Case Report | Journal of Orthopaedic Case Reports 2025 March:15(3):Page 12-16

Mother to Child Non-Vascularized Fibula Transfer in Congenital Pseudoarthrosis of Tibia

Mohammed Sadiq[1], Anudeep Manne[1], Rohith Reddy Kasam[1], Kothalanka Udaya Kumar[1], Libin Bernnabas[1]

Learning Point of the Article:

Maternal fibula as a bone graft for the management of congenital pseudoarthrosis of tibia is a better and cheaper alternative than BMP and allogenic cadaveric bone graft.

Abstract

Introduction: Congenital pseudoarthrosis tibia (CPT) is a relatively rare disease, characterized by anterolateral bowing of the tibia, non-union, and limb length discrepancy. Various surgical treatments have been described in literature for its management with differing favorable outcomes.

Case Report: In this report, we present a case of 3-year-old child with CPT of Crawford type IV, with associated fibular dysplasia. Maternal fibula was harvested and used as a bone graft and was stabilized by intra-medullary fixation. Complete union was achieved at 1 year after the primary surgery. No re-fractures were seen in a follow-up period of 2 years.

Conclusion: Using maternal fibula as an alternative to use as a bone graft in the management of congenital pseudoarthrosis tibia may prove beneficial. Moreover, it is cheaper and readily available and needs less surgical expertise when compared to its alternatives such as use of bone morphogenetic protein 7, allogenic cadaveric grafting, or use of vascularized fibular graft.

Keywords: Allograft, mother's fibula, fixation, congenital pseudoarthrosis tibia.

Introduction

Congenital pseudoarthrosis of tibia (CPT) is characterized by an area of segmental dysplasia in the tibia resulting in anterolateral bowing of the bone [1]. This can result in tibial non-union with limb shortening. The outcome of the condition is extremely unfavorable and, chances of fracture healing without intervention are remote [2].

The condition is relatively rare with an incidence of 1 in 150,000 births [3]. It is most commonly associated with neurofibromatosis type 1, an autosomal dominant condition [4]. According to the Crawford classification, the lesions associated with complete pseudoarthrosis with sclerotic edges and associated fibular dysplasia carry worse prognosis [5].

We present a case of 3-year-old child, who presented to us with Crawford type IV pseudoarthrosis tibia with associated fibular dysplasia. The child posed a challenge as there was a large segment of sclerosis within the tibia and high chances of having significant limb length discrepancy and angular deformity. The child was managed with a non-vascular maternal fibular graft which, to the best of our knowledge, has not been reported previously. Although, there are a few reports of maternal tibia being used as an on-lay graft and maternal periosteal grafting, reports of fibula being used with intra medullary fixation have not been found.

Access this article online

Website: www.jocr.co.in

DOI: https://doi.org/10.13107/jocr.2025.v15.i03.5316

Author's Photo Gallery


Dr. Mohammed Sadiq


Dr. Anudeep Manne


Dr. Rohith Reddy Kasam


Dr. Kothalanka Udaya Kumar


Dr. Libin Bernnabas

[1]Department of Orthopaedics, ESIC Medical College Hospital, Hyderabad, Telangana- 500038, India.

Address of Correspondence:
Dr. Anudeep Manne,
Department of Orthopaedics, ESIC Medical College Hospital, Hyderabad - 500 038, Telangana, India.
E-mail: dr.anudeepmanne@gmail.com

Submitted: 01/12/2024; Review: 22/01/2025; Accepted: February 2025; Published: March 2025

