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CASE REPORT

Case Reports

Lengthening of a Vascularized Free Fibular Graft

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Abstract Wide bone resection is sometimes necessary for bone tumors, and reconstruction is a major challenge. Vascularized fibular graft is one alternative but may result in progressive limb-length discrepancy (LLD) in children with substantial growth left. Progressive distraction lengthening with an external fixator is now a standard procedure to generally correct LLD. However, lengthening of free vascularized fibular grafts for lower limb reconstruction has not been reported frequently and then only in small series or case reports. We report our experience with three patients with lengthening after tibial reconstruction with a free vascularized fibular graft and review the literature.

Introduction

Diaphyseal bone resections may be indicated in the treatment of bone tumors or in congenital pseudarthrosis of the tibia (CPT) [6]. Since its description by Taylor et al. [40] in 1975, free vascularized fibular grafting (FVFG) has become a standard salvage procedure for reconstructing a segmental bone defect after diaphyseal bone resection for either bone tumor [10, 12, 13, 15, 16] or congenital pseudarthrosis [33, 38]. When tumor resection is performed in children, growth-plate sparing is not always possible owing to tumor involvement. This may result in progressive LLD [21, 27, 39].

Progressive bone lengthening (distraction osteogenesis) with an external fixator is a well-established technique to correct LLD that may be indicated in these two conditions [7, 18, 20]. Three cases of FVFG lengthening have been described [21, 29] for humeral malignant bone lesions and infections [21, 29, 39]. Four FVFG lengthenings in the lower limb have been described after surgical treatment of CPT [23, 25].

The purpose of our report is to provide some guiding principles for FVFG lengthening from published cases and from our experience with three cases after tibial diaphyseal reconstruction after tumor and CPT resection.

Case Reports

Patient 1

A 15-month-old girl was diagnosed with a chondroblastic osteosarcoma of the left tibia (Fig. 1A). She had a 10-cm diaphyseal resection of the left tibia sparing both of the proximal and distal growth plates (Fig. 1B). Reconstruction was performed with an ipsilateral vascularized fibular graft stabilized with a plate (Fig. 1C). She received no ancillary therapy. Ten years after surgery, she had multiple myxomas and endocrine overactivity develop. She then was diagnosed with a Carney complex syndrome

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Each author certifies that his or her institution has approved the reporting of these case reports, that all investigations were conducted in conformity with ethical principles of research, and that informed consent for participation in the study was obtained.

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Fig. 1A–E (**A**) The initial AP radiograph shows a chondroblastic osteosarcoma of the tibia. (**B**) An AP radiograph shows the resection of the tibia sparing the proximal and distal growth plates of the tibia. (**C**) A lateral radiograph shows the tibia after the FVFG stabilized with a plate. (**D**) The tibia is shown before lengthening. (**E**) Consolidation of the regenerate can be seen after progressive lengthening with an Ilizarov fixator.

[4, 5]. At that time, the initial diagnosis of chondroblastic osteosarcoma was reviewed histologically and judged rather to have been a chondromyxoid fibroma. At skeletal maturity (16 years), she had a 3-cm lower LLD (Fig. 1D). Because of her short stature resulting from hypercorticism, we started a lengthening program using an Ilizarov fixator. The fixator was mounted using three full rings fixed with two Kirschner wires each. The first ring was proximal to the osteotomy and two others were placed distally. The knee and foot were not included in the frame. An open osteotomy was performed at the level of the proximal metaphysis through the FVFG. The FVFG-derived bone appeared as normal metaphyseal bone and the procedure was not different from a standard osteotomy. The distraction was performed at a 1-mm per day distraction rate. We achieved equalization with an Ilizarov fixator in 1.5 months. No complications occurred during the lengthening procedure. Consolidation of the regenerate on AP and lateral radiographs was used to determine the time of device removal and was achieved 2 months after initiation of distraction. Full weightbearing without walking assistance was allowed with orthosis protection for 3 months. At 9 years followup, the patient had a normal gait and normal knee and ankle range of motion. No malalignment was observed on followup radiographs (Fig. 1E).

