J Child Orthop (2008) 2:279–283
DOI 10.1007/s11832-008-0114-0

ORIGINAL CLINICAL ARTICLE

Continuous decompression with intramedullary nailing for the treatment of unicameral bone cysts

Julio Javier Masquijo · Eduardo Baroni · Horacio Miscione

Received: 22 January 2008 / Accepted: 21 June 2008 / Published online: 24 July 2008

Abstract
Purpose To evaluate the efficacy of decompression of unicameral bone cysts (UBCs) of the long bones with intramedullary nailing and to compare responses to treatment according to location.
Materials and methods We evaluated 48 consecutive patients treated between January 1988 and June 2000. Mean age was 10.3 years. Mean follow-up was 9.8 years. Evaluation was performed according to the radiographic criteria of Capanna.
Results UBCs were located in the proximal humerus ($n = 24$), humeral shaft ($n = 2$), proximal femur ($n = 19$), distal tibia ($n = 2$) and fibula ($n = 1$). A total of 62.5% presented a pathological fracture. Successful results were observed in 89.5% (26 total healing, 17 healing with residual radiolucent areas), and there were four recurrences and, in one case, no response to treatment. There was more healing in the humerus than in the femur (92.3% versus 84.2%), and more tendency to restitution ad integrum, although the difference was not statistically significant ($P = 0.1499$).
Conclusions Intramedullary nailing is a minimally invasive method, which permits early stability and decompresses the cyst allowing healing. Significant differences were not observed among results from different locations.

Keywords Unicameral bone cyst · Intramedullary nailing · Decompression

J. J. Masquijo (✉) · E. Baroni · H. Miscione
Department of Pediatric Orthopaedics,
Hospital de Pediatría Juan P. Garrahan,
Buenos Aires, Argentina
e-mail: otihg@fibertel.com.ar; javimasquijo@yahoo.com.ar

Introduction

The unicameral bone cyst (UBC) is a benign fluid-filled lesion, which is located mainly in metaphyses of long bones in skeletally immature patients [3, 19]. The possible treatment spectrum includes observation, aspiration and injection of corticosteroids [4, 9, 24, 25], curettage and bone grafting [8, 28, 29], subtotal resection with or without bone grafting [11, 20] and percutaneous injections of bone marrow [19, 31, 32]. All these therapeutic options show variable results.

Although the aetiology of UBC remains unknown, various authors agree that the obstruction of the venous return and the increase in the resulting intracystic pressure can be the main factors influencing its size increase [6, 7, 12, 18]. Opening of the medullary canal, creating a connection between the canal and the cyst, would permit permanent decompression, which would favour the healing of the lesion. Although this does not represent a new concept [15], recent studies have shown a high rate of healing using this technique [13, 21].

Our purpose was to evaluate the efficacy of the UBC decompression of the long bones with intramedullary nailing and compare the treatment response according to the location.

Materials and methods

All the patients with a diagnosis of UBC of the long bones, treated with intramedullary nailing between January 1988 and June 2000, were analysed retrospectively. Institutional Review Board approval and informed consent had been obtained previously. As inclusion criteria, the following was used: patients of both sexes with diagnosis of UBC of

the long bones treated surgically with intramedullary nailing. Exclusion criteria: patients with incomplete clinical histories or radiographies and a follow-up less than 5 years. The information was obtained from Hospital records.

Surgical technique

We performed no open or percutaneous biopsies prior to the procedure. A diagnosis of UBC is strongly suspected based on its typical radiographic appearance and is confirmed when an appropriate cyst fluid is demonstrated during surgery. In the case of humeral or femoral location, the insertion of the nail was performed in a retrograde manner. In the case of tibial location, an anterograde insertion was performed through a medial and lateral approach. Once the nail was inserted, the opening of the intramedullary canal was carried out, creating a connection between the canal and the cyst. This favours continuous decompression of the cyst. The election of the material was carried out according to its availability, the size of the intramedullary canal of the affected bone and the age of the patient. The postoperative immobilization was evaluated according to the stability achieved with the fixation, the size of the cyst and the presence of pathological fracture. Sports activities were not allowed until the cyst showed consolidation, demonstrated on X-rays.

Evaluation of the patients

Anteroposterior and lateral radiographs of the affected long bone were performed on the first postoperative day, and 1 month, 3 months, 6 months and 1 year after surgery. Thereafter, the patients were annually called for radiographic control. Results were evaluated according to the criteria of Capanna [2]: (1) healing when the cyst was completely filled with a new bone formation and the cortical margins had thickened; (2) healing with residual radiolucency when most of the cyst was filled with bone and had healed, it was well consolidated with bone, and the cortical margins had thickened but there were still small, residual areas of radiolucency; (3) recurrence when it had healed initially and had become filled with bone but large areas of radiolucency and cortical thinning subsequently developed; and 4) no response to the treatment when there was no evidence of any effect of treatment. Both local recurrences and no response represented a failure of treatment.

