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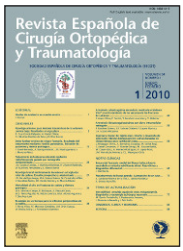

CLINICAL NOTE

# Subperiosteal Aneurysmal Bone Cyst Located in the Fifth Metatarsal Shaft in a 13 Year-Old Girl

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**KEYWORDS**
Aneurysmal bone cyst;
Tumour;
Foot

**Abstract**
*Objective:* We present s a case of aneurysmal bone cyst, unusual and rare, both for its location and its initial clinical and radiographic presentation.
*Case report:* A girl of 13 years consulting for atraumatic pain in the outer edge of right foot, with a normal initial X-ray and then one month later presented with a subperiosteal tumour lesion in the fifth metatarsal shaft, which was definitively diagnosed as an aneurysmal bone cyst, Campanacci type IV.
*Conclusions:* There are few cases in the literature of aneurysmal bone cyst with these characteristics. Such lesions may pose significant diagnostic difficulties with other pathologies, such as tumours, stress fractures or infections, due to its unusual presentation.


**PALABRAS CLAVE**
Quiste óseo aneurismático;
Tumor;
Pie

**Quiste óseo aneurismático subperióstico localizado en el quinto metatarsiano en una niña de 13 años: a propósito de un caso**

**Resumen**
*Objetivo:* Se presenta un caso poco frecuente de quiste óseo aneurismático por su localización y por su forma de presentación clínica y radiográfica inicial.
*Caso clínico:* Mujer de 13 años que consultó por dolor atraumático en el borde externo del pie derecho con radiografía inicial normal y presentación al cabo de un mes de una lesión tumoral subperióstica en la diáfisis del quinto metatarsiano, cuyo diagnóstico definitivo fue de quiste óseo aneurismático tipo iv de Campanacci.

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*Conclusiones:* Existen pocos casos en la literatura médica de quiste óseo aneurismático que presenten estas características. Por su localización poco frecuente, este tipo de lesiones pueden plantear dificultades diagnósticas con otro tipo de enfermedades, como tumores, fracturas de estrés o infecciones.


## Introduction

The WHO defines aneurysmal bone cyst as an expanding osteolytic lesion consisting of blood filled spaces of varying size separated by connective tissue septa that contain trabeculae of the bone or osteoid tissue, and giant osteoclastic cells.[1] It accounts for 6% of all primary bone lesions and appears mostly during childhood. More than three quarters (76%) of the patients are under the age of 20 years and the highest incidence is seen during the second decade of life. The yearly incidence is 0.4 cases per 100,000 inhabitants with a male/female ratio of 1:1.04.[2]

It typically affects the metaphyseal medullary space of the long bones, whereas the cortical or sub-periosteal location is rare. Long bones are generally the most commonly affected. The tibia is the bone that is most often affected, followed by the femur and the humerus.[3]

The clinical course is a quick one; in 72% of the cases, the period that elapses between the onset of symptoms and treatment is less than 6 months. The first symptom tends to be pain associated with swelling in the area and in 25% of the cases, the initial symptom is related to trauma.

We present the case of a patient with an atypically located aneurysmal bone cyst, rapidly progressing both clinically and radiologically from its clinical debut, coursing only with pain and lacking any radiographic manifestation initially.

*Case report*. Thirteen year old female patient presenting at the Emergency Room reporting diffuse pain in her foot right without any history of previous trauma to the area. The pain presented mechanical characteristics, increased on walking, and decreased with rest. An initial radiographic study was performed of the foot with dorso-plantar and oblique projections, with no apparent lesion (fig. 1); the patient was recommended to rest and treated with anti-inflammatories.

After 10 days, the patient returned to the Emergency Department again because of the persistence of the pain in her foot. On this occasion, the pain was located on the outer edge of the foot on the diaphysis of the fifth metatarsal; as a result, new x-rays were taken. This time, periosteal reaction was seen on the external cortex of the diaphysis of the fifth metatarsal and interpreted as a stress fracture (fig. 1). With this diagnosis, a plaster splint was applied and she was to be seen for a check-up 3 weeks later.

