

Large Aneurysmal bone cyst of the proximal humerus, treatment through compound approach.

El Sayed Abd El-halim Abdullah MD and Awad Abdel Mmoniem Rafalla MD

1- El Sayed Abd El-halim Abdullah; Abdullah EA; MD
(Alexandria University, Faculty of Medicine, Orthopedic Surgery department, Assistant professor of Orthopedic Surgery, Egypt).

2-Awad Abdel Mmoniem Rafalla ; Rafalla AAA; MD
(Alexandria University, Faculty of Medicine, Orthopedic Surgery department, Assistant professor of Orthopedic Surgery, Egypt)

Corresponding author ; El Sayed Abd El-halim Abdullah, El Hadra University hospital, Department of Orthopedic surgery and traumatology, Embrozo, Alexandria, Egypt.
Tel 002 01006533993
e-mail sayed_halim@yahoo.com

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Abstract

Background

Although there are many treatment options, the optimal treatment of ABC is controversial.

Methods

The study included thirty four patients suffering from large ABC of the proximal humerus. They were treated by thorough curettage, wash of the cyst by normal saline and hydrogen peroxide and non -vascularized autogenous fibular graft.

Results

After a mean duration of 6 years .The mean overall functional outcome was 97% . 2 recurrent lesions, one had complete recurrence and the other showed partial recurrence

Conclusion

This technique is effective, allows early active mobilization of the limb with minimal rate of recurrence.

Keywords

Aneurysmal bone cysts, Fibular graft, Bone tumors.

Introduction

Aneurysmal bone cysts (ABC) are benign locally aggressive lesions and may extend to soft tissue. They have a great potentiality for local recurrence. [1] Their expanding nature can reach the growth plates resulting in growth arrest.[2] Commonly appear in children and teenagers, they haven't sex predilection, commonly present in the metaphysis of long bone but can arise at any site of the skeleton.[3] Radiographically, they appear as multiple lytic lesions separated by bony trabeculae, eccentrically located with thinning and ballooning of the cortex.[4] These lesions appear as multiple fluid-fluid in magnetic resonance images (MRI).[5] Histologically, they contain blood-filled spaces separated by the septae containing osteoid tissues, osteoclastic giant cells and fibroblasts.[3]

There are different reported options for treatment of ABC including surgical resection of the lesion, curettage with or without bone grafting.[6] Various adjuvant treatments have been used including bone cement, argon beam, phenol, ethanol and cryotherapy.[6] selective arterial embolization, external-beam radiotherapy, percutaneous directed therapies which include sclerotherapy, ablation, bisphosphonate and

doxycycline percutaneous injections.[7,8] Systemic therapy with receptor-activator of nuclear κ B ligand (RANKL) inhibitors (Denosumab).[9] Computed tomography (CT) guided radiofrequency ablation.[5]

The aim of the work was to evaluate the results of treatment of the large aneurysmal bone cysts of the proximal humerus by thorough curettage, wash of the cyst by normal saline and hydrogen peroxide and non -vascularized autogenous fibular graft.

Patients and methods

The study included thirty four patients suffering from large ABC of the proximal humerus. The patients were treated between 2007 and 2017. The large nature was determined according to a radiographic ratio described by Kanellopoulos AD et al 2007 [10] to ascertain the severity of the lesion (fig 1).

Age ranged from seven to seventeen year old at the time of presentation with mean age of twelve year .Twenty were females (59%) and 14 (41%) males. Left arm was affected in twenty one (62%) patients

.Three patients (9%) presented with pathological fracture (fig.2)



Fig.1: The extent of the lesion on the longitudinal axis was divided with the normal expected diameter of the long bone at the site of the lesion. The large lesion was described when it occupied more than 2 times the physiologic diameter of the long bone at the site of the lesion. [10]



Fig.2: Large ABC of proximal humerus with pathological fracture.

All patients were diagnosed by plain X-ray at presentation . MRI was done for clinically and radiologically aggressive lesions in 11 (32%) patients .Core needle biopsy was required in 3 cases (9%) to exclude sarcoma (fig.3).The proximal humeral physis was involved in 4 patients.



A



B

Fig.3: A, plain X-ray and B, MRI of large aggressive ABC of proximal humerus.

Under general anaesthesia and through anterolateral approach, a longitudinal window over the cyst that was large enough to expose most of the cyst was created. All patients had received a thorough curettage of the cyst, thorough wash of the cyst by normal saline and hydrogen peroxide solution 20%. The resultant weak longitudinal cystic segment of the proximal humerus was augmented by impaction of an autologous non vascularized fibular graft . This graft was harvested subperiosteally from ipsilateral leg under tourniquet. Primary rigid impaction of the graft was a necessary (fig.4).Minimal internal fixation was required in 3 patients to stabilize the graft (fig.5).

