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Management of traumatic diaphyseal fracture of humerus with incidental ipsilateral metaphyseal aneurysmal bone cyst in an adult: A case report

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Abstract
In children, an asymptomatic bone cyst usually presents as an incidental pathological fracture as its initial symptom while in adults it usually presents as post traumatic fracture commonly at cyst region. Management with conservative measures in children and adults alike give a satisfactory outcome. Case reports of fixation in adults for pathological fractures are reported. We present a case of skeletally mature young adult sustaining a diaphyseal communited fracture of left humerus with ipsilateral incidental metaphyseal lytic lesion of left proximal humerus. The management dilemma and strategy for management of both the pathologies simultaneously are discussed and detailed.

Keywords: Aneurysmal bone cyst; pathological fracture humerus; diaphyseal fracture humerus

Introduction
Cystic lesions of the skeleton are often detected as an incidental findings and majority of them are benign. Aneurysmal bone cyst is an expansile benign cystic lesion often detected in children and early adulthood [1, 2]. On plain radiography they present as an eccentric expansile lytic lesion in metaphysis and MRI may reveal fluid levels in involved region [1, 3]. There are various philosophies proposed for the causation of cyst including a vascular theory of increased venous pressure in bone [1]. Almost a third of aneurysmal bone cyst could be secondary to giant cell tumor and chondroblastoma [1, 2]. Pain and pathological fracture are the common presentations [1, 2]. Since its first report in 1942, the treatment of the cyst is evolving from the open surgical technique to recent percutaneous technique [1]. The percutaneous intralesional sclerotherapy, embolization, radiation, local cryotherapy with liquid nitrogen or open curettage and highspeed burring of the walls, have all been used with varying outcomes [1, 2, 3]. Management of the cyst becomes challenging when associated with pathological fractures [3]. Many reports in literature address the pathological fractures but mostly in children [1, 3]. In an adult, no known report which address a traumatic diaphyseal fracture of humerus with an incidental ipsilateral metaphyseal aneurysmal bone cyst available to guide us for the management of both entities simultaneously. We report and discuss challenges and considerations in management of this coexistent unusual occurrence.

Case report
A young adult male, twenty-five years old presented to emergency with pain and deformity of left arm after a fall while roller skating. He is right hand dominant person with good functional status prior to injury. The radiographs revealed a comminuted fracture of the diaphyseal shaft humerus along with an incidental finding of lytic lesion in metaphyseal region of proximal humerus (Figure 1). He underwent MRI imaging shows a well-defined lytic lesion of 3.8 x 3.2 cm size in proximal part of humerus in metaphyseal region appearing hyperintense on T2W images, fluid level with few incomplete loculations, spiral fracture involving the diaphysis of humerus with surrounding myofascial edema and subcutaneous edema especially biceps and triceps muscles with no focal area of marrow signal alterations (Figure 2a and 2b). Possibility of benign lesion was considered on MRI and a computerized tomography (CT) further differentiated the metaphyseal cyst and associated ipsilateral diaphyseal fracture as separate

entity (Figure 2c). The skeletal survey did not show any associated lesion in the rest of the skeleton.

A differential diagnosis of simple bone cyst, aneurysmal bone cyst or telangiectatic variant of osteosarcoma was deliberated upon, considering the age of patient and clinical and radiological features of the swelling. Biopsy was done to confirm the diagnosis which was consistent with aneurysmal bone cyst.

Surgical management with curettage, biopsy, phenol and electric burr cauterization with defect reconstruction by autologous cancellous iliac crest bone graft mixed with bio-substitutes of calcium phosphate granules and stabilization with AO proximal humerus interlocking plate (PHILOS) for ipsilateral diaphyseal and metaphyseal lesion done in the same sitting. The curetted material from lytic lesion sent for histopathological examination was consistent with aneurysmal bone cyst.

Wound healing and clinico-radiological progression was uneventful. A supportive slab post-operatively for three weeks was followed by active and passive range of movements for shoulder and elbow. At three months follow up patient had no pain, near normal range of movements for shoulder and elbow region. Radiologically fracture has consolidated with graft incorporation. At one year, patient is progressing clinically with an excellent range of painless shoulder and elbow movements and radiologically has no evidence of recurrence or fixation failure (Figure 3a and 3b). The patient was informed that data concerning the case will be submitted for publication and due consent taken for the same.

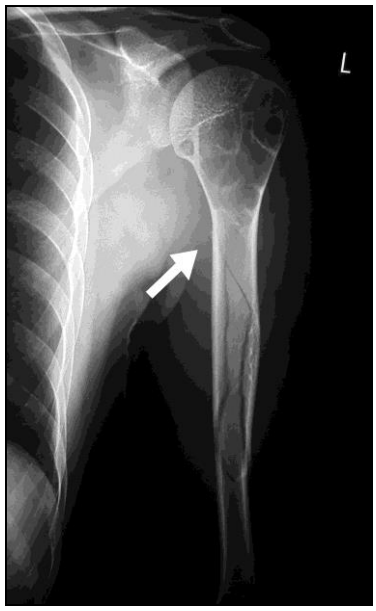

Fig 1: Preoperative anteroposterior radiograph of left shoulder and arm marked with white arrow demarcating metaphyseal cyst with diaphyseal fracture.

