The Egyptian Journal of Hospital Medicine (January 2023) Vol. 90 (2), Page 2446-2450

Can Extended Curettage Alone Be Sufficient for Treating An Aneurysmal Bone Cyst in The Upper Extremity?

Mohamed Abd El Aziz M. Ali*, Mohammad Hassan Abd Ellatif, Ahmed El Nagar
Orthopaedic Surgery Department, Faculty of Medicine, Zagazig University, Egypt
Corresponding author: Mohamed Abd El Aziz M. Ali, **Mobile:** (+20) 01026706518, **Email:** mohamedghith77@gmail.com

ABSTRACT

Background: Different treatment options for aneurysmal bone cysts (ABCs), which have a high risk of recurrence, are discussed. The choice of the treatment approach for patients with ABCs is based on the likelihood of recurrence and any potential side effects. **Objective:** This study aims to evaluate the outcome of extended curettage alone as a method of treatment for patients with ABCs of the upper extremity.

Patients and Methods: From April 2013 to June 2021, 15 primary ABCs in the upper extremity (15 patients) were diagnosed and managed at the Orthopedic Oncology Unit, Zagazig University Hospital. The patients were 9 males and 6 females. The mean age was 13.2 ± 1.6 years (range, 7 years 5 months to 18 years 2 months). ABCs were present in the proximal humerus in 7 cases, distal humerus in 3 cases, radius in 3 cases, glenoid in one case, and distal phalanx of the thumb in one case. All patients were managed by extended curettage and followed up for at least one year.

Results: All lesions healed after a mean period of 16 weeks (range 10–21 weeks). The time to healing was related to the age of patients, size, and behavior of the lesion. Superficial infection was seen in one patient and was managed conservatively. Recurrence developed in one patient at 9 months postoperatively and was managed by repeated extended curettage. There was only one patient who developed a shortening of the humerus of about 1.5 cm due to premature fusion of the proximal humeral epiphysis but without angular deformity.

Conclusion: Extended curettage with high-speed burr is considered an effective technique in the management of ABCs in the upper extremity.

Keywords: Aneurysmal Bone Cyst; Extended Curettage; Upper Extremity.

INTRODUCTION

The medulla of the metaphysis of long bones is frequently affected by benign lesions called aneurysmal bone cysts (ABCs). Rarely do these lesions develop in the cortex or in the outer layers of the diaphysis. Patients with ABCs typically have a shorter lifespan than 20 years [(1)].

ABCs are single lesions that can develop as original lesions or secondary lesions next to other lesions such giant cell tumours, chondroblastomas, or osteoblastomas [(2)]. 1.4% of all primary bone tumours are ABCs, which typically afflict the lower limbs. Only 5% of the lesions, or approximately 22.5% of ABCs, are seen in the hands [(3)].

Patients with ABCs frequently have a slow development of discomfort and edoema, albeit they can also exhibit pathological fractures. An eccentric lytic expansile lesion with bone septae across the cortex, giving the lesion a multi-locular look that is referred to as a "soap bubble appearance," is the characteristic radiographic feature. However, other imaging methods are necessary in addition to X-rays in order to properly diagnose the lesion. The osseous margins of the lesion can be seen on CT images. As ABCs has numerous layers of blood on top of one another, the MRI scans show multiple fluid lines. Additionally, MRI can detect edoema, perilesional extension, and bone septae. Though, the previously mentioned characteristics, such as the "soap bubble look" and "many fluid lines," are not pathognomonic for ABCs because other lesions, such giant cell tumours, osteoblastomas, and telangiectatic osteosarcomas, can also exhibit similar characteristics. Therefore, a biopsy and histological analysis are required for a firm diagnosis of ABCs [(4)].

Treatment attempts to limit the likelihood of recurrence, relieve pain, heal the lesion, and prevent pathological bone fracture [(5)].

A variety of treatment options are discussed, including bone marrow injection, cryoablation, radiotherapy, radionuclide ablation, intralesional curettage with or without adjuvants, en bloc resection, embolization, percutaneous injection of polidocanol or doxycycline inside the lesion, and use of denosumab or bisphosphonates [(6)].

The choice of treatment for patients with ABCs depends on the likelihood of recurrence and potential side effects of the chosen therapy [(5)].

This study aims to evaluate the outcome and effectiveness of extended intralesional curettage alone as a method of treatment for patients with ABCs of the upper extremity.

PATIENTS AND METHODS

This a retrospective study included 15 patients with 15 primary ABCs in the upper extremity. The diagnosis based on a review of clinical, radiologic, and pathologic records of patients

. From April 2013 to June 2021. The patients were treated by intralesional curettage with a minimum follow-up of 2 years. Patients with secondary aneurysmal bone cysts or cysts in the lower extremity were excluded from the study.

