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Case report

Long-term survival after anterior pelvic exenteration and total vaginectomy for recurrent endometrial carcinoma with metastatic inguinal nodes at the time of surgery

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1. Introduction

The pelvic exenteration (PE) was first described by Alexander Brunschwig in 1948 as a palliative procedure for patients with recurrent pelvic malignancy and has since evolved into a potentially curative treatment (Brunschwig, 1948). It is most commonly performed on patients with centrally recurrent cervical cancer, and also on patients with endometrial cancer. It is used less commonly for vulvar, vaginal, and ovarian cancers (Park et al., 2007). Although Brunschwig's initial mortality rate of 23% has improved to approximately 0–7% in modern case series, postoperative morbidity remains high, with complication rates ranging from 22 to 78% (Park et al., 2007; Marnitz et al., 2006; Westin et al., 2014; McLean et al., 2011; Fleisch et al., 2007; Baiocchi et al., 2012; Petruzziello et al., 2014). This fact highlights the importance of rigorous patient selection for PE. The traditional candidate for PE is a patient with a recurrent gynecologic malignancy confined to the central true pelvis without distant foci of disease. While many providers maintain these selection criteria, novel techniques such as the laterally extended endopelvic resection (LEER) are expanding the population of patients who benefit from radical salvage surgery, and studies have demonstrated comparable morbidity, mortality, and 5-year overall survival (Hockel, 2008). There are little data on outcomes of patients who undergo PE with foci of disease outside of the pelvic/paraaortic lymph nodes or fixation to the pelvic sidewall, as this is commonly thought to be a contraindication to exenteration. We present a case of long-term survival after PE for recurrent endometrial cancer in which a metastatic inguinal lymph node was resected at the time of the PE.

2. Case report

A 48-year-old G6P2042 woman was diagnosed with poorly differentiated endometrial carcinoma by endometrial curettage after presenting with heavy menstrual bleeding and severe abdominal cramps. She then underwent a laparoscopic-assisted vaginal hysterectomy, bilateral salpingo-oophorectomy, pelvic and para-aortic lymph node dissection, and peritoneal washings. Pathology revealed an International Federation of Gynecology and Obstetrics grade 3 endometrioid endometrial adenocarcinoma with focal squamous differentiation. Pathology also revealed 37.5 out of 38 mm of myometrial invasion, positive lymph-vascular space invasion, and positive washings. Bilateral pelvic (3 of 36 sampled) and para-aortic nodes (5 of 10 sampled) were positive for metastatic carcinoma. There was no adnexal, endocervical, or vaginal involvement. She received six cycles of adjuvant paclitaxel and carboplatin chemotherapy, as well as radiation therapy with 4500 cGy to the para-aortic lymph nodes and 5040 cGy to the pelvis. Extended field whole pelvic radiation was chosen based on pathologically confirmed metastatic disease, and the decision was made not to give vaginal brachytherapy given that she was to receive whole pelvic radiation.

Approximately 17 months after initial treatment, the patient was found to have a vaginal recurrence diagnosed on biopsy of a vaginal mass. Pathology revealed recurrent endometrioid adenocarcinoma with extensive squamous metaplasia. She underwent a wide local excision with inability to achieve negative margins. A subsequent vaginal Pap smear showed a high-grade squamous intraepithelial lesion, and a biopsy from the peri-urethral area demonstrated metastatic adenocarcinoma. At this time, she was offered and agreed to a PE. Preoperative evaluation with positron emission tomography/computed tomography (PET/CT) demonstrated increased fluoro-2-deoxyglucose (FDG) uptake at the vaginal cuff consistent with known metastasis and a new

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hypermetabolic left inguinal node measuring 1.8 × 2.6 cm. Twenty months after her initial staging procedure she underwent an anterior PE with continent cutaneous urinary diversion, pelvic reconstruction, and left groin lymph node dissection. Anterior PE was performed using a previously published technique involving mobilization and excision of the bladder, entire vaginal tube, urethra, and anterior levator ani muscles via an abdominal and perineal phase (Andikyan et al., 2012). A plastic surgery team then used bilateral myocutaneous gracilis flaps to reconstruct the pelvic floor with creation of a neovagina. Final pathology revealed metastatic adenocarcinoma involving the vagina, with negative surgicopathologic margins and metastasis to one resected left inguinal node. Postoperatively, she received four cycles of adjuvant carboplatin and gemcitabine. She has been followed with surveillance PET/CT scans and remains without evidence of disease on her most recent imaging 10 years after her PE.

