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Research article

The prevalence and risk factors of pubic bone marrow edema in femoroacetabular impingement and hip dysplasia

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ABSTRACT

Femoroacetabular impingement syndrome (FAIS) has been associated with osteitis pubis; however, it is still unclear whether hip dysplasia is associated with osteitis pubis. This study aimed to investigate (i) the incidence of pubic bone marrow edema (BME) on magnetic resonance imaging in symptomatic patients with FAIS, borderline developmental dysplasia of the hip (BDDH) and developmental dysplasia of the hip (DDH) undergoing hip arthroscopic surgery with labral preservation and (ii) the demographic and radiographic factors associated with pubic BME. A total of 259 symptomatic patients undergoing hip arthroscopic surgery between July 2016 and April 2019 were retrospectively reviewed and divided into three groups: FAIS (180 patients), BDDH (29 patients) and DDH (50 patients). Diffuse changes in the pubic bone adjacent to the pubic symphysis were labeled pubic BME, and the prevalence of their occurrence was examined. Multivariate logistic regression analysis was performed to identify factors involved in pubic BME, and odds ratios (ORs) for relevant factors were calculated. There was no significant difference in the prevalence of pubic BME among the three groups (20 [11.1%] of 180 FAIS patients, 6 [20.6%] of 29 BDDH patients and 7 [14%] of 50 DDH patients, $P = 0.325$). Multivariate logistic regression analysis showed that acetabular coverage was not associated with pubic BME, whereas younger age and greater alpha angle were still independent associated factors [age ≤26 years (OR, 65.7) and alpha angle ≥73.5° (OR, 4.79)]. Determining the possible association of osteitis pubis with cam impingement in dysplastic hips may provide insights toward a more accurate understanding of its pathophysiology.

INTRODUCTION

Osteitis pubis is an overuse syndrome characterized by pain and tenderness from the groin to the pubic symphysis caused by exercise [1]. Repeated traction forces caused by the rectus abdominis and adductor muscles attached to the pubic bone cause loading and instability of the pubic symphysis [2–5]. The cam type of femoroacetabular impingement syndrome (FAIS) is reported to be a risk factor for sports-related osteitis pubis [6–8]. Larson *et al.* [9] reported that 75% of American football players had the cam type of FAIS, and 53.6% demonstrated characteristic radiographic findings for osteitis pubis. Phillips *et al.* [10] also reported that osteitis pubis is 5.26 times more likely to occur with the cam type of FAIS than without it. Birmingham *et al.* [11] stated in a cadaver study that the direct force exerted by the impingement between the acetabulum and cam lesion can cause pelvic shear stress, leading to rotational instability of the pubic symphysis and resulting in osteitis pubis. Thus, the intra-articular lesion associated with cam deformity is thought to play a major factor in the onset of osteitis pubis.

In recent years, several studies have shown that a subset of patients with hip dysplasia, such as borderline developmental dysplasia of the hip (BDDH) and developmental dysplasia of the hip (DDH), have cam deformities [12–17]. Therefore, it has been suggested that osteitis pubis may occur in dysplastic hips as well as in FAIS. However, the recent literature has not provided clarity on the frequency and relevance of osteitis pubis in patients with dysplastic hips. It would be useful to know the incidence of osteitis pubis in FAIS, BDDH and DDH and the factors that are associated with these issues to facilitate a more detailed understanding of its pathophysiology.

