Original Article
Clinics in Orthopedic Surgery 2015;7:248-253 • http://dx.doi.org/10.4055/cios.2015.7.2.248

Bone Transport for Reconstruction in Benign Bone Tumors

Chang Seon Oh, MD, Sung Taek Jung, MD, Yong Jin Cho, MD*, Yeong Seub Ahn, MD, Bo Ram Na, MD

Department of Orthopaedic Surgery, Chonnam National University Hospital, Chonnam National University Medical School, Gwangju,
**Department of Orthopaedic Surgery, Yonsei University College of Medicine, Seoul, Korea*

Background: The aim of this study was to assess the results of using the Ilizarov apparatus to transport bones in the treatment of benign bone tumors.

Methods: Seven patients (six males and one female) with benign bone tumors were treated by bone transport with an Ilizarov apparatus at our institution. Their mean age at surgery was 14.4 years (range, 4.8 to 36.9 years). The histological diagnoses were osteofibrous dysplasia (4), giant-cell tumor (1), intraosseous cavernous hemangioma (1), and aneurysmal bone cyst (1). Three radiological indices were used for evaluating the results: an external fixation index, a distraction index, and a maturation index. The bone and functional results were evaluated according to the Association for the Study and Application of the Method of Ilizarov classification.

Results: Five patients had bone union at the reconstructed site, one patient had a local recurrence, and the other had a nonunion at the docking site. The mean length of distraction was 7.3 cm (range, 5.1 to 12.1 cm). The mean external fixation index was 26.0 day/cm (range, 19.8 to 32.5 day/cm), the distraction index was 9.6 day/cm (range, 6.8 to 12.0 day/cm), and the maturation index was 14.9 day/cm (range, 8.0 to 22.5 day/cm). Ultimately, the bone and the functional results were rated excellent in six cases and good in one case.

Conclusions: Bone transport using the Ilizarov apparatus is a good treatment option in patients with bone defects after the resection of an active or aggressive benign bone tumor.

Keywords: *Bone neoplasms, Ilizarov technique, Bone lengthening*

Benign bone tumors are seldom aggressive, but grow rapidly. They can be treated by curettage or a simple lesion resection and consequently, there are few cases of reconstruction. However, some benign bone tumors, like giant-cell tumors, must be resected in order to provide a safe margin to prevent local recurrence. This causes a loss of bone stock, and therefore, reconstruction is required.

Distraction osteogenesis with the Ilizarov apparatus is widely used for the treatment of several orthopedic problems, such as leg length discrepancy (LLD), deformity, osteomyelitis, and congenital or acquired skeletal defects.[1-3] Recently, it has also been used for the treatment of bone defects caused by trauma[4,5] and bone tumor resection.[6,7]

The aim of this study was to assess the results and the indication of treating bone defects in patients with benign bone tumors through bone transport using the Ilizarov apparatus.

METHODS

Between 1997 and 2006, seven patients (six males and one female) with benign bone tumors were treated with bone transport using the Ilizarov apparatus in our institu-

Received October 3, 2014; Accepted February 22, 2015
Correspondence to: Sung Taek Jung, MD
Department of Orthopaedic Surgery, Chonnam National University Hospital, Chonnam National University Medical School, 42 Jebong-ro, Dong-gu, Gwangju 501-757, Korea
Tel: +82-62-227-1640, Fax: +82-62-225-7794
E-mail: stjung@jnu.ac.kr

Table 1. Patient Demographics

Case	Age (yr)	Sex	Diagnosis	Site	Length of defect (cm)	Defect ratio (%)	Follow-up (mo)
1	10.2	Male	Osteofibrous dysplasia	Proximal tibia	5.1	16.0	96.1
2	12.2	Male	Osteofibrous dysplasia	Proximal tibia	5.1	14.0	111.2
3	7.2	Male	Osteofibrous dysplasia	Tibial shaft	5.6	18.7	141.1
4	4.8	Male	Osteofibrous dysplasia	Tibial shaft	12.1	52.4	180.5
5	14.1	Male	Intraosseous hemangioma	Tibial shaft	8.8	21.9	64.4
6	15.8	Female	Aneurysmal bone cyst	Distal femur	8.1	18.9	115.5
7	36.9	Male	Giant-cell tumor	Distal femur	6.2	12.9	34.6

tion. Their mean age at surgery was 14.4 years (range, 4.8 to 36.9 years) and the mean follow-up period was 106.2 months (range, 34.6 to 180.5 months).

