Gynecology and Minimally Invasive Therapy 4 (2015) 23–25

Contents lists available at ScienceDirect

Gynecology and Minimally Invasive Therapy

journal homepage: www.e-gmit.com

Case report

Occurrence of ovarian cancer 13 years after a total hysterectomy and bilateral salpingo-oophorectomy for endometriosis: A case report and literature review

Yen-Hsieh Chiu [a], Ching-Hui Chen [a, b], Li-Hsuan Chiu [a], Yuan-Kuei Yen [a, b], Ching-Wen Chang [a, b], Chii-Ruey Tzeng [a, b], Wei-Min Liu [a, b, *]

[a] Department of Obstetrics and Gynecology, Taipei Medical University Hospital, Taipei, Taiwan
[b] Department of Obstetrics and Gynecology, School of Medicine, College of Medicine, Taipei Medical University, Taipei, Taiwan

ARTICLE INFO

Article history:
Received 18 April 2012
Received in revised form
7 January 2014
Accepted 24 January 2014
Available online 10 July 2014

Keywords:
bilateral salpingo-oophorectomy
endometriosis
ovarian cancer
ovarian remnant syndrome
postoophorectomy

ABSTRACT

This article reports a rare case of cancer derived from ovarian remnant tissue. A 52-year-old woman, with a history of a total hysterectomy and bilateral salpingo-oophorectomy for uterine myomas and endometriosis 13 years ago, presented with a pelvic mass accompanied by high serum levels of CA125 and CA199. Exploratory laparotomy and pathology report revealed an ovarian serous tumor with low malignant potential. Cancer derived from ovarian remnant tissue is extremely rare. However, a malignancy may still originate from the ovarian remnant. Ovarian remnant syndrome after oophorectomy and its possibility of malignancy, especially in case of endometriosis, should not be neglected.

Introduction

Ovarian remnant syndrome (ORS) was a finding of histologically residual ovarian tissue after a bilateral salpingo-oophorectomy (BSO).[1] ORS may be present with a normal ovary or ovary-related diseases, such as the recurrence of disease or the occurrence of a malignancy, which might contribute to some complications.[2] In addition, because ORS is often neglected, it can produce a diagnostic dilemma and therapeutic challenge.[3] We report a case of ovarian cancer diagnosed 13 years after total hysterectomy (TH) and BSO were carried out.

Case report

A 52-year-old woman had previously undergone a TH and BSO for severe dysmenorrhea and continuous pelvic pain, which were secondary to uterine myomas and endometriosis at age 39 years. The patient denied any other previous medical or surgical illness, or having received any hormone therapy. Ultrasound and computed tomography (Fig. 1) revealed an 11-cm cystic pelvic mass with a solid component and rich vascularity. Upper and lower gastrointestinal endoscopy failed to demonstrate any abnormalities. However, she was found to have elevated serum levels of CA-125 (995.3 mIU/mL) and CA-199 (617 mIU/mL).

We performed an exploratory laparotomy and found a >10-cm mass completely occupying the *cul-de-sac* with dense adhesion surrounding the tumor. After careful and meticulous dissection, the tumor was completely removed (Fig. 2). The final pathology revealed the diagnosis of an ovarian serous tumor of low malignant potential (Fig. 3), based on the morphological diagnosis and the assistance of immunohistochemistry, including the positive presence of ovarian stromal tissues within the tumor (Fig. 4), negative staining for calretinin, cytokeratin 5/6, and thrombomodulin, and positive staining for the estrogen and progesterone receptors. The patient is now undergoing regular follow up. No further adjuvant therapy was administered, and the patient remains well at the time of submission.

Discussion

If there is a suspicion of ORS, the following strategies can be used to facilitate a diagnosis: including checking serum levels of

Conflicts of interest: All contributing authors declare no conflicts of interest.

* Corresponding author: Department of Obstetrics and Gynecology, Taipei Medical University Hospital and Taipei Medical University, 252 Wusing Street, Sinyi District, Taipei 110, Taiwan.
E-mail address: weimin@tmu.edu.tw (W.-M. Liu).

http://dx.doi.org/10.1016/j.gmit.2014.05.007

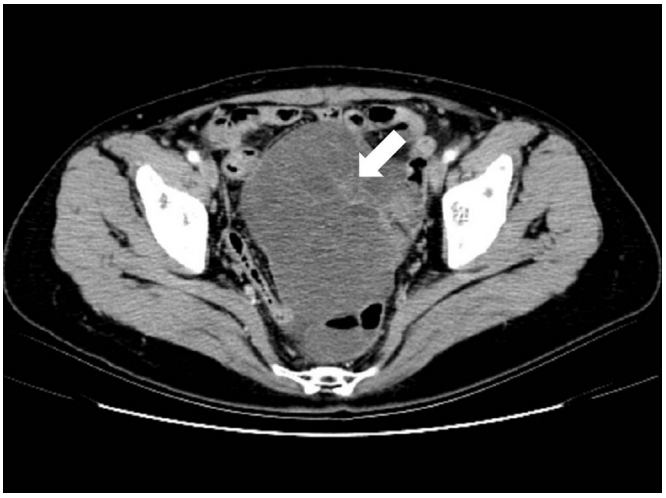

Fig. 1. Postcontrast computed tomographic scan of the pelvis showing a large, cystic, hypodense, heterogeneous, nonenhanced mass (white arrow) measuring about 110 mm × 100 mm × 85 mm contiguous with the sigmoid colon. There is no definitely enlarged sidewall lymph node.

