CASE REPORT

Adamantinoma of the Tibial Shaft Treated with Intercalary Reconstruction Using Ipsilateral Pedicled Vascularized Fibular Graft and Screw Fixation*

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ABSTRACT

Adamantinoma is a rare primary bone tumor, typically affecting the tibia. While traditionally treated with amputation, limb salvage techniques have become the preferred approach, preserving extremity function. This article reports on a 52-year-old female with a growing mass on her left tibia, confirmed as adamantinoma through biopsy. The patient underwent wide resection of a 16 cm tibial segment and reconstruction using an ipsilateral pedicled vascularized fibular graft. The graft was secured with only one cortical screw each proximally and distally, and the medial gastrocnemius muscle was used to cover the graft. A long leg cast was then applied. We demonstrate a successful strategy for treating adamantinoma, focusing on achieving functional recovery and long-term tumor control.

Keywords. adamantinoma, tibial neoplasms, intercalary reconstruction, pedicled fibular graft, limb salvage procedure, vascularized graft hypertrophy

INTRODUCTION

Adamantinoma is a biphasic malignant bone tumor that occurs most commonly at the diaphyseal area of the tibia (80–85%) during the 2nd–5th decade of life with a male predilection. It is rare, accounting for only 0.5–1% of all primary bone tumors. Patients most often experience pain and a palpable mass at the anterior surface of the tibia. Radiographs show an eccentric, lobular, lytic lesion with sclerosing margins at the diaphysis of the tibia. It is likened to a "soap bubble" appearance due to the multifocal radiolucencies. The most common site of metastasis is the lungs.[1-3]

In the United States, 92 patients were diagnosed with adamantinoma from 1973 to 2016.[2] In the University of the Philippines – Philippine General Hospital, adamantinoma accounted for only three out of the 337 primary malignant bone tumors from 1993 to 2000.[3]

CASE

This was a case of a 52-year-old woman with no known comorbidities, who consulted for a one-year history of a mass on her left leg. She had no history of trauma, weight loss, night pains, or infection. She consulted with another institution when the mass became painful and larger. An open biopsy revealed an adamantinoma of the left tibia. She was advised surgical management but then opted to transfer to our institution.

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On examination, there was an approximately 8 x 2 x 3 cm hard, non-movable, tender, non-erythematous mass at the anterior aspect of the leg (Figure 1). The patient could do full range of motion of the ankle and knee and had no sensory deficits. No other lesions were palpated.

Radiographs of the left leg showed a large lytic lesion with permeative borders and neocorticalization with no matrix (Figure 2). An MRI revealed a 2.5 x 3 x 8 cm well-defined lobulated lesion in the anterior aspect of the proximal 1/3 of the tibial shaft with cystic and solid components, demonstrating T1-weighted hypointense and T2-hyperintense signals (Figure 3).

The incision was made starting at the anteromedial proximal tibia, creating an elliptical incision over the biopsy site, extending to the distal 3rd of the leg (Figure 4). Lateral and medial flaps were carefully dissected around the tumor. The tibia was osteotomized 3 cm proximally and 5 cm distally from the tumor to achieve wide resection. The entire resected specimen measured 16 cm in length. (Figure 5). The fibula was approached posterolaterally, marking osteotomy sites 5 cm

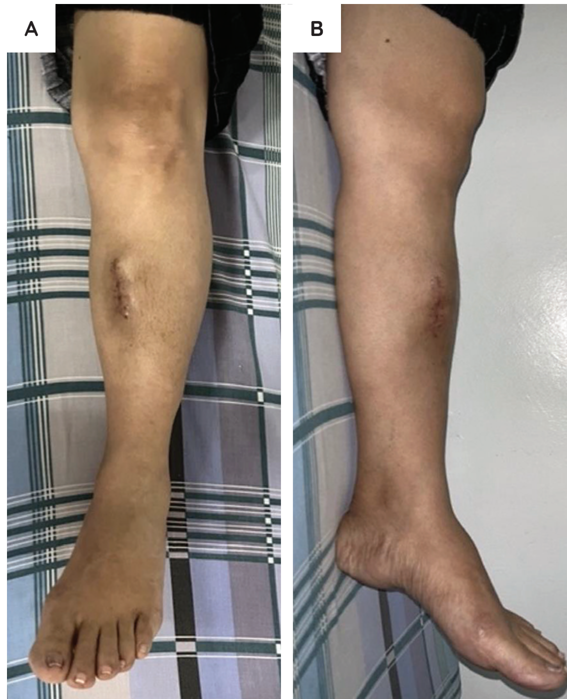

Figure 1. Gross appearance of the mass at the antero-medial middle 3rd of the left leg with the biopsy scar. Anterior **(A)** and medial **(B)** views.

