/210-106). The need for informed consent was waived by the board.

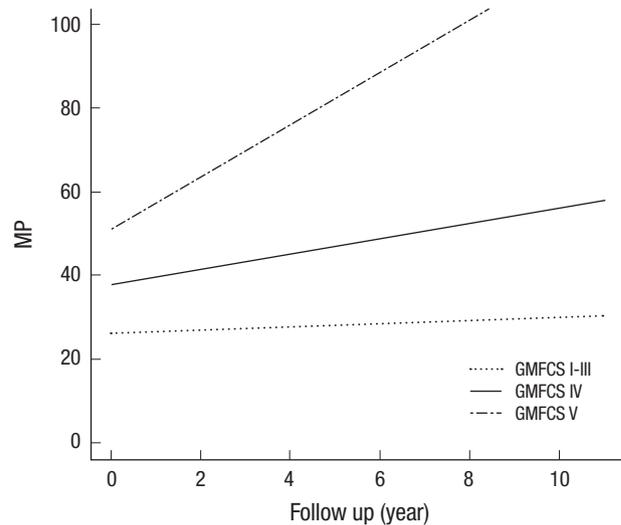
**RESULTS**

After implementation of inclusion and exclusion criteria, 197 patients were included in this study. The mean age of the patients was 8.3 ± 3.6 years, and the mean duration of hip surveillance before any surgical hip intervention, including reconstructive surgery, was 2.0 ± 2.2 years. Of the included patients, 19 (9.6%) had unilateral spastic CP and 178 (90.4%) had bilateral spastic CP. In addition, 100 patients (50.7%) were classified as GMFCS level I-III, 48 (24.4%) as level IV, and 49 (24.9%) as level V (Table 1).

In terms of interobserver reliability, all radiographic measurements showed good-to-excellent reliability for clinical use. Both

intra- and inter-observer reliability were highest for MP (ICC, 0.989 and 0.977, respectively) (Table 2).

We then evaluated MP, NSA, AI, and PO which were adjusted for affecting factors such as sex, duration of surveillance, and laterality, for each GMFCS level. MP increased significantly with the duration of surveillance. In patients with GMFCS levels I-III, IV, and V, MP increased by 0.3%/year (*P* < 0.001), 1.9%/year (*P* < 0.001), and 6.2%/year (*P* < 0.001), respectively (Table 3, Figs. 3 & 4). NSA was significantly affected by duration of surveillance in patients with GMFCS level IV, who showed an increase of 3.4°/year (*P* < 0.001) (Table 4). AI and PO did not show significant annual changes after adjusting for sex, laterality, and duration of surveillance.



**Fig. 3.** Estimation of the progression of migration percentage (MP) by a linear duration of hip surveillance effect according to Gross Motor Function Classification System (GMFCS) level.

**Table 1.** Patient's summary

	Parameters	Values
Patient's Information	No. of patients (Male/Female)	197 (125/72)
	CP type (Unilateral/Bilateral)	19/178
	GMFCS level (I-III/IV/V)	100/48/49
	Follow up duration, yr	2.0 ± 2.2
Radiographic characteristics	MP, %	
	Initial	35.3 ± 24.6
	Last F/U	43.7 ± 27.0
	NSA, °	
	Initial	149.1 ± 9.0
	Last F/U	155.6 ± 9.4
	AI, °	
	Initial	21.4 ± 6.9
Last F/U	22.4 ± 7.8	
PO (-/+/uncheckable)	Initial	133/32/32
	Last F/U	136/40/21

CP, cerebral palsy; GMFCS, gross motor function classification system; MP, migration percentage; NSA, neck shaft angle; AI, acetabular index; PO, pelvic obliquity.

