RESEARCH ARTICLE

Lower Limb Reconstruction Using Tibial Strut Autograft after Resection of Primary Malignant Bone Tumors in Skeletally Immature Patients

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Abstract

Background: Reconstruction of large bone defects in skeletally immature patients remains a surgical challenge. We report the long-term clinical outcomes of a novel surgical technique for lower limb reconstruction using the tibia as a strut autograft following resection of primary malignant bone tumors in skeletally immature patients.

Methods: We retrospectively reviewed the medical records of six patients diagnosed with lower limb primary bone sarcoma. All patients underwent tumor resection and reconstruction using tibial strut autograft. The radiological and clinical outcomes including complications at the recipient and donor sites were assessed.

Results: The mean age at presentation was ten years (range 6-15 years). Two cases had osteosarcoma and four had Ewing sarcoma. The mean length of the resected tumor and tibial autografts were 20.83 and 19.33 cm respectively. Union at both ends was achieved in five grafts while one graft achieved union only at the distal end. The mean time for union of the proximal and distal junctions was 4 and 8.8 months respectively. The mean follow-up period was 8.4 years (range 14 months–20 years). One patient developed a foot drop, and three patients underwent subsequent joint arthrodesis (2 knees and 1 ankle). The mean musculoskeletal tumor society functional score was 80.8%. Two patients had clinically significant leg-length discrepancy that needs further lengthening procedure. Four patients survived with no evidence of disease and two patients died due to their primary oncologic disease. All donor sites regenerated, with the earliest signs of new bone formation at (2-4) weeks post-operatively.

Conclusion: Reconstruction using non-vascularized tibia strut autograft after resection of primary malignant lower limb bone tumors can be a viable alternative method for reconstructing large bone defects in the immature skeleton.

Level of evidence: IV

Keywords: Autograft, Bone tumor, Limb, Outcomes, Reconstruction, Tibia

Introduction

In the last three decades, the advances in the management of primary malignant bone tumors have led to an improvement in patient survival and prognosis. This was attributed to several factors including early diagnosis, effective chemotherapy, accurate preoperative imaging studies, and well-performed tumor resections (1,2). In parallel, the proportion of limb salvage and reconstruction procedures has increased and replaced amputations for primary malignant bone tumors (1). Nearly 90% of patients with extremity bone sarcomas are considered candidates for limb salvage surgery without having an increased risk of local recurrence (3).

Corresponding Author: Mohammed S. Alisi, The University of Jordan, School of Medicine, Queen Rania Al Abdullah St, Al Jubaiha, Amman, Jordan Faculty of Medicine, Islamic University of Gaza, Gaza, Palestine Email: m.elessi2007@hotmail.com Reconstruction options in children after primary malignant bone tumor resections are as varied as they are challenging. Advances in biologic and endoprosthetic design have led to many choices; all considered in the context of prognosis, treatment limitations, and patient/ family expectations (2).

In general, the reconstructive procedures have been chosen considering several factors such as the location and local involvement of the tumor, histologic type, biological behavior of the tumor, life expectancy, metastatic status, and predicted function of the limb (4). The lack of soft tissue coverage, especially in the middle and lower tibia, for large bone defects may



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result in a variety of postoperative complications after reconstruction. This will prompt physicians to look for an appropriate function-preserving alternative after massive resection of such tumors and affected soft tissues (5).

The surgical options for reconstruction in metaphyseal/ metaphyseal-diaphyseal tumors in adolescent patients usually include special endoprostheses that providesatisfactory results shortly after surgery (6). Extendable or expandable mega prostheses have gained enormous popularity in growing patients (7,8).

Other various methods of limb reconstruction have been proposed for diaphyseal tumors. These include fibular autografts (vascularized/ non-vascularized), intercalary allografts, allografts combined with a vascularized fibular graft, irradiated autografts combined with vascularized fibular grafts, composite grafts of devitalized autografts and vascularized fibular grafts, fibular transport, and distraction osteogenesis (9-13). However, it is difficult to determine the best reconstructive option for particular patients.