We hypothesized that osteitis pubis is independent of acetabular morphology but correlates with femoral impingement-prone bone morphology and high levels of sport activity. This study aimed to investigate (i) the incidence of pubic bone marrow edema (BME) on magnetic resonance imaging (MRI) in symptomatic patients with FAIS, BDDH and DDH undergoing hip arthroscopic surgery with labral preservation and (ii) the demographic and radiographic factors associated

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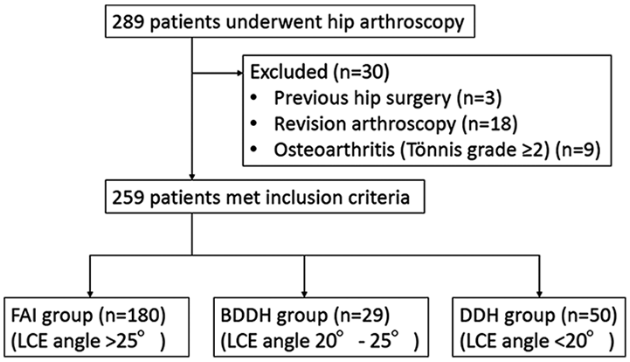

Fig. 1. Patient selection flowchart. Hip arthroscopic surgery patients remaining after application of the exclusion criteria were divided into three groups according to their LCEAs.

with pubic BME in symptomatic patients undergoing hip arthroscopy.

MATERIALS AND METHODS

This retrospective study included 289 consecutive symptomatic patients undergoing hip arthroscopic surgery with labral preservation who were diagnosed with FAIS, BDDH or DDH by a single surgeon (S.U.) between July 2016 and April 2019. The local institutional review board approved the study (Wakamatsu Hospital of University of Occupational and Environmental Health; authorization number H29-220), and all study participants provided informed consent. The indications for hip arthroscopic surgery were persistent groin pain, positive provocative maneuvers, refractory response to nonsurgical treatment, and acetabular labral tearing as detected by 3-Tesla MRI (3T-MRI). The patients were divided into three groups according to the lateral center–edge angle (LCEA): the FAIS group (LCEA >25°), BDDH group (LCEA of 20–25°) and DDH group (LCEA <20°). The diagnosis of FAIS was based on hip pain, a positive flexion adduction internal rotation (FADIR) test, and pincer or cam deformity. Radiographic evidence of a pincer deformity was defined as LCEA ≥40° or acetabular roof obliquity (ARO) ≤0°, and cam deformity was defined as a maximum alpha angle ≥55°. The diagnosis of BDDH and DDH was based on hip pain and a positive FADIR test. Revision cases and cases with arthritic changes of Tönnis Grade 2 or higher were excluded, resulting in 259 included patients: FAIS group ($n = 180$, male/female = 111/69, mean age 34.9 years), BDDH group ($n = 29$, male/female = 11/18, mean age 33.8 years) and DDH group ($n = 50$, male/female = 13/37, mean age 28.7 years) (Fig. 1).

Radiographic evaluation

We assessed the radiographs and computed tomography (CT) images at a PACS workstation to determine which radiographic parameters were predictors of pubic BME. The preoperative radiographic images, including the anteroposterior pelvis view

