

MANAGEMENT STRATEGY FOR UNICAMERAL BONE CYST

Chin-Yi Chuo, Yin-Chih Fu, Song-Hsiung Chien, Gau-Tyan Lin, and Gwo-Jaw Wang
Department of Orthopedic Surgery, Kaohsiung Medical University Hospital,
Kaohsiung, Taiwan.

The management of a unicameral bone cyst varies from percutaneous needle biopsy, aspiration, and local injection of steroid, autogenous bone marrow, or demineralized bone matrix to the more invasive surgical procedures of conventional curettage and grafting (with autogenous or allogeneous bone) or subtotal resection with bone grafting. The best treatment for a unicameral bone cyst is yet to be identified. Better understanding of the pathology will change the concept of management. The aim of treatment is to prevent pathologic fracture, to promote cyst healing, and to avoid cyst recurrence and re-fracture. We retrospectively reviewed 17 cases of unicameral bone cysts (12 in the humerus, 3 in the femur, 2 in the fibula) managed by conservative observation, curettage and bone grafting with open reduction and internal fixation, or continuous decompression and drainage with a cannulated screw. We suggest percutaneous cannulated screw insertion to promote cyst healing and prevent pathologic fracture. We devised a protocol for the management of unicameral bone cysts.

Key Words: unicameral bone cyst, UBC, simple bone cyst, SBC, cannulated screw
(*Kaohsiung J Med Sci* 2003;19:289–95)

The unicameral bone cyst (UBC) or simple bone cyst (SBC) is encountered in the pediatric age group, with the majority of cases occurring in the first two decades of life — with most around the age of 12 years [1,2]. UBC usually appears in the metaphyses of the long bones, especially in the humerus and femur [1,3–5]. Spontaneous healing of a cyst may follow pathologic fracture and there is a tendency for the cyst to disappear after skeletal maturity [3,4,6,7]. There is a greater potential of deformity at the cyst in weight-bearing bones after pathologic fracture that may require internal fixation [6].

The treatment of UBC includes: conservative management to allow spontaneous healing during puberty; curettage and grafting with autogenous or allogeneous cancellous bone, freeze-dried crushed cortical bone allograft [5], or hydroxyapatite (HA) [8]; subtotal resection and grafting [7]; and intralesional injection of steroid [9–12]. More recently, continuous decompression and drainage by application of a cannulated screw [13], percutaneous autogenous marrow grafting, demineralized bone matrix (DBM) local injection [14,15], and flexible intramedullary (IM) nailing in long bone [16] have been reported, with satisfactory results.

The aim of this retrospective case review was to set up a reasonable protocol for UBC management that would lead to a satisfactory clinical result and diminish the risk of re-fracture and cyst recurrence.

Received: December 19, 2002 Accepted: April 20, 2003
Address correspondence and reprint requests to: Dr. Song-Hsiung Chien, Department of Orthopedic Surgery, Kaohsiung Medical University Hospital, 100 Shih-Chuan 1st Road, Kaohsiung 807, Taiwan.
E-mail: shchien@kmu.edu.tw

MATERIALS AND METHODS

In a retrospective review, we assessed 17 cases of UBC seen between November 1989 and September 2001,

and which received different kinds of management. All were diagnosed by plain roentgenography and had pathologic fracture, except one who suffered from a limp in the left hip after falling but had no obvious fracture line. The diagnosis was established by biopsy.

There were 15 males and two females in this series (Table 1). The age of the first manifestation ranged from 6 to 15 years (mean, 10.6 years), except one who was 21 years old. Twelve cysts were in the humerus, three were in the femur, and two were in the fibula. Radiographic evaluation was performed according to the Neer classification with a slight modification by Capanna et al [12]. Each cyst was classified as completely healed, healed with residue, recurred, or no response.

