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Case Report

ATYPICAL SACRAL AND ILIAC BONE INSUFFICIENCY FRACTURES IN A PATIENT RECEIVING LONG-TERM BISPHOSPHONATE THERAPY

Abstract

Insufficiency fractures are a type of stress fracture occurring in bones with low resistance. The most common cause of insufficiency fractures is postmenopausal osteoporosis. Corticosteroid use, radiotherapy, rheumatoid arthritis, Paget disease, osteomalacia, long-term use of bisphosphonates, hyperparathyroidism, diabetes mellitus, primary biliary cirrhosis, renal osteodystrophy are among the other risk factors. These fractures are generally overlooked because the symptomatology is similar to the clinical signs of osteodegenerative diseases, which are common in the elderly, particularly without a history of trauma.

A 73-year-old female patient was admitted to our outpatient clinic with a increase in low back pain intensity and difficulty in walking 3 months ago without a history of any trauma. She had a waddling gait with painful movements of both hips. Lumbar range of motion was normal but painful. On laboratory examination 25(OH) vitamin D3 level was low. Pelvic magnetic resonance imaging showed widespread bone marrow edema at both sacral wings and left iliac bone and signal recordings corresponding to insufficiency fractures in this region.

Herein we present a case of an osteoporosis patient with sacral and iliac bone fractures that were treated conservatively.

Key Words: Fractures, Stress; Ilium; Sacrum; Osteoporosis; Aged; Diphosphonates.

Olgu Sunumu

UZUN SÜRELİ BİFOSFONAT TEDAVİSİ ALAN BİR HASTADA GÖRÜLEN ATİPİK SAKRAL VE İLİAK KEMİK YETERSİZLİK KIRIKLARI

Öz

Yetersizlik kırıkları direnci azalmış kemikte görülebilen bir tip stres kırığıdır. En sık sebep postmenopozal osteoporozdur. Kortikosteroid kullanımı, radyoterapi, romatoid artrit, Paget hastalığı, osteomalazi, uzun süreli bifosfonat kullanımı, hiperparatiroidi, diabetes mellitus, primer bilier siroz ve renal osteodistrofi diğer risk faktörleri arasındadır. Yaşlı popülasyonda sık görülen osteodejeneratif hastalıkların klinik bulgularına benzer semptomatoloji görülmesi nedeniyle, özellikle de bir travma öyküsü yoksa sıklıkla gözden kaçırılır.

Yetmişüç yaşında kadın hasta 3 ay önce travma olmadan oluşan bel ağrısında artış ve yürümede zorluk şikayeti ile polikliniğimize başvurdu. Her iki kalça hareketleri ağrılı ve ördekvari yürüyüşü vardı. Bel hareketleri açık ancak ağrılı idi. Laboratuvarında 25(OH) vitamin D3 düzeyi düşüktü. Pelvik manyetik rezonans görüntülemede bu bölgede yetersizlik kırığını işaret eden her iki sakral kanat ve iliak kemikte yaygın kemik iliği ödemi ile uyumlu bulgular görüldü.

Bu olgu sunumunda sakral ve iliak kemik kırığı tespit edilen, konservatif yöntemlerle tedavi edilen bir osteoporoz hastası sunulmuştur.

Anahtar Sözcükler: Yetersizlik Kırığı; İlium; Sakrum; Osteoporoz; Yaşlı.

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Introduction

Insufficiency fractures are a type of stress fracture that occurs with normal or physiological stress on bones that are weakened by decreased elastic resistance (1). Osteoporosis is the most important risk factor, and the pelvis is the most common location for these fractures. The sacrum and pubic rami are typically affected (2,3). This type of fracture is less common in iliac bones, and only a small number of case reports on iliac bone fractures have been reported in the English literature (3,4).

Insufficiency fractures are generally overlooked because of non-specific clinical symptomatology. Complaints such as pain in the hip and groin and walking problems are suggestive of degenerative diseases in the elderly; therefore, diagnosis of insufficiency fractures causing these symptoms may be delayed (5). Delays in the diagnosis and treatment process may increase disability. The most important components of therapy are a conservative approach and physical therapy (5,6).