After this period of time had elapsed, the splint was removed and she was authorized to carry out gradual loading with orthotics, and she was given another appointment for check-up and X ray one month later. When the patient returned, she was asymptomatic. However, she presented a 2?2 cm area, slightly erythematous, non-fluctuating or painful on palpation and adhered to deep planes located on the outer edge of the foot above the fifth metatarsal. Radiographically, an eccentric, oval, poly-

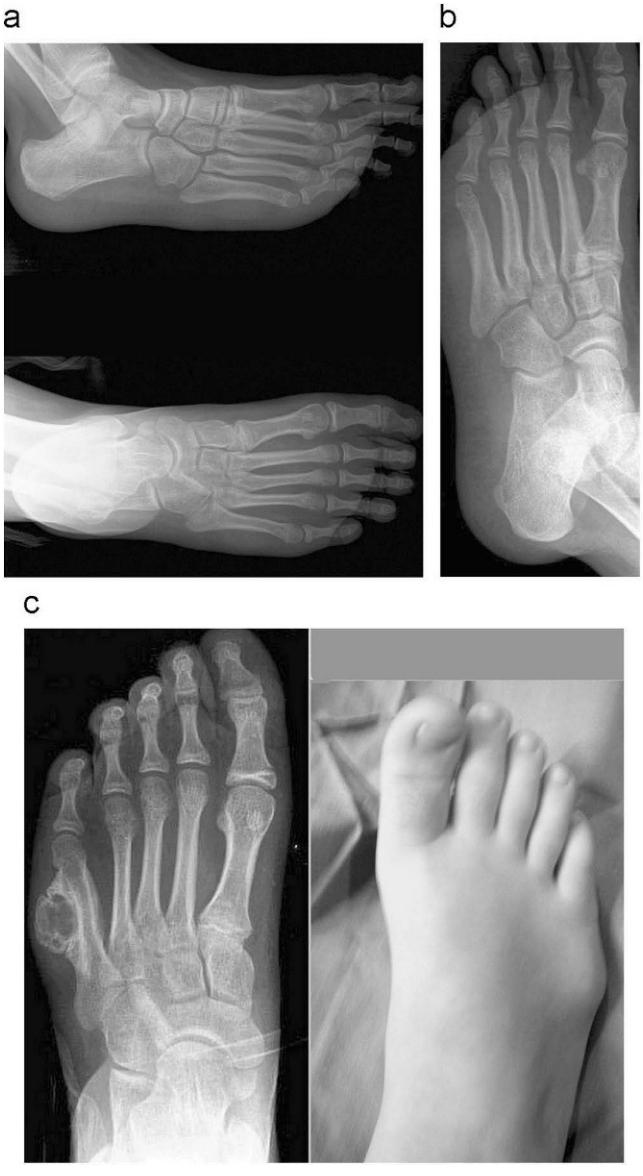


**Figure 1** a) Initial X-rays in which no lesion is detected. b) 10 days later, periosteal reaction in the external cortex of the fifth metatarsal. c) At 2 months with swelling on the external cortex of the fifth metatarsal and medial periosteal separation.

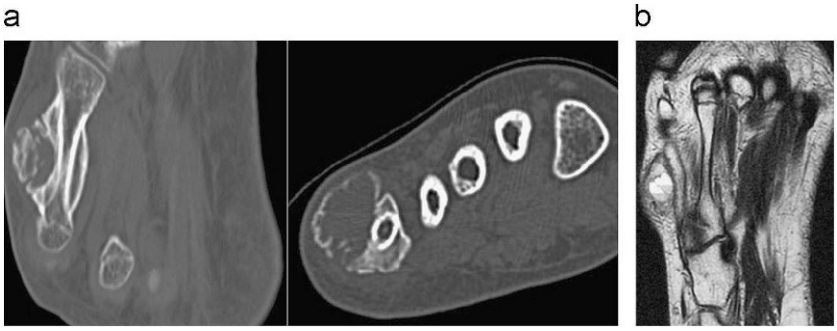


**Figure 2** a) Computerized tomographic slices; bony excrescence on the lateral edge of the fifth metatarsal. b) Magnetic resonance; eccentric exostosis on the lateral edge of the fifth metatarsal; no cortical or medullar lesion, with fluid-fluid levels in the interior and uptake of contrast.

