

Ovarian torsion of mixed epithelial tumor misdiagnosed as a malignancy in postmenopausal woman

A case report

Hyeji Jeon, MD, Aeli Ryu, MD, PhD*, Hui-Gyeong Seo, MD, Si-Hyong Jang, MD

Abstract

Rationale: Adnexal torsion is 1 of the most common emergency gynecological disease. It is more often diagnosed in reproductive age, but rarely in postmenopausal women. The clinical symptoms of adnexal torsion are nonspecific in postmenopausal women. Epithelial ovarian tumors are common in adults, and the risk of malignancy increases with age, especially after menopause. So, it is difficult to diagnose adnexal torsion precisely compared with reproductive women, and most cases of adnexal torsion with postmenopausal women are diagnosed as a malignancy preoperatively. We report a case of ovarian torsion with mixed epithelial tumor misdiagnosed as a malignancy in postmenopausal woman.

Patient concerns: A 65-year-old woman presented lower left abdominal pain, and there was slight abdominal distension, but no tenderness or rebound tenderness on abdominal palpation.

Diagnoses: Radiologic assessments showed a huge multiseptated cystic mass with solid portion in the left ovary, and malignancy was suspected. The test for serum tumor markers revealed normal levels of cancer antigen 125 (CA-125).

Interventions: The patient underwent a laparotomy and there was torsion of the left ovary. We conducted frozen biopsy of left ovary for confirming malignancy before performing staging surgery.

Outcomes: The result of a frozen section biopsy confirmed a borderline Brenner tumor associated with a benign mucinous tumor. Subsequently, total hysterectomy and right salpingo-oophorectomy were performed. The operation was completed without addition procedures

Lessons: Ovarian torsion is benign in most cases and malignancy is rare. Although very rare, ovarian torsion can occur in postmenopausal women, and it should be taken into consideration that the possibility of malignancy is low in postmenopausal women with normal CA-125 levels. Instead of performing staging surgery, it appears to be appropriate to carry out surgery based on the result of intraoperative frozen section biopsy so that we were able to avoid unnecessary surgical procedures.

Abbreviation: CA-125 = cancer antigen 125.

Keywords: adnexal torsion in postmenopausal women, mixed ovarian tumor

1. Introduction

Adnexal torsion is a benign ovarian tumor that mainly occurs in women of reproductive age and rarely occurs in postmenopausal women. Since the clinical symptoms of adnexal torsion are nonspecific in postmenopausal women, accurate preoperative

diagnosis is very challenging.^[1] About 30% of ovarian masses found in postmenopausal women are malignant, and the risk of malignancy increases with age. For these reasons, the possibility of a malignant adnexal mass is greater than the possibility of torsion.^[2,3] On transvaginal ultrasound often used as a screening tool for adnexal tumors, malignancy can be suspected when the size of an ovarian mass is larger than 10 cm or the tumor has papillary projections or solid components. In postmenopausal women with these imaging findings, bilateral salpingo-oophorectomy and hysterectomy are typically performed considering that a mass is malignant.^[4-6]

The authors experienced a case of ovarian torsion with mixed epithelial tumor misdiagnosed as a malignancy while undergoing giant ovarian cyst removal in a postmenopausal woman. We report the case with a brief review of literature. This report was approved by the Soonchunhyang University Research Funds.

2. Case report

A 65-year-old married woman visited our emergency room of Obstetrics and Gynecology Department, Soonchunhyang University Cheonan Hospital, due to a chief complaint of lower left abdominal pain which started 3 days before her visit and aggravated on the day of visit. Her obstetric history was the

Editor: Jianxun Ding.

The authors report no conflicts of interest.

Department of Obstetrics and Gynecology, Department of Pathology, Soonchunhyang University College of Medicine, Soonchunhyang University Cheonan Hospital, Cheonan, Korea.

* Correspondence: Aeli Ryu, Department of Obstetrics and Gynecology, Soonchunhyang University College of Medicine, Soonchunhyang University Cheonan Hospital, Cheonan, Korea (e-mail: bestal@schmc.ac.kr).

Copyright © 2017 the Author(s). Published by Wolters Kluwer Health, Inc. This is an open access article distributed under the terms of the Creative Commons Attribution-Non Commercial License 4.0 (CCBY-NC), where it is permissible to download, share, remix, transform, and build up the work provided it is properly cited. The work cannot be used commercially without permission from the journal.

