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Giant Aneurysmal Bone Cyst of the Right Femur about a Case

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Abstract

Aneurysmal bone cyst (ABC) is a rare bone tumor accounting for 1% - 4% of benign bone tumors. Its etiopathogenesis remains unknown. The main site is the metaphysis of the long bones. ABC occurs almost exclusively in young people, with a slight female predominance. We report an exceptional case of ABC of the femur with involvement of the right hip in a 16-year-old girl in the Medical Imaging Department of Pr Bocar Sidy Sall Hospital of Kati (Mali). We will discuss the clinical and radiological aspects of this pathology. **Observation:** Mrs N C., aged 16, was referred to the orthopaedic-traumatology department for an X-ray of the right thigh, indicated as having a large, disabling thigh. Clinical examination revealed a large thigh measuring 890 mm in circumference, compared with 300 mm on the contralateral side. Radiographically, the femur showed a blown appearance, with the cortex thinned and broken in places. Exceptionally, the lesion affected the entire femur. On CT scan, we found a voluminous fluid collection occupying the entire right femur, including the right femoral head and homolateral ischium. The cortex was interrupted in places, and the adjacent soft tissues were thinned or laminated, but not infiltrated. Histology revealed haemorrhagic and inflammatory elements compatible with the diagnosis of ABC. **Conclusion:** ABC is a tumor entity that is often difficult to diagnose. Medical imaging, topographical distribution and histology form an indissociable whole to establish a diagnosis of certainty.

Keywords

Aneurysmal Bone Cyst, Medical Imaging, Femur

1. Introduction

Aneurysmal bone cyst (ABC) was defined by the WHO in 1972 as a "benign, osteolytic, expansive lesion, occurring most commonly in the metaphysis of long bones, vertebrae, flat bones, consisting of multiple hematic lacunae separated by connective septa bearing osteoclasts and reactive osteogenesis" [1].

Once considered a variety of giant cell tumour, it was first described in 1942 by Jaffe and Lichtenstein [2].

This is a rare bone tumor, accounting for 1% - 4% of benign bone tumors. It is mainly found on the metaphysis of long bones, and occurs almost exclusively in young people, with a slight female predominance. Its etiopathogenesis remains unknown [3].

The symptomatology of ABC is polymorphic, dominated by swelling, pain and/or functional impotence in the event of pathological fracture.

Its radiological diagnosis is sometimes difficult, mimicking other benign or malignant cystic lesions, such as telangiectatic sarcoma, hence the importance of anatomopathology.

Although histologically benign, the prognosis of this lesion is nonetheless quite serious, due to its location, size and proximity to the conjugation cartilage, which poses therapeutic problems, and to the frequency of local recurrence [4].

ABC poses relatively difficult diagnostic and therapeutic problems, as the disease takes on a variety of faces in very different age groups and according to location, which explains the absence of a consensual attitude [5].

The aim of this study is to describe the radiological aspects of an exceptional case of ABC of the femur in a 16-year-old girl and to compare it with the review of the literature.

2. Case History

The case involved a 16-year-old girl with no specific medical or surgical history, admitted to the orthopaedic department with a progressive disabling swelling of the right thigh for 3 years. Questioning revealed moderate pain with no fever or change in general condition. Clinical examination revealed a large right thigh measuring 89 cm in circumference, compared with 30 cm on the contralateral side, with a firm consistency and no inflammatory signs (**Figure 1**). X-rays of the right thigh showed heterogeneous osteolysis, with a "blown" appearance of the femur and local cortical disruption. There was also a mass effect of the lesion, with displacement and thinning of the adjacent soft tissues (**Figure 2**). CT scans of the right hip and thigh without and with iodinated contrast injection revealed a voluminous cystic lesion of the entire right femur, including the femoral head and ischium. It also showed local interruption of the femoral cortical bone, and displacement of the adjacent soft tissues, which were thinned and laminated, but with no evidence of tumour infiltration (**Figure 3**). The patella and the head of the right fibula and tibia were normal.

The definitive diagnosis of ABC was made on anatomopathological examination of the biopsy specimens (**Figure 4**); the specimens examined contained red

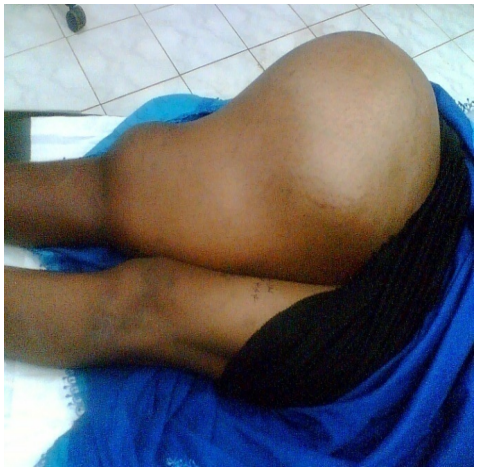

Figure 1. Photograph of the voluminous mass in our patient's right thigh.

