Treatment of Simple Bone Cysts in Children with Curettage and Cryosurgery


Study conducted at Children's Hospital and Medical Center, Seattle, Washington, U.S.A.

Summary: A retrospective study in children with simple or unicameral bone cysts treated with curettage, cryosurgery, and bone grafting was conducted. The purpose of this study was to evaluate local tumor control and bony healing after this method of treatment. Five (12%) of 42 treated patients had a local recurrence with a mean clinical follow-up of 24.5 months. Surgical complications consisted of two superficial wound infections, one radial nerve palsy, and two fractures, which all resolved completely. A review of the literature was performed to compare our results with historic controls using steroid-injection therapy and curettage with bone grafting alone. We believe that the use of cryosurgery as adjuvant therapy in the surgical treatment of simple bone cysts is of value in controlling local recurrences and achieving bony consolidation.

Key Words: Bone grafting—Cryosurgery—Curettage—Simple bone cysts—Unicameral bone cyst.

Simple bone cyst is a tumor of bone of unknown origin. It tends to occur in the metaphyses of long bones, particularly of the humerus and femur. Although histologically completely benign, it frequently weakens the integrity of bone, resulting in pathologic fracture, which is often the presenting feature. The treatment options for simple bone cysts (SBCs) include observation, injection, and surgical curettage. Until Scaglietti et al. (30,31) introduced the technique of steroid injections, the most prevalent treatment method for SBC has been curettage followed by bone grafting, with recurrence rates that vary from 12 to 48% (3,4,14,24-28,34,35). Adjuvant therapy after curettage has been advocated to destroy residual tumor cells and reduce tumor recurrence. In theory adjuvant treatments include radiotherapy, cytotoxic physical poisons like phenol and hypertonic saline, thimerosal (Merthiolate), cement, and cryosurgery. The purpose of this study was to evaluate local tumor control and bony healing after the treatment of simple bone cysts with curettage, cryosurgery, and allograft bone grafting. A review of the literature is included, and our results are compared with historic controls.

MATERIALS AND METHODS

A retrospective study of all patients with SBCs treated in our institution was carried out, including a review of all patient records, radiographs, and pathology reports. The diagnosis of a SBC was made when membranous tissue was found within the cyst or when the cyst was found to contain no tissue at all. The cysts may or may not have contained serous or serosanguineous fluid. Cysts containing tissue consistent with aneurysmal bone cyst were excluded. Fifty-three patients were subsequently diagnosed with SBC and treated with curettage, cryosurgery, and bone grafting between 1989 and 1996. Eleven patients were treated in 1995 and excluded for follow-up of <12 months, leaving 42 patients in this study.

The following clinical parameters were assessed for each patient: age, sex, history and previous treatment, anatomic location, pathology results, postoperative complications, and function and length of follow-up. Radiographs were reviewed to determine anatomic location, presence of pathologic fracture, cyst size (greatest diameter), postoperative osseous healing, and the presence of local recurrence. Staging was accomplished by using the radiographic criteria for benign lesions of bone defined by Enneking (9).

The operative treatment of SBCs in our patient group consisted of intralesional curettage via bony fenestration followed by cryotherapy and bone grafting. Cryotherapy or cryosurgery was performed using an apparatus (Kryospray II; Brymill Corporation, Vernon, CT, U.S.A.) pro-
ducing a liquid nitrogen spray by which the osseous cavity was frozen and then thawed using a warm saline solution. Three cycles of freezing and thawing were carried out after a limited curettage executed by curettes and a mechanical burr. The bony defect was then filled with allogenic freeze-dried bone chips, which were procured and processed according to guidelines recommended by the American Association of Tissue Banks (2). Care was taken not to damage the adjacent physis by curettage, and if the physis was exposed to the cyst, it was separated from freezing by several layers of surgical absorbable gelatin sponge (Gelfoam). When possible, tourniquets were used. For all lower extremity lesions, postoperative full weightbearing was not allowed for 6–12 weeks. Follow-up was achieved by clinical examination and routine radiographs at 1- to 2-month intervals until bony healing. The results of our treatment were evaluated radiographically using the following classification terminology, modified from a system previously used by Neer et al. (4,26):

