CONTRIBUTION TO THE VASCULAR ORIGIN OF THE UNICAMERAL BONE CYST

M. GEBHART[1], P. BLAIMONT[2]

Vascular occlusion between bone cyst and intramedullary venous system seems to be the primum movens in the genesis of a unicameral bone cyst. High speed injection of saline and alternative aspiration injection of the cyst fluid and reaming of the medullary cavity clearly open vascular channels connecting the bone cyst to the intramedullary venous system. This was monitored by pre- and postmanipulative radiographs. This technique was employed in 12 patients with a unicameral bone cyst. All but one went on to healing with increasing diameter of the cortical bone and filling of the cystic cavity.

Keywords : unicameral bone cyst.
Mots-clés : kyste osseux solitaire.

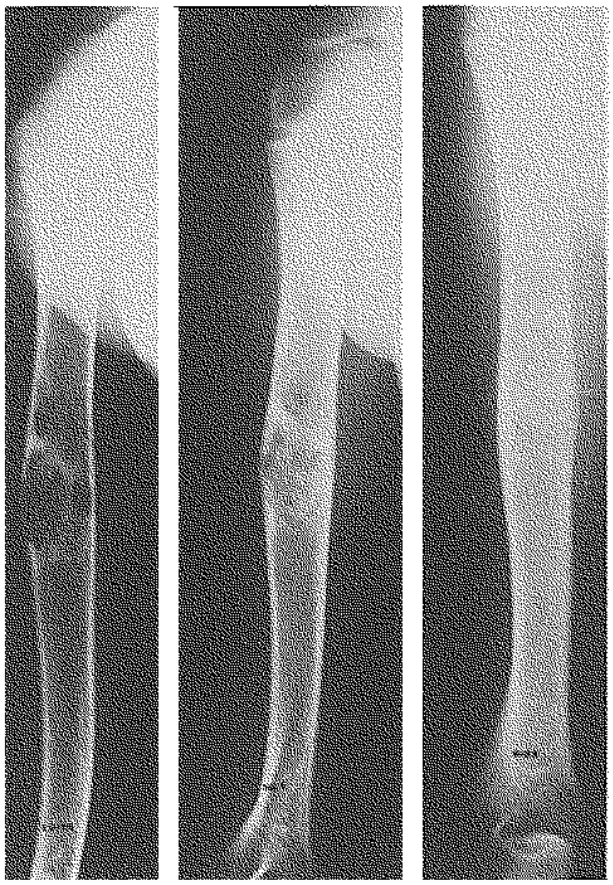
INTRODUCTION

Unicameral bone cyst is a not uncommon bony lesion discovered most often in children and adolescents (3, 9, 13). The preferred location is the proximal humerus (fig. 1), followed by the proximal femur. Less frequent locations are the tibia and fibula, calcaneum, radius and ulna. Almost every other bone which undergoes endochondral ossification during fetal and postfetal development can present with such a malformation. Clinically this lesion is most often silent, and its true incidence is difficult to estimate. Spontaneous fractures and pain are the most classical clinical features. Seldom does this cyst manifest itself by swelling or limitation of motion of the nearest joint.

The etiology at present is unclear. Several mechanisms have been advocated (3, 5) : a traumatic hematoma which undergoes cystic resorption, a cystic degeneration of a preexisting benign tumor, invagination of the synovial membrane of a large joint through the epiphyseal plate during osseous growth and its later separation (7, 8). This latter theory is based on the observation that unicameral bone cysts are located exclusively in the metaphyseal areas of long bones.

Broder (4) was the first to advocate vascular obstruction of the intramedullary venous drainage system of the bone as the possible origin for unicameral bone cyst. Several arguments are in favor of this hypothesis. Histologically, the cyst wall is composed of mesothelial-like cells and entirely separated from the circulation, but with high pressure injection into the cyst, communication with the venous system can be obtained (13). The intracystic fluid has components more similar to serum than to synovial fluid. One exception is the at least ten times higher concentration of alkaline phosphatase (13). Many authors now agree that the classical unicameral bone cyst is a stagnant lake, which contains a serum-like fluid. This fluid has a slightly higher pressure than the intramedullary pressure of the bone marrow of a normal contralateral bone (5). This slightly increased pressure leads to slow bone resorption and to enlargement of the cystic cavity. The cortical bone becomes thinner and thinner until a fracture occurs. The decreased pressure within the cystic cavity following a fracture often leads to healing

