



Pediatric extremity bone sarcoma reconstruction with the vascularized fibula flap: Observational study assessing long-term functional outcomes, complications, and survival

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KEYWORDS

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Pediatric sarcoma;
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Abstract *Background:* Limb salvage is important in pediatric patients with bone sarcomas. The vascularized fibula flap is a versatile option, combined or not with a bone allograft. The authors evaluated the functional long-term outcomes, complications, and survival of using this technique in pediatric patients.

Methods: A retrospective review of 27 pediatric patients reconstructed between 2011 and 2018 with the fibula flap after bone sarcoma resection was conducted. Long-term functional outcomes, complications, and survival were assessed. Variables analyzed were age, sex, Capanna technique, follow-up, complications, additional surgeries, time to weight bearing, length discrepancy, and sport practice.

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Results: Twenty-seven patients with a mean age of 9.3 years were included. The mean follow-up was 44.33 months. The Capanna technique was performed in 15 patients. All extremities but one were salvaged. The overall complication rate was 74.07%. Fibula fracture and nonunion rates were 34.04% and 11.11%, respectively. Partial weight bearing was resumed at a mean of 9.07 months. About 79.17% of patients with a 12-month follow-up achieved full weight bearing. An age below 8 years was significantly associated with a lower major complication rate and a shorter time to weight bearing and full weight bearing. Major complications and additional surgeries were significantly associated with longer periods until weight bearing and full weight bearing.

Conclusions: The fibula flap allows the majority of extremities to be reconstructed. However, a high rate of complications and additional surgeries should be anticipated. Full weight bearing is usually achieved within the first year, with modest functional increase afterward. Less complications and a faster functional recovery are expected in patients below the age of 8 years.

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Introduction

Pediatric bone sarcomas represent 5-6% of all malignancies in children. They arise predominantly in the nonaxial skeleton, with nearly 50% of them located in the femur.¹ The development of new chemotherapy agents, along with the advances in the multidisciplinary treatment of this pathology, has dramatically decreased the mortality in the last three decades, with survival rates ranging between 60% and 92%.¹⁻⁴

Retrospective studies have been unable to find differences in survival between amputation and limb-sparing surgery. In fact, amputation is only considered as the first therapeutic option in 10% of cases.^{5,6} Currently, efforts are focused on preserving stable functional extremities with adequate length, weight-bearing (WB) capacity, and growth potential.⁷

Several bone reconstructive modalities have been described. The most popular is the vascularized fibula flap, with more than 40 years of experience.^{3,8-10} It has proved to be a reliable technique with solid outcomes. It also allows for the reconstruction of epiphyseal segments using the fibular head.^{7,11-14} The Capanna technique combines the stability and mechanical strength of a bone allograft with the capacity of integration and remodeling of a vascularized fibula flap.^{15,16} Notwithstanding, the previous techniques are technically demanding, showing an overall complication rate as high as 64%.³

The present study reports the long-term outcomes of a series of 27 pediatric patients affected by bone sarcomas of the extremities, reconstructed with the fibula flap after oncologic resection.

Methods

After approval from the hospital ethics board, a retrospective review of 27 consecutive pediatric patients reconstructed between 2011 and 2018 with the vascularized fibula flap after oncologic resection of a primary bone sarcoma of the extremities was conducted. All surgeries were performed by a multidisciplinary team consisting of plastic and oncologic orthopedic surgeons. This research adhered

to the STROBE guidelines for observational studies. All patients' parents signed informed consent for the procedure, including consent for taking photographs/videos for scientific purposes.

Surgeries were preoperatively planned by means of virtual reconstruction and 3D printed real-size models. Either angio-computed tomography (CT) or magnetic resonance (MRI) images were 3D reconstructed with AYRA software (Andalusian Public Health Service, Spain). These geometries were then processed by a BQ Witbox 1 (BQ Co., Spain) fused deposition printer. The printed models were fabricated in polylactic acid (PLA).

Extremities were preserved if resection with free margins could be achieved to prevent interference with survival. This was verified with intraoperative biopsies of the resection margins. All patients underwent reconstruction immediately following resection. Defects were reconstructed with a free fibula flap in 25 cases (95.59%), a pedicled fibula flap in one case (3.7%), and a reverse-flow pedicled fibula flap in the remaining case (3.7%).

Chest X-ray, thoracic CT scan, and bone gammagraphy were performed every 6 months during the first 2 years after surgery by a pediatric oncologist to rule out recurrences/lung metastasis and annually from the third to the fifth year.

Patients were followed up every 2-3 months by the orthopedist. Function of the extremity was classified as full weight bearing (FWB) when the infant was able to bear normal load and perform activities of daily living (ADL) autonomously without any orthosis; partial weight bearing (PWB) if orthosis was needed to bear load; or nonweight bearing (NWB) as long as the extremity remained in discharge. Functional status at 12 and 24 months was taken as the reference cut-off points for evaluating function.

The oncologic and functional outcomes of this series were evaluated. Data were retrospectively collected from the hospital medical records. Variables analyzed with the SPSS 17.0 software (SPSS, Inc., Chicago, Ill.) were age, sex, Capanna technique, follow-up time, complications, additional surgeries, time until resumption of WB and its degree, length discrepancy, and sport practice. Chi-square test was used for qualitative variables, whereas the *T* student, Mann

Whitney *U* test, and ANOVA tests were used for qualitative-quantitative analysis. The Kaplan-Meier method was used to estimate survival rates.

Results

Demographic characteristics (Table 1)

Twenty-seven patients with a mean age of 9.3 years (range, 2-17 years) were included, of which fourteen were males and thirteen were females. Sarcomas were located in the upper extremity in four cases (14.81%) and in the lower extremity in 23 (85.19%). Involved bones included 16 femurs (59.26%), five tibias (18.52%), three humerus (11.11%), and one radius, iliac, and calcaneus (3.7% each).