The histological diagnoses included four cases of osteofibrous dysplasia, one giant-cell tumor, one intraosseous cavernous hemangioma, and one aneurysmal bone cyst. The lesions were located in the proximal tibia in two cases, the tibial shaft in three cases, and the distal femur in two cases (Table 1). The mean defect ratio was calculated using the corticotomy method; dividing the affected long bone length by the length of the defect.

Bifocal bone transports were performed in six cases, and a trifocal bone transport was performed in the remaining cases. Bone transport with diaphyseal reconstruction was performed in five cases (tibia), and shortening-distraction with metaphyseal reconstruction was done in two cases (distal femur). The reconstruction procedures were classified *via* distraction osteogenesis.[8]

The distraction was started after a latent period of 8.6 days,[6-11] the distraction rhythm was set at four times, and the length was 0.5 or 1 mm per day. The distraction settings were adjusted according to the quality of bone formation as seen in the serial radiographs during follow-up. Partial weight-bearing was allowed after the full length was reached, and the external fixator was removed after sufficient cortex development had been observed.

We used three indices for evaluating the results: the external fixation index, which was obtained by dividing the duration of external fixation by the length of bone regeneration; the distraction index, which was obtained by dividing the duration of distraction by the length of bone regeneration; and a maturation index, which was obtained by dividing the duration of external fixation, measured from the completion of distraction to the removal of external fixation, by the length of bone regeneration.

The functional and bone results were evaluated according to the Association for the Study and Application of the Method of Ilizarov (ASAMI) classification (Table 2).[4-9]

Table 2. Association for the Study and Application of the Method of Ilizarov Classification

Bone result
Criteria: union, infection, deformity, lower limb discrepancy
Excellent: union, no infection, deformity < 7°, lower limb discrepancy
Good: union plus any two of the others
Fair: union plus one of the others
Poor: nonunion or refracture or none of the others
Functional result
Criteria: significant limping, joint contracture, soft tissue dystrophy, pain and inactivity
Excellent: active individual with none of the other criteria
Good: active individual with one or two of the other four criteria
Fair: active individual with three or four of the other criteria or an amputation
Poor: inactive individual

RESULTS

The mean length of the defect after tumor resection was 7.3 cm (range, 5.1 to 12.1 cm) and the mean defect ratio was 22.1% (range, 12.9% to 52.4%), which is shown in Table 1. The length of distraction was determined by the patient's age, bone age, and unaffected limb length. The mean length of distraction was 6.9 cm (range, 4.9 to 11.0 cm). The external fixation was applied at 185.6 days (range, 106 to 266 days). The mean external fixation index was 26.0 day/cm (range, 19.8 to 32.5 day/cm), the distraction index was 9.6

Table 3. Treatment Results

Case	EF period (day)	EF index (day/cm)	Distraction index (day/cm)	Maturation index (day/cm)	LLD	Complication	ASAMI	
							Bone	Function
1	151	29.6	10.2	17.6	0.2	Recurrence	Excellent	Excellent
2	106	20.8	11.2	8.0	1.2	Nonunion	Good	Excellent
3	160	28.6	12.0	14.6	1.6	-	Excellent	Excellent
4	266	22.0	7.8	12.8	1.1	SI	Excellent	Excellent
5	174	19.8	6.8	11.3	1.1	-	Excellent	Excellent
6	263	32.5	9.0	22.5	2.7	-	Excellent	Good
7	179	28.9	10.3	17.4	0.1	-	Excellent	Excellent

EF: external fixation, LLD: leg length discrepancy, ASAMI: Association for the Study and Application of the Method of Ilizarov, SI: superficial infection.