[1] Department of Surgery, Bordet Institute, Brussels, Belgium.
[2] Department of Orthopedic Surgery, Ixelles Hospital, Brussels, Belgium.
Correspondence and reprints : M. Gebhart.



a b c

Fig. 1. — Example of a unicameral bone cyst of the proximal humerus in a 12-year-old boy.
a) Recurrence of the unicameral bone cyst after curettage and bone grafting 3 years before.
b) Bony filling of the bone cyst at 4 months after high pressure saline solution injection and high speed injection-aspiration of the cyst fluid, plus reaming of the medullary cavity, followed by a low speed injection of methylprednisolone acetate.
c) At 16 months.

of the cyst. One other argument for the venous obstruction theory is derived from the new therapeutic approach advocated by Scaglietti (10, 11). Until then curettage with bone grafting (9) and even en bloc resection with complicated reconstruction procedures (1, 2, 14) were the current treatment modalities for this benign bone disease, and were associated with a high local recurrence rate ; Scaglietti empirically chose to inject the bone cyst with methylprednisolone acetate, and was able to cure about 90% of the patients under the age of 15 years. An explanation for those surprising results is still lacking. Is it the anti-inflammatory action of prednisolone on the cyst wall or is it bone-induction caused by the contact between cyst wall and corticosteroids (2) ? Another very important observation was the fact that simple multiple drilling (12, 5) of the unicameral bone cyst led to similar good clinical results. According to these authors it is mostly the establishment of a valid drainage system which will lead to the osseous filling of the bone cyst. This same venous drainage system could persist after pathological fracture and this feature may be the explanation for the spontaneous healing in some of the affected bones (15%).

Based on this hypothesis, we present a quite similar therapeutic approach to unicameral bone cyst, the principal objective of which is to open up intracystic vascular channels and to make them communicate with the intramedullary venous vessels. This will be confirmed by intraoperative radiographic imaging.

MATERIAL

Twelve patients were treated between 1986 and 1992. There were 8 males and 4 females. The median age was 11.5 years with a range from 5 to 33. The bone cysts were located in the humerus in 5, in the femur in 3, in the calcaneum in 2 and in the tibia or fibula in one patient each. Six patients had pathological fractures before treatment ; 2 had more than one fracture. Seven patients had 2 operative manipulations, 5 had 3.

The healing process was rated in 5 categories :

- 0 : progression of the bone cyst ;
- 1 : status quo ;
- 2 : partial osseous filling with minor cortical remodeling ;
- 3 : total osseous filling with major cortical bone thickening and
- 4 : almost total recovery of a normally shaped bone.

METHODS

Operative technique

An 18-gauge needle was inserted by direct puncture under image intensification into the cyst. Fluid of the

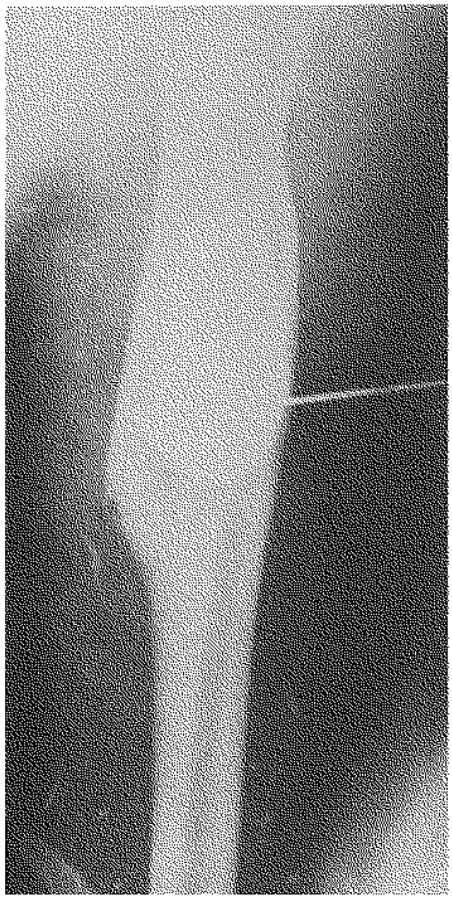


Fig. 2. — Low pressure filling of the bone cyst with dye shows no connection with the intramedullary venous system of the bone.

