A Case of Twisted Ovarian Cyst in a Young Patient and Review of the Literature

ABEF 1 Suriani Idris
EF 1,2 Suzanna Daud
B 1,3 Nurulhuda Ahmad Sani
E 3 Samantha Tee Mei Li

Corresponding Author: Suriani Idris, e-mail: surianiidris@uitm.edu.my
Financial support: None declared
Conflict of interest: None declared

Patient: Female, 16-year-old
Final Diagnosis: Serous cystadenoma ovarian cyst • twisted ovarian cyst
Symptoms: Abdominal pain
Medication: —
Clinical Procedure: Laparotomy salpingo-ophorectomy
Specialty: Obstetrics and Gynecology

Objective: Management of emergency care
Background: Adnexal torsion is a gynecologic emergency that dictates immediate surgical intervention. Twisted ovarian cysts are rare in adolescents, but they can cause significant distress to patients. Idiopathic ovarian torsion is uncommon in adolescents and the incidence is reportedly higher in women aged 20 to 40 years. Most twisted cysts in adolescents are benign. In the past, oophorectomy was commonly performed for a torsed ovary, but currently there is a trend toward ovary preservation. The diagnosis of twisted ovarian cyst is based on pathognomonic symptoms and findings from a focused clinical examination and ultrasound. Because the differential diagnosis includes acute appendicitis and gastroenteritis, referral to a gynecologist may be delayed.

Case Report: We present the case of a huge, twisted ovarian cyst in a 16-year-old girl who presented with an acute abdomen. The initial diagnosis was acute appendicitis, which delayed her referral to a gynecologist. An emergency laparotomy and left salpingo-ophorectomy were performed. The histopathology of the ovarian cyst was reported as serous cystadenoma.

Conclusions: The optimal management of a twisted ovarian cyst in adolescents is the subject of much debate. Here, we review the literature on ovarian torsion in children and adolescents. The patient in the case we present had a twisted ovarian cyst that was managed with salpingo-ophorectomy.

Keywords: Adolescent • Cystadenoma, Serous • Ovary

Full-text PDF: https://www.amjcaserep.com/abstract/index/idArt/933438
Background

Torsion of an ovarian cyst is rare in adolescents, but even though it is uncommon, it can cause significant distress to patients. The estimated incidence of ovarian torsion in females aged 1 to 20 years is 4.9 per 100,000 [1]. Most of these masses are benign; however, malignancy must be considered in the differential diagnosis in any adolescent who presents with an ovarian tumor.

A patient with a torsed ovarian cyst may present with non-specific symptoms, including abdominal pain, vomiting, or lethargy. Torsion of an ovarian cyst also can produce no symptoms and be an incidental finding on ultrasound. In children, there has been a shift toward conservative surgery for future fertility preservation. These patients may be under the care of a gynecologist or a pediatric surgeon. We report the case of a young patient with a twisted ovarian cyst. We also present a review of the literature on ovarian torsion in children and adolescents.

Case Report

A 16-year-old girl presented with sudden onset of severe lower abdominal pain on the third day of her menstrual cycle, which was associated with vomiting. The pain was localized and colicky and she reported having a few similar episodes in the past 6 months, which resolved spontaneously. She had not sought medical attention for the pain because she believed it was normal, probably related to menstruation, and it usually resolved spontaneously. The patient had no diarrhea or abnormal vaginal discharge. Her pain score was 10 and she reported that it got worse when she moved. She had no other bowel or urinary symptoms.

The patient had no history of abdominal trauma. Her menarche occurred at age 13 years and her menstrual cycles were regular, every 28 days with 7 days of flow. She had never taken hormonal contraceptives. She had no other gynecologic symptoms, such as intermenstrual bleeding or dysmenorrhea. She had no significant past medical, surgical, or drug history, nor any relevant family history. She was generally fit and well.

Initially, the patient was diagnosed with acute appendicitis at a district hospital, which delayed her referral to a gynecologist. At admission, the patient was febrile with a temperature of 37.7°C, blood pressure of 108/72 mm Hg, and a pulse rate of 94 bpm. An abdominal examination revealed tenderness on superficial palpation of the suprapubic and right iliac fossa. An abdominal ultrasound was performed and the only notable finding in the report was sediment in the bladder with no hydronephrosis or free fluid. The patient's white blood cell count was 12.9×10^9/L, her hemoglobin level was 11.5 g/L, and her platelet count was 292×10^9/L.

