Original Clinical Article

JOURNAL OF CHILDREN'S ORTHOPAEDICS

Journal of Children's Orthopaedics
2022, Vol. 16(3) 167–173

DOI: 10.1177/18632521221097525
journals.sagepub.com/home/cho

Novel method for the treatment of congenital pseudarthrosis of the tibia using the gastrocnemius flap: A preliminary study

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Abstract

Purpose: Congenital pseudarthrosis of the tibia is a rare disease that is particularly difficult to treat; the most difficult complications include nonunion of the tibia, refracture, and failed surgery. This study aimed to evaluate the efficiency of transposing gastrocnemius flaps for the treatment of congenital pseudarthrosis of the tibia.

Methods: Nine patients (aged 6.2 ± 3.6 years) diagnosed with congenital pseudarthrosis of the tibia in our hospital between March 2013 and March 2018 were enrolled. The tibial pseudarthrosis and thickened periosteum were completely removed, and intramedullary nails were used to fix the tibia. Bone harvest from the iliac, mixed with allogenic bone, was filled in the gap created by excision of the pseudarthrosis site and the surrounding periosteum; the gastrocnemius flap was then used to wrap the pseudoarthrosis site. The plaster cast was fixed postoperatively. The tibial union was evaluated via radiograph, and the plaster cast was removed after 12–24 weeks. Patients began walking approximately 12–14 weeks postoperatively.

Results: Anatomical reduction was achieved in all the patients; the mean bone healing time was 10.1 ± 2.1 months. Bone nonunion was observed in one patient, and no neurovascular injury or wound infection occurred. Limb length discrepancy was in the range 3.2 ± 1.8 cm at 1 year and 4.7 ± 2.7 cm at 2 years after surgery. Two patients underwent replacement of the intramedullary nail, and eight patients exhibited good functional and radiographic outcomes.

Conclusion: This preliminary study proved that using the gastrocnemius muscle flap to cover the pseudarthrosis site was an effective method to promote the tibial union and treat congenital pseudarthrosis of the tibia.

Keywords: Congenital pseudarthrosis of the tibia, gastrocnemius muscle flap, children

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Received 12 January 2022; revised manuscript accepted 18 March 2022

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Introduction

Congenital pseudarthrosis of the tibia (CPT) is a rare disease with an incidence between 1 in 140,000 and 1 in 250,000 live births.[1] While the etiology of CPT remains debatable, it is associated with neurofibromatosis in 50% of patients.[2,3] Although surgery remains the most effective method, treatment for CPT can be challenging.[3] Various methods have been used—including the Ilizarov technique, free fibular bone graft, the Charnley–Williams method, vascularized fibular transplant, and the Masquelet technique;[4] however, there is still no consensus regarding which method offers the best union and lowest refracture rates or even the best age to perform surgery.[5]

Currently, most scholars advocate a combination of two or three methods. Selection of the most appropriate method can minimize patient suffering and help avoid later amputation;[6,7] thus, complete removal of the lesion and promotion of bone healing are the key aims of treatment. There are many complications associated with CPT, such as refractures, nonunion, and ankle valgus; among these, nonunion is considered the most severe, and addressing this complication is the key to treatment success. To resolve this complication, many studies have used techniques, such as bone graft, bone morphogenetic protein-2, bone morphogenetic protein-7, pamidronate,[8–12] and even mesenchymal stromal cells combined with platelet-rich fibrin.[13] There remain many disputes regarding these methods, which provided unideal effects; thus, a single method cannot perfectly resolve the nonunion complications.

In this study, we used gastrocnemius muscle flap transfer to wrap the pseudojoint area and evaluated the effect on bone union. To the best of our knowledge, this is the first study concerning promoting bone union in patients with CPT treated via gastrocnemius muscle flap transfer. This study aimed to describe the procedure and retrospectively evaluate the outcomes of gastrocnemius muscle flap transfer.