Patient 2

A 9-year-old boy presented with a 10-cm left lower LLD. At the age of 2, he had Ewing's sarcoma of the distal part of the left tibia develop. He was treated with chemotherapy and underwent 12-cm resection of the distal tibia involving the distal epiphysis. Reconstruction was performed with an ipsilateral vascularized fibular graft. Eleven centimeters of the proximal fibula with the proximal epiphysis was harvested and transferred. Talofibular arthrodesis was necessary to stabilize the ankle in a neutral position. The arthrodesis was performed between the fibula epiphysis and the talus to preserve the growth potential of the fibula proximal growth plate. This growth plate fused along with the arthrodesis within 3 months after the graft transfer. Osteosynthesis of the tibia and fibula shafts was achieved with wires. Progressive lower LLD developed. A gradual length correction program was started with a monolateral external frame (Orthofix, Verona, Italy) (Fig. 2A–B). The frame was mounted using three proximal pins placed in the native tibia and three distal pins in the FVFG. An open osteotomy was performed in the proximal metaphysis proximal to the FVFG. The FVFG-derived bone appeared as normal metaphyseal bone and the procedure was not different from a standard osteotomy. The distraction was performed at a 1-mm per day distraction rate. We achieved 7-cm lengthening in 2.5 months. Despite the talofibular arthrodesis, the patient had equinus of the hind foot develop during lengthening. Percutaneous Achilles tendon tenotomy was necessary to recover dorsiflexion of the foot and the lengthening was stopped for 10 days. Consolidation of the regenerate was achieved by 3 months after the end of distraction. Full weightbearing was allowed with orthosis protection and crutch assistance for 1 month. Two months after device removal, he fell and sustained a fracture through the regenerate. A cast was applied and the

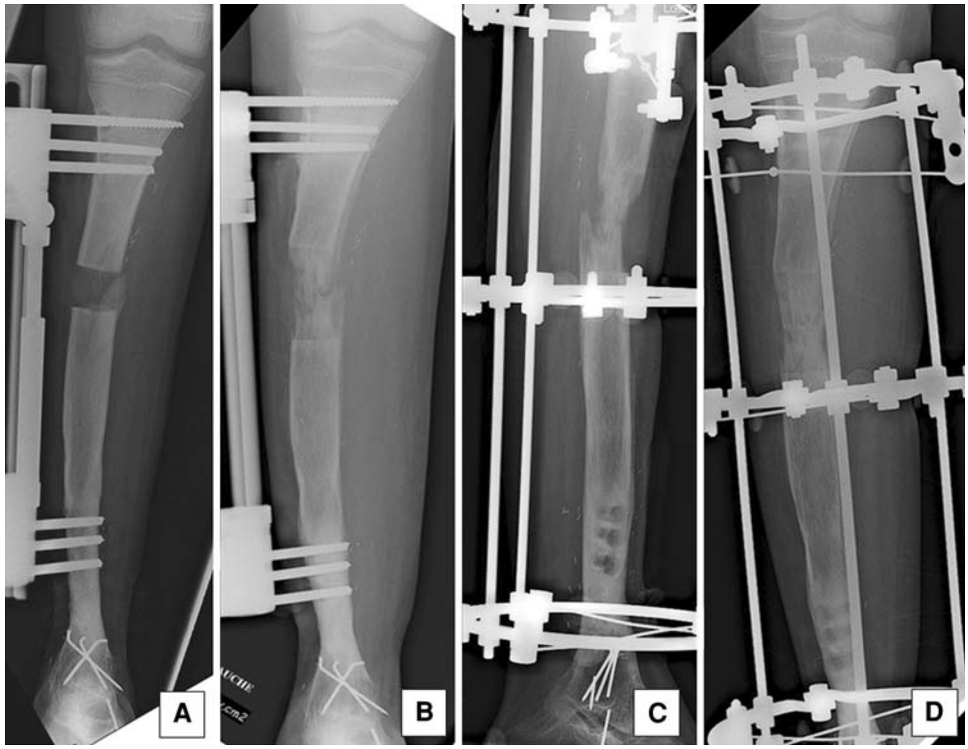

Fig. 2A–D Anteroposterior radiographs show the tibia during the (**A**) first gradual length correction program with a monolateral frame, and (**B**) in this illustration, with more advanced callus formation than in illustration A. (**C**) A radiograph shows the fracture though the regenerate. (**D**) The tibia is seen after stabilization of the regenerate with the Ilizarov fixator.

