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Primary aneurysmal bone cyst of the scapula in adult patient: two case reports and a review of the literature

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Abstract
Aneurysmal bone cyst (ABC) is a rare, benign but locally aggresive bone tumor of unknown origin tumor. It commonly affects children and usually occurs at the metaphysis of long bones. Scapula is a very rare location and ABCs of the scapula have been sparsely described in the literature. Differential diagnosis can be challenging as it shares common radiological and clinicopathological features with other benign and malignant bone tumors. The degree of diagnostic difficulty increases even more when an unusual tumor site has to be taken into account. Here, we describe rare and challenging cases of a primary ABC located at the scapula that was surgically treated. This is the first case report of ABC involving the scapula in adult patient.

Keywords Aneurysmal bone cyst · Scapula · Glenoid · Adult

Introduction

Aneurysmal bone cyst (ABC) is a rare benign, locally aggressive bone tumors that are commonly seen in skeletally immature patients younger than 20 years [1]. ABCs account for 1% of all biopsied primary bone tumors and are most frequently seen in the long tubular bones especially the femur, tibia and humerus [2, 3]. Scapula is an uncommon location and accounts for only 2.3% of all aneurysmal bone cyst locations [2, 4]. ABCs appear on radiographic images as rapidly growing destructive and expansile lesions with a soap-bubble appearance [4, 5]. The purpose of this study is to present a case of a scapular ABCs in two adult patients and one with glenoidal involvement.

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Case report 1

A 27-year-old female presented to our clinic with a severe pain and swelling over the left scapula, since 3 months. She noticed a swelling over the scapula which was increasing in size and the mass was enlarged fast in the last 3 weeks. There was no history of trauma. The mass was hard in palpation. It was a globular swelling with size 6×5 cm, smooth surface and well-defined margins. The shoulder had limited movements in comparison to the normal side. The patient's ASES and VAS scores were 68 and 8, respectively [6]. Radiographic view of left shoulder showed lytic lesion of the superolateral of the left scapula (Fig. 1). Blood investigations were normal. On fat-suppressed T2 axial magnetic resonance imaging (MRI) sections showed a lesion in the left scapula glenoid inferior with multiloculated fluid–fluid levels. The lesion did not comprise a prominent solid component and transition zone was narrow. Postcontrast T2 coronal sections showed a multiseptated lesion on the left scapula glenoid inferior with peripheral soft tissue enhancement. No enhancement was observed in cystic areas (Fig. 2). Axillary neurovascular structures were normal. The lesion was biopsied and final diagnosis of ABC was rendered. Before the excision of tumor, we performed coil embolisation for devascularisation of tumor to reduce operative blood loss (Fig. 3). Surgical excision performed following the embolisation of ABC (Fig. 4). In the surgical treatment, a transverse incision was made just above the mass at the posterior of the scapula,

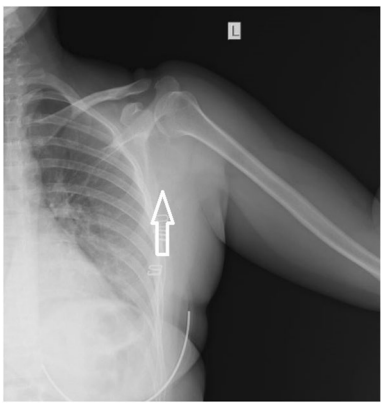

Fig. 1 X-ray view of left shoulder showed lytic lesion of the superolateral scapula with glenoidal involvement

and the mass was reached by excluding the rotator muscles. After intralesional curettage, phenol was used as adjuvant treatment. Arm sling was applied for 4 weeks and with initation of pendulum exercises from the first postoperative day. On histopathological examination, multinucleated giant cells, hemosiderin-laden macrophages and a well-circumscribed lesion with bone around the osteoblastic rim, cystic cavities with blood elements were observed. Also, P63 staining was detected.

