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Sacral Insufficiency Fracture in a Patient with Ulcerative Colitis: Case Report

Ülseratif Kolitli Bir Hastada Sakral Yetersizlik Kırığı: Olgu Sunumu

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Summary

Sacral insufficiency fracture (SIF) is a type of stress fracture that occurs primarily in elderly women without definite trauma history. SIF generally presents as non-specific pelvic or low back pain and is often overlooked. SIFs are secondary to a number of conditions including postmenopausal osteoporosis, steroid-induced osteoporosis and rheumatoid arthritis, which decrease bone resistance and predispose the development of SIFs. Ulcerative colitis (UC) is recognized as a risk factor for development of osteoporosis, but the occurrence of sacral insufficiency fractures (SIFs) in patients with UC has not previously been well emphasized. We presented here an UC patient with a SIF, who was treated with conservative approach successfully. (Turkish Journal of Osteoporosis 2011;17:89-92)

Key words: Sacral insufficiency fracture, stress fracture, ulcerative colitis

Özet

Sakral yetersizlik kırığı (SYK), birincil olarak travma öyküsü olmayan yaşlı kadınlarda ortaya çıkan bir stres kırığı türüdür. SYK genellikle nonspesifik pelvik ya da bel ağrısı şeklinde kendini gösterir ve genellikle gözden kaçmaktadır. SYK kemik direncini azaltan ve SYK gelişimine zemin hazırlayan postmenopozal osteoporoz, steroide bağlı osteoporoz ve romatoid artriti içeren durumlara ikincil olarak ortaya çıkar. Ülseratif kolit (ÜK), osteoporoz gelişimi için bir risk faktörü olarak kabul edilmektedir, ancak daha önceleri ÜK'i olan hastalarda SYK bildirilmemiştir. Burada konservatif tedavi ile başarılı bir şekilde tedavi edilen SYK'ı olan ÜK'li bir olgu sunulmuştur. (Türk Osteoporoz Dergisi 2011;17:89-92)

Anahtar kelimeler: Sakral yetersizlik kırığı, stres kırığı, ülseratif kolit

Introduction

Stress fractures are classified into two categories. Fatigue fractures are caused by abnormal stresses applied to normal bone itself, while insufficiency fractures occur in abnormal bone already weakened by decreased mineralization and with insufficient elastic resistance to withstand normal physiological stress (1). Sacral insufficiency fractures (SIFs) are a well-defined subgroup of insufficiency fractures. Several pathological conditions may decrease bone resistance and predispose the development of SIFs. Among them, postmenopausal osteoporosis seem to be the most commonly associated condition (1,2). They are difficult to diagnose at an early stage since there is usually no significant history of trauma, often accompanied by concurrent lower lumbar degenerative pathology, and routine radiographs of the sacrum do not demonstrate them adequately (3). The incidence of SIFs remains unknown; however, it is apparent that SIFs are more common than widely appreciated and remained largely overlooked as a cause of pain and disability within elderly and other at risk populations.

Ulcerative colitis (UC) is one of the main forms of chronic inflammatory bowel disease (IBD) and is a nontransmural inflammatory disease with episodic progression that is restricted to the colon. Patients with UC can develop extra-intestinal manifestations. The most common types affect the musculoskeletal system, mostly including sacro-iliac joint (4). In the literature, UC is a recognized risk factor for development of osteoporosis (5). The mechanism for development of osteoporosis in UC patients is multifactorial, among them corticosteroid treatment seems to be major risk factor (5,6). Furthermore, recent researches have shown possibly an increased fracture risk in UC patients, mostly including vertebral colon due to decreased bone mineral density (7,8). However, co-occurrence of UC with SIFs has not been reported yet. We present an UC patient with a sacral insufficiency fracture, which is not described in the literature to our knowledge.

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Case Report

A 40-year-old woman was admitted to our outpatient unit complaining of sudden lumbosacral back pain, radiating pain to the left leg since two weeks prior to admission. Conservative treatment was tried at the local clinics for several days including non-steroidal anti-inflammatory drugs (NSAIDs), but there was no symptomatic improvement. She had not experienced any recent trauma. The pain was typically aggravated by weight bearing and relieved by rest, moreover, the pain was so severe that, at admission, she could not walk without using cane.

