Multilobular Cyst as Endosalpingiosis of Uterine Serosa: A Case Report

Yu Chang, Eing-Mei Tsai, Cheng-Hui Yang, Chiu-Hu Kuo, and Jau-Nan Lee
Department of Obstetrics and Gynecology, Kaohsiung Medical University Hospital, Kaohsiung, Taiwan.

A case of endosalpingiosis presented as a multilobular cyst on sonography. The tentative clinical diagnosis was an ovarian tumor; however, laparotomy revealed a degenerative cyst of the uterine myoma with a stalk connecting to the uterus. Histopathologically, it showed characteristics of endosalpingiosis. To our knowledge, such a multilobular cyst of endosalpingiosis originating solely from the uterine serosa has not been reported.

Key Words: endosalpingiosis, uterine serosa, multilobular cyst
(*Kaohsiung J Med Sci* 2003;19:38–41)

Uterine leiomyoma and adnexal tumors are the most frequent neoplasms in women of reproductive age [1, 2]. Pelvic examination is known to be less sensitive than sonography in identifying the nature of pelvic tumors [3]. Sonography allows preoperative planning of surgical procedures according to the morphologic presumption of the tumor's behavior [4]. Clinical workup including computerized tomography and magnetic resonance imaging as well as tumor marker determination are valuable in diagnosis if sonography is inconclusive. However, even under laparoscopic examination, challenges still remain, especially for rare tumors such as serosal cysts, endosalpingiosis, or mesothelial cysts, because diagnosis relies on histopathologic findings. Most of these rare tumors or cysts are congenital and may be of either mesonephric or mullerian origin [5]. Recently, four cases have been diagnosed histopathologically as endosalpingiosis that resembled cystic neoplasm involving multiple pelvic sites [6]. One of these was even interpreted as "suspicious for invasive minimal deviation adenocarcinoma" in frozen section. We report a huge pelvic tumor that initially mimicked an adnexal tumor ultrasonographically and was thought to be a degenerative cyst of the myoma during surgery. Eventually, the histopathologic diagnosis was endosalpingiosis of the uterine serosa.

Case Presentation

A 45-year-old female, gravida 5 para 3 abortus 2, consulted our clinic due to intermittent lower abdominal dullness and irregular menstruation for 3 months. Occasionally, symptoms were aggravated after heavy physical activity. There was no contributory information in her medical history.

A multilobular mass (8.2 x 7.5 x 6.0 cm) with filmy septae and homogenous cystic content appeared behind the uterus in the pelvic cavity on sonography. The morphologic features provided a tentative diagnosis of adnexal tumor. Biochemical examinations and tumor markers including CA125, CA199, carcinoembryonic antigen, α-fetoprotein, and tumor polypeptide antigen were within normal limits.

During laparotomy, an irregular, movable, cystic, and well-encapsulated tumor (the same size as the mass seen on the sonogram) was tan to brown in color

Received: September 13, 2002 Accepted: November 13, 2002
Address correspondence and reprint requests to: Dr. Jau-Nan Lee, Department of Obstetrics and Gynecology, Kaohsiung Medical University Hospital, 100 Shih-Chuan 1st Road, Kaohsiung City 80707, Taiwan.
E-mail: tsaieing@yahoo.com

and localized in the cul-de-sac. A short fibrous stalk (0.3 cm in diameter) connected the cyst and the serous surface of the middle posterior uterine wall (Figure 1). No ascites or pelvic abnormality was observed. Total hysterectomy with tumor resection was performed under the impression of uterine myoma with cystic degeneration.

Sectioning revealed multilobular cysts separated by filmy septae and containing serous or sanguineous fluid. The uterus was normal in size and shape except a small intramural myoma within the anterior wall. Several darkish foci were found in the myometrium with features consistent with adenomyosis.

