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CASE REPORT

A RARE CASE OF ANEURYSMAL BONE CYST OF THE CALCANEUM

***,[1]Dr. Dibakar Ray, [1]Dr. Asik Iqbal [1]Dr. Writtik Porel, [2]Dr. Biswajit Biswas and [1]Dr. Mandal, P.K.**

[1]Orthopaedics BSMCH, West Bengal, India
[2]Pathology, CM and JNMH, KALYANI, West Bengal, India

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ABSTRACT

Aneurysmal bone cyst (ABC) is a benign solitary bone lesion of unknown etiology. ABCs mainly occur in the long bones but only rarely in the bones of the feet. Few cases of ABC involving the calcaneum have been reported.We report an unusual case of ABC in calcaneum in a 23 year old male, as only 1% of this type of tumour is found in the calcaneum. This article discusses the etiology, clinical presentation, radiological features, histological features and preferred examinations of aneurysmal bone cysts.

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INTRODUCTION

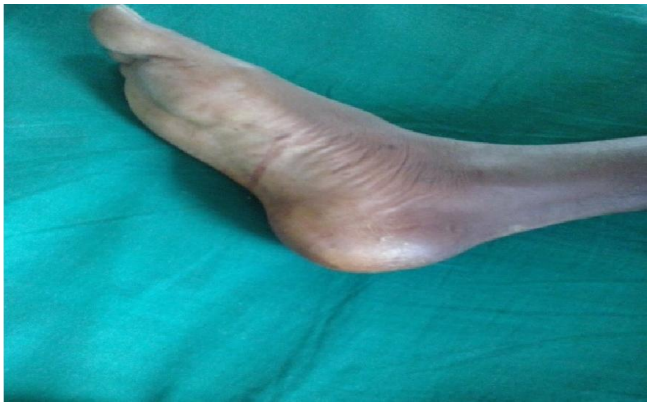
The aneurysmal bone cyst (ABC) is an expansile cystic lesion that most often affects individuals during their second decade of life and may occur in any bone in the body. (Clayer *et al.,* 2008; Segall *et al.,* 2008) Although benign, the ABC can be locally aggressive and can cause extensive weakening of the bony structure and impinge on the surrounding tissues. Jaffe and Lichtenstein first described ABC in 1942 (Jaffe *et al.,* 1942). As defined by the World Health Organization, the ABC is a benign tumor like lesion described as "*an expanding osteolytic lesion consisting of blood-filled spaces of variable size separated by connective tissue septa containing trabeculae or osteoid tissue and osteoclast giant cells (*Brastianos *et al.,* 2009*)*. ABCs both erode and cause 'expansion' of underlying cancellous and cortical bone (Campanacci *et al.,* 1986). Around the lesion there is always a shell formed by periosteal new bone and, although this may be only millimeters thick, it prevents direct extension into the soft tissues (Enneking *et al.,* 1983). The expansile nature of the lesions cancause pain, swelling, deformity, disruption of growth plates, neurologic symptoms (depending on its location), and pathologic fracture (Clayer *et al.,* 2008; Segall *et al.,* 2008; Burch *et al.,* 2008). ABC's in the foot are uncommon. ABC's present about 1% of all primary bone tumors collectively (Duke *et al.,* 2007). Its frequency of occurrence in foot is only about 3% compared to other bones of body (Anand *et al.,* 2007). Occurrence within the calcaneum are rare, and generally present as chronic heel pain and swelling, but may rarely present as pathologic fracture (Unni *et al.,* 2010). A plethora of cystic lesions can occur in the calcaneum, which makes definitive diagnosis difficult based on imaging only. The differential diagnosis includes simple bone cyst, ABC (primary or secondary), chondroblastoma, giant cell tumor (GCT), osteosarcoma, ossifying hematoma or pseudotumor of hemophilia. This mandates histopathological diagnosis prior to the definitive management. We report a rare case of ABC involving calcaneum of 23 year-old male confirmed by histopathology report and radiological report.

