AN UNUSUAL CASE OF ANEURYSMAL BONE CYST OF THE CALCANEUM: A CASE REPORT

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ABSTRACT: Aneurysmal Bone Cyst (ABC, is a locally aggressive benign solitary lesion of bone. Its occurrence within the calcaneum is rare and clinically present with painful ankle swelling. Only a few cases of calcaneal aneurysmal bone cysts have been reported in the literature so far. We report a case of aneurysmal bone cyst of the calcaneum in a 34 year old male who was successfully treated with surgical curettage and bone grafting. Definitive diagnosis and proper management warrants clinical, radiological and histopathological correlation and also to rule out other cystic lesions of calcaneum. **KEYWORDS:** Aneurysmal Bone Cyst, calcaneum, osteoclastic type giant cells, surgical curettage.

INTRODUCTION: Aneurysmal Bone Cyst is a benign tumor of bone and generally occurs during first two decades of life without sex predilection¹. It was first described by Jaffe and Lichtenstein in 1942², mainly affecting the metaphyseal region of long bones and vertebrae.³ Its frequency of occurrence within calcaneum is exceptionally rare, presenting as chronic heel pain and swelling.^{4,5} Various treatment methods are available based on size and site of the lesion.⁶ Our patient had swelling over lateral aspect of right heel and based on clinical, radiological and histopatholgical correlation, a diagnosis of aneurysmal bone cyst of calcaneum was made. He was managed with surgical curettage and calcaneal autologous bone grafting using sandwich technique. Postoperative period was uneventful without any evidence of recurrence.

CASE HISTORY: A 34 year old male presented with chronic heel pain which used to aggravate on walking since 5 months. His past and family history was insignificant. On clinical evaluation, a tender swelling was noted over the lateral aspect of right heel. There was no local rise of temperature and skin over the swelling was stretched out. Radiological examination of right heel revealed two eccentrically situated expansile multiloculated lytic lesion within calcaneum. MRI scan showed thin trabeculae traversing the cystic cavity. There was no breech in the cortex and no involvement of adjacent soft tissues.

Based on these findings, a diagnosis of benign cystic lesion of right calcaneum possibly aneurysmal bone cyst of the calcaneum was offered (Fig. 1 & 2). Subsequently surgical curettage was done followed by autologous bone grafting and gel foam filling was done using Sandwich method.⁷ A curvilinear skin incision was made over lateral aspect of right heel and calcaneum was exposed. A large cortical window was made and entire cyst was curetted out (Fig. 3).

Then cavity was washed with saline and autologous calcaneal bone graft of 1cm thickness was packed at the periphery. A layer of gel foam was applied over the graft and rest of the cavity was filled with Poly Methyl Methacrylate (PMMA) bone cement. Postoperative recovery was uneventful. Patient was advised non weight bearing of affected limb for 8 weeks.

The curetted specimen was sent for histopathological evaluation. On gross, multiple grey brown fragmented granular bits had blood filled cystic spaces. Microscopically, varying sizes of blood filled dilated and ectatic vessels focally lined by fibrous tissue was evident. Solid areas showed reactive osteoid formation and scattered osteoclastic type of giant cells were seen embedded in spindle cell stroma (Fig. 4 & 5). There was no evidence of malignancy. Based on these findings, histological confirmation of aneurysmal bone cyst of calcaneum was given.

Patient was followed up for next 24 months, during which he was clinically asymptomatic and had no evidence of recurrence.

DISCUSSION: Aneurysmal Bone Cysts (ABC) of foot is extremely rare, comprising of only 1% of all primary bone tumors.⁸ They are locally aggressive and expansile lesions and usually impinge on surrounding tissues causing pain, swelling, neurological deficits, disruption of growth plates and rarely pathological fracture.⁶ The exact etio-pathogenesis remains uncertain. Vascular malformations within the bone and genetic association of this tumor with distinctive 17p13 translocation resulting in up regulation of deubiquitinating enzyme USP6 have been speculated.⁹

ABCs are common around knee joint and nearly 70% of cases occur in second decade of life⁴. Our patient was 34 year old male presenting with calcaneal ABC, which makes this case even more unusual.

Calcaneal bone cysts include both neoplastic and non-neoplastic conditions, ranging from simple bone cyst to aggressive telangiectatic osteosarcoma⁵. About one-third of cases are associated with pre-existing giant cell tumors.¹⁰

Therefore exact diagnosis warrants clinical, radiological and histological correlation. Clinically, most of the cases are asymptomatic and detected incidentally¹¹. Only a few cases, like ours present with clinically significant painful swelling associated with difficulty in walking. Radiologically, ABCs presents as eccentrically situated expansile lytic lesion and show multiple fluid levels on MRI scan as a result of blood layering.¹²

Calcaneum, being an unusual site for aneurysmal bone cyst, histological confirmation is necessary to differentiate it from other cystic lesions and also to rule out any associated malignancy. Radiological and histological features of our case were consistent with aneurysmal bone cyst and there was no associated malignancy.

There are various treatment modalities available like surgical curettage supplemented with bone grafting, phenol installation, PMMA cementing and Sandwich technique of autologous bone grafting and filling⁶. Although ABCs have favorable outcome with a cure rate of 90-95%, incidence of recurrence ranges from 50-60%.^{13, 14}

In such cases, wide excision or arterial embolization may be considered. Our patient was managed with surgical curettage followed by calcaneal autologous bone grafting and bi-layering with gel foam and PMMA cement using Sandwich method. Postoperative follow up for next two years was uneventful.

CONCLUSION: ABCs of calcaneum is an extremely rare entity. Proper diagnosis and appropriate therapeutic management is necessary not only prevent the recurrence but also to exclude other cystic lesions of calcaneum. Surgical curettage supplemented autologous bone grafting with multi-layering using Sandwich method is valuable option.

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Fig. 1: X-ray (lateral view) of right foot showing two expansile lytic lesions in calcaneum



Fig. 2: MRI scan of right ankle- showing cystic spaces traversed by thin trabaculae



Fig. 3: Intra-operative photograph showing Aneurysmal bone cyst of right calcaneum

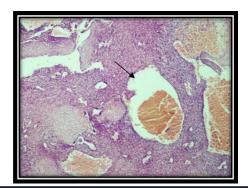


Fig. 4: Photomicrography showing blood filled dilated and ecstatic vessels lined by fibrous tissue. [H& E stain, 100X]

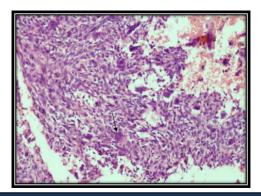


Fig. 5: Photomicrography showing scattered osteoclastic type of giant cells embedded in spindle cell stroma. [H& E stain, 400X]

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