MANAGEMENT OF BONE CYSTS OF THE HUMERUS IN CHILDREN

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- Bone cysts are a common benign tumor, located usually in the metaphysis of long bones.
- Humerus is common location.
- Diagnosis is made after fracture of the cyst, usually after minor trauma. Rarely there are obscure symptoms

- We present a series of 14 children with bone cysts affecting the humerus. Their age ranged from 3-12 years. There were 9 girls and 5 boys.
- In all our cases, cysts were located in the proximal metaphysis of the humerus.

DIAGNOSIS

- Fragility fracture in 12 children
- Unspecified complaint for the shoulder in 2

Radiological investigations

- x ray
- MRI
- CT scan
- NO scintigraphy

- Solitary cyst

classification

- Cysts were classified according to the Capanna classification
- Size, position, contact with growth plate

Skeletal Radiology

Juxtaepiphyseal Aneurysmal Bone Cyst

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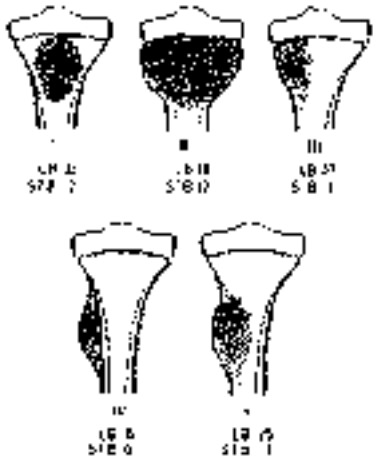
Aberrant bone cases of aneurysmal bone cyst arising in juxtaepiphyseal locations with cross extension of the adjacent growth plate are reported. In five of these patients an abnormality of growth, due to premature fusion of the affected growth plate, ultimately developed. Treatment of these lesions should attempt to avoid this complication, which appears to be more common than has been appreciated in the past. These nine cases represent 23% of 39 cases of aneurysmal bone cyst occurring at a long bone adjacent to a skeletally immature plate. The series was extrapolated from a total of 196 cases of aneurysmal bone cyst in the files of the Istituto Ortopedico Rizzoli, Bologna, Italy.

Key words: Bone tumours, benign – Aneurysmal bone cyst – Growth plate – Epiphyseal invasion



Aneurysmal bone cyst (ABC) is a benign bone lesion of unknown aetiology. We consider it to be reactive rather than neoplastic and believe the condition probably to have a haemodynamic origin. It occurs slightly more often in females, usually before the age of 20 years, and most commonly in long bones. Involvement of the epiphyseal plate is considered to be rare, with only three case reports in the literature [3, 4, 7]. Murray and Hudson illustrated an additional case in the second edition of their radiology text [8]. Our experience with juxtaepiphyseal lesions suggests that the incidence of epiphyseal plate involvement is higher than previously suspected and that growth disturbance may result from the abnormality.

Materials and Methods

The case records, radiographs, and histology of all patients with ABC treated during the years 1969 to 1992 in the Istituto

management

- Treatment for the cyst was performed after the healing of the fracture.
- All cysts were treated with an open procedure, with opening a bony window, removal of the membrane, thorough cleaning using burr,
- filling of the cyst with cortical and cancelous bone that was removed from the iliac crest at the beginning of the procedure.

histology

- Histology clarified that there were 11 aneurysmal cysts and 3 solitary cysts.

results

- Healing of the cyst was achieved in 11 children.
- There was filling of the cyst with bone trabeculae.
- MRI examination was the most accurate for confirming the healing.
- Healing was successful when there was filling of more than 90% of the diameter of the humerus and the remnant of cyst remained unchanged

Recurrence (not achieving healing)

- 3 patients <6 yrs
- 2 Cappanna type.
- There were 3 cases that the cyst remained with further expansion, with only partial healing. They were all aneurusmal cysts, type 2 Capanna, in contact with the growth plate. They were revised with the same surgical approach, adding a flexible intramedullary nail, for permanent drainage of the cyst. They had a complete healing in a 6 months period.