The most common autografts used for reconstruction are the vascularized and non-vascularized fibular grafts. To the best of our knowledge, no previous studies reported the use of the tibia as an intercalary strut autograft for reconstruction of large bone defects in skeletally immature patients.

In this study, we report our experience and the long-term clinical and radiological outcomes of using intercalary tibial autograft for treatment of six skeletally immature patients as a novel technique for reconstruction of large bone defects after resection of primary malignant bone tumors in the long bones of the lower extremity.

Materials and Methods

We retrospectively reviewed the medical records of six skeletally immature patients diagnosed with primary malignant bone sarcomas of the lower limbs and treated by wide resection and reconstruction using tibial intercalary autografts. All surgical procedures were done by the same orthopaedic surgeon at our hospital between the years 2000 and 2013.

After obtaining an approval from our institutional review board, we collected the clinical and radiological data of these patients. The following information is reported: demographic and diagnostic data (age, sex, tumor location, histologic diagnosis, and musculoskeletal tumor society (MSTS) score), therapeutic data (harvesting and reconstruction details, adjuvant/ neoadjuvant chemotherapy, and radiotherapy), length of the autograft, method of autograft fixation, re-operations, the need for a plastic surgeon's assistance in soft tissue coverage, the status of autograft, and the regeneration of the donor site. We also report the postoperative complications (nonunion, fracture, implant failure, or infection) and the time required for the bone union at the proximal and distal ends of the autografts.

All patients underwent complete staging workup in addition to the radiographic investigations which include plain radiographs, whole-body isotope LONG-TERM OUTCOMES OF LIMB RECONSTRUCTION USING TIBIAL STRUT AUTOGRAFT

bone scintigraphy, contrast-enhanced chest and abdomen computerized axial tomography (CAT), contrast-enhanced magnetic resonance imaging (MRI) of the whole tumor-involved bone, and magnetic resonance angiography (MRA) to assess the vascular supply of the affected region.

The diagnosis was confirmed by histopathological examination for all patients preoperatively. Four cases had Ewing sarcoma (ES) and two had osteosarcoma (OS). All the tumors were evaluated according to the Enneking malignant tumor staging system (14). Two patients were stage III and four patients were stage IIB.

We assessed the clinical outcomes by MSTS functional scoring system and the radiological outcomes by plain orthogonal radiographs (14).

All patients and their families had signed a written consent to use of their data and images for publication in this study.

Operative technique

We determined the level and the length of tumor resection, and the length of the autograft from the preoperative images. After surgical resection of the tumor according to the plan, surgical instruments, gowns, gloves, and draping were renewed to prevent tumor contamination of the donor site.

The tibial strut autograft was harvested from the ipsilateral tibia in femoral lesions and from the contralateral tibia in tibial lesions. In contralateral harvestings, a tourniquet was applied on the thigh (220 mmHg) for a maximum of 120 minutes.

A longitudinal incision was made over the anteromedial aspect of the tibia down to the periosteum [Figure 1]. The periosteal sheath was incised longitudinally and raised gently with a sharp periosteal elevator from all around the tibia protected by small blunt bone elevators inserted under the periosteal sheath around the proximal and distal ends of the proposed osteotomy. Multiple 2.5 mm diameter drill holes were made at the proposed level of osteotomy of the tibia proximally and distally. Bone ends were cut by a small sharp osteotome and the tibial graft was removed in one piece involving all the cortices without leaving a strut of normal bone.

The medullary canal to the remaining bones was opened proximally and distally using a curette (size 3mm) to allow the free communication of the bone marrow with the bone defect. The remaining periosteal sheath around the bone defect was tightly closed using Vicryl 2/0 continuous suture to form a periosteal tube. The skin was closed using Vicryl 2/0 subcuticular sutures.

