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Case Report

Unusual presentation of an aneurysmal bone cyst: A case report and literature review ☆,☆☆

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ABSTRACT

Aneurysmal bone cysts (ABCs) are non-neoplastic primary bone tumors, typically involving the long bones and vertebrae in the first 2 decades of life. ABCs require prompt diagnosis and intervention due to their rapidly expansile nature and ability to destroy the adjacent normal bone. ABCs rarely affect the rib. We report a case of a 51-year-old female presenting with chronic dry cough and right upper back pain. A chest X-ray and computed tomography scan revealed an expansile, lytic mass affecting the posterior aspect of the third right rib. The third right rib was resected using a posterolateral, Shaw-Paulson approach. Histopathology of the resected mass confirmed the diagnosis of ABC. There were no intra- or perioperative complications, and follow-up X-ray was normal.

Introduction

Aneurysmal bone cysts (ABCs) are rare, benign tumors of bone, most commonly of the vertebrae and long bones in children and adolescents. ABCs rarely involves the ribs. They are rapidly growing and locally destructive masses which are comprised of multiple, thin-walled, blood-containing cysts [1,2]. Furthermore, ABCs may compress surrounding structures and weaken the bone, irritating surrounding organs and nerves and pathological fractures [3]. Here, we report a 51-year-old woman diagnosed with an ABC of the third right rib and, through a literature review, discuss their salient radiographic features and established and emerging treatment strategies.

Case presentation

A 51-year-old female presented with a 1-year history of right-side upper back pain. The pain was dull and aching

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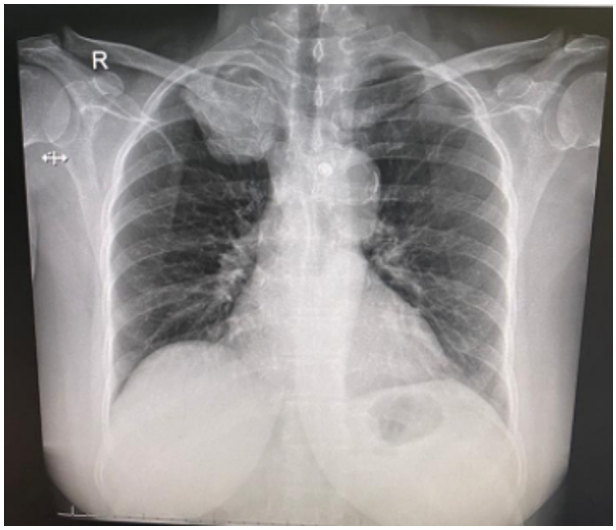

Fig. 1 – Chest X-ray showing a large expansile lesion involving the third rib on the right side.

in character, mild, non-radiating, and relieved by over-the-counter analgesics. There was no history of trauma, but further inquiry revealed a recent history of dry cough without dyspnea. Past medical history revealed chronic hypertension controlled with antihypertensive medication.

A chest X-ray was performed based on the presenting symptoms of dry cough with upper back pain. The X-ray revealed a well-defined opacity projecting over the apex of the right lung (Fig. 1). The lesion was consistent with the patient's history of right upper back pain and was suspicious of a Pancoast tumor. A subsequent computed tomography (CT) revealed a large, expansile lytic lesion involving the posterior part of the third right rib (Fig. 2). The possible diagnosis and treatment options were discussed with the patient. The patient elected for surgery. The third right rib was excised using the Shaw-Paulson (posterolateral) approach. Intraoperatively, the lesion was adherent to the lung and was cystic with a thin wall that was fragmented during the dissection.