fracture healed in 3 months. A second gradual lengthening program was performed at the age of 15 years for persistent 3-cm LLD using a monolateral frame (Orthofix). The distraction was performed at a 1-mm per day distraction rate. We achieved 2.5-cm lengthening in 1 month. After 4 months, the external fixator was removed because of infection around the pins. The regenerate was stabilized with an Ilizarov fixator, but fracture through the regenerate occurred during the procedure (Fig. 2C). The infection was controlled with antibiotics given over 2 months. Consolidation of the regenerate finally was obtained after 3 months with the external device (Fig. 2D). Full weightbearing was allowed with a Sarmiento cast. The patient recovered a normal gait pattern 6 months after device removal. At 10 years followup, this patient was walking with an orthosis with no residual LLD. The radiographs of the tibia showed a residual valgus deformity of 18° and recurvatum of 5° (Fig. 3). The knee was stable with a knee flexion contracture of 10° and maximal flexion range of 130°. The ankle was stiff with residual equinus of 20°. No subsequent operation was planned.

Patient 3

A 12-year-old girl was treated for 11-cm right LLD. At the age of 2, she was diagnosed with congenital pseudarthrosis of the tibia (Fig. 4A). She was treated unsuccessfully by resection of the pseudarthrosis and cortical bone grafting. She had a 9-cm diaphyseal resection and the reconstruction was performed with an 11-cm contralateral vascularized fibular graft. The graft was anchored in the proximal and distal tibial fragments and stabilized with a monolateral external fixator (Fig. 4B). The graft union failed distally with a valgus deformity of the distal tibia. The graft nonunion and deformity were treated successfully with a cancellous bone graft associated with a talofibular arthrodesis. The ankle was fused in a neutral position. A progressive LLD of 11 cm developed during the subsequent years (Fig. 4C). At the age of 12 years, a gradual lengthening correction program was started with an Ilizarov external fixator. The fixator was mounted using three full rings fixed with two Kirschner wires each. The first ring was placed proximal to the osteotomy and the two others distally. An open osteotomy was performed in the proximal metaphysis of the tibia above the FVFG. The knee and foot were not included in the frame. The distraction was performed at a 1-mm per day distraction rate. We achieved 10-cm lengthening in 3 months (Fig. 4D). No complications occurred during the lengthening procedure. Complete consolidation as determined by radiographs and CT scans of the regenerate was achieved by 6 months after initiation of distraction. Full weightbearing was allowed with orthosis protection and crutch assistance for 1 month. She recovered a normal gait and was free of the orthosis 8 months after device removal. At last followup (11 years), there was residual 2-cm LLD, and the knee was stable with a flexion contracture of 10° and maximal flexion range of

Fig. 3A–B (**A**) An anteroposterior radiograph shows a residual valgus of 18°, and (**B**) the lateral radiograph shows a recurvatum of 5°.

140°. The radiographs showed a tibia valgus of 5° but no residual deformity on the lateral views (Fig. 5). The hind foot had a residual valgus of 10° and equinus of 20°.

Discussion

Surgical management of large bone defects remains a challenge in the treatment of bone tumors, traumatic bone defects, and CPT in children. In CPT, resection of the pseudarthrosis followed by reconstruction remains the gold standard for this condition [38].

Depending on the surgeon's preference and the location of the defect, different techniques have been described [3, 11, 13, 25, 32, 33, 41]. Autografts, allografts, induced membrane, and prostheses for joint arthroplasties are among these procedures. Free fibular grafting is one of the standard salvage procedures for reconstructing a segmental bone defect [15, 16]. FVFG is an autologous vascularized graft and is considered by some to be the ideal implant after diaphyseal resection in children [15, 16, 37]. In skeletally immature patients, preservation of the growth plates is not always possible, particularly in bone tumor resection. Therefore, growing patients may need additional lengthening procedures to equalize LLD after growth plate sacrifice or in the case of insufficient growth in congenital disorders.

