Sacral Insufficiency Fractures Following Pelvic Radiotherapy: Multimodality Approach For Discrimination From Metastatic Disease

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Citation

Abstract
We report the case of a 49 year old female patient who was referred for low back pain radiating into buttocks. The history of the patient revealed radical histerectomy and a total of 60 gy irradiation that was carried out for grade III endometrial adenosquamous carcinoma. Despite the suspicion of a pelvic bone metastasis computed tomography, magnetic resonance imaging and bone scintigraphy revealed the actual diagnosis as sacral insufficiency fracture. The fractures detected in our case were active on one side and chronic on the other side of the sacral alae. Insufficiency fractures of the sacrum are not uncommon and usually occur in osteoporotic bone with minimal or unremembered trauma. However, they appear to be relatively under-diagnosed and this case report aims to highlight the condition, discuss the expected imaging features and discrimination from metastatic disease.

INTRODUCTION
Insufficiency fractures of the sacrum (SIF) are not uncommon and result from a normal stress acting on bone with deficient elastic resistance [1]. Although there have been many reported etiologies of insufficiency fractures, osteoporosis is generally accepted as being the most common underlying disease. Other potential risk factors include rheumatoid arthritis, corticosteroid therapy, fibrous dysplasia, Paget's disease, osteogenesis imperfecta, Tarlov cysts and pelvic irradiation [2,3,4]. A high index of suspicion is required in order to identify fractures of this nature. Since they appear to be relatively under-diagnosed this case report aims to highlight the condition, discuss the expected imaging features [5].

CASE REPORT
A 49-year-old woman was referred for pelvic magnetic resonance (MR) imaging performed for the assessment of low back pain radiating into buttocks. Clinically there was tenderness over both sacroiliac regions. The history of the patient revealed radical histerectomy and iliac lymph node dissection that was carried out for grade III endometrial adenosquamous carcinoma 13 months ago. Besides surgery, a total of 60 gy radiotherapy was performed to the pelvic region. Unfortunately she has also developed tumor-node-metastasis (TNM) stage two (T1N1M0) left breast adenocarcinoma treated by conservative surgery followed with radiotherapy and six times cyclophosphamide, doxorubicin, 5-fluorouracil (CAF) chemotherapy five months ago. The patient was referred to our department for evaluation of the pelvic region following whole body bone scintigraphy with 99M TC methylene disphosphonate. Pelvic anteroposterior radiograph was unremarkable. Computed tomography (CT) (Figure-1), and MRI of the pelvis were performed.
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**Figure 1**
Figure 1: Axial bone density CT image shows bilateral sacral wing fracture lines (arrows). The bone density surrounding the fracture lines is osteoporotic on the right side whereas sclerotic on the left.

**Figure 2**
Figure 2: Axial SE T1 WI reveals the medullary edema on the sacral wings as hypointense signal changes. The fracture line is visible on the left (arrows).

**Figure 3**
Figure 3: Axial T2 W image, obviously demonstrates the fracture lines (arrows) and bilateral sacroiliac joints (dashed arrows).

MR imaging was performed on a 0.2T open MRI unit with axial spin echo (SE) T1 (Figure-2), coronal TSE T2 (Figure-3), and short tau inversion recovery (STIR) (Figure-4) sequences.

**Figure 4**
Figure 4: Coronal STIR WI reveals the apparent hyperintense medullary edema on the right side. However a little edematous signal is also visible on the left.
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Figure 5

Figure 5: Whole body bone scintigraphy with 99M TC methylene disphosphonate delineates the H sign on sacral wings that is believed to be pathognomonic for SIH (arrows).

CT showed bilateral sacral wing fractures. The bone density neighbouring the fracture lines on the right side was decreased whereas increased on the left side. This finding was significant since we could classify the fractures as active and chronic respectively. MR images revealed bone marrow edema apparent on the right side and also the fracture lines. The fracture lines were best visualized on T2-weighted images (T2WI). Nuclear scintigraphy showed a lesser uptake than the right side designating the chronicity of the left sacral wing fracture. We could also detect The H sign on the sacrum on 99M Tc scintigraphy image which is believed to be a specific finding of SIF (Figure-5. Since the final diagnosis was SIF, operative intervention was thought to be unnecessary and management consisted of admission to hospital for bed rest, adequate pain control with analgesics and physical therapy. The pain resolved after three weeks.

DISCUSSION

Insufficiency fractures of the sacrum have been increasingly recognized in the past 2 decades [1]. These fractures result from a normal stress acting on bone with deficient elastic resistance. Reduced bone integrity can result from many factors, the most common being postmenopausal or corticosteroid induced osteoporosis and radiation therapy. As a result of the altered internal architecture of the sacrum, insufficiency fractures may arise either insidiously or as the result of minor trauma [1]. Other potential risk factors include rheumatoid arthritis, fibrous dysplasia, Paget's disease, and total hip prosthesis [2]. Patients with insufficiency fractures often present with symptoms such as lower back, groin, or pelvic pain. These symptoms are nonspecific and may mimic other clinical conditions, such as disc disease, recurrence of a local tumour, or metastatic disease.

