Foot and Ankle Surgery 25 (2019) 278–285

Contents lists available at ScienceDirect

Foot and Ankle Surgery

journal homepage: www.elsevier.com/locate/fas

Limb salvage for malignant bone tumours of distal tibia with dual ipsilateral vascularized autogenous fibular graft in a trapezoid-shaped array with ankle arthrodesis and preserving subtalar joint

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ARTICLE INFO

Article history:
Received 27 September 2017
Received in revised form 15 November 2017
Accepted 29 November 2017

Keywords:
Bone defect
Tumour
Vascularized fibular graft
Ankle fusion
Preserving subtalar joints

ABSTRACT

Background: The treatment of malignant tumours of the distal tibia is a challenging surgical problem due to the scarce soft tissue coverage and the instability of the ankle joint that often occurs after resection. However, there is no consensus on the ideal treatment for malignant tumours of the distal tibia.
Methods: We report a new reconstruction for five patients with high-grade osteosarcoma of distal tibia, using dual ipsilateral vascularized autogenous fibular graft in a trapezoid-shaped array and external fixator, with ankle arthrodesis and preserving subtalar joints. The patients were examined clinically and radiographically.
Results: The average follow-up duration was 88 months. The mean wound healing time was 14 days. Bone healing was achieved for all the five patients at an average time of 7 months. There were no complications of mal-union, skin necrosis, post-operative infection, loss of internal fixation, peroneal nerve injury. One patient had a local recurrence, which required amputation 15 months postoperatively. The remaining four patients were able to walk with an average functional score of 81.25% according to MSTS.
Conclusions: Our study shows that this technique is safe and effective to perform implantation of dual ipsilateral vascularized autogenous fibular graft in a trapezoid-shaped array and preserving subtalar joints in terms of the distal tibial reconstruction for malignant bone tumour of the distal tibia. This reconstruction represents a biological alternative protocol for limb salvage in cases of malignant bone tumour of the distal tibia, with encouraging results and with the advantages of lower complications and accelerating recovery.
Level of evidence: Level IV.

1. Introduction

Malignant bone tumours of the distal tibia are rare [1]. Amputation has been the historical standard surgical treatment for malignant tumours in this location [2–5]. Despite the fact that functional scores of patients with below-knee amputation are good, an increasing number of patients refuse amputation for psychological and social reasons [4]. When limb-sparing surgery is performed, various methods are available, such as endoprosthetic replacement, allografts, allograft combined with vascularized bone graft, reimplantation of sterilized autologous bone, or vascularized fibular graft (either free or pedicled) for distal defects [6–14]. When bone sarcomas do occur in the distal tibia, the limb-sparing treatment is a challenging surgical problem due to the scarce soft tissue coverage and the instability of the ankle joint that often occurs after resection. In order to increase the stiffness and stability of the reconstruction, lower the incidence of complications, this study was to evaluate the safety and efficacy of the reconstruction based on implantation of dual ipsilateral vascularized autogenous fibular graft in a trapezoid-shaped array. We studied the feasibility and effectiveness of the technique and report our preliminary results and complications.

2. Methods and patients

Between February 2007 and November 2012, five patients (three males and two females; average ages round up to whole numbers and give range, 31.8 years) with high-grade osteosarcoma of the distal tibia were treated with en bloc excision and reconstruction using unaffected dual ipsilateral vascularized autogenous fibular graft in a trapezoid-shaped array. After the clinical examination and plain biplanar radiographs (Fig. 1A)

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https://doi.org/10.1016/j.fas.2017.11.006
1268-7731/

suggested a malignant lesion, all patients underwent Magnetic Resonance Imaging (MRI) (Fig. 1B) and Emission Computed Tomography (ECT), Computed Tomography (CT) (Fig. 1C), and/or CT angiogram. The CT of chest was also required. CT of abdomen and pelvis depended on the type of tumour.

Core needle biopsy was performed for all five patients, with histopathological examination confirming high-grade osteosarcoma (Fig. 2). The Enneking staging system was used to characterize the bone tumours by Grade (G), Site (T) and Metastasis (M) and by stages I, II and III. Restaging was performed after two to three cycles of preoperative chemotherapy following biopsy. A summary of the patients' profiles was shown in Table 1. All patients were examined postoperatively with standard radiographs and clinical outcomes. Prospective database records included systemic status, residual symptoms, and any necessary additional treatment. Functional evaluation was performed using a modified system of the Musculoskeletal Tumour Society (MSTS), which is based on six parameters: pain, functional activities, emotional acceptance, the use of external support, walking ability and gait.

