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Long-Term Evaluation of Bone Healing and Functional Outcome Following Vascularized Fibular Grafting for Congenital Pseudoarthrosis of the Tibia in Pediatric Population

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Abstract
Congenital pseudoarthrosis of the tibia presents a challenging treatment scenario. In a recent investigation, vascularized fibular grafting (VFG) was employed to address this condition in a cohort comprising five female and three male patients. The mean age of the recipients undergoing VFG was 7.1 years, ranging from 1.8 to 11.6 years, and the subsequent follow-up extended over an average of 11.8 years, with a range spanning from 4.8 to 19.7 years. Among the subjects, five individuals had previously undergone multiple surgical interventions, while the remaining three had not undergone any prior operations. Across all instances, successful bone consolidation was achieved within an average period of 6.7 months. In seven cases, this consolidation was achieved following the initial VFG procedure, whereas one case necessitated a second VFG for the consolidation. Notably, patients with a history of multiple prior operations exhibited a more significant discrepancy in leg length, averaging 7.6 cm, in contrast to those without prior surgical interventions, who displayed an average leg length difference of 0.8 cm. The results underscore VFG as a promising primary treatment approach for CPT, displaying favorable long-term outcomes, particularly in cases where prior surgical interventions had not been undertaken.

INTRODUCTION

Congenital pseudoarthrosis of the tibia (CPT) stands as a formidable challenge in the realm of medical intervention, characterized by a persistent instability and progressive deformity as it advances [1,2]. This condition has a notable association with NF1, also recognized as von Recklinghausen disease. In response, a range of treatment avenues, encompassing both surgical and non-surgical approaches, have emerged. Noteworthy among these are vascularized fibular grafts (VFG), intramedullary stabilizations, and external fixations, all of which have demonstrated relative success in addressing CPT [3-9]. In a preceding study by our team [3], we detailed the efficacious treatment of five CPT patients through the utilization of VFG. Nonetheless, a critical requirement persists for comprehensive, long-term follow-up investigations of VFG's impact. Such studies are crucial for identifying persistent issues like limb-length disparities. These complications, despite apparent bone consolidation in cases of pseudoarthrosis, can significantly compromise functional outcomes [2,10].

This study undertakes an examination of five cases previously documented and supplements them with three fresh cases, collectively aiming to discern the enduring outcomes of VFG treatment. A central

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objective of this study is to draw comparisons between patients with a history of multiple surgical interventions and those who underwent VFG without prior surgical experiences. Notably, the current series does not involve microvascular bone transplantation in any of the preceding surgical procedures, which were primarily centered around intramedullary stabilization accompanied by or devoid of bone grafts.

Methods

Five girls and three boys were among the patients. NF1 was present in six of them (6/8). Five cases of pseudarthrosis involved the right side of the tibia, while three cases involved the left side. First preference was given to ipsilateral VFG, and contralateral VFG was conducted if ipsilateral fibula was not available. Accordingly, ipsilateral VFG was applied in 7 cases and contralateral VFG in 2, in which one case was for the initial VFG trial and the other was for the second trial after ipsilateral VFG failed to consolidate the bone. Prior to attending our hospital, five patients had undergone multiple operations in other institutions. This group of patients has been reported in a study of short-term follow-up [3]. There were 3 to 8 operations performed on these patients, with an average of 3.5. In all these cases, intramedullary nails were used with or without bone grafts. In this current investigation, we expanded the patient cohort to encompass an additional three individuals who had not been subjected to prior treatment. Through a meticulous juxtaposition of patients with antecedent surgical intervention and those without such history prior to undergoing VFG, we scrutinized various aspects. These included the duration required for bone consolidation, as well as the emergence of complications such as differences in leg length.

