

Ipsilateral Fibula Grafting in a Case of Aneurysmal Bone Cyst in Tibia Diaphysis: A Case Report

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Learning Point of the Article:

Tibia diaphyseal Aneurysmal Bone Cysts (ABCs) have a rare presentation for which wide margin resection of the tumor along with ipsilateral fibula strut grafting is a viable surgical method.

Abstract

Introduction: Aneurysmal bone cyst (ABC) is a benign, expansile hemorrhagic tumor usually presenting in the metaphyseal region of long bones. Diaphyseal ABCs are rare and prone to pathological fracture and increased morbidity. Treatment options are centered around the site and size of the lesion.

Case Report: We report a case of a large ABC in the tibia diaphysis in an 18 year female. The patient was treated with wide margin resection followed by an ipsilateral fibular bone graft of 5 cm, which was followed by good post-operative recovery without any recurrence. At 12-month follow-up, the patient had painless mobility with a full functional range of motion of the affected limb.

Conclusion: An early rehabilitation and return to active life can be achieved in diaphyseal ABCs with careful planning and post-operative care.

Keywords: Aneurysmal bone cyst, ipsilateral fibula graft, pathological fracture, bone tumor.

Introduction

Aneurysmal bone cyst (ABC) usually presents as an expansile hemorrhagic tumor in the metaphysis of long bones [1]. Their presentation on the surface diaphysis of long bones is quite rare and more prone to pathological fractures [2]; hence, they pose a unique set of challenges in treatment and rehabilitation. Typically, patients present with complaints of pain at the involved site or pathological fractures. ABCs must be differentiated from other forms of bone cyst. Investigations include radiographs and magnetic resonance imaging (MRI) of the affected site. Radiographs show well-defined expansile osteolytic lesions with sclerosed margins. Multiple fluid levels on an MRI are characteristic of ABC although not specific [3]. Histopathology remains the gold standard in the diagnosis of ABC. There are a wide variety of treatment options available for

treating such lesions. The standard of care for long bones ABCs is curettage and void filling bone grafts with or without local adjuvants [4]. Surgical planning is guided by the location and size of the lesion.

Case Report

An 18-year-old female presented to the outpatient department with a 10 days history of acute onset right leg pain associated with swelling. The pain had gradually progressed in the last 10 days and made it difficult for her to walk. On examination, there was a swelling of approximately 2 cm × 2 cm on the anterior aspect of the right mid tibial shaft. The swelling was tender on palpation with a local rise of temperature. It was not associated with any constitutional symptoms. A plain radiograph of the right leg was

Author's Photo Gallery



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Figure 1: Plain radiograph of tibia in antero-posterior and lateral views.

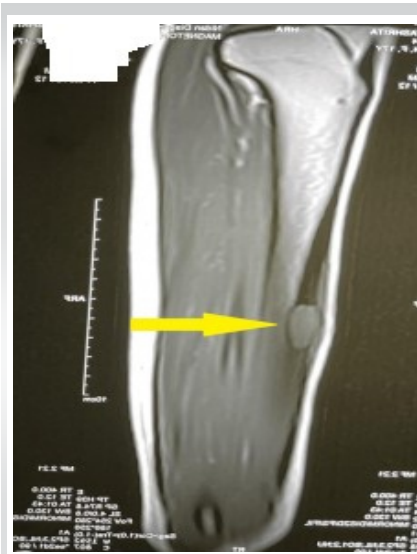


Figure 2: Pre-operative magnetic resonance imaging (sagittal section). Yellow arrow pointing to a well-circumscribed cystic lesion.

obtained in both antero-posterior and lateral views with the arrow showing cystic lesion in the midshaft of the tibia (Fig. 1).

MRI showed a well-circumscribed cystic lesion (Fig. 2).

The lesion was biopsied and sent for histopathological examination which revealed large areas of hemorrhage separated by connective tissue septa, suggestive of an ABC.

