Case report

Utilizing preoperative arterial embolization to minimize blood loss at time of vulvar sarcoma resection: A case report

Vera Schulte a,*, Christianne Persenaire a, Marisa R. Moroney b, Saketh R. Guntupalli b

a University of Colorado School of Medicine, Aurora, CO, United States
b Department of Obstetrics and Gynecology, Division of Gynecologic Oncology, University of Colorado School of Medicine, Aurora, CO, United States

ARTICLE INFO

Keywords:
Embolization
Vulvar cancer
Sarcoma
Quality of life
Surgical morbidity

ABSTRACT

Selective arterial embolization has long been utilized in the field of obstetrics and gynecology as both a prophylactic and therapeutic measure. We present a case of a highly vascularized, poorly differentiated, radiation-related vulvar sarcoma that was resected immediately following selective arterial embolization. Arterial embolization in patients with gynecologic malignancies has been shown to compromise blood supply to the tumor and decrease the risk of hemorrhage and related surgical complications. In this case, the use of embolization prior to surgery appeared to result in a less morbid procedure for the patient with decreased blood loss and improved quality of life thereafter. Based on the presented case, we believe that arterial embolization can be considered a safe preoperative intervention to decrease surgical risk, particularly hemorrhage, at time of resection of highly vascularized vulvar tumors.

1. Introduction

Vulvar cancers comprise approximately five percent of gynecologic malignancies in the United States and afflict approximately 2.6 per 100,000 American women (U.S. Cancer Statistics Working Group, 2022). In its early stages, vulvar carcinoma can be treated surgically, with consideration of adjuvant radiation therapy (RT) pending pathologic evaluation and resection margins. Later-stage vulvar cancers may also be treated surgically with radical vulvectomy or pelvic exenteration, or with chemoradiation versus systemic chemotherapy in the case of unresectable disease.

Use of pelvic radiation, while a critical component of the treatment paradigm, is not without risks. The most cited include localized toxicities, such as radiation proctitis and cystitis, as well as the long-term sequelae of fibrosis and stenosis; more rarely, use of therapeutic radiation can induce malignant neoplasms such as sarcomas, which arise from mesenchymal tissues within the irradiated field, including those of the vulva (Fiset et al., 2014). Risk of these radiation-associated neoplasms increases with escalating radiation dose, younger age at initial exposure, and increased duration of the post-radiation period (Gladddy et al., 2010). While clinically similar to spontaneous vulvar sarcomas, which account for only 1–3% of all vulvar cancers, radiation-induced vulvar sarcomas are associated with significantly lower disease free-survival, and median overall survival is only 12 months (Fiset et al., 2014; Gladddy et al., 2010).

Several case reports in the literature have described malignant vulvar lesions attributable to radiation. A 2001 case report out of the Netherlands presented two different cases of sarcoma following primary treatment of vulvar cancer, the first a case of angiosarcoma, and the second a case of fibrosarcoma; at that time, it was thought to be the first description of a radiation-induced sarcoma arising in association with a genital tract malignancy (Hyde et al., 2001).

Given the relative rarity of these often morbid and aggressive entities, a standardized treatment plan has not been developed and there is little to guide the care of affected patients. This case report highlights the use of vascular embolization prior to surgical resection of a large, radiation-related sarcoma to minimize blood loss and perioperative morbidity.

2. Case

We present the case of a 74-year-old with a history of recurrent vulvar squamous cell carcinoma (SCC) who presented with a rapidly enlarging, exophytic vulvar mass.

The patient was initially diagnosed in 2014 with SCC of unknown origin after right groin swelling prompted a right inguinal lymph node...
dissection; subsequent bilateral pelvic lymphadenectomy, colposcopy, loop excisional surgical procedure, and anoscopy were negative for disease, and positron emission computed tomography (PET/CT) did not show any sites of FDG-avidity. She initially declined adjuvant pelvic radiotherapy, and in 2016, was found to have a right vulvar lesion that prompted radical vulvectomy followed by adjuvant radiation (4500 Gy over 25 fractions, followed by 1400 cGy boost for 7 additional fractions). In 2017 and 2018, she had two additional recurrences that were each treated with radical resection, and she was without evidence of disease until April 2019, when she presented with a new lesion on the right labium majus.

The labial lesion was biopsied, revealing poorly-differentiated sarcoma with surface ulceration, and she underwent repeat radical right vulvectomy with myocutaneous gracilis flap. Final pathology was notable for both multifocal recurrent SCC and high-grade undifferentiated sarcoma with positive margins; immunohistochemical staining of the sarcoma was patchy positivity for muscle specific actin (MSA) and calponin, supporting smooth muscle differentiation. The tumor also demonstrated high-level amplification of c-myc, a finding strongly associated with post-radiation sarcomas, and the patient was started on pembrolizumab, as her tumor was noted to be PD1+. The patient underwent several months of treatment with pembrolizumab. Then, in September 2020, had a biopsy-proven recurrence of invasive squamous cell carcinoma. At this point, the option of hospice care was presented to the patient and discussed in detail. The patient elected to proceed with treatment and was started on cisplatin/paclitaxel/bevacizumab (GOG 240). She discontinued this regimen after five cycles due to poor tolerability.