Case report 2

A 20-year-old female presented to our clinic with a pain and large swelling in the region of the left scapula for 4 months. There was no history of trauma. It was a globular swelling with size 6 × 3 cm, smooth surface and well-limited margins. The shoulder had limited movements in comparison to the normal side. The patient's ASES and VAS scores were 65 and 7, respectively. No axillary or cervical lymph nodes were palpable. Radiographic view of left shoulder showed lytic and expansile lesion of the superomedial of the left scapula (Fig. 5). An MRI showed well-defined heterogeneous mass with septation and multiple fluid–fluid levels. Multi-loculated lesion had cortical expansion and partial destruction on the level of supraspinatus fossa (Fig. 6). The lesion was biopsied by

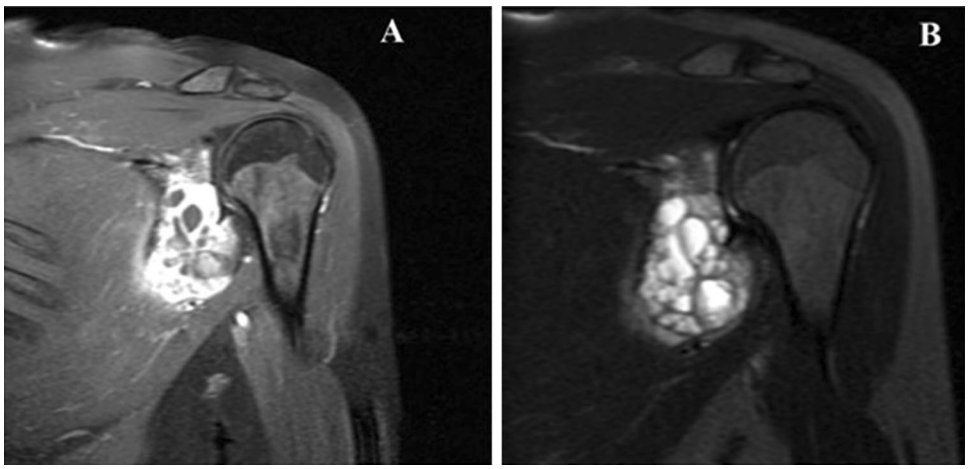

Fig. 2 **a** Contrast enhancement in peripheric soft tissue in inferior glenoid of left scapula in T1 postcontrast coronal sections, multiseptated lesion with none contrast enhancement in cystic areas. **b** The lesion not having solid component and with narrow transition zone having multiloculated liquid–liquid levels in left inferior glenoid of scapula in fat-suppressed T2 coronal sections. (**a** TR:3424.0/TE:90.0, **b** TR:538.0/TE10.0)

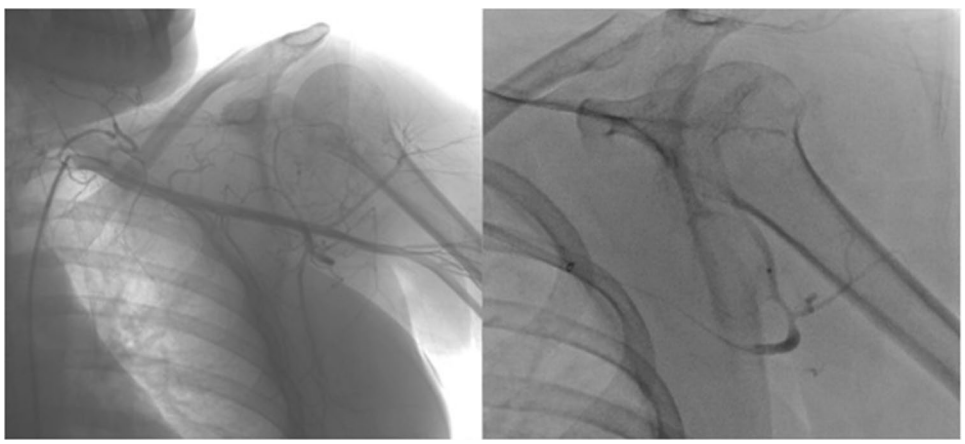

Fig. 3 Angiography showed coil embolisation for devascularisation of tumor to reduce operative blood loss. The area without having blood supply in DSA in left inferior glenoid of scapula

