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GENERAL REVIEW

Rare aneurysmal bone cysts: multifocal, extraosseous, and surface variants

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Abstract
Multifocal, extraosseous, and surface aneurysmal bone cysts are rare variants of the primary lesions. The clinicopathological features are similar, and the optimal treatment is surgical. Although local recurrences may occur, the prognosis is excellent. This review article introduces the readers to a rare diagnosis which they may have been previously unfamiliar with, presents the clinicopathological and imaging features of these rare aneurysmal bone cyst variants, and discusses their diagnosis and treatment. The clinicians who treat patients with aneurysmal bone cysts should be familiar with these uncommon entities and their differential diagnosis.

Keywords Aneurysmal bone cyst · Multifocal · Extraosseous · Surface · Periosteal · Soft tissue

Introduction

Aneurysmal bone cysts (ABCs) are relatively rare neoplastic bone lesion that most commonly occur in patients during their second decade of life. They may affect any bone but usually arise in the metaphysis, cause thinning of the cortex, and eventually protrude from the bone. Recurrence is not uncommon, ranging from 10 to 50%. Secondary ABCs arising in association with other benign or malignant bone tumors have been reported, accounting for approximately 1/5 of the cases [1–13].

There are conflicting reports with respect to whether ABCs represent a neoplastic or a reactive process [14–20]. ABCs were originally thought to be reactive lesions caused by venous hypertension leading to vascular dilatation [21–24]. However, several studies have documented that ABCs are neoplastic lesions in nature [14, 15, 19, 21, 25–29]. Primary ABCs are characterized by recurrent translocations involving the USP6 gene that encodes the ubiquitin-specific peptidase 6 (USP6), on chromosome 17p13. The most common translocation observed in ABCs {t(16;17)(q22;p13)} results in an aberrant expression of USP6 that drives subsequent tumor growth. Other lesions have also shown translocations involving chromosome 16, independent of chromosome 17 [14, 19], but occasionally with partners from other chromosomes such as 1, 2, 6, and 11 [15]. Recently, the X;9 translocation [18] and the USP9X translocation as a novel USP6 fusion partner were discovered [17].

Capanna et al. [1] described 5 types (I–V) of ABCs based on their morphological characteristics on radiographs. Type I includes a central metaphyseal ABCs, type II includes a central ABCs involving the entire segment of bone, type III includes an eccentric metaphyseal ABCs, type IV includes a surface (subperiosteal) ABCs, and type V includes a meta-diaphyseal ABCs [1, 30]. Types I–III in their classification represent ABCs of medullary origin,

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and type V is described as subperiosteal that breaks the cortex and develops both peripherally and toward the center of the bone; a cortical ABC is not classified in this classification [30].

Rarely, ABCs originate in the surface of the bone (cortical or periosteal) or in the soft tissues (extraosseous), and occasionally they may involve multiple bones (multifocal) [31–57]. To enhance the literature, this review article introduces the readers to a rare diagnosis which they may have been previously unfamiliar with, presents the clinicopathological and imaging features of these rare ABCs variants, and discusses their diagnosis and treatment.

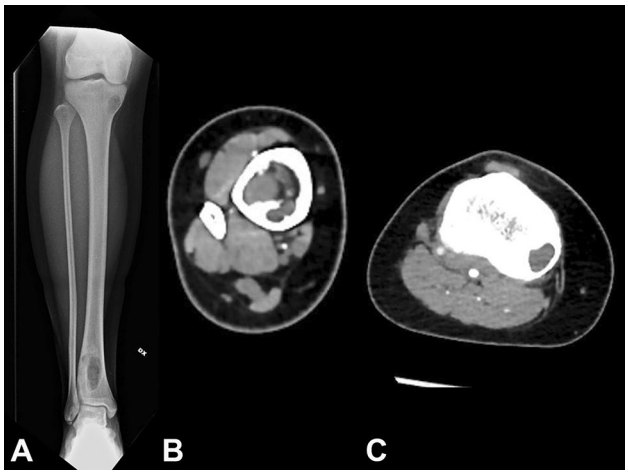

Fig. 1 **a** Anteroposterior radiograph of the right tibia of a 15-year-old girl with multifocal ABCs at the proximal and distal right tibia. The patient was diagnosed with the distal tibia ABC 4 years before and was treated with curettage and bone grafting. At present admission, CT scan shows **b** recurrence of the distal tibia lesion and **c** the second lesion with similar characteristics at the proximal tibia. Both lesions were treated with curettage and bone grafting after biopsy

