Case Report

Journal of Orthopaedic Case Reports 2023 December:13(12):Page 141-147

Cross-beam Non-vascular Fibular Graft Reconstruction in the Management of Long Bone Giant Aneurysmal Bone Cyst – A Case Series

Saravanan Annamalai[1], Velmurugan Kandasamy[2], Srinivasacholan Chandrasekaran[3], Sathish Muthu[4,5,6]

Learning Point of the Article:

The non-vascularized fibular graft can be used as an effective reconstruction tool in the management of bone voids arising in the management of giant aneurysmal bone cysts in long bones.

Abstract

Introduction: Aneurysmal bone cyst (ABC) is a benign expansile osteolytic lesion, characterized by a blood-filled cavity in the bone. Giant ABC (GABC) is an uncommon condition due to the delayed presentation of an ABC that is difficult to handle when it occurs in long weight-bearing bones due to its aggressive nature. The common treatment relies on total resection of tumor with reconstruction of the resultant defect.

Case Report: We present the results of 5 cases of GABCs of long bones managed with non-vascularized fibular graft in a cross-beam fashion along with internal fixation. All patients achieved complete consolidation of the lesion by 12 months along with full incorporation of the graft with good-excellent musculoskeletal tumor society scores. None of the patients had recurrence/pathological fracture till 2 years of follow-up.

Conclusion: We suggest the method of using a non-vascularized fibula graft in a cross-beam fashion to reconstruct the void from the resection of long-bone GABC as a safe, reliable technique with excellent functional and radiological results.

Keywords: Giant Aneurysmal Bone Cyst; Fibular Graft; Resection; Recurrence; Reconstruction.

Introduction

Aneurysmal bone cysts (ABCs) were first reported by Jaffe and Lichenstein in 1942 [1]. ABCs are uncommon bone lesions comprising 1–2% of primary bone tumors. The tumor predominantly occurs in the first two decades with a mean age of presentation around 18 years [2]. The ABC is a lesion of the pseudotumor type, and it can be defined as a cavity filled with blood that has septations made of connective tissue. The mechanism of formation of these tumors is considered to be sudden venous or arteriovenous malformation [3,4]. Pathologically, they contain fusiform cells, multinucleated giant cells, hemosiderin storage areas, and some trabecular bone [5]. The giant ABCs (GABCs) are late presenting ABCs that are very destructive and expansile forms of ABCs which will produce more chance of pathological fracture and disability [6]. They tend to be located eccentrically in the metaphysis of long bones adjacent to the physis region. Although ABCs are benign lesions, there are cases reported of their malignant transformation [7]. They are considered locally aggressive and possess a higher rate of recurrence [1, 8]. The predominant distribution remains to lower limbs (50–60%) of which the tibia (40%), especially the proximal tibia (24%), followed by fibula (20%) [5,9,10]. The clinical feature usually presents with pain, mass, swelling, pathologic fracture, or a combination of these symptoms in the

Author's Photo Gallery


Dr. Saravanan Annamalai


Dr. Velmurugan Kandasamy


Dr. Srinivasacholan Chandrasekaran


Dr. Sathish Muthu

Access this article online

Website:
www.jocr.co.in

DOI:
https://doi.org/10.13107/jocr.2023.v13.i12.4112

[1]Department of Orthopaedics, Thiruvallur Medical College, Thiruvallur, Tamil Nadu, India,
[2]Department of Orthopaedics, Government Kilpauk Medical College, Chennai, Tamil Nadu, India,
[3]Department of Orthopaedics, Government Royapettah Hospital, Chennai, Tamil Nadu, India,
[4]Orthopaedic Research Group, Coimbatore, Tamil Nadu, India,
[5]Department of Biotechnology, Faculty of Engineering, Karpagam Academy of Higher Education, Coimbatore, Tamil Nadu, India,
[6]Department of Orthopaedics, Government Medical College, Karur, Tamil Nadu, India.

