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CLINICAL ARTICLE

Treatment of Aneurysmal Bone Cysts by Minimally Invasive Curettage and Allogenic Bone Impaction Grafting: Mid-to Long-term Results

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Objective: Recurrence is the main hinderance in treatment of aneurysmal bone cysts (ABCs). Therefore, several treatment modalities and adjuvant therapies have been proposed. In this study, we aim to evaluate the long-term effectiveness of percutaneous curettage and allogenic bone grafting as a new, minimally invasive modality in treating ABCs.

Methods: We performed a retrospective review of the medical records of patients diagnosed with primary ABCs at a university hospital over a 10-year period (2000–2010). We selected all patients who were diagnosed with primary ABC in the extremities and pelvis, treated with the same surgical procedure, and were followed for at least 2 years postoperatively. All patients underwent the same procedure of percutaneous curettage and impaction of allogenic pulverized fine bone fragments (harvested from locally stored femoral heads) mixed with autologous bone marrow aspirate from the iliac bone. We reported patient's characteristics (age and gender), site and size of the lesion, presenting symptoms, Capanna classification, follow-up duration, and post-operative complications. Assessment of cyst healing was based on the appearance on radiographs according to the modified Neer classification.

Results: Nineteen patients were included in this study; 10 patients were males and nine were females. The mean age was 9.6 years (range 3–15). The location of the lesions was as follows: femur (eight), tibia (four), pelvis (four), proximal humerus (one), distal radius (one), and calcaneus (one). The most common presenting symptom was pain in the involved area. Pathological fracture was the presenting feature in two patients. The mean follow-up duration was 6.4 years (range 2–18). The earliest radiological sign of incorporation of the allograft was seen at 3 months after surgery. All patients showed bone remodeling and radiographic resolution (classified as either A or B on the modified Neer classification) of their cystic lesions within 6 months. No local recurrence, infection, or pathological fractures occurred during the follow-up period.

Conclusion: Percutaneous curettage and impaction of allogenic bone graft mixed with autogenic bone marrow aspirate is an efficient, minimally invasive, reproducible, and affordable procedure for the treatment of primary ABCs.

Key words: Allograft; Aneurysmal bone cyst; Curettage; Grafting; Minimal invasive

Introduction

A neurysmal bone cysts (ABCs) are tumors of undefined neoplastic nature that are mostly presented late in childhood, with 80% before the age of 20 years. The term ABC was proposed by Jaffe and Lichenstein as a distinct clinical entity.¹ They grow aggressively, containing bloodfilled spaces, and often expand the affected bone.² Despite their benign nature, all modalities of treatment carry a risk of local recurrence.^{3,4}

The ABCs are rare lesions with an incidence of about 1.4/1,000,000 individuals/year. They represent 1-6% of all bone tumors.^{5,6} The most common locations of ABCs are in

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the metaphysis of long bones (67%), posterior elements of the spine (15%), and the pelvis (9%).^{7,8} The clinical presentation of ABCs includes pain, swelling, pathological fractures, neurological impairment (spine), and angular deformity/ leglength discrepancy (juxta-physeal).³

The etiology of ABCs remains uncertain, it has been hypothesized that it is a reactive highly vascular lesion of bone resulting from local hemodynamic impairment rather than a true neoplasm.⁹ They can arise primarily or secondary to an underlying lesion such as chondroblastoma, osteoblastoma, chondromyxoid fibroma, non-ossifying fibromas, or giant cell tumors.^{3,9} More recent cytogenetic analysis has revealed a specific genetic translocation of the *USP6* oncogene in ABCs.¹⁰

The typical appearance of ABCs on radiographs is an expansile, radiolucent lesion in the metaphysis with intralesional septations. Soap-bubble appearance, fluid–fluid levels, and contrast enhancement on MRI are highly suggestive but not pathognomonic.^{3,9}