Medicine (2017) 96:40(e8207)

Received: 19 April 2017 / Received in final form: 30 June 2017 / Accepted: 3 September 2017

<http://dx.doi.org/10.1097/MD.00000000000008207>

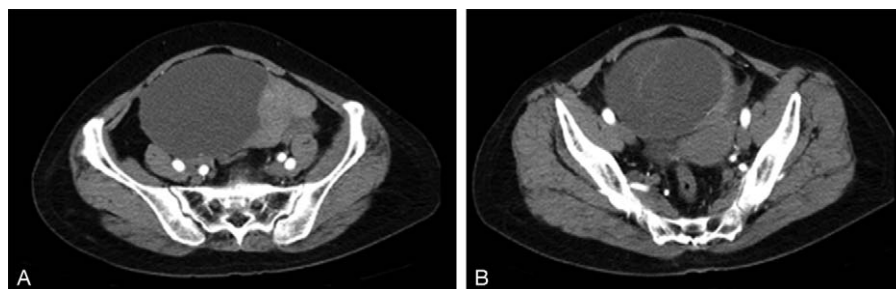


Figure 1. The images of pelvic ultrasonography and computed tomography. (A) A 13×9 cm sized multicystic tumor of left ovary. (B) A 16×9×14 cm sized multiseptated cystic mass with large enhancing solid portion in the left ovary.

patient has had 2 term births, 0 preterm births, 2 abortions, 2 living children and had menopause 10 years ago. At the time of visit, her systemic condition was favorable. Her height was 162 cm and weight was 61 kg. Vital signs were normal with a blood pressure of 110/60 mm Hg, pulse rate of 60 beats/min, body temperature of 36.0°C, respiratory rate of 19 breaths/min, and no apparent specific signs. There was slight abdominal distension, but no tenderness or rebound tenderness on abdominal palpation. In pelvic examination, no vaginal bleeding or discharge was noted.

In radiologic assessment, transvaginal and abdominal ultrasound showed a normal uterus and a 13×9 cm sized multicystic tumor with irregular surface of the left ovary (Fig. 1A). Abdominal and pelvic computed tomography scans revealed a 16×9×14 cm sized multiseptated cystic mass with large enhancing solid portion in the left ovary, and malignancy was suspected (Fig. 1B). In addition, there was a finding of mild dilation of the right distal ureter caused by the compression of the left ovarian mass, and there were no findings of ascites, pleural effusion, and lymph node enlargement. Test results were normal in peripheral blood smear, blood coagulation, liver function, renal function, urinalysis, and C-reactive protein, and her chest x-ray and electrocardiogram were normal. The test for serum tumor markers revealed normal levels of cancer antigen 125 (CA-125) at 8.69 IU/mL (range 0–35 IU/mL) and carbohydrate antigen 19–9 at 3.29 U/mL (range 0–34 U/mL). The patient underwent

exploratory laparotomy with a suspicion of borderline or malignancy of an adnexal mass. A white-gray 17×9 cm sized cystic tumor was detected in the left adnexa, having a torsion of 720° in a counterclockwise direction or no necrosis (Fig. 2). Uterus, right ovary, large intestine, bladder, small intestine, and liver were normal in inspection and palpation of abdominal organs. There were no presentations of ascites or nodules within the peritoneal cavity. We carried out laparotomy because there was the possibility of ovarian malignancy, but torsion of the left ovary was observed. So, we decided to conduct frozen biopsy of left ovary for confirming malignancy before performing staging surgery. Salpingo-oophorectomy on the left side was performed after washing cytology from the peritoneal cavity, and the result of an emergent frozen section biopsy confirmed a borderline Brenner tumor associated with a benign mucinous tumor. Subsequently, total hysterectomy and right salpingo-oophorectomy were performed. The operation was completed without additional procedures such as pelvic lymph node dissection.

Gross pathologic findings revealed that the left ovary showed a well-demarcated oval mass composed of both solid and cystic components, measuring 17.0×16.0×8.5 cm in dimensions. The cystic area showed multilocular appearance and it contained mucin materials. On microscopic examination, the cyst wall was lined by a single layer of benign mucinous cells, which was consistent with benign mucinous cystadenoma. The solid area displayed round to oval tumor cell nests within fibrous stroma and the tumor cells had fine chromatin pattern, obvious nucleoli, and longitudinal grooving with no cytologic atypia or mitotic activity. Histologic features of solid area were benign Brenner tumor (Fig. 3). Therefore, we diagnosed this ovarian tumor as benign mixed Brenner-mucinous tumor.

The patient was discharged in good condition on the 15th postoperative day without specific complications.

