Risk Factors for the Occurrence and Protraction of Anterior Knee Pain in Children and Adolescents: a Prospective Cohort Study of 3 Years

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Research Article

Keywords: anterior knee pain, children and adolescents, physical activity

Posted Date: December 13th, 2021

DOI: https://doi.org/10.21203/rs.3.rs-1095827/v1

Abstract

Background:

Anterior knee pain (AKP) is a common limitation to children's participation in social and physical activities. Therefore, to prevent the occurrence and protraction of AKP, it is crucial to identify risk factors. The purpose of this study was to clarify the factors associated with the occurrence and protraction of AKP in children and adolescents.

Method:

A three-year prospective cohort study was conducted with children and adolescents aged 8–14 in Japan. We recorded the occurrence of AKP, heel buttock distance, straight leg raising angle (SLRA), dorsiflexion angle of the ankle joint, and the Hospital for Special Surgery Pediatric Functional Activity Brief Scale (HSS Pedi-FABS). Logistic regression analysis was performed to calculate the odds ratio (OR) for each predicted risk factor for the occurrence and protraction of AKP among subjects without AKP at baseline.

Results:

We recruited 1,254 children and adolescents for the present study, and 1,133 children and adolescents who did not have AKP at baseline were included in the analysis. Six to nine percent of the subjects developed AKP annually. A high HSS Pedi-FABS score significantly predicted AKP occurrence (in 2017, OR 1.07, 95% CI 1.02–1.12, p = 0.003; in 2018, OR 1.05, 95% CI 1.01–1.10, p = 0.025). Of the participants, 32.9% developed chronic AKP during the follow-up period. When 8-year-old was used as a reference age, 13-year-old subjects (right side, OR 2.37, 95% CI, 1.00–5.61, p = 0.05) and 14-year-old subjects (right side, OR 2.57, 95% CI, 1.00–6.60, p = 0.049; left side, OR 6.32; 95% CI 1.33–30.00, p = 0.020) were at a significantly higher risk of AKP protraction.

Conclusions:

This study showed that a greater physical activity level was a risk factor for the onset of anterior knee pain in childhood. In addition, one-third of the children and adolescents developed chronic knee pain, and elderly adolescents were at a higher risk of protraction.

1. Background

The knee is one of the most common sites of musculoskeletal pain in children and adolescents (1). A cross-sectional study reported a 23% prevalence of knee pain among adolescents aged 10–17 (2). Anterior knee pain (AKP), as typified by Osgood-Schlatter disease (OSD), is a common phenotype in children and adolescents and is usually considered benign and self-limiting (3)(4)(5). Rest, medication,

and physical therapy are typically successful treatments in more than 90% of patients with OSD (6). However, some children experience severe pain, leading them to change sports or limit physical activity and social participation due to chronic knee pain (7). Furthermore, AKP persists even after physeal closure in some cases, ultimately requiring surgery (8). Prevention of AKP occurrence and protraction is desirable; therefore, it is essential to recognize the characteristics of children and adolescents who are likely to develop AKP and those who are likely to have chronic AKP.

Studies have reported several factors to be associated with AKP in children and adolescents. Studies have reported that excess weight is correlated with chronic knee pain (9). One report showed that a higher body mass index increased knee pain and knee joint dysfunction (10). Another study reported lower extremity muscle tightness as an accurate risk factor for knee pain (11)(12). In Denmark, adolescents with knee pain had significantly higher levels of participation in leisure sports than those without knee pain (13). However, since the previous reports are chiefly based on retrospective or cross-sectional studies, it remains unclear whether these factors are the cause or the result of persistent AKP. To date, there are few reports based on adult studies that have investigated the causes of chronic knee pain, and clinicians have little empirical evidence to inform clinical decisions and make recommendations to their patients.

The purpose of this study was to describe the incidence of AKP in Japanese children and adolescents aged 8–14 and to identify the risk factors associated with its occurrence and protraction in children and adolescents.

2. Methods

2-1. study design

This study was approved by the Institutional Review Board and was conducted as a prospective cohort study. The surveillance started in April 2016, and based on the collected preliminary data, the data collection was modified from April 2017. Thus, the data used for the analysis in the current study were from April 2017 to February 2020.

