Percutaneous Reaming of Simple Bone Cysts in Children Followed by Injection of Demineralized Bone Matrix and Autologous Bone Marrow

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Abstract: The authors report the successful treatment of 19 patients (mean age 10 years) with active unicameral bone cysts using a combination of percutaneous reaming and injection of a mixture of demineralized bone matrix and autologous bone marrow. Follow-up ranged from 12 to 42 months (mean 28 months). All patients were asymptomatic at the latest follow-up. Two required a second intervention to accomplish complete cyst healing. Radiographic outcome was improved in all patients according to the Neer classification at the latest follow-up. There were no significant complications related to the procedure, nor did any fracture occur after initiation of the above regimen.

Key Words: bone marrow, unicameral bone cyst, demineralized bone matrix

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Unicameral bone cysts (UBCs) are benign, fluid-filled, expansive lesions that tend to weaken the metaphyseal region of long bones in skeletally immature patients.[1] They are often first noted after a pathologic fracture that usually is nondisplaced and heals uneventfully with conservative treatment. Less commonly, they are diagnosed on a routine radiograph during a workup.[2] Once a UBC is diagnosed, the treating surgeon has to decide whether to treat or follow the patient. The answer is difficult because the natural history of a given cyst cannot be predicted. Various authors have attempted to determine the prognosis based on the patient's age, the site and size of the lesion, or the history of a previous fracture, but for any single cyst these factors are unreliable.[1,3–6]

The etiology of unicameral bone cysts remains unclear. Mira[2] considered them to be intraosseous synovial cysts based on electron microscopy findings. Jaffe and Lichtenstein[7] attributed them to trauma, while Cohen[8] argued that they form in response to venous congestion of the intramedullary space. The latter theory has been supported by others who were able to achieve cyst healing, restoring the venous circulation simply by performing multiple perforations.[5,9–12]

Various treatment modalities have been proposed, ranging from subtotal resection to normal saline injection. The reported results are puzzling, since the aggressiveness of the lesions treated is rarely defined.[9,10,13–20]

Scaglietti[15] described a methylprednisolone injection technique, reporting a 15% to 88% recurrence rate after an average of three injections. Superior results were reported using an osteoinductive preparation consisting of demineralized bone matrix (DBM) and autologous bone marrow.[13,14,21]

The purpose of this study was to evaluate the effectiveness of percutaneous reaming and injection of a mixture of DBM and autologous bone marrow for the treatment of unicameral bone cysts with or without a pathologic fracture.

METHODS

Between November 1999 and July 2002 we treated 19 patients with active unicameral bone cysts. Six patients were female and 13 were male, with a mean age of 10 years (range 4–16 years).The lesion was diagnosed secondary to a pathologic fracture in 14 patients (73.6%); in the other 5 patients (26.4%) the diagnosis was made after radiographic workup for extremity pain related to sports or activities of daily living.

The femur was affected in four patients (21%). In two patients the cyst was located at the distal metaphysis; in the other two it was in the femoral neck. The proximal humerus was affected in 15 patients (79%), while all pathologic fractures occurred in patients with such a UBC.

Surgical intervention was undertaken at least 6 weeks after the fracture to achieve healing of the fracture site. In addition to the plain radiographs, all lesions had been studied before surgery with CT and MRI. All cysts were classified before surgery as active. A cyst was considered active if it was symptomatic with activities of daily living, had fractured once or more, had increased in size within an observation period of 3 to 6 months, or was abutting the physis. Plain radiographs were used to ascertain the aggressiveness of the lesion (epiphyseal extension or physeal abutting). CT was used to assess the thickness of the cortex in all of its perimeters and the concomitant risk for fracture. In every instance the cyst occupied at least 85% of the transverse plane, in concordance

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with Ahn et al.[22] The entry point was dictated by the CT study. The thinnest area was identified and subsequently used to open the entry point to avoid further postoperative iatrogenic weakening of the area that could lead either to the intraoperative use of hardware or to a new pathologic fracture in the immediate postoperative period. MRI was used to confirm the diagnosis and rule out other pathology.[23,24]

