

# Huge Intraosseous Tibial Haemangioma Managed with Embolisation, Excision and Fibular Ilizarov Reconstruction: A Case Report

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## ABSTRACT

**Aim:** Our aim is to report the successful treatment of an intraosseous haemangioma of tibia with an atypical presentation through a multidisciplinary approach of preoperative embolisation, a subtotal resection of the tibia and subsequent reconstruction with the Ilizarov medial fibular translation technique.

**Background:** En bloc excision is the treatment of choice for large tumours of the tibia. However, there is no single recommended method for the reconstruction of the resulting bony defect.

**Case:** A 22-year-old female presented with a massive intraosseous haemangioma of the entire tibia. Sequential, multimodal treatment consisted of (1) preembolisation, (2) en bloc resection and (3) reconstruction of the extensive skeletal defect via the Ilizarov method of fibular medialisation. Radiologic union occurred at 6 months and graft hypertrophy at 22 months. At 45 months, the patient was fully weight-bearing without need for an assistive device.

**Conclusion:** Resection and reconstruction of a large intraosseous haemangioma of the tibia can be treated successfully using a well-planned sequential management of embolisation, resection and Ilizarov fibular grafting.

**Significance:** This report highlights the successful management of an unusually extensive and difficult tumour through appropriate and meticulous perioperative multidisciplinary planning, execution and follow-up.

**Keywords:** Case report, Embolisation, Haemangioma, Ilizarov, Limb salvage, Tibia.

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## BACKGROUND

The goal of the treatment for large tumours in the tibia is complete surgical resection and bony reconstruction without resulting functional deficits. Reconstruction options include distraction osteogenesis, allografts, vascularised fibular autografts, the Capanna technique, non-vascularised autogenous bone grafts or endoprosthetic replacement.

We report the difficult but successful sequential multimodal management of a young lady with an unusually large haemangioma of the tibia treated with preoperative embolisation, complete resection of almost the entire tibia and reconstruction with Ilizarov medial fibular translation.

Informed consent was obtained from the patient prior to writing this report.

## CASE DESCRIPTION

A 22-year-old female from a distant Southern Philippines city was presented with a large, painful mass in her right leg. A biopsy 6 years earlier had revealed an intraosseous haemangioma, but the patient had refused amputation and did not seek further consultation.

The physical examination showed a large, tender, bony mass over the anteromedial portion of the proximal right leg. The patient retained full motion of the ipsilateral ankle and knee joints. There were no other masses (Fig. 1A). The radiographs showed an extensive lesion of the right tibial shaft extending from the proximal epiphyseal line to the distal third of the tibia with an associated huge anterior soft tissue mass. There was a lattice-like pattern with

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horizontally and vertically oriented striations within the entire lesion (Fig. 1B). Magnetic resonance T2-weighted images showed multiple high signal intensity lobules with multiple septations containing several round areas of low signal intensity (Figs 1C to E).

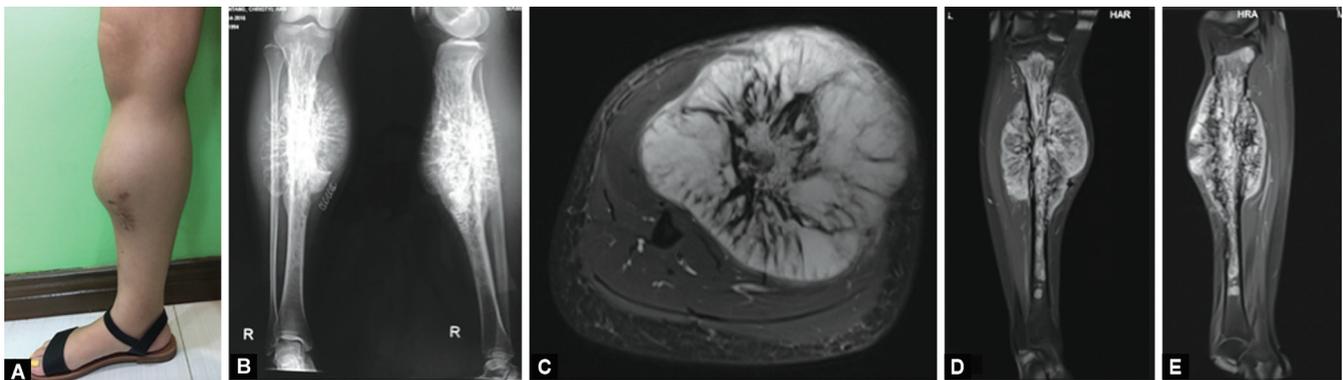
A repeat open biopsy confirmed the histopathology to be that of a haemangioma of the bone. During the biopsy, the mass was noted to be extremely vascular. The continuous intense bleeding was controlled only by packing the biopsy cavity with bone cement.

