CASE REPORT

Lateral Malleolus Reconstruction After Tumor Resection in Children: A Case Report and Literature Review

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Abstract

Background: Pediatric reconstruction of lateral malleolus was necessary and challengeable. Up to now, vascularized fibular was the optimal graft to reconstruct epiphyseal defection. However, the sophisticated microvascular operation has limited the wide application of this technique.

Case Presentation: We present the case of a 9-year-old boy with Ewing sarcoma in left distal fibula. In order to restore the growth capacity, we used reverse-flow vascularized fibular epiphyseal graft with tibialis anterior artery to reconstruct the bone defect after tumor resection with no microvascular anastomosis. More than 4 years after the operation and adjuvant chemotherapy, the patient was free of pain and recurrence, and the function and stability of ankle joint was perfect. Radiology examination revealed satisfied bony union of fibula and normal growth of the fibular head transplant.

Conclusions: The advantage of reverse-flow vascularized fibular epiphyseal graft is requiring no microvascular anastomosis which could not only shorten operating time, but also reduce factitious damage of vessels. This report presented that this technique might be an available option for reconstruction of lateral malleolus in children.

Key words: Epiphysis transfer; Lateral malleolus reconstruction; Proximal fibula; Reverse-flow; Tumor resection

Introduction

Severe trauma and wide malignant tumor resection of distal fibula was the major cause of lateral malleolus defect[1]. Multiple biomedical studies have shown that ankle stability depends on the integrity of the lateral malleolus including fibula, the proximal and distal syndesmotic ligaments, and interosseus membrane[2]. Therefore, reconstruction of distal fibula was indispensable to prevent ankle deformity and abnormal gait. Additionally, in children and adolescents, development depending on growth plate complex was equally important as ankle stability. Growth plate complex, specific structure in immature skeleton, consists of physis, epiphysis, and distal metaphysis[3]. It was commonly located on the terminals of long bone and the border of flat bone. Epiphyseal cartilage proliferation was the origin of bone growth while cartilage ossified constantly until epiphysis closed. Therefore, pediatric growth plate injury could result in growth disturbances, which take its course to late angular deformity and limb length inequalities[4]. Epiphyseal injury on the distal fibula was prevalent in the lateral malleolus defect, which was challenging and necessary to reconstruct. Consequently, vascularized epiphysis transfer has emerged to restore the joint function and the longitudinal growth capacity simultaneously[5].

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In this study, we reported a 9-year-old male with Ewing sarcoma in distal fibula, which was treated by wide resection after six-cycle neoadjuvant chemotherapies and reconstructed with reverse transfer of the vascularized proximal fibula attached with tibialis anterior artery. The prognosis was excellent and without serious complications. Furthermore, given the rarity reports of this method, we systematically reviewed and compared the advantages and disadvantages of the various methods in pediatric lateral malleolus reconstruction.

Case Report

History and Preoperative Preparation

A 9-year-old male was admitted in our hospital on November 15, 2015, diagnosed as Ewing sarcoma in left distal fibula *via* incisional biopsy (Figure 1A, B). The patient presented with pain disappearance and mass regression after six-cycle neoadjuvant chemotherapy using ifosfamide and etoposide alternating with vincristine, doxorubicin, and cyclophosphamide. Radiographic examinations revealed osteolytic disease in the distal fibula and with a soft tissue mass as 2.5 × 8.0 cm approximately. The lesion was located above the epiphyseal line and required wide resection with the distal fibular tip (Figure 1C, D).

Surgical Procedure

The patient was placed supine under general anesthesia, with full exposure of left lower extremity and a pneumatic tourniquet. A straight longitudinal incision was centered over the lateral crus to expose the lateral facet of the fibula and a wide lesion resection was performed. Notably, we took the surgical incision nearby the biopsy incision in order to remove the scar attached to tumor together.

Prolonging the incision proximally to expose the proximal fibula. The common peroneal nerve was dissected meticulously and protected by a rubber loop to prevent inadvertent injury during surgery. The tibialis anterior vessels and deep peroneal nerve were exposed to confirm that the nutrient vessels of fibular head were from tibialis anterior artery. A sufficient cuff of muscles was left around the fibula to preserve the nutritional branch while the fibula was transected with a perfect length. The tibialis anterior vessels were ligatured at the origin and prepared for transplantation (Figure 2A–C).