Students who were enrolled in the 3rd to 9th grades of elementary and junior high schools affiliated with Chiba University in April 2017 were eligible for enrollment in the study. Written informed consent was obtained from all participants and their guardians before participating in the study. None of the students skipped or repeated the grades during the study period. Participants with lower limb trauma at the time of baseline examination, musculoskeletal or neurological disorders that made it impossible to perform the physical examination alone or to walk independently, history of lower limb surgery, or those without baseline data regarding the presence of AKP, were excluded (Fig. 1).

2-2. Data collection

Data were collected at school under the supervision of teachers when required. Subject demographic data such as sex and age were recorded at the time of participation in this study. Their height and weight data were recorded in January, April, and August of each year.

At the beginning of each fiscal year in April, from 2017 to 2019, the participants underwent a direct musculoskeletal examination by orthopedic surgeons and physical therapists. Each examiner received three to four training sessions prior to the medical assessment for accurate and uniform evaluation. The examination was conducted by at least two examiners per subject to allow one examiner to record the measurement, while the other performed the test.

The presence of tenderness was examined by gentle palpation of the supra- and infra-patellar poles and the tibial tubercle in each knee (14). These three inspection sites were chosen as they are anatomically easy to define and are common sites of AKP in children— often diagnosed as OSD or Sinding-Larsen-Johansson disease (SLJD) (13). AKP was marked as positive if the subject felt pain to any degree by gentle palpation in at least one of the sites.

Three items were measured to assess lower limb tightness, as previously reported: 1) heel-buttock distance (HBD, cm) (11), 2) straight leg raising angle (SLRA, degree) (11), and 3) dorsiflexion angle of the ankle joint at the knee in extended position (DFA, degree) (15). With the subject in the prone position, the examiner measured the HBD by bending the subject's knees, individually, as far as possible until the examiner felt resistance. The distance from the heel to the buttock was measured using a standard ruler, and the distance was recorded in centimeters to the first decimal point. Thereafter, the subject was placed in a supine position, and each leg was raised with the knees extended to measure SLRA, individually. The angle of the inspection table with the femoral shaft was measured using a large custom-made protractor and recorded in one-degree increments. Finally, the maximum DFA was measured on each side with the knee extended by setting the stationary arm of the goniometer parallel to the fibular shaft and the movement arm parallel to the fifth metatarsal, and was recorded in one degree increments.

In addition to the direct examination, the participants were required to answer a self-reported questionnaire each month to collect data on the presence of AKP and the degree of physical activity. In this questionnaire, each participant palpated the three inspection sites of their own knees and recorded if they felt pain. The participants received instructions with photos of the inspection sites prior to the study. Physical activity was quantified using the Hospital for Special Surgery Pediatric Functional Activity Brief Scale (HSS Pedi-FABS) (16). The HSS Pedi-FABS is a patient-reported outcome measure, with eight validated items designed to quantify movement in children aged 10–18 years (17). The scale ranges from 0 to 30, depending on how physically active the respondents were during the past month.

2-3. Occurrence and protraction of AKP

Subjects without AKP at baseline were assessed for the occurrence and protraction of AKP. The occurrence of AKP was defined as the first time the subject indicated positive pain on either point of palpation. If AKP occurred in one knee after the occurrence of the other knee was recorded, the latter knee

was not included in the record as an occurrence of AKP. The left and right sides of the knee were recorded individually for each occurrence. If the patient had knee pain in both knees simultaneously, bilateral AKP was considered.

Protracted AKP was defined as when chronic pain that lasted for more than three months or recurrence after the pain had disappeared for more than three months was observed, according to a previous report (18).

2-4. Statistical Analysis

Summary statistics for baseline variables were created using frequencies and proportions for categorical data and mean and standard deviation (SD) for continuous variables.

To analyze the occurrence of AKP in each knee, we used a multivariable nominal logistic regression analysis to calculate the odds ratio (OR) and investigated the effects of each factor. The analysis excluded the occurrence of bilateral AKP. The model was adjusted for HBD, SLRA, and HSS Pedi-FABS. Covariates for adjustment were selected based on clinical significance and previous studies. The analysis was performed for each fiscal year, and subjects with positive AKP were excluded from the analysis for the next fiscal year.

