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Case Report / Olgu Sunumu

Two rare cases during pregnancy: pregnancy-associated inflammatory sacroiliitis and sacral stress fracture

Gebelik sırasında iki nadir olgu: Gebelik ile ilişkili enflamatuvar sakroileit ve sakral stres kırığı

Levent Özgönenel,[1] Ali Önder Atça,[2] Onat Üzümcügil,[3] Hande Başat,[1] Ebru Aytekin[4]

[1]Department of Physical Medicine and Rehabilitation, Kemerburgaz University Medikal Park Hospital, İstanbul, Turkey
[2]Department of Radiology, Kemerburgaz University Medikal Park Hospital, İstanbul, Turkey
[3]Department of Orthopedics and Traumatology, Kemerburgaz University Medikal Park Hospital, İstanbul, Turkey
[4]Department of Physical Medicine and Rehabilitation, İstanbul Training and Research Hospital, İstanbul, Turkey

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ABSTRACT

Hip and low back pain is common during pregnancy or postpartum period. Herein, we report two rare cases in whom pain symptoms started during the last trimester of pregnancy. Both patients had similar clinical presentation; however, a meticulous analyses helped us diagnose pregnancy-associated inflammatory sacroiliitis in the first case, and sacral stress fracture in the second case.

Keywords: Low back pain; pregnancy; sacroiliitis; stress fracture.

ÖZ

Kalça ve bel ağrısı, gebelik sırasında veya doğum sonrası dönemde sık görülür. Bu yazıda, ağrı semptomları gebeliğin son trimesterinde başlayan iki nadir olgu sunuldu. Her iki hastada da benzer klinik tablo mevcuttu; ancak titiz analizler birinci olguda gebelik ile ilişkili enflamatuvar sakroileit ve ikinci olguda sakral stres kırığı tanısında bizlere yardımcı oldu.

Anahtar sözcükler: Bel ağrısı; gebelik; sakroileit; stres kırığı.

Hip and low back pain is common during pregnancy or postpartum period.[1] Pain may originate from the lumbar region, hip or the sacroiliac joint (SIJ). Pain in the low back pain or the buttock has usually a non-specific etiology and a self-limited course.[2] However, there are certain conditions which complicate the diagnosis such as transient hip osteoporosis, hip avascular necrosis, sacral stress fracture, and postpartum sacroiliitis. Herein, we report two cases of pregnancy-associated inflammatory sacroiliitis and sacral stress fracture in each, both of which are rare and often underrecognized in daily clinical practice.

CASE REPORT

Case 1– A 32-year-old woman presented with two-week long pain in her low back which radiated to her left hip 10 days after delivery with cesarean section. The pain was initially mild; however, it became severe in the past two days to the point that she had pain even at rest and unable to walk without an assistance. Her medical history was non-specific. She was afebrile. There was localized tenderness in the left lumbar paravertebral, inguinal, and sacroiliac regions. The flexion, abduction, external rotation (FABERE) flexion, adduction, internal rotation (FADIR), and Gaenslen tests were positive on the left. Erythrocyte sedimentation rate (ESR) was 40 mm/hour and C-reactive protein (CRP) was elevated at 17 mg/dL. There was no leukocytosis or bacteriuria. Initial radiographs of the pelvic and lumbar region were normal. Magnetic resonance imaging (MRI) of the lumbar spine and pelvis revealed central disc bulging in the fifth

Corresponding author / *İletişim adresi*: Ebru Aytekin, MD. Kasap İlyas Mah. Org. Abdurrahman Nafiz Gürman Cd. 34098 Fatih, İstanbul, Turkey.
e-mail / *e-posta*: ebruaytekin@hotmail.com

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lumbar and first sacral levels, bone edema, and sclerosis in SIJ. Bone edema was seen around the upper left SIJ, which was characterized by high-intensity signal on the Short T_1 Inversion Recovery (STIR) sequences and low-intensity signal on the T_1-weighted sequences. Sclerosis was in the left iliac bone characterized by zones with low-intensity signal on T_1- and T_1-weighted sequences. Both sacroiliac articular surfaces were normal (Figures 1a, b).