Surgical Technique

The cyst was initially approached with two percutaneously inserted Jamshidi needles under fluoroscopic guidance at each end of the lesion. Forceful aspiration followed and a cystogram was performed using a 50% solution of Iohexol (Omnipaque, Schepa) in normal saline. Cystography documented the presence of a cyst in all 19 patients (Fig. 1). When the lesion was not of a cystic nature, an open biopsy was performed and the patient was excluded from the study. One of the needles was used to broach the cortex with gentle revolving maneuvers creating an entry point. The needle was withdrawn and a titanium elastic nail (ECMES, DePuy Inc, Leeds, UK) attached to a cannulated T-handle was percutaneously introduced into the cyst (Fig. 2A). Under fluoroscopic control and with gentle sweeping movements of the flattened nail tip, the wall of the cyst was scraped circumferentially in an orderly fashion, taking care to avoid cortex penetration (see Fig. 2B). The nail was then sequentially advanced to the very ends of the cyst, where with twisting maneuvers the medullary canal continuity was satisfactorily restored. If the cyst was abutting the growth plate, that process was performed only toward the diaphyseal end of the medullary canal. At this stage arthroscopic ring curets and rasps were appropriately used to achieve adequate cyst reaming. All instruments were sequentially introduced percutaneously though the same entry hole under fluoroscopic control.

After thorough cyst reaming, the cyst was washed out and the mixture of DBM (Allomatrix, Wright Medical, Arlington, TN) and autologous bone marrow was prepared. Bone marrow was aspirated in 5-mL increments per aspiration site following the principles set by Yandow et al[20] to maximize the concentration of bone progenitor cells in the aspirate. The second needle was withdrawn and a 2.5-mm trocar was introduced through the working entry point for injecting the mixture. The mean volume of the injected mixture was 11 mL (range 5–19 mL). A compression dressing was then applied to the surgical wound. A "collar and cuff" was prescribed for upper extremity lesions; for those of the lower extremity, protected weight bearing with crutches was prescribed as tolerated, gradually advancing to full weight bearing. No metallic implant was used for stability in any patient. One skeletally mature 16-year-old patient with an extensive proximal lesion of the humerus was stabilized with two femoral strut allografts inserted after antegrade reaming through the greater tuberosity, and subsequently the cyst was injected as above (Fig. 3). All patients were discharged the same day.

FIGURE 1. Cystogram shows gradual filling of the cystic cavity.

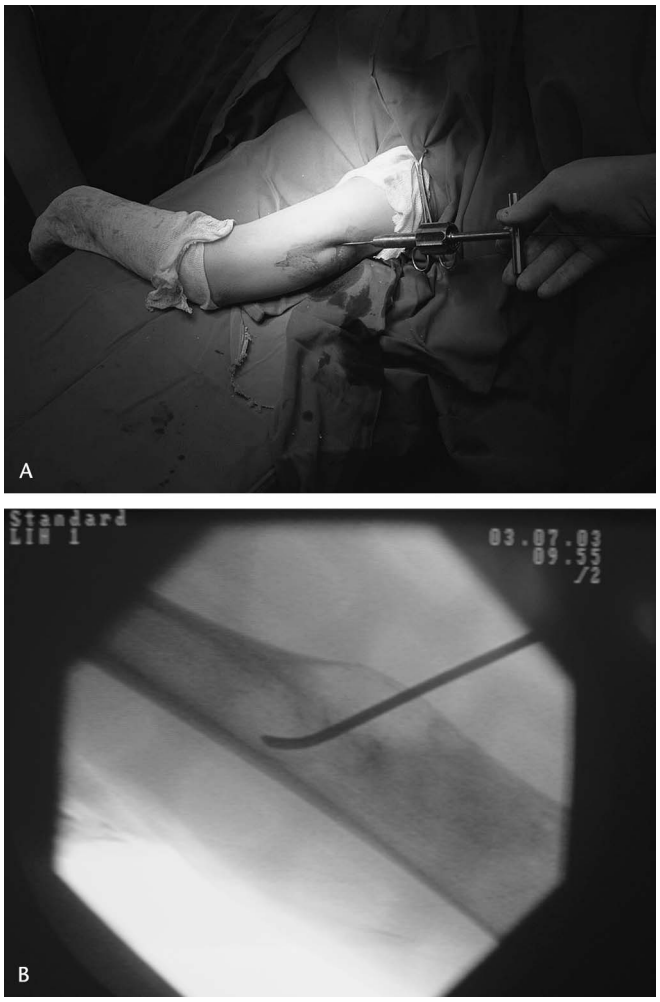

FIGURE 2. A, A titanium elastic nail attached to a cannulated T-handle is introduced into the cyst through the widened hole percutaneously. B, Intraoperative radiograph during percutaneous scraping of the cyst wall using a flexible titanium nail.

