

Osteoporotic Insufficiency Fractures of the Pelvis Simulating a Malignancy in an Elderly Man

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Insufficiency fracture is a common initial presentation of osteoporosis. Fractures of the distal radius are the most common, followed by fractures of the hip, vertebral body, and proximal humerus.¹ Insufficiency fractures of the pelvis are rare, often unrecognized, and reported mostly in females.^{2,3} In their review of the literature, Weber and colleagues³ found that only 17 (7.4%) of 231 insufficiency fractures of the sacrum occurred in males. Although widely regarded as a disease of women, osteoporosis causes significant unrecognized morbidity and mortality in men.⁴ The absolute number of men presenting with osteoporotic fractures is rising because of an increase in the elderly population plus an age-related increased incidence of fractures.⁴ Awareness of this entity, particularly in males, should prevent its being confused with malignant disease, should prevent unnecessary extensive workup, and will allow appropriate management and patient reassurance.

In this article, we report the case of an elderly man who had multiple pelvic metachronous insufficiency fractures with worrisome radiologic features simulating a malignancy. The patient provided written informed consent for us to submit his case data for publication.

CASE REPORT

A man in his mid-70s was referred to our orthopedic oncology service for worrisome lesions on pelvic imaging studies. He complained of low back and groin pain increasing over

the preceding 3 months. Pain onset was insidious, and there was no antecedent trauma. The nonradiating pain worsened to the point that he required an assistive device at all times. He denied constitutional, urinary, and bowel symptoms. He had sustained left wrist and right shoulder fractures after trivial falls 6 and 4 months earlier, respectively, and was treated conservatively; there was no further workup or anti-osteoporotic supplementation. Past medical history included hypertension, thyroid surgery for goiter, and depression. Current medications included antihypertensive agents, a thyroxine supplement, and a selective serotonin reuptake inhibitor (SSRI).

This short patient had a kyphoscoliosis and a dorso-medial deformity at the left wrist. There was generalized tenderness over the lumbosacral spine, the sacroiliac joints, the trochanters, and the symphysis pubis. Spine movements were restricted and painful. Passive straight-leg raise, log roll, pelvic compression-distraction, and Gaenslen sign tests were painful on both sides. Hip rotations on both sides provoked pain in the groin. The distal neurovascular examination was normal.

Pelvic radiographs showed degenerative changes in both hips with protrusio and an undisplaced fracture of the left intertrochanteric region. Osteolysis was also evident at the right pubic and ischial rami (Figure 1). Computed

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Figure 1. Anteroposterior radiograph of pelvis shows osteolysis in right rami, degenerative joint disease with protrusio on both sides, and undisplaced fracture in left trochanter region. Sacral fractures are not appreciable.

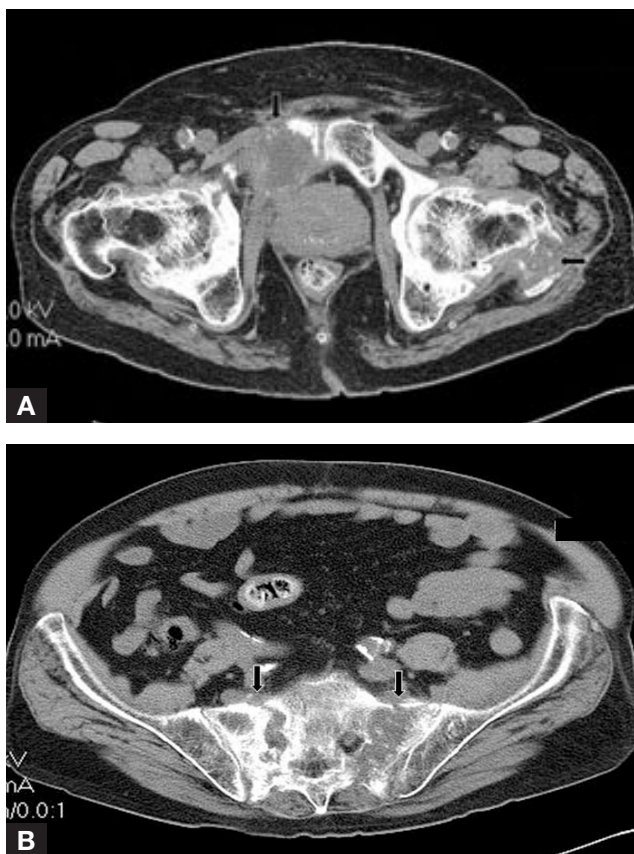


Figure 2. Computed tomography shows (A) osteolysis at right rami and 5×4×4-cm soft-tissue mass with attenuation similar to that of skeletal muscles (similar fragmentation and osteolysis are seen in left trochanteric region) and (B) longitudinal fractures on both sides of sacral body parallel to sacral alae and sacroiliac joints.

