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Case Report

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An uncommon presentation of aneurysmal bone cyst in spine of scapula: a rare case report

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ABSTRACT

Aneurysmal bone cyst is locally aggressive benign osteolytic lesion of the bone. It is more common in second decade of life with slight inclination to female gender. Metaphysis of long are most commonly affected. Incidence in flat-bones is <2% especially in scapula. We present a rare case of aneurysmal bone cyst of scapula with atypical radiological finding that was confirmed with pathological correlation. A 45 years old female, right hand dominant, presented with 3 months old swelling over left shoulder. There was painful restriction of terminal shoulder range of movements. On examination the swelling was 8×7 cm cystic, non-trans illuminant, fixed to scapula, no neurovascular deficit. X-ray shown eccentric lytic lesion over the supraspinous fossa with relatively more prominent soft tissue shadow. MRI image shown well defined cystic swelling with fluid filled level, lacking septations. Excision and extended curettage of lesion revealed hematogenous fluid as content. Histopathology confirmed aneurysmal bone cyst. Our case presented with all atypical features of ABC with regards to age, location and imaging. High degree of clinical suspicion and ruling out other closest possibilities like telanectatic osteosarcoma is necessary when cystic swelling around the scapula is encountered. Excision and extended curettage of the lesion is satisfactory. Periodic follow up with MRI and long-term clinical assessment for recurrence is needed.

Keywords: Aneurysmal bone cyst, Scapula, Benign bone tumor, Rare presentation

INTRODUCTION

Aneurysmal bone cyst (ABC) is locally aggressive benign osteolytic lesion of the bone. It is more common in second decade of life with slight inclination to female gender.[1] Metaphysis of long are most commonly affected. Incidence in flat-bones is <2% especially in scapula.[2,3] MRI with contrast is the specific imaging investigation for diagnosis, though biopsy is essential to confirm and plan the treatment strategy. We present a rare case of ABC of scapula with atypical radiological finding that confirmed with pathological correlation.

CASE REPORT

A 45 years old female, homemaker with right-handed dominance presented with complaints of three months old insidious onset swelling over the back of the left shoulder, progressing to current size. Pain over swelling brought by terminal shoulder range of movement otherwise painless. She does not notice any other similar swelling in the body.

On examination the swelling was present on the left supraspinous fossa region of size 8×7 cm, round to oval with smooth surface, ill-defined margins, non-pulsatile. Palpation reveals tenderness, cystic, non-fluctuant, non-trans illuminant, irreducible and moves with the movement of scapula. Shoulder abduction and forward flexion are 0-1400 respectively further movements are restricted with pain. All blood investigations are found to be normal.

X-ray of left the shoulder AP and scapula Y view shown an eccentric lytic lesion over the superior aspect of the spine of scapula occupying suprascapular fossa with

relatively more prominent soft tissue shadow and no periosteal reaction.

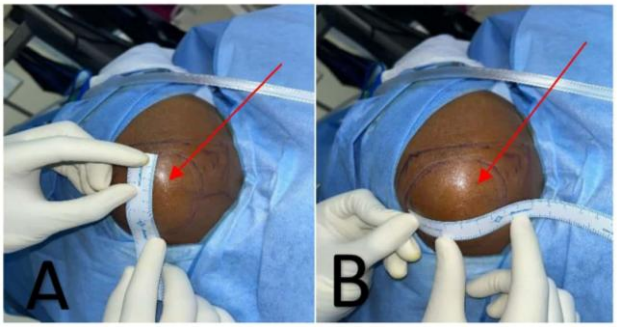

Figure 1: (A) Mediolateral dimension of 8 cm and (B) super inferior dimension of 7 cm.

