

A Rare Case of a Left Ovarian Remnant Syndrome Associated with a Symptomatic Right Ovarian Cyst

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Abstract

Ovarian Remnant syndrome (ORS) is the presence of residual ovarian tissue after an oophorectomy was performed whether associated with a hysterectomy or not. We report the case of a 39-year-old woman with a past surgical history of total abdominal hysterectomy and left salpingo-oophorectomy with an indication of placenta accreta incidentally discovered during a caesarian section. The patient presented with pelvic pain and was diagnosed with ORS. She was successfully managed by laparoscopy with removal of the cyst and the surrounding ovarian tissue, confirmed by histopathological analysis. The post operative period was uneventful, and the patient was discharged after 2 days of good evolution. In our context, surgery remains the main treatment option, however, other treatment options including radiotherapy or medical management need to be considered as either adjunct or main therapy.

Keywords

Pelvic Pain, Ovarian Remnant Syndrome, Laparoscopy

1. Introduction

Ovarian Remnant Syndrome is a rare condition occurring in women who have undergone unilateral or bilateral salpingo-oophorectomy with or without hysterectomy, resulting from the persistence of ovarian tissue left unintentionally after easy or difficult ovarian resection and which becomes functional and/or dystrophic [1]. In other words, Ovarian Remnant Syndrome is the presence of histopathological proven residual ovarian tissue. At first this syndrome referred to only patients who had undergone bilateral oophorectomy, but with time and

more recent studies includes patients with unilateral resection with residual tissues confirmed on the side of the previous resection, ovarian remnants can re-implant themselves anywhere in the abdominal cavity, including the bladder, bowel and ureters [2] [3]. This syndrome was first described in 1970 by Shemwell and Weed [4] and the first case presented in the literature was in 1962 by Kaufmann [5].

This residual ovarian tissue can grow, form cysts or hemorrhage, may be asymptomatic or cause pelvic pain. Risk factors associated with incomplete removal of an ovary and the subsequent development of residual ovarian syndrome include a history of endometriosis, pelvic inflammatory disease, multiple previous surgeries and multiple pelvic adhesions [6]. The recommended treatment is surgical removal by laparotomy or laparoscopy [1] with laparoscopy being more used nowadays. We report a rare case of a left Ovarian Remnant Syndrome associated with a symptomatic right ovarian cyst successfully treated by laparoscopy with uneventful post operative course.

2. Case Report

We report the case of an ovarian Remnant syndrome in a 39-year-old patient with a history of myomectomy by laparotomy, three caesarean sections, a total hysterectomy with left unilateral oophorectomy indicated for placenta accreta incidentally discovered during the last caesarean section and a laparotomy indicated for bowel obstruction. She presented with pain in the right iliac fossa of progressive onset, intermittent, with no relieving nor aggravating factors, evolving over 7 days before her admission, associated with nausea and profuse sweating, relieved by analgesics. The physical examination on admission revealed a renitent pelvic mass in the right iliac fossa of approximately 80 mm in diameter. She had a computerized tomography (CT) scan of the abdomen and pelvis which revealed supra-bladder fluid formation with a thick wall (5 mm), oval in shape measuring $77 \times 71 \times 69$ mm with intense peripheral enhancement, medium-abundance pelvic fluid effusion and left oval ovarian formation with fine and regular wall measuring $34 \times 36 \times 41$ mm not raised (**Figure 1**, **Figure 2**). A laparocystectomy was indicated and the intraoperative findings were a right ovary with a right hemorrhagic cyst. The left ovary was not visualized but we had

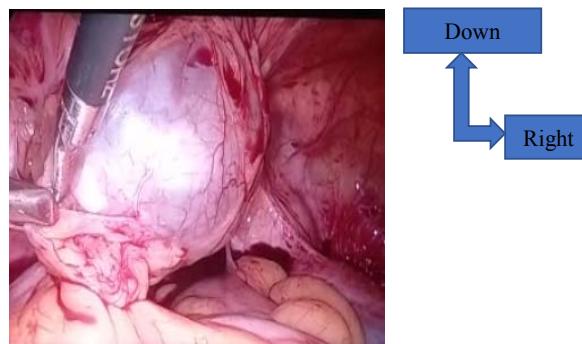


Figure 1. Left parietal cyst.

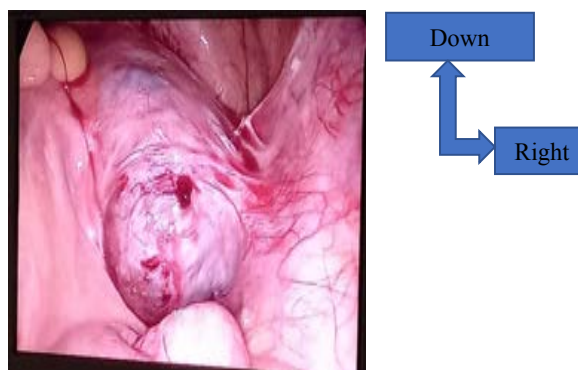


Figure 2. Right ovarian cyst.

a cyst of the anterior abdominal wall which appeared simple surrounded with ovarian tissue and measuring about 4 cm in diameter. The management consisted of the complete removal of the cyst and the surgical specimen sent for anatomicopathological examination confirming the presence of ovarian tissue on the left specimen. A cystectomy was performed on the right side. The post-operative follow-up was unremarkable, and the patient was discharged after 2 days of favorable evolution.