In the analysis of chronic AKP on each side, the odds ratio (OR) was calculated using the generalized estimating equations for the multivariable logistic regression model, and the effect of each factor was investigated. The model was adjusted for age, HBD, SLRA, DFA, and HSS Pedi-FABS. Covariates for adjustment were selected based on clinical significance and previous studies (6). The influence of age was analyzed by comparing each age group from 9 to 14 years, against 8 years as the reference. HBD was analyzed by classifying the participants based on whether their heel could touch their buttock, creating two groups: the heel contact group (HBD = 0mm) and the non-contact group (HBD > 0mm).

All p-values were two-sided. Statistical significance was set at $p < 0.05$. All statistical analyses were performed using SAS software (version 9.4; SAS Institute, Cary, NC, USA).

3. Results

We included 1,254 students and excluded 32 students who were unable to collect data on AKP at baseline (Figure 1). Of the 1,222 students, 1,133 did not have AKP at baseline. Table 1 shows the characteristics of the subjects sorted by the presence of AKP at baseline.

Table 1
Characteristics of the subjects in this study

	Total (n = 1222)	AKP (-) (n = 1133)	AKP (+) (n = 89)
Age (y)			
8	305 (25.0)	292 (25.8)	13 (14.6)
9	110 (9.0)	106 (9.4)	4 (4.5)
10	122 (9.9)	109 (9.5)	13 (14.6)
11	110 (9.0)	102 (9.0)	8 (9.0)
12	282 (23.1)	262 (23.1)	20 (22.5)
13	150 (12.3)	134 (11.8)	16 (18.0)
14	143 (11.7)	128 (11.3)	15 (16.9)
Sex			
Girls	613 (50.2)	582 (51.4)	31 (34.8)
Boys	609 (49.8)	551 (48.6)	58 (65.2)
BMI (Kg/m^2)	17.4 ± 2.5	17.4 ± 2.6	17.4 ± 2.5
Lower limb tightness			
Rt HBD			
Contact group	920 (75.5)	852 (75.5)	68 (76.4)
Non-contact group	298 (24.5)	277 (24.5)	21 (23.6)
Lt HBD			
Contact group	917 (75.3)	851 (75.4)	66 (74.2)
Non-contact group	301 (24.7)	278 (24.6)	23 (25.8)
Rt SLRA (deg)	76.6 ± 13.3	76.8 ± 13.1	74.0 ± 14.7
Lt SLRA (deg)	76.6 ± 13.5	76.9 ± 13.3	73.5 ± 15.3

Data on age and sex are presented as number (prevalence), while BMI, SLRA, DFA, and HSS Pedi-FABS are presented as mean ± standard deviation

AKP, anterior knee pain; BMI, body mass index; HBD, heel buttock distance; SLRA, straight leg raising angle; DFA, dorsiflexion angle of the ankle joint with knee extension;

HSS Pedi-FABS, The Hospital for Special Surgery Pediatric Functional Activity Brief Scale

	Total (n = 1222)	AKP (-) (n = 1133)	AKP (+) (n = 89)
Rt DFA (deg)	15.6 ± 6.9	15.7 ± 6.9	14.0 ± 7.0
Lt DFA (deg)	15.3 ± 6.8	15.3 ± 6.8	14.1 ± 7.1
HSS Pedi-FABS	15.5 ± 8.1	15.5 ± 8.1	15.2 ± 8.7
Data on age and sex are presented as number (prevalence), while BMI, SLRA, DFA, and HSS Pedi-FABS are presented as mean ± standard deviation			
AKP, anterior knee pain; BMI, body mass index; HBD, heel buttock distance; SLRA, straight leg raising angle; DFA, dorsiflexion angle of the ankle joint with knee extension;			
HSS Pedi-FABS, The Hospital for Special Surgery Pediatric Functional Activity Brief Scale			

The prevalence of AKP ranged from 5.7–8.9% (Table 2). Since the minimum number of subjects who newly developed AKP between 2017 and 2019 was 69, the number of explanatory variables was set to six. Statistical analysis was performed using the HBD, SLRA, and HSS-Pedi FABS. Table 3 shows the OR and confidence interval (CI) for each factor at the annual baseline for the occurrence of AKP. HSS Pedi-FABS was a significant risk factor of left AKP (in 2017, OR 1.07, 95% CI 1.02–1.12, p = 0.003; in 2018, OR 1.05, 95% CI 1.01–1.10, p = 0.025). In addition, SLRA was also a significant predictor for the occurrence of right AKP (right SLRA, OR 0.47, 95% CI 0.23–0.97, p=0.041) and left AKP (right SLRA, OR 0.38, 95% CI 0.20–0.76, p = 0.006; left SLRA, OR 2.36, 95% CI 1.20–4.65, p = 0.013) in 2018. Since the OR of AKP was less than 1 when the SLRA increased, it could be interpreted as the decrease in SLRA was a risk factor of AKP occurrence; that is, the tightness of the hamstrings was a risk factor for developing AKP.