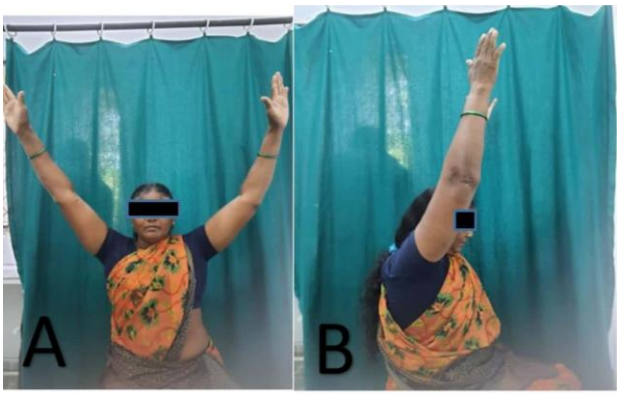

Figure 2: (A) Abduction of 1400 and (B) forward flexion of 1400.

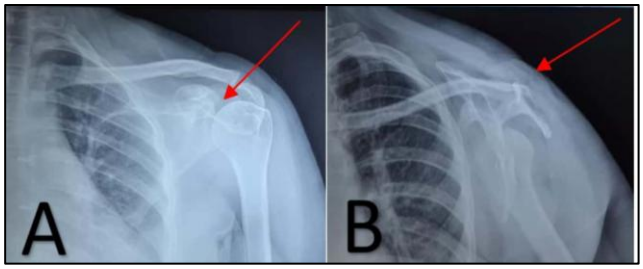

Figure 3: (A) Anteroposterior of left shoulder showing eccentric lytic lesion with sclerotic margin over the spine of scapula with soft tissue extension and (B) capula Y view of left shoulder showing prominent soft tissue lesion.

Ultra sonogram confirmed fluid as content. MRI shown single isointense lesion of size 8 × 7 cm in T1W sequence, mildly hyper intense lesion with small cystic areas within in T2W and STIR sequences. It was not communicating to the shoulder joint and not compressing any neurovascular structures.

She was planned for excision biopsy of the lesion. On operating table with right lateral position affected shoulder on the top. Incision was made over the prominent part of the swelling. Swelling was subcutaneous extending from superior border of scapula to the superior surface of the spine of scapula. Swelling was found to be arising from scapula. Cyst ruptured inadvertently, hematogenous fluid as content was found with loculations. Erosion of supraspinous fossa noted due to pressure caused by the swelling, lesion curreted with burr, cauterized and washed with hydrogen peroxide (H2O2) solution, bone wax applied. Samples sent histopathological examination.

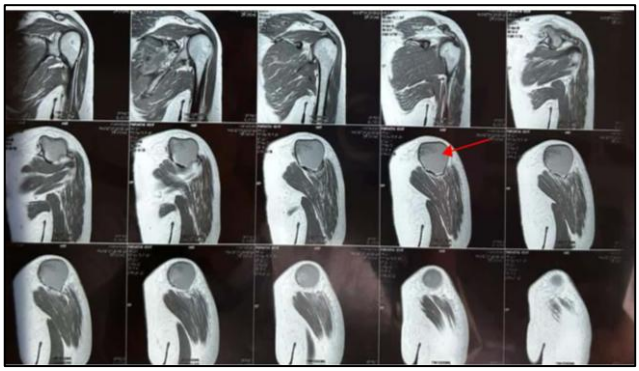

Figure 4: T1W weighted coronal section image showing single well defined iso to hypo intense in the supraspinous fossa noted.

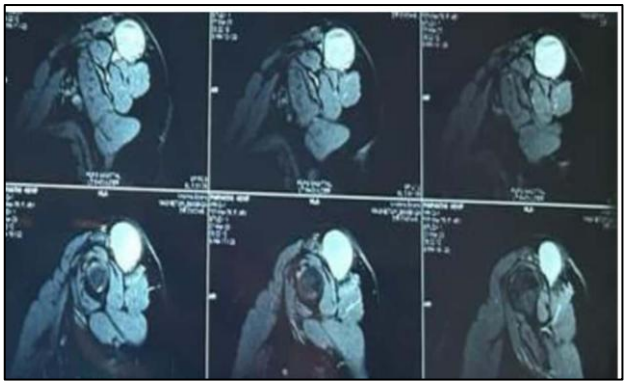

Figure 5: T2W weighted sagittal section image showing single well defined hyper intense lesion with fluid filled level > 2/3rd of the lesion (marked with red arrow) noted and no intra-lesional septations noted.