Herein we present a case of patient who had sacral-iliac bone insufficiency fractures that were treated successfully with physical therapy and vitamin D replacement.

Case Report

A 73-year-old female patient was admitted to our outpatient clinic with a 2-year history of hip and low back pain and difficulty in walking. She had been referred to various clinics for this complaints and had received medical treatments for a diagnosis of lumbar discopathy. She had progressive increase in low back pain intensity and difficulty in walking 3 months ago. She stated that the pain was worse when walking or standing for a long time and that it radiated to both knees and decreased with rest or use of analgesics. She did not report a history of trauma. After that she had received electrotherapy in another center; however, her symptoms returned only 2 weeks after she completed the program. Her medical history revealed hypertension and osteoporosis. She was taking calcium and vitamin D for osteoporosis. She had been treated with bisphosphonates for 10 years and had last received a zoledronic acid infusion 1 year ago.

Her vital signs were stable, and the results of systemic examination were normal. Her pain severity was 8/10 on the visual analog scale (VAS). She had a waddling gait with painful movements of both hips. Lumbar and hip range of motion was normal but painful. The sacroiliac compression test was bilaterally positive. The results of a neurological examination were normal.

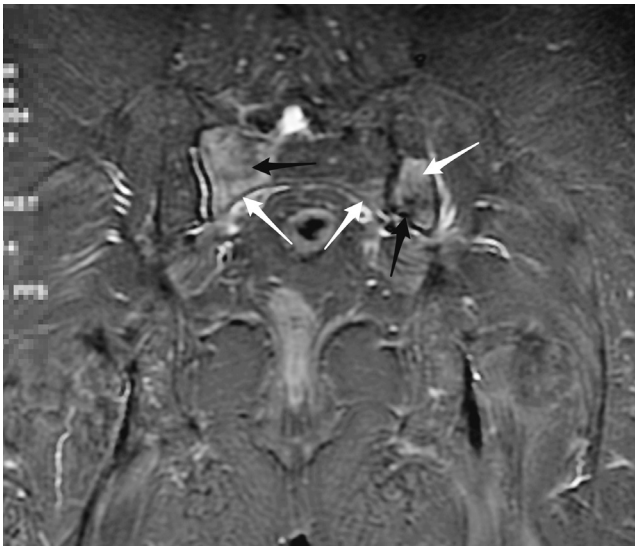

Figure 1— In the Short TI Inversion Recovery (STIR) sequence at the coronal plane, areas of edema are observed in both iliac bones, although more prominently on the right and hyperintense (white long arrows) at the left sacral wing. Hypointense (black short arrows) signal recordings were present, indicating insufficiency fractures within the area of edema.

On laboratory examination, serum calcium was 9,3 mg/dl (8.4-10.2), phosphate 3.1 mg/dl (2.3-4.7), alkaline phosphatase 177 U/l (40-150), PTH of 71 pg/ml (19.8-74.9) and 25(OH) D3 level 10.4 pg/L. Remaining including complete blood count, eritythrocyte sedimentation rate, CRP, renal function tests, protein electrophoresis, tumor markers, hepatic function tests, serum protein and albumin were in normal ranges. A subsequent DEXA scan revealed osteopenia.

Lumbar magnetic resonance imaging (MRI) performed before her referral to our clinic revealed bulging in the L1–2 and L3–4 discs, hypertrophy of the facet joints at the L4–5 vertebrae, and disc degeneration. Roentgenograms of the pelvis and both hip joints revealed degenerative changes. Subsequent pelvic MRI showed widespread bone marrow edema at both sacral wings and left iliac bone and signal recordings corresponding to insufficiency fractures in this region (Figures 1, 2). Because she did not have any diseases such as hypogonadism, hyperthyroidism, hyperparathyroidism, a tumoral disease such as multiple myeloma, or a metabolic disease such as diabetes mellitus and was not using any drugs that may lead to bone mass loss by affecting bone metabolism, the condition of the patient was considered to be the result of atypical sacral and iliac bone insufficiency fractures due to osteomalacia and long-term bisphosphonate therapy.