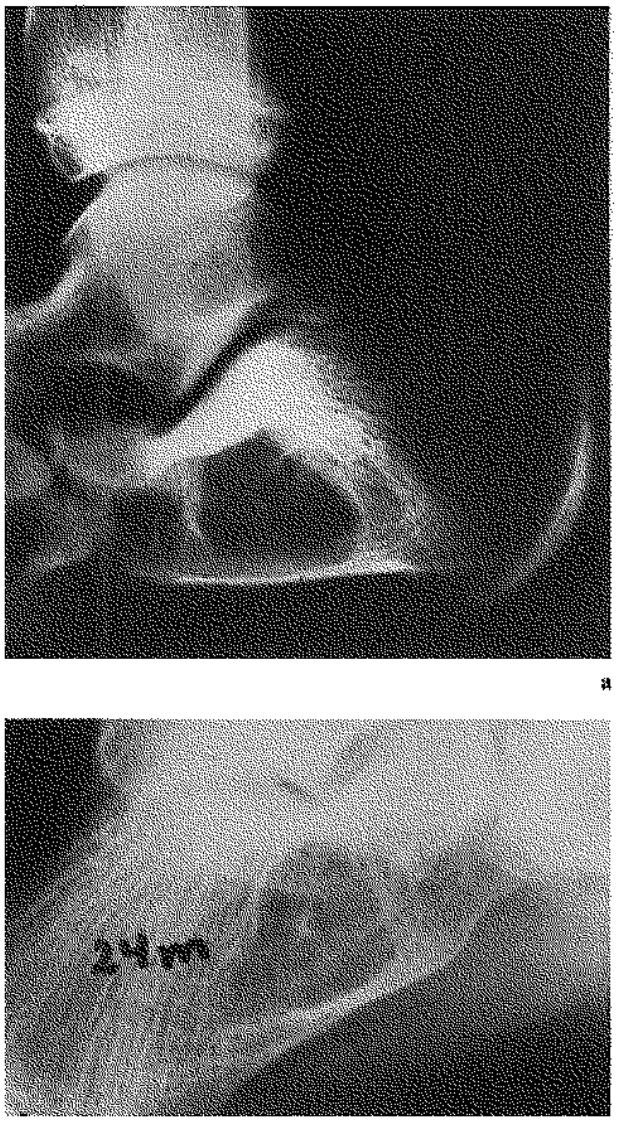
Fig. 3. — The same bone cyst as in fig. 2 after mechanical manipulation. Low pressure injection of contrast medium shows direct leakage of the dye into the venous blood stream of the medullary cavity.

cyst was slowly aspirated and studied for cell content, protein concentration, and alkaline phosphatase. Then contrast material was injected very slowly into the cyst (fig. 2). If the cyst was divided into different chambers, another needle was inserted into the remaining cyst compartments. The next step was to inject under high pressure 100 ml of saline solution, followed by alternative high positive and negative aspiration and injection. A second low speed injection of dye was performed after these manipulations, in order to check venous drainage, and it was documented by radiographs (fig. 3). In order to favor invasion of vessels into the bone cyst, connections between the medullary cavity and the bone cyst were obtained either by drilling holes between both compartments or by introducing a rod into the bone cavity. This rod was then manipulated in order to destroy most of the wall separating the cyst from the medullary cavity. Only at the end of the procedure was methylprednisolone acetate slowly injected. The procedure was repeated at least twice at 6-week intervals. After the second and third injections, the dye left the cystic cavity immediately upon "low" pressure injection (fig. 4).

RESULTS (Table I)

With a median follow-up of 28 months (range from 5 to 72 months) no progressive or stationary disease was observed. Two patients remodeled the bone to normal (fig. 1), 8 patients had a grade 3 (fig. 5) and 2 a grade 2 response to the surgical

Fig. 4. — Low pressure injection of the same bone cyst 6 weeks later shows the persistence of the venous connections even before the mechanical manipulation. The cyst volume is reduced by new bone formed within the cystic cavity.



a

b

Fig. 5. — 13-year-old boy with a solitary bone cyst of the calcaneus.
a) Before injection ;
b) Situation 2 years after treatment.

treatment. In no instance was a recurrence observed. No patients presented with pathological fractures after treatment. It could be suggested from this observation that bone filling of the cyst is related to the direct voiding of the dye into the vascular system of the medullary cavity during the second low pressure injection.

