Rotationplasty: An Option for Limb Salvage in Childhood Osteosarcoma

Herbert S. Schwartz, MD
Frank J. Frassica, MD
Franklin H. Sim, MD

Rotationplasty to reconstruct skeletal defects after resection of primary bone tumors in the distal femur or proximal tibia in skeletally immature patients is becoming more frequently used as a limb-salvage alternative to transfemoral amputation or hip disarticulation. Small series of patients having this procedure are now being reported from both Europe and the United States and the results are encouraging.

Case Report

A 13-year-old girl was seen at the Mayo Clinic with a 3-week history of painful swelling in her right proximal tibia. There was no history of antecedent trauma, fever, or other joint involvement. Physical examination revealed a warm, tender, and swollen proximal anterior tibia with a full range of knee motion. The neurovascular status was intact. A plain radiograph revealed an intramedullary sclerotic lesion without a soft-tissue mass (Fig. 1). Local tumor extension was judged to be minimal, with computed tomography demonstrating a small, extraosseous anterior mass (Fig. 2). A roentgenogram of the chest and CT revealed bilateral pulmonary nodules. Needle biopsy of the tibia, performed the day after presentation, demonstrated a grade 4 osteosarcoma. In order to determine whether the patient had stage IIB or III disease preoperatively, a median sternotomy was performed and both lung nodules were biopsied. Silver stains confirmed that both lung specimens contained noncaseating granulomas, consistent with histoplasmosis. The surgical stage was, therefore, IIB.

Chemotherapy was started according to the Children’s Cancer Study Group protocol. The induction phase consisted of high-dose methotrexate and citrovorum factor, vincristine, and BCD (bleomycin, cyclophosphamide, actinomycin D). The patient tolerated 6 weeks of chemotherapy, and the clinical response was judged to be good. The general criteria that the authors consider for a good response are: a subjective decrease in pain and swelling, mineralization of the soft tissue component of the lesion, and a decrease or cessation in growth of the tumor. If the lesion is found to increase in size during the monthly radiographic examinations, the chemotherapy is stopped and surgery is performed. The patient was subsequently restaged (Fig. 3), and the surgical stage remained the same. CT revealed no extraosseous tumor adjacent to the neurovascular bundle. The extent of the proximal tibial tumor necessitated an extraarticular resection. A determination of the skeletal age of the patient and extrapolation from the Green-Anderson growth charts predicted that approximately 2.5 cm and 1.5 cm of physesal growth remained in the distal femoral and proximal tibial growth centers, respectively.

The goal of surgical treatment for this young girl was to remove the tumor with a wide surgical margin (en bloc resection with a cuff of normal tissue without entering the tumor). The surgical options included a transfemoral amputation or en bloc resection followed by reconstruction. The authors prefer to perform an extraarticular resection for all lesions about the knee in which there is perforation of the
Fig. 1: Anteroposterior (A) and lateral (B) roentgenograms revealing sclerotic lesion in metaphysis of tibia.

Fig. 1A.

Fig. 1B.

Fig. 2: Axial computed tomogram of proximal tibia, demonstrating slight anterior and medial extraosseous extent of lesion.

The surgical procedure of rotationplasty was performed, as described by Kotz and Salzer.³ A rhomboid skin incision was made with the anterior long axis being 5 cm longer than the intended bone cuts proximally and distally. The medial and lateral apexes of the incision met posteriorly. The common peroneal nerve was dissected free and kept in continuity. The popliteal vessels and posterior tibial nerve were isolated and dissected free from proximal to distal, beginning at the adductor hiatus. The anterior tibial vessels were divided at their ostia. An en bloc extraarticular resection was then performed (the biopsy tract was included), and wide margins were
obtained in all dimensions. A tibial bone cut was made, which provided an 8 cm, normal, uninvolved, distal margin. The proximal fibula was included in the resected specimen. A proximal femoral osteotomy was performed at a precalculated level to provide for a limb that was approximately 2.5 cm longer than the contralateral femur (Fig. 4). Thus, at skeletal maturity, both "knees" would be at the same level. Rotation markers were placed on the remaining tibia and femur to guide the reduction, as the tibia was externally rotated 180°. Rigid internal fixation was then applied to the osteotomy sites. The musculature of the anterior aspect of the thigh was sutured to the remaining gastrocnemius complex for anchorage. Deep drains were placed, and closure was done in layers. A bulky compressive dressing was applied. Neurocirculation in the foot was intact.

Study of the pathologic specimen showed that necrosis was present in 55% of the tumor and that the margin obtained in the tibia was 9 cm of normal marrow (Fig. 5). All other margins were free of tumor. The patient’s first pylon cast was placed three days after operation. The patient was walking with lateral supports by 6 months postoperatively (Fig. 6); although the patient was completely pain-free at this time, the osteosynthesis site had not completely

Fig. 3: Anteroposterior roentgenogram of proximal tibial osteosarcoma after chemotherapy showing increased sclerotic definition of lesion.

Fig. 4: Intraoperative photographs showing extremity after extraarticular resection, with remaining neurovascular bundle and foot before (A) and after (B) shortening and external rotation.

Fig. 4A.

Fig. 4B.
Fig. 5: Proximal tibia and fibula in coronal section from resected specimen (A). Gross photograph shows tumor extending into epiphysis and medial periosteal reaction. Radiograph of resected tibial specimen (B).

Fig. 5A.

Fig. 5B.

We have noted that patients on high-dose chemotherapy heal osteotomy sites very slowly. In general, if the patient is doing well and the fixation is solid, any surgical intervention is delayed (bone grafting, etc) until after chemotherapy is completed. At 9 months, she had excellent foot function and was independently ambulating with her prosthesis and without any aids (Fig. 7). The patient and her family had no psychosocial problems with regard to adaptation to the salvaged extremity.