Biopsy is imperative for a definitive diagnosis of ABCs.^{3,9} This will help to exclude malignant lesions such as telangiectatic osteosarcoma. The histopathology usually showed abundant red blood cells filling cyst-like spaces surrounded by septal proliferation of fibroblasts.³

Traditionally the treatment of choice for ABC has been surgical curettage with or without bone graft, with a recurrence rate of 18% to 59%.^{3,11,12} As a result, various adjuvants have evolved to reduce recurrence including the use of high-speed burr, argon beam coagulation, phenol, cryosurgery, cementation, and sclerotherapy. There are no high-level controlled studies to support this, mostly case series from single institutions with rates of recurrence from 4%-40%.³

Alternative modalities include adjuvant radiotherapy (risk of secondary malignancy) and selective arterial embolization, which can be adjuvant or primary, in difficult-toaccess locations (risk of adjacent ischemia). Other emerging techniques include curopsy (limited curettage through a small open biopsy), percutaneous doxycycline, medical bisphosphonate therapy, and RANKL inhibition (Denosumab). These are considered experimental techniques with limited published data.³

Formal surgical excision could carry significant morbidity to the patient and might not be appropriate for lesions in difficult regions such as the pelvis or nearby growth plates (physis). Additionally, the expensive synthetic bone grafts and allografts that are used to fill the defect after excision/ curettage may not be available in all treatment centers. Hence, looking for less invasive, reproducible, and affordable procedure is paramount for the treatment of ABCs.

The purposes of this article are: (i) to describe a simple minimally invasive surgical technique for the treatment of primary ABCs; (ii) to report the clinical and radiological outcomes of 19 patients treated with this surgical technique; and (iii) to provide an affordable alternative option for grafting large bone defects.

Material and Methods

We conducted a retrospective review of patients who were managed surgically for a primary ABC of the extremities and the pelvis. The surgical procedures were performed between the year 2000 and 2010 at a university hospital. We collected patients' data, pathology and operative reports, and all relevant images from the hospital medical records.

Ethical Statement

This study was approved by our institutional review board (No.3207/2020/67). We obtained informed written consent from all included patients/families to have their data and images.

Diagnosis and Selection Criteria

The initial diagnosis was based on the radiographic appearance on plain radiograph, MRI, and CT scan. Frozen section tissue biopsy performed at the beginning of the surgical procedure, and final histopathological examination confirmed the diagnosis.

The inclusion criteria included: (i) diagnosis with primary ABC; (ii) surgical management with percutaneous curettage and impaction of allogenic bone graft (harvested from locally stored femoral heads) mixed with autologous bone marrow aspirate: and (iii) follow up for at least 2 years postoperatively

The exclusion criteria included: (i) patients with insufficient data on their medical records; and (ii) previous treatment for the lesion.

Surgical Procedure

All patients were managed by the same surgeon (senior author) using the same surgical technique of percutaneous curettage and insertion of a paste of allogenic pulverized fine bone fragments mixed with autologous bone marrow aspirate from the iliac bone.

Preparation of the Graft Paste

Allograft was taken from human femoral heads obtained at the time of total or partial hip replacement from consenting donors. Potential donors were screened by interview and questionnaire. Due to the lack and cost of commercially available allogenic bone and bone substitutes in our country, we decided to harvest our own allografts.

Patients who accepted to donate underwent a full medical assessment. Blood tests were carried out for common bloodborne pathogens, including hepatitis B, hepatitis C, and HIV following appropriate counseling. Exclusion criteria for the bone donation were a history of active hepatitis, unexplained jaundice, signs/symptoms consistent with AIDS, a recent history of acute systemic infection, steroid therapy, malignant disease, Alzheimer's disease, and autoimmune/ inflammatory/connective tissue disorders (such as rheumatoid arthritis or systemic lupus erythematosus). Microbiologic samples were taken at the time of allograft harvesting and at the time of graft implantation. Preparation of the graft paste started by removal of residual soft tissues, placement of the graft in a pre-sterilized double plastic container, and then freezing it at -70° C within 2 h of collection for a minimum of 6 months. (Fig. 1).

