Case Report

Giant cell tumor of acromion process with secondary aneurysmal bone cyst

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ABSTRACT

Giant cell tumor (GCT) or osteoclastoma develops after attaining physeal closure with the most common sites of involvement being the ends of long bones. It is uncommon in the small bones of hands and feet and extremely rare in the flat bones. GCT is a locally aggressive tumor, but in rare cases, it may metastasize to lungs, ribs, regional lymph nodes, and skull. Secondary aneurysmal bone cyst (ABC) may also develop in some cases. Histopathological appearance is pathognomonic in most of the cases of GCT with secondary ABC formation, and immunohistochemistry is rarely needed for a confirmatory diagnosis. The usual treatment is excision or curettage with an adjuvant such as bone cement, liquid nitrogen, and phenol. Involvement of the acromion process is extremely rare. Here, we report a case of a young male having GCT of the acromion process with secondary ABC, and it is the second case reported in literature.

Key words: Acromion process, flat bones, giant cell tumor, osteoclastoma, secondary aneurysmal bone

INTRODUCTION

Giant cell tumor (GCT) or osteoclastoma comprises 5% of all primary skeletal tumors. Peak incidence is seen in the third and fourth decades of life. Among all GCTs, 15% have been found in flat bones. Common flat bones involved in GCT are pelvis, skull, sacrum, spine, ribs, patella, sternum, scapula, and rarely acromion. Scapular involvement has been mentioned in <1% of all GCT cases reported in the literature.[1] GCT in flat bones may also occur as a complication of Paget's disease. Rare site of occurrence and atypical presentation of GCT in flat bones may commonly lead to misdiagnosis both clinically and radiologically. GCT may present with variable severity and uncertain origin. Common features of GCT are pain and swelling. Radiologically, GCT of flat bones frequently presents as an expansile lesion within an eggshell-thin rim of new bone and involvement of adjacent soft tissues.[2] It is imperative to differentiate GCT in these locations from hyperparathyroidism and other giant cell containing lesions such as telangiectatic osteosarcoma or giant cell-rich osteosarcoma. Hyperparathyroidism can be excluded after the assessment of serum levels of calcium, phosphate, alkaline phosphatase, and parathormone. Association of GCT with a secondary aneurysmal bone cyst (ABC) has been reported in 14% cases.[3] On PubMed search using keywords: "giant cell tumor," "osteoclastoma," "acromion," and "secondary aneurysmal bone cyst," we found three cases of GCT of acromion by Aoki *et al*. without secondary ABC.[4] Only one case of GCT acromion with secondary ABC reported by Sherwani *et al*. was found.[1] To the best of our knowledge, we report the second case of GCT of the acromion process with secondary ABC in a middle-aged male.

CASE REPORT

A 30-year-old male presented to us with complaints of pain and swelling over the right shoulder for the last 1-year. Pain was associated with limitation of the right

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shoulder movement. Swelling was insidious in onset and gradually progressive in size. On clinical examination, globular swelling of size 7 cm × 6 cm, nontender, fixed to underlying bone, and firm in consistency was present over the superolateral aspect of the right shoulder. Overlying skin was shiny, tense, mobile, and no visible veins or ulcers were present over the swelling. Plain radiograph of the right shoulder showed an expansile lytic lesion involving the acromion process of the right scapula with thinning of the cortex and a prominent overlying soft-tissue shadow [Figure 1]. The lesion was well circumscribed with a thin rim of bone. Magnetic resonance imaging (MRI) could not be performed due to financial constraints of the patient. Blood investigations such as hemogram, kidney function, liver function, serum level of calcium, phosphate, alkaline phosphatase, and parathyroid hormone were done to exclude other causes of expansile lytic lesion and were within normal limits. A diagnosis of GCT of the acromion process was made on the basis of clinicoradiological findings. Fine needle aspiration cytology was done from the margin of the lesion under the guidance of image intensifier, and findings on cytological examination were suggestive of GCT.

The tumor was excised *en bloc* with acromion process, lateral end of the clavicle, and adjacent soft tissue to obtain a clear margin. Blunting of the lateral end of the clavicle was also done to maintain the shoulder contour. *En bloc* tumor was sent for histopathological examination which showed numerous osteoclast-type giant cells surrounded by mononuclear round to polygonal stromal cells along with few areas showing small and large blood-filled cystic spaces. The cyst wall was composed of fibroblasts and few osteoclastic giant cells along with extensive areas of necrosis. There were no atypical cells or mitotic figures. The histopathological picture confirmed the clinical diagnosis of GCT with secondary ABC [Figure 2]. As there was no confusion in diagnosis on histopathology and it correlated well with the clinical and radiological findings, immunohistochemistry was not needed in this case.

Active shoulder and elbow range of motion exercises were advised after 4 weeks of surgery, and the patient achieved a reasonable range of motion at 3 months. The patient was followed up every 3 months for the first 2 years and after that twice a year with plain radiographs [Figure 3]. On 5-year follow-up, there was no sign of recurrence and the patient had a full range of motion at the operated shoulder as compared with the opposite shoulder. He was happy and was doing all his routine activities satisfactorily.