The patient completed her last cycle of chemotherapy 1 year after its initiation. The induction phase was completed with high-dose methotrexate and doxorubicin. The patient was given a maintenance regimen using agents similar to those given to a responder to preoperative chemotherapy. She has been continuously disease-free since surgery.

**Discussion**

In a strict oncologic sense, the major factor in selecting between a limb-sparing procedure and an amputation is the relationship of the tumor to the surrounding soft-tissue margins, especially around the neurovascular structures, because the proximal margins are usually the same in both procedures. Rotationplasty can provide tumor-free margins equal
to or wider than other forms of limb salvage because it is an intercalary amputation. Misplaced biopsy sites, contaminated soft tissues after pathologic fractures, or extraosseous tumor extension may in some circumstances have wider resected margins after a rotationplasty than after alternative types of limb-salvage techniques. In selected patients, the margin of resection can be further widened during rotationplasty by including the popliteal vessels in the resected specimen and by anastomosing the appropriate vessels when the limb is shortened.

Rotationplasty was first described by Borggreve in 1930. In 1950, Van Nes reported his experience with this procedure for the treatment of congenital defects of the femur. He described three patients who had satisfactory results after more than 8 years of follow up. In subsequent years, several others have described the usefulness of this procedure in selected patients who had dysgenesis of the proximal femur along with large limb-length inequalities. They recognized the advantages of saving the foot—that
is, position sense, controlled swing phase of gait, and better general mobility—over the results of above-knee amputation and the use of a prosthesis. The "cosmetic curiosity" resulting from a retained and rotated foot has been previously addressed as a psychosocial disadvantage of this procedure.15

The reported results of rotationplasty for childhood high-grade sarcomas are encouraging. Several series are reported, with a combined total of 39 patients.1-4 Of 12 patients with a minimal follow up of 2 years reported by Kotz,4 none had a local recurrence. In that series, the youngest child receiving a rotationplasty was 6 years of age.

In addition to the question of local control of the tumor, the functional results after the operation are also of concern regarding rotationplasty. Two studies have compared the results of patients with rotationplasty to those of transfemoral amputees.1,17 The children with rotationplasty had pronounced loss of plantar flexor strength on the operated side, compared to knee extension on the nonoperated side. However, these children could use their prostheses efficiently, especially when running and stair climbing. Electromyographic study revealed that the rotated calf muscles performed similarly to knee extensors, adapting well to their new function. Their efficiency, however, was not as good as that in below-knee amputees. The major functional advantages of the rotationplasty, compared with the above-knee amputation, are the ability of the patient to stair climb with both limbs and a smoother gait when running.17 During walking, net energy output in children with rotationplasty was less than that of above-knee amputees.1

The additional advantages of a successful rotationplasty over transfemoral amputations are the increased recreational activities possible, such as skiing and tennis, because of the better control of the prosthesis.4 Also, there are no phantom-pain symptoms, as are commonly seen in amputees.

In summary, rotationplasty offers some advantages over transfemoral amputation, provided that equal tumor control has been achieved. These include smoother gait (especially when running), increased level of activities, and a decreased chance of phantom pain.

Another issue of concern about rotationplasty is the psychosocial adaptation of the patient to the rotated foot and ankle. Of the 39 patients who have undergone this procedure, none has expressed evidence of emotional rejection, such as requests for foot or toe amputations. In the authors' experience, the patients have tolerated the rotated extremity well.

The indications for the use of a rotationplasty begin with the skeletally immature patient who has a primary bone tumor in the distal femur or proximal tibia and in whom limb-sparing surgery will not compromise the oncologic soundness of the operative procedure. The more growth potential that the patient has at the time of operation, the more useful this procedure becomes (the youngest reported patient undergoing rotationplasty being 6-years-old). In these instances, rotationplasty may be the surgeon's only choice to preserve a functional limb. The long-term results of rotationplasty must be directly compared with the proven standards of the transfemoral amputation before the true role of rotationplasty can be determined. However, the procedure remains a viable surgical option and a valuable addition to the options of the orthopedic oncologist.

References

13. Van Nes CP: Rotation-plasty for congenital defects of the

Editorial Discussion

ORTHOPEDICS: In view of the fact that initially the knees remain at a different level and there is a leg length discrepancy, should the very young child be treated by an initial equalization and correction of inequality on completion of growth?

Schwartz et al: Following a rotationplasty, leg length inequality refers to the position of the rotated ankle to the contralateral normal knee. If it is necessary from a cosmetic standpoint to have the rotated ankle at exactly the same level as the opposite knee, then careful follow up with serial scanograms to detect any leg length discrepancy is necessary. If a discrepancy can be predicted, then the appropriate physeal arrest can be performed. From a functional standpoint, a longer operated limb may perform better. Currently, our goal is to have the rotated ankle at the level of the opposite knee; however, we accept a longer operated limb because the function may be superior.

ORTHOPEDICS: Should patients on a chemotherapy protocol be considered more seriously for amputation rather than rotationplasty because of the bone healing problems with chemotherapy?

Schwartz et al: Overall, children on chemotherapy tend to heal osteotomy sites slower than children who are not on toxic chemotherapy regimens. If the fixation remains secure, we follow delayed unions closely and bone graft the osteotomy site after the completion of the chemotherapy, if necessary. To date, this has been necessary in one patient.

The greatest concern in performing reconstructive surgery in patients on high dose chemotherapy is the risk of infection. Prerequisites for the surgery include adequate white blood cell counts, proper nutrition, and the absence of other infections (skin, urinary tract, etc), even if mild. Accordingly, we feel rotationplasty is safe and effective in patients on toxic chemotherapy regimens.