Hip disarticulation with pedicled total leg fillet flap for recurrent fungating sarcoma

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Abstract
Soft tissue sarcomas may be treated with limb-sparing procedures in the majority of cases; however, certain cases involving significant tumor spread and fungation may call for amputation. In the thigh, hip disarticulation typically involves a pedicled gluteus maximus flap or a pedicled anterior quadriceps flap. In this case report, we describe a rare situation in which the anterior flap, posterior flap, and adductor flap musculature were contaminated with tumor; therefore, a hip disarticulation was performed applying a pedicled total leg fillet flap for closure. Eighteen months after treatment, the patient continues to have no local recurrence of disease, a stable flap site, and ambulates with a walker. We present this amputation and closure method as a potentially effective modality in treating extensive oncologic disease of the proximal lower extremity.

Keywords
fillet flap, fungation, hip disarticulation, pedicle flap, sarcoma

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Introduction
Soft tissue sarcomas have undergone a transformation of care in the last several decades. In this modern era, at least 90% of patients may undergo limb-sparing surgery rather than amputation; however, certain situations still benefit from an amputation.1 In particular, amputation may be the most appropriate option in cases with critical nerve neurovascular involvement, significant contamination of the surrounding tissues due to tumor recurrence or prior incomplete surgery, or tumor fungation.2

For tumors of the thigh, amputation often consists of a hip disarticulation. The standard hip disarticulation described by Boyd in 1947 involves a posterior flap pedicled gluteus maximus closure.3 This flap depends on an intact internal iliac artery, which branches into the superior and inferior gluteal arteries supplying the gluteus maximus.4,5 However, this technique is unfavorable in cases where the gluteal musculature is compromised. In such cases, an anterior flap hip disarticulation, involving a portion of or the entire quadriceps muscle group and the superficial femoral artery pedicle, is the typical alternative technique.6 A less common salvage procedure, the medial thigh adductor flap, has been described in cases of posterior gluteal and anterior quadriceps contamination.7 To the best of our knowledge, there is limited literature applying pedicled, island, or fillet flaps of the lower extremity to cover hemipelvectomies or hip disarticulation.8 Even so,
none have employed solely the femoral vascular bundle as a pedicle with a flap composed entirely of filleted leg musculature. In this case report, we describe a rare situation in which the anterior flap, posterior flap, and adductor flap musculature were contaminated with tumor; therefore, a hip disarticulation was performed applying a pedicled total leg fillet flap for closure.

**Case presentation**

A 51-year-old female presented with a fungating metastatic high-grade undifferentiated pleomorphic sarcoma (UPS), also known as malignant fibrous histiocytoma, in her right lower extremity. She had a complex oncological history and was first diagnosed with sarcoma of the right posterior thigh in 2004 at age 39, after which she underwent an excision by another surgeon and was found to have a high-grade liposarcoma with close posterior margins. She received radiation therapy postoperatively, and in 2005, she underwent attempted wide reresection of the right posterior thigh mass with primary closure. Later that year during the routine follow-up, she was found to have a 9-cm mass abutting the uterus and bladder and subsequently underwent a laparotomy and radical resection of the intra-abdominal mass along with small bowel resection. This mass was diagnosed as a liposarcoma. During the intra-abdominal mass resection, a 1-cm retrorectal lesion was also observed. She then underwent four rounds of adjuvant chemotherapy with doxorubicin, ifosfamide, cetuximab, and mesna for bladder protection. In 2007, computed tomography (CT) scan showed that the retrorectal lesion had grown to 10 cm, and CT-guided biopsy demonstrated the presence of liposarcoma. She then underwent a right thoracotomy and radical retrocrural mass resection.

Imaging in 2011 demonstrated local recurrence of the right posterior thigh mass as well as two metastatic mediastinal lesions. The patient underwent wide resection of the right posterior thigh mass with complex closure of the 16 cm incision and a left thoracotomy with resection of the two mediastinal masses. The posterior thigh mass was found to be a new high-grade UPS with negative margins, which was thought to be secondary to radiation. Postoperative wound healing was prolonged taking nearly 1 year. In 2012, imaging showed a new nodule at the superior aspect of the wound. Core biopsy was consistent with recurrent UPS. She underwent resection of the recurrent 4 × 3 cm² thigh tumor with primary closure.

