



# Aneurysmal bone cyst of the fibular malleolus: Report of an additional case at rare site

Jean Marie Vianney Hope<sup>1\*</sup>; Mohamed Sidibe<sup>2</sup>; Jean Claude Sane<sup>3</sup>; Souleymane Diao<sup>2</sup>; Joseph Davy Diouf<sup>2</sup>; Anselme Noaga Nikiema<sup>2</sup>; Amadou Ndiassé Kassé<sup>2</sup>; Mouhamadou Habib Sy<sup>2</sup>

<sup>1</sup>Orthopedics and Trauma Surgery, Department of Rwanda Military Hospital (RMH), University of Rwanda, East Africa

<sup>2</sup>Orthopedics and Trauma Surgery, Department of Grand-Yoff General Hospital, Cheikh Anta Diop University of Dakar, Senegal

<sup>3</sup>Gaston Berger University of Saint-Louis, Senegal

## \*Corresponding Author(s): Jean MV Hope

Fellow West African College of Surgeons (FWACS), Orthopedics and Trauma Surgery, Department of Rwanda Military Hospital (RMH), University of Rwanda, East Africa

Tel: +250789364492, Fax: +250252586420,

ORCID ID: 0000-0003-1127-0586,

Email: zpawlak@xmission.com

## Abstract

Aneurysmal Bone Cyst (ABC) is a benign expansile lesions that may occur in any bone in the body, often affecting individuals during their second decade of life. In the modern English language literature, aneurysmal bone cyst occurring at the level of fibular malleolus is rare. Here, we report an additional such case arising from the fibular malleolus of a 16-year-old female patient successfully treated with en-bloc excision and ankle joint stabilization with tibiotalar joint arthrodesis by two cancellous screws and lateral ligament reconstruction.

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## Introduction

As defined by the World Health Organization (WHO), the Aneurysmal Bone Cyst (ABC) is a benign tumor like lesion that is 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 [1,2]. It most often affects individuals during their second decade of life and may occur in any bone in the body [3,4,5,6]. However, it is generally considered rare, accounting for only 1-4% of all benign bone tumors [7]. The fibular localization is infrequent representing less than 7.3% of all ABCs [8]. The purpose of this study is to report an additional case of ABC of the fibular malleolus of a 16-year-old female patient treated with en-bloc excision and ankle joint stabilization.

## Case report

A 16-year-old girl with no pertinent past personal or family medical history presented with difficulty in movements of ankle joint and gradual distal right leg swelling since 6 months. She attributed these symptoms to a simple fall on her right ankle she sustained while walking. Physical examination was notable for limping and a mass of the lateral aspect of the distal one-fourth of the right leg. The mass was tender on palpation, hard in consistence, immobile in both planes; and hence clinically proving it was arising from the bone. It measured 8cm × 3 cm. Movements of the ankle joint were restricted due to pain. Blood investigations were in normal limits except slight increase in alkaline phosphatase at 167U/L.



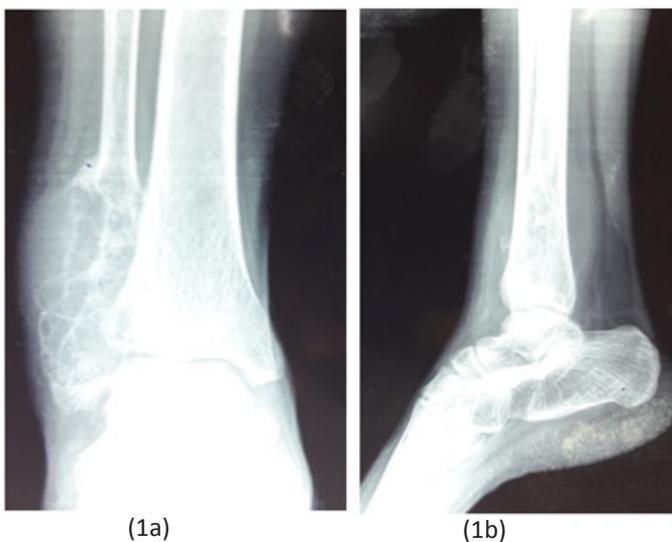
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Plain radiograph of the right ankle joint revealed an eccentrically placed expansile lytic lesion at distal end of fibula with extensive cortical thinning and multiple internal septations (Figure 1 a & b). Reconstructed sagittal and coronal Computed Tomography (CT) images were helpful in improved demonstration of some features, such as the cortical erosion, internal septations with calcified rim and eggshell appearance. Fluid-fluid levels can also be seen. No associated soft tissue mass was present (Figure 2 a & b).

