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Ovarian Remnant Syndrome after Laparoscopic Hysterectomy and Bilateral Salpingo-Oophorectomy for Severe Pelvic Endometriosis

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Abstract

Ovarian remnant syndrome is a rare complication of total abdominal hysterectomy and bilateral salpingo-oophorectomy (BSO). Ovarian enlargement and dense periovarian adhesions are the predisposing factors. Recurrent ovarian remnant syndrome was associated with recurrence of symptomatic endometriosis in a woman who underwent laparoscopic supracervical hysterectomy and BSO for severe endometriosis and extensive pelvic adhesions. After primary surgery she required five additional procedures for complete resection of all ovarian remnants. Definitive surgery for advanced endometriosis with extensive periovarian adhesions may be complicated by ovarian remnant syndrome and reactivation of the disease. Careful retroperitoneal resection of all ovarian tissue is of paramount importance in preventing the syndrome. This, however, may be a limitation of laparoscopic surgery. The choice between laparoscopy and laparotomy in such cases should be individualized and based on the degree of surgical difficulty and the surgeon's level of experience.

Several reports describing reactivation of residual ovarian fragments unintentionally left in situ during laparotomy and bilateral oophorectomy, the ovarian remnant syndrome, were published during the 1970s and 1980s.¹⁻⁷ This is a rare condition that is often underdiagnosed and only seldom reported.⁸ Dense periovarian adhesions and ovarian enlargement by cystic lesions are considered the predisposing factors.⁷ Both of these factors are typically present in

severe endometriosis and are the indication for total abdominal hysterectomy and bilateral oophorectomy (TAH-BSO).

If functional ovarian fragments are left in situ in such cases, they may result in recurrence of the disease and related symptoms.⁷ If they are numerous, their surgical localization and removal may be quite difficult and require several procedures. In addition to recurrent endometriosis and pelvic pain, ovarian

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remnants have been associated with ureteral obstruction,"¹⁰ and ovarian adenocarcinoma.¹¹

Case Report

A 33-year-old nulliparous woman was seen initially because of progressively worsening dysmenorrhea and chronic pelvic pain. Diagnostic laparoscopy revealed stage 4 endometriosis with extensive adhesions and complete obliteration of the cul-de-sac. She was treated with intramuscular depot leuprolide (TAP Pharmaceuticals, Chicago, IL) 3.75 mg monthly for 6 months. During treatment, her estradiol levels were consistently below 20 pg/ml and she became amenorrheic and asymptomatic.

Operative laparoscopy was performed within 3 weeks after the last injection. The procedure consisted of extensive adhesiolysis, neodymium:yttrium-aluminum-garnet laser ablation of peritoneal endometriosis, resection of ovarian endometriomas, and laparoscopic uterosacral nerve ablation. Histologic examination of the surgical specimen confirmed the diagnosis of endometriosis.

After surgery, the patient was prescribed cyclic norethindrone acetate 1 mg plus ethinyl estradiol 20 µg (Loestrin-21; Parke-Davis Pharmaceuticals, Morris Plains, NJ) to suppress endometrial proliferation and to delay the recurrence of the disease. However, she was asymptomatic for only 3 months. The symptoms recurred and became incapacitating, interfering with her employment. Various treatment options were discussed, including definitive TAH-BSO. The woman selected this procedure but opted for a laparoscopic and supracervical approach because it is performed as ambulatory surgery, has a short recovery period, and yields good anatomic results.

At laparoscopic supracervical hysterectomy (SLH) and BSO, both ovaries were enlarged by endometriomas and were adherent to the bowel, uterus, and pelvic sidewall. The left ovary was densely adherent to the rectosigmoid, making dissection between the bowel and the ovary difficult. One month after surgery the patient had an episode of vaginal bleeding associated with recurrence of pelvic pain. The clinical impression was intraperitoneal bleeding from the cervical stump.

Laparoscopic evaluation indicated bleeding from the granulation tissue covering the pelvic portion of the cervical stump and reformation of pelvic adhesions. The granulation tissue was electrocoagulated, and adhesiolysis was performed. Because of the appear-

ance of menopausal symptoms, the woman was given estrogen replacement therapy with micronized oral estradiol 1 mg/day. Two months later she had another episode of vaginal bleeding associated with pelvic pain. Pelvic sonogram demonstrated a right-sided, 30 × 30-mm cystic lesion suggestive of an ovarian cyst and an ovarian remnant. During operative laparoscopy, a cystic ovarian remnant was resected from the right side of the cul-de-sac. Tissue pathology indicated corpus luteum cyst and endometriosis.

The woman did well initially, but then had another episode of vaginal bleeding and pelvic pain. A repeat sonogram 1 month later showed another right-sided, 16 × 34-mm cystic structure indicating reactivation of another ovarian remnant. Another surgical procedure was performed, consisting of vaginal resection of the cervical stump and laparoscopic resection of the ovarian remnant. Again, the histopathologic diagnosis was consistent with corpus luteum cyst and endometriosis of the cervix, and again the patient did well for about 4 weeks, at which time she complained of recurring abdominal pain.

Another pelvic sonogram revealed another ovarian remnant with a 30 × 10-mm cystic follicle at the vaginal apex. With the administration of monthly injections of depot leuprolide 3.75 mg, the follicle disappeared over the next 4 months and the woman improved symptomatically. However, considering the evidence for at least one remaining ovarian remnant, it was decided to perform laparotomy and exploration of the pelvis and retroperitoneal dissection.