The patient continued to do well with close follow-up, but in 2014 was found to have evidence of right posterior thigh sarcoma recurrence on magnetic resonance imaging (MRI) and subsequent biopsy. This was her first presentation to our office. At this time, she was found to have intact sciatic and femoral nerves and appropriate perfusion to the lower extremity. Although amputation was considered at this point, because of her presence of metastatic disease, as well as a reasonably functioning lower extremity without neurovascular invasion of the recurrent tumor and without fungation, she decided against amputation. She then underwent right posterior thigh mass resection and local inferior gluteal rotational flap closure with KCI W.V.A.C.® (San Antonio, Texas, USA) application. She underwent subsequent partial closure and was discharged with a V.A.C., undergoing multiple debridements. In the following year she developed cellulitis with purulent drainage and underwent a formal irrigation and debridement in the operating room. MRI in 2016 demonstrated a new enhancing nodule and the patient underwent a biopsy with interventional radiology, which was consistent with recurrent high-grade sarcoma. In 2017, MRI showed interval growth of the lesions (Figure 1).

Due to concerns for the fungation of the tumor and repeated recurrence, treatment options were discussed with the patient. Amputation and limb sparing were the options; however, amputation was deemed more favorable based on the recurrence history, size, and anatomical position of the masses. After a thorough conversation about the risks, benefits, and alternatives of the procedure, the patient decided that she would like to proceed with an amputation. The case was planned in coordination with plastics and reconstructive surgery for wound flap and coverage.

The patient was placed in the left lateral decubitus position. A sequential compressive device was placed on the contralateral lower extremity and the operation was performed under general anesthesia. Regions of fungating tumor, which included the right thigh and buttock, were sealed off with towels, sutures, staples, and impervious iodine-impregnated incision drape Ioban® 2 (3-M, St. Paul, Minnesota, USA) to minimize risk of sarcoma exposure to the surgical bed. The right lower extremity was then prepared and draped in standard sterile fashion.

An incision was made from the anterior superior iliac spine toward the pubic symphysis and posteriorly coursing proximally, outside of the extensive scar tissue from prior procedures, toward the top of the pelvis on the posterior side. The circular incision was connected in the medial region avoiding the vagina and rectum. An incision was outlined from the femoral vessels in the groin distally toward the knee. Another circumferential mark was made just distal to the knee. The skin and subcutaneous tissues were divided, beginning with developing the proximal circular incision. The incision was made superior to the gluteus maximus, avoiding the region of previous tumor and surgery. The sciatic nerve was identified and carefully transected. An injection of local anesthetic was administered into the epineural sheath to reduce the likelihood of postoperative phantom pain. The hip joint capsule was then incised circumferentially and the ligamentum teres was divided, completing the hip disarticulation.

The femoral artery, vein, and nerve were identified in the groin. The femoral nerve and its branches into the saphenous were maintained to allow for sensation to the distal flap. The dissection was continued distally where
the profunda femoris artery and vein were identified and ligated because they communicated with the tumor region. Additional vasculature was ligated on the plane of the femoral artery and vein. Heavily irradiated tissue was encountered distally, and to reduce the risk of rupturing the femoral vein, the dissection was carried out along a sleeve of tissue alongside the femoral sheath. The dissection was continued toward the popliteal region where the popliteal artery and vein were dissected down distal to the knee. A circumferential incision was then made at the marked site just below the knee, and the knee was disarticulated (Figure 2(a)). The incision was then continued proximally, and the entire thigh was mobilized with the exception of the femoral artery, femoral vein, and femoral nerve with

Figure 1. Axial and sagittal T2 fat saturated MRI demonstrating tumor fungation and extensive involvement of the posterior and posterolateral thigh and buttock musculature. Of key note, the femoral vessels (blue arrow) from the groin all the way to the popliteal fossa were one of the few areas that were not invaded by tumor on the MRI, nor were they exposed during any of her prior surgeries. Also, note that there is no tumor nor any prior surgery in the area below the knee. MRI: magnetic resonance imaging.
corresponding saphenous nerve branches. These neurovascular structures were maintained as a pedicle to the distal lower extremity. The fungating mass was then removed en bloc and sent for pathology analysis (Figure 2(b)).