DISCUSSION

GCT is a locally aggressive benign skeletal tumor characterized by the presence of osteoclast

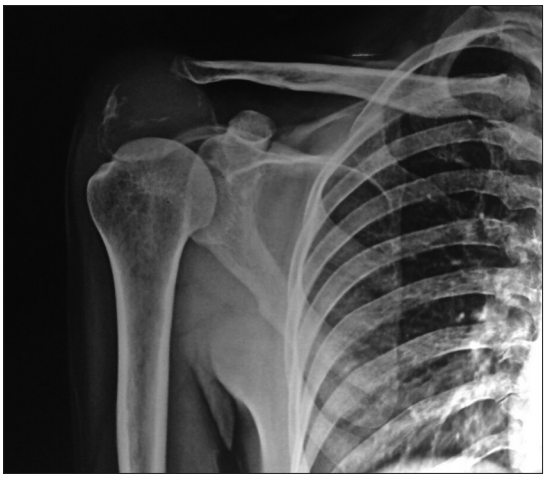

Figure 1: An expansile lytic lesion with soap bubble appearance involving the right acromion process leading to prominent soft tissue

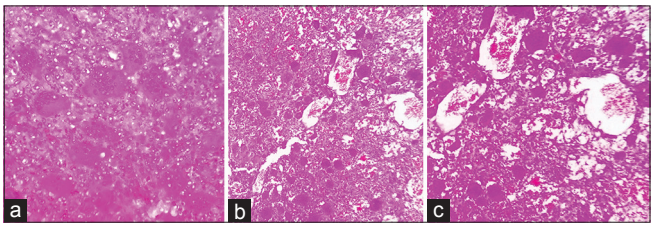

Figure 2: (a) Mononuclear stromal cells and osteoclastic giant cells (H and E, ×100). (b) Blood-filled spaces surrounded by mononuclear stromal cells and few osteoclastic giant cells (H and E, ×40). (c) Blood-filled cystic spaces with cyst wall composed of fibroblasts and few osteoclastic giant cells with some reactive bone formation seen (H and E, ×40)

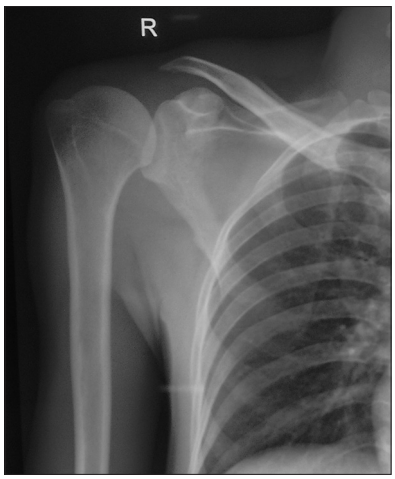

Figure 3: Three-year follow-up X-ray shows postoperative resection of the right acromion process and lateral end of the clavicle. Normal soft tissue is seen

type-multinucleated giant cells in the field of mononuclear stromal cells.[5] This tumor usually develops after attaining the skeletal maturity and is more common in female. The common sites of GCT are the ends of long bones. More than half of the tumor is found around the knee joint that is distal femur and proximal tibia followed by radius.[6] GCT of the smaller bones is more aggressive than the larger bones. Rarely, GCT may transform into malignant tumor

either due to dedifferentiation of the primary GCT or secondary to the previous radiation in <1% of cases.[6,7] Rarely, multicenter GCT also occurs especially in the foot and hands. GCT can metastasize to the lungs in 1–6% cases although metastases to the regional lymph nodes, scalp, and pelvis have been mentioned in the literature. Simultaneous metastasis to lung, scapula, and rib has also been reported by Ismail *et al*.[8]

On plain radiographs, the classical features of GCT are expansile, lytic lesion with well-defined but nonsclerotic margin, eccentric location, and extension to the subchondral bone. In flat bones, GCT shows aggressive radiological features such as wide zone of transition, cortical thinning, expansile lesion, or even cortical bone destruction with soft-tissue involvement. Computed tomography or MRI may reveal fluid levels which indicate the presence of secondary ABC. Diagnosis is confirmed on histopathological examination which shows multinucleated giant cells among with mononuclear cells, which are of even size and round to oval in shape. The nuclei of the giant cells resemble those of the mononuclear cells. These features resemble with the present case.

Treatment of GCT depends on the location and grading of tumor. Treatment with intralesional curettage showed high recurrence rate about 15–25%. The recurrence rate has been reduced to 2.3% by adding the drilling of tumor wall with high-speed burr with cryoablation by liquid nitrogen.[9] GCT could also be treated by curettage adjuvant with phenol, autogenous bone grafting, or bone cementing. Excision of tumor with bone is preferred treatment in expendables bone such as ulna, fibula, and even in small bones of foot and hands. In case of aggressive GCT around larger joints, excision with reconstruction either by arthrodesis or by modular endoprosthesis is treatment of choice. GCT could also be treated by radiotherapy in unapproachable location. In the present case, we excised the acromion process with involved soft tissue and achieved good functional result without recurrence at 5 years of follow-up.

Recently, Thomas *et al*. achieved 90% tumor necrosis in GCT using newer drug denosumab and concluded that it would make surgical excision easier in aggressive tumors or can be used alone for treatment of GCT in inaccessible sites or in high-risk patients for surgery.[10]

GCT with secondary ABC of the acromion is an extremely rare lesion. GCT should be considered in the differential diagnosis of expansile lytic lesion of the acromion. Biopsy is needed for confirmation of the diagnosis. Complete excision of the lesion with wide margin is an effective treatment with good functional outcome.

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Conflicts of interest
There are no conflicts of interest.

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