Before implantation, the femoral heads were placed in hot saline for 1 h to allow for complete thawing before removal of articular cartilage. The heads were then cut into small pieces and ground using a special sterile bone mill with a final product of pulverized fine fragments. This pulverized graft was washed with autoclaved hot saline alternating with a 10% H_2O_2 solution until the color of the fragments become white, which indicated removal of fat and blood sediments.



FIG. 1 showing the steps of allograft preparation. (A) Removal of all cartilaginous tissue by bone nibbler after thawing the femoral heads in hot sterilized saline. (B) Cutting the femoral heads by bone nibbler into small pieces. (C) Using a bone mill to grind the bone pieces. (D) The ground pieces are cleaned by hot normal saline and H_2O_2 until the graft becomes white (devoid of fat and blood elements), then antibiotics and bone marrow aspirate are added to them. (E) The new mixture of the graft was filled with 5–10 cc syringes after cutting the distal end, to be ready for use.

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For each femoral head, we added 1 g of cephalosporin antibiotic powder and 10 cc bone marrow aspirate from the iliac bone. The resultant mixture was a bone paste which we packed by fingers into 5 ml or 10 ml syringes after cutting its distal part to be ready for injection.

Curettage Technique

All procedures were performed under general anesthesia in a supine position with the use of a tourniquet when possible. Under fluoroscopic control, we performed aspiration of the cyst using a Jamshidi needle to verify the nature of the fluid content, followed by a small incision about 1 cm, centered over the cyst.

The tip of the 3 mm curette was advanced into the cyst cavity through the skin incision to open a small bone window of about 1 cm². Under fluoroscopy, percutaneous removal of cyst lining, and curettage was performed using various-sized angled curettes alternating with suctioning the contents and washing the cyst with one-liter normal saline. Initial frozen section sampling followed by routine histopathologic examination for all removed tissue was done.

At the end of curettage, a small raytec gauze inserted over the tip of Kocher forceps was used to clean the wall of the cyst from any remnant lining. The proximal and distal medullary canals were opened with an angled curette or bent Steinman pin for bone cysts in the extremities. The idea behind opening the medullary canal was to allow communication between cyst cavity and the rich bone marrow from the medullary canal which may improve healing potential.

Impaction Technique

Finally, the prepared bone paste syringe was inserted through the bone window into the cyst, deployment and impaction of the graft was done using 1cm pencil punches and gentle tapping by a mallet.

In cases of pelvic lesions, three cases were approached through the ischial tuberosity in a lithotomy position, and the graft was impacted by hand to avoid penetration of the pelvic cavity. None of the patients had any type of internal or external metal fixation around the lesion.

After Surgical Care

Skin wounds were closed routinely, and the extremity was protected in a sling in cases of upper extremity lesions. Hip spica cast was performed for 3 cases of pelvic lesions in younger patients and one pelvic patient was allowed nonweight bearing for 6 weeks. Patients with lower limb lesions were allowed partial weight-bearing on crutches for 8 weeks postoperatively.

Follow-up and Outcomes Evaluation

All patients were followed clinically and radiologically with plain radiographs every month for the first 3 months, then every 6 months until the end of the second year, and thereafter annually. The clinical evaluation included the presence of pain, participation in daily living activities, range of motion of adjacent joints, and the occurrence of a fracture or re-fracture.

Healing of the cysts was assessed on plain radiographs according to the modified Neer classification (Table 1).

Statistical Analysis

We used PASW statistics 18 (IBM, Armonk, NY, USA) for statistical analysis. We tested the variables that might correlate with Neer classification at 6 months postoperatively using the Pearson correlation test. A *P*-value less than 0.05 is considered to be statistically significant.

Results

General Results

The total number of included patients was 19, and 10 were males and nine were females. The age ranges from 3–15 years (mean 9.6 years). The location of the lesions was as follows: femur (eight); tibia (four); pelvis (four); proximal humerus (one); distal radius (one); and calcaneus (one).