**Table 2.** Intra- and inter-observer reliabilities of measurements

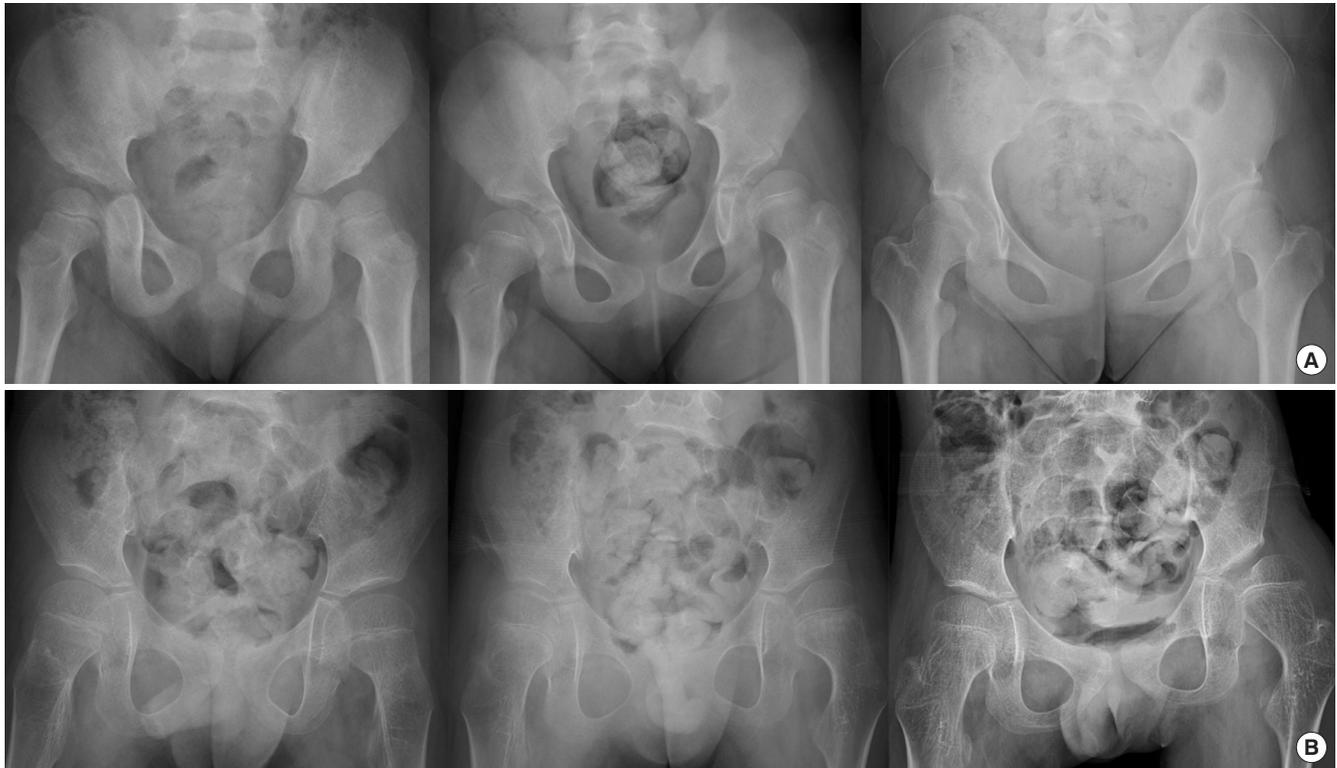
Measurements	Intra-observer reliability		Inter-observer reliability	
	ICC	95% CI	ICC	95% CI
MP	0.989	0.965-0.996	0.977	0.959-0.987
NSA	0.969	0.940-0.984	0.900	0.761-0.954
AI	0.937	0.821-0.971	0.732	0.589-0.841
PO	0.912	0.835-0.954	0.716	0.568-0.831

ICC, intraclass correlation coefficient; CI, confidence interval; MP, migration percentage; NSA, neck shaft angle; AI, acetabular index; PO, pelvic obliquity.

**Table 3.** The estimation of factors affecting MP using Linear Mixed Models

Factors	MP (GMFCS level I-III)			MP (GMFCS level IV)			MP (GMFCS level V)		
	Estimation	SE	<i>P</i> value	Estimation	SE	<i>P</i> value	Estimation	SE	<i>P</i> value
Intercept	27.3	1.5	< 0.001	45.5	4.0	< 0.001	51.7	5.0	< 0.001
Follow up	0.34	0.09	< 0.001	1.9	0.4	< 0.001	6.2	1.5	< 0.001
Sex	-3.3	1.6	0.048	-8.9	4.2	0.037	4.6	5.4	0.396

MP, migration percentage; GMFCS, gross motor function classification system; SE, standard error.



**Fig. 4.** Serial hip radiographs obtained during the duration of hip surveillance. **(A)** Radiographs of a 5-year-old girl with Gross Motor Function Classification System (GMFCS) level III. The left hip seemed to have displacement, but there was no acetabular dysplasia or hip displacement at the last follow-up, 6 years after the initial radiographs were obtained. Migration percentage (MP) decreased throughout the duration of hip surveillance. **(B)** Radiographs of a 7-year-old boy with GMFCS level V. Left hip displacement progressed by a lateral migratory course. Initial MP was 14%, but increased to 50% at the last follow-up, 5 years after the initial radiographs were obtained.