We treated pathologic fractures with splinting, skeletal traction, or casting at the initial visit (except for Cases 1, 7, 14 and 16). Cases 4, 5, 8, 9, 12 and 13 underwent conservative observation after the fracture union (Table 2). In Cases 1, 7, 14 and 16, we followed conventional methods to promote cyst healing and stabilize pathologic fractures. Cases 1 and 14 were

Table 1. Cases of unicameral bone cyst and clinical evaluation

Case	Age (yr)	Sex	Location	Fracture(s)
1	6	Male	PH	+
2	9	Male	PF	+
3	11	Male	MH	+
4	13	Male	DH	+++
5	12	Male	PH	++
6	11	Male	MH	+
7	9	Female	PF	-
8	21	Male	PH	+
9	10	Male	MH	+
10	12	Male	PH	+
11	7	Male	MH	+
12	12	Male	Pf	+
13	15	Male	PH	++
14	9	Female	MF	+
15	14	Male	PH	+
16	11	Male	Mf	+
17	8	Male	MH	+

P = proximal; H = humerus; F = femur; M = middle; D = distal; f = fibula.

Table 2. Fracture (Fr) union time and cyst evaluation after fracture union, management and at final follow-up

Case	Fr union time (mo)	Post Fr union/cyst	Management	Post management/cyst	Total follow-up period/cyst
1	3	*	C+G+pinning	1 yr/healed	12 yr/healed
2	3	6 yr/no response	Screw	4 yr/healed	11 yr/healed
3	6	8 mo/no response	Screw	1 yr 3 mo/healed	7 yr/healed
4	2	1 yr 10 mo/no response	OBS		
	4	3 yr/no response	OBS		
	4	2 yr/recurred	OBS		6 yr/recurred [†]
5	1.5	1 yr 4 mo/residual	OBS		
	3	8 mo/healed	OBS		5 yr/healed
6	6	3 yr 6 mo/recurred	Screw	1 yr 2 mo/healed	4 yr/healed
7		*	C+G+TEN	10 mo/recurred	
			C+G	3 yr/recurred	4 yr/recurred
8	5	9 mo/healed	OBS		3 yr/healed
9	3	2 mo/healed	OBS		2 yr/healed
10	3.5	5 mo/no response	Screw	2 yr/healed	3 yr/healed
11	2	1 mo/no response	Screw	3 yr/healed	5 yr/healed
12	1.5	6 mo/healed	OBS		5 yr/healed
13	2	10 mo/no response	OBS		
	3	5 yr 8 mo/residual	OBS		7 yr/residual [†]
14	3	*	C+G+plating	3 mo/healed	3 yr/healed
15	2	2 mo/no response	Screw	2 yr 2 mo/healed	6 yr/healed
16	3	*	C+G	3 mo/healed	11 yr/healed
17	2	1 mo/no response	Screw	1 yr 8 mo/healed	3 yr/healed

C = curettage; G = grafting; Screw = percutaneous cannulated screw insertion; OBS = observation; TEN = tension band technique. *Cases 1, 7, 14, 16 underwent conventional treatment. [†]Patients' parents refused any operation (Cases 4, 13). Case 7 had no pathologic fracture.

treated with curettage, autografting, and open reduction and internal fixation (ORIF), Case 1 with cross pinning for a proximal humeral fracture and Case 14 with a plate for a femoral fracture. Case 16 was treated with curettage and autografting for a fibular fracture. In Case 7, the cyst was located in the left proximal femur, but there was no fracture. She underwent curettage allografting and the tension band technique. Unfortunately, the cyst recurred 1 year later and the procedure was repeated after removal of the implant.

We managed the seven cases in which the pathologic fracture united but the cyst persisted (Cases 2, 3, 6, 10, 11, 15 and 17) with cannulated screw insertion after introduction of the new concept of decompression and drainage of cysts reported by Ekkernkamp et al in 1990 [13]. The cannulated screw was inserted using an image intensifier through a small skin incision over the anterolateral aspect to the dependent site of the cyst without interference in the growth plate (Table 2).

RESULTS

Of the 16 pathologic fractures (Case 7 had no pathologic fracture), all were united within 6 months (range, 1.5–6 months) regardless of treatment, surgery (Cases 1, 14, 16), or conservative treatment (Table 2). There was no delayed union, nonunion, or malunion except in Case 4, who suffered twice from re-fracture of the humeral shaft and the distal portion that resulted in some deformity. Fourteen cysts were completely healed (Cases 1–3, 5, 6, 8–12, 14–17), one cyst healed with residue (Case 13), and two cysts recurred (Cases 4 and 7); no cysts had no response (Table 2). The total follow-up period ranged from 2 to 12 years (mean, 5.7 years). There was no infection or other operative complications.