RESULTS

No intraoperative or perioperative complication occurred, nor did any fracture occur after surgery. Radiographs

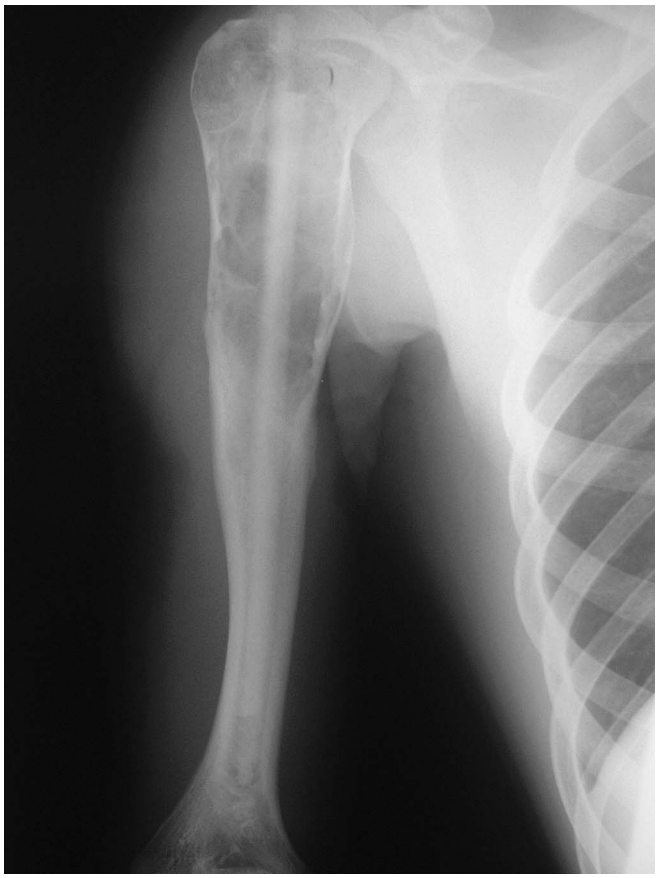

FIGURE 3. A 16-year-old patient with a proximal humerus UBC that underwent additional closed antegrade stabilization with strut grafts.

were obtained 3, 6, and 12 months after surgery and then on an annual basis until skeletal maturity was reached. Mean follow-up was 28 months (range 12–42 months). Six patients had reached skeletal maturity at that time. They were graded by a radiologist according to the Neer classification (Table 1).[25] The patients were examined for adjacent joint range of motion and were asked about pain with sports or activities of daily living. All cysts were graded as Neer stage I or II. All patients were asymptomatic within 6 weeks after surgery.

Two patients required a repeat procedure at 12 and 16 weeks after surgery since it was felt that inadequate reaming was performed at one end of the lesion, which was possibly recurring at that point (Fig. 4). Both patients were asymptomatic and the lesions were located at the proximal and

TABLE 1. Neer Classification

Stage	
I	100% cyst filling with new bone
II	Partial cyst filling, increased cortical thickness; osteolytic foci less than 3 cm
III	Recurrence; cortical thinning, osteolytic areas >3 cm
IV	No response to treatment