Address of Correspondence:
Dr. Sathish Muthu,
Orthopaedic Research Group, Coimbatore, Tamil Nadu, India.
E-mail: drsathishmuthu@gmail.com

Submitted: 17/09/2023; Review: 03/10/2023; Accepted: November 2023; Published: December 2023

DOI: https://doi.org/10.13107/jocr.2023.v13.i12.4112

This is an open access journal, and articles are distributed under the terms of the Creative Commons Attribution-NonCommercial-ShareAlike 4.0 License https://creativecommons.org/licenses/by-nc-sa/4.0/, which allows others to remix, tweak, and build upon the work non-commercially, as long as appropriate credit is given and the new creations are licensed under the identical terms

© 2023 Journal of Orthopaedic Case Reports| Published by Indian Orthopaedic Research Group

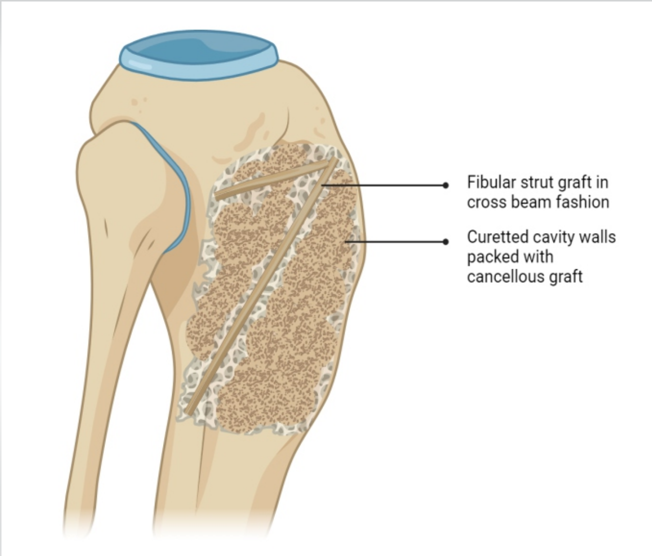

Figure 1: Schematic representation of the cross-beam configuration of non-vascularized fibular strut graft to prevent the collapse of the curetted cavity.

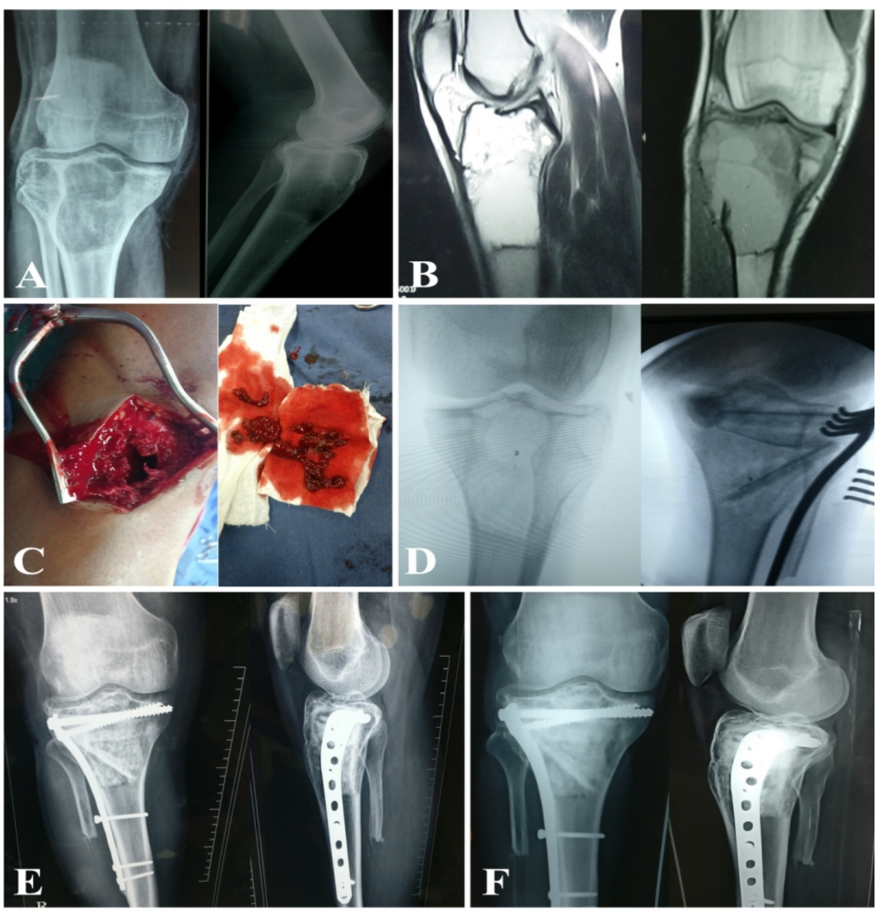

Figure 2: Illustrative case of proximal tibia giant aneurysmal bone cyst in a 20-year-old male (a) pre-operative radiographs; (b) magnetic resonance imaging; (c) intraoperative images showing the blood-filled cavity; (d) intraoperative curettage and placement of the non-vascularized fibular graft in cross-beam fashion; (e) 3-month follow-up radiograph; (f) 24-month follow-up radiograph demonstrating successful consolidation of the graft into the debrided cavity.

