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Case Report

A case report on the rare presentation of aneurysmal bone cyst on proximal radius with management

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ABSTRACT

Aneurysmal bone cyst (ABC) is a non-neoplastic vaso-cystic tumor of the second decade. A 16-year-old male patient presented to the outpatient department with gradual dull aching pain and progressive increasing swelling, which was localized to right lateral elbow and upper part of the forearm. There was no restriction in the range of motion. After getting an x-ray, a lytic lesion is found at the proximal radius. MRI and cytology, are both investigations done to confirm the diagnosis. Two injections of 10 ml polidocanol (3%) were administered percutaneously two months apart under the guidance of an image intensifier. After one year X-ray showed marked sclerotic, however, the tumor size remained almost the same. Although ABC is a non-aggressive benign lesion. ABC responds to treatment very well. recurrence is also common. Proximal radius is a less common site for ABC and the case report suggests that it is curable. We found no recurrence. Treatment with polidocanol is less expensive, less morbid, has a good functional outcome, early discharge from the hospital, and highly effective.

Keywords: ABC, Proximal radius tumour, Polidocanol, Fibrosing agent

INTRODUCTION

Aneurysmal bone cysts (ABC) are rare benign tumors comprising 1-6% of primary osseous tumors but can be locally destructive. It was first reported by Jaffe and Lichtenstein in 1942, in the lesions associated with the spine and pelvis.[1] Though it has the name ABC, there is no true cyst or aneurysmal lesion.[2,3] Although many hypothesis have been developed over the years till today the etiology of ABC is unclear. Many authors have proposed that it can be primary or secondarily in a known precursor.[2] Most cases are found amongst children and young adults most of whom are less than 20 years with slightly increased incidence in females (1 to 1.3 times more common than males).[4] The majority of the lesions are found in the metaphysis of long bones (distal femur, proximal tibia, proximal humerus, and distal radius), and (mostly from the posterior element) and pelvis. Rarely it can involve craniofacial bones and epiphyses.[5] Histologically, they are multi-cystic cavernous spaces filled with blood, containing septa trabeculated bone or osteoid tissue. The whole cavity is enclosed in a subperiosteal shell of reactive bone. The stroma is composed of fibroblasts, spindle cells, osteoid, and multinucleated giant cells.[3,5] ABCs may be primary or secondary (lesions aroused from osteoblastoma, chondroblastoma, giant cell tumors, and others).[6] Association of USP-6 gene up-regulation and CDH11 with ABC is found, and define it is a primary neoplasm. Spontaneous regression may be seen in the active primary ABCs but rarely in aggressive and secondary lesions.[6,7]

However it has two types (primary and secondary) and diagnosis is made with the help of various investigations

like an x-ray (eccentrically located radiolucent cystic lesions circumscribed by a very thin layer of the cortical bone, soap bubble appearance), Computed tomography (presence of periosteal rim surrounding the lesions), magnetic resonance imaging (on T2-weighted images high signal levels and layering in the blood means fluid-fluid level) and biopsy.[5] Mahnken et al found that with help of MRI and x-ray we can achieve high specificity and sensitivity to confirm the diagnosis.[8] Historical treatment for ABC include curettage followed by reconstruction, EN bloc excision, radiotherapy and high speed burr. Argon beam coagulation, phenol, cryosurgery are other alternatives. New modalities like sclerotherapy by polidocanol, curopsy, and percutaneous doxycycline have recently been proposed as a treatment for ABCs.[5]

It presents as pain and swelling at the lesion, sometimes pathological fracture. Although long bones are the most common site, the proximal radius is a less common site of ABCs. Here is a case report of a 16-year-old male patient having ABC at the proximal radius

CASE REPORT

A 16-year-old male patient visited the department of orthopaedics with a chief complaint of pain and swelling in the upper elbow on the right side for 2 months. The pain was gradual in onset, progressive, localized to right lateral elbow and upper part of forearm. It was dull aching in character, mostly aggravated by movements and lifting of objects but relieved by taking rest, and oral analgesics. There was no other swelling in any other body parts. The swelling was of a gradual onset; it was initially small and progressed to the present size (5×3 cm). The swelling was associated with occasional paraesthesia of the right side of the elbow. The patient did not give any previous history of trauma. Family history and other history were unremarkable (Figure 1 A).