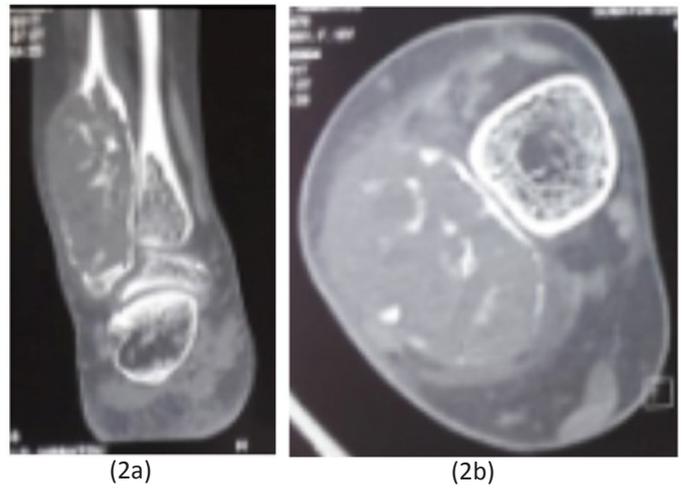
Biopsy was then carried out, and the specimen sent to the lab for histological examination. The gross appearance was that of a blood-soaked sponge surrounded by a thinned sub periosteal shell and containing cystic blood-filled cavities. The tissue within showed brownish intertwining septa. The stroma contained proliferative fibroblasts, spindle cells, areas of osteoid formation, and an uneven distribution of multinucleated giant cells that tended to surround the fluid-filled cavities in a “pigs at the trough” formation. Biopsy report also revealed the absence of malignant cells (Figure 3).

Under epidural anesthesia and tourniquet control, the patient was placed in supine position on operating table. The external approach centered on the lateral part of the middle of the fibula was performed. The skin incision begun to the lateral aspect of the middle of the leg and extended distally 1 cm below the end of lateral malleolus (Figure 4). She underwent en-bloc excision of the tumor and ankle joint stabilization by tibiotalar arthrodesis with two cancellous screws (Figure 5 a & b). The peroneus brevis tendon was transected proximally at its myotendinous junction and then sutured to the calcaneofibular and anterior talofibular ligaments in sequence and then tenodesed to the lateral distal tibia. No adjuvant therapy was administered. Postoperatively, the patient was immobilized in a cast for 6 weeks followed by gradual weight bearing of ankle joint. The patient made an eventful recovery with solid ankle joint fusion. The incision was by primary intension. The Musculoskeletal Tumor Society (MSTS) score was 94.7% and the American Orthopaedic Foot and Ankle Society (AOFAS) score was excellent. She was followed up to 5 years without recurrence.

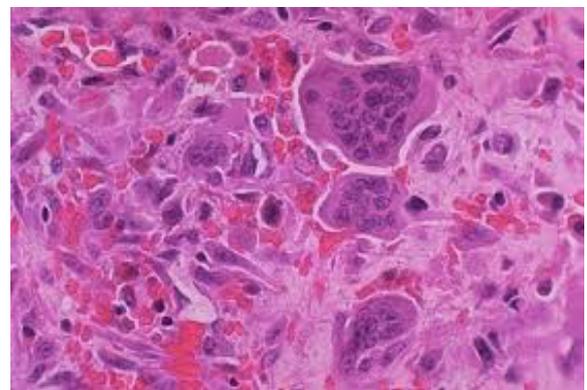
**Figures**



**Figure 1:** Anteroposterior (1a) and lateral (1b) radiographs of the right ankle showing an eccentrically placed expanded osteolytic lesion of the fibular malleolus. It has well-defined margins and multiple internal



**Figure 2:** Reconstructed sagittal (2a) and coronal (2b) CT showing erosion of the margin of the expanded lesion, fluid-fluid levels, internal septations with calcified rim and eggshell appearance. No associated soft tissue mass was seen.



**Figure 3:** Photomicrograph of the excised ABC, at 40x magnification, showing highly vascular tumor with blood-filled spaces without endothelial lining. The tissue within shows intertwining septa. The stroma contains proliferative fibroblasts, spindle cells, areas of osteoid formation, uneven distribution of multinucleated giant cells that tend to surround the fluid-filled cavities in a “pigs at the trough” formation. No malignant cells are found.



**Figure 4:** Skin incision.



**Figure 5:** Anteroposterior (1a) and lateral (1b) postoperative radiographs showing the ankle joint arthrodesis with two cancellous screws.