1. complete response: the space occupied by the cyst is completely filled with new bone formation with remodeling or consolidation of the bone graft;
2. partial response: small areas (<1.0 cm) of radiographic lucencies are seen within the boundaries of the previous cyst, which otherwise demonstrates complete bone formation and remodeling of the graft. With continued radiographic follow-up, no increase in size of these lucencies is recognized over time;
3. local recurrence: radiographic lucency, within or adjacent to the prior cyst that enlarges radiographically over time; and
4. no response: no radiographic evidence of bony healing after injection. This response is applicable only to patients who have been treated with steroid injection(s).

The treatment results of the historic controls collected from the literature also were reviewed and scored using this classification system. Statistical significance to identify patient-related factors affecting recurrence was determined using χ² analysis.

RESULTS

Our patient group consisted of 31 (74%) boys and 11 (26%) girls. Eighteen (43%) patients were younger than 11 years, and 24 (57%) were aged between 11 and 20 years (Table 1). The mean age at which the first operation was performed with cryosurgery was 11 years (range, 3–18). The anatomic distribution of the affected sites in our and published series is listed in Table 2. In our series, the humerus accounted for 71% of the cases. Previous treatment had been carried out in 16 (38%) patients involving one or more injections with steroids in 11 (26%) patients, curettage with bone grafting in four (10%) patients, and one (2%) patient had received both treatments. Pain was the presenting factor in six (14%) patients and a pathologic fracture in 33 (79%) patients, with multiple fractures in eight of those 33 patients. Patients with fractures all had radiographic signs of cortical thinning and endosteal erosion and were all staged according to Enneking's criteria as active benign tumors (9). The mean longitudinal length of the cyst in long bones was 6.2 cm (range, 1–13). The mean clinical follow-up was 24.5 months (range, 13–64). A "complete response" to treatment (Fig. 1) was observed in 21 (50%) patients and a "partial response" in 16 (38%) patients. Five (12%) patients had a local recurrence (Fig. 2) and were subsequently further treated as summarized in Table 3. All patients except one were controlled with a second similar surgical treatment. Postoperative complications included two superficial wound infections involving serous wound drainage, which resolved within 10 days with oral antibiotics. One patient with a humeral diaphyseal SBC had a transient partial radial nerve palsy postoperatively, which resolved

### TABLE 1. Epidemiology of simple bone cysts

<table>
<thead>
<tr>
<th>Author (ref.)</th>
<th>No.</th>
<th>Male</th>
<th>Female</th>
<th>0–10</th>
<th>11–20</th>
<th>21–30</th>
<th>&gt;30</th>
<th>Pathologic fracture (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Campanacci et al. (4)</td>
<td>416</td>
<td>295</td>
<td>121</td>
<td>54</td>
<td>312</td>
<td>42</td>
<td>8</td>
<td>45</td>
</tr>
<tr>
<td>Inoue et al. (14)</td>
<td>23</td>
<td>15</td>
<td>8</td>
<td>7</td>
<td>11</td>
<td>2</td>
<td>3</td>
<td>22</td>
</tr>
<tr>
<td>Oppenheim &amp; Galloeno (27)</td>
<td>53</td>
<td>43</td>
<td>10</td>
<td>124</td>
<td>49</td>
<td>3</td>
<td>1</td>
<td>&gt;50^</td>
</tr>
<tr>
<td>Spence et al. (35)</td>
<td>144</td>
<td>103</td>
<td>41</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Spence et al. (34)</td>
<td>177</td>
<td>134</td>
<td>43</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Neer et al. (26)</td>
<td>175</td>
<td>120</td>
<td>55</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Bovill &amp; Skinner (3)</td>
<td>32</td>
<td>17</td>
<td>15</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Mylle et al. (25)</td>
<td>59</td>
<td>37</td>
<td>22</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Morton (24)</td>
<td>76</td>
<td>51</td>
<td>25</td>
<td>37</td>
<td>24</td>
<td>6</td>
<td>9</td>
<td></td>
</tr>
<tr>
<td>Scaglietti et al. (31)</td>
<td>163</td>
<td>111</td>
<td>52</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Pentimalli et al. (28)</td>
<td>40</td>
<td>26</td>
<td>14</td>
<td>17</td>
<td>23</td>
<td>—</td>
<td>—</td>
<td></td>
</tr>
<tr>
<td>Schreuder et al. (33)</td>
<td>42</td>
<td>31</td>
<td>11</td>
<td>18</td>
<td>24</td>
<td>—</td>
<td>—</td>
<td></td>
</tr>
<tr>
<td>Total patients</td>
<td>1,400</td>
<td>983</td>
<td>417</td>
<td>259</td>
<td>443</td>
<td>53</td>
<td>21</td>
<td></td>
</tr>
</tbody>
</table>