affected area. The symptoms are usually present for several weeks to months before the diagnosis is made, and the patient may also have a history of a rapidly enlarging mass. Neurologic symptoms associated with GABCs may develop secondary to pressure or tenting of the nerve over the lesion, typically in the spine [3, 7, 10]. Pathologic fracture occurs in about 8% of ABCs, but the incidence may be as high as 21% in ABCs that have spinal involvement [5, 11].

The ABCs are commonly managed with extended curettage, intralesional sclerotherapy injection, intralesional-doxycycline injection, systemic bisphosphonates, denosumab, or selective arterial embolization in areas where surgical exposure is difficult [12-14]. In contrast, GABCs of long bone need total resection of tumor with the reconstruction of the defect using bone graft with or without internal fixation depending on the size and site of the lesion. Among the various cancellous and cortical bone grafts, the non-vascularized fibula, a strong, long, tubular bone, can be used as a strut graft in place of a large defective cavity not only to fill the void but also for additional mechanical stability so that patient can be mobilized as early as visible consolidation of the graft with the native bone [15-17].

Previous reports on the use of non-vascularized fibula for the reconstruction of ABCs have been shown to give radiologically and functionally appealing results [6, 18, 19]. However, literature on the use of non-vascularized fibula for reconstruction in the management of GABCs remains limited. Hence, the main aim of this article is to present the role of non-vascularized fibula in the management of GABC in long bones of limbs in a particular cross-beam fashion taking advantage of its mechanical strength.

Case Report

We report the results of 5 patients (M:F=3:2) who were diagnosed with GABCs in the long bones and managed with extended curettage and reconstruction with non-vascularized fibular graft supplemented with buttress plating. The age group of patients ranged between 17 and 44 years. The size of the lesion ranged from 18 cm2 to 72 cm2. The lesions were more common in the long bones of the lower limb as discussed earlier

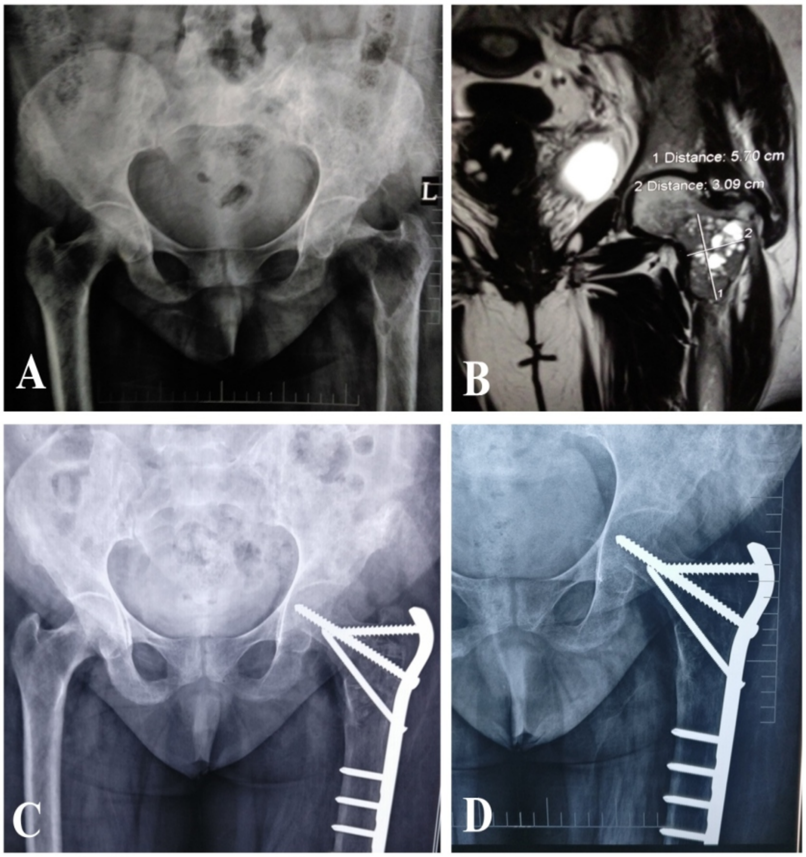

Figure 3: Illustrative case of proximal femur giant aneurysmal bone cyst in a 44-year-old female (a) pre-operative radiographs; (b) magnetic resonance imaging; (c) immediate post-operative radiograph; (d) 24-month follow-up radiograph demonstrating successful consolidation of the graft into the debrided cavity.

