

Reconstruction of calcaneum using the deep circumflex iliac artery bone graft following wide local excision due to Ewing's sarcoma: A case report on a rare disease presentation and its unconventional management

Zoha Sajid Qureshi, Mamoon Rashid, Sakina Malik, Sohail Hafeez

ABSTRACT

Introduction: Ewing's sarcoma of the calcaneum, although rare, is a significant health threat as it is quick to spread locally and systemically reducing the overall survival rates. Conventionally, below knee amputation was used as its only management. Currently, due to numerous advancements, wide local excision following reconstruction is the treatment of choice worldwide.

Case Report: This case report provides clinical and functional outcomes of using deep circumflex iliac artery bone graft for calcaneal reconstruction. The follow-up duration in our study was two years. Normal ambulatory status, post-procedure, was achieved within nine months with a good range of motion at ankle joint both actively and passively. No donor site morbidity was seen and radiological imaging confirmed a healthy and viable graft with negative oncological margins.

Conclusion: Our choice of graft proved to be a suitable option for calcaneal reconstructions as the curvature of iliac crest resembles calcaneal tuberosity. The ability of

its osseous flap to naturally fit the area of defect makes it the most appropriate choice for the reconstruction of the heel of the foot.

Keywords: Calcaneal tumors, Deep circumflex iliac artery bone graft, Ewing's sarcoma, Limb salvage

How to cite this article

Qureshi ZS, Rashid M, Malik S, Hafeez S. Reconstruction of calcaneum using the deep circumflex iliac artery bone graft following wide local excision due to Ewing's sarcoma: A case report on a rare disease presentation and its unconventional management. *Int J Case Rep Images* 2023;14(2):85–90.

Article ID: 101416Z01ZQ2023

doi: 10.5348/101416Z01ZQ2023CR

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Received: 24 May 2023

Accepted: 26 August 2023

Published: 12 October 2023

INTRODUCTION

Ewing's sarcoma is an osteolytic, malignant bone tumor that commonly occurs in the second decade of life and usually involves the diaphysis of long bones. It is rarely seen in the small bones of the hand and feet but the incidence of calcaneal Ewing sarcoma has been reported to be less than 1% [1]. The proximity of various structures in the foot and the aggressive nature of the tumor often result in an extraosseous presentation [2]. Because of the presence of only a limited literature and resources, the management of calcaneal tumors remains a challenge in surgical oncology [3]. As a result, below knee amputation was commonly practiced for its management [4].

However, the recent advances such as diagnostic oncological modalities and reconstructive procedures, i.e., microvascular plastic surgeries, have made limb salvage an optimal management approach, as it conserves the functionality and durability of the compromised limb [4–6]. To date no consensus has been established among surgeons worldwide regarding the best reconstructive technique to be used. This case report elaborates upon the use of deep circumflex artery bone flap as an effective and novel technique to reconstruct the calcaneum following wide local excision of the Ewing sarcoma. Moreover, it highlights clinical and functional outcomes at 24th month follow-up of the patient.

CASE REPORT

A 22-year-old female, who was a known case of Ewing's sarcoma of right calcaneum, presented with moderate pain and progressive swelling over the right hindfoot for one year. Physical examination revealed a firm, immobile, and tender mass at posterior aspect of the right ankle with intact distal neurovascular status. During walking, right-sided limp was observed with compromised weight-bearing of the heel of the foot. No regional lymph nodes were palpable. TRU-CUT biopsy of the mass confirmed Ewing's sarcoma of the calcaneum. The magnetic resonance imaging (MRI) of the right foot with contrast revealed a large, heterogeneous, avidly enhancing lesion of the right calcaneum, extending slightly superiorly into the subtalar joint causing erosion of sub articular cartilage with a positive periosteal reaction in the area of the lesion (Figure 1). Following discussion in our sarcoma multi-disciplinary tumor board, the patient underwent six cycles of neo-adjuvant chemotherapy (poor tumor response of chemotherapy), and then wide local excision of the calcaneum and reconstruction with a deep circumflex iliac artery free flap was planned. The procedure was explained and a written consent was obtained from the patient.

After completing pre-operative protocols, the procedure was performed in supine position under pneumatic tourniquet control. A posterolateral incision was given and the calcaneum was circumferentially dissected based on "Enneking staging system" (Figure 2). Eleven frozen sections were taken from the soft tissue margins and were found to be clear. For flap elevation, an oblique skin incision was made starting from the right pubic tubercle and extending toward the anterior superior iliac spine and iliac crest. The external oblique fibers were separated and the round ligament was retracted medially. A 6×6 cm of iliac crest along with internal oblique muscle was harvested with right deep circumflex iliac artery and vein (Figures 3 and 4). Hemostasis was secured and the wound closed in layers. Posterior tibial nerve was preserved and skin was closed. The graft along

with talonavicular and calcaneocuboid joints was then fixed with 5 K-wires from plantar aspect into the talus. The end-to-end anastomosis between deep circumflex iliac vessels and posterior tibial vessels was done. Flap circulation was ensured and the wound was closed in layers. No noteworthy perioperative complications were observed.