2.1. Surgical technique

The procedure was carried out with four main steps: 1. harvesting the vascularized autogenous fibular graft, 2. en bloc intra-articular excision of the tumour, 3. reconstruction of the defect with autogenous fibular graft, and 4. arthrodesis of the ankle joint and fixed by external fixation and screws.

The anatomic landmarks were based on the medial and lateral malleoli and the anterior tibial crest. The patients were placed in the supine position. Two incisions were used, one for procuring fibula and one for tumour excision and reconstruction. A longitudinal anterior incision from the proximal of tibia (5 cm above the margin of lesion) to the ankle was taken according to the site of biopsy and the position of the tumour determined by CT, MRI. The incision could be extended onto the dorsal side of the foot in order to gain adequate access to the talus. Previous biopsy tracts were incorporated into the incision and completely excised with the specimen. Meticulous dissection was carried out, and tumour-free margins were obtained in all cases.

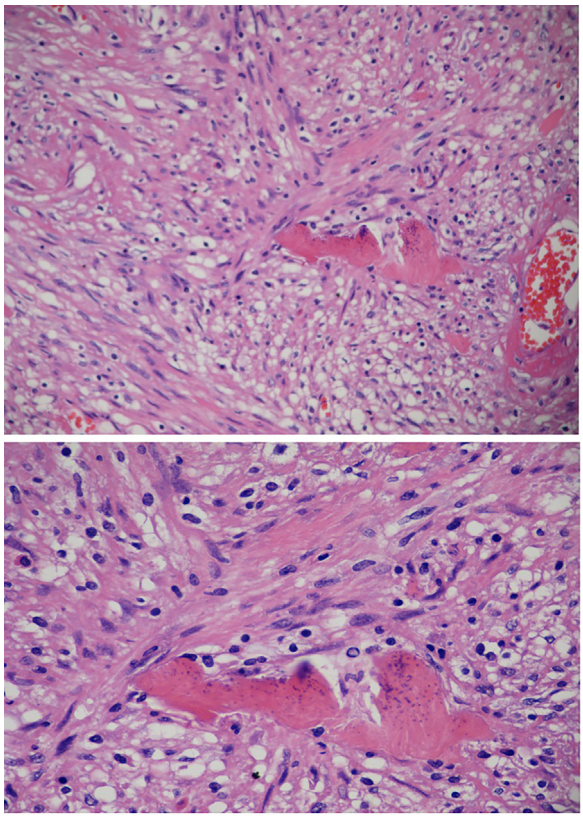

Fig. 2. Pathology showing a diagnosis of high-grade osteosarcoma.

2.2. Harvesting the vascularized autogenous fibular graft

The ipsilateral fibula was separated just above the tumour margin and then again separated proximally to obtain a vascularized pedicled segment. Enough proximal fibular length was left to protect the common peroneal nerve (at least 5 cm from the tip of fibular head) (Fig. 3A).