Tourniquet was released to confirm the bleeding of the muscular cuff and fibula and then reapplied. Thereafter, the

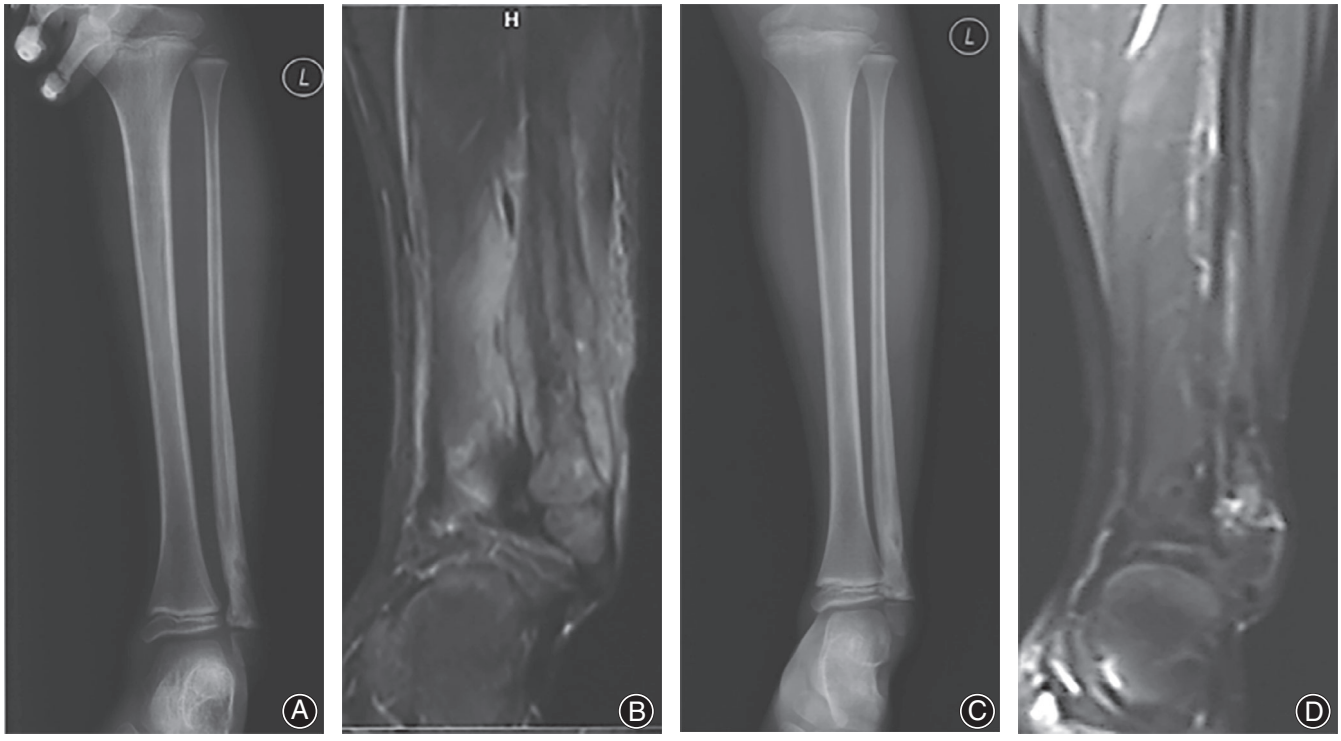

Fig. 1 Preoperative radiographic examination of patient. (A) X-ray before neoadjuvant chemotherapy in August 2015. This revealed an osteolytic disease with periosteal reaction in the distal fibula, which was diagnosed as Ewing sarcoma. (B) MRI showed osteolytic disease in the distal fibula with a soft tissue mass before neoadjuvant chemotherapy in August 2015. (C) X-ray revealed an increase in ossification which suggested a good response to chemotherapy in December 2015. (D) MRI showed tumor regression and the tumor size was 2.5 × 8.0 cm approximately surrounding the distal fibula after chemotherapy in December 2015