Table 2
The number of new-onset AKP for each year

	2017 (n = 770)	2018 (n = 543)	2019 (n = 436)
No AKP	371 (48.2)	261 (48.1)	169 (38.8)
Rt AKP (+)	44 (5.7)	34 (6.3)	39 (8.9)
Lt AKP (+)	44 (5.7)	41 (7.6)	30 (6.9)
Bi AKP (+)	38 (4.9)	16 (2.9)	32 (7.3)
No data of AKP	273 (35.5)	191 (35.2)	166 (38.1)
Data was presented as number (prevalence)			
AKP, anterior knee pain; Rt, right; Lt, left; Bi, bilateral			

Of the 322 subjects with newly developed AKP, 106 developed protracted AKP during the study period. Table 4 shows the ORs and CIs of each factor for AKP protraction. When 8-year-old was used as a reference age, 13-year-old subjects (right AKP, OR 2.37, 95% CI, 1.00–5.61, p = 0.05) and 14-year-old subjects (right AKP, OR 2.57, 95% CI, 1.00–6.60, p = 0.049; left AKP, OR 6.32; 95% CI 1.33–30.00, p = 0.02) were at a significantly higher risk of AKP protraction. In addition, DFA was a significant predictor of left AKP protraction (right DFA, OR 1.56, 95% CI 1.13–2.16, p = 0.007; left DFA, OR 0.62; 95% CI, 0.45–0.86; p = 0.004). Since the OR of AKP was less than 1 when the DFA increased, it could be interpreted as the decrease in DFA was a risk factor of AKP protraction; that is, the tightness of the gastrocnemius muscle was a risk factor for chronic AKP. Furthermore, HSS Pedi-FABS was also a significant predictor of AKP protraction (right AKP, OR 0.97, 95% CI, 0.95–1.00, p = 0.042; left AKP, OR 0.96; 95% CI 0.94–0.99, p = 0.012).

4. Discussion

The most important finding of our study was that children and adolescents aged 8–14 years, with higher levels of physical activity have a generally higher risk of developing AKP. In addition, the prevalence of chronic knee pain within three years was 32.9% in children and adolescents aged 8–14 years, and older adolescents were at a higher risk of protracted AKP than younger children. These results suggest that AKP occurrence may be prevented by controlling the amount of physical activities and that more care should be taken in the treatment of older adolescents who have AKP.

Most previous studies examined non-specific knee pain in children and adolescents, or young athletes with AKP, and compared the characteristics and sports activity levels between subjects with and without knee pain (12)(19). Our study is unique in that we focused on children and adolescents without AKP at baseline and conducted a longitudinal study to investigate the risk factors for the occurrence and protraction of AKP.

There have been several studies on the risk factors for AKP in children and adolescents. Some researchers have reported that quadriceps tightness is associated with AKP (11)(12). A cross-sectional study compared lower limb tightness in adolescents with and without OSD and revealed that HBD was significantly higher in those with OSD. However, this study found no differences in SLRA (12). Our study showed that there was no obvious association between the new onset of AKP and HBD. There was an association between SLRA and the occurrence of AKP in one of the analyzed years. The difference between the current study and the previous report may be that the previous study focused only on athletic adolescents, and even the non-OSD subjects had tighter lower limbs—with a mean HBD of 6 cm and SLRA of 71 degrees—compared to our study subjects (mean HBD 1.3 cm, mean SLRA 77 degrees). Other researchers showed the association between AKP and physical activity (20)(21). In a cross-sectional study, Tomaru et al. showed that the longer the exercise time, the higher the proportion of OSD in elementary and junior high school students (21). High HSS Pedi-FABS was a significant risk for AKP occurrence in our study. OSD caused repetitive traction at apophysis in tibial tuberosity (6). This

suggested that AKP was induced by inflammation in the knee as a result of increased load on the knee components due to excessive exercise.

