

Primary aneurysmal bone cyst of talus

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Aneurysmal bone cyst (ABC) of the talus is an extremely rare lesion; less than 20 cases have been reported in PubMed till 2012. We report a primary ABC of the talus in a 20-year-old male that was managed by extended intralesional curettage with phenol as an adjuvant and autologous cancellous iliac crest bone grafting. The patient had excellent functional outcome and there was no recurrence at 2 years of follow-up.

Key words: Aneurysmal bone cyst, curettage, talus

INTRODUCTION

Aneurysmal bone cyst (ABC) is a benign tumor like condition of bone, which consists of blood filled cystic cavities and is locally destructive. Talus is an extremely rare site for aneurysmal bone cysts. A search of the PubMed database reveals less than 20 such cases till 2012. We report an aneurysmal bone cyst of talus in a 20-year-old male that was managed by extended intralesional curettage and autologous iliac crest bone grafting. There was no recurrence at 2 years of follow-up.

CASE REPORT

A 20-year-old male, student by occupation, presented to the outpatient department of Government Medical College, Jammu, India in December 2009 with complaints of insidious onset, intermittent dull aching pain in the left ankle, since last 7 months. He denied history of swelling, fever, chills, sweats, weight loss or similar complaints in other joints of the body. There was no history of recent or remote trauma. The family, drug, occupational, sexual and recreational histories were non-contributory. General physical examination was unremarkable. On examination of the foot, there was no visible or palpable swelling, local temperature was normal and tenderness to deep palpation was noted on the dorsal aspect of ankle. The distal neurovascular status was normal.

Serum biochemistry studies were normal. Anteroposterior (AP) and lateral radiographs of the ankle [Figure 1] and a Computed Tomography (CT) Scan [Figure 2] revealed a radiolucent, expansile, lytic lesion with lobulated borders occupying the head and neck of the left talus. No periosteal reaction or calcifications were noted. Gadolinium enhanced Magnetic resonance Imaging (MRI) of the foot and ankle revealed an eccentric, cystic lesion in the left talus, which was hypo-intense on T1 weighted sequences and hyperintense on T2 weighted sequences and showed localized cortical erosion. The lesion showed subtle enhancement with gadolinium. There was no extension into the subtalar joint or adjacent soft tissues [Figure 3].

CT guided fine needle aspiration from the lesion revealed numerous osteoclast giant cells in an hemorrhagic background.

On the basis of the clinical findings, imaging studies and cytological examination a provisional diagnosis of giant cell lesion of bone was made. We considered giant cell tumor, aneurysmal bone cyst and simple bone cyst amongst the diagnostic differentials. However, we decided to plan the treatment of the lesion along the lines of giant cell tumor. The nature of the lesion, the diagnostic differentials, different treatment options and possible outcomes were discussed at length with the patient prior to obtaining an informed and written consent.

The patient was taken up for extended intra-lesional curettage with phenol as an adjuvant and autologous iliac crest bone grafting. The talus was exposed by the standard antero-lateral approach to the ankle. Per-operatively, cortical erosion on the lateral aspect of the talar head and neck could be identified. A window was made on the lateral surface of the talar neck to

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Received: 19-01-2012; **Revised:** 14-07-2012; **Accepted:** 23-08-2012

facilitate curettage. The interior of the talus revealed presence of a spongy, blood filled mass. Thorough curettage was performed and the cavity was enlarged in all directions using a high speed burr and phenol was used as an adjuvant. Finally, the ensuing cavity was packed with autologous cancellous iliac crest graft.

Post-operatively, the foot was immobilized in a non weight bearing below knee cast for 12 weeks.

Histopathological examination of the curettage material revealed presence of multiple blood filled cavities enclosed by fibrous septae without an endothelial lining [Figure 4]. Numerous osteoclast giant cells arranged irregularly in clusters, as well as scattered individually were also identified. A diagnosis of primary aneurysmal bone cyst was made.

Radiographs at 12 weeks showed good incorporation of the bone graft after which patient was permitted

to bear weight [Figure 5]. Initially, the patient had restriction of dorsiflexion, which improved completely with physiotherapy. There was no recurrence after two years of follow-up.

DISCUSSION

Aneurysmal bone cyst (ABC) is a benign tumor like condition of the bone. Many authors believe it to be a result of local circulatory disturbances and therefore do not consider it as a true neoplasm.^[1,2] Most cases occur between the ages of 10-20 years and show a slight female preponderance. The favored site of presentation is the vertebrae, flat bones and metaphysis of long bones.^[1] Talus is an unusual site for ABC, in fact the most common tumors of talus are intra-osseous ganglion cysts.^[3]

It has been said that ABC is associated with distinctive 17p13 translocations that result in up-regulation of USP6,



Figure 1: Lateral radiograph of the ankle showing a lytic lesion in the talar head and neck



