Ipsilateral Nonvascularized Autograft and Periosteal Repair for the Treatment of Pediatric Tumors of the Distal Fibula

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abstract

Resection of the distal fibula is used to treat malignant or benign locally aggressive tumors involving this region. Current reconstructive techniques are limited by undesirable functional deficits. The authors present a part-retrospective and part-prospective case-comparison of 2 patient groups (study and control) treated for malignant or benign locally aggressive primary bone tumors of the distal fibula. Patients in the study group underwent a novel surgical technique involving distal transportation of the ipsilateral proximal portion of the fibula after periosteal dissection and osteotomy with subsequent proximal periosteal reconstruction. Patients in the control group underwent other limb-salvage or amputation procedures. Patients were evaluated at follow-up appointments for oncologic, radiographic, and functional outcomes. The groups had similar demographic and perioperative data. Operative time was longer in the study group. Radiographic union occurred in 75% of the study group vs 50% of the control group. Musculoskeletal Tumor Society scores were similar between groups. According to American Foot and Ankle scores, activity limitations and support requirements were greater in the study group; according to Foot and Ankle Disability Index scores, patients in the control group reported more difficulty with squatting. All patients in the study group were without recurrence at follow-up. One patient in the control group had recurrence of disease. Complications occurred in 3 of 4 study group patients and in all control group patients. With the authors’ technique for distal fibular reconstruction involving transporting the ipsilateral remaining proximal fibula as a nonvascularized autograft, the regenerative capability of the periosteum allows for restoration of the proximal fibula, preserving osseous stock. [Orthopedics. 2016; 39(4):e687-e694.]

Distant diaphyseal fibular resection is used in the treatment of malignant or benign locally aggressive tumors involving this region. Reconstruction is critical in preventing ankle instability and/or valgus deformity. Several techniques are available for reconstruction, including arthrodesis, vascularized fibular graft, fibular allograft, and soft tissue reconstruction. However, deficits in motion with arthrodesis, peroneal nerve injury or articular incongruity with proximal fibular autografts, and nonunions and infection with fibular allografts have limited the success of these techniques. Another option is below-knee amputation, which eliminates ankle instability and deformity at the expense of limb loss.

The authors describe a novel procedure to reconstruct the distal fibula that addresses the limitations of currently...
used techniques. In this study, 4 patients treated with the authors’ technique involving ipsilateral nonvascularized fibular transport autograft with periosteal repair were compared with 6 patients treated with standard reconstructions or amputation to determine procedure effectiveness; radiographic, functional, and oncologic outcomes; and complications in both groups.

**Materials and Methods**

This retrospective case-comparison of 2 groups of patients included 4 patients in the study group and 6 in the control group who were treated at 2 centers for malignant or benign locally aggressive primary distal fibular bone tumors between 1990 and 2013. Inclusion criteria for the study group were patients older than 2 years with at least 2 years of follow-up who were treated with ipsilateral, nonvascularized, fibular autograft transport and periosteal repair for distal fibular reconstruction. The control group included all patients who were treated with other limb-salvage techniques or amputation to determine procedure effectiveness; radiographic, functional, and oncologic outcomes; and complications in both groups.

**Surgical Technique**

The patient is positioned in lateral decubitus with the affected extremity up. A direct lateral approach is used to expose the tumor located in the distal fibula (A). A wide resection of the tumor is performed using proximal and distal osteotomy sites (B). The periosteum is incised longitudinally and dissected off the native proximal fibula, preserving it intact while exposing the bone. An osteotomy is then performed in the proximal fibular diaphyseal portion with sufficient length to reconstruct the bone previously resected distally (C). The fibular autograft obtained is transported distally; 2 perpendicular holes in the proximal end of the autograft are drilled and used to anchor the subsequent periosteal repair (D). The fibular autograft is fixed to the remaining distal fibula using plates and screws, tension-band constructs, or intramedullary devices such as retrograde flexible nails (E).

In this institutional review board–approved protocol, patient, tumor, perioperative, and radiographic data were collected retrospectively from medical records. Patients were then contacted and asked to complete functional questionnaires prospectively.

