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AN UNUSUAL CASE OF PATHOLOGICAL FRACTURE DUE TO OSTEOID OSTEOMA IN THE DIAHYSEAL REGION OF HUMERUS: A CASE REPORT

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ABSTRACT

Introduction: Osteoid osteoma is reported to occur in the cortex of the shafts of long bones in 80-90% of cases. It is also reported in the epiphyseal and metaphyseal regions of both small and large bones of the axial and appendicular skeletons, especially the femur, tibia, and humerus. We report a case of pathological fracture found to have an osteoid osteoma in the diaphyseal region of humerus. **Case Presentation:** A 20 years male was admitted in our institute with complaints of severe pain in right arm with inability to move the shoulder and elbow since last evening. There was a history of trivial trauma prior to this complaint. Radiographs revealed a fracture at the middle one third of right humerus. There was a suspicious widening of the bone around the fracture area. Patient was taken up for operative fixation of the fracture. Open reduction and internal fixation with locking compression plate was performed. Intra-operatively, it was found that the bone was very brittle and cortex was paper thin. Excision biopsy of the pathological bone was performed. The fixation was augmented with a fibular strut bone graft and cancellous bone grafts from the iliac crest. Post-operatively, the fixation was augmented with a plaster slab. Biopsy reported histological features of osteiod osteoma with areas of bloody infiltrates - presumably from fracture haematoma. **Conclusion:** Osteoid osteoma is not a very uncommon condition. But there may be unusual presentations. Radiologically, a classical thickening of the bone cortex with nidus may not be appreciable in all the cases, as was seen in our patient. Hence, histopathological examination of bone fragments from all suspicious fractures must always be done.

KEYWORDS: Pathological fracture, osteoid osteoma.

INTRODUCTION

Osteoid osteoma is a small, benign but painful lesion with specific clinical and imaging characteristics. It is a benign osteoblastic tumour that Bergstrand first described in 1930.[1] Jaffe described it in 1935 and was the first to recognize it as a unique entity.[2] It represents 2 – 3% of all bone tumours and 10 to 20% of all benign bone tumours. In a Mayo Clinic review of 11,087 primary bone tumors that were subjected to either – biopsy or complete surgical resection, osteoid osteoma accounted for 13.5% of all benign tumors.[3] Osteoid osteoma is reported to occur in the cortex of the shafts of long bones in 80-90% of cases. Physical examination may disclose focal tenderness; however, signs of inflammatory disease, including erythema and warmth, are almost always absent. The results of laboratory analyses are typically normal. The standard treatment traditionally has been surgical resection. But when it is associated with a pathological fracture, in addition to complete removal of the nidus, the fracture needs to be fixed securely. We report a case of pathological fracture found to have an osteoid osteoma in the diaphyseal region of humerus.

Case Presentation

A 20 years male was admitted in our institute with complaints of severe pain in right arm with inability to move the shoulder and elbow since one day. There was a history of trivial trauma prior to this complaint. Radiographs revealed a fracture at the middle one third of right humerus (Fig.1). There was a suspicious widening of the bone around the fracture area. Patient was taken up for operative fixation of the fracture. Intra-operatively, it was found that the bone was very brittle and cortex was paper thin (Fig.2). Excision biopsy of the pathological bone was performed. The native bone was reinforced by an intramedullary fibular strut bone graft and cancellous bone grafts from the iliac crest (Fig.3). Open reduction and internal fixation with locking compression plate was achieved (Fig.4). Post-operatively, a U-slab was given (Fig.5). Biopsy reported histological features of osteiod osteoma with areas of bloody infiltrates - presumably from fracture haematoma (Fig.6).

Figure Legends

Figure 1: Radiographs at presentation.

Figure 2: Intra-operative picture showing fracture site with thinned-out and brittle bone.

Figure 3: Harvested fibular strut bone graft and cancellous bone graft from the iliac crest.

Figure 4: Intra-operative picture showing internal fixation with locking compression plate.

Figure 5: Post-operative radiograph showing the bone grafts and fixation.

Figure 6: Microscopic findings of the histo-pathological specimen from the fracture site showing mass of irregular osteoid tissue surrounded by dense reactive bone with areas of bloody infiltrates.