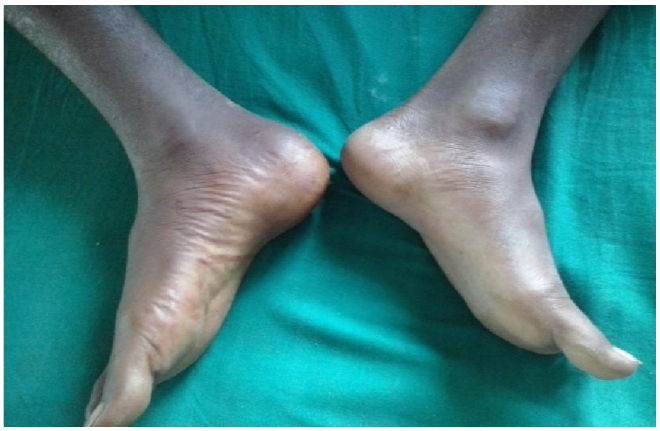
Corresponding author: Dr. Dibakar Ray,
Orthopaedics BSMCH, West Bengal, India.

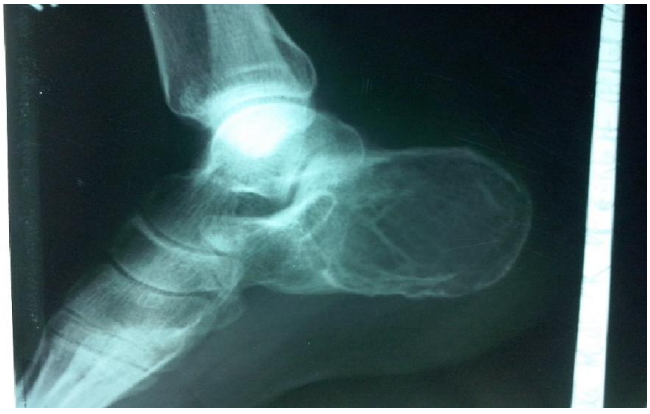
Case report

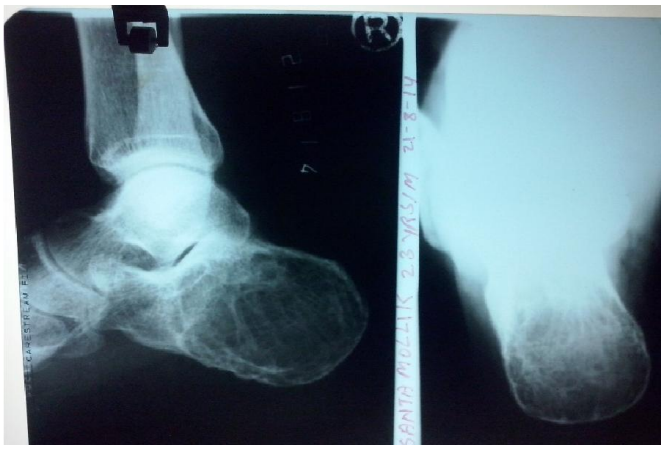
A 23 year-old male, mason by occupation,presented with a chief complaint of swelling in the right heel during the last 6 months. An increase in swelling was associated with pain in heel from the last 3 months. He had difficulty in walking because of pain. He did give history of blunt trauma 2 years prior to the onset of symptoms. Clinical evaluation revealed swelling over the medial and lateral aspect of the heel and the skin over the swelling was stretched. Tenderness was present on palpation but there was no local rise of temperature. The swelling was bony hard in consistency and arising from calcaneum. There were no distal neurovascular deficits or any significant lymphadenopathy. Radiographic examination of his ankle revealed an eccentric, expansile, multiloculated lytic lesion of the calcaneum with thin trabeculae traversing the cystic cavity. There was no breach in the cortex. Based on clinical and radiological findings, a diagnosis of benign cystic lesion of right calcaneum was made. Plain antero- posterior and lateral radiograph views showing eccentric expansile lytic lesion with thin shell of cortex and trabeculae traversing the

