

Bone transport for reconstruction of large bone defects after tibial tumor resection: a report of five cases

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Abstract

This study was performed to explore the clinical efficacy of bone transport using external fixation for treatment of large bone defects after tibial tumor resection in five patients. Bone transport started 14 days postoperatively at 1 mm/day and was adjusted according to the callus-to-diameter ratio. The bone transport time, bone graft fusion, relapse, and metastasis were recorded. Clinical efficacy was evaluated using the Musculoskeletal Tumor Society (MSTS) scoring system. The tumors included osteosarcoma (n=2), Ewing sarcoma (n=1), malignant schwannoma (n=1), and hemangioma (n=1). The average bone defect length after resection was 11.6 cm. The five patients were followed up for an average of 50.8 months, and the average bone transport time was 15.5 months. Three patients who underwent postoperative chemotherapy were followed for 22.7 months, and two who did not undergo chemotherapy were followed for 4.75 months. Four patients underwent iliac bone grafting, and one underwent vascular pedicle fibular transplantation. The average MSTS score was 21.2 (19.3 for patients who underwent chemotherapy and 24.0 for patients who did not). No relapse or metastasis was observed. Bone transport is effective for reconstruction of large bone defects after tibial tumor resection as well as tibial malignancies with high doses of chemotherapy.

Keywords

Bone transport, large bone defect, reconstruction, tibia, tumor, external fixation

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Introduction

Reconstruction of large bone defects after tibial tumor resection is challenging. Because of the poor coverage of soft tissue in the tibia, especially in the middle and lower tibia, large bone defects result in a variety of postoperative complications after reconstruction. Clinical treatment experience is also limited due to the low incidence of tibial tumors.¹ Reconstruction methods such as tumor-type prostheses or allogeneic bone grafts are often associated with complications including infection and instability.^{2,3} Fibular surgery using vascular pedicles requires complex microsurgical skills and achieves poor mechanical stability.⁴ Bone transport has been widely used to treat large segmental bone defects after traumatic surgery.⁵ This technique is less risky and is associated with fewer postoperative complications and better long-term outcomes after the biological reconstruction has healed.⁶ Limited studies have shown that bone transport can be applied for reconstruction of large bone defects after tumor resection.^{7,8} However, whether high-dose chemotherapy of malignant tumors will affect the long-term outcomes of this procedure remains unknown. To address this knowledge gap, we herein present five clinical applications of bone transport for reconstruction of large tibial bone defects including one case of Ewing sarcoma and two cases of osteosarcoma resection with chemotherapy.

Case reports

From January 2011 to March 2013, five patients (two male, three female) with malignant tibial tumors (periosteal osteosarcoma, osteosarcoma, Ewing sarcoma, malignant schwannoma, and hemangioma) were treated at our hospital. Written informed consent was obtained from the patients for publication of their cases,

including any necessary photographs. The surgical staging of all tumors was IIB, except for the hemangioma (G0T2M0).⁹ The median age of the patients was 25 years (range, 10–65 years). The median length of the resected tibial segment was 12 cm (range, 8–15 cm) (Table 1). The study protocol was approved by the ethics review committee of the Second Affiliated Hospital, School of Medicine, Zhejiang University.

Four patients were treated with a uniplanar unilateral external fixator (Orthofix, Lewisville, TX, USA), and one patient was treated with ring external fixation. Two patients underwent both preoperative and postoperative chemotherapy, one patient underwent postoperative chemotherapy only, and two patients underwent no chemotherapy. High-dose methotrexate (12 g/m²) or ifosfamide (1.8 g/m²), cisplatin (120 mg/m²), and adriamycin (60 mg/m²) were used for chemotherapy.

The callus-to-diameter ratio (CDR) was calculated as the diameter of the callus divided by the diameter of the original diaphysis.¹⁰ The distraction rate was reduced from 1.0 to 0.5 mm at 0.25 mm/day if the CDR was <80%. Follow-up observation of the bone turnover time, bone graft fusion rate, tumor recurrence, metastasis rate, and Musculoskeletal Tumor Society (MSTS) functional score¹¹ were used to evaluate clinical efficacy and outcomes.

The average bone transport time for all patients was 15.5 months (range, 4.5–28 months). The average bone transport time for patients who underwent chemotherapy was 22.7 months (range, 16–28 months). The average bone transport time for patients who did not undergo chemotherapy was 4.75 months (range, 4.5–5 months). Four patients had iliac bone grafts and one underwent vascular pedicle fibular transplantation. The average MSTS score for all patients was 21.2 (range, 18–26). The average MSTS score for