### Discussion

Previously considered as a giant cell tumor variety, Jaffe and Lichtenstein first described ABC as its own entity in 1942, when they noted a peculiar blood-containing cyst of large size [9]. The true etiology and pathophysiology remain a mystery. Most investigators believe that ABCs are the result of a vascular malformation within the bone; however, the ultimate cause of the malformation is a topic of controversy. Three commonly theories are proposed. Firstly, ABCs occur as reaction secondary to another bony lesion including mainly giant cell tumors, fibrous dysplasia, osteoblastoma, chondromyxoid fibroma, non-ossifying fibroma, chondroblastoma, osteosarcoma, chondrosarcoma, unicameral or solitary bone cyst, hemangi endothelioma and metastatic carcinoma. ABCs in the presence of another lesion are called secondary ABCs. Secondly, ABCs may arise de novo; those that arise without evidence of another lesion are classified as primary ABCs. Thirdly, ABCs may arise in an area of previous trauma as results of either arteriovenous fistulas, venous blockage or local hemorrhage that initiates the formation of reactive osteolytic tissue [4,10,11,12,13]. The third theory may explain the occurrence of ABC in our case.

The age distribution of those with ABC is distinct, with the majority of lesions occurring in skeletally immature patients. It most often affects individuals during their second decade [3,4,5,6,14]. Some authors reported sex predilection with female predominance [15,16]. These findings concur with our case (16 years old female patient). Patients with an ABC usually present with pain, a mass, swelling, a pathologic fracture, or a combination of these symptoms in the affected area. The symptoms are usually present for several weeks to months before the diagnosis is made, and the patient may also have a history of a rapidly enlarging mass [17,18,19]. This is in line with our case who presented with pain, swelling and limited range of motion of the ankle joint over 6 months.

ABCs may affect any bone in the body; the long tubular bones being the most common sites, followed by the spine and the flat bones. In a published review of 897 cases of ABC, the reported rates of occurrence were high in tibia (17.5%), followed by femur (15.9%), vertebra (11.2%), pelvis (11.6%) and humerus (9.1%) while fibular localization represent 7.3% [8]. Other few cases of fibular localization have been reported [20,21,22,23]. Lesion at distal end of fibula is definitely a rare entity as pre-

sented in our case.

Radiographic findings usually consist of an eccentric lesion that appears cystic or lytic. Images may show expansion of the surrounding bone with a blown-out, ballooned, or soap-bubble appearance. Some views may show an eggshell-appearing bony rim surrounding the lesion. One may see the cystic spaces and, rarely, partially ossified septa [24]. These radiological findings have been found in our case. Capanna et al [25] described five morphologic types of ABC on the basis of radiographic findings whereas Lodwick radiographic grading with bone destruction distinguishes three grades [26]. The staging system of benign musculoskeletal neoplasms adopted by the Musculoskeletal Tumor Society (MSTS) was described by Enneking in 1986, who classified benign lesions into three broad categories with stage 1: latent (inactive); stage 2: active and stage 3: aggressive [27]. On the basis of radiographic findings, our case was classified Capanna morphologic type V and Lodwick grade III. Based on broad categories of Enneking, it was described as stage 2.

Some authors [22,28] have stressed the importance of physal preservation in the treatment of aneurysmal bone cysts even if lesions are likely to recur. By contrast, when a fibular location is considered, the expression “expendable bone” is sometimes used, justifying more aggressive treatment [28]. But, the mainstay of treatment has been intralesional curettage. Other surgical options include selective arterial embolization, and curettage with locally applied adjuvants such as liquid nitrogen, argon beam photocoagulation, or phenol [29,30]. The main reconstructive problems after resection are restoration of fibular length and instability of the ankle [21,22]. In our case, we performed en-bloc excision of the entire distal fibula. Stabilization of the tibiotalar joint was done by tibiotalar joint arthrodesis by two cancellous screws. We furthermore performed the reconstitution of lateral ligaments. No recurrence was seen after 5 years.

### Conclusion

We report an additional rare case of Aneurysmal Bone Cyst (ABC) arising from fibular malleolus of a 16-year-old female patient successfully treated with en-bloc excision and ankle joint stabilization with tibiotalar joint arthrodesis by two cancellous screws and lateral ligament reconstruction. The clinical and imaging features alone are non-specific, and the definitive diagnosis relies on pathological examination.

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### Ethical Approval

This article does not contain any studies with human participants or animals performed by any of the authors.

### Informed Consent

The informed consent was obtained from the child’s parents to publish the information, including his photograph.

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