

# A Rare Case of Aneurysmal Bone Cyst of the Distal Humerus in an 11-Year-Old Child

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## ABSTRACT

Aneurysmal bone cysts (ABCs) are expansile, blood-filled, intramedullary lesions occurring in the metaphysis of young adults. Their etiology is still unclear. These lesions have often been associated with other neoplasms. Traditional surgical treatment included en bloc resection or intralesional curettage with bone grafting or cement insertion. The recurrence rate has been reported to be almost 25%. This was a case of an 11-year-old child with ABC of the distal humerus treated by intralesional sclerosant injections. The patient showed good radiological and functional recovery post-procedure. This was done while avoiding all the major complications associated with surgery.

**Key words:** Aneurysmal bone cyst, humerus, polidocanol, sclerosant

## INTRODUCTION

Aneurysmal bone cysts (ABCs) are expansile, blood filled, intramedullary lesions occurring in the metaphysis of young adults. They are benign, vasocystic tumors. The lesions are commonly seen in young adults in the age group of 10–20 years. They have a slight female predominance.<sup>[1]</sup> They may be either a primary pathology or be secondary to other benign antecedent tumors.<sup>[2]</sup> A history of trivial trauma is seen associated with many cases. They occur in the metaphysis of long bones with the distal femur and proximal tibia being the common sites.<sup>[1,3]</sup> The etiology is often debated and unclear. The standard treatment includes curettage of the lesion followed by

bone grafting or cement insertion. This is combined with adjuvant therapy. The rates of recurrence are still high in these cases.<sup>[4]</sup> The action of the sclerosants results in damage to the endothelium of blood vessels. This triggers the coagulation cascade and results in thrombosis. It is a comparatively newer modality and has shown promising results with lower recurrence rates. Polidocanol (hydroxypolyaethoxydodecan) is a sclerosing agent which has been found to be effective in the treatment of varicose veins. It has been shown to have faster pain relief and better functional outcomes. This case report studies the outcome of an ABCs of the distal humerus in an 11-year-old child treated with polidocanol therapy.

## CASE REPORT

An 11-year-old female child presented to the outpatient department of a tertiary care center in rural India. She had complaints of gross swelling over the left elbow which was associated with intermittent pain and severe restriction of elbow movements for 9 months. The antecedent history was that of trivial fall while playing. The swelling was first noticed by the patient 20 days after the fall and was of approximately 2 cm in

### Access this article online

**Website:**  
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**DOI:**  
10.15713/ins.mmj.53

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diameter. It gradually progressed to present size. The patient had taken over-the-counter analgesics during this period and had not sought any other treatment. This was not associated with recurrent fever or any constitutional symptoms.

On examination, the swelling was found to be bony hard in consistency. There was no localized increase of temperature or focal tenderness over the lesion. The lesion itself was 7 cm × 5 cm in size. The medial epicondyle and the olecranon process could be palpated. The lateral epicondyle was grossly enlarged and involved with the swelling. The elbow arc of motion was restricted from 30° to 80° [Figure 1]. There was no neurological involvement. The wrist and shoulder movements were not affected. No other bony swellings were found over the body.

## Investigations

A plain X-ray of the elbow joint in the anteroposterior and lateral view was taken. It showed the presence of an eccentric radiolucent lesion over the medial aspect of the distal humerus. It had well-defined borders and multiple osseous septations were visible in the lesion [Figure 2]. The inflammatory blood markers were mildly elevated. A magnetic resonance imaging (MRI) was planned. It showed a lobulated lesion of the distal humerus with multiple fluid levels. There were few hypointense areas suggestive of hemorrhagic areas with a peripheral hypointense rim [Figure 3 a-f]. Based on these findings, a provisional diagnosis of ABCs was suggested.

The patient was then posted for open biopsy. A biopsy was taken from the posterior aspect of the lesion using a bone biopsy needle. The cortex of the lesion was paper thin and was broken on needle insertion. The biopsy gave small fragments of bony wall and a hemorrhagic aspirate. Histopathological examination showed presence of hemorrhagic cells without any atypical/malignant population. The aspirate yielded

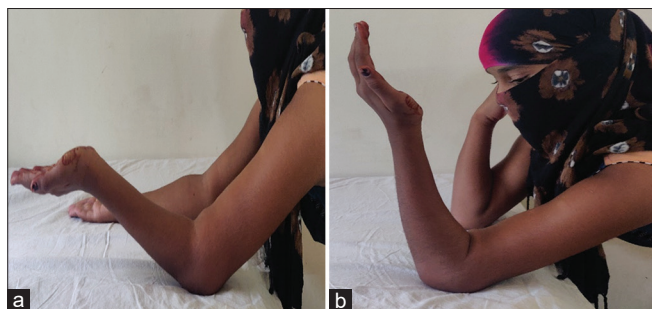
no growth on culture media. A second sample sent for histopathology showed presence of osteoclastic giant cells, histiocytes, and the presence of fibrosis. This was consistent with the radiological diagnosis of ABC.

