

## Case Report

# A Novel Technique of Vascularised Fibular Epiphyseal Transfer Combined with Cryosterilized Autograft and 3D printing in a 2-year-Old Child with Ewing's sarcoma: A case report

### ABSTRACT

Vascularized fibular epiphyseal transfer (VFET) though technically challenging, is a technique which can be used in skeletally immature patients to preserve the limb length in tumors near physis. As limb salvage at a young age adds to the complexity of the procedure since the growth potential of the limb has to be considered to restore the optimal function of the salvaged limb. This case report documents the treatment of an Ewing's Sarcoma of the left proximal tibia in a 2-year-old boy, following VFET modified with free freezing liquid nitrogen and three-dimensional printing. We succeeded in improving the patient's oncological and functional outcome because our patient was mobilized early and no postoperative complications occurred. This technique can thus be considered for the biological reconstruction of very young children as it has the least influence on limb lengths and growth potential.

**Keywords:** Ewing's sarcoma, free freezing liquid nitrogen, vascularized fibular epiphyseal transfer

### INTRODUCTION

The procedure of salvaging a limb at a young age is more complex since the growth potential of the limb needs to be considered to ensure that it is restored to its optimal function. In the last 40 years, vascularized fibular graft has been used widely in tumor surgery for biological reconstruction, first described in the mid-1970s.<sup>[1]</sup>

Nonmicrosurgical techniques are of limited use in the pediatric population as they can lead to the obliteration of the epiphyseal plate resulting in progressive limb deformity, and limb length discrepancy.<sup>[2]</sup> In pediatric cases, vascularized transplant of the proximal fibula with its open physis allows for an adequate reconstruction for the bone loss and the possibility of conserving the growth potential.<sup>[3]</sup>

Vascularized proximal fibular epiphysis and a part of the proximal diaphysis have been used in children as effective

biological alternatives to reconstruct the radius, proximal humerus, and proximal femur.<sup>[4-9]</sup>

This paper details a novel approach to surgical treatment of Ewing's sarcoma of the proximal tibia in a 2-year-old with physeal involvement using this principle. We transplanted vascularized proximal fibular physis into the paired proximal tibial frozen autograft here so that the limb could retain its potential growth.

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## CASE REPORT

A 2-year-old child was referred to our center after an open biopsy done in a nononcology center, with a history of swelling over left proximal part of leg following a trivial trauma. On examination, the patient had left proximal leg swelling and tenderness with a restricted and painful range of motion of knee, a 4 cm linear scar over the lateral aspect of proximal leg, with no distal neurovascular deficit. A radiograph of the left knee [Figure 1] showed an osteolytic lesion over the metaphyseodiaphyseal region of the proximal tibia. Magnetic resonance imaging (MRI) of the left joint revealed a homogeneously expansile osteolytic lesion within the upper metaphyseal region of the tibia with soft tissue extension and extension into the physis [Figure 2]. The lesion was sparing the epiphysis and the differential radiological diagnoses were round cell tumor/osteosarcoma. Positron emission tomography scan showed hypermetabolic activity at the left upper tibia measuring 32 mm × 28 mm for a length of about 42 mm with no signs of skip or distant metastasis, suggestive of primary osseous neoplasm. Open biopsy/curettage done from the referred center was reported as A1A2 section showing bony trabeculae with an infiltrating tumor, which is cellular and is composed of small round cell with occasional pseudorosettes. Tumor cells were immunopositive for CD99, Vimentin, MIC2 and focally positive for S100, BCL2, CD117. Hence, immunohistochemistry also favored a diagnosis of Ewing's sarcoma. Translocation studies: Negative for *EWS/FLI1*, t (21;22) and t(7;22) which suggested an atypical Ewing's sarcoma. Six cycles of neoadjuvant chemotherapy (VAC-IE) were given. Postneoadjuvant chemotherapy and MRI (3 TESLA) were performed which revealed significant interval regression of the lesion. A 1:1 three-dimensional (3D)-printed model of the tibia and fibula were used in preoperative planning.



Figure 1: Plain X-ray showing lytic lesion in the proximal femur

The patient was planned for free liquid nitrogen frozen autograft and proximal fibula pedicled physeal transplant.