In her past medical history, she had been diagnosed with UC for 12 years. She had been treated with sulphasalazine (2 g/day for 12 years), corticosteroids (4 mg/day of prednisone during the previous 3 years) and NSAIDs (100 mg/day of indomethacin if required). A physical examination revealed sacral tenderness on the left side to palpation. Abduction and external rotation of left hip was limited due to pain. Other ranges of motion (ROM) of all joints were within normal limits. There were no signs of nerve-root compression or any neurological deficit. She did not complain morning stiffness.

Anteroposterior plain radiograph of the pelvis revealed irregular contour and erosions of the bones comprising both sacroiliac joints (bilateral stage 3 sacroiliitis) (Figure-1). In the laboratory investigations including total blood count, routine biochemical tests, immunoglobulins, complement levels, 25(OH) vitamin D3 and parathyroid hormone level were within normal range, rheumatoid factor was negative. Erythrocyte sedimentation rate (ESR) and C-reactive protein (CRP) were also in normal limits. T-score of the lower lumbar spine was -2.4, femur neck was -2.8 on bone densitometry. Initially, the patient's pain considered to belong to possible acute exacerbation of sacroiliac joints involvement due to UC. However, the normal levels of inflammatory markers, lack of morning stiffness and the nature of the pain made this less likely. Thus, magnetic resonance imaging (MRI) applied and showed low signal intensity on T1-weighted images and high signal intensity on T2- weighted images on the left sacrum wing, which is parallel to sacroiliac joint. T2-weighted images with fat suppression showed hypointense fracture line within hyperintense edema, which is demonstrated sacral insufficiency fracture in our patient. Bilateral sacroiliitis also revealed on MRI (Figure 2a, 2b).

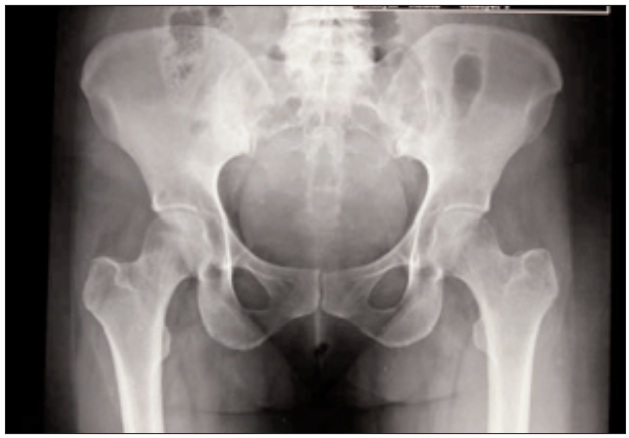

Figure 1. Anteroposterior radiograph of the pelvis shows no obvious fracture. Stage 3 bilateral sacroiliitis including irregular contour and erosions of the bones comprising both sacroiliac joints

After the diagnosis, the patient received paracetamol 2000 mg/day for the treatment of pain, and calcium carbonate 1000 mg/day and vitamin D3 800 IU/day daily for the treatment of osteoporosis. Transcutaneous electrical nerve stimulation was applied to the painful area. Range-of-motion (ROM) exercises for two weeks applied, followed by non-weight bearing crutch walking and muscle strengthening exercises. On the control visit at the third month, clinical outcome showed gradual pain relief, and she was able to walk without using crutch.

Discussion

SIFs often arise insidiously, resulting from axial stresses transmitted from the spine to a sacral ala with deficient elastic resistance. Sacral insufficiency fractures occur mostly in women aged >55 years, whose bone strength is inadequate to withstand normal repetitive stress. Several conditions may compromise bone density and strength, predisposing patients to SIFs; postmenopausal osteoporosis being the main cause among them. Corticosteroid-induced osteopenia and radiation therapy are also implicated as common risk factors for SIFs (9). Other underlying pathologies include: Cushing's syndrome, primary biliary cirrhosis, osteomalacia, rheumatoid arthritis, myelomatous disease, and endocrine disorders such as hyperparathyroidism (1,9).