The most frequent histopathologic diagnosis was Ewing sarcoma ($n = 16$, 59.26%), followed by conventional osteosarcoma ($n = 11$, 40.74%). Every patient received neoadjuvant and adjuvant chemotherapy (QT) under protocol. Radiotherapy was not administered in any case.

Follow-up and oncologic outcomes (Table 1)

The mean follow-up was 44.33 months (range, 5-95 months). Five patients (18.52%) developed lung metastasis. They underwent surgical resection and QT. Unfortunately, the disease progressed in two of them and they both eventually died. One of the patients also developed local recurrence and had his lower limb amputated. The remaining 22 patients were free of disease at last follow-up.

Follow-up time was significantly associated with the appearance of metastasis ($p 0.02$). Overall survival was 92.59%. The Kaplan-Meier analysis revealed a 91% survival rate at 30 months of follow-up, and a sustained 80% disease-free survival rate after 20 months (Figures 1 and 2).

Procedural details (Table 1)

The mean length reconstructed was 13.37 cm (range, 6-22 cm). The most selected recipient vessels were the descending branch of the lateral circumflex femoral vessels ($n = 8$), followed by the superficial femoral vessels ($n = 6$). There were 23 osseous and four osteocutaneous flaps. The configuration of the fibula included 25 single and two double-barreled constructs. The Capanna technique was performed in 15 cases (55.55%), all of them involving the lower extremity. The fibula head was included in three cases - for reconstruction of a femoral head, a humeral head, and a distal radius.

Complications (Table 2)

The overall complication rate was 74.07%: 20 out of 27 patients experienced complications. Age ≥ 8 years appeared as a significant ($p 0.03$) risk factor for major complications, with more than eightfold increased risk in this group (OR 8.4 (1.26-56.07)) compared to <8 years (73.7% versus 25%).

Early minor complications were observed in five patients (18.52%), including pressure ulcers ($n = 2$), wound dehiscence ($n = 1$), peroneal nerve palsy ($n = 2$), and partial braquial palsy ($n = 1$); they all healed conservatively.

Late major complications were found in 16 cases (59.26%). Ten patients (34.04%) experienced fractures, arising as the most common complication. Two of them were treated conservatively, resuming WB after observing callus formation in radiographic controls. Eight patients required open reduction and internal fixation (ORIF) (Figure 3(A) and (B)). There were also three cases of nonunion (11.11%), which eventually needed bone grafting. Other major complications included one case of tibia valga, *agenu varo*, a hardware infection, and a radiocarpal volar subluxation in a distal radius reconstruction. Neither partial nor total flap losses were encountered.

Major complications were significantly associated ($p 0.02$) with a longer period until WB resumption, with an estimated difference of 7 months (2-11 months). Similarly, they were associated ($p 0.03$) with a longer period until FWB (difference 6.36 months [0.58-12.15 months]), along with the need for additional surgeries ($p 0.03$, difference 6.43 months [0.66-12.2 months]).

Assessment of the 15 lower extremity Capanna reconstructions revealed a 40% ($n = 6$) and a 13.33% ($n = 2$) fracture and nonunion rates, respectively. With the exception of one fracture, the remaining required ORIF for correction. The Capanna technique was not significantly associated with any of the overall ($p 0.66$), major ($p 0.45$) and minor ($p 1$) complication rates or additional surgeries ($p 0.45$). Similarly, no significant differences were found regarding functional outcomes ($p 1$).

Additional surgeries (Table 2)

Fourteen patients (51.85%) required additional surgeries, including the correction of three nonunions by means of bone grafting and internal fixation, the ORIF of eight fractures, a wrist arthrodesis, one case of hardware removal, and a posteromedial tibial epiphysiodesis for the correction of a *genu varo*.

Functional outcomes (Table 2)

All extremities but one were salvaged. PWB was resumed when callus was detected in radiological controls, at a mean of 9.07 months (range, 1 to 33 months). A minimum follow-up of at least 12 months to assess final WB capacity and aptness to perform ADL was available in 24 patients. Two patients showed follow-up periods inferior to 12 months, whereas another died six months after surgery. Consequently, not enough data were available to evaluate their functional outcomes.

Nineteen patients (79.17%) achieved FWB at a mean of 12.7 months (range, 4-33 months) and were able to perform ADL autonomously, increasing to 85.71% if a 24-month follow-up was considered. Eight patients (32%) had resumed sports practice. Follow-up time was significantly associated with this resumption ($p 0.02$). Three patients (12.5%) achieved PWB. Two patients (8.33%) remained NWB because

Table 1 Patient demographic data and procedural characteristics.

Patient	Age (years)	Sex	Histology	Status at last follow-up	Follow-up (months)	Bone involved	Defect length (cm)	Flap vascularity	Recipient vessels	Flap laterality	Flap configuration	Fixation
1	17	M	Ewing sarcoma	FOD	95	Tibia (distal)	11	Free	Posterior tibial	Contralateral	Double barrel	Internal
2	7	F	Ewing sarcoma	FOD	90	Humerus (proximal)	9	Free	Humeral	Contralateral	Single barrel	Internal
3	7	M	Ewing sarcoma	LM, Deceased	6	Femur (mid)	14	Free	DB. LFC	Contralateral	Capanna	Internal
4	13	F	Ewing sarcoma	FOD	83	Humerus (proximal)	14	Free	Humeral	Contralateral	Single barrel	Internal
5	11	F	Osteosarcoma	FOD	79	Femur (distal)	18	Free	DB. LCF	Contralateral	Capanna	Internal
6	6	F	Ewing sarcoma	FOD	76	Tibia (proximal)	7	Free	Posterior tibial	Contralateral	Capanna	Internal
7	3	M	Ewing sarcoma	FOD	72	Iliac	9	Free	Superior gluteal	Homolateral	Double barrel	Internal
8	6	F	Ewing sarcoma	FOD	68	Calcaneus	6	Pedicled	-	Homolateral	Osteocutaneous single barrel	Internal
9	9	F	Ewing sarcoma	FOD	57	Femur (mid)	11.5	Free	DB. LCF	Contralateral	Osteocutaneous single barrel	External
10	9	F	Osteosarcoma	FOD	56	Femur (mid)	16	Free	Superficial femoral	Contralateral	Capanna	Internal
11	12	F	Osteosarcoma	FOD	52	Tibia (proximal)	16.5	Free	Anterior tibial	Contralateral	Single barrel	Internal
12	9	M	Ewing sarcoma	LM	50	Tibia (mid)	6	Pedicled	-	Homolateral	Single barrel	Internal
13	9	F	Ewing sarcoma	FOD	49	Radius (distal)	9	Free	1. Peroneal AV: prox. radial. 2. DGA: distal radial. 3. Peroneal V: cephalic	Contralateral	Osteocutaneous single barrel with fibular head	Internal
14	9	M	Ewing sarcoma	FOD	43	Femur (proximal)	13.2	Free	Anterior tibial AV to DB. LCF (2 veins)	Homolateral	Capanna with fibular head	Internal and external