FVFG is associated with a high rate of complications [2, 31, 43]. Common complications of FVFG are mechanical failure (range, 23%–24%) [2, 26], delayed union (range 5%–23%) [15, 31, 41], thrombosis (1%) [2], infection

Fig. 4A–D (**A**) An initial radiograph shows the congenital pseudarthrosis of the tibia and fibula. (**B**) Reconstruction of the tibia with a FVFG was done. (**C**) The tibia is shown before lengthening. (**D**) Consolidation of the regenerate is evident after progressive lengthening with an Ilizarov fixator.

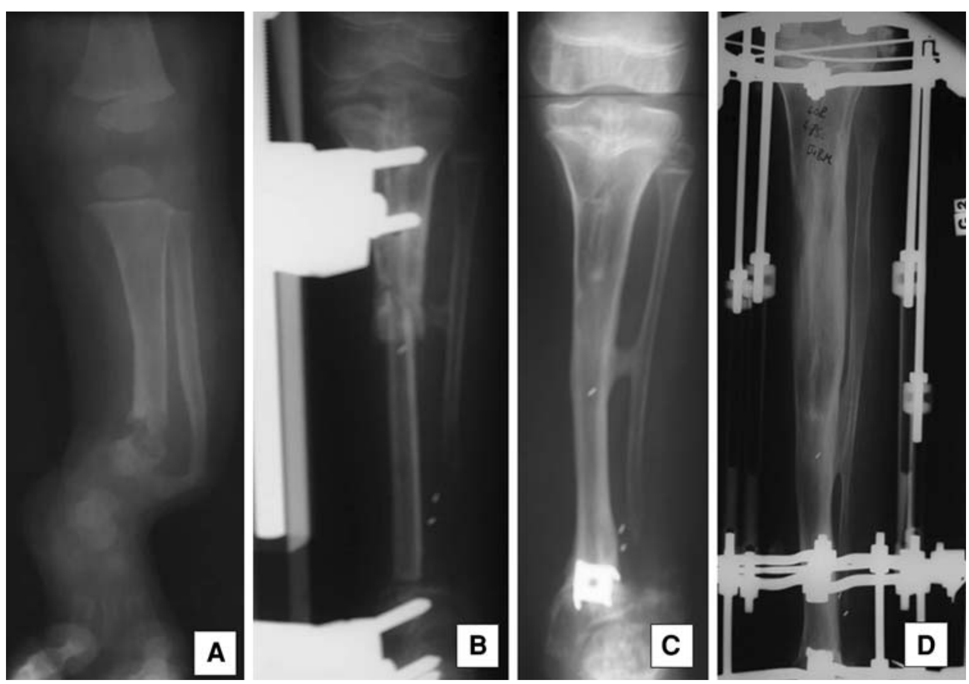

Fig. 5A–B After frame removal, (**A**) an AP radiograph shows a residual valgus of 5°. (**B**) No malalignment can be seen on the lateral radiograph.

(10%) [43], or associated complications at the donor site (range, 8%–20%) [14]. A combination of massive allograft and FVFG seems to decrease the early postoperative fracture rate [15, 35].

Delayed complications such as LLD have not been reported frequently (Table 1), although some cases of LLD after FVFG for traumatic bone defects have been reported [30, 37]. In these cases, the LLD developed because of traumatic epiphysiodesis or multiple surgical procedures. LLD is a rare complication after FVFG performed after tumor resection in children [15, 36]. When they occur, LLD after tumor resection (range, 1–3 cm) may not require a subsequent lengthening procedure or contralateral epiphysiodesis [3, 10]. In other circumstances, more substantial LLD (7–8 cm) after bone tumor resection may require a lengthening procedure over the FVFG [21, 39]. CPT reconstruction with FVFG also provides delayed complications such as LLD or axial deformities [38]. Four cases of lengthening procedures performed after CPT reconstruction with FVFG have been reported [23, 25]. A lengthening procedure after FVFG is rare.

