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ORIGINAL REPORT

Aneurysmal Bone Cyst: A Review of 150 Patients

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A B S T R A C T

Purpose
We have reviewed a series of 150 aneurysmal bone cysts treated over the last 20 years.

Patients and Methods
The lesions were principally located in the tibia, femur, pelvis, humerus, and spine and, in most cases, presented the imaging appearance originally described by Jaffe and Lichtenstein as a blowout with thin cortices.

Results
Only one of the patients was believed to have an osteoblastoma of the spine with secondary development of an aneurysmal bone cyst, and none of the patients developed additional lesions. The patients were treated primarily with curettage and implantation of allograft chips or polymethylmethacrylate, but some patients were treated with insertion of autografts or allografts. The local recurrence rate was 20%, which is consistent with that reported by other centers.

Conclusion
Aneurysmal bone cysts are enigmatic lesions of unknown cause and presentation and are difficult to distinguish from other lesions. Overall, the treatment is satisfactory, but it is possible that newer approaches, such as improved magnetic resonance imaging studies, may help diagnose the lesions and allow the physicians to plan for more effective treatment protocols.

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INTRODUCTION

Despite a descriptive history of more than 60 years, the nature, character, and optimal treatment of aneurysmal bone cysts remain obscure. The lesion was first described by Jaffe and Lichtenstein[1] in 1942, was subsequently further defined by both of these authors,[2,3] and became known as Jaffe-Lichtenstein disease. Despite attempts on the part of investigators to establish a relationship of the disorder to other entities, the term aneurysmal bone cyst remains purely descriptive. It does not provide any concept of pathogenesis or causation mechanisms, and efforts on the part of a number of investigators to discover a genetic or neoplastic cause have failed.[4-18] Examination of the tissue at the time of surgery has, in the past, demonstrated a blood-filled cavity within an expanded region of the bone, and the cells that line the cyst wall show fibrous components, macrophages, giant cells, and islands of bone.[1,7-9,19-24] The term aneurysmal seems to relate to the blowout distension, and the word cyst reflects the fact that the tumor often presents as a blood-filled cavity.[7,9,25] Occasionally in prior studies, there have been findings suggesting the possibility that the aneurysmal cyst is actually a result of hemorrhagic degradative events occurring in patients with other lesions including giant cell tumor, hemangioma, chondroblastoma, osteoblastoma, nonossifying fibroma, fibrous dysplasia, chondromyxoid fibroma, eosinophilic granuloma, and other tumors.[1,7,9,19-24] Of greater concern is the possibility that the lesion is not

an aneurysmal cyst but a partially necrotic and extremely vascular telangiectatic osteosarcoma, which has a high rate of metastasis.[7,9]

Many reports have appeared that describe patients with aneurysmal bone cysts.[3,6,8,14,16,22,24,27,28,31-36] The lesions are more common in patients in the first two decades of life rather than in later years[7,8,28,31,37] and seem to be slightly more frequent in females than males.[7,8,28-30] The major sites of occurrence, according to a number of authors, are the femur, tibia, humerus, spine, and pelvis, and although tumors arising in the small bones of the hands and feet are seen occasionally, there are fewer of such instances in most series.[3,7,9,14,16,24,32-36] The tumors are usually metaphyseal or diaphyseal and are most often eccentrically located, which is an important distinguishing radiographic feature from unicameral bone cyst in patients in their second decade.[7-9,12,20,24,38,39] The appearance of the lesions support the word blowout and show marked thinning of the cortex over the site, with only minimal bone formation; all of which are sometimes best seen on a computed tomography (CT) or a magnetic resonance imagining (MRI) scan.[38,39] Controversy exists regarding optimal treatment, and regardless of techniques reported, there remains a recurrence rate that ranges from 5% to greater than 40%.[7,24,28-34] At present, curettage and insertion of bone graft or polymethylmethacrylate are the principal techniques used,[7,20,24,29,31,33,34] but in the past, radiation has been used.[40] In several trials, sclerosing substances, bone substitutes, and other agents seemed to be less effective than conventional curettage.[41-43] Our purpose in presenting this material is to review the data obtained over the last 20 years regarding patients

