CASE REPORTS

ENCASEMENT OF POPLITEAL VASCULAR BUNDLE DUE LOCAL OSTEOSARCOMA RECURRENCE TREATED WITH WIDE RESECTION AND VASCULAR RECONSTRUCTION

João Vale[1*], Sara Diniz[1], Carlos Pereira[2], Vânia Oliveira[1,3], Pedro Cardoso[1,3]

[1] Department of Orthopaedics, Centro Hospitalar Universitário de Santo António, Porto, Portugal
[2] Department of Angiology and Vascular Surgery, Centro Hospitalar Universitário de Santo António, Porto, Portugal
[2] Instituto de Ciências Biomédicas de Abel Salazar, Universidade do Porto, Porto, Portugal

* Corresponding author: jsoaresvale@gmail.com

Abstract

Introduction: *Osteosarcoma is the most common primary bone malignancy. The prognosis of patients after local recurrence is generally poor and management of the locally recurrent disease is not well defined, especially in patients who have undergone limb-sparing surgery.*

A 20-year-old male presented a local recurrence of conventional osteosarcoma at the popliteal fossa with encasement of popliteal vascular bundle after previous tumor-wide resection and reconstruction with proximal tibia endoprosthesis. A wide resection "en bloc" of the lesion included part of the popliteal vessel. A bypass of both popliteal vessels, the vein with polytetrafluoroethylene (PTFE) prosthesis, and the artery with contralateral saphenous vein were performed to allow a limb salvage surgery.

Local management of recurrent osteosarcoma in a previously reconstructed limb is highly individualized.

This case confirms that preservation of lower limb function is possible using reconstruction techniques of bone and vessels in the sarcoma of the musculoskeletal system.

Keywords: *Osteosarcoma recurrence; Encasement of popliteal vascular bundle; Popliteal vessels reconstruction; Polytetrafluoroethylene prosthesis; Limb salvage surgery*

INTRODUCTION

Osteosarcoma is the most common primary bone malignancy. The prognosis of patients after local recurrence is generally poor and management of the locally recurrent disease is not well defined, especially in patients who have undergone limb-sparing surgery.

It has a bimodal age distribution; the first peak occurs during adolescence, is the most frequent bone cancer occurring in children and adolescents aged 10–20, and is rare before five years[1]. It can happen in any bone, but it most often occurs near the metaphyseal growth plates of the long bones of the extremities, usually the femur and tibia.[1]

The clinical presentation varies from case to case and largely depends on the type of osteosarcoma, its anatomic site, and the patient's age. Pain and soft tissue swelling or mass development are the most common presenting symptoms.[3] For diagnostic purposes, conventional radiography usually provides adequate information. Computed tomography (CT) and magnetic resonance imaging (MRI) is crucial for determining a tumor's intraosseous and extraosseous extension.[3] Osteosarcomas can be graded based on cellularity, cytologic atypia (nuclear

pleomorphism), and mitotic activity. Grading has clinical, therapeutic, and prognostic importance.[3] Approximately 20 % of patients have metastatic disease at the time of diagnosis, and the most common site of metastasis is the lung.[4]

The outcomes of patients with bone tumors of the lower extremities have historically been poor; in "pre-chemotherapy era," patients were treated only by surgery, and the majority were amputated (90%), and the cure rate was 11%.[5,6] Treatment strategy of giving preoperative chemotherapy followed by surgery and adjuvant therapy has dramatically improved the survival rates of patients with osteosarcoma over the past decades.[1] Death rates for osteosarcoma have been declining by about 1.3% per year from 1990–2004, and cure rates were escalated from <20% before 1907 to current levels of 65–75%.[1] Accompanying this escalation has been the ability to offer limb salvage to approximately 80% of patients.[1] However, limb salvage surgery must be individualized and consider the tumor stage and location, the presence of metastasis, and suitability for chemotherapy.[6]

Patient's prognosis after local osteosarcoma recurrence is generally poor, despite aggressive multimodal treatment. Managing the locally recurrent disease is not well defined, especially in patients undergoing limb-sparing surgery.[7] Surgical resection and metastasectomy showed more prolonged survival, while the role of chemotherapy for recurrent disease is unclear and much less defined than surgery.[4]

We present a case report with local recurrence of conventional osteosarcoma at the popliteal fossa with encasement of the popliteal vascular bundle after the previous wide resection and reconstruction with proximal tibia endoprosthesis

