

Case Report

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Successful Resection of Ewing Sarcoma Involving the Whole Fibula in a Pediatric Patient with Long Vascular Bypass and Reconstruction of Knee and Ankle Ligaments: A Case Report and Literature Review

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ABSTRACT

Introduction: Ewing sarcoma is the second most prevalent primary malignant bone tumor affecting pediatric and adolescent patients. Ewing sarcoma can affect any bone, but it primarily affects bones in the lower limb and pelvis. The femur is the most commonly affected bone. Approximately 8% of primary Ewing sarcoma originates from the fibula, and involvement of the entire fibula is a rare presentation. The standard treatment of Ewing sarcoma involves neo-adjuvant multimodal chemotherapy followed by local treatment using wide local resection (with or without radiation) and adjuvant chemotherapy.

Case Presentation: A nine-year-old male patient, who is medically free, presented to the orthopedic oncology clinic complaining of left leg pain and swelling, accompanied by nighttime pain and weight loss. Clinical examination revealed swelling and tenderness in the entire leg, particularly the lateral aspect, and distal neurovascular examination was normal. Radiographic assessment, including plain X-rays and whole leg MRI with contrast, suggested the presence of a potentially malignant lesion affecting the entire fibula. CT-guided biopsy and histopathology confirmed the diagnosis of Ewing Sarcoma of bone. CT Angiogram of the left lower limb was performed to assess vascular involvement, which revealed the anterior tibial artery and peroneal artery were found be encased by the tumor. Following neo adjuvant chemotherapy, the patient underwent total fibular resection, long vascular bypass and reconstruction of the ankle and knee ligaments. Postoperative follow-up and recovery yielded maintained ankle and knee stability, resulting in a satisfactory functional outcome.

Conclusion: Ewing sarcoma involving the entire fibula is exceptionally rare, with limited experience reported in the literature. In the present pediatric case, complete tumor resection, long segment vascular bypass, and concurrent reconstruction of the ankle and knee ligaments were performed, resulting in durable stability of both joints. This experience underscores that, when guided by a multidisciplinary team, total fibular resection can offer an effective and function-preserving procedure in pediatric bone sarcoma.

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Abbreviations

MRI: Magnetic Resonance Image

CT: Computed Tomography

IV: Intravenous

CFL: Calcaneo-Fibular Ligament

Keywords: Ewing Sarcoma, Fibula, Whole Fibula, Vascular Bypass, Reconstruction of Ankle and Knee Ligaments, Limb Salvage Surgery

Introduction

Ewing Sarcoma is a malignant, small round cell sarcoma that ranks as the second most prevalent primary bone sarcoma in the pediatric and adolescent populations. It constitutes approximately 10-15% of all malignant bone tumors [1]. In the appendicular skeleton, Ewing Sarcoma frequently affects the distal femur, tibia, and humerus. Fibula Ewing Sarcoma accounts for 8% of primary Ewing Sarcoma of bone [2]. When presenting in the fibula, the proximal third of the fibula is the most common site

affected whereas involvement of the entire fibula is an uncommon presentation [3]. Ewing Sarcoma typically presents with pain, swelling, and constitutional symptoms. The diagnosis of Ewing Sarcoma, like other malignancies, is a multidisciplinary approach that involves plain radiographs, whole limb MRI with intravenous contrast, and histopathological assessment through a Tru-Cut biopsy. Systemic staging is performed by computed tomography of the chest and bone scan or positron emission tomography scan [4]. Bone marrow aspiration is an additional consideration for its prognostic value in the case of Ewing Sarcoma [5]. The standard treatment regimen includes neoadjuvant chemotherapy followed by local control through either surgery or radiation therapy, and adjuvant chemotherapy.

Case Presentation

A nine-year-old male patient presented to the orthopedic oncology clinic with left leg pain persisting for eight months. The pain improved with simple analgesia but was accompanied by nighttime pain disrupting sleep and weight loss. A recent swelling in the left leg had also developed and was progressively increasing in size.

On physical examination, the patient had a tender swelling over the left leg, preserved ankle and knee joint range of motion, and

intact neurovascular status. Systemic examination revealed no abnormalities.

Initial radiographic assessment showed a potentially malignant lesion involving the entire fibula. The patient was admitted, administered analgesia, and immobilized with axillary crutches.

