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Treatment of primary malignant bone tumours of the distal tibia

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Abstract We treated 15 patients with primary malignant bone tumours of the distal tibia of which 14 were treated by limb salvage surgery. Reconstructions were done by allografts with or without microvascular fibula transfer, by bone transport, by fibula transfer alone or by endoprosthetic replacement. The most successful methods were bone transport and endoprosthetic replacement. However, serious complications with deep infections leading to secondary amputation occurred in four patients and in all reconstruction groups. After a mean follow up of 7 years, no local recurrence occurred, and all patients were alive and free of disease. After radical resection, bone transport in defects less than 15 cm is a viable option. In larger defects in children, allograft with vascularised fibula is an acceptable alternative, but amputation still has a role in this group. In adults, endoprosthetic replacement with proper soft tissue coverage is a viable option in cases with large bony defects.

Résumé Nous avons traité 15 malades avec une tumeur maligne primitive du tibia distal avec pour 14 d'entre eux une chirurgie conservatrice du membre. Les reconstruc-

tions ont été faites par allogreffe avec ou sans transfert de péroné vascularisé, transport osseux, transfert du péroné seul ou par remplacement prothétique. Les meilleures méthodes étaient le transport osseux et le remplacement prothétique. Des complications sérieuses avec infection profonde menant à une amputation secondaire, se sont cependant produites chez quatre malades et dans tous les groupes de reconstruction. Après un suivi moyen de 7 ans, aucune récurrence locale n'est survenue et tous les malades sont vivants et libres de maladie. Après une résection radicale, le transport osseux dans les défauts de moins de 15 centimètres est une option valable. Dans plus grands défauts chez les enfants, l'allogreffe avec péroné vascularisé est une alternative acceptable, mais l'amputation a encore un rôle dans ce groupe. Chez les adultes le remplacement prothétique avec couverture cutanée adéquate est un choix valable dans les grands défauts osseux.

Introduction

Primary malignant bone tumours of the lower leg differ from malignant bone tumours in other parts of the body. Malignant tumours in the lower leg are less commonly encountered, and the resultant death rates are markedly lower than for tumours of other parts of the body [2, 17].

For many decades, amputation was the method of choice. The less malignant behaviour and advances in oncological treatment tempt one to use limb salvage reconstruction methods such as allograft and/or microvascular fibula transfer, bone transport, prosthesis or composites [3, 5, 6, 11]. However, limb salvage surgery of the distal tibia presents several difficulties, including soft tissue coverage and ankle joint biomechanics. Resection and reconstruction with an ankle arthrodesis using bone grafts or bone transport is a good and safe technique although it also includes disadvantages such as loss of function and non-union [3, 6, 14]. An additional vascularised free fibula graft may limit these problems [2]. A tumour prosthesis replacing the ankle joint is commercially available, but there are few reports detailing endoprosthetic replace-

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ment. Prosthetics allow quick functional recovery and maintains ankle joint movement [1, 8, 11]. However, as with all prostheses, there is a risk of loosening and problems with talar collapse. So far, long-term results are not convincing [1, 11].

Here we report the outcome of 15 patients treated for malignant bone tumours of the distal tibia. We compared the various techniques of reconstruction with oncological and functional outcome.

Patients and methods

Between March 1991 and August 2004, 15 patients (nine male and six female), with primary malignant bone tumours of the distal tibia were treated in the Department of Orthopaedics, University of Münster. Histopathological diagnosis was obtained by open biopsy. There were nine osteosarcomata; four Ewing's sarcomata; one chondrosarcoma; and one adamantinoma. In 13 patients, the tumour was stage II-B; chondrosarcoma and adamantinoma were stage I-B.

We performed a limb salvage reconstruction in 14 patients. One patient (case 12) was treated with primary amputation because of infiltration of the tumour into the ankle joint and the neurovascular bundle. Five patients were treated with allograft and microvascular fibula transfer

(group A) and three patients with bone transport (group B). Three patients were treated with allograft alone (group C), of which one was a total tibial allograft. Two patients were treated with fibula transfer alone (group D), and one patient was treated with a modular endoprosthesis of the MUTARS-System (group E) [8].

Bone transport was conducted in two phases, and it was not started until chemotherapy had been completed. Meanwhile, a cement spacer was inserted in the resected area (Fig. 1).

Mean age at the time of surgery was 24.4 (11.2–45.2) years and mean follow-up 7.0 years (0.5–14.0). When this study was completed, all patients were alive and free of disease. Thirteen patients were treated with neoadjuvant and adjuvant chemotherapy according to the COSS-Study for Osteosarcoma and the EURO-Ewing study for Ewing's sarcoma. Patients with chondrosarcoma and adamantinoma did not receive any chemotherapy.