TABLE 1 Modified neer classification						
Classification	Description	Details				
А	Healed	Cyst filled with new bone with small radiolucent area (<1 cm)				
В	Healed with a defect	Radiolucent area (< 50% diameter) with enough cortical thickness				
С	Persistent cyst	Radiolucent area (≧50% diameter) with thin cortical rim				
D	Recurrent cyst	Cyst reappearing in the obliterated area or in the increased residual radiolucent area				

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The most common presenting symptom was pain in the involved area. Pathological fracture was the presenting feature in two patients; they were undisplaced fractures in the proximal tibia in both cases and were treated conservatively in a long leg cast after the procedure.

The smallest lesion was located in the proximal tibia with a size of 9 cm² (3 × 3 cm). The largest lesion was located in the ilium and acetabulum with a size of 70 cm² (10 × 7 cm). More than 50% of included cases have cyst size \geq 30 cm².

The details of all patients' age, gender, location of the lesion, presenting feature, size of the cyst, and Capanna type are illustrated in (Table 2). The mean volume of the final graft paste injected was 25 ml (range 15–50 ml). The histopathology results for all lesions confirmed the diagnosis of primary ABC.

Follow-Up

The mean follow-up duration was 6.4 years (ranges from 2– 18 years). Individual follow-up duration for all patients is illustrated in Table 2.

Radiographic Outcomes

On radiographs, there was an initial decrease in density of the bone graft which later progressed to fill the cavity and even obliterate the margins of the original lesion. The earliest radiological sign of incorporation of the allograft was seen at 3 months after surgery. All cysts were classified as either A or B (on the modified Neer classification) within 6 months of the surgical procedure (Table 2).

Using the Pearson correlation test, we tested the correlation between these variables (age, gender, location, cyst size in cm^2) and Neer classification at 6 months postoperatively. The only variable that has a significant correlation with Neer

TABLE 2 Illustrates the demographic, clinical, and radiological data of all patients

	Age/ year	Gender	Location	Main symptoms	Size/cm	Capanna type	Neer classification at 6 months	Follow- up/year
1	12	Male	Proximal humerus	Pain	6 × 4	1	A	4
2	12	Male	Distal radius	Pain / swelling	5.5 imes 5.5	П	В	12
3	6	Female	Ischium and acetabulum	Hip pain/limping	7×4.5	-	В	6
4	12	Male	llium and acetabulum	Hip pain/limping	10×7	-	В	17
5	4	Female	Ischium and acetabulum	Limping	7.5 imes 5	-	В	18
6	3	Female	Ischium and acetabulum	Limping	7.1 imes 3.5	-	В	10
7	8	Male	Femur neck	Hip pain after falling down	5×4	I	А	3
8	12	Female	Femur neck	Hip pain after falling down	6×5.5	I	В	5
9	9	Female	Femur neck	Hip pain after falling down	8 × 6	I	В	4
10	7	Male	Femur neck	Hip pain	6 imes 4.5	I	А	18
11	11	Male	Trochanteric region	Hip pain after falling down	5.5 imes 5.5	I	А	3
12	3	Male	Trochanteric region	Pain	6×5	I	В	3
13	7	Male	Femur diaphysis	Pain	7×4	I	А	4
14	15	Male	Distal femur	Pain	6×7	V	В	3
15	14	Female	Proximal tibia	Pathological fracture	7×7	П	В	2
16	10	Female	Proximal tibia	Pathological fracture	6.5 imes 6.5	П	В	3
17	15	Female	Proximal tibia	Pain	3×3	Ш	А	2
18	9	Male	Distal tibia	Pain/swelling	4×4	Ш	А	2
19	14	Female	Calcaneum	Pain/swelling	6×4	П	А	3

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classification was the size of the cyst in cm² (P = 0.003). In other words, increased preoperative size of the ABC will delay healing by advancing the Neer classification toward stage B (Pearson correlation 0.65). Figures 2 and 3 showing the results of treatment of an ischial and femoral neck ABCs respectively.