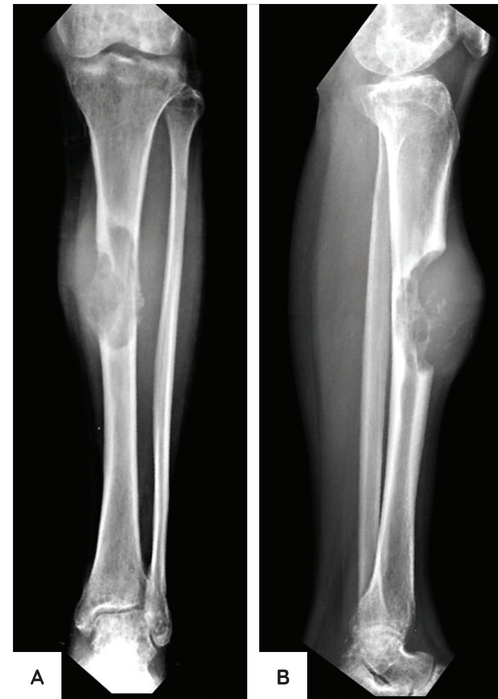

Figure 2. Anterio-posterior (AP) **(A)** and lateral **(B)** radiographs showing the tumor on the tibial shaft.

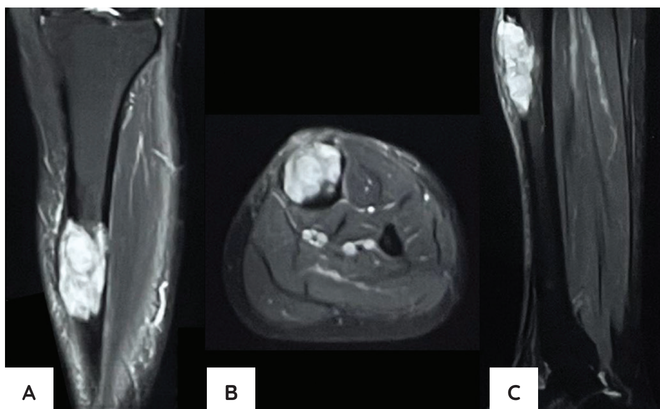

Figure 3. MRI T2 weighted images showing coronal **(A)**, axial **(B)**, and sagittal **(C)** cuts.

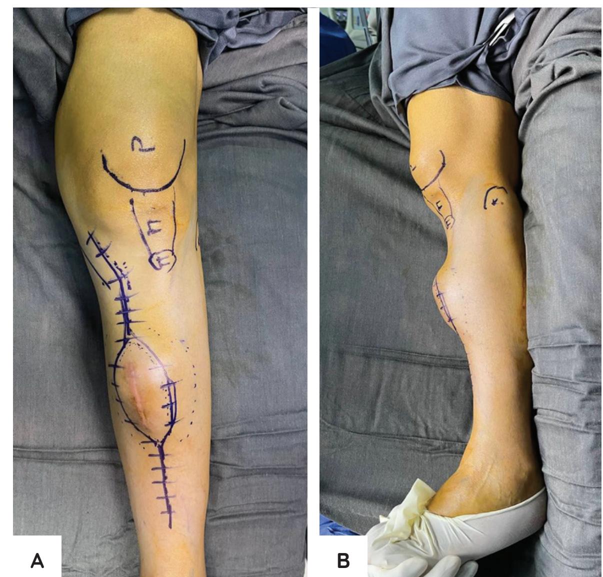

Figure 4. Landmarks and planned anteromedial approach **(A)**. Fibular head marked on its lateral aspect **(B)**.

below the fibular head and 8 cm above the lateral malleolus to reduce donor site morbidity and maintain ankle stability (Figure 6). A straight incision was made just posterior to the posterolateral septum. The interval was developed between the peroneus and soleus. The fibula was released from the superficial posterior compartment. The peroneus longus and brevis muscles were released from the fibula through an extraperiosteal plane, while the muscles of the anterior compartment were released close to the fibula. The anterior tibial artery and common peroneal nerve were identified and protected. Osteotomies were made on the previously marked levels. Dissection was done going proximally, following the middle of the V shape in the tibialis posterior muscle, preserving the proximal vascular bundle on the side of the fibula (Figure 7A). A total of 19 cm of pedicled fibular graft was harvested, tunneled towards the defect, and secured with two cortical screws (Figure 7B). The medial gastrocnemius was harvested from the superficial compartment and applied as soft tissue coverage for the graft (Figure 8). A long leg splint was applied and the patient was advised no weight

