Ovarian remnant syndrome: a case report and review of the literature

D. Vavilis, A. Loufopoulos, T. Agorastos, M. Vakiani, L. Intzesiloglou, A. Karavida, J. N. Bontis

2nd Department of Obstetrics and Gynecology, Aristotle University, Thessaloniki (Greece)

Summary

The ovarian remnant syndrome in an unusual complication of bilateral oophorectomy, usually presenting with pelvic mass and pain. A case of the syndrome is described in a 35-year-old woman with a history of abdominal hysterectomy and bilateral oophorectomy. We suggest that ovarian remnant syndrome should be considered in the differential diagnosis of chronic pelvic pain after recorded oophorectomy.

Key words: Ovarian remnant syndrome; Chronic pelvic pain; Bilateral oophorectomy

Introduction

Ovarian remnant syndrome is an unusual complication of bilateral oophorectomy, often when the latter is combined with hysterectomy [1]. Diagnosis of ovarian remnant syndrome is consistent with the detection of functioning ovarian tissue in a patient with a history of bilateral oophorectomy [2].

Although the incidence of ovarian remnant syndrome is unknown, it seems that there is an apparent increase in its incidence. This is probably the result of a more widespread use of ultrasonography, computed tomography and magnetic resonance imaging.

Case Report

A 35-year-old woman, para 2, gravida 2, was admitted in our deparment with a history of pelvic pain and a pelvic mass which was found ultrasonographically.

Two years before she had undergone a right salpingo-oophorectomy because of an ovarian mass of 6.5x3.5x4.7 cm. Histopathologic examination of the mass revealed a granulosa cell tumor. Five months after the operation the patient was re-admitted to our department complaining of acute pelvic pain. Ultrasonographic examination showed a solid mass in the left ovary and a small amount of free fluid in the pouch of Douglas. The patient underwent laparotomy and adhesiolysis, hysterectomy and left salpingo-oophorectomy. Histopathologic examination of the left ovarian mass revealed a ruptured corpus luteum cyst.

Postoperatively, transdermal oestrogen replacement therapy was started to prevent the menopausal syndrome.

The most recent admission was 19 months after the second operation because of chronic abdominal pain. A pelvic mass of 6x5x4 cm was identified with ultrasonographic examination. The mass was cystic with thin echogenic internal septations. Hormonal replacement therapy had been discontinued by the patient five weeks before. Serum oestradiol levels were 154 pg/ml (normal range in reproductive age: 25-155 pg/ml) while FSH and LH levels were 20mIU/ml (normal range in reproductive age: 3-20 mIU/ml) and 35mIU/ml (normal range in reproductive age: 2-15 mIU/ml), respectively. The patient underwent exploratory laparotomy. There were multiple dense adhesions between bowel loops and between the bowel-omentum-parietal peritoneum. In the left iliac fossa a twisted small bowel loop was found attached to the parietal peritoneum and omentum. A hemorrhagic collection was included in this structure within which there was an irregular and rough surface mass of 3 cm in diameter approximately. During adhesiolysis an extensive division through the small bowel wall happened accidentally and finally 20 cm of the small bowel were excised along with the mass. In the afternoon of the operation day the patient underwent urgent laparotomy due to intra-abdominal bleeding and hemostasis was achieved in the region of the sigmoid colon. On the 4th postoperative day the patient developed paralytic ileus, which was successfully treated conservatively. On the 9th postoperative day the serum levels of E2, FSH and LH were 12 pg/ml, 48 mIU/ml and 25 mIU/ml, respectively. Histologic examination of the mass revealed ovarian tissue with several corpi lutei, one of which was hemorrhagic (Figure 1).

The patient was discharged on the 14th postoperative day in good condition. Two months later the patient was well (had no complaints) and pelvic ultrasonographic examination did not reveal any abnormal findings.

Discussion

The troublesome reappearance of functioning ovarian tissue after bilateral oophorectomy has been labeled the ovarian remnant syndrome [1]. Although only a small series of such cases have been reported until now [1-5], it is believed that the syndrome has a greater incidence than was appreciated in the past. This could be explained by the better diagnostic approach that ultrasonograph offers today [2]. On the other hand the widespread use of laparoscopic surgery has possibly contributed to the increase in frequency of this syndrome because small fragments of ovarian tissue can be implanted in the peritoneal cavity.

The demographic profile and clinical presentation of the patients with this syndrome show that usually there is a history of endometriosis or pelvic inflammatory

Revised manuscript accepted for publication May 8, 2000

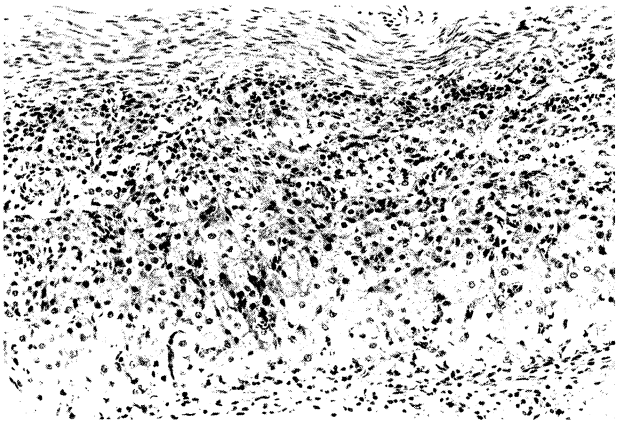

Figure 1. — Ovarian tissue. Corpus luteum H-E, X160.

disease with multiple abdominal operations. Most of these patients complain of chronic pelvic pain, backache, symptoms of irritable bowel syndrome or dyspareunia [2, 5].