Multifocal ABCs

Multifocal ABCs are characterized by the occurrence of multiple lesions within the same bone (mono-ostotic) or within multiple bones (poly-ostotic) [53–57]. The first case of metachronous multifocal ABCs in a skeletal location different from that of the primary site was described in 1997 by Sundaram et al. [57]. Since then, including the two cases presented herein (Figs. 1, 2), seven cases with multifocal ABCs were reported in the related literature (Table 1) [53–57]. Male patients aged 2–44 years were more commonly affected [53–57]. The pathogenesis of multifocal ABCs is unknown [15, 16].

Pathology

In the reported multifocal ABCs, histological sections showed blood-filled honeycombed dilated vascular beds separated by fibrovascular membranes consisting of a moderately dense cellular proliferation of bland fibroblasts, with scattered, multinucleated, osteoclast-type giant cells and reactive woven bone or osteoid rimmed by osteoclast. The woven bone frequently followed the contours of the fibrous septa. Numerous mitoses were commonly present; however, atypical mitoses were absent. The cystic spaces had endothelial-like CD32-negative cells. Using anti-p35 antibody, approximately 5% of the cell nuclei were immunohistochemically stained. Giant osteoclast-like cells were predominantly located in the areas of extravasated red blood cells [56].

Clinical presentation

Clinical presentation was more common with pain and/or a pathological fracture. Occasionally, pain was associated with swelling, tenderness, or a palpable mass. The duration of symptoms ranged from some weeks to 8 months.

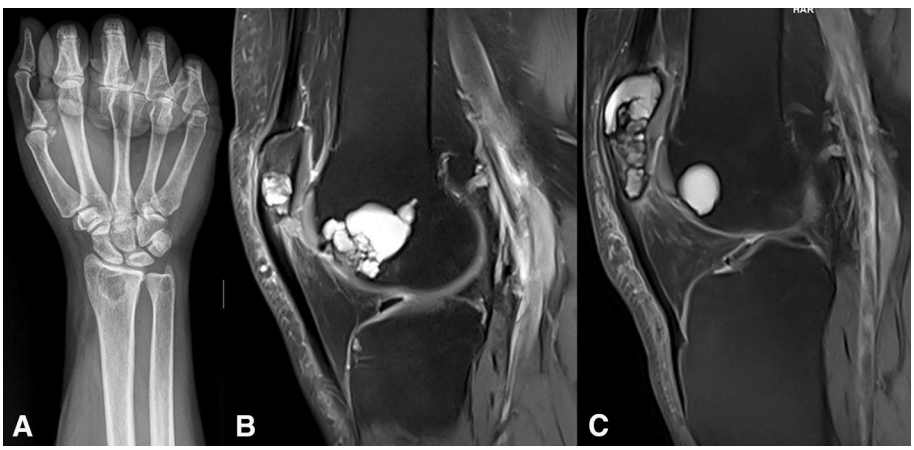

Fig. 2 **a** Anteroposterior radiograph of the right wrist, **b** T2-weighted MR imaging of the left knee and **c** T2-weighted MR imaging of the right knee of a 44-year-old man with multifocal ABCs

Table 1 Summary of the most important published studies on multifocal ABCs

Study	Age (years)/ gender	Location	Treatment	Recurrence	Follow-up
Amer et al. [53]	9/M	Scapula and proximal tibia	Doxycycline, curettage, and bone grafting	Not reported	3 years
Donigan et al. [54]	18/M	Humerus and T8 (pathological fracture)	Curettage and bone grafting (humerus); resection and bone grafting (T8)	No	1.5 years
Niemeier et al. [55]	14/F	Proximal femur and elbow	Curettage and bone grafting	Yes (femur), at 4 months (repeat curettage and adjuvants)	2.5 years
Scheil-Bertram et al. [56]	2/M	Proximal humerus, clavicle, left distal radius, and right distal radius	Curettage	Yes, at age 3 and 7 years (repeat curettage)	10 years
Sundaram et al. [57]	9/M	Tibia (recurrent) and pubic ramus	Curettage and bone grafting (tibia); embolization and resection (pubic ramus)	No	Not reported
Current report	44/M	Femoral condyles (bilateral), patellae (bilateral), proximal tibia (right), and distal radius (right)	Curettage and bone grafting (scheduled)	-	-
	15/F	Distal tibia and proximal tibia	Curettage and bone grafting	No	1 year

The proximal humerus was the most commonly involved bone followed by the tibia, radius, proximal femur, clavicle, elbow, pubic ramus, scapula, and vertebra [53–57].