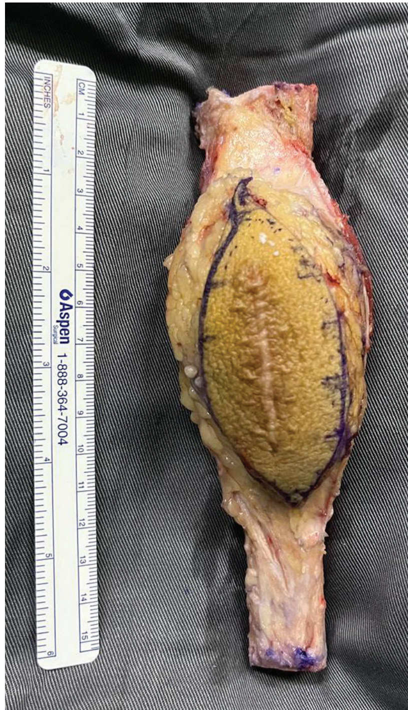

Figure 5. Resected mass measuring 16 cm in length.

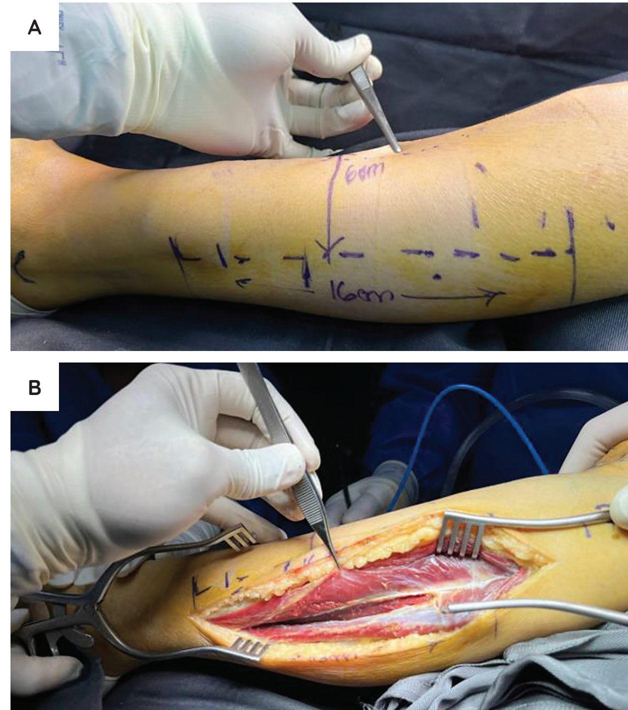

Figure 6. Markings of the lateral aspect of the left leg with a 6 cm skin island **(A)**. Interval between peroneus and soleus **(B)**.

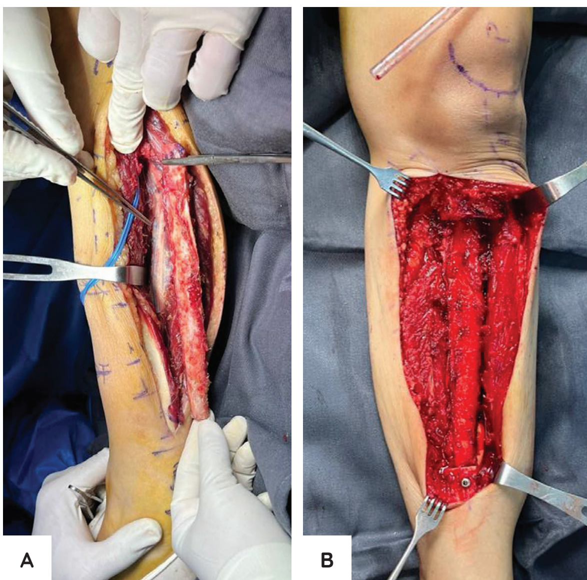

Figure 7. The 19 cm pedicled fibular graft was harvested **(A)** and tunneled to the defect and secured with two cortical screws **(B)**.

