Limb salvage for osteosarcoma of the distal tibia with resection arthrodesis, autogenous fibular graft and Ilizarov external fixator

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We report the results of limb salvage for non-metastatic osteosarcoma of the distal tibia using resection arthrodesis, autogenous fibular graft and fixation by an Ilizarov external fixator.

In six patients with primary osteosarcoma of the distal tibia who refused amputation, treatment with wide *en bloc* resection and tibiotalar arthrodesis was undertaken. The defect was reconstructed using non-vascularised free autogenous fibular strut graft in three patients and a vascularised pedicular fibular graft in three, all supplemented with iliac cancellous graft at the graft-host junction. An Ilizarov external fixator was used for stabilisation of the reconstruction.

In five patients sound fusion occurred at a mean of 13.2 months (8 to 20) with no evidence of local recurrence or deep infection at final follow-up. The mean post-operative functional score was 70% (63% to 73%) according to the Musculoskeletal Tumour Society scoring system. All five patients showed graft hypertrophy.

Union of the graft was faster in cases reconstructed by vascularised fibular grafts. One patient who had a poor response to pre-operative chemotherapy developed local tumour recurrence at one year post-operatively and required subsequent amputation.

Primary malignant bone tumours of the distal tibia are very rare.¹ Amputation has been the standard surgical treatment with satisfactory functional results obtained using an appropriate prosthesis.² With advances in limb salvage surgery, reconstruction has been attempted using autograft, allograft, implanted prostheses and composites. When the ankle is involved, limb salvage presents unique difficulties both in terms of the biomechanics and obtaining soft-tissue cover.³ Casadei et al⁴ reported that arthrodesis is the best reconstructive procedure for the ankle in patients for whom limb salvage is indicated.

The aim of this study was to assess the results of treating a group of patients with high grade osteosarcoma of the distal tibia using a specific protocol for limb salvage, of wide *en bloc* intra-articular excision and reconstruction of the defect with autogenous fibular strut graft, stabilised using an Ilizarov external fixator.

Patients and Methods

A series of six patients with high grade non-metastatic primary osteosarcoma of the distal tibia (Fig. 1) who had refused amputation,

were treated by wide *en bloc* excision with reconstruction of the defect by autogenous strut fibular graft with stabilisation provided by an Ilizarov external fixator (Smith & Nephew, Memphis, Tennessee). Following clinical diagnosis, imaging studies including plain radiographs, MRI, CT scans, bone scans and angiography were carried out to assess tumour mass, soft-tissue involvement, infiltration of adjacent tissues and to determine the margins of resection. Carefully planned open biopsy was undertaken for all patients, with histopathological examination confirming conventional high grade osteosarcoma. All of the tumours were graded as Enneking stage IIB.⁵ Restaging was undertaken after three cycles of pre-operative chemotherapy at three week intervals, each formed of adriamycin 75 mg/m² and cisplatin in 105 mg/m² for three days.

The mean age of the patients at the time of surgery was 18.8 years (16 to 23). Three patients were male and three female. The mean follow-up was 35 months (28 to 42).

Surgical technique. The operation consisted of three main steps: wide *en bloc* intra-articular excision, reconstruction of the defect by autogenous fibular strut graft and fixation of the graft using an Ilizarov external fixator.

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Fig. 1a

Fig. 1b

Radiographs taken at presentation showing an osteolytic lesion of the right distal tibia, from a) anteroposterior and b) lateral views.

Fig. 2

Radiographs after *en bloc* excision, non-vascularised fibular graft and application of an Ilizarov external fixator.

The mean length of the bony defect after excision of the tumour segment was 17 cm (15 to 19). In three patients the fibular graft was taken as a free non-vascularised segment through a lateral approach with subperiosteal dissection. In the others the fibular graft was transferred with its attached muscles and intact nutrient peroneal blood vessels. The mean length of the fibular graft was 19.8 cm (18 to 22). The fibula was inserted proximally at the tibial osteotomy site into the intramedullary canal and distally into a trough in the talus to achieve tibiotalar fusion. All patients had supplementary iliac crest cancellous bone graft placed around the graft host junctions.