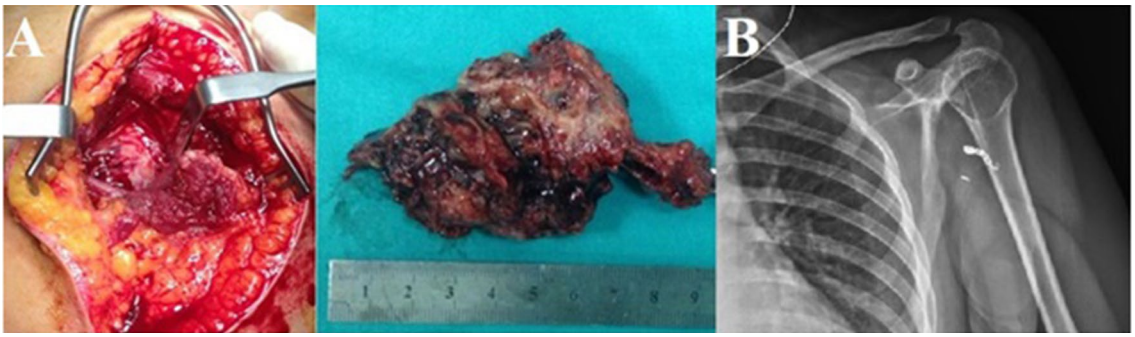

Fig. 4 **a** Intraoperative appearance of tumor and gross appearance of excision material. **b** The image after the excision operation

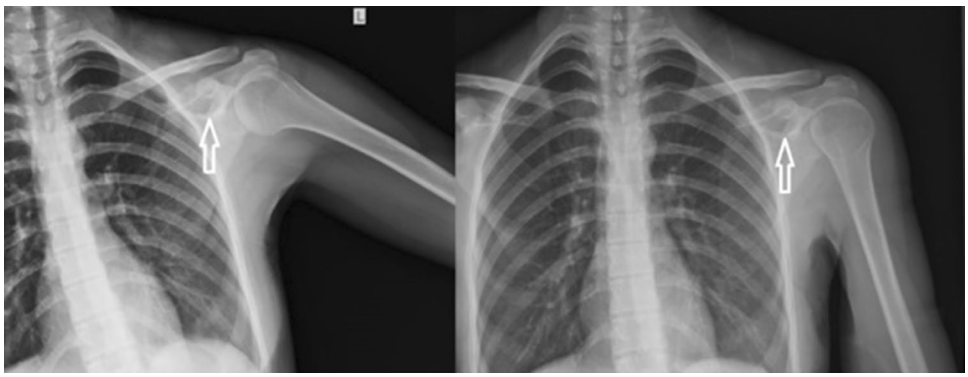

Fig. 5 X-ray view of left shoulder showed lytic and expansile lesion of the superomedial of the left scapula

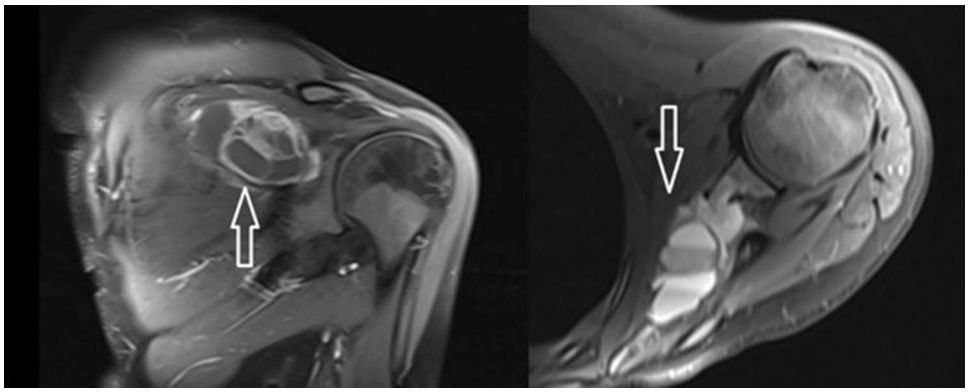

Fig. 6 **a** Postcontrast coronal T1-weighted MR image revealed a expansile hypointense multicystic lesion with fluid/fluid levels. (TR:660.0/TE:11.0). **b** Axial T2-weighted MR image showing a well-defined expansile, multicystic, hiperintense lesion with blood/fluid levels (TR:3200.0/TE:47.0).

tru-cut method and final diagnosis of ABC was rendered. In the surgical treatment, a transverse incision was made through the posterior of the scapula and directly above the mass. Surgery involved intralesional curettage through a cortical window large enough over the lesion followed by high-speed power burring, brushing and pulse lavage of the lesion (Fig. 7). Arm sling was applied for 4 weeks and with initiation of pendulum exercises from the first postoperative day. In the examination of the histopathological sections, fibroblastic and vascular proliferation followed by reactive bone formation in the environment, cystic spaces in which septa-like structures were found and solid are as dominated by fibrohistiocyte proliferation, multinuclear giant cells rarely chronic inflammatory cells and stromal spindle cells were observed.

