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ORIGINAL ARTICLE

TWO YEAR FOLLOW UP OF GIANT ANEURYSMAL BONE CYSTS MANAGED WITH AN AUTOLOGOUS NON VASCULARIZED FIBULAR STRUT GRAFT

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ABSTRACT:
Introduction: The aim of this study is to highlight the results of non-vascularized fibular graft for reconstruction of bone defects after extended curretage of giant aneurysmal bone cyst (ABC) of the extremities. **Materials and Methods:** Between 2013 to2016, twelve patients, with giant aneurysmal bone cysts were treated. The cysts were located in the proximal humerus, proximal radius and proximal tibia. All patients were given extended curettage of the cyst followed by non vascularized fibular bone graft, with the graft length ranging from 5 to 9 cm and ipsilateral cancellous iliac crest graft. All patients required supplementary fixation with a TENS or Kirschner wire or plating. **Results:** At the final follow-up, bony union was achieved in each case, and there was norecurrence, limitation of range of motion or disability. Non-vascularized fibular graft is an optimal and valuable method for the reconstruction of bone defects after resection of giant ABC in the extremities. **Conclusion:** Extended curettage of the lesion along with fibular strut grafting supplemented with cancellous bone chips with or without primary internal fixation seems to be a reasonable option for giant aneurysmal bone cysts with good functional outcome and excellent patient satisfaction at long term follow up with minimal donor site morbidity.
Keyword: Graft; Humerus; Aneurysmal bone cysts

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INTRODUCTION

Despite a descriptive history of more than 60 years, the nature, character, and optimal treatment of aneurysmal bone cysts remain obscure. The lesion was first described by Jaffe and Lichtenstein [1] in 1942, was subsequently further defined by both of these authors [2, 3] and became known as Jaffe-Lichtenstein disease. It is a rare tumor and accounts for 1% to 2% of all primary bone tumors. [4] Aneurysmal bone cysts usually occur in the first two decades of life and exhibit a slight female preponderance.[5]Aneurysmal bone cystshows an evident predilection for long bones and the vertebral column, particularly the femur, humerus, tibia, and fibula.[6] It was suggested that ABC corresponds to a hemodynamic disturbance because of primaryor secondary venous malformation of bones.[7] Several treatment modalities have been described for ABCsuch as sole curettage, curettage with cementation or bone grafting, fibrosing agents (phenol) or bone marrow injections, arterial embolization, adjuvant cryotherapy or radiotherapy, demineralized bone matrix applications and segmental or en bloc resections.[8] If the cyst's transverse diameter on radiographic examination is equal to or more than three times the diameter of the adjacent normal bone, it is labeled giant ABC.[9] Giant ABC is an uncommon pathological lesion and can be challenging because of the destructive effect of the cyst on the bones and the pressure on the nearby structures. Curettage of the diseased segment is a useful surgical technique in the treatment of benign bone lesions. The fibula is a strong, long, tubular bone that can be used as a non-vascularized bone graft for restoration of mechanical continuity and cancellous bone graft from iliac crest to increases the healing potential of the limb after bone defects resulting from tumor curettage. The purpose of this article is to evaluate the clinical and radiological

results of the curettage/resection and reconstruction of giant ABCs using non-vascularized fibular along with cancellous bone graft. [9,10,11]

MATERIALS AND METHODS

After proper pre anaesthetic check up between Janauary 2013 to July 2016, 12 patients with diagnosis of giant ABC of the extremities were treated with extended curettage of the cyst followed by nonvascularized fibular bone grafts trut or split augmented with cancellous bone chips with or without internal fixation . All patients had painful swelling ranging from 6–12 months' duration. Lesions were biopsied before or after the final surgery.

Operative technique

Surgery was performed in three steps:

Step 1: Harvesting of the fibula from the ipsilateral leg. Care was taken not to injure the superficial peroneal nerve or leaving short distal segment less than 6 cm to prevent an adverse effect on ankle joint stability. The length of the required fibula for bridging the bone defect was estimated from the Magnetic resonance scans. The leg wound was sutured and covered by sterile dressing before opening the site of the lesion to avoid donor site contamination by tissues of the tumor.

Step 2: Thorough curettage was performed in and around the pathological lesion.

Step 3: The third and most vital step was impaction of the harvested fibula into the cyst. In some cases the fibula got impacted snuggly and no internal fixation was required while in some cases the incorporated fibula was fixed with either Kirschner wire or titanium elastic nail or plate.

The patients were followed up by plain radiography regularly for 2 years. Solid union was seen in all the cases in 6 to 9 months time with no case of non union. Full range of motion of adjacent joints were seen except in 3 cases which had slight limitation. No recurrence was seen in 2 years follow up period. All the patients were followed up for an average period of 2 years (1.5 – 2.9 years). At each follow- up visit, patients were thoroughly examined for donor and recipient site complications. X-rays were taken to assess union at each graft host junction. Functional assessment was done using MSTS Score. [10] Weight bearing was allowed only after radiological union.

