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What Is the Outcome of Allograft and Intramedullary Free Fibula (Capanna Technique) in Pediatric and Adolescent Patients With Bone Tumors?

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Abstract

Background After bone tumor resection, reconstruction for limb salvage surgery can be challenging because of the resultant large segmental bony defects. Structural allografts have been used to fill these voids; however, this technique is associated with high complication rates. To circumvent the complications associated with this procedure, massive bony allografts have been supplemented with an intramedullary vascularized free fibula. However, few studies have examined the outcomes using this technique in the pediatric and adolescent populations.

Questions/purposes The purpose of this study was to examine the revision-free survival using he Capanna technique for limb salvage for pediatric lower limb salvage. We attempted to answer the following questions: (1) What was the overall limb salvage rate along with incidence of reoperation and complications? (2) How did pediatric and adolescent patients functionally perform after this technique? (3) What was the incidence of late complications including infection and fracture? (4) What was the incidence of limb length discrepancy?

Methods Eighteen pediatric patients who underwent lower extremity limb salvage with the use of cadaveric allograft and intramedullary free fibular transfer (Capanna technique) were identified. There were nine boys males and nine girls with a mean age of 11 years (range, 5–18 years) and mean followup of 8 years (range, 2–15 years), respectively. All patients had at least 2 years followup. Three patients have not been seen in followup during the past 5-years; however, all had made it to their 5-year clinical followup. The patients' medical records were reviewed for clinical and functional outcomes as well as postoperative complications. Time to union was recorded through an evaluation of radiographs. Mankin functional outcome and Musculoskeletal Tumor Society (MSTS) rating scale were recorded for each patient.

Results The overall limb salvage rate was 94%. Fourteen patients underwent an additional surgical procedure. Six patients underwent additional procedure(s) to treat a symptomatic nonunion. Seventeen of the patients had a good or excellent Mankin score with a mean MSTS rating of 93% at last followup. Six of the patients underwent a limb length modification procedure.

Conclusions Use of large allografts in conjunction with intramedullary vascularized free fibulas appears to be a reliable method for the reconstruction of large bony tumors of the lower extremity in this population, although we did not directly compare this with allografts alone in our study. The use of locked plates may improve union times. The proportion of patients achieving limb preservation was high and complication rates are acceptable.

Level of Evidence Level IV, therapeutic study.

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Introduction

Through advances in surgical technique, adjuvant chemotherapy, and radiotherapy, limb salvage surgery has become the treatment of choice for a majority of patients with long bone sarcomas [41]. To provide the patient with an adequate oncological margin during the tumor resection, large osseous defects can be created that hamper functional limb salvage. Traditionally, structural allografts have been used to fill these defects, providing structural support using cortical bone; however, they are associated with a high complication rate as a result of the avascular nature of the allograft [5, 11, 13, 24, 25, 27, 31].

Free vascularized fibular grafts have been used since the 1970s for reconstruction after resection of bony tumors [10, 39]. They provide an osteogenic environment and have shown the ability to survive infection, chemotherapy, and radiotherapy [10, 39]. Although they possess the ability to unite and remodel with the host bone and increase in strength, free fibulas initially lack the structural support of large cortical allografts [1]. To circumvent the complications associated with this procedure, Capanna and Campanacci [12] supplemented cortical allografts with intramedullary free fibular grafts. The combination of the osteogenic potential of the vascularized free fibula and the structural support of the cortical allograft make the use of these grafts particularly attractive in the reconstruction of bony defects in young patients. Previous reports have noted early success with this technique [1, 11, 21, 37]; however, few reports have examined survival and complications associated with reconstruction in the pediatric and adolescent patient population, including the requirements for late limb lengthening and donor site morbidity [31]. The purpose of this study was to examine the outcomes using this technique for lower extremity limb salvage surgery in the pediatric and adolescent patient populations with a focus on long-term limb function and growth.

This study attempts to answer the following questions: (1) What was the overall limb salvage rate along with incidence of reoperation and complications? (2) How did pediatric and adolescent patients functionally perform after this technique? (3) What was the incidence of late complications including infection and fracture? (4) What was the incidence of limb length discrepancy?

