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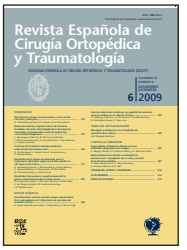

CASE REPORTS

Vascular reconstruction during limb preserving surgery in the treatment of lower limb sarcoma: A report on four cases

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KEYWORDS
Limb preserving surgery;
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Abstract
Purpose: To report on the experience acquired by our center in the field of surgical repair of the great vessels during tumor resection surgery in the lower limb and the pelvis.
Materials and Methods: We present 4 consecutive cases of lower limb musculoskeletal tumor resection surgery associated to great vessel reconstruction performed in the same surgical procedure. The procedures were performed in our hospital between 1990 and 2009. We analyzed tumor type, technique for vascular reconstruction and immediate and long-term complications.
Results: We studied a Ewing sarcoma of the pelvis; a pelvic chondrosarcoma, a parosteal distal femur sarcoma and an atypical thigh schwannoma. In two cases, repair was achieved through an intraoperative vascular lesion. In the other two cases, a vascular resection was carried out owing to the inexistence of a dissection plane between the tumor and the vessels. Limb preservation was achieves in three of the four cases.

PALABRAS CLAVE
Cirugía conservadora de la extremidad;
Lesión vascular;
Sarcoma

Reconstrucción vascular durante cirugía conservadora de la extremidad en el tratamiento de sarcomas del miembro inferior

Resumen
Objetivo: Exposición de la experiencia de este centro en reparación quirúrgica de vasos mayores durante la cirugía de resección de tumores en la extremidad inferior y la pelvis.
Material y método: Se presentan 4 casos consecutivos de cirugías de resección tumoral del miembro inferior asociadas a reconstrucción vascular de vasos mayores en el mismo acto quirúrgico en tumores del aparato locomotor realizadas en el hospital entre 1990 y

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2009. Se analizan el tipo tumoral, la reconstrucción vascular y las complicaciones inmediatas y a largo plazo que presentaron.
Resultados: Se estudió un sarcoma de Ewing de pelvis, un condrosarcoma de pelvis, un sarcoma parostal de fémur distal y un schwannoma atípico de muslo. En 2 casos la reparación se realizó por lesión vascular intraoperatoria. En los otros 2 casos se realizó una resección vascular por no haber plano de disección entre el tumor y los vasos. Se logró conservar la extremidad en 3 de los 4 casos.

Introduction

Limb preserving surgery has become the surgical treatment of choice for the majority of lower limb bone or soft tissue sarcomas[1,2]. With the use of adjuvant chemotherapy, radiation therapy protocols or both, as well as advanced Imaging techniques, it is possible to obtain wide surgical margins in the majority of cases without the need to resort to amputation[1,3]. Complete tumor resection is essential to obtain local control of the disease and improve survival[1-3]. When the tumor includes the major vessels in the limb, their preservation becomes much more difficult[1,3].

If the tumor originates in the vessels themselves, its resection becomes inevitable[1]. If the tumor is adjacent to the vessels and infiltrates or surrounds them, it may be necessary to resect the vessels together with the tumor in order to obtain a tumor-free surgical margin in accordance with the recommended procedures[1,4]. The adventitia is regarded as an adequate surgical margin provided that the surgeon longitudinally splits the adventitia opposite the tumor thereby obtaining a rim of healthy tissue at the vessel-tumor interface, as reported by a few authors[1,5]. Nevertheless, resection of the vascular segment provides a wider margin[1,4]. For this reason, it is believed that vessels must be resected when a wide margin cannot be obtained otherwise[4]. Finally, it should not be forgotten that tumor-free vessels may sustain an injury that requires repair by means of vascular surgery techniques.

The purpose of this study is to present the results obtained in 4 cases operated in our center where it was necessary to reconstruct the vascular axis.

Clinical cases

Case 1

Fifteen-tear-old male with no significant disease history, who presented with an Ewing sarcoma in the left hemipelvis.

A plain film showed increased bone density in areas ii and iii of the left hemipelvis, with osteolysis in the ischial tuberosity (fig. 1A). In the computerized tomography (CT) the soft tissue mass showed intra- and extrapelvic involvement, surrounding an osteoblastic lesion of the iliac body and the left ischiopubic ramus (fig. 1B). A bone scan was performed, which revealed increased uptake in the ilioischial area of the left hemipelvis.