(continued on next page)

Table 1 (continued)

Patient	Age (years)	Sex	Histology	Status at last follow-up	Follow-up (months)	Bone involved	Defect length (cm)	Flap vascularity	Recipient vessels	Flap laterality	Flap configuration	Fixation
15	2	M	Ewing sarcoma	FOD	39	Femur (mid)	12	Free	DB. LCF	Contralateral	Single barrel	Internal
16	9	F	Osteosarcoma	LM, Deceased	30	Femur (mid and distal)	19	Free	Superficial femoral	Contralateral	Capanna	Internal
17	10	F	Osteosarcoma	FOD	36	Femur (mid and distal)	16	Free	Superficial femoral	Contralateral	Capanna	Internal
18	13	M	Osteosarcoma	FOD	33	Femur (mid)	17	Free	Superficial femoral	Contralateral	Capanna	Internal
19	11	M	Osteosarcoma	LM	32	Femur (distal)	14.5	Free	Superficial femoral	Contralateral	Capanna	Internal
20	12	M	Ewing sarcoma	FOD	28	Femur (mid and distal)	22	Free	DB. LCF	Contralateral	Capanna	Internal
21	13	F	Osteosarcoma	FOD	27	Femur (mid)	13	Free	DB. LCF	Contralateral	Capanna	Internal
22	9	M	Ewing sarcoma	LM	25	Femur (proximal and mid)	17.5	Free	Deep femoral	Contralateral	Capanna	Internal
23	9	M	Osteosarcoma	FOD	22	Femur (mid and distal)	15.5	Free	Superficial femoral	Contralateral	Capanna	Internal
24	5	M	Ewing sarcoma	FOD	18	Tibia (distal)	12.2	Free	Posterior tibial	Contralateral	Osteocutaneous single barrel	External
25	4	M	Osteosarcoma	FOD	16	Humerus (proximal)	13	Free	Humeral	Contralateral	Single barrel with fibular head	Internal
26	9	M	Ewing sarcoma	FOD	10	Femur (distal)	12	Free	DB. LCF	Contralateral	Capanna	Internal
27	10	F	Osteosarcoma	FOD	5	Femur (mid)	17	Free	DB. LCF	Contralateral	Capanna	Internal

M: male; F: female; FOD: free of disease; LM: lung metastasis; A: artery; V: vein; Prox.: proximal; DGA; descending genicular artery.

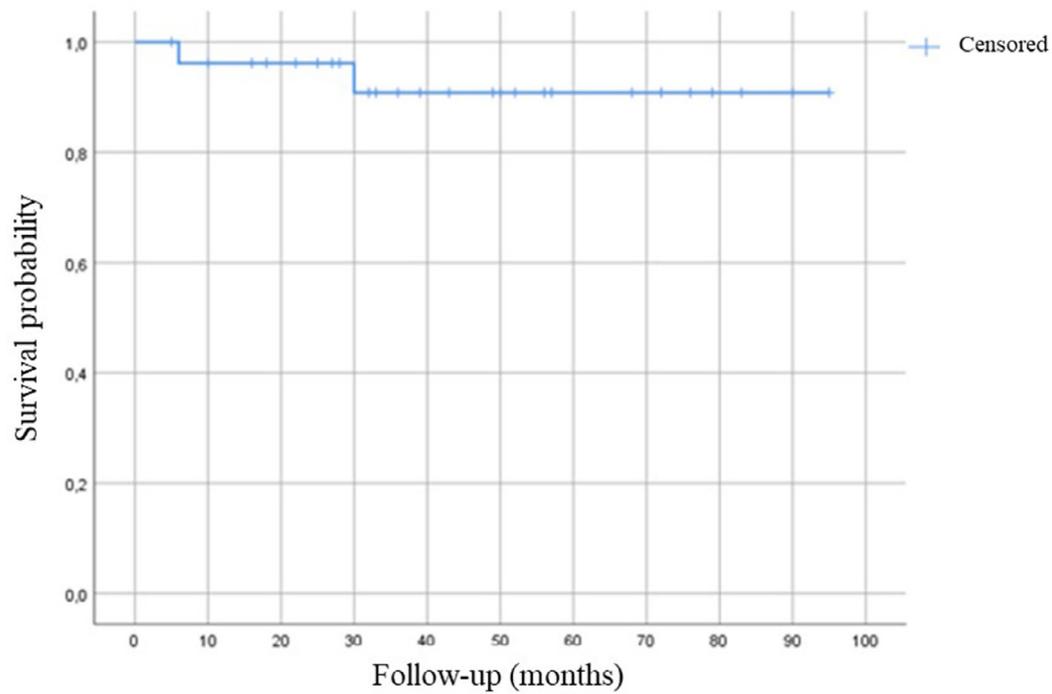


Figure 1 Survival after pediatric extremity bone sarcoma resection and reconstruction with the vascularized fibula flap.