Patients and Methods

After approval from our institutional review board, we performed a retrospective study based on chart review of all patients undergoing intercalary bony reconstruction using an intramedullary vascularized free fibula and cortical allograft (Capanna technique) over a 15-year period (1997–2012). In addition, all patients had at least 2 years of clinical and radiographic followup.

Patients were identified through a review of our institutions medical and surgical index as those undergoing intercalary bony reconstruction using an intramedullary vascularized free fibula and cortical allograft.

After tumor resection, a cortical allograft was selected and cut to match the defect. The fibula was then harvested in the standard fashion through a lateral approach. The amount of fibula harvested depends on the size of the defect, and an attempt was made to keep the fibular graft at least 4 to 6 cm longer than the allograft while maintaining sufficient distal fibula to prevent ankle instability. The medullary canal of the allograft was reamed to accept the fibula. The free fibula was then passed into the medullary canal of the allograft. A burr was used to create a trough to pass the fibular vessels through (Fig. 1) and a Kirschner wire was used to transfix the fibula into the allograft to prevent rotation during placement. The composite allograft/fibula construct was slotted into the ends of the host bone, acting as an intramedullary rod. Our preferred internal fixation construct was designed to achieve circumferential compression at the host-allograft junction. Plates were placed on each side of the bone, 180° opposite each other. The construct might consist of two long plates or a long plate on one side and then two shorter plates on the opposite side, each small plate spanning across one host-allograft junction. Using plates with combined compression and locking capability, first maximal compression across the host-allograft junction was achieved; then selective locking screw placement was used to maximize fixation. The microvascular anastomoses were then performed after bony fixation (Fig. 1). In patients requiring additional skin coverage, a separate slot is created in the allograft to allow the skin paddle of the fibula graft to exit from the construct. In younger patients (< 12 years of age), tethering of the distal tibiofibular syndesmosis of the donor site is performed using a single screw to prevent valgus angulation of the ankle.

After this, the leg of the donor site is initially placed in a bulky posterior slab splint and changed to a fiberglass cast for 6 weeks. For tibia reconstructions, the recipient site is immobilized in a cast or removable splint for 3 months, after which the extremity is placed in a brace until the osteotomy site is healed. Femoral reconstructions were immobilized in a long-leg cast or splint. Patients were allowed to begin partial weightbearing at a mean of 3 months (range, 2–22 months) postoperatively; however, full weightbearing was delayed until a mean 10 months (range, 3–22.5 months).

Patients' medical records were evaluated for pertinent demographic information, postoperative complications, and the pathology of the tumor specimen. Union was