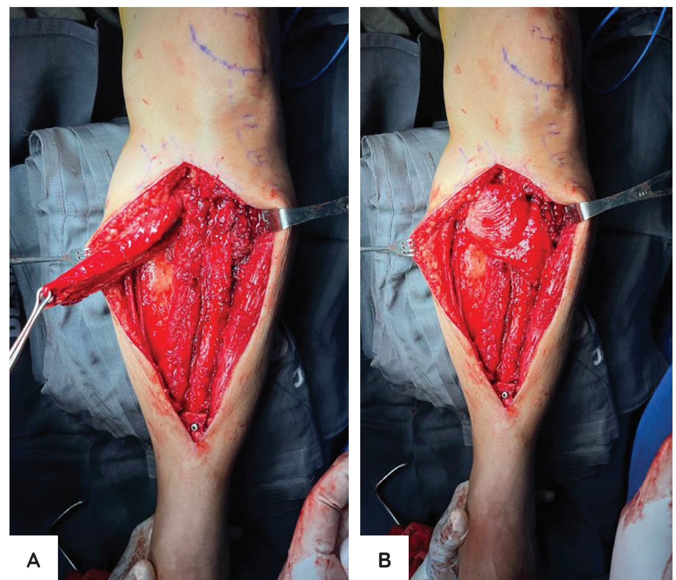

Figure 8. The medial gastrocnemius flap was harvested **(A)** and used for soft tissue coverage of the vascularized fibular graft **(B)**.

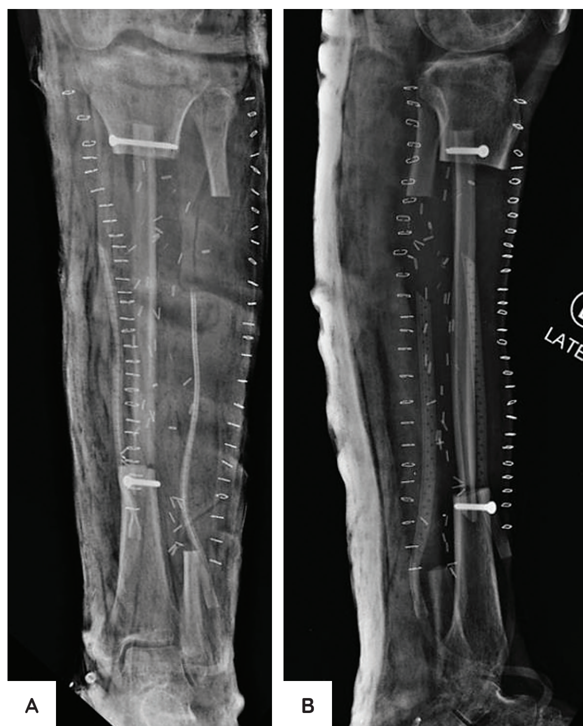

Figure 9. AP **(A)** and lateral **(B)** immediate postoperative radiographs showing the fibular strut graft fixated with two cortical screws.

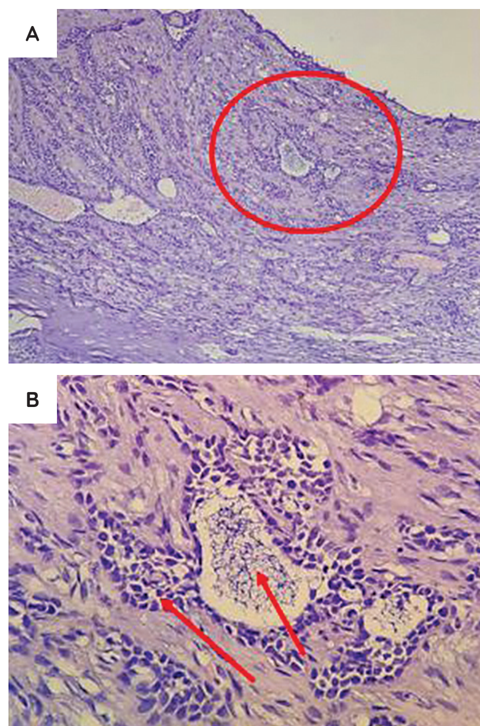

Figure 10. LPO **(A)** and HPO **(B)** Arrow shows basaloid cells in a palisading pattern, cribriform nests with dispersed chromatin pattern.

bearing. Immediate post-operative radiographs are seen in Figure 9. The splint was converted to a long leg cast at two weeks post-op.

