Active unicameral bone cysts in the upper limb are at greater risk of fracture

Inn Kuang Tey, Arjandas Mahadev, Kevin Boon Leong Lim, Eng Hin Lee, Saminathan Suresh Nathan
Department of Orthopaedic Surgery, KK Women and Children’s Hospital, Singapore

ABSTRACT

Purpose. To elucidate the natural history of unicameral bone cyst (UBC) and risk factors for pathological fracture.

Methods. 14 males and 8 females (mean age, 9 years) diagnosed with UBC were reviewed. Cyst location, symptoms, and whether there was any fracture or surgery were recorded. Cyst parameters were measured on radiographs, and included (1) the cyst index, (2) the ratio of the widest cyst diameter to the growth plate diameter, and (3) the adjusted distance of the cyst border from the growth plate.

Results. There were 11 upper- and 11 lower-limb cysts. 13 patients had pathological fractures and 9 did not. 20 patients were treated conservatively with limb immobilisation; 2 underwent curettage and bone grafting (one resolved and one did not). Seven cysts resolved (5 had fractures and 2 did not). The risk of fracture was higher in the upper than lower limbs (100% vs 18%, p<0.001). Fractured cysts were larger than unfractured cysts (mean cyst index, 4.5 vs. 2.2, p=0.07). Active cysts were more likely to fracture.

Conclusion. Conservative management had a 30% resolution rate. Surgery should be considered for large active cysts in the upper limbs in order to minimise the fracture risk.

Key words: unicameral bone cyst, active, fracture risk, upper limb, lower limb

INTRODUCTION

Unicameral bone cyst (UBC), also known as simple/benign/juvenile unicameral bone cyst, is the commonest benign bone tumour in the paediatric population. It is a fluid-filled cavitating lesion in the metaphysis of long bones and is distinct from an aneurysmal bone cyst. On radiography, a UBC appears as a centrally located, lytic lesion with well-defined margins and may expand concentrically but never penetrates the cortex. The cyst wall is lined by a fibrous membrane and contains clear yellow, serous fluid. In contrast, an aneurysmal bone cyst appears as an expansile, lytic lesion with periosteal lifting. On
magnetic resonance imaging, an aneurysmal bone cyst shows intralesional septations, double density fluid levels, and a low signal on T1-weighted and a high signal on T2-weighted images.\[^2\] Histologically, the wall of an aneurysmal bone cyst is lined by fibrous membrane, contains haemorrhagic fluid and may co-exist with a giant cell tumour, chondroblastoma, osteoblastoma, fibrous dysplasia, non-ossifying fibroma or chondromyxoid fibroma. We aimed to elucidate the natural history of UBCs and risk factors for pathological fracture.

**MATERIALS AND METHODS**

Between December 1997 and April 2007, 14 males and 8 females (mean age, 9 years; standard deviation [SD], 4 years) diagnosed with UBC were reviewed. Cyst location, symptoms, and whether there was any fracture or surgery were recorded.

The cyst parameters were measured on radiographs (Fig. 1), and included (1) the cyst index\[^3\]—the cyst area (the best-fit trapezoid around the cyst) divided by the diaphyseal diameter\[^2\], (2) the ratio of the widest cyst diameter to the growth plate diameter, and (3) the adjusted distance of the cyst border from the growth plate relative to the growth plate diameter, as the magnification of radiographs for each patient was not standardised. Cysts were latent/active when growing away from/adjacent to the growth plate. Upper- and lower-limb cysts as well as fractured and unfractured cysts were compared using the \(t\)-test. A \(p\) value of <0.05 was considered statistically significant.

**RESULTS**

There were 11 upper- and 11 lower-limb cysts. 13 patients had pathological fractures and 9 did not. 20 patients were treated conservatively with limb immobilisation; 2 underwent curettage and bone grafting (one resolved and one did not). Seven cysts resolved (5 had fractures and 2 did not). On presentation, the mean cyst index was 3.6 (SD, 4.2), the mean ratio of the widest cyst diameter to the growth plate diameter was 0.6 (SD, 0.3), and the mean adjusted distance of the cyst border from the growth plate was 0.62 (SD, 0.52) cm.

