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Treatment of pathological fractures due to simple bone cysts by extended curettage grafting and intramedullary decompression

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Objective: Effectiveness and morbidity of curettage grafting and intramedullary decompression in the treatment of pathological fractures due to simple bone cysts (SBCs) were evaluated.

Methods: Between 2005 and 2012, 34 children with SBCs were treated with extended curettage grafting and intramedullary decompression. Average age of the patients (23 male, 11 female) at surgery was 11.7 years (range: 6–21 years). The lesions were localized in the humerus (19), femur (12), tibia (2), and ulna (1). Pathological micro- or displaced fractures occurred in 31 patients. Surgical procedure included extended curettage by using cauterization and high-speed burring, bone grafting, and intramedullary decompression with elastic nails. Six patients had been treated conservatively or surgically in other institutions previously. Radiographic and functional results were evaluated by Capanna criteria and MSTS scores respectively. Early and late complications and additional surgical procedures were recorded.

Results: The average follow-up was 37 months (range: 18–89 months). The average time to heal for pathological fractures was 8 weeks (range: 6–12 weeks). Radiographic evaluation revealed Grade 1 healing in 28 patients (82%) and Grade 2 healing in 6 patients (18%). The average MSTS score based on final follow-up was 28.5 (range: 17–30); excellent (32 patients; 94%) and good (2 patients; 6%) functional results were obtained. There was no early or late infection, refracture or implant failure. Malunion developed following treatment of 2 humeral and 2 femoral lesions. With the exception of 2 implant removals, no patients required additional surgical intervention.

Conclusion: Complete cyst healing and satisfactory functional results can be obtained by curettage grafting and intramedullary decompression. This technique restores bone integrity by allowing early motion and prevents refracture and subsequent deformity in the majority of patients

Keywords: Curettage and grafting; intramedullary decompression; intramedullary nailing; simple bone cyst.

Simple bone cysts (SBCs), defined by Virchow in 1876, are benign fluid-filled lesions. They typically are located in the metaphysis of long bones in children and adolescents.[1] The proximal humerus and femur are the most commonly involved regions, followed by calcaneus and ilium.[2,3]

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The pathogenesis of SBCs is still unclear. The most commonly accepted hypotheses include abnormal growth due to damage of the growth plate and increase in intraosseous pressure secondary to venous obstruction.[4–6] Prostaglandin (PG) E2 and interleukin (IL) 1-B enzymes within the cyst fluid may also cause bone destruction.[7]

There is no standard approach in the treatment of SBCs. The main objectives include prevention of pathological fracture, eradication of the lesion, and pain relief. Even though various treatment methods have been developed based on the accepted hypotheses for pathogenesis, none of them demonstrated a significant advantage over the others.[8]

Curettage grafting and intracystic steroid injections are the conventional methods used in the treatment of SBCs. Curettage grafting has been widely used with high rates of partial healing and recurrence.[9–11] Total or subtotal resection of the cyst can provide more favorable results; however, these aggressive surgical procedures are associated with high morbidity and intraoperative complications.[12–14] Intracystic steroid injection has been accepted as a standard method due to the simplicity of application and low rate of complications.[15] The high rate of partial healing, which can be achieved by repeated injections, is the principal disadvantage of steroid injections. Recently, bone marrow, demineralized bone matrix (DBM), calcium sulphate pellets, and fibrin injections have been used with varied results.[16–25] Even though short-term results are promising, these new techniques are associated with high rates of partial healing and healing with residual lesion.

The high rates of partial healing obtained by conventional and new methods catalyzed the development of percutaneous surgical techniques. However, healing with residual cyst must be recognized as a disadvantage of percutaneous techniques.[24,26–28]

Intramedullary decompression is a breakthrough in the treatment of SBCs. By providing continuous drainage between the medullary canal and the cyst cavity, intracystic pressure can be reduced and increased healing rates can be obtained. Elastic intramedullary nailing has been frequently used in the treatment of SBCs.[29–32] Elastic nails reduce intracystic pressure by intramedullary decompression, stimulate healing, and allow early postoperative motion by providing immediate stability, particularly in patients with pathological fractures.[33–38]

The conventional and new methods used in treatment of SBCs are frequently associated with partial healing, healing with residual lesion, and recurrence. In addition, there is still debate as to whether it is more advantageous to provide acute stability by internal fixation or to wait for fracture healing in order to eliminate the need for stabilization in cases with pathological fractures.

