

Anterior Knee Pain in Patients with Cerebral Palsy

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Background: The aim of this study was to identify the risk factors for anterior knee pain in patients with cerebral palsy.

Methods: This prospective study investigated the risk factors for anterior knee pain in 127 ambulatory patients with spastic cerebral palsy in terms of walking pain, resting pain, and provocative pain. Demographic data analysis and physical examination for measuring the knee flexion contracture and unilateral and bilateral popliteal angles were performed. Patellar height was measured on radiographs, and patella alta was identified. The risk factors for anterior knee pain were analyzed using multivariate analysis with a generalized estimating equation.

Results: Seventy-seven patients were found to have patella alta based on the radiographic measurements (60.6%). Overall, sixteen patients (12.6%) had either unilateral or bilateral anterior knee pain. Of these, 6 patients showed a visual analogue scale (VAS) ≤ 3 , 9 patients showed $3 < \text{VAS} \leq 7$, and one patient showed a VAS > 7 . Age was found to be a significant risk factor for walking pain and resting pain with odds ratios (ORs) of 1.08 (95% confidence interval [CI], 1.02 to 1.14) and 1.09 (95% CI, 1.03 to 1.15), respectively. In the multivariate analysis, knee flexion contracture was a significant protective factor with an OR of 0.92 (95% CI, 0.85 to 0.98).

Conclusions: Approximately 12.6% of ambulatory patients with spastic cerebral palsy were found to have anterior knee pain in our hospital-based cohort study. Age was found to be a significant risk factor for anterior knee pain while walking and resting.

Keywords: Patellofemoral pain syndrome, Cerebral palsy, Patella alta, Popliteal angle

An abnormal knee extensor apparatus is known to be associated with deterioration of gait in patients with cerebral palsy by causing abnormal knee joint motion.¹⁾ Furthermore, previous studies have reported that spasticity causes abnormal knee joint kinematics and kinetics, and that knee flexion deformity caused by hamstring contracture increases the contact force in the patellofemoral joint.^{2,3)}

Eventually, this condition is believed to cause a serious problem in patients with cerebral palsy, i.e., anterior knee pain.

Several surgical procedures have been devised to correct the knee flexion deformity and to treat the weakened knee extensor apparatus in patients with cerebral palsy, such as distal hamstring lengthening,⁴⁻⁶⁾ extension osteotomy of the distal femur,¹⁾ anterior hemiepiphysal stapling of the distal femur,⁷⁾ and patellar tendon advancement.⁸⁾ The rationale behind all of these procedures includes the correction of structural flexion deformities of the knee joint and restoration of the knee extensor mechanism. Previous studies reported that these procedures effectively restore knee biomechanics and improve abnormal

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gait patterns, including a crouch gait.¹⁾ However, it is not known whether these procedures are effective in reducing or eliminating anterior knee pain in patients with cerebral palsy.

Therefore, the aim of this study was to identify the risk factors for anterior knee pain in patients with cerebral palsy. These risk factors were examined comprehensively in terms of demographics, physical examination findings, and radiographic measurements; and anterior knee pain was evaluated with respect to walking pain, resting pain, and provocative pain.

METHODS

This prospective study was approved by the Institutional Review Board of our hospital (a tertiary referral center for cerebral palsy), and informed consent was obtained from patients or legal guardians. The study included consecutive patients with spastic cerebral palsy who were able to walk (gross motor function classification system [GM-FCS] level I–III)⁹⁾ and communicate effectively with other people (communication function classification system [CFCS] level I–III),¹⁰⁾ registered at our hospital between June 2009 and January 2010. The exclusion criteria applied were as follows: (1) a neuromuscular disease in addition to cerebral palsy and (2) previous surgery that might cause a change in the patellar height, such as patellar tendon advancement and supracondylar extension osteotomy of the femur.

Demographic Data and Physical Examination

Demographic information was collected, including data on gender, age, extent of involvement (bilateral vs. unilateral), and GMFCS level.⁹⁾ Physical examination was performed by an orthopedic surgeon with 8 years of experience, and it included the assessment of knee flexion contracture, unilateral popliteal angle, and bilateral popliteal angle in the supine position. Knee flexion contracture was defined as the angle between the long axis of the thigh and that of the lower leg when the examiner gently and passively extended the knee. While measuring the popliteal angle, the test limb was flexed to 90° at the hip and the knee was extended passively with the contralateral hip and knee in extension. The angle between the longitudinal axis of the lower leg and the vertical line passing through the thigh was defined as the unilateral popliteal angle. The same measurement was performed in the contralateral hip in 90° of flexion, and this was referred to as the bilateral popliteal angle. Angles were measured by a research assistant using a standard goniometer with an arm length of 18 cm

in one degree increments.