with preponderance for the proximal tibia (n = 2) and proximal femur (n = 2) while one case was noted in the proximal humerus. All the lesions were insidious in onset and slow growing with duration ranging from 4 months to 1 year before presentation. All the lesions were biopsied and found to be of Grade II (n = 3) and Grade III (n = 2) as per Enneking's grading of the lesion. All the patients were pre-operatively evaluated for the functional status with musculoskeletal tumor society (MSTS) score. The detailed clinical characteristics of the cases are presented in Table 1.

Surgical technique

The lesion is approached following the general principles of tumor dissection and the entire lesion is debrided with extended curettage using phenol and electrical cauterization of the bleeding walls. The cavity is further washed with hydrogen peroxide for chemical cauterization. Usually, the fibula is harvested from the ipsilateral side. An 8–10 cm section of the middle third of the fibula is harvested subperiosteally following the posterolateral approach, leaving 5 cm of the proximal fibula and 8 cm of the distal segment to avoid joint instability. The debrided cavity is supported with the fibular strut in a cross-beam fashion as illustrated in Fig. 1 to prevent the collapse of the cavity walls and aid in maintaining

Case No	Age/Sex	Site	Size (cm)	Duration at presentation	Pre-operative grade	Surgical procedure	Follow-up (months)
1	20/Male	Proximal Tibia	9×6	12 months	Grade III	Extended curettage, fibula grafting, and fixation with proximal tibia-locking compression plate	30
2	44/Female	Proximal	6×3	6 months	Grade II	Extended curettage, fibula grafting, and fixation with proximal femur locking compression plate	24
3	17/Male	Proximal Tibia	6×6	4 months	Grade II	Extended curettage, fibula grafting, and fixation with proximal tibia buttress plate	28
4	18/Female	Proximal Humerus	9×8	7 months	Grade III	Extended curettage, fibula grafting, and fixation with PHILOS plate	24
5	23/Male	Proximal Femur	7×5	6 months	Grade II	Extended curettage, fibula grafting, and fixation with proximal femur locking compression plate	32

PHILOS: Proximal humerus internal locking system

Table 1: Characteristics of the patients and their lesions included in the series with their surgical management and follow-up duration

Case No	Age/Sex	Site	Size (cm)	Pre-operative score	3 months	6 months	9 months	12 months	24 months
1	20/Male	Proximal Tibia	9×6	26	16	24	24	26	26
2	44/Female	Proximal Femur	8×7	18	14	16	18	18	20
3	17/Male	Proximal Tibia	6×6	18	12	16	18	18	20
4	18/Female	Proximal Humerus	9×8	24	14	18	24	24	25
5	23/Male	Proximal Femur	7×5	25	16	20	25	25	27
Mean scores				2.2	14.4 (P<0.001)*	18.8 (P<0.001)*	22.2 (P<0.001)*	22.2 (P<0.001)*	23.6 (P<0.001)*
*Paired student t -test compared to baseline mean, MSTS: Musculoskeletal tumor society									

Table 2: Significant improvements in the MSTS score at serial follow-up compared to baseline.

the space for faster consolidation and weight bearing. The remainder of the cavity is filled with cancellous grafts harvested from the contralateral iliac crest regularly. No cement or artificial bone substitutes were utilized to fill the cavity. Further, the construct is augmented with a buttress plate and screws to prevent further collapse of the subchondral bone till the fibular strut graft incorporates with the native bone.