**Distal fixation method** depends on the patient’s age and the size of the remaining distal fibula, if present. One such method involves fixation with plates and screws, preferably locking plates given the generally shortened length distal fibula and the nonvascularized nature of the autograft. Alternative fixation methods include tension-band constructs and intramedullary devices such as retrograde flexible nails.

After distal fixation is complete, the periosteum is repaired in its anatomic tubular shape with a continuous stitch using absorbable suture (the authors prefer chromic). The periosteum is then reattached to the proximal end of the mobilized fibular autograft with the same type of suture through the 2 perpendicular predrilled holes in the fibular autograft proximal end (Figure 1).

The final closure by layers is performed in the standard fashion. A sterile dressing is then applied, and the patient is immobilized in a bivalved, short-leg cast. Sample pre- and postoperative radiographs are shown in Figure 2.

**Aftercare**

Patients were made nonweight bearing for 8 weeks postoperatively. All pa-
tients had postoperative follow-up at 2, 6, and 12 weeks, with radiographs obtained at 6 and 12 weeks. Subsequent follow-ups were based on diagnosis but were scheduled at least every 3 months until bony union was achieved. Weight-bearing status was advanced as appropriate based on radiographs and clinical progression.

Variables and Outcome Measures

Functional outcome instruments included the Musculoskeletal Tumor Society (MSTS) score (clinician administered). Although not validated, it is the accepted functional scoring system in orthopedic oncology. Pain, functional activities, and emotional acceptance are graded with 3 additional factors adapted to either the upper or lower extremity. Each category is graded from 0 (worst) to 5 (best), with a maximum total score of 30. Scores are reported as a percentage of the maximum possible and represent the proportion of expected patient normal function.

The American Foot and Ankle (AFA) score and Foot and Ankle Disability Index (FADI) are commonly used in orthopedic foot and ankle literature. The AFA (clinician-administered and patient-rated components) comprises 9 categories: pain=40; activity limitations, support requirement=10; maximum walking distance, blocks=5; walking surfaces=5; gait abnormality=8; sagittal motion=8; hindfoot motion=6; alignment=10; and ankle-hindfoot stability=8. A higher score implies better function, with a score higher than 80 reflecting good to excellent function. The maximum possible score is 100.

The FADI is a patient-rated instrument designed to evaluate functional deficits or foot and ankle disability because of a given condition. It is commonly used to evaluate therapeutic response to interventions ranging from physical therapy to surgical procedures. It contains 26 questions pertaining to specific activities and the degree of difficulty that a patient experiences while performing them. The scale ranges from 0 to 100, with a higher score representing better function.

The FADI also has an optional sports module of 8 additional questions. This module is also scored from 0 to 100, with a maximum possible score of 12.5 per category. In the current study, patients took both the FADI and the FADI sports module, with 100 as the maximum possible score for each.

Statistical Analyses

Due to the expected small sample size and the focus on surgical technique, no a priori power analysis was performed, and reported statistics are purely descriptive. No inferential statistical analyses were performed and no P values reported.

Patient Demographics

There were 3 males and 1 female in the study group and 2 males and 4 females in the control group. The most common tumor was Ewing’s sarcoma. In the study group, the right ankle was affected in 2 patients and the left in 2 patients. All patients in the control group had left ankle involvement. Three patients in each group completed the functional evaluation and questionnaires. Patients who did not complete the questionnaires are noted in Table 1.

RESULTS

Demographics and Perioperative Data

Perioperative data between groups were comparable with regard to estimated blood loss, length of hospital stay, follow-up duration, and fibular resection length. Patients in the study group had a younger age of symptom onset (8 vs 12.6 years) (Figure 3, Table 1). Margins were positive in 2 patients; they were subsequently treated with below-knee amputation (patient 4) and radiation (patient 8). In addition, patient 1 in the study group had close margins, requiring re-excision with subsequent negative surgical margins. Margins were negative in all other patients. Operative time was longer in the study group by approximately 90 minutes (Figure 3, Table 1).

All patients with Ewing’s sarcomas had standard chemotherapy pre- and postoperatively, consisting of vincristine, doxorubicin, and cyclophosphamide alternating with ifosfamide and etoposide.