The histopathologic examination revealed biphasic stromal and epithelial components, consisting of cords, trabeculae, solid, and cribriform nests observed under low power (LPO) and high power (HPO) magnification, consistent with adamantinoma. In Figure 10, the arrow indicates a basaloid cell arranged in a palisading pattern. Both the proximal and distal margins were free of tumor involvement. Follow-up radiographs after two months showed callus formation at both the distal and proximal fixation sites (Figure 11). At 28 weeks post-op, the long leg cast was converted to a walking boot and the patient was advised partial weight bearing with the help of axillary crutches to stimulate hypertrophy of the graft (Figure 12). At 44 weeks post-op, the proximal and distal tibia were completely united with beginning hypertrophy of the graft (Figure 13). The patient was able to fully extend and flex her knee up to 100 degrees (Figure 14). At 76 weeks post-op the patient had started full weight bearing without crutches and a pneumatic walking boot. There was no pain or local recurrence. Radiographs showed complete union of the graft to the tibia. Hypertrophy was seen on the entire length of the fibular graft (Figure 15). At 100 weeks post-op, the patient walked without any assistive device and exhibited full range of motion of both the ankle and knee joints (Figure 16). Radiographs showed further hypertrophy of the entire fibular graft with no signs of local recurrence (Figure 17).

DISCUSSION

This patient's mass was located at the anterior proximal 3rd of the tibia shaft, consistent with the disease's epidemiology. The most common presentation of Adamantinoma is pain and swelling of the affected site and in 72% of the cases, they may present with a palpable mass on the anterior tibia, as seen in our patient.[1]

Radiographically, adamantinoma presents as an eccentric/central lobular lytic lesion with well-defined sclerotic margins of overlapping radiolucency located at the tibial diaphysis. It is described as having a "soap bubble" appearance.[1] Our patient's radiographs showed a gradually enlarging multilobulated lytic lesion with signs of neocorticalization at the proximal left tibia.

Histologically, adamantinoma can be classified into osteofibrous-like and classic adamantinoma.[1] Classic adamantinoma can be classified into four patterns: namely basaloid which consists of solid masses of basaloid cells, tubular which contains cuboidal epithelial cells with central discohesion, spindle-cell which exhibits uniform spindling, and squamous which resembles squamous carcinoma.[1] Osteofibrous-like adamantinoma, on the other hand, consists of osteofibrous tissue containing clusters of epithelial cells and tests positive during cytokeratin staining.[4] Other characteristics of Adamantinoma include a low mitotic rate, foci calcification, giant cells, xanthoma, and spindle cells.[1,4] While its pathogenesis is still in question, the accepted theory is that the skin basal epithelium

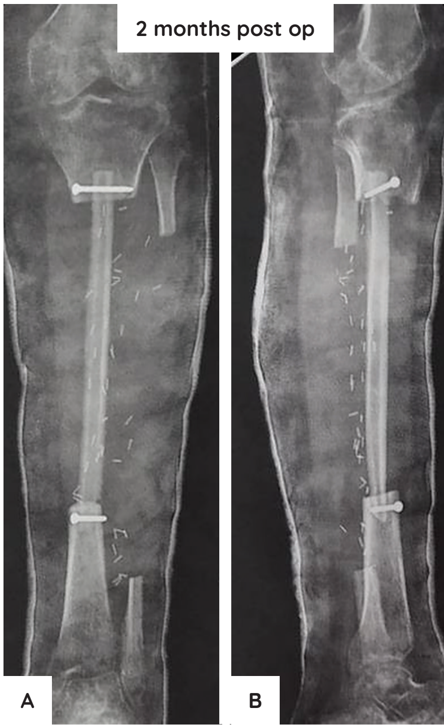

Figure 11. AP (A) and lateral (B) radiographs at two months post op showing hypertrophy of the fibular graft.