3. Discussion

The diagnosis of residual ovarian syndrome begins with a clinical suspicion obtained through a careful history and knowledge of the syndrome itself. It is supported by a thorough pelvic examination, imaging investigations and laboratory tests [1]. Dense peri-ovarian adhesions and ovarian enlargement can make it difficult to identify ovarian tissue, and they are considered predisposing factors for this disease. These two risk factors are present in severe endometriosis, and it was often found to be the primary indication for initial oophorectomy in patients who developed ORS later on in life [5] [6].

Patients with this syndrome most often present with symptoms within the first 5 years after oophorectomy, although some reports have shown that patients whose first surgeries dated back more than 20 years ago [5]. Patients most often present with pelvic pain and less frequently with asymptomatic pelvic masses [5]. In the largest cohort of patients (183), reported by the Mayo Clinic, patients who were treated surgically for this syndrome, presenting symptoms include chronic pelvic pain (84%), dyspareunia (26%), cyclic pelvic pain (9%), dysuria (7%) and tenesmus (6%) [7]. In pre-menopausal women, pain symptoms may be caused by an increase in the volume of residual ovarian tissue which may lead to compression of adjacent structures. Pain can also be due to hormonal stimulation ectopic endometriotic implants. In premenopausal patients in whom oophorectomy is performed unilaterally, the manifestation of pain alone often triggers suspicion [3].

Imaging can be used to aid in diagnosis. Modalities include pelvic ultrasound, CT and MRI. In one study, a complex pelvic mass corresponding to the syn-

drome was identified in 93%, 92% and 78% of cases by ultrasound, CT and MRI respectively [7]. Similarly in the most recent study, 90% of adnexal mass was detected by ultrasound [7] which alone seems to be sufficient to highlight the pelvic mass. It is relevant to mention that FSH and E2 levels can be used for the diagnosis of ovarian remnant syndrome; however, patients are often presenting with symptomatic adnexal masses that will require surgical intervention independent of the results. The laboratory investigations are of less use in patients with a history of unilateral oophorectomy.

The therapeutic options are medical treatment, radiotherapy but the main treatment remains surgery. The pharmacological approach is geared toward suppression of ovarian function and includes oral contraceptives, danazol, and GnRH analogs. A recent study used GnRH in a patient without success, which confirms the limited application of the medical approach [3]. Radiation therapy has also been used as a therapeutic approach. The overall results, however, have been inconsistent. In addition, irradiation without tissue diagnosis has been discouraged due to the discovery of malignancy in residual tissues and potential deleterious effects on adjacent tissues [1] [3] [4]. Surgical removal with histological confirmation of ovarian tissue remains the reference method [4]. Surgical removal is the main treatment and in addition excision of remaining tissue may require retroperitoneal dissection [6], ovarian fragments can be difficult to locate and reappearance of new fragments is possible. Surgery can be performed by laparotomy, laparoscopy or robotics [5]. The minimally invasive approach has been the most frequently used, as shown by recent studies with the aim of reducing the morbidity associated with laparotomy [3] [5] [8] [9]. However, all the authors emphasize the difficulties of this, due to the surgical history and the modification of the anatomical relationships, with a significant risk of visceral complications, involving the digestive and urinary systems, and vascular complications. Price *et al.* [9], in an analysis of the literature, reported a complication rate of around 16% to 30% after a laparotomy. Regarding the approach, laparoscopy can be proposed as first-line treatment, as it reduces the risk of occurrence of postoperative adhesion complications compared to laparotomy [10] [11]. It also leads to lower blood loss and a shorter hospital stay. It has been proven that there are increased risks of a woman with BRCA-1/BRCA-2 mutations developing ovarian cancer over a lifetime ranges from 16% to 54% [3] and in this population if there is a significant risk of developing from the remaining ovarian tissue. If some ovary tissue is left in this population, there is a significant risk of ovarian cancer and clinical treatment is not indicated [3].

In the first series by Nezhat and Nezhat [2] comprising 13 patients treated by laparoscopic surgery, 9 had a complete improvement of their painful symptoms.

4. Conclusion

Ovarian remnant syndrome is a very difficult diagnosis to pose for many physicians and it may go unnoticed in many situations. High risk factors associated to ORS include dense pelvic adhesions from multiple prior surgeries, endometri-

osis and other conditions which favor the appearance of adhesion leading to functional ovarian tissue embedding on adjacent tissue. Surgical excision remains the treatment of choice in ORS as malignancy can be associated with the remnant tissue especially in high-risk patients with BRCA1 and BRCA2 mutations. In many reports literature reports failure in the medical treatment of ORS but in some cases showed to be efficient highlighting the need for research on more therapies.

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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