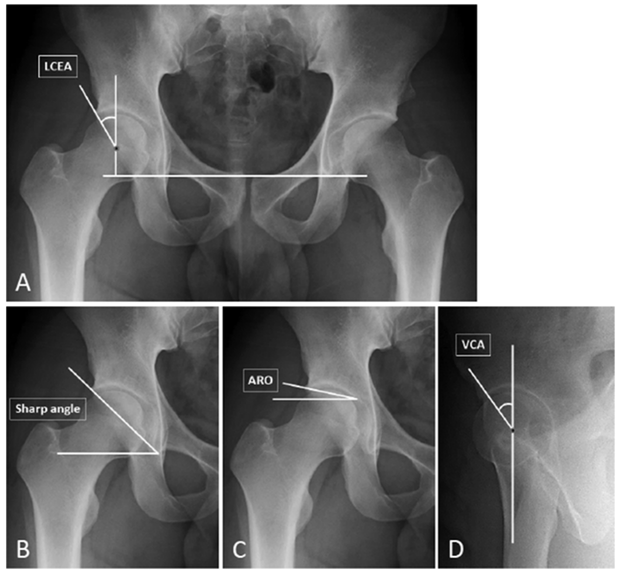

Fig. 2. Measurement of each parameter in the anteroposterior pelvic view (A–C) and the false-profile view (D). (A) LCEA: the angle between the line connecting the center of the bone head and the lateral edge of the acetabulum and the line perpendicular to the line connecting the bilateral teardrop lines. (B) Sharp angle: the angle between the line connecting the inferior margin of the teardrop and the lateral margin of the acetabulum and the tear drop line. (C) ARO: the angle between the line connecting the inner and outer scleral edges of the acetabular loading zone and the tear drop line. (D) VCA angle: the angle between the line connecting the center of the femoral head to the anterior margin of the sclerotic zone of the acetabular loading area and the perpendicular line to the horizontal line.

in the supine position, false-profile view and Dunn view (flexion 45°; abduction 45°), and preoperative CT scans (SOMATOM Sensation 16; Siemens Medical Solutions, Erlangen, Germany) were taken with a 2-mm slice thickness including the area from the pelvis to the knee in all patients. The LCEA, Sharp angle and ARO were measured on the anteroposterior pelvis view images (Fig. 2A–C), and the vertical center anterior (VCA) angle was measured on the false-profile view images (Fig. 2D). Alpha angles were measured on the Dunn view images according to the methods described by Nötzli *et al.* [18] (Fig. 3). Femoral neck anteversion (FNA) was assessed by measuring the angle of FNA based on the posterior condylar axis of the femoral condyle on CT axial images. Evaluation of the pubic symphysis was performed using 3T MRI (Signa EXCITE HD; GE Healthcare, Chicago, IL, USA) images taken preoperatively. We assessed the presence of pubic BME in terms of the T2 fat suppression (T2 FS) in the coronal image (repetition time (TR)/echo time (TE), 4140/91; matrix, 360 × 448; slice thickness, 4.0 mm; field of view (FOV), 360 × 360 mm) and the short TI inversion recovery in the axial image (TR/TE, 4600/67; matrix, 360 × 320; slice thickness, 4.0 mm; FOV, 360 × 360 mm), based on a study by Branci *et al.* [19], and defined an involved region ≥1 cm (T2 FS in the coronal image) as a positive finding of the presence of pubic BME [20] (Fig. 4). Two examiners (H.S. and Y.M., each with >10 years of experience in orthopedic surgery) evaluated each parameter and the

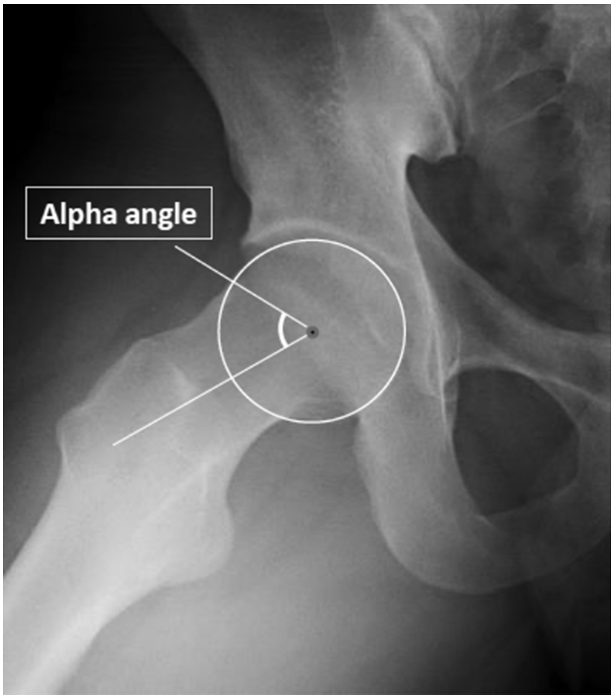

Fig. 3. Alpha angle in the Dunn view (flexion, 45°; abduction, 45°): the angle between the line passing through the center of the femoral head and the center of straightest portion of the neck and the line connecting the point where the anterior margin of the neck protrudes from the circle indicating the femoral head and the center of the femoral head.

presence of pubic BME twice at 1-month intervals, without disclosing the clinical findings or imaging reports. Patient characteristics were defined as age at surgery, sex and degree of sporting activity. The degree of sporting activity was divided into four levels based on the Hip Sports Activity Scale (HSAS) described by Naal *et al.* [21]: low activity (Grade 0–2), moderate activity (Grade 3–4), high activity (Grade 5–6) and very high activity (Grade 7–8).

