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Original Article

Prevalence of pistol-grip deformity in patients with axial spondyloarthritis

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Abstract

Objective: To our knowledge, the prevalence of pistol-grip deformity (PGD) has not previously been studied in patients with axial spondyloarthritis (axSpA). This study aimed to evaluate PGD prevalence in patients with axSpA and to assess its relationship with the clinical and demographic factors.

Methods: A total of 158 patients with axSpA in whom diagnosis was established according to the Assessment of SpondyloArthritis International Society criteria in rheumatology department and 193 age- and sex-matched controls admitted to the emergency room and had anteroposterior (AP) pelvic X-rays were included in the study. PGD was identified by determining the non-spherical shape of the femoral head on AP hip or pelvic X-rays.

Results: Prevalence of PGD was significantly higher in patients with axSpA than in controls (20.3% vs. 8.8%, respectively, p=0.002). PGD was also found to be more frequent in patients with radiographic axSpA than in those with non-radiographic axSpA (26/106 [24.5%] vs. 6/52 [11.5%]); however, this difference did not reach statistical significance (p=0.056). The presence of PGD was significantly associated with the presence of hip arthritis (odds ratio [OR], 3.1; 95% confidence interval [CI], 1.2-8.7; p=0.023), ever smoking (OR, 4.5; 95% CI, 1.4-13.6; p=0.008), and male sex (OR, 38.7; 95% CI, 5.1-292.7; p<0.001) in univariate analyses. In multivariate model, ever smoking (OR, 2.9; 95% CI, 1.10-10.05; p=0.03) and male sex (OR, 27.0; 95% CI, 3.5-208.4; p=0.002) were associated with PGD.

Conclusion: PGD was significantly more common in patients with axSpA. Presence of PGD correlated significantly with hip arthritis, smoking, and sex. We assume that new bone formation could be the possible reason for increased PGD prevalence. Femoroacetabular impingement should be considered as a secondary cause of hip pain in patients with axSpA.

Keywords: Pistol-grip deformity, femoroacetabular impingement, spondyloarthritis, X-rays

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Introduction

Femoroacetabular impingement (FAI) causes mechanical hip pain owing to premature contact of the proximal femur with the acetabular wall (1). FAI is classified as pincer-type, cam-type, or mixed-type, which consists of properties of both pincer and cam-types. Pincer-type impingement is the acetabular cause of FAI, whereas cam-type is characterized by a non-spherical femoral head, which causes reduction of the femoral head-neck offset. Patients with FAI mostly report hip and/or trochanteric pain, limited hip range-of-motion, and a positive test for anterior impingement. The diagnosis of impingement syndromes generally depends on both clinical and imaging evidences. FAI is diagnosed by evaluating the morphology of the femoral head, acetabular roof, femoral head-neck junction, and contour of the acetabular rim on X-rays. Many different plain radiographic parameters, such as pistol-grip deformity (PGD), decreased femoral version, increased alpha angle, increased center-edge angle, and crossover signs, have been previously described to diagnose FAI. PGD has been reported as a distinctive feature of cam-type impingement on anteroposterior (AP) X-rays. Atypical extension of the more horizontally directed femoral epiphysis results with flattening of the concave surface of lateral femoral head in PGD (Figure 1) (2-6). The mild subclinical slipped femoral epiphysis, delayed epiphyseal closure, Legg-Calve-Perthes disease, bone remodeling, and osteophyte formation were suggested as the etiological factors for the formation of PGD, which caused early osteoarthritis (7-9). Doherty et al. (2) have reported a significantly higher prevalence ratio of PGD (17.71%) in patients than in the control group (3.61%), and a relative risk of 7 (odds ratio [OR] of 6.95) was estimated for its association with hip osteoarthritis.

