

Case Report

Biological Reconstruction of the Calcaneus after Sarcoma Resection: Is It Really Necessary? Case Report and Review of the Literature

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Abstract

Osteosarcoma and Ewing's sarcoma are the most common primary, non-haemopoietic malignant bone tumors in children and adolescents. However, they rarely occur in the calcaneus with only few cases reported in the literature. Below knee amputation is considered to be the standard treatment leading to good functional results. However, in some rare instances a calcanectomy alone is possible and reconstructive options have to be considered.

Herein, we report a case of an 8-year-old boy with calcaneal Ewing's sarcoma, who presented with heel pain followed by swelling for more than one month. Following neo-adjuvant chemotherapy and preoperative radiation therapy a wide resection was performed. Reconstruction of the calcaneus was done using an allograft which got resorbed within two years of follow-up. The patient had no evidence of disease at a follow-up of 101 months.

Reviewing the literature for similar cases or different reconstructive methods showed different treatment modalities for calcaneal malignancies besides below knee amputations such as custom made prosthesis, autograft- or allograft reconstruction or total calcanectomy.

INTRODUCTION

One to three percent of osseous tumors occur in the foot and ankle according to the literature, and calcaneus is the first or second most common site of involvement before or after involvement of the metatarsals [1-5].

Although intraosseous lipomas or bone cysts are most frequent, malignancies like osteosarcoma, Ewing's sarcoma or chondrosarcoma might appear at this anatomical site, additionally. Therefore, errors in diagnosis and treatment can make limb salvage impossible and a biopsy is always needed for final diagnosis [1,3,6]. It has also been shown that bone sarcomas of the calcaneus present a different biological behavior in contrast to other locations [3,7].

Nevertheless, primary malignant tumors of the foot, especially Ewing's sarcomas, should be treated with neo-adjuvant and adjuvant chemotherapy, radiotherapy, and wide resection [3,5,8].

After major resections reconstruction is often difficult, because the calcaneus is exposed to high load [9]. Therefore, some authors favor amputation which has been reported to lead to good functional results [4-7,10,11]. However, in some rare instances a calcanectomy alone is possible and reconstructive options have to be considered.

Herein, we present the case of an 8-year-old boy with calcaneal Ewing's sarcoma managed with neo-adjuvant and adjuvant chemotherapy, preoperative radiation therapy and wide resection as well as allograft reconstruction.

CASE PRESENTATION

In February 2006, an 8-year-old boy was admitted to our department due to pain and swelling in the area of his right ankle without any kind of trauma. The boy hobbled, the right ankle was swollen and over-warmed. There was also a leucocytosis in the laboratory check.

Diagnostic imaging (plain radiographs, MRI and CT) showed blastic/cloudy and osteolytic areas in the calcaneus with destruction of the cortex (Figure 1A-D). Therefore, as differential diagnosis osteosarcoma, Ewing's sarcoma or chronic osteomyelitis were assumed.

An open biopsy was performed showing a small blue-cell tumor in the histology. Further, the genetical analysis resulted in chromosomal aberrations in the EWSR-1 gene. Consequently, the diagnosis Ewing's sarcoma was given. All other staging investigations (bone scan and chest CT) were negative for metastases.

The boy was treated with neoadjuvant chemotherapy according to the EURO E.W.I.N.G 99 protocol. For further treatment, a local radiotherapy with totally 44,8Gy was applied.

All imaging procedures following chemotherapy and radiation therapy showed regression in tumor size and an open re-biopsy in June 2006 revealed a regression grade I according to the classification system of Salzer-Kuntschik [12,13].

Due to the good response, a wide resection of the calcaneus and the surrounding soft tissues was performed. The reconstruction

of the heel was done using an allograft (Figure 2). Furthermore, reconstruction of the peroneal tendons and the Achilles tendon was done by plastic surgeons.

The patient was doing well postoperatively and the chemotherapy protocol was finished. Plain radiographs of the allograft showed tendencies of osteointegration in the talus as well as a stable osteosynthesis 3 months postoperatively (Figure 3A).