There are no guidelines for a lengthening procedure of a FVFG, but progressive fibular lengthening, as a single weightbearing bone, has been described after congenital tibial deficiency (CTD) reconstruction [42]. Lengthening procedures in Kalamchi Types 1 and 2 [24] CTD must be distinguished. In Type 1, the osteotomy is performed in the centralized fibula [42], whereas in Type 2, it is performed in the persistent proximal tibia [17, 22]. These two different types of reconstructions of CTD are equivalent to partial or complete tibial defect reconstruction with FVFG. However, fibula centralization in CTD does not involve a microsurgical procedure and has the specific problem of knee instability. LLD in CTD is always important. The reported amounts lengthened ranged from 2 to 15 cm [17, 22, 42], with callus distraction initiated at a rate of 0.75 mm per day [42] or 1 mm per day [17]. The healing index was approximately 1 cm per month in all cases. The various authors encountered no major complications and bone consolidation was completed in all cases.

Therapeutic options for LLD after FVFG depend on the importance of the discrepancy. Nonoperative management or contralateral epiphysiodesis in small LLD seems a safe and reasonable option. In Patient 1, a contralateral epiphysiodesis would have been an acceptable alternative, but it

Table 1. Summary of published series

Study	Number of patients	Age at FVFG (years)	Indication	Topography	LLD (cm)	Lengthening procedure after FVFG
Jupiter et al. [23]	1 (case report)	18	CPT	Lower limb	8	Lengthening with Ilizarov fixator
Kanaya et al. [25]	3	4.9 (range, 2–12)	CPT	Lower limb	4.9	Lengthening with Ilizarov fixator
Lim et al. [29]	1 (case report)	1	Neonatal osteomyelitis	Humerus	6	Lengthening with Ilizarov fixator
Ilizarov et al. [21]	1 (case report)	13	Tumor resection	Humerus	7	Lengthening with monolateral frame
Shaw et al. [39]	1 (case report)		Tumor resection	Humerus	8	Lengthening with monolateral frame

FVFG = free vascularized fibular grafting; LLD = limb-length discrepancy; CPT = congenital pseudarthrosis of the tibia.

was not accepted by the patient. The lengthening procedure was performed for aesthetic purposes taking into account the patient's very short stature. For LLD greater than 4 to 5 cm, a lengthening procedure should be discussed. After a FVFG reconstruction, lengthening procedures can only be attempted using external devices such as monolateral or circular frames. We do not have the experience of using an internal lengthening nail in these cases.

Our prerequisite for lengthening was fibular graft hypertrophy evaluated with standard radiographs. The main issue is to appreciate the proper time to perform lengthening on a transferred bone. The mean age of our patients at surgery was 12 years (range, 9–15 years), and the mean interval between the end of reconstruction and lengthening was 10 years (range, 7–13 years). FVFG hypertrophy has been well studied. De Boer and Wood [8] reported 80% hypertrophy (as a percentage increase in diameter) of the graft at 2 years' followup. For El-Gammal et al. [9], there was no increase of hypertrophy 3 years after FVFG. In their experience, hypertrophy was magnified in mechanically loaded extremities and younger patients. Hypertrophy increased faster in their patients who received chemotherapy compared with those who did not but declined afterward to end at the same rate. The distraction program must be performed when maximum hypertrophy of the fibula has been achieved. Three years after the FVFG seems to be a safe delay.