The interesting finding reported here was the intra-operative monitoring of the vascular pattern before and after the high pressure injection with oscillating movements of the intracystic fluid. Figure 2 shows the cystic cavity after low pressure injection of dye prior to the injection-aspiration manipulations. No contrast leakage was observed. After the high pressure injection and aspiration, the intracystic fluid became bloody. Figure 3 shows the same bone cyst after high pressure injection-aspiration manipulations. Here again, the dye was injected at low speed, and direct efflux of the contrast medium into the intramedullary and peripheral venous system could be observed. More interesting is that even 6 weeks later, during the low pressure injection of the dye before the high pressure injection-aspiration

Table I

	Age	Sex	Localization	Previous fracture	Number of injections	Follow-up in months	Cyst healing* (0, 1, 2, 3, 4)	Vascular drainage of cyst at 2nd and 3rd injection direct voiding
1	11	F	prox humerus	yes	3	65	3	yes
2	14	M	humeral diaphysis	yes	3	72	4	yes
3	8	M	calcaneus	no	2	60	3	yes
4	13	M	calcaneus	no	2	24	3	yes
5	12	M	prox humerus	yes	2	7	3	yes
6	5	F	prox femur	yes	2	8	4	yes
7	12	M	prox humerus	yes	2	18	3	yes
8	11	M	prox humerus	yes	3	5	3	yes
9	16	M	distal femur	no	3	48	2	yes
10	8	F	prox fibula	no	2	18	3	yes
11	33	F	prox femur	no	3	32	2	no
12	24	M	prox tibia	no	2	36	3	yes

* *cyst healing* : 0 : progression
1 : no difference
2 : partial osseous filling
3 : total osseous filling + increased diameter of the cortex
4 : almost complete healing

maneuver, the dye leaked directly into the intramedullary and peripheral venous system in 10 of the 11 patients (fig. 4). The cystic cavity was reduced by ingrowing bone.

DISCUSSION

Although the pathological, clinical and radiological features of unicameral bone cyst have been well studied for a long time, its etiology is still unknown and open to speculation. Two hypotheses are emerging : one advocates the synovial origin, the other favors vascular occlusion of the intramedullary venous drainage system. The latter hypothesis is based on histological examination of the cyst wall and especially the fact that revascularization either through a pathological fracture, multiple hole drilling or injection of corticosteroids occurs. Corticosteroid injections may have this revascularization effect through introduction of a needle, the increased pressure during the injection itself or through an anti-inflammatory action which opens the vascular channels. Our therapeutic approach tends to favor the rupture of the vascular occlusions and to connect the cystic lake with the intramedullary venous drainage system. Clear differences in the vascular dynamics of the bone cyst prior to and after the mechanical disturbance were observed.

Initially, the unicameral bone cyst can be considered as a stagnant lake filled with a serum-like fluid, and a low pressure injection of dye does not show connections between the cystic cavity and the intramedullary venous system. The mechanical high pressure injection-aspiration manipulation is effective in opening the channels that connect the cyst with the intramedullary venous system. One can observe direct efflux of the

contrast medium from the cyst into the intramedullary venous drainage system. The opening is reinforced by reaming with a 3-mm diameter rod.

These vascular channels remained open in at least 11 of the 12 patients in whom the same procedure was performed 6 weeks later. Our twelve patients went on to healing, but the follow-up is still short for some of them. It would be interesting to see if the healing tendency of those bone cysts is directly related to the persistence of the vascular connections between cyst and intramedullary blood flow.

The intracystic injection of methylprednisolone acetate led to a high healing rate in other studies. Thus, it is still unclear if the corticoid by itself opens up the vascular channels of the cyst wall or if this occurs after the mechanical aggression, which consists in a needle puncture and an injection. According to our experiment, there are no arguments in favor of a direct action of the corticosteroid, because this product is directly released into the systemic circulation. We think that the mechanical aggression (puncture, injection, high speed injection and aspiration and reaming) is the primum movens of the healing process. This is correlated with the fact that spontaneous healing of bone cysts occurs after pathological fractures or after multiple drilling of the bone cyst. Both situations lead to decreased pressure within the cyst. The establishment of an efficient drainage system would be the best explanation for the subsequent cyst healing.

CONCLUSION

Recent advances in the treatment of unicameral bone cysts by injection of corticosteroids or by mechanical aggression consisting in multiple bone drilling have given arguments in favor of a malfunction of the venous drainage system between bone cyst and intramedullary blood circulation. An operative procedure consisting in high pressure injection-aspiration was designed to open up the hypothetical vascular channels. This event was monitored by low pressure intracystic injections of dye prior to and after the mechanical manipulations. This study shows clear differences in the vascular perfusion dynamics : prior to the mechanical manipulation, the cyst fluid is entirely separated from the intramedullary venous system. After mechanical manipulation, the cyst is integrated into the venous intramedullary system and this phenomenon persists until at least six weeks later.