In the current study, we aimed to assess the effectiveness of a combined method including extended curettage grafting and continuous intramedullary decompression in the treatment of SBCs.

Patients and methods

Between 2005 and 2012, 34 patients with SBC were treated with extended curettage grafting and intramedullary decompression with titanium elastic nails. Patients with a minimum follow-up of 18 months were included. Data was obtained from our extensive orthopedic oncology files which include clinical and radiological evaluations (conventional radiography, magnetic resonance imaging [MRI], and whole body bone scintigraphy) at presentation and during follow-up, and operative and pathology reports. Data was collected on age and gender of the patients, localization of the lesions, radiological and histopathological findings, radiographic stage, duration of symptoms, presence or absence of pathologic fracture at presentation, previously-applied treatment modalities, surgical procedure, clinical and radiographic results at final follow-up, and complications.

The demographic data, radiographic staging, and details of surgical treatment are provided in Table 1. There were 23 males and 11 females included in this study. The average age at surgery was 11.7 years (range: 6–21 years). The lesions were located in the metaphyseal or metaphyseal-diaphyseal regions of the humerus (n=19; 56%), femur (n=12; 35%), tibia (n=2; 6%), and ulna (n=1; 3%). The majority of the humeral and femoral cysts were localized in the proximal metaphysis and frequently extended to diaphysis. Isolated diaphyseal involvement was occasionally observed in the humerus and ulna. Radiographic examination usually revealed a lytic lesion with mild expansion and thinning of the cortex. MRI indicated homogeneous hypointense and isointense unicameral cysts on T1- and T2-weighted sequences, respectively. However, the lesions with previous pathological fractures frequently had a multiloculated appearance resembling an aneurysmal bone cyst. The cysts were classified as Stage 1 (latent; 3 cysts; 9%), Stage 2 (active; 30 cysts; 88%), and Stage 3 (aggressive; 1 cyst; 3%).

Seven patients in this series required a biopsy procedure prior to definitive treatment. Since the preoperative imaging studies supported the diagnosis of SBC, single-stage surgery was possible in the remaining 27 patients.

Table 1. Demographic data, staging, surgical treatment, radiographic and clinical follow-up results and complications.

No	Age	Gender	Location	Stage	Pathological fracture at presentation	Treatment prior to C+G+EIMN	Surgical treatment	Follow-up (months)	Radiographic healing*	Clinical results (MSTS score)	Complications
1	17	Male	Humerus	Active	+		C+G+EIMN	25	Grade 1	30	
2	7	Male	Femur	Active	+		C+G+EIMN	21	Grade 1	29	
3	15	Male	Femur	Active	+		C+G+EIMN	44	Grade 1	30	
4	18	Female	Humerus	Active	+		C+G+EIMN	32	Grade 1	30	
5	17	Male	Humerus	Active	+		C+G+EIMN	20	Grade 1	30	
6	16	Female	Tibia	Active	+		C+G+EIMN	24	Grade 1	27	
7	10	Male	Femur	Active	+		C+G+EIMN	39	Grade 1	30	
8	13	Male	Humerus	Active	+		C+G+EIMN	42	Grade 1	30	15’ varus malunion
9	16	Male	Humerus	Latent	–		C+G+EIMN	24	Grade 1	28	
10	9	Female	Humerus	Active	+		C+G+EIMN	36	Grade 1	30	
11	14	Male	Humerus	Active	+	Cast/splint	C+G+EIMN	34	Grade 1	28	
12	11	Male	Humerus	Active	+		C+G+EIMN	35	Grade 1	29	
13	7	Female	Tibia	Latent	–		C+G+EIMN	31	Grade 2	24	
14	7	Male	Femur	Active	+		C+G+EIMN	22	Grade 1	28	
15	11	Male	Femur	Active	+		C+G+EIMN	30	Grade 1	30	
16	21	Female	Humerus	Active	+		C+G+EIMN	31	Grade 2	27	
17	14	Female	Humerus	Active	+	Cast/splint	C+G+EIMN	26	Grade 1	28	
18	6	Female	Femur	Active	+		C+G+EIMN	36	Grade 1	28	
19	13	Female	Femur	Aggressive	+		C+G+EIMN	24	Grade 1	30	
20	12	Male	Femur	Active	+		C+G+EIMN	22	Grade 2	17	20’ varus malunion
21	17	Male	Humerus	Active	+		C+G+EIMN	18	Grade 1	29	
22	7	Male	Humerus	Active	+	Steroid injection	C+G+EIMN	39	Grade 1	30	
23	8	Male	Femur	Active	+	C+G	C+G+EIMN	48	Grade 1	21	10’ varus malunion
24	9	Male	Humerus	Active	+		C+G+EIMN	36	Grade 1	29	
25	9	Male	Humerus	Active	+		C+G+EIMN	29	Grade 1	30	
26	7	Female	Humerus	Active	+	Cast/splint	C+G+EIMN	47	Grade 2	29	
27	14	Male	Humerus	Active	+		C+G+EIMN	62	Grade 1	30	
28	16	Female	Humerus	Latent	–		C+G+EIMN	34	Grade 2	30	
29	11	Male	Humerus	Active	+		C+G+EIMN	68	Grade 1	28	
30	14	Male	Humerus	Active	+	Cast/splint	C+G+EIMN	50	Grade 2	29	10’ varus malunion
31	9	Female	Femur	Active	+		C+G+EIMN	89	Grade 1	30	
32	8	Male	Femur	Active	+		C+G+EIMN	43	Grade 1	30	
33	7	Male	Ulna	Active	+		C+G+EIMN	32	Grade 1	30	
34	9	Male	Femur	Active	+		C+G+EIMN	60	Grade 1	30	