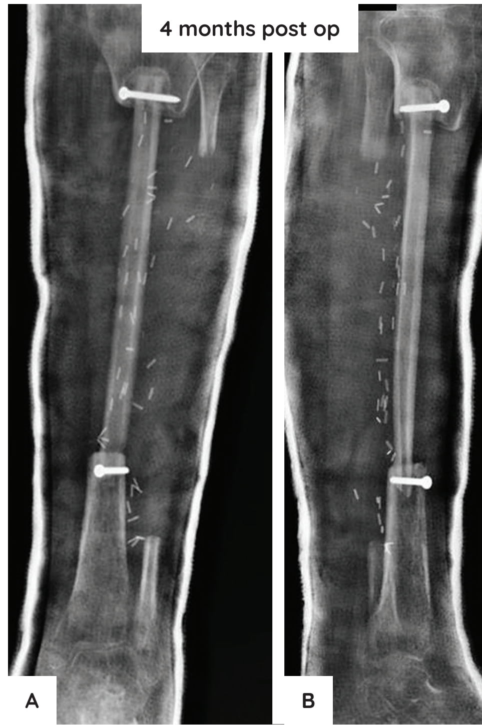

Figure 12. AP (A) and lateral (B) radiographs at four months post op showing hypertrophy of the fibular graft.

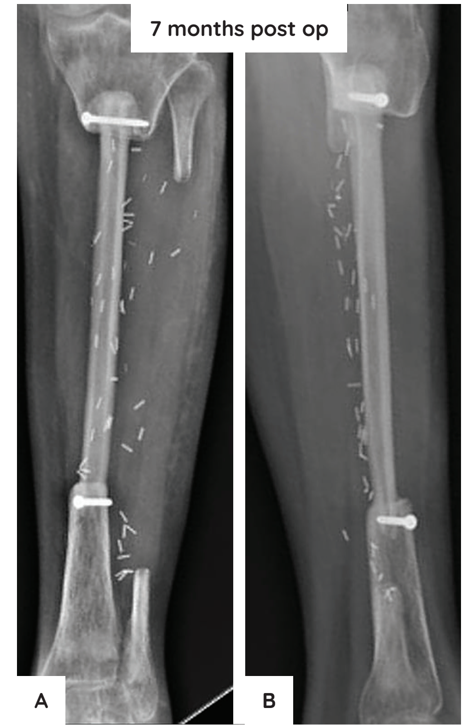

Figure 13. AP (A) and lateral (B) radiographs at seven months post op showing further hypertrophy of the fibular graft.

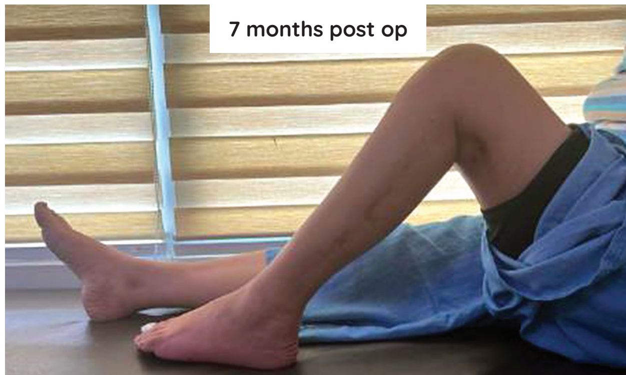

Figure 14. Follow-up at 7 months: knee flexion up to approximately 100 degrees.

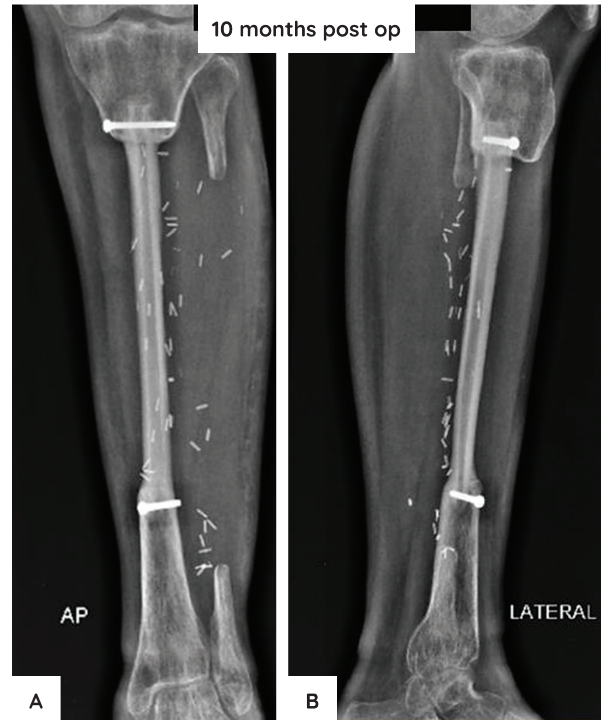

Figure 15. AP (A) and lateral (B) radiographs at 10 months showing hypertrophy on the entire length of fibular graft.

is displaced during embryonic development, moving the bone and skin closer to the anterior tibia, making this site the most common.[4] This patient's histologic findings include basaloid cells in a palisading pattern and cribriform nests.