Eight months later, the x-rays started to show beginning resorption of the allograft and the screws used for allograft fixation started to irritate the skin of the heel (Figure 3B-D). Furthermore, there was a progressive malposition of the foot in supination. Sixteen months following index surgery a revision was performed to shorten the screws as well as to do a split tendon transfer of the anterior tibial muscle for correction of the supine malposition. Eighteen months later, the plain radiographs and the CT scan showed a progression in resorption of the allograft. Nevertheless, the patient was walking on the screws without pain and good function in the ankle. Further follow-ups showed a fracture of one screw without any significance (Figure 3E). In September 2012 the screws were removed and the patient was able to ambulate without any complications (Figure 3F). The patient had no evidence of disease at a follow-up of 101 months.

DISCUSSION

Life expectancy of patients with malignant bone tumors has improved within the last decades due to the framework of standard therapy optimization. Whilst investigations proved

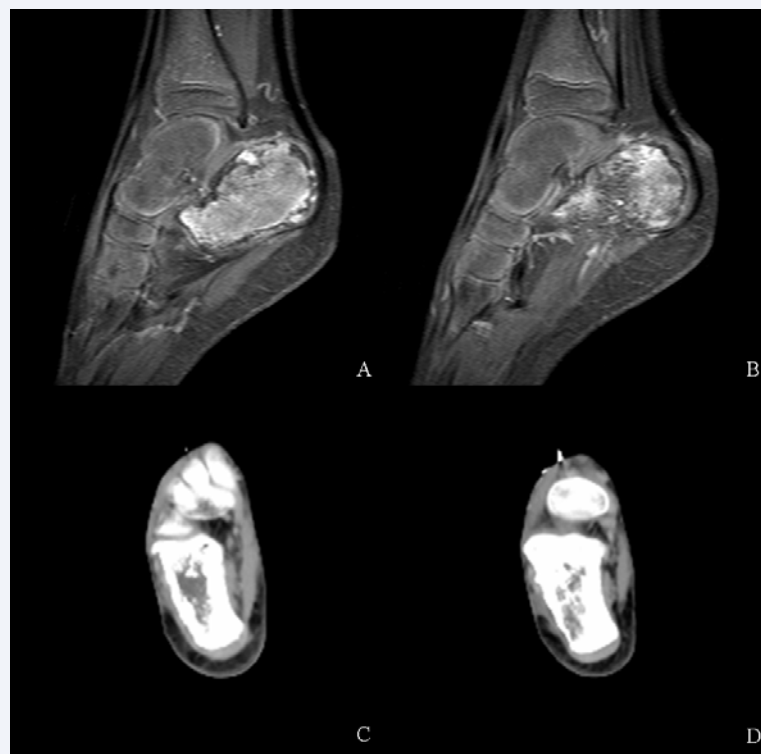


Figure 1 (A&B) - MRI of the right calcaneus of an 8-year-old boy showing an infiltration with intensive contrast enhancement and erosion as well as destruction of the cortex. (C&D) - CT scan of the affected bone showing a partial sclerotic and partial lytic expansion in the right calcaneus.



Figure 2 Intraoperative photograph of the reconstructed calcaneus which was done with a homologous femoral head.

that limb salvage is possible in up to 90% of all musculo-skeletal malignancies, the question which reconstruction modality after resection allows best outcome is still elusive although limb salvage procedures are associated with a higher rate of revision surgeries than amputations [2,4,5].

The functional outcome after limb-sparing procedures or amputation is discussed controversially. Rougraff et al. [14] and Renard et al. [9] reported that patients after limb-salvage procedures have a better functional outcome than patients after amputation.

Limb-salvage of primary malignant calcaneal tumors is performed for stage I (low-grade) and stage IIA (high-grade, intracompartmental) lesions according to Enneking [15]. In some cases it is possible to perform a limb-sparing surgery for IIB lesions, too. In these cases a regression grade I after

chemotherapy according to Salzer-Kuntschik [12,13] is seen as a precondition.

For primary and secondary malignant tumors of the calcaneus several reconstruction techniques and treatment modalities have been reported in the literature. Some authors tend to prefer total calcaneotomy or below knee amputation, while others prefer surgical reconstruction [2,4,5,16-25].

Partial or total calcaneotomy without any reconstruction is a relatively rare orthopedic procedure although it is an alternative procedure to transtibial amputation [4,5,20,24,25]. Resection of the major portion of the calcaneus does not prevent the patient from weight bearing on the foot, while subtotal calcaneotomy is superior to total calcaneotomy, because it provides more stability concerning talo-navicular subluxation [20].