Neoadjuvant treatment was administered with alternating cycles of chemotherapy in November 1990. Drugs administered were etopoxide (VP16), ifosfamide (IFx), vincristine with cyclophosphamide (VCR/CTX), adriamicyn (ADR) and vincristine (VCR); a satisfactory response was obtained with a significant reduction of the tumor volume (fig. 1C). The patient did not receive radiation therapy. At that moment, the patient had a Karnofsky index of 80%, was pain-free and could only ambulate with the help of 2 walking canes because of a 35° flexum deformity in the left hip.

The patient was operated in December 1990 by resecting areas ii and iii of the left hemipelvis. He had been subjected to a preoperative angiogram that showed increased vascularization in the area occupied by the tumor; no signs of iliac vessel compression were detected.

Intraoperatively, an irreparable vascular lesion occurred of the left external iliac artery and vein, which made the excision of that section necessary. During the same procedure, a vascular reconstruction was performed by means of an end-to-end arterial and venous bypass with a synthetic expanded polytetrafluoroethylene (PTFE) graft. Finally, the pelvic resection was stabilized by means of a pelvifemoral arthrodesis with an L plate (fig. 1D).

Postoperatively, the patient developed an abscess-induced deep *Staphylococcus aureus* infection, which made a reoperation necessary to perform a surgical drainage and lavage of the area. The hardware was removed but it was not necessary to withdraw the expanded PTFE graft.

The patient evolved favorably and is at present disease-free. The attempted arthrodesis failed and a stable and well-tolerated iliofemoral pseudoarthrosis was created (fig. 1E). The main sequela following surgery was a paresis of the left external popliteal sciatic nerve.

Case 2

34-year-old woman without a history of disease who was referred to us by her local hospital due to the appearance of a lytic lesion in the acetabular region of her right hemipelvis (fig. 2A and B). A trocar puncture biopsy was performed which revealed what seemed to be an enchondroma. Subsequently, the lesion was scraped off and the area filled with allograft. The pathologic diagnosis of the excised tissue and the inspection of the first few samples indicated that the lesion was in actual fact a grade II chondrosarcoma. A decision was made to reoperate the patient one month later to widen the margins of the resection.

Figure 1 A) Radiograph of the patient described in Case 1 showing increased bone density in areas ii and iii of the left hemipelvis and osteolytic lesions in the isichial portion of the bone. B) Computerized tomography (CT) of the patient described in Case 1 showing bony involvement caused by an Ewing sarcoma as well as an intrapelvic soft tissue mass created by the tumor (arrow). The study was performed prior to administration of chemotherapy. C) Post-chemotherapy CT. D) Left pelvifemoral arthrodesis following tumor resection in the patient described in Case 1. E) Current follow-up radiograph of the same patient. He presents with a well tolerated iliofemoral pseudoarthrosis.

Eighteen months later, the patient developed recurrence of the tumor, with the lesion extending to the acetabulum and the right iliopubic ramus. A new biopsy was carried out that confirmed recurrence of a grade II chondrosarcoma. In October 2005 a wide excision of the neoplasm was made, resecting areas ii and iii of the right hemipelvis and of the femoral head and neck. The report from the Pathology Department revealed that the pelvic osteotomy was tumor-free. A cryopreserved structural graft, fixed with reconstruction plates, and a bipolar prosthesis were used for the reconstruction (fig. 2C). Surgery required careful dissection of the common iliac vessels, the external iliac vessels and the superficial and deep femoral vessels, together with the crural nerve. This surgery did not reveal tumor infiltration of the vascular axis.

In March 2007 a new recurrence of the tumor was detected: a 4x8 cm intrapelvic neoplasm located in the vicinity of the right iliac bone, more specifically at the bifurcation of the common iliac vessels. A wide excision of the mass was performed.