Statistical methods

Data obtained was expressed in absolute numbers and percentages. Statistical analysis using Student's *t*-tests was carried out to detect differences in the response to the treatment among the different locations. A $P < 0.05$ was considered statistically significant.

Results

Between January 1988 and June 2000, 54 consecutive patients with a diagnosis of UBC of the long bones were treated using the intramedullary nailing technique. Of these, 48 cases were included for analysis—32 males and 16 females—and the 6 remaining patients were lost to follow-up. The mean age of these patients was 10.3 years (range 5–18 years). There was a minimum follow-up of 5 years for all cases, with a mean follow-up of 9.8 years (range 5–19 years). The cysts were located in the proximal humerus in 24 patients, in the humeral shaft in 2, in the proximal femur in 19, in the distal tibia in 2 and in the fibula in 1.

In 62.5% ($n = 30$) of the patients, pathological fractures were demonstrated, in 25% ($n = 12$) pain or functional impotence with no radiographic images of fracture through the cyst, and in the 12.5% remaining ($n = 6$) there were incidental radiographic findings. None of the fractures involved the physis. However, one case of humeral location proximal to the physis evolved with early physeal closure and marked length discrepancy. Previous treatment had been received by 39.5% ($n = 19$) of the patients, 6 of whom had received more than one treatment. Ender's nails were used in 44 cases (1 nail in 23 patients, 2 in 15 and 3 in 6), Rush nails were used in 3 cases, and Steinman in 1 case.

According to the radiographic criteria of Capanna [2], in 89.5% of the cases successful results were reported—26 patients presented a total response (healing), 17 a partial response (little residual areas of radiolucency smaller than 1 cm in the region of the cyst and with the radiographic follow-up an increase in the size of said areas was not observed) and 4 local recurrences (Fig. 1). At the beginning, the patients that experienced recurrence evolved in a favourable way with bone formation; but then the osteolitic image began expanding such that a re-treatment was recommended. All were treated with a new intramedullary nailing and all healed, none experiencing recurrence again (Table 1). Although one case located in the proximal aspect of the femur showed no response, no fractures were suffered at 5 years of follow-up. The result distribution by location can be observed in Table 2. We found no difference in the rate of consolidation with or without pathological fracture. When analysing the two regions with more frequency of presentation (humerus and femur), it could be observed that, although the femur presented a minor response to the treatment (92.3% versus 84.2%), the difference was not statistically significant ($P = 0.1499$). However, in our

Fig. 1 6+0 female patient: pathological fracture through proximal humerus bone cyst (a), immediate postoperative with Enderś nail (b), 6 months follow-up (c), 1 year follow-up (d), 5 years follow-up (e), and complete healing at 9 years follow-up (f)

Table 1 Recurrences of treatment with intramedullary nailing

Name	Age (years)	Location	1st Treatment	2nd Treatment	Evolution
CJ	5 + 3	Proximal femur	Ender's (2)	Change of nails	1
GS	10 + 0	Humeral shaft	Ender's (1)	Change of nail	2
BM	6 + 0	Proximal femur	Ender's (2)	Change of nails	1
PV	8 + 1	Proximal humerus	Ender's (1)	Change of nail	2

1 complete healing, *2* healing with residual radiolucency

series, the humerus has shown more tendency towards total healing than the femur (61.5% versus 42.1%).

The complication rate of the technique was of 4%. A patient with humeral location had radial neurapraxia and recovered spontaneously in 2 months. In one of the cases, the migration of surgical material (Ender's) could be observed. This required its re-positioning. In four cases, a second surgery had to be carried out due to the growth of the affected member. (In three cases, a change of nails was required and in the remaining patient a longer nail was added.) The osteosynthesis was removed in seven cases (five humerus, two femur) due to pain at the insertion site.

Discussion

Despite the continuous publication of studies about UBC, very little is known about its aetiology and pathogenesis [14]. Several authors agree on the fact that venous obstruction is the main cause, with the resulting rise of internal pressure in the cyst, which would lead to the increase in its size [6, 7, 12, 18]. The increased pressure inside the cyst would cause the osseous re-absorption of the cyst wall and subsequent accumulation of exudates in the centre of the metaphyses. Besides, it was suggested that cyst fluid would play an important role in its pathogenesis. The interleukin 1, the prostaglandins and proteolytic enzymes present in the cyst would start a great resorptive activity in the affected bone [1, 26]. According to these findings, the continuous drainage of the cyst fluid and the continuous decompression of the cyst seem to be a reasonable option for the treatment of these lesions.

