A Case Report of Premature Ovarian Failure by Asynchronous Bilateral Ovarian Torsion with Dermoid Cyst

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Abstract

Adnexal torsion is a serious condition and delay in surgical intervention may result in loss of ovary. Adolescents who have suffered from adnexal torsion may be at risk for asynchronous torsion of the contralateral adnexa. We report the case of asynchronous bilateral ovarian torsion in a 36-year-old woman who at the age 18 year had undergone a left adnexectomy by ovarian torsion. Due to delay of diagnosis, during the urgent surgery, the detorsion involved the spontaneous amputation of the adnexa. The final histological examination indicated the presence of dermoid cysts the size of 7 X 6 cm, widespread haemorrhagic necrosis, and absence of follicular tissue. The hormonal replacement therapy was prescribed to the patient for premature ovarian failure. When ovarian torsion is suspected, especially in young women with a single ovary, laparoscopy should be performed without delay to avoid catastrophic events. In the adolescent age group, conservative treatment of ovarian torsion is mandatory.

Keywords: Asynchronous bilateral ovarian torsion; Dermoid cyst; premature ovarian failure; Whirlpool sign.

Introduction

The adnexal torsion is the fifth most common gynecologic emergency. Thirty percent of all cases of adnexal torsion occur in females younger than 20 years [1].

This disease is considered a gynecological emergency because a diagnostic delay could permanently compromise the interested ovary. Early recognition and restoration of blood flow are important to avoid irreversible ovarian damage.

Asynchronous Bilateral Ovarian Torsion (ABOT) is defined as torsion of each ovary at different points of time. The possibility of ABOT is rare, but it is described in the literature [2]. The symptoms of ovarian torsion are often non-specific and may include acute onset of lower abdominal pain, nausea, and vomiting [3]. If torsion is prolonged, the adnexa can become necrotic and even infected, at which time the patient may exhibit signs of peritonitis [4].

In fertile women, an ovarian mass is a primary risk factor for torsion. Adnexal masses that twist are usually benign, with dermoid cyst and serous cystadenoma being the most commonly found pathology [5]. Torsion is unusual in patients affected by endometriosis or by malignant lesions, most probably because of the increased likelihood of local inflammation and adhesions that fix the mass.

Ovarian torsion can occur in females of all ages; however, women in their reproductive years have the highest prevalence.

Factors contributing to ABOT are still unknown. Higher recurrence rate has been noticed in patients with previously normal adnexa, especially in prepubertal girls [2]. The main factor that makes this entity stand out is the complete loss of ovarian tissue which can be devastating for patients. The loss of ovarian tissue can be a result of delay to surgery, of unrecognized fear of untwisting the adnexa, or failure to protect any residual ovary from subsequent torsion. The loss of ovarian tissue is an extremely rare event, and there are no guidelines or consent statements with regards to surgical approach to ABOT [6].

The most typical ultrasound findings of a twisted ovary were reported to be an enlarged and displaced ovary, ovarian edema, free fluid in the pelvis and the “Whirlpool Sign” which is thought to be due to the twisting of the vascular pedicle in cross-section [7].

Recognizing ultrasound signs of adnexal torsion is important so that surgery is not delayed.

There are conservative and definitive options for treatment of ovarian torsion. Conservative treatment includes only untwisting the adnexa and confirming viable adnexal tissue, untwisting the adnexa and aspirating any associated cyst, or untwisting and removing any associated cyst. Definitive treatment includes salpingectomy and/or oophorectomy. Age, future fertility, menopausal status and evidence of ovarian disease are all factors considered in the management decision [8].

We present the case of asynchronous bilateral ovarian torsion in a young woman resulting in Premature Ovarian Failure (POF).

**Case presentation**

A 36-year-old woman presented to emergency gynecologic unit with persistent pain on the right lower abdominal quadrant. In the previous 4 days, the woman had been admitted to another hospital with a diagnosis of haemorrhagic corpus luteum. Pain medications had been administered and a period of observation had been decided. The patient, however, continued to complain of pain in the right flank and decided to self-discharge from the hospital.

At the age of 18, the patient had undergone a left adnexitomy for a voluminous dermoid cyst. She had two term normal vaginal deliveries and had regular menstrual cycle.

In the gynecology department, we performed a transvaginal and transabdominal ultrasound. Ultrasonography indicated the presence of a voluminous unilocular cyst the size of 12.7x8.6 cm. The cystic formation extended from the pelvis on the right to the abdomen.