Table 1. Demographic Data for 150 Patients With Aneurysmal Bone Cyst

Characteristic	No. of Patients
Age, years	
Mean	18
Standard deviation	12
Range	3-62
Sex	
Male	69
Female	81
Follow-up, years	
Mean	6
Standard deviation	7
Range	1-20
Anatomic sites	
Tibia	37
Proximal	25
Middle	5
Distal	7
Femur	26
Proximal	9
Middle	4
Distal	13
Fibula	16
Proximal	7
Middle	2
Distal	7
Pelvis	13
Humerus	10
Proximal	7
Middle	2
Distal	1
Clavicle	10
Foot	8
Ulna	5
Proximal	1
Middle	3
Distal	1
Lumbar spine	5
Cervical spine	3
Scapula	3
Sacrum	2
Ribs	2
Dorsal spine	1

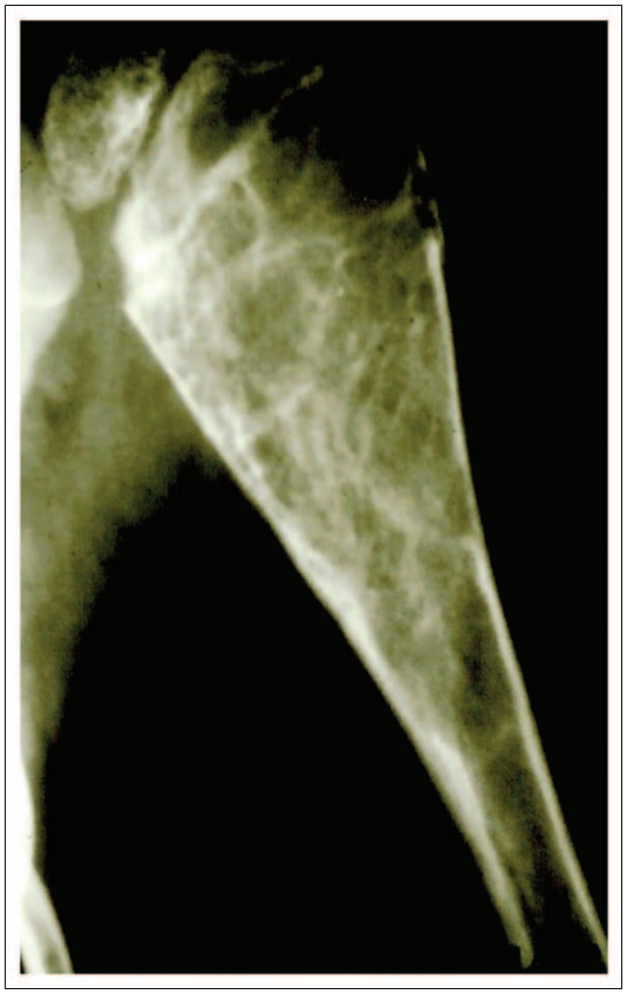

Fig 1. An x-ray of an aneurysmal cyst arising in the proximal humerus in a child. Note the irregularity of the cortices and the expansion of the bone. The tumor was painful not only because of its structure but also because of a small pathologic fracture on the lateral side.

with aneurysmal bone cysts to define the appearance, anatomic site, complications, and results of various forms of treatment.

PATIENTS AND METHODS

The Orthopedic Oncology Service at the Massachusetts General and Children's Hospitals has maintained a computerized database containing information regarding more than 17,000 patients with bone and soft tissue tumors treated over the last 30 years.[44] Study of the system provided information regarding 223 patients with aneurysmal bone cyst, but only 150 of these patients who had been observed for as long as 20 years had sufficiently accurate descriptions of treatment protocols, definition of pathology, and recent assessment of outcomes to allow statistical analysis (Table 1). The system used to gather information regarding the patients did not, in any way, violate patient confidentiality and was approved by the hospitals' institutional review boards.

Sixty-nine of the 150 patients were male, and 81 were female. The mean age of the patients was 18 ± 12 years, with a range of 3 to 62 years. The mean follow-up time for the patients was 6 ± 7 years (range, 1 to 20 years). The anatomic locations for the lesions are listed in Table 1, and as noted, the most common sites were the tibia, femur, fibula, pelvis, humerus, clavicle, foot, and lumbar spine. The patients' complaints at the time of the initial visit were almost always concerning pain at the site and sometimes indicated pain and numbness extending down the limb. The patients were often disabled by the pain and almost always presented with tenderness over the site of the lesion. Examples of the imaging studies are shown in Figures 1 and 2, and as noted, the lesions are metaphyseal or diaphyseal in location, are usually eccentric with thin cortices, and, in most cases, present the classic blowout appearance (Fig 3). Gross structure is shown in Figure 4, which displays the thin cortices, the expansion, and the chambers often filled with blood. The histologic patterns are shown in Figures 5 and 6. The patterns demonstrate thin cortices; blood elements in the central portion of the tumor; and an array of benign-appearing macrophages, lymphocytes, fibroblasts, bone-forming cells, and giant cells, which are sometimes atypical in terms of structure and nuclear distribution.