gonadotropins and estradiol; using an ovulation-inducing drug to stimulate ovary growth; and evaluating final images.[4] Ultrasound and ovulation-inducing drugs are the most economical and convenient tools for diagnosing ORS in premenopausal women.[5] The main cause of ORS is secondary to the incomplete removal of one or both ovaries. Surgical difficulty contributes to the incomplete removal, including disease itself, such as endometriosis, or any inflammatory or surgical process; and problems related to the surgical techniques.[6,7] To avoid ORS, the main principle is clear identification and the complete removal of the ovary tissue during the surgical procedure.

ORS seldom causes severe sequelae, but the majority of patients suffer from chronic pelvic pain and recurrence of diseases. However, severe life-threatening diseases may also occur. Cancers might be the most serious sequelae, and mostly were malignancies.[2,8–10] This leads to an idea that oophorectomy may not comprehensively protect against the subsequent development of ovarian carcinoma.[8]

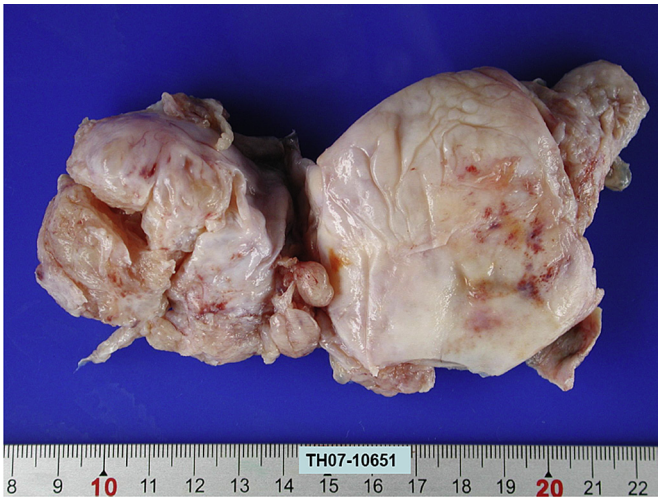

Fig. 2. Excision of the tumor by exploratory laparotomy revealing cystic tissue fragments with a gray, smooth external surface.

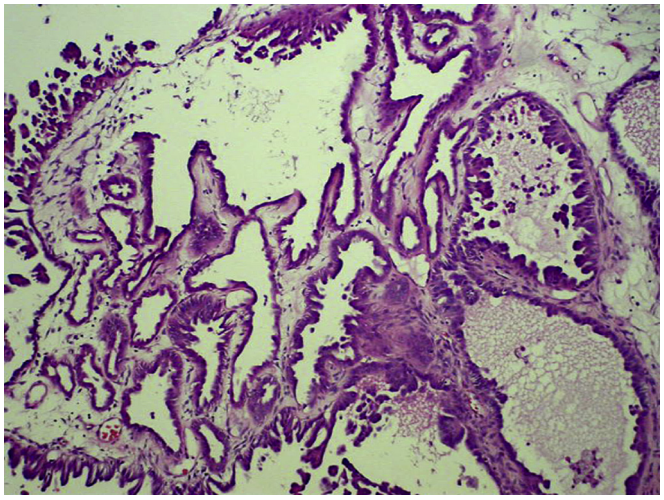

Fig. 3. Focal stratification and tufting of epithelial cells with a micropapillary pattern noted in areas of the tumor. No stromal invasion can be seen.

In addition, half of the cases had a history of endometriosis. The patient in this report also had a history of endometriosis, but was complicated by the borderline malignancy. This is believed to be the first documented case of a serous tumor of low malignant potential arising from an ovarian remnant.

Distinguishing between an epithelioid peritoneal mesothelioma and papillary serous carcinomas involving the peritoneum may be very difficult due to overlapping morphological features.[11] If ovarian stromal tissue exists in the tumor, an ovarian origin is favored. However, sometimes it is not easy to find the ovarian stromal tissue. Therefore, immunohistochemistry using several markers may facilitate establishing a correct diagnosis.[11] In this study, we used five markers to aid in the diagnosis. Because this tumor was negative for calretinin, cytokeratin 5/6, and thrombomodulin, but positive for estrogen and progesterone receptors, a diagnosis of a serous ovarian tumor was made.