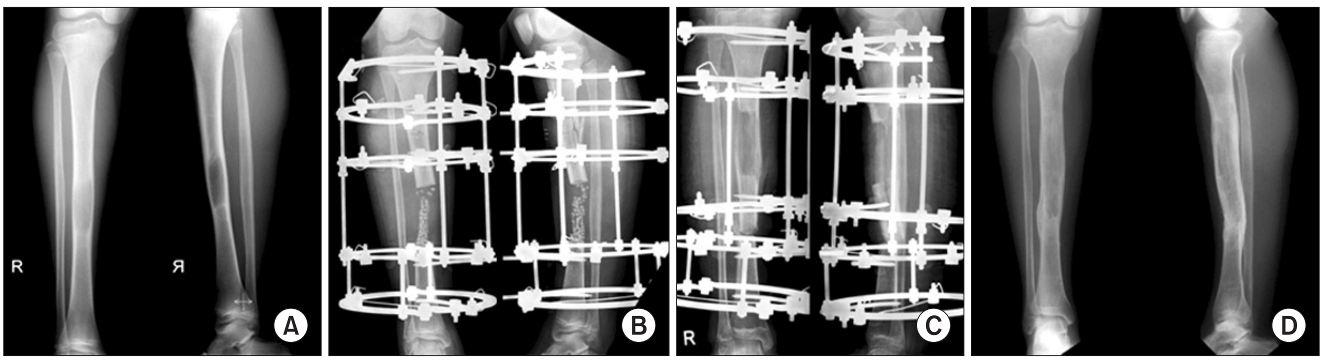

Fig. 1. (A) A 14-year-old male with an osteolytic lesion of the tibia shaft. (B) We resected the lesion with calcium sulfate grafting and applied the Ilizarov apparatus. The histological diagnosis was cavernous hemangioma of the bone. (C) Distraction was done. (D) Union of the transported bone was achieved.

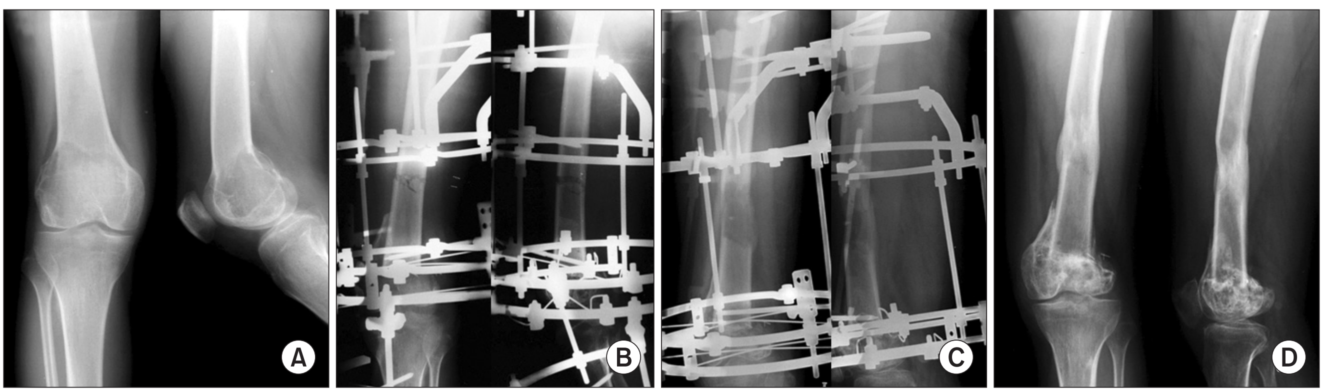

Fig. 2. (A) A 36-year-old male patient with an osteolytic lesion of the distal femur. (B) Tumor was resected and proximal femur was docked to the distal femur. The histological diagnosis was giant cell tumor. (C) Lengthening was done at proximal from the docking site. (D) Docking site and distracted bone union was achieved.