The resected specimen was sent for histopathological assessment. Grossly, the resection had multiple fragments of bony tissue, which were brownish. Microscopy revealed

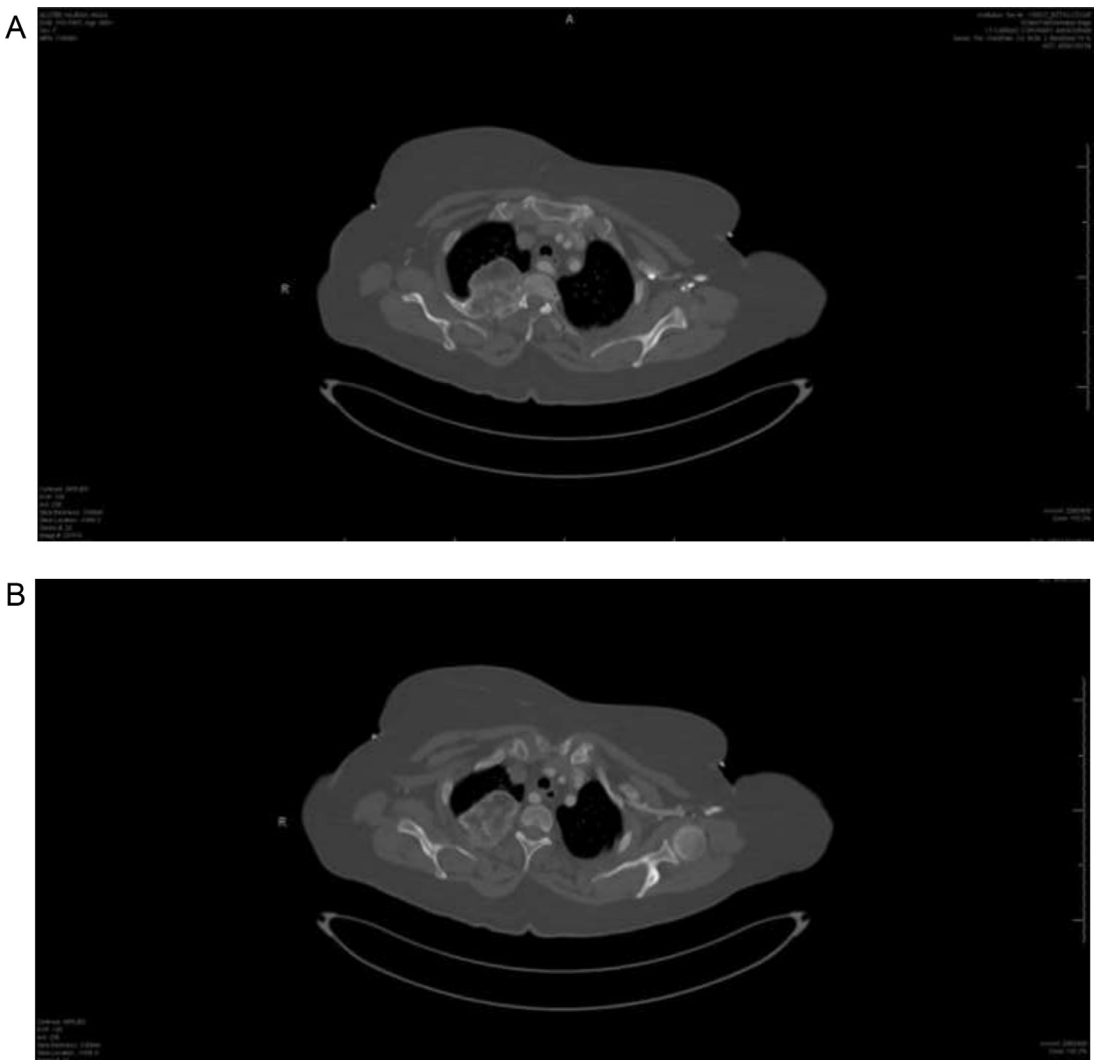

Fig. 2 – Contrast-enhanced CT scan demonstrating a large expansile lytic lesion of the right third rib with no cortical destruction. No adjacent soft-tissue and lung parenchymal involvement is seen.