C+G: Curettage and grafting; EIMN: elastic intramedullary nailing.
*Radiographic healing due to Capanna criteria (clinical and radiographic results reflect the last follow-up evaluations of the patients).

Histopathological examination revealed a single layer of endothelial cells which lined the inner side of the cyst wall, confirming the diagnosis of SBC.

Patients were admitted directly to our institution or referred from other institutions due to pathological fracture, pain, and progression of the lesions. A pathological fracture occurred in 31 patients. Except for 3 displaced fractures associated with 2 femoral and 1 humeral cysts, microfractures were observed. Two patients had been previously treated by open curettage grafting or intracystic steroid injections in other institutions. In addition, 4 patients had been managed by conservative treatment for pathological fractures. Partial healing, recurrence, progression of the lesion, and pathological fracture were the indications for resurgery in these patients. The patients were usually operated on within 1 week of their admission (range: 3 days–3 weeks).

The surgical procedure was begun as an incisional biopsy in 27 patients with no previous biopsy procedure. A small window was opened on the cyst wall, and the cyst fluid was aspirated. Tissue samples were obtained from the cyst content or inner side of the cyst wall for frozen section. After malignancy was ruled out by frozen section, the surgical procedure was continued. Patients underwent extended intralesional curettage grafting followed by elastic intramedullary nailing. Following curettage, the surgical margins were extended by high-speed burring and cauterization. A deltopectoral approach was used for proximal humeral lesions, and the incision was extended to the lateral arm if the lesion involved the diaphysis. Proximal femur and tibia were approached by lateral and anterior longitudinal incisions, respectively.

The ulnar cyst was approached by longitudinal incision over the lesion.

Initially, a large cortical window was created in the thinnest area of the cortical shell, and a through curettage was performed. The curettage material was sent for pathological examination. Gross appearance varied from a clear yellow fluid to a hemorrhagic fluid. The lesions with previous fractures had a septated and multicameral appearance. Cauterization and high-speed burring of the interiors of the cyst wall were performed several times in order to extend the surgical margins beyond the reactive zone of the lesion. Since most of the lesions were accompanied by pathological fracture, adequate care was taken when using the electrocautery and high-speed burr to avoid soft tissue damage. In patients with displaced fractures, a gross reduction of the fragments was obtained first, and the described procedures were then performed again through a cortical window, crossing the fragments. The cyst defect was washed off by pulsatile lavage to clean debris. Local adjuvants such as liquid nitrogen or phenol were not used in any case.

The resultant defect was filled with cancellous allograft (allograft cancellous cubes), cancellous autograft (morselized iliac crest), a combination of both, and a combination of cancellous graft and structural fibular allograft. Fibular allograft was used in 4 patients with a large metaphyseal-diaphyseal lesion. The average amount of cancellous graft (autograft, allograft, or combination) was 60 cc (range: 30–150cc).