Patients with UC have an increased prevalence of reduced bone mineral density compared with healthy controls (10). In the literature, the reported prevalence of osteoporosis/osteopenia varies from 7% to 56% in IBD (11,12). A population-based study of UC patients, estimated the prevalence of osteopenia at 15% and osteoporosis at 8% (13). Low bone density is receiving increasing attention as a complication of UC over the past decade since an increased risk of fractures among UC patients has been reported (5,7,8). In a study, it was suggested that there was a trend for an increased relative risk of vertebral fractures among patients with UC compared with control subjects (14). A large matched cohort study from Manitoba estimated that the risk of osteoporotic fracture in UC patients was increased 45% relative to that of matched control subjects, particularly of spine, hip and wrist (8). Co-occurrence of UC with SIFs has not been reported in the literature yet.

The precise incidence of sacral insufficiency fractures remains unknown. In a prospective study for 2 years by Weber et al, incidence in women aged over 55 years was 1.8% (15). In reality, the sacral insufficiency fractures may be more common than the usually expected because of diagnostic difficulties and vague clinical symptom and signs. The clinical presentation is often variable. Prominent features include sudden, intractable, low back or pelvic pain coupled with a significant reduction in mobility and independence. Symptoms are exacerbated by weight-bearing activity and generally improve with rest. The pain, which may be severe, radiate to the groin, buttocks, and thighs. Gait is usually slow and antalgic. Neurological examination is often unremarkable. Physical examination may reveal sacral tenderness on lateral compression. SI joint tests, although not specific for SIFs, are often positive (1,9).

Musculoskeletal system manifestations in UC are characterized by axial and/or peripheral joint involvement. The spectrum of axial involvement ranges from inflammatory lower back pain with or without radiological evidence of sacroiliitis (SI). Inflammatory back pain (IBP) in UC is usually difficult to localize, insidious in onset, frequently monolateral, more intense at rest, associated with stiffness but relieved by movement (4). In the studies prevalence of SI in UC patients ranged from 24.2% to 43%, and SI is one of the most frequent joint inflammations found in UC patients (16,17). SI seems more related to duration of UC. The clinical picture of SI includes IBP and stiffness, on the other hand it was suggested that asymptomatic SI might be present in 10% to 50% of patients with IBD (18). Exception of IBP and morning stiffness, additionally existing of acute onset of pain, which was aggravated by weight bearing, made the diagnosis of exacerbation of SI less likely, although the imaging investigations revealed SI in our patient.

The mechanism for development of osteoporosis in UC patients seems to be multifactorial. This decreased bone mineral density is thought to be mainly the result of treatment with glucocorticoids in IBD (10). Other causes include malabsorption, increased age and the inflammatory process itself (10,19). It has been suggested that the frequency of osteopenia in patients receiving >5 mg cumulative dose of prednisolone equivalents is higher compared to patients who received a lower dose which affect mainly the trabecular skeleton (20). However, Tsironi et al. showed that bone loss occurs more prominent at the cortical skeleton rather than trabecular skeleton. This is possibly an indicator that factors other than corticosteroid use play a significant role in bone loss in IBD including UC (19). Additionally some studies suggested that, any use of corticosteroids, increased duration of use, and increased cumulative dose were not associated with fracture risk in UC (21,22). Thus, inflammation may have a key role in the pathogenesis of osteoporosis in IBD patients. High levels of inflammatory cytokines such as tumor necrosis factor alpha and interleukin-1 beta in the gut mucosa and of interleukin- 6 (IL-6) in the peripheral blood of IBD patients have been related to decreased bone formation in experimental models. Tumor necrosis factor alpha has been proved to promote activation of osteoclasts by increasing the expression of RANK-L in osteoblasts. Furthermore, it directly inhibits apoptosis of osteoclasts and stimulates osteoblast apoptosis, thereby increasing bone resorption and reducing bone formation. IL-6 serum level has also been found to predict bone loss in postmenopausal women (10,23,24). In a study, a highly significant inverse correlation between IL-6 serum level and bone mineral density in UC patients was found (24). In our patient, corticosteroid treatment, and inflammation due to UC might have caused osteoporosis that lead to SIFs.