Statistical analysis

Intraclass correlation coefficients and corresponding 95% confidence intervals (CIs) were calculated to quantify the inter- and intraobserver reliability of each radiographic parameter and the evaluation of pubic BME.

First, each image parameter was compared among the three groups using ANOVA, and the frequency of occurrence of pubic BME was compared using Fisher's exact test. Next, we examined whether there were differences in patient characteristics and imaging parameters in the presence or absence of pubic BME. Fisher's exact test was used to compare sex, HSAS grade and pubic BME, and other parameters were compared using the Mann–Whitney U test. In addition, multivariate logistic regression analysis with the presence of pubic BME as the dependent variable and patient characteristics and radiographic parameters as independent variables was performed to identify the factors involved. We then calculated the area under the curve (AUC) and identified the cutoff value for each factor by receiver operating characteristic (ROC) curve analysis. Finally, the stratified incidence of relevant factors was examined, and ORs were calculated from the cutoff values. Statistical analysis was performed with SPSS (version 26.0; IBM, Armonk, NY, USA), and a P value of <0.05 was considered significant. We performed *post hoc* analysis to determine whether the sample size was sufficient to compare the frequency of pubic BME among the three groups.

RESULTS

The intra- and interobserver reliability of each radiographic parameter and the determination of pubic BME were in substantial to almost-perfect agreement (Table I). *Post hoc* analysis revealed an effect size (Cramer's V) of 0.28 for the frequency of occurrence of pubic BME among the three groups, an alpha level of 0.05, a total sample size of 259 and 2 degrees of freedom, resulting in a total power of 0.98, which indicates the adequacy of the sample size in this study.

A comparison of radiographic parameters among the FAIS, BDDH and DDH groups showed significant differences in demographic and acetabular dysplastic parameters (LCEA, Sharp angle, ARO and VCA). There was no difference in the alpha angle among the three groups, nor was there a difference in the percentage above 55°. Pubic BME was found in 20 patients (11.1%) with FAIS, 6 (20.6%) with BDDH and 7 (14%) with DDH, with no significant difference among the three groups ($P = 0.325$) (Table II). In comparisons according to the presence or absence of pubic BME, pubic BME was associated with younger age, male sex, higher HSAS grade and less FNA than no pubic BME, while there was no significant difference in LCEA, Sharp angle, ARO or VCA (Table III). Multivariate logistic regression analysis showed that sex, sports activity, femoral neck version and acetabular coverage were not factors associated with pubic BME, whereas younger age and a greater alpha angle were still independent associated factors (Table IV). ROC curve analysis of the factors involved showed that the cutoff value of age was 26 years old (AUC, 0.867; 95% CI, 0.820–0.914; sensitivity, 67.3%; specificity, 97.0%; $P < 0.001$) and the alpha angle was 73.5° (AUC, 0.697; 95% CI, 0.594–0.799; sensitivity, 51.5%; specificity, 81.9%; $P < 0.001$) (Fig. 5). The incidence of pubic BME, stratified according to relevant factors, showed a tendency for younger age and greater alpha angle to increase the frequency of pubic BME [age ≤26 years (OR, 65.7; 95% CI, 8.81–490.4; $P < 0.0001$) and alpha angle ≥73.5° (OR, 4.79; 95% CI, 2.23–10.2; $P < 0.0001$)] (Fig. 6).