3. Discussion

The most common histologic type of ovarian tumor is epithelial tumor which is subdivided into 5 subtypes including serous, mucinous, endometrioid, clear cell, and Brenner tumors.^[7] Mucinous tumor is a relatively common and benign condition accounting for approximately 10% to 15% of all ovarian tumors, but Brenner tumor is a very rare epithelial tumor accounting for about 2% of all cases.^[8,9] Brenner tumors are composed of epithelial cells that resemble the epithelium of the urinary bladder, and about 20% often manifest as a mixed epithelial tumor with other ovarian cancers, mainly associated with mucinous tumor.^[10] The risk of malignancy increases with age in epithelial ovarian tumors. The malignancy rate of ovarian



Figure 2. Intraoperative picture shows that a white-gray left ovarian cystic tumor, approximately 17×9 cm size, had a torsion of 720° in a counterclockwise direction.

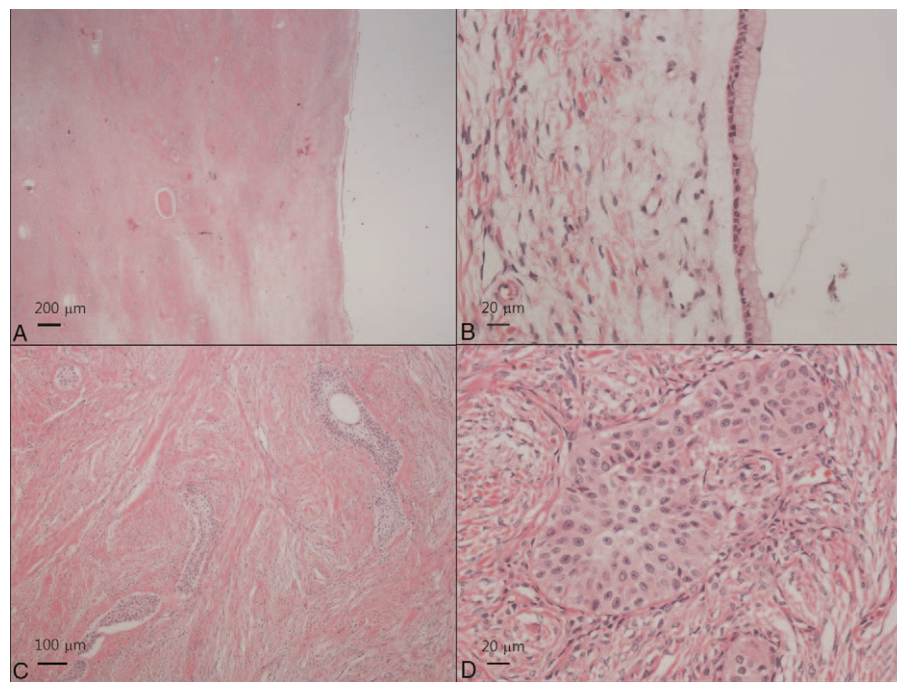


Figure 3. (A) The tumor consisted of solid (left side) and cystic (right side) areas (H&E, $\times 40$). (B) Cystic spaces were covered with single layered mucinous cells (H&E, $\times 400$). (C) Solid areas showed dense fibrous stroma and round to oval-shaped epithelial cell nests (H&E, $\times 100$). (D) Tumor cell nests in solid areas showed no abundant eosinophilic cytoplasm with a single prominent nucleolus and frequent nuclear groove (H&E, $\times 400$).

tumors is known to range between 6% and 11% in premenopausal women, whereas the risk increases up to about 30% in postmenopausal women, especially in women above 40 years of age.^[2,3]

Tumor marker test and ultrasound are relatively easy diagnostic methods of screening for ovarian cancers. CA-125 is a tumor marker used in discriminating malignant from benign ovarian tumors and CA-125 levels increase in women with epithelial ovarian tumors. Although reproductive age women with elevated levels of CA-125 are more likely to develop malignant ovarian tumors, the usefulness of elevated CA-125 levels decrease in discriminating malignant from benign ovarian tumors in women of childbearing age because an increase in CA-125 can be associated with benign conditions such as endometriosis, uterine leiomyoma, and pelvic inflammatory disease, pregnancy, and menstruation.^[11] CA-125 level and ultrasonography have been shown to have a low positive predictive value when used in a screening test for adnexal tumors in postmenopausal women.^[12] Ultrasonography is a useful tool for diagnosing adnexal lesions and has been commonly performed for discriminating malignancy in ovarian masses and adnexal torsion. Malignancy can be suspected when the size of an ovarian mass is larger than 10 cm or the tumor has papillary projections or solid components on ultrasound.^[13] Since mucinous tumors are usually large in size, manifest as multicystic lesions, and contain characteristic internal components such as septations with low-level internal echoes, discrimination between benign and malignant pathology is difficult with sonographic findings alone.^[14] Brenner tumors often appear as a solid mass with internal echoes and are generally similar to fibroma and uterine leiomyoma, and borderline or malignant ovarian tumors contain mixed cystic components and need to be discriminated from other malignant ovarian masses.^[15] Absence of arterial flow in the twisted ovarian pedicle was identified in