An abdominal ultrasound revealed an adnexal mass that measured 11×7 cm and contained a hyperechoic area, and free fluid in the pouch of Douglas to a depth of 1 cm. Also, fluid-debris was seen in the peripherally displaced follicular cysts. The adnexal mass was separate from the uterus, which measured 5×3 cm. The patient's right ovary was normal. An emergency laparotomy was scheduled, based on a preoperative diagnosis of a hemorrhagic or twisted dermoid cyst.

Intraoperatively, the patient was found to have a blue-black, gangrenous-looking left ovarian mass that measured 12×10 cm (Figure 1) and a twisted left fallopian tube. There was also a 100-mL hemoperitoneum. The patient's contralateral fallopian tube and uterus were normal, with no signs of endometriosis. Detorsion of the ovarian cyst was performed, but after the procedure, no viable ovarian tissue was left. Therefore, the team proceeded with a left salpingo-oophorectomy.

On examination, the patient's temperature was 37.7°C, her blood pressure was 108/72 mm Hg, and her pulse rate was 94 bpm. An abdominal examination revealed tenderness on superficial palpation of the suprapubic and right iliac fossa. However, there was no guarding of the abdomen. A mass of the size of a 20-week gravid uterus was found, which could be moved from side to side. No pelvic examination was performed. A complete blood count showed that the patient had a white blood cell count of 12.9×10^9/L, her hemoglobin level was 11.5 g/L, and her platelet count was 292×10^9/L.

Intraoperatively, the patient was found to have a blue-black, gangrenous-looking left ovarian mass that measured 12×10 cm (Figure 1) and a twisted left fallopian tube. There was also a 100-mL hemoperitoneum. The patient's contralateral fallopian tube and uterus were normal, with no signs of endometriosis. Detorsion of the ovarian cyst was performed, but after the procedure, no viable ovarian tissue was left. Therefore, the team proceeded with a left salpingo-oophorectomy.

The patient had no complications and was discharged on postoperative day 2. The histopathology report revealed an infarcted ovarian cyst with features suggestive of serous cystadenoma and an ischemic/infarcted left fallopian tube.

Figure 1. Gangrenous-looking left ovary and fallopian tube.
Discussion

Patients with ovarian cysts can present with no symptoms or a variety of them as a result of cyst rupture, hemorrhage, or torsion [2]. Idiopathic ovarian torsion is uncommon, particularly in girls younger than age 18 years [3]. It is even rarer for a huge ovarian cyst to be twisted, as was the presentation in the patient in our case.

The findings of ovarian mass associated with tenderness and tachycardia often suggest ovarian torsion [2]. However, the absence of tachycardia in our patient did not exclude torsion.

In the present case, the palpable mass corresponded to a 20-week-size gravid uterus. Hence, the possibility of ovarian malignancy should have been kept in mind. Oltmann et al reported an overall malignancy rate of 1.8% in twisted ovaries in children and found that there was a greater likelihood of malignancy in ovaries that measured ≥8 cm [4].

Even when a patient has obvious symptoms, diagnosing ovarian torsion in the emergency room can be difficult. The presence of characteristic symptoms (abdominal pain, nausea, and vomiting) in a young patient with an ovarian cyst >5 cm, coupled with a high level of awareness and clinical suspicion, is what most often leads to a diagnosis of twisted ovarian cyst.

There are several lessons to be drawn from the present case study. First, in a child or a woman who has a combination of abdominal pain (especially in the iliac fossa region), nausea, and vomiting, an urgent gynecological evaluation is required. Second, a thorough abdominal examination by an experienced clinician and imaging – preferably bedside ultrasound by an experienced sonographer – are the best ways to detect a pelvic mass. Third, the presence of a painful ovarian or adnexal mass or cyst should raise the possibility of ovarian torsion, which is a gynecological emergency. Finally, any subsequent pain in a patient should be treated as a surgical emergency.

Most ovarian cysts are diagnosed preoperatively using transabdominal ultrasound. A study by Kiechli-Kohlendorfer et al [5] found that 12 patients younger than age 17 years who were diagnosed with ovarian torsion had gray-scale and color Doppler pelvic sonography that showed an enlarged ovary with peripheral cysts. High-resolution sonography detected fluid-debris, which is a sonographic sign of ovarian torsion with or without associated teratoma [5].

Similarly, in the present case, transabdominal ultrasound showed peripherally displaced follicles (no image available). In addition, free fluid also was seen in the pouch of Douglas, a clue to diagnosis of ovarian torsion, as previously described by Badin et al [6].