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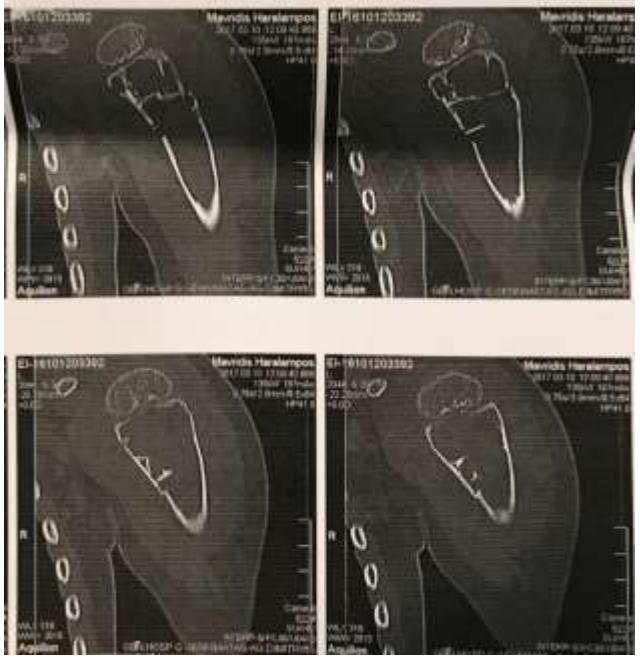

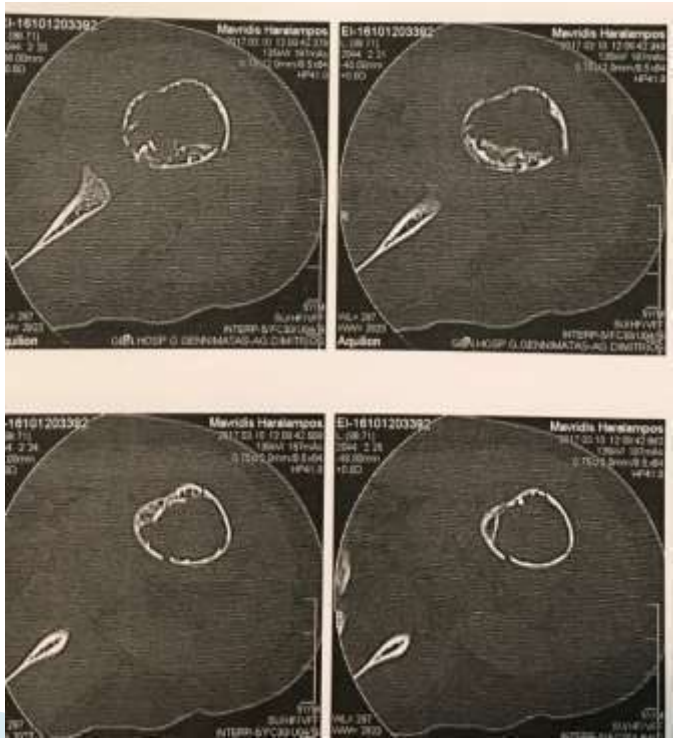

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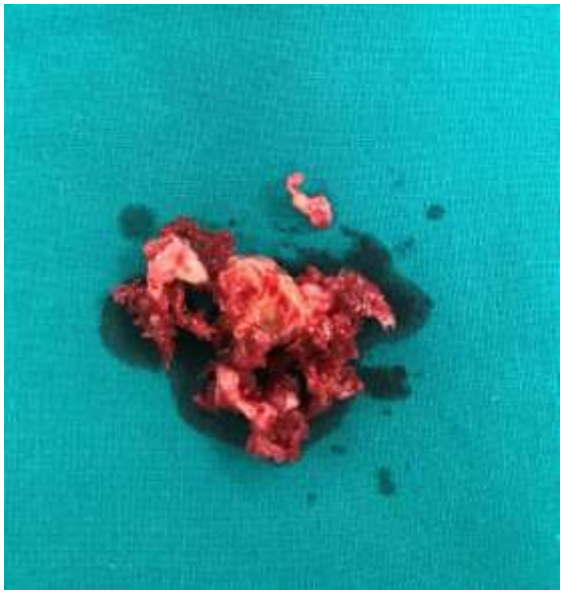