In the group under conservative observation of UBC after pathologic fracture union (Cases 4, 5, 8, 9, 12 and 13), only the cysts of Cases 4 and 13 did not heal but recurred or displayed residue after three and two fracture unions, respectively (Table 2). Case 4 had gross deformity over the distal humerus due to multiple fractures and malunion. In the other four cysts, the average period for cyst healing was 6.3 months (8, 9, 2 and 6 months, respectively) after fracture union.

Four cases were initially treated with conventional methods. Cases 1 and 14 underwent curettage, autografting, and ORIF (Table 2). The cysts healed within 1 year and 3 months, respectively. Case 7 underwent curettage, allografting, and the tension band technique. However, the cyst recurred 1 year later. After repeat curettage and allograft, the cyst recurred in the 3 years of follow-up. Case 16 underwent curettage and autografting and the cyst healed in 3 months.

Of the seven cases treated with cannulated screw insertion after pathologic fracture union, the cysts all healed within 4 years, as shown by serial radiographic studies. The mean time to healing was 26 months (range, 1 year 2 months to 4 years) (Table 2). In the cyst healing process, decreased cavity size, thickened cortex, and increased bone density were noted at follow-up.

DISCUSSION

Initial treatment of UBC is to stabilize the pathologic fracture and to obliterate the cyst. Treatment has evolved from curettage and grafting (autogenous or allogeneous cancellous bone, freeze-dried cortical bone, or HA) [5,8], through subtotal resection with grafting (strut or cancellous bone) [7], to more conservative procedures such as intralesional steroid injection [9–12], percutaneous autologous marrow grafting [4,15], continuous decompression and drainage by percutaneous insertion of a cannulated screw [13], or DBM intralesional injection [14,15]. Flexible IM nailing in the weight-bearing long bone has been reported to produce a satisfactory result [16].

Curettage and bone grafting have unreliable results, with recurrence rates from 30 to 50% [6,8]; curettage and HA grafting have been reported to have a 78% complete healing rate [8]. However, there is a risk that curettage of active cysts might damage the growth plate and cause shortening of the extremities [17].

In 1979, Scaglietti et al found a favorable outcome in 90% of 72 UBCs treated with methylprednisolone [18]. Capanna et al had an 80% favorable outcome in 90 patients with steroid injection, and mentioned that other investigators had obtained as high as a 96% favorable result with this simple non-surgical intralesional injection [12].

However, Hashemi-Nejad and Cole concluded that the healing response to intralesional corticosteroid is unpredictable and usually incomplete even after

multiple injections [9]. This is especially true in weight-bearing bones, which may need more aggressive curettage or subtotal resection and grafting [7].

In 1996, Lokiec et al reported 10 cases of UBC treated with percutaneous autologous marrow grafting after curettage [4]. All healed within 4 months after one injection. In 1998, Killian et al treated 11 UBC patients with percutaneous injection of DBM using a two-needle technique and a custom large-bore needle [14]. The cysts were obliterated in nine of the 11 patients within 4 to 5 months of a single injection, and at 2 years' follow-up, no cysts were deemed active or recurrent.

Since 1995, we have used the percutaneous insertion of a cannulated screw, as reported by Ekkernkamp et al [13], to continuously drain the fluid through the cyst wall. The treatment is based on the concept that the lesion is caused by interstitial fluid that is unable to escape from the bone due to venous obstruction and blockage [16].

The last three methods have relatively high rates of satisfactory results and cyst healing through the percutaneous approach. Due to the simple and safe approach through a small skin incision, there are no major operative complications with these methods.