## Surgical technique

Patient in the supine position, after painting and draping left lower limb under appropriate antibiotics coverage, a single incision spanning the knee and leg anteromedially was made involving the previous biopsy scar [Figure 3]. Soft tissue dissected and tibia isolated, wide marginal resection of the tumor was done with supraphyseal cut, preserving as much of the epiphyseal bone and articular cartilage as possible [Figures 4 and 5]. Distal diaphyseal and proximal osteotomy was stabilized by 2 Kirschner wires.

Processing method for Liquid nitrogen treatment of disease-affected bone: The standard free-freezing method was performed. Disease-affected bone was completely extracorporealized [Figure 6], immersed into Liquid Nitrogen for 20 min [Figure 7], then at room temperature for 15 min followed by 10 min in distilled water as a part of thawing to kill the tumor cells.<sup>[10]</sup>

*Dissecting the recurrent branch of the anterior tibia artery:* The main aim of this step is to harvest proximal epiphysis and a variable amount of diaphysis of the fibula with the anterior tibial vessels as a vascular pedicle.<sup>[11,12]</sup> This artery supplies the epiphysis through a recurrent epiphyseal branch and proximal two-thirds of the diaphysis through musculoperiosteal branches [Figure 8], which is preserved during the dissection. The fibula graft is exposed by detaching extensor digitorum longus muscle, together with peroneus longus muscle, at its proximal insertion at the level of the emergence of peroneal nerve into the anterior compartment of the leg. The common peroneal nerve is identified and dissected at the neck of the fibula. The proximal cuff of muscle must be left attached to the fibular head as it contains the recurrent epiphyseal branch of the anterior tibial artery on which this transfer is based.<sup>[11,12]</sup>

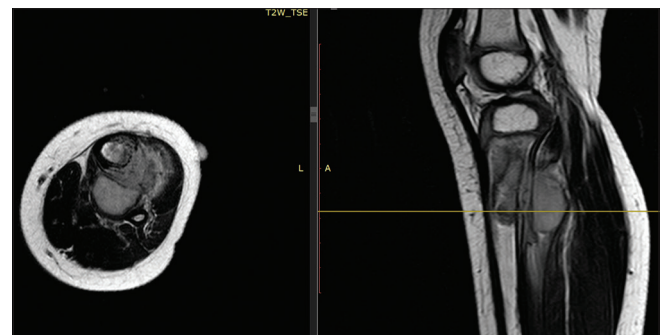
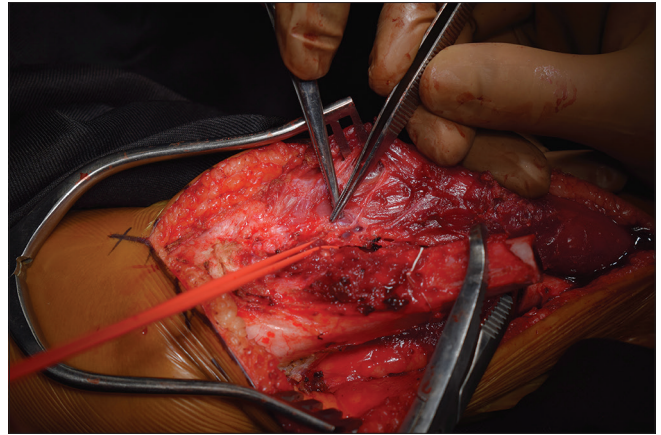


Figure 2: The MRI (3T) showing a homogeneously expansile osteolytic lesion in the upper metaphyseal area of the tibia, extending into soft tissue and the physis while sparing the epiphysis