The curretted samples were sent to histopathology for secondary staging and confirmation of the diagnosis. All the lesions were confirmed to be ABCs with cyst walls made up of fibroblasts, woven bone, and osteoclastic giant cells. All patients were immobilized with appropriate methods in the immediate post-operative period. Isometric exercises were initiated in 1st week, mobilization exercises in 2nd week, and strengthening exercises in the 3rd week. From 3rd-week onward, non-weight-bearing walking was started with walker support. Patients were maintained on non-weight bearing until consolidation of the lesion became radiologically evident which occurred by a mean duration of 17 weeks (range 14–19 weeks) following which partial and full weight bearing was gradually resumed as tolerated. Patients were serially followed up with functional scoring with MSTS score and radiological evaluation every 3 months for 1 year and every 6 months thereafter. The scores improved significantly from the pre-operative mean score of 2.2 (poor grade) to 23.6 (excellent grade) at 2-year follow-up as shown in Table 2. It is also noted from Table 2 that the post-operative scores do not depend on the age, sex, location, or size of the lesion among the included patients. None of the included patients reported any complications, donor site morbidity, or recurrence at 2-year follow-up. The additional appropriate internal fixation gave good mechanical stability resulting in complete consolidation of the lesion within 12 months. Fibular grafts were fully incorporated by mean duration of 13.8 (range 12–15) months. All patients went back to their daily activities by mean duration of 10.2 (range 9–12) months. Fig. 2 and 3 show illustrative case examples in the proximal tibia and proximal femur, respectively.

Discussion

ABCs are benign lesions with a <1% chance of malignant transformation [5]. While the majority of these tumors are primary bone lesions, 30% are secondary to certain other bone lesions such as giant cell tumors, chondroblastoma, eosinophilic granuloma, browns tumors, chondromyxoid fibroma, non-ossifying fibroma, and telangiectatic osteosarcoma [20, 21]. Overall, GABCs are the late presenting ABCs making them presented in large sizes with aggressive local expansions mimicking malignant lesions [6]. Radiologically, they are found to be sharp expansile lesions with thin sclerotic margins with evident fluid levels [22].

The management of ABCs depends on the time of diagnosis. In 25% of cases, lack of early radiological or medical diagnosis gives management difficulty in these types of tumors [12, 13]. This delayed presentation often makes the tumor locally aggressive massive lesions presenting with mechanical symptoms to be labeled as GABCs [6]. In small ABCs, the routine management ranges from minimally invasive curettage and bone grafting which may be allogenic or autologous depending on the size of the lesion. These tumors have a high rate of local recurrence with routine curettage ranging from 18% to 59% [2, 23, 24]. To prevent the high recurrence, various adjuvants such as a high-speed burr, argon beam coagulation, phenol, liquid nitrogen, cementation, and sclerotherapy have been used to be called extended curettage [25-27]. Alternatively, arterial embolization and radiotherapy have also been used [28]. However, radiotherapy is associated with a risk of malignant transformation [14]. They are usually reserved for lesions in difficult-to-reach areas for surgical management.

GABCs that present with large expansible lesions around the metaphysis of long weight-bearing bones possess a high risk of pathological fracture and present with severe pain due to local soft-tissue compression. Often, these cases present with pathological fractures where management not only depends on the excision of the lesion but also needs mechanical strength to provide hassle-free mobility in the future [10]. However, none of the included cases presented with pathological fractures while pain and swelling were their presenting complaints. In these conditions, compared to vascularized fibular graft that demands surgical expertise with significant donor site morbidity, non-vascularized fibular grafts are a good option for reconstruction of the void arising from extended curettage of the lesion [19, 25]. Furthermore, vascularized fibular graft needs an extensive approach with increased operating time which may increase the risk of other complications [29]. The geometric orientation of the non-vascularized fibula is of great importance to maintain the anatomic structural integrity of the void developed from the curettage. The authors consider the cross-beam concept of graft placement bridging the diagonal ends of the cavity with the cortical strut of the fibula to prevent further collapse and early incorporation with the native bone. The rest of the dead space can be filled with either iliac crest autograft or cancellous femoral head allograft depending on the availability. Apart from the fibula, other void management strategies such as the sandwich technique of using cement and autologous bone grafts, complete cement filing, and the usage of artificial bone substitutes and tissue engineering techniques are also in vogue [30-32]. They have other limitations such as cost, delayed bone ingrowth, and reduced osteogenic potential compared to the current technique described.