We used the same lengthening protocol as that for native bone [7]. We usually perform two consecutive, short lengthening procedures to prevent complications. In Patient 3, we performed a one-step 10-cm lengthening, but it now is a rare situation. The therapeutic strategy now would consist either of two consecutive lengthening procedures of 5 cm each or one lengthening of 6 to 7 cm followed by a contralateral epiphysiodesis of the tibia. In Patient 2, a contralateral epiphysiodesis would have been a safer alternative to a second lengthening, but the growth potential of the proximal tibia was not sufficient at that time. We arbitrarily used either an Ilizarov fixator or a monolateral frame and achieved the desired distraction without joint instability. The knee was intact and stable before the distraction and the ankle previously was stabilized with arthrodesis in Patients 2 and 3. For these reasons, the knee and foot were not included in the frame, but range of motion was maintained with daily physiotherapy. In Patients 2 and 3, the lengthening procedure resulted in satisfactory equalization of limb length, but both patients had a 10°-knee contracture develop. Despite the ankle arthrodesis, a hind foot equinus deformity developed that could have been prevented with inclusion of the foot in the frame. An Achilles lengthening procedure was performed in Patient 2 but without major improvement of ankle dorsiflexion. We performed no subsequent Achilles lengthening or subtalar joint arthrodesis to correct the equinus but still obtained a good functional outcome. However, equinus at the subtalar level can progress to painful osteoarthritis of the subtalar joint, which may lead to an arthrodesis. In Patient 3, the equinus deformity compensated the residual 2-cm LLD. Compared with nontransferred bone lengthening, we found no difference in the healing index of the regenerate.

In all three of our patients, the osteotomy site was located at the proximal half of the bone. The osteotomy was performed in the FVFG (Case 1) or in the native bone proximally to the transferred fibula (Patients 2 and 3). Jupiter et al. [23] performed two osteotomies for angular correction, one in the fibula graft and one in the native bone. Shaw et al. [39], Lim et al. [29], and Ilizarov et al. [21] performed the osteotomy in the remodeled fibula graft after humeral reconstructions. Kanaya et al. [25] provided no details of their procedure, but they reported malalignment of the ankle and severe frontal deformity in all cases after lengthening. Bone consolidation was always achieved, whatever the site of osteotomy.

As established by Ilizarov, we used a distraction rate of 1 mm per day [19]. As suggested, a distraction rate of 0.75 or 1 mm per day is appropriate for CPT [42], however Ilizarov et al. [21] recommended a lower distraction rate (0.5 mm per day) for humeral lengthening to obtain good consolidation of the regenerate. In our patients, the average healing index was 25 days per cm (range, 21–25 days/cm), which is similar to published results [1, 21, 34]. FVFG lengthening was not associated with a higher healing index than standard long bone lengthening, in our experience.

In all series, the complication rates were low. The complications are similar to those encountered with any distraction procedure [34]. We did not experience specific complications when performing lengthening over FVFG.

In case reports involving the upper limb, the main complication was delayed consolidation after a fracture through the regenerate. We observed a similar complication in Patient 2. Pin tract infection is a complication of lengthening with an external fixator and usually is treated with local care and antibiotics. In Patient 2, device removal was necessary because of bone suppuration. The second fracture through the regenerate could have been avoided if the changing procedure had been performed on a traction table.

Residual deformities were observed at the end of the lengthening. Frontal plane deformity is a common complication of lengthening with a monolateral frame [28]. In Patient 2, the tibia deformity of 18° valgus and 5° recurvatum need subsequent correction using an Ilizarov fixator to improve the mechanical stress in the lateral compartment of the knee. The osteotomy will be placed in

the middiaphysis and hinges at the same level. The deformity could have been corrected with application of hinges on the Ilizarov fixator when the fracture through the regenerate occurred, but the fracture was only diagnosed on postoperative radiographs. In Patient 3, the valgus deformity was insignificant and no correction is planned. In our experience, the valgus deformity is decreased by use of a circular external fixator.

Wide bone resection is sometimes necessary in specific pediatric orthopaedic conditions such as CPT or bone tumors. Reconstruction is the main challenge in this situation and transferred vascularized fibular graft is a relevant procedure. After a review of the literature and of our cases, it is difficult to provide formal guidelines for lengthening procedures after a FVFG, but we highlight some key points.

FVFG reconstruction after CPT resection seems complicated more frequently by considerable LLD (greater than 5 cm) than reconstruction after bone tumor resection. After FVFG reconstruction, the minimum delay before secondary lengthening is 3 years. In our experience, the lengthening protocol should be similar to that for a native bone procedure in lower limbs, but the basic principles of progressive lengthening must be followed carefully.

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