

Desmoid Tumor of Ilio-Acetabular Region with Articular Cartilage Breach: A Case Report

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What to Learn from this Article?

Very rare presentation of Desmoid tumor and successful treatment of the same

Abstract

Introduction: Desmoid tumor of bone is a rare benign tumor. It is reported commonly in mandibular and meta-diaphyseal region of long bones. We report involvement of unusual site in ilio-acetabular region with breach in articular cartilage.

Case Report: A 40 year old female presented with pain in the left hip. Radiologically, a lytic lesion at ilio-acetabular region was seen. Intra-operatively breach in acetabular roof was seen which was missed in the scan. Curettage and defect reconstruction was done. Histopathology reported as desmoid tumor. 20 months post-operatively patient was symptom free.

Conclusion: Desmoid tumor is a rare bone tumor. This case report emphasizes about the rarity of the lesion in this location and the rare chances of breach in articular cartilage of the joint. The chances of recurrences are high with intralesional curettage.

Keywords: lytic lesion, Desmoid tumor, ilio-acetabular region, breach in articular margin

Author's Photo Gallery



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Introduction

Bone desmoid tumor was an unknown entity until when Jaffe HL reported a case in 1958. It is a rare tumor comprising about 0.1-0.3% of benign tumors of bone [1]. It is a locally aggressive benign tumor reported commonly in mandible and meta-diaphyseal region of long bones. We report a case of desmoid tumor of ilio-acetabular region in a 40 year old female.

Case Report

A 40 year old female came with complaint of pain in the left hip region which aggravated on squatting and climbing stairs. Radiological evaluation showed a lytic lesion in the left ilio-acetabular region, surrounded by a sclerotic margin superiorly. There were no trabeculations and no frank breach in the cortex (Fig. 1). MRI scan was done which showed a well defined homogenous lytic lesion with no break in the cortex and no soft tissue involvement (Fig. 2). Hematological reports were not significant. A provisional diagnosis of giant cell tumor was made and planned for excision.

Through an ilio-inguinal incision the lesion was approached through inner table of ilium. A firm, ivory white mass with a rubbery consistency was excised. A defect on the roof of the acetabulum was found which was missed by the scan (Fig. 3). Thorough curettage was done and the roof defect was

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Figure 1: Plain radiograph of pelvis with both the hips which shows a well defined lytic lesion over the left ilio-acetabular region. There appears no obvious breach in the cortex or the articular margin

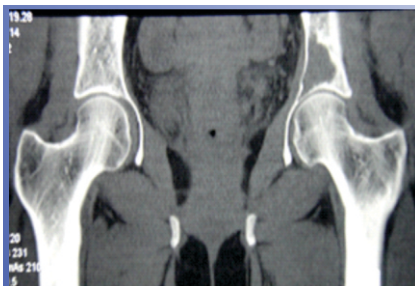


Figure 2: A CT scan of the pelvis was taken to rule out any breach in the cortex. There appears no obvious discontinuity

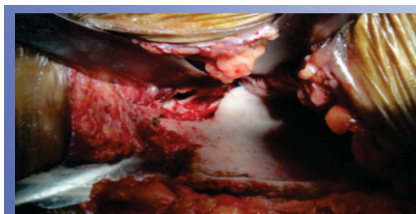


Figure 3: An intra-operative photograph seeing the pelvis from the cephalad aspect. The iliac crest margin is seen in the lower aspect of the photograph. There appears a hollow curetted lesion within which a breach in the roof of the acetabulum is seen

reconstructed with a cortical graft harvested from the inner table of the ilium. The curetted lesion was filled with tricalcium phosphate.

Histopathological finding showed multiple spindle shaped fibroblast cells with small and elongated nuclei in the background of dense collagen fibres. There were no nuclear atypia or mitotic activity. The findings were suggestive of desmoid tumor (Fig 4.)

Patient was kept non weight bearing for 3 months with gradual return to full weight bearing walking. At follow up of 20 months patient is symptom free and Xray showing no evidence of lytic lesion (Fig 5). However, on CT scan there appears a persistent lytic area which we suspect to be a recurrence (Fig 6). Hence patient is kept on regular follow-up.