tomography (CT) confirmed these findings along with a 5×4×4-cm soft-tissue mass at the pubis with attenuation similar to skeletal muscles (Figure 2A). Additional longitudinal fractures, parallel to the sacral alae, were also found (Figure 2B). Magnetic resonance imaging (MRI) showed hypointense signal on T₁-weighted and hyperintense signal on T₂-weighted and short tau inversion recovery images at both sacral alae, the right rami, and the left trochanter (Figure 3). Technetium-99m hydroxymethylene diphosphonate (oxidronate sodium) bone scintigraphy showed increased uptake in the areas just mentioned and in the right proximal humerus, the left trochanter, and the left distal radius (Figure 4).

Given the radiologic findings, the possibility of a malignancy had been considered. At a different institution, results from a CT-guided biopsy of the pubis led to an inconclusive diagnosis (the reason for referral to our service). The laboratory workup, which included serum tumor markers, protein levels, 25-hydroxyvitamin D, parathyroid hormone, serum and urine electrophoresis, immunoglobulins, calcium, and phosphorus levels, was normal except for increase alkaline phosphatase (206 IU/L; normal, 20-125 IU/L) and increased erythrocyte sedimentation rate (36

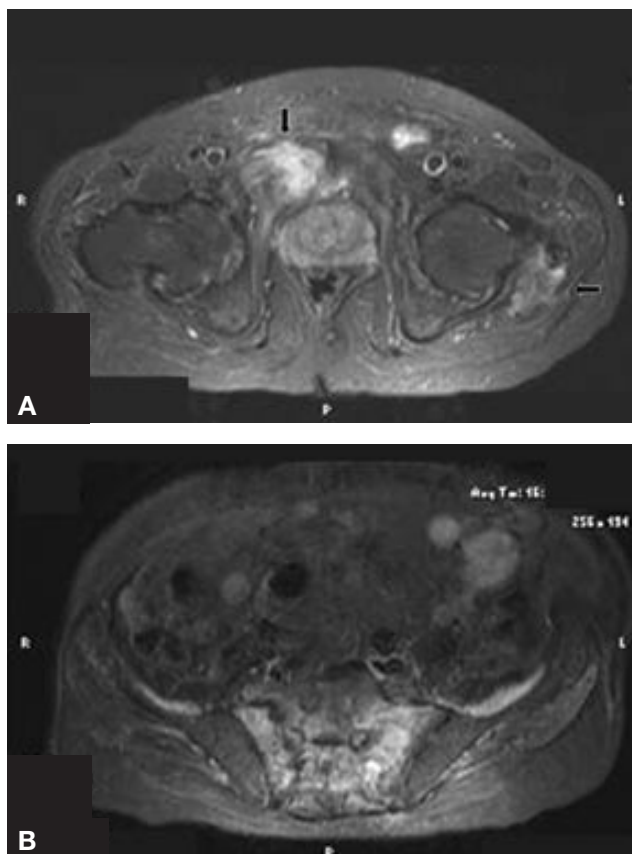


Figure 3. Magnetic resonance imaging shows hyperintense signal in (A) right rami and left trochanter and (B) bilateral sacrum on short tau inversion recovery axial image (TR 5878, TE 45).

mm/h; normal, ≤20 mm/h). Chest and abdomen CT did not show any other significant focus of disease. The pathology indicated a benign reactive process with focal fibrosis, hemosiderin-laden macrophages, calcifications, fibrin, and granulation tissue with no evidence of malignant cells (Figure 5). Dual-energy x-ray absorptiometry of the hip showed bone mineral density of 0.62 g/cm² with a T score of −3.6 and a Z score of −2.5 below the respective SDs.

Given the features mentioned, the patient was diagnosed with osteoporotic insufficiency fractures of the pelvis and was treated with gradual mobilization after 6 weeks of bed rest and osteoporosis treatment (bisphosphonates, calcium, and vitamin D supplements). Earlier, the patient had refused operative intervention for the trochanteric fracture. He improved dramatically and at 4 months was ambulating without an assistive device. Radiographs showed gradual healing of the pubic fracture. At last follow-up (18 months), he was asymptomatic except for hip discomfort after prolonged walking.