The Ilizarov frame had a proximal partial ring connected to a full ring by three or four connecting rods, a full ring in the middle overlying the fibular graft and a distal foot assembly composed of a calcaneal half ring connected to a forefoot half ring. One or two wires were inserted under direct vision into the fibular graft, tensioned and fixed to the middle full ring to allow manipulation of the graft and compression at both ends during the follow-up period, if needed (Fig. 2).

Post-operatively, all patients were allowed immediate full weight-bearing up to their tolerance.

The degree of tumour necrosis, in response to the pre-operative chemotherapy, was rated according to the Huvos grading system.⁶ Three patients had a grade IV response (complete tumour necrosis), two had a grade III response (more than 90% necrosis) and one had a grade II response (less than 90% necrosis).

The patients were examined clinically and radiologically every six weeks during the first six months after surgery for evidence of infection and local recurrence, every three months for the following two years and then every six months. A CT scan of the chest was performed every three

months in the first year and then every six months to detect pulmonary metastases. A bone scan was performed every six months in the first year and then annually until the last follow-up.

The Ilizarov external fixator was removed after graft union. This was followed by protection of the limb in a below-knee brace for an additional four to six months.

Union of the graft proximally and distally was assessed according to Hsu et al⁷ with graft union defined as uninterrupted external bony borders between the fibular graft and the recipient bone in addition to obscured or absent osteotomy lines at both junctions.

We assessed graft hypertrophy by the system of de Boer and Wood,⁸ using a graft hypertrophy index I_n as:

$$I_n = \frac{(G_2/R_2) - (G_1/R_1)}{(G_1/R_1)} \times 100\%$$

where G_1 is the graft diameter at the proximal junction at operation, R_1 is the host bone diameter at the proximal junction at operation, G_2 is the graft diameter at the proximal junction at follow-up and R_2 is the host bone diameter at the proximal junction at follow-up.

The most recent follow-up was used for this index. A positive index value confirms hypertrophy and a negative value represents graft atrophy. We recorded hypertrophy only if the graft index had increased by more than 20%.

The functional evaluation was performed using a modified system of the Musculoskeletal Tumour Society,⁵ which is based on six parameters: pain, functional activities, emotional acceptance, the use of external support, walking ability and gait.

Results

The mean post-operative functional score was 70% (63% to 73%). Table I shows the details of the functional results.

Fig. 3

Full union of the graft and a hypertrophy index of 40% at the 42-month follow-up.

Table I. Details of the functional results

Patient number	Pain	Functional activities	Emotional acceptance	External support	Walking ability	Gait	Total functional score (%)
1	4	3	5	3	3	3	70
2	4	3	4	3	3	2	63
4	5	3	5	3	3	3	73
5	4	3	5	3	3	3	70
6	5	3	5	3	3	3	73

Five patients had good joint stability after sound fusion. Two patients required secondary iliac crest cancellous bone grafting at the proximal end to achieve union. The mean time to graft union was ten months (8 to 12) in patients with vascularised fibular grafts and 18 months (16 to 20) in those with a free non-vascularised graft.

All the patients showed graft hypertrophy at final follow-up with the exception of one patient who suffered a local recurrence. We believe that graft hypertrophy is a function of the mechanical stimulation of early weight-bearing permitted by the Ilizarov external fixator (Fig. 3). Table II summarises the details of the study group including complications, follow-up and results.

None of the patients had developed clinical signs or symptoms of arthritis of the mid-foot or the subtalar joints at their final follow-up.

Complications. A local recurrence occurred in one female patient one year post-operatively. This patient showed a poor response to pre-operative chemotherapy (grade II according to Huvos grading⁶) and was not compliant with the post-operative chemotherapy regimen. Subsequently she was treated by above-knee amputation and showed no

Table II. Details of this study group

Patient number	Type of fibular graft	Length of defect after resection (cm)	Length of fibular graft (cm)	Follow-up period (mths)	Complications	Time to graft union (mths)	Graft hypertrophy index (%)
1	Non-vascularised	19	22	42	None	16	40
2	Non-vascularised	18	22	31	Stress fracture of fibular graft	20	25
3	Non-vascularised	16	19	36	Local recurrence	---	---
4	Vascularised	18	20	33	Pin track infection	8	40
5	Vascularised	15	18	28	None	10	50
6	Vascularised	16	18	40	None	12	50

local recurrence or pulmonary metastases at one year after amputation.