Anterior Knee Pain Evaluation

The patients included in the study could act as effective sender and receiver with either familiar or unfamiliar partners in communication (CFCS level I–III). All patients were questioned regarding the presence of anterior knee pain, which was evaluated in terms of walking pain, resting pain, and provocative pain. Provocative anterior knee pain was examined by performing a patellar compression test, whereby the examiner compressed the patella into the trochlear groove of the femur. The presence of anterior knee pain was assessed using visual analogue scale (VAS).¹¹⁾

Patella Alta Based on Radiographic Measurements

Patellar height was evaluated on lateral knee radiographs that were taken at 70 kVp and 7 mAs using a UT 2000 unit (Philips, Eindhoven, The Netherlands) in approximately 30° of knee flexion at a source-to-image distance of approximately 100 cm. All radiographic images were digitally acquired using a picture archiving and communication system (PACS; Impax, Agfa, Antwerp, Belgium), and assessments were subsequently carried out using the PACS software. A previous report demonstrated that the Insall-Salvati (IS)¹²⁾ and the Koshino-Sugimoto (KS)¹³⁾ methods have reasonable reliability and validity with respect to depicting the patellar height in children and adolescents,¹⁴⁾ and therefore, IS and KS ratios were used to determine the patellar height. On lateral knee radiographs, the IS method was used to measure patellar tendon length relative to patellar bone length, and the KS method was to measure the ratio of the distance between the proximal tibial physis and the center of the patella to the distance between the distal femoral physis and the proximal tibial physis. Patella alta was defined as an IS ratio of > 1.20 or a KS ratio of > 1.33.^{12,13)}

Consensus building and reliability sessions were held before taking radiographic measurements. The two observers (both orthopedic surgeons with 10 and 8 years of experience, respectively) agreed on the definitions of radiographic landmarks and measurements. Reliabilities were determined using intraclass correlation coefficients (ICCs), and the assessment was performed using 51 randomly selected joints of 51 subjects after performing the required sample size calculation. Interobserver reliability was determined between the two orthopedic surgeons who individually measured the IS and KS ratios, and were unaware of each other's findings. Intraobserver reliability was determined by having one of the observers repeat the above radiographic measurements 3 weeks later. Order of measurements was randomized for each observer.

Statistical Analysis

Sample size estimation was performed to determine the minimal sample size for reliability testing. Reliability was determined using ICCs and a two-way random effect model. When ICCs were calculated, single measurement and absolute agreement were assumed. Using an ICC target value of 0.8, Bonett's approximation was used in the setting of 0.2 as a width of 95% confidence interval (CI) for the two raters.¹⁵⁾ The minimal sample size was calculated as 51.

Associations between risk factors and anterior knee pain were assessed using the generalized estimating equation (GEE)¹⁶⁾ models to calculate adjusted odds ratios (ORs) and 95% CIs. This procedure takes into account the correlation between the left and right knees in subjects. Multivariate analysis using GEE models was used to identify the risk factors that significantly affected anterior knee pain. Univariate analysis using GEE models was performed initially in order to reduce the number of variables entered into the multivariate analysis. Variables significant at the $p < 0.1$ level in the univariate study and clinically important factors were included as potential risk factors in the multivariate analysis. Statistical analyses were conducted using SPSS ver. 15.0 (SPSS Inc., Chicago, IL, USA), and statistical significance was accepted for $p < 0.05$.

RESULTS

One hundred and twenty-seven ambulatory patients (254 knees) with spastic cerebral palsy were finally included in this study. Mean patient age was 14.5 years (standard deviation, 7.9 years), and there were 89 male and 38 female patients. Thirty-three patients had unilateral anterior

knee pain and 94 patients had bilateral anterior knee pain. Thirty-nine patients were classified as GMFCS level I, 65 patients were classified as GMFCS II, and 23 patients were classified as GMFCS III (Table 1).

Seventy-seven patients were found to have patella alta based on the radiographic measurements (60.6%). Of these, 26 patients had unilateral patella alta and 51 patients had bilateral patella alta. The interobserver reliabilities of IS and KS were 0.910 (95% CI, 0.821 to 0.956) and 0.889 (95% CI, 0.801 to 0.940), respectively, and the intraobserver reliabilities of IS and KS were 0.947 (95% CI, 0.894 to 0.974) and 0.922 (95% CI, 0.840 to 0.963), respectively.

