



Aneurysmal bone cyst of the clavicle: a series of 13 cases

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Background: Aneurysmal bone cyst (ABC) is a benign but locally aggressive bone tumor occurring most commonly in the first 2 decades of life. The clavicle is a rare location for tumors, and ABCs of the clavicle have been sparsely described in the literature. We present the largest known series of ABCs of the clavicle to describe this rare condition.

Methods: Patients were identified retrospectively from an orthopedic oncology database to obtain demographic, diagnostic, and treatment information. A literature search was performed to identify all English language reports of ABC of the clavicle.

Results: We identified 13 patients with ABC of the clavicle, 77% of which were in the acromial end. Most patients (77%) presented with pain or swelling or both. The initial treatment in 11 patients was by curettage, with or without allograft bone packing, and 1 underwent resection/reconstruction of the lateral clavicle. Seven patients (58%) had 1 or more recurrences at an average of 6 months, for which 2 were treated with partial resection of the clavicle.

Conclusions: ABC of the clavicle is a rare condition that we found to occur most frequently in the acromial end of the bone. Most patients were initially treated with curettage and showed a higher rate of recurrence than in other locations. The unique anatomy of the clavicle makes resection a feasible and very functional option, especially if the coracoclavicular ligaments can be preserved. However, the potential resulting deformity may cause patients to opt for a less aggressive and, consequently, less effective treatment method.

Level of evidence: Level IV; Case Series; Treatment Study

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Aneurysmal bone cyst (ABC) is a benign, locally aggressive bone tumor that is most commonly seen in the skeletally immature, with 80% occurring in patients aged younger than 20 years. The literature reports either no or a slight female predilection.^{2-4,6,18,19} ABCs account for 1% to 3% of all benign osseous tumors and are most frequently seen in the long tubular bones, vertebrae, pelvis, and scapula.^{4,6,19}

ABCs appear on radiographic images as a blowout of the periosteum. The lesions are osteolytic and “expansile,” with a soap-bubble appearance.^{4,6,19,26} ABCs share many clinical and radiologic features with other lesions, most concerning telangiectatic osteosarcoma, and a biopsy specimen is necessary to make the distinction.^{4,18,19} The differential diagnosis of ABC also commonly includes giant cell tumor of bone, chondromyxoid fibroma, and unicameral bone cyst.^{4,6}

The pathogenesis of primary ABCs has been long debated. Theories range from vascular malformation or disturbance to trauma, but more recent research has shown several rearrangements of chromosome 17p13.^{6,19,21} Approximately 30% of ABCs are secondary to other benign bone tumors, such as chondroblastoma, giant cell tumor, nonossifying fibroma, or fibrous dysplasia, and do not show the characteristic translocation.^{6,19}

Clinical presentation typically includes pain or swelling, or both, lasting weeks to years.^{4,19} Pathologic fracture is seen infrequently.¹⁹ Several treatment modalities are commonly used for ABCs. The most effective is complete surgical resection of the lesion, but the risk of functional impairment due to location and lesion size necessitates other options, namely curettage with or without bone grafting or cement packing.^{4,9,18,19} Nonoperative treatments include sclerotherapy and radiotherapy. Radiotherapy has proven effective but is seldom used due to the risk of radiation-induced sarcoma.^{6,9} Recurrence rates vary according to treatment type but may be 40% or higher.¹⁸

The clavicle is an uncommon location for primary tumors in general; thus, ABC of the clavicle is particularly rare. Only a handful of cases have been reported in the literature to date, and many of those come from broader studies that do not explicitly describe the clavicular cases. We present a series of 13 ABCs of the clavicle all treated at a single institution and discuss the presentation, treatment, and recurrence in this site

compared with that of more typically located ABCs, such as those of the femur and tibia.^{6,18,19}

Materials and methods

This retrospective case series looked at all patients treated at an urban hospital for ABC of the clavicle. Patients were identified from an orthopedic oncology database from 1976 to 2015. All patients who were treated at our institution for an ABC of the clavicle were included. Diagnosis of ABC was made by biopsy specimen. Patient records were reviewed to collect sex, age, diagnosis, treatment, and recurrence data. Descriptive statistics were used to present and summarize the data. All data were prepared with STATA 13.1 software (StataCorp, College Station, TX, USA).

Of the 13 patients identified, 7 were male. Mean age at diagnosis was 17 years (range, 8–46 years). Average follow-up time was 42 months (range, 0–237 months). Two patients were lost to follow-up after the primary treatment, and 1 patient was lost to follow-up after the diagnosis.