After closure, 40-80 ml of aspirated iliac crest bone marrow was injected into the periosteal tube using plastic green cannula size G 16 to prevent the collapse of the periosteal tube and to promote new bone formation. This step was repeated after one week via percutaneous approach under general anesthesia.

Stabilization of the leg of the donor site was achieved by a monoplanar external fixator in four patients, a multiaxial ring fixator in one patient, and an intramedullary nail in one patient. The aim was to keep the leg stable in

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Figure 1. Steps (from A to D) for harvesting the tibia as strut autograft.

a straight position and under tension to allow the new regeneration of the tibia within the periosteal tube.

The non-vascularized tibia strut graft was implanted in the bone defect resulting from tumor resection. This was fixed proximally and distally by different methods according to each case [Table 1].

Postoperative measures and outcomes

Prophylactic antibiotics were administered to all patients on induction of anesthesia and continued for seven days postoperatively. All patients were transferred to the intensive care unit for 48 hours postoperatively until hemodynamic stability was ensured. Hemoglobin levels and systolic arterial blood pressure were maintained above 9 mg/dl and 100 mmHg, respectively.

On postoperative day three, range of motion

exercises were started under the guidance of the rehabilitation team. Patients started to walk with the aid of a walker frame, allowing full weight-bearing on the intact lower limb in patients with femoral tumors. Patients with tibial lesions were allowed to mobilize using wheelchair in the first 2 weeks then weightbearing as tolerated on the donor limb. No weight bearing was allowed on the reconstructed limb until early signs of radiological union were observed. Patients then progressed to partial weight-bearing and later to full weight-bearing according to the signs of graft healing.

Four weeks post-operatively, after all surgical wounds were healed, patients were referred to the oncology service to continue the planned adjuvant chemotherapy and radiotherapy.

External fixators on the donor site were removed after

| Table 1. Demographic and clinical data for each patient | | | | | | | | |
|---|------------------------|----------------|--------------------------|-----------------------------------|-------------------------|------------------------|-------------------------------|---|
| | Age (years)/ Gender | Location | Diagnosis/ MSTS stage | Length of resected specimen | Length of bone graft | Fixation proximally | Fixation distally | Plastic surgery cover |
| 1 | 6 / Male | Distal femur | OS/ IIB | 19 cm | 14cm | DCP | Ex. Fix. | - |
| 2 | 12/ Male | Femur shaft | ES/ IIB | 29 cm | 25cm | DCS-IMR | 95° angled blade plate | - |
| 3 | 9/ Male | Distal tibia | ES/ III | 16 cm | 16cm | DCP | T-plate, screw, bone graft | Bilateral fasciocutaneous flaps + FTSG |
| 4 | 15/ Male | Tibial shaft | ES/ III | 20 cm | 20cm | IMN-DCP | IMN + DCP | Fasciocutaneous Flap + STSG |
| 5 | 12/ Female | Proximal tibia | ES/ IIB | 24 cm | 24cm | IMN | IMN | - |
| 6 | 6/ Male | Distal femur | OS/ IIB | 17 cm | 17cm | DCP + Ilizarov | T-plate + Ilizarov | - |

OS: Osteosarcoma, ES: Ewing's sarcoma, MSTS: Musculoskeletal Tumor Society system, IMR: intramedullary rod, IMN: intramedullary nail, DCP: Dynamic compression plate, DCS: dynamic compression screw, Ex. Fix.: External fixator, FTSG: Full-thickness skin graft, STSG; Split-thickness skin graft

adequate consolidation had been observed on plain radiographs.

Patients were evaluated in the clinic clinically and radiographically every (4-8) weeks in the first year, every three months during the second year, every six months during the next three years, and then annually. Conventional radiographs of the recipient site were obtained to evaluate evidence of graft union, bony consolidation, tumor recurrence, and complications. Union was defined as the disappearance of the autografthost junction or bridging bone across three cortices on AP and lateral views. Radiographs of the donor site were performed at 2 weeks, 6 weeks, and then every 8 weeks to evaluate bone regeneration in the tibial periosteal tube.