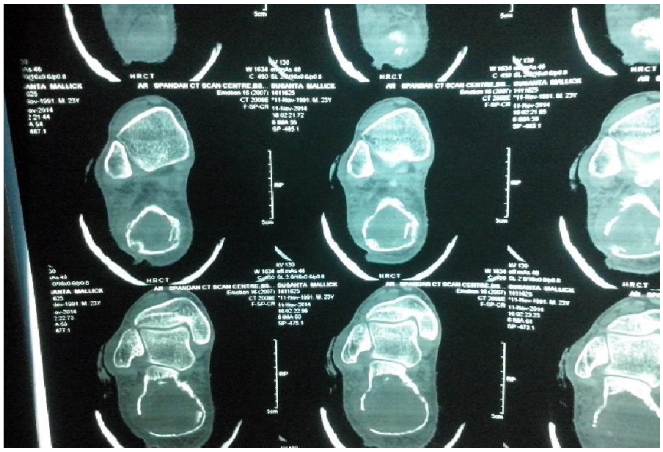
cyst. The patient was referred for computed tomography (CT) of his right foot.

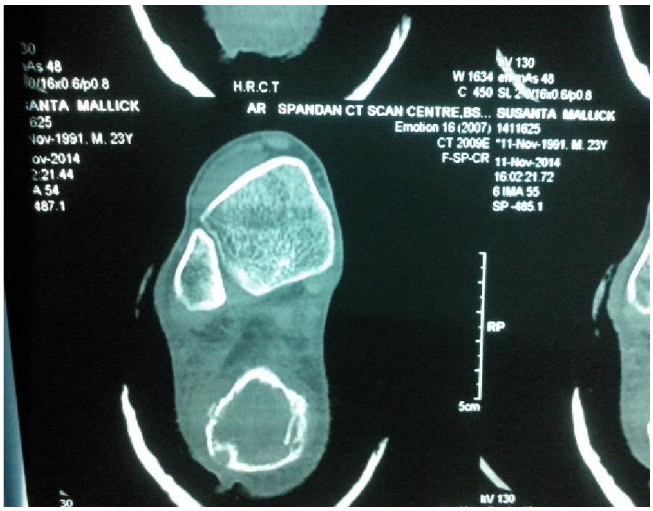




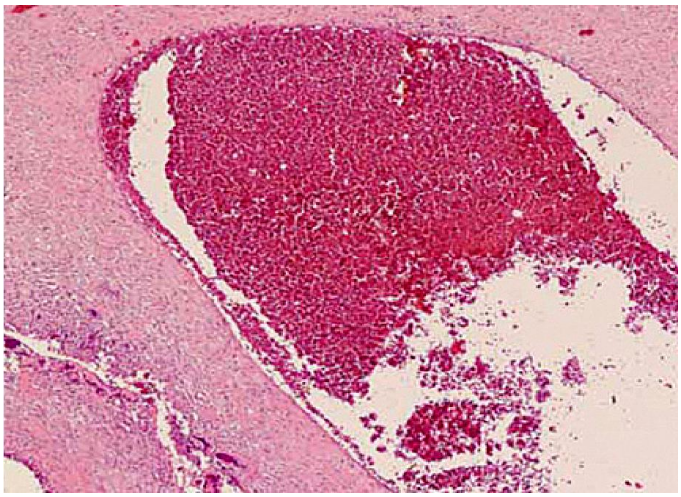


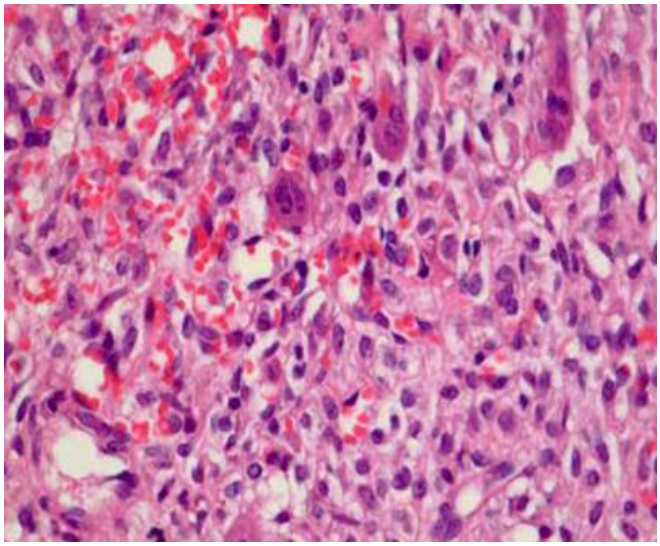






Three millimeter axial cuts were employed and these demonstrated a well-defined, expansile lytic lesion withfew septations and thinned out cortex noted the calcaneum on the right side. There is no significant soft tissue swelling around the lesion. The mass lesion measured 5cm x 6cm in diameter. Open biopsy of the cyst was made to confirm the diagnosis. The cyst grossly consisted of cavities filled with brown altered blood. Histopathological report revealed large blood filled cavities lined by fibrous septa, with occasional osteoclastic giant cells, hemosiderin laden macrophages with a thin rim of bone.





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Hence the diagnosis of ABC involving the right calcaneum was made. The patient is currently awaiting further evaluation and treatment.

DISCUSSION

ABC is an entity on its own having unique clinical, radiological and diagnostic behavior (Campanacci *et al.,* 1986). The exact aetiology of this tumour is unknown. The descriptive name is derived from the macroscopic appearances of blood-filled, expansile , sponge-like tumour containing numerous giant cells (Sutton *et al.,* 1987). These lesions can develop de novo or as a result of cystic changes in a pre-existing lesion such as a chondroblastoma, osteoblastoma, giant cell tumour or fibrous dysplasia (Greenspan *et al.,* 1992). A giant cell tumour is the most common cause, occurring in 19%-39% of cases and, whatever the aetiology, aneurysmal