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ORIGINAL ARTICLE

Ovarian remnant syndrome: a retrospective evaluation of surgical management

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Abstract The objective of the study is to report on patient characteristics, surgical findings, pathology, and recurrence of ovarian remnants. This is a retrospective case series completed at an academic tertiary care hospital. Seventeen patients were identified between September 2005 and December 2015 with ovarian remnant syndrome using a diagnosis code search at one institution. All patients underwent surgical excision. Pathology confirmed ovarian tissue. Three recurrences were treated non-surgically. All patients had a history of endometriosis and previous surgeries. The average number of laparotomies, laparoscopies, and cesarean sections was 1.29 (range, 0–3), 2.47 (range, 0–6), and 0.59 (range, 0–3), respectively. Ten patients (58.8 %) had a prior bilateral salpingoophorectomy. Seven patients (41.2 %) had a prior unilateral salpingoophorectomy. Five patients (29.4 %) had one prior excision; two patients (11.8 %) had two prior excisions of their ovarian remnant. Fifteen excisions were performed laparoscopically and two with planned laparotomy. There were no intraoperative complications. All cases had pathologically confirmed ovarian tissue. Three patients had recurrent disease. Treatments included medical suppression, ovarian artery embolization, and radiation. Surgical expertise, often utilizing minimally invasive techniques, allows for the dissection needed to remove ovarian remnants. Recurrence is possible. Medical or other procedural treatments may be appropriate alternatives or adjuncts to treatment.

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Introduction

Ovarian remnant syndrome (ORS) is a gynecologic diagnosis that occurs after a woman has had a previous oophorectomy. ORS was first described in 1970 by Shemwell and Reed who used a feline model to demonstrate that ovarian specimens are able to re-implant and become functional even after ligation of the blood supply [1].

ORS is, by definition, the presence of histologically confirmed residual ovarian tissue. Historically, it included only patients with prior bilateral salpingoophorectomy. Currently, however, this diagnosis includes patients with unilateral oophorectomy with ovarian tissue noted on the side of previous resection [2, 3]. This residual tissue often causes pelvic pain or a pelvic mass which leads to surgical exploration and excision.

Risk factors associated with the development of ORS include endometriosis, pelvic inflammatory disease, and pelvic adhesive disease [2]. In a 2012 review by Kho and Abrao, it was noted that endometriosis was the most common indication for initial oophorectomy in patients who then went on to have ORS.

The work-up and evaluation of patients presenting with suspected ovarian remnant syndrome include a thorough history and physical exam (paying particular attention to previous intra-abdominal pathology notes during prior surgery), imaging, and consideration of serum FSH and E2 levels. Common radiographic imaging performed includes computed tomography (CT), ultrasound, and/or MRI. We have found that pelvic ultrasound alone is often sufficient to identify a cystic adnexal structure, and CT can be incorporated if unable

to identify structures using ultrasound alone. FSH and E2 levels can play a role in the diagnosis and treatment of ovarian remnant syndrome; however, patients are often presenting with symptomatic adnexal masses that will require surgical intervention independent of the results. One potential benefit can include the avoidance of extensive continued exploration in search of ovarian tissue in patients with no evidence of ovarian function. The laboratory values are of less use in patients with a history of unilateral oophorectomy.

The gold standard of treatment includes surgical excision with confirmation of ovarian tissue on histology [4]. Surgical management was previously performed by laparotomy; however, recently, laparoscopy has been used. Alternative treatments to surgical intervention have also been reported in the literature and include medical suppression and radiation therapy [2]. Although not previously documented, we will also report a case of ovarian artery embolization. In this study, we will present a retrospective review of 17 patients who underwent surgical excision for treatment of ORS over 10 years. We will report on patient demographics, clinical presentation, previous treatments, and surgical and pathologic outcomes, as well as follow-up and recurrence.

