Coexistence of Ipsilateral Sacroiliitis and Simple Cyst in the Iliac Bone: A Case Report

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Abstract

Here, a 36-year-old female patient with inflammatory sacroiliitis and simple bone cyst of the ipsilateral iliac bone is presented. Simple bone cysts occur in the developing skeleton, usually asymptomatic; however, they are benign lytic bone lesions that can cause pathological fractures. The prevalence of simple bone cysts in the whole body has been reported as 0.30/100000. Only 2% of these rare cysts are found in the pelvis, and according to our knowledge, it is the first case in which iliac simple bone cyst is seen together with sacroiliitis on the same side.

Keywords: Low back pain, Sacroilitis, Simple bone cyst

Özet

Burada, ipsilateral iliak kemiğinde inflamatuar sakroileit ve basit kemik kisti bulunan 36 yaşında bir kadın hasta sunulmaktadır. Basit kemik kistleri gelişmekte olan iskelette asemptomatik olarak ortaya çıkar; ancak patolojik kırıklara neden olabilen iyi huylu litik kemik lezyonlarıdır. Tüm vücutta basit kemik kisti prevelansı 0,30/100000 olarak bildirilmiştir. Bu nadir kistlerin sadece %2`lik kısmı pelviste bulunur ve bilgilerimize göre bu olgu, iliak basit kemik kistinin aynı tarafta sakroileit ile birlikte görüldüğü ilk vakadır.

Anahtar Sözcükler: Basit kemik kisti, Bel ağrısı, Sakroileit

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Introduction

Pain originating from the sacroiliac joint constitutes 13-30% of all low back pain (1). Sacroiliac joint dysfunction, rheumatological diseases causing inflammatory sacroiliitis, trauma, pregnancy, and sports-related pain are the causes of pain originating from the sacroiliac joint (2). Simple bone cysts (SBC) are seen in the developing skeleton, usually asymptomatic; however, they are benign lytic bone lesions that can cause pathological fractures (3). The prevalence of SBC in the whole body has been reported as 0.30/100000 (4). Only 2% of these rare cysts are located in the pelvis (5).

Here, a case with inflammatory sacroiliitis and SBC in the ipsilateral iliac bone is presented.

Case Report

A 36-year-old female patient who applied to our polyclinic had low back and left hip pain for 8 years; but intensified in the last 3 months. Back and neck pain have been added to low back pain in the last year. Low back pain decreased with movement and increased with rest. The patient did not have night pain, but had morning stiffness lasting up to two hours. It was learned that she had hypothyroidism and used levothyroxine 25 mcg/day. The patient's family history was unremarkable. In the systemic examination, skin rash, peripheral arthritis, uveitis, oral and genital aphthae, diarrhea, constipation were not detected.

On physical examination, cervical spine movements were painful; but there was no limitation. Movements of the lumbar spine were painful and limited. Chest expansion was 2.5 cm. The fingertip-to-floor distance was 10 cm, the Modified Schober was 5 cm, the lumbar lateral flexion was 9 cm, and the intermalleolar distance was 104 cm. Gaenslen, Mennel, and compression tests were positive in the left sacroiliac joint. The FABER test was positive on the left. There were tenderness in the bilateral 1st and 7th costochondral joints, bilateral iliac crests, bilateral spina iliaca posterior superiors, and the 5th lumbar spinous process.

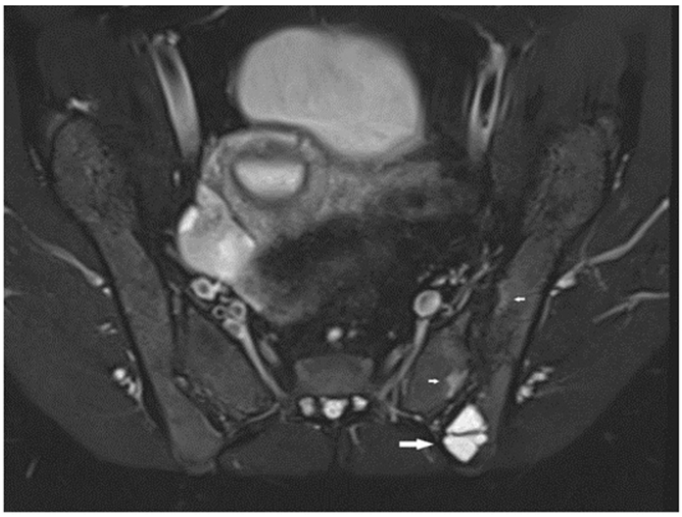
In laboratory examination, hemogram, biochemical tests and inflammatory markers were within normal limits. Brucella agglutination test was negative. In the pelvic anteroposterior radiograph, there were irregularities suggesting sacroiliitis in the left sacroiliac joint, sacral and iliac wings, and increased sclerosis (Figure 1).

