Int J Clin Exp Pathol 2022;15(7):289-295
www.ijcep.com /ISSN:1936-2625/IJCEP0139958

Case Report

Chronic ovarian torsion after vaginal hysterectomy: a case with metastatic serous ovarian cancer

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Received October 20, 2021; Accepted March 16, 2022; Epub July 15, 2022; Published July 30, 2022

Abstract: Ovarian torsion refers to partial or complete rotation of the ovary on its ligamentous support, often resulting in partial or complete obstruction of its blood supply. It is one of the most common gynecologic surgical emergencies and may affect females of all ages, but it is relatively rare in postmenopausal women. Chronic adnexal torsion with complete occlusion of the ovarian blood supply results in necrosis and loss of ovarian function. Clinical symptoms and signs are not specific and definitive diagnosis is often challenging. In this case report, we present a 65-year-old woman with chronic ovarian torsion that was detected 7 years after vaginal hysterectomy. Approximately 3% of postmenopausal cases with adnexal torsion are associated with malignancy. Ovarian torsion incidence is low in postmenopausal ovarian cancer due to the progression of accompanying inflammation, which causes immobility of the ovarian mass.

Keywords: Ovarian torsion, serous ovarian cancer, metastatic, postmenopausal, vaginal hysterectomy

Introduction

Ovarian torsion is a surgical emergency that is rare in postmenopausal women and is not easy to diagnose due to the lack of specific clinical findings. The incidence of adnexal torsion is unknown. Ovarian torsion accounted for 2.7% of emergency surgeries. Risk factors for ovarian torsion include the presence of a mobile ovarian mass and a history of previous ovarian torsion, but normal ovaries can undergo torsion [1].

Although the incidence of adnexal mass is highest in young women, the suspicion of malignancy increases when torsion is detected in older women since approximately 30% of adnexal masses are associated with malignancy. More attention should be paid to the detection of malignancy in postmenopausal women by ultrasound examination, especially in cases with a large mass, thick septa, and in the presence of solid components accompanied by acid [2].

Preserving the ovaries during a hysterectomy performed for benign reasons is an increasingly preferred approach in women under the age of 50. However, prophylactic salpingo-oophorectomy has been shown to reduce the risk of ovarian cancer, breast cancer, and the rate of possible adnexal surgery. It is also important to note that the majority of adnexal masses requiring surgery after some time of hysterectomy are ultimately benign ovarian masses [3]. For malignant masses diagnosed by frozen section, further surgery and treatment are required.

Primary surgical cytoreduction followed by systemic chemotherapy is the preferred initial management for women with metastatic serous ovarian cancer. Some patients who are not candidates for surgery may be considered to achieve neoadjuvant chemotherapy. Following the documentation of treatment response, the patients may undergo interval debulking followed by additional chemotherapy cycles. In general, chemotherapeutic treatment is preferred to begin as soon as feasible after surgery. The standard approach to treatment for advanced-stage serous ovarian cancer is to use a platinum agent with a taxane. The efficacy of the combination regimen was demonstrated in meta-analyses that it resulted in a 55% relative risk reduction for mortality as com-

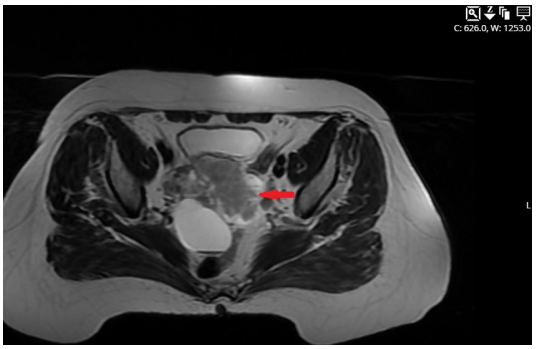

Figure 1. MRI image of ovarian mass, axial section.