DISCUSSION

The main finding of our study was that pubic BME occurred to the same extent in both FAIS and hip dysplasia. Moreover, the degree of acetabular dysplasia was weakly associated with pubic BME, and younger patient age (≤26 years, 65.7 times) and greater alpha angle (≥73.5°, 4.79 times) were more likely to be associated with pubic BME. Regarding the validity of the measurements, nearly perfect agreement on the alpha angle was observed in terms of both intra- and interobserver reliability in

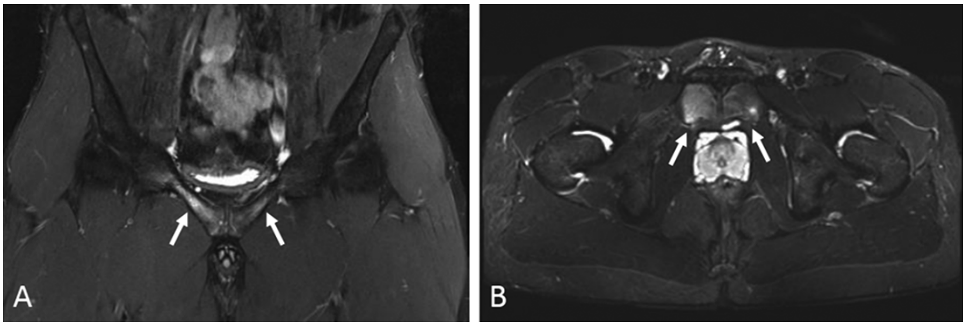

Fig. 4. The diffuse changes of the pubic bone adjacent to the pubic symphysis (white arrows) give rise to an abnormal signal in the T2 fat suppression image in the coronal image (A) and short T1 inversion recovery in the axial image (B).

Table I. Intra- and interobserver reliability of radiographic measurements

Measurement	*Intraobserver 1 (95% CI)*	*Intraobserver 2 (95% CI)*	*Interobserver (95% CI)*
LCEA	0.973 (0.899–0.993)	0.990 (0.961–0.997)	0.874 (0.687–0.950)
Sharp angle	0.895 (0.605–0.974)	0.838 (0.390–0.959)	0.799 (0.506–0.920)
ARO	0.975 (0.904–0.994)	0.997 (0.989–0.999)	0.937 (0.844–0.975)
VCA angle	0.886 (0.570–0.971)	0.984 (0.938–0.996)	0.847 (0.609–0.940)
Alpha angle	0.761 (0.151–0.940)	0.968 (0.883–0.992)	0.879 (0.665–0.954)
FNA	0.890 (0.712–0.959)	0.995 (0.982–0.999)	0.936 (0.773–0.978)
Pubic BME	1.000 (1.000–1.000)	0.871 (0.612–0.958)	0.932 (0.852–0.968)

this study, although some studies have found that interobserver reliability is lower than intraobserver reliability [22, 23]. We believe that our relatively high reliability is the result of conducting measurements using computer software (PACS workstation) and closely following the method of Nötzli *et al.* [18]. As some studies have reported similar levels of reliability to our study using similar methods [24, 25], the validity of our measurement of the alpha angle seems to be satisfactory.

Although pubic BME was expected to occur less frequently in dysplastic hips than in FAIS in this study due to the higher proportion of women and lower levels of sports activity, we found that pubic BME occurred as frequently in dysplastic hips as in FAIS. The multivariate logistic regression analysis showed that a greater alpha angle remained the final independent associated factor, rather than sex or sports activity, and there was no difference in the percentage above 55° among the three groups. This may be the reason why there was no difference in the occurrence of pubic BME among the three groups. The incidence of cam deformity in dysplastic hips varies widely in previous reports. Anderson *et al.* [12] reported cam deformity in 10% of dysplastic hips, whereas Clohisy *et al.* [13] reported it in 75% and Kraeutler *et al.* [15] reported it in as many as 96%, similar to the results of the present study. These studies, including the present one, included a small number of subjects, and imaging positions vary in the literature. For further validation of our results, a study with a large number of images taken in the same position is necessary.