The ankle immobilization period was two months after which the patient started partial weight bearing. Additionally, passive movements of the right foot and ankle began without any significant difficulty and the patient gradually attained full weight bearing six months post-operatively. Complete planter flexion and dorsiflexion were restored in eight months and the patient returned to regular household work within 10 months of procedure. No special shoes were needed and the formed contour easily fit the everyday footwear.

On the two-year follow-up, the ankle and hind foot were well aligned with satisfactory ankle joint stability (Figure 5). Improved inversion was observed with up to 20 degrees of motion, whereas, all other foot movements were well within normal ranges both actively and passively. Besides a mild limp on the terminal stance, no other gait cycle alterations were observed. "Musculoskeletal Tumor Society Score" and "American Orthopedic Foot and Ankle Society Score" in two year follow-up were calculated to be 28% and 96%. Follow-up MRI confirmed adequately positioned viable bone flap with good arthrodesis. No signs of recurrence of malignancy were noted. The patient reported satisfaction with clinical results and good emotional acceptance of the outcome.

For this study, institutional review board and ethics committee (IRB) approval was obtained from relevant institution.

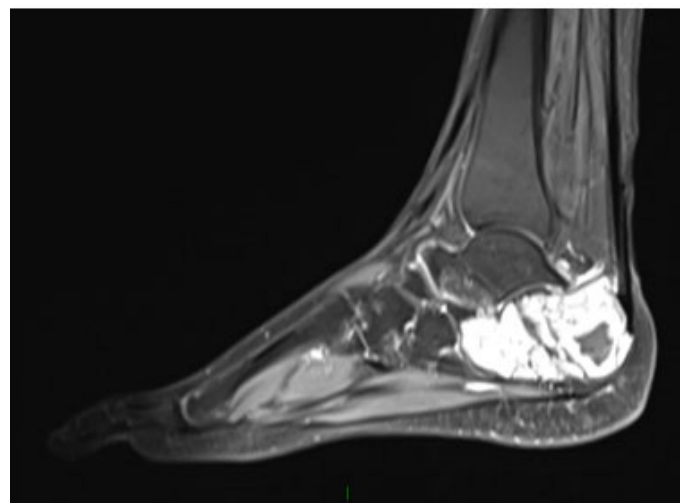


Figure 1: The magnetic resonance imaging (MRI) of the right foot with contrast revealed a large, heterogeneous, avidly enhancing lesion of the right calcaneum.



Figure 2: Intra-operative image from the site of calcaneal removal, defect in the right ankle after tumor extirpation.



Figure 5: Follow-up investigation (X-ray of right foot lateral view) after two years in our patient showing calcaneus replaced with iliac crest graft support with wires. A viable graft, that is firmly placed, aligns with natural foot contours.

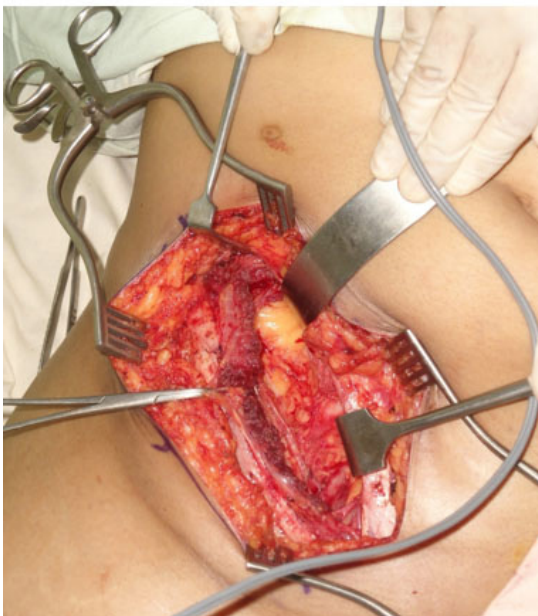


Figure 3: Extracted calcaneum using wide resection margins.



Figure 6: Preoperative radiograph of our patient showing lytic lesion of calcaneum (right foot lateral view) with erosion of overlying cortex suggestive of a malignant condition.



Figure 4: Intra-operative image from the site bone flap harvest, showing deep circumflex iliac artery (DCIA) flap harvest with exposed iliac crest.