As shown in Table 2, 130 patients were treated with curettage, and of these patients, 101 had lesions that were packed with allograft bone, and 20 had lesions that were packed with polymethylmethacrylate cement. Twenty of the patients had an excision or resection of the lesion, and 11 of these patients received an intercalary allograft transplantation.

Statistical studies used analysis of variance and Mantel-Haenzel and Fisher's exact tests using χ^2 analysis. The systems were provided by BMDP Statistical Software (Los Angeles, CA). $P < .05$ was considered statistically significant.

RESULTS

None of the patients died of disease, and there were no amputations. The principal problem that the patients encountered with their treatment of the aneurysmal bone cysts was local recurrence, which occurred in 30 (20%) of the 150 patients at 1.2 ± 0.7 years (range, 0.3 to 3 years) after discovery of the lesion. There was no statistical difference for sex. Patient age did not seem to have a significant effect on outcome, although the rate of local recurrence was slightly increased in younger patients (Table 3). Anatomic site seemed to make some difference in rates of local recurrence. The recurrence rate for the 10 patients with lesions of the clavicle was 50%, and the recurrence rate for the 13

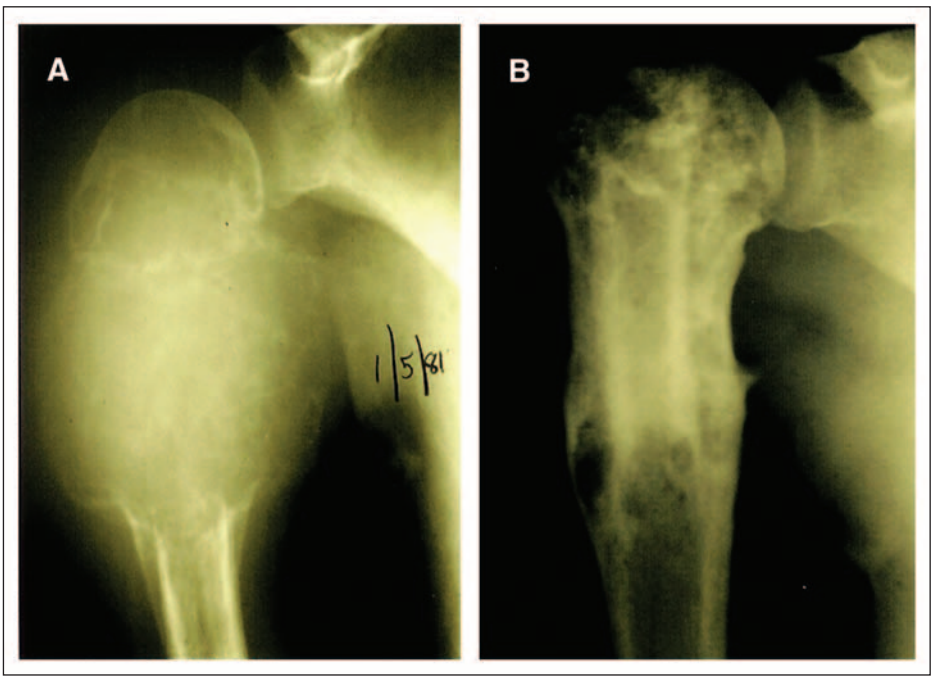

Fig 2. An enormous aneurysmal bone cyst of the proximal humerus. Note the size of the lesion and the blowout appearance with thin cortices and periosteal new bone on the shaft. Despite the suggestion that this was a giant cell tumor, the histology was classic for an aneurysmal bone cyst. The lesion was curetted out, and a segment of allograft radius was inserted (B).