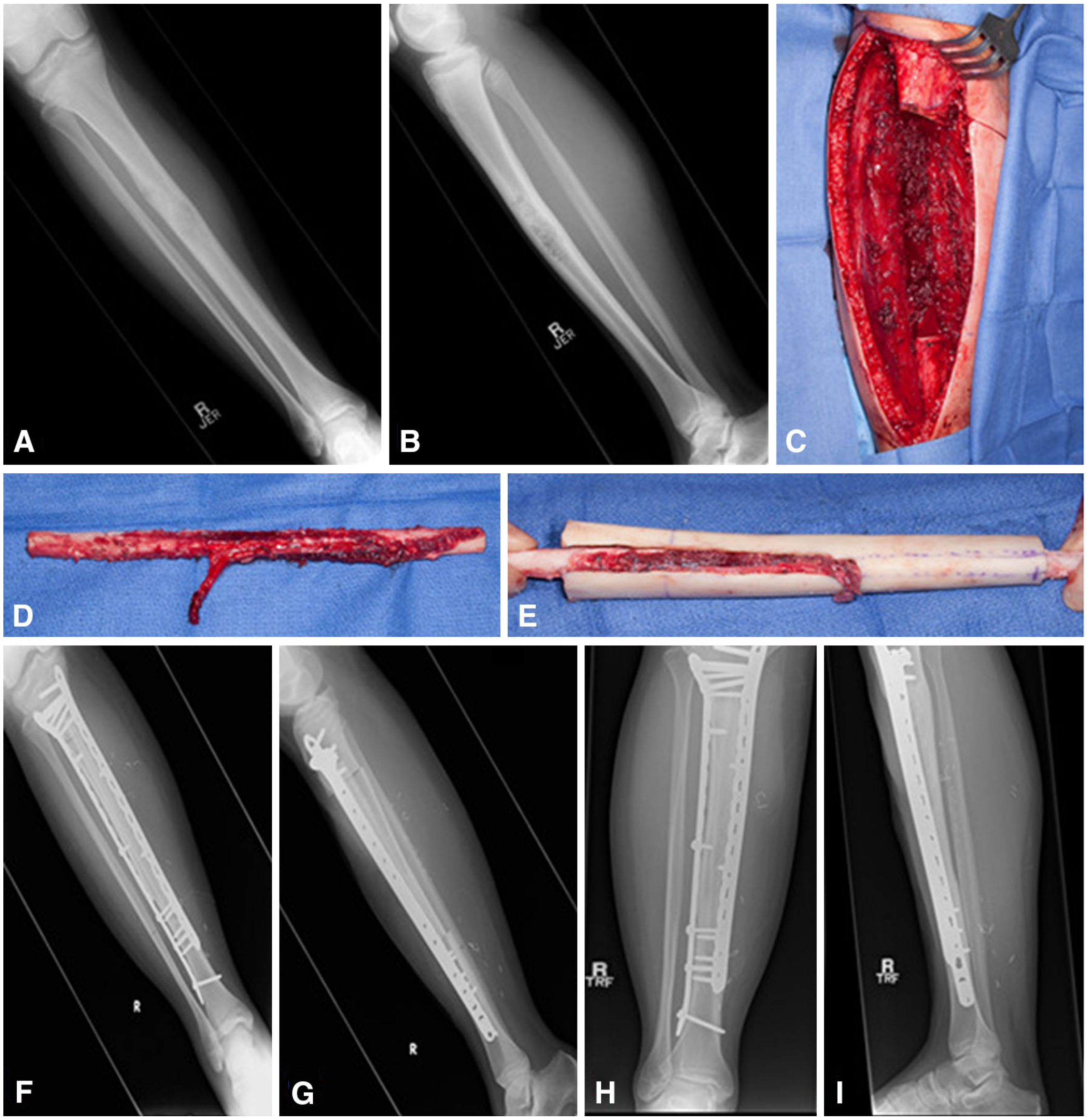

Fig. 1A–I Preoperative radiographs show a teenage patient with a tibial adamantinoma (**A–B**). The decision was made to treat the patient using the Capanna technique with an intraoperative photograph of the postresection defect (**C**). The bony defect was filled by combining a free vascularized fibula (**D**) and a massive bone allograft (**E**). Comparison of the immediate postoperative (**F–G**) and 2-year followup radiographs show union of the fibula/allograft composite (**H–I**).

judged through evaluation of the patients' radiographic studies by the treating orthopaedic and plastic surgeon and defined as bridging callus on three of the four cortices. If there was question of union or the junction site was difficult to evaluate as a result of surgical hardware, a CT scan of the extremity was performed. Clinical and functional outcomes were measured using the Mankin extremity score as well as the Musculoskeletal Tumor Society (MSTS) score from information gathered from the patient's medical record [19, 28]. Late complications were defined as fracture, infection, or graft failure occurring greater than 24 months after the initial surgery [35]. Failure of limb salvage was defined as any patients undergoing amputation or removal of the allograft and/or vascularized fibula for complication. Revision surgery was defined as a reoperation where the allograft and/or vascularized fibula were revised but retained. Contralateral limbs were evaluated by the treating surgeon for any evidence of ankle instability, flexor hallucis contracture, chronic pain, or paresthesia. Reconstructed limbs were followed for any need of secondary procedure, including limb lengthening. ROM was documented by the treating surgeon at the preoperative and postoperative visits. Limb length inequalities were measured using standing scanogram radiographs and growth modulation was recommended if the inequality was greater than 2 cm.

Survival was estimated using the Kaplan-Meier survival method. All statistic calculations were made using JMP Version 9 (Statistical Analysis Software, Cary, NC, USA) with statistical significance set at a p value < 0.05.

Over the 15-year period, there were nine boys and nine girls who underwent bony reconstruction using the Capanna technique. The mean age at the time of surgery was 11 years (range, 5–18 years) (Table 1). The mean followup of these patients was 8 years (range, 2–15 years).