pared with monotherapy using neither platinum nor taxane [4]. Angiogenesis inhibitors (e.g bevacizumab) [5, 6] and poly (ADP-ribose) polymerase (PARP) inhibitors (e.g. niraparib) [7] may be used in maintenance treatment in selected patients at higher risk of recurrence since these agents afford a progression-free survival (PFS) advantage. Moreover, median PFS was prolonged with high-dose chemotherapy, hematopoietic cell transplantation [8], dose-dense chemotherapy [9], tyrosine kinase inhibitors [10], and PARP inhibitors compared with standard therapy, but there has been no demonstrable improvement in overall survival. BRCA carriers who have responded to platinum-based chemotherapy are advised to use olaparib for maintenance treatment [7]. In the SOLO-1 study, olaparib monotherapy demonstrated a 70% reduction in the risk of disease progression or death compared to placebo. But the optimal agent in this setting is not clear, as no comparative efficacy trials have been documented. Another popular attempt at additional chemotherapy is HIPEC (hyperthermic intraoperative intraperitoneal chemotherapy) which has major concerns due to potential morbidity and lack of randomized trials so it requires further evaluation. Until now, all attempts to incorporate additional chemotherapy agents into the first-line treatment of advanced ovarian cancer have not been successful.

Adnexal torsion after hysterectomy is a rare event, and its incidence after vaginal, abdominal, or laparoscopic hysterectomy has not been determined. Ravid et al. reported the rate of adnexal torsion after vaginal hysterectomy as 0.06% [11]. Within the experiences of our clinic and doctors, who have been experienced in the field of gynecological oncology for 25 years, the current case is the first. When we consider the total rates of our 25-year cases, and that over a thousand gynecological cases are done per year, the fact that a single case has been seen in a clinic with so many cases shows us how rare it is. Therefore, we present this case as an example of torsion due to ovarian cancer after vaginal hysterectomy.

A 65-year-old woman had chronic ovarian torsion that was detected 7 years after vaginal hysterectomy. The final pathology report revealed a malignant serous ovarian tumor. We also review the topic in the light of a brief literature review.

Case presentation

A 65-year-old woman was admitted to our outpatient clinic with groin pain and fever that had been present for 2 weeks. She was para 2 with two live births delivered by vaginal delivery and had been in menopause for 13 years. The patient's consent was obtained. There was no family history of the tubo-ovarian tumor. Her Body Mass Index (BMI) was 29.7 kg/m^2. She had tenderness in the right pelvic area without defense or rebound on physical examination. The gynecologic examination did not reveal vaginal bleeding or any type of vaginal discharge. We noted that she had undergone a vaginal hysterectomy due to uterine prolapse 7 years ago. On transvaginal ultrasound, the uterus was not observed; there was a right adnexal mass which was 93×61 mm in diameter with irregular borders and solid areas. Color doppler ultrasound showed blood flow within the adnexa. 25 mm of free fluid was observed in the pouch of Douglas. The left ovary could not be observed. She underwent a magnetic resonance imaging (MRI) scan. A lobulated contoured mass originating from the vaginal stump was obliterating the rectovesical area (90×88×64 mm in size), and the mass consisted of cystic areas and a heterogeneous irregular solid component possessing contrast enema. A mass (13×10 mm in size) compatible with lymphadenopathy was detected [**Figure 1**] on the left iliac area. Laboratory analysis of tumor markers revealed the following: Ca 125 = 137.9 U/ml (0-35 U/ml), Ca 72-4 = 20.6 U/ml (0-8.2 U/ml), Ca 15-3 = 54.7 U/ml (0-34.5 U/ml), and Ca 19-9 = 9.9 U/ml (0-34 U/ml). Other blood tests, including complete blood

Table 1. Patient results of blood examination

Blood Test	Patient's results	Reference
Hemoglobin	13.6 g/dL	12-16 g/dL
White blood cell count	7130/mm^3	4370-9680/mm^3
Platelet	270000/mm^3	150000-450000/mm^3
Sedimentation	11	0-20
C-reaktif protein	12.00 mg/L	0-5 mg/L
AST	24	0-34
ALT	29	10-49
Albumin	44.80 g/L	32-48 g/L
Ca-125	137.9 U/ml	0-35 U/ml
Ca 72-4	20.6 U/ml	0-8.2 U/ml
Ca 15-3	54.7 U/ml	0-34.5 U/ml
Ca 19-9	9.9 U/ml	0-34 U/ml

Note: Ca: Carbohydrate antigen

count indices, C-reactive protein levels, and erythrocyte sedimentation rate were all within normal limits. The patient's results of blood examination are presented in **Table 1**.

We performed an elective diagnostic laparotomy to rule out occult malignancy. We did not notice any pathology related to the left tube and ovary. However, the bladder peritoneum, sigmoid colon, small intestines, and omentum had formed a conglomerate mass in the right adnexal region. Additionally, we observed a 10-cm solid mass in the right adnexal region, which was adherent to the bladder and the ovarian fossa posteriorly, and to the sigmoid colon and lateral pelvic wall. The right ovary and tube were found to be twisted-with 3 rotations around their axis after dissection of adhesions. The right adnexa was detorsed, and bilateral salpingo-oophorectomy was performed.