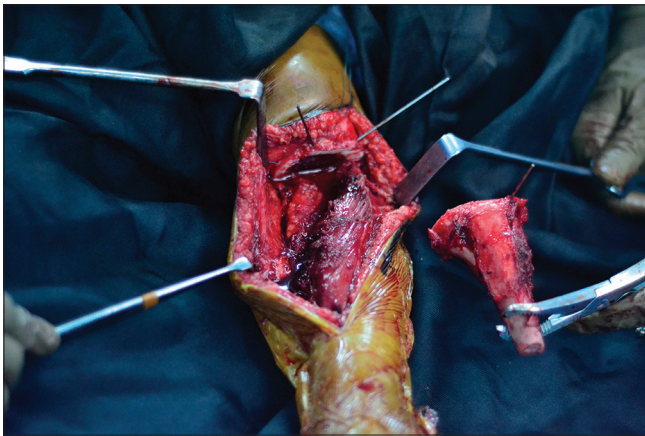




**Figure 3:** Skin incision planning, incorporating the prior surgery scar and bone landmark



**Figure 4:** Dissection of the neurovascular bundles in the anterior compartment



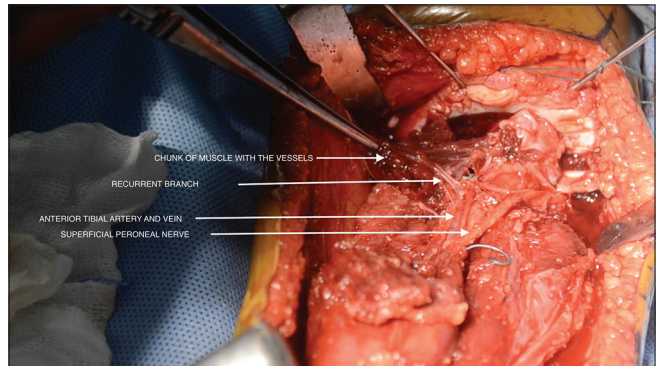
**Figure 5:** En bloc removal of the diseased segment of tibia



**Figure 6:** Preparation of the excised segment for liquid nitrogen treatment



**Figure 7:** Liquid nitrogen poured to the container used for immersion of disease-affected bone



**Figure 8:** Picture depicting the vascular supply to the fibula being preserved during transposition

Dissection of the neurovascular bundle is carried out from distal to proximal because in the distal part only occasional motor branches of peroneal nerve surround the anterior tibial vascular pedicle, and its dissection is more manageable at this level. The proximal dissection of the pedicle is carried

out until the origin of the anterior tibial artery is identified. It becomes tedious as we go proximally where many motor branches cross the recurrent epiphyseal branch of the anterior tibial artery. In this case, we were able to separate the motor branches from the vessels as it was not crossing over each other closely. While dissecting the diaphyseal region, the interosseous membrane and a longitudinal strip of muscle were harvested for preserving maximum possible periosteal branches.



The tibial autograft was then placed in its location after making a customized slot with the aid of 3D-printed fibula to accommodate the transposed native fibula [Figure 9]. The harvested native fibula was transposed into this slot and fixed proximally with the help of K wires. The distal end was inserted into the medullary canal of the tibia and fixed with a medial 2.5 mm T plate. The lateral collateral ligament was repaired using a suture anchor [Figure 10]. The stability of the construct was confirmed under the image intensifier and the wound was closed in layers.

Frozen sections from the surrounding tissue of resected specimen were negative for malignant cells. Postoperative histopathological examination revealed residual viable Ewing's sarcoma, with adjacent stroma showing therapy-related changes with tumor regression score-picci system grade 1.

The postoperative period was uneventful. Knee was immobilized in a POP (plaster of paris) slab. Started on nonweight bearing mobilization POD1 (postoperative Day). Weight-bearing was delayed by 2 months. There was no local wound complications and started her on adjuvant chemotherapy as soon as the wound healed.

Follow-up: There was no limb length discrepancy and the patient's condition was clinically and radiologically satisfactory till the last follow-up. She was walking with slab support and satisfactory knee bending. The patient was given physiotherapy at home by our home care team. But unfortunately, after 3 months' postoperative while on adjuvant chemotherapy, the patient developed sepsis and succumbed to death.

## DISCUSSION

Biological reconstruction of proximal tibia following oncological resection in a 2-year-old is challenging. In this case, we chose

the technique of vascularized fibular epiphyseal transfer (VFET) as it confers the ability to restore both joint function and axial limb growth. Other options include Rotationplasty, expandable prosthesis<sup>[13]</sup> and amputation. Rotationplasty first described in 1930 by Borggreve has the advantage of being a single biological and durable reconstruction with early rehabilitation and an active lifestyle with long-term good outcomes.<sup>[14]</sup> Associated complications include arterial occlusion, deep vein thrombosis, impaired venous drainage and pressure sores in acute stages, and traumatic fractures in later stages.<sup>[15]</sup> Due to high physical and psychological morbidity, this procedure<sup>[16]</sup> was deferred. The expandable prosthesis was deferred to avoid multiple surgeries, the chance of stress shielding of the diaphysis, and increased risk of infection during long-term follow-up.<sup>[17]</sup> Amputation which was once considered to be the standard treatment in bone sarcomas has now been reserved for only special cases where there is a contraindication to limb salvage procedures.<sup>[18]</sup> Since there was no encasement of the major neurovascular bundle by the tumor or any extensive soft tissue or joint involvement, amputation was deferred.

