



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

Stefania Malmusi^{1*}; Mirvana Airoud¹; Rosamaria Pellegrini²; Maria Cristina Galassi¹

¹Department of Obstetrics and Gynecology, Sassuolo Hospital, Modena, Italy.

²Department of Obstetrics and Gynecology, University of Modena and Reggio Emilia, Modena, Italy.

*Corresponding Author(s): Stefania Malmusi

Department of Obstetrics and Gynecology, Sassuolo Hospital, 2, Francesco Ruini Street, 41049 Sassuolo (MO), Italy.

Tel +39-0536-846365;

Email: s.malmusi@ospedalesassuolo.it

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.

Received: Mar 08, 2021

Accepted: Mar 23, 2021

Published Online: Mar 26, 2021

Journal: Annals of Obstetrics and Gynecology

Publisher: MedDocs Publishers LLC

Online edition: <http://meddocsonline.org/>

Copyright: © Malmusi S (2021). *This Article is distributed under the terms of Creative Commons Attribution 4.0 International License*

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].



Cite this article: Malmusi S, Airoud M, Pellegrini R, Galassi MC. A Case Report of Premature Ovarian Failure by Asynchronous Bilateral Ovarian Torsion with Dermoid Cyst. *Ann Obstet Gynecol.* 2021; 4(1): 1027.

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 adnexectomy 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 tubercle 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 sebum 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.

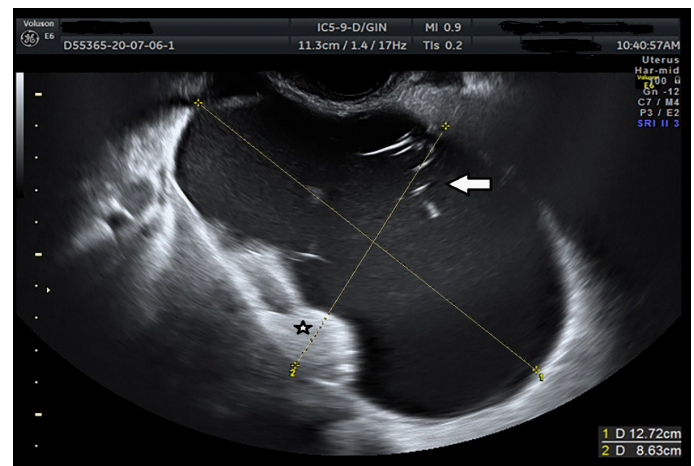


Figure 1: Unilocular cyst (12.7X8.6 cm) with echogenic tubercle projecting into the cystic lumen with acoustic shadowing (star) and hyperechoic lines and dots (arrow).

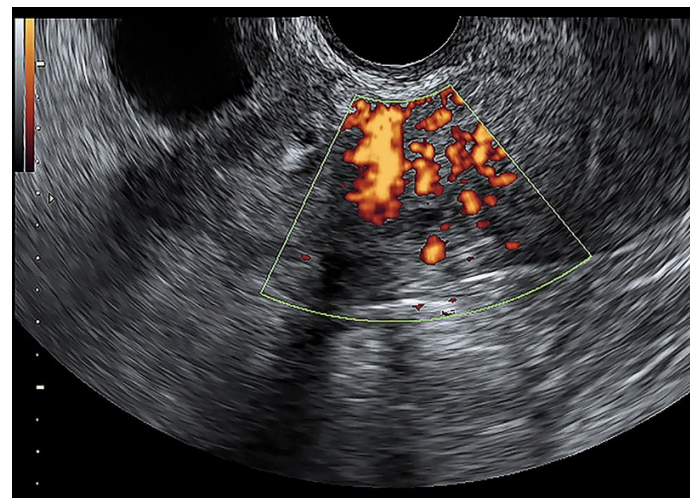


Figure 2: Ultrasound image of “Whirlpool Sign”.

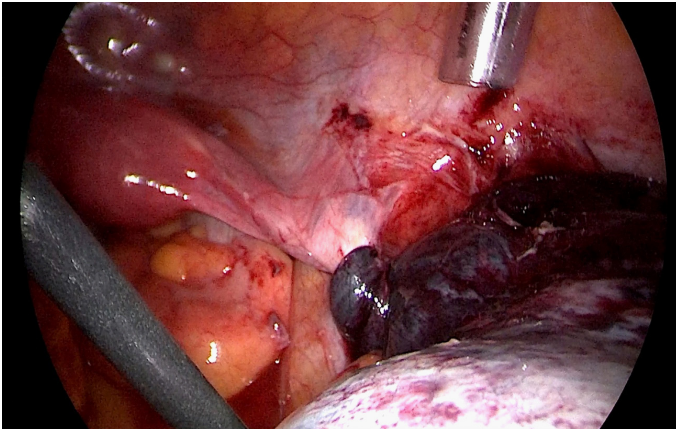


Figure 3: Laparoscopic image of right adnexal torsion (torted twice).

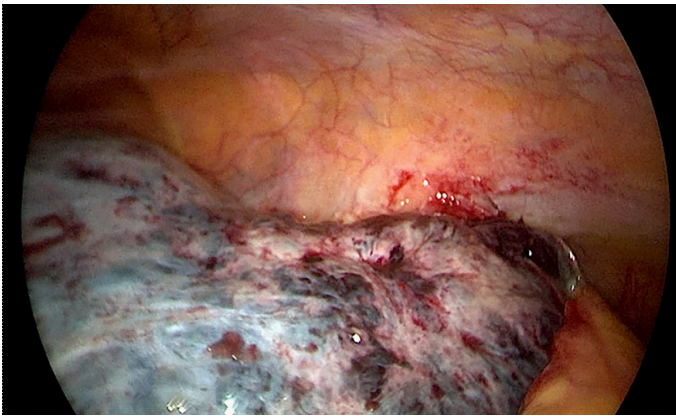


Figure 4: Laparoscopic image of cyanotic, and apparently necrotic right ovary.



