Insufficiency fracture of the supra-acetabulum that required differentiation from a pathological fracture secondary to a malignant bone tumor: a case report

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Abstract
Background: The supra-acetabulum is a common site for malignant bone tumors, which can be difficult to differentiate from insufficiency fractures. We report a rare case of a stress fracture of the supra-acetabulum that required differentiation from a malignant bone tumor.

Case presentation: A 74-year-old Japanese man presented to the hospital because of right hip joint pain. X-rays showed no obvious abnormalities. Magnetic resonance imaging showed an abnormality in the right supra-acetabulum, and he was referred to our department. A linear, low-signal region and its surrounding equal signal region were observed at the same site in the T1-weighted image, and a linear low-signal region and high signal region were observed in the surrounding area in the T2-weighted image. On the contrast-enhanced magnetic resonance imaging, the lesion was still unclear and the whole area was gradually enhanced. A computed tomography-guided needle biopsy was performed, but no tumor cells were observed, therefore the lesion was presumed to be a fracture healing. The bone density was 66% for the lumbar spine (young adult mean, L2–4), and blood biochemistry showed an increase in alkaline phosphatase and total type I procollagen N-terminal propeptide.

Conclusion: This case was diagnosed as an insufficiency fracture of the supra-acetabulum in a male patient with primary osteoporosis by biopsy specimen. Initially, a pathological fracture associated with a malignant lesion was considered. On magnetic resonance imaging, the boundary around the fracture line was unclear and a signal change that was gradually enhanced by gadolinium was observed. This is likely to be bone marrow edema associated with the stress fracture, and we believe this to be a useful finding that may help in differentiating a stress fracture from a pathological fracture secondary to a malignant lesion.

Keywords: Supra-acetabulum, Insufficiency fracture, Malignant bone tumor
Supra-acetabular fractures due to bony insufficiency in men are not common [3].

In addition, supra-acetabulum is more commonly the site of primary malignant bone tumors and bone metastases from other malignant tumors [4]. Sakamoto et al. [1] described that a biopsy for the supra-acetabular lesion was needed for the purpose of diagnosis and to rule out a neoplastic lesion. We present a case of supra-acetabular IF that was difficult to diagnose.

**Case presentation**

A 74-year-old Japanese man presented to the previous hospital complaining of right hip pain. There was no history of trauma. His history included appendicitis and hypertension. X-rays showed no obvious abnormalities (Fig. 1), so further examinations were performed. The computed tomography (CT) scan showed evidence of sclerosis with a periosteal reaction in the right supra-acetabulum (Fig. 2). Magnetic resonance imaging (MRI) with T1 weighting showed a linear low signal and an equivalent signal surrounding it at the right supra-acetabulum, and the linear low signal and high signal surrounding it were seen on the T2-weighted image. The lesion was not suppressed by short TI inversion recovery. Contrast-enhanced MRI showed the lesion gradually enhanced (Fig. 3). A malignant bone tumor at right supra-acetabulum was suspected by CT and MRI, and he was referred to our department. Blood testing was carried out for differential diagnosis considering bone metastasis from carcinoma or secondary osteoporosis. There were no inflammatory findings in the blood results, nor were there any abnormalities in his electrolytes, thyroid hormones, or tumor markers. Moreover, nuclear examinations were performed to distinguish between benign or malignant and to check whether there were other bone lesions. A bone scintigram showed accumulation in the right supra-acetabulum. Positron emission tomography showed accumulation of standardized uptake value (SUV)max 3.5 in the right supra-acetabulum.

The differential diagnoses considered were a primary malignant bone tumor such as osteosarcoma, a right supra-acetabular metastasis, and a right supra-acetabular IF. A bone biopsy was performed for diagnosis. The CT-guided needle biopsy showed no tumor cells, but there was formation of osteoids and infiltration of inflammatory cells, indicating the healing process after a fracture (Fig. 4). The pathology confirmed an IF of the right supra-acetabulum.
We performed some follow-up imaging to examine the patient for osteoporosis. The L2–L4% young adult mean (YAM) was 66% (normal value ≥ 80%), which suggested osteoporosis. Blood results included alkaline phosphatase (ALP) level at 558 IU/L (normal value 50–350), and the total type I procollagen N-terminal propeptide (totalP1NP) was 115 µg/L (normal value 18.1–74.1). These results are compatible to a healing fracture.

The patient was started on oral bisphosphonate once a month from 2 months after the first visit. Four months after the first visit, he no longer complained of hip pain and sclerosis could be seen on images of the right supra-acetabulum (Fig. 5). X-ray at 2 years after the first visit showed the shadow of the sclerosis faded.

No adverse and unanticipated event occurred.

