International Journal of Medical Imaging
2013; 1(2): 19-22
Published online August 30, 2013 (http://www.sciencepublishinggroup.com/j/ijmi)
doi: 10.11648/j.ijmi.20130102.11

Unicameral bone cyst: a case report and literature review

Redouane Roukhsi[1, *], El Mehdi Atmane[1], Monsef Elabdi[2], Tahir Nebhani[3], Abdelilah Mouhcine[1], Abdelghani El Fikri[1], M'barek Mahfoudi[1]

[1]Department of Radiology, Military hospital Avicenne, Marrakech, Morocco
[2]Department of Orthopedics, HMIMV, Rabat, Morocco
[3]Emergency department, Military Training Hospital Med V, Rabat, Morocco

Email address:

redouan9990@hotmail.com (R. Roukhsi), atmanemehdi@yahoo.fr (E. M. Atmane), elabdimonsef@gmail.com (M. Elabdi), propre_9@hotmail.com (T Nebhani), abdelilahmouhsine@yahoo.fr (A. Mouhsine) , abdelghani@123.live.fr (A. E. Elfikri), mahfoudi@gmail.com (M. Mahfoudi)

To cite this article:

Redouane Roukhsi, El Mehdi Atmane, Monsef Elabdi, Tahir Nebhani, Abdelilah Mouhcine, Abdelghani El Fikri, M'barek Mahfoudi. Unicameral Bone Cyst: A Case Report and Literature Review. *International Journal of Medical Imaging.* Vol. 1, No. 2, 2013, pp. 19-22. doi: 10.11648/j.ijmi.20130102.11

Abstract: Unicameral bone cysts (UBCs) are benign, osteolytic lesions which are often asymptomatic and are commonly seen in the proximal of humerus and femur. We report such a lesion in a eleven years old boy who presented to orthopedic clinic with a 10 months history of painful limping.Clinic, radiographic and pathologic findings and cyst aspiration evocated solitary bone cyst. Child was treated successfully with repeated intralesional steroid injection.

Keywords: Unicameral Bone Cyst, Osteolytic Lesions, Benign

1. Introduction

A UBC is a relatively common benign lucent bony lesion, which is seen in childhood and typically remains asymptomatic.

They usually found in children in the 1st and 2nd decades, and are more common in males (M/F = 2 – 3 / 1). These lesions are usually asymptomatic and found incidentally, although pain, selling and stiffness of the adjacent joint also occur. They are typically intramedullary and are most frequently found in the metaphysis of long bones, abutting the growth plate. Plain radiographs typically show a symmetric lesion with cortical thinning and expansion of the cortical boundaries. CT and MRI add little to the diagnosis, but are however helpful in eliminating other entities that can potentially mimic a simple bone cyst. Once diagnosed, unicameral bone cysts continue to be a treatment dilemma. Traditional methods, such as prednisolone therapy, usually involve multiple anesthetics and injections and are associated with high recurrence rates. Major surgical procedures, such as wide exposure, curettage, and bone grafting, may be somewhat more effective, but still carry with them significant morbidity and recurrence rates.

2. Case Report

We report such a lesion in a eleven years old boy who presented to our clinic with a 10 months history of painful limping. There was no history of any specific infection or trauma. She did not have any similar complain previously. The pain was constant throughout the day but was alleviated by analgesics. There was a moderate tenderness in the proximal of tibia in palpation without any warmth or change in color of the skin in the suspicious area. The child was not febrile and there was no sign or symptom of systemic illness. Hematological investigations (include leukocyte count, cell profile, erythrocyte sedimentation rate, calcium, phosphorus and alkaline phosphatase) were normal. The plain radiography of the right knee demonstrated a small wellcircumscribed eccentric lesion in the proximal medial metaphyseal region of the tibia without disruption of the cortex (Figures 1 a, b).