Results

The mean age at diagnosis was ten years (range 6-15 years). Five patients were males and one patient was female. In all patients, bone tumors were localized to the lower extremity. The two cases of OS were in the distal femur (one in the metaphyseo-diaphysis and the other in the metaphysis). Of the four cases of ES, three were in the tibia (one in the diaphysis and two in the metaphyseo-diaphysis) and one in the femoral diaphysis.

The mean length of the resected tumor specimen was 20.83 cm (range 16-29 cm). The length of the tibia autografts matched the length of the resections; the mean length of the harvested autografts was 19.33 cm (range 14-25 cm). Table 1 summarizes the details of the clinical and demographic data of each patient. The outcomes and complications encountered postoperatively are shown in Table 2.

Two patients developed a hematoma at the recipient site despite the use of a wide-bore drain. Cultures from the hematomas revealed negative bacterial growth in LONG-TERM OUTCOMES OF LIMB RECONSTRUCTION USING TIBIAL STRUT AUTOGRAFT

both patients.

Four patients (two femoral OS and two tibial ES) were alive with the preservation of the limb at the final followup. Two patients died of tumor-related reasons without failure of the autograft. The first patient had femoral ES who died after 14 months due to local recurrence and lung and bone metastases. The other patient had tibial ES who died after five years due to multiple lung and bone metastases.

On the last follow-up, the alive patients (4 cases) had a mean of 24.25/30 (80.8%) score on the MSTS87 functional assessment. They reported as mean scores the following: Pain (4.5/5), function (3.5/5), emotional acceptance (3.75/5), need for support (4/5), walking ability (4.75/5), and gait (3.75/5).

We observed no limb-length discrepancy (LLD) in one patient with preservation of both physes during oncologic resection of the tumor. In the remaining three patients, LLD developed as a result of the loss of one or both physes. One patient with femoral OS had lengthening of the tibia one year after the initial surgery (9 cm) and lengthening of the femur 5 years later (7 cm). However, at maturity, he has had a 1.5 cm LLD [Figure 2]. One patient with distal tibial ES developed a 3 cm LLD and he is currently using a 1.5 cm shoe lift. The other patient with femoral OS is waiting for a lengthening procedure of his femur.

Osseous consolidation was considered complete when the graft united to the recipient site with signs of new bone formation. Eleven out of twelve (91.6 %) junctions between tibial autografts and the recipient sites united. One patient had a residual non-union of the proximal junction; high-dose adjuvant radiotherapy might have contributed to that [Figure 3].

All patients received neoadjuvant and adjuvant chemotherapy. None of the patients received

| Table 2. Follow-up duration, complications, and clinical outcomes | | | | | | | | |
|---|-----------|---|-------------------|-----------------|--|---------------|-------------------------------|--|
| | Follow up | Complications | Proximal union | Distal union | Further surgical procedures | Prognosis | MSTS functional score % | |
| 1 | 20 Years | Hematoma, pin tract infections, varus distal femur, valgus tibia donor site | 3 months | 12 months | 3 procedures for limb length- ening in both femur ant tibia (total 25cm), femoral nailing | No recurrence | 86.6 | |
| 2 | 14 months | Valgus tibia donor site, proximal local recurrence, lung and bone metastases | 6 months | 10 months | - | Dead | - | |
| 3 | 3 Years | Hematoma, foot drop, anterolateral bowing leg (donor site), partial skin necrosis, LLD 3 cm | Not united | 10 months | Bone graft for nonunion, tibial retrograde rod, strut fibula autograft, ankle arthrodesis | No recurrence | 66.6 | |
| 4 | 15 Years | Varus leg (donor site). | 4 months | 5 months | - | No recurrence | 100 | |
| 5 | 5 Years | Varus ankle, LLD 3 cm, metastasis. | 4 months | 4 months | - | Dead | - | |
| 6 | 6 Years | LLD 5 cm | 3 months | 12 months | Planned for lengthening | No recurrence | 70 | |

(LLD: limb-length discrepancy, MSTS: Musculoskeletal Tumor Society)

neoadjuvant radiotherapy. Three patients with Ewing's sarcoma received adjuvant radiotherapy as they have poor chemotherapeutic response.