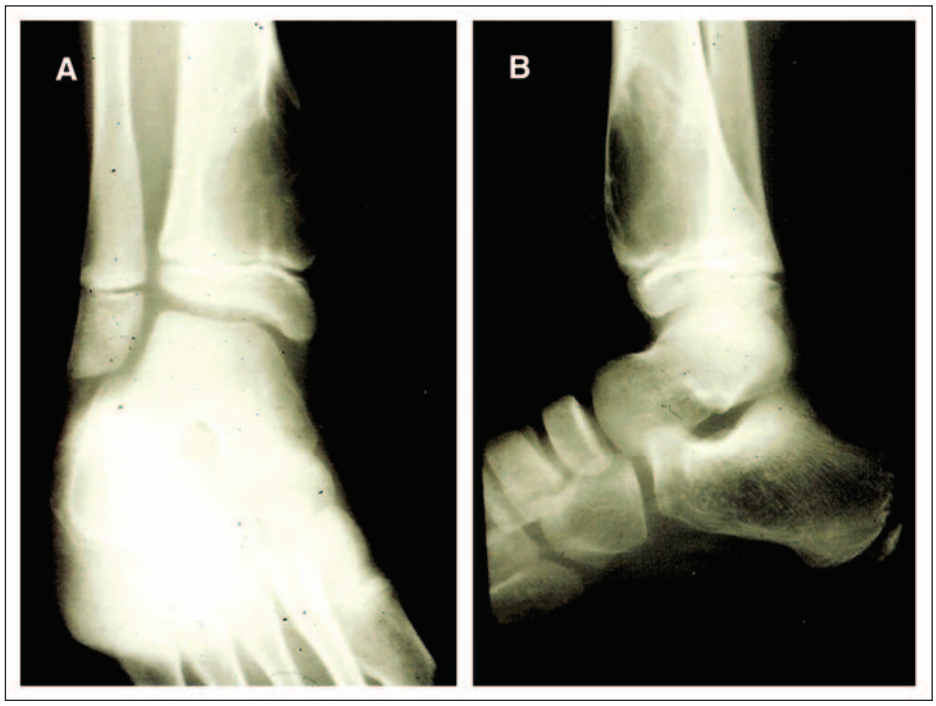

Fig 3. A typical site and structure for an aneurysmal bone cyst of the distal tibia in a 12-year-old female patient. The eccentric location and the marked thinning of the cortex are characteristic.

patients with tumors in the distal femur was 46%, whereas other sites showed few or no recurrences (Table 3).

Of considerable interest was the recurrence rate data for the various operative procedures performed by our group over the 20+ years of experience in the treatment of patients with aneurysmal bone cysts. The recurrence rate for the 121 patients who were treated with curettage and packing with either allograft chips or polymethylmethacrylate was 22%, which was considerably greater than the 5% recurrence rate for patients who underwent resection and either an autograft or an allograft implantation (Table 2).

The patients who developed a recurrence required subsequent surgery, which consisted of another curettage and packing or resection of the site and autograft or intercalary allograft implantation. Of the 150 patients, 34 required a second operation and 13 required a third operation for recurrences or structural problems related to failed systems. One of the patients was treated with sclerotherapy, and two patients were treated with local injections of alcohol and other agents, all of which were successful. Three of the patients developed fractures through the weakened bony part, which, although not related to recurrence, required

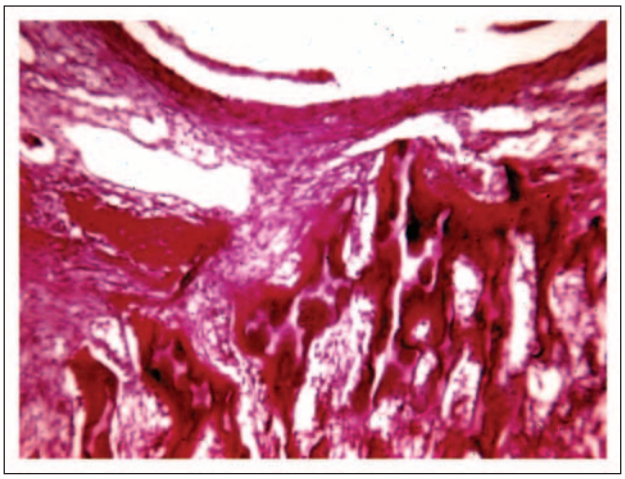

Fig 4. Classic gross appearance of an aneurysmal bone cyst of a rib. The blood-filled chamber is irregular in structure, and there are islands of bone and fibrous tissue. The bone is expanded and irregular in shape with the typical blowout appearance.