Discussion

Jaffe was first to describe about desmoplastic fibroma of bone in 1958. It is a rare benign tumor of bone slowly growing and non metastatic [2]. Few cases have been reported since then and common occurrence being in mandible and meta-diaphyseal region of the long bone [3]. Recognition of desmoplastic fibroma is important because on radiology and histology, the lesion may be mistaken for an indolent, benign fibrous lesion or more aggressive spindle-cell sarcomas.

It commonly occurs in the age group between the adolescent to 40 years with no specific sex predominance. Patients commonly come with complaint of pain; however there are few reported cases of pathological fracture especially those involving tibia and femur. Such fracture in pelvis has not been reported [4].

Radiologically, the lesion is lytic type with rare trabeculations. However; they may be seen in recurrent cases. The lesions are well defined with no sclerosis or periosteal reaction except if there is associated pathological fracture. If a soft tissue lesion is invading onto the bone, they are eccentric in position with sclerosis due to endosteal bone formation. MRI helps in delineating the lesion and knowing soft tissue involvement, if any. Significant T2 shortening of a non-sclerotic fibro-osseous lesion should place desmoplastic fibroma high among the diagnostic considerations [5]. Radiologically they have to be differentiated from giant cell tumor, fibrous dysplasia (long bone lesions), fibrosarcoma, brown tumors, and chondromyxoid fibroma and hence a tissue diagnoses is must [6].

Pathologically, the findings were similar to the other reported cases. The mass is soft rubbery whitish in color. However unlike other reports we did appreciate a breach in the articular cartilage. This is important as the chances of joint involvement become high.

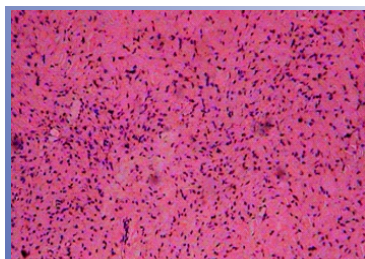


Figure 4: Histopathology specimen picture showing spindle shaped fibroblast cells with small and elongated nuclei over a background of collagen fibers. There appears no nuclear atypia or mitotic activity



Figure 5: A 20 month post operative X ray picture showing well maintained articular margin and the joint space. The lytic lesion seems filled up

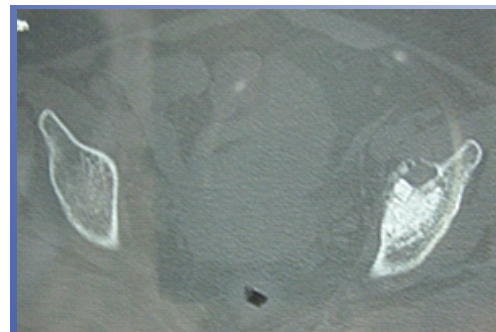


Figure 6: CT scan of the hip taken at 20 month post op, however shows lytic lesion anteriorly which seems to be a recurrence

Histo-pathological findings were quite representative of desmoid tumor. Spindle shaped cells with small and elongated nuclei over a background of collagen fibers. There were no nuclear atypia or mitotic activity [6]. The differentiation from a low grade fibrosarcoma is difficult though [1].

As these are rare tumors, there is no standard protocol for treatment of these lesions. All kinds of procedures like intralesional, marginal or wide resection are reported as a treatment. The chances of recurrences have been reported to be high in intralesionally resected cases. But if wide resection would result a major functional deficit, an attempt of intralesional curettage with bone grafting seems warranted [7].

Conclusion

Desmoid fibroma of ilio-acetabular region is very rare hence a great sense of suspicion is required for the appropriate management. This case report emphasizes about the rarity of the lesion in this location and the chances of breach in articular cartilage of the joint. However, long term follow up is required to know about the recurrences.

Clinical Message

Desmoid tumor, though rare, should be kept as one of the important differentials for the lytic lesion around pelvis. One should also define whether there is any associated cortical or articular margin breach (as seen in our case report) and then decide further management regarding reconstruction.

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