Received: 25/09/2022
Accepted: 28/11/2022

Ethical approval:

An approval of the study was obtained from Zagazig University Academic and Ethical Committee.. After explaining our research objectives, written informed consent was obtained from all study participants. This study was conducted in compliance with the code of ethics of the world medical association (Declaration of Helsinki) for human subjects.

The age and sex of patients at the time of presentation were identified. All patients suffered from gradual onset of pain and swelling **(Fig. 1)**. Plain radiographs were done for all cases revealing eccentric expansile lytic lesions **(Fig. 2).**

Fig. (1): clinical photos showing large thumb swelling

Fig. (2): Plain radiographs showing ABC (a) glenoid, (b) distal radius, (c) distal phalangex of the thumb

Computed tomography (CT) scan was done in 5 cases to assess the lesion for subtle corticaldestruction or fracture **(Fig. 3).** MRI were done for all cases to identify multiple fluid levels and the extension to the epiphysis **(Fig. 4).**

Fig. (3): Computed tomography of the glenoid

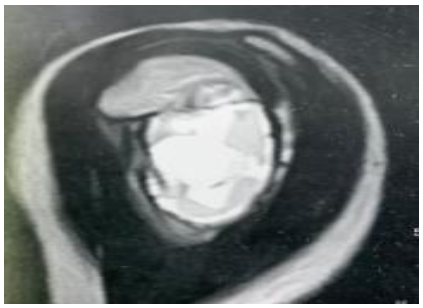

Fig. (4): MRI of the proximal humerus showing multiple fluid levels

Histologic diagnosis using guided core biopsy was examinated. All patients underwent intralesional curettage that was extended with the use of high-speed burr and nothing was used for cavity filling after curettage.

Surgical technique:

After the revision of all preoperative data, all cases were managed under general anesthesia. with the use of a tourniquet when feasible. An image intensifier was used to identify the cyst and the physis. After full skin preparation and draping, the lesion was exposed by the appropriate approach according to its site to provide easy access to the lesion, enable the creation of a sufficient window, and provide clear visualisation for curettage, then a wide side window, and extended curettage was done. Curettes of different sizes and shapes were used to remove the cystic content and the lining membrane and break the bony septae.

The curette was applied with caution close to the physis. The medulla was opened and the margin of excision was extended using a high-speed burr. Moreover, the high-speed burr was used in a tangential to avoid injury to physis. After that, a gauze soaked with H_2O_2 was applied to the cyst for ten minutes then the cyst was irrigated with jets of normal saline to achieve more adequate curettage. The remaining periosteal flap was collapsed and double breasting sutures were done. The wound was closed in multiple layers over a suction drain. In the patient with ABC affecting the distal phalanx, extended curettage was done and the remaining periosteal flap was sutured in double breasting maneuver over K-wire **(Fig. 5)**.

Fig. (5): Intraoperative photo showing the remaining periosteal flap after exteneded curettage of the distal phalangex of the thumb.

Follow up:

After intralesional curettage of ABCs, in the case of humeral and glenoid lesions, the limb was kept in an arm sling. Early passive range of motion (ROM) of the shoulder, elbow, and wrist was started after the removal of sutures as tolerated. Strengthening exercises and active ROM were delayed till the complete healing of the lesion. A below elbow plaster cast was applied for about 6 weeks in patients with lesions affecting the distal radius. For distal phalangeal lesions, a posterior splint was applied for 4 weeks.

A routine follow-up plan that comprised clinical and radiologic evaluation every 6 to 8 weeks during the first postoperative year and every 3 months during the second year was used to monitor all patients for at least two years. Then annually to assess healing, diagnose local recurrence and identify shortening and deformity occurring as a result of injury of the physis. Patients were evaluated clinically (pain, swelling shortening, and deformity).

Complications and occurrences of local recurrences were recorded. Histological analysis supported the occurrence of the condition. The functional outcome was assessed using the Musculoskeletal Tumor Society (MSTS) scoring system.

Statistical analysis

Data analysis was conducted using SPSS software. Mean and range were used to convey quantitative data.

RESULTS

This study included 15 patients (9 males, 6 females), with a mean age of 13.2 ± 1.6 years (range, 7 years 5 months to 18 years 2 months). ABCs were located in the proximal humerus in 7 cases, distal humerus in 3 cases, radius in 3 cases, glenoid in one case, and distal phalanx of the thumb in one case. Pain and swelling were the main presenting symptoms, with gradual onset of pain.

Plain radiographs were done for all cases revealing eccentric expansile lytic lesions. Computed tomography (CT) scan was done in 5 cases to assess the lesion for subtle cortical destruction or fracture. MRI was done for all cases to identify multiple fluid levels and the extension to the epiphysis. Needle biopsy and histopathological examination were done in all cases to confirm the diagnosis. All patients were managed by extended intralesional curettage and followed up for at least two years. All lesions healed after a mean period of 16 weeks (range 10–21 weeks) **(Fig. 6).** The time to healing was related to the age of patients, size, and behavior of the lesion. Healing of the lesion occurred earlier in young patients, with small lesions and less aggressive lesions.