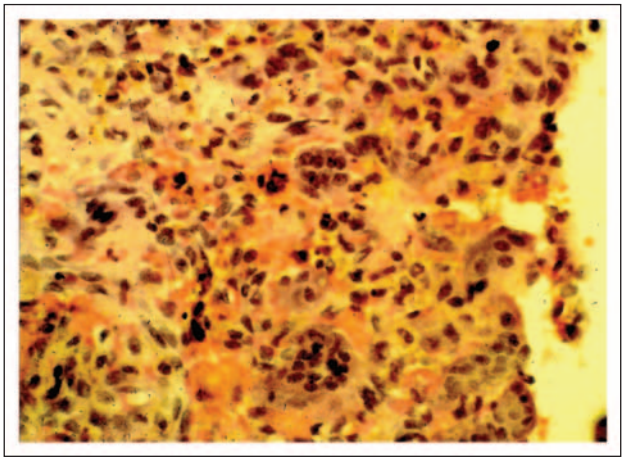

Fig 5. Histologic appearance of the margin of the tissue from the rib lesion shown in Figure 4. The bone is irregular in structure, and a fibrous layer separates it from the region of the blood-filled cavity.

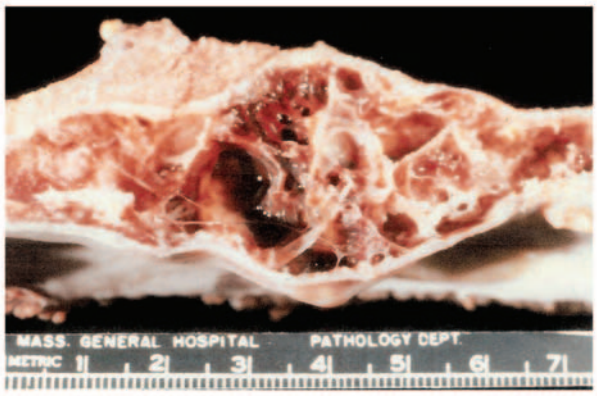

Fig 6. A high-power view of the tissue shown in Figure 5 showing the cellular components, which include fibroblasts and many monocytic cells along with blood cells. Several giant cells are present, but the appearance of the tissue does not suggest a giant cell tumor.

further surgery. The 11 allograft implantations were all rated as good or excellent at 6 ± 6 years (range, 1 to 20 years), and only one of the grafts sustained a fracture, which healed after replacement of the fixation. Only one of the patients in our series was found to have tumor suggestive of an osteoblastoma of L4 at the time of biopsy. The lesion was curetted and packed with allograft chips, and there has been no recurrence reported to date.

Table 3. Effect of Treatment on Recurrence Rate

Treatment Method	No. of Patients	Recurrences	
		No.	%
Curettage and pack with allograft chips	101	21	21
Curettage and pack with PMMA	20	5	25
Resect or excise and autograft implantation	9	1	11
Biopsy, curettage, and no implantation	9	3	33
Resect or excise and allograft implantation	11	0	0

Abbreviation: PMMA, polymethylmethacrylate.

Fifty-seven of the 150 patients had the DNA content of the tissue obtained at surgery studied by flow cytometric analysis.[45] The mean value for the diploid peak was 92 ± 4.3 (range, 86 to 98), and the mean value for the G2+M was 4.7 ± 3.2 (range, 0. to 14). The average mean DNA index was 1.06 ± 0.04 (range, 1 to 1.2). No aneuploid peaks were encountered. These values can be interpreted as being characteristic of benign tumors.[45]

DISCUSSION

As stated in the Introduction, despite the long experience of radiologists, pathologists, and orthopedists with aneurysmal

Table 2. Recurrence Rate for 150 Patients According to Various Parameters

Parameter	No. of Patients	Recurrences		Proximal		Middle		Distal	
		No.	%	No. of Patients	No. of Recurrences	No. of Patients	No. of Recurrences	No. of Patients	No. of Recurrences
Sex*									
Male	69	13	21						
Female	81	21	19						
Patient age*									
< 10 years	29	7	24						
10-20 years	76	16	21						
> 20 years	45	7	16						
Anatomic site									
Tibia	37	4	11	25	3	5	0	7	1
Femur	26	9	35	9	1	4	2	13	6
Fibula	16	4	25	7	1	2	0	7	3
Pelvis	13	2	15						
Humerus	10	2	20	7	2	2	0	1	0
Clavicle	10	5	50						
Foot	8	2	25						
Ulna	5	1	20	1	1	3	0	1	0
Lumbar spine	5	0							
Cervical spine	3	1	33						
Scapula	3	0							
Sacrum	2	0							
Ribs	2	0							
Dorsal spine	1	0							