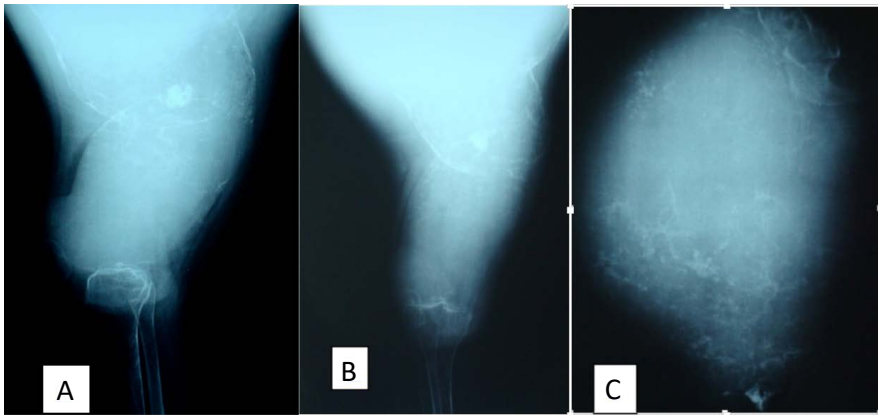

Figure 2. X-ray of the right thigh, side (A) and front (B and C), showing the significant heterogeneous osteolysis of the femur and the sequestrations giving the lesion a "cloudy appearance".

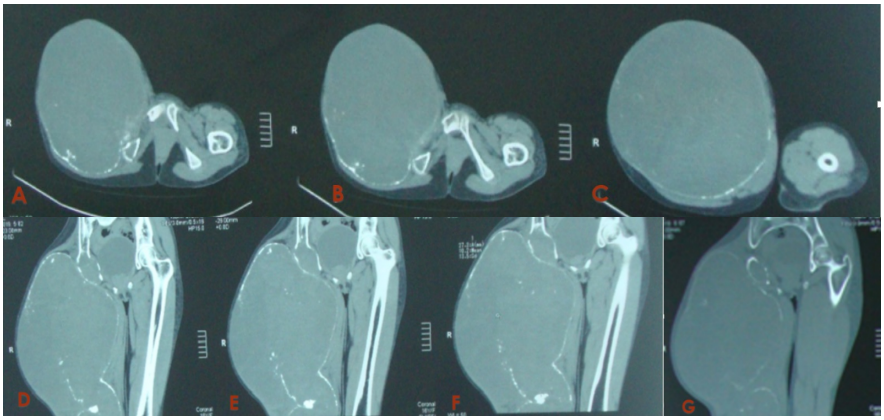

Figure 3. CT scan of the right femur and hip in axial sections (A, B and C) with coronal reconstruction (D, E, F and G) in parenchymal and bone windows (G) without IV injection of contrast medium, showing a voluminous fluid collection occupying the entire right femur and extending to the right femoral head and homolateral ischium. Note the local interruption of the cortical bone and the displacement of adjacent soft tissue, which was thinned or laminated but not infiltrated.

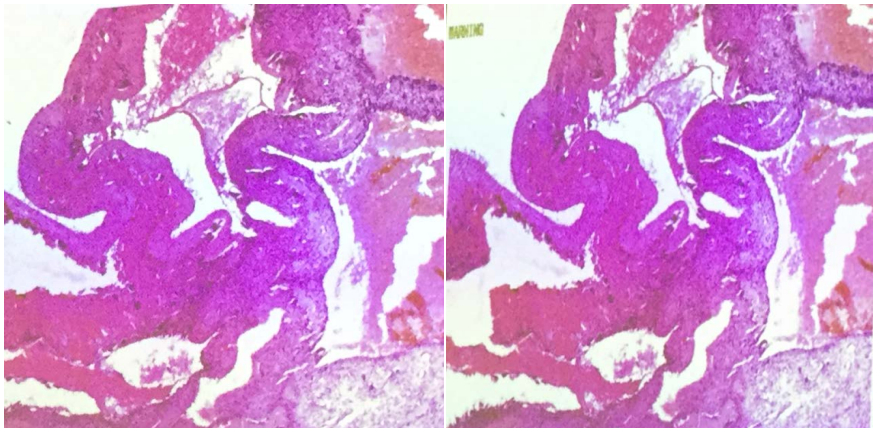

Figure 4. Hemorrhagic and inflammatory staging compatible with the diagnosis of aneurysmal cyst.

blood cells, histiocytes, fibroblasts, osteoclastic giant cells and an amorphous substance.

The patient was lost to follow-up after diagnosis. Sepsis secondary to cyst infection was fatal.

3. Discussion

Aneurysmal bone cyst (ABC) is a rare bone tumor representing less than 4% of benign bone tumors, first described by Jaffe and Lichenstein [2]. It is half as common as giant cell tumors, and four to seven times rarer than osteosarcoma [2].