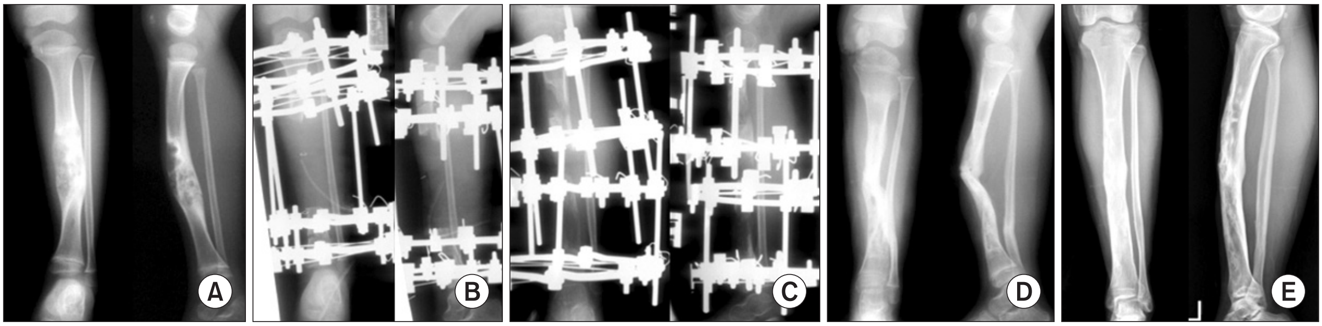

Fig. 3. (A) A 4-year-old male patient with an osteolytic lesion of the tibia shaft. (B) Tumor was resected with tricorticotomy. The histological diagnosis was osteofibrous dysplasia. (C) Bone transport was done at proximal and distal corticotomy site. (D) Bone union and angular deformity. For protection of stress fracture, we applied long leg splint. (E) The last follow-up radiograph shows the remodeled tibia without recurrence.

day/cm (range, 6.8 to 12.0 day/cm) and the maturation index was 14.9 day/cm (range, 8.0 to 22.5 day/cm) (Table 3). All the patients were able to perform their daily activities without problems. The bone and functional results were rated excellent in six cases and good in one case. The radiographs of representative cases are shown in Figs. 1-3.

There were complications in three patients, including pin tract infection, local recurrence, and nonunion. The pin tract infection was treated successfully with oral antibiotics for one week. One patient had local recurrence of the tumor due to an insufficient resection. The patient was subsequently treated with a tumor resection and a fibular autogenous bone graft. Nonunion occurred in one patient and was treated by osteosynthesis with a fibular autogenous bone graft. There was no instance of LLD over 3 cm shortening (mean, 1.1 cm; range, 0.1 to 2.7 cm) at the final follow-up.

DISCUSSION

Bone defects are a common orthopedic problem, resulting from trauma or malignant or benign bone tumors. Malignant bone tumors can lead to bone defects, soft tissue defects, and limb length discrepancy after a surgical resection. After the malignant bone tumor is resected, there are many modalities of treatment to reconstruct the defect, including allografts, autoclaved autografts, vascularized fibular or iliac bone grafts, irradiated bone grafts, prosthesis, and distraction osteogenesis.[6,7,10,11] Each technique has its own advantages and disadvantages.

Allografts are a commonly used technique for the reconstruction of bone defects after tumor resections. The technique is beneficial because of biology and the graft is customized to the proper size and form. However, there are also some disadvantages, including the potential transmission of a disease, allograft rejection, late fracture, infection, and nonunion.

For this reason, autogenous bone grafts are also commonly used for reconstruction. There are advantages to this solution, such as the capability to cover long defect lengths, a resistance to infection, and its biological properties. Laffosse et al.[12] reported in a study of 13 patients with long bone defects that autologous vascularized fibular graft treatment yielded good union results. However, there are some disadvantages, including the fact that it is a technically demanding procedure, takes a long time, graft fracture, nonunion, and size limitations.

Prosthetic replacement is a useful reconstructive method after resection a malignant bone tumor, but there are few reports of its use in benign bone tumors. Shin et al.[13] reported that prosthetic replacements for aggressive benign bone tumors are a good treatment option for reconstruction of the bone defect. However, late complications such as infection, loosening, and breakage are common and troublesome.