Traditionally, adamantinoma has been treated with amputation, but different treatment options have evolved, including distraction osteogenesis, intercalary allografts, nonvascularized fibular auto-grafts, bridging custom-made prosthesis/endoprosthesis, free vascularized fibular grafts, and pedicled fibular grafts.[1,5] Intercalary allografts and non-vascularized grafts can generate new bone but are limited to defects under 6–8 cm and rely on creeping substitution for bone formation, which can weaken the graft and lead to complications. Vascularized fibula autografts support

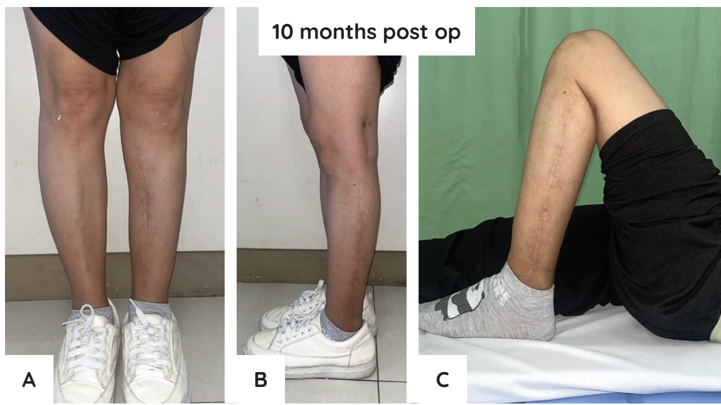

Figure 16. Able to bear weight without any assistive device **(A)** and **(B)** with full range of motion 0-120 degrees **(C)**.

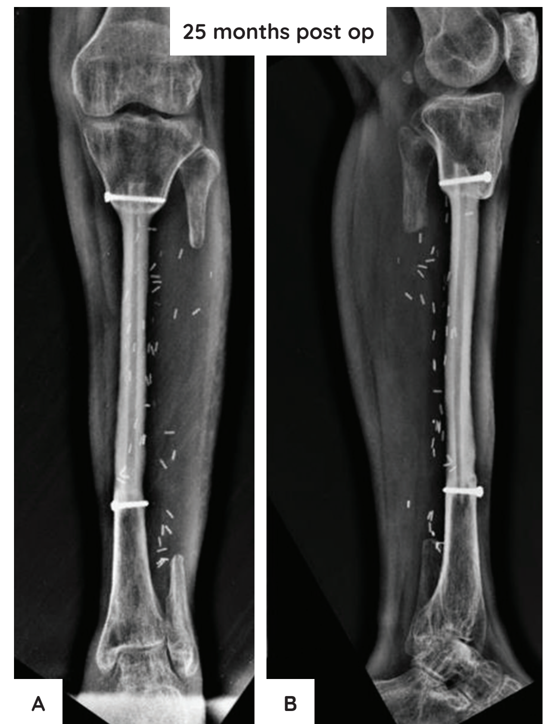

Figure 17. AP **(A)** and lateral **(B)** radiographs at 25 months showing hypertrophy on the entire length of the fibular graft.

healing via primary bone union and gain strength as they hypertrophy.[6-8] For lower limb cases, full weight-bearing was typically achieved on an average of 7.5 months to 18 months.[9]

In this case, following tibial resection, the bone defect was reconstructed using an ipsilateral pedicled vascularized fibular graft, which was then secured in the medullary cavity with proximal and distal screws. Pedicled fibular grafts retain their native blood supply, making them particularly effective for local reconstructions near the donor site. This approach minimizes the risk of ischemic complications and allows for simpler and shorter surgery without the need for microsurgical vascular reconnection. There are many options for securing the VFG to the tibia—screws, plates, intramedullary nails, or external fixators may be used. Graft hypertrophy is more robust in grafts that are mechanically loaded and not bypassed by either plate fixation or intramedullary nail.[10] For this case, pedicled fibular graft was secured using two cortical screws, one proximal and one distal, to provide rotational stability. The limb was immobilized in a long leg cast for seven months