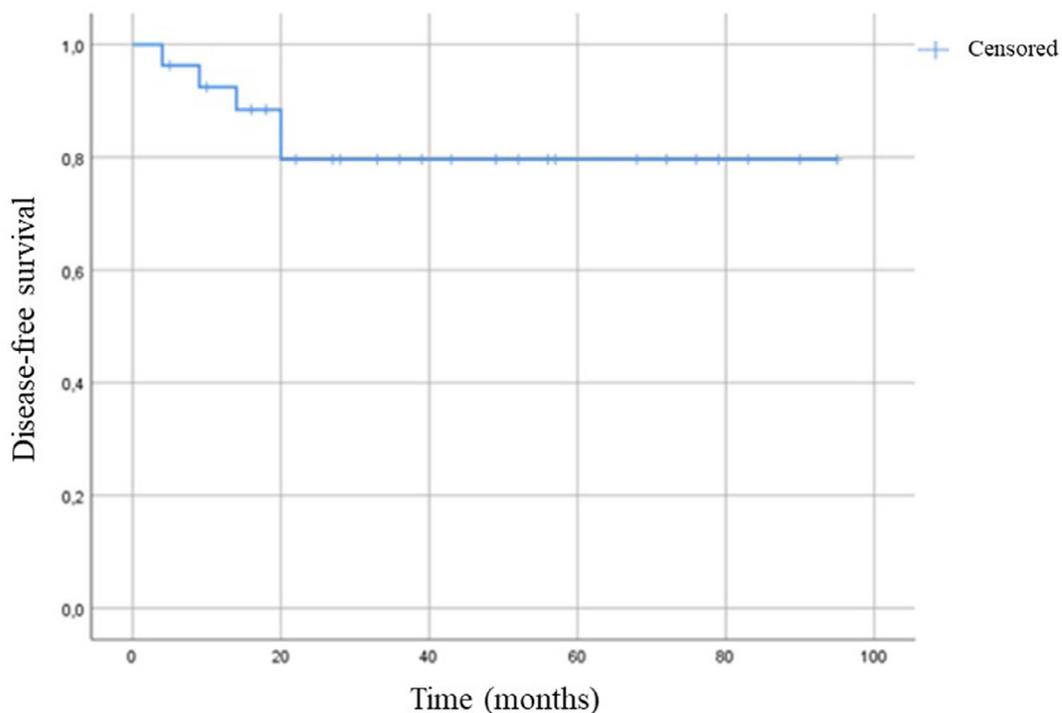


Figure 2 Disease-free survival after pediatric extremity bone sarcoma resection and reconstruction with the vascularized fibula flap.

of fractures. The later five patients needed assistance to perform ADL. Twenty out of these 24 patients underwent reconstruction of the lower extremity. Evaluating the lower extremity specifically, FWB was achieved at a mean of 13.87 months (range, 5-33 months).

Considering age and functional outcomes, patients below 8 years showed statistically significant (p 0.04) lower periods until WB resumption, with an estimated difference of 7 months (0.01-12 months); and FWB (p 0.02) with a difference of 7.47 months (1.18-13.75 months).

Table 2 Complications, additional surgeries, and functional outcomes.

Patient	Complications	Additional surgeries	Time to bone union (months)	Weight-bearing capacity	Time to FWB (months)	Sports practice	Limb discrepancy (cm)
1	Fracture	No	8	FWB	12	Yes	5.5
2	No	No	3	FWB	4	No	No
3	Incomplete braquial palsy	No	Deceased	-	-	-	-
4	No	No	3	FWB	18	No	No
5	Nonunion	Bone-graft	33	FWB	33	No	5.5
6	Genu varo	Tibial epiphysiodesis	8	FWB	9	Yes	No
7	Pressure ulcer	No	3	FWB	9	Yes	No
8	Wound dehiscence	No	6	FWB	10	Yes	2
9	Fracture	ORIF	4	FWB	17	Yes	No
10	No	No	3	FWB	10	Yes	No
11	Nonunion	Bone graft	4	FWB	18	No	1.5
12	Fracture	ORIF	4	PWB	-	No	No
13	Radio-carpal subluxation	Wrist arthrodesis	3	FWB	5	No	No
14	Pressure ulcer, fracture	No	5	PWB	-	No	5
15	No	No	5	FWB	5	Yes	2
16	Fracture	ORIF, amputation	3	FWB	19	No	No
17	Nonunion	Bone graft	4	FWB	10	No	3
18	Fracture	ORIF	4	FWB	17	Yes	2.4
19	Fracture	ORIF	3	FWB	9	No	2.5
20	Fracture	ORIF	7	PWB	-	No	1
21	No	No	9	FWB	12	No	No
22	Transient peroneal palsy	No	6	FWB	12	No	No
23	Fracture	ORIF	3	FWB	20	No	1.5
24	Fracture	ORIF	4	NWB	-	No	No
25	Transient peroneal palsy	No	4	FWB	5	No	No
26	No	No	2	FWB	5	No	1
27	Infection	Hardware removal	4	NWB	-	No	No

ORIF: open reduction and internal fixation; FWB: full weight bearing; PWB: partial weight bearing; NWB: nonweight bearing.