**Table 4.** The estimation of factors affecting NSA using Linear Mixed Models

Factors	NSA (GMFCS level I-III)			NSA (GMFCS level IV)			NSA (GMFCS level V)		
	Estimation	SE	P value	Estimation	SE	P value	Estimation	SE	P value
Intercept	148.1	1.1	< 0.001	154.1	1.8	< 0.001	161.4	1.5	< 0.001
Follow up	0.05	0.15	0.755	3.4	0.8	< 0.001	1.1	0.7	0.1
Sex	-1.3	1.2	0.272	-0.7	1.9	0.711	-2.5	1.6	0.1

NSA, neck shaft angle; GMFCS, gross motor function classification system; SE, standard error.

## DISCUSSION

This study investigated the rate of progression of hip displacement in patients with CP, the factors influencing this progression, and the relationship between GMFCS level and degree of progression. Our results suggest that there was an annual increase in MP before reconstructive hip surgery and that progression rates differed according to GMFCS level. Moreover, MP tended to increase as GMFCS level increased, indicating that hips become more unstable with less favorable function (Fig. 3). These results are concurrent with those of previous studies (25,26).

This study has some limitations to be addressed before discussing the results. First, the data were collected retrospectively, which introduces the possibility that a strictly uniform protocol might not be maintained. Second, measurements were obtain-

ed from radiographs of internally rotated hips, and the assessment may have been affected by the degree of internal rotation. Ideally, internal hip rotation radiographs should be obtained with the femoral head and neck located perpendicular to the direction of the radiation beam. In clinical practice, simple hip radiographs are generally obtained without knowledge of the degree of femoral anteversion. However, internal rotation of the femur facilitates determination of the femoral NSA to within 10° (20). Additionally, previous studies, including the study by Reimers and Bialik (27), have shown that MP measurements of hip do not differ significantly in neutral and internally rotated positions. Third, in some hip radiographs, triradiate cartilage were equivocal because of the bony maturation process in some radiographs; therefore, in some cases AI could not be measured. Lastly, some hip radiographs did not include the L4 and L5 spinous processes or these processes were rotated; therefore, in

some cases PO could not be measured. In these cases, LMM was applied by missing value.

The natural history of spastic hip disease involves progressive lateral displacement of the hip secondary to impaired mobility and spastic hypertonia of the hip adductor and flexor musculature (28). To determine the extent of hip displacement, the most accepted and reproducible measurement is MP (29,30), which is a measure of containment of the femoral head within the acetabulum in the coronal plane (31). Reimers and Bialik (27) described MP as an important index of hip displacement and the most important factor in preoperative planning. Since MP is the most valid, reliable, and useful linear measure of hip displacement in children with CP (29,30), we focused on MP as a measurement of hip displacement during the duration of hip surveillance.

In this study, we also noted that GMFCS level was a significant factor affecting the progression of hip displacement. Terjesen (32) reported that hip displacement occurs in 63% of children with GMFCS level IV or V, and that MP increases as functional level increases by 0.2%/year for those with GMFCS level I, and by 9.5%/year for those with GMFCS level V. The differences between children with no gait function (GMFCS levels IV and V) and those with gait function were significant. However, his paper presented only descriptive statistics. The strength of this study is the use of proper statistical methods (LMM), which enable us to adjust for factors affecting the results, such as sex, age, laterality, and date of the radiographs.

A prior study of normal children found a pattern of slowly decreasing NSA by age, from a mean of 135°-140° at birth to 125° at skeletal maturity via an indirect measurement method. However, in patients with CP, NSA increased by 30°-50° greater than normal (33). In this study, initial and final NSA were 149.1° ± 9.0° and 155.6° ± 9.4°, respectively. We consider that increased NSA might be associated with persistent fetal alignment, which may be caused by delayed walking and limitations in gross motor function (14,15,26,34,35).

In summary, our study retrospectively evaluated the rate of progression of hip displacement in patients with CP. Our results suggest that routine radiographic surveillance is important, especially in patients with GMFCS level IV or V.

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This study is from the master's degree thesis of the first author (JYP).

## DISCLOSURE

The authors have no potential conflicts of interest to disclose.

## AUTHOR CONTRIBUTION

Conception and coordination of the study: Park MS. Consideration of ethical issues: Park JY, Moon SY. Data collection and analysis: Park JY, Choi Y, Cho BC, Moon SY, Chung CY. Statistical analysis: Lee KM, Sung KH, Kwon SS. Manuscript approval: all authors.

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