CLINICAL CASE

A 20-year-old male diagnosed with proximal tibia conventional (high grade) osteoblastic osteosarcoma (Figure 1). He was treated with three cycles of neoadjuvant chemotherapy with cisplatin and doxorubicin, followed by surgical treatment. One month later, he underwent a wide resection of the proximal tibia and fibula, followed by reconstruction with proximal tibia endoprosthesis (MUTARS - Implantcast®), reinsertion of the patellar tendon with Trevira tube (polyethylene terephthalate - Implantcast®), and soft tissue cover with internal gastrocnemius rotated flap (Figure 2). The histology report showed conventional osteoblastic osteosarcoma of the left proximal tibia G3 ypT1 R0. Chemotherapy-associated tumor necrosis was a Grade 3 according to the Rosen Grade, with 95-97% necrosis of the tumor area. Chemotherapy was resumed one month after surgery, completing three more cycles with cisplatin and doxorubicin regimen, completing six cycles.

Six months after surgery, he presented with pain and swelling at the popliteal fossa, suggestive of clinical findings of recurrence. CT scan and positron emission tomography (PET) scan suggested local recurrence. The biopsy confirmed osteoblastic neoplasia with atypia, compatible with previously diagnosed osteoblastic osteosarcoma. Left leg CT-angiography showed unresectable involvement of the popliteal vascular bundle (Figure 3).

The case was discussed in the multidisciplinary group meeting. To not compromise the oncologic outcome, a wide resection through an "en-bloc" resection was proposed, and since the vascular bundle needed to be sacrificed, a vascular reconstruction to allow a limb salvage was also decided.

A posterior approach through the popliteal fossa (gently curved incision starting laterally over the biceps femoris muscle) brings the incision obliquely across the popliteal fossa, turning downward over the medial head of the gastrocnemius muscle and run the incision inferiorly into the calf) was performed. Following identifying the limits to allow a wide resection, the proximal and distal parts of popliteal vessels were identified and clamped. After this, a wide resection "en bloc" of the lesion, around 6 cm of the popliteal artery and vein, was removed. Then, under systemic heparinization, a bypass of either of the popliteal

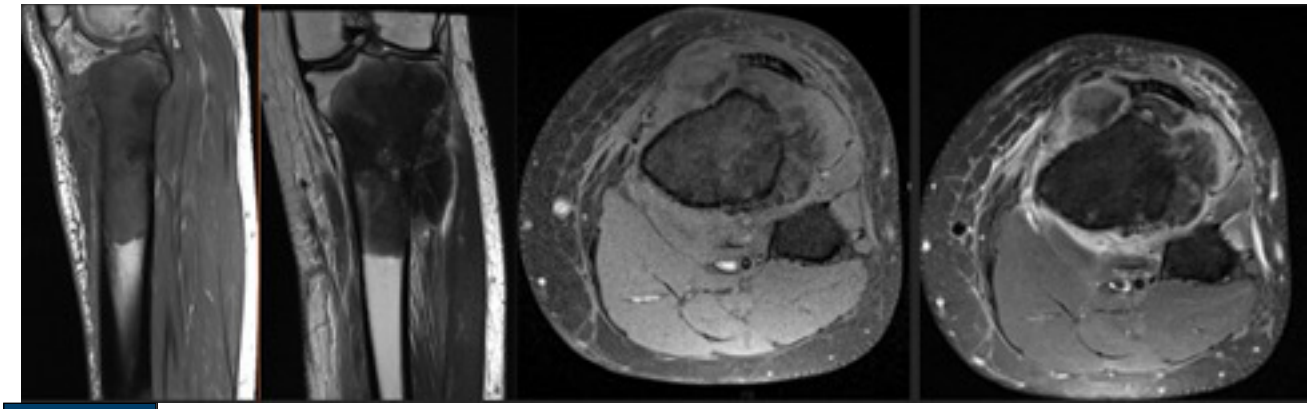

Figure 1 *MRI showing a proximal tibia conventional (high grade) osteoblastic osteosarcoma with a large soft tissue component, very close to the head of the fibula*

vein, with polytetrafluoroethylene (PTFE) prosthesis, and the popliteal artery, with contralateral saphenous vein, were performed (Figure 4). The artery bypass was performed with the interposition of an inverted saphenous vein of the contralateral leg in an end-to-end manner. The venous bypass was performed with a PTFE prosthesis.