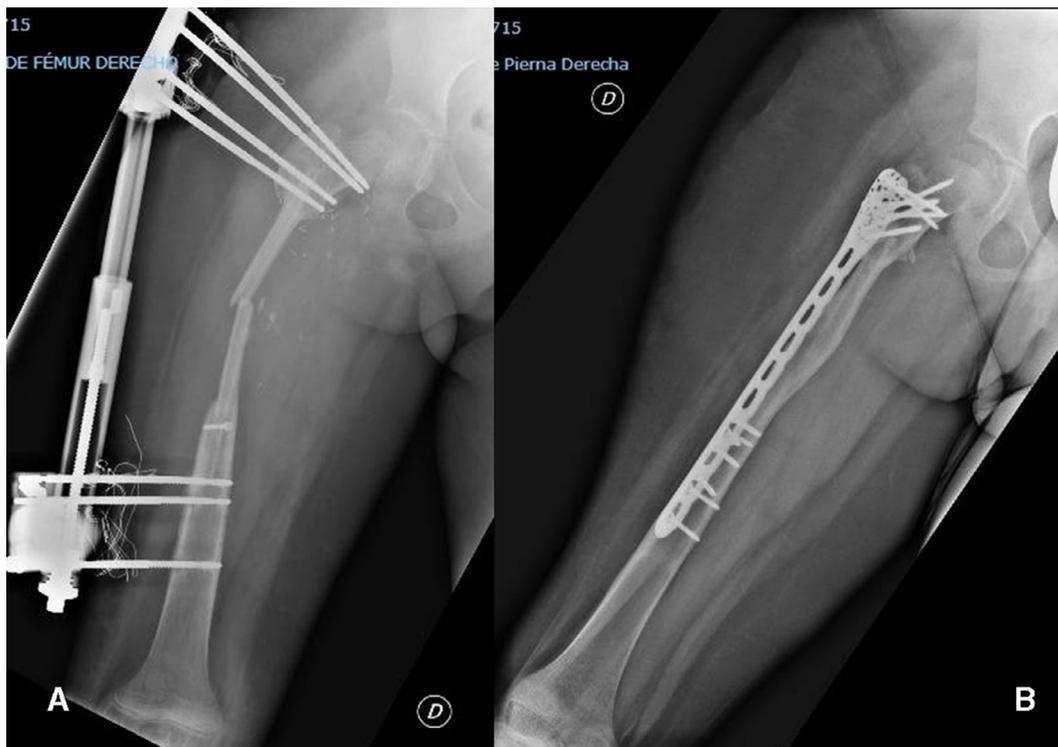


Figure 3 (A) Displaced fracture of the vascularized fibula 9 months after surgery. (B) 12 months after secondary surgery and ORIF, consolidation is appreciated.



Figure 4 X-ray showing a lytic lesion located in the middle third of the right femur.

Limb length discrepancies ≥ 2 cm were observed in seven patients, with a mean of 3.7 cm (range, 2-5.5 cm). The limbs were corrected with shoe lifts. Two additional patients were ambulated with a limp not related to length discrepancies: a Trendelenburg limp as a consequence of the resection of the gluteal musculature and a *genu varo*.

Discussion

Improvements in the multidisciplinary treatment of pediatric bone sarcomas in the last 30 years have drastically enhanced the survival rates¹⁻⁴. After resection with safety margins, the aim is to preserve functional limbs with growth potential⁷. The preferred reconstructive technique for intercalary defects of the long bones is the fibula flap: after four decades of experience, it has demonstrated its superiority in comparison to other alternatives, such as bone grafts, allografts, or endoprostheses. It offers a plentiful source of autologous bone with remodeling potential and low donor-site morbidity^{7-14,17}. However, these surgeries are not exempt from complications³.



Figure 5 18 months after surgery showing integration, consolidation, and hypertrophy of the construct.

Outcomes and complications

Amputations were avoided with the exception of one case. Twenty-five out of 27 patients were alive at last follow-up. About 79.17% of the patients with a minimum follow-up of 12 months achieved FWB and could perform ADL autonomously. Another 12.5% of patients achieved PWB with the help of orthosis. These results are similar to previously published studies,^{3,7,18} reporting FWB and PWB rates of 72-85% and 20-22%, respectively. If a longer follow-up period of 24 months is considered, the FWB rate increases to 85.71%. This indicates that the maximum functional recovery in pediatric extremity bone sarcoma reconstruction with the fibula flap takes place in the first year after surgery, with discrete gain afterward.

A long follow-up is essential to interpret the results.^{3,15} These are complex surgeries with lengthy NWB periods and a long recovery time. They also need intense rehabilitation to achieve optimal outcomes. Complications such as fractures may appear, which delay the recovery and may require additional surgeries. In addition, Starnes-Roubaud et al.¹⁹ demonstrated the negative impact of adjuvant therapies



Figure 6 MRI revealing an Ewing sarcoma of the calcaneus.

such as QT in pediatric microsurgery, leading to poor wound healing, bony nonunions, and infections.

Reported complication rates in pediatric lower extremity sarcoma reconstruction with the fibula flap are as high as 64–69%.^{3,20} About 35% of patients develop early minor complications, whereas 29% of patients experience nonbony major complications.³ Major bony complications include fractures (41%), nonunions (18%), and infections (14%). About 29% of patients need additional surgeries to solve nonbony major complications.³ These rates are lower considering the upper extremity, showing a 56.9% overall complication rate and a 11.7% fracture rate. On the contrary, the need for additional surgeries increases to 34.5%.²¹

The overall complication rate in the present study was 74.07%. Early minor complications appeared in 18.52% of patients and healed with conservative management. Sixteen patients (59.26%) experienced bony or articular major complications, including fractures (34.04%) and nonunions (11.11%). Fourteen patients (51.85%) underwent additional surgeries for their correction. At last follow-up, infection rate remained 3.7%. These complication rates are slightly below the average in comparison with other studies.^{3,20}

This study demonstrated that major complications are significantly associated with longer periods until WB