In April 2021, she underwent radical left vulvectomy with pedicled myocutaneous gracilis flap complicated by wound infection and breakdown necessitating short interval debridement and repair. Six weeks thereafter, she was admitted with vulvar bleeding and drainage and was found to have a $14 \times 15 \text{ cm}$ friable, fungating vulvar mass arising from the right labial reconstruction. The lesion was biopsied, notably with significant blood loss, and she was started on palliative doxorubicin and radiation. Biopsy revealed pleomorphic sarcoma with evidence of osteosarcomatous differentiation. The patient was highly symptomatic of this mass, as it left her unable to walk and precluded her from completing her activities of daily living (ADLs). Palliative care met with the patient to rediscuss goals of care and quality of life measures. The patient was presented with options including transition to hospice care, directed chemotherapy, or surgical resection. She chose to proceed with a palliative resection.

Given the potential for significant hemorrhage, computed tomography (CT) angiography of the pelvis was obtained and revealed a large arterial supply to the mass originating predominantly from branches of the deep femoral artery. Based on these findings, the patient was preemptively transfused two units of packed red blood cells, then taken to the interventional radiology (IR) suite, where she underwent image-guided particle embolectomy of the tumor feeder vessels with polyvinyl alcohol (PVA); these vessels were noted to arise from the profunda femoris, right circumflex femoral artery, and the right and left internal pudendal arteries. She was then taken to the operating room, where she underwent uncomplicated resection of what by then was a $20 \text{ cm}$ vulvar mass. Intraoperatively, a vessel sealing device was used to achieve better hemostasis, and the total estimated blood loss (EBL) was 1200 mL. She received one additional unit of packed red blood cells while in the operating room. Due to resection of the urethra, a suprapubic catheter was placed at the time of the operation.

The patient’s postoperative course was uncomplicated, and she was discharged home on post-operative day nine with significant improvement in her mobility and ability to perform ADLs. The patient elected to proceed with two more cycles of chemotherapy post-operatively, eventually transitioned to hospice care, and passed away two months thereafter.

3. Discussion

Arterial embolization has a range of practical uses in the field of obstetrics and gynecology. For example, uterine artery embolization (UAE) is often utilized in the management of postpartum hemorrhage, uterine arteriovenous malformations, and both prophylactically and therapeutically in the care of patients with benign leiomyomas. In studies of the latter, this intervention has been found to be 82–92 % effective at controlling bleeding and minimizing morbidity at the time of fibroid-related surgery (Badawy et al., 2001). In patients with placenta accreta spectrum disorder, UAE has similarly been used to lessen blood loss at time of delivery and planned peripartum hysterectomy. Benefits of embolization in these settings include low complication rates, avoidance of surgery, and fertility preservation (when desired and where feasible), as well as decreased blood loss and rates of transfusion.

In gynecologic oncology specifically, applications of selective artery embolization have been similarly useful. Since the 1970s, embolization has been to control hemorrhage of pelvic malignancies, particularly in the setting of locally advanced cervical cancer. A small case series of arterial embolization in cervical carcinomas found that the procedure was 100 percent successful in quickly controlling an active hemorrhage (Alméciga et al., 2020). Nevertheless, despite the efficacy of embolization in these settings, limitations of the procedure exist, and complications do and can occur; these are most often related to distortion of normal anatomy, tumor invasion of larger arteries, and/or post-radiation adhesions and scarring (Josephs, 2008).

Compared to other pelvic malignancies, data for the use of embolization in the management of vulvar cancers is limited. A case study from 2020 used selective arteriography and embolization in a patient with vulvar SCC and found that this technique led to ischemia and spontaneous separation of the exophytic mass from the surrounding labia, thus decreasing the need for a more morbid intervention (Zhao and Xue, 2020). However, there is a paucity of data to guide the use of this modality in the treatment of vulvar masses, and the literature is even more scarce when it comes to its potential utility in the care of patients with vulvar sarcomas.

Given the perceived benefit of preoperative arterial embolization in this case of a highly-vascularized, radiation-related vulvar sarcoma, we believe that this intervention can be considered as a means of decreasing morbidity associated with surgical resection of vulvar sarcomas.

4. Conclusion

While selective arterial embolization has long been used in the care of patients with a wide variety of gynecologic malignancies, its utility in minimizing perioperative morbidity and mortality in the resection of hypervascular vulvar lesions, specifically radiation-induced vulvar sarcomas, has not been well-established. This case report offers insight into the efficiency and effectiveness of embolization in minimizing the surgical risk associated with the resection of large, vascular vulvar tumors; however, it is clear more research is needed to determine the optimal approach to the care of these patients.

Author contributions

V.S. and C.P. performed literature review and drafted the manuscript in consultation with M.M. and S.G. S.G. supervised the project.

Declaration of Competing Interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.
Appendix A. Supplementary material

Supplementary data to this article can be found online at https://doi.org/10.1016/j.gore.2022.101089.

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