The possibility of the disease is very often overlooked in patients with previous oophorectomy. Similar to other ovarian tumor, ORS is very often asymptomatic and probably mostly being suspected when a pelvic tumor is found in imaging. ORS should be suspected when patients with previous oophorectomy developed pelvic pain,

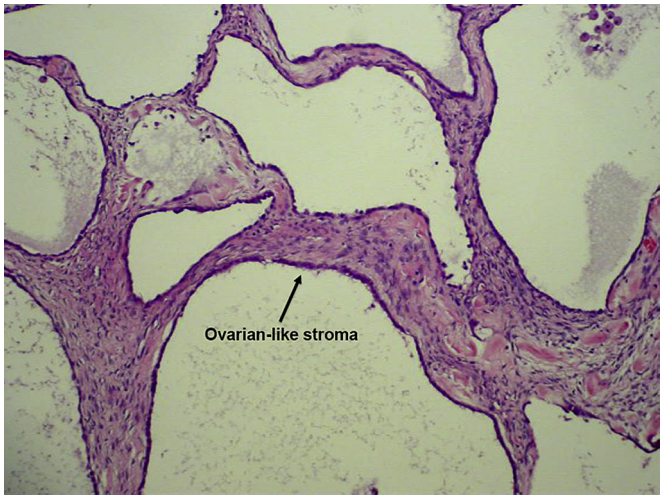

Fig. 4. Microscopic picture showing a multicystic neoplasm composed of numerous variably sized cysts or torturous glandular structures lined by single-layered epithelioid to flat lining epithelia among the ovarian stroma.

especially when the procedure was performed for endometriosis. As a result, regular checkup and CA125 monitoring in patients with BSO for endometriosis in early detection of ORS might be considerable.

Conclusion

The point of interest in this case was the difficulty and challenge of the clinical diagnosis (TH and BSO),[12] because ORS associated with other diseases is frequently and easily neglected. Physicians sometimes overlook this possibility when a woman presents with a pelvic mass after a TH and BSO for benign diseases, therefore ovarian diseases should be kept in mind in the differential diagnosis.

References

1. Shemwell RE, Weed JC. Ovarian remnant syndrome. *Obstet Gynecol*. 1970;36:299–303.
2. Donnez O, Squifflet J, Marbaix E, Jadoul P, Donnez J. Primary ovarian adenocarcinoma developing in ovarian remnant tissue ten years after laparoscopic hysterectomy and bilateral salpingo-oophorectomy for endometriosis. *J Minim Invasive Gynecol*. 2007;14:752–757.
3. Webb MJ. Ovarian remnant syndrome. *Obstet Gynecol*. 1989;29:433–435.
4. Rana N, Rotman C, Hasson HM, Redwine DB, Dmowski WP. Ovarian remnant syndrome after laparoscopic hysterectomy and bilateral salpingo-oophorectomy for severe pelvic endometriosis. *J Am Assoc Gynecol Laparosc*. 1996;3:423–426.
5. Kaminski PF, Sorosky JI, Mandell MJ, Broadstreet RP, Zaino RJ. Clomiphene citrate stimulation as an adjunct in ovarian remnant syndrome. *Obstet Gynecol*. 1990;76:924–926.
6. Magtibay PM, Magrina JF. Ovarian remnant syndrome. *Clin Obstet Gynecol*. 2006;49:526–534.
7. Dereska NH, Cornella J, Hibner M, Magrina JF. Mucinous adenocarcinoma in an ovarian remnant. *Int J Gynecol Cancer*. 2004;14:683–686.
8. Hamid R, May D. Ovarian malignancy in remnant ovarian tissue. *Int J Gynaecol Obstet*. 1997;58:319–320.
9. Kho RM, Magrina JF, Magtibay PM. Pathologic findings and outcomes of a minimally invasive approach to ovarian remnant syndrome. *Fertil Steril*. 2007;87:1005–1009.
10. Narayansingh G, Cumming G, Parkin D, Miller I. Ovarian cancer developing in the ovarian remnant syndrome. A case report and literature review. *Aust NZ J Obstet Gynaecol*. 2000;40:221–223.
11. Comin CE, Saieva C, Messerini L. h-Caldesmon, calretinin, estrogen receptor, and Ber-EP4: a useful combination of immunohistochemical markers for differentiating epithelioid peritoneal mesothelioma from serous papillary carcinoma of the ovary. *Am J Surg Pathol*. 2007;31:1139–1148.
12. Lu YF, Huang BS, Lai CR, Yen MS, Chen YJ, Wang PH. Endometrial stromal sarcoma occurring 20 years later after total hysterectomy for myomas. *Gynecol Min Invas Ther*. 2014;3:19–22.