The basic rationale in the management of GABCs involves the preservation of the length of the long bone. Hence, we used fibular graft along with cancellous iliac crest autograft to improve better uptake of the non-vascularized fibular graft. The cortical nature of fibular graft in cross-beam architecture provides immediate stability and maintains the length necessary. The implanted graft will aid in the osteogenic induction with the help of the overlying periosteum and surrounding soft tissues where in our cases two involve the proximal tibia, two proximal femurs, and one proximal humerus. All these regions have a good local blood supply and along with autologous iliac crest grafts add to the osteogenic potential to form a favorable environment for new bone formation and incorporation of the fibular graft and reconstitution of the bone void.

Although the malignant transformation potential of the ABCs remains limited, the particular malignant transformation potential in GABCs remains unknown [5]. We did not note any recurrence or malignant transformation on histopathological examination in our series. However, long-term follow-up of these lesions is needed to account for their recurrence rate and risk of malignant transformation.

Conclusion

GABCs should be diagnosed and managed with extra care. Non-vascularized fibular graft with iliac crest autograft remains a safe and satisfactory method in GABC management with excellent functional and radiological results. Long-term follow-up studies are required to provide information regarding the recurrence and risk of malignant transformation in these GABCs.

Clinical Message

- GABCs are late presenting ABCs with pressure symptoms as their presenting complaint.
- The cross-beam architecture of a non-vascularized fibula graft aids in maintaining the length of the void developed from extended curettage.
- Non-vascularized fibular graft with iliac crest autograft remains a safe and satisfactory method in GABC management with excellent functional and radiological results.
- Long-term follow-up studies are required for early detection of recurrence due to their aggressive nature.

Declaration of patient consent: The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given the consent for his/ her images and other clinical information to be reported in the journal. The patient understands that his/ her names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Conflict of interest: Nil **Source of support:** None

References

1. Jaffe HL, Lichtenstein L. Solitary unicameral bone cyst: With emphasis on the roentgen picture, the pathologic appearance and the pathogenesis. Arch Surg 1942;44:1004-25.

2. Mankin HJ, Hornicek FJ, Ortiz-Cruz E, Villafuerte J, Gebhardt MC. Aneurysmal bone cyst: A review of 150 patients. J Clin Oncol 2005;23:6756-62.

3. Mendenhall WM, Zlotecki RA, Gibbs CP, Reith JD, Scarborough MT, Mendenhall NP. Aneurysmal bone cyst. Am J Clin Oncol 2006;29:311-5.

4. Leithner A, Windhager R, Lang S, Haas OA, Kainberger F, Kotz R. Aneurysmal bone cyst. A population based epidemiologic study and literature review. Clin Orthop Relat Res 1999;363:176-9.

5. Stevens KJ, Stevens JA. Aneurysmal bone cysts. In: StatPearls. Treasure Island, FL: StatPearls Publishing; 2023.

6. Abuhassan FO, Shannak A. Non-vascularized fibular graft reconstruction after resection of giant aneurysmal bone cyst (ABC). Strategies Trauma Limb Reconstr 2010;5:149-54.

7. Brindley GW, Greene JF, Frankel LS. Case reports: Malignant transformation of aneurysmal bone cysts. Clin Orthop Relat Res 2005;438:282-7.

8. Schajowicz F, Sissons HA, Sobin LH. The World Health Organization's histologic classification of bone tumors. A commentary on the second edition. Cancer 1995;75:1208-14.

9. Kaplanoğlu V, Ciliz DS, Kaplanoğlu H, Elverici E. Aneurysmal bone cyst of the calcaneus. J Clin Imag Sci 2014;4:60.

10. Babazadeh S, Broadhead ML, Schlicht SM, Powell GJ, Tymms GM. Pathologic fracture of a calcaneal aneurysmal bone cyst. J Foot Ankle Surg 2011;50:727-32.

11. Kitamura T, Ikuta K, Senba H, Komiya N, Shidahara S. Long-term follow-up of a case of aneurysmal bone cyst in the lumbar spine. Spine J 2013;13:e55-8.

12. Deventer N, Deventer N, Gosheger G, de Vaal M, Vogt B, Budny T. Current strategies for the treatment of solitary and aneurysmal bone cysts: A review of the literature. J Bone Oncol 2021;30:100384.

13. Mohaidat ZM, Al-Gharaibeh SR, Aljararhih ON, Nusairat MT, Al-Omari AA. Challenges in the diagnosis and treatment of aneurysmal bone cyst in patients with unusual features. Adv Orthop 2019; 2019:2905671.