The exact time to bony union was identified using plain orthogonal radiographs. All the proximal junctions united at a mean duration of four months (range 3-6 months), except one proximal end. The mean duration of the distal junction union was 8.8 months (4-12 months). Bony union appeared to be earlier and stronger at the proximal compared to the distal junction, probably due to richer proximal blood supply and better muscle coverage.

Three patients underwent joint arthrodesis due to tumor encroachment on the growth plate and impossibility to preserve the articular surface of the bone; one distal femoral OS, one proximal tibial ES, and one distal tibial ES. One patient needed an ankle fusion due to difficulty in preserving the distal tibia epiphysis, however, he developed a foot drop due to tumor invasion of the peroneal nerve branches and the anterolateral muscles of the leg.

All donor sites consolidated [Figure 2-5]. The earliest sign of new bone formation was at (2-4) weeks and well-formed bone regeneration was observed at four months in all cases. The external fixation on the donor site was removed after complete regeneration. The mean duration of external fixation on the donor site was six months (range 5-8 months).

One varus and two valgus tibias and one anterolateral bowing of the tibia were documented as complications at the donor site [Figure 2; 3; and 5]. LONG-TERM OUTCOMES OF LIMB RECONSTRUCTION USING TIBIAL STRUT AUTOGRAFT



Figure 2. Immediate postoperative images and the outcome after 20 years for patient number 1

A. AP view radiograph of the distal femur and the donor site in the second postoperative day showing the tibial autograft in the distal half of the femur fixed proximally by a plate and screws and distally by two Schanz screws attached to external fixator that is also fixing the leg at the same time (donor site).

B. Long film standing radiographs after 20 years of follow-up showing well-remodeled right femur and tibia, fused right knee, 15-degree varus angulation of the right distal femur, mild valgus deformity of the right leg, and pelvic tilt to the right side due to 2 cm limb-length discrepancy.

C. A clinical photograph of the lower limbs from the front showing the preserved right lower limb with evidence of scars of the previous surgeries.



Figure 3. Preoperative and postoperative images for patient number 3

A. AP view radiograph of both legs on the third postoperative day showing the right leg with the tibial graft fixed proximally and distally by plates and screws. The left leg radiograph shows the defect in the tibia after graft harvesting and the monoplanar external fixator to stabilize the leg.

B. AP view radiograph 14 weeks postoperatively showing regeneration of the left tibia at the site of graft harvesting.

C. Radiographs three years later. The AP view of the right leg shows nonunion of the proximal graft junction and the fused ankle with an intramedullary rod. The left tibia appeared well-regenerated with mild lateral bowing.

Figure 4. Radiograph and clinical photo of patient number 4 after 15 years of follow-up.

Standing AP view radiograph of both legs (A) showing the well regenerated, remodeled right tibia with mild varus deformity. The left tibia appeared fully remodeled and the graft is well-incorporated. Photograph (B) in standing position showing symmetrical leg alignments and scars of previous surgeries.

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the right ankle with evidence of scars of previous surgeries.

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Discussion

С

Among neoplasms affecting humans, primary malignant bone tumors are rare. They represent an incidence rate of roughly ten cases per one million inhabitants per year. During childhood (<15 years), the percentage of malignant bone tumors accounts for 6% of all childhood malignancies (15).