Radiographic and Functional Outcomes

Radiographic union was noted in 3 (75%) of 4 patients in the study group at an average of 9 months postoperatively. Patient 1 in the study group did not achieve complete union. At last follow-up, there was still a 1-cm space between the proximal osteotomy site and the distally transported fibular autograft. However, this patient remains asymptomatic. Notably, patient 4 achieved radiographic union prior to amputation.

In the control group, union was noted in 3 (50%) of 6 patients at 27 months postoperatively. Union was not ap-
# Table 1

## Study and Control Group Patient Data

<table>
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<tr>
<th>Parameter</th>
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<td>Wide resection of DF and reconstruction with IPFA</td>
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<td>Abbreviations: BKA, below-knee amputation; DF, distal fibula; F, female; IFA, intercalary fibular autograft; IPFA, ipsilateral proximal fibular autograft; L, left; M, male; MEA, Megan E. Anderson, MD; MCG, Mark C. Gebhardt, MD; N/A, not applicable; R, right.</td>
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*Patient did not return the Musculoskeletal Tumor Society, American Foot and Ankle, or Foot and Ankle Disability Index questionnaire. Note that patient 4 is included in the study group under intention-to-treat analysis. This patient did not return any questionnaires and thus would not have any functional responses that could be influenced by his subsequent below-knee amputation.
applicable to the remaining patients in the control group: 1 received a below-knee amputation and the remaining 2 had soft tissue reconstruction only.

Patients in the study group had similar total MSTS scores and MSTS subcategory scores compared with patients in the control group. Average total MSTS score was 23 points for both groups (Figure 4).

American Foot and Ankle scores were comparable between the groups in all but 1 category; patients in the study group had slightly more activity limitation and support requirement. Patient 7 (below-knee amputation) was not included in the AFA scoring because the score contains categories assessing alignment, sagittal and hindfoot motion, and ankle-hindfoot stability (Figure 5).

Foot and Ankle Disability Index scores were comparable between the groups. Slightly more ease with squatting was observed in the study group (Figure 6).

**Oncologic Outcomes**

One patient in the control group had a recurrence (osteofibrous dysplasia). There were no local tumor recurrences in any other patients at follow-up. Patients with malignant diagnoses are all alive without evidence of disease.
Complications were common in both groups. With the authors’ described technique, 1 patient had a close margin requiring tumor bed reexcision. Another developed a valgus deformity requiring multiple subsequent corrective surgeries. Finally, 1 patient suffered wound dehiscence requiring closure revision with subsequent amputation for recurrence.

In the control group, all patients developed complications. These ranged from symptomatic hardware requiring removal to valgus deformity. Detailed information is included in Table 2.

**Discussion**

The fibula is affected in only 2.4% of primary bone tumors. Tumors most commonly affecting the fibula include giant cell tumor of bone (GCT) and Ewing’s sarcoma, which have an incidence of 1% and 8%, respectively. Occurrence of fibular osteosarcoma has also been reported (2%-5.6%); its occurrence specifically in the distal fibula is extremely low at 0.47%. Because the fibula is a dispensable bone, wide resection is considered easier to achieve when compared with other locations. Resections in the middle portion are not technically demanding and do not require reconstruction. In contrast, management of proximal and distal fibular tumors is more difficult. Resections in the proximal fibula are challenging due to the proximity of the common peroneal nerve and the anterior tibial artery. In addition, important knee-stabilizing structures insert in the fibular head. Reconstruction of these structures is crucial, with the lateral collateral ligament being the most important. Similarly, distal fibular resections are difficult because soft tissue coverage is particularly challenging. Moreover, there are concerns regarding residual ankle instability and ankle biomechanics, although this is controversial because some authors have reported good results without osseous reconstruction.
In 2005, Nathan et al. evaluated ankle instability in pediatric and adult patients who underwent fibular harvesting for vascularized fibular autograft reconstruction for oncologic procedures. They demonstrated a higher prevalence of ankle instability in pediatric patients in whom the sum of their age plus the residual fibula was less than 16.