Due to a large size (4 cm) and location (cortical midshaft tibia) which would lead to a large post-excision defect an ideal bone graft had to be chosen. A fibular graft from the contralateral side was considered for the case. However, the parents of the patient were reluctant for any operative procedure on the contralateral side. Hence it was decided to proceed with an ipsilateral fibular graft. After a written informed consent patient was taken up for surgery under spinal anaesthesia. After thorough dissection, the involved part of the bone was resected with a 1 cm margin

followed by curettage. The resected piece was measured intraoperative (5 cm). A similar length bone graft was obtained from the midshaft of the fibula and placed at the tibial defect (Fig. 3).

Post-surgery patient was put on an above-knee slab and advised against weight-bearing. The post-operative wound healed well. Partial weight-bearing was started at 6 weeks. The patient was kept on monthly follow-up. Gradually, the patient was symptomatically better. Radiological signs of graft uptake were visible on X-rays. At 7-month post-surgery patient was able to bear weight without a walker.

On radiographs, some areas of osteopenia were noticed in the distal tibia and talar region which gradually improved back to normal in subsequent follow-up (Fig. 4).

At the end of 1-year follow-up, patient had complete graft uptake on radiographs and had pain-free ambulation with a full active range of motion of the affected leg without any limitation of activities of daily living (Fig. 5).

Discussion

Since its first description as a separate pathological entity by Jaffe, ABCs have been studied extensively [5, 6]. The median age of presentation of ABCs is 13 years. The patient, in this case, was aged 18 years. ABCs have a wide variety of presentations. Although the usual presentation is in the metaphyseal part however there have been several instances of it being reported in the diaphyseal area or with physeal extension [7]. The etiopathogenesis of ABC however remains controversial with no consensus.

Earlier it was believed to be reactive due to underlying arteriovenous malformations. The bone void arising out of the increased venous pressure was filled with blood, resulting in a typical ABC appearance. Though recent study by Ye et al. has shown that ABCs may be independent neoplasms with activation of the USP6 gene [8], ABC does not however have the potential for malignancy.

When encountering a lesion suggestive of ABC in the tibia, it is important to consider several differential diagnoses which mainly include unicameral bone cyst, telangiectatic osteosarcoma and giant cell tumour and chondroblastoma. Out of all the differentials since telangiectatic osteosarcoma is a malignant bone tumour, it is evident that it possesses the biggest diagnostic obstacle [9-11].



Figure 3: Intra-op images (a) Bone defect post excision and curettage (b) Fibular graft from contralateral side (c) Defect filled with fibular graft.



Figure 4: Follow up radiograph (Arrow pointing to areas of osteopenia).



Figure 5: One year follow up images (a) Radiographs at 1 year follow up (b) Clinical photograph of the patient with healed scar mark.

Histologically, ABCs are characterized by innumerable osteoclastic giant cells scattered throughout the lesion. These giant cells are multinucleated and are located centrally in the blood-filled intertrabecular spaces. The stroma of the tumor consists of fibrous tissue as well as spindle-shaped fibroblasts which are found in it [12].

The size and site of the presentation of ABC are the most crucial factor for determining the treatment course [13]. Historically, studies have shown that en bloc excision of the cyst has excellent outcomes in terms of recurrence. Studies have reported up to 100% localized control after en bloc excision [9, 14, 15]. Due to the large size and location of the lesion, it was decided to proceed with en bloc excision, followed by bone grafting. Curettage of ABCs have returned highly variable recurrence rates with some studies reporting a recurrence rate up to 59% [16]. In major weight-bearing bones such as the femur and tibia, the choice of treatment will have a grave impact on the patient's functioning. Any recurrence can lead to the requirement for surgery and severe limitation in functional mobility. Diaphyseal fracture on progression

involves the adjacent cortex and invariably may lead to pathological fracture. Excellent radiological and functional outcomes can be achieved in diaphyseal ABCs with careful planning and post-operative care.

Conclusion

An early rehabilitation and return to active life is possible with wide margin excision of the tumor combined with ipsilateral fibula strut grafting for surgical management of tibia diaphyseal ABCs. This surgical method has not been mentioned in the literature with a follow-up of more than a year in the management of diaphyseal ABCs.

Clinical Message

Excellent functional results with no recurrence can be achieved by this method for management of diaphyseal ABCs.

Declaration of patient consent: The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given the consent for his/ her images and other clinical information to be reported in the journal. The patient understands that his/ her names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Conflict of interest: Nil **Source of support:** None

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