The main aim of primary malignant bone tumor surgery resection is to obtain local control while maximizing the functional outcomes without compromising the long-term survival of the patient. This can be achieved by limb salvage surgery or amputation with wide surgical margins. Numerous studies have shown no difference between long-term survival in patients treated with limb salvage surgery or amputation if negative surgical margins were achieved (16).

The availability of advanced imaging techniques, improvement of surgical margins with neoadjuvant chemotherapy, and advances in the surgical techniques have made limb salvage surgery an accepted method of local control.

Many tumors of the metaphyseal-diaphyseal region of long bones can be resected with joint preservation resulting in intercalary bone defects. There are many options for the reconstruction of these defects as biological and non-biological techniques. Each of these



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techniques has its advantages and disadvantages; every patient must be carefully evaluated and the reconstructive option should be selected individually [Table 3] (17).

Endoprosthetic reconstruction gives a good functional result with early stability, mobilization, emotional acceptance, and rapid restoration of function due to the advantage of early weight-bearing (18). However, treating skeletally immature pediatric patients with such reconstructive methods is enormously challenging especially in patients younger than ten years of age. First, the size of the remaining bone can be too small for endoprosthetic reconstruction (19). Second, even when the remaining bone is large enough for endoprosthetic reconstruction there is a greater possibility of future implant failure in younger patients (19).

All of our cases were not amenable for endoprosthetic reconstruction either due to the age of the patient (three under the age of ten years), the location of the tumor (two diaphyseal, and one large tibial metaphyseodiaphyseal tumor), or inadequate soft tissue coverage (in two cases). Problems such as infection, mechanical LONG-TERM OUTCOMES OF LIMB RECONSTRUCTION USING TIBIAL STRUT AUTOGRAFT

insufficiency, and aseptic loosening limit the long-term survival of the endoprosthesis, especially in young patients with continuing skeletal development (20,21).

Extendable or expandable mega prostheses have gained enormous popularity as they have the ability of longitudinal extension with a tube within a tube design which lengthens like a telescope offering a great alternative to amputation in growing pediatrics. However, the affordability and non-availability in the developing countries are real concerns.

The optimal reconstructive method after resection of diaphyseal primary malignant bone tumor remains controversial (18). Despite unpredictable healing, allograft has traditionally been one of the treatment choices for limb salvage, particularly for diaphyseal lesions. When successful, allograft reconstruction can ultimately provide good outcomes (22,23).

Performing intercalary segment reconstruction after primary malignant bone tumor resections results in both mechanical and biological challenges. Fixation must be solid enough to avoid early mechanical failure (24). The solid fixation using the new modality of limited

| Table 3. Advantages and limitations of different reconstruction methods for diaphyseal defects | | | | | | | |
|--|--|---|--|--|--|--|--|
| Method of reconstruction | Advantages | Limitations | | | | | |
| Endoprosthetic replacement | Early full weight bearing Well controlled size and length by custom made prosthesis No external fixation Good for long femur defects | Higher failure rate if bone other than femur Higher failure rate if short residual normal bone remains for fixation May need flap coverage in tibial lesions High reoperation rate Loosening, wear and breakage are concerns in young patients High cost | | | | | |
| Allograft | Different allograft sizes and lengths are available | The patient must remain non- or partially weight bearing for few months Higher risk of infection, non-union or delayed union, and fracture Less impressive and higher risk of complications for femur defects The incidence of allograft-host junction nonunion is considered to be higher in patients who receive adjuvant therapy | | | | | |
| Vascularized fibula autograft | Patient`s own tissue Better incorporation rates than allograft | Donor site morbidity Relatively short segment Hypertrophy of the graft sufficient to allow full weight bearing can take years in adults Requires microvascular surgery setup Prolonged surgery time | | | | | |
| Non-vascularized fibula auto- graft | Patient`s own tissue No need for microvascular surgery setup Better incorporation rates than allograft | Limited size and length Donor site morbidity | | | | | |
| Extracorporeal irradiated autologous bone graft | Provides size-matched bone | Non-availability of radiation centers Risks of infection due to long surgical hours | | | | | |
| Tibia strut autograft | Patient`s own tissue No need for bone bank or radiation facility Well-matched size and length Complete remodeling of donor site | Delayed weight bearing Prolonged surgery | | | | | |

contact locking plates, if were available at the time of our treatment of those patients, could have helped the early recovery of at least three patients. We noticed a more rapid recovery after using intramedullary nailing in two patients.

