

A Rare Case of Aneurysmal Bone Cyst of Talus

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Abstract: An aneurysmal bone cyst (ABC) is a benign solitary lesion of unknown etiology, mainly occurs in the long bones and spine but, are rarely reported in the bones of the foot and ankle. Very few cases of ABC involving the talus reported up today. We report an unusual case of ABC of talus in a 23-year-old female. Clinical presentation, histological features, radiological features, treatment options, and prognosis have discussed.

Keywords: Aneurysmal bone cyst, talus, curettage, bone grafting

1. Introduction

The aneurysmal bone cyst (ABC) is an expansile cystic lesion that commonly occurs in the long bones and spine but rarely reported in other bones of the body. The most common age of occurrence is during the second decade of life.^{1,2}

Jaffe and Lichtenstein were the first to described ABC.³ As defined by the World Health Organization. The ABC is a benign tumor-like lesion described as “an expansile osteolytic lesion consisting of blood-filled spaces of variable size separated by connective tissue septa containing trabeculae or osteoid tissue and giant osteoclast cells.”⁴

ABCs erode and cause ‘expansion’ of underlying cancellous and cortical bone⁵. Around the lesion, there is always a shell formed by periosteal new bone; although this may be only millimeters thick, it prevents direct extension into the soft tissues⁶. The expansile nature of the lesions can cause pain, swelling, deformity, disruption of growth plates, neurologic symptoms (depending on its location), and pathologic fracture.⁷

ABC’s in the foot and ankle are uncommon. ABC’s present about 1% of all primary bone tumors collectively. Its frequency of occurrence in foot and ankle is only about 3% compared to other bones of the body.² Occurrence within the talus is rare, and generally present as chronic ankle pain and swelling.⁸

We report a rare case of ABC involving talus in a 23-year-old female confirmed by histopathology report. We performed curettage and bone grafting of the cyst and found no signs of recurrence at one year follow up.

2. Case Report

A 23-year-old female presented with a chief complaint of difficulty in walking because of pain and swelling in the left ankle during the last seven months. An increase in swelling was associated with pain in the heel from the last two months, which preceded an episode of trauma while driving a two-wheeler.

Clinical evaluation revealed mild swelling over the anteromedial aspect of the ankle. Joint line tenderness was present on palpation of both ankle and subtalar joints all around, but there was no local rise of temperature. There was a terminal restriction of both subtalar and ankle joint movements due to pain (**Fig.1**). There were no distal neurovascular deficits or any significant lymphadenopathy.



Fig.1.Range of movements A.Dorsiflexion
B.plantarflexion restricted in the left ankle joint.

Radiographic examination of ankle joint Antero Posterior and Lateral views



Figure 2 X-RAY-Antero posterior and lateral view
Radiographic examination of her ankle revealed an expansile, multiloculated lytic lesion of the talus involving head, neck, and body with thin trabeculae traversing the cystic cavity. There was no breach in the cortex or collapse of the bone. Articular margins and joint spaces are standard (**Fig.2**). Based on clinical and radiographic findings, we thought of differential diagnosis of 1.bone cyst 2.giant cell tumor of the talus.



Fig.3 CT scan of the ankle revealed evidence of a well defined sharply marginated lytic lesion of the talus with medullary expansion and cortical thinning. There was no cortical breach or matrix calcification. A decreased talonavicular joint with normal tibiotalar and the talocalcaneal joint observed.

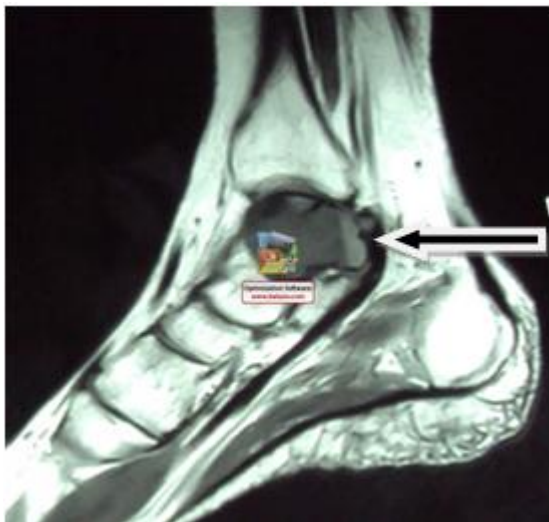


Fig.4 MRI of the ankle added finer details such as fluid level in the lesion and healthy articular cartilage

The CT and MRI findings of lesion described at talus excluded the possibility of GCT and left us with a differential diagnosis of ABC or a simple bone cyst of the talus.