Imaging

Radiographs showed radiolucent eccentric expansile lesions emanating from the cortex with well-defined sharp margins that were sclerotic. The tumors contained a thin shell of subperiosteal reactive bone. Computed tomography (CT) scan showed a periosteal location of the epicenter of the lesion with a pressure effect and erosion of the cortical surface (scalloping), and an incomplete shell-like periosteal reaction partially mineralized. The lesions had a low internal density, protruded from the bone cortex and were composed of soft tissue surrounded by a thin osseous layer [53–57]. Magnetic resonance (MR) imaging showed the typical fluid–fluid levels (blood and serum levels) on T1-weighted images. T2-weighted images showed a complete rim of low signal intensity as well as fluid–fluid levels with hypointense signal intensity of the dependent fraction and hyperintense signal intensity of the non-dependent fraction. Contrast-enhanced T1-weighted images showed enhancing cyst walls and internal septations as well as absence of larger portions of solid material. Localized edema of adjacent soft tissues was present in all cases [53–57].

Differential diagnosis

The differential diagnosis of multifocal ABCs should include unicameral (simple) bone cyst that arises in patient under 20 years of age in the metaphysis of a bone, fibrous dysplasia that could present with a multiloculated and ground-glass appearance, intraosseous lipoma with well-defined borders and fat tissue signal intensity, chondroblastoma that arises in the epiphysis and has sclerotic margins and occasionally calcifications, and telangiectatic osteosarcoma composed of cystic cavities containing necrosis and hemorrhage [53–57].

Treatment

In the reported multifocal ABCs, treatment was curettage or resection and bone grafting. Prophylactic osteosynthesis with a plate and screw may be necessary for stability of the bone until healing. Preoperative percutaneous intralesional chemoablation with doxycycline (280–370 mg) [53] and selective arterial embolization [57] followed by curettage and bone grafting have also been reported.

Prognosis

All reported cases had no evidence of ABC at 12 months to 10 years follow-up. Two patients experienced local recurrences [55, 56]; a 2-year-old boy had 2 recurrences at the age of 3 years and 7 years and developed multiple ABCs

over the 10-year follow-up [56]. This patient also had multiple complex cardiovascular malformations including aortic isthmus stenosis, hypoplastic thoraco-abdominal aorta, and bilateral renal artery stenosis that may be combined in a specific phenotypic condition.

Extraosseous ABCs

The first case of an extraosseous ABC in a soft tissue location different from that of the primary site was described in 1972 by Salm and Sissons [47]. They described the lesion as a giant cell tumor of the soft tissues, with pathologic features remarkably similar to aneurysmal bone cysts, indicating that, unbeknownst to the authors, these cases may have been the first cases of extraosseous ABCs to be described in the literature [21, 36, 47]. Since then, 25 cases with extraosseous (soft tissue) ABCs were reported in the related literature (Table 2). Female patients aged 3–57 years were more commonly affected [33–52, 58, 59]. The pathogenesis, age, gender, racial predilection, and histology of extraosseous ABCs were similar to the bone lesions [14–19].

Pathology

In the reported cases, histological sections showed blood-containing cysts that were separated by multiple internal fibrous septa of valuable thickness composed of spindle fibroblasts, occasional multinucleated osteoclast-like giant cells along with hemosiderin containing set in a collagenous stroma, features characteristic of an ABC. No endothelial or epithelial lining, neither significant atypia was observed. Fluorescence in situ hybridization (FISH) using break-apart probes clearly showed a USP6 rearrangement, confirming the diagnosis of ABC [33, 35–52, 58].