The prevalence of chronic knee pain in children and adolescents has been reported to be 31–40%, with a follow-up period of 1–5 years (22)(23). In our study 32.9% of Japanese children and adolescents suffered from chronic AKP, which is in line with results from other cohorts. Some previous reports have studied the risk factors for chronic knee pain in children and adolescents. In a cross-sectional study including 967 children and adolescents in Finland, the relationship between age, sex, weight, and the frequency of chronic knee pain was investigated (24). The investigators concluded that adolescents aged 14 or 15 had more chronic knee pain than children aged 9 or 10, and more than half of the subjects with chronic knee pain participated in some form of sport. A prospective cohort study of risk factors for persistent knee pain, including 768 adolescents between 12 and 15 years, found that a high level of sports participation was a risk factor for chronic knee pain (19). Our study showed that higher age was a risk factor for AKP protraction. This may be due to the physical development stage, but another possibility is how the adolescents deal with their physical problem. It has been reported that adolescents tended to choose inadequate coping strategies for knee pain (25)(26). In many cases, knee pain is bearable and adolescents would try to continue their athletic activities. We assume that elder subjects in our study refused or were not allowed to stop their activities at the beginning of AKP, thus leading to protraction of the pain. However, our data also showed that low sports participation levels correlated with AKP protraction. The 14-year-old subjects in this study were 9th-grade students. Most of the athletic students in this grade had their final competition in early summer before the preparation period for high school admission exams. Therefore, the 14-year-old subjects were highly athletic until summer, and their physical activity level was greatly reduced over the following two-thirds of the year. This may be the reason for the protraction observed in elder subjects and lower physical activity at the same time.

The strength of this study is that it was a prospective cohort study, and a larger number of subjects than in previous reports were included. In addition, collecting knee pain data every month likely incurs less recall bias than annual studies. However, this study has some limitations. First, although we evaluated knee pain based on tenderness—assumed to be a common point of tenderness observed in OSD and SLJD (6)—we were not able to reach a definite diagnosis. The possibility of other underlying diseases causing AKP could not be ruled out. Second, the proportion of missing AKP data was relatively high with 35.2–38.1% of subjects lacking data on AKP at least once during the follow-up period (Table 2). This rate was calculated by omitting subjects with more than one instance of missing data, which may be a strict cut-off for children and adolescents. The rate of missing data count to the total data count was only 7.1%. Therefore, the results may be biased in performing sensitivity analyses for the occurrence and protraction of AKP. We also performed a complementation analysis by referring to the previous month of missing data. Complementation was considered impossible if missing data continued for more than two months in a row. The results are shown in Appendices 1 and 2 and are similar to the initial analysis; however, no correlation was observed between DFA and AKP protraction. The association between the protraction of AKP and DFA is unclear. Third, this study was conducted in a single public school, thus

causing some bias in the results. Finally, we do not know if the subjects had a history of temporal knee pain before participating in the study.

5. Conclusions

We conducted a prospective cohort study to investigate the risk factors for the occurrence and protraction of AKP in Japanese children and adolescents aged 8–14. AKP occurred in 5.7–8.9% of children and adolescents each year, and high level of physical activity was found to be a risk factor for the occurrence of AKP. In addition, 32.9% had chronic knee pain within three years, and elderly adolescents were at higher risk of protracted AKP than children.

List Of Abbreviations

anterior knee pain, AKP

Osgood-Schlatter disease, OSD

Sinding-Larsen-Johansson disease, SLJD

heel buttock distance, HBD

straight leg raising angle, SLRA

dorsiflexion angle of the ankle joint with knee extension, DFA

The Hospital for Special Surgery Pediatric Functional Activity Brief Scale, HSS Pedi-FABS

standard deviations, SD

Odds Ratio, OR

confidence interval, CI

Declarations

 Ethics approval and consent to participate

The Ethics Committee approved this study of the Graduate School of Medicine Chiba University (IRB no 2297).

Informed consent was obtained from the participating children or their guardians. All study methods were carried out according to the relevant guidelines and regulations.

 Consent of publication

Not applicable

 Availability of data and materials

Requests for data not shown in the body of this manuscript can be made to the corresponding author.

 Competing interest

The authors declare that they have no competing interests.

 Funding

Not applicable

 Authors' contributions

RA helped in the collection, analysis, and interpretation of data, the conception and design of the study, drafting the article, and critically revising the final approval of the version to be submitted. ST helped in the statistical analysis and design of the study. SW helped in the data collection. YO helped with data collection. SK helped with data collection. SY helped in the conception and design of this study. SO helped in the conception and design of the study. TS helped in the conception and design of the study.

