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CLINICAL RESEARCH

Is Curettage and High-speed Burring Sufficient Treatment for Aneurysmal Bone Cysts?

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Abstract

Background To decrease the recurrence rate after intralesional curettage for aneurysmal bone cysts, different adjuvant treatments have been recommended. Liquid nitrogen spray and argon beam coagulation have provided the lowest recurrence rates, but unlike the high-speed burr, these adjuvants are not always available in operating rooms.

Questions/purposes We asked: (1) Is high-speed burring alone sufficient as an adjuvant to curettage with respect to recurrence rates? (2) What is the complication rate from this technique? (3) What are the risk factors for local recurrence?

Methods A retrospective review of the database of the University Musculoskeletal Tumor Unit and the private files of the senior author (EHW) for a period of 19 years (1993–2011) was performed to identify all patients histologically diagnosed with primary aneurysmal bone cyst. During that period, patients with aneurysmal bone cysts were treated with intralesional curettage, burring, and bone grafting if the lesions showed an adequate cortical wall or a wall with thinned out portions which could be reconstructed with bone grafting. Based on those indications, we treated 54 patients for this condition. Of those, 18 were treated using approaches other than burring because they did not meet the defined indications, and an additional five patients were lost to followup before 2 years, leaving 31 patients for analysis, all of whom were followed up for at least 2 years (mean, 7 years; range, 2–18 years).

Results Of these 31 patients, one had a recurrence (3.2%). Complications using this approach occurred in three patients (9.7%), and included growth plate deformity (1) and genu varus (2) secondary to collapse of the reconstructed condyle. With only one recurrence, we cannot answer what the risk factors might be for recurrence; however, the one patient with recurrence presented with a large lesion and a pathologic fracture.

Conclusions Curettage, burring, and bone grafting compare favorably in the literature with other approaches for aneurysmal bone cysts, such as cryotherapy and argon-beam coagulation. We conclude that high-speed burring alone as an adjuvant to intralesional curettage is a reasonable approach to achieving a low recurrence rate for aneurysmal bone cysts.

Level of Evidence Level IV, therapeutic study. See the Instructions for Authors for a complete description of levels of evidence.

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Each author certifies that his or her institution approved the human protocol for this investigation and that all investigations were conducted in conformity with ethical principles of research.
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Introduction

Aneurysmal bone cysts are benign but locally aggressive lesions, which account for approximately 2% to 3% of all benign bone tumors [1, 17, 18]. They usually occur during the first two decades of life and are most commonly located in long bones [1, 17, 18]. Standard treatment of accessible extremity aneurysmal bone cysts is intralesional curettage but local recurrence rates are high, ranging from 12% (four of 34) to 60% (26 of 44) [2, 7, 12, 15]. To decrease recurrence rates, different adjuvant methods have been advocated after curettage to increase the zone of necrosis and reduce the likelihood of leaving residual tumor cells. These include phenol, alcohol, polymethylmethacrylate, burring, liquid nitrogen, and more recently argon beam coagulation [4, 7, 15].

Among these adjuvants, lower recurrence rates ranging from 4% (one of 27) to 7.5% (three of 40) have been associated with argon beam coagulation and liquid nitrogen spray [4, 11, 14, 15]. There is concern, however, regarding the relatively high incidence of postoperative complications, especially pathologic fracture, from both procedures [10, 15]. However, curettage with high-speed burring alone has been reported to have a recurrence rate of 12% (four of 34) and no serious postoperative complications [7]. A second study from the same institution 12 years later showed a rate of 20.6% (seven of 34) for a combined recurrence rate of 14% (seven of 50). The complication rate continued to be low at only 6% (two of 34) [15]. Percutaneous sclerotherapy with intralesional injections of polidocanol have reported recurrence rates of 2.8% (two of 72) to 4.5% (two of 44), but these rates were achieved only after a mean of two to three injections [12, 13]. In our hospital, accessible aneurysmal bone cysts have been treated since 1993 with intralesional curettage and high-speed burring followed by bone grafting. Liquid nitrogen spray and argon beam coagulation are not readily available. En bloc resection is reserved for large aggressive lesions and lesions in expendable bones, whereas arterial embolization is used for aneurysmal bone cysts in inaccessible locations such as the pelvis or spine. However, as noted, there are few studies reporting on this approach of curettage and adjuvant burring [7, 15], even though it may be important insofar as other adjuvant treatments may not be available in all hospitals.

We therefore asked (1) if high-speed burring alone was sufficient as an adjuvant to curettage with respect to recurrence rates, (2) what the complication rate is from this technique, and (3) what the risk factors are for local recurrence.