DOI: https://doi.org/10.13107/jocr.2025.v15.i03.5316

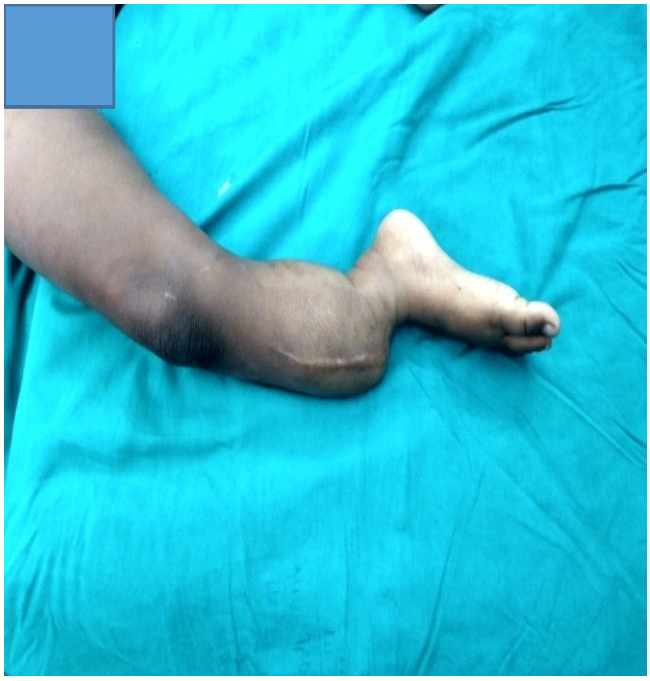

Figure 1: Clinical picture of the right lower limb at the time of first surgery. There was a scar noted on the anterior aspect of the leg from a previous surgery, details of which are not available. The age of the child at this point was 3 years.

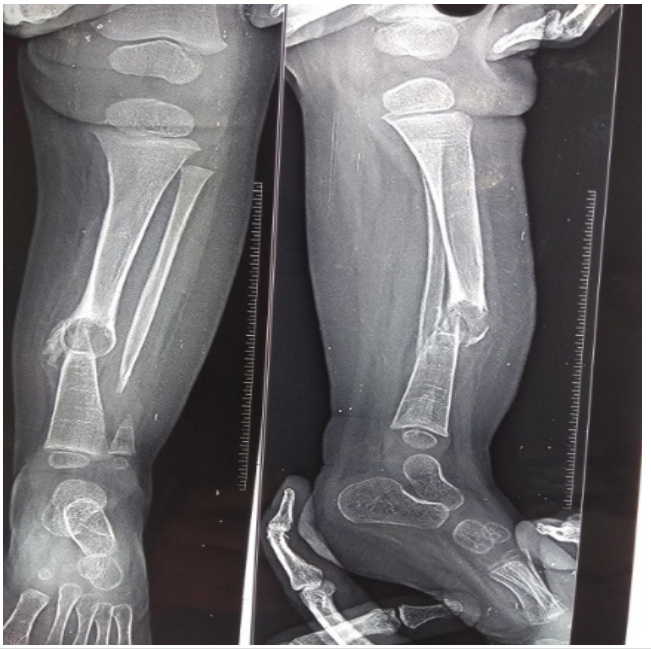

Figure 2: Plain radiographs showing presence of cystic lesion with formation of pseudoarthrosis between middle and distal 1/3rd of tibia, with significant narrowing of the bony ends. Fibula is also dysplastic. Magnetic resonance imaging shows presence of fibrotic soft-tissue interposition between the fragments. Computed tomography scan also reveals the presence of pseudoarthrosis.

Case Report

A 3-year-old male child presented to our outpatient department at ESIC Medical College and Hospital, Kalaburagi, Karnataka, India. At presentation, the child had an anterolateral bowing of the right leg with abnormal mobility present between the distal and proximal segments of the leg. The femoral length on the right side was 2 cm longer than the left, while the tibial segment was 6 cm shorter. There was a scar on the anterior aspect of the leg, due to some previous intervention attempted by some surgeon when the child was 2 years old (Fig. 1). However, no implant was placed and further details of the surgery were not available.

Radiographs showed the presence of a pseudoarthrosis at the junction of middle and lower 1/3rd of the right leg, with sclerotic margins of the bony ends (Fig. 2). The fracture was complete, and there was an anterolateral deformity between the fractured ends. The lower 1/3rd of fibula was also dysplastic. According to the Crawford classification, it was of type IV. Magnetic resonance imaging showed the presence of soft-tissue interposition between the fractured ends and computed tomography scan showed the presence of well-formed pseudoarthrosis.

Considering that the bony edges were tapering and sclerotic, we anticipated that a large segment of the bone had to be removed to get good bleeding in the bony margins. As, the child was only 3 years old, getting adequate native bone to bridge the gap was difficult. The only option left to us was to do a maternal bone transfer as cadaveric allograft facility is not available in our setup. Parents were explained about the expected problems, and available options. Mother consented for the transfer of her fibula to bridge the gap.