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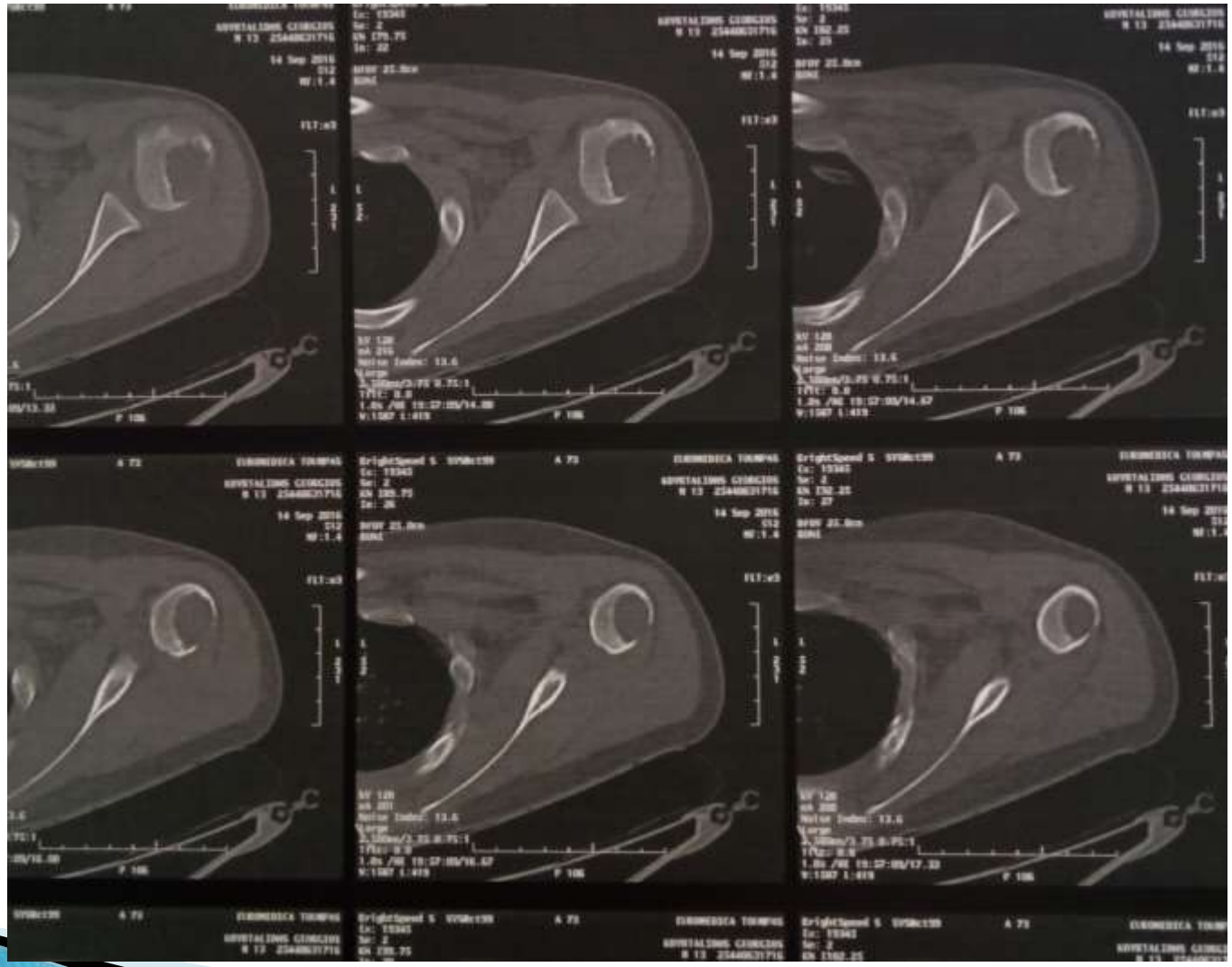

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- Treatment
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Orthop Rev (Pavia). 2015 Dec 28; 7(4): 6182.

Current Strategies for the Treatment of Aneurysmal Bone Cysts

Panagiotis Tsagozis and Otte Brosjö

- En block surgical excision
- Intralesional curretage with local adjuvants(phenol, cryotherapy, bone cement and high speed burr)
- Embolization
- Sclerotherapy
- Medical treatment biphosphonates **denosumab**

Acta Orthop Belg. 2016 Aug;82(3):474-483.