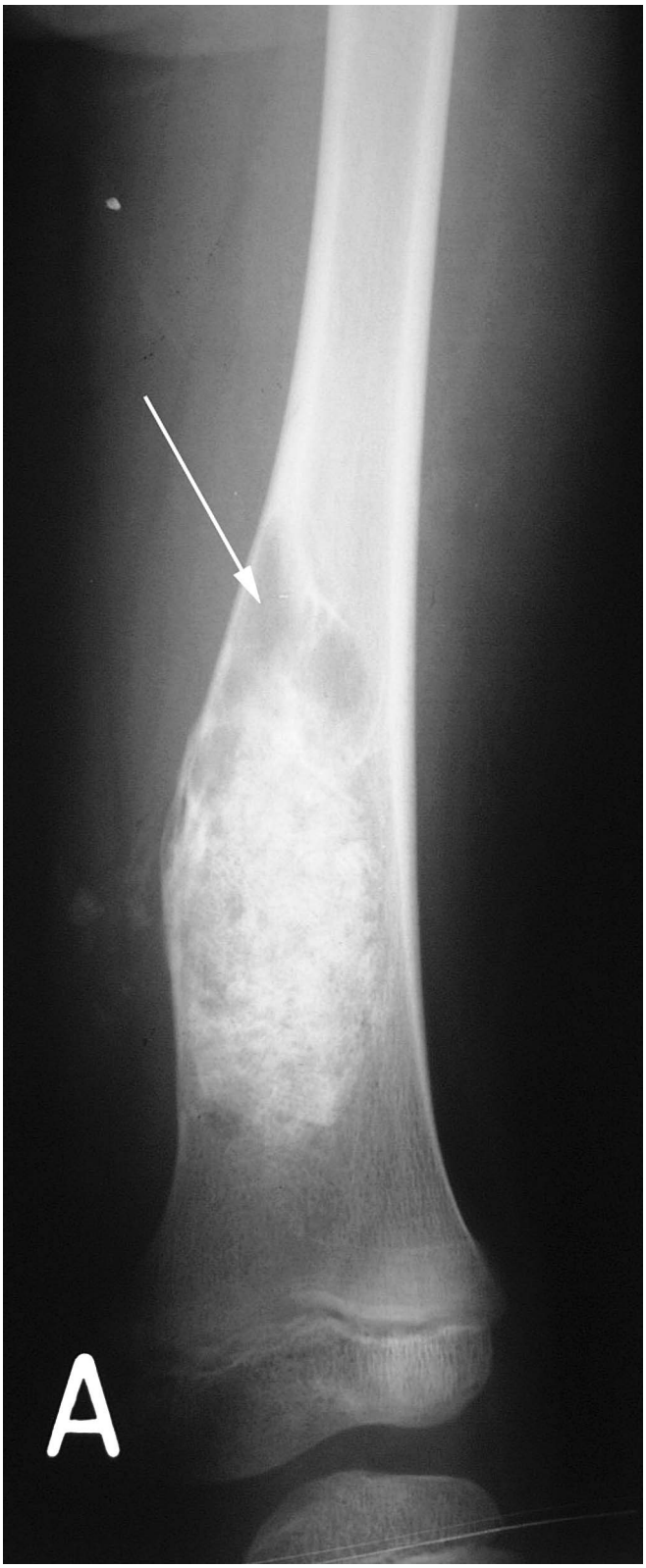

FIGURE 4. Large lesion in the distal femur of a 4.5-year-old boy, 8 weeks after percutaneous reaming and grafting with bone marrow and DBM. Possible recurrence at the proximal edge (arrow). Repeat procedure led to cyst involution.

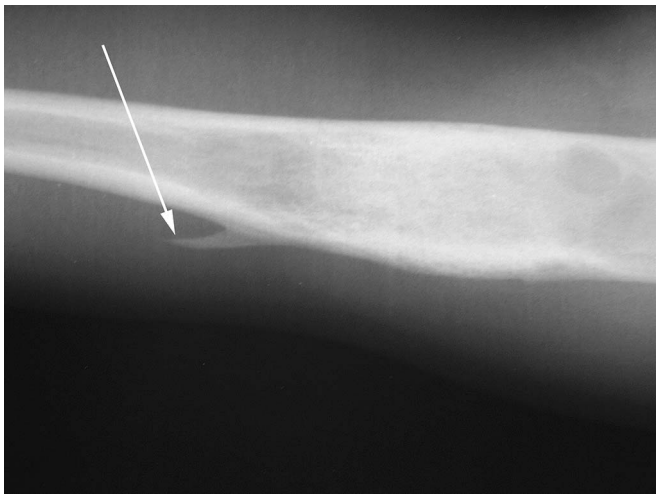

FIGURE 5. Radiographic aberration at the working entry point to the cyst, resembling an exostosis (arrow). All patients were asymptomatic. This finding can be attributed to spillage from excessive mixture that leaked after the trocar was withdrawn.

distal end of the femur. Both cysts were initially large lesions, occupying almost 100% of the cross-sectional area. The possible recurrence site was always located at the non-curetted edge of the cyst and its diameter was less than 1 cm while the rest of the cyst was radiographically healing. After the second intervention, the lesions resolved.

Six patients developed a radiographic aberration at the working entry point to the cyst, resembling an exostosis; it was observed. All six patients were asymptomatic even to deep palpation. This finding can be attributed to leakage of the mixture upon trocar withdrawal (Fig. 5).