bone cyst has a close relationship to all the giant cell variants (Sutton *et al.,* 1987). Some reports suggest that aneurysmal bone cyst is not a true neoplasm sinceit has been shown to arise in association with other abnormalities of theskeleton (Sutton *et al.,* 1987). Trauma is also considered an initiating factor in the pathogenesis of some cysts in well-documented cases involving acute fracture. Local hemodynamic alterations to venous obstruction or arteriovenous fistulae that occur after an injury are important in the pathogenesis of an aneurysmal bone cyst (Anand *et al.,* 2007). Aneurysmal bone cysts are seen predominantly in children: 90% of these lesions occur in patients under 20 years (Greenspan *et al.,* 1992). An aneurysmal bone cyst may occur in patients between the ages of 10-30 years with a peak incidence in those aged 16 years (Anand *et al.,* 2007). Three quarters of aneurysmal bone cysts occur before epiphyseal fusion has commenced. ABC has slight female predominance (Sutton *et al.,* 1987).

The location is usually metaphyseal when the long bones are involved. Purely diaphyseal lesions are seen in 8% of cases. Extension into the epiphysis is even more rare (Dahnert *et al.,* 1990). Aneurysmal bone cysts have a predilection for the long bones and lumbar spine (Sutton *et al.,* 1987). Those tumours arising in the spine occur slightly later, between 10 and 20 years. The neural arch is more commonly involved than the body, half of these cases involving more than one vertebra. The prognosis is entirely benign apart from secondary neurological lesions due to spinal canal compression (Sutton *et al.,* 1987). Regarding the location of the lesions any bone may be affected.