Methods

Prior to starting this retrospective study, approval was obtained from the Penn State Hershey Institutional Review Board. Patients were identified by a diagnosis code search at one institution. The diagnosis code is not exclusive to remnants, and the list received was then manually searched for ovarian remnant cases. A total of 17 patients were identified between September 2005 and December 2015. After identification, all patients were included in our study based on inclusion and exclusion criteria.

Inclusion criteria included female patients, over the age of 18, with a prior bilateral salpingoophorectomy or unilateral oophorectomy with recurrence on the same side as previous excision. These patients also had clinical symptoms, such as pain, suspicious for an ovarian remnant, and/or a pelvic/adnexal mass on imaging. Finally, pathologic confirmation of ovarian tissue in the surgical specimen was noted for all included patients. If pathology did not identify ovarian tissue, the patient would have been excluded, although this did not occur for any of our patients.

A retrospective review of the patients' medical records was completed. Informed consent was not obtained as this was a retrospective study. We obtained patient characteristics including age, body mass index, previous surgical history, previous history of endometriosis, presenting symptoms, pre-operative imaging, hormone profiles, mode of original oophorectomy, and previous attempts at ovarian remnant treatment or excision.

Data obtained from each operative report included location of the ovarian remnant, estimated blood loss, intraoperative complications, and operating time. At our institute, we do not follow a strict surgical protocol as first described by Webb in 1989 [5]. This surgical approach included high ligation of the gonadal vessels above the level of the pelvic brim, ureterolysis to the level of entrance into the bladder, ligation of the anterior division of the internal iliac artery, bilateral stripping and excision of the pelvic sidewall peritoneum, and wide excision of the tissue surrounding the remnant ovary. In contrast to this method, we tailor each surgery to the patient's specific situation. It is altered based on site of recurrence and procedure necessary to complete the surgery. With this, we are able to avoid extensive dissection in some areas that may cause subsequent problems. An example of this is an ovarian remnant located at the pelvic brim. Complete ureterolysis to the bladder entrance would not be necessary, and this potentially decreases the risk of devascularizton that comes with complete dissection. Final pathology was reviewed and reported. Postoperative follow-up, including complications, was also obtained from the medical records.

Findings

Seventeen patients underwent surgery for an ovarian remnant between September 2005 and December 2015 in the Division of Minimally Invasive Gynecologic Surgery at Penn State Milton S Hershey Medical Center. Patient demographics can be found in Table 1. Patients' ages ranged from 30 to 54 years with a mean of 37.94 years. Body mass index (BMI) ranged from 20.25–41.10 kg/m^2 with a mean of 32.79 kg/m^2.

Table 1 Patient characteristics

	Mean	Range
Age (years)	37.94	30–54
BMI (kg/m^2)	32.79	20.25–41.10
Number of cesarean sections	0.59	0–3
Time to presentation (months)	33.23	5–132
	N = 17(%)	
History of BSO, performed by:		
Laparotomy	3 (17.6)	
Laparoscopy	5 (29.4)	
Laparotomy and laparoscopy	2 (11.8)	
History of USO, performed by:		
Laparotomy	3 (17.6)	
Laparoscopy	4 (23.5)	
History of prior ovarian remnant excision		
1 excision	5 (29.4)	
2 excisions	2 (11.8)	

All patients in this study had a history of endometriosis. All patients had multiple previous surgeries by both laparoscopy and laparotomy. The average number of prior exploratory laparotomies was 1.29 (range, 0–3). The average number of prior laparoscopies was 2.47 (range, 0–6). These numbers do not include cesarean sections. The average number of prior cesarean sections was 0.59 (range, 0–3).

Of the 17 patients, ten patients (58.8 %) had prior bilateral salpingoophorectomies (BSO). Of the patients who had BSO, three (17.6 %) were completed by prior laparotomy, five (29.4 %) by prior laparoscopy, and two (11.8 %) with completion in staged procedures including first unilateral salpingoophorectomy (USO) by laparotomy, followed by a second USO by laparoscopy. Seven patients (41.2 %) had prior USO with the ovarian remnant occurring on the side of prior excision. Of the patients with prior USO, three (17.6 %) were completed by laparotomy and four (23.5 %) were completed by laparoscopy. Four of these seven patients went on to have removal of the contralateral ovary at the time of their remnant excision.