The most common oncologic diagnosis was osteosarcoma ($n = 9$) followed by Ewing's sarcoma ($n = 6$). Nine cases involved reconstruction of the tibia, and nine cases were for femoral reconstruction (Table 1). In one of these cases, the procedure was performed after an allograft failure in which the tumor was removed 30 months prior. In the remaining cases, the Capanna procedure was performed at the time of tumor resection. Six patients underwent a local pedicled muscle flap in addition to a skin graft for wound coverage. One patient underwent skin paddle incorporation into the allograft construct. All local flaps and skin paddle use were for tibial reconstruction. Six of the tibial reconstructions called for soft tissue coverage compared with none of the femoral reconstructions.

All resections spared the articular surface with preservation of the epiphysis. In three cases, the physeal plate was removed with the resected specimen. Only one patient was skeletally mature at the time of the surgical procedure. The mean defect, allograft, and free fibula length was 15 cm (range, 10–30 cm), 15 cm (range, 10–30 cm), and 19 cm (range, 13–23 cm), respectively. In one patient the structural cortical allograft was longer than the free fibula graft. At least one surgical plate (up to three) was used for surgical stabilization in all patients. Ten patients had a locked reconstruction.

Ipsilateral fibula was used in six of patients, and all were for femoral reconstruction. To prevent a valgus ankle deformity, tethering of the donor syndesmosis site was performed in 12 patients. There were five donor site complications. Complications at the donor site included wound dehiscence ($n = 1$), a prominent syndesmosis screw undergoing removal ($n = 3$), and one case of intraoperative posterior tibial artery injury requiring vein graft repair. There were no reported cases of flexor hallucis contracture, ankle instability, or transient or permanent deep peroneal nerve palsy.

Accounting for All Patients/Study Subjects

After the surgical procedure, patients were followed for recurrence for every 3 to 4 months for the first 2 years, then every 6 months for Years 2 to 5, and then annually for Years 5 to 10 with radiographs of the involved extremity and, depending on the tumor pathology, a CT scan of the

Table 1. Clinical and functional outcomes after intramedullary vascularized fibulas with massive bone allograft

Sex	Age (years)	Location of reconstruction	Pathology	Knee ROM at followup	Mankin score	MSTS rating
Female	14	Proximal tibial metaphysis	Osteosarcoma	135	Excellent	100
Female	10	Femoral shaft	Osteosarcoma	130	Good	67
Female	16	Proximal tibial metaphysis	Osteosarcoma	135	Excellent	87
Female	6	Proximal tibial metaphysis	Ewing's sarcoma	150	Good	90
Female	14	Proximal tibial metaphysis	Osteofibrous dysplasia	150	Excellent	100
Female	10	Distal femoral metaphysis	Osteosarcoma	120	Excellent	93
Male	5	Proximal tibial metaphysis	Ewing's sarcoma	110	Good	83
Male	7	Distal femoral metaphysis	Ewing's sarcoma	142	Excellent	100
Male	10	Distal femoral metaphysis	Osteosarcoma	90	Excellent	100
Male	15	Proximal tibial metaphysis	Osteosarcoma	120	Excellent	100
Male	18	Femoral diaphysis	Chondrosarcoma	110	Excellent	93
Female	7	Distal femoral metaphysis	Osteosarcoma	130	Excellent	100
Female	8	Femoral diaphysis	Ewing's sarcoma	130	Excellent	100
Male	12	Proximal tibia metaphysis	Ewing's sarcoma	140	Excellent	100
Male	9	Tibial diaphysis	Ewing's sarcoma	140	Excellent	97
Female	12	Femoral diaphysis	Osteosarcoma	100	Good	87
Male	15	Tibial diaphysis	Adamantinoma	140	Excellent	100
Male	11	Distal femoral metaphysis	Osteosarcoma	140	Failure	77

MSTS = Musculoskeletal Tumor Society.

chest. This surveillance occurred at our institution or if the patient was not able to return for followup, the images were sent to our institution for review.

In our patient cohort, three patients have not been seen for greater than 5 years since their last followup visit. One of these patients has previously had greater than 10 years of clinical followup. The other two patients have since been lost to followup but have had at least 5 years of clinical followup. One of these patients is an international patient, so returning for followup would be difficult.