Elastic intramedullary nails were inserted retrograde through oval windows opened on the lateral cortex of the distal femur, posterior cortex of the distal humerus, and anterior cortex of the distal tibia. Intramedullary fixation and decompression of the ulnar lesion were performed via an antegrade nail inserted percutaneously through the olecranon. Based on the affected bone and size of the lesion, 2–4 elastic nails (2.0–3.5 mm in thickness) were used. One nail was sufficient for the ulnar lesion. Intramedullary nails were inserted under fluoroscopic guidance; in patients with an open growth plate, the nails did not pass the physeal line.

Patients with humeral lesions required a sling postoperatively. Active wrist and finger movements and passive elbow motion were allowed in the early postoperative period. Active elbow and passive shoulder movements were started at 2 and 4 weeks, respectively, postoperatively and gradually increased. Patients with femoral lesions were followed by a walking spica cast with toe-touch weight bearing for 4–6 weeks. Range of motion (ROM) exercises were initiated, and weight bearing was gradually increased. Full weight bearing with no external support was achieved at the end of the 3rd month. A similar protocol was followed for the tibial lesions by using a long leg splint.

Patients were evaluated radiographically and clinically at 3, 6, 12, and 24 months, as well as at final follow-up. Capanna criteria were used to determine radiographic cyst healing. According to these criteria, in Grade 1 healing, the cystic cavity is completely filled with bone, and the cortical edges thicken; in Grade 2 healing, even though a significant consolidation is observed, small residual cystic areas remain; in Grade 3 healing, a large residual cyst is present; and in Grade 4 healing, there is no response to treatment.[39]

The functional scoring system of the Musculoskeletal Tumor Society (MSTS) was used to evaluate functional results.[40] By this scoring system, pain, functional capability, and emotional status of the patients were evaluated. A total of 6 parameters were considered: walking distance, gait pattern, use of support for lower extremity lesions, hand position, hand skills, and weight lifting for upper extremity lesions. Even though the MSTS scores were measured separately for upper and lower extremity lesions, the average score of the whole study group was taken into consideration. The results were rated as perfect for 23 and above, good for 15–22, moderate for 8–14, and bad for 8 and below.

Early or late postoperative complications including superficial or deep infection, wound problem, loss of reduction, nonunion, refracture, failure of metallic implant, partial or complete injury to growth plate (secondary to surgical procedure or penetration and/or irritation of elastic nails), deformity, and limb length discrepancy were recorded. The need for additional surgical intervention for early or late complications was investigated.

Results

Average follow-up was 37 months (range: 18–89 months). Radiographic and clinical (functional) results for each patient at final follow-up are given in Table 1. All pathological fractures healed uneventfully in 8 weeks on average (range: 6–12 weeks). According to final follow-up radiographs, Grade 1 and Grade 2 healing were achieved in 28 patients (82%) and 6 patients (18%), respectively. (Figure 1a–f, Figure 2a–f) All cases in this series responded well to treatment and none experienced recurrence. Periodic follow-up radiographs revealed that the healing process started at 3 months and was completed at 12 months in the majority of patients. The healing process continued for between 12–24 months in a minority of patients.

Fig. 1. A humeral simple bone cyst with a pathological fracture in a 9-year-old girl **(a, b)**. Postoperative early **(c, d)** and 36 months **(e, f)** follow-up radiographs of the patient. Grade 1 healing was achieved according to Capanna criteria.

Average MSTS score based on final follow-up of patients was 28.5 (range: 17–30). The MSTS scores were perfect in 32 patients (94%) and good in 2 patients (6%). No patients in this series produced moderate or bad MSTS scores. Functional improvement of patients increased progressively until the end of the 12th month.

No patients experienced superficial or deep infection, wound problem, or loss of reduction in the early postoperative period. Physeal damage and subsequent deformity or limb length discrepancy were not observed. Malunion developed after surgical treatment in 2 humeral and 2 femoral lesions (Table 1). Since 10–15° of malalignment did not lead to any functional disability, proximal humeral angular deformities did not require additional surgical intervention. A varus deformity of 10° and 20° developed following surgical treatment of 1 intertrochanteric and 1 subtrochanteric lesion, respectively. A significant clinical problem was not observed in the 1st case. The 2nd case showed slight limping and shortening due to varus malalignment. At 30-month follow-up, this 12-year-old child compensated for limb length discrepancy by using a shoe lift. The patient remains under close monitorization for possible corrective osteotomy.