For the literature review, the PubMed database was searched using the terms “aneurysmal bone cyst” and “clavicle” or “shoulder.” We included articles in English that reported 1 or more cases of ABC arising from the clavicle.

Results

Presentation and diagnosis

Patients presented with a combination of the following symptoms: pain/tenderness (n = 10 [77%]), swelling (n = 10 [77%]), and decreased range of motion (n = 3 [23%]; **Table I**). Time from onset of symptoms to presentation to an orthopedic surgeon ranged from 0 to 12 months. Radiographic imaging showed lytic, expansile lesions in all patients; none were solid (**Fig. 1**). All ABCs in this series were primary lesions.

Table I Patient presentation of aneurysmal bone cyst in the clavicle

Patient	Sex	Age (yr)	Presentation	Location (thirds)
1	M	13	Pain, swelling,* pathologic fracture	Lateral
2	F	11	Pain, swelling, pathologic fracture, decreased range of motion	Lateral
3	M	25	Pain	Medial
4	M	10	Pain	Lateral
5	M	10	Pain, swelling	Medial
6	F	22	Tenderness, swelling	Lateral
7	M	19	Swelling	Lateral
8	M	16	Pain, swelling	Lateral
9	F	12	Pathologic fracture, swelling	Lateral
10	F	8	Pain, swelling, pathologic fracture	Medial two-thirds
11	F	8	Unavailable	Lateral
12	F	27	Pain, swelling, decreased range of motion	Lateral
13	M	46	Pain, swelling, decreased range of motion	Lateral

M, male; F, female.

* Symptom developed after initial presentation but before diagnosis of aneurysmal bone cyst.

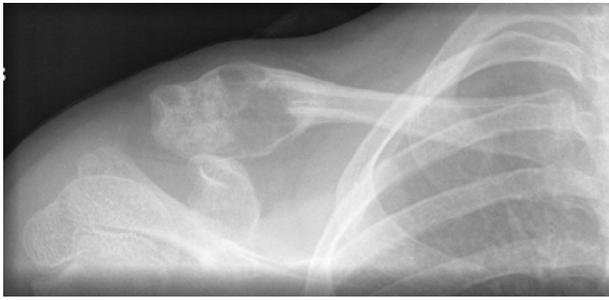


Figure 1 Osteolytic, expansile lesion in the acromial end of the clavicle.

The lesion occurred in the lateral third of the clavicle in 10 patients (77%), medial third in 2 (15%), and the medial two-thirds in 1 (7%). The lesion was located on the right side in 8 patients (62%). Four patients (31%) sustained a pathologic fracture of the clavicle.

The initial diagnosis of ABC was made via x-ray imaging or magnetic resonance imaging. The final diagnosis was made by a specimen from an open biopsy and intralesional resection in the same surgical event in 3 patients (23%). A specimen was obtained from an incisional biopsy or core biopsy before surgical treatment in 9 patients (69%). Information about method of diagnosis was unavailable for 1 patient.

Treatment

Of the 12 patients for which clinical information was available, the lesion was initially treated by curettage alone (n = 5 [42%]), curettage with allograft packing (n = 4 [33%]), curettage with autograft packing (n = 2 [17%]), or partial resection with reconstruction (n = 1 [8%]). No adjuvant phenol or nitrogen was used.

There were 9 total recurrences (Table II): 7 patients (58%) had 1 or more recurrences, and 2 patients had more than

1 recurrence. Average time to recurrence was 6 months (range, 1-17 months). Recurrences were treated with curettage (n = 1 [8%]), curettage with allograft packing (n = 3 [25%]), partial resection of the clavicle (n = 2 [17%]), and excision of the lesion (n = 1 [8%]). All patients who underwent partial resection of the clavicle or resection of the recurrence with negative margins had no subsequent recurrences. The 1 patient who underwent complete resection for the initial procedure did not have a recurrence, and the 2 patients who had resection after a recurrence did not have subsequent recurrence. Two patients were lost to follow-up after recurrence was identified.

Literature review

We identified 26 articles from 1957 to 2015 that mentioned 1 or more cases of ABC of the clavicle, together reporting 62 cases. The 8 articles that focused exclusively on ABC of the clavicle were all single case reports. The largest number of cases from a single study included 10 clavicular ABCs. Ten studies, with a total of 12 cases, reported all of the following data points: age, sex, location within the clavicle, and recurrence. The average age at time of diagnosis was 12.6 years (range, 3-26 years), 10 (80%) were female, and 2 (20%) were male. The medial third of the clavicle was involved in 9 (75%) and the lateral third in 3 (25%). One case reported a recurrence.

