Aneurysmal Bone Cyst—An Unusual Presentation of Wrist Pain

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Abstract

Keywords

- aneurysmal bone cyst
- expansile
- metaphysis
- distal ulna
- curettage

Introduction Aneurysmal bone cyst (ABC) is a cystic benign expansile lytic bone lesion, resembling multiple intraosseous blood-filled spaces. It is most commonly seen in the first two decades of life. Most frequent sites of involvement are metaphysis of proximal and distal femur, proximal tibia, and posterior elements of spine. It is seen more frequently in females. ABC of distal ulna is rarely reported in the literature.

Case Presentation We are reporting a case of ABC of a rare location—distal ulna—in a young male, which was successfully treated with extended curettage, and the cavity was filled with bone graft, with K-wire stabilization.

Aneurysmal bone cysts (ABCs) are benign locally destructive blood-filled reactive lesions of bone.¹ They are not considered to be true neoplasms. ABC accounts for 1% of all bone tumors.² They are most frequently seen in the first two decades of life.^{3,4} ABCs show slight female predominance. Though it can involve any bone of the body,⁵ the most common locations of ABC are distal femoral metaphysis and proximal tibial metaphysis. Other frequent sites of involvement are proximal femoral and humeral metaphysis and posterior elements of spine. Vertebral lesions account for 15 to 20% of the cases. ABCs involving distal radius and ulna are very rare.

ABC was first described by Jaffe and Lichtenstein in 1942 as a distinct entity.⁶ Although its etiopathogenesis is uncertain, it is believed to occur from circulatory disturbance within the bone.⁷ ABC can be primary, when arise de novo or secondary, when associated with giant cell tumor, osteoblastoma, chondroblastoma, chondrosarcoma, unicameral bone cyst chondromyxoid fibroma, non-ossifying fibroma, osteosarcoma, etc. Patients usually present with complaints of mild-to-moderate pain and swelling that has been present from weeks to months. Neurological deficits or radiculopathy can be seen in spinal lesions.

We are reporting a case of ABC of a very rare location—distal ulna, presenting with wrist pain and swelling, managed

successfully by extended curettage with satisfactory clinical outcome.

Case Presentation

A 14-year-old male patient presented to the outdoor patient department of our institute with complaints of pain and swelling over anterior aspect of right distal forearm for four and a half months. The pain and swelling were insidious in onset, without any history of trauma or fever. Pain was of constant, dull-aching type, and swelling has gradually increased up to the present size. The patient took conservative treatment from elsewhere, but got no relieve in symptoms.

Local examination revealed diffuse fusiform swelling of bony hard consistency and tenderness over anterior aspect of right distal ulna. The swelling was irreducible and incompressible with no visible pulsations. No skin changes were noted. Dorsiflexion of the wrist was restricted and painful. Ulnar deviation of the wrist was painful. There were no signs of neurovascular deficit. Routine anteroposterior (AP) and lateral radiographs of the right wrist with forearm revealed a normal wrist joint with expansile lytic lesion in the distal ulna involving the metaphysis and diaphysis with well-defined margins (► Fig. 1).

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Fig. 1 Preoperative radiograph, showing expansile lytic lesion in the distal ulna.

Magnetic resonance imaging (MRI) of the wrist (**Fig. 2**) showed a large well-defined, oblong, intramedullary, multiloculated fluid-containing expansile cystic lesion of $4.9 \times 2.4 \times 2.3$ cm in the distal ulnar metaphysis and diaphysis with narrow zone of transition. Multiple small fluid levels were seen in the periphery of the lesion. There was mild surrounding soft tissue edema. The MRI findings lead to differential diagnosis of simple bone cyst with secondary hemorrhage and ABC.