The bulging in the L_5-S_1 region was not consistent with her clinical presentation and the patient was diagnosed with pregnancy-associated inflammatory sacroiliitis. A written informed consent was obtained from the patient. As the patient was not nursing, she was treated with indomethacin 25 mg tid around-the-clock for three weeks and, then, as needed. A cane and lumbosacral corset was given to assist her ambulation. We also recommended cold pack application to the sacroiliac region twice a day. The patient was instructed to perform bed exercises (ankle rotation-pumps, knee bends, quadriceps sets, hip abduction-extension sets). At three months during follow-up, she was able to perform all daily activities without pain. In her repeated sacroiliac MRI, bone edema regressed with no sign of erosion or destruction in the SIJ (Figures 2a, b).

Case 2– A 28-year-old woman presented with pain in the low back and right hip starting at 37th week of pregnancy. The pain radiated to the right leg and was aggravated by movement. She delivered at 38th week of pregnancy via cesarean section without any relief in pain. Her vital signs were within normal ranges. She had localized tenderness in the low back and right sacroiliac regions. Lumbar flexion, extension, and hip rotations were painful and restricted. Pelvic and lumbar radiographs revealed only hyperlordosis. Pelvic and lumbar MRI showed a vertical fissure surrounded by medullary bone edema in the right sacrum involving the sacral ala and medial to the SIJ, which was diagnosed with sacral stress fracture. There was also bone edema around the lower part of left SIJ, which could be early bone edema related to the sacral stress fracture. Patchy bone edema characterized by high-intensity signal on STIR sequences and low-intensity signal on T_1-weighted sequences were also noted. A hypointense fracture line was seen within the area of edema (Figures 3a, b). A written informed consent was obtained from the patient. Physical therapy with intermittent ultrasound, magnetotherapy, hot-cold packs, and electrostimulation was initiated. Paracetamol 500 mg three times daily and local diclofenac patch were prescribed in addition to diclofenac Na 25 mg once daily, if she had severe pain, as she was still nursing. Vitamin D replacement therapy was given, as her serum 25-hydroxy vitamin D [25(OH)D] level was very low (7.4 ng/mL). She was instructed to wear a lumbosacral corset during ambulation. After 20 sessions of physical therapy and drug therapy, she was free of pain.