## Treatment

Intralesional sclerosant injections were planned after duly explaining to the relatives the pros and cons of surgery and sclerosant therapy. A written informed consent was taken. Four milliliters of polidocanol injection (60 mg/2 ml – Asklerol 3%) were injected into the lesion using a wide bore needle using the same posterior approach used for biopsy. It was followed by a period of immobilization for the cortex break that occurred during biopsy. Post-immobilization physiotherapy was started for elbow range of motion. Two weekly X-rays were taken to monitor the progress of treatment. A repeat dose of sclerosant was again given after 2 months of first dose. Physiotherapy protocol was continued. Serial clinical and radiological monitoring of the case was done.

## Outcome

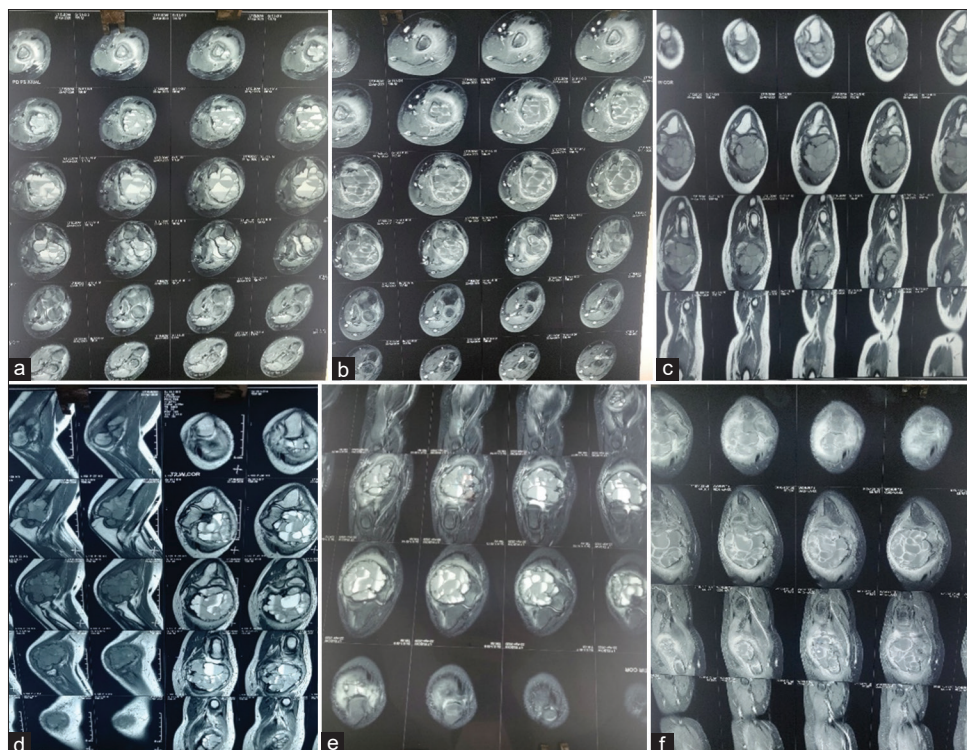
The patient was followed up serially up to a period of 3 months. Regular radiographs were taken and elbow range of motion was monitored. The patient was completely free from pain post-procedure. The elbow arc of motion had improved till 20° to 90° [Figure 4]. Serial radiographs showed presence of complete sclerosis of the lesion at 3 months follow-up. There was significant reduction in size on serial radiographs [Figures 5-7].



**Figure 1:** (a and b) Elbow range of motion at presentation



**Figure 2:** X-ray on presentation showing an eccentric, expansile, lytic lesion in the distal humerus



**Figure 3:** (a-f) Magnetic resonance imaging on presentation showing a lobulated lesion of the distal humerus with multiple fluid levels