Six patients had abnormal radiographic findings of the knee other than that of patella alta. The abnormal radiographic findings were as follows: inferior pole fragmentation in two cases, ossicles on the tibial tuberosity in two cases, superior pole fragmentation in one case, and elongated patella in one case. However, none of these patients had anterior knee pain.

Ten patients (six patients with unilateral anterior knee pain and four patients with bilateral anterior knee pain) had anterior knee pain while walking (7.9%) and six patients (three patients with unilateral anterior knee pain and three patients with bilateral anterior knee pain) had anterior knee pain while resting (4.7%). Thirteen patients (five patients with unilateral anterior knee pain and eight patients with bilateral anterior knee pain) had provocative anterior knee pain during the patellar compression test. Overall, sixteen patients (12.6%) had either unilateral or bilateral anterior knee pain. Of these, six patients showed a VAS ≤ 3 , nine patients showed $3 < \text{VAS} \leq 7$, and one patient showed a VAS > 7 .

With respect to walking pain, age ($p = 0.013$) and patella alta ($p = 0.997$) were the potential risk factors, and they were further analyzed in the multivariate analysis. On performing the multivariate analysis with GEE, age was found to be a significant risk factor for anterior knee pain while walking, with an OR of 1.08 (95% CI, 1.02 to 1.14) (Table 2).

With respect to resting pain, age ($p = 0.008$), knee flexion contracture ($p = 0.078$), and patella alta ($p = 0.622$) were the potential risk factors. In the multivariate analysis with GEE, age was a significant risk factor with an OR of 1.09 (95% CI, 1.03 to 1.15) (Table 3).

With respect to provocative pain, age ($p = 0.131$), knee flexion contracture ($p = 0.024$), and patella alta ($p = 0.150$) were the potential risk factors. In the multivariate analysis, knee flexion contracture was a significant protective factor with an OR of 0.92 (95% CI, 0.85 to 0.98) (Table 4).

Table 1. Patient Demographics and Physical Examination

Variable	Value
No.	127 (254 knees)
Mean age (SD)	14.5 (7.9)
Gender (male : female)	89 : 38
GMFCS level (I/II/III)	39/65/23
Extent of involvement (unilateral/bilateral)	33/94
Mean flexion contracture of knee (°) (SD, range)	1.8 (5.1, -5 to 30)
Mean unilateral popliteal angle (°) (SD, range)	42.4 (15.9, 10 to 90)
Mean bilateral popliteal angle (°) (SD, range)	30.6 (14.6, 0 to 85)

GMFCS: gross motor function classification system; SD: standard deviation.

Table 2. Odd Ratios of Risk Factors of Anterior Knee Pain While Walking

Risk factor	Crude ORs (95% CI)	<i>p</i> -value	Adjusted ORs (95% CI)	<i>p</i> -value
Age	1.07 (1.02–1.14)	0.013	1.08 (1.02–1.14)	0.008
Gender (male/female)	1.18 (0.26–5.26)	0.828	-	-
FC	0.95 (0.85–1.07)	0.403	-	-
PA (U)	1.00 (0.94–1.07)	0.983	-	-
PA (B)	1.01 (0.93–1.09)	0.804	-	-
Patella alta	1.00 (0.20–5.16)	0.997	1.34 (0.26–6.80)	0.723

OR: odds ratio, CI: confidence interval, FC: knee flexion contracture, PA (U): unilateral popliteal angle, PA (B): bilateral popliteal angle.

Table 3. Odd Ratios of Risk Factors of Anterior Knee Pain While Resting

Risk factor	Crude ORs (95% CI)	<i>p</i> -value	Adjusted ORs (95% CI)	<i>p</i> -value
Age	1.08 (1.02–1.14)	0.008	1.09 (1.03–1.15)	0.004
Gender (male/female)	0.66 (0.12–3.71)	0.637	-	-
FC	0.94 (0.88–1.01)	0.078	0.91 (0.82–1.00)	0.055
PA (U)	1.00 (0.94–1.06)	0.977	-	-
PA (B)	0.99 (0.94–1.05)	0.859	-	-
Patella alta	1.36 (0.40–4.66)	0.622	1.87 (0.44–8.05)	0.399

OR: odds ratio, CI: confidence interval, FC: knee flexion contracture, PA (U): unilateral popliteal angle, PA (B): bilateral popliteal angle.