Figure 5: Macroscopic appearance of right adnexa.

Discussion

Adnexal torsion is reported to be the fifth most common gynecologic emergency, with a prevalence of 2.7% [9]. It was previously reported that 70% of cases are diagnosed in women between 20 and 39 years of age, few occur in postmenopausal women, and about 15% of cases occur during infancy [10].

Asynchronous bilateral ovarian torsion presents a special sub-group of patients and 29 cases were reported in literature. Most publications reported cases of ABOT in the premenarchal age group [2]. Conservative treatment of ovarian torsion (with or without ovarian pathology predisposing to torsion) is manda-

tory, particularly in the pediatric age group, because ABOT is a rare but potentially catastrophic event.

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 pul-taceous 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 salpingo-oophorectomy. 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 cleansing, 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.

Acknowledgments

Stefania Malmusi, MD: Conception and design of the study. Analysis and interpretation of data. Drafting of the article.

Mirvana Airoud, MD: Substantial contributions to acquisition of data, to analysis and interpretation of data. Drafting of the article.

Rosamaria Pellegrini, MD: Substantial contributions to acquisition of data and interpretation of data.

Maria Cristina Galassi, MD: Approval of the version to be published; revising it critically for important intellectual content.

Disclosure

Stefania Malmusi reports non-financial support outside the submitted work.

Mirvana Airoud, Rosamaria Pellegrini and Maria Cristina Galassi report no conflict of interest for the present article.

References

1. Ashwal E, Hirsch L, Krissi H, Eitan R, Less S, Wiznitzer A, et al. Characteristics and management of ovarian torsion in premenarchal compared with postmenarchal patients. *Obstet Gynecol.* 2015; 126: 514-520.
2. Lucchetti MC, Orazi C, Lais A, Capitanucci ML, Caione P, Bakhsh H. Asynchronous bilateral ovarian torsion: Three cases, Three Lessons. *Case Rep in Pediatr.* 2017; 6.
3. Sheizaf B, Ohana E, Weintraub AY. Habitual adnexal torsions- recurrence after two oophoropexies in a prepubertal girl: A case report and review of the literature. *J Pediatr Adolesc Gynecol.* 2013; 26: e81-e84.
4. Tandulwadkar S, Shah A, Agarwal B. Detorsion and conservative therapy for twisted adnexa: Our experience. *J Gynecol Endosc Surg.* 2009; 1: 21-26.
5. Huchon C, Fauconnier A. Adnexal torsion: A literature review. *Eur J Obstet Gynecol Reprod Biol.* 2010; 150: 8-12.
6. Raicevic M, Saxena AK. *World J Pediatr.* 2017; 13: 416-420.
7. Moro F, Bolomini G, Sibal M, Vijayaraghavan SB, Venkatesh P, Nardelli F, et al. Imaging in gynecological disease: Clinical and ultrasound characteristics of adnexal torsion. *Ultrasound Obstet Gynecol.* 2020; 56: 934-943.
8. Sasaki KJ, Miller CE. Adnexal Torsion: Review of the Literature. *J Minim Invasive Gynecol.* 2014; 21: 196-202.
9. Breech LL, Hillard PJA. Adnexal torsion in pediatric and adolescent girls. *Curr Opin Obstet Gynecol.* 2005; 17: 483-489.
10. Khalil RM, El-Dieb LR. Sonographic and MRI features of ovarian torsion. *Egypt J Radiol Nucl Med.* 2016; 47: 621-629.
11. Rabinovich I, Pekar-Zlotin M, Bliman-Tal Y, Melcer Y, Vaknin Z, et al. Dermoid cysts causing adnexal torsion: What are the risk factors? *Eur J Obstet Gynecol Reprod Biol.* 2020; 251: 20-22.
12. Mage G, Canis M, Manhes H, Pouly JL, Bruhat MA. Laparoscopic management of adnexal torsion. A review of 35 cases. *J Reprod Med.* 1989; 34: 520-524.
13. Sola R, Wormer BA, Walters AL, Heniford BT, Schulman AM. National trends in the surgical treatment of ovarian torsion in children: an analysis of 2041 pediatric patients utilizing the Nationwide Inpatient Sample. *Am Surg.* 2015; 81: 844-848.
14. Rossi BV, Ference EH, Zurakowski D, Scholz S, Feins NR, Chow JS, et al. The clinical presentation and surgical management of adnexal torsion in the pediatric and adolescent population. *J Pediatr Adolesc Gynecol.* 2012; 25: 109-113.
15. Hubner N, Langer JC, Kives S, Allen LM. Evolution in the management of pediatric and adolescent ovarian torsion as a result of quality improvement measures. *J Pediatr Adolesc Gynecol.* 2017; 30: 132-137.
16. Bardin R, Perl N, Mashiach R, Ram E, Orbach-Zinger S, Shmueli A, Wiznitzer A, Hadar E. Prediction of Adnexal Torsion by Ultrasound in Women with Acute Abdominal Pain. *Ultraschall Med.* 2020; 41: 688-694.