Several studies have reported an association between the alpha angle and osteitis pubis [6, 7, 9, 10], which are consistent with our findings. On the other hand, the association between dysplasia and the pubic symphysis was outside the scope of our investigation. In addition, several studies have reported associations with extra-articular lesions other than the pubic symphysis. Jacobsen *et al.* [26] reported that approximately 50% of patients with dysplastic hips had pain associated with the iliopsoas and abductor muscle of the hip, and Moulton *et al.* [27] reported that an increased anterior opening angle of the acetabulum was associated with tendinitis of the gluteus maximus. These findings may be the result of increased torque in the muscles surrounding the hip joint; this increased torque is compensatory for the hip instability resulting from acetabular shallowness and a reduced loading area [28]. In the present study, the degree of dysplasia did not differ with respect to the presence or absence of pubic BME, suggesting that hip instability was not associated with pubic BME. Osteitis pubis is also known as overuse syndrome, which occurs in active young adult athletes. In this study, young age remained a relevant factor. We thought of reasons why youth might be relevant, such that the stability of the pubic symphysis is more immature at a younger age, making it more prone to instability and inflammation, but we could not find any literature to prove this. Although sports activity was not a significant independent factor in the multivariate analysis, it was close to the level of significance and may be associated to some extent.

There are several limitations to this study. First, the study was single center rather than multicenter, and it was limited to surgical cases. However, a sufficient number of cases were obtained for comparisons between groups and for multivariate analysis according to the power analysis. Second, the study included patients with groin pain but did not confirm that the pain was from an intra-articular rather than an extra-articular source in all cases. As reported by Kheterpal *et al.* [29], the subject group would be stronger if a hip source of symptoms was proven with an anesthetic exam. However, the study included patients undergoing hip arthroscopic surgery, and all had acetabular labral tearing. We do not believe that this limitation influenced the results. Third, there were differences in demographic data between groups, including age, sex and sporting activity levels.

Table II. Comparison of demographic and radiographic parameters among the FAIS, BDDH and DDH groups

	FAIS (n = 180)	*BDDH (n = 29)*	*DDH (n = 50)*	*P*
Age, years	34.9 ± 14.8 (14–74)	33.8 ± 15.7 (13–64)	28.7 ± 12.6 (14–54)	0.031
Sex, no.				<0.0001
Male	111 (61.7)	11 (37.9)	13 (26.0)	
Female	69 (38.3)	18 (62.0)	37 (74.0)	
HSAS				<0.0001
0–2	80 (44.4)	14 (48.3)	30 (60.0)	
3–4	41 (22.8)	10 (34.5)	13 (26.0)	
5–6	6 (3.3)	5 (17.2)	5 (10.0)	
7–8	53 (29.4)	0 (0)	2 (4.0)	
LCEA, degrees	31.5 ± 5.5 (26–49)	22.4 ± 1.1 (21–25)	15.5 ± 3.3 (4–20)	<0.0001
Sharp angle, degrees	41.0 ± 4.0 (31–54)	43.6 ± 2.8 (38–51)	46.4 ± 3.2 (37–55)	<0.0001
ARO, degrees	5.2 ± 4.6 (−13–17)	9.6 ± 4.2 (1–21)	15.7 ± 4.9 (4–26)	<0.0001
VCA angle, degrees	33.9 ± 7.9 (14–59)	24.7 ± 8.5 (6–46)	15.2 ± 8.1 (−15–32)	<0.0001
Alpha angle, degrees	65.6 ± 10.5 (41–86)	65.0 ± 9.0 (43–78)	65.4 ± 11.6 (39–88)	0.957
No. of >55°	153 (85.0%)	26 (89.6%)	41 (82%)	0.70
FNA, degrees	17.1 ± 11.8 (−13–50)	17.8 ± 10.8 (−1–38)	21.6 ± 12.6 (−8–46)	0.07
Pubic BME, no.	20 (11.1%)	6 (20.6%)	7 (14.0%)	0.325

Data are presented as a median (range) or no. (%).