The time from initial oophorectomy to presentation of ovarian remnant averaged 33.23 months (range 5–132 months). One patient was medically treated with Lupron prior to her surgical management. She discontinued medical management secondary to return of pain despite continued treatment. Five patients (29.4 %) had one prior attempt at surgical excision for their ovarian remnant. Two patients (11.8 %) had two prior surgical excisions. All prior excisions were confirmed to have ovarian tissue on pathology.

Prior to surgical excision, all patients underwent or presented to their consultation with imaging. CT scan was completed in four patients (23.5 %). Transvaginal ultrasound was completed in 13 (76.5 %). Sixteen of the 17 patients (94.1 %) had an adnexal mass or cystic structure confirmed on imaging. One CT scan was negative. However, this patient had cyclic symptoms consistent with an ovarian remnant. We did not routinely obtain pre-operative FSH on patients with previous BSO as they all presented with pain requiring intervention, and this lab value would not be useful in patients with previous USO.

Of the 17 cases, 15 cases were performed laparoscopically. Two cases were completed by planned exploratory laparotomy secondary to a history of multiple laparotomies in both patients and a history of necrotizing fasciitis complicating the second patient. At the time of surgery, 11 remnants (64.7 %) were found on the left side. Five (29.4 %) were found on the right side of the pelvis. One remnant was found attached to the omentum. This remnant was seen on pre-operative imaging and noted to be located in the region of the left adnexa; however, the side walls were clear bilaterally. To ensure there was no ovarian tissue missed, the left colon was dissected from the sidewall and the left retroperitoneum was inspected. We did not perform a strict surgical protocol, as each case was tailored to the specific patient. However, in all but one of the cases of left-sided remnants, the descending colon was mobilized, the retroperitoneal space was entered, and the remnant was revealed near the pelvic brim. The one exception to this was a remnant identified on the left at the level of the vaginal cuff. Right-sided remnants were found at varying locations along the sidewall. They were located in the retroperitoneal space, near the lateral aspect of the vaginal cuff, along the ureter, in the infra-ovarian fossa, and at the pelvic brim.

There were no intraoperative complications such as bowel or genitourinary injuries. Ureteral stents were not placed during any of these procedures. Mean estimated blood loss was 50 ml (range 5–100). Mean surgical time was 90.7 min (range 44–264). Surgical time was recorded from skin incision to skin closure. All cases had pathologically confirmed ovarian tissue.

Post-operatively, three patients required treatment of a recurrence. One patient was initiated on a course of leuprolide and is now pain free per current medical record review. A second patient experienced a post-operative complication of a pulmonary embolism and treated with anticoagulation. She then had recurrence of her symptoms and a pelvic mass seen on imaging. In some circumstances, radiation therapy has been used to manage ovarian remnants [6]. This treatment does, however, have side effects including pelvic adhesion formation, enteritis, and colitis [2]. After thorough counseling of the risks and benefits involved with radiation verse additional surgery, our patient opted to undergo radiation treatment. Her ovarian remnant was treated with a total of 20 Gy in ten fractions. She had follow-up imaging using CT scan which showed complete resolution of the soft tissue mass in her left pelvis. The third patient also declined further surgical intervention and underwent left ovarian artery coil embolization. Post procedure, she has been followed by CT angiogram. The volume of her left-sided remnant has progressively decreased from 10.5 to 2 cm^3 over the course of the 4-month follow-up. We are continuing to follow her progress.