In December 2007 a further procedure was carried out because of a new recurrence in area i of the right hemipelvis. During the withdrawal of the hardware, a sizeable tear of the external iliac vessels occurred because of a decubitus lesion of the external iliac artery against the fixation plate. The artery was repaired with an end-to-end PTFE by pass. As a result of this vascular complication, a marginal excision of the neoplasm was carried out in this case, resecting the anterior portion of the iliac crest; the hemipelvis was provisionally stabilized with molded polymethylmetacrylate (fig. 2D). None of the studies conducted prior to this surgery has revealed any kind of vascular involvement.

Two further vascular procedures were necessary in the immediate post-op period: implantation of a side-to-side bypass with a contralateral external saphenous vein graft to treat thrombosis of the previously implanted synthetic

Figure 2 A) Plain film prior to the pelvic lesion in the patient described in Case 2. B) Computerized tomographic view. C) X-ray of the patient described in Case 2 following resection of areas ii and iii of the right hemipelvis that shows the reconstruction with bone bank allograft and a bipolar prosthesis. D) Radiograph taken following the surgery at which the iliac artery was ruptured. Note the result obtained following resection of area i and emergency stabilization with polymethylmetacrylate and screws.

bypass and a popliteal thrombectomy to treat thrombosis distally to the graft.

In June 2008, a follow-up TC revealed the appearance of an irregular cyst-like mass in the greater pelvis which reached up to the sacrum. We consulted the gynecology and the general surgery departments as there was infiltration of the right ovary and uterine tube as well as digestive tract involvement. The patient's right lower limb was affected by chronic ischemia given malfunctioning of the vascular bypass. In July 2008 the hip was disarticulated by means of a right external hemipelvectomy that included the ovary and the uterine tube, and the pathologic study revealed that a free resection margin had been achieved.

On completion of this study (May 2009) a positron emission tomography found no tumor recurrences or distant metastases. The patient was able to ambulate with a prosthesis.

Case 3

Forty-four-year-old male with no relevant history of disease who, in May 1994, was diagnosed with an osteochondroma on the posterior surface of the right distal femur (fig. 3A). The patient was referred to us from his local hospital to be treated for lesion enlargement and pain intensification. He was subjected to a simple excision with scraping of the tumor bed; subsequent evolution was satisfactory. .

In March 1999 the tumor recurred with a definitely malignant x-ray appearance. An incisional biopsy was carried out that revealed the presence of a parosteal osteosarcoma. A decision was made to perform a wide resection of the tumor, filling the defect with structural bone bank allograft (fig. 3B). As a sequela to this surgery, the patient presented with transient paresis of the EPS nerve, which resolved spontaneously in the following weeks. At 4 months from surgery the bone graft already showed signs of incorporation and the evolution of the case was favorable.

In July 2007 a new recurrence of the osteosarcoma was detected and confirmed by a puncture biopsy. The tumor involved all of the distal third of the right femur, it infiltrated the soft tissues and, according to the preoperative MRi and CT studies, affected the vascular bundle. The aforementioned studies revealed the absence