Discussion

The clavicle is a rare site for primary tumors, and although most types of lesions have been reported to occur in the clavicle, the infrequency of all complicates the diagnostic process.^{5,17,23,28,31} Because of the diverse histologic possibilities, many of which have clinical and radiologic commonalities, a biopsy specimen is an essential tool for definitive

Table II Recurrence

Patient	Recurrences (No.)	Time to recurrence (mo)	Initial treatment	Treatment for recurrence
1	1	5	Curettage/allograft	Curettage/allograft
2	2	3, 2	Curettage	(1) Curettage; (2) resection of lateral half of clavicle
3	0	—	Curettage/allograft	—
4	2	3, 4	Curettage/allograft	(1) Curettage/allograft; (2) curettage/allograft
5	0	—	Curettage	—
6	1	1	Curettage	Resection of distal three-fourths of clavicle
7	0	—	Curettage/allograft	—
8	1	6	Curettage	Unknown
9	1	17	Curettage/autograft	Unknown
10	0	—	Curettage	—
11	Unknown	Unknown	Unknown	Unknown
12	1	12	Curettage/autograft	Excision of lesion
13	0	—	Resection of lateral clavicle with coracoclavicular reconstruction	—

diagnosis. Although ABC is not a malignant lesion, its aggressive nature often calls for meticulous management. Furthermore, a secondary ABC may mask a primary malignancy.⁶ For these reasons, it is imperative that the orthopedic surgeon be familiar with this uncommon condition and able to recognize when it should be included in the differential for clavicular complaints.

The clavicle differs from other tubular bones structurally and in development. It is the first bone to undergo primary ossification and the last to complete fusion, usually in the early to midsecond decade.^{20,23} Although the sternal and acromial ends both develop epiphyses, the epiphysis at the acromial end is relatively thin, whereas the sternal end is the site of a secondary ossification center and contributes most to growth.^{17,20,23} The clavicle has almost no medullary cavity, and the vascular supply is minimal.²⁸ This could be a contributing factor to the infrequency of clavicular tumors.

The ABCs in our series occurred in the acromial (lateral) end of the clavicle in 77% of patients. Smith et al²⁸ reported 5 of 6 clavicular ABCs arising in the acromial end. Previous studies have identified the acromial end as the most common location for clavicular lesions in general.^{23,28} This raises the question of whether the developmental differences of the acromial end make it more prone to tumorigenesis.

ABCs most commonly present with pain or swelling, or both.⁴ Because the clavicle is subcutaneous in its entirety, even a small amount of swelling is easily detectable.¹⁷ Three of our patients presented with pain before a mass was visible, whereas 8 had a visible mass at the time of presentation. Unfortunately, a painful mass is a clinical finding common to both malignant and benign conditions and offers little diagnostic insight. The onset of pain in 6 of our patients was associated with activity or trauma, and in the cases of sports-related traumas, the ABCs were incidental findings. The differential diagnoses for our patients included fibrous dysplasia, eosinophilic granuloma, brown tumor, unicameral bone cyst, enchondroma, lymphoma, and telangiectatic osteosarcoma. All lesions in this series were de novo and exhibited the classically described radiologic features.

Although most ABCs arise in the first 2 decades, they can be seen much later in life as well. Smith et al²⁸ reported 2 clavicular cases at ages 61 and 63. Our cohort's average age at diagnosis was 17 years; however, 1 patient presented at age 46. This patient was initially treated conservatively for arthritis of the acromioclavicular joint. When no improvement was seen, further workup led to the correct diagnosis of ABC. A complaint of shoulder pain in this age range is highly suspect for mechanical causes or malignancy, but these cases illustrate that although uncommon, ABC should not be excluded from the differential.

The best form of treatment for ABC is still debated.^{3,6,18} Despite the relatively substantial recurrence rate of curettage, with or without grafting or cement (31% in a review of the literature by Cottalorda et al⁶), it is the most commonly used treatment for ABCs.^{3,4,18,19} Adjuvant therapies, including

phenol and nitrogen, have reportedly varied effects, with some data showing them to increase recurrence.¹⁶

Wide resection offers the lowest rate of recurrence but is often avoided due to the issues of functional impairment and need for reconstruction.^{3,4,6,17} However, because the clavicle is an expendable bone that can be resected partially or totally without causing major significant disability, wide resection may be a more feasible option for aggressive ABCs of the clavicle than in other sites,^{4,17,23} especially if the coracoclavicular ligaments can be completely or partially preserved.