Approximate frequencies by site are listed below:

- Skull and mandible (4%)
- Spine (16%)
- Clavicle and ribs (5%)
- Upper extremities (21%)
- Pelvis and sacrum (12%)
- Femur (13%); (24%)
- Foot (3%)
- Lower leg (23%) (Anand *et al.,* 2007)

The most common site is the metaphyseal region of the knee. Histologically, the lesion consists of multiple blood-filled sinusoid spaces alternating with more solid areas. The solid tissue is composed of fibrous elements containing numerous multinucleated giant cells and is richly vascular. The sinusoids have fibrous walls often containing osteoid tissue or even mature bone (Greenspan *et al.,* 1992). The clinical manifestation depends on the specific site of involvement. A common presentation includes pain of relatively acute onset that rapidly increases in severity over 6-12 weeks. The local skin temperature may increase, a palpable bony swelling may be present and movement in an adjacent joint may be restricted; spinal lesions may cause neurologic radiculopathy or quadriplegia and patients may have moderate to severe headaches (Anand *et al.,* 2007; Dahnert *et al.,* 1990). Diagnostic imaging plays a role in patient management. At radiography, aneurysmal bone cysts are well-defined, cortically based, rapidly expansile lytic lesions. They can grow large enough to involve the medullary cavity, although their tendency is for eccentric expansion, a so-called blister lesion (Levine *et al.,* 2003) . A typical area of bone resorption occurs with slight or marked expansion. The size of the lesion can vary between 2 cm and in gross examples as much as 20 cms in diameter. The overlying cortex is thinned and may be expanded to such a degree that in places it can be identified only by tomography or CT (Sutton *et al.,* 1987). Radiographs usually are adequate for diagnosis. The classic description of an aneurysmal bone cyst in tubular bones includes an eccentric radiolucency and a purely lytic or, occasionally, trabecular process, with its epicenter in the metaphysis of an unfused long bone. The trabeculae in the cyst may create a soap-bubble appearance in the lesion (Anand *et al.,* 2007). The margins of the lesion are well defined, with a smooth inner margin and a rim of bone sclerosis. The tumor does not usually extend into the epiphyseal plate until after complete fusion, when it may occasionally do so. The expansion or ballooning of the cortex occasionally may result in the loss of the sharp definition of its margin and should then be interpreted as an aggressive lesion.
Bone cysts of the calcaneum are rare lesions. These may include a wide spectrum of non-neoplastic cysts, benign or malignant neoplastic lesions ranging from simple bone cyst, ABC (primary or secondary), chondroblastoma, giant cell tumor (GCT), and an osteosarcoma (especially telangiectatic)

(Unni *et al.,* 2010). Clinically, calcaneal cysts are often symptomatic and present with heel pain, although some of these lesions may remain asymptomatic and are detected as incidental findings. Even though there are many typical radiograph, computed tomographic (CT) scan, and magnetic resonance imaging (MRI) findings to confirm a diagnosis of ABC, an open biopsy must be performed because of the high frequency of accompanying tumors (Unni *et al.,* 2010) . When a biopsy is performed, the sample should ideally include material from the entire lesion; a limited biopsy could easily cause a coexisting lesion to be missed, leaving the patient with a morbid prognosis. There are various methods of treatment based on the site and size of the lesion, which include curettage, which may be supplemented with various adjuvant therapies such as bone grafting, use of liquid nitrogen, phenol instillation and Poly (methyl methacrylate) (PMMA) cement. Radiotherapy valuable in spinal lesions where surgery may be considered hazardous. An increasing role, however, has developed for trans-catheter embolisation in the successful management of these tumours. Aneurysmal bone cyst is a particularly good example of the importance of radiological investigation being complete before biopsy is undertaken, since these lesions, not surprisingly, bleed considerably and the pre-operative demonstration of the vascular nature of this tumour is very important (Sutton *et al.,* 1987). Although relatively rare, there is no reason to assume that ABCs of the feet will respond to treatment or recur any differently from ABCs that occur elsewhere in the body. Surgical curettage is sufficient to treat most ABCs of the feet, including the calcaneum (Chowdhry *et al.,* 2010).

To conclude, ABC of the calcaneum is an extremely uncommon entity. Proper diagnosis entails correlating the clinical presentation, anatomical location, radiological profile, and histopathological appearance. This is imperative not only to exclude other more common histological mimics, but also for choosing the appropriate therapeutic regimen and prognosticating the disease outcome. In a case of calcaneal cystic lesion, ABC should be considered as one of the differential diagnosis. Hence histological diagnosis is essential.

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