Clinical presentation

Clinical presentation was more common with pain and swelling, occasionally with a palpable growing mass. The

Table 2 Summary of the most important published studies on extraosseous ABCs

Study	Age (years)/ gender	Location	Type	Treatment	Recurrence	Follow-up
Pietschmann et al. [21]	26/F	Thigh	Extraosseous	Excision	No	36 months
	38/M	Upper arm	Extraosseous	Excision	No	29 months
Ajilogba et al. [33]	12/F	Thigh	Extraosseous	Excision	No	Not reported
Amir et al. [35]	15/F	Groin	Extraosseous	Excision	No	2 years
Baker et al. [36]	41/F	Upper arm	Extraosseous	Excision	No	3 years
Hao et al. [37]	10/F	Shoulder	Extraosseous	Excision	No	8 months
Karkuzhali et al. [38]	23/M	Fibula	Extraosseous	Resection	No	Not reported
		Thigh	Extraosseous	Excision		
Lopez et al. [40]	26/F	Thigh	Extraosseous	Excision	Yes, at 5 months (re-excision)	2 years
Lòpez-Barea et al. [41]	57/F	Arm	Extraosseous	Excision	No	18 months
Nielsen et al. [43]	8/M	Shoulder	Extraosseous	Excision	Yes (re-excision)	16 months
	29/F	Groin	Extraosseous		No	4.5 years
	3/F	Upper arm	Extraosseous		No	9 months
	28/M	Upper arm	Extraosseous		No	10 years
	30/F	Thigh	Extraosseous		No	Not reported
Petrik et al. [44]	7/M	Common carotid artery	Extraosseous	Excision	No	Not reported
Rodriguez-Peralto et al. [45]	20/F	Shoulder	Extraosseous	Excision	No	2 years
Sahu et al. [46]	12/F	Palm	Extraosseous	Excision	No	24 months
Salm et al. [47]	32/M	Thigh	Extraosseous	Excision	Not reported	Not reported
	45/F	Abdominal wall	Extraosseous		Yes, at 2 months (re-excision)	10 years
Samura et al. [48]	51/F	Pelvis	Extraosseous	Excision	No	14 months
Shannon et al. [49]	29/F	Retroclavicular space	Extraosseous	Excision	No	18 months
Sukov et al. [50]	11/M	Thigh	Extraosseous	Excision	Not reported	Not reported
	36/F	Thigh	Extraosseous			Not reported
Wang et al. [51]	21/M	Thigh	Extraosseous	Excision	No	8 months
Ellison et al. [58]	10/F	Thigh	Extraosseous	Excision	No	13 months

duration of symptoms ranged from a few weeks to 2 years. The thigh was the most common locations followed by the upper arm, tibia and fibula, shoulder, groin and pelvis, carotid artery, abdominal wall, toe, and palm [33, 35–52, 58].

Imaging

The MR imaging appearance of extraosseous ABCs was identical to bone ABC. Septations usually had low T1 and T2 signal as a result of their fibrous (and sometimes calcified) nature and enhanced on post-contrast images, producing a "honeycomb"-type appearance. The extraosseous ABCs capsule/margin should be well defined and low in signal from its calcified, sclerotic rim [36, 37]. A mature extraosseous ABCs with a calcified periphery should produce the characteristic "doughnut" sign of bone ABCs on bone scintigraphy as a result of osteoblastic activity in the calcified rim and central photopenia from central cystic spaces [60]. Extraosseous ABCs may have a connection with the bone but there is no significant periosteal reaction neither a pressure effect nor erosion of the cortical bone [36, 37].

Differential diagnosis

The differential diagnosis of extraosseous ABCs should include other benign or malignant soft tissue tumors including hematoma, myositis ossificans, myxoma, giant cell-rich tumors of the soft tissue, giant cell tumor of the tendon sheath, brown tumor of hyperparathyroidism, Morel–Lavallée lesions, intramuscular hemangioma, arteriovenous malformations, periarticular calcinosis that shows fluid–fluid levels but also calcium deposits, soft tissue leiomyoma, clear cell hidradenoma, synovial sarcoma, malignant fibrous histiocytoma, myxoid sarcoma, and extraskeletal telangiectatic osteosarcoma [33, 35–52, 58, 61].

The differential diagnosis of extraosseous ABCs from giant cell tumor of soft tissue is particularly difficult. Both can occur at any age, although giant cell tumors usually occur in those over 20 years of age, while extraosseous ABCs usually occur in patients under 20 years. Radiographically, giant cell tumors of soft tissue do not tend to have a calcified rim and they rarely exhibit cystic changes, necrosis, and/or hemorrhage with formation of fluid–fluid levels [62, 63]. Extraosseous ABCs and mature myositis ossificans appear very similar on radiographs and CT; both entities feature a thin rim of ossification and a lucent/hypodense center [33, 37]. However, in myositis ossificans, clinical history often yields an antecedent history of trauma, fluid–fluid levels, and honeycomb septations enhancement should not be seen, and its imaging features vary with the age of the lesion; as the lesion matures in its later stages, MR imaging contrast enhancement will no longer be present [33, 37].

