
CASE REPORT

Post-traumatic Postoperative Osteolysis: an Under-recognised Cause of Recrudescence of Osteolysis in Quiescent Osteoblastic Pagetic Bone

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ABSTRACT

Post-traumatic postoperative osteolysis is a rare cause of recrudescence of osteolysis in previously quiescent sclerotic pagetic bone. This condition can develop rapidly following trauma and operation, and mimic sinister causes like malignancy. Awareness of this rare disease entity can help in prompt diagnosis and treatment for reversal of osteolysis. Here we report on a middle-aged man who developed post-traumatic postoperative osteolysis over the pagetic humerus after internal fixation for pathological fracture. To the best of our knowledge, this is the first reported case for this condition to occur in the humerus.

Key Words: Osteitis deformans; Osteolysis; Osteosclerosis

中文摘要

創傷後術後的骨質溶解：於已靜止骨質硬化的變形性骨炎，骨質溶解重現的一個罕見病因

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創傷後術後的骨質溶解是已靜止骨質硬化的變形性骨炎骨質溶解重現的一個罕見病因。病症可於創傷及術後迅速發展，易被混淆為一些如癌症等更嚴重的病因。認識此罕有病症有助及時診斷並讓病人接受逆轉骨溶解的治療。本文報告一名中年男性因肱骨變形性骨炎導致病理性骨折，於接受內固定手術後出現創傷後術後骨質溶解。據我們所知，這是首宗發生在肱骨的類似病例。

INTRODUCTION

Paget's disease of the bone (osteodystrophia deformans, osteitis deformans) is a chronic bone disorder characterised by disruption in osteoclastogenesis leading to excessive bone remodelling. Paget's disease of the bone can be monostotic or polyostotic. The

radiological finding is a continuum from osteolysis to osteosclerosis, representing evolution from the active phase, mixed phase to the inactive phase. Sometimes, recrudescence of osteolysis occurs in osteoblastic pagetic bone, which is usually related to neoplasm or osteomyelitis. Post-traumatic postoperative osteolysis

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is an under-recognised disease entity that can develop following trauma or operation. Here, we report a middle-aged male patient who developed post-traumatic postoperative osteolysis over a pagetic humerus after internal fixation for pathological fracture.

CASE REPORT

In April 2013, a 59-year-old man presented to the orthopaedic surgeons with acute right upper arm pain and deformity following a minor fall. His clinical history was unremarkable. On physical examination, the mid portion of his right arm was tender and slightly swollen. He had no fever. Apart from his alkaline phosphatase level, which was elevated to 199 U/L (reference range, 56-119 U/L), the results of the blood tests were normal, including white blood cell count, erythrocyte sedimentation rate (ESR), C-reactive protein (CRP), urate and creatinine levels, calcium and phosphate levels, thyroid function test, and tumour marker level (α -fetoprotein, carcinoembryonic antigen, prostate-specific antigen). There was no abnormal band in protein



Figure 1. An anteroposterior radiograph of the right humerus taken at presentation shows sclerosis, cortical thickening, and coarsened trabeculation at the distal two-thirds of the humerus to the distal subarticular region, with a displaced transverse fracture at the mid shaft.

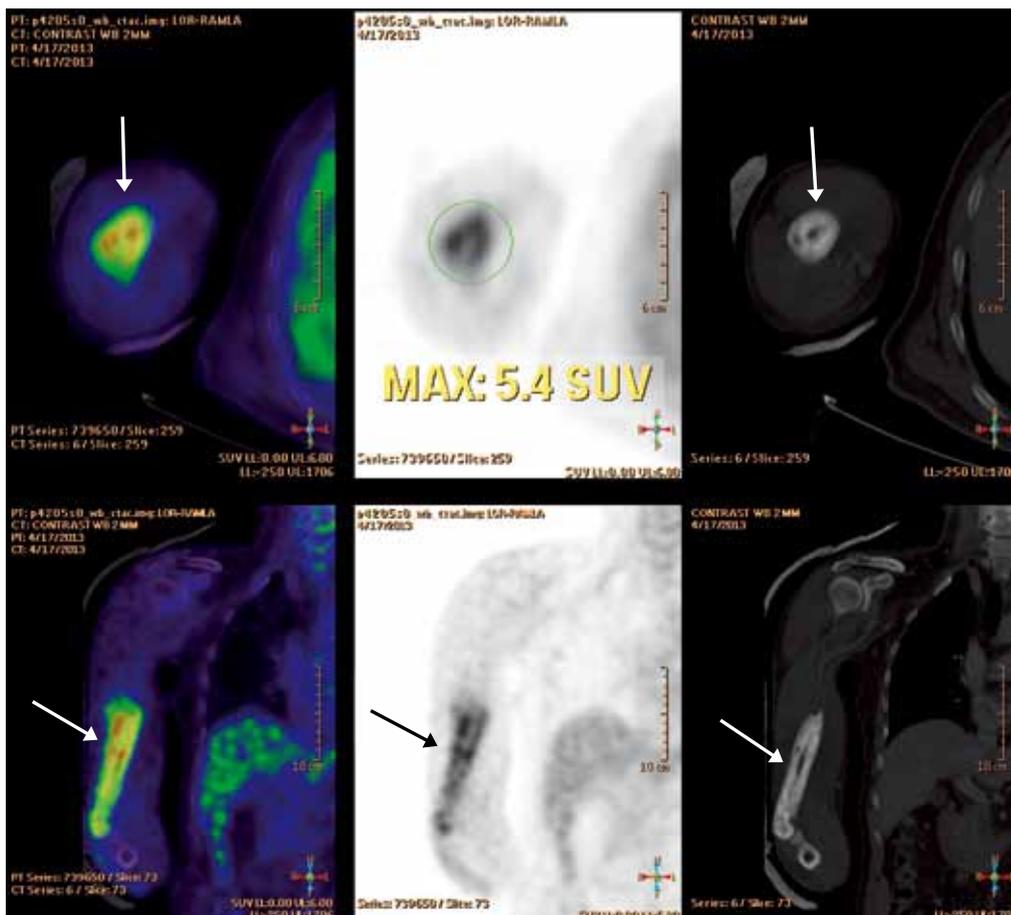


Figure 2. Positron emission tomography-computed tomography shows slightly elevated ^{18}F -fluorodeoxyglucose uptake distal to the fracture site (arrows). There is no associated soft tissue mass.