Fig. (6): Follow up reavled all cysts healed and bone remodelled; plain radiograh (a) distal phalangex (b) distal radius,(c) glenoid and computed tomography of the glenoid (d).

Superficial infection was seen in one patient, which was managed conservatively. Recurrence developed in one patient at 9 months postoperatively and was managed by extended curettage. One patient with a huge lesion in the proximal humerus adjacent to the physis developed a shortening of the humerus of about 1.5 cm due to physeal affection, without angular deformity. There were no cases of deep infection, nerve injury, or deformity.

DISCUSSION

Aneurysmal bone cysts are extremely uncommon, having a frequency of about 1.4% [(7)]. The lengthy bones of the lower extremities are where they often develop. Only 5% of the lesions, or approximately 22.5% of ABCs, are seen in the hands [(3)]. Although ABCs can occur at any age, the majority of occurrences (80%) occur in those under the age of 20 [(8)]. Distention of the bone with blood-filled areas without endothelial lining is a feature of ABCs. Numerous spindle cells, benign giant cells, and fine strands of newly woven bone can all be found in the hollow lining. ABCs are diagnosed with imaging investigations such plain X-rays, CT scans, and MRI, but a histological evaluation of the lesion is required for a conclusive diagnosis. Giant cell tumours, telangiectatic osteosarcoma, fibrous dysplasia, simple bone cyst, osteoblastoma, and plasmacytoma are among other diagnoses [(9)].

A variety of treatment options are discussed, including bone marrow injection, cryoablation, radiotherapy, radionuclide ablation, intralesional curettage with or without adjuvants, en bloc resection, embolization, percutaneous injection of polidocanol or doxycycline inside the lesion, and use of denosumab or bisphosphonates [(6)]. The choice of treatment for patients with ABCs depends on the likelihood of recurrence and potential side effects of the chosen therapy [(5)].

A substantial chance of recurrence exists with ABCs [(10,11)]. Imaging investigations and clinical assessments are used to find ABC recurrence. Recurrence of symptoms (such as pain or swelling) is a clinical indicator of recurrence. Imaging investigations such as X-rays that reveal expanding focal lytic regions inside the lesion and MRIs that reveal numerous fluid levels are also indicative of recurrence [(12,13)]. The most crucial element in determining recurrence is proper surgical technique with appropriate tumour removal. Therefore, by executing a big window to expose the whole cavity, employing varied sizes of curettes, having good visibility of the interior walls, and utilising high-speed burrs, the rate of recurrence can be decreased [(14)].

In our study, there were 15 patients with ABCs in the upper extremity managed by curettage with burring. Healing occurred in all cases with symptom relief. Healing was evaluated radiographically by the decrease in ABC size, increased sclerosis with a decrease in the lytic component, and increased cortical thickening and remodeling of areas of bone. Complications occurred in 3 patients including superficial infection in one patient, shortening in another one, and recurrence in the third patient. Recurrence was attributed to inadequate curettage.

Gibbs *et al.* [(15)] reported cure rates of around 90% of ABCs patients treated by curettage with burring without the use of adjuvants, which contrasts with our data. Cure rates of around 82% were attained in patients with ABCs treated with curettage and burring in the trial by **Dormans *et al.*** [(16)].

In the **Wang *et al.*** [(17)] investigation, one patient out of 31 with ABCs treated with high-speed burr, curettage, and bone graft had recurrence.

However, there was no discernible difference in the 5-year disease-free survival associated with or without the use of a high-speed burr in the comparative analysis carried out by **Lin *et al.*** [(18)]. **Deventer *et al.*** [(13)] examined intralesional curettage (34 patients), percutaneous polidocanol sclerotherapy (32 patients), and en bloc resection as treatment options for ABC (8 patients). In 44.1% of the patients treated with intralesional curettage, they found a local recurrence rate requiring further care. In the group receiving sclerotherapy, 90.6% of patients had chronic disease. Serial sclerotherapy resulted in acceptable healing with a reduction in lesion volume in 71.9% of patients, even if full healing was only achieved in 9.4% of instances.

Curopsy, also known as percutaneous curettage performed during a biopsy, was reported by **Reddy *et al.*** [(19)] as an appropriate approach for successfully

removing the membrane lining various cyst quadrants. They noted a 9.6-week duration to bone repair and an estimated tumour recurrence rate of 18.6%.

In the research by **Crowe *et al.*[(20)],** curettage using a high-speed burr, adjuvants, and bone graft were used to treat 11 patients with ABCs distal to the elbow. Ten patients were said to have integrated the bone transplant and recovered without the need for additional surgery. Revision bone grafting was necessary for one patient.

CONCLUSION

Extended curettage with high-speed burr is considered an effective treatment method for ABCs in the upper extremity.

Supporting and sponsoring financially: Nil.
Competing interests: Nil.

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