On examination skin over the swelling appeared normal and there was no evident ulceration, suppuration /discharge. No visible pulsations were noted. On palpation, there was no local rise in temperature, the swelling was over the lateral aspect of the elbow with restricted movement, mild tender, smooth-surfaced, firm to bony hard in consistency, non-compressible and non-reducible. The range of motion (ROM) in the left elbow joint was 0-130° of flexion, 45° of pronation, and 45° of supination. Distal neurovascular status was normal.

All laboratory investigations were within normal limits. The radiograph showed eccentric ballooning expansion and radiolucency with septate loculations (Figure 1 B). MRI showed that the lesion was centered within the metaphysis and appeared expansile with surrounding cortical thinning, without invasion into the adjacent physis. it was adjacent to the diaphyseal location of the proximal radius extending up to the subarticular location. There were multiple blood-filled cystic spaces with fluid levels. There were no underlying soft tissue elements noted, suggesting this was a primary ABC (Figure 1 C). Aspiration was performed which yielded blood, which on cytological examination showed scattered osteoclastic giant cells (Figure 2 A). The diagnosis of an unusual aggressive ABC was set, then we decided to manage this case with a 3% polidocanol injection. For every cm^3 of the tumor's volume, approximately 1 ml of 3% polidocanol is needed. Given that the highest dose that can be administered at once is 10 ml and that this equation only applies to tumours smaller than 3 cm. We chose to administer 10 ml (300 mg) of polidocanol percutaneously while using a fluoroscopic C-arm. Initially, 2 ml of 2% lignocaine with adrenaline was infiltrated into the intended injection site followed by the aspiration of blood from the cavity, and then proceeded to inject 10 ml of 3% polidocanol into the tumor through a 16 G cannula. Injected fluid reflux was prevented by locking the cannula for some time and by injecting 1 ml saline. The post-injection period was uneventful and the patient was discharged the next day with analgesics. The patient was reviewed every two weeks. A second injection of same dose was given after six weeks and x-ray prescribed (Figure 2 B). After 2 months of the second injection, the pain subsided and the range of elbow movements improved. After one year, he is painless with a full range of elbow movements. X-ray showed marked sclerotic however, tumor size remained almost the same (Figure 3).

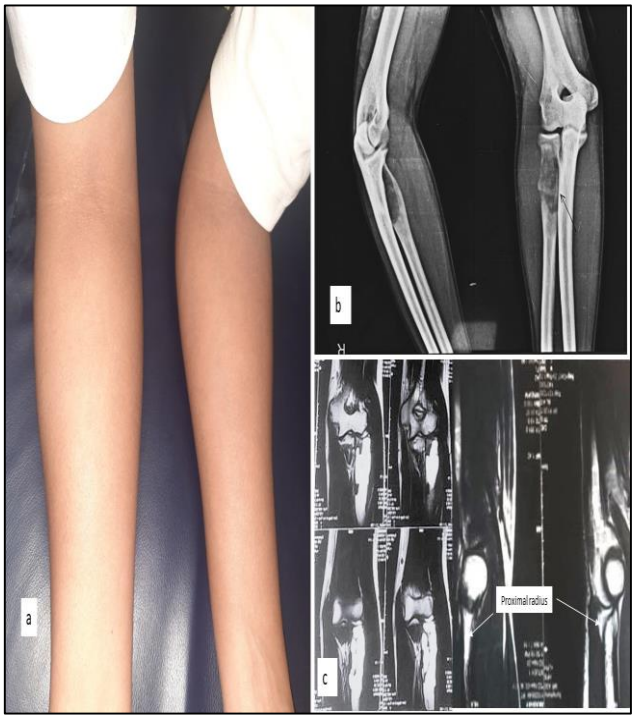

Figure 1 (A-C): clinical picture of elbow showing swelling at proximal radius; plain radiograph showing eccentric ballooning expansion and radiolucency with septate loculations; MRI of involved lesion showing multiple blood-filled cystic space lesion within the metaphysis and appeared expansile with surrounding cortical thinning on T2 weighted of coronal and sagittal.