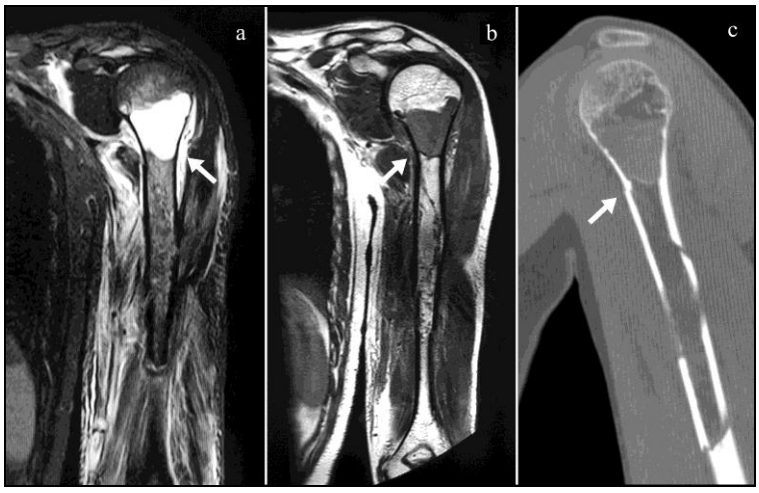

Fig 2: Preoperative MRI images 2a and 2b shows a well-defined lytic lesion marked with white arrow, appearing hyperintense on T2W image with few loculations and mild subcutaneous edema and Preoperative CT scan image 2c demarcates lytic lesion and fracture.

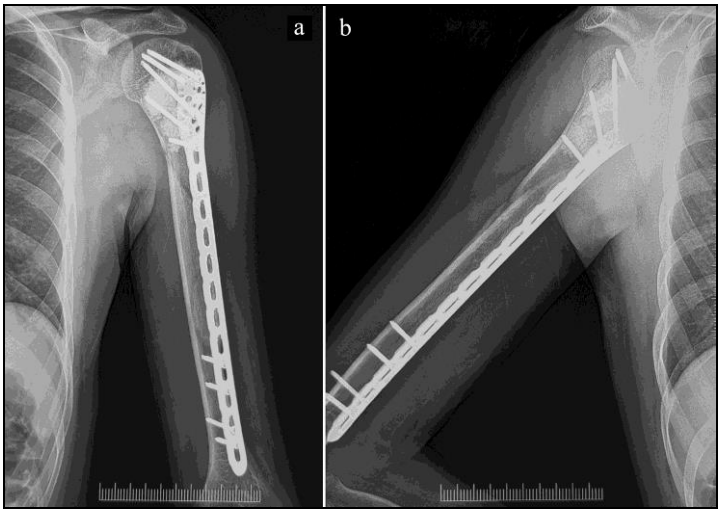

Fig 3: Radiologically at 12 months anteroposterior 3a and lateral 3b view show no recurrence or implant failure

Discussion

The treatment of aneurysmal bone cyst should be individualized depending on the location, aggressiveness and extent of the lesion. There are several treatment modalities with curettage grafting or en-bloc resection as the treatment of choice for accessible lesions [1, 3, 4]. Other treatment modalities include percutaneous intralesional sclerotherapy and cryotherapy. Cryotherapy has been proposed as an adjuvant therapy with surgical treatment to achieve local control [2]. Radiation and selective arterial embolization have also been used for less accessible or recurrent lesions [2, 3, 4]. Chemical cauterization with phenol is recommended for relatively large primary lesion to kill any surface tumor cells of the curetted cavity [4].

Large defects after curettage or resection of aggressive type of ABC with decreased mechanical support are difficult to treat. Various reconstructive options are available to fill these defects and provide bone integrity. An autogenic bone graft using cortico-cancellous graft from the anterior superior iliac spine, fibula or rib, either vascularized or non-vascularized, has been suggested as the best treatment method for the large defects [5]. Autogenic or allogenic bone grafts and many different bony substitutes like calcium oxalate granules have been used [5]. The goal of surgery in ABC with pathological or traumatic fracture should be to achieve adequate clearance of lesion, provide stability to allow potential healing with no recurrence. In addition, trying to obtain functionally mobile, active and stable joint and extremity is also important.

Management by immobilization alone predisposed to shoulder, elbow joint stiffness and an uncertainty regarding progression of cyst near an adjacent fracture remained. We used cortico-cancellous iliac ipsilateral autologous bone grafting with mixture of calcium oxalate granules after meticulous curettage of the cystic lesion with fixation of fracture with PHILOS. This allowed simultaneous management of cystic lesion and diaphyseal fracture and prevented further extension of fracture in the adjacent pathological lesion. It aided in early rehabilitation and allowed for an excellent functional outcome. A good pre-operative assessment planning, meticulous surgical technique with adequate fracture stability and patient cooperation for post-operative rehabilitation will ensure desirable functional outcome

Management of cystic lesion in young adults should be carefully evaluated prior to aggressive surgical management. In a cystic metaphyseal diaphyseal lesion with wide transition zone, as in our case, radiological diagnosis should be made and biopsy should be done to confirm the diagnosis. Failure to follow this protocol can lead to misdiagnosis of a metastatic tumor. However, possibility of late recurrence requires close follow-up.

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