We used three different types of management to treat UBC after pathologic fracture union: close observation, conventional curettage and bone grafting with or without ORIF, and the newer concept of cannulated screw insertion. Of the six cases who underwent close observation, four cysts healed (mean, 6.3 months) after one or two pathologic fractures (Case 5 had two, Cases 8, 9 and 12 had one), but the cysts of Cases 4 and 13 did not heal after three and two fracture unions at 6 and 7 years of follow-up, respectively. Due to these fractures, Case 4 had malunion over the distal humerus and gross deformity. Although most cysts heal after pathologic fractures and disappear after skeletal maturity, they may recur and pathologic fractures can occur.

In the second group, who underwent conventional treatment, the cyst recurred in one of the four cases (Case 7) even after the procedure had been repeated twice (Figure 1). This is comparable with the papers we reviewed that reported unreliable results and a variable recurrence rate.

In the third group, who underwent cannulated screw insertion, the seven cysts healed within 4 years (mean, 26 months) without any re-fractures. This included Case 3, who had undergone curettage and



Figure 1. Case 7 suffered from a limp in the left hip without any fracture but unicameral bone cyst was suspected and proved by pathology. (A) Initial manifestation at roentgenography. (B) Post curettage, allografting, and tension band technique. (C) The cyst recurred at 4 years, after two treatments of curettage and allografting.

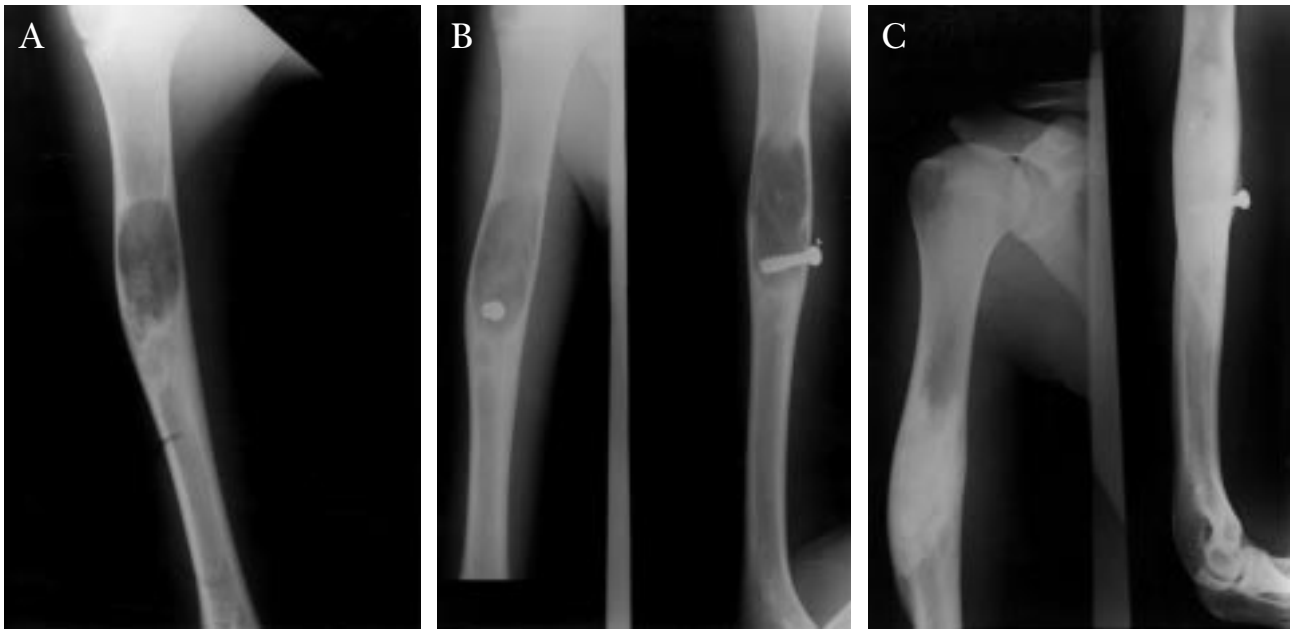


Figure 2. Case 3. (A) Recurrent cyst within 6 months after curettage and bone grafting performed at another hospital. (B) Percutaneous cannulated screw insertion through an anterolateral skin incision. (C) The cyst healed in 1 year 3 months.

bone grafting at another hospital, after which the cyst recurred within 6 months followed by pathologic fracture. At our hospital, a percutaneous cannulated screw was inserted and the cyst healed in 1 year 3 months (Figure 2). With this type of management, the morbidity of autograft harvesting and opportunity for malunion and re-fracture are reduced. Therefore, we believe that percutaneous cannulated screw treatment is an effective and simple procedure to prevent cyst recurrence and re-fracture.