Treatment

In the reported extraosseous ABCs, treatment was excision or resection. Three of the reported cases experienced local recurrences [40, 43, 47]. All patients with a local recurrence were treated with re-excision, without any evidence of ABC re-recurrence at their last follow-up.

Surface ABCs

Surface ABCs extend beyond the confines of the outline of the bone and arise within the cortex of the bone or beneath the periosteum, limited by periosteum externally and endosteum internally [64–66]. They were initially referred to as parosteal type in the radiographic classification of ABCs [67] and were named subperiosteal giant cell tumor [31] or subperiosteal osteoclasia [32]. In the radiographic classification developed by Capanna et al. [1, 30], surface ABCs were described as types IV and V. According to this classification, type IV lesions include subperiosteal forms that develop away from the bone so that the cortex is either intact or superficially eroded. Type V lesions develop both peripherally toward the periosteum and centrally toward the medulla so that the cortex is penetrated [30, 64–66]. Maiya et al. [64] described a cortical metaphyseal ABC that showed similar degrees of intraosseous and extraosseous extension and tended to be more aggressive than subperiosteal ABCs. They also described a mixed pattern ABC that was not possible to distinguish their precise origin in that they either presented with features suggestive of one pattern and then developed features of the other. Fifty cases of surface ABCs have been reported in the related literature (Table 3) [34, 39, 42, 52, 64–66]. In our practice, we have treated seven patients with surface ABCs; six patients had periosteal ABCs (Figs. 3, 4), and one patient had a mixed pattern ABC (Fig. 5), as previously reported by Maiya et al. [64].

Imaging

Radiographs showed a round periosteal lesion with well-defined circumscribed borders with or without peripheral calcification (Fig. 6). CT scan showed an abnormality of the soft tissue protruding from the bone cortex with a thin rim and a density, suggesting the presence of a calcified mass and slightly irregular wall. The erosion of the cortex did not extend to involve the medulla [31, 34, 39, 52, 64–66]. MR imaging showed a well-defined circumscribed lesion sometimes scalloping the outer cortex without breaching it. The peri-tumoral soft tissue showed significant swelling and enhancement along the periosteum with blood-filled spaces of varying sizes with numerous fluid–fluid levels, T1/T2 hypointense septa, and T1/T2

Table 3 Summary of the most important published studies on surface ABCs

Study	Age (years)/gender	Location	Type	Treatment	Recurrence	Follow-up
Alrayes et al. [34]	29/F	Tibia	Periosteal	Excision	No	2 years
Kobayashi et al. [39]	7/M	Humerus	Periosteal	Resection	No	3 years
Muller et al. [42]	39/M	Toe	Periosteal	Amputation (distal phalanx)	No	Not reported
Woertler et al. [52]	13–55/F (four patients), M (two patients)	Femur (3 patients), tibia (2 patients), and humerus (1 patient)	Periosteal	Not reported	Not reported	Not reported
Maiya et al. [64]	3–41/F (16 patients), M (seven patients)	Forearm bones, tibia, femur, fibula, hand bones, and clavicle	Periosteal, cortical, mixed (intra- and extraosseous)	Curettage	No	Not reported
Van Royen et al. [65]	19/M	Femur	Periosteal	Curettage	Not reported	Not reported
Yalcinkaya et al. [66]	11–44/F (five patients), M (five patients)	Femur (7 patients), tibia (2 patients), and humerus (1 patient)	Periosteal	Curettage	No	98.4 months
Current report	16/F	Humerus	Periosteal	Embolization	No	7 years
	29/F	Humerus	Periosteal	Curettage	No	2 years
	17/F	Pelvis	Periosteal	Embolization	No	5 years
	13/F	Humerus	Periosteal	Curettage	No	5 years
	17/M	Fibula	Periosteal	Curettage	Unknown	Lost to follow-up
	28/F	Humerus	Periosteal	Sclerotherapy	No	5 years
	19/F	Tibia	Mixed (intra- and extraosseous)	Excision and curettage	No	2 years