Patients and Methods

A retrospective review of the database of the University Musculoskeletal Tumor Unit and the private files of the senior author (EHW) for a period of 19 years (1993–2011) was performed to identify all patients histologically diagnosed with a primary aneurysmal bone cyst. All patients had rereview of their histologic evaluations by an experienced bone pathologist (AVD). Secondary cysts were excluded. This study was approved by the research ethics board of our university.

During that period, patients with primary aneurysmal bone cysts were treated in one of three ways: (1) intralesional curettage, burring, and bone grafting; (2) marginal excision with or without curettage; or (3) embolization. Lesions which on radiographs showed an adequate cortical wall or a wall with thinned out portions which were deemed reconstructible with cortical bone graft were subjected to intralesional curettage and burring. Otherwise, an en bloc marginal excision was performed. Patients with cysts in expendable bones, such as the proximal fibula and distal ulna, likewise underwent marginal excision. Arterial embolization with or without radiation was recommended for lesions that were unresectable or located in inaccessible areas such as the sacrum or pelvis. To be included in the study, patients had to have received complete surgical treatment by our Unit and be followed for at least 24 months to ensure that all local recurrences were detected.

Of 54 patients identified with primary aneurysmal bone cysts, five with less than 2 years followup and five who underwent embolization alone were excluded, leaving 44 patients for evaluation. An additional 13 patients were treated using marginal resection. Because these patients represented a different clinical problem, and could be defined as such, they were excluded from analysis here, leaving a total of 31 patients available for analysis in the current study at a minimum of 2 years (mean, 7 years; range, 2–18 years), except for one 49-year-old man with a distal humerus lesion who died of a myocardial infarction 19 months postoperatively. At the time of death, his humerus was fully healed with full ROM and no recurrence.

Of the 31 patients who underwent intralesional curettage, 12 (39%) were 20 years old or younger. Fourteen patients (45%) presented with a pathologic fracture. Two patients received prior treatment at another hospital and were referred for a local recurrence. Fifteen of 31 patients underwent internal fixation. No bone cement or bone substitute was used and no patient received adjuvant treatment (Table 1).

Radiologic classification was based on the Enneking staging system for benign bone tumors [6]. A Stage 1 or latent lesion is surrounded by a mature rim of cortical-like reactive bone without bony deformation. With a Stage 2 active lesion, there is a clear demarcation between the lesion and cortical bone, but there is no cortical-like

Table 1. Patient and tumor variables for the 31 patients

Variable	Numeric value
Mean age	25.7 years (8–62 years)
Sex	
Male	15 (48%)
Female	16 (52%)
Anatomic location	
Clavicle	1 (3%)
Humerus	5 (16%)
Radius	2 (6%)
Metacarpal	1 (3%)
Pelvis	3 (10%)
Femur	12 (39%)
Tibia	4 (13%)
Calcaneus	3 (10%)
Enneking stage	
1	4 (13%)
2	15 (48%)
3	12 (39%)
Pathologic fracture	
Yes	14 (45%)
No	17 (55%)
Growth plate	
Open	9 (29%)
Closed	22 (71%)

reactive shell. The margin can be irregular, and there often is some expansion of the overlying cortex (Fig. 1A–B). An aggressive Stage 3 lesion is characterized by an ill-defined border, incomplete reactive bone margins, and cortical destruction, some exhibiting soft tissue extension [6, 7]. In 13 patients, growth plates were open; nine cysts abutted these plates.

With intralesional surgery, a window was created through the thinned out cortex overlying the cyst after which a methodic, thorough curettage of the lesion was done. In all cases, the window was enlarged to approximate the length of the lesion to allow complete observation and easy access to the entire cavity with curettes and burr. The curettage was followed by meticulous burring of the cavity walls until all ridges and septations in the cavity were flattened. The cavity was washed with normal saline solution and a second cycle of curettage and burring was repeated. Cavity defects were filled with bone graft using either autogenous or allograft bone or a combination of the two. If an area in the cortex was excessively thinned out or a defect inadvertently made with the burring, a larger piece of bone graft was shaped to cover the defect (Fig. 1C–D).

Postoperatively, all patients were seen every 3 to 4 months for 2 years and had repeat radiographs. Patients then were seen twice a year for the third to fifth years and annually thereafter. Patients who had not followed up in the last 3 months were contacted via mail, cellular phone calls, SMS messaging, or through referring physicians. Travel was inconvenient for some patients because our country is an archipelago. If patients were unable to come to the hospital, our study coordinator would go to the patients to examine them and have their radiographs taken at a facility near their residence. These radiographs then were brought back to us for review.

Results

Local Recurrence

There was one local recurrence among the 31 patients (3.2%) who underwent curettage and burring. The single recurrence after intralesional surgery occurred 24 months postoperatively in a patient who presented with a pathologic fracture through an extensive aneurysmal bone cyst of the humerus. This patient underwent intralesional surgery, had repeat curettage, burring, and bone grafting, and has remained disease-free for 11 years.