Patients were followed-up every 2 weeks in first 2 months. The patients underwent a control MRI for relapse at 3, 6 and 12 months. Six months postoperatively, shoulder movements were similar to those of the normal side. After 1 year, case 1 was painless with full range of shoulder movement with no relapse (Fig. 8). Case 2 had moderate pain and minimal abduction limitation with no relapse. ASES and VAS scores of the patients at the first-year follow-up were 92–1 and 87–4, respectively.

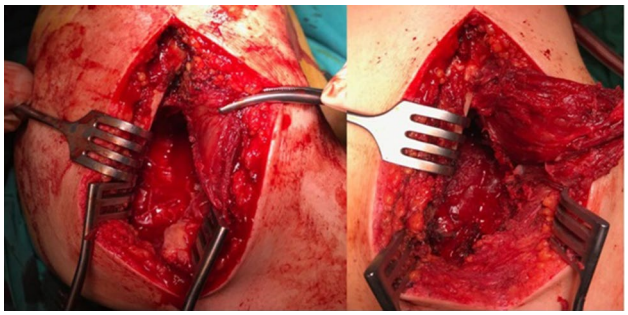

Fig. 7 Intralesional curettage performed through a cortical window over the lesion. Followed by adjuvant therapy with high-speed power burr

Fig. 8 Preoperative abduction limitation and 1-year follow-up showing good function with painless good range of motion of left shoulder

Discussion

ABCs are rare benign, locally aggressive bone tumors that are first described by Jaffe and Lichtenstein in 1942 [1, 7]. They represent only 1% of all primary bone tumors. Most patients are under 20 years of age. ABCs are most common during the first decades of life, with a reported median age ranging from 10.2 to 13 years [8, 9]. Leithner et al. [10] reviewed 1096 patients in 1999 and they report primary ABCs were found to have an estimated annual incidence of 0.14 per million individuals with a slight male predominance below the age of 14 years.

ABC can exist primary neoplasm or as a secondary lesion to an underlying a primary tumor such as chondroblastoma, giant cell tumor, fibrous dysplasia, osteoblastoma and osteosarcoma [9]. Therefore, pre-operative evaluation and histopathologic studies are required to make sure that the ABC is not secondary. ABCs' diagnosis is based on history, physical examination and radiologic studies. ABCs located in the scapula are usually present with pain and swelling [11]. In our two cases, the complaints were pain and swelling. In the long bones, the radiographic appearance of ABC is metaphyseal eccentric or concentric lesion with elevating the periosteum as a blowout and eroding the cortex [2, 12]. MRI is useful in evaluating intramedullary extension of the lesion and soft tissue involvement [12]. Radiological imaging findings of our cases were consistent with the literature.

The pathogenesis of ABC is controversial. Some have been considered as reactive process resulting from a local increased venous pressure leading to a dilated vascular bed within the involved bone [13]. In recent years, a novel translocation of *t*(X; 9) (q26–q32) was described in a solid variant of ABC [14].

For ABC, it is necessary to determine an algorithm based on the literature reviewed; first of all, in recent genetic and immunohistochemical studies, we should note that primary ABC is a tumor and not a reactive tumor mimicking lesion. Direct radiography and imaging examinations following physical examination; even CT scan and MRI sometimes do not provide a clear diagnostic criterion for the diagnosis of ABC. Differential diagnosis between ABC and unicameral bone cyst (UBC) is sometimes not clear in radiological imaging methods. Double density fluid level, septation, low signal on T1 images and high intensity on T2 images strongly suggest the bone cyst is an ABC, rather than a UBC. Performing a biopsy is mandatory before any treatment. Treatment methods are varying widely in the literature. General treatment methods include curettage, resection, intracystic injections and embolization [3].