At the hospital ward, 48 hours after the surgery, an eco doppler confirmed the arterial and venous bypass patency. After discharge, edoxaban (60 mg once a day) was prescribed.

The histology report was compatible with extra-osseous recurrence of previously diagnosed osteosarcoma G3rpT1R0, corresponding to an enlarged resection with free margins.

One month after surgery, the patient started a 4-cycle chemotherapy course with a high dose of ifosfamide for four months.

During follow-up, the patient was asymptomatic for one year with no clinical signs of local recurrence or complication related to the arterial and venous bypass (no significant edema or vascular claudication). During follow-up, seven months after surgery, he suspended edoxaban. 11 months after surgery, an eco doppler was performed, revealing a permeable arterial bypass and the venous bypass with PTFE was occluded but well compensated.

After rehabilitation, he recovered his knee range of motion and walking capacity autonomously. This allowed him to perform daily activities without limitations and even return to sport activities with low to moderate energy.

One year after surgery, he presented a local recurrence confirmed by Tru-Cut needle biopsy. The re-stating study showed distant lung, bone and ganglion metastases. Two years after local surgery, the patient remains under palliative treatment, with good function in affected limb.

DISCUSSION

The quality of life in patients with appendicular malignant bone tumors may be compromised if the salvaged limbs have sustained muscle or nerve damage that may cause agony and financial burden later. Furthermore, an endoprosthesis or other hardware in the limb can complicate surgical planning and may require an amputation even more proximal than would otherwise be indicated.[7]

A meta-analysis of six high-quality studies indicates that amputation and limb salvage surgery provide similar functional outcomes and quality of life for patients with osteosarcomas of the lower extremities.[6] However, Eiser et al. reported that those with a limb salvage procedure reported better daily competence and were less likely to require aid when walking.[8]

Local recurrence in osteosarcoma is clinically distinct from metastasis, although associated with a similar reduction in survival.[9] One study reports local recurrence of 9% (18/200) in patients that underwent limb-sparing surgery at a median of 2.5 years of follow-up.[7] Twelve (67 %) of those died within 15 months of local recurrence.[7]

Five- and ten-year survival in patients with local recurrence were 29% and 10%, respectively, and five-year overall survival rate was significantly better for patients with late relapse compared to patients with early relapse (within one year after primary diagnosis).[9,10]

Durnali et al., in a retrospective study with 240 patients with high-grade osteosarcoma concluded that surgical resection of recurrent disease and metastasectomy extend survival. The role of chemotherapy for recurrent disease is unclear and much less defined than that of surgery, although in a few studies about relapsed patients, salvage chemotherapy correlated with more prolonged survival.[4]

Figure 2 *Status after wide resection including proximal tibia and fibula, and reconstruction with proximal tibia endoprosthesis (MUTARS®).*

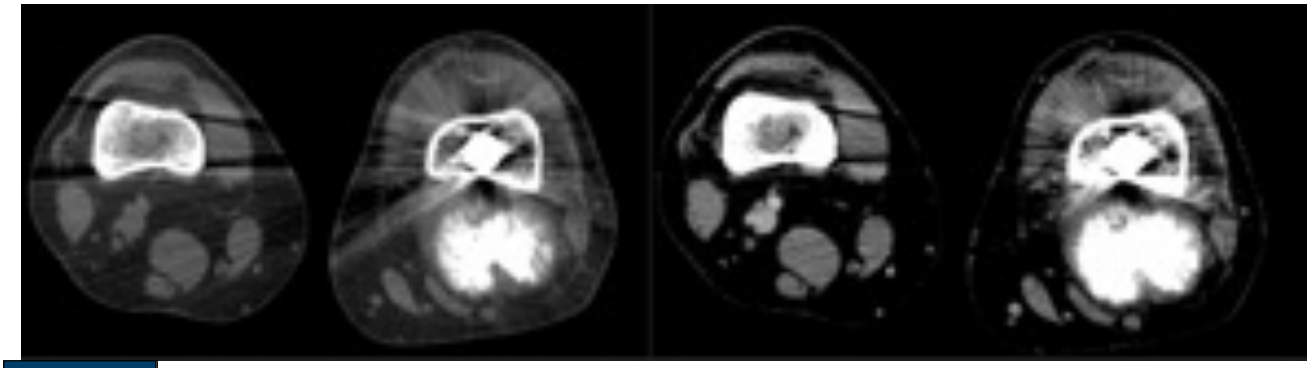

Figure 3 *Presence of a mass of extensively calcified in soft tissues measuring 62 mm in maximum axial diameter, contacting the ipsilateral popliteal artery.*