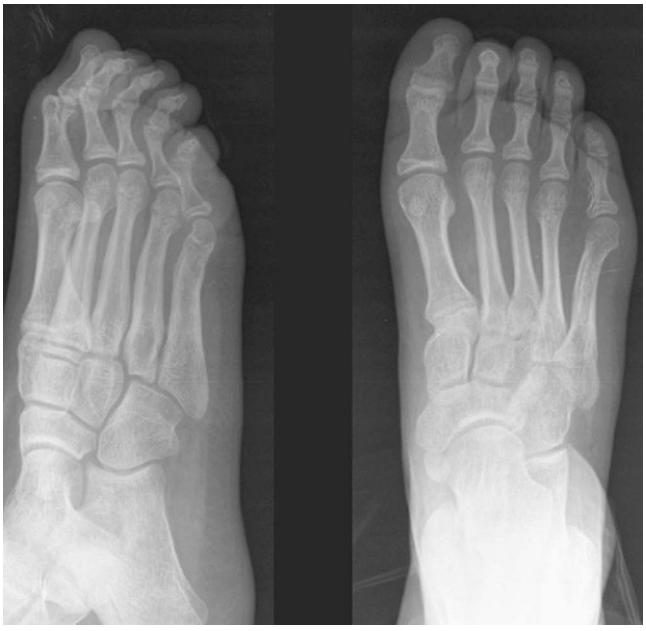

**Figure 3** X-rays, 6 months postoperative.

lobulated swelling was seen underneath the periosteum of the lateral cortex of the diaphysis of the fifth metatarsal (fig. 1) and with no lesion of the cortex or of the medullary space. We also observed separation of the periosteum from the medial cortex. In light of these findings, the decision was made to hospitalize the patient for further study and treatment.

Both a CAT scan and an MRI of the foot were order to complete the study. The CAT scan (fig. 2) revealed a large bony excrescence on the lateral edge of the diaphysis of the fifth metatarsal that had an irregular, albeit well-defined contour with areas filled with a soft tissue density and without any calcifications inside, the characteristics of which pointed toward a cortical swelling of external periosteal growth associated to an adjacent cortical reaction. The MRI (fig. 2) revealed the presence of an eccentric exostosis toward the lateral aspect of the fifth metatarsal without significant alteration in the nearby soft tissue, as well as the absence of any cortical lesion or medullar involvement with fluid-fluid levels inside the exostosis and uptake of the contrast, following gadolinium administration, without clear characteristics of aggressiveness. The differential diagnosis posed included parosteal osteochondromatous proliferation, periosteal chondroma, and superficial osteosarcoma. The bone trace denoted a hyperintense lesion on the fifth metatarsal of the foot without any peripheral lesions to indicate metastasis.

An excisional biopsy of the lesion was performed; the pathology report regarding the specimen indicated the gross appearance of the tumour to be indicative of an aneurysmal bone cyst and, under the microscope, to be mature laminar and trabecular bone tissue that presented a central cystic formation delimited by spindle cells, lacking mitosis or atypias, accompanied by giant multinucleated cells with red blood cells and an expansive growth, thereby confirming the diagnosis of aneurysmal bone cyst.

The clinical course following tumour resection was favourable (fig. 3) and no relapse of the tumour has been seen after 2 years since surgical treatment.