Microscopically, there was a cystic formation with an outer smooth muscular layer and inner epithelial lining. The epithelium was lined with ciliated and non-ciliated columnar or cuboidal cells, which were benign and mitotically inactive, mimicking the features of tubal epithelium (Figure 2). The epithelial lining was mostly a single layer with some foci of papillary infoldings. The histopathologic picture contributed to the diagnosis of endosalpingiosis with serous cystadenoma transformation.

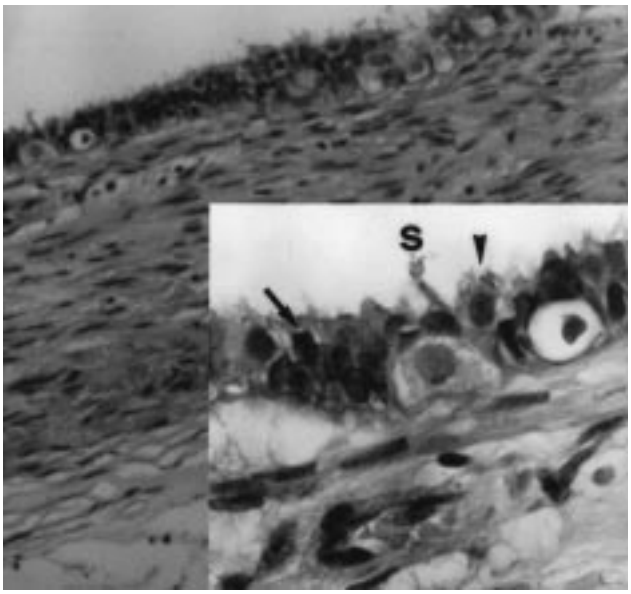

Figure 2. *Cystic wall composed of single-layered epithelium and scanty muscular tissues. (Hematoxylin & eosin, x 40). Inset: Epithelium with serous-secretory characteristics (S) shows mitotically inactive cuboidal cells (arrow) with ciliation (arrowhead). (Hematoxylin & eosin, x 400)*

Discussion

We report a case of multilobular, cystic tumor (8.2 x 7.5 x 6.0 cm in size) arising primarily from the uterine serosa with a connecting stalk. No endometriotic lesions were found in the pelvis. A variety of epithelial-lined cystic structures over the uterine serosal aspect should be taken into consideration for differential diagnosis [7]. This case was interesting due to the tumor origin and size, the histopathologic evaluation, and the clinical diagnosis.

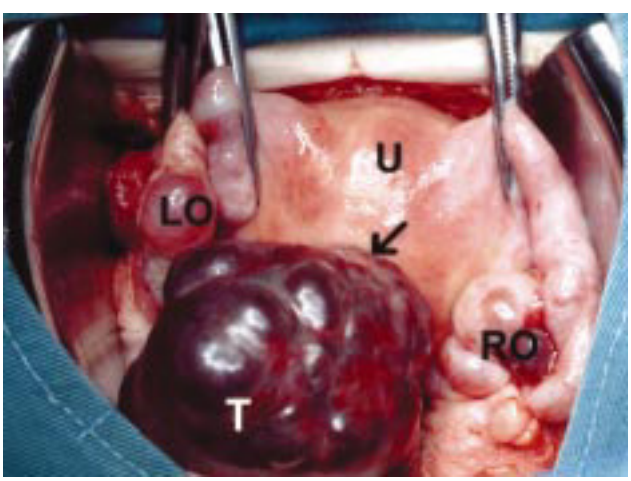

Figure 1. *A multilobular cyst (T) independent of bilateral adnexae with a short stalk (0.3 cm in diameter) connecting the cyst to the middle posterior fundus (arrow). U = uterus; RO = right ovary; LO = left ovary.*

Endometriosis of the uterine serosa was considered first due to its high prevalence in women of reproductive age. Although there were several hemorrhagic areas of this cyst, endometriosis was unlikely due to the absence of typical glandular-stromal components and hemosiderin-laden macrophages microscopically.