electrophoresis. Radiographs showed sclerosis, cortical thickening, and coarsened trabeculation at the distal two-thirds of the humerus to the distal subarticular region (Figure 1). A displaced transverse fracture was seen at the mid shaft of the humerus. There was no periosteal reaction, sequestrum, involucrum, or associated soft tissue mass. Overall features were indicative of sclerotic phase of Paget's disease at the humerus, complicated by pathological fracture. Radiographic skeletal survey showed monostotic involvement. Positron emission tomography-computed tomography showed slightly elevated ^{18}F -fluorodeoxyglucose uptake distal to the fracture site and there was no associated soft tissue mass (Figure 2).

Closed reduction and internal fixation of the humerus with an intramedullary nail was performed 11 days after presentation. The distal fragment was noted to be extraordinarily hard during operation. Histology of the reaming material showed disjointed trabecular bone in a mosaic jigsaw puzzle pattern without obvious osteoclastic bone resorption. These features were in keeping with Paget's disease in the inactive phase.

In the postoperative radiographs taken 1 day after operation, subtle osteolysis was evident around the



Figure 3. An anteroposterior postoperative radiograph of the humerus taken 1 day after closed reduction and intramedullary nail insertion. Subtle osteolysis around the fracture is evident.

fracture (Figure 3). The osteolysis then progressed rapidly and excessively, and peaked at 4.5 weeks after operation. There was almost complete bone loss at the portion of humerus distal to the fracture (Figure 4). Only mild osteolysis was evident proximal to the fracture. Despite the apparent extensive destruction in the radiographs, the patient only had mild upper limb weakness. There was no evidence of infection. The upper arm bruise and swelling gradually resolved. In the serial radiographs, alignment of the humerus was maintained and there was no implant failure. The patient started treatment with pamidronate infusion 90 mg/day for 3 days, and physiotherapy for active and passive mobilisation. In the follow-up radiographs taken 6.5 weeks after operation, there was evidence of bone remineralisation (Figure 5). The bone mass has almost fully reconstituted in the radiograph taken 5 months after operation (Figure 6). Overall, the features supported the diagnosis of post-traumatic postoperative osteolysis.

DISCUSSION

In the active phase of Paget's disease, osteolysis starts



Figure 4. (a) Anteroposterior and (b) lateral radiographs taken 4.5 weeks after operation. There is almost complete bone loss distal to the fracture.



Figure 5. An anteroposterior radiograph of the humerus taken 6.5 weeks after operation shows evidence of bone remineralisation (arrows).



Figure 6. An anteroposterior radiograph of the humerus taken 5 months after operation shows the almost fully reconstituted humerus.

from the subarticular region and progresses towards the other end in long bones. The leading edge is characteristically sharp with a flame or wedged shape, the so-called ‘blade of grass’ appearance.¹⁻³ The mixed phase is most commonly encountered in radiology practice, in which there is coarsened trabeculation and cortical thickening from osteoblastic activity, which can be associated with bone enlargement.^{1,4,5} In the inactive phase, sclerosis develops at a variable rate and obliterates areas of bone with coarsened trabeculations. Bone enlargement is most common at this stage.^{1,4,5} Similar to the histological changes, the occurrence and progression of the radiological stages can vary at different regions of the involved bone(s).^{1,4-6}

The recrudescence of osteolysis in osteoblastic pagetic bone is a concern. The best-known cause is neoplasm, which can be caused by malignant transformation, giant cell tumour, concurrent malignancy or metastasis.¹ Another consideration would be osteomyelitis, in which there would be clinical evidence of sepsis or local infection. Massive osteolysis can also happen after immobilisation in traumatised pagetic bone.⁷ The occurrence of spontaneous rapid extensive osteolysis

in pagetic femora following arthroplasty, revision arthroplasty, or internal fixation has been reported.⁸⁻¹⁰ Bisphosphonates are widely regarded as the medical treatment of choice for Paget’s disease.¹¹ These drugs induce apoptosis of osteoclasts and inhibit osteoclastic activity via inhibition of enzymes of the mevalonate pathway, thus reversing osteolysis.

The differential diagnosis of recrudescence of osteolysis in pagetic bone depends on the clinical, laboratory, and radiological findings. In patients with malignancy or osteomyelitis, there should be constitutional and / or local symptoms and signs.¹⁰ Elevation of white blood cell count, ESR, and CRP support a diagnosis of osteomyelitis. On imaging, both of these conditions can be associated with periosteal reaction, soft tissue mass, and swelling.¹ Computed tomography and magnetic resonance imaging are more sensitive than radiography for detecting these findings.

CONCLUSION

In post-traumatic postoperative osteolysis, the osteolysis can be rapid and massive.⁸⁻¹⁰ Awareness of this rare entity helps in early diagnosis and commencement of correct treatment, and reversal of osteolysis.

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