

Case Report

Management of distal femur aneurysmal bone cyst in an adult by extended curettage with bone substitute and internal fixation: an illustrative case report

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ABSTRACT

Aneurysmal bone cysts (ABCs) are benign osteolytic vascular neoplasm characterised by several sponge like blood filled or serum filled non endothelialized spaces of various diameter. ABCs can present a diagnostic challenge, as they share several histological and radiographic characteristics with other aggressive lesions like giant cell tumors and malignant telangiectatic osteosarcomas. In this article, we present a case of 34-year gentleman with left distal femur aneurysmal bone cyst treated by extended curettage augmented with bone substitute and internal fixation. In ABC management, curettage is the treatment of choice inspite of high recurrence rate. This is because of early return to functional activities and less morbidity. En bloc excision results in a lower recurrence rate but more extensive reconstructive surgery is needed which is associated with higher morbidity

Keywords: Aneurysmal bone cyst, Bone curettage, Bone substitute

INTRODUCTION

Aneurysmal bone cysts (ABCs) are benign osteolytic vascular bone tumour characterised by aggressive local expansion and bone destruction.¹ Many hypotheses have been proposed to explain the cause and pathogenesis of aneurysmal bone cyst, and until very recently the most commonly accepted idea was that aneurysmal bone cyst was the consequence of an increased venous pressure and resultant dilation and rupture of the local vascular network. However, studies by Panoutsakopoulos et al and Oliveira et al uncovered the clonal neoplastic nature of aneurysmal bone cyst. Primary cause has been regarded arteriovenous fistula within bone.²

The lesion may arise de novo or may arise secondarily within a pre-existing bone tumor, because the abnormal

bone due to change in hemodynamics. An aneurysmal bone cyst can arise from pre-existing giant cell tumour, fibrous dysplasia, chondromyxoid fibroma or chondroblastoma. A giant cell tumor is the most common cause, occurring in 19% to 39% of cases.

Patients typically present during the second decade of life and ABCs represent approximately 9% of benign bone tumors.³ It is essential to differentiate it from unicameral bone cyst and telangiectatic osteosarcoma.⁴

Tissue biopsy with histologic interpretation is mandatory for appropriate diagnosis. If the lesion is small and does not involve articular surface, curettage with bone grafting is the recommended initial approach to management.⁵ Adjuvant therapies are multiple which includes electrocautery, cryotherapy, intra-lesional sclerotherapy,

preoperative embolization. There is also medical management in the form denosumab to prevent fracture.⁶

In this article, we discuss about a 34 year gentleman with left distal femur aneurysmal bone cyst managed by extended curettage with bone substitute and internal fixation. This paper will also review the different radiographic types of ABCs, discuss the radiographic and histologic differentiation from telangiectatic osteosarcoma, and discuss the recommended management.

CASE REPORT

A 34 year gentleman came to outpatient department with complaints of left knee pain for past one month. No history of any previous trauma. No history of any fever or any constitutional symptoms. On physical examination, he has localised warmth and bony tenderness over distal femur over metaphyseal and epiphyseal region and also pain while walking.

X ray showed osteolytic lesion over metaphysis extending into epiphysis of distal femur. Lesion is central, expansile with multiple septations and does not

involve articular surface. No evidence of any pathological fracture or any periosteal reaction. MRI showed a 5×3.7×3.5 cm osteolytic lesion with sclerotic margins at the metaphysis of distal femur extending into the epiphysis with free fluid levels and blood noted within the lesion. There were no underlying soft tissue elements noted, suggesting this was a primary aneurysmal bone cyst.



Figure 1: Pre-operative X-ray.