Local staging was performed using MRI and CT angiogram. A multidisciplinary discussion with the musculoskeletal radiologist revealed a large tumor involving the entire fibula with a large soft tissue component. The tumor is encasing the anterior tibial artery, Tibio-peroneal trunk, and peroneal nerve (Figure 2, A-F). A CT-guided biopsy confirmed Ewing sarcoma of the fibula.

Systemic staging revealed no evidence of distant metastasis. The case was reviewed by the pediatric oncology tumor board as part of a multidisciplinary management strategy. Consequently, the decision was made to initiate the patient on neoadjuvant chemotherapy, followed by local control and adjuvant chemotherapy. Patient presented for local control, which was decided to be total fibular resection, long Vascular bypass grafting and ligament reconstruction of the ankle and knee joints.

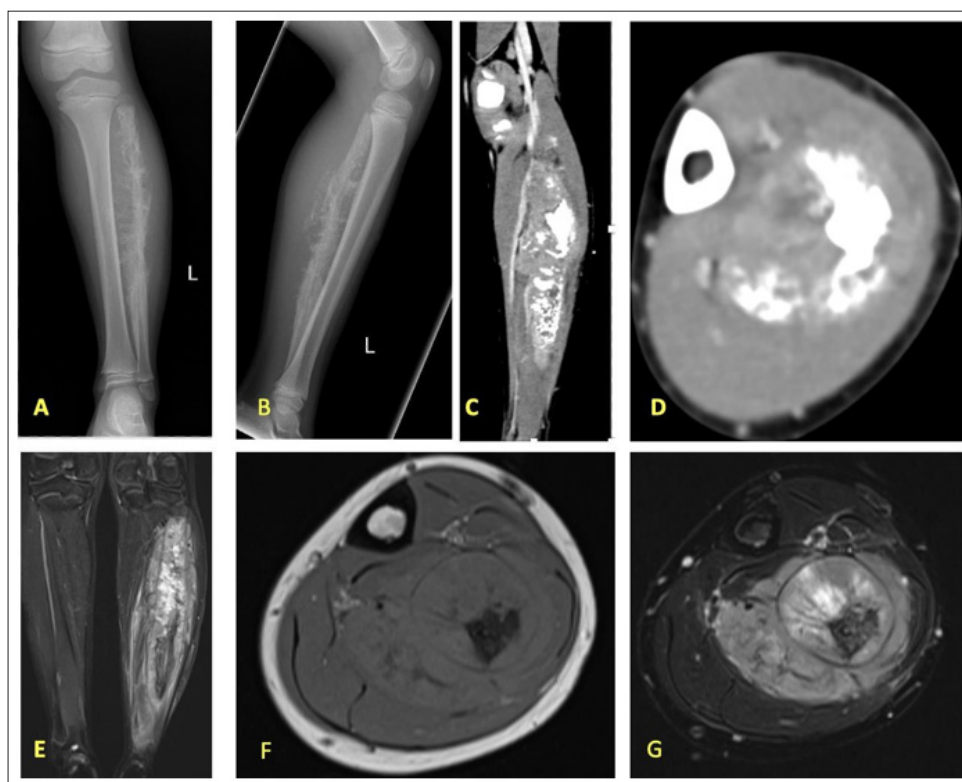


Figure 1: Pre Chemotherapy Evaluation. (A-B) AP and Lateral Views of the Tibia show Extensive Aggressive Looking Lesion Involving the Entire Fibula Shaft with Aggressive Periosteal Reaction and Sunburst Appearance as well as Soft Tissue Mass. (C-D) Coronal and Axial views of the CT Angiogram Showing Encasement of the Tibioperoneal truck and Posterior Tibial Artery. The Peroneal Artery courses inside the Tumor Mass and becomes Untraceable. (E-F) Coronal STIR, Axial T1 and Axial STIR Weighted Magnetic Resonance Images of the Left Leg showing the Aggressive Fibular mass as Heterogeneously Hyperintense on STIR and Isointense to slightly Hyperintense to Muscle on T1 Weighted Images.