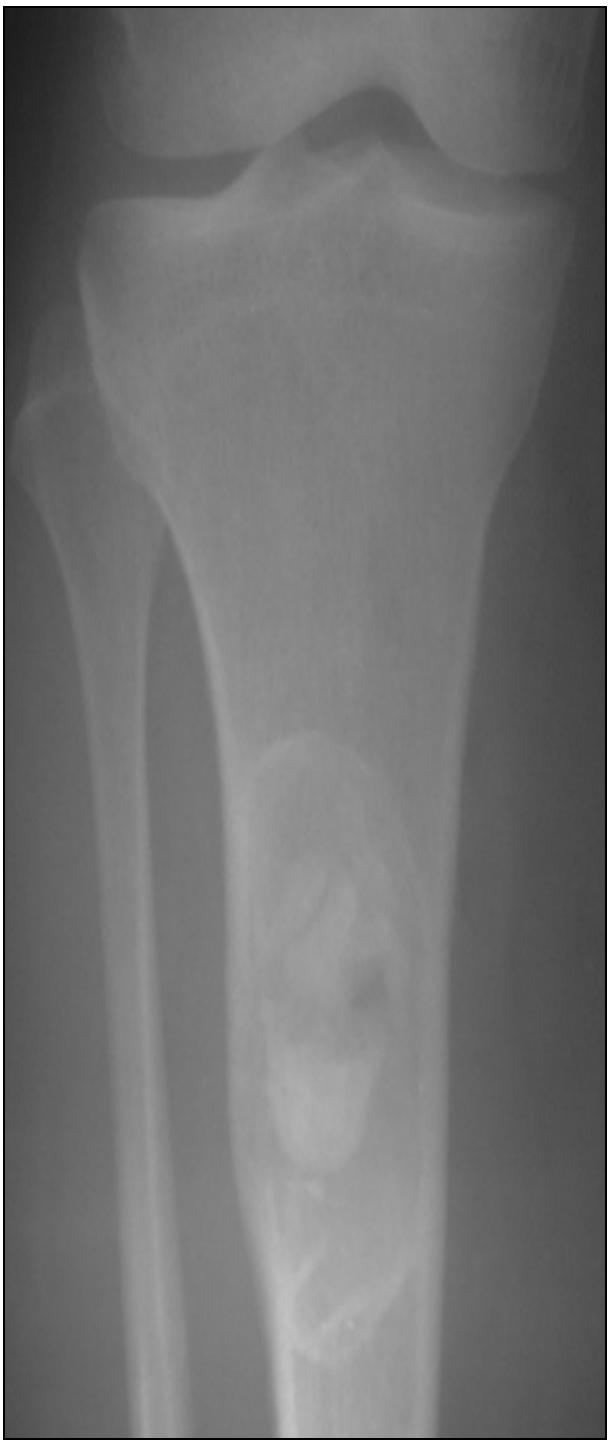

Figure 1 a: *X-ray of the left tibia. Front= geographical osteolysis of the upper end of the left tibia.*

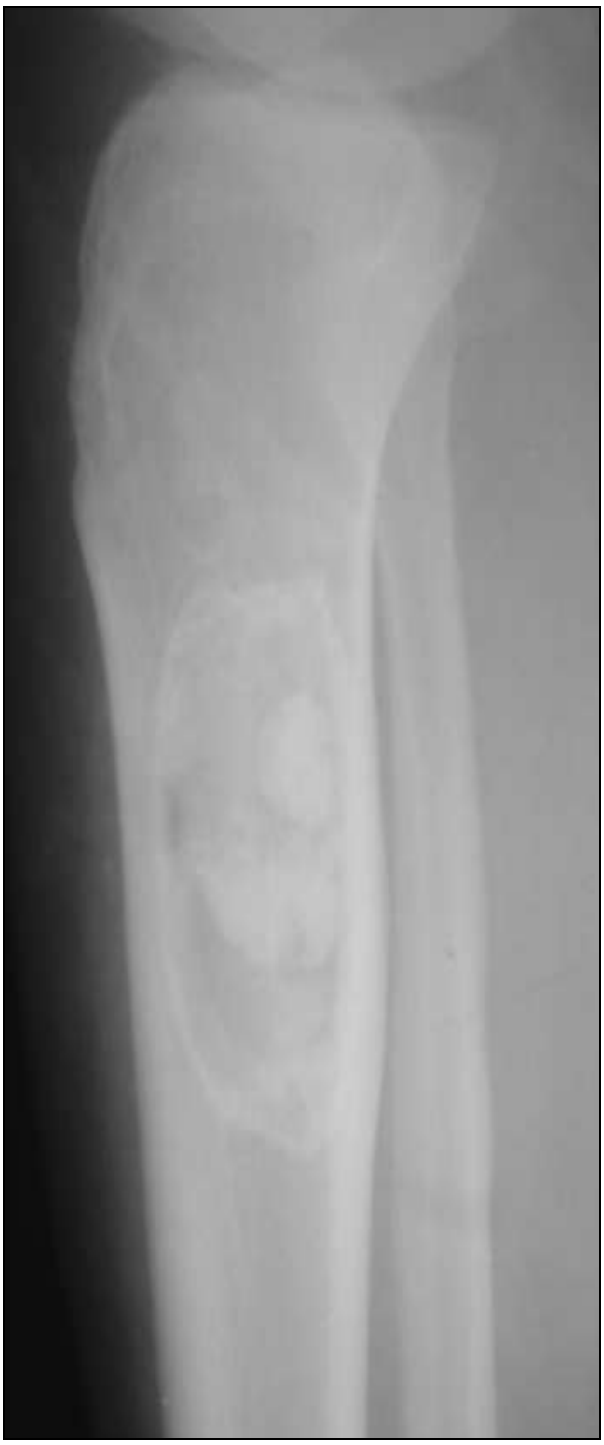

Figure 1 b: *X-ray of the left tibia of Profile= geographical osteolysis of the upper end of the left tibia.*

Computerized tomography Scan (Figure 2) shows that osteolysis is intra medullary cystic walls with recasts, although limited by the marginal sclerosis, without periosteal reaction and without invasion of the adjacent soft tissues.

