

## Extrasosseous Aneurysmal Bone Cyst of the Ankle

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### Abstract

**Introduction:** Extrasosseous aneurysmal bone cyst (ACB) represents an exceptional and extremely rare variant of aneurysmal bone cysts. This presentation deserves to be known by clinicians and radiologists to avoid diagnostic delays.

We present an original case of extrasosseous ABC arising in the soft tissues of the ankle.

**Case report:** A 16-year-old patient, without pathological medical history, was explored for a discomfort of the left foot when wearing shoes, painless and progressive swelling of the left ankle that has been evolving for two months. The standard X-ray of the ankle and ultrasound of the soft parts of the ankle were suggestive of myositis ossificans. The patient was operated with a complete curettage and spongy graft. The histological examination confirmed the diagnosis of ABC of soft tissues. The evolution was favorable.

**Conclusion:** The localization in the soft parts of the ankle of these benign bone tumors has not been reported before. Our observation is, to our knowledge, the first reporting this location.

**Keywords :** Extrasosseous aneurysmal bone cyst, aneurysmal bone cyst, ankle, soft tissues.

### 1. INTRODUCTION

The aneurysmal bone cyst (ABC) is a relatively recent nosological entity of individualization, since its first description dates back to 1942 by Jaffe HL and Lichtenstein L [1]. It has since been recognized and defined by the "Histopathological Classification of Primary Bone Tumors" of the World Health Organization (WHO) initially established in 1972 and revised in 1993, as "a benign, expansive osteolytic lesion, occurring most often in the metaphysis of long bones, vertebrae and flat bones, consisting of multiple hematological gaps separated by connective septa containing osteoclasts and reactive osteogenesis"; ABC is thus considered as a "tumor-like" lesion (group IX of primary bone tumors according to the WHO classification" [2]. It can be primitive in 70% of cases, developing in anteriorly normal bone, or secondary in 30% of cases emerging from other benign or malignant bone lesions [3-5].

It is a rare tumor representing only 1-2% of all bone tumors and about 10% of benign bone tumors [6]. Its incidence is estimated at 0.14 to

0.32/100,000/year, and is particularly common before the age of 20 and in female patients [4-6].

Beside classical ABC, atypical and unusual forms of this tumor (solid ABC and extrasosseous or soft tissue ABC) can be seen and represent a real diagnostic challenge for clinicians [5].

We report an original observation of extrasosseous ABC arising in the soft tissues of the ankle; location not previously reported in the English medical literature.

### 2. CASE REPORT

A 16-year-old patient, without pathological medical history, was explored for a discomfort of the left foot when wearing shoes, painless and progressive swelling of the left ankle that has been evolving for two months. No concept of recent trauma or old fracture has been reported.

The somatic examination noted a deformed left ankle with subcutaneous swelling, hard, painless, non-mobile, and opposite the medial face of the calcaneus. The ankle joint was supple, painless at mobilization, and with preserved area of mobility.

Apart from this swelling, the somatic examination was without abnormalities as well as the basic biological assessment: total blood count, erythrocyte sedimentation rate, C-reactive protein, serum calcium, phosphoremia, alkaline phosphatase, creatinine, fasting glucose, uric acid, ionogram, transaminases, muscle enzymes, lactate dehydrogenase, and electrophoresis of serum proteins.

The standard X-ray of the ankle showed an extra-osseous opacity, rounded, well limited by a thin bone shell, containing some bony trabeculae inside, located opposite the medial face of the calcaneus, and measuring about 3 cm diameter (Fig 1). No associated lytic bone lesions were noted.



**Fig1. X-ray of the ankle:** extra-osseous opacity 3 cm in diameter, limited by a thin bone shell, containing some bony trabeculae on the medial side of the calcaneus.

Ultrasound of the soft parts of the ankle revealed a hypoechoic mass, encapsulated by a hyperechoic shell, developing in the soft parts opposite the medial aspect of the calcaneus, and which does not seem to adhere to the cortical bone of the latter (Fig 2).