The best approach to manage a local relapse near a prosthesis or allograft remains unknown, and it is unclear whether further limb salvage should be attempted with the potential risk of obtaining marginal or even unintentional positive resection margins.[7] Conversely, it is unknown whether amputation at this point will affect an already poor prognosis and does not appear to provide significant survival benefit.[7,9]

Nathan et al. based on the prognostic strata, determined an algorithm for the patient with local recurrence.[9] In this case, without metastasis who develop local recurrence within one year of primary resection advocate chemotherapy before resection to improve resection margins and treat micrometastatic disease.[9]

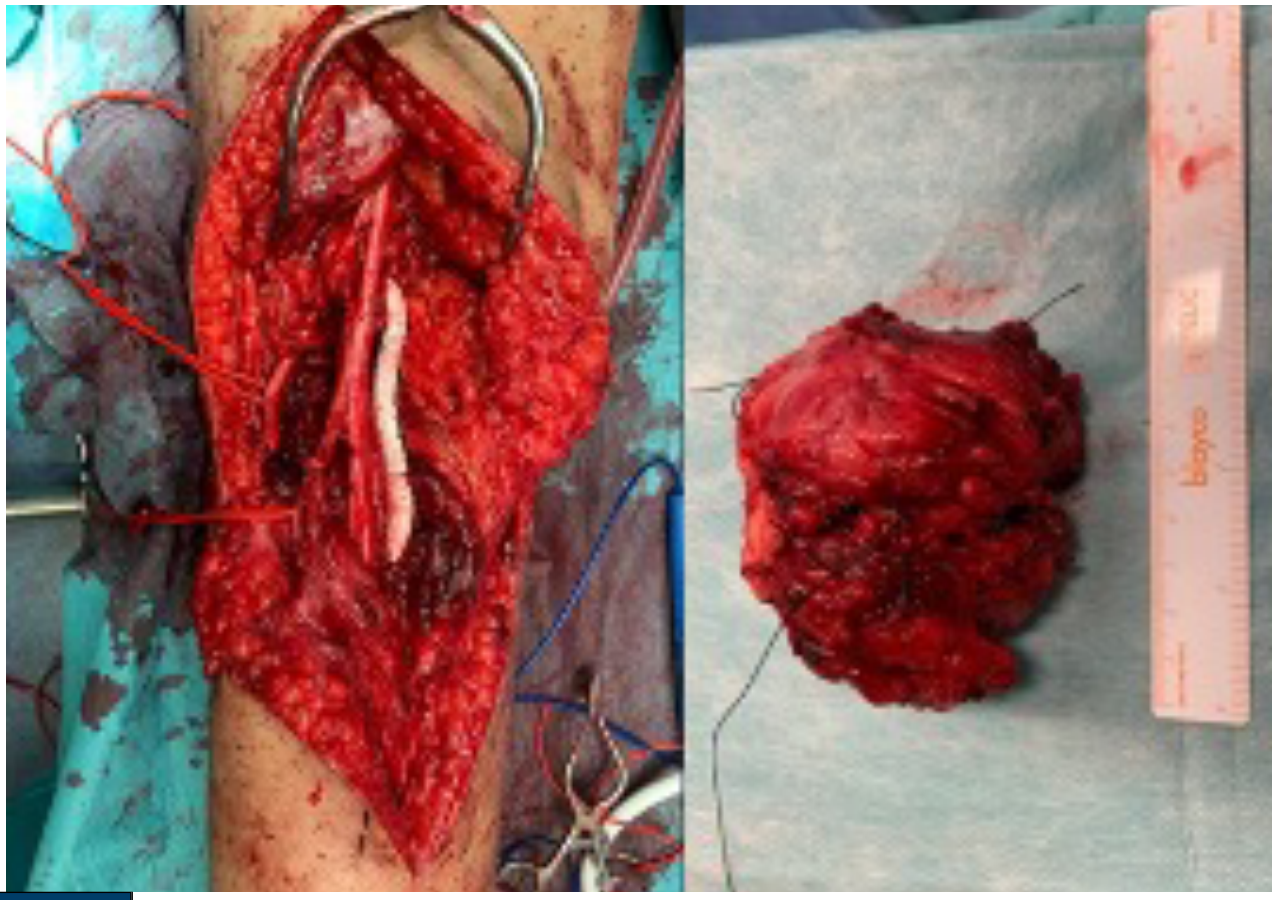

Figure 4 *Bypass of popliteal vein with PTFE prosthesis, and popliteal artery with contralateral saphenous vein.*