At our institution the Capanna technique is reserved for patients in whom the projected limb length discrepancy will be 4 cm or less and we are able to preserve the articular surface with our resection. If there is projected to be less than a 4-cm limb length difference, surgical procedures such as epiphysiodesis or lengthening of the allograft can be performed to offset this difference. A relative contraindication to limb-sparing surgery is a very immature skeletal age (< 8 years); this is because these patients will likely need multiple surgical procedures as a result of the projected limb length discrepancy of > 4 cm. In these patients we would recommend an expandable prosthesis or modified amputation such as a rotationplasty. If the tumor involves the joint or epiphysis, then we typically perform an osteoarticular allograft or an expandable prosthesis. We recommended limb length correction surgery if the projected limb length discrepancy would be 2 cm or greater. Likewise, a patient has to have a fibula, which can be transferred.

We are unable to comment on the exact number of patients who met the criteria for a Capanna and were treated with a different procedure. However, if a patient met our criteria of projected limb length inequality of less than 4 cm, would be able to preserve the physis/articular surface, and there was a viable fibula for transfer, we chose to perform this technique for limb salvage at our institution. We feel the addition of a biological graft is extremely important in these patients as a result of their higher activity demands and longer lifespan.

Results

The overall limb salvage rate was 94%, although many of the grafts required reoperation after the procedure. Fourteen patients underwent an additional surgical procedure, seven of which underwent multiple procedures, at a mean of 15 months (range, 1–49 months) after the initial procedure (Fig. 2A).

Seventeen of the patients achieved a good to excellent Mankin functional outcome score and the mean MSTS rating was 93% (range, 67%–100%). There was one failure according to the Mankin score [6]. Two patients died from metastatic disease at 55 and 73 months postoperatively. There was no difference in the patients' mean pre- and postoperative knee ROM (125° ± 30° versus 128° ± 30°, p = 0.7).

There was a total of six nonunions during the course of the study. There were two nonunions at the proximal host-allograft junction, two at the distal junction, and two occurred at both junctions. All six patients underwent a repeat operation to treat a symptomatic nonunion causing the patient pain at the allograft and host junction. Five of these nonunions occurred when the patient was undergoing chemotherapy at the time of the original reconstruction. Four of the nonunions developed in the tibia and two were in the femur. Wound healing complications occurred in two patients, two patients underwent removal of painful hardware, two patients underwent removal of screws in the distal epiphysis to allow for growth, one patient had a free latissimus flap to cover exposed hardware, and one patient underwent below-knee amputation for tumor recurrence. Over the course of the study, fractures of the allograft occurred in seven patients at a mean 40 months (range, 5–130 months) after the procedure (Fig. 2B). Four of the patients were treated nonoperatively with cast immobilization for 12 weeks, whereas the other three were treated with open reduction and internal fixation (ORIF). After treatment, all the fractures successfully healed. Late allograft fracture occurred in three patients (Fig. 3). Two of these patients had ORIF, whereas the other patient was successfully treated with a period of protected weightbearing. Two fractures occurred while the patients were participating in a sport, and one occurred through a screw hole in the allograft. All patients returned to their preinjury status after treatment. There were no cases of late infection.

At last followup, a mean 1-cm (range, 0–8 cm) limb length inequality was observed. Four patients underwent limb length adjustment surgery including femoral lengthening (n = 1), tibial lengthening (n = 1), femoral shortening (n = 1), and tibial epiphysiodesis (n = 1) at a mean age of 11 years (range, 9–13 years) and 48 months (range, 24–80 months) after the limb salvage procedure. The mean limb lengthening correction was 3.5 cm (range, 2.6–5.5 cm). Seven patients had a limb length discrepancy treated nonoperatively. Six patients underwent a procedure to correct a genu valgus (n = 5) or varus (n = 1) deformity of the resected limb. All five cases of valgus deformity occurred in the femur.