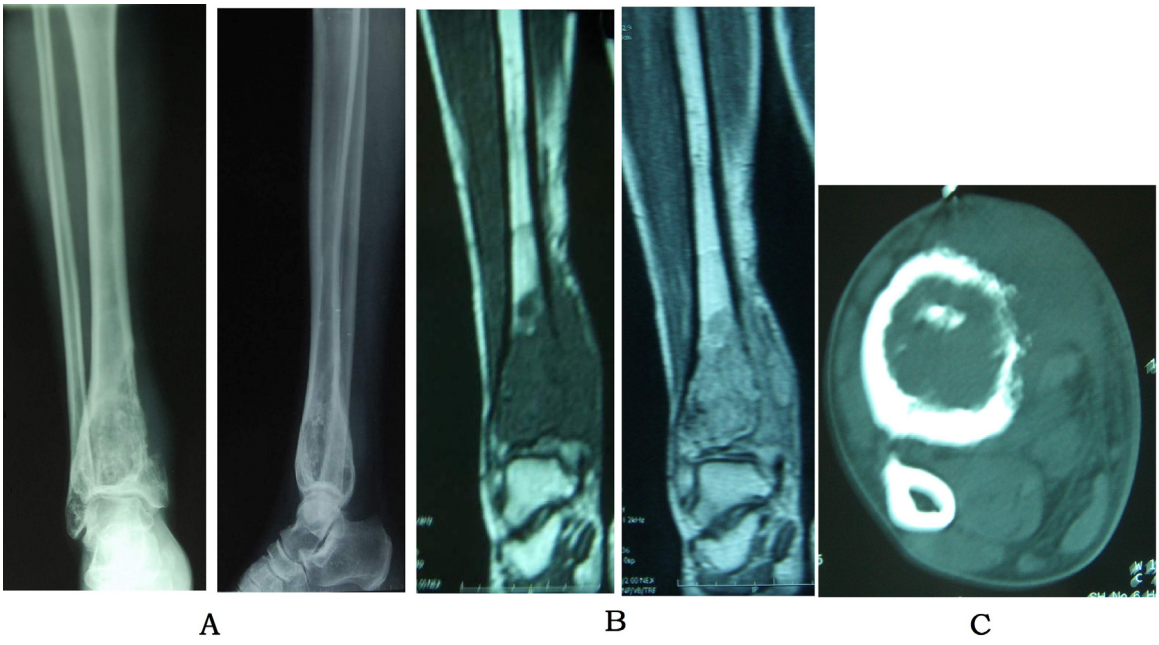

Fig. 1. Pre-operative X-ray (A), MRI (B), CT (C) showed the lesion located in the distal of the right tibia.

Table 1
Summary of the patients' profiles.

Patient	Location	Age	Gender	Diagnosis	Enneking stage
1	Distal tibia	34	M	Osteosarcoma	II A
2	Distal tibia	39	M	Osteosarcoma	II A
3	Distal tibia	16	F	Osteosarcoma	II B
4	Distal tibia	25	F	Osteosarcoma	II A
5	Distal tibia	45	M	Osteosarcoma	II B

2.3. Mobilized and dissected the anterior tibial artery, and great saphenous vein

The anterior tibial artery and the great saphenous vein were exposed and cut off, then clamped for anastomosis after the procedure of "reconstruction the defect by dual-vascularized autogenous fibular grafts".

2.4. En-bloc, intra-articular excision of the bone tumour

The tibia was sectioned 3 cm above the proximal margin of the tumour, usually 15–17 cm above the ankle. A specimen of bone marrow from the proximal side of the tibial stump was evaluated by frozen section to confirm that the margin was negative. Then en bloc intra-articular excision of the bone tumour was performed.

2.5. Installed the external fixator

The length of limb and the correct position was confirmed firstly. Then the unilateral external fixator was installed. Two screws were inserted into the tibia proximally and the other two screws were inserted into the calcaneus. The frame should be installed after the tumour resection and before the vessels were anastomosed.