This case reports conventional osteosarcoma of proximal tibia treated initially with limb-sparing surgery with wide resection and reconstruction with proximal tibia endoprosthesis. Although a free margin (G3 ypT1 R0) and high chemotherapy-associated tumor necrosis (Grade 3 according to Rosen) were obtained, a soft tissue local recurrence at the popliteal fossa with encasement of popliteal vascular bundle occurred. Different studies report local recurrence after negative margins and favourable local response to chemotherapy. This may suggest that finding the involved margin contributing to local recurrence after a favorable response to preoperative chemotherapy is more difficult.[9] Furthermore, there is a lack of evidence showing the histopathological correlation between tumor percent necrosis and local recurrence.[9,11] However, tumor percent necrosis greater than 90% after neoadjuvant chemotherapy had a significantly better overall survival, as it has been correlated with lower rates of pulmonary metastasis.[9,10] In this case, a lesion at first deemed unresectable was successfully managed due to multidisciplinary teamwork with tumor resection and arterial and venous revascularization. For the venous bypass, using a saphenous venous graft was excluded due to the requirement of a larger gauge and a longer extension, which could make this procedure more complex or even impossible. The PTFE prosthesis was chosen to solve these problems. Wide margin resection and vascular reconstruction showed oncological effectiveness until present and an excellent functional outcome, providing less morbidity and better quality of life.

CONCLUSION

Local management of recurrent osteosarcoma in a previously reconstructed limb is highly individualized. Preservation of lower limb function is possible using reconstruction techniques of bone, nerves and vessels to improve functional quality of patients even in those whose clinical prognosis is poor.

REFERENCES

1. Jaffe N, Bruland ØS, Bielack S. Pediatric and Adolescent Osteosarcoma: Editorial summation. Cancer Treat Res 2009; 152: 573–575.
2. Linabery AM, Ross JA. Trends in childhood cancer incidence in the U.S. (1992-2004). Cancer 2008; 112: 416–432.
3. Greenspan A, Jundt G, Remagen W. Differential Diagnosis in Orthopaedic Oncology. Second Edi. 2007.
4. Durnali A, Alkis N, Cangur S, et al. Prognostic factors for teenage and adult patients with high-grade osteosarcoma: An analysis of 240 patients. Med Oncol; 30. Epub ahead of print 2013. DOI: 10.1007/s12032-013-0624-6.
5. Mario Campanacci; Gaetano Bacci Fbpp, Angel0 Minutillo; Claud10 Franceschi. The Treatment of Osteosarcoma of the Extremities: Twenty Years' Experience at the Istituto Ortopedico Rizzoli. Am Cancer Soc 1981; 48: 1569–81.
6. Mei J, Zhu XZ, Wang ZY, et al. Functional outcomes and quality of life in patients with osteosarcoma treated with amputation versus limb-salvage surgery: a systematic review and meta-analysis. Arch Orthop Trauma Surg 2014; 134: 1507–1516.
7. Loh AHP, Navid F, Wang C, et al. Management of Local Recurrence of Pediatric Osteosarcoma Following Limb-Sparing Surgery. Epub ahead of print 2014. DOI: 10.1245/s10434-014-3550-8.
8. Eiser C, Darlington ASE, Stride CB, et al. Quality of life implications as a consequence of surgery: Limb salvage, primary and secondary amputation. Sarcoma 2001; 5: 189–195.
9. Nathan SS, Gorlick R, Bukata S, et al. Treatment algorithm for locally recurrent osteosarcoma based on local disease-free interval and the presence of lung metastasis. Cancer 2006; 107: 1607–1616.
10. Aggerholm-Pedersen N, Maretty-Nielsen K, Keller J, et al. The importance of standardized treatment in high-grade osteosarcoma: 30 years of experience from a hospital-based database. Acta Oncol (Madr) 2015; 54: 17–24.
11. Kawaguchi N, Ahmed AR, Matsumoto S, et al. The Concept of Curative Margin in Surgery for Bone and Soft Tissue Sarcoma. Clin Orthop Relat Res 2004; 5: 165–172.