Data not used in totals.

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This table provides an overview of the epidemiology of simple bone cysts, detailing the numbers and ages of patients, the distribution of pathologic fractures, and other relevant information. The percentages of the total number of patients are also provided, along with the distribution of patients based on their ages.
over 6 weeks without deficit. Two patients had nondisplaced postoperative fractures that occurred after accidents. Both fractures healed with conservative treatment. Limb-length discrepancies of >2 cm were not observed. All patients, including those who had successful treatment of local recurrence, had excellent function of the treated limb recorded during their follow-up examinations. No specific patient-cyst characteristics (age, gender, size, previous treatment, and pathologic fracture) could be identified as significant risk factors for a local recurrence, when evaluated by $\chi^2$ analysis ($p < 0.05$).

![FIG. 1. Simple bone cyst of the humerus in an 11-year-old girl. A,B: Preoperative radiographs showing pathologic fracture. C,D: Postoperative radiographs at 2 months after curettage, cryosurgery, and bone grafting. E,F: Postoperative radiographs at 13 months demonstrating complete consolidation of the grafted site and humeral remodeling.](image-url)
TABLE 3. Patients with local recurrence

<table>
<thead>
<tr>
<th>Patient</th>
<th>Age (yr)</th>
<th>Gender</th>
<th>Center</th>
<th>Site</th>
<th>Time to recurrence (cm) First Second Third</th>
</tr>
</thead>
<tbody>
<tr>
<td>A</td>
<td>16</td>
<td>Male</td>
<td>11</td>
<td>-</td>
<td>9</td>
</tr>
<tr>
<td>A</td>
<td>10</td>
<td>Male</td>
<td>11</td>
<td>-</td>
<td>4</td>
</tr>
<tr>
<td>A</td>
<td>9</td>
<td>Male</td>
<td>11</td>
<td>-</td>
<td>2</td>
</tr>
</tbody>
</table>

DISCUSSION

A complete response...

Survey of patients with local recurrence: Further treatment and outcome. Patients with local recurrence were classified into two groups: those with complete response and those with incomplete response. The results are presented in Table 3. The complete response group showed no recurrence of disease, while the incomplete response group experienced a recurrence of disease. The time to recurrence and the size of the site are also shown in Table 3. The time to recurrence ranged from 9 to 11 months, with an average of 10 months. The size of the site ranged from 2 to 6 cm, with an average of 4 cm.

An initial decrease in intracranial pressure (6.8 mmHg) was observed after cranial decompression in all patients, indicating that the cysts were not located directly adjacent to the brainstem. The cysts were localized in the midline on sagittal and coronal images, indicating that they were located in the brainstem. The radiographic findings were consistent with the clinical symptoms of the patients.

Histologically, the cysts were lined by a foreign soft-tissue material with no intracranial bone cyst. The distribution of the cysts was consistent with the images obtained from the patients. The expansion of the cysts was associated with some limited expansion of the bone, which was subsequently reabsorbed.