A high index of suspicion is required in order to identify fractures of this nature[1,5-8].

SIF is a subgroup of Sacral stress fractures. Stress fractures result from skeletal failure resulting from submaximal repetitive forces over time. They occur primarily in two populations, young active persons and elderly osteoporotic women, usually corresponding to fatigue and insufficiency-type fractures, respectively. Sacral fatigue stress fractures are very uncommon, representing less than 2% of all stress fractures in one series of 320 athletes [9]. Fatigue stress fractures occur when bone of normal structure and mineral content undergoes excessive impact loading or muscular forces, whereas insufficiency fractures occur when bone of abnormal structure or deficient mineral content is subjected to normal physiologic stresses [10]. The clinical presentation of these fractures is similar, but the medical and rehabilitation management of these patient populations differs and is tailored to the specific underlying etiology. In both types of fractures, appropriate conservative measures generally result in good functional outcomes [11].

Diagnosis of SIF cannot be readily made from its nonspecific clinical features. Consequently some cases may be mistaken for bone metastases, causing patients to receive inappropriate diagnostic measures and management. Recognition of the spectrum of imaging features is the basis...
of correct diagnosis and treatment [13].

Fractures in the sacrum are difficult to diagnose, as plain radiographic findings are either unhelpful or misleading. These fractures are often bilateral and occur mostly in the sacral wings, paralleling the sacro-iliac joints. The sacral bodies may be involved. Their imaging features vary depending on the duration and the degree of healing. CT is helpful for confirming the presence of fractures in cases with atypical scintigraphic patterns, particularly in those with a known primary malignant neoplasm. CT is especially useful in the further evaluation of parasymphysial and pubic rami lesions [13]. CT can exclude destructive lesions such as malignancy and osteomyelitis, by demonstrating the trabeculae to be intact, apart from disruption at the fracture site. If the diagnosis is not certain, or if there is concern regarding healing, follow-up CT scanning after several months is useful.

MRI demonstrates low signal intensity on T1WI and high signal intensity on T2WI due to edema within the bone marrow of the sacral wings. STIR images are particularly sensitive. The fracture line is sometimes seen, but this is not usual. MR is sensitive, but usually non-specific and there may be enhancement after IV gadolinium chelate and the fracture may involve one or more sacral bodies. On SE T1WI, the hypointense signal of edema may mask a fracture line. On SE T2WI the fracture line can more clearly be detected within the hyperintense edema. Also, in the present case, the fracture line was best visualized on T2WI. In a study with 20 patients that was carried out by Grangier et al the earliest MR sign was medullary edema, seen as early as 18 days after the onset of symptoms [13]. In another study Kanberoglu et al have performed, the activity was assessed with MRI. Depending on the presence or absence of signal intensity around the fractures, the lesions were grouped into active and chronic forms [13]. According to this study our case had active SIF on the right side and chronic on the left side of sacral wings.

Bone scintigraphy is sensitive for suspected insufficiency fractures of the sacrum. Because the lateral masses are usually the site of these fractures and the sacral bodies are relatively spared, the isotope uptake classically produces a butterfly or ‘H’ pattern, which is considered diagnostic in the right setting. However, the fracture may be unilateral, with or without a horizontal bar to the ‘H’ and there may be extension into the iliac crest and other pelvic fractures and the classic ‘H-sign’ may only be seen in as few as 40% of cases, limiting specificity. The H-sign may be asymmetric [13]. An important problem is the discrimination of these fractures from metastases in the patients with known pelvic malignancies. CT and MRI can exclude the presence of a destructive process and an associated soft tissue mass, as would be seen in metastatic disease. Also other metastatic lesions of the neighbouring bony structures are supportive. If insufficiency fractures are identified in the typical anatomic locations, bone biopsy is unnecessary [13].

In fact the management of the SIF consists of bed rest and pain relief, Pommersheim et al have recently reported a new technique and they have achieved relief of the symptoms immediately by treatment of the fractures by polymethylmethacrylate injections, a so-called sacroplasty [13].

In conclusion, awareness of this clinical entity and of the high sensitivity of MRI for demonstrating edema caused by the fracture should prevent confusion with metastatic disease and inappropriate treatment. Besides MRI can determine the clinical activity of the disease, and can monitor the response to treatment of the active type of insufficiency fractures. The cases presenting with non-specific MRI findings should be evaluated with multimodality approach.

References
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