Recently, distraction osteogenesis using external fixation has been reported as a management technique for posttraumatic bone defects[4,5] and bone defects after malignant tumor resection.[6,7] There are many advantages to this technique, such as biomechanical stability, a bloodless technique, regeneration of new bone, and a gradual lengthening of the soft tissues.[14] Therefore, we tried to use the bone transport technique with the Ilizarov apparatus to reconstruct bone defects after resecting benign bone tumors and saw good results.

Karita et al.[15] treated two patients with osteofibrous dysplasia using *en bloc* marginal excision of the lesion and bone transport, a distraction osteogenesis procedure. They reported good results and recommended the treatment as a way to reconstruct large bone defects after tumor resection.

Tsuchiya et al.[14] also reported good functional results in 10 patients with benign bone tumors of the extremities that were treated by resecting the tumor and reconstructing with external fixation. They recommended an Ilizarov frame or Taylor spatial frame instead of unilateral fixators because limb-length discrepancy and deformity can be treated simultaneously with these frames. In this study, we use an Ilizarov apparatus for reconstruction in all cases.

The mean defect ratio was 17.1% (range, 12.9% to 21.9%), except for one outlier with the largest defect (52.4%). It is not the same condition in children because the total limb length is shorter than in adults. We used the trifocal bone transport in one case and its defect ratio was 52.4%. Trifocal bone transport is used in cases of longer defects as compared to bifocal bone transport.[16] If we use the trifocal bone transport when there is a large defect ratio, we can shorten the period of treatment. But, we only have one such case, so we cannot make conclusions about the indication of trifocal bone transport. Thus, more research about the correlation between the defect ratio and bifocal or trifocal bone transport is needed. The most common complication of external fixation is a pin track infection. In our cases, one case had a pin track infection that was managed by oral antibiotics.

Nonunion or refracture are not rare. The external fixation period is important for healing distracted bones because it promotes stability and maturation. We had one case of nonunion and reviewed the maturation index. The mean maturation index was 14.9 day/cm but only 8.0 day/cm in the case of nonunion. If the duration of the external fixator is too short, the limited maturation time can lead to the instability of the distracted bone, causing nonunion or refracture. Therefore, it is important that external fixation is maintained for maturation of distracted bone. Also Green et al.[17] recommended bone grafting at the docking site in order to shorten the period of treatment and promote the rate of union formation. In our cases, we did an autogenous bone graft at the docking site in all cases and all but one case achieved a union.

The mean LLD was 1.1 cm at last follow-up. The case of the longest length discrepancy is a pathologic fracture with an aneurismal bone cyst in the distal femur. The lengthening after shortening was done, but we could not get a full lengthening because of leg pain. In the other cases, lengthening was stopped after correction of LLD. The residual LLD was caused by stopping the lengthening early and occurred during growth. Long periods of external fixation can be uncomfortable for the patient. Today, we can shorten the period of external fixation by converting to internal fixation.[18,19] Internal fixation greatly reduces the patient's time in external fixation during the consolidation phase, gives additional stability to protect the tibia against refracture, and is expected to prevent nonunions.

Bone transport using the Ilizarov apparatus is a good treatment option in patients with bone defects after resection of a benign bone tumor. It offers a good alternative to other conventional methods. Furthermore, bifocal or trifocal osteotomies can be used to reconstruct large bone defects. Some complications such as a pin track infection, nonunion, and local recurrence still need to be addressed.

Bone transport is only indicated in limited cases, not in all benign bone tumors. We think this is the most suitable course of treatment for active or aggressive benign bone tumors where a large bone defect is expected, but more study is needed.

CONFLICT OF INTEREST

No potential conflict of interest relevant to this article was reported.

REFERENCES

1. Ilizarov GA. The tension-stress effect on the genesis and growth of tissues. Part I. the influence of stability of fixation and soft-tissue preservation. Clin Orthop Relat Res. 1989;(238):249-81.