Two different surgical teams operated on two different tables. One team performed the fibular graft harvesting, while the other team comprising the author performed the procedure on the child. Surgical exposure revealed, frank non-union with sclerotic bone margins, and thick fibrotic tissue interposition. The fibrotic tissue was cleared thoroughly. The tapering bony edges were resected until good bleeding bone was visible. The gap between the resected bony edges was about 5 cm (Fig. 3).

The maternal fibula was completely denuded of soft-tissue attachments and intramedullary canal was reamed to clear all the marrow. Maternal fibula measuring exactly 5 cm was placed between the bone ends and stabilized with an intramedullary nail. Antigrade titanium elastic nail was passed starting from the lateral side. Additional iliac crest graft form the child was placed adjacent to the fibular ends (Fig. 4). External fixator was applied to provide additional stability. Wound closure was done in layers and an above knee posterior slab was applied.

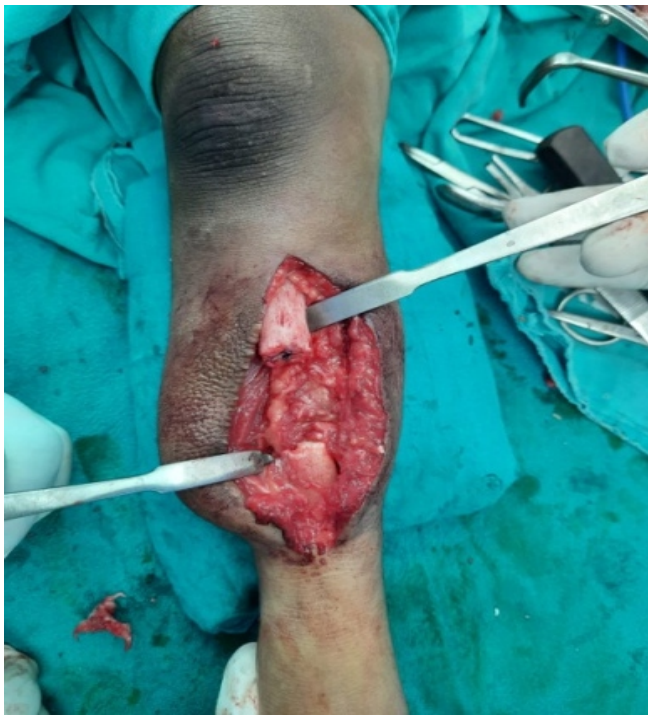

Figure 3: The interposing fibrous tissues were removed and sclerotic margins were excised. The gap between the bony ends was measured to be about 5 cm. Same length of fibula was harvested from the mother and cleared of intramedullary contents and placed between the bony ends. The diameter of the fibula was exactly matching the recipient tibia.

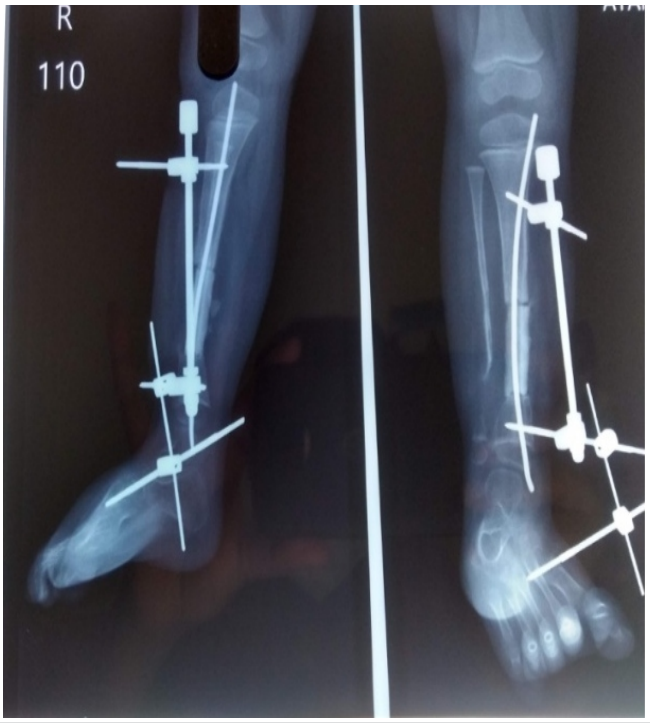

Figure 4: Immediate post-operative radiograph showing maternal graft well engaged between the bony ends of the recipient and is stabilized with an intramedullary nail and external fixator. At 3 months, follow-up bridging callus was seen at the proximal end while the distal end appeared to have delayed union and ends appeared sclerotic.