We sent a sample from the mass in the right ovary for the frozen section intraoperatively. The frozen section report revealed that the specimen was unable to differentiate between benign or malignant tissue so the operation was completed without additional surgical staging. The final pathology report revealed the following: high-grade serous carcinoma, right adnexal torsion, serous tubal intraepithelial carcinoma in the right tube, serous carcinoma implantation on the serosa of the left ovary, and serous carcinoma metastasis in the abdominal wall. The tumor in the right ovary had a size of 110×70×50 mm and demonstrated signs of torsion macroscopically. The cross-sectional surface of the ovary was completely hemorrhagic. Immunohistochemical staining was positive for WT-1 and mutant P53, which were applied for definitive diagnosis (**Figures 2** and **3**). Our diagnosis was malignant adnexal mass. The tumor was diagnosed as stage 3A. The patient was discharged from the hospital on the 3rd postoperative day. Her medical data were re-evaluated by the multidisciplinary oncology council, and she was referred to the medical oncology department for adjuvant therapy.

Discussion

The current case report showed that ovarian torsion in a postmenopausal woman is an important issue because of its risk of malignancy. Ovarian torsion is usually a benign phenomenon and malignant ovarian tumors associated with torsion are rare. The incidence of ovarian torsion is low in postmenopausal women as bilateral salpingo-oophorectomy is usually performed together with hysterectomy to prevent ovarian cancer [12-14]. Ovarian torsion during the premenopausal period is unlikely to be malignant, with an incidence of 2.4-5%. The possibility of torsion is low in malignant masses, as malignant masses that are rich in inflammatory mediators and adherent to adjacent structures are more likely to be immobile. Conservative treatment is preferred in reproductive-aged women to preserve ovarian function. However, the risk of ovarian malignancy of the twisted ovary is higher in postmenopausal women. Therefore, bilateral salpingo-oophorectomy is performed to rule out occult malignancy, and ultimately, to prevent a recurrence. Laparotomy with a radical approach is preferred most commonly in postmenopausal women possessing a 22% to 30% risk of malignancy [2, 12-15].

Adnexal torsion is a gynecological emergency associated with acute abdominal pain due to the rotation of the ovary, fallopian tube, or cystic mass around the vascular axis. Torsion may occur due to prolonged rotation of the utero-ovarian ligament or an increase in the size of an adnexal mass, resulting in tissue necrosis. Although adnexal torsion is often detected in the reproductive period, it is rare in postmenopausal women, with an incidence of 2.7%. The main clinical symptom is acute, nonspecific

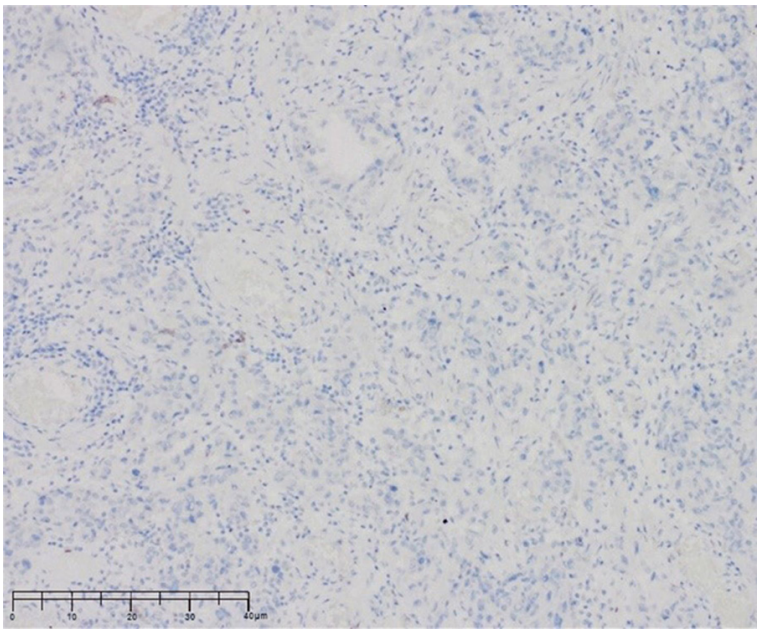

Figure 2. Serous tubal intraepithelial carcinoma. p53 staining shows positive immunoreactivity with malignant cells (×4).