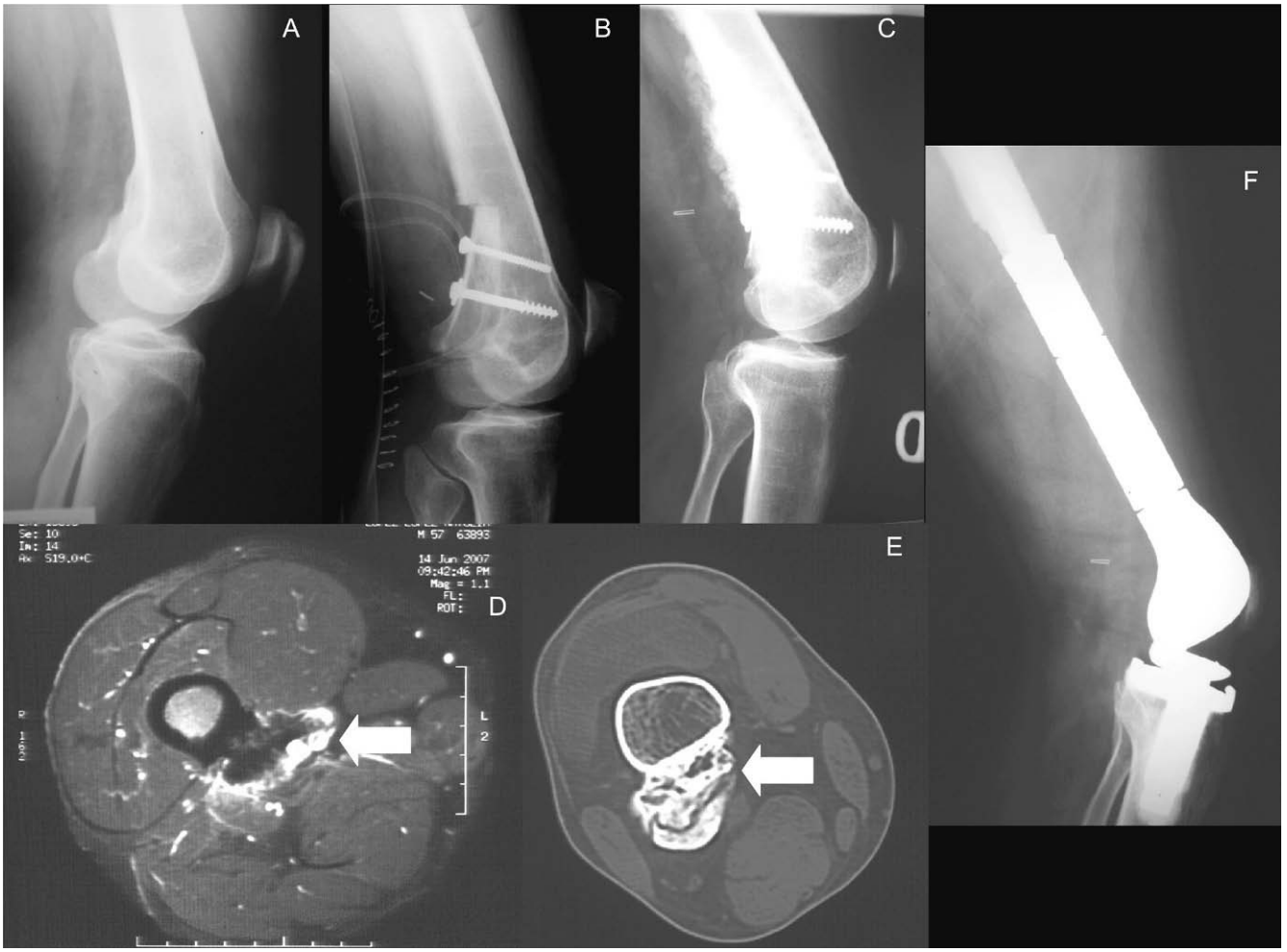

Figure 3 A) Distal femur radiograph of the patient described in Case 3 that shows the lesion initially described as an osteochondroma. B) Follow-up radiograph of the same patient where we resected the parosteal osteosarcoma and performed a reconstruction with structural bone bank allograft. C) Radiograph of the same patient showing recurrence of the parosteal osteosarcoma over the allograft. D) Magnetic resonance view showing the relationship of the femoral vessels with the tumor (arrow). E) Computerized tomography showing tumor growth around the femoral vessels. F) Follow-up radiograph of the tumor resection surgery showing the reconstruction carried out by means of a tumor knee prosthesis.

of a dissection plane between the vessels and the tumor (fig. 2C, D and E). After careful consideration, it was decided to carry out a wide resection of the tumor. An excision was made of around 20 cm of the distal femur and of the soft tissue tumoral mass, together with the section of the femoral artery and vein engulfed by the tumor. The joint was reconstructed by means of a 220 mm-long tumor prosthesis (fig. 2F), while the arterial axis was addressed by an end-to-end graft of around 20 cm taken from the contralateral saphenous vein from the superficial femoral portion to the second popliteal portion. Reconstruction of the vein was not deemed necessary.

A pathologic study showed infiltration of the adventitia by the tumor; the infiltration did not however reach the vascular wall or cause endovascular lesions.

Postoperative evolution was satisfactory with the patient achieving a good functional level with no signs of recurrence at 12 months from surgery.

Case 4

Forty-nine-year-old male with no history of disease who in December 2002 was referred to us from another hospital for the appearance of a soft tissue mass in the right groin.