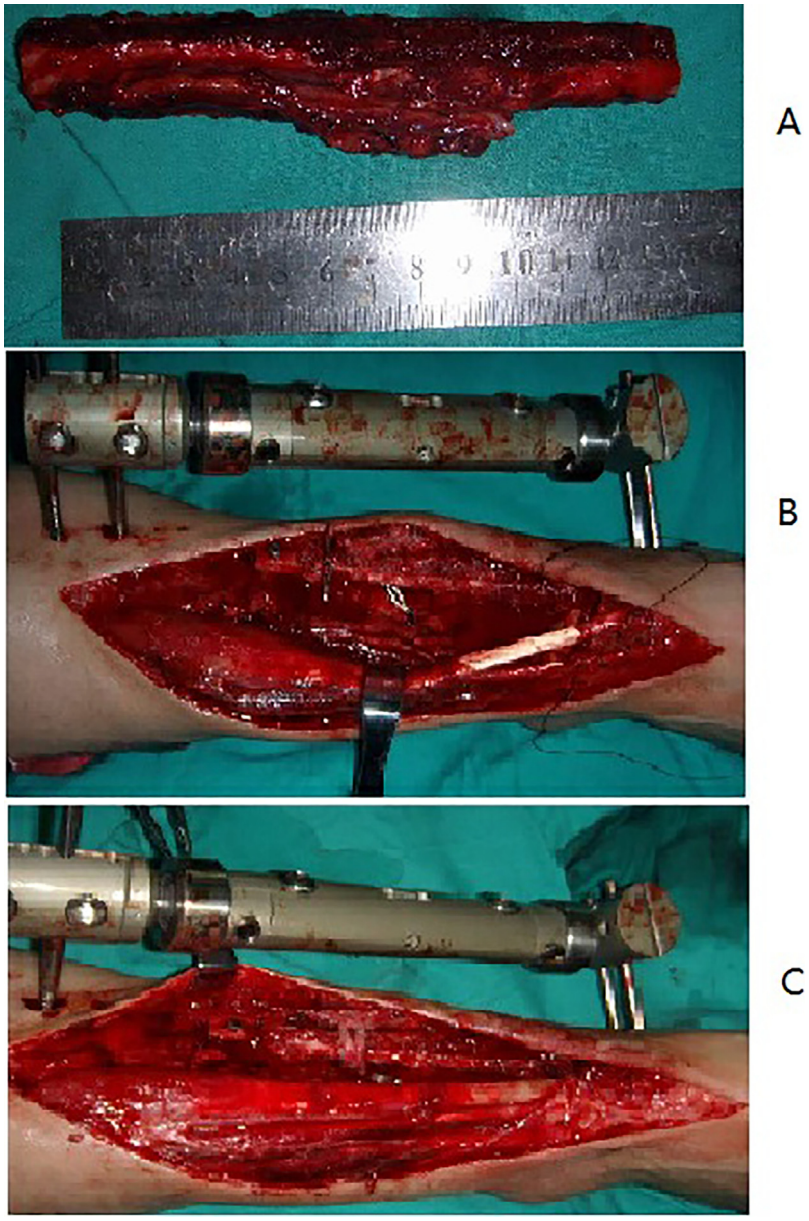

Fig. 3. Harvested the fibular graft, anastomosed the vessels and reconstructed the defect of the distal tibia successfully.

2.6. Reconstruction the defect by dual-vascularized autogenous fibular grafts

The proximal osteotomized free vascularized fubula is then placed on the medial side of the talus distally and medial side of the remaining tibia proximally. The cartilages of the two sides of the talus were eradicated, then were fused together with the adjacent double fibulae. Supplementary autogenous iliac crest graft was obtained and placed at all the fusion sites around the graft host junctions to promote fusion, both proximally and distally. Put iliac crest autogenous graft to two pairs of junctions medial and lateral sides. The two junctions were fibula-tibia junctions and fubula-talus junctions. The ankle joint was fused but the subtalar joint was not fused. Using two screws to fix the proximal of fibula and tibia junction. Using a long lag screw to fix the fibula and talus distally (Fig. 3B).

2.7. Anastomosed fibular artery/vein with anterior tibia artery and great saphenous vein

Anastomosis was achieved by connecting the fibular artery with anterior tibia artery, fibular vein with great saphenous vein (Fig. 3C). Because the diameter of fibular vein is bigger than anterior tibia vein, thus the great saphenous vein was selected to connect with anterior tibia vein since the two veins have similar diameter. The microvascular anastomosis was performed by end-to-end coaptation [15,16].

The distal fibula is still vascularized by the branch of posterior tibial artery and should be considered a vascularized autogenous graft (Fig. 4A) [17]. The blood supply of the residual distal lateral fibula was intact so the fibula on the lateral side was already vascularized because anterior tibial artery goes down and becomes arteria dorsalis pedis, then communicates with peroneal artery and posterior tibial artery. They can provide adequate blood supply to the distal of the fibula.

2.8. Wound closure

Wound closure is achieved where there is adequate soft tissue, by suturing the extensor hallucis longus across the fibular graft and attaching this to the peroneus longus to provide some cover for anterior area. The wound is closed over a single vacuum drain. We recommended a subcuticular absorbable suture where the skin edges are healthy and suitable to accept this technique. When it is difficult to close the wound, skin graft is recommended. When there is insufficient soft tissue, then a combined procedure with a plastic surgical team should be performed with fabrication of a free flap or fascio cutaneous turn-down flap from the calf. Transposition of the medial head of the gastrocnemius is recommended to close the soft tissue defect because it could provide necessary blood supply for wound healing and reduce the risk of infection [18,19].

The limb was then bandaged. Postoperative elevation was essential. Prophylactic antibiotics (Cefradine, 2 g, iv, qd for 2 days) were recommended. We gave dipyridamole, Dextran 40, Papaverine to keep anastomosis patent and for DVT prophylaxis treatment.

2.9. The rationale of the method

The present rationale was to achieve a biological reconstruction of the affected site, avoiding the use of endoprostheses or allografts, but with sufficient biomechanical strength and durability for the construct to withstand the stress of normal weight bearing. The patient's fibular autograft was taken as vascularized