The diagnosis of SBC or unicameral bone cyst was first described as a specific entity by Aitchison in 1876 (33). The classification refers to a unicameral bone cyst that is radiographically unique and is not locally reabsorbed or eroded. The cysts are located in the brainstem or spinal cord and are often associated with a decrease in intracranial pressure. The cysts are lined by a foreign tissue material and are not associated with any intracranial bone cyst.

Improving treatment of SBC or unicameral bone cyst requires further study.
synovial cyst, based on electron microscopic analysis of cyst membranes. High levels of oxygen scavengers have been isolated in SBC fluid and implicated in the associated bone destruction (17).

The demographic characteristics of our series do not differ significantly from the typical picture demonstrated in the literature for SBCs. The majority (57%) of patients in the literature with SBCs are between the ages of 11 and 20 years, 33% are younger than 11 years, and 10% involve patients older than 20 years (Table 1). There is a clear male predominance: 70 versus 30% females (Table 1). Fifty-seven percent of the cysts are located in the humerus, and 25% in the femur. SBCs frequently are seen with a pathologic fracture (68%), often caused by a minimal trauma (Table 1). Pain without radiographic evidence of fracture is the second most common presentation, and the rest are discovered incidentally. When a fracture is present, normal fracture healing will occur, and in a small percentage of cases, the SBC may heal; however, most SBCs will recur without treatment and are at risk for a second fracture (1,15). Our series of patients with SBCs was exceptional only in the relatively high number of humeral cysts and a slightly higher fracture incidence.

Treatment alternatives for SBC in the past have most commonly included steroid injection or operative curettage. Because recurrences after curettage alone remain significant, several forms of local adjuvant treatment have been tried. Phenol as an adjuvant treatment after intralesional curettage of SBCs has been reported, with a recurrence rate of 20% (5,26,32). Cementation, or the use of cement, is another form of adjuvant treatment used in the past for giant cell tumor (22,29). To our knowledge, no data are available on its use as an adjuvant for SBCs. Marcove (20) used cryosurgery as adjuvant treatment in 23 patients with SBCs and recorded two (9%) recurrences.

We summarized the published results of the treatment of SBCs in Table 4. As possible, the literature treatment results have been classified into A, B, C, and D categories, as has our own series. The distinction between a complete response (A) and a partial response (B) is subjective to potential intra- and interobserver bias. The categories of local recurrence (C) and no response (D) represent patients requiring further treatment. As described by this review of the literature, curettage with bone grafting for SBCs is associated with an overall mean recurrence rate of 29% (range, 12-48%). Using cryosurgery as local adjuvant therapy, we have been able to achieve a recurrence rate of 12% (five of 42 patients).

Scaglietti et al. (31) introduced the technique of steroid injection, which remains widely used. In their initial series, 76% of the patients failed to respond after the first injection and required more injections. On the average, three to four injections over a period of 12-20 months were needed to achieve complete healing in 55% with "rare recurrences" and partial healing in 45% associated with a higher recurrence rate (30,31). Other series reported local recurrence together with no response after injection as high as 25% (3,4). The disadvantages of steroid injections include the need for repeated injections and multiple visits for follow-up over long periods. The ability to produce a completely healed bony defect remains poorly defined. The advantage of injections is the minor extent of the treatment and its minimal associated morbidity.

Cryosurgery with liquid nitrogen has been used in orthopaedic oncology for the treatment of benign active and aggressive tumors (18,20). This form of adjuvant treatment is delivered by spraying liquid nitrogen into the curetted cavity to extend the surgical margin. The associated temperature injury will kill residual tumor cells.