Physical examination led to the identification of a firm subfascial mass of around 8?4 cm that hurt on palpation and which was located at the root of the right thigh, with no related adenopathies or evidence of collateral circulation.

An MRI was performed, which showed the solid mass in intimate association with the superficial femoral vessels (fig. 4A). A fine needle puncture-aspiration was carried out, which identified a low to intermediate-grade fusocellular sarcoma. An arteriogram and a phlebogram were also carried out and showed permeable superficial femoral vessels in spite of the compression exerted by the tumor

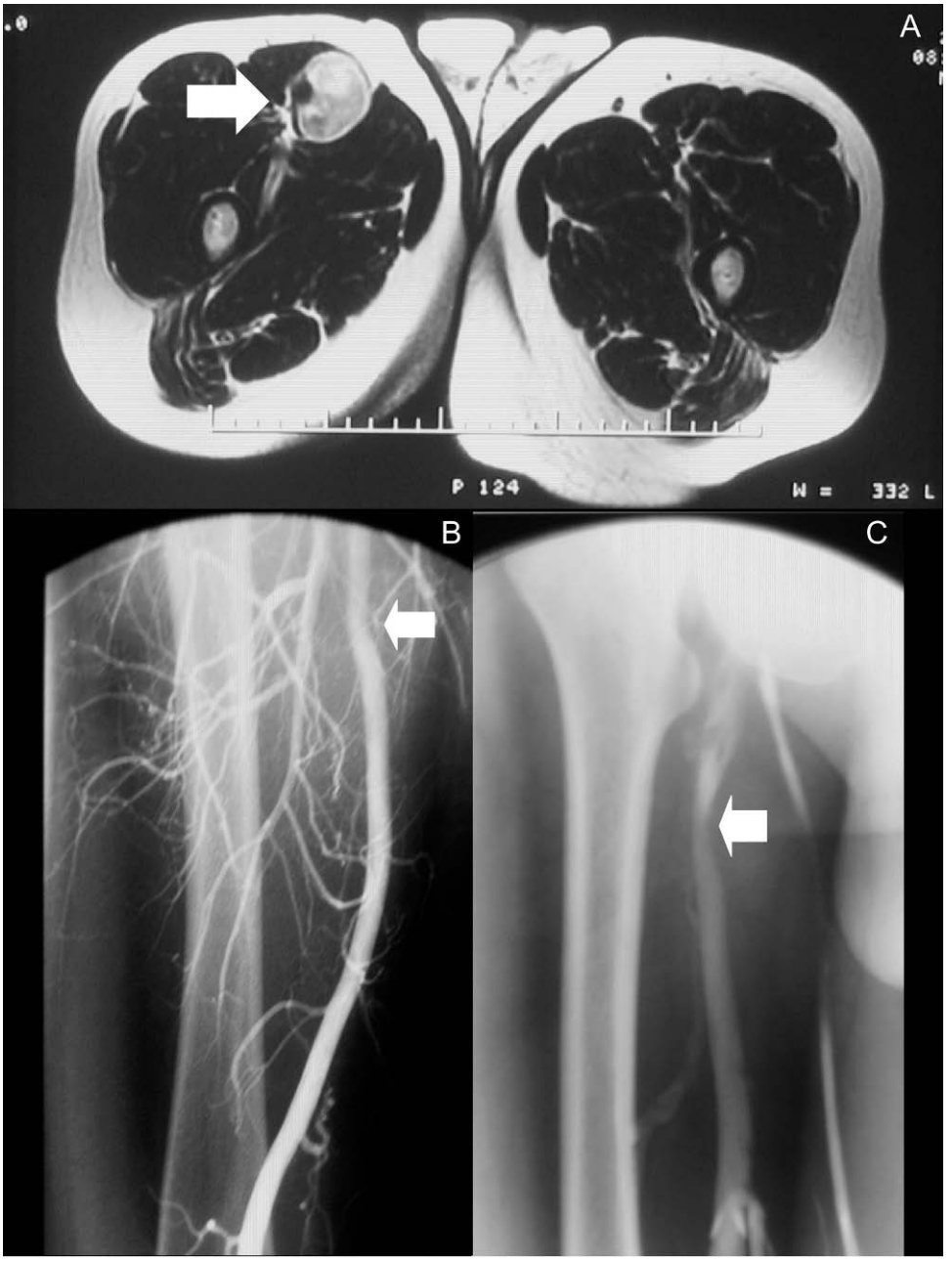

Figure 4 A) Magnetic resonance view of the patient described in Case 4 showing the relationship of the tumor and the femoral vessels. B) Arteriogram of the same patient showing the compression exerted by the tumor on the superficial femoral artery. C) Phlebogram of the patient described in Case 4.