Aneurysmal bone cyst (ABC) : treatment options and proposal of a follow-up regime.

Hauschild O, Lüdemann M, Engelhardt M, Baumhoer D, Baumann T, Elger T, Südkamp NP, Herget GW.

- The aim of this study was to describe treatment -options and develop a follow-up regime for the -**aneurysmal bone cyst**, a neoplastic bone lesion with a **noticeable recurrence rate.** Reports of **28 patients** and a mean follow-up of 42.2 months treated multidisciplinary were analysed. Data were complemented by a literature review including 790 patients. **Patient age was from seven to 57 years**, in line with the literature (1-69 years). Lesions most frequently affect long bones, spine and pelvis ; pain is the most common symptom**. Treatment modalities vary, recurrences -occurred in 26.1% in our series**, rates ranged from 0-60% in the literature, with the vast majority within 2 years. With regard to the findings we propose, irrespective of treatment, **a follow-up** regime including clinical survey and imaging, **best with MRI**, at 3 months, 6 months and at half-yearly intervals within the first two and yearly within the third to fifth year

Bone cysts: unicameral and aneurysmal bone cyst.
Mascard E, Gomez-Brouchet A, Lambot K.
Orthop Traumatol Surg Res. 2015 Feb;101(1 Suppl):S119-27. doi: 10.1016/j.otsr.2014.06.031. Epub 2015 Jan 8. Review

- **UBCs resolve spontaneously between adolescence and adulthood**; the main concern is the risk of pathologic fracture. Treatment in non-threatening forms consists in intracystic injection of methylprednisolone. When there is a risk of fracture, especially of the femoral neck, surgery with curettage, filling with bone substitute or graft and osteosynthesis may be required.
- ABCs are potentially more aggressive, with a risk of bone destruction. Diagnosis must systematically be confirmed by biopsy, identifying soft-tissue parts, as telangiectatic sarcoma can mimic ABC. **Intra-lesional sclerotherapy with alcohol is an effective treatment**. In spinal ABC and in aggressive lesions with a risk of fracture, surgical treatment should be preferred, possibly after preoperative embolization. The risk of malignant transformation is very low, except in case of radiation therapy.

Orthop Traumatol Surg Res. 2016 Apr;102(2):213-6. doi: 10.1016/j.otsr.2015.11.016. Epub 2016 Feb 10.

Aneurysmal bone cyst: A 19-case series managed by percutaneous sclerotherapy.

Batisse F[1], Schmitt A[2], Vendeuvre T[3], Herbreteau D[4], Bonnard C[5]

- Between 2006 and 2014, 19 patients (7 females, 12 males, aged 3 to 17 years) with ABC treated by sclerotherapy were included. Six received Ethibloc(®), 9 Aetoxisclerol(®), 2 liquid absolute alcohol, and 2 absolute alcohol gel. Assessment used fluoroscopy in 17 cases and CT in 2. Ossification was assessed on MRI and pain on a visual analog scale and HEDEN score.
- **RESULTS:**
- Ossification was complete in 11 cases (84.6%) and partial in 2 (15.4%). Eighteen patients (94.7%) were pain-free at 3 months. There was no recurrence, at a minimum 2 years' follow-up. One case of skin necrosis was observed, associated with use of liquid absolute alcohol; there was 1 case of arterial reflux of Ethibloc(®) under CT control

Orthop Traumatol Surg Res. 2016 Apr;102(2):213-6. doi: 10.1016/j.otsr.2015.11.016. Epub 2016 Feb 10.

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Batisse F[1], Schmitt A[2], Vendeuvre T[3], Herbreteau D[4], Bonnard C[5]

- Sclerotherapy enables minimally invasive treatment of lesions that are deep, difficult of access to surgery and potentially damaging. Use of absolute alcohol gel and fluoroscopic control seems to improve the risk/benefit ratio, limiting complications by vascular extravasation of the sclerosing agent, thanks to real-time visualization of diffusion. Its clinical and radiological efficacy makes sclerotherapy and alternative primary treatment choice in ABC.