## Discussion

The aneurysmal bone cyst is a benign tumoural lesion that is generally intramedullary or metaphyseal. Only rarely does it present a superficial and sub-periosteal location. In the work by Capanna et al.[4] about their series of 121 cases, the sub-periosteal type represented a mere 7% of their cases; of the 238 cases at the Mayo Clinic, only 7% were superficial lesions,[5] and Maiya et al.[6] recorded 144 cases, of which only 23 were superficial and 11 of them, sub-periosteal.

Insofar as the most commonly affected bone, the long bones rank first followed by the vertebrae.[2,3,7-9] Consequently, although any bone in the skeleton can be affected, a location on the coccyx is rare, although aneurysmal bone cysts on the metatarsals,[10] the metacarpals,[11-13] the phalanges of the thumb,[14,15] the patella,[16] and tarsal bones[17] have been reported, but none was located sub-periosteally. Thompson[18] reported a sub-periosteal case located in the neck of the astragalus and Rios et al.[19] described another Campanacci type V located on the astragalus in a 14-year old child associated with a chondroblastoma.

The pathogenesis of this tumour is not clear. It has been suggested that traumatic injuries might be a causative factor in sub-periosteal locations.[20] Capanna et al.[4] found a history of trauma in 23%of their cases.

Campanacci et al.[3] developed a radiological classification system for aneurysmal bone cysts consisting of 5 types. The case reported here corresponds to a type IV; that is, it is a sub-periosteal form with superficial erosion of the underlying cortex, in which the elevation of the periosteum could only be seen by means of soft x-rays or angiography and, subsequently, the periosteum formed a layer of reactive bone that characteristically affected the diaphysis. This is the least common subtype. Among the 198 cases collected by Campanacci et al.,[3] only 9 were type IV. Woertler et al.[21] reported 6 cases of sub-periosteal forms, albeit all located on long bones.

The diagnosis is more complicated in sub-periosteal forms because of their atypical location, but their radiographic characteristics, similar to intramedullary forms, tend to point the diagnosis in the direction of aneurysmal bone cyst. Although MRI may be of use in establishing their diagnosis given the characteristic presence of septa and fluid levels, these findings are not pathognomonic and a pathology study is still needed to confirm the diagnosis. Histologically, sub-periosteal and intracortical forms are identical to intramedullary forms. They should be included in the radiographic differential diagnosis of other intracortical or sub-periosteal lesions as relatively uncommon forms of aneurysmal bone cysts. Thus, the differential diagnosis should be established including osteoid osteoma, haemangioma, osteosarcoma, and chondromyxoid fibroma.[22]

The treatment of choice is surgical, since spontaneous healing of the cyst is uncommon. Classically, standard treatment consisted of curettage followed by filling with bone graft; however, due to the high rate of relapse, adjuvant treatments tend to be administered, such as cementing, cryotherapy, and embolization. Cementing with polymethyl methacrylate followed by replacement with bone graft 4-6 months later appears to be more effective than just curettage and bone graft. Adjuvant cryotherapy also increases the cure rate and embolization can be used prior to surgery to selectively interrupt vascularization of the cyst, and thereby facilitate the subsequent surgery, particularly in areas having limited access, such as the spine or the pelvis. In the sub-periosteal subtypes (types IV and V), total or partial resection is indicated and both are aimed at ensuring the cyst is cured[3]. In our case, a total resection of the cyst was carried out with subsequent pathology study that confirmed the diagnosis.

One of the main problems besetting these lesions is their tendency to relapse; as a result, the use of high-speed drills has been indicated during the curettage of the wall of the cyst to reduce the chance of it recurring.[23]

In conclusion, aneurysmal bone cysts, although they are benign tumours, can present clinical and radiographic characteristics of aggressiveness that complicate their diagnosis. They should be suspected when facing well-delimited, expansive lesions that grow on the cortex or underneath the periosteum with radiographic characteristics resembling those of typical intramedullary aneurysmal bone cysts.

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