Clinical Outcomes

All patients were fully active and pain-free after 6 months of the surgery. The two pathological fractures healed. Physical examination revealed painless, full range of motion of all joints around the involved area at the last follow-up visit for every patient. No complications such as pathological fractures or infections were encountered.

Discussion

Goals of Treatment

The goals of treatment of ABCs are to relieve pain, halt progression, prevent pathologic fracture, and avoid local recurrence. Variant adjuvant treatment modalities have been described in addition to intralesional curettage to minimize the recurrence of ABCs. These include, amongst others, liquid nitrogen, phenol, bone cement (polymethyl methacrylate), radiation, and the use of a burr.^{13–15} Embolization is used in certain inaccessible or hypervascular lesions.^{16–17} Autografts and allografts were used to promote healing and ossification in ABCs.^{18–22}

These goals were achieved successfully in all patients by one graft technique in a single-stage surgery, without the

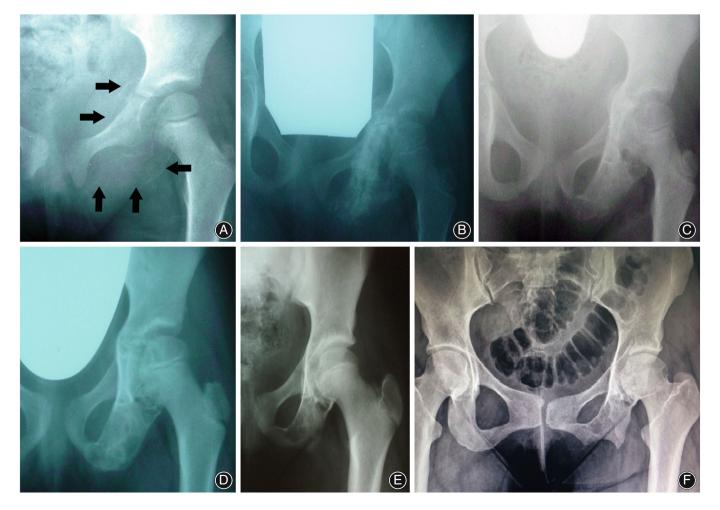


FIG. 2 showing the preoperative and postoperative radiographs for Case-5 in Table 2. The preoperative plain radiograph showed an expansile osteolytic lesion involving the left ischium, left inferior pubic ramus, and extending to the acetabulum (A: black arrows). Follow-up radiographs at 2-months (B), 7-months (C), and 2-years (D) showing sequential filling of the defect with new bone. Remodeling of the acetabulum, inferior pubic ramus, and ischium is clear at 8-year follow-up (E). A small residual cystic lesion is present in the ischium as evident on radiograph taken 18-years postoperatively (F).

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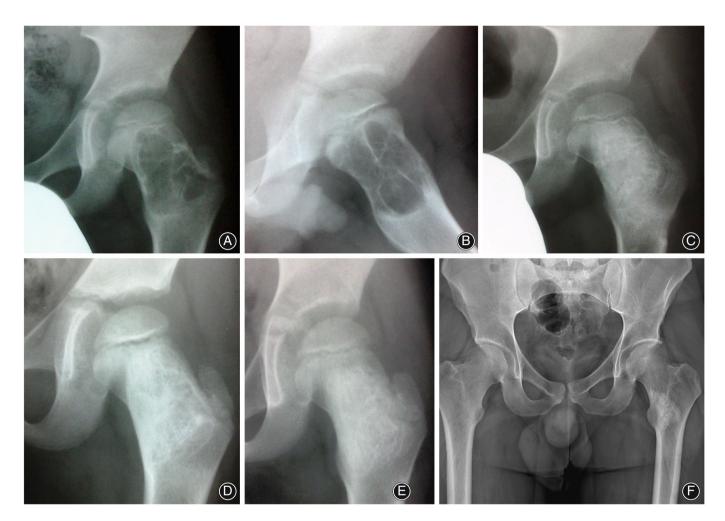


FIG. 3 showing the preoperative and postoperative radiographs for Case-10 in Table 2. Preoperative plain radiographs showed a multilocular cystic lesion involving almost all the femoral neck of the left hip as appeared on the AP (A) and lateral (B) views. Follow-up radiographs at 2-months (C), 6-months (D), and 12-months (E) showing sequential filling of the defect with new bone and complete remodeling at the 18-year follow-up radiograph (F).

