



Soft Tissue Aneurysmal Bone Cyst: A Case Report

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The authors present an unusual case of a primary soft tissue tumor having histologic features identical to an intraosseous aneurysmal bone cyst. A retrospective chart and radiographic review of a 44 year-old male was performed with a 17 month follow-up. Initially presenting as a painful, palpable nodule on the medial left ankle; a discreet, encapsulated mass was confirmed with ultrasound and magnetic resonance imaging. Surgical excision followed and pathologic analysis of the specimen diagnosed an aneurysmal bone cyst in soft tissue. Aneurysmal bone cysts usually appear in the metaphyses of long bones or in vertebral bodies. The primary soft tissue tumor is rare, with only 17 cases having been reported in the English literature. This is the first report of a primary soft tissue aneurysmal bone cyst in the distal lower extremity. In this case, as in the majority of reported cases, complete resolution was obtained with surgical excision.

Key words: Aneurysmal Bone Cyst, tumor of the foot, Magnetic Resonance Imaging.

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Aneurysmal bone cyst (ABC) is classically described as a benign lesion developing mostly in the metaphyses of long bones and in vertebral bodies.¹ They are characterized by blood-filled spaces separated by connective tissue septa containing fibroblasts, osteoclast-like giant cells, and reactive woven bone.² ABC is considered a non-neoplastic lesion, although some cytogenetic studies have found reproducible chromosomal abnormalities.^{3,18} The majority of ABCs arise de novo (primary ABC), but some have been found associated with other bony lesions secondarily.^{4,15}

Lesions usually appear on radiograph as lytic and expansile. Computed tomography (CT) or magnetic resonance imaging (MRI) modalities often show a heterogeneous lesion with fluid-filled cystic spaces surrounded by a thin rim of bone.⁵ Standard treatment of ABCs with marginal, yet complete surgical excision has shown acceptable results.^{3,5,6}

While traditionally classified as a more common benign tumor of bone, ABCs have rarely been described as being found in soft tissues. A search of the English literature found a total of 17 cases of soft tissue ABC, with no primary lesions previously described in the distal lower extremity.^{3,4,7-16}

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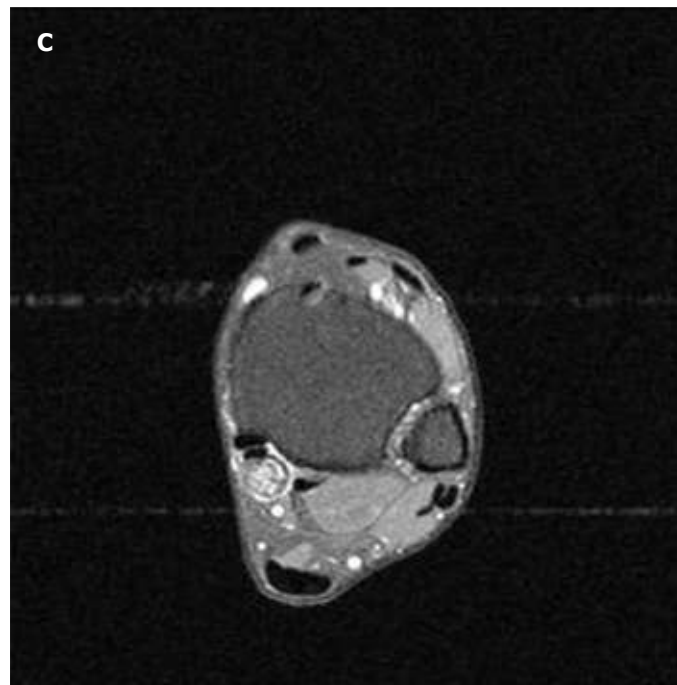


Figure 1A, 1B and 1C Magnetic resonance images (MRI) showing the heterogeneous, well-encapsulated mass immediately posterior to the flexor digitorum longus tendon. (A) Coronal Proton-dense T2. (B) Sagittal T1 image. (C) Axial T1 Image.

Case Report

The patient is a 44 year-old male who originally presented with complaints of a palpable “knot” in his medial left ankle, separate from and incidental to his chief complaint of hallux limitus on the contralateral limb. Initial exam showed a firm, semi-mobile, deep mass approximately 2cm proximal and immediately posterior to the medial malleolus. This was mildly tender to palpation, but did not restrict motion of the lower extremity. Ultrasound exam revealed a 5 x 10mm mass posterior to the tibialis posterior tendon. The mass was heterogeneous and ill-defined, without evidence of attachment to adjacent structures.