Discussion

With increasing survival rates, limb salvage surgery has become one of the goals of treatment for bony tumors of the extremities [3, 23, 41]. Current options for limb salvage surgery include large cortical allografts, endoprostheses,

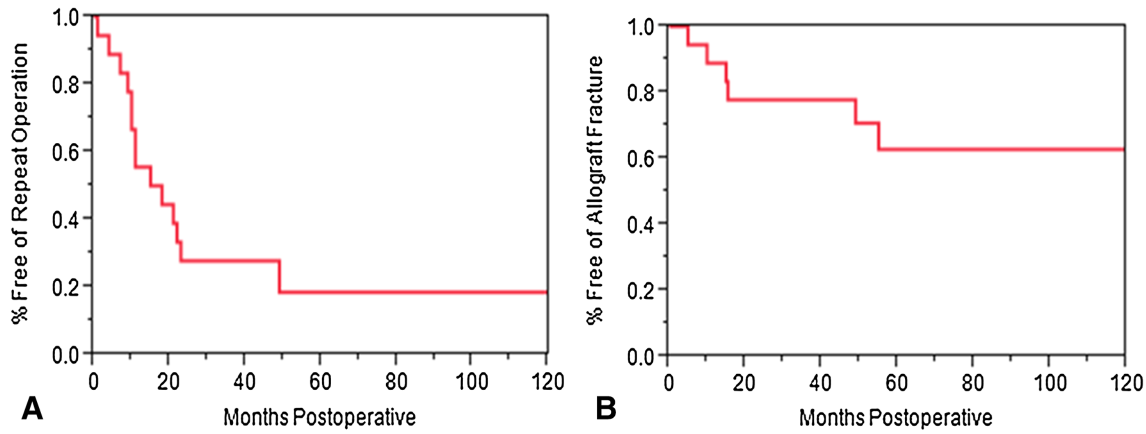

Fig. 2A–B Kaplan-Meir survival with reoperation for any cause (**A**) and allograft fracture (**B**) as an outcome in patients undergoing the Capanna technique for limb salvage is shown.

distraction osteogenesis, and vascularized fibular graft [3, 9, 20, 24, 34, 35]. Massive structural allografts have been most frequently used for limb reconstruction; however, complications such as allograft infection and fracture are common as a result of the avascular nature of the graft [24]. The Capanna technique was developed to add biology to the avascular graft to help reduce these complications [12]; however, reports of the use of the Capanna technique in the pediatric and adolescent populations are limited [31]. In our series we were able to use the Capanna technique to provide a successful means of limb salvage in the pediatric and adolescent populations with an acceptable complication profile.

There are several limitations to this study. There is significant selection bias and we are unable to comment if there were other patients who met our criteria for the Capanna technique but were treated with a different method of limb salvage. Although all patients had made it to the 2-year clinical followup point, this study included patients over a 15-year period. Two patients have been lost to followup and it is possible they could have sustained a complication and been treated elsewhere. The retrospective nature of the study limits the amount of data we were able to collect from the medical record and contains obvious constraints on the analysis presented. Although there were no instances of allograft infection, or removal of the grafts for complications, with longer followup, late limb or graft failure might occur. Although this study was performed at one institution, the procedure was performed by multiple surgical teams, including members who were fellowship-trained microvascular surgeon(s). Likewise, union was judged by the treating orthopaedic and plastic surgeons' review of the radiographs and clinical symptoms; as such, the data are limited by assessor bias. In addition, we had a relatively small series of patients with no comparison group so we cannot definitively state that these results are comparable or superior to others or to allograft reconstruction alone.

In our series we noted a 94% rate of overall limb salvage using the Capanna technique in pediatric and adolescent patients with all but one of the patients maintaining their original reconstruction. The rate of retention of the original reconstruction observed in our study is greater than the previously reported rates for the use of allografts alone in the pediatric population. Muscolo and colleagues [32] noted a 95% rate of limb salvage; however, only 68% of patients had their original allograft reconstruction. Likewise, Campanacci and colleagues [9] reported 76% of pediatric patients had retained their original limb salvage reconstruction at last followup. In oncological processes near the epiphysis and articular surface, a physeal-sparing procedure allows for the maintenance of the articular surface and epiphysis depending on the location of the tumor and that is typically not achievable with endoprostheses or osteoarticular allografts [15]. In selected patients, intercalary allograft reconstruction can be performed in combination with partial epiphyseal preservation; however, this is associated with a high complication rate [32, 34]. The preservation of the articular cartilage is thought to provide joint stability, longevity of the joint, and, if the growth plate can be preserved and not hampered by fixation devices, limb growth may be unimpeded [34]. Because the Capanna technique allows for the preservation of these structures along with augmenting bony union, it provides a satisfactory biological reconstruction technique for pediatric patients [21].