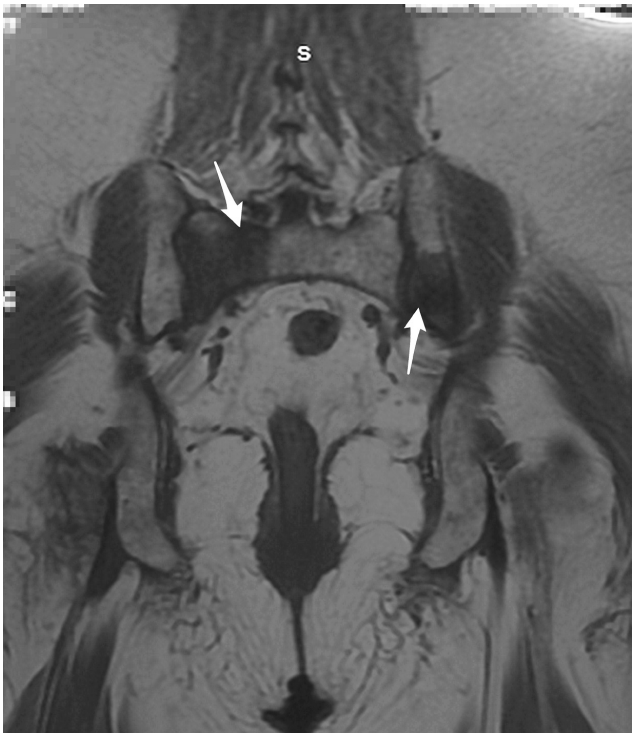

Figure 2— An area of edema in the right iliac bone and a hypointense edema area at the left sacral wing were observed on T1-weighted sequences at the coronal plane. Hypointense (white arrows) signal recordings are present, indicating insufficiency fractures within the area of edema.

A nonsteroidal anti-inflammatory drug (Naproxen 750mg/day, 7days), hotpack (20min/day), and electrotherapy (transcutaneous electrical nerve stimulation, 20 min/day) was started for pain relief. Joint range of motion exercises, progressive resistance exercises, and balance-coordination exercises were planned as physiotherapy program. Therapeutic doses of vitamin D (50000 IU/week, 8 weeks) was started and dietary calcium intake was increased with close follow-up of serum calcium levels. At the end of 3 weeks, her VAS score had decreased from 8 to 1, and her walking distance had increased. She was discharged with a home exercise program.

At the follow-up examination 3 months after discharge, her VAS score was 2, her vitamin D level was normal, and there were no recently developed symptoms. The patient was safely ambulatory for long distances without any support.

Discussion

Stress fractures are classified as fatigue and insufficiency fractures. Fatigue fractures are due to abnormal stress on normal bone, whereas insufficiency fractures are due to normal stress on weakened bone (1). Pelvic insufficiency fractures occur rarely as a diagnostic entity in cases of back and hip pain; however, they are one of the most significant conditions in terms of morbidity and mortality (5,6). These pelvic and hip fractures are still frequently overlooked in the elderly. It was reported that the incidence had increased 5.3-fold by 2002, and the mortality rate approached that of hip fractures (7). Moreover, the incidence is considered to be underestimated because of uncertain clinical signs and difficulty in diagnosis (5).

The vertebrae, tibia, femur, fibula, calcaneus, ramus pubis, sacral wing, and ilium are the areas where insufficiency fractures are observed. The pelvis is the most frequently affected region, whereas ilium fractures are relatively less common (3,4). Soubner et al. (3) reported only one case of ilium fracture out of 91 bone fractures in a series of 60 patients with insufficiency fractures. Fractures generally occur in elderly postmenopausal osteoporosis patients. Corticosteroid use, radiotherapy, rheumatoid arthritis, Paget disease, osteomalacia, hyperparathyroidism, diabetes mellitus, primary biliary cirrhosis, and renal osteodystrophy are among the other risk factors (8). Long-term use of bisphosphonates is among the recently described risk factors (1,9). In our case, the patient had a known history of osteoporosis for 10 years and osteomalacia.