After one month of observation, the patient expressed continued pain and possible increasing size. An MRI exam was performed to show a 1.8 x 1cm heterogeneous mass abutting the flexor digitorum longus tendon. (Figs. 1A, 1B and 1C) A decision was made to perform a complete excisional biopsy.

Surgical dissection revealed a firm, purplish-brown, well-encapsulated mass within membranous soft tissue and with no defined attachment to surrounding structures. (Fig. 2A)

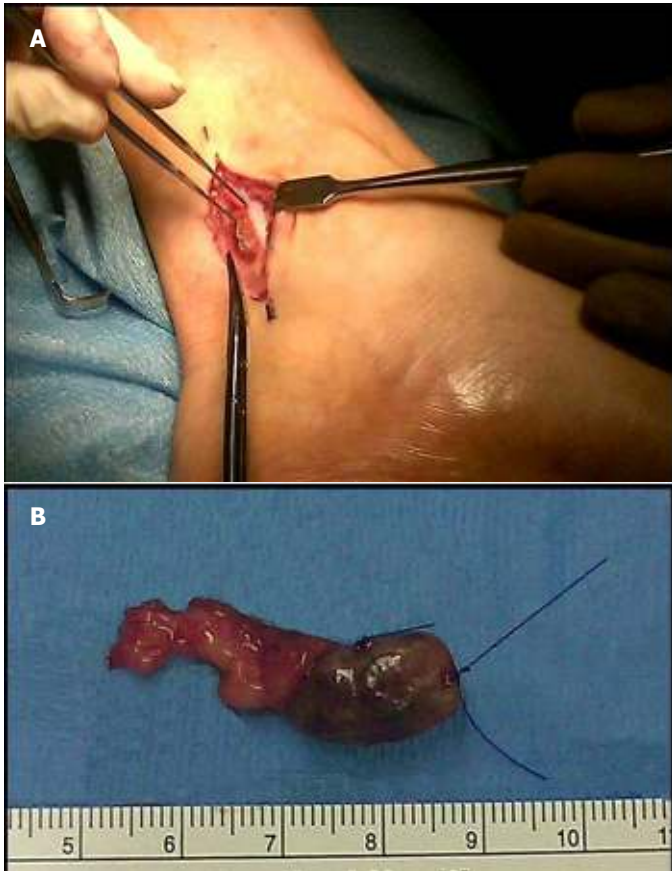


Figure 2A and 2B Intra-operative (A) and gross specimen photos (B) showing the excised mass with attached membranous soft tissue, and its location posterior to the tibialis posterior tendon.

The mass was tagged with suture to identify orientation and submitted to pathology as a single specimen. (Fig. 2B) The patient healed uneventfully with full weight-bearing in the immediate post-operative phase.

The specimen margins were inked per orientation and the mass was sectioned to reveal a *“variegated red-yellow cut surface with calcified tissue and extensive hemorrhage.”* Microscopic analysis proved inconclusive and slides were sent to an outside pathology lab for independent review by multiple pathologists. The final pathology report is as follows: *“The specimen is a circumscribed nodular heterogeneous lesion.”*

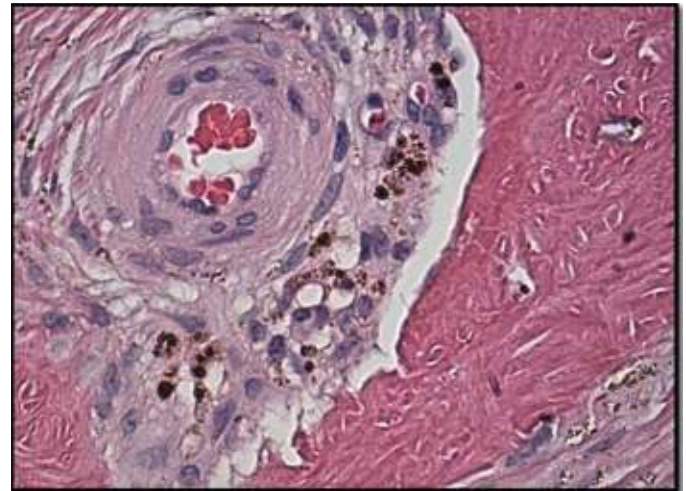


Figure 3 Prepared H&E stained slide showing histologic features of aneurysmal bone cyst in soft tissue at 40X magnification.