The response to the chemotherapy was evaluated by the classification of Salzer–Kuntschik [15] where response grade I is for no viable tumour cells, grade II is for some viable tumour cells or an area of viable tumour cells below 0.5 cm in diameter, grade III is less than 10% viable tumour cells, grade IV is 10–50% of viable cells, grade V is for over 50% of viable tumour cells and grade VI is no response to chemotherapy. For one patient (case 5) the response grade was missing.

Fig. 1 **a** Post-operative radiograph showing the resected area with a temporary cement spacer. **b** Ongoing bone transport with external fixation. **c, d** Final results after completed bone transport.

Functional results were assessed using the Casadei rating system [6], which is very similar to the Musculoskeletal Tumour Society (MSTS) rating system. However, it evaluates the problems in the ankle joint more precisely, namely union, pain, ambulation, deformity, psychological acceptance and functional activity, and complications are all listed.

Results

In all patients, resection was wide and no local recurrence had occurred. One patient (case 5) developed a distant metastasis to the third thoracic vertebrae, which was treated aggressively with adjuvant chemotherapy and additional

Table 1 Details of patients with distal tibial malignant tumours. *A* Treatment group A: allograft and microvascular fibula transfer, *B* treatment group B: bone transport, *C* treatment group C: only allograft, *D* treatment group D: only microvascular fibula transfer, *E*

treatment group E: endoprosthesis, *OS* osteosarcoma, *ES* Ewing's sarcoma, *CS* chondrosarcoma, *AD* adamantinoma, *n.b.a* not before amputation, *N.E.D.* no evidence of disease

Case	Age (years)	Gender	Primary histology	Stage	Response to chemotherapy ^a	Treatment	Follow-up (years)	Months to weight bearing (months)	Status	Casadei	Complication	Treatment of complications
1	35	M	OS	II-B	V	A	5.7	n.b.a.	N.E.D	Poor	Infection, necrosis	Amputation
2	16	F	OS	II-B	I	B	1.3	12	N.E.D	Good	Wound healing problems	Conservative
3	41	M	OS	II-B	II	C	1.4	9	N.E.D	Good	Wound healing problems	Conservative
4	13	F	OS	II-B	I	D	0.5		N.E.D	Fair	None	–
5	16	F	ES	II-B	Missing	D	14	n.b.a.	N.E.D	Poor	Infection	Amputation
6	37	M	ES	II-B	I	B	9.1	n.b.a.	N.E.D	Poor	Non-union of bone-transport, infection	Fibula transfer, amputation
7	16	M	OS	II-B	II	A	11.5	15	N.E.D	Fair	Wound healing problems	Conservative
8	11	F	ES	II-B	II	A	11.4	12	N.E.D	Poor	Shortening valgus deformity, infection, osteomyelitis	Bone transport, debridement
9	43	M	CS	I-B	–	C	10.9	12	N.E.D	Fair	None	–
10	14	F	OS	II-B	I	B	5.1	12	N.E.D	Good	None	–
11	20	M	AD	I-B	–	C (Total tibial allograft)	8.8	n.b.a.	N.E.D	Poor	Allograft fracture, Infection	Knee exarticulation
12	45	M	OS	II-B	V	Primary Amputation	9.2	–	N.E.D	–	None	–
13	13	F	OS	II-B	III	A	10.2	12	N.E.D	Fair	Allograft non-union, breakage of plate	Re-osteosynthesis
14	12	M	OS	II-B	II	A	2.7	16	N.E.D	Fair	Shortening, valgus deformity	Conservative
15	34	M	ES	II-B	V	E	2.9	6	N.E.D	Good	Wound healing problems	Conservative

^aAccording to Salzer and Kuntschik

wide surgery. Since then, the patient has remained free of disease.

In group A, the defect of the distal tibia was on average 16.4 cm (14.5–19 cm), and an arthrodesis of the ankle joint was performed in four patients. In group B, the defect was on average 12.8 cm (12.5–13 cm), and all patients had an arthrodesis. In the group C, the defect was on average 11.5 cm (11–12 cm), excluding the patient who received a total tibial allograft, and both patients had an arthrodesis. In group D, the defect was on average 13.5 cm (13–14 cm), and both patients had an arthrodesis. In group E, the whole tibia was resected and replaced with a prosthesis with functioning ankle joint.

Response to chemotherapy according to Salzer–Kuntschik was grade I in four patients, grade II in four, grade III in one and grade V in three. Details are summarised in Table 1.