Operative technique

A summary of the method of VFG is as follows: Previous operation, angiography was performed to determine the vascular anatomy. Vascularized fibulas (peroneal vessels) were dissected. For the donor, usually the fibula closest to the pseudo-orthrosis site was used. Resection of the fibrous tissue surrounding the tibial pseudoarthrosis was complete, while resection of the sclerotic bone ends was minimal.

Statistical analysis

Quantitative data pertaining to bone consolidation and leg-length discrepancy underwent statistical analysis utilizing the Mann-Whitney U-test. Similarly, for qualitative information concerning ankle pain, Fisher's exact test was employed. To establish statistical significance, a threshold of $p < 0.05$ was adopted.

RESULTS

PATIENTS

During the VFG surgical intervention, the patients had an normal age of 7.1 years. Upon concluding the final follow-up, the patients had an average age of 19.4 years (ranging from 8.2 to 27.4 years), and the mean duration of postoperative follow-up encompassed 11.8 years (ranging from 4.8 to 19.7 years). Throughout this extended observation period, there were no instances of recurrence.

Bone consolidation

A complete bone consolidation, observed in all eight patients (8/8), was the outcome following VFG. In the subset of five patients with a history of multiple surgical interventions, all cases (5/5) exhibited successful bone consolidation after their initial VFG operation, negating the need for subsequent surgical procedures. In contrast, among the three patients without prior surgical intervention, one patient failed to achieve bone consolidation after the initial ipsilateral VFG operation performed at the age of 1.8 years. Subsequently, at the age of 7.4 years, this patient underwent a second VFG operation on the contralateral side, resulting in bone consolidation five months thereafter. Incorporating this case of second VFG operation into the dataset, the mean duration for bone consolidation across all eight cases ranged from 5 to 9 months. In this particular instance, consolidation was accomplished 7 months after the second operation.

Among the three patients with no history of prior surgery, two achieved bone consolidation within a span of 10 months. A comparison between patients who had undergone multiple operations and those without previous surgery indicated a bone consolidation period of 6.9 months for the former and 6.4 months for the latter. Notably, these outcomes did not exhibit significant variation ($p = 0.78$).

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Sex/Age	Number of previous operations	Donor site	Term until union	Age at follow-up (term)	Leg-length discrepancy (before VFG)
F/7.5 y	9	I	8 m	27.1 y	5.3 cm
F/8.9 y	4	I	7 m	21.7 y	5.9 cm
F/8.9 y	8	I	6 m	27.4 y	10.3 cm
M/8.8 y	5	C	6 m	22.3 y	0.7 cm
F/11.6 y	7	I	8 m	18.6 y	15.8 cm
M/1.8 y	0	I	Non union		
M/3.3 y	0	I	9 m	8.2 y	0.0 cm
F/6.3 y	0	I	5 m	15.4 y	2.1 cm

Complications

Leg-length discrepancy

In aggregate, an average leg-length discrepancy of 4.8 cm was observed across all cases, with variations spanning from 0 to 15.8 cm. Notably, cases that had undergone multiple surgical interventions exhibited a notable leg-length discrepancy of 7.6 cm (ranging from 0.7 to 15.8 cm), while those without prior surgical history had a comparatively minimal discrepancy of 0.8 cm (ranging from 0.0 to 2.1 cm) ($p = 0.08$). Specifically, among the cases undergoing multiple surgeries, 4 out of 5 displayed a discrepancy exceeding 5.1 cm, whereas all 3 cases without prior surgery maintained a discrepancy within this threshold. This discrepancy was already evident prior to the application of VFG, with cases undergoing multiple operations showcasing a before operative leg-length discrepancy of 6.9 cm. Intriguingly, no significant disparity was discernible between the pre- and post-operative states of VFG (measuring 6.9 cm and 7.6 cm, respectively) within the multiple-operation group ($p = 0.85$). This pattern indicates that the existing discrepancy was attributed to the preceding surgical procedures conducted before VFG.