Based on this theory, some authors initially tried the decompression through drilling of multiple holes with Kirschner wires [27] and cannulated screws [30]. Although Chigira [6] reported good results in the short term, 42% (three of seven cases) presented with late recurrences. Although the intramedullary nailing of long bones does not represent a new concept [15], recent studies have demonstrated a healing rate between 94 and 100% [10, 13, 21], making the treatment of these lesions once more a topic for investigation. The different elasticity modulus between the bone and the metal deform the nail slightly during ambulation. This deformation prevents the formation of new bone tissue in direct contact with the nails. Consequently, a small layer of loose connective tissue is created around the nails [23]. This technique has the advantage of being carried out through a small incision, diminishing the blood loss and with no necessity of creating an area of cortical thinning, which reduces the hospital stay and accelerates return to normal activity. Moreover, it can be associated with other procedures during the intervention, such as corticosteroids or autologous bone-marrow injections.

Table 2 Result distribution by location

Location	*n*	Capanna				Healing rate (%)
		1	2	3	4	
Humerus	26	16 (61.5%)	8 (30.8%)	2 (7.7%)	–	92.3
Femur	19	8 (42.1%)	8 (42.1%)	2 (10.5)	1 (5.3%)	84.2
Tibia	2	1 (50%)	1 (50%)	–	–	100
Fibula	1	1 (100%)	–	–	–	100

Capanna *1* complete healing, *2* healing with residual radiolucency, *3* recurrence, *4* no response to treatment

In our sample, 89% (43/48) of the cases had a successful result, comparable to the results obtained by Roposch [21, 22], De Sanctis [10], Knorr [16, 17], Santori [23] and Catier [5] (Table 3). We did not find significant differences in the response to the treatment according to the location. Humerus presented more tendencies to restitution ad integrum and a major percentage of healing than femur. In our experience, only 8% of our patients required a second

Table 3 Rates of healing with intramedullary nailing according to different authors

Author	Year	*n*	Capanna				Healing (%)	Mean follow-up
			1	2	3	4		
Catier [5]	1981	2	2	–	–	–	100	[a]
Santori [23]	1988	11	[b]	[b]	[b]	[b]	100	10.5 months
Knorr [17]	1996	5	5	–	–	–	100	[a]
Roposch [21]	2000	32	14	16	2	–	94	4.5 years
Knorr [16]	2003	15	10	5	–	–	100	[a]
Roposch [22]	2004	12	2	9	–	–	100	4.8 years
De Sanctis [10]	2006	47	31	16	–	–	100	6.5 years

n number of patients, Capanna *1* complete healing, *2* healing with residual radiolucency, *3* recurrence, *4* no response to treatment

[a] Not specified in the study

[b] Did not use Capanna's criteria for evaluation

intervention to change the nailing, a rate noticeably less than the 28% reported by Roposch [21].

In conclusion, this study represents the series with the largest number of UBC cases treated using this method and with the longest follow-up period. Intramedullary nailing represents an effective treatment of choice for UBC in the long bones. It is a minimally invasive, well-tolerated method, which allows an acceptable stability, obviates the need for a plaster cast and decompresses the lesion, thus permitting its healing. There were no significant differences in the results in the different locations.