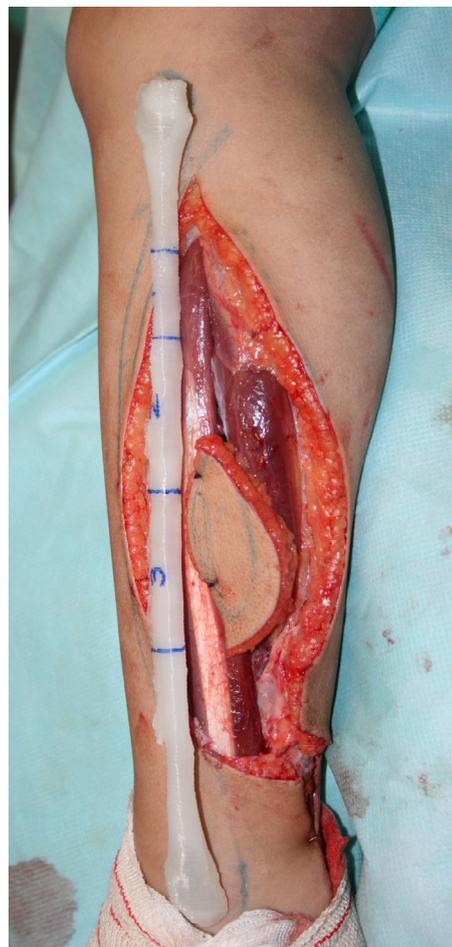


Figure 7 Intraoperative planning with a 3D-printed PLA 1:1 model. Single-barreled reverse flow pedicled osteocutaneous fibula flap.

resumption and FWB, along with the need for additional surgeries. On the contrary, major complications are not associated with poorer functional outcomes or inferior FWB rates (p 0.41) (Figure 3(A) and (B), online video content 1, which illustrates functional outcomes 24 months after ORIF of a fractured fibula for femoral reconstruction).

The age appeared as a relevant feature concerning both complications and functional outcomes, with an age <8 years significantly associated with a lower major complication rate and a shorter period until WB resumption. This finding may be related to a superior bone osteointegration and remodeling capacities, along with a higher tolerance to the deleterious effects of QT, characteristic of younger individuals. Previous studies concerning the free fibula flap for bone reconstruction in pediatric patients do not stratify by age groups. Consequently, they do not provide any information about how the age may affect the functional outcomes.^{7,11-13,18,19}

Reconstruction with the Capanna technique

This technique combines a nonvascularized osseous allograft with a vascularized fibula flap for the reconstruction

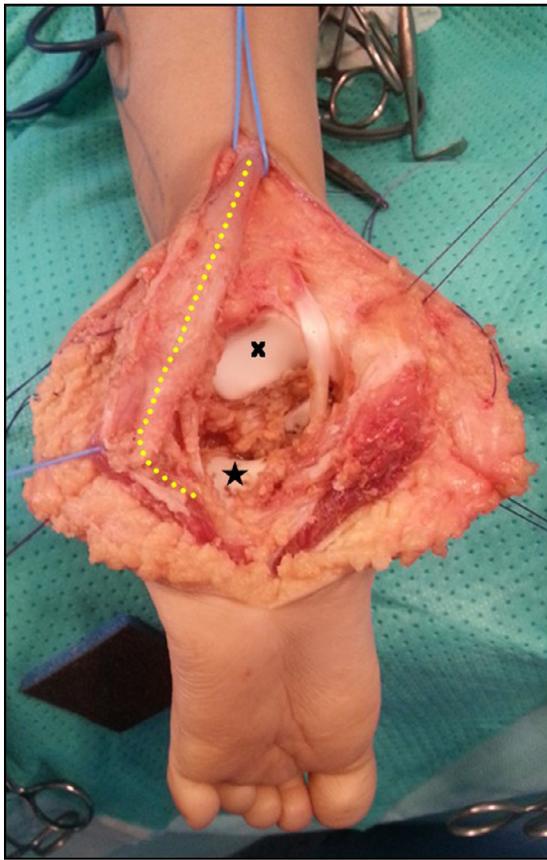


Figure 8 *En bloc* resection of the calcaneus, posterosuperior cortical margin preserved for insertion of the Achilles tendon (yellow dots). Cross: talus; star: cuboid.

of large intercalary defects. It takes advantage of the combined individual properties of each component and allows for an earlier WB.²²⁻²⁴ High complication rates have been reported when using massive bone allografts alone, including 5-30% of infections, 12-63% of nonunions, and 7-21.4% of fractures.^{19, 25-28}

Recent studies concerning the Capanna technique for the reconstruction of bony defects of the lower extremities after tumor resection have reported encouraging results. In their pediatric series of 12 patients, Weichman et al.¹⁵ reported a mean of 14.3 months until FWB was achieved, preventing amputation in all cases. They also reported a 67% overall complication rate, including a 16.7% of construct removal due to infections, and a 25% of both fracture and nonunion rates. Additionally, 58.7% of patients needed subsequent surgeries. According to Halim et al.,¹⁶ these patients reported a mean of 7.2 months until FWB was achieved, and an 8.3% amputation rate in their series of upper and lower extremity reconstruction after bone sarcoma resection. A 16.7% of both construct removal and fracture rates in the later series must be highlighted, along with a 33% of additional surgeries.

In the present pediatric cohort, 15 lower extremity Capanna reconstructions were performed, showing an overall complication rate of 54.33%, including a 40% fracture rate and a 13.33% of nonunion rate. Seven patients (46.67%) underwent subsequent surgeries for functional purposes.

Amputations were prevented in all cases but one. None of the constructs needed to be removed, and just one case of hardware infection was encountered.

These results are more favorable than previously reported series regarding overall complication, infection, nonunion, and construct removal rates, as well as the need for additional surgeries.^{15,16} Fracture rate remained slightly above the average, although it should be noted that the present cohort of Capanna reconstructions dealt exclusively with pediatric lower WB extremities receiving neoadjuvant and adjuvant QT. Besides, no statistically significant differences were encountered between Capanna and non-Capanna reconstructions regarding additional surgeries and minor, major, and overall complication rates.