Post-operatively, an arm sling was used to stabilize the limb for a period from four to six weeks, after that active exercises and use of the limb was started.

The patients were evaluated both clinically according to the musculoskeletal tumor society system (MSTS) [10] and radiographically at the end of follow up.

An approval was given by the institutional review board (IRB) and informed consent was obtained from each patient or parents.

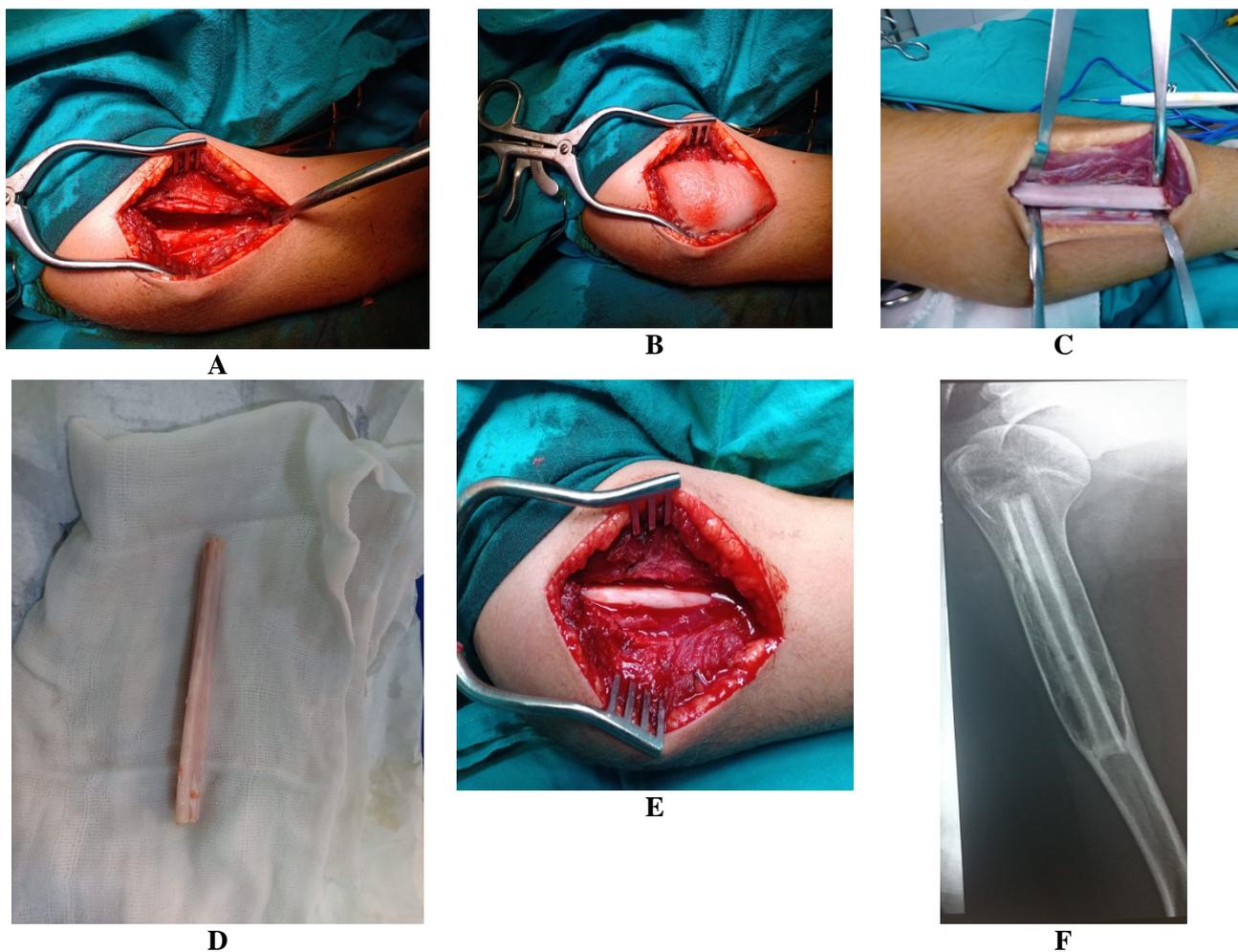


Fig.4: A,Cortical window with thorough curettage. B, Wash with saline and hydrogen peroxide. C, Subperiosteal harvesting of fibular graft. D, non-vascularized fibular graft. E, impaction of the fibular graft. F, follow up X-ray showing healed lesion.

Statistical analysis was done using the statistical program for social sciences (SPSS)-version 20 (Armonk, NY: IBM Corp). [11] t-test was used to analyze the relations between the obtained results and the different variables. P<0.05 was considered significant

Results

The patients were followed up for a mean duration of 6 years (range from 2 to 10 years). The average time of healing that allowed the patients to do active exercises without pain was 7 weeks (range from 6 to 8 weeks). The average time for complete healing of the lesion was 3 months (range from 2.5 to 5 months).