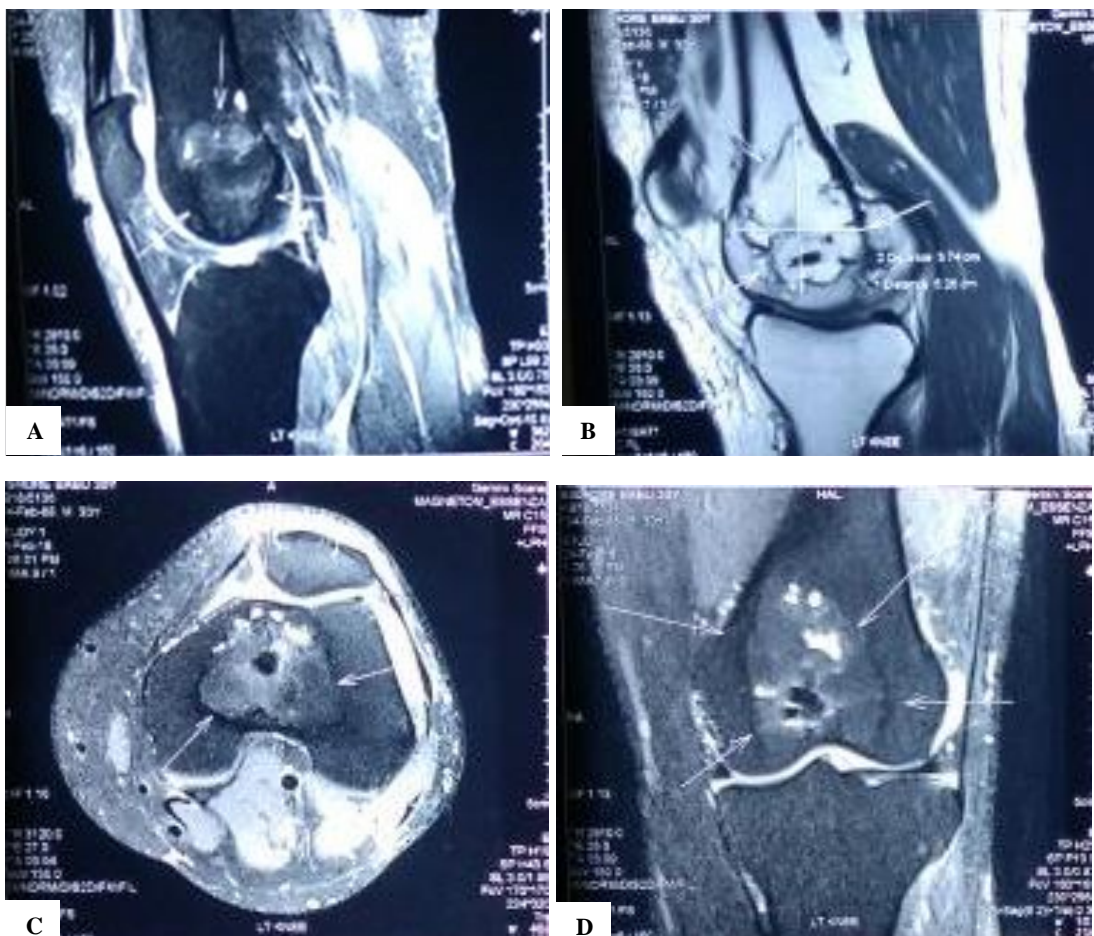


Figure 2: Magnetic resonance imaging; (A) T2 weighted saggital section, (B) T1 weighted saggital section, (C) axial section, (D) coronal section.

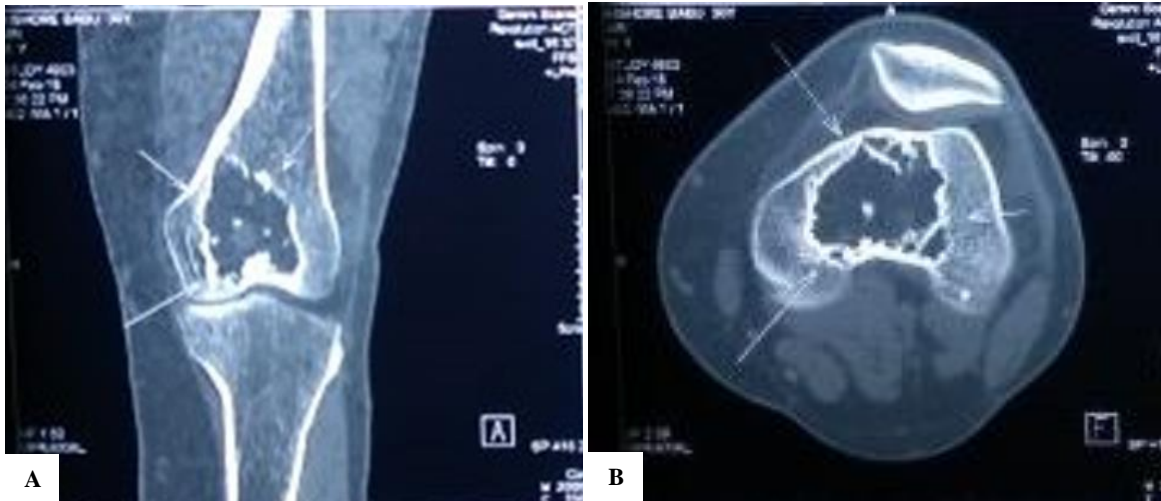


Figure 3 (A and B): CT films.



Figure 4: Post-operative X-ray.

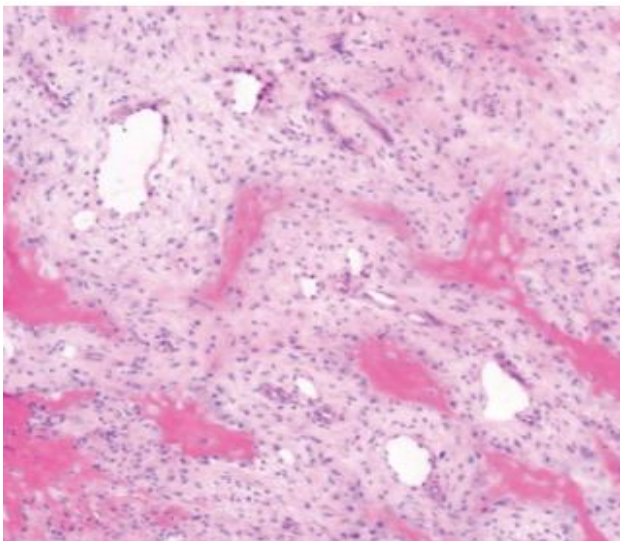


Figure 5: Histology shows fibroblasts mixed with osteoid and multinucleated giant cells, no significant cellular atypia was seen.