This study has limitations. Firstly, the relatively small sample size of this series hampers the generalization of the results. Although reaching statistical significance, they must be interpreted cautiously. Secondly, a mean follow-up of 44.33 months might be insufficient for oncologic surveillance: recurrences and/or distant metastasis may appear with longer periods. In fact, follow-up time was significantly associated with the appearance of metastasis. Consequently, the reported oncologic outcomes may vary with time. The relatively low incidence of pediatric bone sarcomas hinders the realization of large sample-sized prospective controlled studies.

Finally, the oncologic nature of the disease carries the need for pre- and postoperative QT, which may jeopardize healing and osteointegration, leading to higher rates of major complications and additional surgeries. Unfortunately, for ethical reasons, a prospective controlled study is not feasible.

Case 1

A 9-year-old girl presented with right thigh pain for two months. X-ray showed a lytic lesion in the midfemoral shaft (Figure 4). Biopsy revealed a conventional osteosarcoma. Resection and immediate Capanna reconstruction were preoperatively planned by means of an MRI and virtual reconstruction and a PLA 1:1 3D-printed model. A 16-cm intercalary defect was reconstructed. No complications were encountered (Figure 5). The patient achieved PWB and FWB at 3 and 6 months after surgery, respectively (online video content 2, which illustrates functional outcomes 24 months after surgery).

Case 2

A 6-year-old girl presented with pain and swelling of her left heel for four weeks. An MRI revealed a tumor invading the calcaneus (Figure 6). Biopsy was positive for Ewing sarcoma. An ipsilateral reverse-flow pedicled osteocutaneous fibula flap was preoperatively evaluated by means of virtual reconstruction and 3D-printed PLA 1:1 models (Figure 7). Only the posterosuperior cortical margin of the calcaneus and the insertion of the Achilles tendon were preserved (Figure 8).

No complications were encountered. The patient achieved PWB and FWB at 6 and 10 months after surgery, respectively (Figure 9). She is able to walk and run. She uses



Figure 9 30 months after surgery. (A) Radiologic control showing the reverse flow pedicled osteocutaneous fibula flap. (B) 2 cm length discrepancy.

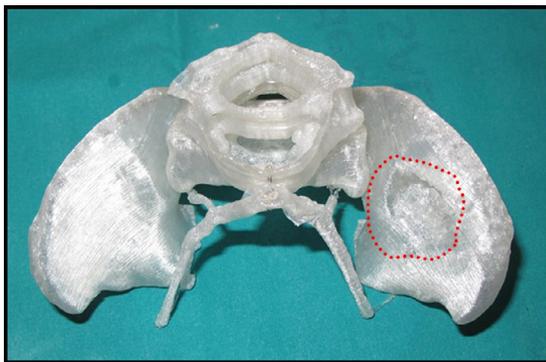


Figure 10 3D polylactic acid 1:1 model used for preoperative planning of the resection and immediate reconstruction.



Fig. 12 X-ray 14 months after surgery showing consolidation and hypertrophy of the construct.

a shoe lift to compensate a 2-cm height discrepancy (online *video content 3*, which shows functional outcomes without shoe lift, 20 months after surgery).

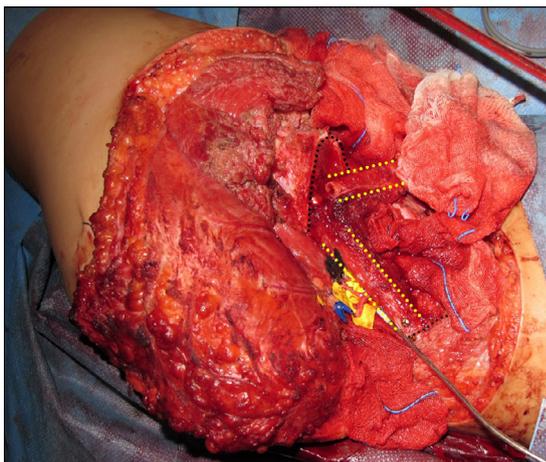


Fig. 11 Hemipelvectomy, resection margins: acetabular roof and sacroiliac joint (black dots). Double-barreled free fibula flap (yellow stripes). Anastomosis to the superior gluteal vessels (clamps).

Case 3

A 3-year-old boy presented with a history of two months of pain at his left iliac region. X-ray showed a lytic lesion in the left iliac bone. Biopsy was positive for Ewing sarcoma. Resection from the left sacroiliac joint to the acetabular roof and reconstruction with a double-barreled free fibula flap were preoperatively planned by means of a CT-scan and virtual reconstruction and a 3D-printed PLA 1:1 model (Figure 10). Anastomoses were performed end-to-end to the superior gluteal vessels (Figure 11).

No complications were encountered. The patient achieved PWB and FWB at 3 and 7 months after surgery, respectively. He walks with a Trendelenburg limp. However, he is able to walk and run, and has resumed sports practice (Figure 12) (online video contents 4 and 5, which illustrate the functional outcomes 30 months after surgery and the consolidation and hypertrophy of the fibula, respectively).

Conclusion

Extremity reconstruction after bone sarcoma resection in the pediatric population remains a challenge. The fibula flap allows the majority of extremities to be salvaged with satisfactory long-term outcomes. Full weight bearing is usually achieved within the first year, with modest functional increase afterward. Notwithstanding, a high rate of complications and additional surgeries should be anticipated. Less complications and a faster functional recovery are expected in patients below the age of 8 years.

Declaration of Competing Interest

None.

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Supplementary materials

Supplementary material associated with this article can be found, in the online version, at doi:[10.1016/j.bjps.2019.08.009](https://doi.org/10.1016/j.bjps.2019.08.009).