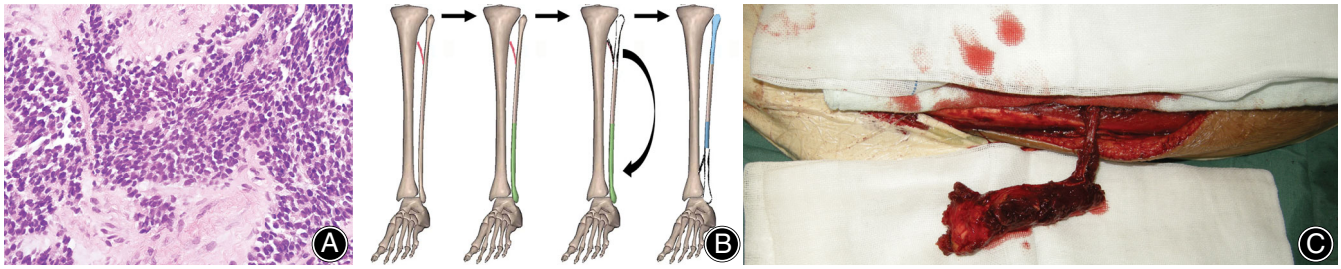

Fig. 2 (A) Hematoxylin–Eosin stain of the tumor specimen was consistent with the preoperative diagnosis as Ewing sarcoma. (B) Diagram of reverse-flow vascularized fibular epiphyseal graft. The proximal fibula (white part) was removed to repair the bony defect (green part) after tumor resection. Allografts (blue part) were used to replace the donor site and connect the remaining fibula and vascularized graft. (C) Intraoperative view of reverse-flow vascularized fibular epiphyseal graft. The tibialis anterior vessels were harvested with the proximal fibula attached to sufficient muscle