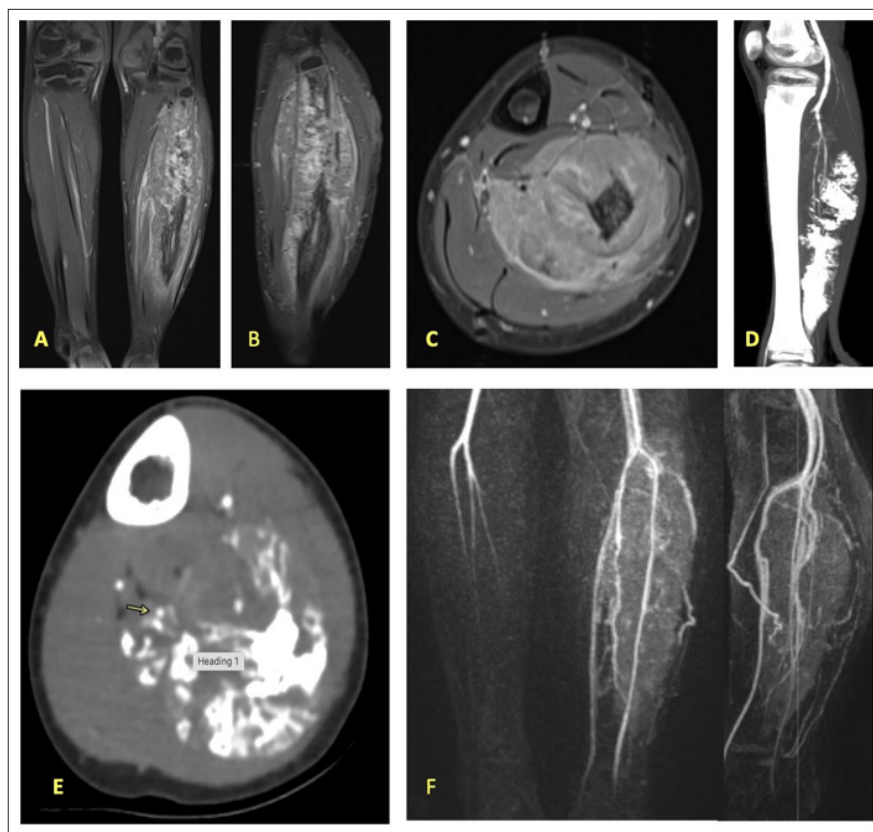


Figure 2: Post Chemotherapy Evaluation. (A-C) Coronal, Sagittal and Axial T1 Post Gadolinium Enhanced Magnetic Resonance Images of the Left Leg show the Aggressive Fibular Mass with Avid Heterogenous Post Contrast Enhancement (D-E) Sagittal MIP, Maximum Intensity Projection, and Axial CTA Images Showing Shrinkage of the Tumor Mass with Persistent Encasement of the Tibioperoneal Trunk Indicated by the Arrow. (F) Coronal and Sagittal Magnetic Resonance Digital Subtraction Angiogram Images Showing the Aggressive Hyper Vascular Fibular Mass with its Arterial Relationship

Surgical Technique

Under general anesthesia, the patient was positioned in the left lateral decubitus position with adequate padding of all bony prominences. The left lower limb was prepared and draped in a standard surgical manner. A direct lateral approach to the fibula was employed. Skin incisions were made over the fibula from the mid-popliteal fossa to the lateral malleoli. Superficial dissection with hemostasis was maintained.

The sural nerve was initially identified and protected. The lateral gastrocnemius muscle was identified and released to expose the soleus muscle, which was dissected more proximally to identify the popliteal artery, the Tibio-Peroneal trunk, and the anterior tibial artery. The common peroneal nerve was completely encased within the tumor; therefore, it was sacrificed. Additionally, the saphenous vein was identified and protected throughout the surgery for potential use as a vascular graft.

The anterior tibial artery was found to be involved within the tumor proximally, as predicted from preoperative angiography assessment. Consequently, it was clamped proximally. Intraoperative evaluation of the proximity of the posterior tibial artery to the tumor margin was deemed unsafe, necessitating its sacrifice as well. Distally, the anterior tibial artery was identified just above the ankle joint, clamped.

The proximal and distal ends of the fibula were exposed, with preservation of the calcaneofibular ligament, talofibular ligament, and peroneal tendons to facilitate subsequent reconstruction. The

lateral collateral ligament was also preserved proximally after being detached from the fibular head. Proximal and distal fibular disarticulations were performed, followed by en bloc resection of the entire fibula and its associated structures. Negative surgical margins were confirmed intraoperatively by frozen section analysis.