The cyst had mixed, non-uniform echogenicity with a densely echogenic tubercole projecting into the cystic lumen with acoustic shadowing (suspected for Rokitansky nodule) (Figure 1). Another characteristic was the presence of hyperechoic lines and dots and fat-fluid level with supernatant hyperechoic serum and down hypoechoic fluid (Figure 1). The mass was sure to the targeted pressure of the transvaginal probe. Free pelvic liquid was also displayed. Color Doppler sonogram revealed circular vessels in a whirlpool mass close to the right side of the uterus. This finding was attributable to the “Whirlpool Sign” which was compatible with a twisted structure (Figure 2). The abdomen was sore at palpation and Blumberg sign was positive. Based on these findings, ovarian torsion caused by adnexal mass was the likely diagnosis. Immediately we performed a video-laparoscopy. The right ovary was found to be torsed twice (Figure 3), cyanotic, and apparently necrotic (Figure 4). We tried the detorsion, but during untwisting, spontaneous amputation of ovary occurred. The adnexa was removed inside to endobag. At the inspection, the adnexa showed important hemorrhagic tissue associated to hair fragments (Figure 5). The pain resolved completely after surgery and the histopathologic diagnosis was the presence of dermoid cyst, haemorrhagic necrosis and absence of ovarian follicle. On discharge from hospital, we prescribed HRT to the patient for the premature ovarian failure.

![Figure 1: Unilocular cyst (12.7X8.6 cm) with echogenic tubercle projecting into the cystic lumen with acoustic shadowing (star) and hyperechoic lines and dots (arrow).](image1)

![Figure 2: Ultrasound image of “Whirlpool Sign”](image2)
Certain factors have been identified in literature as indicative of risk for ovarian torsion and ABOT. Adnexal masses that twist are usually benign: mature cystic teratomas, haemorrhagic cysts, and cystadenomas commonly predispose the ovary to swing on its vascular pedicle. Torsion is uncommon in patients with malignant ovarian mass, endometriosis, most probably because increased likelihood of local inflammation and adhesions that fix the mass [5]. Other reported risk factors are previous adnexal torsion (11-19%), conditions that are associated with enlarged ovaries such ovarian stimulation (0.8-1.3%) in the context of assisted fertilization programs, and polycystic ovary syndrome [5].

Mature cystic teratomas (dermoid cyst) account for 10 to 20% of all ovarian neoplasms. Torsion occurs in about 15% of dermoid cysts as a result of their high fat content, and pultaceous material along with bony tissue and teeth which makes the cyst heavy causing them to freely float in the abdominal cavity, leading to torsion of the adnexa [11].

Our patient had undergone a left adnexectomy at the age of 18 for ovarian torsion by dermoid cysts. Until two decades ago, the standard approach to twisted adnexa was salpingooophorectomy. In 1989, Mage et al. proved that detorsion and preservation of adnexa was an alternative mode of treatment and ever since it has become the method of approach by many ever since, replacing excision of adnexa whenever possible [12]. Despite this and many other early reports of success with detorsion, most patients continue to undergo oophorectomy. A recent examination of pediatric patients in the National Inpatient Sample (NIS) demonstrated that 15% of patients underwent detorsion, 6% underwent detorsion with oophoropexy, and 78% underwent oophorectomy [13]. The advantages of ovarian preservation are clear, as adnexal torsion occurs most often in adolescent girls and women of childbearing age. Detorsion of adnexa helps to preserve ovarian function and maintain fertility. It reduces the risk of premature ovarian failure and the ensuing complications [5]. The viability of an ovary declines as time elapses from the onset of pain to surgical detorsion; two retrospective studies suggest that a sharp decrease in ovarian function occurs 72 hours after the onset of symptoms [14,15]. In the case of our patient, it had been 4 days from hospitalization to surgery. Since the patient had only one ovary, it was important to preserve this structure in consideration of her young age. However, during the ovarian detorsion, we witnessed the spontaneous amputation of the adnexa. The final histological examination indicated the presence of dermoid cysts the size of 7 x 6 cm, widespread haemorrhagic necrosis, and absence of follicular tissue.

Ultrasound is the primary mode of evaluation of a female patient with lower abdominal pain because it is non-invasive, accessible, and cost-effective. In a retrospective cohort analysis of 322 women, presenting to a tertiary medical center with acute abdominal pain, the sensitivity of ultrasound for adnexal torsion was 84.4%, and the positive predictive value was 81.4% [16]. The most typical ultrasound findings of a twisted ovary were reported to be an enlarged ovary, unusual location of the ovary, ovarian edema, free fluid in the pelvis and “Whirlpool Sign”. Edematous ovary and/or tube, as well as positive whirlpool sign had the highest sensitivity and positive predictive value [16]. The presence of free pelvic fluid is usually associated with haemorrhagic ovarian tissue. In a recent study of Moro et
al, the free fluid in pelvis was present in the 71.3% of 315 patients with adnexal torsion. Another, the “whirlpool sign” was present in 78.8% of cases [7].

Ultrasound examination, persistent abdominal pain and the patient’s clinical history allowed us to suspect an ovarian torsion with dermoid cyst associated. Despite our rapid diagnosis and our intention to proceed with ovarian laparoscopic cleaning, the damage to the adnexa was such as to cause amputation of the same.

We can hypothesize that the ovary could have been saved if the patient had been operated on promptly. The patient’s symptoms, her medical history and ultrasound examination are essential information, which together can help in an appropriate diagnosis.

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