Functionally, all the surviving patients in our cohort achieved a high functional level after their procedure as suggested by the good to excellent Mankin scores of all the surviving patients and the high average MSTS rating. We did have one failure secondary to tumor recurrence resulting in amputation. All surviving patients were able to

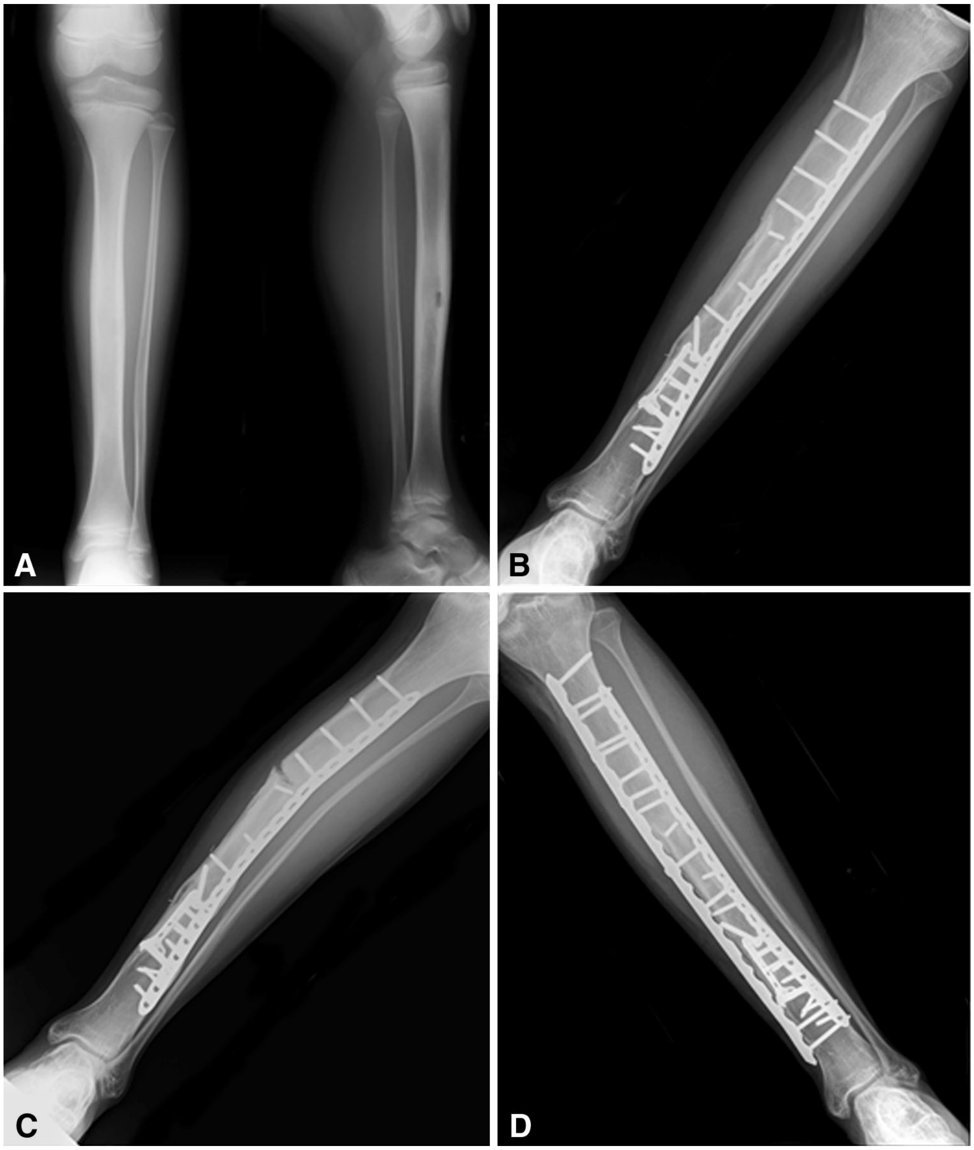

Fig. 3A–D Preoperative radiographs (**A**) show a patient with a tibial Ewing's sarcoma. The patient underwent the Capanna technique for limb salvage surgery and at 20 months postoperatively union was noted (**B**). The patient sustained a tibial fracture (**C**) 42 months after surgery while participating in sports. He was treated with ORIF and subsequently went on to heal (**D**) and return to sports.