(fig. 4B and C). The extension study (a thoracoabdominopelvic CT) showed no metastatic lesions.

In January 2003 a wide excision was made of the neoplasm, which included an en bloc resection of around 7 cm from the superficial femoral vessels as there was no dissection plane that permitted their preservation and compliance with the standard oncologic criteria. The superficial femoral vein was ligated, whereas the artery was reconstructed with a PTFE interposition graft.

According to the pathologic study, the neoplasm was an atypical schwannoma, with tumor-free resection margins and with no tumor infiltration of the vascular wall.

In terms of late complications, the patient developed a lymphocele with cutaneous fistulization, which resolved spontaneously.

After a 5-year follow-up, the patient is now disease-free, with a single lymphedema as the only sequela, which he controls with physical measures.

Discussion

Patients who develop tumors in their limbs often present with involvement of the greater vessels as a result of either invasion of the vascular wall by the adjacent neoplasm or confinement due to the pressure exerted by the growing tumor[1,6]. Another possibility is that, given the complexity of surgery, an irreparable lesion could be caused to a vessel intraoperatively making reconstruction necessary by vascular surgery techniques. Between 1990 and 2009, only 4 cases in our Center have required vascular reconstruction out of a total 1.351 musculoskeletal tumors resected over the same period. None of these 4 patients received pre- or postoperative radiation therapy.

One of these patients (case 3) showed tumor infiltration of the adventitia; in another one (case 4), growth of the tumor around the vessels made it safer to resect it *en bloc* than having to dissect them and separate them from the tumor since in the latter case the surgical margins obtained would not have been wide enough. In the other cases a severe lesion to the vessels occurred in the course of tumor resection.

In the experience of the authors of this study, the most common reason for vascular repair has been an attempt to obtain wide surgical margins in cases in which the surgeon failed to detach the tumor from the vessels or in which an irreparable lesion was caused intraoperatively to any of the major vessels of the limb. Vascular infiltration by primary bone or soft tissue sarcomas is rare. The authors have had no cases in which the limb was rescued through vascular reconstruction under these circumstances. However, it is fairly frequent to find this situation in tumor recurrences as in the patient described in Case 3[1,2,7]. Moreover, in these cases, surgical manipulations can contribute to the development of vascular involvement.

The number of cases in which limb preservation surgical techniques have been carried out in conjunction with arteriovenous reconstruction is very small when compared with the amount of tumors excised in our Department during the study period. This is in line with the experience of other authors who estimate that only 10%of soft tissue sarcomas present with vessel involvement, with less than 50%of them requiring vascular repair[1].

Limb preservation surgery has become the treatment of choice for the surgical management of the majority of patients with bone or soft tissue sarcomas of the limbs[1,2]. When the main artery is involved and must be resected, it is essential to reconstruct it since resection without revascularization normally leads to the loss of the limb[6]. On the other hand, venous reconstruction does not normally follow any protocol as regards the indication of reconstruction, but repair is preferable in order to prevent distal venous hypertension and its sequelae[6]. In the 4 cases we reconstructed the arterial axis and in one of them (case 1) we also reconstructed the venous trunk. Resection without reconstruction of the venous trunk seems to increase the incidence of distal edema, venous claudication, itching and cutaneous inflammation of the involved limb[7]. Nonetheless, venous reconstruction often leads to complications, such as graft occlusion and edema. In a series of 17 patients, Nishinari et al obtained a venous graft survivorship at 2 years of 79.3%. The case in our study that underwent venous reconstruction still maintains good graft patency after 10 years' follow-up. All patients presented with edema in the operated limb that was well tolerated when physical measures were applied. There was a single instance of an intense lymphedema in the limb of one patient (case 4), who initially developed a lymphocele with cutaneous fistulization that subsequently progressed to a lymphedema with satisfactory long-term evolution (table 1).