DISCUSSION

The etiology of UBCs is unclear, but two main theories related to their emergence have been proposed: the mechanical theory (secondary to intramedullary venous congestion) and the biologic theory (due to bone absorption).[1,4,11,26–28] This study attempted to assess the efficacy of a comprehensive treatment plan that addresses both groups of etiologic factors. To accommodate the biologic pathogenetic factors, a mixture of autologous bone marrow with DBM was injected in a mechanically reamed and perforated cyst to decrease the intramedullary pressure.

UBCs are uniformly characterized by a thick plug of cortical bone at each end obstructing the medullary canal. According to Cohen,[8] this mechanical factor is related to the pathogenesis of the cyst by blocking venous channels of outflow. This pathogenetic theory was supported by studies that reported success rates of up to 97.4%[9] simply by breaching the cortical intramedullary plug. Some authors performed that by implanting flexible or rigid intramedullary nails,[10] while others reported an 8.33% recurrence rate just by reaming the canal.[11] On the other hand, the combination of bone marrow and DBM led to involution of 23 bone cysts followed for 50 months.[14] Killian et al[13] reported complete cyst obliteration after percutaneous injection of DBM in 9 of 11 patients. In contrast to these data, Hashemi-Nezad[29] reported the rate of successful outcomes of intralesional steroid injection to be as low as 13% 5 years after surgery.

Our study had an overall success rate of 89.5% (17/19) with the first attempt that reached 100% with a second intervention. The number of patients in our study was relatively small, but the results were uniformly good. In the literature, when only one of those factors was addressed, the results of treatment were good (Fig. 6). Our approach addresses both biologic and mechanical etiologic factors, so it is impossible to ascertain the effect of each separate factor on each given cyst. It is merely a confirmation of a rather logical guess that since two factors had a strong curative effect upon UBCs when acting separately, the combined treatment should yield an even more favorable outcome. Our study included only active cysts in younger patients (mean age 10 years), which are known to be aggressive and resistant to treatment, especially with one attempt.[8,22]

The decision to pursue surgical intervention in patients with UBCs should be highly individualized. Initially, the aggressiveness of the cyst needs to be established. Painful

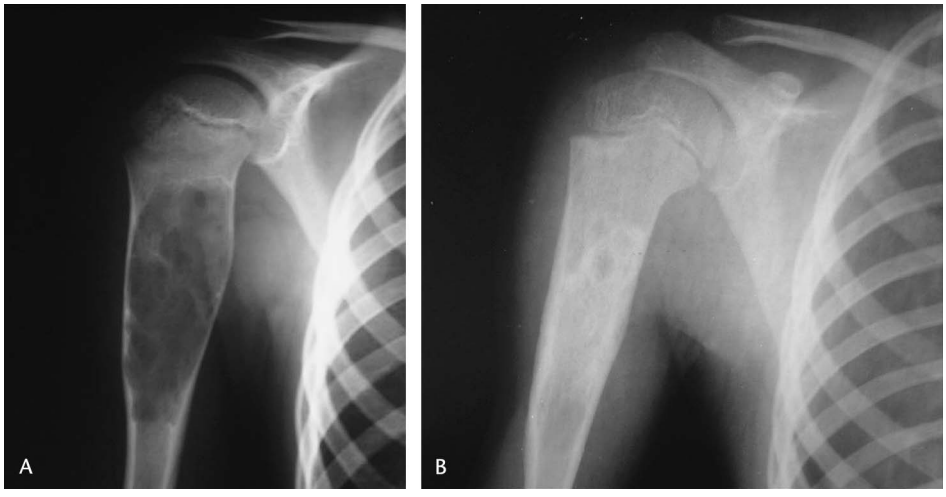

FIGURE 6. A, Admission radiograph of a 4.5-year-old boy. B, Same patient, 23 months after surgery.

cysts and cysts that fractured at least once as well, as well as those located at the neck of the femur, even in asymptomatic patients, should be treated operatively.

In conclusion, in this study we described a percutaneously applied technique that offers the merits of low morbidity with no hospital stay and produced complete healing in active and rather aggressive cysts. The postoperative morbidity was low, patient comfort was enhanced, and the hospital stay was minimal. This method attempts to single-handedly eliminate both the mechanical and biologic causative factors of UBCs.

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