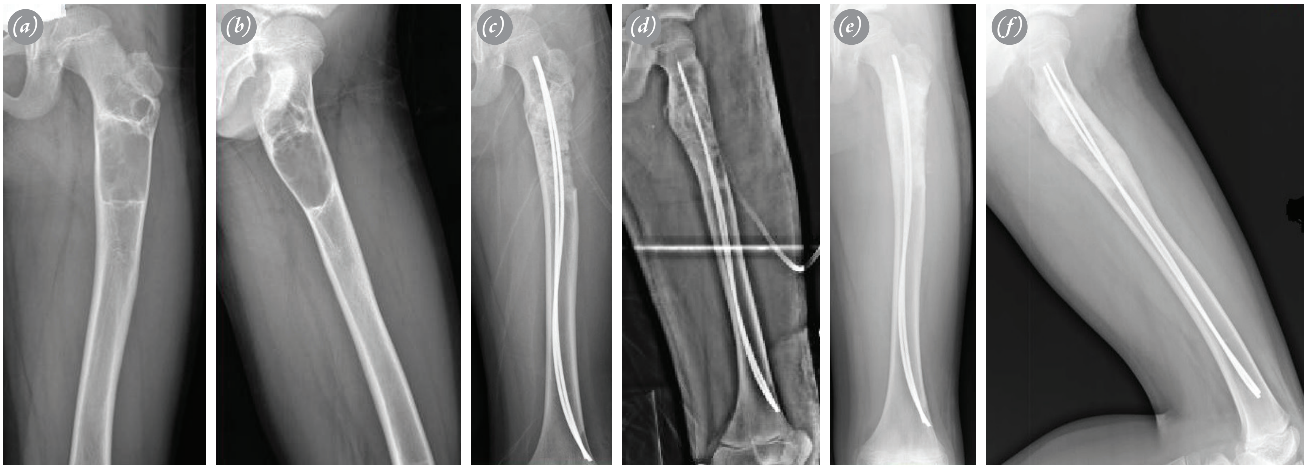

Fig. 2. A femoral simple bone cyst in an 11-year-old boy. Preoperative **(a, b)**, postoperative 1 month **(c, d)**, and 30 months **(e, f)** follow-up radiographs. Grade 1 healing was achieved according to the Capanna criteria.

Elastic intramedullary nails failed in no patients. Except for removal of intramedullary nails in 2 skeletally mature children with long-term follow-up, no patients required additional surgical intervention.

Discussion

Surgical treatment of SBCs should prevent pathological fracture and subsequent deformity while eradicating the lesion sufficiently to preclude recurrence. Conventional methods including open curettage grafting and intracystic corticosteroid injections are associated with high rates of partial healing, recurrence, and surgical morbidity.[9–14] Healing with partial opacification of the cyst cavity is also seen in relatively new techniques such as percutaneous intramedullary decompression and bone marrow or DBM injections.[22–28] The mixed results of isolated or combined use of bone marrow and DBM injections prevent the use of these techniques in the standard treatment of SBCs.

Cleaning the cyst membrane, from which the cyst fluid and destructive enzymes originate, is essential to eradicate SBCs. Intracystic corticosteroid injections can prevent bone destruction by affecting PG E2; however, corticosteroids do not penetrate the cyst membrane.[41] Bone marrow and DBM injections have osteogenic potential and ensure cyst healing by stimulating bone production but have no influence on the cyst membrane.[16–18]

Percutaneous or open mechanical destruction of the membrane covering the inner wall of the cyst is believed to produce a more permanent solution. Canavese et al. reported that destruction of the cyst membrane will contribute to the healing process, and this technique may be preferred to intralesional injections.[19] Likewise, Sakamato emphasized the positive effect of curettage on bone consolidation.[42] While high rates of partial healing and reoperation have been reported following open curettage grafting,[10,43] this technique is still widely used.[22,24,28]

Intramedullary decompression has been used effectively in the surgical treatment of SBCs. Chigara et al. demonstrated high intracystic pressure and suggested venous obstruction as an etiological factor in the formation of SBCs. The authors achieved perfect results in 7 patients who underwent decompression by multiple drilling.[6] Cannulated screws were placed in the cyst cavity to provide continuous drainage and decompression.[24,44] Compared to conventional methods, better results were achieved in these studies. Nonetheless, it has been stated that this method should not be preferred for lesions in weight bearing long bones, particularly if they are associated with a pathological fracture.