2. Ilizarov GA. The tension-stress effect on the genesis and growth of tissues: Part II. The influence of the rate and frequency of distraction. Clin Orthop Relat Res. 1989;(239):263-85.

3. Ilizarov GA, Green SA. The transosseous osteosynthesis: theoretical and clinical aspects of the regeneration and growth of tissue. Berlin: Springer-Verlag; 1992.

4. Song HR, Cho SH, Koo KH, Jeong ST, Park YJ, Ko JH. Tibial bone defects treated by internal bone transport using the Ilizarov method. Int Orthop. 1998;22(5):293-7.

5. Robert Rozbruch S, Weitzman AM, Tracey Watson J, Freudigman P, Katz HV, Ilizarov S. Simultaneous treatment of tibial bone and soft-tissue defects with the Ilizarov method. J Orthop Trauma. 2006;20(3):197-205.

6. Shalaby S, Shalaby H, Bassiony A. Limb salvage for osteosarcoma of the distal tibia with resection arthrodesis, autogenous fibular graft and Ilizarov external fixator. J Bone Joint Surg Br. 2006;88(12):1642-6.

7. Watanabe K, Tsuchiya H, Yamamoto N, et al. Over 10-year follow-up of functional outcome in patients with bone tumors reconstructed using distraction osteogenesis. J Orthop Sci. 2013;18(1):101-9.

8. Tsuchiya H, Tomita K, Minematsu K, Mori Y, Asada N, Kitano S. Limb salvage using distraction osteogenesis: a classification of the technique. J Bone Joint Surg Br. 1997;79(3):403-11.

9. Paley D, Catagni MA, Argnani F, Villa A, Benedetti GB, Cattaneo R. Ilizarov treatment of tibial nonunions with bone loss. Clin Orthop Relat Res. 1989;(241):146-65.

10. Kapukaya A, Subasi M, Kandiya E, Ozates M, Yilmaz F. Limb reconstruction with the callus distraction method after bone tumor resection. Arch Orthop Trauma Surg. 2000;120(3-4):215-8.

11. Nishida J, Shimamura T. Methods of reconstruction for bone defect after tumor excision: a review of alternatives. Med Sci Monit. 2008;14(8):RA107-13.

12. Laffosse JM, Accadbled F, Abid A, Kany J, Darodes P, Sales De Gauzy J. Reconstruction of long bone defects with a vascularized fibular graft after tumor resection in children and adolescents: thirteen cases with 50-month follow-up. Rev Chir Orthop Reparatrice Appar Mot. 2007;93(6):555-63.

13. Shin KH, Park HJ, Yoo JH, Hahn SB. Reconstructive surgery in primary malignant and aggressive benign bone tumor of the proximal humerus. Yonsei Med J. 2000;41(3):304-11.

14. Tsuchiya H, Morsy AF, Matsubara H, Watanabe K, Abdel-Wanis ME, Tomita K. Treatment of benign bone tumours using external fixation. J Bone Joint Surg Br. 2007;89(8):1077-83.

15. Karita M, Tsuchiya H, Sakurakichi K, Tomita K. Osteofibrous dysplasia treated with distraction osteogenesis: a report of two cases. J Orthop Sci. 2004;9(5):516-20.

16. El-Alfy B, El-Mowafi H, Kotb S. Bifocal and trifocal bone transport for failed limb reconstruction after tumour resection. Acta Orthop Belg. 2009;75(3):368-73.

17. Green SA, Jackson JM, Wall DM, Marinow H, Ishkanian J. Management of segmental defects by the Ilizarov intercalary bone transport method. Clin Orthop Relat Res. 1992;(280):136-42.

18. Oh CW, Shetty GM, Song HR, et al. Submuscular plating after distraction osteogenesis in children. J Pediatr Orthop B. 2008;17(5):265-9.

19. Kocaoglu M, Eralp L, Bilen FE, Balci HI. Fixator-assisted acute femoral deformity correction and consecutive lengthening over an intramedullary nail. J Bone Joint Surg Am. 2009;91(1):152-9.