References

1. Abed R, Grimer R. Surgical modalities in the treatment of bone sarcoma in children. *Cancer Treat Rev* 2010;**36**:342-7.
2. Wilkins RM, Cullen JW, Odom L, et al. Superior survival in treatment in primary non-metastatic pediatric osteosarcoma of the extremities or trunk: an analysis of 1702 patients treated on neoadjuvant cooperative osteosarcoma study group protocols. *Ann Surg Oncol* 2003;**10**:498-507.
3. Piper M, Irwin C, Sbitany H. Pediatric lower extremity sarcoma reconstruction: a review of limb salvage procedures and outcomes. *J Plast Reconstr Aesthet Surg* 2016;**69**:91-6.
4. Yaw KM. Pediatric bone tumors. *Semin Surg Oncol* 1999;**16**:173-83.
5. Rougraff BT, Simon MA, Kneisl JS, et al. Limb salvage compared with amputation for osteosarcoma of the distal end of the femur. A long-term oncological, functional, and quality-of-life study. *J Bone Jt Surg Am* 1994;**76**:649e56.
6. Weber KL. What's new in musculoskeletal oncology. *J Bone Joint Surg Am* 2005;**87**:1400-10.
7. Sainsbury DC, Liu EH, Alvarez-Veronesi MC, et al. Long-term outcomes following lower extremity sarcoma resection and reconstruction with vascularized fibula flaps in children. *Plast Reconstr Surg* 2014;**134**:808-20.
8. Taylor GI, Miller G, Ham FJ. The free vascularized bone graft: a clinical extension of microvascular techniques. *Plast Reconstr Surg* 1974;**55**:533-44.
9. Taylor GI, Corlett RJ, Ashton MW. The evolution of free vascularized bone transfer: a 40-year experience. *Plast Reconstr Surg* 2016;**137**:1292-305.
10. Sparks DS, Saleh DB, Rozen WM, et al. Vascularised bone transfer: history, blood supply and contemporary problems. *J Plast Reconstr Aesthet Surg* 2017;**70**:1-11.
11. El-Gammal TA, El-Sayed A, Kotb MM. Reconstruction of lower limb bone defects after sarcoma resection in children and adolescents using free vascularized fibular transfer. *J Pediatr Orthop B* 2003;**12**:233-43.
12. Ghert M, Colterjohn N, Manfrini M. The use of free vascularized fibular grafts in skeletal reconstruction for bone tumors in children. *J Am Acad Orthop Surg* 2007;**15**:577-87.
13. Kurlander DE, Shue S, Schwarz GS, et al. Vascularized fibula epiphysis transfer for pediatric extremity reconstruction: a systematic review and meta-analysis. *Ann Plast Surg* 2019;**82**:344-51.
14. Scaglioni MF, Chang EI, Gur E, et al. The role of the fibula head flap for joint reconstruction after osteoarticular resections. *J Plast Reconstr Aesthet Surg* 2014;**67**:617-23.
15. Weichman KE, Dec W, Morris CD, et al. Lower extremity osseous oncologic reconstruction with composite microsurgical free fibula inside massive bony allograft. *Plast Reconstr Surg* 2015;**136**:396-403.
16. Halim AS, Chai SC, Wan Ismail WF, et al. Long-term outcome of free fibula osteocutaneous flap and massive allograft in the reconstruction of long bone defect. *J Plast Reconstr Aesthet Surg* 2015;**68**:1755-62.
17. Feuvrier D, Sagawa Y Jr, Béliard S, et al. Long-term donor-site morbidity after vascularized free fibula flap harvesting: clinical and gait analysis. *J Plast Reconstr Aesthet Surg* 2016;**69**:262-9.
18. Schwarz GS, Disa JJ, Mehrara BJ, et al. Reconstruction of oncologic tibial defects in children using vascularized fibula flaps. *Plast Reconstr Surg* 2012;**129**:195-206.
19. Starnes-Roubaud MJ, Hanasono MM, Kupferman ME, et al. Microsurgical reconstruction following oncologic resection in pediatric patients: a 15-Year experience. *Ann Surg Oncol* 2017;**24**:4009-16.
20. Shea KG, Coleman DA, Scott SM, et al. Microvascularized free fibular grafts for reconstruction of skeletal defects after tumor resection. *J Pediatr Orthop* 1997;**17**:424-32.
21. Landau MJ, Badash I, Yin C, et al. Free vascularized fibula grafting in the operative treatment of malignant bone tumors of the upper extremity: a systematic review of outcomes and complications. *J Surg Oncol* 2018;**117**:1432-9.
22. Donati D, Capanna R, Campanacci D, et al. The use of massive bone allografts for intercalary reconstruction and arthrodeses after tumor resection: a multicentric European study. *Chir Organ Mov* 1993;**78**:81-94.
23. Capanna R, Campanacci DA, Belot N, et al. A new reconstructive technique for intercalary defects of long bones: the association of massive allograft with vascularized fibular autograft: long-term results and comparison with alternative techniques. *Orthop Clin N Am* 2007;**38**:51-60 vi.

24. Lee JE, Kim MB, Han DH, et al. One-Barrel microsurgical fibula flap for reconstruction of large defects of the femur. *Ann Plast Surg* 2018;**80**:373-8.
25. Gebhardt MC, Flugstad DI, Springfield DS, et al. The use of bone allografts for limb salvage in high-grade extremity osteosarcoma. *Clin Orthop Relat Res* 1991;**270**:181-96.
26. Mankin HJ, Springfield DS, Gebhardt MC, et al. Current status of allografting for bone tumors. *Orthopedics* 1992;**15**:1147-54.
27. Muscolo DL, Ayerza MA, Aponte-Tinao L, et al. Intercalary femur and tibia segmental allografts provide an acceptable alternative in reconstructing tumor resections. *Clin Orthop Relat Res* 2004;**426**:97-102.
28. Ortiz-Cruz E, Gebhardt MC, Jennings LC, et al. The results of transplantation of intercalary allografts after resection of tumors: a long-term follow-up study. *J Bone Joint Surg Am* 1997;**79**:97-106.