REFERENCES

1. Agerholm J. C., Goodfellow J. W. Simple cysts of the humerus treated by radical excision. J. Bone Joint Surg., 1965, 47-B, 714-717.
2. Bensahel H., Baum C. Traitement des kystes osseux solitaires du col du fémur chez l'enfant. J. Chir., 1974, 107, 61-66.
3. Boseker E. H., Bickel W. H., Dahlin D. C. A clinicopathologic study of simple unicameral bone cysts. Surg. Gynecol. Obstet., 1968, 127, 550-560.
4. Broder H. M. Possible precursor of unicameral bone cysts. J. Bone Joint Surg., 1968, 50-A, 503-507.
5. Chigira M., Maehara S., Arita S., Udagawa E. The etiology and treatment of simple bone cysts. J. Bone Joint Surg., 1983, 65-B, 633-637.
6. Cohen J. Simple bone cysts ; studies of the cyst fluids in six cases with a theory of pathogenesis. J. Bone Joint Surg., 1960, 42-A, 609-616.
7. Mirra J. M. Bone tumors : Diagnosis and treatment. Lippincott, Philadelphia, 1980, pp. 462-477.
8. Mirra J. M., Bernard G. W., Bullough P. G., Johnston W., Mink G. Cementum like bone production in solitary bone cysts. Clin. Orthop. 1978, 135, 295-307.
9. Neer C. S., Francis K. C., Marcove R. C., Terz J., Carbonara P. N. Treatment of unicameral bone cyst. J. Bone Joint Surg., 1966, 48-A, 731-745.
10. Scaglietti O., Marchetti P. G., Bartolozzi P. The effects of methylprednisolone acetate in the treatment of bone cysts. J. Bone Joint Surg., 1979, 61-B, 200-204.
11. Scaglietti O., Marchetti P. S., Bartolozzi P. 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., 1982, 165, 33-42.
12. Siegel I. M. Brisement force with controlled collapse in treatment of solitary unicameral bone cyst. Arch. Surg., 1966, 92, 109-114.
13. Spence K. F., Sell K. W., Brown R. H. Solitary bone cyst : treatment with freeze-dried cancellous bone allograft. J. Bone Joint Surg., 1969, 51-A, 87-96.
14. Zenker H., Stürz H. Die juvenile Knochenzyste. Behandlungsvorschläge und neue Ergebnisse. Z. Kinderchir., 1983, 38, 48-56.

SAMENVATTING

M. GEBHART, P. BLAIMONT. Bijdrage tot de vasculaire origine van solitaire botcyste.

Een vasculaire occlusie tussen een botcyste en het intramedullair veneus systeem kan de origine van de solitaire botcyste zijn. Injectie van fysiologisch serum onder hoge druk en alternatieve aspiratie-injectie van de inhoud van de cyste alsmede uitboren van de medulla openen vasculaire kanalen tussen de cyste en het intramedullair veneus systeem. Dit werd gevolgd met roentgen opnamen vóór en ná behandeling. Deze techniek werd gebruikt bij 12 patiënten met een solitaire botcyste. Elf op twaalf patiënten genazen met toename van de diameter van de cortex en vulling van de cystische holte.

RÉSUMÉ

M. GEBHART, P. BLAIMONT. Contribution à l'étude de l'origine vasculaire des kystes osseux solitaires.

Une obstruction vasculaire entre le kyste osseux et le système veineux intramédullaire semble être à l'origine de la formation d'un kyste osseux solitaire. Des injections sous pression d'une solution physiologique et des manipulations d'injection et d'aspiration alternatives, de même que l'alésage du canal médullaire ouvrent des canaux vasculaires mettant en communication la cavité kystique et le système veineux intramédullaire. Ceci a été démontré par des injections de produit de contraste dans la cavité kystique avant et après les manipulations. Cette technique a été utilisée chez 12 patients présentant un kyste osseux solitaire. Tous les patients sauf un ont totalement ou partiellement été guéris, avec augmentation de l'épaisseur de l'os cortical et comblement de la cavité kystique par de l'os spongieux.