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Epiphyseal extension of a unicameral bone cyst

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Abstract Epiphyseal extension of a unicameral bone cyst is rare. We report a case of a 13-year-old boy with three pathological fractures through a unicameral bone cyst with epiphyseal involvement in the proximal humerus. These lesions initially tends to expand the humeral epiphysis laterally and progress medially. They also commonly cause a slip of the epiphysis in a medial direction. They also have a greater association with growth retardation and lesser degree of recurrence than their metaphyseal counterpart.

Key words Unicameral bone cyst · Epiphyseal extension

Case report

A 13-year-old athletic boy sustained three pathological fractures through a cystic lesion in the left proximal humerus over a period of 1.5 years. He was treated symptomatically for his fractures over this time period. The patient was subsequently transferred to our institution for diagnostic biopsy and treatment of his humeral lesion. Physical examination revealed visible left deltoid atrophy with a palpable deformity over the left proximal humerus and sensory and motor impairment of the left axillary nerve. Radiographically the plain films showed a large expansile lytic lesion extending from the metaphysis to the epiphysis with evidence of old and new callous formation. There was expansion of the lat-

eral epiphysis extending medially with a slip of the epiphysis in the medial direction (Fig. 1). The physis was irregular in its inner two-thirds. Computed tomography revealed a 7-cm, expansile lytic lesion extending from the diaphyseal region to the epiphysis (Fig. 2). The cortex was discontinuous, with extreme thinning at various levels. Periosteal reaction consistent with multiple sites of healing fractures was noted as well. Magnetic resonance imaging at this time demonstrated internal septations and irregularity of the lesion without fluid-fluid levels; sagittal STIR images demonstrated penetration of the lesion through the physis (Fig. 3). The differential diagnosis included aneurysmal bone cyst, giant cell tumor, fibrous dysplasia, and unicameral

bone cyst with epiphyseal extension.

At surgery, most of the lesion was cystic containing a minimal amount of blood. An open biopsy was performed with curettage and subsequent instillation of allograft demineralized bone graft with methylprednisolone.

Histologically the sections revealed fibromembranous tissue with acellular eosinophilic material and giant cells (Fig. 4). In addition, scattered foci of calcification and hyaline cartilage from the growth plate were noted. These microscopic findings are diagnostic of a unicameral bone cyst with epiphyseal involvement.