Postoperatively wound inspection was done t 2nd day and the patient was put on daily calcium supplements and weekly dose of bisphosphonates (35 mg of alendronate). The patient was discharged on the 5th day and was kept in regular monthly follow-ups. X-rays were done every month to look for the progress of the union. On the 3rd follow-up after surgery, the proximal end of the fibular graft showed signs of healing but the distal end showed signs of non-union (Fig. 4). Three months after the index surgery, bone marrow injection was injected adjacent to the distal end of the fibula and an additional pin was passed retrograde, across the ankle through the fibular graft. One year after the primary surgery, good healing of the pseudoarthrosis was achieved (Fig. 5). Child started walking bearing weight after 1 year, that is, at the age of 4 years. Finally, there was a shortening of <5 mm and there was no angular deformity. The patient was kept in observation for 2 years and there were no complications or re-fractures noted.

Discussion

Management of CPT surgically remains a challenge [1, 6, 7]. The ideal treatment should be able to provide a long-term bony union of tibia and fibula, should avoid limb length discrepancy or mechanical axis deviation, and should prevent joint stiffness and avoid re-fractures [8, 9]. Surgical options include, excision of the pathological tissue including the periosteum with a stable fixation using intra-medullary or extra-medullary fixation [10]. Addition of vascularized or non-vascularized bone grafting has also been described in the past. If surgical methods do not yield acceptable results, amputation can also be considered in failed cases [11].

Maternal bone graft has been used in many pediatric conditions requiring large segment bone grafting. Ansari et al. reported the use of maternal fibular graft in a child of 6–1/2 years with an aneurismal bone cyst (ABC) of the forearm measuring 11 cm [12]. Palanisamy and Balachandran reported a good incorporation of maternal fibula in a case of fibrous dysplasia of humerus in a 4 year old boy [13]. Maternal fibula has also been used for management of benign lytic lesions following excision and marginal resection [14]. Literature suggests that fresh allogenic graft elicit both acellular and humoral immune response, which leads to development of enhancing factors that block detectable immunity and prevent graft rejection. Accordingly, fresh viable allografts could be employed without prior freezing or tissue matching for human leukocyte antigen (HLA) antigens [15]. In our case, since it was maternal graft which was cleared of the marrow contents, there was no need for HLA typing.

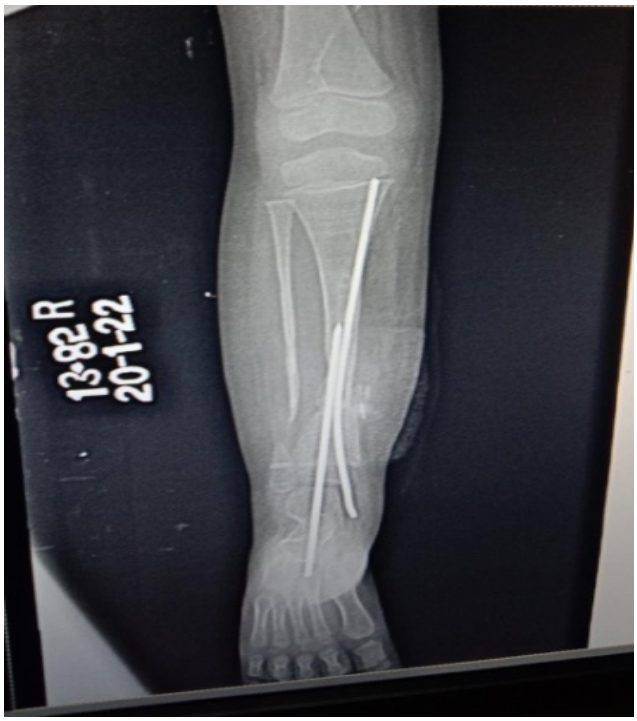

Figure 5: Since there was no union seen at the distal end of the maternal graft, bone marrow injection with retrograde pinning across the ankle was done. Complete union was seen at 9 months after the surgery. Child started walking with minimal shortening and angulation at 12 months after the first surgery.

However, the use of non-vascularized maternal fibula in CPT is rare and we have not come across such reports. D. Paley has reported the use of maternal periosteum by a surgeon, based on personal communications [16]. Some authors have described maternal tibial struts placed as on-lay grafts [17].