A literature review in 2012 noted that current studies support reconstructive surgery after distal fibular resection because improved functional outcomes are obtained in almost all patients with benign tumors and in more than half of patients with malignancies. Of the studies referenced, the largest available case series come from Dieckmann et al. and Capanna et al.; each study involved 11 patients. Dieckmann et al. reported results in patients treated with complete fibular resection for bone sarcomas or metastatic lesions, followed by tibiotalocalcaneal arthrodesis using retrograde nails or tibiotalar arthrodesis with screws. Good results were obtained in 9 patients, with 2 failures occurring in osteopenic patients. Advantages of this technique included immediate stability and avoidance of extrinsic fixation or allograft material use in the surgical area. Despite these advantages, this technique is rarely indicated in pediatric patients because subsequent limb discrepancy or residual angular deformities after growth plate injury require further surgical treatment. Capanna et al. focused on different reconstructive techniques. Seven of the 11 patients in their study recovered normal function, whereas the remaining 4 presented residual decreased mobility. In addition, 1 patient developed talar lateral subluxation. Despite these complications, all patients were pain free at follow-up.

Since these 2 case series, efforts have been made to evaluate treatment alternatives for distal fibular tumors. However, no techniques have been proven to be optimal in the pediatric population. The current authors present a new technique in which an ipsilateral fibular autograft can be obtained to reconstruct and increase the length of the residual fibula while simultaneously decreasing the risk of ankle instability and talar lateral subluxation.

Advantages of this technique include proximal fibular gap restoration after mobilization of the autograft as the repaired periosteum remains intact, facilitating proximal fibular osseous regeneration. This has the additional advantage of facilitating future fibular lengthening if the wide resection involved growth plate excision. This technique also avoids allograft use, likely reducing infection risk.

Although there is no articular surface in this autograft, if the entire distal fibula is removed, the autograft can also serve as support for a soft tissue interposition arthroplasty, allowing recreation of the talofibular joint, or for tibiofibular arthrodesis. Importantly, the period of nonweight bearing or partial weight bearing in the postoperative period is similar to what is used in other reconstruction techniques involving vascularized autografts or allografts.

As expected, the authors’ technique had longer operative times given its additional steps. However, this had no effect on final estimated blood loss. Radiographic union was achieved in most patients predictably at approximately 9 months.

The attained functional outcomes were comparable between groups. The observed differences in terms of less activity limitation and support requirement and ease in squatting with the authors’ technique need to be confirmed with a larger sample size. All patients in the study group were without local tumor recurrences at follow-up. Oncologic goals do not have to be compromised to perform the presented technique.

This study is limited by lack of sufficient power to conduct inferential statistics. Nevertheless, comparable results between the study and control groups, as well as the authors’ observations of the patients’ clinical courses, strongly suggest that this technique is a safe alternative. In addition, there were fewer complications in the study group. The advantages of this technique in harnessing the regenerative capabilities of the periosteum and preserving osseous stock and the distal fibular growth plate raise the possibility that, given sufficient statistical power, the patients’ favorable outcomes in the study group may be confirmed in future investigations. Limitations of this technique include the lack of articular surface of the autograft when reconstructing the entire distal fibula. There is no available literature comparing this technique vs arthrodesis or reconstruction with a reverse proximal fibula autograft. Therefore, the current authors cannot recommend one technique over the others, although they have perceived these techniques, including their own, to be suboptimal for articular reconstruction at the talofibular level.

CONCLUSION

Distal fibular reconstruction after wide resection is a challenging problem in pediatric patients. The authors present a reconstructive technique transporting the proximal ipsilateral fibula with periosteal repair. This technique restores native bone stock and allows subsequent correction of limb discrepancy or angular deformities that is often required in the pediatric population. Restoration of bony support in the lateral column of the ankle increases lateral stability and decreases residual valgus deformity. Although described as an intercalary reconstruction, this procedure is versatile and can be used as an interposition arthroplasty if the distal fibular articular surface requires reconstruction, or it can be used to aid a stable arthrodesis.

REFERENCES
2. Jamshidi K, Mazhar FN, Masdari Z. Recon-