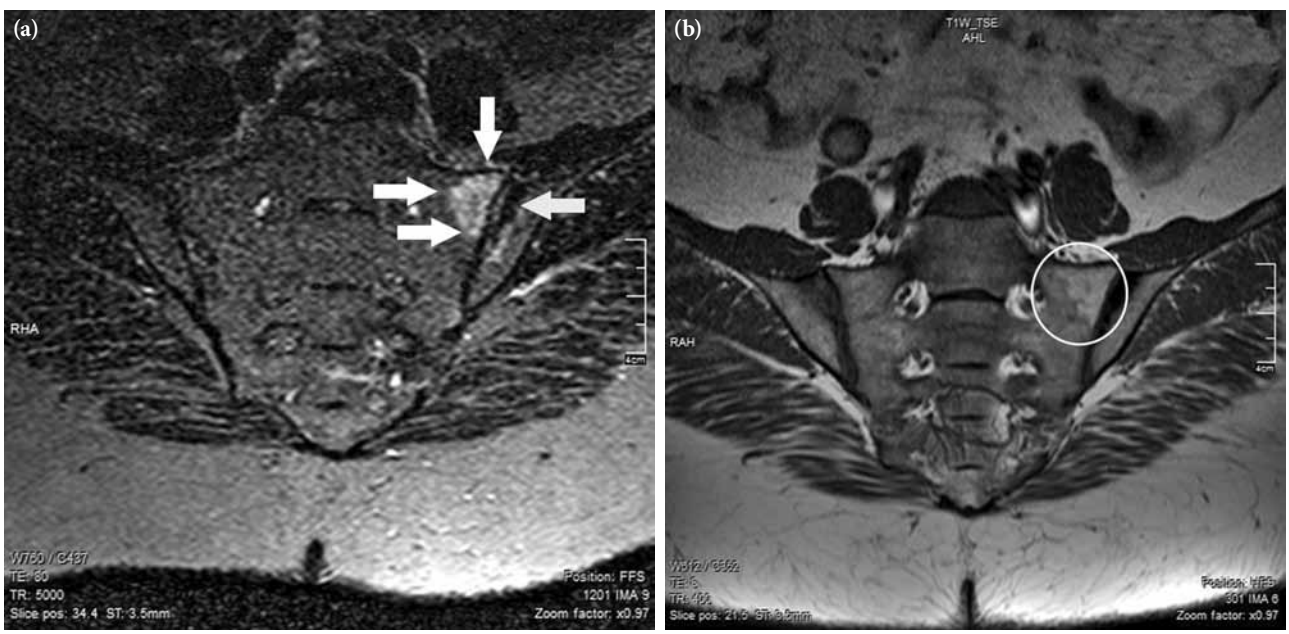

Figure 1. (a) Bone edema (high-intense) in a triangular shape and sclerosis (low-intense) in the left iliac bone on short T_1 inversion recovery sequences. **(b)** Bone edema and sclerosis characterized by low-intensity on T_1-weighted sequences.

Figure 2. (a) A short T_1 inversion recovery magnetic resonance image showing regression of bone edema. **(b)** A T_1-weighted magnetic resonance image showing regression of bone edema, subchondral fatty marrow changes, and sclerosis.

DISCUSSION

Weight gain, hormonal, and postural changes associated with pregnancy may disrupt the pelvic stability.[4] Increased lumbar lordosis, pelvic tilt to anterior, and softening of the ligaments by relaxin give rise to biomechanical changes in the pelvic region.[4] Sacroiliac joint is a non-weight-bearing joint with a small range of motion which has to endure extra-burden during pregnancy due to biomechanical changes, thereby, leading to pubic instability, inflammation, bone edema, and stress fractures.[5] The latter can easily occur, if there is also accompanying insufficient bone mass due to osteopenia or vitamin D deficiency.[5]

The first case had severe pain and her CRP was very high, leading us to include pyogenic sacroiliitis