Figure 2: CT Scan of the talar lesion

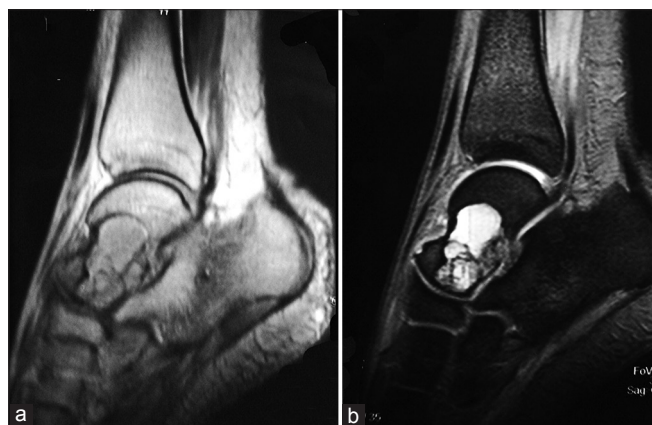


Figure 3: Magnetic resonance Imaging Scan (a) T1 weighted sagittal section showing an eccentric, cystic, hypointense lesion occupying the talar head and neck with localized cortical erosion. There is no soft tissue extension, (b) The lesion shows subtle enhancement with gadolinium

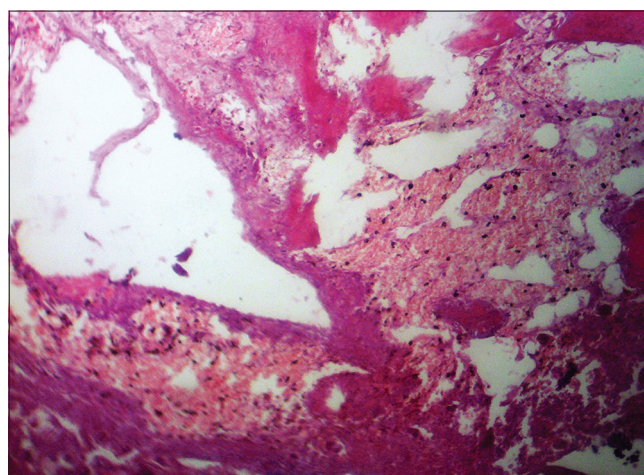


Figure 4: Sections from the curettage material showing multiple blood filled cavities enclosed by fibrous septae without an endothelial lining. Numerous osteoclast giant cells arranged irregularly in clusters, as well as scattered individually are also seen. (H and E, x400)



Figure 5: Lateral radiograph of the ankle at 2 years of follow-up. The graft has incorporated completely within the talus and there is no evidence of recurrence

a deubiquitinating enzyme.^[4]

Aneurysmal bone cysts that arise de-novo are termed as 'primary' whereas those occurring in conjunction with another tumor are termed as 'secondary'. Secondary ABCs may be seen with fibrous dysplasia, osteoblastoma, chondromyxoid fibroma, nonossifying fibroma, chondroblastoma, osteosarcoma, chondrosarcoma, unicameral bone cyst, hemangioendothelioma, and metastatic carcinoma. Therefore, a diagnosis of ABC merits a thorough search any associated pathology, which if present, would dictate the line of management. Other differentials include simple bone cyst (SBC) and giant cell tumor (GCT). The presence of blood filled cavities surrounded by proliferating fibroblasts and osteoclast giant cells differentiates ABC from a SBC. Giant cell tumor (GCT) tends to occur in the skeletally mature population and involves the epiphyses of long bones. GCTs have been reported to occur in the talus^[5] and can sometimes present with a secondary ABC.^[6] It may be very difficult to differentiate talar GCTs from ABCs on imaging studies. However, the presence of mononuclear stromal and regular distribution of giant cells favors the diagnosis of GCT. In addition, the giant cells in GCT tend to be larger and contain more nuclei.^[1]

Curettage and bone grafting is the standard treatment for ABCs in long bones.^[2] However, talar lesions can be challenging to treat. Many authors have described excellent results with intralesional curettage and bone grafting for lytic lesions that were well localized within the talus.^[7-9] Partial or total talectomy along with tibio calcaneal

arthrodesis has also been described for lesions that show extensive destruction of the talus and soft tissue or subtalar extension.^[5,8] Luna *et al.*, have described the use of external fixation in place of a traditional cast after curettage and bone grafting for ABC of talus.^[7]

In conclusion, the possibility of an aneurysmal bone cyst must not be forgotten when a patient presents with a lytic lesion in talus. Since, it may be difficult to differentiate talar ABCs from GCTs on the basis of imaging studies or fine needle aspiration alone, it is prudent to follow the principles of treatment of GCT viz. extended intralesional curettage and use of adjuvants in order to minimize recurrences. Even though the histological picture may be diagnostic of an ABC, a meticulous search should be made to rule out the secondary nature of such a lesion. With primary ABCs, an excellent prognosis can be expected after intralesional curettage and bone grafting.

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How to cite this article: Sharma S, Gupta P, Sharma S, Singh M, Singh D. Primary aneurysmal bone cyst of talus. *J Res Med Sci* 2012;17:1192-4.

Source of Support: Nil, **Conflict of Interest:** None declared.