When treating UBC, the pathologic fracture and cyst must usually be dealt with simultaneously. UBC is always encountered in the pediatric age group and usually appears in the metaphyses of the long bones. If pathologic fracture occurs, conservative treatment may be simpler than surgery and operative complications can be avoided. There is no problem about fracture union. Spontaneous healing of a cyst may follow pathologic fracture and there is a tendency for the cyst to disappear after skeletal maturity. If the cyst persists, re-fracture with malunion may occur.

From a review of the published papers and our own experience, we set up a protocol for pathologic fractures and cyst healing with a procedure that is less invasive, simpler and safer, to obtain a higher rate of cyst healing with satisfactory clinical results and a lower complication rate, compared to

conventional methods (Figure 3). In the protocol, because the first main manifestation of UBC is frequently a pathologic fracture, we treat the fracture with conservative methods (splinting, skeletal traction, or casting). Solid fracture union is expected within a few months. Our data revealed that cysts healed within 2 to 9 months (mean, 6.3 months) after pathologic fracture union if spontaneous cyst healing occurred. Thus, we recommend monitoring cysts for 6 months to allow them to heal spontaneously. The children can participate in ordinary daily activities, but must be prevented from falling or overusing their extremities.

If there is no sign of cyst healing within 6 months after fracture union, or for early prevention of cyst recurrence and re-fracture, we suggest percutaneous cannulated screw insertion at the dependent site to allow drainage of the cyst fluid and to lower intralesional pressure in order to elicit cyst healing. If the UBC is diagnosed incidentally without a pathologic fracture, we also suggest immediate use of this simple procedure to decrease the risk of pathologic fracture.

This was a retrospective study. Due to the limited number of cases, we feel that additional cases and a better understanding of UBC may prove our protocol to be effective and safe.