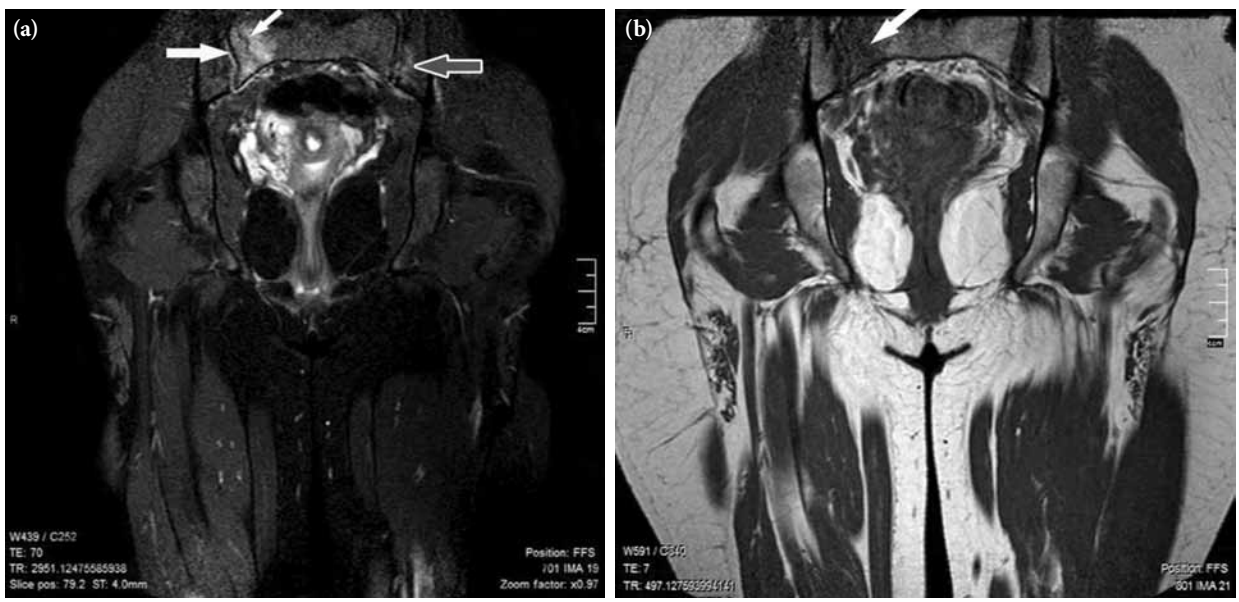

Figure 3. (a) Sacral stress fracture on short T_1 inversion recovery sequences. Vertical fissure surrounded by medullary bone edema medial to the right sacroiliac joint. Bone edema around the lower part of the left sacroiliac joint. **(b)** Same image of Figure 3a on T_1-weighted sequences. Vertical fissure surrounded by medullary bone edema are characterized by low-intensity signal.

and spondyloarthritis in her differential diagnosis. Pyogenic sacroiliitis may be accompanied by a history of intravenous drug abuse, infective endocarditis, urinary tract infection, abortion or delivery.[6] There is commonly high fever along with severe back pain. Laboratory findings include leukocytosis, elevated ESR and CRP levels, and occasionally positive blood and urine cultures.[6] Bone edema, sclerosis, and erosions in SIJ are typical radiological changes similar to those in postpartum sacroiliitis.[7] On the other hand, our first case had no sign or medical history of any infection. There was no leukocytosis and bacteriuria in her laboratory findings. Elevated CRP could possibly be due to the recent cesarean section.[8] Most importantly, the sacroiliac articular surfaces were normal on the MRI. The second case had no signs of infection, either. Therefore, we did not consider pyogenic sacroiliitis in her differential diagnosis.

Spondyloarthritis was included in the differential diagnosis of our cases. The personal and family histories were negative for chronic back pain, stiffness, peripheral arthritis, inflammatory bowel disease, or uveitis. Although spondyloarthritis may flare up during the postpartum period, the symptoms started during the past days of pregnancy in our case, which is atypical of a flare-up. Additionally, the ventrocranial part of the SIJ was affected in our case, as opposed to the typical dorsocaudal synovial involvement in early sacroiliitis of spondyloarthritis.[9]

Although the patient's history and clinical presentation were atypical for spondyloarthritis (SPA), axial SPA was unable to be excluded, since our case had sacroiliitis, back pain, and elevated ESR and CRP levels. Therefore, SPA should be considered during follow-up.

Osteitis condensans ilii (OCI), a condition characterized by bilateral well defined triangular sclerosis of SIJ, was also considered in the differential diagnosis for both cases. This clinically benign condition predominantly affects the iliac portion of the SIJ, and the acute phase reactants are usually within normal limits.[10] However we excluded this diagnosis, as because laboratory parameters were not in normal limits and radiological involvement of the SIJ were not typical for OCI.

Sacral stress fractures usually occur in elderly with osteoporosis or in individuals with a normal bone density who are exposed to repetitive stress, such as in long-distance runners or marching soldiers.[2,11] Biomechanical and hormonal (relaxin-induced) changes during pregnancy may also cause bone edema and sacral stress fractures, in particular, in the presence of insufficient bone mass due to osteopenia or vitamin D deficiency, as noted in our second case.[12] Sacral stress fractures most commonly involve the sacral ala and are located medial to the SIJs and may be uni- or bilateral.[13] In our case, the location of the lesion was typical for a sacral stress fracture. There was one fracture on the right side in our case; however, the bone edema around the left SIJ could be a signal for an early fracture, as well. In the literature, both uni- and bilateral fractures are reported with relatively equal frequencies.[14]

In conclusion, hip and low back pain is common during pregnancy or postpartum period. A careful analysis of the clinical, laboratory, and radiographic findings may help the clinician to detect the underlying cause and tailor an appropriate treatment.

Declaration of conflicting interests

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