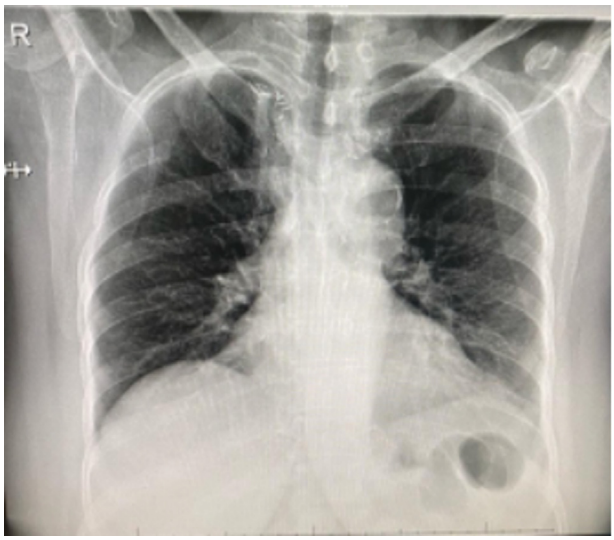

Fig. 3 – Normal chest X-ray 3 months after surgical resection.

osteoclast-like cells adhering to the bony fragments to reactive lung parenchyma via fibrous and hemorrhagic tissue. The patient recovered well postoperatively with only mild pain upon discharge. Upon follow-up 3 months later, a chest X-ray was normal (Fig. 3).

Discussion

ABCs account for approximately 9.1% of all bone tumors [4]. The pathogenesis of ABCs remains uncertain. Some reports suggest that an increase in venous pressure from a tumor-induced arteriovenous malformation or trauma ultimately progresses to bleeding and bone resorption, forming fluid-filled ABCs [3,6]. In about 30% of ABCs, a pre-existing tumor may be identified, most commonly a giant cell tumor [6]. Oliveira et al. demonstrated higher TRE17/USP6 expression in ABCs, an oncogene overexpressed in other bone tumors such Ewing's sarcoma [5].

An X-ray typically shows a solitary radiolucent expansile lesion surrounded by a sclerotic layer of new bone and a transitional zone between the two. A thick periosteal reaction or matrix calcification is also usually seen. CT scans can better define the tumor and identify a potential soft tissue extension or a cortical breach [6]. On magnetic resonance imaging (MRI), the cystic nature of ABCs can be appreciated as fluid-fluid levels and multiple internal septations. Fluid within the cyst can be of varying signal intensity, likely representing oxidized blood breakdown products of varying age [7]. Hence, ABCs must be differentiated from giant cell tumors, which require histopathologic evaluation of a biopsy specimen. ABCs histologically show a multiloculated blood-filled mass with cystic spaces separated by fibrous septations. The fibrous walls contain osteoclast-like multinucleated giant cells, reactive woven bone, and proliferating fibroblasts.

Surgical intervention is curative in most cases. While some reports performed en bloc resection [1], we resected the affected rib entirely. Some invasive treatment options aside from operative measures, like curettage bone grafting, have been performed for other lesions such as giant cell tumors [8]. Novel, less invasive treatment alternatives are under research. Denosumab—monoclonal antibody suppressing osteoclast activity—administration significantly improved outcomes in 9 patients with ABCs but with a higher recurrence rate compared to surgical resection [9]. Another study used autologous bone-marrow-derived mononuclear cells with β-tricalcium phosphate and absorbable atelocollagen to form new bone within the cysts [10].

ABCs should be treated promptly to minimize the chance of possible complications, including pathologic fractures and compression of surrounding structures. Although ABCs are benign lesions, their malignant transformation into osteosarcoma has been reported. Malignant transformation of primary ABCs occurs almost exclusively in the context of prior radiation exposure [11]. However, very rare instances where primary ABCs transform into malignancies, most commonly fibroblastic osteosarcomas, without prior radiation exposure have also been reported; we found seven such cases [11–17].

Conclusion

ABCs are non-neoplastic primary bone tumors that rarely involve the ribs. A diversity of less invasive management options for ABCs have been added to the literature. However, these novel treatment modalities require validation through more robust clinical studies. Despite the encouraging results of these interventions in alleviating symptoms, the risk of recurrence after these treatment strategies is higher than total surgical excision. Surgical excision, therefore, is still the treatment modality of choice for ABCs.

Author contributions

All authors worked in conceptualizing and drafting the final manuscript. All authors have revised and approved the final version for submission.

Patient consent

Written informed consent was obtained from the patient for publication.

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