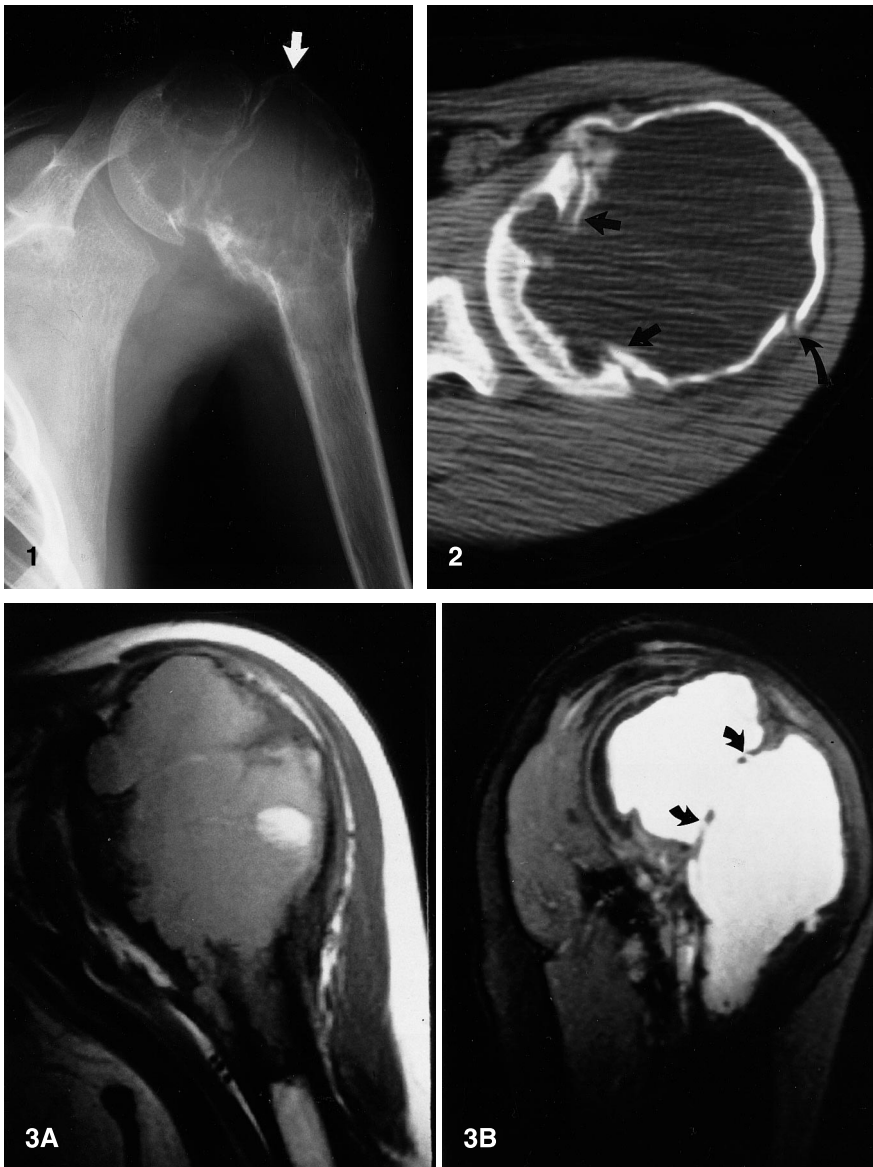


Fig. 1 Plain radiograph of the left shoulder demonstrates a lytic, expansile lesion of the proximal humeral metaphysis, extending through the physis into the epiphysis. Note the deformity and callous formation in the medial aspect of the humerus, secondary to a healing pathological fracture. A linear lucency is seen extending vertically through the lytic lesion, representing an acute fracture. Observe the expansion of the lateral aspect of the epiphysis (*arrow*) and the medial slippage

Fig. 2 Axial CT image through the proximal humerus demonstrates the lytic lesion with low attenuation coefficient, with the range of fluid. Note the transphyseal extension of the lesion (*arrows*). Note also the fracture of the posterior wall of the lesion (*curved arrow*)

Fig. 3 **A** Coronal T1-weighted image (TR500, TE20) and **B** sagittal STIR image (TR1500, TE19, TI150) demonstrate the intramedullary extension of the lesion and its cystic nature. Observe the “neck” of the lesion as it crosses the physis (*arrows* in **B**)

Discussion

Unicameral bone cysts are benign metaphyseal lytic lesions that originate in the long bones of skeletally immature persons. These cysts normally originate in the metaphysis immediately adjacent to the growth

plate and may, with continued growth away from the epiphysis, involve the diaphysis. These lesions are three times more common in males [1]. Unicameral bone cysts are located most commonly in the humerus followed by the femur (55% and 26%, respectively, of reported lesions) [2]. Unicameral bone cysts are rarely found in adults, and when found are commonly located in unusual locations such as the ileum, radius, or rib [3]. There have been several hypotheses as to the etiology of these lesions, but a definite cause has not been well established [4–9]. Despite the classic location for these lesions being metaphyseal, there have been infrequent reported cases of these lesions involving the epiphysis [2, 3, 10–16].

The first reported case of epiphyseal involvement of a unicameral bone cyst was documented by Jaffe and Lichtenstein in 1942 [14]. Since this time, there have been several reports of unicameral bone cysts with epiphyseal involvement [2, 3, 10–16]. The two largest and most complete reports were by Malawer and Markle [15] and Capanna et al. [10]. Malawer and Markle reported one case, reviewed the literature, analyzing seven previously reported cases, and commented on clinical behavior and appropriate management of these lesions. They concluded that unicameral bone cysts with epiphyseal extension are clinically distinct entities presenting in an older age group after closure of the growth plate. They also surmised that these lesions were less aggressive than conventional metaphyseal unicameral bone cysts, with a lower rate of recurrence and morbidity following curettage [15].

Capanna et al. reviewed 607 cases of unicameral bone cysts and found that 12 had epiphyseal or apophyseal extension across an open growth plate. Ten of these cases featured lesions involving the humerus. The involvement of the epiphysis by the unicameral bone cyst expanded more towards the lateral portion of the hu-