Vascular reconstruction was performed by the vascular team through harvesting and reversing the small saphenous vein. The distal end of the small saphenous vein was anastomosed to the popliteal artery proximally (end-to-end) using 6/0 Prolene sutures. Conversely, the proximal part of the vein was anastomosed distally to the anterior tibial artery (end-to-side) using 7/0 Prolene sutures, as the posterior tibial artery distally is very small to accommodate vascular anastomosis. The dorsalis pedis artery distal to the anastomosis was palpable, and distal pulse was biphasic on Doppler assessment.

To execute the reconstruction, the calcaneofibular ligament and anterior talofibular ligament were anchored to the distal tibia via suture anchors. Additionally, the peroneal tendons were augmented around the ankle by suture anchors to the tibial epiphysis. Proximal reconstruction was achieved by attaching the lateral collateral ligament to the proximal tibia and reinforced with the tensor fascia lata for additional stability (Figure 3, A-E).

The surgical wound was thoroughly irrigated, a drain was placed, and layered closure was performed.

The limb was immobilized with a cast to be placed for six weeks.

Post-operative radiographs showed complete resection of the fibula with stable congruent ankle and knee joints (Figure 4, A-B).

At six weeks postoperatively, the cast was removed to initiate range of motion and strengthening exercises. Final histopathological analysis of the resected specimen confirmed complete fibular excision with negative surgical margins (Figure 4, D-E).

Regular follow-up examinations have consistently demonstrated excellent stability of the ankle and knee joints, allowing the patient to walk independently. No evidence of local recurrence has been observed two years after surgery and radiographs showed maintained alignment of the knee and ankle joints with no evidence of deformity (Figure 4, C).

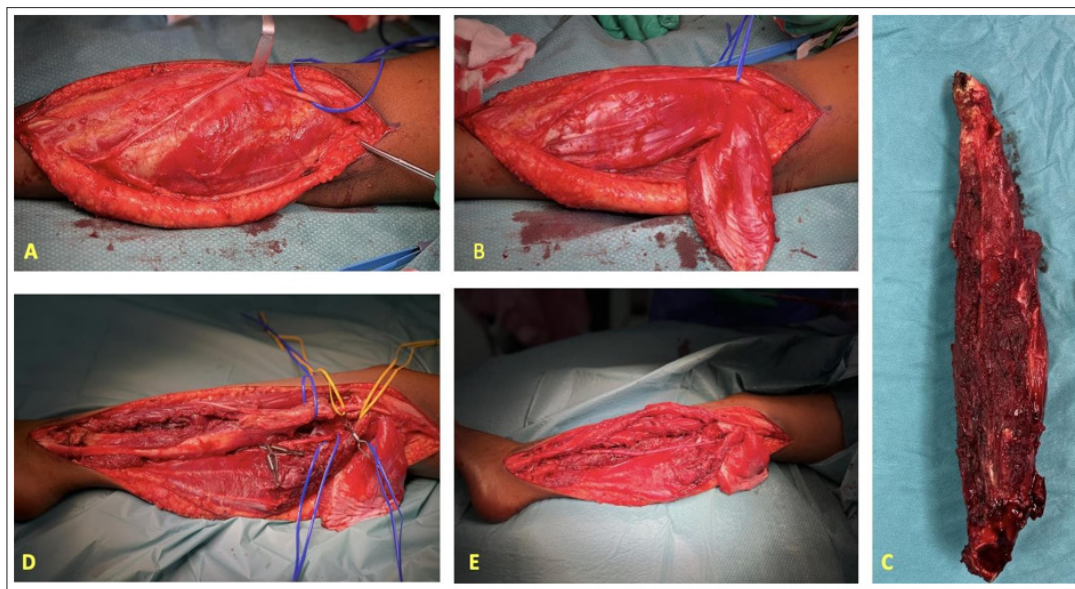


Figure 3: Surgical Technique. (A) Standard Lateral Approach to the Fibula with Identification of common Peroneal Nerve and Popliteal Artery. (B) Lateral Gastrocnemius was Identified. (C) Resected Fibula. (D) Popliteal Artery Identified and Clamped. (E) Vascular Bypass Graft using the great Saphenous vein to Bypass the Popliteal to Anterior Tibial Artery.