Complications

One patient had genu valgus develop after curettage and burring of an aneurysmal bone cyst adjacent to the proximal tibial growth plate. Two patients had genu varus develop secondary to collapse of either the medial femoral condyle or medial tibial plateau after placing weight on the affected leg. One patient underwent a high tibial osteotomy but the other refused additional surgery.

Risk Factors for Recurrence

With only one recurrence, we did not have enough patients to analyze for risk factors for recurrence.

Discussion

For treatment of aneurysmal bone cysts, the traditional method of intralesional curettage has resulted in recurrence rates ranging from 12% (four of 34) to 60% (26 of 44) [11]. Marginal or en bloc resections are associated with much lower local recurrence rates but entail removal of large amounts of bone and can be difficult when the cyst is adjacent to either an osteoarticular segment or a growth plate [3]. Curettage therefore has remained the preferred

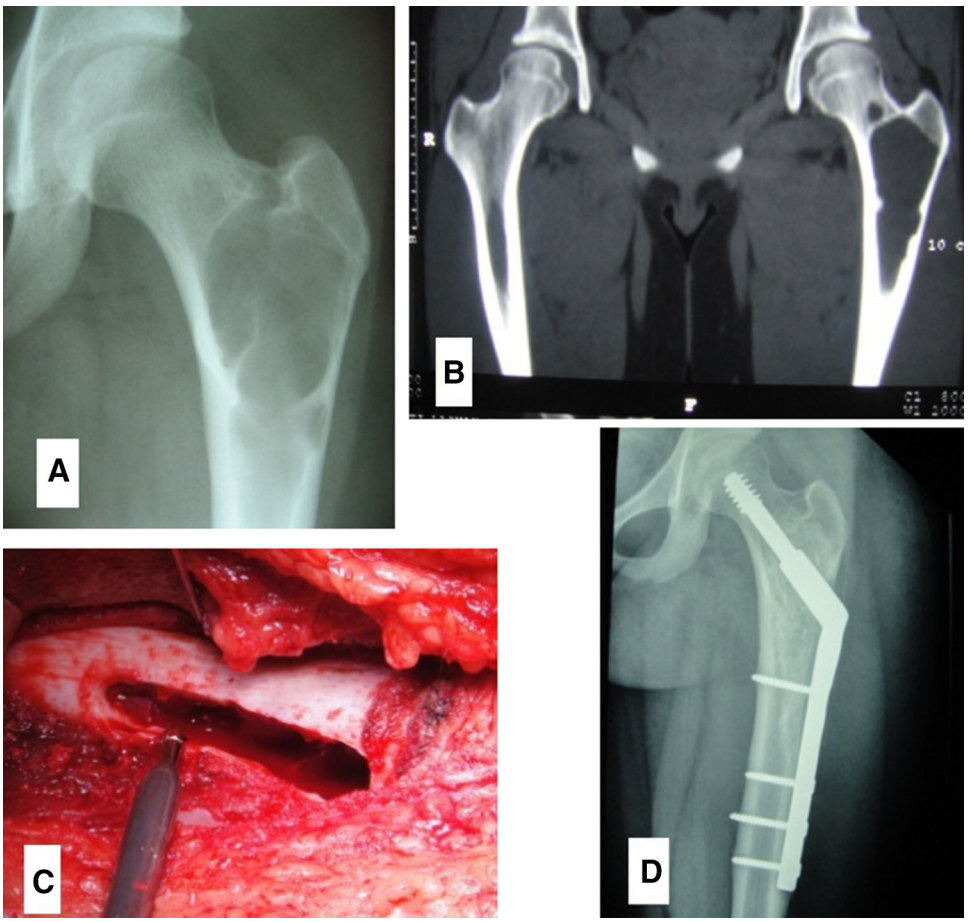

Fig. 1A–D (**A**) A preoperative radiograph shows an Enneking Stage 2 lesion of the left proximal femur. (**B**) The CT image shows the same Enneking Stage 2 lesion of the left proximal femur. (**C**) Thorough curettage and burring was achieved through a generous cortical window. (**D**) The postoperative radiograph shows allograft bone and a compression hip screw.

treatment but has been augmented with different adjuvants, all of which seek to increase surgical oncologic margins and thus decrease the recurrence rate. These adjuvants include phenol, alcohol, polymethylmethacrylate, high-speed burring [4, 5, 7, 8, 15], liquid nitrogen [10, 11, 14], and most recently argon beam coagulation [4, 15].