Manfrini described the first case in a 4-year-old child with a proximal femoral Ewing's sarcoma,<sup>[19]</sup> in which a 4-year follow-up showed remodeling of the fibula that allowed the construct to form a pseudo proximal femur and flap showed an adequate growth. Other studies include Innocenti *et al.*<sup>[4]</sup> where they successfully achieved an average growth rate of more than 1 cm/year ranging between 0.75 and 1.33 cm with a vascularized fibula graft incorporating the proximal physis. Stevenson *et al.*<sup>[7]</sup> in their retrospective study found that VFET preserves function and growth in young children following excision of the proximal humerus for a malignant bone tumor.

VFET is a technically demanding technique, which can lead to severe complications both in the donor as well as the recipient site.<sup>[20]</sup> In our case, since the lesion was small, we

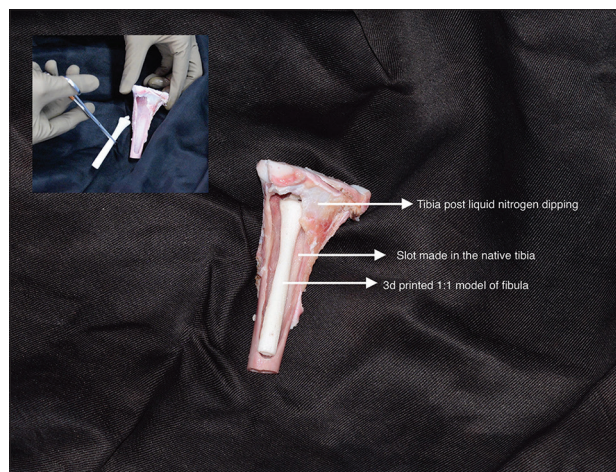


Figure 9: Postliquid nitrogen treatment the tibia is slotted to accommodate the three-dimensional printed fibula as trial

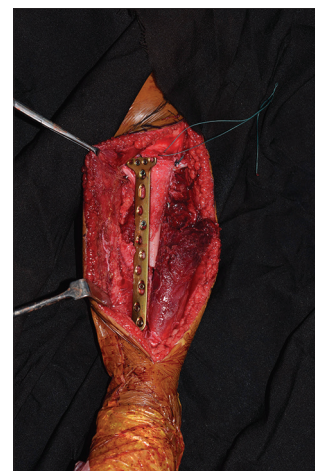


Figure 10: Final construct of tibia with fibula and locking plate

were able to dissect out and separate the epiphyseal recurrent branch without sacrificing any motor branches of the peroneal nerve. Our modification of the standard technique involved using a 3D printed fibula to assess accommodation of the transposed fibula into the free frozen proximal tibia. This strategy shielded us from any mismatch or intraoperative difficulties with vascular manipulation throughout the procedure. The absence of long-term follow-up owing to the patient's mortality after adjuvant treatment due to sepsis is one of our study's shortcomings.

## CONCLUSION

We are reporting the first recorded case of proximal tibia Ewing's sarcoma in a 2-year-old child. The emphasis here is on the surgical process rather than the outcome, which should be considered in future reconstructive treatments for children with these disorders. By combining VFET with free frozen autograft, this novel technique has the potential to address limb length discrepancies in tumors of the physeal region in skeletally immature patients. Further systematic studies are recommended to validate the method fully.

## Ethical committee approval

Ethical approval was waived off by the local Ethics Committee of the institution in view of the retrospective nature of the study.

## Declaration of patient consent

Parents of the patient gave consent for publication of case data.

## Financial support and sponsorship

Nil.

## Conflicts of interest

There are no conflicts of interest.

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