DISCUSSION

The foot, with its distinctive anatomical formation and dynamic arches, is functionally unique as it is an essential component of weight bearing [7]. The calcaneus is the largest tarsal bone present in the posterior foot compartment and is an important component of the lateral arch of the foot. Not only does it sustain high tensile forces but it also maintains the hind foot height, gait cycle's stance phase, and stability on standing [8].

Ewing's sarcoma of the calcaneum, although rare, causes lytic, condensed lesions with permeating margins, cortical destruction, and aggressive periosteal reaction in the respective small bone that damages its structural and functional integrity [9] (Figure 6). Due to its highly aggressive nature, Ewing sarcoma, regardless of its location, is quick to advance in stage and grade, hence adequate local control is crucial. Distance metastasis is the single most important unfavorable prognostic factor (<30% survival rate) that necessitates early local control and systemic intervention [10].

As the calcaneum only has the planter surface that is close to the major neurovascular bundle, wide resection margins could be achieved in most patients [5]. Hence, in the recent years, limb salvage procedures have become the mainstay of treatment for calcaneal malignancies as compared to limb amputation. Since the traumatic loss of foot warrants severe physiological and psychological impacts such as phantom pain, anxiety, depression, and sometimes death, better prognosis is seen with limb salvage surgeries [11].

The choice of reconstructive technique to be used depends on multiple factors; however, it is crucial that appropriate talar and calcaneal inclinations are achieved along with adequate hind foot height to reduce maximum pressure on the ankle to achieve stable weight bearing [12]. Attempts of mechanical reconstruction have been made using prosthetics but some studies have reported the partial collapse of the central part of the calcaneum [5]. The popularity of vascularized free tissue transfer has recently increased as it provides sufficient tissue volume to cover complex heal defects and the osteoprogenitor cells in the graft potentiates wound healing by resisting bone resorption [13]. There is no consensus on the most appropriate bone flap to be used for reconstruction. The vascularized rib flap is known to have an insufficient bone volume; moreover, it does not provide stable anchoring point for Achilles tendon fixation, rendering 3D reconstruction of calcaneum inadequate [5, 12]. Some literature also suggests the use of vascularized scapular graft, but they are highly associated with complications at the donor site following flap elevation [14]. Most studies are in the favor of vascularized fibular graft, which can be used to cover a large defect up to 20 cm, but it is difficult to contour the straight bone flap to fit the curved defect [15].

The vascularized iliac crest free flap takes precedence over other choices as it not only provides the sufficient bony component (up to 4×6 cm in height and width), but its natural curvature also resembles calcaneal tuberosity, and thus the flap can be tailored to fit the given defect [16, 17]. The thickness and length of iliac crest flap helps to maintain natural height of the hind foot and the crest can be used as a weight bearing surface to provide sturdiness to the heel during locomotion. Aesthetically, the resulting contour also imitates the natural ankle curvature, which allows easy everyday shoe usage. The broad surface area

of the bone, serving as a durable platform for multiple muscles and tendons, preserves the function of planter flexion and extension of the ankle which is crucial for maintaining gait cycle [13]. Vascularized grafts are not only less prone to resorption but they also promote early arthrodesis, and keep the graft viable [14, 18]. The flap elevation from iliac crest causes the donor site to remain relatively concealed facing less morbidity resulting in a higher emotional acceptance score on Musculoskeletal Tumor Society (MSTS) evaluation.

This case report aims to contribute in the existing literature by providing unbiased evidence of usefulness of iliac crest bone graft for foot and ankle malignancies. Although short follow-up duration is a limitation in our report, we believe the appealing short-term results may warrant satisfactory long-term prognosis.

CONCLUSION

Calcaneal malignancy, i.e., Ewing's sarcoma occurs infrequently, but their best prognosis is observed with reconstructive approaches. Deep circumflex iliac artery bone graft is an optimal choice for calcaneal reconstruction as shown by satisfactory clinical and functional outcomes. Additionally, befitting flap shape and size along with comparatively less donor site morbidity, makes deep circumflex iliac artery osseous flap a well suited option compared to the rest. It is evident that when used as a combined modality, reconstructive procedures can help to improve patient care standards.

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Author Contributions

Zoha Sajid Qureshi – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Mamoon Rashid – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Sakina Malik – Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Sohail Hafeez – Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Guarantor of Submission

The corresponding author is the guarantor of submission.

Source of Support

None.

Consent Statement

Written informed consent was obtained from the patient for publication of this article.

Conflict of Interest

Authors declare no conflict of interest.

Data Availability

All relevant data are within the paper and its Supporting Information files.

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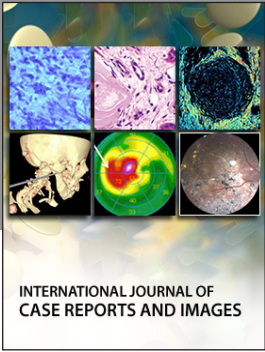
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