Other ultrasound findings also have been studied for predicting adnexal torsion, such as enlargement of the ovary, ovarian and/or tubal edema, a whirlpool sign (a clockwise or counter-clockwise wrapping of the hypoechoic vessels around the central axis with or without color Doppler), ovarian blood flow on Doppler, and free fluid in the cul-de-sac [6]. An edematous ovary and/or tube and a positive whirlpool sign have the highest sensitivity and positive predictive value for adnexal torsion [6].

When a definitive diagnosis cannot be established, computed tomography (CT) and magnetic resonance imaging (MRI) can narrow the differential diagnosis [7]. However, these tests are not routinely done in an acute setting because of their cost, and the technology is not readily available in all hospitals. Nevertheless, it is essential to note that in the presence of complex or solid lesions, MRI or CT scans are recommended to distinguish an ovarian malignancy from a benign lesion [8].

Other investigations, such as with serum tumor markers, can identify different types of ovarian pathology in a child with an ovarian tumor, but these markers can be elevated in association with both benign and malignant lesions [9]. Furthermore, a huge cyst in a very young child should heighten suspicion before surgery about the possibility of malignancy. Elevated beta-human chorionic gonadotropin (beta-HCG), alpha-fetoprotein (AFP) and cancer antigen-125 (CA-125) levels are significantly associated with ovarian malignancy, but an elevated carcinoembryonic antigen (CEA) level is not [9]. Because of our patient’s acute presentation, waiting for results of tumor marker testing was unnecessary.

In the present case, the patient’s leucocyte counts were normal, whereas in some reported cases of torsion of the ovary, an elevated total leukocyte count was found [2]. This may be due to an inflammatory reaction of the peritoneum to the twisted adnexal mass. However, to date, no correlation has been found between leukocytosis and tissue necrosis [10].

Most patients suspected of having ovarian torsion will undergo emergency surgery with oophorectomy as the procedure of choice for a necrotic-looking ovary. However, the gross appearance of a necrotic ovary does not correlate with the future ability of the ovary to form follicles. Therefore, appearance should not dictate ovarian resection [3]. Successful conservative surgery for ovarian torsion, including detorsion of the affected ovary with enucleation done simultaneously or following detorsion, has been reported [4,11,12].

Previously, surgical management has been done variously by pediatric surgeons and gynecologists. The latter professionals are more likely to favor ovarian preservation for torsion than are general pediatric surgeons [3]. Recently, however, Alberto et al [13] found decreasing rates of oophorectomy...
performed by pediatric surgeons. This indicates that pediatric surgeons potentially have shifted their management toward the new American College of Obstetricians and Gynecologists recommendations for tissue preservation.

Ovarian detorsion and preservation may be the procedure of choice in children with ovarian torsion. McGovern et al reported that 379 of 979 cases of adnexal torsion were successfully managed conservatively with detorsion and adnexal preservation (with or without cystectomy, ovariopexy [oophoropexy]) or with additional cyst aspiration [14]. Further, detorsion of a twisted ovarian cyst does not increase the risk of pulmonary embolism (PE) more than excision of ovarian torsion without untwisting [13]. PE can still occur in cases in which an adnexal mass is resected without untwisting. Therefore, detorsion of an adnexal mass can be safely performed during the surgery.

Abes et al reported another surgical approach, in which they performed oophoropexy in both detorsed and contralateral ovaries. There were no complications postoperatively [15]. This procedure could be beneficial, especially in young patients, because there is a risk of recurrence of torsion in the contralateral ovary. However, it is not practiced routinely because the latest systematic review suggested a lack of evidence to support oophoropexy in patients who first presented with ovarian torsion, although previous reports indicated that it might be beneficial.

The vast majority of ovarian cysts diagnosed in patients of all ages are benign. As in the present case, histopathological examination showed serous cystadenoma. Another study [5] revealed findings of benign ovarian cystic teratoma.

Serous cystadenomas are the most frequent types of epithelial tumors. They occur in adults of all ages, with reported mean ages ranging from 40 to 60 years. These tumors are bilateral in 12% to 23% of cases, which accounts for the dilemma in managing patients with them, given the risk of recurrence. Treatment options include unilateral salpingo-oophorectomy or ovarian cystectomy [16]. Recurrence is uncommon, although it remains to be considered in these patients in the future. Recurrence risk can be increased by either incomplete resection or the development of a new primary tumor [16].

Oltmann et al found a 1.8% incidence of malignancy in 707 patients with ovarian torsion. The types of malignancy included juvenile granulosa cell tumors, dysgerminomas, serous borderline tumors, and undifferentiated adenocarcinomas.