Functional results according to Casadei are also presented in Table 1. The overall results were good in 4/14 patients: two were treated with bone transport, one with allograft alone and one with endoprosthesis. Results were fair or poor in 10/14 patients. Significant complications that led to amputation developed in four patients and in all, deep infection was the final cause of amputation.

In three patients, non-union of the bone transport or the allograft–host junction developed. In one patient (case 6), autologous microvascular fibula transport healed the non-union but was later infected. In one patient with non-union of the allograft–host junction (case 13) a re-osteosynthesis including autologous bone grafting ended up successfully. In one patient (case 8), shortening and severe valgus deformation followed the bone transport. However, callostasis and the docking region did not unite. After numerous surgical interventions, the patient ended up with osteomyelitis, which so far has been treated conservatively. Four patients experienced delayed wound healing and one patient shortening, including minor valgus deformity.

Mean duration until patients were allowed to carry full weight was 12 months. Patients with complications leading to amputation were never allowed full weight bearing. In one patient (case 4), partial weight bearing is still advised due to the short follow up.

Discussion

The purpose of the current study was to evaluate the oncological outcome and the advantages and disadvantages of different reconstruction methods. The analysis, although being one of the largest studies of distal tibial malignant bone tumours, is limited by the small number of patients and by the extended period of accrual. However, some guidelines can be discussed.

All patients were alive and free of disease at the end of the study. In the limb salvage patients, there was no local recurrence although most tumours were high-grade and in some patients the response grade was low. This finding is in agreement with previous reports [2, 17] observing that distal tibial malignant tumours may have a better prognosis compared with malignant tumours in other locations.

The reconstruction methods included bone transport, allograft alone or together with a vascularised fibular graft, fibula transfer alone and endoprosthetic replacement. In our series, bone transport offered the best functional results. In one case, the bone transport became infected and was later followed by an amputation. The resected limb had been irradiated post-operatively, and bone transport in irradiated bones is a controversial issue [7, 10]; our experience does not support this procedure. Bone transport is a demanding procedure in which many problems may occur. The number of problems increases as the length of the gap increases. Bone transport has generally been used with gap lengths less than 10 cm, but bone transport in gaps up to 20 cm has been reported [4, 13, 16]. In our series, the length of bone transport did not exceed 15 cm. In one patient, a larger defect was successfully treated with endoprosthetic replacement. Such procedures offer a functional and cosmetic limb, but long-term survival is questioned and the procedure is not recommended for children. Furthermore endoprostheses of the distal tibia are associated with many problems, such as talar collapse, limited function and stem loosening [1, 11, 12]. The inclusion of a trans-talar and trans-calcaneal stem improves primary rotational stability. This may secondarily improve osteointegration [8]. Results with the distal tibial endoprosthesis from MUTARS are promising [8], and prosthesis is therefore recommended for adult patients.

Our results of allograft reconstruction are inferior to those of previously reported allograft series. Casadei et al. [6] reported good results in 12 patients with distal tibial allograft arthrodesis. In our series, only one of eight had good results, in four the functional result was fair and in three poor. Infection, fracture and non-union were less prevalent in Casadei's study than in ours. Several factors may account for the differences: In our study, the larger resections for malignant tumour, the universal use of chemotherapy and the use of gamma irradiation of allografts may all have provoked the complications and subsequent failures.

Our results did not give any viable solution for the treatment of large defects in children. Special consideration should be given to the growth potential and the long-term durability. Although allograft reconstruction in our series was followed by complications, we believe that allograft with or without a vascularised fibula transfer is a viable option in children. Although amputation is associated with significant psychological, physiological and social costs to the patient, it still achieves a safe oncological margin and good functional results. Hence, amputation must be considered carefully in cases with large high-grade tumours in the distal tibia of children.

The major complication leading to secondary amputation was infection. It occurred in 29% of cases, which is a high infection rate. The use of local rotational flaps has been shown to effectively diminish infection in tumour surgery [9]. However, in the distal tibia, there are no obvious rotational flaps. Free-muscle flap, either primarily or at the earliest sign of wound healing problems, is also a valuable tool and may decrease the infection rate.

Reconstruction after limb-sparing surgery for high-grade bone sarcoma in the distal tibia is challenging. Bone transport is a viable option in defects shorter than 15 cm. In larger defects in adult patients, endoprosthetic replacement may give good results. In children, allograft with vascularised fibula transfer or fibula transfer alone provides an acceptable alternative, but here again bone transport seems promising [2, 13, 16].

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