In anticipation of increased intraoperative bleeding, the patient was referred for embolisation 2 days before surgery. At surgery, the tumour mass was exposed with an incision beginning medial parapatellar proximally, extending distally to include the previous biopsy site and proceeding distally over the medial tibia just above the ankle joint. After detaching the medial gastrocnemius muscle origin and pes anserinus insertions, the popliteal vessels were identified and protected. Given the long duration of the tumour, the extensive bleeding on biopsy and that the patient resided far away from the treating establishment, thus making regular follow-up monitoring impractical, the agreement reached pre-operatively with the patient was to achieve a marginal resection as far as possible even if it meant a longer segment of bone would be resected distally in order to decrease the chance of a local recurrence. Proximally, however, based on the magnetic resonance images, barely 1 cm of normal tibia remained and resection at this level would have included the tibial tubercle. A cut was therefore made 4 cm distal to the plateau, preserving the tibial tubercle. At this level, the cortex had not been infiltrated; but since tumour had extended beyond this point within the intramedullary canal, curettage and high-speed burring of the medullary contents were undertaken, leaving only the outer cortical shell of the proximal tibial epiphysis. Distally, the tibia was cut 1 cm beyond the intramedullary extent of the tumour, leaving 3 cm of distal tibia (Fig. 2A). The entire resected specimen measured 24 cm in length and 12 cm in maximum diameter (Fig. 2B).

It was decided that this huge tibial defect would be reconstructed with the ipsilateral fibula. The remaining proximal and distal tibia were aligned and fixed with tensioned transfixation wires clamped to an Ilizarov circular frame. Added stability was achieved with olive wires. The fibula was then fixed with three olive wires with the olives on the lateral side of the fibula. On the medial side, Ilizarov components were configured to pull the fibula via olive wires anteromedially. The fibula was percutaneously osteotomised at two levels corresponding to the cuts of the remaining portions of the proximal and distal tibia (Figs 3A to C).

Gradual medial translation of the fibula at the rate of 1 mm/day was initiated after a week of latency. Bone grafting at the two docking sites of the fibula to the tibia was undertaken a month later. At 6 months, radiographs showed healing at both distal and proximal ends (Fig. 4). The fixator was then removed and a posterior splint applied to protect the leg.

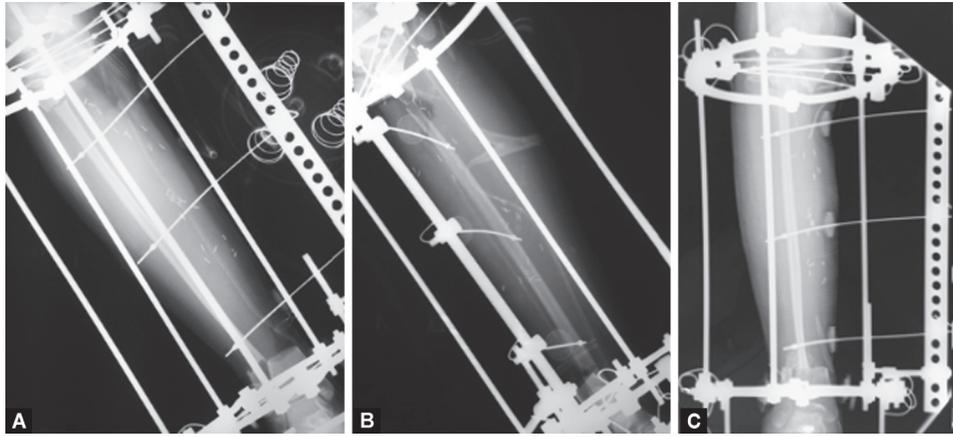
Weight-bearing with bilateral axillary crutches commenced at 16 months, and fibular hypertrophy was noted on radiographs at 22 months. Radiographs at 38 months showed union at proximal and distal graft-host sites with fibular hypertrophy (Fig. 4). At review, the patient has no pain, has full knee extension and has up to 100° of knee flexion (Fig. 5). There is a 1.5-cm leg length discrepancy, which does not affect walking. And at latest follow-up at 45 months, the patient is fully weight-bearing without any assistive device. There are no signs of local recurrence, and the patient has resumed previous activities with an MTS score of 26, scoring 5 for pain, 4 on function, 4 on emotional, 4 on supports, 4 on walking and lastly, 5 on gait. The final histopathology confirmed the diagnosis of haemangioma.