There is also no evidence to justify performing an oophorectomy during detorsion because of concern about leaving behind a malignancy, as the incidence of malignancy is low. Oophorectomy should only be considered in cases in which intraoperative findings clearly suggest malignancy [3]. Unfortunately, our patient had a left-sided oophorectomy because her ovary appeared necrotic during surgery.

The prognosis is good in young patients who have a twisted ovarian cyst. The systemic review showed that gross appearance intraoperatively did not correlate with long-term viability or ovarian function. There are reported cases of spontaneous pregnancy after ovary detorsion and of retrieval of oocytes from previously twisted ovaries [1].

Conclusions

Ovarian torsion can lead to non-specific symptoms, which can delay the diagnosis. Clinicians must be careful not to miss the diagnosis of acute ovarian torsion in a young patient and they should bear in mind that even a huge cyst could still be twisted. Current evidence on management of ovarian torsion supports ovarian detorsion rather than oophorectomy because the ovary may retain viability after detorsion and the risk of malignancy is low. Unfortunately, that procedure could not be performed on our patient because her condition was misdiagnosed and identification of her ovarian torsion therefore was delayed.

Acknowledgements

The authors thank Associate Professor Bahiyah Abdullah, the head of our department, who provided support.

Declaration of Figures’ Authenticity

All figures submitted have been created by the authors who confirm that the images are original with no duplication and have not been previously published in whole or in part.

References:

1. Guthrie BD, Adler MD, Powell EC. Incidence and trends of pediatric ovarian torsion hospitalizations in the United States, 2000-2006. Pediatrics. 2010;125(3):532-38
2. Bottomley C, Bourne T. Diagnosis and management of ovarian cyst accidents. Best Pract Res Clin Obstet Gynaecol. 2009;23(5):711-24
3. Dasgupta R, Renaud E, Goldin AB, et al. Ovarian torsion in pediatric and adolescent patients: A systematic review. J Pediatr Surg. 2018;53(7):1387-91
4. Oltmann SC, Fischer A, Barber R, et al. Pediatric ovarian malignancy presenting as ovarian torsion: Incidence and relevance. J Pediatr Surg. 2010;45(1):135-39
5. Kiechl-Kohlendorfer U, Maurer K, Unsinn KM, et al. Fluid-debris level in follicular cysts: A pathognomonic sign of ovarian torsion. Pediatr Radiol. 2006;36(5):421-25
6. Bardin R, Perl N, Mashlach R, et al. Prediction of adnexal torsion by ultrasound in women with acute abdominal pain. Ultrascall Med. 2020;41(6):688-94
7. Iraha Y, Okada M, Iraha R, et al. CT and MR Imaging of gynecologic emergencies. Radiographics. 2017;37(5):1569-86
8. Al Jama FE, Al Ghamdi AA, Gasim T, et al. Ovarian tumors in children and adolescents – a clinical study of 52 patients in a university hospital. J Pediatr Adolesc Gynecol. 2011;24(1):25-28
9. Oltmann SC, Garcia N, Barber R, et al. Can we preoperatively risk stratify ovarian masses for malignancy? J Pediatr Surg. 2010;45(1):130-34
10. Oelsner G, Shashar D. Adnexal torsion. Clin Obstet Gynecol. 2006;49(3):459-63
11. Galinier P, Carfagna L, Delsol M, et al. Ovarian torsion. Management and ovarian prognosis: A report of 45 cases. J Pediatr Surg. 2009;44(9):1759-65
12. Huchon C, Fauconnier A. Adnexal torsion: A literature review. Eur J Obstet Gynecol Reprod Biol. 2010;150(1):8-12
13. Alberto EC, Tashiro J, Zheng Y, et al. Variations in the management of adolescent adnexal torsion at a single institution and the creation of a unified care pathway. Pediatr Surg Int. 2021;37(1):129-35
14. McGovern PG, Noah R, Koenigsberg R, et al. Adnexal torsion and pulmonary embolism: Case report and review of the literature. Obstet Gynecol Surv. 1999;54(9):601-8
15. Abeş M, Sarihan H. Oophoropexy in children with ovarian torsion. Eur J Pediatr Surg. 2004;14(3):168-71
16. Seidman JD, Ronnett BM, Shih LM, et al. Epithelial tumors of the ovary, in Blaustein’s Pathology of the Female Genital Tract. Kurman RJ, Hedrick Ellenson L, Ronnett BM, eds. Springer International Publishing; 2019:841-966