Clinical presentation of FAI may resemble to that of some other diseases, which can involve the hip joint, such as ankylosing spondylitis (AS), hip dysplasias, or diffuse idiopathic skeletal hyperostosis. Hip involve-

ment in axial spondyloarthritis (axSpA) is a common and disabling problem. Prevalence of hip involvement in AS has been reported as 19%-36%, and the possibility of hip involvement increases if the disease starts at a younger age (10-12). Hip involvement, such as synovitis, entheseal inflammation, and bone-marrow involvement, may finally lead to secondary osteoarthritis (10-13). Resnick (8) has proposed that PGD develops as a result of osteophytes and degenerative alteration of the femoral head. He suggested that PGD was an acquired malformation caused by osteophyte formation and remodeling of the femoral head because of a degenerative process.

The prevalence of PGD has been estimated at approximately 10%, and it could be more frequent in men than in women. Several studies have investigated the prevalence of PGD and cam-type FAI in different populations and reported a close relationship between the cam-type FAI and sports; however, we could not find an article regarding the association and prevalence of PGD in patients with axSpA in the English literature (2, 7, 14-16).

Therefore, this study aimed to evaluate the prevalence of PGD in patients with axSpA and to assess the related clinical and demographic factors.

Methods

Study population

A total of 158 patients with axSpA in whom diagnosis was established according to the Assessment of SpondyloArthritis International Society (ASAS) criteria (17) in our rheumatology department and 193 age- and sex-matched controls that had AP pelvic X-rays after their admission to the emergency room were included in the study. According to the ASAS criteria, patients with axSpA can be divided to 2 subgroups: (a) radiographic (r-axSpA) and (b) non-radiographic (nr-axSpA).

Main Points

- Femoroacetabular impingement may contribute to hip-joint pain in axial spondyloarthritis (axSpA). Therefore, it should be included in differential diagnosis of hip involvement in this population.
- Pistol-grip deformity, which was one of the radiographic paramaters of the femoroacetabular impingement, was significantly more common in patients with axSpA.
- Smoking and male sex were significantly associated with the presence of pistol grip deformity.

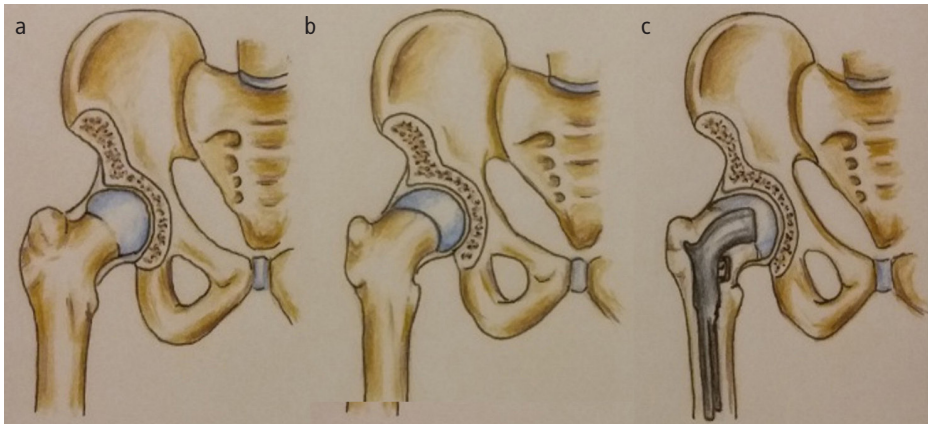

Figure 1. a-c. Normal femoral head is spherical in shape (a). Aspherical femoral head shape which gives the appearance of a "pistol-grip" deformity (b, c) is a typical radiographic sign of cam-type femoroacetabular impingement.