References

1. Bumci I, Vlahoviæ T (2002) Significance of opening the medullar canal in surgical treatment of simple bone cyst. J Pediatr Orthop 22:125–129. doi:10.1097/00004694-200201000-00026
2. Capanna R, Dal Monte A, Gitelis S (1982) The natural history of unicameral bone cyst after steroid injection. Clin Orthop Relat Res 166:204–211
3. Capanna R, Campanacci DA, Manfrini M (1996) Unicameral and aneurysmal bone cysts. Orthop Clin North Am 27(3):605–614
4. Carrata A, Garbagna P, Mapelli S, Zucchi V (1983) The treatment of simple bone cysts by topical infiltrations of methylprednisolone acetate: technique and results. Eur J Radiol 3(1):3–8
5. Catier P, Bracq H, Canciani JP (1981) The treatment of upper femoral unicameral bone cysts in children by Ender's nailing technique. Rev Chir Orthop Reparatrice Appar Mot 67(2):147–149
6. Chigira M, Maehara S, Arita S (1983) The aetiology and treatment of simple bone cysts. J Bone Joint Surg Br 65:633–637
7. Cohen J (1960) Simple bone cysts: studies of cyst fluid in six cases with a theory of pathogenesis. J Bone Joint Surg Am 42:609–616
8. Cohen J (1977) Unicameral bone cysts. A current synthesis of reported cases. Orthop Clin North Am 8(4):715–736
9. De Palma L, Santucci A (1987) Treatment of bone cysts with methylprednisolone acetate. A 9 to 11 year follow-up. Int Orthop 11(1):23–28. doi:10.1007/BF00266054
10. De Sanctis N, Andreacchio A (2006) Elastic stable intramedullary nailing is the best treatment of unicameral bone cysts of the long bones in Children?: prospective long-term follow-up study. J Pediatr Orthop 26:520–525
11. Fahey JJ, O'Brien ET (1973) Subtotal resection and grafting in selected cases of solitary unicameral bone cyst. J Bone Joint Surg Am 55(1):59–68
12. Gerasimov AM, Toporova SM, Furtseva LN (1991) The role of lysosomes in the pathogenesis of unicameral bone cysts. Clin Orthop Relat Res 266:53–63
13. Givon U, Sher-Lurie N, Schindler A, Ganel A (2004) Titanium elastic nail—a useful instrument for the treatment of simple bone cyst. J Pediatr Orthop 24(3):317–318. doi:10.1097/00004694-200405000-00014
14. Glaser DL, Dormans JP, Stanton RP (1999) Surgical management of calcaneal unicameral bone cysts. Clin Orthop Relat Res 360:231–237. doi:10.1097/00003086-199903000-00027
15. Imhauser G (1968) Management of juvenile bone cysts using intramedullary nailing. Z Orthop Ihre Grenzgeb 105(3):110–111
16. Knorr P, Schmittenbecher PP, Dietz HG (1996) Treatment of pathological fractures of long tubular bones in childhood using elastic stable intramedullary nailing. Unfallchirurg 99(6):410–414
17. Knorr P, Schmittenbecher PP, Dietz HG (2003) Elastic stable intramedullary nailing for the treatment of complicated juvenile bone cysts of the humerus. Eur J Pediatr Surg 13(1):44–49. doi:10.1055/s-2003-38288
18. Komiya S, Minamitani K, Sasaguri Y (1993) Simple bone cyst: treatment by trepanation and studies on bone resorptive factors in cyst fluid with a theory of its pathogenesis. Clin Orthop Relat Res 287:204–211
19. Lokiec F, Ezra E, Khermosh O, Wientroub S (1996) Simple bone cysts treated by percutaneous autologous marrow grafting. A preliminary report. J Bone Joint Surg Br 78(6):934–937. doi:10.1302/0301-620X78B6.6840
20. McKay DW, Nason SS (1977) Treatment of unicameral bone cysts by subtotal resection without grafts. J Bone Joint Surg Am 59(4):515–519
21. Roposch A, Saraph V, Linhart WE (2000) Flexible intramedullary nailing for the treatment of unicameral bone cysts in long bones. J Bone Joint Surg Am 82-A(10):1447–1453
22. Roposch A, Saraph V, Linhart WE (2004) Treatment of femoral neck and trochanteric simple bone cysts. Arch Orthop Trauma Surg 124(7):437–442. doi:10.1007/s00402-004-0702-5
23. Santori F, Ghera S, Castelli V (1988) Treatment of solitary bone cysts with intramedullary nailing. Orthopedics 11(6):873–878
24. Scaglietti O, Marchetti PG, Bartolozzi P (1979) The effects of methylprednisolone acetate in the treatment of bone cysts. Results of three years follow-up. J Bone Joint Surg Br 61-B(2):200–204
25. Scaglietti O, Marchetti PG, Bartolozzi P (1982) Final results obtained in the treatment of bone cysts with methylprednisolone acetate (depo-medrol) and a discussion of results achieved in other bone lesions. Clin Orthop Relat Res 165:33–42
26. Shindell R, Connolly JF, Lippiello L (1987) Prostaglandin levels in a unicameral bone cyst treated by corticosteroid injection. J Pediatr Orthop 7:210–212
27. Shinozaki T, Arita S, Watanabe H, Chigira M (1996) Simple bone cysts treated by multiple drill-holes. 23 cysts followed 2–10 years. Acta Orthop Scand 67:288–290
28. Spence KF, Sell KW, Brown RH (1969) Solitary bone cyst: treatment with freeze-dried cancellous bone allograft. A study of one hundred seventy-seven cases. J Bone Joint Surg Am 51(1):87–96
29. Spence KF Jr, Bright RW, Fitzgerald SP, Sell KW (1976) 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 58(5):636–641
30. Tsuchiya H, Abdel-Wanis ME, Uehara K, Tomita K, Takagi Y, Yasutake H (2002) Cannulation of simple bone cysts. J Bone Joint Surg Br 84(2):245–248. doi:10.1302/0301-620X.84B2.12473
31. Wientroub S, Goodwin D, Khermosh O, Salama R (1989) The clinical use of autologous marrow to improve osteogenic potential of bone grafts in pediatric orthopedics. J Pediatr Orthop 9(2):186–190
32. Yandow SM, Lundeen GA, Scott SM, Coffin C (1998) Autogenic bone marrow injections as a treatment for simple bone cyst. J Pediatr Orthop 18(5):616–620. doi:10.1097/00004694-199809000-00012