Diagnostic criteria for this syndrome are the following:

1) Documentation of previous bilateral oophorectomy in the course of multiple previous operations, and

2) Histopathologic documentation of ovarian tissue obtained at current operation.

Serum FSH and E2 levels are usually normal and lend support to the diagnosis, although some cases have been reported with postmenopausal FSH levels [2, 5].

As regards the etiopathogenetic mechanism of this syndrome, Shemmell and Weed in an experimental study on cats in the early 70's demonstrated that fractions of ovarian cortex that had been implanted in the peritoneum kept their functioning ability [6]. More recently, Minke *et al.* in an experimental study on rats showed that devascularized ovarian tissue may reimplant on intact or abraded peritoneal surfaces where in time it may resume functioning [7].

The current management of this syndrome is surgical. In the past conjugated oestrogens [8] or combinations of estradiol and medroxyprogesterone acetate [9] were used in order to suppress the ovaries via reduction of FSH levels us well us radiotherapy [6]. The latter showed good results but was not used widely due to the increased risk of bowel complications. The surgical management of the syndrome is technically difficult because vital organs such as the ureters and pelvic vessels are frequently adherent to the ovarian remnant tissue [2, 10]. A surgical approach can be done by means of laparotomy or laparoscopy. Those who are in favour of laparotomy support the fact that laparoscopy cannot adequately expose the abdominal cavity [2] or that a malignant lesion could exist within the ovarian remnant [11, 12]. Hence the laparoscopic approach is contraindicated. Those who opt for laparoscopy, on the other hand, state that the laparoscopic approach is completely safe and effective [13]. Finally the use of GnRH agonists or antagonists as an alternative treatment is an attractive therapeutic approach to the syndrome.

Our case fulfilled all the criteria for inclusion in this syndrome: there was a history of bilateral oophorectomy and ultrasonographic findings of a pelvic mass, while E_2 and FSH levels were within the normal range of a premenopausal woman. Finally, the histopathologic examination confirmed the diagnosis.

Ovarian remnant syndrome, although it is an uncommon pathologic condition, should be considered in the differential diagnosis of chronic pelvic pain after recorded bilateral oophorectomy.

References

[1] Symmonds R. E., Pettit P. D. M.: "Ovarian remnant syndrome". *Obstet. Gynecol.*, 1979, *54*, 147.

[2] Pettit P., Lee R.: "Ovarian remnant syndrome; diagnostic dilemma and surgical challenge". *Obstet. Gynecol.*, 1988, *71*, 580.

[3] Riva J. M., Mikuta J. J.: "Ovarian remnant syndrome". *Postgrad. Obstet. Gynecol.*, 1984, *4*, 1.

[4] Webb M. J.: "Ovarian remnant syndrome". *Aust. N.Z. J. Obstet. Gynecol.*, 1989, *29*, 433.

[5] Steege J.: "Ovarian remnant syndrome". *Obstet. Gynecol.*, 1987, *70*, 64.

[6] Shemell R., Weed J.: "Ovarian remnant syndrome". *Obstet. Gynecol.*, 1970, *36*, 299.

[7] Minke T., DePond W., Winkelmann T., Blythe J.: "Ovarian remnant syndrome: study in laboratory rats". *Am. J. Obstet. Gynecol.*, 1994, *171*, 1440.

[8] Utian W. H., Katz M., Davey D. A.: "Effect of premenopausal castration and incremental doses of conjugated equine estrogens on plasma follicle-stimulating hormone, luteinizing hormone, and estradiol". *Am. J. Obstet. Gynecol.*, 1978, *132*, 297.

[9] Schiff I., Tulchinsky D., Cramer D.: "Oral medroxyprogesterone in the treatment of postmenopausal symptoms". *JAMA*, 1980, *244*, 1443.

[10] Price F. V., Edwards R., Buchsbaum H. J.: "Ovarian remnant syndrome: difficulties in diagnosis and management". *Obstet. Gynecol. Surv.*, 1990, *45*, 151.

[11] Bruhwiler H., Luscher K. P.: "Ovarialkarzinom bei ovarian remnant syndrome". *Geburtsh. U. Frauenheilk.*, 1991, *51*, 70.

[12] Glaser D., Burrig K. J., Mast H.: "Ovariankarzinom bei ovarian remnant syndrome?". *Geburtsh. U. Frauenheilk.*, 1992, *52*, 436.

[13] Kamprath S., Rossover M., Schneider A.: "Description of a laparoscopic technique for treating patients with ovarian remnant syndrome". *Fertil. Steri.*, 1997, *68*, 663.

Address reprint requests to:
D. VAVILIS, M.D.
37 Kassandrou St.
54633 Thessaloniki (Greece)
e-mail: vavilis@ccf.auth.gr