In general, positive results have been obtained by intramedullary decompression of SBCs. Dormans et al. achieved complete healing in 22 of 24 patients (91.7%) who underwent percutaneous intramedullary decompression, curettage, and injection of calcium sulphate pellets.[20] On the other hand, the need for reoperation may arise in patients who undergo percutaneous surgery. In another study reported from the same institution, Mik et al. reported 55 patients who were managed by percutaneous decompression and curettage grafting. In this series, 2nd and 3rd surgeries were required to achieve healing in 11 patients (20%) and 2 patients (2%), respectively.[26] Hunt et al. obtained healing in 20 humeral SBCs by percutaneous curettage grafting.[27] However, healing was possible following 2nd operation in 5 patients (25%) and 3rd operation in 1 patient (4%).

Many studies support the use of elastic intramedullary nails in the treatment of SBCs.[33–38] Bumci et al. suggested that a connection between the cyst cavity and the medullary canal alters the microcirculation and stimulates bone formation.[29] Givon et al. reported that intramedullary nails can decompress the lesion by providing a channel between the cyst and the medullary canal without creating a large defect on the cyst wall.[30]

Roposch et al. stated that elastic intramedullary nails can destruct the cyst membrane and provide continuous drainage of the cyst cavity in addition to permanent stability.[33] The authors performed intramedullary nailing in the management of 32 SBCs with acute pathological fractures. Complete healing, healing with residual lesion, and recurrence rates were 44% (14 patients), 50% (16 patients), and 6% (2 patients), respectively. The same author, in another study, applied retrograde elastic intramedullary nails to 11 proximal femoral cysts and achieved complete healing in 2 patients (18%) and healing with residual lesion in remaining 9 patients (82%).[45] In a series of 47 patients managed with only intramedullary nails, De Sanctis et al. obtained complete healing in 31 patients (66%) and healing with residual lesion in 16 patients (34%).[34] No patients in this series experienced recurrence. Kokavec et al. compared various treatment methods used in the management of SBCs and achieved healing with residual lesion in 2 of 5 patients (40%) who underwent intramedullary nailing.[22]

The decompression effect of intramedullary nails contributes positively to cyst healing. Despite the fact that good results have been generally reported, a high rate of healing with residual lesion is a significant disadvantage for the isolated use of intramedullary decompression. In the current study, by combining intramedullary nailing and curettage grafting, complete and partial radiographic healing was achieved in 28 patients (82%) and 6 patients (18%), respectively. No recurrence was observed, and no patients required additional surgical intervention. When compared to the isolated use of intramedullary decompression,[33,34,45] similar functional results but lower rate of healing with residual lesion were achieved. Furthermore, in contrast to other percutaneous curettage grafting series in which a considerable number of reoperations were required to achieve high healing rates,[20,26,27] no patients in this series required additional intervention to eradicate the lesion. High healing rates were possible by combining the continuous intramedullary decompression effect of elastic intramedullary nails with the destructive effect of open curettage on the cyst membrane.

General opinion regarding treatment of pathological fractures due to SBCs involves fracture healing followed by management of the lesion. It is thought that the need for internal fixation may be reduced if interventions for eradication of the lesion are made following fracture union. This approach requires a long preoperative and postoperative immobilization period in children. Some studies in the literature state that the bone cyst and the pathological fracture can be treated simultaneously, especially in high-stress localizations.[46,47] Roposch and De Sanctis achieved high rates of cyst healing in children with pathologic fractures via acute intramedullary nailing.[33,34] In the current study, lesions with pathological fractures were acutely managed by surgical treatment without waiting for fracture healing. In addition to eradication of the lesion, acute intramedullary fixation contributed to early motion and mobilization, especially in the upper extremities.

The lack of a control group to compare the results of our combined surgical method was the primary disadvantage of the current study. Our relatively aggressive approach involving extended curettage grafting and intramedullary decompression was compared with the previous series in the literature, in which various surgical methods were used in terms of cyst healing and need for reoperation. The comparison of various methods in a single study would be more useful in order to determine a standard treatment approach for SBCs.

By using a combined treatment, we aimed to eliminate the disadvantages of isolated use of curettage grafting and intramedullary decompression. The bone cysts were eradicated by extended curettage through the large cortical window, and bone formation was stimulated by grafting. Elastic intramedullary nails contributed to cyst healing by providing continuous decompression of the cyst cavity. Additionally, they stabilized the fracture and allowed for early motion and mobilization. We believe that open curettage grafting and intramedullary nailing is effective in the treatment of SBCs, with high healing and low reoperation and complication rates.

Conflics of Interest: No conflicts declared.

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