Methods

Patients

Nine children with CPT who underwent gastrocnemius flap transfer during surgery at our hospital between March 2013 and 2018 were enrolled in this study. All the patients had type IV CPT according to Crawford's classification.[14] Preoperatively, all the patients were diagnosed with CPT and were either unable to walk or could only walk with the assistance of a walking aid. All surgeries were performed by the same pediatric orthopedic surgeon (with over 5 years of experience). The ethics committee of our hospital approved this study, and written informed consent was obtained from the parents or guardians of all patients before surgery.

Surgical procedure

Surgery was performed under general anesthesia and in the supine position. The first step was autologous bone graft harvesting from the iliac crest, wherein a triangular bone was used to fill the gap created after the excision of the pseudarthrosis site. An incision measuring approximately 5 cm in length was made from the anterior superior ileac spine. The subcutaneous fat and iliac cartilage were exposed accordingly. A scalpel was used to cut the iliac cartilage down from the middle; subsequently, the iliac was exposed, and an adequately long tricortical bone osteotomy was performed such that shortening the tibia was not required. Furthermore, the wound was irrigated, and the iliac cartilage was subsequently securely reattached to the ilium. The subcutaneous tissue and skin are closed. In the second step, a tourniquet was applied to the leg. Incisions were generally placed on the inside of the calf (to expose nonunion) and the medial head of the gastrocnemius muscle. Nonunion of the tibia was completely exposed, and lesioned tissues—including the fibrous connections at the pseudojoint, periosteum, surrounding abnormal muscles, and fascia, as well as the tapered ends of the bone—were removed entirely (Figure 1(a)–(c)). The extent of resection was determined by the surgeon during surgery based on the macroscopic evaluation of the bone and the reappearance of a permeable intramedullary canal. The longitudinal length of the excised periosteum was measured. The two ends of the tibia were expanded, and the appropriate Rush nail was selected to penetrate from the tibial tuberosity through the ankle joint to the talus, and assistance was required to maintain the length of the calf side when the nail was inserted. The autologous bone was transplanted together with the allogeneic bone. Coverage of the pseudojoint area by the gastrocnemius flap—along with autologous cancellous bone/artificial bone or periosteal grafting—can help restore healthy periosteum at the CPT site; the flap ensures adequate blood supply and nutrient delivery to the pseudojoint area. Rush nail, an intramedullary nail (manufactured by Kangding Medical Company, Shanghai, China) matching the diameter of the medullary canal was initially inserted through the anterior tibial tuberosity; if unsuccessful, it was introduced through the anterior aspect of the proximal tibial epiphysis and subsequently through the medullary cavity past the pseudojoint area and ankle joint, and finally through the talus to fix the tibia (Figure 1(b)). While searching for the anatomical landmark separating the soleus and the gastrocnemius medial head, we observed that some parts of the medial head of the gastrocnemius muscle had been cut off, the locations of which we are unsure of. Generally, we assessed the patients for pseudoarthrosis and evaluated the height. False joint resection of the osseous length is related to the location of the false joint if it interrupts in the tibia; in addition, we assessed the closure of the medial head of the proximal muscle fascicle. The medial head of the gastrocnemius muscle was cut off from the