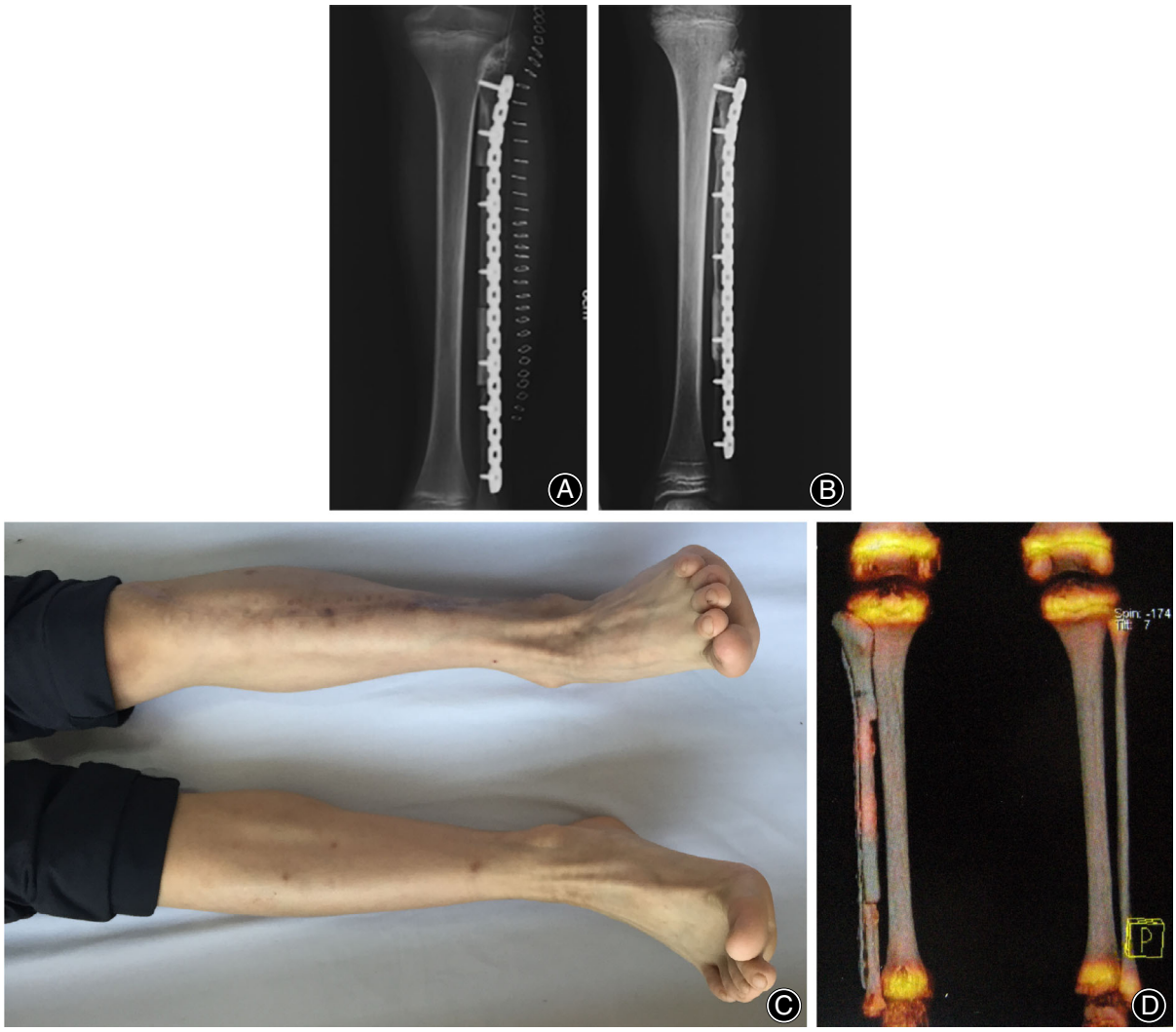

Fig. 3 Radiographic examination of patient at 4 years after operation. The patient was free of pain and recurrence, and the function and stability of ankle joint was perfect. (A) X-ray after reconstruction revealed that all segments were fixed with an osteosynthesis metallic fibular plate. (B) X-ray of satisfied bony union of fibula and normal growth of the fibular head transplant compared with the other side. (C) Ankle dorsiflexion of the injured limb was excellent as the other side. (D) The back view of SPECT/CT showing high nuclide absorption in the epiphysis of fibular head revealed the survival of graft

vascularized proximal fibular graft was reversed to repair the bony defect and replace lateral malleolus with fibular head. An allograft stored in our bone bank was prepared and bifurcated. One part was used for replacement of the donor site, and the other was inserted between the remaining fibula and vascularized graft to maintain the continuity of fibula (Figure 3A). Under a satisfied restoration, all segments were fixed with an osteosynthesis metallic fibular plate. An above-knee plaster cast was applied postoperatively for several weeks depending on follow-up.

Follow-up

The patient had suffered foot drop post operation immediately, which might have been caused by compression of the peroneal nerve, and was restorable gradually. More than 4 years after the operation and adjuvant chemotherapy, the patient was free of pain and recurrence. For the reconstructed ankle joint, the passive valgus and varus movement was stable, and the active functions were comparable to that of the normal side with the Kofoed Ankle Score of 100. Additionally, the appearance of the left knee was intact as per preoperation and the Knee Society Score presented an excellent function (Figure 3B). X-ray showed satisfactory bony union of fibula and normal growth of the fibular head transplant compared with the other side (Figure 3C). SPECT/CT showed high nuclide absorption in the epiphysis of fibular head and revealed survival of graft (Figure 3D).

Discussion

Several studies have documented that the absence of lateral malleolus could lead to lateral displacement of the talus and valgus instability, which required appropriate techniques to reconstruct the distal fibula[6]. Ligamentoplasty, as suturing of distal peroneal tendons to lateral side of tibia, has been performed by Capanna *et al.* and showed satisfactory prognosis with no pain but reduced mobility and slight joint instability[7]. In a case series, Jamshidi *et al.* suggested that reconstruction of distal fibula with osteoarticular allograft after tumor resection could achieve bony union with the mean time of 12 months and full ankle function except asymptomatic valgus deformity in one patient[8]. Additionally, primary ankle arthrodesis and prosthetic ankle replacement were feasible in limb salvage for treatment of distal intraarticular tibial tumors[9,10]. Nevertheless, these techniques were not available in pediatric reconstruction.

