PRIMARY ANEURYSMAL BONE CYST OF TALUS — A RARE CASE REPORT

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ABSTRACT

Aneurysmal bone cyst (ABC) of the talus is an extremely rare lesion. Less than 20 cases have been reported in PubMed till 2012 and none from Nepal. We report a rare case of primary ABC of talus in a 17 year old male that was managed by extended intra-lesional curettage and autologous cancellous iliac crest bone grafting. The patient had excellent functional outcome and there was no recurrence at 1 year of follow-up.

KEYWORDS

Aneurysmal bone cyst, bone graft, curettage, talus

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INTRODUCTION

Aneurysmal bone cysts (ABC) are tumor-like benign, expansile, osteolytic lesions of bone of unknown etiology. A current theory is that they are intraosseous arteriovenous malformations surrounded by a thin layer of periosteal bone.

It represents 1.0% of all primary bone tumors collectively.² It was first described by Jaffe and Lichtenstein in 1942.³ This tumor has a predilection for metaphyseal regions of long bones (femur – 22.0%, tibia – 17.0%, humerus – 10.0%) and spine (12.0%) and pelvis (9.0%).⁴ They occur rarely in the bones of the feet, making up only 5.0% to 9.0% of lesions.¹

Primary ABC of the talus is an extremely rare lesion and less than 20 cases have been reported in PubMed till 2012 and none from Nepal. We report a rare case of primary ABC of talus.

CASE REPORT

A 17-year-old male patient, student by occupation presented to Patan hospital OPD with complains of pain over right ankle for last six months. It was mainly localised on the medial side, insidious in onset, dull aching in character and intermittent in timing. The pain was also associated with minimal swelling on



Fig. 1: Pre-operative radiographs

the medial aspect of ankle which was again insidious in onset and gradually progressive. There was past history of trauma over the same ankle while playing football one year back which was managed conservatively with plaster cast for around one month. There was no history of fever, night cries/sweats, weight loss or similar complaints in other joints of the body. Rest of the history and general physical examination were unremarkable. On local examination of ankle and foot, the local temperature was normal, minimal tenderness was present just below medial malleolus region on deep palpation and about 2x2 cm size, fixed bony swelling was present just below medial malleolus. The range of motion of his ankle was restricted due to pain.

His blood investigations including serum biochemistry were within normal limits. Antero-posterior and lateral radiographs of his ankle showed a well-defined, lytic lesion involving the body of the talus with sharply demarcated margins. The lesion was expansile and causing thinning of the cortex. Fine thin septa were noted within the lesion. There was no evidence of any cortical break or soft-tissue extension (Fig. 1). Computer Tomography Scan (CT-Scan) of his ankle was also done and it showed a lytic, mildly expansile lesion with internal trabeculations measuring 5x3.7 cm in the talus. No cortical breach or erosion seen. Features were likely of ABC (Fig. 2).

On the basis of the clinical findings and imaging studies, a provisional diagnosis of ABC of talus was made. We considered Simple bone cyst and Giant cell tumour amongst the diagnostic differentials. 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 surgery and extended intra-

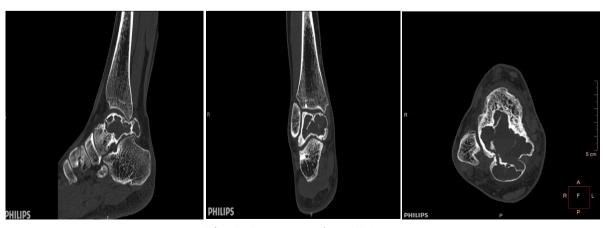


Fig. 2: Pre-operative CT-Scan

lesional curettage and autologous iliac crest bone grafting was performed. The talus was exposed by the postero-medial approach to the ankle. 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. Extended curettage was done using distilled water as an adjuvant. Finally, the ensuing cavity was packed with harvested autologous cancellous iliac crest graft. Post-operatively, the foot and ankle was immobilized in a non weight bearing below knee plaster cast.

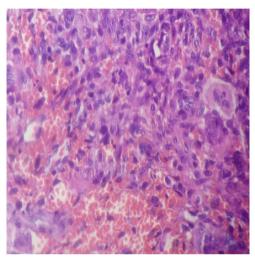


Fig. 3: Histopathology of curettage material showing cystic spaces filled with blood and separated by fibrous septa, lined by fibroblasts. (H&E, 40x)



Fig. 4: Post-operative radiographs

Histopathological examination of obtained curettage material showed cystic spaces filled with blood and separated by fibrous septa, lined by fibroblasts. Solid areas showed peripherally located multinucleated giant cells and fibroblasts. The findings were consistent with ABC. (Figure 3) Post-operative radiographs at one and two months follow up showed that the cyst appeared to be obliterating and bone graft appeared to be incorporating well with talus (Fig. 4). The plaster cast was removed at

three months and gradual weight bearing was allowed. The reason for delay was to prevent collapse of talus.

Post-operative radiographs at six months and one year follow up also showed progress and no recurrence (Fig. 5). At one year follow up, the patient does not feel any pain and the range of motion of his ankle is also within normal limits.



Fig. 5: Post-operative radiographs at one year follow up

DISCUSSION

Aneurysmal bone cyst 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. 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. Talus is an unusual site for ABC, in fact the most common tumors of talus are intraosseous ganglion cysts. ABC may be associated with distinctive 17p13 translocations that result in a deubiquitinating enzyme.

Aneurysmal bone cysts that arise de-novo are termed as 'primary' whereas those occurring in conjunction with another tumor are termed 'secondary'. Secondary ABCs may be seen with fibrous dysplasia, osteoblastoma, chondromyxoid fibroma. nonossifying chondroblastoma, fibroma. 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.9 Other differentials 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 and can sometimes

present with a secondary ABC. 9,10 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.

Curettage and bone grafting is the standard treatment for ABCs in long bones.⁶ 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.¹¹⁻¹³ Partial or total talectomy along with tibiocalcaneal arthrodesis has also been described for lesions that show extensive destruction of the talus and soft tissue or subtalar extension.^{12,14} Luna *et al*¹¹, have described the use of external fixation in place

of a traditional cast after curettage and bone grafting for ABC of talus.

In conclusion, the possibility of ABC should be considered in patients presenting with a lytic lesion in the talus, although it is considered rare. On imaging alone, differentiating talar ABCs from GCTs is difficult. Histology is required for the diagnosis of an ABC. Primary ABCs treated by intralesional curettage and bone grafting provide an excellent prognosis.

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