Figure 6: Six week follow up X-ray.



Figure 7: One year follow up X-ray.

Left distal femur aneurysmal bone cyst extended curettage with bone substitute packing with locking compression plating was done. First the femur was exposed through lateral incision, periosteal window in the bone was created. Tissue samples were analysed by frozen section and, once determined to be a benign lesion, the lesion was thoroughly curetted. Chemical adjuvant such as phenol was used. After extended curettage, void in the bone was filled with artificial bone substitute and then distal femur locking compression plating was done.

The pathological report demonstrated a cystic lesion with fibroblasts admixed with osteoid and multinucleated giant cells without cellular atypia suggestive of aneurysmal bone cyst

At the six-week follow-up, X-rays demonstrated consolidation of the bone substitute with callus formation at the fracture site. The patient was able to return to full preoperative weight-bearing activities. Patient now had a follow up of one year with no tumour recurrence and doing all day to day activities.

DISCUSSION

Aneurysmal bone cyst has been widely regarded a reactive process of uncertain cause since its initial description by Jaffe and Lichtenstein in 1942. It is second most common tumor of spine and commonest benign tumor of pelvis in pediatric population. Incidence is

slightly more in males than females (1.3:1). The distal femoral metaphysis is an important weight-bearing location where the patient's first presentation is commonly following trauma and fracture which can complicate treatment.

Histologically they are classified in two variants. The classic (or standard) form (95%) has blood filled clefts among bony trabeculae. Osteoid tissue is found in stromal matrix. The second one is solid form (5%) show fibroblastic proliferation, osteoid production and degenerated calcifying fibromyxoid elements.

Table 1: Buraczewski and Dabska’s phases of development of aneurysmal bone cyst.

Stage	Description
Initial phase	Osteolysis without peculiar findings
Growth phase	Rapid increase in size of osseous erosion
	Enlargement of involved bone
	Formation of shell around central part of lesion
Stabilisation phase	Fully developed radiological pattern

In 1986, a radiographic classification system of ABCs was presented by Campanacci et al.⁸ These criteria distinguish amongst five unique ABC types based on their radiographic appearance.

Table 2: Campanacci classification of aneurysmal bone cyst.

Type	Osteologic site	Skeletal site	Radiographic appearance
1	Central	Long bone metaphysis and metadiaphysis	Cystic lucency with cortical attenuation
2	Central expansive	Long bone metaphysis and metadiaphysis	Cystic lucency with severe cortical distension
3	eccentric	Long bone metaphysis	Variable lucency
4	subperiosteal	Long bone diaphysis	Periosteal elevation
5	Subperiosteal expansive	Cancellous bone	Periosteal elevation with cortical destruction

As per the Campanacci criteria, our case comes in type 1 ABC. The extended curettage with bone substitutes provides appropriate management for this type of tumours.

Making the appropriate diagnosis is important for the management of an ABC. The most important alternative diagnosis that one must consider is telangiectatic osteosarcoma (TOC). TOC presents as multiple cystic compartments on imaging, with fluid levels making it easy to confuse with an ABC.⁹ The age of presentation between these two conditions is also similar, further complicating differentiation.

Radiographically, TOCs tend to grow more rapidly and invade nearby structures more than aneurysmal bone cysts.^{10,11} However, biopsy is performed for appropriate diagnosis. The primary means of differentiating these two tumors histologically is the presence of cellular atypia which is present in telangiectatic osteosarcomas and absent in aneurysmal bone cysts.

There are different methods of management of ABCs, ranging from medical therapy alone to curettage with grafting with or without internal fixation and even wide resection with or without fusion. Each method has its own advantages and disadvantages. According to Schreuder et al study, recurrence rate in wide resection was 0 percent while in curettage and bone cement was 31

percent.¹² However the morbidity and functional disability in wide resection and reconstruction is very high compared to curettage part. Hence appropriate quick diagnosis makes the tumour amenable for curettage which aids in easy return for functional activities.

CONCLUSION

The diagnosis and management of aneurysmal bone cysts is a challenge to the surgeons. Lesions of the distal femur can present as pathologic fractures and must be differentiated from the other aggressive dangerous tumours. Many treatment options are available, but curettage with bone grafting with or without internal fixation provides an acceptable management between tumor-free survival and treatment morbidity and mortality.

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