Figure 2: *CT shows that osteolysis is intra medullary cystic walls with recasts, although limited by the marginal sclerosis, without periosteal reaction and without invasion of the adjacent soft tissues.*

Clinic, radiographic, pathologic findings, and cyst aspiration evocated: solitary bone cyst. Child was treated successfully with repeated intralesional steroid injection.

3. Discussion

UBCs are benign, fluid-filled lesions that occur mostly in long bones of male children aged 5 to 15 years. 94% of UBCs occur in the proximal humerus and proximal femur, with the proximal humerus being affected 2-3 times more frequently than the proximal femur [1]. The remaining 6% occur in other bones including the calcaneus (2%), ilium (2%), talus, tibia, fibula, metatarsals, ischium, pubic rami, sacrum, vertebral bodies, forearm, and craniofacial bones [2].

Komiya and Inoue have the only longitudinal study (with serial radiographs over 6 y) that documents the development of a UBC over time3. Initially, a small erosive lesion of the endosteal humeral metaphysis appeared, and over time, the lesion progressively enlarged into a typical UBC [3]. The lesion analyzed by these authors was somewhat unusual in that it was located in the distal humerus.

These cysts are sometimes classified as either "active" or "latent". An active cyst is adjacent to the growth plate and tends to enlarge. A latent cyst is one that is more apt to heal with treatment because the growth plate has migrated away from the cyst.

The radiological features on plain x-rays include a centrally located, expansile lesion of the metaphysic. Cortical thinning without disruption is seen. The lesion appears as a well defined osteolytic area with a thin sclerotic margin. It fills and perhaps slightly expands the juxta-epiphyseal metaphysis of the bone. The lesion is relatively symmetrical with respect to the midline axis of the bone. The lesion is not eccentric and does not break out through the cortex or form any extra osseous mass. There is no periosteal reaction visible unless there has been a previous fracture [4].

Computerized tomography Scan can be useful to evaluate the extent of the cyst, especially if pelvic bones are affected, and will help to differentiate lipomas from fluid-filled cysts. Typically Magnetic Resonance Imaging will demonstrate lowsignal intensity on T1 weighted images and high-signal intensity on T2weighted images in the typical simple bone cyst [5].

The most characteristic histopathologic finding is the thin membranous lining of the cyst, composed primarily of flattened to plump epithelium-like cells; the lining may also possess osteoclast-type giant cells, cholesterol cells and fat cells. Hemosiderin, fibrin, calcification, and reactive bone may be seen in focal areas of the cyst [6].

The major differential diagnoses include aneurysmal bone cysts, monostotic fibrous dysplasia, and atypical eosinophilic granuloma [7]. All of these lesions may be radiolucent. However, features typically associated with these lesions usually help differentiate them from simple bone cysts.

Unicameral bone cysts continue to be a treatment dilemma. Traditional methods, such as prednisolone therapy, usually involve multiple anesthetics and injections and are associated with high recurrence rates. Major surgical procedures, such as wide exposure, curettage, and bone grafting, may be somewhat more effective, but still carry with them significant morbidity and recurrence rates. Newer techniques involving percutaneous grafting with allograft or bone substitutes or a combination of the two are promising in light of their low complication rate and lower reoperation rate.

There is a variety of treatment modalities for unicameral bone cysts, with variable outcomes reported in the literature. Although good initial outcomes have been reported, the success rate has often changed with longer-term follow-up. An asymptomatic lesion with satisfactory maintenance of cortical thickness may require only observation. A lesion with precarious cortical thinning may demand surgical intervention. Some authors have suggested the use of a cyst index aimed at predicting the future risk of a pathologic fracture. Andre Kaelin and Dean Mac Ewen discussed this concept and defined their cyst index as the area of the UBC measured via its widest dimensions divided by the diameter of the diaphysis of the same bone [8]. However, treatment should be strongly considered for lesions that have resulted

in a fracture or marked weakening of bone. Some evidence exists that spontaneous healing of a UBC may occur following fracture. Such healing occurs in only a minority of cases. Growth disturbance secondary to a UBC is also a concern [9].

Prognosis for a unicameral bone cyst is generally good. Most of these cysts do heal with proper treatment and if left alone, most heal spontaneously by the time the skeleton ceases to grow. Recurrence can, however, occur. Continuous follow-up care is essential for the successful treatment of this kind of bone cyst.

Complications are Injury to the growth plate (physis) may occur secondary to direct cyst expansion, pathologic fracture, or unintended mechanical disturbance during surgical intervention. Direct cyst expansion across the growth plate and into the epiphysis of the proximal humerus has been well documented by Gupta and Crawford via MRI.[10] Growth arrest has also been reported following treatment either by local injection of steroid or curettage and bone grating.[11] Growth disturbance leading to angular deformity or disturbed longitudinal growth has been estimated to possibly occur in approximately 14% of cases [12, 13]. Steroid injection has been a successful treatment, even in the setting of cyst extension into the epiphysis [14].

Conflict of Interests

Authors have no conflict of interests.

Authors' Contributions

All authors planned and conducted the study procedure and wrote. All authors read and approved the final draft of the manuscript.

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