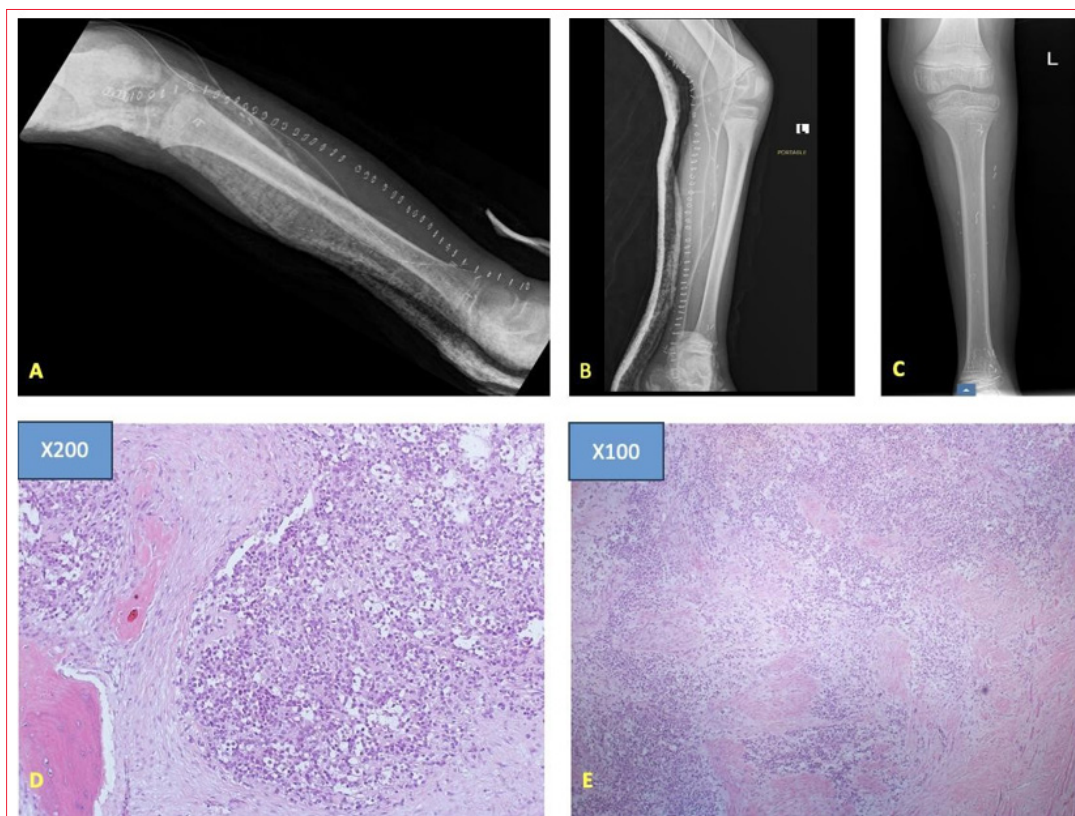


Figure 4: (A-B) Immediate Post-Operative Radiographs in AP and Lateral Projection. (C) AP view Two Years Post-Surgery. (D-E) Histological Assessment of the Resection Specimen after Receiving Neoadjuvant Therapy Showing Residual Sheets of Monomorphic Round Blue Cells Infiltrating the Bone (A, X 200) and Adjacent Soft Tissue (B, X 100).

Discussion

Ewing sarcoma ranks as the second most prevalent primary malignant bone tumor in children and adolescents, predominantly affecting long bones such as the femur, tibia, and pelvis. Primary involvement of the fibula is uncommon, comprising less than 10% of cases. In pediatric patients, Ewing sarcoma of the fibula presents distinct surgical challenges owing to the fibula's anatomical location, its proximity to vital neurovascular structures, and its significance in stabilizing both the knee and ankle joints.

When located in the fibula, the tumor often extends to involve nearby critical structures, including the common peroneal nerve, lateral collateral ligament (LCL), and the popliteal trifurcation. The proximal fibula also serves as the attachment site for key stabilizing structures such as the arcuate ligament, biceps femoris tendon, and iliotibial band, which are essential for knee stability, further complicating surgical management [6]. The distal fibula serves as an anatomic stabilizer for the ankle joint and provides attachment to the lateral ankle ligaments, which further enhance the stability of the ankle joint.