Therefore, all authors certify that they have participated sufficiently in taking public responsibility for the content. All authors read and approved the final manuscript.

 Acknowledgment

The authors appreciate all the assistance and cooperation from the teachers and school nurses of Chiba University Elementary and Secondary School, physical therapists for their support in data collection, and Yumi Akagi for data management.

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Figures

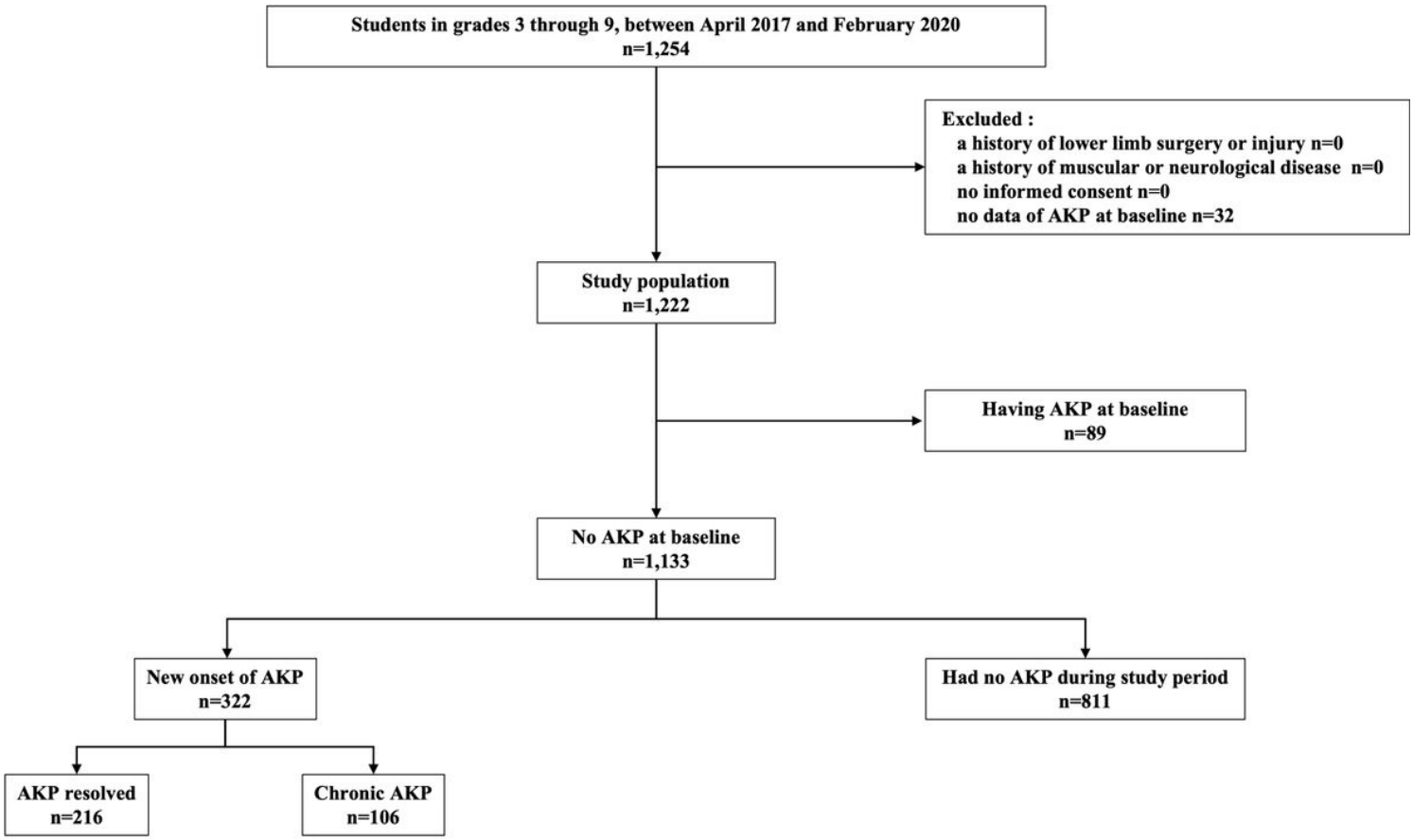

Figure 1

Flow diagram of this study

Supplementary Files

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