Adenomatoid tumors originating from the uterine serosa are seen in about 1% of reproductive-age women [8]. They usually infiltrate the myometrium and extend even toward the endometrium. After careful sectioning in this case, neither the uterus nor the tumor demonstrated the diffusely infiltrating nature of adenoid and/or tubular conformations.

In past decades, increasing case reports of multilobular peritoneal inclusion cyst (MPIC) or cystic mesothelioma, probably involving uterine serosa, have emphasized the recurrence or persistence of disease behavior. Despite the same embryologic origin, the present case is unlikely to be MPIC for several histologic reasons: the uniformity of the cellular appear-

ance mimicking tubal metaplasia; single-layered secretory epithelium accounting for most of the cyst; and the absence of papillary formation in the cystic mesothelioma. Moreover, the integrity of the peritoneum and the absence of inflammatory process or pelvic adhesion exclude the diagnosis of MPIC [9].

Endosalpingiosis, a benign lesion, refers to the presence of tubal-like gland or epithelium (typically < 5 mm in diameter) involving the peritoneum in addition to the serosa of the uterus [10]. The diagnosis is almost incidental and made by microscopic examination. Recently, Clement and Young reported four cases of endosalpingiosis presenting as masses similar to neoplasms [6]. Two of these were endosalpingiosis involving multiple sites on the uterus, adnexa, and other pelvic organs, while the other two were nominated as the first reported cases of transmural endosalpingiosis of the uterus.

The present multilobular cystic tumor arose from the uterine serosa and had a benign histologic appearance with multiple fluid-filled cysts lined with mitotically inactive and single-layered tubal epithelium containing ciliated secretory cells. These microscopic features are consistent with endosalpingiosis. However, endosalpingiosis originating solely from the uterine serosa with such a multilobular cyst has never been reported, notably with initial clinical misinterpretation twice.

Acknowledgments

The authors would like to thank Prof. HG Hsu and Dr. MC Lin, Department of Pathology, School of Medicine, National Taiwan University, for their kind review of our specimen.

References

1. Buttram VC Jr, Reiter RC. Uterine leiomyomata: etiology, symptomatology, and management. *Fertil Steril* 1981; 36:433–45.
2. Koonings PP, Campbell K, Mishell DR Jr, Grimes DA. Relative frequency of primary ovarian neoplasms: a 10-year review. *Obstet Gynecol* 1989;74:921–6.
3. Rulin MC, Preston AL. Adnexal masses in postmenopausal women. *Obstet Gynecol* 1987;70:578–81.
4. Kurjak A, Jukic S, Kupesic S, Babic D. A combined Doppler and morphopathological study of ovarian tumors. *Eur J Obstet Gynecol Reprod Biol* 1997;71:147–50.
5. Thor AD, Young RH, Clement PB. Pathology of the fallopian tube, broad ligament, peritoneum and pelvic soft tissues. *Hum Pathol* 1991;22:856–67.
6. Clement PB, Young RH. Florid cystic endosalpingiosis with tumor-like manifestations: a report of four cases including the first reported cases of transmural endosalpingiosis of the uterus. *Am J Surg Pathol* 1999;23:166–75.
7. Hendrickson MR, Kempson RL. Non-neoplastic conditions of the myometrium and uterine serosa. In: Fox H, Wells M, eds. *Haines and Taylor Obstetrical and Gynecological Pathology*. New York: Churchill Livingstone, 1995:511–8.
8. Tiltman AJ. Adenoid tumors of the uterus. *Histopathology* 1980;4:437–43.
9. Weiss SW, Tavassoli FA. Multicystic mesothelioma: an analysis of pathologic finding and biologic behavior in 37 cases. *Am J Surg Pathol* 1988;12:737–46.
10. Clement PB, Young RH. Pathology of endometriosis and lesions of the secondary mullerian system. In: Clement PB, Young RH, eds. *Atlas of Gynecologic Surgical Pathology*. Philadelphia, PA: W.B. Saunders Co, 2000:424–43.