TABLE 3 Capanna classification				
Туре	Morphological characteristics			
 V V	Central Central affecting the entire bone diameter Eccentric Subperiosteal Subperiosteal extending to soft parts			

use of any adjuvant therapy. Our results showed no late recurrence of any cysts.

Advantages of our Technique

A thorough curettage of the bone cavity with complete disruption and removal of the entire membrane lining is performed until a healthy, raw bleeding bone surface is left behind. The percutaneous technique of bone paste injection is a minimally invasive one and preserves the integrity of the surrounding tissues. The allograft paste is expected to provide the osteoconductive scaffold on which the red marrow will stimulate osteogenesis, new bone formation, cyst healing and reduce the incidence of recurrence.

Resection through the margins of lesions in the proximal femur, pelvis, or children with open growth plates is often too extensive/aggressive for the treatment of benign lesions. Besides, pelvic cysts are difficult to access with concerns of their proximity to neurovascular structures and vulnerability of the integrity of the acetabulum.

The surgical curettage of pelvic lesions carries about a 14% recurrence rate, but still, it is not considered an unacceptable rate.²³ Four of our cases were poorly accessible pelvic lesions and were treated successfully without the need for embolization or any adjuvant therapy.

Generally, a high radiographic Capanna stage (Table 3),¹² occurrence in a long bone, the presence of a

pathological fracture, and violation of an open growth plate showed some trend toward an increased risk of local recurrence, which was not the case in our patients, likely because our series is small.

To the best of our knowledge, no previous studies described the concept of usage of minimal invasive curettage and the impaction of a hand-made mixture of allograft with bone marrow aspirate in treating ABCs. One article shared part of our concept published in 2005.²⁴

Our technique is a minimally invasive method in which the curettage was performed through a small incision (about 1 cm) and the allograft paste was injected through the syringe even for difficult areas of the pelvis. It helps to decrease morbidity and enhance the rehabilitation process. Besides, it is affordable, the allografts were taken from nonprofit donors and patients do have to pay for that. The tools used to store and prepare the allograft can be easily provided.

Clinical Implications and Future Perspectives

The absence of recurrent lesions and other complications reported over a long-term follow-up in ABCs of different sizes and locations in both genders as shown in our case series adds strength to this study and emphasizes the effectiveness of our surgical technique.

In addition to being easy, safe, minimally invasive, and reproducible; we believe that our technique can be used effectively in countries with limited resources in which the use of high-cost synthetic bone grafts or commercial allografts may not be affordable. Furthermore, the concept of allogenic bone graft impaction might be used in treatment of other benign bone cystic lesions.

Further large comparative studies should be implemented to get more evidence about the effectiveness of our technique against other treatment modalities for ABCs and other cystic bone lesions.

Conclusion

Minimally invasive curettage and impaction of morcellated femoral head allografts mixed with autogenous bone marrow aspirate is a simple, efficient, and affordable technique for the treatment of primary ABCs. It can induce bone healing and consolidation without the need for adjuvant therapy or internal fixation.

This method helps to avoid extensive surgery and blood loss. It is also convenient for the treatment of poorly accessible lesions such as in the pelvis without the need for embolization.

Author Contributions

A ll authors contributed to the study conception and design. Material preparation, data collection and analysis were performed by Mohamad Yasin, Omar Samarah, and Yazan Hammad. The first draft of the manuscript was written by Mohammed Alisi and Freih Abu Hassan and all authors commented on previous versions of the manuscript. All authors read and approved the final manuscript.

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