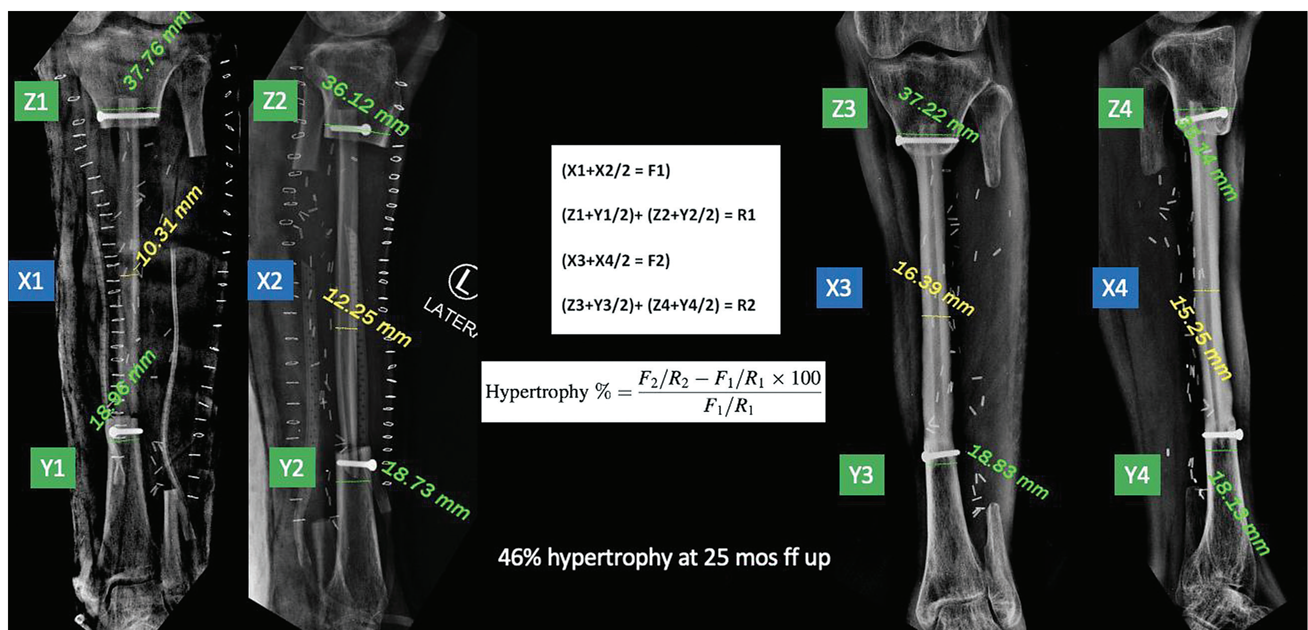

Figure 18. Graft hypertrophy was measured in relation to host bone according to El-Gammal et al. where F1 = mean fibular graft anteroposterior and lateral width at the midpoint postoperatively, R1 = mean recipient bone anteroposterior and lateral width at a fixed point away from the graft-host junction postoperatively, F2 = mean fibular graft anteroposterior and lateral width at the midpoint at follow-up, and R2 = mean recipient bone anteroposterior and lateral width at a fixed point away from the graft-host junction at 25 months follow-up.

before shifting to a pneumatic walking boot. During this time, repeat radiographs already showed an initial bone bridge between the tibia and the fibular graft which led us to start partial weight-bearing with axillary crutches. Full weight-bearing was allowed after repeat radiographs showed complete union and consolidation of the fibular graft at 19 months post-op. The graft hypertrophy index of 46% hypertrophy at 25 months (Figure 18) was calculated according to the formula used by El-Gammal et al.[9] The fibular graft is expected to have 50% hypertrophy at 18–36 months.[11] Mechanical stress is key to balancing bone formation and resorption, with increased stress promoting bone growth. However, excessive loading can cause stress fractures. Careful monitoring guided our weight-bearing progression to prevent this from happening.

At 25 months post-op, the patient was ambulating without any assistive device, had a full range of motion, was free of pain, local recurrence, and stress fractures, and had an MSTS score of 28 out of 30. Radiographs confirmed both periosteal and endosteal hypertrophy and consolidation at the graft-host interface.

CONCLUSION

We present a rare case of adamantinoma of the tibia which was successfully treated with resection and reconstruction using an ipsilateral vascularized fibular graft, secured with simple cortical screws and casting. At 25 months, the patient was able to resume her daily activities without pain or restriction of motion.

ETHICAL CONSIDERATION

Patient consent forms were obtained before manuscript submission.

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AUTHOR DISCLOSURE

The authors declared no conflict of interest.

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