Conclusion

Through this report, we have described an alternative treatment for CPT in young children with frank pseudoarthrosis and long segment bony sclerosis. Such children if intervened surgically would require a lot of bone graft between the fractured ends. Maternal fibula would be ideal in such situation as the diameter of fibula actually match the diameter of pediatric tibia. It additionally provides good structural support until the process of bony remodeling is complete. It is a cheaper readily available and needs less surgical expertise when compared to its alternatives such as use of bone morphogenetic protein 7, allogenic cadaveric grafting, or use of vascularized fibular graft.

Clinical Message

Management of pseudoarthrosis tibia has always been a challenge for clinician with no definitive standardized method with proven results. Our experimentation with maternal fibula as an alternative for bone graft may prove beneficial for the management. Since we have found success in one case, it requires further research and scientific evidence to make it a standardized procedure in congenital pseudoarthrosis tibia.

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. Andersen KS. Radiological classification of congenital pseudarthrosis of the tibia. Acta Orthop Scand 1973;44:719-27.

2. Mahnken AH, Staatz G, Hermanns B, Gunther RW, Weber M. Congenital pseudarthrosis of the tibia in pediatric patients: MR imaging. AJR Am J Roentgenol 2001;177:1025-9.

3. Hefti F, Bollini G, Dungl P, Fixsen J, Grill F, Ippolito E, et al. Congenital pseudarthrosis of the tibia: History, etiology, classification, and epidemiologic data. J Pediatr Orthop Part B 2000;9:11-5.

4. El-Rosasy MA. Congenital pseudarthrosis of the tibia: The outcome of a pathology-oriented classification system and treatment protocol. J Pediatr Orthop Part B 2020;29:337-47.

5. Crawford AH. Neurofibromatosis in children. Acta Orthop Scand Suppl 1986;218:1-60.

6. Boyd HB. Pathology and natural history of congenital pseudarthrosis of the tibia. Clin Orthop Relat Res 1982;166:5-13.

7. Gutmann DH, Aylsworth A, Carey JC, Korf B, Marks J, Pyeritz RE, et al. The diagnostic evaluation and

multidisciplinary management of neurofibromatosis 1 and neurofibromatosis 2. JAMA 1997;278:51-7.

8. Karol LA, Haideri NF, Halliday SE, Smitherman TB, Johnston CE 2nd. Gait analysis and muscle strength in children with congenital pseudarthrosis of the tibia: The effect of treatment. J Pediatr Orthop 1998;18:381-6.

9. Johnston CE 2nd. Congenital pseudarthrosis of the tibia: Results of technical variations in the Charnley-Williams procedure. J Bone Joint Surg Am 2002;84:1799-810.

10. Shah H, Rousset M, Canavese F. Congenital pseudarthrosis of the tibia: Management and complications. Indian J Orthop 2012;46:616-26.

11. Weber M. Neurovascular calcaneo-cutaneus pedicle graft for stump capping in congenital pseudarthrosis of the tibia: Preliminary report of a new technique. J Pediatr Orthop Part B 2002;11:47-52.

12. 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.

13. Palanisamy JV, Balachandran R. Incorporation of allogenous fibular graft in pediatric humerus following segmental resection. IOSR J Dent Med Sci 2016;7:1-4.

14. Salunkhe AA, Raiturker PP. Benign lytic lesions of the bone, staging and management correlations. Indian J Orthop 2003;37:8.

15. Langer F, Czitrom A, Pritzker K, Gross A. The immunogenicity of fresh and frozen allogeneic bone. J Bone Joint Surg Am 1975;57:216-20.

16. Paley D. Congenital pseudarthrosis of the tibia: Biological and biomechanical considerations to achieve union and prevent refracture. J Child Orthop 2019;13:120-33.

17. Heikkinen ES, Poyhonen MH, Kinnunen PK, Seppänen UI. Congenital pseudarthrosis of the tibia. Treatment and outcome at skeletal maturity in 10 children. Acta Orthop Scand 1999;70:275-82.

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

Sadiq M, Manne A, Kasam RR, Kumar KU, Bernnabas L. Mother to Child Non-Vascularized Fibula Transfer in Congenital Pseudoarthrosis of Tibia. Journal of Orthopaedic Case Reports 2025 March;15(3): 12-16.