Synthetic expanded PTFE grafts were used 4 times in this series; on another 2 occasions contralateral autologous saphenous vein grafts were used. The use of autologous vascular grafts is recommended for vascular reconstruction, mainly for venous reconstruction, provided that they are of a caliber similar to the vessel to be replaced. Otherwise, synthetic grafts must be used[1,2,8]. The authors of this study have no experience of the use of bone bank vascular grafts in these types of surgery.

The appearance of complications is common in surgeries that require resection of the greater vessels and these are not subsequently reconstructed, or when the reconstruction is not successful[2]. As regards vascular complications in the immediate post-op period, we ha done instance of thrombosis of the synthetic graft, which was resolved with a new bypass and a thrombectomy (case 2). In this patient, vascular resection and reconstruction were not performed as elective procedures but rather as a result of an accidental

Table 1 Vascular and non-vascular complications (EPS, LLL, RLL)

Case	Tumor	Location	Complications	
			Non-vascular	Vascular
1	Ewing sarcoma	Left hemipelvis	Infection and abscess	LLL edema
			EPS nerve palsy	
2	Grade II chondrosarcoma	Right hemipelvis	—	Bypass thrombosis
				Right popliteal artery embolism
3	Parosteal osteosarcoma	Right femur	—	RLL lymphedema
4	Atypical schwannoma	Right thigh	—	Lymphocele with fistula
				ELL lymphedema

EPS: external popliteal sciatic nerve; RLL: right lower limb; LLL: left lower limb.

vascular tear in a badly damaged area subjected to several prior surgeries. The fact that reconstruction was an emergency non-elective procedure may have played a role in the subsequent vascular complications and in the poor prognosis of this case together with the type of tumor developed by the patient (a pelvic chondrosarcoma) and the numerous local recurrences that occurred. In this case, as the surrounding vessels and tissues were badly damaged at the time of reconstruction a radical type of procedure was selected given the impossibility to obtain a satisfactory functional result through conservative surgery. In the other patient where the vascular reconstruction was not pre-programmed (case 1), the final result was far more satisfactory, doubtless because it was a primary procedure and the tissues were in very good condition.

Appropriate pre-operative planning is essential to achieve a good surgical result since this allows the surgeon to anticipate the potential vascular complications that may arise. The tumor resections and vascular reconstructions described were carried out in collaboration with the Department of Vascular Surgery of our hospital. Participation of vascular surgeons in the resection of tumors where there is involvement of major vascular structures is fundamental for prevention and treatment of iatrogenic injuries; vascular surgeons should also participate in the subsequent reconstruction of the vessels[9]. Therefore, hospitals that carry out oncologic orthopedic procedures must have vascular surgeons in their staff.

Non-vascular complications reported in the medical literature include neurologic impairment, flap necrosis, wound infection, hematoma and enteric fistulae[2]. In our series, we had one case of wound infection and abscess formation, which was resolved with surgical lavage and hardware removal and a sequela in the form of EPS nerve paresis at 10 years' follow-up in the same patient.

To facilitate surgical planning it is necessary to carry out a thorough extension study that includes imaging techniques. MRi provides multi-planar high-resolution anatomic images without having to irradiate the tissues[10]. Anterial or venous angiography makes it possible to visualize tumoral vascularization and its relationship with vascular trunks[10]. For the authors of this study, apart from being essential for correct surgical planning, this test allows the surgeon to perform preoperative embolizations in certain hypervascular tumors located in complex anatomic regions such as the pelvis. Angio-MRi constitutes an alternative to angiography since it provides tridimensional vessel reocnstructions[10]. These techniques make it possible to identify degree of tumor involvement of the different vascular structures and, if appropriate, plan for vascular resection and subsequent vessel reconstruction.

Although the number of patients in this series is too limited to be able to carry out a statistical analysis, the authors believe that vascular reconstruction allows the surgeon to carry out a lower limb preserving procedure with good functional results in the context of tumor resection with a related vascular lesion. Careful planning of the resection is essential to prevent intra- and postoperative complications. Inclusion of vascular surgeons in the surgical team is also fundamental.

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