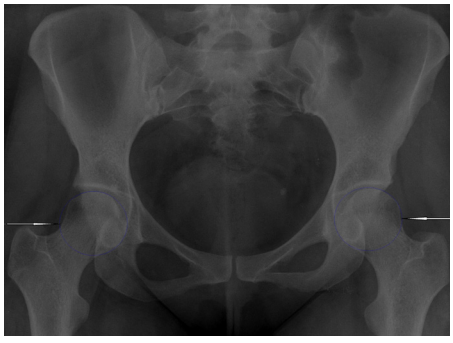

Figure 2. A 30-year-old woman's normal hip joints are shown on anteroposterior pelvic radiograph. Bilateral heads of the femur circumscribed by spherical blue circles (arrows) are spherical in shape. The sacroiliac joints are normal.

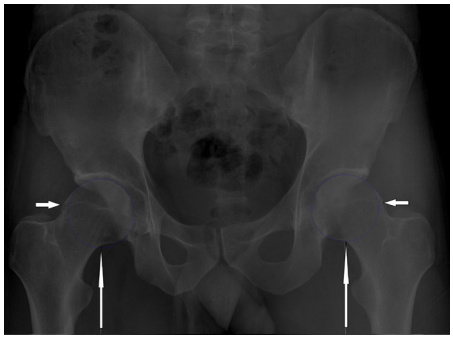

Figure 3. A 25-year-old man with a diagnosis of axial spondyloarthritis. Anteroposterior radiograph of the pelvis shows sphericity of the bilateral femoral heads (pistol-grip deformity) (short arrows) as the area that extrudes laterally from the blue circle (long arrows). This finding is compatible with cam-type impingement. Erosion and sclerosis with irregular joint narrowing, which is compatible with grade 3 sacroiliitis, are also seen in both the sacroiliac joints.

Demographic and clinical characteristics of the patients were evaluated. Clinical characteristics included duration of the disease, hip arthritis (chronic hip pain and/or radiological findings), Bath ankylosing spondylitis disease activity index (BASDAI), ankylosing spondylitis disease activity-C reactive protein score (ASDAS-CRP), erythrocyte sedimentation rate (ESR), human leukocyte antigen-B27 (HLA-B27) positivity, Bath ankylosing spondylitis functional index (BASFI), Bath ankylosing spondylitis metrology index (BASMI), modified stoke ankylosing spondylitis spine score (mSASSS).

Patients with grade 3-4 hip osteoarthritis (presence of prominent osteophytes on the joint margins, sclerosis of the subchondral bone, marked narrowing of the joint space, and altered shape of the femur), hip arthroplasties, and acetabular protrusions were excluded. We also excluded patients with improperly (positional and technical) taken radiographs. The study was approved by the Ethics Committee of İzmir Katip Çelebi University School of Medicine, and owing to its retrospective nature, the requirement to obtain informed consent was waived (Approval Date: July 21, 2016; Approval Number: IRB no: 212).

Radiographic assessment

All AP pelvic X-rays, which were close to the enrollment date, were evaluated by an experienced musculoskeletal radiologist (O.T. experienced with musculoskeletal radiology for 13 years). PGD was identified by observing the non-spherical femoral head on AP pelvic X-rays (Figure 1) (3). The femoral head was circumscribed by a spherical circle tool of the archived images by picture archiving and communication system (PACS) and classified as spherical or non-spherical on the basis of its complete or incomplete encirclement, respectively (Figures 2 and 3). The same radiologist determined the presence or absence of PGD on AP pelvic X-rays.

Table 1. Demographic and clinical characteristics of the study population.

Characteristics	Patients with axial spondyloarthritis (n=158)	Controls (n=193)
Age (years), mean±SD	41.5±12.5	40.3±14.7
Male, n (%)	87 (55.1)	116 (60.1)
Education level (years), mean±SD	9.6±4.4	N/A
Smoking status (smokers), n (%)	102 (65.4)	N/A
Duration of disease (years), mean±SD	13.0±10.5	N/A
BASDAI score, mean±SD	4.2±2.4	N/A
BASFI score, mean±SD	3.3±2.7	N/A
ASDAS-CRP score, mean±SD	2.8±1.2	N/A
HLA-B27 positivity, n (%)	75/121 (62.0)	N/A
BASMI score, mean±SD	3.1±1.8	N/A
mSASSS score, mean±SD	8.9±18.4	N/A
ESR (mm/h), mean±SD	25.5±21.9	N/A
CRP (mg/dL), mean ±SD	14.0±22.0	N/A