Table III. Comparison of parameters by pubic BME status

	With pubic BME	*Without pubic BME*	*P*
No. of patients	33	226	
Age, years	18.4 ± 3.4 (13–29)	35.8 ± 14.4 (14–74)	<0.0001
Sex, no.			<0.0001
Male	27 (81.8)	108 (47.8)	
Female	6 (18.2)	118 (52.2)	
HSAS			<0.0001
0–2	4 (12.1)	120 (53.1)	
3–4	6 (18.2)	58 (25.7)	
5–6	4 (12.1)	12 (5.3)	
7–8	19 (57.6)	36 (15.9)	
LCEA, degrees	26.6 ± 8.3 (12–44)	27.5 ± 8.0 (4–49)	0.53
Sharp angle, degrees	42.6 ± 3.9 (35–52)	42.3 ± 4.3 (31–55)	0.69
ARO, degrees	7.6 ± 6.4 (0–22)	7.7 ± 6.2 (−13–26)	0.93
VCA angle, degrees	28.2 ± 11.7 (5–48)	29.4 ± 10.8 (−15–59)	0.53
Alpha angle	71.5 ± 10.9 (43–86)	64.6 ± 10.2 (39–88)	<0.0001
FNA	12.9 ± 13.4 (−12–36)	18.8 ± 11.6 (−13–50)	0.007

Data are presented as a median (range) or no. (%).

Table IV. Multivariate logistic regression model: risk factors for pubic BME

Risk factor	*Odds ratio (95% CI)*	*P*
Age, years	0.817 (0.733–0.911)	<0.0001
HSAS	1.206 (0.983–1.480)	0.072
Alpha angle, degrees	1.061 (1.017–1.108)	0.006

It is difficult to align demographic data across the three groups in general, since FAIS is generally more common in men with high levels of sporting activity [30], and DDH is more common in women, regardless of sporting activity level [31]. However, the results of the present study do not change the fact that DDH with a greater alpha angle might also potentially cause pubic BME. Fourth, we used MRI signal changes to evaluate osteitis pubis in this study, but the clinical and imaging findings may be divergent. In general, in osteitis pubis, early signal changes in the pubic branch from the peripubic symphysis are seen on MRI [32, 33]. However, this signal change is thought to occur before the onset of symptoms [34] and may also progress asymptomatically [19, 35]. Since many studies of the association between FAIS and osteitis pubis have considered MRI findings, including those of asymptomatic cases, superior in identifying osteitis pubis [6, 8–10, 36], we also used pubic BME on MRI to assess osteitis pubis in this study. We believe that this does not weaken the conclusions of the present study. Fifth, the assessors were not blinded to the hip joint parameters when assessing pubic BME, which may have resulted in information bias. However, the determination of pubic BME was clear, with good intra- and interobserver reliability; thus, the effect of the change seems to be small. Sixth, this study was a retrospective study, and sufficient information about the duration of the disease, clinical symptoms, examination findings and past sports history was unavailable. These items were not included in this study and could have been examined in more detail. However, the results of this study are unlikely to be invalidated. Seventh, we could not elucidate the mechanisms that greater alpha angle would be associated with pubic BME. More detailed studies such as cadaver studies or motion analysis studies are needed in the future. Finally, this study was limited to preoperative evaluations. Follow-up studies are needed to determine how pubic BME develops postoperatively.