The risk of fracture was higher in the upper than lower limbs (100% vs 18%, \(p<0.001\)). Fractured cysts were larger than unfractured cysts (mean cyst index, 4.5 vs. 2.2, \(p=0.07\)). Cysts were larger in the upper than lower limb but not statistically significant (mean cyst index, 4.9 vs. 2.3, \(p=0.16\)). The mean ratios of the widest cyst diameter to growth plate diameter were not significantly different between upper- and lower-limb cysts (0.6 vs. 0.6, \(p=0.96\)) and between fractured

---

\(D\)\(^2\)\(\times L+1\)\(H\)\]

![Figure 1](image-url)  
*Figure 1  Measurement of the (a) cyst index \(\frac{1}{2}(L+1)\times H\), (b) the ratio of the widest cyst diameter to growth plate diameter (A/B), and (c) the distance of the cyst border from the growth plate.*
and unfractured cysts (0.6 vs. 0.5, p=0.45) [Tables 1 and 2]. The mean adjusted distances of the cyst border from the growth plate were not significantly different between fractured and unfractured cysts (0.77 vs. 0.44 cm, p=0.17). Active cysts were more likely to fracture. The mean growth away from the growth plate in fractured and unfractured cysts were 0.01 and 0.67 cm, respectively. In unfractured cysts, the mean adjusted distance increased (i.e. grew away) with time. In fractured cysts, the distance remained more or less constant over time (Fig. 2).

DISCUSSION

UBC is common in children between the ages of 5 and 15 (mean, 9) years, but its aetiology is unknown.4,5 The proximal humerus is the commonest site, followed by the proximal femur. Males are affected twice as often as females.6,7 UBCs are classified into active (juxtaposed to the cartilaginous growth plate) or latent (migrating away and separating from the growth plate by a normal area of cancellous bone).8 Latent cysts are more likely to resolve, with or without treatment. Active cysts are unlikely to regress and have a higher risk of fracture and growth retardation. Because this may lead to angular deformity and limb length shortening,9 such cysts should be treated more aggressively.10,11 UBCs typically present with pathological fractures (usually following minor trauma) or pain secondary to impending fracture. Occasionally, they may be completely asymptomatic and are detected incidentally on radiographs.

Discrepancy between upper- and lower-limb cysts can be explained by the hypothesis that connections of upper-limb cysts with the physis cause decreased growth. The rapid growth of the cyst exceeds the longitudinal growth of the metaphysis. The upper limbs grow more slowly than the lower limbs. Thus, UBCs in upper limbs should be treated more aggressively to minimise the risk of pathological fracture.

In our study, only one of the 7 UBCs that resolved was treated by curettage and bone grafting. This suggests a role for conservative treatment, especially after cysts result in fracture. Up to a third of UBCs resolved spontaneously. 15% of all reported cysts heal spontaneously,12-17 supporting the concept of maturing UBCs.

In our study, conservative management had a 30% resolution rate, but some studies do not recommend watchful waiting.18,19 Surgery should be considered for large, active cysts in the upper limbs in order to minimise the fracture risk.

| Characteristics | No. of cysts |    |    |    | Fractured (n=13) | Unfractured (n=9) | p value |
|-----------------|--------------|----|----|----|----------------|----------------|---------|
| Resolved        | 5            | 2  |    |    |                |                |         |
| Did not resolve | 8            | 7  |    |    |                |                |         |
| Upper limb      | 11           | 0  |    |    |                |                |         |
| Lower limb      | 2            | 9  |    |    |                |                |         |

Figure 2 Line graph showing the trend of unfractured cysts growing away from the growth plate while fractured cysts remain near the growth plate.