The remainder of the case was performed alongside a plastic surgeon. Dissection was carried out to the level of the bifurcation of the popliteal vessels—the tibioperoneal trunk and the anterior tibial vessels. Disarticulation of the proximal tibia-fibula joint was carefully performed to maintain the anterior tibial vessels. A small segment of the proximal fibula was resected. The vessels distally were then dissected and the musculature of the leg was stripped off of the tibia as the lower leg was filleted with attention toward maintaining the vasculature. The peroneal and tibial vessels were clamped and divided at the level of the ankle.

The tibia and fibula were resected and sent as separate specimens to pathology (Figure 2(c)). The transarticular ankle joint was also disarticulated and sent as a specimen to pathology. All bony structures were removed distally, and the right foot was amputated to create the final fillet flap (Figure 2(d)).

The defect in the right hip measured $25 \times 30$ cm$^2$ and involved the iliac crest posteriorly near the gluteal crease and extended inferiorly and medially to the groin crease. Mobilization of the fillet flap was performed along with careful coiling of the vessels. The flap was placed onto the right hip and trunk defect and repaired to the surrounding defect with the posterior calf located superiorly (Figure 2(e)). Two #10 flat Jackson–Pratt drains were inserted into the wound, and the fillet flap was inset and

Figure 2. Intraoperative photographs. (a) Hip disarticulation with dissection of thigh musculature (yellow arrow is pointing toward the disarticulated femoral head and blue arrow to the residual acetabulum). (b) En bloc resection of entire thigh including the recurrent sarcoma. (c) Preservation of femoral vessels with removal of distal lower extremity bony structures. (d) Final lower extremity pedicle flap. (e) Placement of pedicled flap. Note carefully coiled femoral vessels curving alongside medial and inferior border of acetabulum (blue arrow). (f) Postoperative appearance of flap.
closed sequentially in layers beginning with the fascial layer and continuing circumferentially (Figure 2(f)). The total area of repair was 90 cm. The wound was dressed with bacitracin ointment and Mepilex® (Möllycke Health Care, Gothenburg, Sweden).

The flap was warm and viable, and the patient was stable in the immediate postoperative period. She was placed on prophylactic cefazolin for 3 days and also required transfusion of two units of packed red blood cells within 24 h of the surgery for a hemoglobin/hematocrit of 7.0/21.0 g/dL, which subsequently increased to 9.4/27.9 g/dL. She was seen by the acute pain service and placed on gabapentin for phantom limb pain. She also had a documented history of seizure disorder, and she had a seizure during her hospital stay on postoperative day 7, after which neurology titrated her medications appropriately.

After discharge to a rehabilitation facility, she obtained a right hip abscess with blood cultures positive for methicillin-sensitive Staphylococcus aureus, which was treated by drainage with interventional radiology and an

Figure 3. Postoperative photographs. (a to d) Six-week postoperative visit. Note patient ambulates with walker. (e and f) Six-month postoperative visit. Note significant reduction in bulk of flap.
8-week course of intravenous nafcillin. She had a mechanical fall at 5 months postoperatively, which resulted in some buttock bruising that healed well. At 8 months postoperatively, she underwent repeat thoracotomy with peri-aortic and periesophageal mass resection. Of note, she also underwent thoracotomies in 2011 and 2015 for similar masses. Her postoperative course was otherwise uneventful. She ambulated with a walker at 6 weeks postoperatively, and her flap decreased in bulk significantly by the 6-month postoperative time point (Figure 3). She continues to do well and ambulates with a walker at 18 months after the procedure.

