

Case Report



Limb Salvage Surgery with Customized Titanium Prosthesis for Management of Calcaneus Osteosarcoma

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Abstract

Introduction: Malignant bone tumors in adolescents and young adults are relatively rare, with osteosarcomas, chondrosarcomas, and Ewing's sarcomas being the most representative. However, primary malignant neoplasms in the foot are even rarer, with calcaneal osteosarcoma representing less than 1% of the total osteosarcoma locations. Various salvage surgery strategies have been reported, including the use of allografts and vascularized flaps for calcaneal reconstruction, as well as partial or total calcaneal replacement, all with good functional outcomes. **Case presentation:** An 18-year-old young adult, with no pathological history, he was diagnosed with an osteoblastic osteosarcoma of the calcaneus. As part of the limb salvage surgery, a total calcaneus replacement was proposed using a custom-made porous titanium prosthesis printed in 3D. **Conclusion:** The replacement of the calcaneus following resection for osteosarcoma remains an area of innovation in oncological surgery. The few reported cases highlight the viability of custom prostheses as a promising alternative to preserve function and improve the quality of life for patients.

Keywords: *Osteosarcoma, calcaneus, salvage surgery, custom-made prosthesis.*

Introduction

Malignant bone tumors represent approximately 3% of tumors in adolescents and young adults, with osteosarcoma, chondrosarcomas, and the Ewing sarcoma family of tumors being the most common and representative ^[1]. Osteosarcoma is a mesenchymal tumor characterized by unique tumor cells that produce immature osteoid ^[2]. It can occur in any bone but has a predilection for the metaphysis of long bones, most commonly the distal femur, proximal tibia, and proximal humerus ^[1,2].

Conversely, the distribution of primary malignant neoplasms in the foot and ankle is even rarer, with osteosarcoma localized in the calcaneus representing less than 1% of all osteosarcoma locations ^[3]. It is often underdiagnosed because pain or inflammation is attributed to trauma or secondary to a sports injury. The clinical presentation is very non-specific but is characterized by the presence of pain at the tumor site, which is initially painless but as the tumor grows and alters bone architecture, it will cause pain. Additionally, the pain can be exacerbated by physical activity or at rest, especially at night ^[1,2,4]. Other associated symptoms include a sensation of mass, edema, weight loss, and fever, with the latter two being characteristic of advanced disease ^[1,2].

Diagnosis begins with the clinical presentation, followed by imaging studies that can provide information about the lesion's size, tissue characteristics, and relationship with other anatomical structures. This is the case with CT scans, which better characterize

bone lesions, and MRI, which is useful for evaluating soft tissue characteristics and bone lesions.

Diagnostic confirmation is performed through a biopsy and histopathology ^[1,5,6], followed by extension studies to evaluate for metastatic disease and thus direct pharmacological and surgical treatment.

Over time and with the development of new technologies, in addition to chemotherapy, limb salvage surgery has become the surgical procedure of choice for osteosarcomas in the last two decades ^[7]. However, foot tumors, due to their behavior and different compartmentalization, often cause early involvement of adjacent bones and soft tissues. This is why salvage surgery is difficult to achieve and requires detailed preoperative evaluation ^[7].

Various treatment techniques have been reported, from total calcanectomy and reconstruction with allografts, partial and total prostheses, vascularized iliac flaps, and pedicled osteocutaneous fibular flaps, all with good functional results ^[9-15].

In this article, we report the presence of an osteoblastic osteosarcoma of the calcaneus in an 18-year-old young adult who was managed with complete tumor and calcaneal resection, replaced by a custom-made 3D printed calcaneal prosthesis.

Case Presentation

We present the case of an 18-year-old young adult patient, with no pathological history, who consulted for pain and edema in the right

heel that increased with sports activities. Initially, a foot X-ray was requested, which showed an irregular, osteolytic image with cortical expansion, located on the calcaneal body and affecting more than 60% of it (**Fig 1**). Further studies included a contrast-enhanced MRI to better evaluate the lesion's characteristics. Additionally, CT scans of the abdomen, pelvis, and thorax were performed to rule out metastatic lesions.

A partial tumor resection was performed and replaced with methyl methacrylate (**Fig 2**). In addition, pathology samples were taken, which confirmed the diagnosis of a malignant tumor lesion of fusiform and oval mesenchymal cells with nuclear hyperchromatism, prominent nucleoli, and marked nuclear pleomorphism with frequent atypical mitoses (up to 10 in 10 high-power fields), abundant osteoid material, without the presence of necrosis, and SATB2 positive immunohistochemistry in neoplastic cells, with CD99 and smooth muscle actin negative. Subsequently, after receiving chemotherapy, the patient underwent total calcanectomy through a posterior calcaneal approach, following prior planning and preparation of a custom-made 3D printed calcaneal prosthesis (**Fig 3**).

The patient was on the operating table, under spinal anesthesia, positioned in the lateral decubitus position, with a pneumatic tourniquet applied. After asepsis and antisepsis, a U-shaped incision was made around the heel, creating the thickest possible flap. Dissection was performed layer by layer, preserving neurovascular structures and main ligaments. Kirschner wires were positioned at the subtalar and calcaneocuboid joint levels as references for subsequent release of intertarsal ligaments, thus achieving complete en bloc resection of the calcaneus. Adjacent structures were verified without the presence of secondary lesions. Subsequently, a trial prosthesis was positioned, showing adequate adjustment to the adjacent joints, proceeding to place the definitive titanium prosthesis. It was stabilized to the talus with two 5.0 mm screws and to the cuboid with two 3.5 mm screws. Once stabilized, the Achilles tendon was inserted, hemostasis was controlled, the previously designed flap was closed, the wound was covered with a negative pressure system, and it was immobilized with a sural splint. A post-surgical X-ray was taken, showing adequate prosthesis position with appropriate fixation (**Fig 4**).