Conclusion

Ovarian remnant syndrome is a diagnosis that proves to have a difficult treatment course for both patients and physicians. Our patient population all presented with symptoms of pain and had a history complicated by endometriosis and multiple prior surgeries. Almost half of our patients had at least one prior surgical excision for an ovarian remnant. Predisposing factors leading to ORS include dense pelvic adhesions from multiple prior surgeries and pelvic and bowel inflammatory disease. Endometriosis is one of the inflammatory conditions that can lead to increased adhesions and functional ovarian tissue embedding on adjacent tissue [7].

Pre-operative evaluation can include multiple modalities. Although we did not routinely complete pre-operative lab work with our patients, pre-operative FSH may help to confirm the diagnosis prior to proceeding to the operating room in patients with previous BSO. However, functioning ovarian tissue may not produce estradiol levels that are capable of suppressing gonadotropins, and in these cases, FSH >40 does not exclude a diagnosis of ovarian remnant [2]. Imaging has also been noted to help aid in the diagnosis, and in our current patient population, 94.1 % of the patients were found to have a cystic lesion on transvaginal ultrasound or CT scan.

Contributing to the thought that ovarian remnant syndrome is a multifactorial problem, including both pathologic factors and surgical skills, the initial oophorectomy in our subset of patients was performed by both laparotomy and laparoscopy. ORS did not occur after one type of excision alone. In our study, we predominantly used laparoscopy as our treatment of choice. There has been increased literature regarding treatment with a laparoscopic approach over the past several years. Notably, laparoscopic techniques for management and outcomes have been published by Nezhat et al. in 2005 and Arden and Lee in 2010. These studies demonstrated that laparoscopy can produce favorable outcomes when surgically excising ovarian remnants. It is thought that this is due to advanced expertise with laparoscopic instruments over time. If the surgeon has the necessary skills to complete the dissection, the patient will receive all of the benefits of laparoscopy over laparotomy. When laparoscopy is used, whether a strict surgical protocol is followed or dissection is tailored to the patient, it is important to take special care in identifying and restoring anatomy, especially the ureter. Furthermore, complete excision usually requires extensive retroperitoneal dissection to ensure all ovarian tissue is removed.

If surgery is not an option, there has been a small amount of data on alternative treatments. Medical suppression can be achieved by oral contraceptive pills, danazol, gonadotropin releasing hormone agonists, and progestins [7]. Many patients, however, do not respond well to these treatments. We have limited, but satisfactory experience with medical management, as only one patient was treated with a gonadotropin releasing hormone agonist.

Scant data is available on alternatives such as radiation therapy. A case report by Haglund reports on three patients with imaging evidence of ovarian remnants. They were not candidates for further medical or surgical interventions and were successfully treated with external beam radiation. We have treated one patient who was no longer a surgical candidate. She now has no radiologic findings of an ovarian remnant and improved pain. Prior to her radiation treatment, she did have tissue diagnosis from her previous excisional procedure that ruled out a malignant process.

A novel management technique that was used at our institute is ovarian artery embolization. Coil embolization has been used with success to treat pelvic congestion syndrome and fibroids [8]. With this in mind, it may treat the pain and decrease the size of the ovarian remnant. The patient treated with this method had three attempts at excision of her ovarian remnant, with histologic confirmation of benign ovarian tissue at each. After coil embolization, her remnant has continued to decrease in size over a 4-month follow-up. We plan to continue to follow her results and report on this in greater detail. Further information is needed to determine long-term success, failures, and complications of this technique.

Recurrence or persistence of an ovarian remnant is possible. Three out of our 17 patients (17.6 %) had continued adnexal mass and pain. These patients are the ones who went on to subsequent medical treatment. This is consistent with a recurrence risk between 8 and 25 % [2].

Our small series of patients adds to the literature that a laparoscopic approach at ovarian remnant excision is feasible and safe. Surgical expertise is needed to complete the dissection needed to remove ovarian remnants. Despite this, recurrence is still possible. In select patients, medical or other procedural treatments may be appropriate alternatives and/or adjuncts to treatment.

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M Pacis: project development and manuscript writing/editing.

G Harkins: project development, data analysis, and manuscript writing/editing.

Compliance with ethical standards

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