The mean overall functional outcome was 97% (range 90% to 100%) (fig.6). There was significant difference between the pre-operative limb functions and those at the final follow up ($p=0.002$). We didn't find significant relation between the final functional outcome and age, sex and side of the lesions ($p=0.231$, $p=0.341$, $p=0.211$ respectively). There were 2 (6 %) recurrent lesions, one had complete recurrence and the other showed partial recurrence (fig.6).



Fig.5: Minimal internal fixation with screws in a case of ABC with pathological fracture.



Fig.6: Thirteen years old patient with ABC. **A**, Pre-operative X-ray shows large ABC proximal humerus. **B**, X-ray after 3 years shows complete healing of the lesion.

Discussion

Aneurysmal bone cysts are locally aggressive benign lesions. The optimal treatment of ABC is controversial. Although, there are many treatment options, still curettage with or without bone graft the predominate treatment option but it has a rate of recurrence of 20 %.[12] In the study of Kececi et al 2014 [13] to evaluate the effect of phenol as an adjuvant after curettage, they found slight difference in the rate of recurrence with or without phenol in 2 studied groups (14 %, 16 % respectively).

Other methods like Argon beam coagulation, cryosurgery and cementation have either the risk of osteonecrosis and fracture or leave a biologically inert implant in a meta-epiphyseal location in children or

young adults.[6] In the study of Grahneis et al 2019 [3] they reported high local recurrence with repeated percutaneous polidocanol injections. Ethibloc and polidocanol haven't been recommended in many centers due to the reported complications like pulmonary embolism, skin necrosis, pain, swelling, and fever.[14,15] Shiels et al. 2016 [16] reported a healing rate of 94% with percutaneous doxycycline injection, however the patients required multiple sessions of injections (2-14 sessions per patient) with risk of skin necrosis due to extravasation of the drug. Denosumab, a human monoclonal antibody against the kappa B ligand (RANKL) which promotes osteoclast activation is also being used in ABC. It is recently used, but the studied cases were few with a short follow up, also some authors reported sever acute hypercalce-

mia after denosumab withdrawal in children.[3,17,18] Repeated arterial embolization have also been used with success rates of 80% and higher.[19]

Aiba et al 2018 [20] reported success rate of 90 % in a case series of patients with ABC who were treated with endoscopic curettage under arthroscopic guidance. Recurrence was occurred in 10% of cases. These results were comparable to the results of our study. But urged careful consideration of this method in patients less than 10 years and when the lesion comes in contact with the physis.

Curettage and high speed burr was used for treatment of ABC in the study of Steffner RJ et al 2011 [21], they reported considerable rate of post-operative fracture, and concluded that, this method didn't decrease the rate of recurrence. However, their results were improved when they add argon beam coagulation to curettage and high speed burr.

We reported 100% healing rate and overall final functional score of 97%. Our technique can manage all types of ABC including those with pathological fractures. We think that pathological fractures couldn't be managed by the recent percutaneous methods due to leakage of the agent into soft tissues with subsequent inefficacy and complications. The fibular graft acting as an osteoconductive material also it provided immediate mechanical stability that allowed early active motion before complete healing of the lesion. Fibular graft had many advantageous in comparison with iliac bone graft; 1. It can fill large lesions, 2. It didn't need internal fixation in most of cases, 3. It provides primary stabilization, 4. It allowed early mobilization of the limb. 5. We didn't report any donor site morbidity in our study.

Thorough wash with saline and hydrogen peroxide was used after curettage and before fibular graft impaction to kill the residual tumor cells.

The internal fixation was needed in 3 cases because the graft wasn't stable after impaction. It was in form of screws in one case and Kirschner wires in two cases.

We reported 2 cases with recurrence during follow up period. One had a small focal recurrence after 3 years of initial treatment, it was discovered accidentally during radiographic follow up and was treated by simple curettage, the lesion then healed with excellent functional outcome. The second case had complete recurrence of the lesion after 2.5 years and she was treated by formal technique with fibular graft harvested from the site of previous one due to complete regrowth of the donor site, the lesion completely healed with excellent functional outcome.

Although, the proximal humeral physis was involved in 4 cases in the study, we didn't report significant effect on the limb length, appearance and function. This was attributed to: 1. The age of these patients was ranged between 15-17 years, these patients had minimal remaining growth potential of the limb. 2. The minimal limb length discrepancy (LLD) in the upper limb is accepted and isn't noticeable as in the lower limb. 3. Minimal LLD in upper limb doesn't disturb the function of the limb.

Conclusion

Although there are many methods for treating ABC, our technique for management of ABC of proximal humerus is effective, allows early active mobilization of the limb with minimal rate of recurrence. The fibular graft is easy harvested without any complications.

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