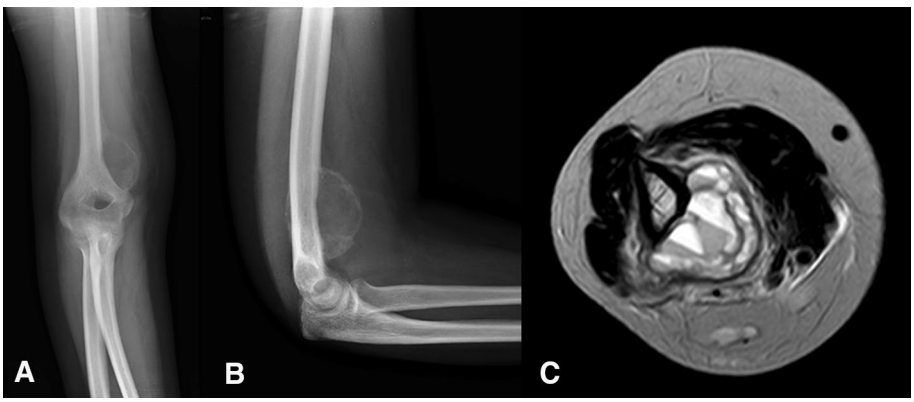

Fig. 3 **a** Anteroposterior and **b** lateral radiographs and **c** axial T1-weighted MR imaging of the right elbow of a 29-year-old woman with a surface (periosteal) ABC of the distal humerus. The lesion was treated with curettage after biopsy, without any evidence of recurrence at the last follow-up

hypointense calcified rim with some septal enhancement. In contrast, the lesions showed a predominantly high signal intensity with well-defined margins on T2-weighted images; the signal intensity of the intralesional septa was low to intermediate, while the signal intensity of the separated areas was high [31, 34, 39, 52, 64–66].

Diagnosis

The differential diagnosis of surface ABCs should include ossifying subperiosteal hematoma, periosteal chondroma, periosteal ganglioma, subperiosteal giant cell reparative granuloma, osteoid osteoma, intracortical hemangioma,

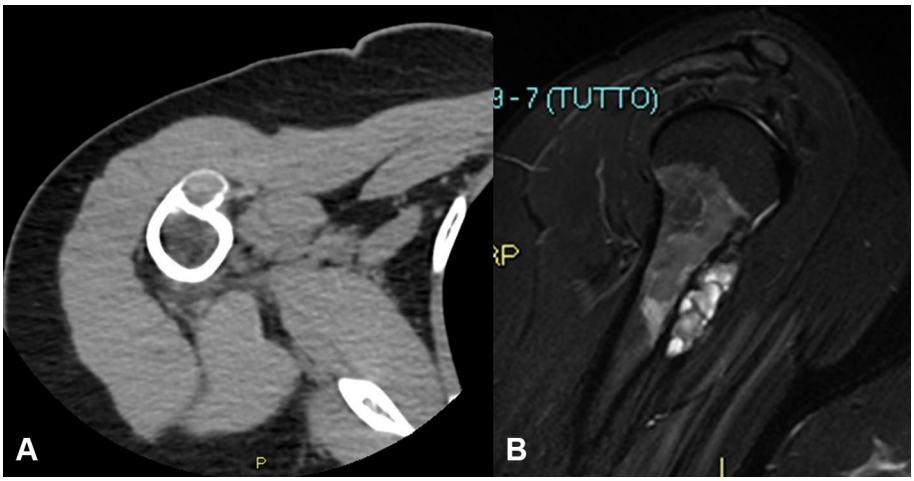

Fig. 4 **a** CT and **b** sagittal T2-weighted MR imaging of the right arm of a 17-year-old girl with a surface (periosteal) ABC of the proximal humerus. The lesion was treated with embolization after biopsy, without any evidence of recurrence at the last follow-up