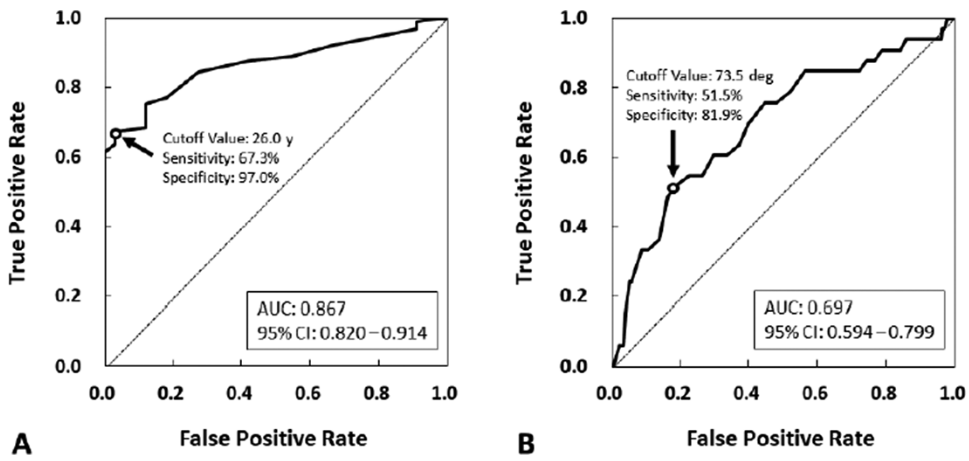

Fig. 5. The ROC curve of relevant factors of pubic bone marrow edema for age (A) and alpha angle (B).

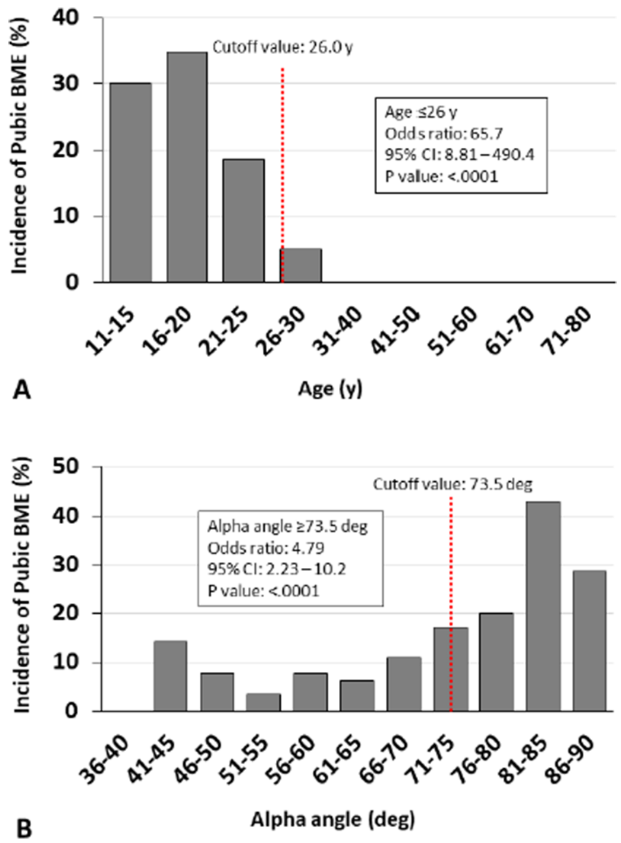

Fig. 6. The stratified incidence of pubic BME according to relevant factors for age (A) and alpha angle (B).

CONCLUSION

There was no significant difference about pubic BME on MRI among the three groups. Acetabular morphology was not a risk factor for pubic BME, but younger age (age $\leq$26 years) and greater alpha angle (alpha angle $\geq$73.5°) were risk factors for pubic BME. The surgeon should be aware of the possibility of osteitis pubis as well as FAIS in dysplastic hips, especially those with a large cam lesion.

DATA AVAILABILITY

The data supporting the conclusions of this article are included within the article.

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CONFLICT OF INTEREST STATEMENT

None declared.

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