The results of intercalary segmental defects reconstruction after bone tumor resection were good from an oncologic and radiological point-of-views. One or more operative procedures are sometimes needed to obtain bone union (24).

Good results have been reported after use of osteoarticular allografts for limb reconstruction after massive resection of malignant bone tumors (25). However, several disadvantages are associated with their use. These include nonunion, late fracture, the potential transmission of disease, allograft rejection, and higher risk of infection in patients receiving adjuvant chemotherapy and radiotherapy (5,9,21,25). Furthermore, they require a bone-banking system that is not available in all countries (26). Allografts and bone banks were not available in our country at the time of these surgeries.

To reduce the number of failures, the use of allografts should be considered for reconstructions of bone defects less than 15 cm in length, especially in older patients, and applying bridging osteosynthesis with the use of LONG-TERM OUTCOMES OF LIMB RECONSTRUCTION USING TIBIAL STRUT AUTOGRAFT

plate fixation (25). The bone defects in our patients were more than 16 cm in length.

The segmental prostheses provide immediate stability and early weight-bearing capability. However, the risk of complications such as loosening, rotational instability, breakage and disassembly of the prostheses, infection, implant failure, and fracture of the adjacent bone may be higher than those for biological reconstructions with a 10-year implant survival of 63% (18,27,28).

Indeed, the intercalary prosthesis is not recommended to reconstruct defects exceeding 10 cm in length (28). The unavailability of the segmental prosthesis, the sizes of defects of more than 16 cm, and the younger age of the patients (<12 years) made this option not viable for the patients included in our series.

Vascularized fibular autografts have been previously described as a primary biological reconstruction method for treatment of metaphyseo-diaphyseal defects after resection of primary malignant bone tumors of the long bones of the lower extremity. It is a highly demanding surgical procedure that needs an expert vascular surgeon with complex microsurgical skills. Furthermore, patients need a long period of non-weight bearing due to poor mechanical stability if used for intercalary reconstruction of lower limb tumor defects in larger bones (15, 29-31).

| Table 4. A comparison between the outcomes of this study and other studies reporting different reconstruction techniques for diaphyseal defects | | | | | | | | |
|---|--------------------|--------------------|-----------------------------|---|-------------------|-----------------------|------------------|---|
| Study | Number of patients | Mean age (year) | Mean defect length cm | Method of Reconstruction | Mean follow up | Mean MSTS score | Survival rate | Others |
| The current study | 6 | 10 (6-15) | 20.83 | Tibia strut autograft | 8.4 years | 80.8%. | 66.6 % | Mean time for proximal and distal junction's union was 4 and 8.8 months respectively |
| Salunke A. et al 2019 (33) | 16 | 20 (9-45) | 14 | Non-vascularized fibula | 36 months | 28 | 75% | Mean Union time 8.5 mon 2 Nonunion 2 surgical site infections 1 local recurrence |
| Salunke A. et al 2019 (33) | 12 | 20 (9-45) | 16 | Extracorporeal irradiated autologous bone graft | 24 months | 28 | 83% | Mean Union time 8.75 months 1 Nonunion 1 Implant failure |
| Hanna S. et al 2010 (27) | 23 | 41.3 (10- 68) | NA | Endoprosthetic replacement | 97 months | 87% | 77% | Rate of re-operation was 26% |
| Aldlyami E. et al 2005 (4) | 35 | 29 (8-75) | 19 | Endoprosthetic replacement | 107 months | NA | 65% | 5 local recurrences |
| Muscolo D. et al 2004 (34) | 59 | 28 (4-66) | NA | Allograft | 5 years | NA | 79% | Infection and fracture rates were 5% and 7% respectively |

(MSTS: Musculoskeletal Tumor Society, NA: not available)

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Combining an allograft with vascularized fibula graft achieves better results, although anastomoses are demanding and the procedure is time-consuming and not devoid of complications at the donor site (32).