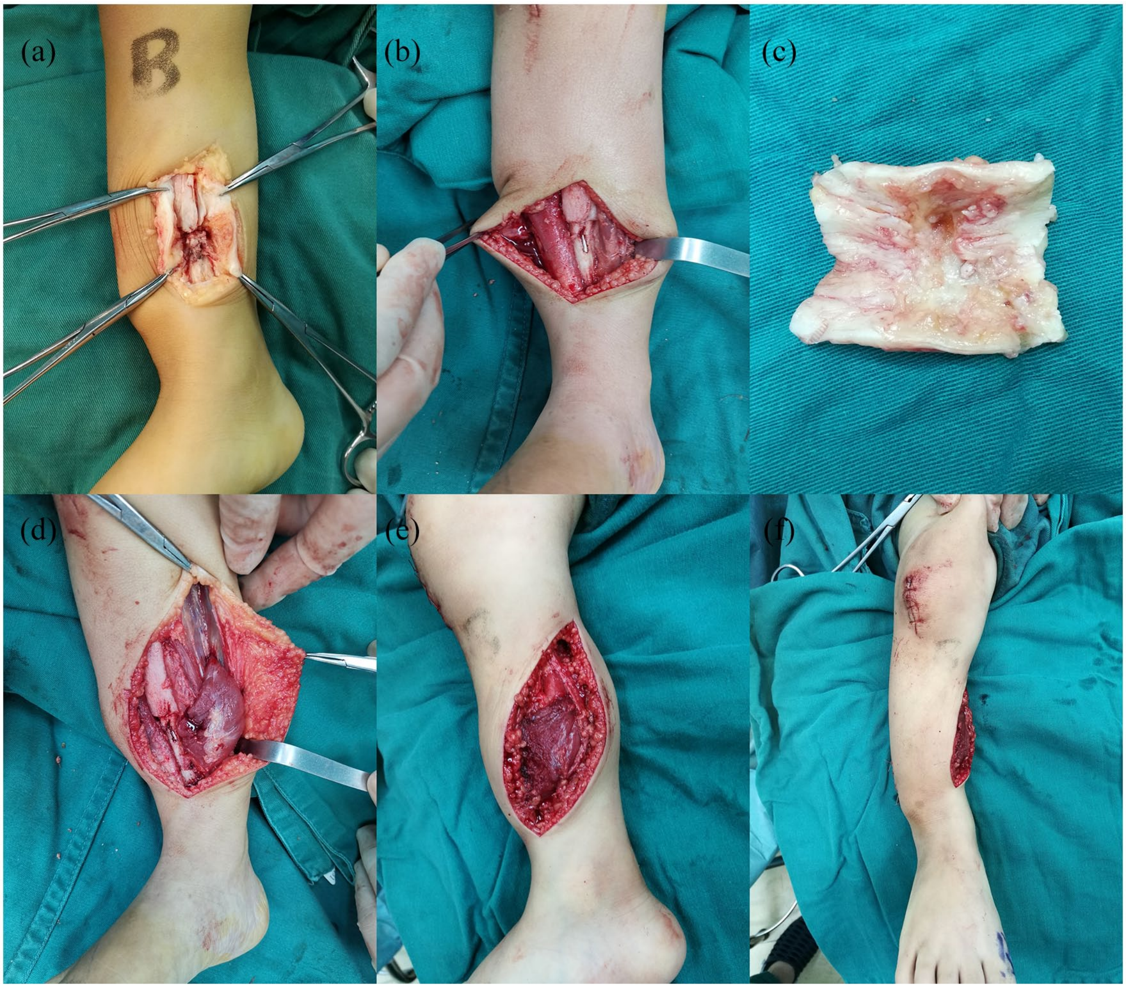

Figure 1. A 5-year-old boy with right tibial pseudarthrosis: (a) shows the lesion of the tibia, completely exposing the periosteum, abnormal muscles and fascia, and tapered ends of the bone; (b) shows that the lesion was completely removed, and the intramedullary nail was placed; (c) shows the abnormal tissue of the lesion; (d) shows part of the gastrocnemius flap transferred to the tibia; (e) shows the gastrocnemius flaps transferred to the tibia; the blood supply was adequate; (f) shows the appearance of the tibia.

proximal end, and about one-third to half of the medial head of the gastrocnemius muscle was transected; subsequently, it was longitudinally split, and the covering bone graft was laterally pulled over. Furthermore, the anterior tibial muscle was pulled medially (for suture and fixation) to form muscle coverage for the bone graft area. The muscle was then freed from the medial head of the gastrocnemius triceps to the pseudarthrosis site, ensuring that the blood supply to the muscle flap was preserved. An autologous bone graft was implanted, and the muscle flap was used to completely cover the nonunion area (Figure 1(d)); the tourniquet was subsequently loosened to confirm that the muscle flap had adequate blood supply (Figure 1(f)). A drainage tube was inserted, and the wound was closed without performing skin graft after the gastrocnemius flap transfer. The Rush intramedullary nail is particularly strong and can effectively support the tibia and maintain the tibia's vertical line; however, a sufficiently long cast is necessary to prevent the end of the bone from rotating because of the frequent occurrence of the valgus ankle, which is observed in approximately 45% of patients.[15] The limb was immobilized with a plaster slab, which was replaced with a full plaster cast 1 week later.