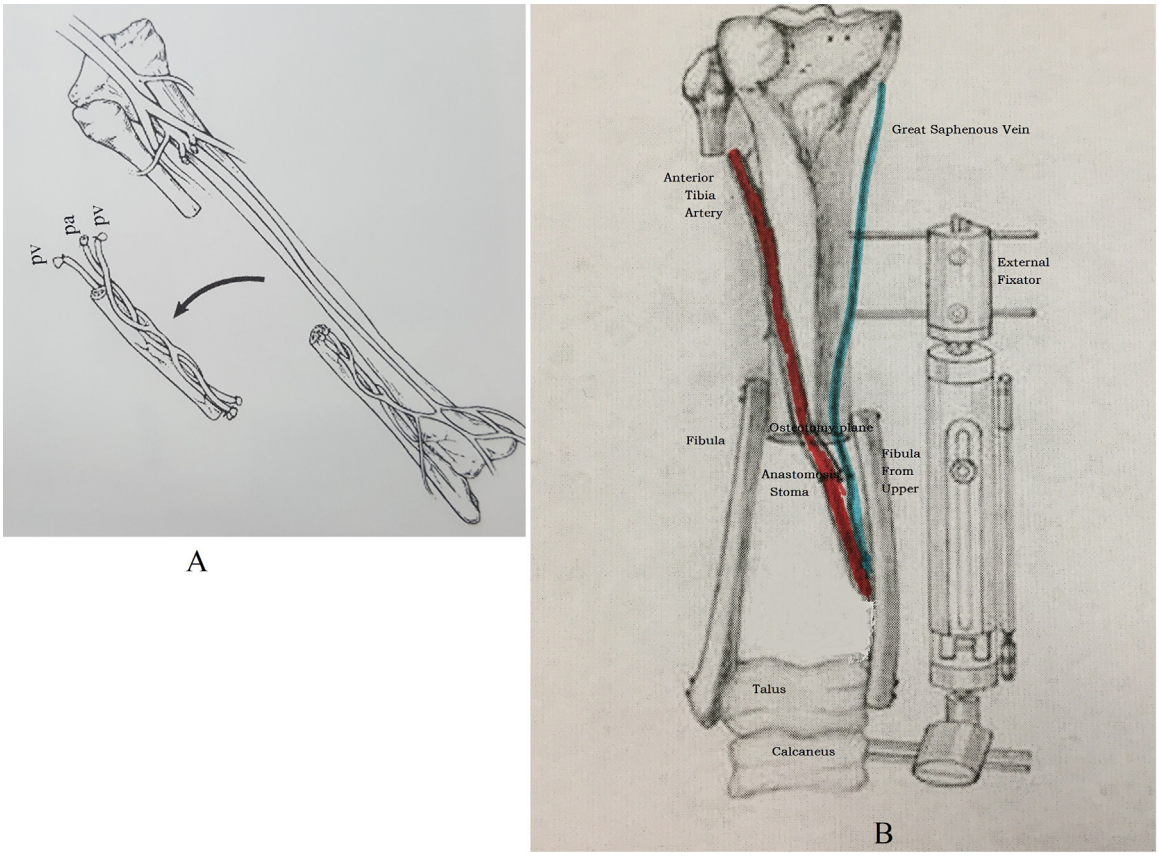

Fig. 4. Schematic representation of blood supply to the distal half of the fibula (A). Diagram of the surgical design. It showed the reconstruction and vessels anastomosis (B).

segment with its attached muscles and intact nutrient peroneal blood vessels. The fibular was put parallel proximally at the tibial osteotomy site and distally into parallel closely with the talus to achieve tibiotalar fusion and tibiofibular fusion. The surface between the fibula and the tibia was polished by a high speed bur to accelerate bone healing. The dual inlay grafts can sustain more stress stimulation which is very important for graft hypertrophy. The graft hypertrophy is a result of the mechanical stimulation of early weight-bearing permitted by external fixation and the result of revascularization. Such stabilizing method has the advantage of providing the transferred fibular grafts with a state of relative stability instead of absolute stability. In the biomechanical environment of relative stability, stress shielding is minimized and fibular graft hypertrophy can be promoted [20]. The reconstruction structure looked like a trapezoid-shaped array. We believe that this trapezoid structure can provide better stability. The external fixator stabilized the fibular grafts and tibia. The diagram showed the design conception and outcome of the distal tibia with resection, dual vascularized autogenous fibular graft, ankle arthrodesis preserving subtalar joint, and unilateral external fixator (Fig. 4B).

2.10. Postoperative management

Postoperative chemotherapy was given to all the five patients by four-drug regimen of cisplatin, adriamycin, high-dose methotrexate and ifosfamide. The degree of tumour necrosis was rated according to the Huvos grading system [21]. The Huvos grading system has served as a model for chemotherapy response systems. A Grade III and IV response is characterized by an extensive or complete destruction of cells within the primary tumour and is associated with better survival. While a Grade I or II response is indicative of minimal destruction of tumour; these patients are more likely to develop distant metastases and have a poor survival.