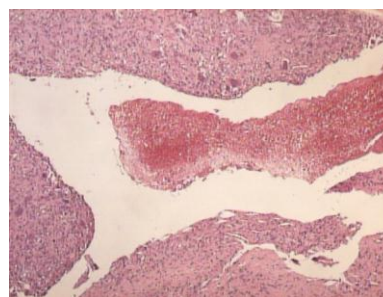


**Fig2. Ultrasound of the soft parts of the ankle:** well encapsulated mass limited by a hyperechoic bone shell, opposite the medial face of the calcaneus.

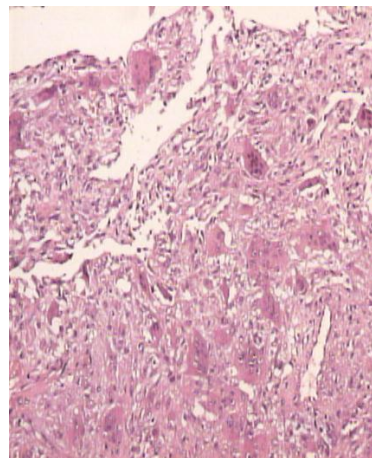
The patient was transferred to the department of orthopedic surgery with the diagnosis of myositis

ossificans. The procedure consisted of a complete curettage with spongy graft. The postoperative course was simple.

Anatomopathological examination of the operative specimen showed macroscopically the presence of multiple rounded hemorrhagic gaps separated from each other by connective septa. Microscopically, cavities were filled with blood and without endothelial lining (Fig 3), septa lined with fibroblasts, osteoclastic giant cells, and reactive bone tissue lined by osteoblasts (Fig 4). The electron microscopy study confirmed the presence of cells containing peripheral filaments suggesting their myofibroblastic differentiation. The histological aspect was compatible with the diagnosis of ABC in its extraosseous form.



**Fig3. Histological examination (Hematoxylin and Eosin×100):** Cystic cavity filled with blood without endothelial lining.



**Fig4. Histological examination (Hematoxylin and Eosin ×100):** wall of a cystic cavity rich in multinucleated giant cells (osteoclasts).

### 3. DISCUSSION

The soft tissues of the foot and ankle are generally unusual sites for the development of both benign and malignant primary tumors [7]. Cystic bone lesions are particularly rare in these soft tissues [8]. Of all these tumors, the ABC remains exceptional [7,8].

Extrasosseous or soft tissues ABC is an extremely rare variant of ABC [5,9,10]. His first description goes back to Salm R and Sissons HA in 1972 [11], and was long confused with giant cell tumor

of soft tissue [12] or myositis ossificans [13]. Since then only sporadic cases have been reported in the literature [9-10,14-17]; in fact, Lopez LV et al's review of English-language literature in 2017 found only 26 cases [9].

These forms of ABC are still unknown by health practitioners explaining the often significant diagnostic delay and often inadequate management [8,14]. They represent a real diagnostic challenge for both clinicians and radiologists [9].

These soft tissue ABCs can be seen at any age and in both sexes; the most frequent locations are the soft tissues of the lower limbs, arms, and shoulders [9,17]. The ankle remains an exceptional and unusual location; in fact, no case of extrasosseous ABC of the ankle has been found in the literature according to the 2017 review by Lopez LV et al [9].

The clinic is classically reduced to soft tissue pain and tumefaction, sometimes with palpation of an expansive mass [4]. Standard radiography may paradoxically remain normal [15]. Ultrasound and nuclear magnetic resonance imaging will be of great diagnostic value in these situations [4,5,15]. The diagnosis of certainty remains histological [7,9,16,17]. The treatment of choice for these particular forms remains surgical excision and the evolution is usually favorable [7,9,16,17].

#### 4. CONCLUSION

Soft tissue ACB or extrasosseous ACB represents an exceptional and extremely rare variant of aneurysmal bone cysts. This presentation deserves to be known by clinicians and radiologists to avoid diagnostic delays. The localization in the soft parts of the ankle of these benign bone tumors has not been reported before. Our observation is, to our knowledge, the first reporting this location.

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