| Cyst parameters | Upper limb | Lower limb | p value | Fractured cyst | Unfractured cyst | p value |
|-----------------|------------|------------|---------|----------------|----------------|---------|
| Mean (SD) cyst index | 4.9 (5.6) | 2.3 (1.4) | 0.16 | 4.5 (5.1) | 2.2 (1.5) | 0.07 |
| Mean (SD) ratio of the widest cyst diameter to growth plate diameter | 0.6 (0.2) | 0.6 (0.3) | 0.96 | 0.6 (0.3) | 0.5 (0.3) | 0.45 |
| Mean (SD) adjusted distance of the cyst border from the growth plate (cm) | 0.6 (0.5) | 0.6 (0.6) | 0.84 | 0.77 (0.62) | 0.44 (0.32) | 0.17 |
REFERENCES

1. Carnesale PG. Benign bone tumors. In: Canale TS, editor. Campbell's operative orthopaedics. 10th ed. St Louis: Mosby; 2003:796–9.
2. Sullivan RJ, Meyer JS, Dormans JP, Davidson RS. Diagnosing aneurysmal and unicameral bone cysts with magnetic resonance imaging. Clin Orthop Relat Res 1999;366:186–90.
3. Kaelin AJ, MacEwen GD. Unicameral bone cysts. Natural history and the risk of fracture. Int Orthop 1989;13:275–82.
4. Capanna R, Campanacci DA, Manfrini M. Unicameral and aneurysmal bone cysts. Orthop Clin North Am 1996;27:605–14.
5. Lokic F, Wientroub S. Simple bone cyst: etiology, classification, pathology, and treatment modalities. J Pediatr Orthop B 1998;7:262–73.
6. Zehetgruber H, Bittner B, Gruber D, Krepler P, Trieb K, Kotz R, et al. Prevalence of aneurysmal and solitary bone cysts in young patients. Clin Orthop Relat Res 2005;439:136–43.
7. Campanacci M, Capanna R, Picci P. Unicameral and aneurysmal bone cysts. Orthop Clin Orthop Relat Res 1986;204:25–36.
8. Neer CS, Francis KC, Johnston AD, Kiernan HA Jr. Current concepts on the treatment of solitary unicameral bone cyst. Clin Orthop Relat Res 1973;97:40–51.
9. Moed BR, LaMont RL. Unicameral bone cyst complicated by growth retardation. J Bone Joint Surg Am 1982;64:1379–81.
10. Hecht AC, Gebhardt MC. Diagnosis and treatment of unicameral and aneurysmal bone cysts in children. Curr Opin Pediatr 1998;10:87–94.
11. Capanna R, Campanacci DA, Manfrini M. Unicameral and aneurysmal bone cysts. Orthop Clin North Am 1996;27:605–14.
12. Adams AW. Report of a case of solitary fibrocystic disease of the humerus exhibiting spontaneous resolution: with a review of the literature and a consideration of the etiology and treatment. Br J Surg 1926;13:734–41.
13. Baker DM. Benign unicameral bone cyst. A study of forty-five cases with long-term follow up. Clin Orthop Relat Res 1970;71:140–51.
14. McLachlin AD. Treatment and results in localized osteitis fibrosa cystica (the solitary bone cyst). J Bone Joint Surg Am 1943;25:777–90.
15. Morton KS. Unicameral bone cyst. Can J Surg 1982;25:330–2.
16. Siegel IM. Brisement force with controlled collapse in treatment of solitary unicameral bone cyst. Arch Surg 1966;92:109–14.
17. Stewart MJ, Hamel HA. Solitary bone cyst. South Med J 1950;43:927–34.
18. Neer CS, Francis KC, Johnston AD, Kiernan HA Jr. Current concepts on the treatment of solitary unicameral bone cyst. Clin Orthop Relat Res 1973;97:40–51.
19. Neer CS 2nd, Francis KC, Marcove RC, Terz J, Carbonara PN. Treatment of unicameral bone cyst. A follow-up study of one hundred seventy-five cases. J Bone Joint Surg Am 1966;48:731–45.