Figure 1: Radiograph showing osteolytic lesion and cortical expansion



Figure 2: A-D Images of the tumor after partial resection and application of methylmethacrylate. Lateral radiograph (Fig. 2-A) and Harris projection (Fig. 2-B). Axial (Fig. 2-C) and sagittal (Fig. 2-D) MRI.

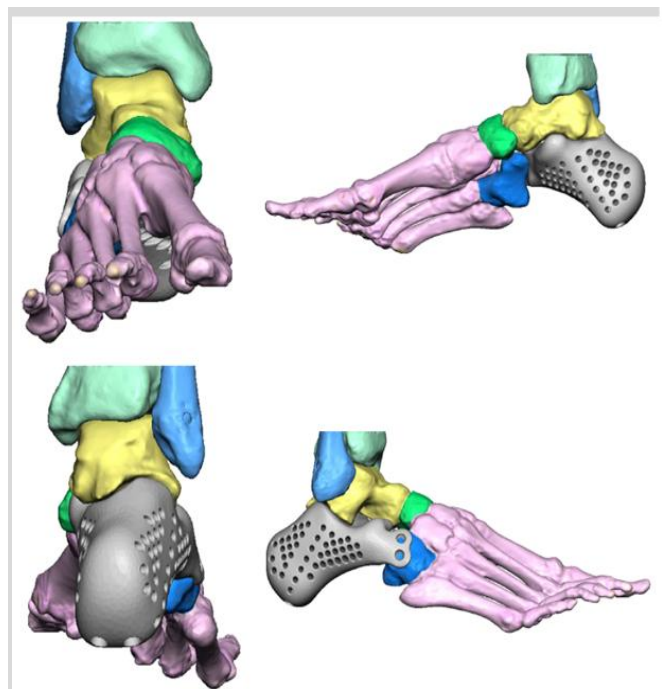


Figure 3. Planning of a custom-made calcaneal prosthesis



Figure 4: A-C. Postoperative radiographs. Anteroposterior projection of the foot (Fig. 4-A), anteroposterior projection of the ankle (Fig. 4-B), and lateral projection of the ankle (Fig. 4-C).

Discussion

Osteosarcomas are mesenchymal bone tumors with various histological types, including osteoblastic, fibroblastic, chondroblastic, and telangiectatic [8]. This malignant pathology is relatively rare, primarily affecting adolescents and young adults in the metaphysis of long bones like the femur and tibia. The localization of osteosarcomas in the foot is even rarer, accounting for less than 1% of cases, with the calcaneus and metatarsals being the most common sites of origin [3,8].

Salvage surgery for calcaneal osteosarcomas has been an utilized alternative in recent decades. Its objective is wide resection of the lesion to achieve clear margins, provided there's no neurovascular bundle compromise in the tarsal tunnel, with the aim of maintaining normal foot function. Furthermore, various reconstruction strategies are needed for the calcaneus, as it's a fundamental structure for the different phases of gait.

Diverse reconstructive techniques have been reported. Lingala [11] published a technique on total calcanectomy as salvage surgery with return to ambulation at 3 months. Muscolo [9] proposed a total calcanectomy and allograft replacement technique, showing a 32-year follow-up with adequate results in range of motion and pain. Arienzo [10] reported a hemi-calcanectomy with hemi-calcaneal reconstruction using a custom-made prosthesis, showing excellent results in terms of functionality. Woon Park [12] and Chou [13,14] reported on total calcanectomy replaced with a custom-made prosthesis, showing excellent results in terms of pain reduction,

adequate ranges of motion, and ambulation at 3 and 6 months, respectively.

In the presented case, the important role of 3D printing technologies in oncologic orthopedic surgery can be highlighted. The prosthesis design was assisted by software and preoperative biomodels with important characteristics, such as the selection of porous titanium as the biomaterial due to its high resistance, biocompatibility, and osseointegration. Similarly, the structural design was crucial as it allowed for fixation to the talus and cuboid to increase in situ stability and decrease long-term complications like loosening, wear, periprosthetic fractures, and chronic pain. Another feature was the mesh network on the posterior part of the prosthesis, which facilitated the suturing of ligaments and soft tissues, thus promoting the prosthesis's functionality and stability. The results and prosthesis selection in our case were very similar to those obtained by Woon Park [12], however, the prosthesis design and its characteristics differ from the results obtained by Chou [13,14] and Arienzo [10]. Nevertheless, there are few case reports and patient outcomes with calcaneal osteosarcoma for whom calcaneal replacement was chosen as a limb salvage treatment option. For this reason, this case represents a valid treatment option as part of surgical management and as a salvage measure for calcaneal malignancy.

Conclusion

Calcaneal replacement following osteosarcoma resection remains an area of innovation in oncologic surgery. The few reported cases highlight the feasibility of customized prostheses as a promising alternative for preserving function and improving patients' quality of life. However, the need for new prospective studies with a larger number of patients and long-term follow-up studies to evaluate their impact on quality of life, functional outcomes, and implant survival is emphasized. Meanwhile, these cases represent the potential of new technologies to transform the management of complex tumor locations.

In the present case, the use of a custom-made calcaneal prosthesis, elaborated with porous titanium, allowed for precise reconstruction and stable fixation, which translated into favorable clinical outcomes regarding pain and functionality.

Declarations

Informed Consent

The patient signed an informed consent authorizing the use of their clinical data for the elaboration and publication of this case report.

Conflicts of Interest

None declared by the authors.

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