Only 2 patients in our series opted for resection after recurrence. The first patient elected to have partial resection of the clavicle at the first recurrence, and the second chose partial resection to treat the second recurrence. Both had undergone curettage without packing. Conversely, resection was recommended in a third patient after 2 recurrences treated by curettage and allograft packing, but the patient elected to curette the lesion once again. Although resection is the most effective way to prevent recurrence, this option may be less appealing to the patient due to the resulting physical deformity. Because many of these patients are young, patients and surgeons alike may be motivated to continue pursuing a method associated with lesser morbidity and physical change.

Average time to recurrence of ABC has been cited between 6 months and 1 year, and almost all occur within 4 years of primary treatment.^{2,4,9,18} In our series, the average time to recurrence was 6 months, with the latest at 17 months. The average follow-up for this study was 66 months, which should be sufficient to capture all recurrences. Mankin et al¹⁸ reported 50% recurrence in 10 cases of clavicular ABC, which was higher than the rate of recurrence at any other site in their study. We documented recurrence in 58% of our patients. It is possible the clavicle is a site of higher recurrence, although there are not enough data to statistically support this conclusion. Recurrence rate is also heavily influenced by treatment method and may reflect the preference of the treating surgeon at a given institution.

The clavicle is a rare site for ABC, which is reflected in its scarce representation in the literature (Table III). ABCs are most commonly reported at the femur and tibia.^{18,19} Although our literature search revealed 26 articles that included at least 1 ABC of the clavicle, the scope of many of these articles was much broader and had little information pertaining to the clavicular cases. As a result, recurrence rate, treatment, and even basic demographic data were unavailable for many of the 62 cases. Average age at diagnosis in the late-second decade agreed with our findings; however, this metric was available for only a fraction of the 62 cases. Furthermore, the 8 publications that focused specifically on clavicular ABCs were all single cases often reporting new or unusual treatment modalities that could not be compared across studies.

This study had several limitations. Owing to the retrospective nature and extensive time period from which records were collected, availability of data and follow-up time varied.

Table III Aneurysmal bone cyst of the clavicle in the literature

Author	Year	Cases (No.)	Location
Sherman et al ²⁶	1957	4	Not reported
Dabska et al ⁷	1969	1	Not reported
Biesecker et al ²	1970	3	Not reported
Ruiter et al ²⁴	1977	6	5 lateral, 1 medial
Hudson ¹⁵	1984	1	Not reported
Herring et al ¹⁴	1986	1	Medial
Smith et al ²⁸	1988	6	5 lateral, 1 distal diaphysis
Barlow et al ¹	1994	1	Not reported
Freiberg et al ⁹	1994	1	Medial
Vedantam et al ²⁹	1996	1	Not reported
Green et al ¹¹	1997	2	Not reported
Hemmadi et al ¹³	1999	1	Lateral
Foerster et al ⁸	2001	1	Middle
Scheil-Bertram et al ²⁵	2004	1	Lateral
Cleeman et al ⁵	2005	1	Medial
Mankin et al ¹⁸	2005	10	Not reported
Kaila et al ¹⁶	2007	2	2 medial
Yadavrao et al ³⁰	2007	1	Not reported
Chadha et al ⁴	2008	1	Medial
Gilbert et al ¹⁰	2008	1	Medial
Kapoor et al ¹⁷	2008	2	1 lateral, 1 medial
Ren et al ²³	2012	9	Not reported
Parashari et al ²²	2012	1	Medial
Shiels et al ²⁷	2013	2	1 lateral, 1 not reported
Heidt et al ¹²	2014	1	Lateral
Yashavnta et al ³¹	2014	1	Medial

Secondly, because of the small sample size, statistical analysis was not appropriate.

Despite the limitations, we present the largest single-institution series to date of a rare condition and provide descriptive information that may be useful in the diagnosis and treatment of future patients with ABC of the clavicle.

Conclusion

ABC of the clavicle is a rare condition that presents with nonspecific symptoms. Although benign, the aggressive nature and potential to be secondary to malignancy calls for timely and accurate diagnosis. The rate of recurrence was high in our series, which may reflect the use of curettage as the primary treatment modality in most patients. The most effective way to prevent recurrence is resection, for which the clavicle is a better candidate than many sites due to its unique anatomy, especially if the coracoclavicular ligaments can be preserved partially or in their entirety. However, because ABC is benign, patients and surgeons may opt for a less aggressive treatment with higher potential for recurrence.

Disclaimer

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