Hemosiderotic synovium and a pool of muscular-walled, variably sized vessels are seen at the periphery of the nodule. The nodule had a vaguely multilobulated configuration with blood-filled pseudocystic spaces alternating with spindle cell areas and areas of bone formation. The center of the lesion is occupied by well-formed bone with spicules that radiate outward. The outer rim of the nodule demonstrates a discontinuous thin shell of bone. Numerous giant cells are seen within the blood filled spaces. Prominent hemosiderin deposition is seen within the intervening spindle-cell areas. The histologic features are those of so-called aneurysmal bone cyst of soft tissue. (Fig. 3) No neoplasm is seen. The patient remains without symptoms or signs of recurrence at a follow-up of 17 months post-operatively.

Discussion

Unlike the more common primary bone lesion, soft tissue aneurysmal bone cyst is a rarely occurring pathology. A search of English language databases revealed a total of 17 cases of soft tissue ABC as reported by 12 authors.^{3,4,7-16}

Case	Published	Author	Age	Sex	Location	Size (cm)
1	1972	Salm and Sissons	32	M	Thigh	6
2	1972	Salm and Sissons	45	F	Abdominal wall	3.5
3	1992	Amir <i>et al.</i>	15	F	Groin	2
4	1993	Petrik <i>et al.</i>	7	M	Common carotid A.	3
5	1994	Rodriguez-Perralto <i>et al.</i>	20	F	Shoulder	6
6	1996	Lopez-Barea <i>et al.</i>	57	F	Upper arm	7
7	1997	Shannon <i>et al.</i>	29	F	Retroclavicular tissues	4.5
8	2002	Nielson <i>et al.</i>	8	M	Shoulder	8
9	2002	Nielson <i>et al.</i>	29	F	Groin	4
10	2002	Nielson <i>et al.</i>	37	F	Upper arm	2.5
11	2002	Nielson <i>et al.</i>	28	M	Shoulder	9
12	2002	Nielson <i>et al.</i>	30	F	Thigh	4
13	2005	Ajilogha <i>et al.</i>	12	F	Thigh	4
14	2007	D'Costa <i>et al.</i>	60	F	Breast	10
15	2007	Karkuzhali <i>et al.</i>	23	M	Proximal fibula	3
16	2008	Sahu <i>et al.</i>	12	F	Palmar side thenar hand	4.5
17	2009	Fellig <i>et al.</i>	25	M	Cerebello-pontine angle	3.3
18	2011	McCann <i>et al.</i>	44	M	Distal lower extremity	1.8

Table 1 Summary of reported cases of soft tissue aneurysmal bone cyst (ABC).

Salm and Sissons⁷ first described two cases of extraosseous giant-cell tumors having histologic features identical to bony ABC in 1972. Since then, 16 cases (including this report) have been reported occurring in various locations, and are summarized in Table 1. Patients are made up of eleven females and six males ranging in age from 7 – 60 years with a median age of 28. Tumors showed histologic features identical to the bony ABC and ranged in size from 1.8cm to 10cm with a median of 4cm. Lesions were most often located in the deep soft tissues of the upper extremity or shoulder⁷, or thigh.³ All cases described were treated by surgical excision. Recurrence rate is unknown due to the small number of cases and inadequate follow-up.

Recently, Karkuzhali *et al.*,¹⁵ described a case of multiple soft tissue ABC's arising following excision of a bony ABC from the proximal fibula 20 months prior, possibly suggesting a neoplastic course. Cytogenetic studies^{3,18} have shown an association with translocations at the 16q22 and 17p13 loci, providing further evidence of a possible neoplastic etiology. It has been proposed that the lesion is first formed as a reaction to ectopic bone, but is more likely the result of a primary arteriovenous malformation.³

Despite the usual appearance of aneurysmal bone cyst in long bone metaphyses, the foot and ankle surgeon should be aware that it may also manifest as a soft tissue lesion of the lower extremity.