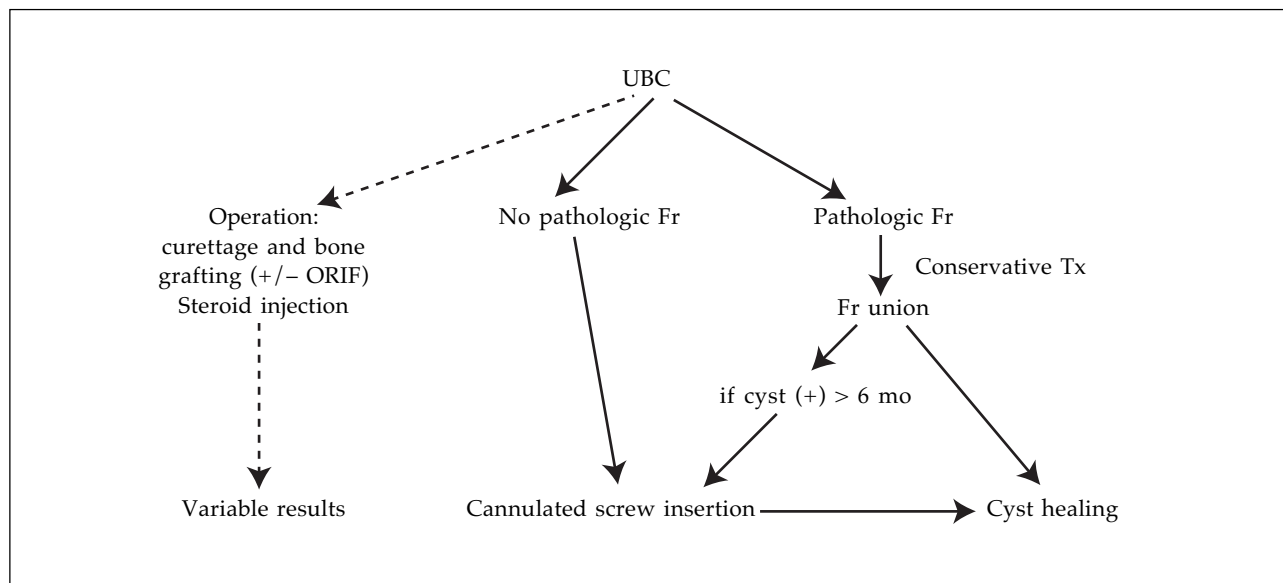


Figure 3. Conventional treatment and steroid injection (dashed arrow) compared with our protocol (solid arrow) for the management of unicameral bone cyst (UBC) and its pathologic fracture. Fr = fracture; Tx = treatment; ORIF = open reduction and internal fixation.

REFERENCES

- Morton KS. Unicameral bone cyst. *Can J Surg* 1982;25:330-2.
- Biermann JS. Common benign lesions of bone in children and adolescents. *J Pediatr Orthop* 2002;22:268-73.
- Makley JT, Joyce MJ. Unicameral bone cyst (simple bone cyst). *Orthop Clin North Am* 1989;20:407-15.
- Lokiec F, Ezra E, Khermash O, Wientroub S. Simple bone cysts treated by percutaneous autologous marrow grafting: a preliminary report. *J Bone Joint Surg Br* 1996;78:934-7.
- Spence KF Jr, Bright RW, Fitzgerald SP, Sell KW. Solitary unicameral bone cyst: treatment with freeze-dried crushed cortical-bone allograft. A review of one hundred and forty-four cases. *J Bone Joint Surg Am* 1976;58:636-41.
- Neer CS, Francis KC, Johnston AD, Kiernan HA Jr. Current concepts on the treatment of solitary unicameral bone cyst. *Clin Orthop* 1973;97:40-51.
- Fahey JJ, O'Brien ET. Subtotal resection and grafting in selected cases of solitary unicameral bone cyst. *J Bone Joint Surg Am* 1973;55:59-68.
- Inoue O, Ibaraki K, Shimabukuro H, Shingaki Y. Packing with high-porosity hydroxyapatite cubes alone for the treatment of simple bone cyst. *Clin Orthop* 1993;293:287-92.
- Hashemi-Nejad A, Cole WG. Incomplete healing of simple bone cysts after steroid injections. *J Bone Joint Surg Br* 1997;79:727-30.
- Shindell R, Connolly JF, Lippiello L. Prostaglandin levels in a unicameral bone cyst treated by corticosteroid injection. *J Pediatr Orthop* 1987;7:210-2.
- Shindell R, Huurman WW, Lippiello L, Connolly JF. Prostaglandin levels in unicameral bone cysts treated by intralesional steroid injection. *J Pediatr Orthop* 1989;9:516-9.
- Capanna R, Alessandro DM, Gitelis S, Campanacci M. The natural history of unicameral bone cyst after steroid injection. *Clin Orthop* 1982;166:204-11.
- Ekkernkamp A, Muhr G, Lies A. Continuous decompression. A new method in the treatment of juvenile bone cyst. *Unfallchirurg* 1990;93:539-43. [In German]
- Killian JT, Wilkinson L, White S, Brassard M. Treatment of unicameral bone cyst with demineralized bone matrix. *J Pediatr Orthop* 1998;18:621-4.
- Rougraff BT, Kling TJ. Treatment of active unicameral bone cysts with percutaneous injection of demineralized bone matrix and autogenous bone marrow. *J Bone Joint Surg Am* 2002;84:921-9.
- Roposch A, Saraph V, Linhart WE. Flexible intramedullary nailing for the treatment of unicameral bone cysts in long bones. *J Bone Joint Surg Am* 2000;82:1447-53.
- Crawford AH. Growth arrest resulting from unicameral bone cyst. *J Pediatr Orthop* 1998;18:824-5.
- Scaglietti O, Marchetti PG, Bartolozzi P. The effects of methylprednisolone acetate in the treatment of bone cysts. Results of three years follow-up. *J Bone Joint Surg Br* 1979;61:200-4.