Anacak et al. [19] reported intraoperative extracorporeal irradiation for primary malignant calcaneal tumors and re-implantation of the irradiated bone. Advantages of this method are the elimination of fitting and rejection problems. Furthermore, it is much cheaper than allogenic bone grafts or custom-made prostheses. One of the drawbacks of this procedure is the lack of material for histological analysis to define chemotherapeutical response.

The most common treatment modality for malignant calcaneal tumors is total calcaneotomy and reconstruction with a homologous allograft. Especially young patients seem to benefit from such a procedure as shown in the current case despite the slow on-going resorption of the allograft [17,23].

Musculo et al. [16] also reported two cases of total calcaneotomy and replacement with a massive allograft with a follow-up of 32 and 9 years. In both cases the used allograft was still working at time of latest follow-up but also showed signs of resorption which might be due to the weight bearing function

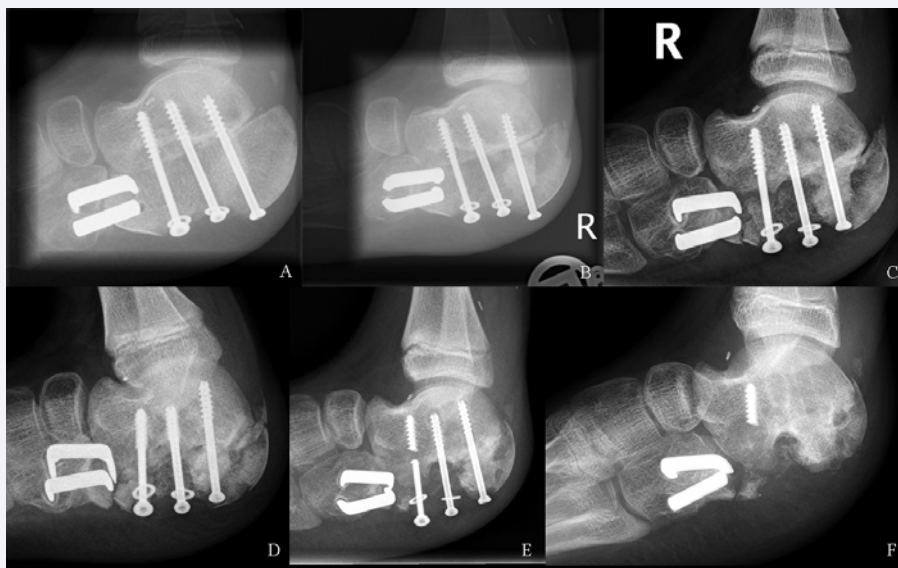


Figure 3 (A) - Plain radiographs of the right calcaneus 3 months following reconstruction. (B-D) - X-rays showing the progression of allograft resorption at 10, 24 and 30 months of follow-up, respectively. (E) - Sixty months postoperatively, an implant failure of a screw used for allograft fixation got apparent. (F) - Plain radiograph after removal of all screws 71 months following calcaneal reconstruction.

of the bone. Nevertheless, this procedure always showed good functional outcome in spite of higher complication rates (infection, resorption and local recurrence) [16,17,21,22].

The data about biological reconstructions of the calcaneus using autografts are also controversial [2,26,27]. Li et al. [2] showed satisfactory local tumor control and functional restoration of the lower limb following biological reconstruction using distally pedicledosteocutaneous fibular in five cases, while Brenner et al. [26] related that the vascularized rib is not a good choice for reconstruction following total calcanectomy because of high morbidity rate at the donor site and insufficient bony reconstruction [2].

In 2007 Chou et al. [18] reported a case of calcaneal reconstruction with a custom made prosthesis following resection of an osteosarcoma in a 31-year-old female patient. There was a good functional outcome at a follow-up of 12 years. On the other hand, there are no further reports about calcaneus prostheses, therefore it is not possible to make a statement if this method is reliable or not.

As presented in our case and according to the literature total calcanectomy might be indicated in some cases of calcaneal malignancies. Reports on the different reconstruction methods are sparse but in most cases satisfactory functional results have been described. Therefore the authors think that if oncologically safe, total calcanectomy and biological reconstruction seem to be another possible and reliable alternative to below knee amputation.

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