Figures.

Figure 1

Figure 2

Figure 3

Figure 4

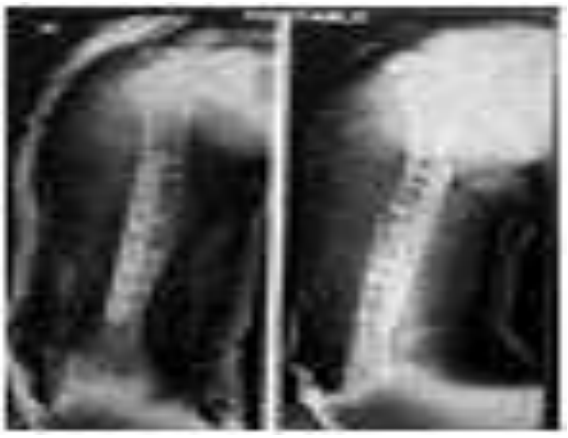

Figure 5

Figure 6

DISCUSSION

Osteoid osteoma was described by Jaffe[2], who classified it as a benign tumour in 1935. Before that, the lesion had always been considered as infectious and inflammatory one and

had been classiffied with the chronic bone abscesses or Brodie's abscess. Jaffe pointed out that there was no histologic evidence of infection. The tumour consists of an area of calcified osteoid in a stroma of loose, vascular connective tissue without polymorphonuclear or round-cell infiltration. Surrounding this osteoid nidus is a zone of sclerotic but otherwise normal bone. Cultures of osteoid osteomas have not produced consistent growth of organisms. Jaffe's concept of osteoid osteoma is now almost universally accepted.[4]

Symptoms of osteoid osteoma can be very variable. In addition, stress fracture, intracortical abscess, intracortical hemangioma, chondroblastoma, osteoblastoma, and compensatory hypertrophy of the pedicle may mimic osteoid osteoma. [5] Pain is the principal symptom which is continuous, deep, aching, and intense with varying quality and severity typically localized to the site of the lesion. It is usually worse at night. It is located preferentially in long bones with a predilection for lower limbs, including the tibia and femur.[6] Osteoid osteomas are characterised by the presence of a bony nidus which is usually well-demarcated and may contain a variable amount of calcification. The term nidus, which was described as the "core" or "nidus-like focus" by Jaffe in 1953, refers to the tumor itself and is composed of bone at various stages of maturity within a highly vascular connective tissue stroma. The center of the nidus usually is the most highly mineralized part, and it may display various amounts of mineralization. Surrounding the nidus is a zone of sclerotic but otherwise normal bone.[7, 8, 9]

Magnetic resonance imaging (MRI) is considered by some authors the most sensitive examination for the diagnosis of osteoid osteoma. The nidus usually has a low to intermediate signal intensity on T1-weighted and a low to high signal intensity on T2-weighted magnetic resonance images. However, this investigation lacks specificity, and the nidus cannot be viewed in nearly 50% of cases.[10] The diagnosis of osteoid osteoma can be confirmed only with pathologic examination. Complete surgical excision is the most predictable way to cure osteoid osteoma and should be the goal of surgical intervention. En bloc resection allows the entire nidus resection and therefore healing.[11]

A pathological fracture through the oseoid osteoma site has rarely been reported in literature although formation of osteoid osteoma following a stress fracture has been reported.[12] Pathological fractures are common in secondary bone tumours and some benign bone tumours where the bone is weakened by the tumour by expansion and invasive destruction. Direct trauma to long bones can result in fractures and that was the case in our patient.

CONCLUSION

Osteoid osteoma is not a very uncommon condition. But there may be unusual presentations. Osteoid osteoma may rarely present with a pathological fracture. This creates problems in making a correct diagnosis. Radiologically, a classical thickening of the bone cortex with nidus may not be appreciable in all the cases, as was seen in our patient. Hence, histopathological examination of bone fragments from all suspicious fractures must always be done.

Clinical Message

Diagnosing the cause of a pathological fracture can be challenging. Certain tumours may present in unusual fashion. Although rare, osteoid osteoma may present as a pathological fracture, without any classical radiological findings. A detailed evaluation is essential.

Competing Interests

The authors declare that they have no competing interests.

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