Discussion

The complex nature of this hip disarticulation laid in the compromised nature of the surrounding thigh and buttock tissues. The principal options were a gluteal posterior flap or an anterior quadriceps flap, as the extensive vascularity of the gluteal and posterior thigh region allows for myriad use in reconstructive surgery, including for sacral neuro- of the gluteal and posterior thigh region allows for myriad use in reconstructive surgery, including for sacral neurogenic tumors. However, these were not viable options due to gross contamination from tumor and prior surgery in these areas. The entire length of femoral vessels, from the groin all the way to the popliteal fossa, was one of the areas that did not involve gross tumor and was not previously operated on. Furthermore, there was never any tumor involvement nor was there any surgery on this patient in the area below the knee. This allowed for the unique possibility of utilizing a pedicled flap, with the pedicle consisting of the femoral artery and vein, involving the uncontaminated tissues below the knee.

The other main option was a free flap such as latissimus dorsi. While this would have been the leading option had there been contamination of the femoral vessels, in the context of extensive radiation and multiple prior surgeries, we believe this would have been at higher risk of failure due to challenges of microvascular surgery. In comparison, a pedicled flap would not require microvascular surgery. However, there was still risk of clotting in the vessels due to kinking or coiling up. Also, there was risk of local tumor recurrence due to retention of the femoral vessels. Even though the femoral vessels showed no gross tumor involvement on MRI nor was there any prior surgery in this area, given the extensive surgeries, radiation, and history of non-healing wounds in this patient, the femoral vessels could in principle have been contaminated. We want to emphasize that this approach could only be considered since there was reasonable belief that there was no contamination of the femoral vessels.

We also considered attempting to maximize patient function by including bone below the knee such as the tibia and fibula in the pedicled flap. This could have been accomplished with a rotationplasty, which involves rotating a distal segment 180° and fixing it to a proximal segment to leave a functional and durable salvage limb. We thus contemplated rotating the leg 180° and performing a hip arthroplasty into the tibia, allowing the tibia to articulate with the acetabulum. Another option was to retain the tibia and perform a hip arthroplasty into it. The reason we decided against these options was to avoid the placement of hardware. Given the patient’s history and the complex nature of this surgery, infection or wound healing problems were likely to occur postoperatively, and actually did occur. The presence of hardware would have increased this risk; moreover, the risk of requiring a revision was deemed undesirable.

In addition, a small case series showed that a pedicled total leg fillet flap provided viable coverage of extensive stage IV pressure sores of the trochanteric, sacral, and ischial regions in paraplegic patients. In our patient, the viable leg musculature and need for a salvage technique to provide closure made a pedicle total leg fillet flap the optimal choice. We found that preservation of the femoral vessels allowed for creation of a pedicle, and the total leg fillet flap was successfully applied to the hip region.

The results of this case are similar to prior literature on the use of various flaps to cover hip disarticulations and hemipelvectomies in terms of wound healing and overall patient function. A case described using a free leg fillet flap for closure of a hemipelvectomy performed for sarcoma involving a wound defect of similar size demonstrated appropriate wound healing. However, this flap utilized anastomoses of the popliteal vasculature to the iliac vessels, which was not needed in our case due to the preservation of the femoral vasculature in its entirety. Another case used a pedicled leg fillet flap to cover a hemipelvectomy and abdominal defect after femoral prosthesis resection and aortic bifemoral Y-prosthesis. In contrast to our case, this situation did not involve any oncologic concerns and therefore a large amount of myocutaneous tissue was included in the flap. Thus, our study is the first to show that a pedicled total leg fillet flap with preserved femoral vasculature may be applied for closure after hip disarticulation with equivalent outcomes to other methods of complex closure of hip defects.

Conclusion

In cases of hip disarticulation where extensive soft tissue compromise forbids closure with a local flap, consideration for salvage should include preservation of the femoral vessels and performing a pedicled fillet soft tissue flap from the leg.

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