Scand J Surg. 2018 Mar;107(1):76-81
Treatment of Aneurysmal Bone Cysts with Bioactive Glass in Children.
Syvänen J[1], Nietosvaara Y[2], Kohonen I[3], Koskimies E[1], Haara M[2], Korhonen J[4], Pajulo O[1], Helenius I[1].

- Aneurysmal bone cysts represent about 1% of primary bone tumors. The standard treatment is curettage, followed by local adjuvant treatments and bone grafting. **The problem is the high recurrence rate**
- A total of **18 consecutive children (mean 11.3 years at surgery;** 10 males; 11 lower, 6 upper limb, 1 pelvis; 15 with primary surgery) **with histologically proven primary aneurysmal bone cysts operated with curettage and bioactive glass filling** between 2008 and 2013 were evaluated after a mean follow-up of 2.0 years (range, 0.7-5.1 years).
- **Two (11%) patients showed evidence of aneurysmal bone cyst recurrence** and both have been re-operated for recurrence. Bone remodeling was noted in all patients with remaining growth and no growth plate disturbances were recorded. **Two patients needed allogeneic blood transfusion.** No intraoperative or postoperative complications were recorded.

Acta Orthop Traumatol Turc. 2018 May;52(3):232-235. doi: 10.1016/j.aott.2017.04.005. Epub 2017 May 18.

Aneurysmal bone cyst healing response with intramedullary lengthening nail.

Acan AE[1], Basci O[2], Havitcioglu H

- We report the treatment process of a pediatric patient with deformity and shortening in the arm after a recurrent aggressive aneurysmal bone cyst (ABC) in the proximal humerus. The patient was treated with curettage of the lesion and lengthening on an intramedullary nail following an osteotomy just distal to the ABC. The period of lengthening was approximately 50 days. At the end of the treatment the lengthening goal was achieved without any neurovascular complication. There was a minimal loss in shoulder hyperabduction due to the deformity of the humeral head

Acta Orthop Traumatol Turc. 2018 May;52(3):232-235. doi: 10.1016/j.aott.2017.04.005. Epub 2017 May 18.

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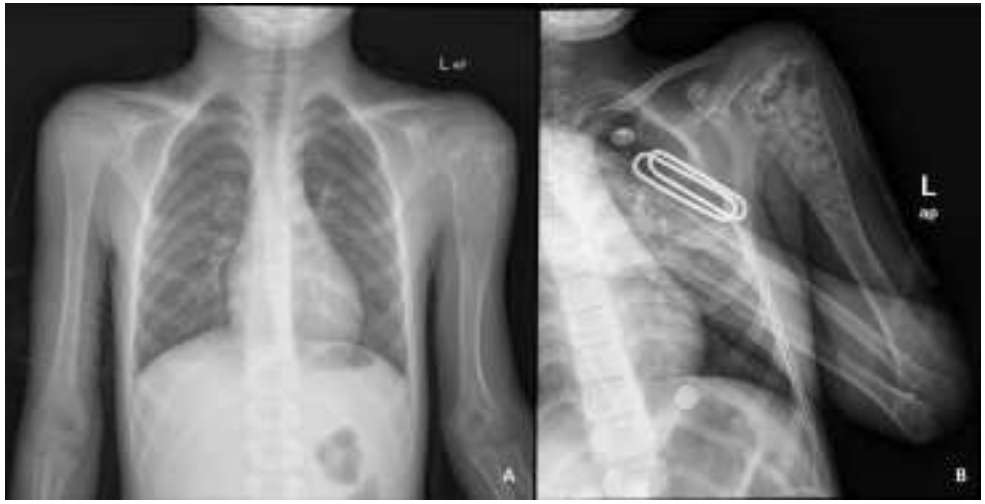

Clin Orthop Relat Res. 2013 Aug; 471(8): 2675–2683.