On the donor sites, we did not notice any major complications. Some sort of angular deformities was evident in most patients in addition to the simple pin tract infections that were treated by regular pin tract care. We believe that use of rigid external fixator is important for early rehabilitation and also may enhance bone regeneration.

The ideal reconstruction option should be similar to human biology, resistant to infection, have good strength, and be mechanically stable. We introduce this method as a valuable procedure in skeletally immature children. The technique we used avoided the use of allografts, endoprostheses, and extendable endoprosthesis that were not available in our country at the time of surgeries. Therefore, we suggest that this surgical technique is a valid option in countries with limited resources.

The outcomes of the described surgical technique are comparable in terms of reconstruction success, patient satisfaction, and complication rates with the outcomes of other different reconstruction methods that have been described for diaphyseal defects. [Table 4] (4,27,33,34).

The only other available option for our cases was amputation, which is technically less challenging than limb salvage procedures. Several complications are associated with amputation in young patients such as phantom pain, bleeding, infection, bone overgrowth, soft tissue-stump migration, and muscle imbalance (35). The concept of amputation is also not easily acceptable by the patients and their parents in our community. Hence, we opted for another method of limb reconstruction to overcome these difficult challenges.

No previous study has highlighted the effectiveness of subperiosteal tibial autograft reconstruction after resection of primary malignant bone tumors of the long bones of the lower extremity. A similar technique using the fibula has been described by Yeung et al (13). Additionally, subperiosteal resection of benign cystic bone tumors has been reported with successful complete regeneration of the bone defect without bone grafting (36). Preservation of the integrity of the periosteum after subperiosteal bone resection constitutes a valuable matrix for bone regeneration in children and young adolescents. It plays a major role in the complete filling of the bone defects and avoids the morbidity associated with other techniques (36).

The beneficial effect of the periosteum in the regeneration of the tibia after subperiosteal resection as a large long bone for intercalary strut bone graft has not been discussed before. On the assumption that the inner layer of the periosteum has an osteoblastic capability

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that allows invasion of the hematoma in the tightly sutured periosteal tube by osteoprogenitor cells, we chose our technique of tight closure of the periosteum. We achieved excellent results after two occasions of filling the newly developed periosteal tube with blood and osteogenic bone marrow cells.

Healing of the remaining defect occurred in a progressive manner that suggests osteogenesis occurred initially at the margins of the cavity and moved toward its center over the following weeks (36). We noticed the progressive calcification and ossification until the defect was transformed into a solid new reformed bony mass.

We used intramedullary nail to bridge the defect in the donor site after harvesting the tibia graft in one case. The aim was to avoid external fixation. The healing time did not differ from other cases in which external fixation was used in the donor site. Though, this finding cannot be generalized based on the result of one case.

This study has certain limitations. First, the sample size was small to determine the true incidence of the complications associated with our technique. Second, this article is a retrospective descriptive study; it does not compare different reconstructive techniques at the same institution. We believe that large comparative studies are needed to evaluate our technique. Though, the small number of malignant bone tumor cases may limit the applicability of these studies.

The use of non-vascularized tibial autografts serves as a considerable alternative for reconstruction of various types and sizes of bone defects following resection of primary malignant bone tumors in the lower extremities of the immature skeleton.

This new technique has several advantages. However, it is cost-effective in countries with limited resources. Additionally, it does not require the availability of a bone bank and can replace the use of an expensive prostheses. It also provides excellent psychological coping possibility for the patients and their families who do not accept amputation.

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