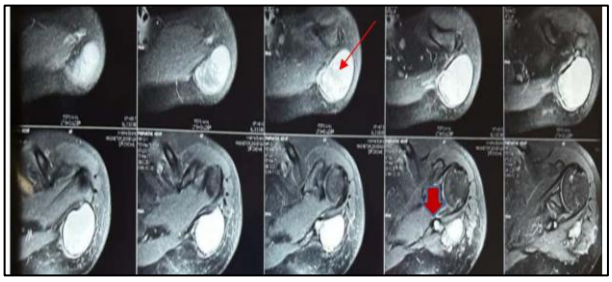

Figure 6: T2W axial section images showing well defined hyper intense lesion (marked with thin red arrow) not communicating with shoulder joint; no suprascapular nerve encasement seen and erosion of spine of scapula seen (marked with broad red arrow).

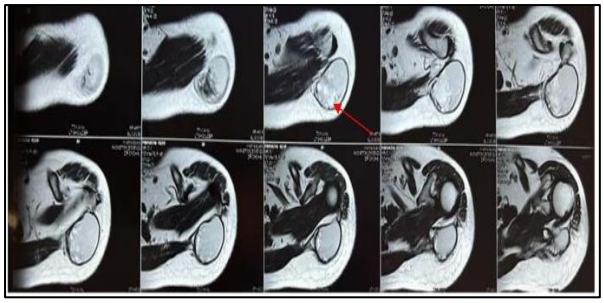

Figure 7: T1W axial section images showing intralesional sclerosis.

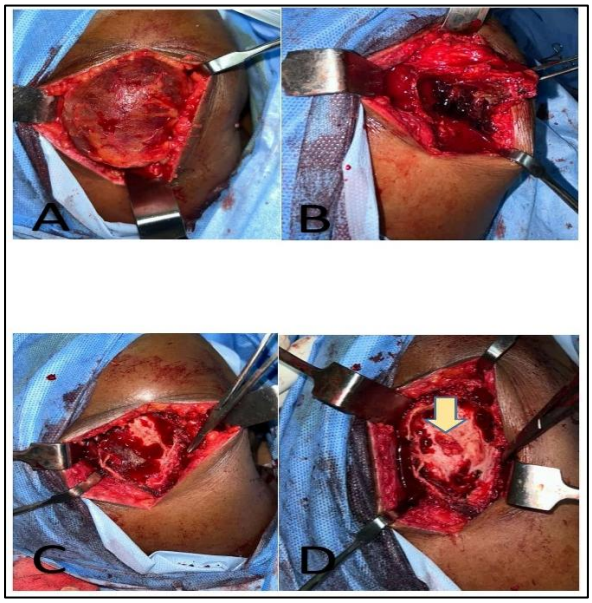

Figure 8: (A) 8×7 cm subcutaneous swelling occupying supraspinous fossa; (B) hematogenous fluid as content noted; (C) erosion of scapula due to pressure noted and (D) erosion of spine of scapula noted (marked with yellow arrow).

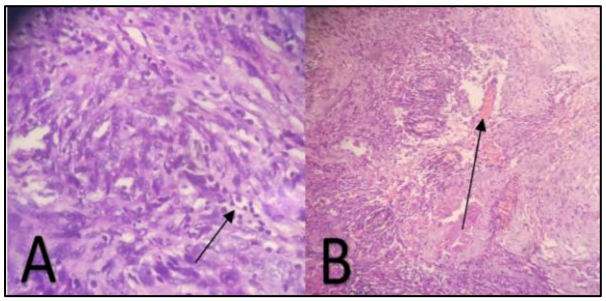

Figure 9: (A) Multinucleated giant cells (marked with black arrow) and (B) dilated congested blood vessels.