Discussion

Vascularized fibular grafting (VFG) has proven effective in treating congenital pseudoarthrosis of the tibia (CPT) in 94% of cases, resulting in successful bone consolidation [4]. In all instances treated with VFG, bone consolidation was achieved within an average of 6.6 months and without any recurrence. While previous research has indicated a potential gender-based influence on the consolidation period, as seen with boys having an average consolidation term of 13 months and girls, 9 months [2], such a trend was not evident in our study. At our institute, VFG stands as the primary treatment modality, with Ilizarov bone transport being considered an alternative option due to the favorable bone consolidation results following VFG. The Ilizarov bone transport approach has shown promise in promoting primary healing in CPT [11], though it has been accompanied by complications such as refractures and postoperative deformities. There remains a necessity for further exploration through long-term follow-up investigations of the Ilizarov bone transport method.

The influence of age on the outcomes of VFG, particularly bone consolidation, has been highlighted in previous studies. Older patients have exhibited successful outcomes, with a report indicating that patients over 10 years of age had more success compared to those under 9 years old [12]. An EPOS (European Pediatric Orthopedic Society) Multicenter Study [6] also underscored the impact of age on final outcomes, showing that younger patients tend to have better results. Consequently, it is advised that operation not be undertaken on patients younger than 3 years of age, with a recommended delay until the age of 5 [6]. Another study on bone consolidation in CPT has suggested that the optimal age range for swift consolidation lies between 3.5 and 7.5 years [4].

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Notably, an instance of unsuccessful bone consolidation in our study involved a patient aged 1.9 years. The subsequent success achieved after the patient's second contralateral VFG at the age of 7.3 might corroborate the notion that a younger age might adversely affect bone consolidation. Dealing with CPT can engender tibial deformities, leading to limb-length discrepancies. [2,4]. It's important to note that the end outcome is not solely determined by bone consolidation. In cases of chronic lower-extremity dysfunction and clinical symptoms, amputation might be considered [7]. Among cases involving multiple prior operations, a majority exhibited limb-length discrepancies exceeding 5 cm, while none of the cases without prior operation exhibited such discrepancies. The average discrepancy was 7.5 cm for the former group and 0.7 cm for the latter. While the small number of cases precludes statistically significant differences ($p = 0.07$), it's worth noting that surgical procedures that pose a risk to the growth plate might contribute to limb-length disparities, potentially explaining the deformity observed before VFG [12].

Among the cases examined, ankle pain was experienced by three individuals. Notably, these instances occurred later in life than the VFG procedure itself (mean of 18.3 years post-VFG) and were associated with multiple prior operations [5,13]. It's crucial to acknowledge that individuals who were unoperated upon before VFG were more prone to experiencing long-term ankle pain, even when short-term follow-up had not indicated such discomfort. Prior research has aligned with these findings, associating post-VFG ankle pain with multiple surgeries involving intramedullary nails [14]. Such pain can stem from degenerative changes due to ankle valgus deformities or the intramedullary rod's interaction with the ankle joint [14-16]. Following VFG consolidation, fractures are not uncommon [13,17,18]. First fractures tend to take place before the age of 1 year [4]. In this study, one case experienced a stress fracture at the age of 7.4 years, which was effectively managed through casting.

Conclusion

Based on the comprehensive outcomes observed during the extensive follow-up period, it is evident that VFG has demonstrated remarkable efficacy. Notably, even in cases characterized by a history of multiple surgical interventions, instances of enduring limb-length disparities and persistent ankle discomfort were prevalent. Conversely, patients who received VFG as their initial treatment encountered fewer challenges associated with these concerns. Consequently, it becomes evident that VFG should rightfully be regarded as the primary and optimal treatment approach for addressing congenital pseudoarthrosis of the tibia.