Percutaneous Doxycycline Treatment of Aneurysmal Bone Cysts With Low Recurrence Rate: A Preliminary Report

William E. Shiels, II, DO and Joel L. Mayerson, MD

- Doxycycline is an antibiotic that heals microcystic and macrocystic lymphatic malformations that are associated with elevated VEGF and MMPs. In bone and soft tissue malignancy cell cultures, doxycycline demonstrates antitumoral properties with inhibition of MMP and angiogenesis ,inhibition of osteoclastic function, and induction of osteoclastic apoptosis .
- Doxycycline enhances osteoblastic healing of bone defects in dogs
- We retrospectively reviewed 20 patients with 21 ABC sites who underwent percutaneous treatment with doxycycline (off-label use) delivered in a protein foam delivery system from 2006 to 2010
- Twenty of 20 patients demonstrated reduction in lytic destruction and bony healing. All patients demonstrated cortical thickening. One patient demonstrated recurrent minimal lytic destruction after 20 months of observation.
- In this series, patients undergoing percutaneous doxycycline treatment of ABCs demonstrated a healing response and a recurrence rate of 5% at more than 24 months.

ΒΙΒΛΙΟΓΡΑΦΙΑ

✓ Juxtaphyseal aneurysmal bone cysts
Rizzo M, Dellaero DT, Harrelson JM, Scully SP.
Clin Orthop Relat Res. 1999 Jul;(364):205-12.

✓ Aneurysmal Bone Cysts Recur at Juxtaphyseal Locations in Skeletally Immature Patients
BPatrick P. Lin, MD, Christopher Brown, S, A. Kevin Raymond, MD, Michael T. Deavers, MD, and Alan W. Yasko, MD
Clin Orthop Relat Res. 2008 March; 466(3): 722–728.

✓ Juxtaepiphyseal aneurysmal bone cyst.
Capanna R, Springfield DS, Biagini R, Ruggieri P, Giunti A.
Skeletal Radiol. 1985;13(1):21-5.

✓ Aneurysmal bone cysts: do simple treatments work?
Reddy KI, Sinnaeve F, Gaston CL, Grimer RJ, Carter SR.
Clin Orthop Relat Res. 2014 Jun;472(6):1901-10.

Clin Orthop Relat Res. 2014 Jun;472(6):1901-10. doi: 10.1007/s11999-014-3513-1. Epub 2014 Feb 15.
Aneurysmal bone cysts: do simple treatments work?
Reddy KI[1], Sinnaeve F, Gaston CL, Grimer RJ, Carter SR

- we introduced a novel biopsy technique we call **"curopsy,"** which is a percutaneous limited curettage at the time of biopsy, obtaining the lining membrane from various quadrants of the cyst leading to consolidation (curopsy = biopsy with intention to cure).
- Of the 102 patients who had curopsy and observation, 83 (81%) required no additional treatment and the lesion resolved. Of the 88 patients who underwent curettage (with or without adjuvant therapy) after core needle biopsy, the success rate was 90% (79 of 88). Local recurrences in both groups (curopsy or curettage) were treated successfully with additional curettage in all but one case.

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✓ Treatment of aneurysmal bone cysts with percutaneous sclerotherapy using polidocanol. A REVIEW OF 72 CASES WITH LONG-TERM FOLLOW-UP
S. Rastogi, M. K. Varshney, V.Trikha, S. A. Khan, B. Choudhury, R. Safaya
J Bone Joint Surg [Br] 2006;88-B:1212-16

✓ Primary Aneurysmal Bone Cyst of the Epiphysis
Gilbert Chan, Alexandre Arkader, Raymond Kleposki, and John P. Dormans
Clin Orthop Relat Res. 2010 April; 468(4): 1168–1172.

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Distal femoral aneurysmal bone cyst, crossing the open growth plate

Nikolaos A. LALIOTIS, Chrysanthos K. CHRYSANTHOU, Iordanis Petrakis, Panagiotis Konstantinidis

GLOBAL JOURNAL FOR RESEARCH ANALYSIS for 15th November, 2017 issue.

Original Research Paper | Medicine

DISTAL FEMORAL ANEURYSMAL BONE CYST, CROSSING THE OPEN GROWTH PLATE AND EXTENDING INTO THE EPIPHYSIS.