BASFI: bath ankylosing spondylitis functional index; BASDAI: bath ankylosing spondylitis disease activity index; ASDAS-CRP: ankylosing spondylitis disease activity score-C-reactive protein; HLA-B27: human leukocyte antigen-B27; BASMI: bath ankylosing spondylitis metrology index; mSASSS: modified stoke ankylosing spondylitis spine score; CRP: C-reactive protein; ESR: erythrocyte sedimentation rate; SD: standard deviation.

Table 2. Univariate and multivariate analysis of factors associated with PGD.

	Univariate analysis			Multivariate analysis		
Variables	OR	95% CI	p	OR	95% CI	p
Sex (man vs. women)	38.7	5.1-292.7	<0.001	27.0	3.5-208.4	0.002
Smoking (ever vs. never)	4.5	1.4-13.6	0.008	2.9	1.10-10.05	0.03
Hip involvement (present vs. absent)	3.1	1.2-8.7	0.023	2.0	0.64-6.71	0.21

PDG: pistol-grip deformity; CI: confidence interval; OR: odds ratio.

Statistical analysis

Data were given as mean and standard deviation (SD) or percentages, as appropriate. The chi-squared test was used for comparisons of categorical data. The Spearman's rank correlation test was used for bivariate correlations between variables. A logistic regression model was constructed to determine the factors associated with the presence of PGD. All the tests were 2-tailed, and for all measurements, a p-value of <0.05 was considered statistically significant. All statistical analyses were performed by using the Statistical Package for the Social Sciences version 16.0 software (SPSS Inc; Chicago, IL, USA).

Results

Baseline characteristics of study population

The axSpA group included 106 patients with r-axSpA and 52 patients with nr-axSpA. Demographic, clinical, and laboratory characteristics of the patients with axSpA and controls are summarized in Table 1.

Trauma was found to be the most common reason for emergency admission in the control group (153/193; 79.3%).

Among the patients with axSpA, 13.1% had hip arthritis, 80.3% were using non-steroidal anti-inflammatory agents, and 12.3% were using tumor necrosis factor-α inhibitors.

Frequency of PGD and related factors

Of the 158 patients with axSpA, 32 had PGD as did 17 of the 193 controls. The presence of PGD was significantly higher in patients with axSpA (32/158 [20.3%] vs. 17/193 [8.8%] and p=0.002). PGD was also found to be more frequent in patients with r-axSpA than in those with nr-axSpA (26/106 [24.5%] vs 6/52 [11.5%]); however, this difference did not reach statistical significance (p=0.056). PGD was only present in 2 female control subjects and 1 female patient with axSpA (Figures 2 and 3).

The presence of PGD correlated significantly with the presence of hip arthritis (r=0.190, p=0.018), ever smoking (r=0.227, p=0.004), and sex (r=0.424, p<0.001). No correlation was found between the presence of PGD and other clinical parameters (BASDAI, ASDAS-CRP, ESR, and CRP) and severity markers (HLA-B27, BASFI, BASMI, and mSASSS).

Multivariable analyses for PGD

We constructed a logistic regression model to determine the predictive factors for the presence of PGD. To construct the model, we included the variables, which showed significant associations in univariate analysis. This regression modeling revealed male sex (OR, 27.0; 95% CI, 3.5-208; p=0.002) and ever smoking (OR, 2.9; 95% CI, 1.10-10.05; p=0.03) as the factors that had independent associations with the presence of PGD (Table 2).

Discussion

In this study, we assessed the prevalence of PGD in patients with axSpA and found that it was significantly more common in these patients. Although it was statistically insignificant, PGD was more frequently seen in patients with r-axSpA than in those with nr-axSpA. Our findings also showed that the presence of PGD correlated significantly with hip arthritis, smoking, and male sex.