The scapula is an uncommon location for ABC [1, 2]. Scapular primary ABCs are very rarely reported. For the literature review, the PubMed database was searched using the terms "aneurysmal bone cyst" and "scapula" and "glenoid". There were eight articles from the beginning of year 1985. All nine patients from those eight articles were under 16 years of age [2, 15–21]. We consider the following three reasons why our cases are seen at an older age when compared with the literature: the lesion has developed in the usual age range, has been silent for years and has given symptoms in adulthood. Or this is because it may also be seen in adulthood in a rare localization such as scapula, and this may be the first time we identified it. Third, it may be that the tumor changes character and that we are detecting the first examples.

Patients with ABCs typically present with pain and/or swelling [22]. Pathological fracture is seen rarely [22]. Many treatment modalities have been described for ABCs. The most effective approach is complete surgical excision. If the risk of functional impairment due to location and lesion size requires other choices, curettage with bone grafting or cementation may be performed [5, 22, 23]. Because of the high risk of recurrence after curettage alone, some

Table 1 Updated review of the literature about primary aneurysmal bone cyst of the scapula

References	Year	Article title	Age/sex
Chang et al. [16]	2016	Treatment of aneurysmal bone cysts by percutaneous CT-guided injection of calcitonin and steroid	13 years, boy
Shiels WE 2nd et al. [17]	2013	Percutaneous doxycycline treatment of aneurysmal bone cysts with low recurrence rate: a preliminary report	8 years, girl
Beslikas et al. [2]	2012	A giant scapular aneurysmal bone cyst in a child	7 years, boy
Mavrogenis et al. [18]	2011	Aneurysmal bone cyst of the acromion treated by selective arterial embolization	8 years, boy
Megas et al. [15]	2009	Aneurysmal bone cyst of the scapula. A case report	15 years, boy
Kaila et al. [19]	2007	Aneurysmal bone cyst of the paediatric shoulder girdle: a case series and literature review	12 years, girl 14 years, girl
Ruggieri et al. [21]	1995	Aneurysmal bone cyst of the acromion: a case report	6 years, girl

authors reduced the rate of local recurrence using a variety of additional treatment methods, such as phenol, polymethylmetachrylate, liquid nitrogen, or the use of high-speed mechanical burr [3]. Also, selective arterial embolization is an effective treatment. It is commonly used preoperatively to reduce the vascularity of tumor [24]. It can be performed together with surgery or as a treatment alone. Advantage of this procedure include that it does not prevent the possibility of surgical intervention in case of failure and the complications of procedure are ischemia to neural structures and vital structures, such as viscera. In two of our cases, we achieved successful results similar to the literature and did not encounter any complications related to treatment in at least 1 year follow-up after curettage, phenol, burr and embolization applications.

Jamshidi et al. [25] reported that their clavicular ABC patients who underwent curettage had better functional outcomes but more relapse rates than patients treated with segmental resection. The functional scores of our cases which we performed curettage were also good.

Cottalorda and Bourelle [3] reported the recurrence rates of the treatment of ABCs in 2007. According to their study, wide resection showed the lowest rates (4%) for recurrence and curettage and bone grafting has the greatest probability for recurrence (30.6%). Basarir et al. [26] presented a retrospective analysis of 56 ABCs and they report the recurrence rate was 16%. De silva et al. [4] reported that 82% of recurrences are seen in first 1 year of treatment.

Differential diagnosis consists of giant cell tumor, chondromyxoid fibroma and telangiectatic osteosarcoma [15, 27]. The distinction is very challenging, however, patient features contribute very much to it. The treatment choices are curettage and grafting or curettage and cementation [28]. In difficult sites for surgery, selective arterial embolization is an effective option. In less functional bones such as clavicle or fibula and in eccentric lesions, en bloc resection is the treatment of choice.

An updated review of the literature about primary aneurysmal bone cyst of the scapula is provided in Table 1.

In conclusion, scapular aneurysmal bone cysts are rare. According to other common localizations, it is more difficult to diagnose the scapular localizations. ABC should be kept in mind and should be considered when a scapular swelling is faced. It typically occurs in childhood. The most commonly used method of treatment is curettage. Periodic follow-up with MRI is recommended in the postoperative period.

Compliance with ethical standards

Conflict of interest The authors declare that they have no conflicts of interest.

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