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ABSTRACT We report the case of an aggressive aneurysmal bone cyst, in a 7 year-old girl, crossing the middle of the growth plate and extending into the epiphysis. Plain radiographs, bone scan and MRI were the imaging methods that were used to reach the diagnosis and to rule out other aggressive bone lesions that invade into the epiphysis. The aneurysmal bone cyst was operatively treated with curettage, while preserving both the periosteum and the growth plate. Upon her latest follow-up visit, 5 years postoperatively, the radiographic and clinical examination of the patient yielded satisfactory results, without signs of recurrence and severe growth disturbances.

KEYWORDS Aneurysmal bone cyst, Benign bone tumor, Children, Growth plate

Introduction

Aneurysmal bone cyst (ABC) is characterized as a benign and expansive osteolytic lesion, active and aggressive, that usually originates at the metaphyseal - diaphyseal area of the long bones [1]. Its incidence is 0, 14/100,000 of the population per year and specifically ranges between 70% - 80% in the second decade of life [2]. An ABC may appear as a primary condition or as a secondary response to other lesions, such as giant cell tumors, chondroblastomas, chondromyxoid fibromas, chondrosarcomas, telangiectatic osteosarcomas or metastatic disease [3].

Localization of the lesion predominantly involves the metaphysis of a long bone, with asymetrical involvement [4]. Juxtaepiphyseal ABC has been well described, as an aggressive lesion that affects the growth [5]. The invasion of an ABC through the growth plate is extremely rare, with only few cases being reported, affecting the fibula, upper and lower tibia and distal ulna and metatarsals [6-12]. The epiphysis is an infrequent area for an ABC to be located entirely and as a primary condition, with only three cases being reported in the literature [13-15].

We report a 7 year-old girl with a distal femoral aneurysmal bone cyst crossing the growth plate and extending into the epiphysis. We present the clinical and radiological evaluation of the patient. She was treated with curettage and careful preservation of the periosteum and the growth plate. After 5 years of follow-up, she remains asymptomatic and the ABC is inactive.

Case Report

An otherwise fit 7-year-old girl presented to the outpatient department of our clinic complaining of pain and discomfort of her left knee joint. Her clinical manifestations were increased diameter of the distal femur, antalgic abnormal gait, as a mild limp. On physical examination, the distal femur was painful on palpation, however full knee range of motion could be achieved. On precise evaluation, she was expressing discomfort when performing solely weight standing on the affected side. Despite the swelling, the skin remained normal and there were no obvious signs of a neurovascular compromise in the left lower extremity.

Plain radiographs revealed the existence of a metaphyseal eccentric, radiolucent, expansive lesion, extending through the adjacent physis into the distal femoral epiphysis (Fig. 1), without evidence of a pathologic fracture. There was a clear line of demarcation of the lesion, in the distal femoral epiphysis. This lytic region with "bubbly appearance" indicates bony septas inside the lesion, whereas marked cortical thinning can be noticed. No periosteal reaction was observed. The growth plate appeared intact, apart from the central whyle, well formed in both AP and lateral projections.

Figure 1: Initial plain left knee joint radiograph of a 7-year-old female with an expansile, radiolucent eccentric lesion located in the metaphyseal and epiphyseal region of the left distal femoral bone, anteroposterior (a) and lateral (b) projection, note that this radiolucent lesion crosses the growth plate and extends into the distal femoral epiphysis (black arrow)

The patient underwent an MRI scan (Fig. 2a1, 2a2). The MRI scan clarified the extent of the distal epiphyseal involvement and showed double density fluid - fluid level, distinguishing blood from serum and apparent bony septation. Remarkable feature was the central lesion of the growth plate through which the lesion extended into the epiphysis, with intact articular cartilage. These imaging findings were consistent with an aneurysmal bone cyst. The bone scan indicated increased uptake in the perimeter of the lytic area, with decreased uptake in its center, which is suggestive of an ABC. Blood tests revealed normal values for ESR, CRP, alkaline phosphatase.

CONCLUSION

- ANEURYSMAL BONE CYSTS are benign BUT with HIGH RECURRENCE RATE
- Young age <5 and contact with GP are bad prognostic factors
- We propose the use of intramedullary flexible nail as part of the initial surgical clearance of an aneurusmal bone cyst that is in contact with the growth plate or in extensive cysts.
- Adequate follow up is important to diagnose recurrence of an aneurusmal bone cyst.