DISCUSSION

Insufficiency fractures of the pelvis remain an unsuspected but not uncommon cause of low back pain in the elderly.^{2,5} The most common presentation of an insufficiency sacral fracture is severe back or buttock pain, reported in 64% and 43% of patients, respectively, though up to 22% of

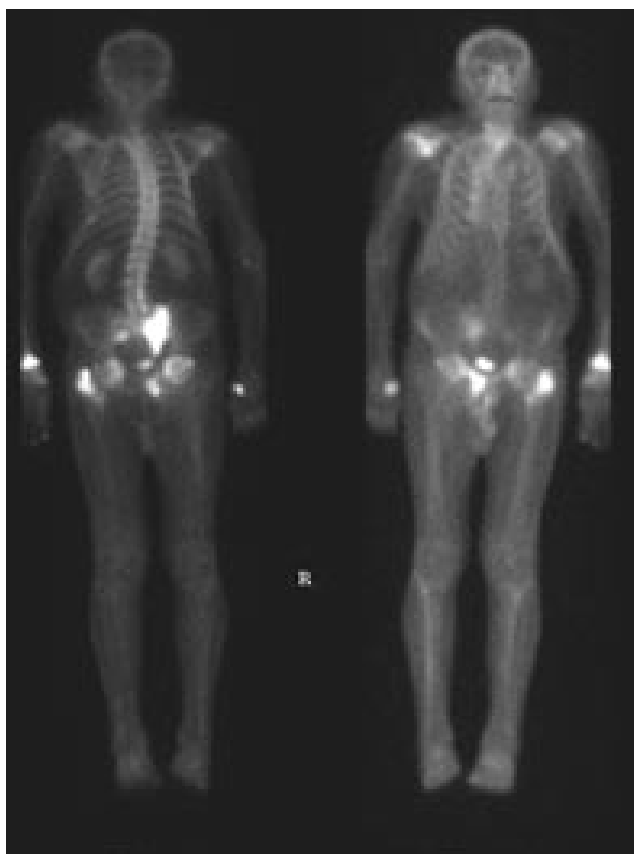


Figure 4. Technetium-99m hydroxymethylene diphosphonate (oxidronate sodium) bone scintigraphy shows increased uptake in right rami, sacrum, left trochanter, right proximal humerus, and left distal radius. Scoliosis is evident.

patients are thought to be asymptomatic at diagnosis.⁶ Antecedent minor trauma has been reported in 32% of cases.⁷ Approximately 80% of patients complain of sacral tenderness, and 14% have neural symptoms.⁷ Groin pain is common in symphyseal lesions. Most patients with pelvic insufficiency fractures respond to simple nonoperative modalities and become pain-free within a year. After a short period of bed rest and analgesics, management consists of gradual mobilization and antiosteoporotic treatment.⁸⁻¹⁰ Improvement and resolution usually take 2 weeks to 24 months (reported up to 131 months) and depend on the cause and associated fractures.^{7,11}

The group with the highest risk for pelvic insufficiency fractures is postmenopausal osteoporotic women who undergo local radiation, though there are other predisposing factors, including corticosteroid use, connective tissue disorders, obesity, multiparity, diabetes mellitus, metabolic bone disease, fibrous dysplasia, Tarlov cyst, spine deformities, total hip arthroplasty, excessive pelvic anteversion, and even pregnancy and lactation.^{3,5,7,9,10,12-16} Risk factors identified in our patient included old age with sedentary lifestyle, hypertension, hyperthyroidism and (later) thyroxine supplementation, and depression with SSRI intake.^{1,17-19}

The differential diagnosis of pelvic insufficiency fractures includes malignancy (primary, recurrent, or metastat-

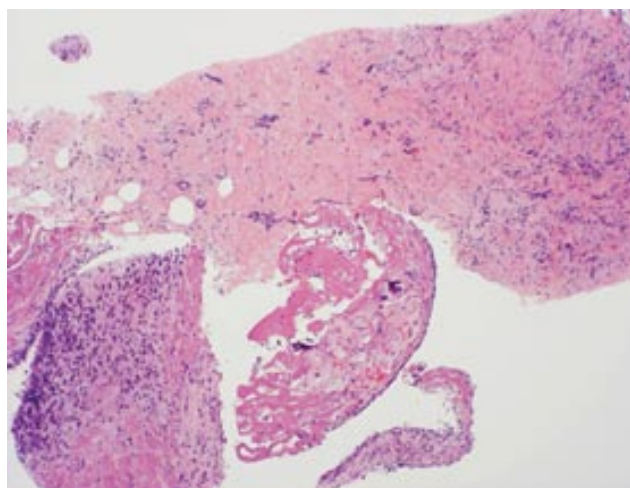


Figure 5. Computed-tomography-guided biopsy from right pubic ramus mass shows benign reactive process with focal fibrosis, hemosiderin-laden macrophages, calcifications, fibrin, and granulation tissue with no evidence of malignancy (hematoxylin-eosin, original magnification x40).