AxSpA is a chronic inflammatory disease that can involve the peripheral joints during the course of the disease. Hip involvement is one of its frequent manifestations, and inflammatory arthritis could eventually lead to secondary degenerative changes, namely osteoarthritis of the joint. Other factors that can cause mechanical hip joint pain may also coexist and complicate the disease process.

FAI is an important cause of early hip osteoarthritis. Ganz et al. (1) stated that early diagnosis and treatment of FAI is of particular importance in preventing secondary hip joint osteoarthritis. Establishing the diagnosis depends on taking a careful history, performing physical examination, and obtaining a proper radiological evaluation. Although advanced imaging methods, such as magnetic resonance imaging (MRI) or computed tomography (CT) scans, may provide more details, conventional radiography is still the primary preferred imaging modality in FAI. Stulberg et al. (18) have defined an abnormal head-neck junction on AP pelvic X-rays and referred to it as "PGD." Several studies have investigated the association between PGD and osteoarthritis through evaluation of AP pelvic X-rays (2, 15, 18).

Gosvig et al. (15) have investigated the prevalence of FAI-related anatomical malformations

in their cross-sectional study. PGD was diagnosed in 19.6% of men and 5.2% of women in the general population. They have also reported that 30.2% of men and 5.0% of women with hip osteoarthritis had PGD. Hack et al. (14) have investigated the prevalence of cam-type morphology in 200 asymptomatic volunteers, and PGD prevalence was found 14% in this study. They have also reported that the prevalence of a cam-type deformity was significantly higher in men than in women (24.7% vs. 5.4%). Goodman et al. (7) have reported not only a PGD prevalence of 8% in examination of 2,665 adult human skeletons but also a higher prevalence of severe osteoarthritis in the PGD group than in the non-PGD group (38% vs. 26%). Doherty et al. (2) have reported the prevalence of PGD as 17.71% in patients with hip osteoarthritis (30.26% for men, 4.67% for women) and 3.61% in control subjects (6.37% for men, 0.39% for women). Klit et al. (16) have reported the prevalence of PGD as 20.3% in patients with hip osteoarthritis who were derived from the Copenhagen osteoarthritis sub-study group. We found the prevalence of PGD as 20% in patients with axSpa and 9% in the control group.

Jung et al. (19) have evaluated the pelvic CT scans of 755 hips, and the prevalence of cam-type FAI was found to be 14% in men and 6% in women. Ahn et al. (20) have evaluated 45° Dunn view X-rays of the male volunteers, and cam-type FAI prevalence was reported as 30%. Similar to previous studies, we also found that PGD prevalence was significantly higher in men than in women. Moreover, the male sex was found to be independently associated with the presence of PGD. This higher prevalence of PGD in men (8% in control subjects, 20% in oatuebts with axSpA) correlates well with the fact that PGD is primarily a deformity of males as reported in previous studies (2, 7, 14, 15, 19, 20). This result is also compatible with the results of the Copenhagen osteoarthritis study, which has reported the prevalence of cam-type deformity in 17% of men and 4% of women (21).

Hashimoto et al. (22) have reported a PGD prevalence of 41% in patients undergoing total hip replacement. Sahin et al. (23) have reported the prevalence of cam-type FAI as 76.3% in patients with chronic hip pain and 42.9% in control subjects, which was quite higher than the reported prevalence in the literature. They used α angle in establishing the diagnosis of cam-type FAI. Differences in imaging methods, population groups (for example, unselected or athlete), sex of patients, and morphological features of interest (for example, cam or pincer) may be the possible explanations of inconsistency between the reported prevalences in the literature.