At this procedure, the stumps of both ovarian vessels were identified and resected high in the pelvis. The uterus and rectosigmoid were explored, and another ovarian remnant was removed. Histopathology indicated ovarian tissue, endometriosis, and nonpathologic vessels. In approximately 2 months the woman had another episode of pelvic pain, and sonogram revealed a 36 × 16-mm cyst in the cul-de-sac. Serum estradiol level without hormone replacement was 156 pg/ml. During the next 8 weeks the patient was managed symptomatically. She experienced some symptomatic improvement, and the cyst decreased in size to 11 × 12 mm. However, because of persistent pain she went to another city and underwent another operative laparoscopy.

Aggressive retroperitoneal dissection was performed to remove all ovarian tissue. Two corpus luteum cysts were removed; one from the right pelvic brim (5 cm) and the other one (4 cm) from the left side of

the cul-de-sac. For the next 2 years the woman remained symptom free and is currently receiving estrogen replacement therapy without pelvic symptoms.

Discussion

This entire episode was very frustrating to the woman and to her medical team. It appears that at the time of laparoscopic SLH and BSO for severe pelvic endometriosis, numerous ovarian remnants were inadvertently left in situ. They subsequently, and in sequence, became functional, causing several episodes of pelvic pain and reactivation of the disease. This necessitated one laparotomy and four laparoscopic procedures within 15 months. One can only speculate at this point to what extent the initial surgical approach contributed to the outcome, and whether a traditional TAH-BSO by laparotomy would have resulted in more complete ovarian resection.

At the time of hysterectomy, the patient had severe endometriosis, bilateral ovarian endometriomas, and extensive adhesions between the ovaries, uterus, bowel, and pelvic sidewall. Laparoscopic dissection of the ovaries was difficult, and apparently fragments of the adherent ovarian cortex were left in situ. An alternative explanation is that fragments of cut or torn ovarian cortex were not completely removed from the peritoneal cavity through laparoscopic ports, but were lost between the pelvic organs and subsequently implanted in new locations. It was shown in cats that small ovarian fragments under similar circumstances may implant in the peritoneal cavity and become hormonally active.¹

The ovarian cortex in young women contains thousands of microscopic, primordial follicles. Each one of these follicles can respond to gonadotropic stimulation with about a 100-fold increase in size, and produce levels of estrogen and progesterone characteristic of a normal menstrual cycle. It seems unlikely that ovarian remnant syndrome can be completely prevented when a cortex of a massively enlarged ovary is densely adherent to pelvic structures. Our earlier report described seven women with ovarian remnants and recurrent endometriosis after TAH-BSO by laparotomy.⁴ The extent of the disease at the time of hysterectomy in all seven was comparable with or less than that in this woman. Yet after the definitive surgery, three of the seven patients underwent three laparotomies each for sequentially reactivated ovarian remnants and recurring endometriosis.

This case of ovarian remnant syndrome after laparoscopic surgery is in many respects similar to those reported after laparotomy.¹⁻¹¹ The recurrence of pelvic symptoms is typically associated with enlargement of the ovarian fragment and development within the fragment of a hormonally functional follicular or corpus luteum cyst and with reactivation of endometriosis. The diagnosis of ovarian remnant syndrome is facilitated by measuring levels of gonadotropins and estradiol, and by pelvic sonograms. These are frequently omitted in clinical practice, and the symptoms are typically attributed to disease recurrence or to pelvic adhesions.² Inactive ovarian fragments (without ovarian cystic structures) may be difficult to localize at laparotomy or laparoscopy, as demonstrated by our patient, and sequential reappearance of new fragments may be unavoidable. Ovulation-inducing drugs have been successful in stimulating the enlargement of such fragments, which facilitates their localization and subsequent removal.¹²

With increasing popularity of laparoscopic surgery and with increased complexity of the procedures, there will undoubtedly be an increase in the frequency of ovarian remnant syndrome. Laparoscopic dissection of densely adherent structures is more difficult than laparotomy, and because of lack of tactile sensation, may leave behind fragments of the ovarian cortex that otherwise could be palpated and removed.

Nine women had ovarian remnant syndrome after bilateral oophorectomy. Two of them had laparoscopic oophorectomy performed with the endoloop technique to ligate the infundibulopelvic ligament.¹³ The authors suggested electrodesiccation rather than the endoloop procedure to ensure complete removal of ovarian tissue and to eliminate the possibility of leaving remnants. This, however, may not be an adequate solution. In our patient electrocoagulation was used exclusively to obliterate the ovarian vasculature.

This case illustrates the difficulties and potential complications of definitive surgery for severe endometriosis, in which complete resection of all ovarian tissue is of paramount importance. The ovarian remnant syndrome may result from unintentional incomplete resection of the ovaries attached to the rectosigmoid and/or cul-de-sac. To avoid such a possibility, meticulous removal of all fragments of the ovarian cortex, retroperitoneal dissection as in cancer surgery, copious irrigation, better surgical handling of ovarian tissue, and use of surgical specimen bags during laparoscopic procedures may be of value.

It can be argued that advanced endometriosis with extensive periovarian adhesions may not be suitable for laparoscopic resection; however, laparotomy does not guarantee a better outcome. Each procedure has some advantages and some disadvantages. The choice of approach should probably be individualized and based on the degree of surgical difficulty and the surgeon's level of experience. Regardless, whether the surgery is by laparotomy or laparoscopy, resection of all ovarian tissue should be the primary consideration.

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