ic), lumbosacral radiculopathy, posttraumatic osteolysis, radiation-induced bone atrophy, radiation-induced sarcoma, and metabolic bone disease.²⁰ In patients with a history of malignancy and radiation, metastatic disease is often the first consideration, though differentiating features have been described.^{13,21,22} Degenerative disease of the spine is more common in this age group and thus attributed as a diagnosis in patients without a defined malignant lesion. In some patients, biopsy or serial imaging studies may be needed to confirm the diagnosis.^{13,20,21} However, biopsy should be done judiciously, as it can lead to infection and irreparable devitalized bone.^{13,23}

The diagnosis is difficult for several reasons.^{2,3,13,20} Findings on routine radiographs may be subtle and may be overlooked because of the complex anatomy and overlying bowel shadow and calcification. Stress fractures through cancellous bone, particularly in an osteopenic skeleton, are difficult to detect. A biopsy specimen from an insufficiency fracture may contain immature bone, cellular stroma, necrotic debris, and inflammation. These histopathologic findings out of context may be interpreted as a sarcoma or osteomyelitis. However, pelvic insufficiency fracture is an uncommon diagnosis simply because many are unaware of its existence.

Typically, the sacral fracture is vertical in the alae and parallel to the sacroiliac joint. Transverse fractures develop secondarily and can displace with time. They may also be located just lateral to the margins of the lumbar spine. This distribution suggests that body weight transmitting through the spine to the pelvis may be at least partially responsible for the fracture.² Presence of additional fractures in the spine or pelvis (up to 80%), particularly with a history of osteoporosis or local radiotherapy, helps confirm that the pathologic process in the sacrum is indeed a fracture.^{2,3,13,15,21,24} The high incidence of pubic fractures (up to 47%)⁷ in this group suggests that they are often concomitant with sacral fractures, though the sacral fractures have been reported to

occur earlier, and other fractures are thought to be caused by mechanical failure of the pelvic ring.^{13,15,24} Bilateral or multiple pelvic fractures can delay healing.²¹ Bone scintigraphy is the most sensitive study, and commonly it demonstrates an H-shaped increased uptake (Honda sign) in up to 45% of cases of sacral fractures (though other patterns have been described).^{7,15,16} CT, with proper window setting, further confirms and defines the fracture and excludes a destructive process. MRI findings are nonspecific but are helpful in demonstrating early medullary edema, fracture lines, and absence of a soft-tissue mass. Soft-tissue mass and osteolysis, which were found in our patient's case, are typically not features of insufficiency fracture but have been (rarely) reported in the pubis, the ribs, and the clavicle.^{2,12,13,22-26}

Osteoporosis must be considered in the differential for men just as often as for women, particularly in an elderly population. Results from 2 large studies have confirmed that similar factors affect bone density in both sexes.^{17,18} In a recent wrist insufficiency fracture study, Freedman and colleagues²⁷ found that, compared with males, significantly more females underwent diagnostic interventions and subsequent osteoporotic treatment—an indication that there is less awareness about male osteoporosis. Lifetime risk for any osteoporotic fracture at age 50 has been estimated to be 40% to 53% for women and 13% to 22% for men. Osteoporotic fracture is one of the most significant risk factors for a new fracture. There are a relative risk of 2 for a new fracture after any fracture and a relative risk of 4 for a vertebral fracture after a vertebral fracture.²⁸ In males, distal forearm fractures correlate with lower bone mineral density, and males specifically are at increased risk for osteoporosis.⁴ Of these males, 41.8% were found to be osteoporotic in at least one site (a finding comparable to that in studies of females with distal forearm fractures), and 51% had identifiable causes for osteoporosis.

CONCLUSIONS

Our patient's distal radius fracture, sustained 6 months before referral to our service, should have led to recognition of osteoporosis. Proper diagnosis and management at that time could have prevented other problems. A second insufficiency fracture, of the proximal humerus, was sustained within 2 months, but, again, the diagnosis of osteoporosis was missed. This resulted in still other insufficiency fractures, leading to more morbidity, more diagnostic uncertainty, and mental agony and eventually a surgical procedure. It is therefore imperative that orthopedic surgeons recognize osteoporosis in men, as well as women, who present with insufficiency fractures so that further morbidity and mortality can be prevented.

AUTHORS' DISCLOSURE STATEMENT AND ACKNOWLEDGMENTS

The authors report no actual or potential conflict of interest in relation to this article.

The authors thank Carlos A. Muro-Cacho, MD, and Enma Saiz, MD, for interpretation of our patient's histologic slides.

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