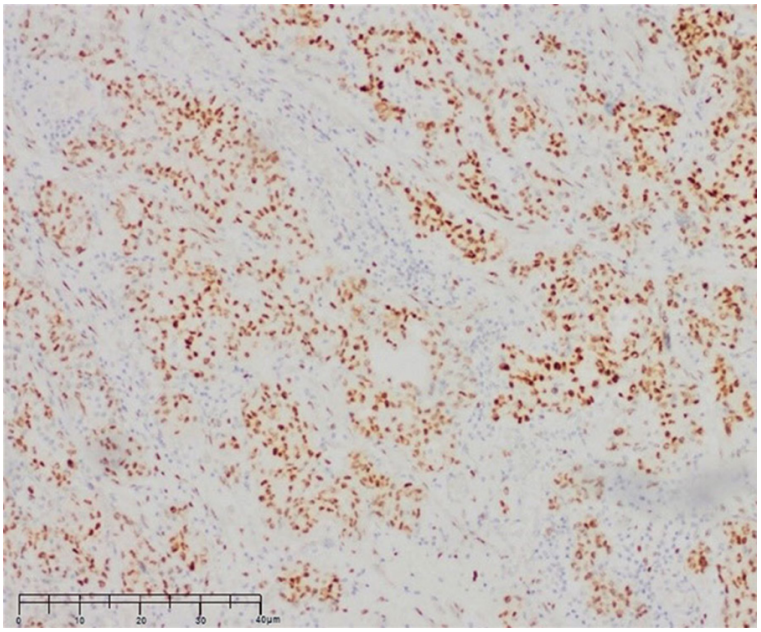

Figure 3. High-grade serous ovarian carcinoma. Malignant cells show severe nuclear atypia and significant nuclear pleomorphism with large, bizarre forms (WT1 ×40).

pain in the lower abdomen, which usually lasts for a few hours and can last 2 days. Although severe abdominal pain can develop and persist in some patients, it may also be recurrent and intermittent in cases with partial adnexal torsion. Moreover, adnexal torsion may trigger autonomic nervous system reflexes, including nausea, vomiting, anxiety, and tachycardia, in an estimated 7% of individuals [2, 16, 17]. Abdominal pain caused by torsion may present with different clinical manifestations during the reproductive and menopausal periods. While the pain often progresses to a sharper and more severe form in the reproductive period, it may be persistent in the postmenopausal period. Therefore the presence of acute abdominal pain in women of reproductive age enables early diagnosis of torsion and the time from the onset of torsion to surgery shortens. However, nonspecific clinical findings prolong this period resulting in a delay in diagnosis in postmenopausal women. As the lymphatic flow and adnexal blood flow are obstructed, the size of the mass can increase rapidly, thereby making it possible to palpate such masses in the pelvis. White blood cell count may increase as a result of adnexal ischemia and necrosis, but there are no specific laboratory findings indicative for torsion. A complete blood count showing leukocytosis may prove valuable [2, 12]. Symptoms and laboratory examinations are often non-diagnostic.

Our patient also had non-specific signs of partial torsion. The main clinical findings suggesting malignancy in our patient were elevated Ca125 level and an increase in ovarian size with the presence of irregular borders and solid areas on ultrasonography. Since color Doppler ultrasound showed blood flow, she had not undergone operation urgently, and we performed elective diagnostic laparotomy to rule out occult malignancy.

According to Cohen et al., the preoperative diagnosis of ovarian torsion can be confirmed in only 44% of women. They reported that torsion must be confirmed during surgery and requires simultaneous diagnosis and treatment [18]. Laparoscopic surgery is most often performed in reproductive-age women, while laparotomy is usually preferred in postmenopausal women who may have malignant tumors to allow staging of tumor [2, 11].

Most adnexal masses that require reoperation after some time of hysterectomy are benign and originate from the ovary. Casiano et al. found that the incidence of oophorectomy after hysterectomy was 9.2%. The blood flow of the ovaries may be impaired after hysterectomy, which could impact the functions of the ovaries and could result in adnexal pathology. It must also be noted that women who suffer from adnexal lesions after hysterectomy often report to clinics with considerable delay [3, 19]. Our patient was also admitted to our clinic 7 years after vaginal hysterectomy.