NOTE. Recurrence rate: 30 recurrences (20%) at 1.2 ± 0.7 years (range, 0.3 to 3 years).
*Not significant.

bone cysts, there is limited knowledge regarding the cause of the lesion, its natural history, and the results of treatment.[1,4,5-7,9-17] The concept that the lesion represents a vascular degenerative process for some benign bone lesions is an attractive one, but the pathologic findings, with rare exception, do not really support this proposal. Few pathologic specimens contain tissues that are highly characteristic or diagnostic of giant cell tumor, chondroblastoma, hemangioma, osteoblastoma, nonossifying fibroma, fibrous dysplasia, chondromyxoid fibroma, and other tumors.[6,7,9-11,15-17,19,21,26,28,29] Furthermore, the recurrences after surgical treatment do not show evidence of such lesions, particularly aggressive lesions such as giant cell tumor. Only one of our 150 patients showed a finding suggestive of an osteoblastoma at the site of the aneurysmal bone cyst of L4. At best, it was a difficult decision based on the similarity of the repair process to the histologic pattern for osteoblastoma, and the lesion has not recurred at more than 2 years since the surgery.

Of some importance is the difficulty that can occur in diagnosing these lesions. The imaging studies, even CTs and MRIs, sometimes do not provide clearly diagnostic criteria for the diagnosis of aneurysmal bone cyst, and aneurysmal bone cyst is sometimes added on to a list of diagnoses including eosinophilic granuloma, giant cell tumor, nonossifying fibroma, unicameral bone cyst, fibrous dysplasia, chondroblastoma, chondrosarcoma, chondromyxoid fibroma, Ewing's tumor, and, in older patients, metastatic carcinoma or myeloma.[7-9,12,20,24,38,39] The lesions are often eccentric and irregular in structure and sometimes show calcification in the central areas. As a rule, the cortex is thin, but there is rarely a cortical defect or a soft tissue mass. CT and MRI are often helpful in defining the extent of the lesion and establishing the diagnosis. A biopsy is often helpful, and many of our patients underwent a needle biopsy before definitive treatment. Needle biopsies are sometimes a problem because the material obtained may consist of mostly blood elements. Often, an open biopsy and frozen section are necessary to establish the diagnosis.

The recurrence rate in this series was 20%, which should be considered high compared with other series and other benign tumors.[7] Part of this problem could conceivably be related to the methods of treatment over the 20 years during which these patients were treated. The recurrence rate for patients treated in the earlier years was higher than for patients who were treated more recently (approximately 26% *v* 17%, respectively). In the past, we used curettage alone, but our principal current approach was biopsy followed by curettage and then implantation of allograft chips or, more recently, polymethylmethacrylate. Autograft implantations or utilization of intercalary allografts were quite successful but were, for the most part, used for patients with lesions that were large or seemed to threaten the integrity of the bone and were used less frequently for patients who experienced failure of their primary procedure.

Although none of our patients died or required an amputation, a number of them had some relatively minor disability as a result of the tumor and its treatment. Thus, it is our opinion that aneurysmal bone cyst is sometimes an aggressive lesion that is difficult to treat. Lesions that occur in the proximal femur should perhaps be treated more aggressively, partly because of the high rate of local recurrence and the risk of fracture. The most appropriate techniques for some of these tumors are primary resective surgery and allograft implantation. Patients with lesions of the proximal or mid fibula or clavicle or body of the scapula could be treated by resection alone, and lesions of the foot might be best treated by resection and arthrodesis using autograft. According to our series, such treatment would at least reduce the local recurrence rate considerably and probably reduce the degree of even minimal disability reported by some of these young patients.

In this last analysis, aneurysmal bone cyst remains an enigma, not only regarding causation, but also regarding clinical and imaging diagnosis and optimal treatment. There is perhaps some hope for implantation of newer agents, such as the bone substitutes, to aid in healing of the lesions. Although there are now some markers that are alleged to be specific for aneurysmal bone cyst,[1,9,12-15,18] there is still no system to establish the diagnosis or to support different methods of treatment and, thus, reduce the problems encountered by the patient and the surgeon.

Authors' Disclosures of Potential Conflicts of Interest

The authors indicated no potential conflicts of interest.

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