Table 1. Patient characteristics and treatment outcomes

Patient	Sex	Age (y)	Pathologic diagnosis	Location within tibia	Surgical stage	Chemotherapy	Bone defect (cm)	Bone migration (mo)	Follow-up (mo)	MSTS score	Graft or transplantation	Nails used (n)
1	M	29	Periosteal osteosarcoma	Proximal	II B	Postoperative	11	16	51	22	Iliac bone graft	2 nails at both proximal and distal ends
2	F	16	Osteoblastoma-like osteosarcoma	Distal	II B	Pre- and postoperative	15	28	56	18	Iliac bone graft	2 nails at both proximal and distal ends
3	M	10	Ewing sarcoma	Distal	II B	Pre- and postoperative	12	24	65	18	Fibular transplantation	2 nails at both proximal and distal ends
4	F	25	Malignant schwannoma	Distal	II B	None	8	5	42	22	Iliac bone graft	2 nails at proximal end, 3 nails at distal end
5	F	65	Hemangioma	Middle	G0T2M0	None	12	4.5	40	26	Iliac bone graft	Ring external fixator and 2 nails each at distal and proximal end

M, male; F, female; MSTS, Musculoskeletal Tumor Society

patients who underwent chemotherapy was 19.3 (range, 18–22), and that for patients who did not undergo chemotherapy was 24.0 (range, 22–26).

The average follow-up time was 50.8 months (range, 40–65 months). Four patients developed complications including nail infection, exudation, and loosening. One patient had a wound that did not heal at the junction of the vascular pedicle fibular graft and tibia, but ankle fusion was achieved. This patient was able to walk without further surgery. One patient developed a weak traction callus, and re-implantation was performed. No recurrence or metastasis was observed. Two typical cases are illustrated in Figures 1 and 2.

Discussion

Osteotomy is generally performed proximal to the metaphysis because the cancellous bone is rich in blood and strongly osteogenic. Performing osteotomy close to the metaphysis is beneficial for healing during bone transport. Periosteal integrity should be maintained during osteotomy to facilitate bone formation during bone transport. The timing and speed of bone transport are also important. Reports of bone transport after surgical treatment of trauma suggest that a distraction length of 1.0 mm/day is appropriate in most cases. Ilizarov^{12,13} reported that a distraction length of 1.0 mm/day is optimal because 0.5 mm/day can lead to premature bone healing, while 2.0 mm/day produces only fibrous connective tissue at the bone ends without osteogenesis. However, in patients with cancer, particularly patients undergoing chemotherapy, a distraction length of 1.0 mm/day cannot be fully applied because chemotherapy affects osteogenesis and often requires adjustment of the distraction setting. This is the main difference in the application of bone transport between patients with trauma and those with cancer.

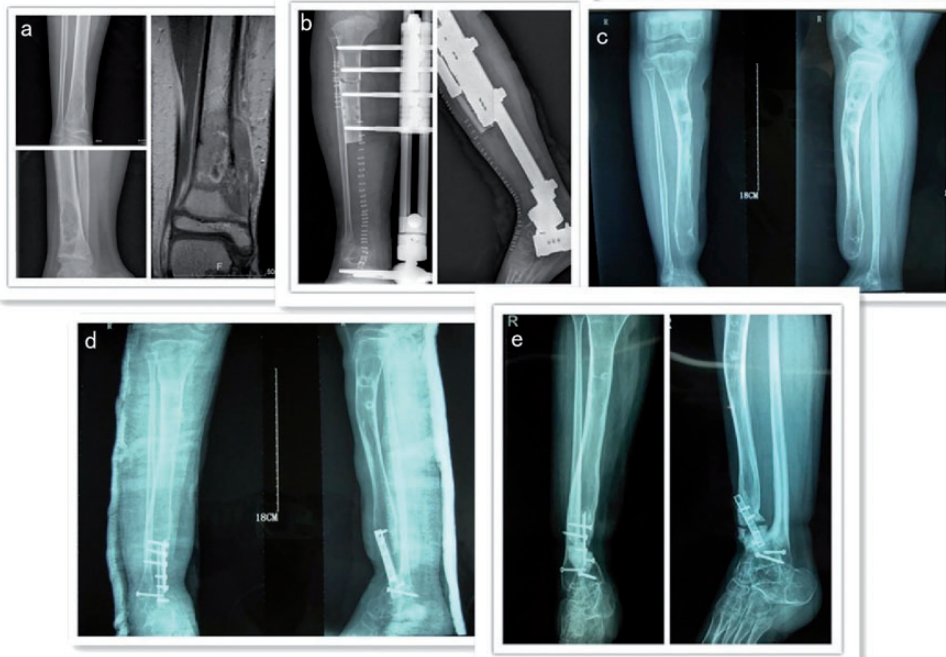


Figure 1. Patient 3. A 10-year-old boy with juvenile sarcoma in the lower end of the right tibia was admitted due to a 3-month history of ongoing swelling and pain in the right leg. (a) A preoperative radiograph and magnetic resonance image showed right lower tibial bone destruction with a soft tissue mass and no metastasis; percutaneous biopsy suggested Ewing sarcoma in the right lower tibia. The patient underwent two cycles of chemotherapy preoperatively and postoperatively. (b) After excision of a large bone segment, external superarticular fixation with an Orthofix single arm was performed on the 12-cm bone defect; bone transport was initiated on postoperative day 14 at a distraction rate of 1 mm/day twice daily. Monthly radiographs were taken, and the distraction rate was adjusted according to the callus-to-diameter ratio. (c) Bone defects of about 2.8 cm were present for 24 months after bone transport. (d) Vascular pedicle fibular grafting was performed. (e) Five months after transplantation, the fibula fused with the lateral malleolus and talus, forming a pseudarthrosis with the tibia. The patient could walk with no further surgery. The follow-up time was 65 months with no recurrence or metastasis. The patient's Musculoskeletal Tumor Society functional score was 18