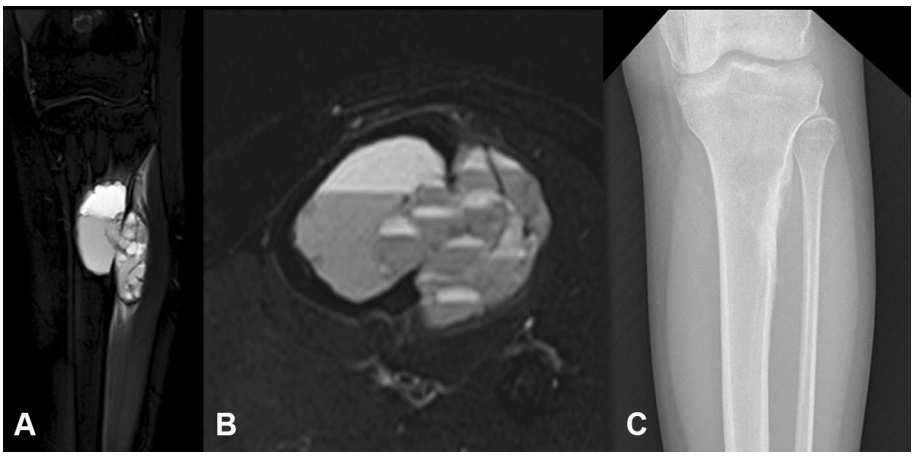

Fig. 5 **a** Coronal and **b** axial T2-weighted MR imaging of the left tibia of a 19-year-old girl with a mixed pattern (intra- and extraosseous component) ABC of the proximal tibia metaphysis. The lesion was treated with curettage after biopsy, **c** without any evidence of recurrence at the last follow-up

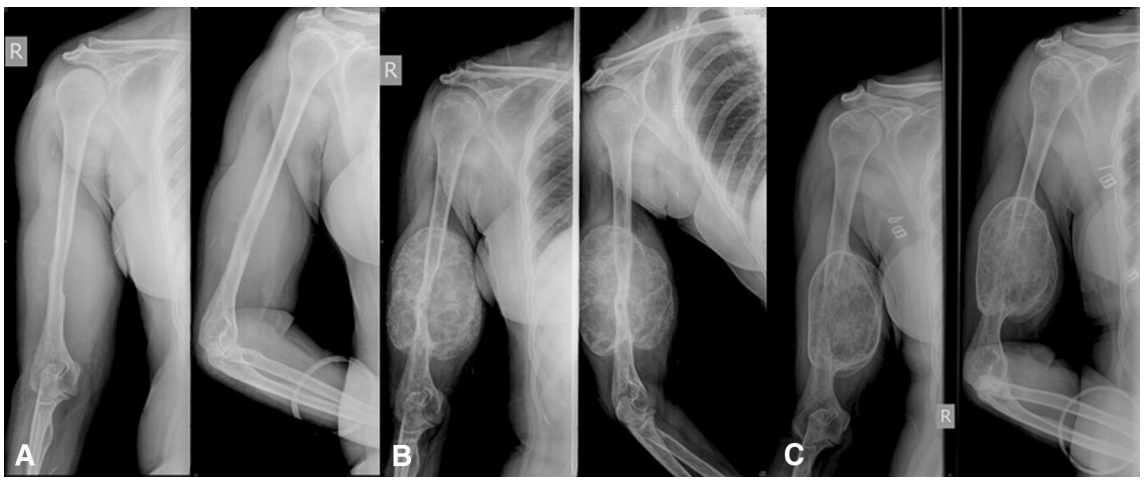

Fig. 6 **a** Anteroposterior and lateral radiographs of the right humerus of a 28-year-old woman with a periosteal ABC. The lesion was treated with sclerotherapy after biopsy, and the patient was followed only with radiographs. **b** Anteroposterior and lateral radiographs of the right humerus 6 months and **c** 5 years after sclerotherapy show complete ossification of the bone lesion

post-traumatic cysts, Ewing's sarcoma, periosteal osteosarcoma, low-grade intracortical osteosarcoma, high-grade surface osteosarcoma, telangiectatic osteosarcoma, and periosteal metastatic lesions such as from primary lung cancer [64–66].

Treatment

In the reported surface ABCs, treatment was curettage, excision, or resection. In one patient with involvement of the distal phalanx of a toe, amputation of the involved phalanx was done [26]. None of the reported cases had any evidence of local recurrence at their last follow-up [34, 39, 42, 52, 64–66].

Conclusion

Multifocal, extraosseous, and surface ABCs are rare variants of primary bone ABC. The clinicopathological features are similar, and the optimal treatment is surgical. Although local recurrences may occur, the prognosis is excellent. The clinicians who treat patients with ABC should be familiar with these uncommon presentations and their differential diagnosis.

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Compliance with ethical standards

Conflict of interest All authors declare that they have no conflict of interest.

Ethical approval All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki Declaration and its later amendments or comparable ethical standards.

Informed consent Informed consent was obtained from all individual participants included in the study.

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