Table 4. Odd Ratios of Risk Factors for Provocative Anterior Knee Pain

Risk factor	Crude ORs (95% CI)	<i>p</i> -value	Adjusted ORs (95% CI)	<i>p</i> -value
Age	1.04 (0.99–1.10)	0.131	1.05 (1.00–1.10)	0.066
Gender (male/female)	0.60 (0.15–2.44)	0.479	-	-
FC	0.93 (0.87–0.99)	0.024	0.92 (0.85–0.98)	0.015
PA (U)	0.99 (0.95–1.04)	0.670	-	-
PA (B)	1.01 (0.97–1.06)	0.611	-	-
Patella alta	1.76 (0.82–3.78)	0.150	2.10 (0.86–5.13)	0.105

OR: odds ratio, CI: confidence interval, FC: knee flexion contracture, PA (U): unilateral popliteal angle, PA (B): bilateral popliteal angle.

DISCUSSION

Approximately 12.6% of ambulatory children and young adults with spastic cerebral palsy were found to have anterior knee pain in our hospital-based cohort study. Age was found to be a significant risk factor for anterior knee pain while walking and resting. Although knee flexion contracture was found to be a significant protective factor

for provocative anterior knee pain, its clinical implication was not clarified because of the small difference in knee flexion contracture between the patients with and without pain (approximately 1.5°).

Before discussing the clinical implications of the results of this study, its limitations should be mentioned. First, this was a hospital-based cohort study, and the patients included were children and young adults. Therefore,

care should be taken while generalizing our results to patients with cerebral palsy. Second, patients with previous surgeries affecting knee biomechanics were excluded, because it was believed that these patients were more likely to have anterior knee pain. Therefore, this exclusion may have caused a selection bias. Third, age was found to be the most important factor for the development of anterior knee pain in patients with cerebral palsy. Therefore, a longitudinal study with a long-term follow-up is warranted to investigate the factors that contribute to anterior knee pain.

In our study population, 60.6% of patients had patella alta according to the radiographic measurements. The presence of patella alta is believed to cause weakness of the knee extensor apparatus by diminishing the lever arm function,¹⁷⁾ and it may also be importantly associated with various knee problems. Therefore, patella alta needs to be defined and measured appropriately. A recent study showed that the methods commonly used to measure the patellar height are not applicable to children and adolescents due to the different degrees of ossification of bony landmarks.¹⁴⁾ In the present study, patellar height was measured using two different methods (IS and KS) rather than using a single uniform method considering the applicability of the methods. Therefore, the definition of patella alta used in this study is believed to be more accurate than that used in previous studies.^{17,18)} In addition, the GEE model considering the correlation between the right and left knee within a subject is believed to support the study results.

In the present study, anterior knee pain was evaluated in terms of walking pain, resting pain, and provocative pain. It was interesting to note that the significant risk factors differed slightly according to these three pain characteristics. The most important risk factor for walking and resting anterior knee pain was the patient age. In view of the study design and results, walking and resting anterior knee pain appear to be subjective symptoms that cannot be resolved by treatment. Provocative anterior knee pain has more objective characteristics, but no treatable risk

factors were identified in children and adolescents with cerebral palsy.

In the present study, six patients had abnormal radiographic findings of the knee extensor apparatus other than patella alta, including inferior pole fragmentation in two cases, ossicles on the tibial tuberosity in two cases, superior pole fragmentation in one case, and elongated patella in one case. However, none of these patients had anterior knee pain. This finding is somewhat at odds with a previous study, in which abnormal radiographics were found among patients with intractable anterior knee pain.¹⁹⁾ Another study reported that the radiographic findings were usually chronic in patients with cerebral palsy, and required no treatment,¹⁸⁾ which concurs with the findings of this study. Therefore, a further study with a longitudinal design including comprehensive risk factors is needed to investigate the risk factors for anterior knee pain in patients with cerebral palsy.

Although this study included a relatively large number of patients and data were analyzed using the GEE model, only children and adolescents were included. Furthermore, because a small number of patients with anterior knee pain were included, the risk factors for anterior knee pain identified in this study may not be generalizable. A longitudinal study that includes older patients and investigates candidate risk factors, such as torsional alignment of the limb, postural and rotational position of the foot, and gait parameters, more comprehensively is needed.

CONFLICT OF INTEREST

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

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