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Solid aneurysmal bone cyst of the humerus mimics metastasis or brown tumor☆,☆☆,☆☆☆

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ABSTRACT

Solid aneurysmal bone cyst (ABC) is a rare subtype of ABC that most commonly involves the small bones of the hands or feet. We present a case of a solid ABC of the distal humerus in a 52-year-old man with a history of chronic kidney disease and renal cell carcinoma. On imaging with plain radiographs, CT, and MRI, this expansile lucent lesion with solid internal enhancement had an appearance that overlapped with metastasis or brown tumor of hyperparathyroidism. On 18F-FDG PET-CT, this lesion was hypermetabolic with an SUVmax of 9.9. Only 37 cases of solid ABC have previously been reported to involve the long bones in the literature, and only 4 in the humerus. We review the clinical, imaging, and histopathological findings and differential diagnosis of solid ABC, and highlight the usefulness of identifying the USP6 gene rearrangement on FISH to distinguish this lesion from other lesions with secondary ABC formation.

1. Introduction

Solid aneurysmal bone cyst (ABC) is a reactive non-neoplastic lesion with varied imaging characteristics [1]. Giant cell reparative granuloma was first described as a non-neoplastic hemorrhagic process of the jaw or the short tubular bones of the hands and feet by Jaffe in 1953 [2, 3]. The term solid variant of ABC was first used in 1983 by Sanerkin et al. to describe four lesions characterized histologically by florid fibroblastic and fibrohistiocytic proliferation, areas of osteoclast-type giant cells, foci of osteoblastic proliferation with osteoid production, occasional foci of calcifying fibromyxoid tissue, and aneurysmal sinusoids [4]. Histologically, giant cell reparative granuloma and solid or solid variant of aneurysmal bone cyst cannot be distinguished morphologically. The terms have been used interchangeably in the literature, although the more recent literature favors the term solid aneurysmal bone cyst for extra-gnathic locations and giant cell reparative granuloma (GCRG) in gnathic locations [5]. This distinction is supported by the development of Ubiquitin specific protease 6 (USP6) fluorescence in situ hybridization (FISH), with the USP6 gene rearrangement identified only in primary ABC and not secondary ABC or gnathic GCRGs [6–8]. The term solid ABC will be used throughout the remainder of this article when discussing extra-gnathic locations. Although aneurysmal bone cysts account for 1–6% of primary bone tumors, solid ABC is a rare subtype that accounts for only 3.4–7.5% of all ABCs [9].

These lesions tend to occur in the 2nd and 3rd decades of life with 74% of patients under 30 years of age at presentation [10, 11]. The most common site for solid ABC is within the small bones of the hands and feet, most commonly affecting the phalanges of the hand, followed by the metacarpals, metatarsals, carpal bones, tarsal bones, and phalanges of the feet [12]. There is an equal sex distribution in the hands and feet [11]. Involvement of the spine and flat bones has also been rarely reported [13–23]. Prior to 2001, only 32 lesions in 31 patients with solid ABCs of the long bones had been described in the English language literature, with only 5 additional reported cases in the interim [1, 24–28]. In the long bones, the lower extremity is involved twice as frequently as the upper extremity.

On radiographs, solid ABCs can range from an appearance indistinguishable to classical ABC to an osteolytic lesion with a wide zone of transition, cortical destruction and soft tissue component [29]. The

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differential diagnosis includes giant cell tumor, brown tumor, and malignancy. In the long bones, solid ABCs can be juxta-articular, metaphyseal, or diaphyseal, and are eccentric in two-thirds of cases [1]. Lesion size is typically between 1 and 7 cm and one-third of cases are non-aneurysmal. Mineralization may occasionally be seen in some cases, which can be helpful for differentiation from giant cell tumor [1, 11]. Unusual findings include pathological fracture or periosteal reaction. Areas of cortical thinning and destruction can better be assessed on CT. The lesion attenuation is similar to muscle. On MRI, these lesions generally are iso- to slightly hyperintense to muscle on T1-weighted sequences and heterogenous but predominantly hyperintense with scattered foci of low signal intensity on T2-weighted sequences. Although cystic components may be present, they are typically less common and constitute a smaller proportion of the lesion compared with giant cell tumor. Adjacent bone marrow edema can be seen in 50% of cases.

2. Case report

A 52-year-old man presented to his primary care physician with a complaint of right medial elbow pain, progressively worsening over the past 6 months. The patient described it as a burning pain, worse with activities with some decreased range of motion. There was no associated numbness or tingling and the patient had full function in his right hand. The patient denied fevers or weight loss.

The patient had a history of chronic kidney disease due to gouty nephropathy status post deceased donor renal transplantation 8 years prior. He was on hemodialysis for 5 years prior to the transplant. He also had renal cell carcinoma of his native left kidney status post nephrectomy 7 years prior. At presentation, the patient had recurrent stage III chronic kidney disease with a baseline creatinine of 1.4.