Several mechanisms were proposed as the cause of PGD in previous studies. High-impact sports in particular, including soccer, ice hockey, or basketball, might predispose an individual to the development of a cam-type deformity because of the high shear stress applied to the femoral head (24-26). Agricola et al. (27) have investigated the presence of the cam-type deformity among elite soccer players and reported a higher prevalence (26%) in soccer players than in controls (17%). Fukushima et al. have reported the prevalence of PGD as 44% in Japanese professional baseball players (28). The high stresses occurring during sports activities that lead to reactive bone formation was assumed as the underlying pathophysiological mechanism that leads to the cam-type deformity (26, 28, 29). Bone disease in axSpA is a complex process with both bone loss and new bone formation. New bone formation, which may lead to bridging and ankylosis between the bones, contributes to the structural progression of the disease (30). New bone formation having an impact on clinical features of axSpA was the probable underlying pathophysiological mechanism of increased cam-type deformity in our study.

In our study, we found that the prevalence of PGD was significantly higher in patients with axSpA than in controls. Large joints, including hips, are frequently involved after sacroiliac and spinal involvements in AS. Hip involvement occurs in 25%-50% of patients with AS. Inflammation and pathological new bone formation in hip joints may be observed. Secondary osteoarthritis is also seen in involved hips of patients with AS at later stages, and new bone formation can also be seen because of osteoarthritis, which results in further functional impairment. To the best of our knowledge, this is the first study in English literature that reported a higher prevalence of PGD in patients with axSpA. In this study, although we assumed that new bone formation was the possible reason for increased PGD prevalence, it would be difficult to distinguish whether this process could be attributed to AS or secondary osteoarthritis (10-13). Further studies evaluating the prevalences and possible underlying causes of PGD in patients with axSpA are needed to accurately interpretate our results.

We found that the presence of PGD correlated significantly with the presence of smoking and male sex. Smoking negatively affects the repair process of fractures. It delays regeneration of the bone in fracture repair and increases the risk of non-union and osteomyelitis, probably as a result of decreased oxygen delivery to the tissues (31). Decreased oxygen delivery caused by smoking may result in high stresses in the bone tissue that may lead to reactive bone formation and PGD. Therefore, we can assume that male smokers with axSpA are at a higher risk of developing PGD.

This study had several limitations. First, the prevalence of cam-type FAI might have been underestimated because advanced imaging techniques, such as MRI or CT, are more sensitive than X-rays for detecting the morphological characteristics associated with FAI. The Dunn view radiography of the hip would also be useful for diagnosis of cam-type FAI. However, it would not be ethical to expose asymptomatic volunteers to unnecessary radiation or to perform MRI scans in all participants owing to its high cost. Second, we did not conduct the study in a normal population; it was a patient-based study. The lack of inter-observer and intra-observer correlations, which might have decreased the consistency of evaluation, was also a major limitation of our study. We determined PGD according to the definition of PGD reported by Stulberg et al. (18) who defined an abnormal head-neck junction on AP pelvis radiography as "PGD." To decrease inconsistency, the femoral head was circumscribed by a spherical circle tool of PACS and classified as spherical or non-spherical on the basis of its complete or incomplete encirclement, respectively (3). The radiologist who determined the presence or absence of PGD had 13 years of experience in musculoskeletal radiology. Another limitation was the relatively small sample size. Further studies including more subjects will provide more accurate results.

In conclusion, PGD was significantly more common in patients with axSpA. The presence of PGD correlated significantly with hip arthritis, smoking, and male sex. We believe that new bone formation is the possible reason for the increased PGD prevalence. The prevalence of PGD in patients with r-axSpa was higher than in those with nr-axSpA; however, the difference was not statistically significant. Further studies are required to determine a significant difference between them. FAI should be considered as a secondary cause of hip pain in patients with axSpA.

Ethics Committee Approval: Ethics committee approval was received for this study from the Ethics Committee of İzmir Katip Çelebi University School of Medicine (Approval Date: July 21, 2016; Approval Number: IRB no: 212).

Informed Consent: Informed consent was obtained from the patients who participated in this study.

Peer-review: Externally peer-reviewed.

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