On account of the particularity in children, reconstruction was required to restore the joint function and the longitudinal growth capacity of the transferred segment simultaneously. Necessarily, it's paramount to repair the growth plate complex *via* vascularized epiphysis transferred with a portion of diaphysis. Three sites are suitable for epiphysis transfer: the iliac crest, the distal portion of the scapula, and the proximal fibula[11]. However, the iliac crest and the distal portion of the scapula are lined with hyaline cartilage and lack of articular surface, which have limited the function and raised uncertain resistance to transfer. Therefore, variable diaphysis segment and similar structure morphology made proximal fibula become the most perfect graft in pediatric lateral malleolus reconstruction compared with other donor sites.

As early as in 1905, Huntington assumed that vascularized fibular graft, fibula with its own nutrient supply, could form satisfied bony union and restore weightbearing[12]. Benefited from microvascular anastomotic in free bone graft, this technique was verified in essential experimental research since 1970s[13]. Since then, the vascularized fibular epiphyseal graft (VFEG) had been promoted and rapidly applied to pediatric reconstruction of mandible, long bones in extremities, and pelvis in the past 20 years[14]. Considering the rarity of distal fibular defection, only a few reports using VFEG in immature patients have been published. In long-term follow-up, radiographic examination revealed that VFEG could provide not only good bony stability, but also excellent growth potential. However, the critical part step in the VFEG was sophisticated microvascular techniques which extremely limited the wide application of VFEG and survival of graft[15].

The reverse-flow VFEG operative technique used in this patient is a one-stage procedure requiring no microvascular anastomosis which cannot only shorten operating time, but also reduce factitious damage of vessels[16]. However, it was controversial as to which artery was chosen to nourish graft. The arterial blood supply for the fibular head consisted of lateral inferior geniculate artery or the tibialis anterior artery nourishing the upper part of epiphysis and peroneal artery supplied metaphysis and diaphysis. Disappointedly, peroneal artery was concluded to be insufficient to support the physeal growth plate, which could cause limb shortening. The tibialis anterior artery was firstly used and recommended to supply epiphysis by Taylor[17], and had been confirmed *via* further experimental and clinical studies. On the other hand, Yang *et al.* recommended to use inferior lateral genicular artery to supply the graft in the reconstruction of type III radial longitudinal deficiency[18]. To assist surgeons in choosing the optimal pedicle, Morsy *et al.* performed an anatomical and high-resolution computed tomographic angiography study to describe the detailed vascular anatomy of the proximal fibula[19]. Based on research of 28 lower extremities, the authors suggested that both arteries could be used successfully as the pedicle for VFEG, although the tibialis anterior artery would be preferred in cases where a longer length of the diaphysis or pedicle was required. In our experience, the most important thing is confirming the nutrient vessels of fibular head. Then in this case, choosing tibialis anterior artery was on account of length at the bony defect and the minimum injury for peroneal nerve. SPECT/CT revealed that the fibular graft was surviving and normally growing compared with the other side, which verified the feasibility of reverse-flow VFEG.

The complications in this case were similar as reported. Peroneal palsy was the most common complications in tibialis anterior artery flaps on account of their intimate location between the motor branch of the peroneal nerve and the tibialis anterior vascular bundle at the fibular

head. Ben Amotz *et al.*[20] demonstrated that the majority of patients had temporary motor dysfunction of peroneal nerve and only 2.6% were permanent. Additionally, the complications in donor site, mainly ankle valgus and proximal migration of the lateral malleolus, could also result in unbalanced force of tibia and instability of ankle joint, which could be prevented by reconstruction of proximal tibia with tibiofibular syndesmotic screw, tibial autograft, prosthesis or artificial bone graft[21–23]. In our study, peroneal palsy and foot drop occurred temporarily and were self-restoring, and no associated complication occurred in donor site after reconstruction with an allograft proactively. We recommended to use SPECT/CT to evaluate the survival of vascularized fibula graft and allograft at different stages.

In our experience, reverse-flow VFEG has provided a viable option to reconstruct lateral malleolus after wild resection or severe trauma. Due to requiring no microvascular anastomosis, it could reduce operative risk and shorten the operative time with restorable complications.

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