On physical examination, there was fullness about the medial aspect of the right elbow that was tender to palpation. He was non-tender at the common extensor origin. His elbow range of motion was from 10 to 120 degrees with 80 degrees of pronation and 80 degrees of supination. There was no varus or valgus instability.

Based on the patient's history and clinical assessment, a differential diagnosis of primary bone neoplasm, metastasis, infection, and gout were considered. Radiographs were ordered and a referral to an orthopaedic oncologist was made.

Plain radiographs of the right elbow demonstrated an expansile lytic lesion of the medial aspect of the right distal humeral metaphysis extending to the supracondylar ridge with marked cortical thinning (Fig. 1). The lesion measured up to 2.9 × 5.5 cm. Given the patient's reported history a renal cell carcinoma metastasis was favored, but the differential also included primary bone tumor and brown tumor of hyperparathyroidism. The lack of articular or periarticular involvement made gout unlikely and lack of subchondral extension made giant cell tumor less likely.

Subsequently, a CT with and without intravenous contrast was performed (Fig. 2). The lesion measured up to 4.9 × 3.1 × 2.8 cm in greatest longitudinal, anteroposterior, and transverse dimensions, respectively. The CT better demonstrated the degree of generalized marked cortical thinning with a few areas where the cortex was imperceptible or destroyed, however no soft tissue component was identified. On pre-contrast images, the lesion had a mean measurement of approximately 40 Hounsfield units (HU), mildly hypodense compared to muscle (~60 HU). There was mildly heterogenous but avid solid internal enhancement on post-contrast images with a mean attenuation of approximately 100 HU.

An MRI with and without intravenous contrast was then obtained (Fig. 3). The expansile mass of the medial aspect of the distal humeral metaphysis was slightly hyperintense to muscle on T1-weighted images and heterogeneously hyperintense on short tau inversion recovery (STIR) images. There was avid solid internal enhancement on the post-contrast images, although small cystic foci were identified. There is mild adjacent bone marrow edema.

Next, a whole body (vertex to toes) 18-fluorodeoxyglucose (FDG) positron emission tomography (PET)-CT scan was obtained (Fig. 4). This examination showed that this lesion was intensely hypermetabolic with a maximum standardized uptake value (SUVmax) of 9.9. However, there were no other areas of abnormal tracer uptake on this 18F-FDG PET-CT scan. Given this high metabolic activity on the PET-CT scan, the primary differential diagnosis was a metastatic focus from the patient's renal cell carcinoma (or other unknown primary) versus a primary bone sarcoma.

The decision was made to take the patient to surgery for an incisional biopsy. The tumor bled profusely at the incisional biopsy site, but hemostasis was achieved with electrocautery and with absorbable gelatin powder (Gelfoam®). The patient tolerated the procedure well without intraoperative complications. Intraoperative frozen section reviewed by the pathologist showed a giant cell rich tumor.

Permanent sections stained with hematoxylin eosin stain showed multinucleated giant cells, cellular spindle stroma and reactive bone formation (Fig. 5). No cytological atypia was seen. Higher magnification showed numerous giant cells and few mitotic figures (Fig. 6). The histological differential diagnosis included giant cell tumor of bone, brown tumor of hyperparathyroidism and solid aneurysmal bone cyst. Carcinoma with multinucleated giant cells was excluded as there was no cytological atypia and immunostain for pancytokeratin performed

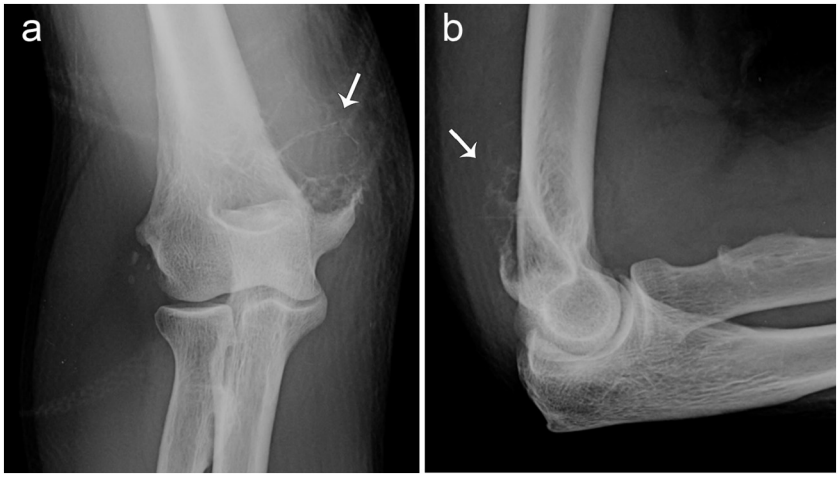

Fig. 1. Anteroposterior (a) and lateral (b) radiographs of the right elbow demonstrate an eccentric expansile lytic lesion (arrows) of the medial aspect of the distal humeral metaphysis.