**Figure 4:** (a and b) Improved elbow range of motion seen at 4 months follow-up

## DISCUSSION

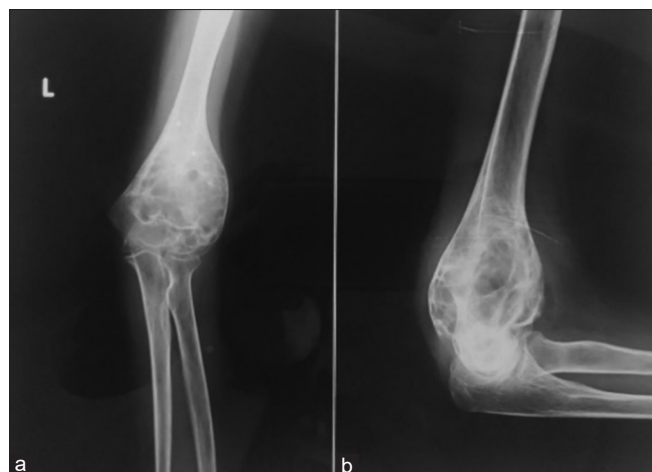
ABCs have a predilection for young children and adolescent. Most of the lesions are detected in patients <30 years age. The median age of the affected patients is 13 years.<sup>[5]</sup> The very name of the condition is a misnomer. The lesions are deficient of an endothelial wall. They are benign in nature. The lining of the lesions is formed by proliferative fibroblasts, giant cells, and trabecular bone. They are described as expansile lesions producing



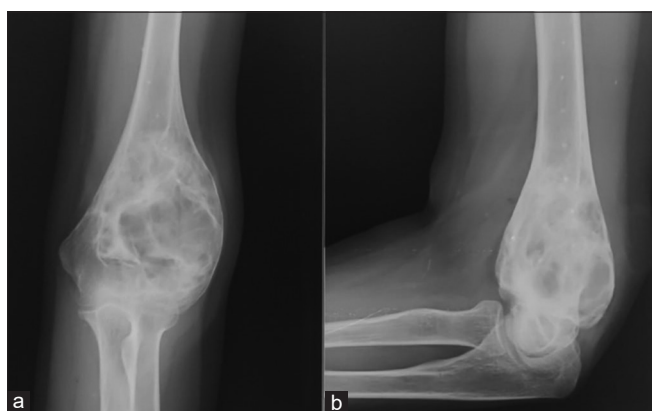
**Figure 5:** Follow-up radiograph taken 1 month post 1<sup>st</sup> sclerosant injection

blood-filled cavities within the bone and are neither cystic nor aneurysmal.<sup>[6,7]</sup> X-rays show the presence of a characteristic soap bubble appearance. They classically appear as eccentrically located radiolucent cystic lesions circumscribed by a thin layer of the cortical bone. An





**Figure 6:** Follow-up radiograph taken at 2 months following 1<sup>st</sup> sclerosant injection



**Figure 7:** Follow-up radiograph taken at 1 month post 2<sup>nd</sup> sclerosant injection

MRI done in conjunction improved the specificity and sensitivity of diagnosis. It reveals multiple fluid levels, internal septations, peri-lesional extension, and edema.<sup>[8]</sup> An accurate histological diagnosis is mandatory. An incisional biopsy remains the standard for the histological studies.<sup>[9]</sup> Earlier treatments included *en bloc* resections and radiotherapy but many studies have shown them to have significant adverse effects.<sup>[10,11]</sup> The current standard of care for ABCs is curettage. The need for a bone graft is based on the resultant void post-curettage. The recurrence rates vary across the spectrum of existing literature, some case series have noted rates as high as 59%.<sup>[12]</sup> Hence, it is often combined with adjuvant therapies such as argon laser, cryosurgery, and phenol injections. Sclerotherapy is a comparatively recent modality for the treatment of ABCs. It induces sclerosis of the vascular network of the lesion and helps to achieve local control. Two drugs are

preferred for this – Ethibloc and Polidocanol. Ethibloc has been documented to have local inflammatory reactions in up to 94% of patients. The other disastrous adverse effects include aseptic bone necrosis, pulmonary embolism, deep venous thrombosis, and cerebellar infarct leading to death.<sup>[13-16]</sup> In comparison with curettage, polidocanol sclerotherapy had a higher healing rate of 93.3% compared to 84.8% of curettage.<sup>[14]</sup> A good clinical response was found with an average of three polidocanol injections.<sup>[17]</sup> Polidocanol was found to have faster pain relief and was associated with better functional outcomes while avoiding surgery.<sup>[14]</sup>

## CONCLUSION

This case of an Aneurysmal Bone Cyst was well managed with sclerosant injections. A substantial reduction in the size of the lesion was observed on serial followup. Gradual sclerosis of the lesion was seen radiographically. Unfortunately due to the late presentation of the patient, the elbow joint was already involved and a significant reduction in range of motion was present. Through physiotherapy functional range of motion was restored so the patient could carry out her activities of daily living. Two injections of Polidocanol were enough in this case to provide satisfactory resolution of the lesion.

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**How to cite:** Godbole SR, Borkar SS, Nandanwar AY, Hegde V, Patil S, Desai S. A Rare Case of Aneurysmal Bone Cyst of the Distal Humerus in an 11-Year-Old Child. *MIMER Med J* 2020;4(2):37-41.

**Source of Support:** Nil. **Conflicts of Interest:** None declared.

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