There is a slight female predominance, and the cysts appear 3 times out of 4 before the age of 20, and 9 times out of 10 before the age of 30 [1].

Our patient (female) was 16 years old.

Age has a diagnostic value, as ABC is a frequent pathology in children and adolescents. Proximal or distal involvement of the long bones (femur and tibia) is reported in the literature [6] [7]. As our patient was seen late, the entire femur was affected, as well as the coxofemoral joint, but the knee joint was intact. The soufflure was responsible for mass displacement and thinning of the soft tissues.

The most frequent topography on long bones is metaphyseal and metaphysodiaphyseal, usually near the extremities [7].

We found involvement of the entire femur, with a "cloudy appearance" of bone sequestration.

Several ABC sites have been described in the literature, including the clavicle [8] and vertebrae [9] [10]. In a series published by Ariel M. *et al.* in 2004 (n = 238), the authors reported ABC of virtually the entire axial skeleton, including multifocal involvement [11].

The symptomatology of ABC is variable, dominated by swelling and pain [11] in cases of pathological fracture. Sometimes the lesion is discovered by chance [11] [12].

In our case, the clinical presentation was marked by significant swelling of the right thigh, associated with functional impotence. In the literature, pain is gen-

erally moderate, irregular, rarely nocturnal, and sometimes increased by exertion or fatigue. It usually precedes the discovery of the swelling, but is sometimes revelatory. [9] [11] [12] [13]. The delay between the onset of symptoms and diagnosis is less than 6 months in 72% of cases [14]. In our observation, the evolution of the pathology was 3 years, different from that of the study carried out by Campana R. *et al.* [14]. This difference could be explained by the delay in diagnosis in our case, linked to the traditional treatments carried out by the patient, which could explain the volume of the cyst.

On standard radiography, in the typical form [10] [13] as in our observation, the ABC presented as a large proximal epiphyseal-metaphyseal lytic lesion of the right femur. The affected femur was blown, with local cortical fracture. The lesion involved the homolateral coxofemoral joint and was responsible for a mass effect with displacement and thinning of the adjacent muscles. A CT scan of the femur and right hip without and with injection of contrast medium was performed to complement the radiographic work-up. It showed a voluminous lytic lesion with local interruption of the cortical bone of the coxofemoral joint and the proximal and the proximal third of the right femur. The lesion displaced the adjacent soft tissues, which were thinned and laminated but not infiltrated. CT scans allow a better assessment of the thinned cortical bone than on standard radiographs. The ABC presents a very clear boundary with the soft tissues, which are certainly compressed but not invaded [10], as observed in our case.

Magnetic resonance imaging was not performed in our patient, due to lack of availability. According to some authors, it would allow a better appreciation of the dimensions and relationships of the cyst, as well as the internal architecture of the lesion [6] [10] [15]. The ABC is characterized on MRI by a T2 hypersignal, sharp borders with a peripheral hyposignal border and numerous septal delineations [6] [10] [15].

Biopsy of the lesion yielded a mixed serosanguinolous fluid in our case. Pathological examination confirmed the diagnosis of ABC, showing red blood cells, histiocytes, fibroblasts, osteoclastic giant cells and an amorphous substance on biopsy samples. Histologically, the aneurysmal bone cysts are associated with a number of elements, including plurinucleated giant cells and hemosiderin deposits. Reactive osteogenesis occurs in immature septa, or more frequently in more mature trabeculae bordered by a more or less visible osteoblastic border [7].

On imaging, ABC prompts discussion of a number of differential diagnoses, including essential bone cyst [15], chondromyxoid bone fibroma [8] [15], giant cell tumour [8] [13] and, above all, telangiectatic osteosarcoma and clear-cell chondrosarcoma [6] [8] [11] [15].

Management of ABC varies, with some cysts healing spontaneously or being treated by puncture followed by injection of steroids, alcohol or Ethibloc® [16], with a risk of recurrence. Surgical resection of the lesion through healthy bone considerably reduces the risk of recurrence, and is the only truly curative treatment for ABC. The location of the cyst, its volume and its degree of aggressiveness guide the surgical strategy [6]. The more recent use of innovative treat-

ments such as calcitonin injections [17] appears promising. In our case, the patient was lost to follow-up after diagnosis. She was readmitted to the emergency department two months later with septicemia secondary to cyst infection, which had a fatal outcome.

4. Conclusion

Aneurysmal bone cysts are benign but aggressive tumors of children, adolescents and young adults. Its symptomatology is variable, and the diagnosis of pseudotumoral forms is relatively difficult. Imaging is essential for diagnosis, but only anatomopathology of biopsy specimens can confirm the diagnosis. The management of ABC requires a multidisciplinary approach in a specialized environment, in order to avoid infectious complications with potentially fatal outcomes, as in our case.

Conflicts of Interest

The authors declare that they have no conflict of interest.

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