Basic research has been done to investigate the extent of histologic changes in frozen bone segments in vivo. Malawer et al. (19) showed in experiments with dogs that liquid nitrogen was capable of inducing trabeicular and bone necrosis, extending 7-12 mm around the circumference of a cavity representing a bone cyst. The minimal temperature necessary for a cytotoxic effect is believed

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**TABLE 4. Treatment results from published series**

<table>
<thead>
<tr>
<th>Author (ref.)</th>
<th>Steroid Injection</th>
<th>Curettage ± bone graft</th>
<th>Follow-up</th>
<th>Comments</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>N</td>
<td>A</td>
<td>B</td>
<td>C</td>
</tr>
<tr>
<td>Campanacci et al. (4)</td>
<td>141</td>
<td>71</td>
<td>36</td>
<td>21</td>
</tr>
<tr>
<td>Inoue et al. (14)</td>
<td>—</td>
<td>—</td>
<td>—</td>
<td>—</td>
</tr>
<tr>
<td>Oppenheim &amp; Galleno (27)</td>
<td>50</td>
<td>15</td>
<td>2</td>
<td>1</td>
</tr>
<tr>
<td>Spence et al. (35)</td>
<td>—</td>
<td>—</td>
<td>—</td>
<td>—</td>
</tr>
<tr>
<td>Spence et al. (34)</td>
<td>—</td>
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<td>—</td>
<td>—</td>
</tr>
<tr>
<td>Nee et al. (26)</td>
<td>—</td>
<td>—</td>
<td>—</td>
<td>—</td>
</tr>
<tr>
<td>Bovill &amp; Skinner (3)</td>
<td>12</td>
<td>6</td>
<td>3</td>
<td>1</td>
</tr>
<tr>
<td>Mylle et al. (25)</td>
<td>20</td>
<td>5</td>
<td>13</td>
<td>1</td>
</tr>
<tr>
<td>Morton (24)</td>
<td>—</td>
<td>50</td>
<td>52*</td>
<td>—</td>
</tr>
<tr>
<td>Scaglietti et al. (31)</td>
<td>198</td>
<td>90b</td>
<td>73b</td>
<td>—</td>
</tr>
<tr>
<td>Permali et al. (28)</td>
<td>20</td>
<td>14</td>
<td>5</td>
<td>1</td>
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<tr>
<td>Total patients</td>
<td>374</td>
<td>201</td>
<td>132</td>
<td>25</td>
</tr>
<tr>
<td>Percentages of totals:</td>
<td>54</td>
<td>35</td>
<td>7</td>
<td>4</td>
</tr>
</tbody>
</table>

In reviewing these articles, we used our modification of the original Nee et al. classification of treatment results (A, B, C, and D; see text) for simple bone cysts. The definition was applied consistently to patients treated with steroid injection or curettage.

*a* Not defined in A or B.

*b* Recurrences mentioned but not quantified.
Cryosurgery as Adjuvant for Simple Bone Cyst

Although the benefit of the cryosurgical technique has been recognized for some time, it has not been practiced widely in orthopaedic oncology because of limited experience and concerns regarding a high incidence of complications (1,2). Although complications caused by nitrogen embolism have been reported, they were not observed in our patients (20,33). One patient in our series did have a partial radial nerve palsy that completely resolved spontaneously. We agree with Marcove (22) that the complication rate has been significantly reduced with greater experience.

All patients except two in this series had their bony defects grafted with allogenic bone chips. In general, bony healing was rapid and relatively complete (13). There were no cases of postoperative osteolysis that were not associated with tumor recurrence. We did not recognize any other complications attributed to the use of this graft material. Patients and their families were carefully informed regarding the small risk of potential disease transmission with allogenic bone grafts, and all patients receiving allograft consented to its use. Based on this experience, we continue to encourage patients and their families to consider allogenic freeze-dried chips as the graft of choice.

We believe that the use of cryosurgery as an adjuvant therapy in the surgical treatment of SBCs is of significant value. With this technique, we experienced a local recurrence rate of 12%, which is lower than the overall mean recurrence rate of published series of 29% for curettage and bone grafting. Local recurrences that did occur were typically recognized (four of five) within 12 months after surgery. These results were achieved in a group of patients who had a relatively high fracture rate and for whom prior treatment had failed in 38% of the patients. Considering this relatively high-risk group of patients, we think that our local tumor control is an improvement over past published techniques.

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REFERENCES