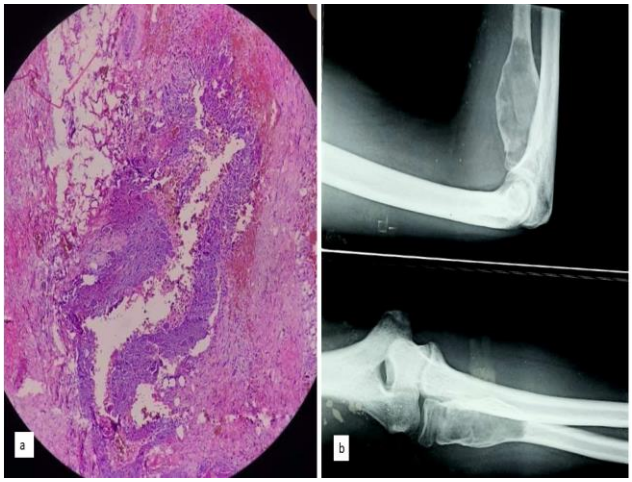

Figure 2 (A and B): blood-filled cystic spaces separated by septa containing woven bone, bland fibroblasts, and multinucleated osteoclastic giant cells; plain radiograph showing moderate sclerosis and thickening of cortex.

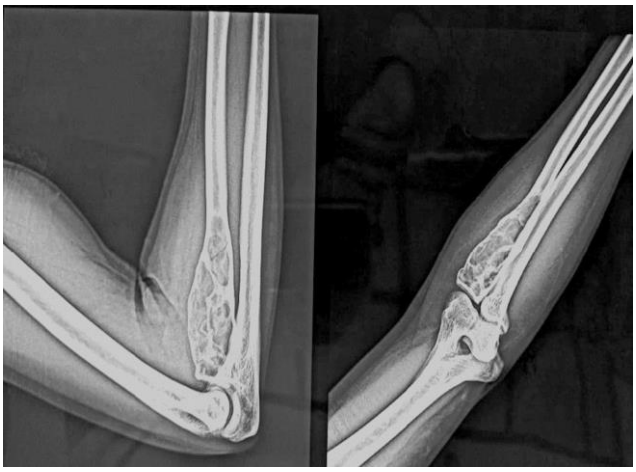

Figure 3: X-ray showed marked sclerotic changes however, tumor size remained almost the same.

DISCUSSION

Polidocanol (hydroxypolyaethoxydodecan) is a sclerosant agent, it initiates coagulation cascade by damaging the endothelium of the ABC. Rastogi et al conducted a study on 72 patients with a follow-up of 34 months and found a positive result of 84.5% after the administration of three injections of polidocanol.[9] This decreases the vascular system of the lesion; provides faster relief of symptoms with a decrease in morbidity and further need for surgery. Varshney et al did a comparison of polidocanol sclerotherapy versus curettage with high-speed burr followed by bone graft on 94 patients.[10] They found polidocanol therapy superior to an excision in terms of higher healing, good functional score, early discharge from the hospital, less blood loss, fewer chances of physis damage, and less invasive procedure. Puri et al conducted a study on 56 patients to analyze the effectiveness and complication of percutaneous use of sclerosant therapy (polidocanol).[11] Mean age of the study was 20 years at a follow-up of 62 months. In our case, the age of the patient was 16 years. The result of the study suggests that the effect of the single injection was 44% as compared to two injections which were 78%. We used 2 injections of polidocanol 2 months apart and followed up for 1 year. As far as no study compare the different type of fibrosing agent and their complications. Puthoor et al compared two groups of patients.[12] Group 1 having 31 patients and group 2 having 17 patients, were followed for 2 years. Group 1 was treated with the help of sclerotherapy using polidocanol but group 2 was treated with help of extended curettage. 100 percent healing was achieved in goup1 as compared to 82% in group 2 but in our case, 100 percent result was achieved and no recurrence was found. Puri et al and Puthoor et al commented on the better safety, high effectiveness, good cosmesis, and less invasive property of polidocanol.[11,12] Some study also provides evidence of complications of a sclerosing agent like pain, hypopigmentation, and local site necrosis. Which do not need severe attention, and mostly healed spontaneously. However, ABC has many modalities of treatment and all have a better outcome.

CONCLUSION

Although ABC is a non-aggressive benign lesion. ABC responds to treatment very well. Recurrence is also common. Above mention case report suggested that the proximal radial is a less common site for ABC but curable. We found no recurrence in this case till now. Polidocanol therapy offers a good functional outcome, is less expensive, less morbid, and requires a shorter hospital stay for the patients.

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