References

1. Boyd HB: Pathology and natural history of congenital pseu- darthrosis of the tibia. Clin Orthop Relat Res 1982:5-13.
2. Morrissy RT, Riseborough EJ, Hall JE: Congenital pseudarthrosis of the tibia. J Bone Joint Surg Br 1981, 63-B(3):367-375.
3. Uchida Y, Kojima T, Sugioka Y: Vascularised fibular graft for con- genital pseudarthrosis of the tibia. Long-term results. J Bone Joint Surg Br 1991, 73(5):846-850.
4. Gilbert A, Brockman R: Congenital pseudarthrosis of the tibia. Long-term followup of 29 cases treated by microvascular bone transfer. Clin Orthop Relat Res 1995:37-44.
5. Kanaya F, Tsai TM, Harkess J: Vascularized bone grafts for con- genital pseudarthrosis of the tibia. Microsurgery 1996, 17(8)::459-69; discussion 470-1.
6. Grill F, Bollini G, Dungl P, Fixsen J, Hefti F, Ippolito E, Romanus B, Tudisco C, Wientroub S: Treatment approaches for congenital pseudarthrosis of tibia: results of the EPOS multicenter study. European Paediatric Orthopaedic Society (EPOS). J Pediatr Orthop B 2000, 9(2):75-89.
7. Dobbs MB, Rich MM, Gordon JE, Szymanski DA, Schoenecker PL: Use of an intramedullary rod for treatment of congenital pseudarthrosis of the tibia. A long-term follow-up study. J Bone Joint Surg Am 2004, 86-A(6):1186-1197.
8. Paley D, Catagni M, Argnani F, Prevot J, Bell D, Armstrong P: Treat- ment of congenital pseudoarthrosis of the tibia using the Ili- zarov technique. Clin Orthop Relat Res 1992:81-93.
9. Guidera KJ, Raney EM, Ganey T, Albani W, Pugh L, Ogden JA: Ili- zarov treatment of congenital pseudarthroses of the tibia. J Pediatr Orthop 1997, 17(5):668-674.
10. Leung PC: Congenital pseudarthrosis of the tibia. Three cases treated by free vascularized iliac crest graft. Clin Orthop Relat Res 1983:45-50.

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www.jchr.org

JCHR (2023) 13(2), 433-437 | ISSN:2251-6727

11. Kristiansen LP, Steen H, Terjesen T: Residual challenges after healing of congenital pseudarthrosis in the tibia. Clin Orthop Relat Res 2003:228-237.
12. Romanus B, Bollini G, Dungl P, Fixsen J, Grill F, Hefti F, Ippolito E, Tudisco C, Wientroub S: Free vascular fibular transfer in con- genital pseudoarthrosis of the tibia: results of the EPOS mul- ticenter study. European Paediatric Orthopaedic Society (EPOS). J Pediatr Orthop B 2000, 9(2):90-93.
13. Weiland AJ, Weiss AP, Moore JR, Tolo VT: Vascularized fibular grafts in the treatment of congenital pseudarthrosis of the tibia. J Bone Joint Surg Am 1990, 72(5):654-662.
14. Inan M, El Rassi G, Riddle EC, Kumar SJ: Residual deformities fol- lowing successful initial bone union in congenital pseudoar- throsis of the tibia. J Pediatr Orthop 2006, 26(3):393-399.
15. Andersen KS: Congenital pseudarthrosis of the tibia and neu- rofibromatosis. Acta Orthop Scand 1976, 47(1):108-111.
16. Anderson DJ, Schoenecker PL, Sheridan JJ, Rich MM: Use of an intramedullary rod for the treatment of congenital pseudar- throsis of the tibia. J Bone Joint Surg Am 1992, 74(2):161-168.
17. Dormans JP, Krajbich JI, Zuker R, Demuynk M: Congenital pseu- darthrosis of the tibia: treatment with free vascularized fibu- lar grafts. J Pediatr Orthop 1990, 10(5):623-628.
18. Minami A, Kaneda K, Itoga H, Usui M: Free vascularized fibular grafts. J Reconstr Microsurg 1989, 5(1):37-43.