Follow-up

The plaster cast was removed after 8–12 weeks of wear, once anteroposterior and lateral radiographs of the tibia confirmed continuous callus formation at the pseudarthrosis site. Walking with partial weight-bearing was initiated 12–14 weeks post-surgery, and all the patients were assisted with braces or plaster fixation. During this time,

Table 1. Demographic and clinical characteristics of patients and the follow-up findings.

Number	Sex	Age (years)	Side	CT	OL (mm)	HT (m)	FT (years)	NF-1	AF	Re
1	F	8.5	R	IV	25	10	4.3	+	+	−
2	F	12.8	L	IV	31	9	3.4	+	+	−
3	M	8.2	L	IV	22	8	2.6	+	+	−
4	M	2.9	R	IV	19	11	2.7	+	+	−
5	M	5.8	R	IV	25	12	3.9	−	+	−
6	M	4.2	R	IV	21	14	3.3	+	+	−
7	M	8.5	R	IV	27	8	2.4	+	+	−
8	M	2.6	R	IV	24	9	2.3	+	+	−
9	F	2.3	R	IV	22	NA	3	−	+	−

R: right; L: left; CT: Crawford type; OL: osteotomy length; HT: healing time; FT: follow-up time; AF: ankle fixation; NF-1: neurofibromatosis type 1; NA: not available; Re: refracture.

the bone callus was observed from the anterior-posterior and lateral radiographs of the tibia. If the bone callus was dense and the fracture lines could not be observed, the braces or plaster was removed, and full weight-bearing was initiated. The follow-up was initiated every 3–4 weeks. The Rush nails were passed through the ankle and driven into the calcaneus to provide stability while the tibia healed; notably, the nails can be left in situ in the tibia for a long time, as they prevent refracture and do not affect lower limb function.

Results

In total, nine patients (seven boys, two girls; aged 6.2 ± 3.6 years) were treated using the gastrocnemius flap and Rush nails (Table 1); six had pseudarthrosis on the left side, and three had pseudarthrosis on the right side. The mean follow-up duration was 3.1 ± 0.7 years. The mean intraoperative blood loss was only 10 mL, and straightening of the tibial vertical line was achieved in all the patients. The mean bone healing time was 10.1 ± 2.1 months. None of the patients exhibited deformities of the ankle joint or developed wound infection. The mean length of hospitalization was 5.2 ± 1.8 days. While eight patients achieved satisfactory bone union according to the follow-up radiographs, one patient had nonunion. In two patients, the intramedullary nails required replacement as they were too short (Figure 2(e) and (f)), and none of the patients experienced refracture.

Discussion

This study aimed to describe the use of the gastrocnemius muscle flap to cover the area of pseudarthrosis during treatment for CPT and evaluate the promotion of bone union using this technique. Although the gastrocnemius muscle flap has been successfully used to improve blood flow for several conditions (e.g. nonunion and bone infection),[16,17] its use in patients with CPT has not been previously reported. The primary outcome demonstrated that this method was useful, and 8/9 patients achieved union after the first surgery.

CPT is a particularly challenging disease for pediatric orthopedic surgeons,[5] as there are many complications that require surgery, including fractures, leg-length discrepancies, and ankle valgus.[5] The first surgical treatment aims to achieve bone union at the pseudarthrosis site, restore leg alignment, prevent recurrent fracture, and preserve the ankle function. The goal of all surgical procedures is to achieve bone healing; however, the mean union rate in previous studies has only been 72%. A success rate of slightly over 50% is not very promising for either the patients' guardians or surgeons. In our preliminary study, the healing rate of the tibia was 88.9% (8/9), which is higher than that observed in previous studies.