Symptoms of the disease may be confused with complaints of degenerative conditions. Early diagnosis is difficult in the absence of a history of major trauma. Persistent, sometimes sudden back pain, sacral-pelvic pain, and groin pain are the frequently observed symptoms (5). Back pain may radiate to the gluteal region, hips, and lower extremities. Thus, it may be confused with osteoarthritis, trochanteric bursitis, spinal stenosis, and lumbar disc herniations (6,10).Generally, patients complain of difficulty in walking because of the increasing severity of pain. Neurological symptoms are generally absent. Gotis-Graham et al. (11) reported pain at the sacrum and hips as the most common symptom and a 5%–6% incidence of radiculopathy. In our case, the patient had experienced back pain and pain in both hips for years. Our patient's diagnosis was also delayed and she was prescribed several medications with the misdiagnosis of lumbar disc herniation previously.

The most preferred diagnostic methods are conventional radiography, MRI, computed tomography (CT), and bone scintigraphy (12). Conventional radiography is used primarily. However, vascular calcifications, gas shadows, and fecal material may pose difficulty in interpreting the images, and the fractures can be detected in only 40% of cases (13). Bone scintigraphy is the most sensitive diagnostic method (96%). Abnormal uptake may be detected 72 h after the onset of symptoms (14). MRI is preferred as a sensitive modality for detecting changes at spinal structures and bone marrow. Decreasing signal intensity on T1-weighted sequences and increasing signal intensity on T2 images indicate edema at the fracture line. Fat-suppressed and contrast-enhanced sequences may aid in the differential diagnosis by discriminating the pathologic structure (10,15). CT may show the fracture line; however, exposure to radiation and limited diagnostic information in evaluation of transverse fractures limit its usage (15). Conventional radiography of our patient revealed no pathological finding except for degenerative changes. MRI revealed widespread bone marrow edema at both the sacral wings and left iliac bone and signal recordings probably due to insufficiency fractures within this area.

Treatment of insufficiency fractures is generally conservative. Analgesics, bed rest, and reduced load are the first choices (5,6). Pain is controlled by analgesics and physical therapy methods. Osteoporosis and osteomalacia should not be overlooked (6,8). The patients should be mobilized as soon as possible to avoid the negative effects of immobilization. Early rehabilitation and loading exercises should be applied as much as can be tolerated by the patient (8,12). Assisted mobilization devices may be used if needed. Babayev et al. (6) stated that pelvic insufficiency fractures are rarely life threatening but may lead to functional disability and emphasized the importance of rehabilitation. In our patient, a nonsteroidal anti-inflammatory drug, electrotherapy, and hotpack treatment were used for pain control and range of motion exercises, progressive resistance exercises, and balance-coordination exercises were planned as physiotherapy program. 50000 IU 25 (OH) Vitamin D weekly was prescribed along with increased dietary calcium intake with close monitoring of serum calcium levels and in 8 weeks. The patient was able to do her routine daily activities at the follow-up examination 3 months after discharge.

Sacroplasty or surgical therapy is recommended when conservative treatment fails or when neurological loss and/or pelvic instability occurs (12). Our patient was successfully treated with conservative measures and thus did not require surgery.

In conclusion, we should always bear in mind the possibility of insufficiency fractures in differential diagnosis of elderly patients presenting with bone and muscle pain. Osteoporosis and osteomalacia should not be overlooked. Nonspecific symptomatology may delay the diagnosis, and diagnosis also may be easily overlooked by plain radiography, which will in turn increase the disability, because early diagnosis and proper rehabilitation may help in reducing functional disability.

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