Pathology report showed bony tissues with degeneration, fibro-cartilaginous tissue shows moderate lymphocytes and plasma cell with fibroblast proliferation, congested capillary vessels with endothelial proliferation multinucleated giant cells and hemosiderin laden macrophages confirms the diagnosis of ABC.

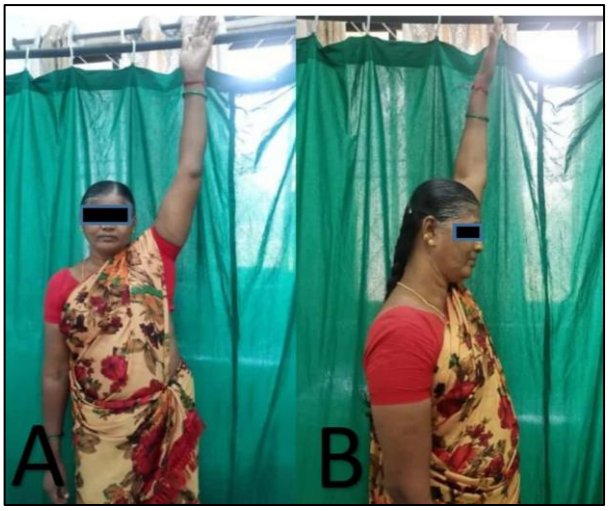

Figure 10: (A) Abduction of 1800 and (B) forward flexion of 1800.

Postoperatively patient immobilized in arm sling pouch for two weeks, wound healed without complication. Shoulder range of movements started at third week. Shoulder range of movements improved to abduction 0-1700 forward flexion to 0-1600 internal rotation 0-400 external rotation 0-300. Sixth month follow up did not show any recurrence of the swelling.

DISCUSSION

ABC is benign locally aggressive blood-filled reactive lesion of the bone. It was first described by Jaffe and Lichtenstein in 1942.[4] Typically, any bone may be involved, most commonly involves the metaphysis of the long bone (67%) viz. Distal femur (18%), proximal tibia (8.3%), proximal humerus (6.3%), flat bones (20.4%), hands and feet (7.7%).[5] Most commonly affects children and adolescent < 20 years with slight preponderance to female gender. It constitutes 1-6% of biopsied bone tumors. Due to its preponderance to growing bone can affect the growth plate causing deformity and limb length discrepancy. In our case it is in 5th decade. Scapular ABC is reported to be very rare (<2%) compared to other unusual location.

Traditionally ABC is considered to be the result of vascular malformations. Etiology is theorized to be, either it is primary or secondary to various other separate bone lesion like chondroblastoma, osteoblastoma, chondromyxoid fibroma, chondrosacrcoma, fibrous dysplasia, giant cell tumor. Recent genetic studies prove them to be neoplastic, 69% of primary ABC contain t (16,17) causing TRE17/USP16 oncogene upregulation leading to NF-KB and matrix metalloproteinase production. Matrix metalloproteinase breaks the extracellular matrix leading to rapid tumor growth, these translocations are absent in secondary lesions.[6]

Patients with ABC typically presents with pain and swelling of the involved limb, rarely with a pathological fracture. Unusual location like diaphysis or epiphysis and cancellous bone like scapula as in our case creates confusion in diagnosis.