color Doppler test performed to differentiate ovarian torsion.^[14] However, despite blood flow to the ovaries in color Doppler ultrasound, the likelihood of torsion cannot be completely excluded.^[16]

Adnexal torsion is a gynecological emergency associated with acute abdominal pain that results from the rotation of the ovary, fallopian tube, and cystic mass. The cause of torsion has been suggested to be excessively elongated utero-ovarian ligament or the increased volume and weight of an adnexal mass.^[1,17] Adnexal torsion more commonly occurs in women of reproductive age, but this gynecological emergency rarely affects postmenopausal women at approximately 2.7%.^[1] The leading clinical symptom is acute pain in the lower abdomen. Usually, severe abdominal pain persists, but repeated relapse and remission may be seen in partial adnexal torsion. Abdominal pain occurs during elevation, exercise, and during sexual intercourse, and secondarily produce autonomic nervous system reflexes including nausea, vomiting, anxiety, tachycardia, and others.^[17] Abdominal pain resulting from torsion has different clinical manifestations in the reproductive and menopausal periods. Abdominal pain progresses to sharp and severe pain in women of reproductive age, whereas is constant and dull in postmenopausal women.^[4] Therefore, acute abdominal pain arising in women of childbearing age enables early diagnosis of torsion and shortens duration from the onset of torsion to surgery. On the contrary, nonspecific clinical signs in postmenopausal women extend the duration until the first surgery.^[4,5] When torsion occurs, the size of a mass increases rapidly as lymphatic flow and blood supply to the adnexa diminishes. Thus, a large mass in the pelvis is easily palpated on abdominal examination. Elevated white blood cell count may be observed with the development of ischemia and necrosis of the adnexa in torsion, but there are no specific laboratory markers that indicate the diagnosis of torsion.

A 65-year-old postmenopausal woman in this case report had an ovarian mass manifested as a large multicystic lesion containing a solid portion in radiologic assessment. An operation was conducted to exclude the possibility of a malignant tumor. Moreover, laparotomy was carried out by keeping in mind the possibility of staging surgery if frozen section biopsy revealed evidence of malignancy, but the analysis showed torsion of the left ovary. Though torsion was suspected preoperatively because of an ovarian mass accompanying abdominal pain, her clinical presentations were nonspecific compared with reproductive age women. There are limitations to confirm torsion based on clinical findings alone. Considering severity of her condition, we thought that it was more appropriate to design a treatment plan by diagnosing the condition as a malignant tumor rather than torsion. According to Cohen et al,^[18] the preoperative diagnosis of ovarian torsion was confirmed in only 44% of the women, and torsion should be confirmed at surgery and requires diagnosis and management simultaneously.^[19] Laparoscopic surgery has been most commonly used in reproductive age women, whereas laparotomy has been often performed in postmenopausal women with a malignant tumor by keeping staging surgery in mind.^[4]

Ovarian torsion is benign in most cases, and malignant ovarian tumors are rare because malignancy causes inflammation and decreased mobility of the uterus adnexa due to adhesion formation in peripheral tissues. Eitan et al reported that histological examination did not reveal any malignancy in reproductive age women with torsion of the adnexa, whereas malignancy was diagnosed in 22% of postmenopausal women.^[4,20–22]

Since the symptoms of adnexal torsion are nonspecific in postmenopausal women, for an ovarian mass associated with abdominal pain, the choice of a surgical approach is determined based on condition severity in preparation for malignant tumor treatment instead of torsion. Although very rare, ovarian torsion can occur in postmenopausal women, and it should be taken into consideration that the possibility of malignancy is low in postmenopausal women with normal CA-125 levels. Instead of performing staging surgery, it appears to be appropriate to carry out surgery based on the result of intraoperative frozen section biopsy to exclude the possibility of malignancy. By carefully conducting intraoperative frozen section biopsy, instead of performing staging surgery, we were able to avoid unnecessary surgical procedures and treatment by considering the possibility of a malignant mass and keeping in mind normal CA-125 levels in a postmenopausal woman with a giant ovarian tumor. The authors experienced a case of ovarian torsion with mixed epithelial tumor preoperatively misdiagnosed as a malignancy in a postmenopausal woman. The patient was successfully treated and we report the case with a brief review of literature.