The diagnosis of ovarian torsion is usually made by the clinical examination findings and diagnostic imaging modalities, in which ultrasound examination with color Doppler analysis takes great part. Ultrasonography is cheap, free of ionizing radiation exposure, and an easily available imaging technique that can be used for the diagnosis of ovarian torsion. On ultrasound, the most common findings of ovarian torsion are an enlarged adnexa with an ovarian mass, characterized by ovarian stromal edema with or without peripherally displaced antral follicles, the whirlpool sign, and free fluid in the pelvis [20], although these are not pathognomonic for torsion. Color doppler signals in the twisted ovary are slightly less common signs since the severity of the vascular impairment is variable. The number of twists and the tightness at the neck of the torsion can cause partial or complete vascular obstruction to determine the vascularization of the twisted organ [21]. Vascularization does not exclude torsion, with up to 60% of cases of ovarian torsion having normal blood flow [22]. Normal blood flow on color doppler ultrasound may lead to delay in the diagnosis of ovarian torsion; absence of blood flow may occur only as a late finding. Measured doppler findings can still be unreliable due to the dual blood supply from the uterine and ovarian arteries. Doppler ultrasound is not sensitive enough to be used as a rule-out test for ovarian torsion [23]. Moreover, ultrasonography is clinician-dependent and experience is important, so its role in the early diagnosis of adnexal torsion is not yet fully established.

Pena et al. stated that color doppler ultrasonography examination could not detect 60% of adnexal torsion; however, arterial flow disturbances are examined in the doppler examination with a positive predictive value of 100%; therefore, it may not detect previous venous deterioration [22]. They concluded that Doppler ultrasound was inefficient in both the establishment and ruling-out of adnexal torsion diagnosis. Additionally, it has been reported that neither computerized tomography nor MRI appear to contribute to the diagnosis of adnexal torsion [17]. The diagnosis is made definitively only during surgery.

Magnetic resonance imaging (MRI) is an additional imaging method commonly used in the emergency department to narrow the differential diagnosis. It may be the initial preoperative work-up test for some patients. The use of MR imaging is recommended to help detect the twisted vascular pedicle or thickened tube in subacute or chronic cases and patients with a suspected pelvic mass. MR imaging features of adnexal torsion include fallopian tube thickening which appears as a beak-like protrusion adjacent to the mass, smooth wall thickening of the twisted adnexal cystic mass, ascites, and uterine deviation to the twisted side. It can also present the imaging findings in adnexal torsion that are specific to hemorrhagic infarction by showing signal intensity changes within the lesions [24]. Recent studies have reported the usefulness of diffusion-weighted MR images in the evaluation of ovarian torsion [25]. Considering all these evaluations and our current case, torsion should be kept in mind in the differential diagnosis of patients presenting with acute or chronic pelvic and abdominal pain and whose ovaries and tubes were not removed after hysterectomy.

Post-hysterectomy adnexal torsion is a rare event and its incidence after vaginal hysterectomy is not established. Ravid et al. reported the rate of adnexal torsion after vaginal hyster-

ectomy as 0.06% [11]. They stated that the rate of adnexal torsion was higher after laparoscopic hysterectomy compared to other types of hysterectomy. The authors describe this situation as laparoscopy not using sutures, preservation of humidity, less adhesion formation, and greater ovarian mobility. In another study, only 1 of 132 cervical cancer patients reported suspected torsion after radical hysterectomy and ovarian transposition [26]. Since we use sutures during vaginal hysterectomy and this suture creates an inflammatory reaction, we may cause adhesions around the ovary to keep the ovary immobile. According to our clinical experience; this is the first case presented as chronic ovarian torsion underlying carcinoma after vaginal hysterectomy. We reviewed the patients who had been operated on due to torsion after hysterectomy between 2004 and 2021, and we noticed that all of the patients had undergone abdominal or laparoscopic hysterectomy.

Conclusion

Adnexal pathologies detected some time after hysterectomy carry a high possibility of malignancy, especially in postmenopausal women. Although these cases are considered rare in the literature, the risk for chronic or partial torsion should not be ignored, and physicians should be aware that alterations in physiologic and anatomic structures may contribute to malignant changes in the ovary. Regarding our experience and the literature review on this topic, it may be feasible to recommended bilateral salpingectomy with or without oophorectomy to such patients together with hysterectomy. The patient, especially postmenopausal cases with high risk for malignancy, should be informed about the risks and benefits of bilateral salpingectomy.

Disclosure of conflict of interest

None.

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