The standard treatment approach consists of neoadjuvant chemotherapy, surgical resection with or without radiation therapy, and subsequent adjuvant chemotherapy. The surgical objective is the complete excision of the tumor with histologically negative margins, which substantially enhances local control and long-term survival [7]. Advancements in surgical techniques have progressively facilitated limb-sparing resections, even in anatomically intricate locations [8, 9].

Achieving wide surgical margins during fibular resection presents a significant challenge due to the close proximity of vital neurovascular structures and joint stabilizers. Furthermore, preserving these margins for sarcomas originating from the fibula can be particularly demanding, even for highly experienced orthopedic oncologists.

Malawer described two main surgical techniques for fibular resection based on tumor type and extent [9]. Type I resection, used for benign tumors, involves removing the proximal fibula with a 1–2 cm segment of normal diaphysis and preserving the neurovascular bundle. Type II, used for malignant tumors, involves a wide intra-compartmental resection, including a 3 cm segment of normal diaphysis, the anterior and lateral muscle compartments, the common peroneal nerve, anterior tibial artery, and the proximal tibiofibular joint. Reconstruction with a gastrocnemius muscle flap is common after type II resections. Type III and IV resections may preserve the superficial peroneal nerve or the proximal tibiofibular joint, respectively.

Proximal fibular tumor resection is complex due to nearby neurovascular structures. In nine patients treated between 1992–2002, wide en-bloc resection achieved good tumor control without recurrences, though some developed peroneal nerve palsy requiring tendon transfers. Wide resection is recommended for large or recurrent tumors, with careful reconstruction to optimize function [10].

Some studies suggest that satisfactory knee function can be maintained even in the absence of LCL reconstruction, particularly when other stabilizing structures such as the popliteus, lateral gastrocnemius, and posterior joint capsule are preserved. Postoperative fibrosis and scarring following radiotherapy may also contribute to joint stability. However, when distal fibular resection is involved, maintaining ankle stability is more difficult.

If unreconstructed, patients may experience varus instability, varus collapse, altered gait, and early-onset ankle arthritis. Techniques to restore ankle stability after fibular resection include ankle arthrodesis, soft tissue reconstruction, or bone grafting. Arthrodesis can be achieved through tibiotalar or tibiotalarocalcaneal fusion using intramedullary nails or screws, although this sacrifices ankle motion and carries the risk of delayed union, especially in patients undergoing chemotherapy or radiotherapy.

There is a scarcity of data on outcomes of primary fibular sarcomas in literature. Only few case series and case reports have discussed the survival and functional outcome of high-grade sarcomas at this location [11].

Despite these challenges in the surgical management of this case, the outcome was remarkably favorable. The patient exhibited no local recurrence, demonstrated stable joint function, and maintained independent ambulation at the two-year follow-up. This outcome highlights the effectiveness of a multidisciplinary approach combining limb-salvage surgery, vascular reconstruction, and joint stabilization in managing complex and rare presentations.

Outcomes of Limb Salvage Surgery in Primary Fibular Sarcomas, between 2014 and 2018, fourteen patients with primary fibular sarcomas were reviewed, with thirteen undergoing limb salvage surgery, mostly involving Malawer type II resection. At a mean follow-up of 37 months, 10 patients remained disease-free, while distant or combined relapse occurred in four cases. Despite the difficulty of achieving wide margins, local recurrence rates were not higher than those reported for sarcomas at other sites, highlighting the effectiveness of limb salvage in managing fibular sarcomas [11].

Reconstruction techniques must be customized to the patient's anatomy and age, particularly in pediatric cases where growth potential must be preserved. Close postoperative monitoring is essential to detect recurrence and ensure functional recovery. This case underscores the recommendation that centers treating bone sarcomas should implement team-based, individualized treatment strategies to enhance outcomes in technically challenging cases.

Conclusion

Ewing sarcoma of the entire fibula is an uncommon presentation, and traditional management has often involved above-knee amputation. However, with advancements in surgical techniques and multidisciplinary collaboration, limb preservation with functional restoration is achievable even in complex cases. The proximity of the fibula to key neurovascular and stabilizing structures poses a significant challenge, particularly in children, but individualized, team-based care can yield favorable oncologic and functional outcomes.

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