3. Discussion

Our case report demonstrates that the historically strict selection criteria of patients appropriate for PE need not exclude select patients who on an individual basis may benefit from the procedure. There is a paucity of data on patient outcomes after PE for patients with extra-pelvic disease to guide management. In our extensive review of the literature, we identified one other case report of a PE performed in a patient with known extra-pelvic disease from Taiwan (Wu et al., 2004). In that case, a patient with recurrent endometrial carcinoma with previously resected inguinal lymph nodes and previously irradiated neck lymph nodes was offered a PE after an isolated central recurrence 10 months after her last remission. She remained disease free 5 years after treatment.

Most of the current literature on morbidity/mortality and predictors of long-term survival following PE are institutional reviews that span up to 2 decades in patient selection. The rarity of the procedure and lack of prospective and randomized studies leave many patient selection and management questions unanswered. The ability to achieve negative surgicopathologic margins is uniformly considered a necessity to consider PE as curative procedure. PET/CT has shown high sensitivity (100%) and specificity (73%) in preparation for exenteration to define the extent of disease and exclude those with unresectable disease (Husain et al., 2007). In our case, one small easily resectable focus of disease was discovered by PET/CT, which was amenable to complete resection.

Identifying predictors of long-term disease-free survival is crucial for patient selection in the face of a known morbid procedure. However, reported risks and predictors of good outcome vary widely and are often contradictory. A greater interval between diagnosis and recurrence is thought to confer a survival advantage. One institutional review reported an 83% 5-year survival rate in those who had a PE for recurrence >5 years after initial diagnosis, and another reported poorer overall survival (8 months vs 33 months) among those with a disease-free interval <2 years (Marnitz et al., 2006; McLean et al., 2011). A review of 160 cases of pelvic exenteration at MD Anderson identified positive pelvic and para-aortic lymph nodes, lymph-vascular space invasion, and positive margins as poor predictors of recurrence-free survival (Westin et al., 2014). In a single surgeon case review, only tumor size >4 cm was associated with recurrence after surgery, although margin status and positive nodes negatively affected overall survival. The most consistently reported risk factor for poor survival after PE for recurrent disease is the presence of positive pelvic or para-aortic lymph nodes. In a separate review of 448 PEs at MD Anderson, 10.9% of patients had positive nodes, and those with positive nodes had about half the 3- and 5-year survival rates as those with negative nodes (36% vs 64% at 3 years, 26% vs 57% at 5 years) (Rutledge and McGuffee, 1987). Some gynecologic oncologists will abort a planned PE upon diagnosis of metastatic pelvic or para-aortic lymph nodes at the time of surgery.

There are still many unanswered questions, even after careful patient selection, with regards to PE. The choice of anterior, posterior, or total PE is highly surgeon dependent and lacks prospective data to guide management. We have previously reported on the excellent results achieved in carefully selected patients with anterior exenteration and total vaginectomy in patients with smaller anterior recurrences (Andikyan et al., 2012). This patient's small, isolated vaginal recurrence without evidence of posterior compartment or pelvic sidewall involvement made her an ideal candidate for an anterior PE. Adjuvant treatment after pelvic exenteration, usually with chemotherapy, can be considered and is feasible (Andikyan et al., 2013). Survival benefit is unproven, and adjuvant treatment is often considered for postoperative patients with high-risk features, including positive margins, positive lymph nodes, and/or lymph-vascular space invasion. In our patient, adjuvant chemotherapy was chosen due to the likelihood that her recurrence represented a systemic process given its location outside the pelvis.

Our patient's long-term survival after PE with a single focus of resectable metastasis outside the pelvis serves as an example of the benefit of individualized treatment planning in patients with recurrent gynecologic malignancy. We caution against a “one size fits all” algorithm for treating recurrent gynecologic malignancy, even in the face of a multifocal recurrence. Disease outside the pelvis is often cited as the single “absolute contraindication” to PE. However, when considered in the context of histology, disease burden, ability to achieve complete resection and the wishes of the patient, extra-pelvic disease need not limit treatment options for the well-informed and appropriately selected patient. Understanding the potential prolonged recovery, high probability of morbidity, and quality of life issues that are prevalent after PE are critical to ensuring proper patient selection for any PE (Dessole et al., 2016).

Informed consent

This patient has provided informed consent for publication of her case.

Conflict of interest statement

The authors have no conflicts of interest to disclose.

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