The mechanism of CPT is not fully understood; however, over 50% of cases are associated with neurofibromatosis type 1.[18] Vessels play an important role in the formation of the tibial pseudarthrosis.[19] In previous multicenter studies, during treatment via vascularized bone transfer, wide resection of the dystrophic tibia is followed by replacement with healthy vascularized bone, and the rate of primary union following vascularized transfers was >70%.[20,21] However, intramedullary fixation is difficult to achieve via this method due to the risk of injury to the vascularized transplant. In this study, the gastrocnemius muscle flap was used to cover the pseudojoint, and the vessels—together with muscle—were transferred around the lesion; this may be one of the factors (with regard to the method employed) that promoted bone healing.

We have only applied our technique in nine patients thus far due to the rarity of CPT; nevertheless, the treatment was successful in 8/9 patients, which is very encouraging. In patients with CPT, the abnormal periosteum can form a fibrous constricting band that would decrease blood flow, leading to bone atrophy.[2] We believe that pseudojoint formation occurs primarily due to changes in the surrounding periosteum and soft tissues; thus, we opted to completely remove the periosteum, adipose tissue, and other

Figure 2. Radiographs of an 8-year-old boy with right tibial pseudarthrosis: (a) and (b) show the preoperative anteroposterior (AP) and lateral radiographs of the tibia; (c) and (d) show the postoperative AP and lateral radiographs; (e) and (f) show the 1-year postoperative AP and lateral radiographs indicating union of the tibia.

soft tissues around the pseudojoint. This alters the osteogenic microenvironment, representing the key element in the physiopathology of this disease.[4,22]

Many fixation techniques, such as intramedullary nailing[23] and the Ilizarov technique,[24] have been used to fix the tibia. The union rate of the Ilizarov technique varies between 32% and 85% (mean = 57%). A multicenter study concluded that the Ilizarov technique was the best technique for the treatment of CPT, owing to its ability to provide stable fixation and restore length.[4] However, the limitations included a high rate of recurrent fractures, axial deformities, and pin-site infections.[4,24] Intramedullary rod fixation is a more traditional treatment and the basis of various methods with a success rate in CPT ranging from 12% to 75% (mean = 40%). Some studies focused on modifying the intramedullary nailing technique—via transplanter intramedullary locking nailing[25] and "telescopic rods."[23] All surgeries in this study used Rush nails—which were stable, economical, and without pin-end infection—buried under the skin.

Although the optimal treatment for CPT remains controversial, there is consensus regarding some matters.[20,26] Irrespective of the technique used, realignment of the tibial segment and stable internal fixation are essential for union. Furthermore, intramedullary fixation is the best technique to maintain alignment during growth and prevent the risk of recurrent fracture, while leg-length discrepancies <5 cm can be treated by contralateral epiphysiodesis; for larger leg-length discrepancies, the Ilizarov technique is indicated. In this study, one patient exhibited nonunion of the bone after treatment; while the reason is unclear, this may have occurred due to the young age of the patient at the time of surgery.

This study has several limitations; first, this was a retrospective analysis study. To overcome the associated limitations, a multicenter or randomized study should be conducted in the future. Second, this study had a very small sample size; therefore, more patients should be enrolled in the future. Third, the follow-up period was short; hence, a future study with a longer follow-up duration is required to evaluate the outcomes. Finally, the mechanism of this method is not fully understood; thus, further study is necessary to elucidate the detailed process of this technique.

Conclusion

Using the gastrocnemius flap to cover the pseudarthrosis area appears to be an effective method to promote CPT union and can, therefore, be considered effective for the treatment of CPT.

Author contributions

G.N. and Y.S. conceived the study, participated in its design, and drafted the manuscript. W.C. and Y.S. helped in collecting the clinical data and help drafted the manuscript. All authors read and approved the final manuscript.

Declaration of conflicting interests

The author(s) declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

Funding

The author(s) received no financial support for the research, authorship, and/or publication of this article.

Ethical approval and consent to participate

The study was approved by the ethics committee of our hospital. Informed and signed consent was required from the parents or guardians before the operation.

Availability of data and material

The data sets used and/or analyzed during this study are available from the corresponding author on reasonable request.

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