The patients were examined clinically and radiographically every month during the first six months after surgery for evidence of local relapse or other complications. Follow-up was then conducted at intervals of three months in the next two years and then at intervals of six months thereafter. A CT scan of the chest was carried out every 6 months in the first year then every 12 months for pulmonary surveillance. A standard bone scan was performed every six months in the first year and then annually thereafter.

The external fixator was removed after graft union. Protection of the limb was then achieved with a walking boot for an additional four to six months. Bony union was defined as a solid callus bridge between the fibula graft and the host tibia on plain X-rays. Bone healing of graft and osteotomy sites including uninterrupted external bony borders between the fibular graft and the recipient bone, obscured or absent osteotomy lines at both junctions, and clinical resumption of normal function and weight-bearing without discomfort [22,23].

3. Results

The average duration of operation was 6.8 h (range 5–8 h) and the average blood loss was 550 ml (range 400–600 ml). The distal tibia defect lengths ranged from 13 to 15 cm (mean 14 cm). The mean wound healing time was 14 days (range 11–16 days). The clinical and follow-up data were shown in Table 2. The average follow-up period was 88 months (range 47–124 months). Bony union was achieved in all five patients at a mean time of 7.14 months (5.6–8.5 months) postoperatively (Figs. 5 and 6), and the external fixator was then removed. Tibiotalar arthrodesis with screws was successfully performed in five cases. The ankle was immobilized between 12 and 16 weeks (median 14 weeks). The motion of subtalar joint was preserved. None of the patients had developed symptoms of arthritis of the mid foot or the subtalar joint. All five patients were alive at last follow-up after the date of reconstruction. During the follow-up period, there was no complications of mal-union, skin necrosis, post-operative infection, peroneal nerve injury. One young patient (patient 3) aged 16 years had an amputation performed 15 months after the primary operation because of local recurrence. The remaining four patients showed good response to chemotherapy, with a tumour necrosis greater than 90% (Huvos Grade III–IV). They exhibited no signs of local recurrence or distant metastases during the clinical follow-up. They could stand, walk and crouch with good joint stability. There was no discrepancy of leg length, and their gaits were nearly normal. The mean MSTS score was 81.25% (range 76–86%) (Table 3).

4. Discussion

Free vascularized fibular graft (FVFG) has been widely used in malignant bone tumours, trauma, as well as for those defects causing congenital pseudarthrosis [24]. It can be an ideal method to repair long bone defects. This technique accelerates bone healing and prevents the formation of non-union characteristics. Several reports have demonstrated that limb reconstruction of bony defects of the middle or distal tibia after bone tumour resection using pedicled vascularized fibula can be performed inexpensively and has a low rate of late complications [25,26].

Ipsilateral vascularized fibula transfer (IVFT) has been successfully used in the treatment of tibial nonunion, traumatic bone defects, bone tumour defects and congenital pseudarthrosis with favorable results and minor complications [27–34]. Long-term follow-up of pedicled vascularized fibula in reconstruction of bony defects of the middle or distal tibia after bone tumour resection has illustrated that it is an useful tool for limb salvage [27–34]. When ipsilateral vascularized fibula from the affected site is available, IVFT has the advantages of shorter operative and convalescent time, less complications, better outcomes, the operative invasion could be limited to only one leg [27–34]. Our results are consistent with the literature. We had sound union between 5.6 and 8.5 months and no complications of mal-union, skin necrosis, post-operative infection, peroneal nerve injury. Still, careful control of

Table 2
Clinical and follow-up data.

Patient	Length of bone defect (cm)	Length of fibular graft (cm)	Follow up period (months)	Chemotherapy	Huvos grade	Days for wound healing	Complications	Time to graft union (months)	Limb discrepancy (cm)
1	15	17	124	Pre/post	IV	12	None	6.5	No
2	14	16	98	Pre/post	III	15	None	8.5	No
3	15	17	47	Pre/post	II	16	Local recurrence	8.1	NA
4	13	15	57	Pre/post	III	16	None	5.6	No
5	14	16	114	Pre/post	IV	11	None	7.0	No

Pre/post = pre and postoperative; NA = not available.