14. Elsayad K, Kriz J, Seegenschmiedt H, Imhoff D, Heyd R, Eich HT, et al. Radiotherapy for aneurysmal bone cysts : A rare indication. Strahlenther Onkol 2017;193:332-40.

15. Enneking WF, Eady JL, Burchardt H. Autogenous cortical bone grafts in the reconstruction of segmental skeletal defects. J Bone Joint Surg Am 1980;62:1039-58.

16. Wilson PD. A clinical study of the biomechanical behavior of massive bone transplants used to reconstruct large bone defects. Clin Orthop Relat Res 1972;87:81-109.

17. Sailesh SS, Muthu S, Ismail ND. Hemi-fibular grafting for metacarpal giant cell tumor-surgical technique. J Orthop Case Rep 2020;10:80-4.

18. Ansari MT, Gautam D, Kotwal PP. Mother's fibula in son's forearm: Use of maternal bone grafting for aneurysmal bone cyst not amenable to curettage-a case report with review of literature. SICOT-J 2016;2:18.

19. Yadav SS. Dual-fibular grafting for massive bone gaps in the lower extremity. J Bone Joint Surg Am 1990;72:486-94.

20. Hakim DN, Pelly T, Kulendran M, Caris JA. Benign tumours of the bone: A review. J Bone Oncol 2015;4:37-41.

21. Rapp TB, Ward JP, Alaia MJ. Aneurysmal bone cyst. J Am Acad Orthop Surg 2012;20:233-41.

22. Duran S, Kose CC, Guresci S. An extra-articular ganglion cyst with multiple fluid-fluid levels. Curr Med Imaging 2024;20: e080823219538

23. Park HY, Yang SK, Sheppard WL, Hegde V, Zoller SD, Nelson SD, et al. Current management of aneurysmal bone cysts. Curr Rev Musculoskelet Med 2016;9:435-44.

24. Capanna R, Bettelli G, Biagini R, Ruggieri P, Bertoni F, Campanacci M. Aneurysmal cysts of long bones. Ital J Orthop Traumatol 1985;11:409-17.

25. Rahman MA, El Masry AM, Azmy SI. Review of 16 cases of aneurysmal bone cyst in the proximal femur treated by extended curettage and cryosurgery with recons using autogenous nonvascularized fibula graft. J Orthop Surg (Hong Kong 2018;26(12)

26. Yenigül AE, Sofulu Ö, Erol B. Treatment of locally aggressive benign bone tumors by means of extended intralesional curettage without chemical adjuvants. SAGE Open Med 2022;10:20503121221094199. doi: 10.1177/20503121221094199.

27. Solooki S, Keikha Y, Vosoughi AR. Can ethanol be used as an adjuvant to extended curettage in order to reduce the recurrence rate of aneurysmal bone cyst? Rev Brasil Ortop 2017;52:349-53.

28. Cevolani L, Staals E, Campanacci L, Dozza B, Martella C, Spinnato P, et al. Aneurysmal bone cyst: Is selective arterial

embolization effective as curettage and bone grafting? J Surg Oncol 2023;128(8):1428-36.

29. Gorski SM, Dong C, Lenze U, Haug M, Krieg AH. Vascularized and non-vascularized fibula grafts in tumour reconstruction: Single centre experience with mid to long-term results. Anticancer Res 2022;42:5443-7.

30. Liu Q, Long F, Zhang C, Liu Y, He H, Luo W. Biological reconstruction of bone defect after resection of malignant bone tumor by allograft: A single-center retrospective cohort study. World J Surg Oncol 2023;21:234.

31. Migliorini F, La Padula G, Torsiello E, Spiezia F, Oliva F, Maffulli N. Strategies for large bone defect reconstruction after trauma, infections or tumour excision: A comprehensive review of the literature. Eur J Med Res 2021;26:118.

32. Nishida J, Shimamura T. Methods of reconstruction for bone defect after tumor excision: A review of alternatives. Med Sci Monit 2008;14:RA107-13.

Conflict of Interest: Nil
Source of Support: Nil

Consent: The authors confirm that informed consent was obtained from the patient for publication of this case report

How to Cite this Article

Annamalai S, Kandasamy V, Chandrasekaran S, Muthu S.Cross-beam Non-vascular Fibular Graft Reconstruction in the Management of Long Bone Giant Aneurysmal Bone Cyst – A Case Series. Journal of Orthopaedic Case Reports 2023 December;13(12): 141-147.