**Figs 1A to E:** (A) Gross appearance of the mass at anteromedial aspect of the leg and previous biopsy scar. (B) Anteroposterior and lateral radiographs of the tibia. (C) Axial. (D) Coronal. (E) Sagittal T2-weighted images showing high-intensity signal lobules with striations extending from proximal epiphyseal border to distal tibia with soft tissue expansion



**Figs 2A and B:** (A) Resection of tibia. (B) Resected mass measuring 24 cm in length and 12 cm in maximum diameter



**Figs 3A to C:** (A) Anteroposterior. (B) Lateral views of the leg showing Ilizarov construct and the olive wires at the fibula. (C) Follow-up radiographs show callus formation at graft–host junction



**Fig. 4:** Radiographs at 38 months after surgery showing union at proximal and distal graft–host site with fibular hypertrophy

## DISCUSSION

The intraosseous haemangioma constitutes about 1% of all bone tumours. A haemangioma presenting in a long bone, such as the tibia, is uncommon. There have been only six cases of tibial haemangioma reported in the English literature, all of which have been 4 cm or smaller in dimension.<sup>1,2,4,6–8</sup> The goal of the treatment is complete tumour removal with either curettage or resection, and subsequent reconstruction.<sup>9–11</sup>

While a marginal resection would have been the preferred option for both ends of the patient’s tumour, achieving such a margin proximally would have resulted in removal of the tibial tubercle and leaving only a wafer-thin piece of tibial plateau. The decision was therefore made to osteotomise just distal to the tubercle and remove intramedullary tumour through curettage and high-speed burring. Distally, the tibia was osteotomised at 3 cm proximal to the distal joint line, providing a margin of 1 cm from the most distal intramedullary extent of the tumour.



**Fig. 5:** Patient at 38 months postoperative, with flexion up to 100°

The extremely small remaining tibial fragments precluded the option of a traditional bone transport method of reconstruction. Bridging this 24-cm longitudinal gap by bone transport would have meant an inordinate amount of time in the external fixator. Transporting an entire intact fibula was therefore deemed a better reconstruction alternative. Gradual fibular translation allowed the fibula to retain its vascularity and thus in turn allowed the possibility of bony hypertrophy. In order to facilitate the union of the tibial ends to the fibula and, subsequently, a timely removal of the fixator, early bone grafting at the docking sites was done. Meselhy et al. described this method of gradual medial fibular transfer using the Ilizarov external fixator to reconstruct the large tibial defects. In their study, the average segmental tibial defect was 13.2 cm, and the longest was 18 cm. Fixator time was 8 months and union was achieved in 100% of patients, all 14 of whom were eventually able to fully weight-bear on the affected extremity without external support.<sup>5</sup>

In our patient, union of the transposed fibula proximally and distally to the host tibia was observed at 6 months, after which the Ilizarov fixator was removed and the leg protected with a splint. The patient was allowed to weight-bear with crutches at 16 months. Fibular hypertrophy was visualised at 22 months, consistent with the observation of hypertrophy occurring once a limb is mechanically loaded.<sup>3,5</sup> At 45 months, the patient was fully weight-bearing on the affected leg without any assistive device, with knee flexion from 0 to 100°, and with an MTS score of 26 out of 30.

## CONCLUSION

We present a case of a massive haemangioma of the tibia successfully treated via multimodal treatment, including pre-operative embolisation, meticulous tumour resection and Ilizarov fibular translation. At 45 months, the patient remains free of disease and has resumed routine activities without pain or restriction of motion.

## CLINICAL SIGNIFICANCE

This report highlights the difficult but successful management of an unusually extensive tumour through meticulous multidisciplinary perioperative planning, execution and follow-up.

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