Figure I. Anteroposterior pelvis X-ray

Arrows: Left iliac and sacral wing irregularities suggesting sacroiliitis and increased sclerosis

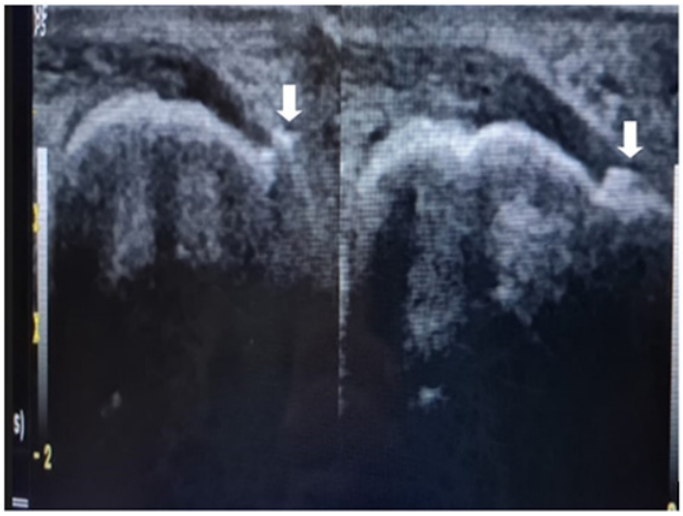
In the sacroiliac joint magnetic resonance imaging (MRI), hyperintense bone marrow edemas in the anterior of the iliac wing in the axial STIR sequence and in the posterior of the sacral ala in the left sacroiliac joint and areas of hyperintense fat accumulation evaluated in favor of structural changes on T1-weighted images were observed. These findings were evaluated as acute and chronic inflammatory sacroiliitis. In addition, MRI examination revealed a 25x18 mm lesion in the posteroinferior aspect of the left iliac bone, which was hyperintense on STIR (Figure 2) and hypointense on T1-weighted images, consistent with benign, naturally septated SBC. HLAB-27 was negative. In the ultrasonographic evaluation of both feet, bilateral Achilles enthesophyties were determined (Figure 3).

Figure II. Sacroiliac joint MRI STIR sequence axial plan view



Thick arrow: Bone cyst with hyperintense appearance in posterior iliac wing
Thin arrows: Bone marrow edema with subchondral hyperintense appearance in the sacral region and iliac wing

Figure III. Bilaterally foot ultrasonography



Arrows: Achilles enthesopathy

The Bath Ankylosing Spondylitis Disease Activity Index (BASDAI) was 5.6, and the Bath Ankylosing Spondylitis Functional Index (BASFI) was 1.8.

With the diagnosis of axial spondyloarthropathy and iliac bone cyst, the patient was started on a physical therapy program consisting of hot pack TENS, ultrasound, breathing, posture, flexors stretching, extensors strengthening exercises, and 3x50 mg/g indomethacin treatment.

In the follow-up two weeks later, BASDAI was 2.3 and BASFI was 0.6. The same medical treatment was continued. Written informed consent was obtained from the patient.

Discussion

SBC are benign, usually asymptomatic lesions that are seen at the age of 5-15 years and slightly more frequently in men, although they can occur at any age (6). Two-thirds of SBC are located in long bones such as the proximal femur or proximal humerus (7). Rarely, cysts located in the calcaneus or pelvis are also encountered (8-10). Our case was female, and the cystic lesion adjacent to the sacroiliac joint was a very rare site of involvement (11).

In the differential diagnosis of sacroiliitis, causes such as inflammatory rheumatic diseases, acne and isoretinoic acid-related arthritis, specific and nonspecific infections, sarcoidosis, hyperparathyroidism, and paraplegia are included. Our case presented with inflammatory low back and hip pain. In accordance with the physical examination and laboratory findings, and in the imaging methods requested with a preliminary diagnosis of sacroiliitis, SBC was detected on the same side of the pelvis with sacroiliitis. Because SBC are usually painless, most cases have pathological fractures, while those found in the ileum and calcaneum are usually diagnosed incidentally.

Although SBC are mostly seen in childhood, in a study examining 16 cases located in the pelvis, it was shown that 5 patients had cysts in the posterior part of the ilium adjacent to the sacroiliac joint, and these posterior lesions occurred in the older patient group (8). In another study evaluating 75 SBC, it was argued that those with pelvic localization were seen in older patients, while another study reported four cases of simple pelvic cysts seen in the adolescent group (5, 12). It can be thought that pelvic lesions may remain asymptomatic for a longer period of time, since they usually occur in the non-weight-bearing parts of the ileum.

The presence of accompanying sacroiliitis on the same side was effective in detecting SBC at a relatively early age in our case.

In conclusion; SBC in the pelvis are an extremely rare condition. Our case is interesting because, to our knowledge, it is the first case in which sacroiliitis and SBC of the iliac bone were seen together with ipsilateral sacroiliitis. Diagnosis and follow-up of SBS are important because they carry a risk of bone fracture.

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