Around 57% of lesion around the scapula are malignant, whose incidence increase with age. Out of the malignant lesions osteosarcoma being most common followed by chondrosarcoma and Ewing's sarcoma. Most common soft tissue sarcoma is of high grade undifferentiated pleomorphic sarcoma, liposarcoma and leiomyosarcoma. Whereas, osteochondroma being most common benign bone tumor, ABC is the second most common followed eosinophilic granuloma, fibrous dysplasia, arteriovenous malformation, giant cell tumor, osteoid osteoma. Among the benign soft tissue tumor lipoma being the commonest followed by elastofibromatosis, enchondroma, ganglion cyst.[7]

X-ray features of typical ABC are eccentric, expansile, lytic, metaphyseal in location with periosteal reactions depending on the activity of the lesion with erosion of the cortex. In our case the X-ray shown a more prominent soft tissue shadow with no calcified peripheral shell relative to the size of bony lesion. Bony lesion was occupying superior aspect of the spine of scapula, eccentric, lytic without periosteal reaction. Ultra sonogram confirmed fluid as content of the swelling. Typically, MRI of an ABC will show double density fluid filled level, septations, low signal intensity in T1W a high signal intensity in T2W images.

In our case MRI shown a well-defined iso to hyper intense lesion with narrow zone of transition in T1W image. A mildly hyper intense cystic lesion with fluid level, mineralization and sclerotic margin in T2W images. There were neither any neurovascular structure encasement nor communication to shoulder joint. No obvious septations seen within the lesion.

Atypical radiological features are suggestive of underlying primary bone tumors with secondary ABC like changes. Bone lesions associated with a soft tissue mass may represent a giant cell tumor or osteosarcoma with secondary ABC like changes. Similarly, a solid variant ABC can involve diaphysis of long bone, endosteal scalloping, variable degree of mineralization, lacking internal septations, shows intense enhancement on MRI with contrast, surrounding soft tissue and bone marrow edema. Aggressively growing primary ABC can even present atypically with wide zone of transition, epiphyseal in location, aggressive periosteal reaction, adjacent soft tissue extension without calcified peripheral shell. The uncommon presentation, location and atypical radiological features in our case mandates us to rule out other closest possibilities like unicameral bone cyst (UBC), telangectatic osteosarcoma.

Commonest location of UBC being proximal humerus and femur, which constitutes 80%, metaphyseal central with sclerotic rim. They are not expansile with a pathognomonic pathological fracture, classically described as" fallen leaf sign", lacks septation or fluid filled level, this rules out the possibility of UBC radiologically.

Primary ABC has four phases of growth initial phase, growth phase, stabilization phase and healing phase. Hence its radiological appearance depends upon the phase of the growth. Since our case had cortical erosion and soft tissue extension, it needed to be differentiated from telangectatic osteosarcoma. Most of the radiological features were similar in most of both the conditions. Telangectatic osteosarcoma is relatively a rare tumor in flat bones, only 7.9% were reported in non-appendicular skeleton. Major differentiating feature is fluid filled level, in ABC almost >2/3rd of the cavity is filled with fluid compared to telangectatic osteosarcoma (TOS) <2/3rd of the cavity is filled with fluid level.[8]

Non aggressive growth pattern, narrow zone of transition in T1W MRI images, less soft tissue involvement are features more suggestive of ABC ruling out the possibility of TOS which can be confirmed without pre-operative biopsy.[9,10]

We planned for excision and extended curettage of the lesion. Post-operatively shoulder range of movements improved. No recurrence was noticed till sixth month post-operatively.

Histopathology report shown fibro collagenous and fibro adipose cyst wall composing of numerous capillaries to dilated blood vessels and occasional one show endothelial cell proliferation with areas of hemorrhage, few clusters of atypical spindle cells surrounded by lymphocytic aggregates. All these features are suggestive of aneurysmal one cyst.[11]

CONCLUSION

Our case presented with all atypical features of ABC with regards to age, location and imaging. High degree of clinical suspicion and ruling out other closest possibilities like Telanectatic osteosarcoma is necessary when cystic swelling around the scapula is encountered. MRI with contrast can reveal many information. FNAC prior to definitive surgery will help in preoperative planning. Excision and extended curettage of the lesion is satisfactory. Periodic follow up with MRI and long-term clinical assessment for recurrence is needed.

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Ethical approval: Not required

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