Discussion

IFs are caused by normal stresses for bone with weakened mechanical strength [5]. They commonly occur in the spine, pelvis, and lower extremities. In the pelvis, they are produced in the pubis, ilium, and sacrum [6]. Supra-acetabular IFs are rare, and this case of an IF of the supra-acetabulum required differentiation from a pathological fracture due to a malignant bone tumor.

Most IFs occur in older women with postmenopausal osteoporosis. Rheumatoid arthritis, radiation therapy, and steroid therapy are also risk factors [7]. Osteoporosis affects women four times more frequently than men [8, 9], and it is said that osteoporosis in males is more often secondary osteoporosis [10, 11]. In this case, there were no electrolyte or hormonal abnormalities, and it was suspected that the less common male primary osteoporosis was the underlying cause of the fracture.
Supra-acetabular IFs were firstly described by Cooper et al. [12]. There are some reports that it was difficult to distinguish from malignant bone tumors by X-ray or MRI [1, 13]. On contrast-enhanced MRI, malignant bone tumors are enhanced early [14, 15]. In this case, there were some MRI findings: fracture line curvilinear superior acetabulum; low signal intensity on T1- and T2-weighted images; significant bone marrow edema; the enhancement increased only gradually; the unclear border of the contrasted edge; and absence of associated soft tissue masses. These findings may be features for the differentiation of stress fractures from malignant bone tumors.

Conclusion
This case of an IF of the supra-acetabulum required differentiation from a pathological fracture due to a malignant bone tumor. We propose that the signal change that was gradually lightly enhanced on contrast-enhanced MRI may be useful in differentiating IF from pathological fractures secondary to malignant bone tumors.

Author contributions
SM, NY, KH, AT, SM, KI, HY, YA, SS, and HT determined the treatment plan. SM and NY conducted the follow-up. SM was a major contributor in writing the manuscript. HT oversaw the study. All authors read and approved the final manuscript.

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Availability of data and materials
Medical imaging data will not be shared, because it is not fully anonymous.

Declarations
Ethics approval and consent to participate
This study was approved by the Medical Ethics Committee of Kanazawa University. Participant outcomes were collected independently from the participant with written informed consent.

Consent for publication
Written informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

Competing interests
The authors declare that they have no competing interests.

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References
1. Sakamoto A, Yamamoto T, Tanaka K, et al. Insufficiency fracture in the para-acetabulum, with features mimicking those of a malignant bone tumor. Clin Med. 2008;1:73–6.
2. Robinson SP, Hammoud S, Sculco TP. Insufficiency fracture of acetabular medial wall. J Arthroplasty. 2002;17:68–70.
3. Rosa MA, Maccauro G, D'Arienzo M. Bilateral acetabular fracture without trauma. Int Orthop. 1999;23(2):120–1.
4. Brown TS, Salib CG, Rose FS, et al. Reconstruction of the hip after resection of periacetabular oncological lesions: a systematic review. Bone Joint J. 2018;100:22–30.
5. Pentecost RL. Fatigue, insufficiency, and pathologic fractures. JAMA. 1964;187:1001.
6. Cooper KL. Insufficiency stress fractures. Curr Probl Diagn Radiol. 1994;23:29.
7. Schreiber S. Insufficiency fracture of the acetabulum. Clin Rheumatol. 1992;11:440–2.
8. Center JR, Nguyen TV, Schneider D, et al. Mortality after all major types of osteoporotic fracture in men and women: an observational study. Lancet. 1999;353:878–82.
9. Feldstein A, Elmer PJ, Orwoll E, et al. Bone mineral density measurement and treatment for osteoporosis in older individuals with fractures: a gap in evidence-based practice guideline implementation. Arch Intern Med. 2003;163:2165–72.
10. Riggs BL, Melton LJ, Robb RA, et al. A population-based assessment of rates of bone loss at multiple skeletal sites: evidence for substantial trabecular bone loss in young adult women and men. J Bone Miner Res. 2008;23:205–14.
11. Orwoll ES. Osteoporosis in men. Endocrinol Metab Clin North Am. 1998;27:349–67.
12. Cooper KL, Beabout JW, McLeod RA. Supra-acetabular insufficiency fractures. Radiology. 1985;157:15–7.
13. Theodorou SJ, Theodorou DJ, Schweizer ME, et al. Magnetic resonance imaging of para-acetabular insufficiency fractures in patients with malignancy. Clin Radiol. 2006;61:181–90.
14. Moulopoulos LA, Maris TG, Papanikolaou N, et al. Detection of malignant bone marrow involvement with dynamic contrast-enhanced magnetic resonance imaging. Ann Oncol. 2003;14:152–8.
15. Kayhan A, Yang C, Soylu FN, et al. Dynamic contrast-enhanced MR imaging findings of bone metastasis in patients with prostate cancer. World J Radiol. 2011;28:241–5.

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