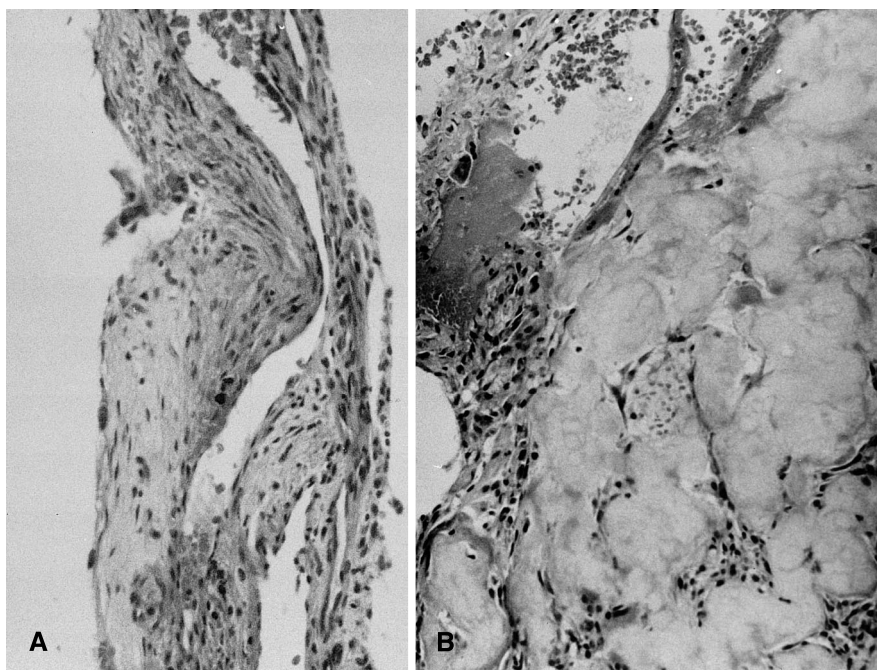


Fig. 4 Photomicrograph showing fibromembranous tissue (A) with acellular eosinophilic material (B)

meral head. Only in those cases where more than one-third of the head was cystic did the lesions involve the medial portion of the humeral head. Capanna et al. also reported that slip of the epiphysis relative to the expanding metaphysis commonly occurred, and in every case this took place in a medial direction. Another finding described in all the reviewed cases was the penetration of the physis in the middle causing a central bulging [10]. Our case showed similar features, with an expansion of the lateral aspect of the epiphysis extending medially and a slip of the epiphysis in a medial direction. MRI provided the most clear demonstration of direct communication between the metaphyseal and epiphyseal components of the lesion.

Growth retardation or shortening as a result of a unicameral bone cyst has been reported to be a relatively infrequent occurrence, with only scattered cases reported in the literature, the majority of which are

thought to be related to surgical curettage of the epiphysis [2, 3, 9, 12, 13, 17, 18]. This finding appeared to be more common in patients with epiphyseal involvement of unicameral bone cysts. Capanna et al. reported that four in ten patients with epiphyseal involvement of unicameral bone cysts in the humerus had 2.5 cm or more of shortening [10]. Although shortening can be associated with surgical damage to the physis, none of these four cases was treated surgically. Malawer and Markle commented on one case of shortening in the presence of a humeral epiphyseal unicameral bone cyst, which was thought to be a result of a previous fracture [15]. Our patient, who had three previous fractures and surgical curettage of his epiphyseal lesion, had no evidence of shortening on physical examination.

Interestingly, it is unclear whether unicameral bone cysts with epiphyseal extension are more likely to recur secondary to their close relationship to the epiphysis. Classically, unicameral bone cysts have been categorized as either active or latent based on several characteristics [14, 19, 20]. First, cysts are considered

active when they are located within 1 cm of the epiphyseal plate as well as having an elevated intracystic pressure significantly greater than standard venous pressure. Also, with injection of dye into an active cyst, the radiopaque material is rapidly cleared into the venous circulation. This classification has profound clinical implications, with active cysts being much more likely to recur than latent cysts.

Campanacci et al. examined outcome after treatment of unicameral bone cysts. They specifically looked at whether cysts abutting the physis had a greater tendency to recur. They found that out of 141 cases, steroid injection gave complete healing in 50% of cases, incomplete healing in 25% of cases, recurrence in 15%, and no response in 10%. Specifically in lesions abutting the growth plate treated by steroid injection (70 cases), results showed 41% displayed total healing, 26% incomplete healing, 20% recurrence, and 7% showed no response. Thus they showed that lesions abutting the growth plate were more resistant to treatment [1].

Capanna et al., using similar criteria for results, showed that 6 of 12 patients with epiphyseal or apophyseal involvement of a unicameral bone cyst treated with curettage and/or steroid injection showed complete healing, while the remaining 6 showed incomplete healing [10]. Malawer and Markle reported that five cases showed complete healing and the remaining two cases showed incomplete healing. A problem with the data in this report is that four of their seven cases had closed epiphyses, making recurrence less likely [15]. After 6 months of follow-up, our patient showed residual lucencies of the lateral aspect of the epiphysis. This was present in the radiographs obtained a few days after surgery, and therefore indicated residual cyst rather than recurrence.

In summary, epiphyseal extension of a unicameral bone cyst is an uncommon entity. Epiphyseal involve-

ment of a unicameral bone cyst occurs most commonly in the humerus followed by the femur. The cysts tend to expand the epiphysis of the humeral head beginning laterally and progressing medially. Unicameral bone cysts with epiphyseal involvement also have a propensity to penetrate the physis centrally and cause a slip of the humeral epiphysis in a medial direction. Although not present in our case, these epiphyseal lesions seem to have a greater association with growth retardation than their metaphyseal counterparts. Whether this risk is secondary to fracture damage to the physis or damage from the cyst itself is not clear. Also, from the limited available data, unicameral bone cysts with epiphyseal involvement show similar results to conventional unicameral bone cysts for complete healing; however, there was no evidence of recurrence in any case with epiphyseal extension. These limited results illustrate that unicameral bone cysts, with a higher incidence of shortening and a lower incidence of recurrence.

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