Initial diagnosis is difficult to be made. Blood test results are usually unremarkable, apart from mild-to-moderate elevation of the serum alkaline phosphatase level. Routine plain radiography of the lumbar spine and pelvis is limited because it generally shows no demonstrable fracture line at the beginning of symptoms, and the curved anatomy of the sacrum, demineralization of surrounding bone and overlying bowel gas may conceal the fracture lines (3). In addition, in our case, we could not find the fracture line at initial plain radiographs and there was no susceptible blood test result.

In a suspected case of the SIFs, MRI and bone scan are very useful. MRI is by far the most sensitive screening investigation. It can pick up signal from bone marrow edema that results from fracture inflammatory and reparation processes. T1-weighted images demonstrate low signal intensity while T2-weighted images demonstrate high signal intensity. T2-weighted short tau inversion recovery (STIR) images and T2-weighted images with fat suppression are particularly sensitive to demonstrate a fracture line. However, the appearance of low signal areas in T1- weighted sequences may mimic metastatic disease, and MRI may account negative findings at an earlier stage (25). Bone scintigraphy is a very sensitive method of determining the pathology and location of SIFs. The characteristic H-shaped or butterfly-shaped uptake pattern is seen generally within 72 h of onset of symptoms. But, the characteristic 'H-shape' configuration, representing a combined bilateral vertical and horizontal sacral fractures, was present in 15 to 68% of cases reported before (1,9). Additionally, bone scan results may be dismissed as sacroiliitis or metastatic disease, hence, a definitive diagnosis should not be based on scintigraphy alone (1,9). In our patient, characteristic fracture line on the left sacrum wing which is parallel to sacroiliac joint was detected, and lead us to the diagnosis of SIF. Bone scintigraphy was not performed.

SIFs are frequently bilateral and arise predominantly in the sacral ala parallel to the sacro-iliac joints, on the other hand unilateral involvement is not uncommon. Fractures usually display a vertical pattern extending parallel to the sacro-iliac joint and lateral to the sacral foramina; neurological deficits are therefore uncommon. Bilateral vertical sacral fractures are likely to be connected by a horizontal fracture through the upper sacral body, which progresses to the typical "H" pattern on imaging studies. This horizontal component of fracture is considered a secondary development caused by continued stress, and classified further according to the degree of angulation and displacement in the sagittal plain. Associated fractures are seen in 25 to 80% of patients having sacral insufficiency fractures. Pelvis biomechanics dictate that disruption of the bony skeleton at one site may lead to increased stresses in other parts of the pelvic ring, resulting in fracture. Pubic rami fractures are most common, followed by vertebral compression fractures and iliac wing fractures (1,9,26). In our patient, involvement was unilateral, and the fracture line was parallel to the left sacro-iliac joint. There was no associated fractures.

The majority of SIFs are treated conservatively with bed rest and NSAIDs, followed by gradual mobilization, in order to avoid complications of immobilization, with walking aids. This results in the recovery of the vast majority of patients as in our patient. Moderate weight-bearing exercises, within the confines of pain tolerance, have been suggested. During this period, transcutaneous electrical stimulation and ultrasound can provide symptomatic benefit. Following initial management of SIFs, anabolic or antiresorptive medication, calcium and vitamin D supplementation are often prescribed to reduce the risk of further insufficiency fractures (9). Pelvic insufficiency fractures rarely lead to pelvic deformity, operative management is suggested when severe instability of the pelvic ring is observed, or for the patients who failed with conservative treatment (27).

In conclusion, pelvic insufficiency fractures are uncommon in patients with UC, and they may be silent clinically. The presence

of SIF may also complicate the definite diagnosis. Although most patients with pelvic insufficiency fractures respond well to simple conservative treatments, continued stress can lead to associated fractures that may concluded in pelvic instability, which require operative management. In order to avoid misdiagnosis and improper treatment modalities, physicians should suspect possibility of the sacral insufficiency fracture in patients who are at risk of primary or secondary osteoporosis. The clinical course of the pain and selection of appropriate radiological investigation modalities will lead to more rapid and accurate diagnosis.

Disclosures: None

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