return to their previous activities including basketball, running, volleyball, baseball, golf, and karate. As a result of this, we feel children can be counseled that a return to some athletic activity is possible after limb salvage. This appears to be a higher percentage of patients achieving this level of function than has been previously reported after the Capanna technique or with the use of massive allografts alone where good to excellent Mankin scores were only seen in 49% to 74% of patients [2, 6, 17, 21].

Although initially structurally superior to fibular grafts, the use of large cortical intercalary allografts for lower extremity limb salvage has been reported to have a high rate of complications as a result of the avascular nature of the graft including nonunion, infection, and fracture [5, 17, 29, 33, 34, 36, 43]. Previous studies have shown that the outer surface of the allografts becomes populated with living cells; however, the inner allograft remains acellular [4, 6, 22]. Because the allograft is acellular and lacks a blood supply, when complications such as fracture and infection occur, the body is unable to heal the injury like native vascularized bone [5, 7, 8, 13, 17, 26, 27, 33, 34, 36]. In our patient cohort, nonunions occurred in six patients. A majority of our patients were receiving chemotherapy during the time their nonunion developed, and they all occurred in the tibia, which previous studies have shown to increase the risk of nonunion [11, 18].

Similar to nonunion, seven patients sustained an allograft fracture. Allograft fracture is a recognized complication that occurs after the use of massive bone allografts in up to 20% of patients [5, 8, 30, 35, 36, 40]. Because the free fibular grafts provide an intrinsic blood supply, bony union can occur through the process similar to normal fracture healing [14, 16, 24, 38, 42]. Although fractures were common in our series, the intramedullary free fibula allowed the fractures to heal with ORIF or nonoperatively, which could not occur in an allograft alone. Infection has been shown to occur in 5% to 18% of allografts [2, 8, 33, 36, 43]. Although a majority of our patients in our series were undergoing chemotherapy, which has been shown to increase the risk of infection [11], we did not have any postoperative infectious complications. This is a lower rate of postoperative infection than has previously been shown for the Capanna technique, which has ranged from 4% to 33% [1, 11, 21, 37]. Late donor site complications were minimal and no patient developed ankle instability. Late allograft fracture occurred in three patients, and similar to the early fracture patients, all fracture healed without revision of the allograft.

Previous literature examining the use of the Capanna technique in the pediatric and adolescent populations did not examine limb length discrepancies [38]. The average limb length discrepancy in our study is similar to previously reported limb length inequalities in children undergoing allograft procedures [39]. Musculo and colleagues [39] showed a 2-cm limb length inequality when using an intercalary allograft in children younger than 10 years of age; however, the authors acknowledge their followup was limited. Likewise Campanacci and colleagues [9] noted an average 3-cm limb length discrepancy with five patients undergoing a growth modulation procedure. Parents and patients should understand that limb discrepancies may occur in over half of patients once they reach skeletal maturity. Fortunately, most limb length differences were less than 2 cm. If the discrepancy is projected to be significant, corrective procedures can be performed, including lengthening and growth modulation. These are important factors to pass along to parents as they contemplate this reconstruction option.

Use of large allografts supplemented with intramedullary vascularized free fibulas provides a reasonable option for reconstruction of large bony defects in the lower extremity after limb salvage surgery, especially in younger patients in whom the risks of allograft fracture and infection have been reported to be high. Although there was a high rate of reoperation, the results of this procedure in terms of function and tumor control may outweigh the risk of complications. The Capanna technique should be considered in pediatric and adolescent cases of segmental defects of the lower extremity. Because there are no direct comparison studies to allografts alone, more than half of which avoid the added complexity of the vascularized fibular grafting procedure, further comparison studies are necessary to elucidate which surgical techniques optimize long-term outcomes and minimize complications. In the meantime, surgeons, patients, and their families will have to be treated in an individualized manner.

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