References

- [1] Hibbard LT. Adnexal torsion. *Am J Obstet Gynecol* 1985;152:456–61.
- [2] Webb PM, Jordan SJ. Epidemiology of epithelial ovarian cancer. *Best Pract Res Clin Obstet Gynaecol* 2017;41:3–14.
- [3] Kinkel K, Lu Y, Mehdizade A, et al. Indeterminate ovarian mass at US: incremental value of second imaging test for characterization: meta-analysis and Bayesian analysis. *Radiology* 2005;236:85–94.
- [4] Cohen A, Solomon N, Almog B, et al. Adnexal torsion in postmenopausal women: clinical presentation and risk of ovarian malignancy. *J Minim Invasive Gynecol* 2017;24:94–7.
- [5] Ganer Herman H, Shalev A, Ginath S, et al. Clinical characteristics and the risk for malignancy in postmenopausal women with adnexal torsion. *Maturitas* 2015;81:57–61.
- [6] Practice Bulletin No. 174 summary: evaluation and management of adnexal masses. *Obstet Gynecol* 2016;128:1193–5.
- [7] Chen VW, Ruiz B, Killeen JL, et al. Pathology and classification of ovarian tumors. *Cancer* 2003;15:2631–42.
- [8] Koonings PP, Campbell K, Mishell DR Jr, et al. Relative frequency of primary ovarian neoplasms: a 10-year review. *Obstet Gynecol* 1989;74:921–6.
- [9] Ziadi S, Trimeche M, Hammedi F, et al. Bilateral proliferating Brenner tumor of the ovary associated with recurrent urothelial carcinoma of the urinary bladder. *N Am J Med Sci* 2010;2:39–41.
- [10] Waxman M. Pure and mixed Brenner tumors of the ovary: clinicopathologic and histogenetic observations. *Cancer* 1979;43:1830–9.
- [11] Van Calster B, Timmerman D, Bourne T, et al. Discrimination between benign and malignant adnexal masses by specialist ultrasound examination versus serum CA-125. *J Natl Cancer Inst* 2007;99:1706–14.
- [12] Fung MF, Bryson P, Johnston M, et al. Screening postmenopausal women for ovarian cancer: a systematic review. *J Obstet Gynaecol Can* 2004;26:717–28.
- [13] American College of Obstetricians and Gynecologists' Committee on Practice Bulletins—Gynecology. Gynecologists' Committee on Practice, Practice Bulletin No. 174: evaluation and management of adnexal masses. *Obstet Gynecol* 2016;128:e210–26.
- [14] Chu LC, Coquia SF, Hamper UM. Ultrasonography evaluation of pelvic masses. *Radiol Clin North Am* 2014;52:1237–52.
- [15] Green GE, Morteale KJ, Glickman JN, et al. Brenner tumors of the ovary: sonographic and computed tomographic imaging features. *J Ultrasound Med* 2006;25:1245–51. [quiz 1252–1254].
- [16] Willms AB, Schlund JF, Meyer WR. Endovaginal Doppler ultrasound in ovarian torsion: a case series. *Ultrasound Obstet Gynecol* 1995;5:129–32.
- [17] Huchon C, Fauconnier A. Adnexal torsion: a literature review. *Eur J Obstet Gynecol Reprod Biol* 2010;150:8–12.
- [18] Cohen SB, Weisz B, Seidman DS, et al. Accuracy of the preoperative diagnosis in 100 emergency laparoscopies performed due to acute abdomen in nonpregnant women. *J Am Assoc Gynecol Laparosc* 2001;8:92–4.
- [19] Mage G, Canis M, Manhes H, et al. Laparoscopic management of adnexal torsion. A review of 35 cases. *J Reprod Med* 1989;34:520–4.
- [20] Koonings PP, Grimes DA. Adnexal torsion in postmenopausal women. *Obstet Gynecol* 1989;73:11–2.
- [21] Sommerville M, Grimes DA, Koonings PP, et al. Ovarian neoplasms and the risk of adnexal torsion. *Am J Obstet Gynecol* 1991;164:577–8.
- [22] Eitan R, Galoyan N, Zuckerman B, et al. The risk of malignancy in postmenopausal women presenting with adnexal torsion. *Gynecol Oncol* 2007;106:211–4.