Our study has several limitations. It is a retrospective study of a heterogeneous group of patients with no true comparison group; selection bias commonly is a question in studies of this design, and may have been a factor here. Offsetting this was that we applied this procedure using clear and defined indications, and had specified indications for which patients should receive other treatments (such as embolization or marginal resection). We did lose five of the 31 patients (16%) to followup; however, even if these patients experienced local recurrences and presented elsewhere for treatment, our recurrence rate would be within ranges of other studies using adjuvant treatments [4, 7, 11].

We found only one recurrence in the 31 patients treated with curettage and high-speed burring. We have been able to achieve a recurrence rate of only 3% (one of 31), which is one of the lowest recurrence rates for intralesional surgery in a cohort of patients with at least 24 months followup. Gibbs et al. [7] reported a 12% (four of 34) recurrence rate after curettage and high-speed burring in a cohort of 34 patients. A more recent study of 34 patients including 18 of the original patients included in the study by Gibbs et al. reported a 20.6% (seven of 34) recurrence rate [15]. However, argon beam coagulation, when used with curettage and burring, achieved a recurrence rate of 7.5% (three of 40) and a rate of 10% (four of 30) among patients with at least 24 months followup [15]. Similar encouraging results were reported by Cummings et al. [4] for 17 patients with aneurysmal bone cysts treated with curettage and argon beam coagulation without any recurrences. However, this latter patient population included patients with less than 24 months followup. Liquid nitrogen or cryosurgery also has been advocated as an adjuvant. After the initial 18% (nine of 51) recurrence rate reported by Marcove et al. [10], Peeters et al. [11] and Schreuder

et al. [14] reported even lower rates of 5% (four of 80) and 3.7% (one of 27), respectively. This discrepancy in recurrence rates has been attributed to a difference in cryosurgical techniques—pouring of liquid nitrogen [10] against liquid nitrogen spray [11, 14], the latter technique possibly enabling a larger surface area for freezing compared with the former technique.

There are several reasons to possibly explain our low recurrence rate. First, tumor removal was pursued aggressively in each case through wide, often generous exposure, meticulous curettage, and thorough burring. We accessed the tumor cavity through an extensive cortical window at least the length of the intramedullary lesion, removing as much cortical shell as necessary. Extra effort was taken to reconstruct cortical defects resulting not only from tumor lysis, but also from this aggressive approach to curettage and burring. Defects were spanned with large pieces of bone graft. The number of patients requiring internal fixation with metal implants (15 of 31 [48%]) may be a reflection of this aggressive surgical approach. Second, the epidemiologic profile of our patients is slightly different from the literature. We have a relatively smaller population of young patients, with 35% (11 of 31) of patients in the first two decades of life, compared with the younger age profile more common in the literature [4, 9, 11, 14, 16, 17]. Growth plates were still open at presentation in only nine patients, six of whom had cysts abutting the plates. This likely allowed more aggressive curettage and burring of lesions because there was less fear of injuring growth plates.

Concern has been raised regarding the postoperative complications encountered with the use of liquid nitrogen, including postoperative fractures, infections, and nerve palsy [10, 11, 14]. Our results suggest that there may be fewer complications associated with high-speed burring compared with the other adjuvants. There were only two patients (6.5%) in our series of 31 who experienced collapse of either the medial femoral or medial tibial condyle, resulting in varus angulation of the knee. This collapse can be attributed to cortical thinning resulting from tumor growth and surgical treatment. All 31 of our patients have resumed their original activities and have full ROM. Steffner et al. [15] reported a 19% (eight of 42) complication rate after using argon beam coagulation, the majority of which were pathologic fractures (12.5%; five of 40). Liquid nitrogen is similarly associated with an increased incidence of postoperative fractures, with Marcove et al. [10] reporting an almost 10% (five of 51) fracture rate. Pathologic fractures from argon beam coagulation and cryosurgery are attributed to dessication or osteonecrosis inflicted on surrounding bone [15].

Although we did not have enough patients to identify risk factors for recurrence in a statistically robust way, we do observe that our one patient with recurrence had a large lesion and presented with a pathologic fracture. Risk factors for recurrence after treatment have included pathologic fracture, Enneking stage, surgical margins, local recurrence on presentation, and proximity to growth plate [11, 15]. Additional studies will need to see to what degree those risk factors may apply to treatment with curettage and burring, and we suspect they likely do apply.

We showed that the high-speed burr, even when used as a sole adjuvant method for intralesional curettage and bone grafting, is a reasonable approach for achieving low recurrence and complication rates in the treatment of aneurysmal bone cysts. This is a potentially viable approach, but it needs to be compared with other approaches in properly designed, multicenter trails. The high-speed burr is readily available in most operating rooms, whereas other adjuvants such as the argon beam and liquid nitrogen spray, are not regular operating room armamentarium.

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