The advantages of bone transport for treatment of tibial tumors after a large bone defect has been removed are as follows. First, the bone transport method is easy to perform. It is relatively simple and requires a shorter surgery than other reconstruction methods such as the use of a tumor-type prosthesis, allogeneic bone, and vascular pedicled fibula. Second, the surgical risks associated with bone transport are minimal because this method

involves no complicated internal fixation or prosthesis foreign body at the wound site. These factors reduce the risk of wound infection, and when wound infection does occur, it is easy to control. Third, this technique produces stable and strong bones of the appropriate length, providing patients with a better quality of life. Fourth, bone transport is a biological reconstruction technique in which a large segment of active bone formation provides sufficient



Figure 2. Patient 5. A 65-year-old woman with a right middle tibial hemangioma was admitted due to a 2-month history of right leg pain. (a) A preoperative radiograph and magnetic resonance image showed left middle tibial osteolytic destruction and a soft tissue mass with no metastasis. (b) After excision of a bone segment, external fixation with an Orthofix ring was performed on the 12-cm bone defect; bone transport was initiated 14 days postoperatively at a speed of 1 mm/day on each side. Monthly radiographs were taken to adjust the traction speed according to the callus-to-diameter ratio. (c) Follow-up was performed 4.5 months after bilateral bone transport. (d) The external fixation strut was removed with the iliac bone graft and gypsum external fixation. (e) Bone graft fusion occurred after 3 months. The patient was followed up for 40 months with no recurrence or metastasis. The Musculoskeletal Tumor Society score was 26

strength and stability. This reconstruction method can provide a long-term or even lifelong solution while reducing possible second-phase renovation of the bone for the prosthesis or allogeneic graft.

Bone transport is not without limitations. First, the treatment time can be lengthy, especially in patients with malignant tumors who require high doses of chemotherapy. The bone transport speed and traction frequency may be low, resulting in a long transport time. In the present study, patients who underwent chemotherapy experienced a relatively longer transport time than patients who did not undergo

chemotherapy. The impact of chemotherapy on bone transport remains controversial. Watanabe *et al.*⁶ reported that chemotherapy has no effect on distraction osteogenesis, but they did not use a high-dose chemotherapy regimen in patients with malignant tumors. Our experiences suggest that high-dose methotrexate, cisplatin, and doxorubicin may inhibit bone formation, promote bone deterioration, and prolong the osteogenesis time, resulting in decreased bone transport speed and stretch frequency and even may even cause callus distress osteogenesis failure. These outcomes could be attributed to the effect of chemotherapy on

growth of local blood vessels. Fortunately, however, chemotherapy does not affect the final osteogenesis. Chemotherapy can also reduce the limb force and decrease muscle strength. The occurrence of varying degrees of clubfoot in patients who have undergone bone transport could be attributed to the length of the bone transport or the migration of the muscle end point. Clubfoot might also be related to the instability of the single-arm external fixation extension. Our experiences suggest that the use of a ring external fixator could improve traction stability. The patients in this study were effectively fixed with a ring-shaped strut, although the strut was larger, which may be inconvenient in daily life. Unfortunately, after vascular pedicle fibular transplantation in Patient 3, a false joint formed between the tibia and fibula because of hypertrophic nonunion and caused instability; however, it finally fused with the ankle. The patient was able to walk without further surgery. We did not observe any tumor relapse or metastasis in association with bone transport. This result is also consistent with the findings of other studies, although our sample size is limited.¹⁴ Lou *et al.*¹⁵ reported local recurrence of a sarcoma in one of five patients because of a poor response to chemotherapy. We also believe that the rate of tumor relapse is more closely related to the degree of tumor resection than to the bone transport procedure. If the bone defect is close to the joint, less space is available for fixing the nails, and the patient is more suitable for joint fusion surgery.

Our experiences suggest that bone transport can be applied in patients with bone defect lengths of ≤ 15 cm, the ability to perform fixation at the distal segment after tumor excision, a sufficient length at one side of the bone for bone transport, no local infection, and acral tissue integrity. Similarly, our results are consistent with the findings of other studies.¹⁴⁻¹⁸ Patients with extensive tumor resection or resection

involving vascular nerves, lung metastases, or other extrapulmonary metastases may not be suitable for bone transport. Bone transport is an effective biological remodeling method for the treatment of bone defects after tibial tumor resection. It offers a possible alternative for patients with tibial malignancies requiring high-dose chemotherapy.

In this case report, four of five patients with large bone defects after tibial tumor resection were successfully treated with bone transport. The bone transport time in the first three patients who underwent chemotherapy was longer than that in the two patients who did not undergo chemotherapy. Further studies with more clinical cases and a control group are needed to confirm our findings.

Declaration of conflicting interest

The authors declare that there is no conflict of interest.

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