One patient had a stress fracture at the proximal end of the non-vascularised fibular graft six months post-operatively. The graft was 22 cm long and the defect after tumour resection was 18 cm. The patient was treated by application of autogenous iliac cancellous graft at the site of the fracture, and full union of the graft occurred after 20 months.

Pin track infection occurred in one patient and was treated by oral antibiotics and routine cleaning of the wire sites.

At the final follow-up the fusion was solid without any evidence of local recurrence or deep infection in five patients.

Discussion

Reconstruction after massive bone loss around the ankle is difficult because of the limited surrounding soft tissue and problems achieving adequate fixation. Accordingly, below-knee amputation has been the standard surgical procedure for malignant bone tumours involving the distal tibia, providing satisfactory function with a below-knee prosthesis.⁹

In spite of the fact that functional scores of a below-knee amputee are likely to be 90% or more, some patients refuse amputation for psychological or social reasons.¹⁰

Advances in chemotherapy have also made limb salvage surgery of the distal tibia increasingly possible.¹¹ Limb salvage surgery consists of two main parts; *en bloc* resection of the tumour, and subsequent skeletal reconstruction. Various techniques have been described for reconstruction including endoprosthetic replacement, osteoarticular allograft, autograft, distraction osteogenesis or re-implantation of the tumour-bearing bone segment after devitalisation of the tumour cells by heating (autoclaving, pasteurisation and microwave-induced hyperthermia), freezing (using liquid nitrogen) or extracorporeal irradiation.¹²

Ankle endoprosthetic replacement has occasionally been carried out in patients with distal tibial tumours with a report of many complications and high rates of failure.¹¹ Other reports also show a deterioration of functional outcome with time, together with significant medium-term morbidity.^{2,13}

Among the many options of reconstruction, arthrodesis was regarded as the best, providing excellent stability of the ankle and avoiding problems relating to the prosthetic implantation.¹¹ Casadei et al⁴ reported good functional and oncological results in patients with malignant bone tumours of the distal tibia, treated by resection and arthrodesis with autogenous bone graft.

The autogenous fibular graft has some important advantages over other donor sites because of its length, geometrical shape and mechanical strength. The fibula is a long and straight tubular bone which is not difficult to harvest. The anatomy is predictable and its size and shape allow tibial

intramedullary insertion. Donor site morbidity has been reported as minimal with grafts up to 20 cm long.¹⁴

Non-vascularised autogenous fibular grafts hypertrophy, but only after they have been revascularised and repaired completely. Vascularised bone graft does not have to undergo creeping substitution and does not depend on the quality of the host bed for its success. In contrast, non-vascularised bone grafts need good vascularity of the host bed.⁷

However, achieving stability after using fibular graft for reconstruction is difficult. The use of internal fixation carries a high rate of failure with infection being a disastrous complication. These techniques also involve long periods of non-weight-bearing which is a particular concern for tumour patients.¹⁵

Bishop et al³ achieved success with a reconstruction using a vascularised free fibular graft for the treatment of malignant tumours in the distal tibia. A successful fusion was obtained in nine of 11 patients.

Our technique has several advantages. The fibular strut graft is easy to harvest and causes minimal donor-site morbidity. It can also fit snugly inside the medullary canal of the tibia. The Ilizarov external fixator allows stability and longitudinal compression to be established early in the post-operative period, avoiding the problems of internal fixation of the fibular graft, enhancing graft hypertrophy and allowing early weight-bearing.

The vascularised fibula was superior to the non-vascularised graft with reduced time to union and faster hypertrophy.

The functional outcome was better than reconstruction with an endoprosthesis¹³ and more long-lasting.² It was also comparable with that reported for below-knee amputation.¹⁰

We believe that wide *en bloc* resection arthrodesis, with reconstruction of the defect by autogenous fibular graft and fixation using an Ilizarov external fixator, together with pre- and post-operative chemotherapy, offers an effective alternative protocol for limb salvage in cases of osteosarcoma of the distal tibia.

This technique could be similarly applied to defect reconstruction after a wide range of bone pathologies which involve the distal tibia.

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