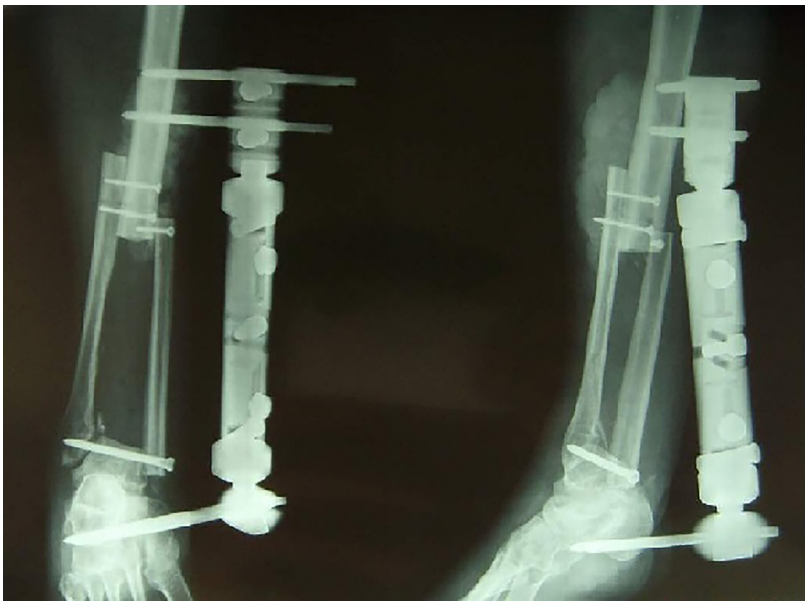

Fig. 5. Postoperative X-ray showed the stability of the reconstruction of the distal tibia defect.

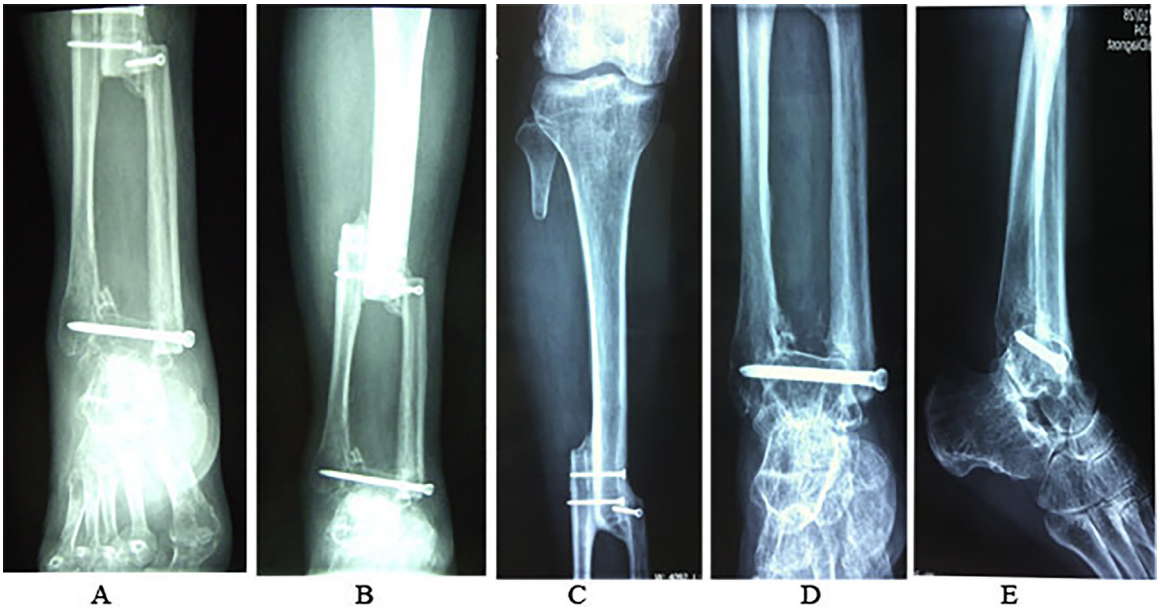

Fig. 6. X-ray at 12 months (A,B), 60 months (C–E) postoperatively showing the union of the junctions of the tibia-fibula and the fibula-talus.

Table 3
Postoperative MSTS functional score of patients.

Patient	Pain	Functional activities	Emotional acceptance	Gait	External support	Walking ability	Total functional score	Functional score (%)
1	4	4	5	4	4	4	25	83
2	5	4	5	4	4	4	26	86
4	4	3	4	4	4	4	23	76
5	4	4	4	4	4	4	24	80

alignment at the operative site is necessary to prevent malalignment of the lower leg.

