

A Case of Ovarian Remnant Syndrome

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Abstract

Ovarian Remnant Syndrome (ORS) is a possible, though rare, result of incomplete resection of ovarian tissue during oophorectomy. Patients with abdominal adhesions, whether secondary to a history of endometriosis, pelvic inflammatory disease, inflammatory bowel disease, or previous surgical intervention are at particular risk for the development of this disease process. Commonly, patient's present with symptoms that mimic a menstrual cycle. Our case presents a patient with the typical symptomology of ORS, but equally with cystic structures at the vaginal cuff mimicking incarceration of bowel at the vaginal cuff.

Keywords: Endometriosis; Cyclic pain after oophorectomy; Incomplete ovarian tissue resection; Abdominal adhesions; Cyclic vaginal bleeding after oophorectomy

Introduction

Ovarian Remnant Syndrome (ORS) is defined as the incomplete removal of ovarian tissue in patients with a history of unilateral or bilateral oophorectomy [1]. ORS is a rare condition and its incidence is largely unknown due to most publications consisting solely of case reports/series with few review papers.

The most common risk factor associated with the development of ORS is abdominal adhesions. These are typically secondary to endometriosis, which ironically, is the leading indication for initial oophorectomy. Abdominal adhesions may otherwise form as a consequence of inflammatory bowel disease, pelvic inflammatory disease, or multiple pelvic/abdominal surgical procedures [2,3]. Whichever the indication, surgical intervention with incomplete resection of ovarian tissue can lead to the development of ORS.

This typically presents with symptoms of cyclic pelvic pain, cyclic vaginal bleeding, and other symptoms that mimic a menstrual cycle [4]. Physical examination alone has not been found to be adequate for diagnosis. Serum levels of FSH and estradiol, along with imaging may be of aid. Often, the diagnosis is difficult to achieve [5]. Treatment of endometriosis is the most common indication for oophorectomy [3].

Case

The case is of a 40 year-old surgically-induced menopausal female who presented for evaluation of pelvic pain and vaginal bleeding since bilateral salpingo-oophorectomy 8 months prior. According to the patient, she began experiencing recurrent vaginal bleeding and pelvic pain just 6 weeks after

her surgical procedure that had initially been performed for similar complaints. She did have a notable gynecological history of known endometriosis for which the patient had undergone abdominal hysterectomy 10 years prior. Her pelvic pain was cyclic in nature, worsened with vaginal exams, and unrelieved with defecation. The patient did admit to constipation, with bowel movements occurring every two to three days. She denied currently being sexually active and declined any sexual activity since her surgery.

The patient had previously presented to the Emergency Department for vaginal bleeding. An examination confirmed an intact vaginal cuff with small amount of bleeding. Review of systems and physical exam were otherwise negative. She then presented to her gynecologist for increased pelvic pain. A transvaginal ultrasound demonstrated cyst-like structures just superiorly adjoining the vaginal cuff. A repeat pelvic examination with a speculum was worrisome for a vaginal cuff dehiscence with cystic structures possibly representing an incarcerated bowel loop. She was then urgently scheduled for an exploratory laparotomy for further evaluation.

Intra-operatively, dense adhesions were encountered while entering the abdomen. Upon dissection, the bowel was easily moved from the vaginal cuff. With visualization of the cuff itself, there were multiple cystic structures adherent to the vaginal apex. The cysts appeared simple in nature, and measuring ~1cm in diameter. These were resected and sent to pathology for further assessment. Frozen pathology results demonstrated chronic granulation tissue with a small amount of ovarian tissue identified. Final pathology showed evidence of ovarian follicular cysts without endometriosis. The patient had an uncomplicated post-operative course and was discharged after two days in the hospital.

Conclusion

A patient presenting with menstrual-like symptoms (as opposed to menopausal symptoms) following surgically-induced menopause should prompt evaluation for ovarian remnant syndrome, especially when she has known risk factors for the development of the disease such as endometriosis and multiple abdominal surgeries.

Endometriosis is the most common indication for oophorectomy [2]. This disease is a known risk factor for the development of abdominal adhesions. Other associations with the development of abdominal adhesions include pelvic inflammatory disease, inflammatory bowel disease, multiple pelvic or abdominal surgical procedures [3]. Incomplete resection of ovarian tissue can lead to remnants of ovaries being left behind. Poor surgical technique for conducting blunt dissection of fibrotic ovarian adhesions to peritoneum or pelvic viscera has also been indicated as a factor in developing ORS [3].

The most recent review paper identified six published case series on ORS since 2006 [2]. Most patients were diagnosed within five years of their oophorectomy and each of these studies reported pelvic pain as the most common initial presentation [2]. In the largest published report describing 186 patients with ORS, *Magtibay et al.* [4], found that patients most commonly presented with chronic pelvic pain (84%), pelvic mass (66%), dyspareunia (26%), cyclic pelvic pain (9%), cyclic vaginal bleeding (8%), dysuria (7%), and painful defecation (6%).

Although postmenopausal serum hormone levels do not exclude ORS diagnosis, ovarian remnant is more likely in suspected patients with Follicle-Stimulating Hormone (FSH) levels <35 IU/dL and estradiol levels >35 pg/mL post oophorectomy [3]. As such, women presenting with symptoms concerning for ORS should have laboratory measurement of her hormones. This should be emphasized because some hormone replacement therapy may be pre-emptively initiated shortly after oophorectomy to prevent menopausal symptoms. However, when ovarian tissue remains and hormone replacement is added, patient can receive supraphysiologic levels of estrogen, which could inherently increase the patient's risk of hypercoagulability, gynecological and non-gynecological cancers, among others. As such, it is imperative to ensure that all ovarian tissue be respected and patient undergo confirmation, either clinically or serologically, of surgically-induced menopause prior to introduction of hormone replenishment.

Other diagnostic tools including imaging such as pelvic ultrasound and less commonly, computed tomography and magnetic resonance imaging, have been shown useful in demonstrating a pelvic mass to aid in diagnosing ORS [2]. Definitive diagnosis of ORS requires histological confirmation of ovarian tissue. Most (42%) ovarian remnants comprises of corpus luteum tissue histologically [4]. Definitive treatment requires complete surgical excision of ovarian implants. This has been associated with a high incidence of complications, such as bowel or bladder injury [6].

While our patient had a presentation that could have represented ovarian remnant syndrome, her physical examination findings of possible bowel incarceration at the vaginal cuff prompted urgent surgical intervention. It was only during this time that a diagnosis was made. Otherwise, the patient could have had persistent symptoms and never received an appropriate diagnosis.

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