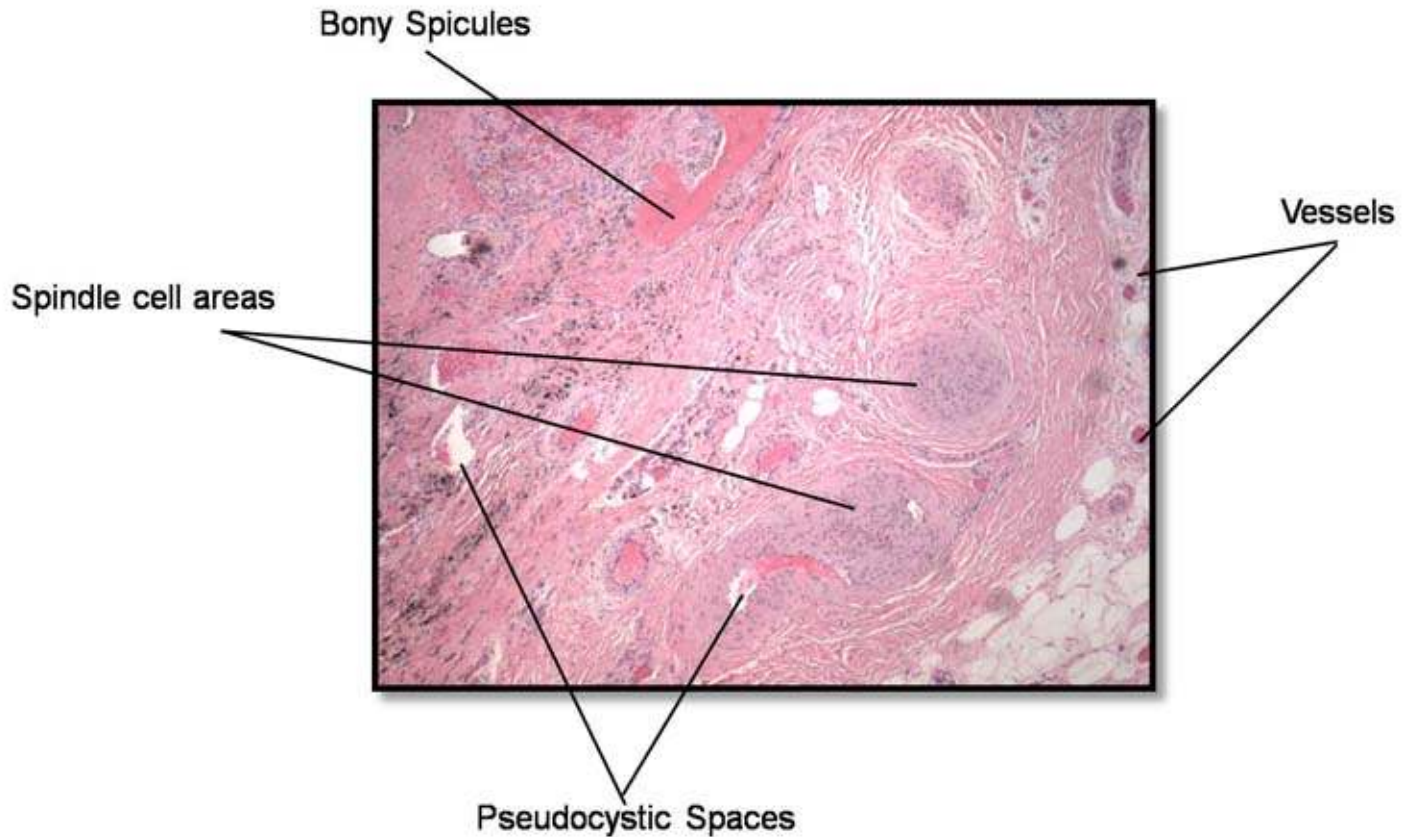


Figure 4 Pathologic identification of soft tissue ABC.

The differential diagnosis of soft tissue ABC can include nodular fasciitis and ossifying fibromyxoid tumor. Nodular fasciitis lesions may histologically mimic soft tissue ABC, but do not contain the characteristic pseudocystic spaces or thin shell of bone. Ossifying fibromyxoid tumors often show spicules of bone, but are without osteoclastic-like giant cells characteristic of soft tissue ABC.³ Definitive diagnosis is made based on microscopic pathology, which is undistinguishable from the classic bony lesion. (Fig. 4)

The exact etiology of soft tissue ABC's remains unknown. While the lesion can be aggressive in growth, it is benign and can be successfully treated with surgical excision.

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