There were nine females and three males. The cysts were present in the proximal humerus in three cases, the proximal radius in six cases, proximal tibia in three cases. Their mean age at the time of presentation was 24.3 years (13 to 38) and the mean follow-up was 2 years. In majority of the cases that is in 9 patients, the chief complaint was painful swelling with pain being dull in nature, being exaggerated by movements and relieved on taking medications while 3 patients presented with a pathological fracture. Functional outcome was assessed by Musculoskeletal Tumour Society (MSTS) Score in which results were expressed as proportion of the expected normal function. Average MSTS score was 78.5% in this study with 7 patients having excellent score and 5 patients having good score. Primary internal fixation was performed in 7 cases while 5 cases did not require any internal fixation.

RESULTS:

Table 1: Demographics and clinical data of the patient

Total cases	**12**
Mean age	24.3 yrs (13 to 38)
Sex (Male :Female)	3:9
Site	Proximal Humerus : 3, Proximal radius :6 Proximal tibia: 3
Mean size of lesion(in cms)	7.4cm x 5.6 cm
Fibular Graft length (in cms)	7.2 cms (5.5-9.1 cm)
Radiological type **(central or subperiosteal)**	Central :11 Subperiosteal : 8
Ennekings Scale(I,II,III)	I: 3, II: 4, III: 5
Clinical presentation	Pain and Swelling: 9, Pathological fracture: 3,
complications	limitation in range of motion 3 cases
Follow up (in yrs)	2 yrs (1.5 to 2.9 yrs)

Surgical procedure performed	**Number**
Curettage+Fibular graft +cancellous bone	5
Curettage+ fib. Graft+canc. Bone +TENS/K wire	7

FUNCTIONAL OUTCOME:

MSTS SCORE	**Number**
Good	5
Excellent	7

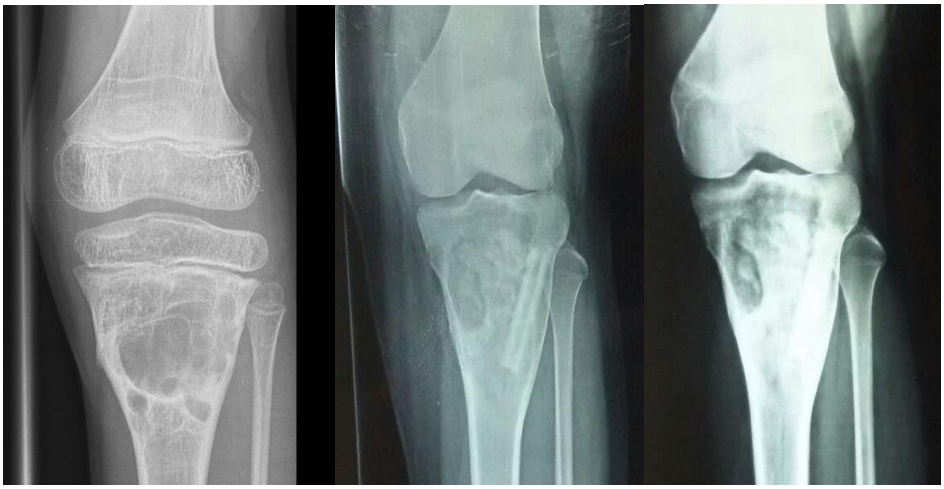

Figure 1: Giant ABC involving the proximal tibial metaphysis, treated with extended curettage with fibular strut graft with cancellous bone chips and final follow up showing well healed lesion with full incorporation of the graft