Treatment was done by extended curettage of the lesion up to approximately 1 cm proximal to the lesion. Ulna was

Fig. 3 Intraoperative image after curettage.

approached dorsomedially. **Fig. 3** shows the intra operative images after curettage. The thin shell of cortical bone was left intact. The residual cells were killed by chemical cauterization with phenol, and the cavity was filled with bone graft from the iliac crest. This was stabilized with K-wire, and plaster of Paris (POP) slab was applied. **Fig. 4** shows the immediate postoperative radiograph. Excised specimen was sent for histopathological examination, which revealed the diagnosis of aneurysmal bone cyst. POP cast was applied after stitch removal and was removed after 3 weeks. K-wire was removed after 2 months (**Fig. 5**).

Postoperative follow-up at 1 year showed excellent graft incorporation with no deformity at the wrist and good range of movements at wrist (**Fig. 6**). There was almost full dorsiflexion and palmar flexion at the wrist without any pain, and the patient could easily carry out his activities of daily living. There were no any signs of secondary infection.

Discussion

The ABC is an expansile, lytic benign bony lesion, leading to thinning of the cortical bone, without breaching it. ABC results from specific pathophysiological alteration, which can result from tumor or trauma-induced anomalous hemodynamic process. The most common precursor lesion is the

Fig. 2 Preoperative MRI images, revealing multiloculated fluid-filled lesion. MRI, magnetic resonance imaging.

Fig. 4 Immediate postoperative X-ray.

Fig. 5 Postoperative X-ray after K-wire removal.

giant cell tumor, followed by osteoblastoma, chondroblastoma, angioma, fibrous dysplasia, fibroxanthoma, chondromyxoid fibroma, unicameral bone cyst, fibrous histiocytoma, eosinophilic granuloma, and even osteosarcoma.⁸ Approximately, 80% of patients presenting with ABC are <20 years of age.⁸

More than half of lesions involve the long bones, and one-third involve the spine. The pelvis accounts for 10% all lesions.⁹ The long bones principally involved are tibia, femur, pelvis, and humerus.¹ Although it most commonly involves the metaphysis, three cases of primary epiphyseal ABC have been reported in proximal tibia, distal tibia and distal ulna.^{10–12} Diaphyseal ABC has been reported in humerus.¹³

Here, we are reporting a case of aneurysmal bone cyst, involving the distal ulnar metaphysis and meta-diaphyseal region. There is only one case of ABC of distal ulna in the literature, which is primarily involving the epiphysis.¹²

Radiological examination reveals a radiolucent expansile lytic lesion that elevates the periosteum, but remains contained within thin shell of cortical bone. The lesion is mostly eccentric situated in the metaphyseal area of long bones.¹⁴ MRI findings include an intact hypointense rim completely surrounding the lesion, with multiple intralesional septations and cysts with fluid–fluid levels of varying intensity.¹⁵ Definitive diagnosis can be made by histopathological examination. Grossly, ABC is a cavitary lesion, with blood-filled

Fig. 6 X-ray and clinical images at the final follow-up.

septate spaces. The entire lesion is surrounded by a thin rim of bone. Microscopically, it consists of hemorrhagic tissue with cavernous spaces, separated by cellular stroma.¹⁶

Surgical treatment with extended curettage and grafting with bone graft or bone graft substitute is usually recommended. Arterial embolization can be used as treatment modality at locations, where curettage is difficult.¹⁶ After treatment, the rate of recurrence of ABC is 10 to 30%,^{17,18} with higher recurrence in very young children.¹⁹ Postsurgical treatment, with local application of alcohol, phenols, or liquid nitrogen to kill the residual cells, reduces the risk of relapse and recurrence.²⁰ Recurrence can be treated with the same approach as primary.

The common differential diagnoses for ABC are giant cell tumor, telangiectatic osteosarcoma, chondroblastoma, fibrous histiocytomas, and chondromyxoid fibromas which can be distinguished on the basis of clinical, radiological, and histological aspects.

Conclusion

In conclusion, we present a rare non-neoplastic tumor of long bones—aneurysmal bone cyst, involving a rare site—the distal ulna. Accurate preoperative diagnosis helped in planning the treatment and achieving a satisfactory outcome.

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Conflict of Interest

None.

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