Double-strut FVFG can withstand a higher torque and mechanical stress than conventional single-strut FVFG and has been widely used to manage large bone defects in the lower extremities. The vascularized double-barrel fibular graft represents one type of fistulous bone graft with blood supplies, facilitating bony union and reducing the treatment period [24,35–39]. The single fibula double-turnover operation prevents the insufficient or weak intensity bone healing that occurs in large bone defect repair. It resulted in clinically acceptable bone activity and biological mechanics [24]. The present study reported that the

patients have nearly normal walk abilities and gaits which shows that dual ipsilateral vascularized autogenous fibular graft in a trapezoid-shaped array can provide stable and strong reconstruction frame for patients' walking ability and is an effective reconstruction method for distal tibia after bone tumour resection.

Due to the missing lateral ligament at the distal part of the fibula, instability of the ankle can occur despite reconstruction, some authors recommend an arthrodesis of the ankle joint since ankle arthrodesis could provide excellent stability of the reconstruction and the results remain satisfactory after ankle arthrodesis [3,5,40–42]. Ankle arthrodesis after distal tibial resection may be performed with allografts, autografts, vascularized grafts or a combination of them. The use of bone grafting procedures for ankle arthrodesis has been the method of choice by most authors. When the subtalar joint needs to be sacrified, an arthrodesis of the subtalar joint is an acceptable disadvantage in their clinical experience [3,5,40–42].

The subtalar joint allows inversion and eversion of the foot, but plays no role in dorsiflexion or plantar flexion of the foot [43]. It is considered a plane synovial joint, also commonly referred to as a condyloid joint. The subtalar joint can also be considered a combination of the anatomic subtalar joint discussed above, and also the talocalcaneal part of the talocalcaneonavicular joint. This is the more common view of the subtalar joint when discussing its movement. When both of these articulations are accounted together, it allows for pronation and supination to occur. Moore et al. reported that the allograft ankle arthrodesis was performed using a retrograde intramedullary tibial nail, the nail was inserted through the plantar surface of the calcaneus, crossing the subtalar joint, neotibiotalar anastomosis, and the junction of the allograft and native tibial shaft. They thought that because of the mechanical weakness of the nail at that location, nail fracture might occur at the subtalar joint, thereby restoring subtalar motion [44]. In the present study, ankle arthrodesis with screws was successfully performed. The subtalar joint was preserved; thus, the functions of subtalar joint was remained. The functional limitations of the present study with ankle arthrodesis and without subtalar joint arthrodesis were significantly fewer than patients with a tibiotalocalcaneal arthrodesis as published [3,5,40–42]. An extended ankle/hindfoot or tibiotalocalcaneal fusion was not recommended on basis of our current results.

Few studies have evaluated the functional results after reconstructing bone defects with vascularized fibula grafts following tumour resection in lower extremities. Khira and Badawy [25] reported the MSTS scores ranged from 24 to 27.5 points using pedicled vascularized fibular graft with Ilizarov external fixator for reconstructing a large bone defect of the tibia after tumour resection. Petersen et al. [6] investigated on 8 consecutive cases with limb-sparing tumour resection and reconstruction by vascularized fibular grafts. All patients with the fibula graft in situ had an average total MSTS score of 24. In the present report, the final follow-up functional outcome postoperatively was graded as excellent, and the MSTS score range of 23–26 points was comparable with others published [6,25,45–48].

Our technique has several advantages over the current methods. First, unaffected dual ipsilateral vascularized autogenous fibular grafts are easy to harvest and cause minimal donor-site morbidity. Second, the functional outcome is favorable because dual ipsilateral vascularized autogenous fibular graft in a trapezoid-shaped array can provide stable and strong reconstruction frame for patients' walking ability in the future. Third, since there is no use of prosthesis or allograft, the auto-graft reconstruction is an economical, safe, and effective method without complication of the rejection reaction and nonunion, infection, osteolysis, fracture [49]. Fourth, the functions of subtalar joint was preserved. The authors consider this method to be stable and strong enough for distal tibial reconstruction, despite the risk of external fixator pin tract infection, delayed weight bearing on the biological reconstruction, and the need for experienced doctors to perform vascular anastomosis.

The limitations of the present report included the low number of cases, but due to the rarity of distal tibia tumours, no large series have been reported for this type of procedure. Longer follow-up is still needed to confirm the oncological results.

In conclusion, dual ipsilateral vascularized autogenous fibular graft in a trapezoid-shaped array with resection, ankle arthrodesis and external fixation offers an effective and economical biological alternative procedure in cases of malignant bone tumours of the distal tibia.

Conflict of interests statement

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.

Acknowledgement

Not applicable.

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