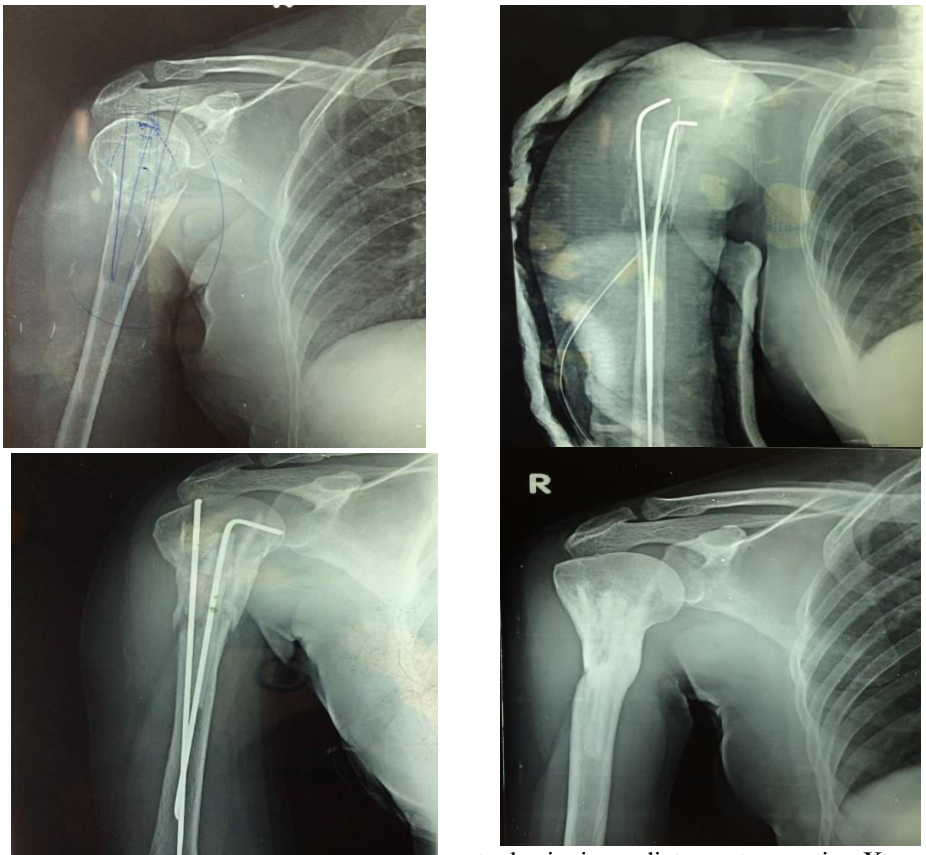

Figure 2: Giant ABC involving the proximal humeral metaphysis, immediate post operative X ray of the same case treated with extended curettage with fibular split graft and stabilized with kirshner wires, three month follow up showing the lesion being healed and hardware in proper place and 2 year follow up showing solid union with full incorporation of the graft with removal of hardware

Figure 3: Giant ABC involving proximal radius with sparing of radial head(pre op X ray and MRI), 1month post op X ray and 6 months follow up xray showing solid union of the incorporated fibula with native bone.

DISCUSSION

It is well known that ABC is classified as an aggressive benign bone tumor, which means if not treated properly, it may recur, or if left untreated, it may enlarge and become giant ABC. Although the pathogenesis of ABC is still unknown, the two broad types are either primary (70%) or secondary (30%).Primary ABCs arise de novo. An ABC is termed as secondary when it develops in association with some other neoplasm. Radiographically, the diagnosis of an ABC shows five classic findings[9]. First, the neoplasm typically has a soap-bubble appearance. Secondly, it may present as an eccentric lesion outlined by a thin layer of subperiosteal new bone. Third common feature is that it can also present as a centric lesion. Fourth, it reveals a metaphyseal lesion that occupies a large percentage of the bone with trabeculations at the edges. Fifth, it manifests soft tissue expansion and destruction of the cortex. None of these criteria could fit any of our patients; consequently, we suggest that, if the cyst's transverse diameter on radiographic examination is equal to or more than three times the diameter of the adjacent normal bone, we can call it giant ABC; in these cases, it can be treated by extended curettage and fibular strut or split graft. Non-vascularized autogenous bone grafts are biologically active grafts with relatively low donor site complications that will be replaced completely by living bone and that are capable of remodeling to fulfill the function need[12–15]. Its cortical composition provides immediate stability to the surrounding joints[16]. The long term possible complications of this procedure include fracture of the graft, non-union, failure of incorporation of the graft, and stiffness or degenerative arthrosis in the joints.[17, 18]Dell et al[19] and Brown[20] found no substantial difference between non-vascularised and vascularized grafts in the time to consolidation or in the incidence of union. Vascularised grafts were transiently stronger than conventional grafts in the first six months, but there was no difference thereafter.

Enneking et al[21] found primary union in 63% of the long bone reconstructions within the first 12 months, Yadav et al[22] found union after 8 to 10 months in 60% and AH Krieg et al[14] got primary union in 89% in mean period of 24 weeks. In our case the success of integration of the strut fibular graft can be attributed to the rich blood supply of the recipient site& the surrounding well-vascularized soft tissues of the ABC. We achieved primary union in 95.77% of the reconstructions in a mean period of 29 weeks (24- 36 weeks).

Enneking et al [21] did not found any correlation between the length of the non vascularised fibular graft and the rate of healing. Similar results were observed in our study as well. The complication rate at the donor site for vascularised grafts has been reported to vary between 7% and 35%.[23] It appears to be higher than for non-vascularised grafts whosecomplication rate has been reported to vary between 4% and 12%.[21,22,24]Functional outcome in the form of MSTS score in our study is 78.5% which is comparable to K.Shah etal [25]

CONCLUSION:

Extended curettage of the lesion along with fibular strut grafting supplemented with cancellous bone chips with or without primary internal fixation seems to be a reasonable option for giant aneurysmal bone cysts with good functional outcome and excellent patient satisfaction at long term follow up with minimal donor site morbidity. The rich blood supply of the recipient site and highly vascularized soft tissue envelope surrounding the lesion contribute primarily to the uneventful healing and the successful incorporation of the graft.

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