Keeping in mind the benign nature of the lesion, an open biopsy of the cyst along curettage and bone grafting has planned. Biopsy of the cyst through an anterior-medial approach has done, which revealed cavities filled with the altered brown color of blood. Curettage and bone grafting has done with cancellous bone graft harvested from the ipsilateral ilium to fill the cavity after a thorough saline wash. The histopathological report revealed large blood-filled cavities lined by fibrous septa, with occasional osteoclastic giant cells (**Fig. 5A and 5B**). Hence the diagnosis of ABC involving the left talus was made.



Fig 5A and 5B histological slides reveal large blood-filled cavities lined by fibrous septa(A), with occasional osteoclastic giant cells, hemosiderin-laden macrophages with a thin rim of bone(B)

Postoperatively the patient was advised non-weight bearing for eight weeks and later mobilized with partial weight-bearing walking for a further four weeks and then followed by full weight bearing on the affected limb.

At six months follow up, the patient was pain-free and returned to her regular activities with the restoration of full painless range of motion of both ankle and subtalar joints. At one year follow-up, the patient is clinically asymptomatic with radiologically no evidence of recurrence and proper incorporation of graft material (**Figs. 6A, 6B**)



Figure 6:Follow-up radiographs showing incorporation of graft material at six months(A), one year (B)

3. Discussion

ABC is an entity on its own, having unique clinical, radiological, and diagnostic behavior.⁵ The exact etiology of ABCs is unknown. Most investigators believe that ABCs are the result of a vascular malformation within the bone; however, the ultimate cause of the malformation is a matter of controversy.⁹

The concept of an ABC as a secondary phenomenon occurring in a pre-existing lesion and based on the fact that in approximately one-third of the cases, a pre-existing lesion identified, the most common of which is a giant-cell tumor.^{10, 11} About 50-70% of ABCs occurs in the second decade of life, with 70-86% occurring in patients younger than 20 years.

On histology, the ABC characterized by blood-filled cavities lined by fibrous septa. The stroma contains proliferative fibroblasts, spindle cells, areas of osteoid formation, and an uneven distribution of multinucleated giant cells. The tissue within the septations includes cavernous channels that do not contain a muscular or elastic layer in their walls. Areas of new and reactive bone formation can also found in the ABC. Mitotic figures are

familiar to ABCs, but no atypical figures should be evident.¹²

Localized fibrocystic disease is a generic term for a group of nine lesions that have many features in stock, but each of them with a distinct entity. The lesions are a unicameral bone cyst, fibrous dysplasia, fibrous metaphyseal defect, non-osteogenic fibroma, aneurysmal bone cyst, giant cell tumor, osteoblastoma, chondroblastoma, and chondromyxoid fibroma. All nine lesions referred to as giant cells variants.⁸

Clinically, talar cysts are often symptomatic and present with ankle pain, although some of these lesions may remain asymptomatic and detected as incidental findings. Even though there are many typical radiographs, computed tomography (CT), and magnetic resonance imaging (MRI) findings to confirm a diagnosis of ABC, an open biopsy to be performed because of the high frequency of accompanying tumors.¹³ When a biopsy is to perform, the sample should ideally include material from the entire lesion; a limited biopsy could easily cause a coexisting lesion to get missed, leaving the patient with a morbid prognosis.¹²

There are various methods of treatment based on the site and size of the lesion, which includes curettage, which may supplement with various adjuvant therapies such as bone grafting, use of liquid nitrogen, phenol installation, and Poly Methyl Metha Acrylate (PMMA). Other modalities, such as wide excision or arterial embolization, to consider. Although relatively rare, there is no reason to assume that ABCs of the foot and ankle will respond to treatment or recur any differently from ABCs that occur elsewhere in the body. Surgical curettage along with bone grafting is sufficient to treat most ABCs of the foot and ankle.¹⁴

Despite a favorable outcome of ABCs with an overall cure rate of 90-95%,¹⁵ one of the most common problems encountered during management is a frequent recurrence. The incidence of recurrence has been noted to vary between 59% in cases treated with intralesional excision¹⁶ and 0% in cases with resection. Recurrence usually happens within the first year after surgery. Therefore, a patient of ABC needs to be observed for at least this period to exclude any recurrence. It is beneficial to detect recurrence early when the lesion is still small and more comfortable to treat.

To conclude, ABC of the talus is an extremely uncommon entity and remains an enigma, not only regarding causation but also regarding clinical presentation, imaging diagnosis, and optimal treatment. Proper diagnosis entails correlating the clinical presentation, anatomical location, radiological profile, and histopathological appearance. The biopsy is too imperative not only to exclude other more common histological mimics but also to choose the appropriate therapeutic regimen and to prognosticate the disease outcome. There is still no system to establish the diagnosis or to support different methods of treatment and thus reduce the problems encountered by the patient and the surgeon. Curettage and bone grafting are still a valuable

option for treatment, prevention of recurrences and in reducing the morbidity.

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