





Contents lists available at ScienceDirect

Gynecology and Minimally Invasive Therapy

journal homepage: www.e-gmit.com



Case report

Laparoendoscopic single-site surgical cystectomy of a twisted ovarian dermoid cyst during early pregnancy: A case report and literature review



Dah-Ching Ding a, b, *, Yu-Hsiun Chang b, c

- ^a Department of Obstetrics and Gynecology, Buddhist Tzu Chi General Hospital, Tzu Chi University, Hualien, Taiwan, ROC
- ^b Institute of Medical Sciences, Tzu Chi University, Hualien, Taiwan, ROC
- ^c Department of Pediatrics, Buddhist Tzu Chi General Hospital, Tzu Chi University, Hualien, Taiwan, ROC

ARTICLE INFO

Article history Received 25 August 2015 Received in revised form 18 December 2015 Accepted 29 December 2015 Available online 4 February 2016

Keywords: dermoid cyst laparoscopy pregnancy single port torsion

ABSTRACT

In pregnancy, the most frequently discovered ovarian tumor is mature cystic teratomas. Acute ovarian torsion in a pregnant patient is rare and is difficult to diagnose. Recent studies have demonstrated that laparoscopy during pregnancy is safe and confers many advantages over laparotomy. We report a patient with acute ovarian teratoma torsion treated with ovarian cystectomy via a single-port laparoscopy and review of the literature. A 17-year-old woman, gravida 1, in her 12th week of pregnancy, came to our emergency room with severe abdominal pain and nausea. Torsion of ovarian teratoma (5 cm in diameter) was suspected. The patient underwent emergent laparoendoscopic single-site surgery (LESS) under general anesthesia, and the detorsed cyst was successfully excised and removed intact through the single port. This was enabled by the endobag technique, thus preventing spillage of the cyst content into the abdominal cavity. The fetus tolerated surgery well without complications and was term delivered uneventfully. The literature review revealed that the advantages of using LESS in pregnant patients are that this technology is safe for both the mother and the fetus, and it allows easy removal of specimen. However, it also has several drawbacks: technically challenging and limitation of working space. In conclusion, LESS seems to be a viable alternative to multiport laparoscopic surgery for the treatment of adnexal masses in pregnancy.

Copyright © 2016, The Asia-Pacific Association for Gynecologic Endoscopy and Minimally Invasive Therapy. Published by Elsevier Taiwan LLC. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/).

Introduction

Ovarian torsion is a twisting of the ovary in its vascular pedicle, and torsion will cause ovarian infarction if the twisted ovary is not unwound. It accounts for 3% of gynecologic emergency cases.¹ Ovarian cystic torsion during pregnancy has been reported in the literature.² Prompt diagnosis and treatment are crucial for the preservation of the ovary. Nevertheless, clinical presentation of ovarian torsion is usually indistinct, often presenting a diagnostic challenge to clinicians.

Surgical intervention should weigh the benefits and risks of surgery to both the mother and the fetus. Nevertheless, the occurrence of adverse events during surgery is low. Laparoscopy during any trimester of pregnancy is feasible, safe, and confers many advantages over laparotomy including shorter recovery, less pain, and shorter hospital stays.³ Recently, laparoendoscopic single-site surgery (LESS) has emerged as an option for treatment of a multitude of benign and malignant gynecologic conditions.^{4,5} Compared with the conventional laparoscopic approach, use of only one 1.5- to 2.0-cm umbilical incision may enable superior exteriorization of an intact ovarian cyst or mass.

Detorsion of a twisted ovary has been reported without complication in pregnant or nonpregnant women.^{6,7}

We report a case of acute torsion of ovarian teratoma in the 1st trimester of pregnancy that was successfully managed with a LESS technique.

E-mail address: dah1003@yahoo.com.tw (D.-C. Ding).

Conflicts of interest: None of the authors has any financial or personal relationships with people or organizations that could inappropriately influence their work.

^{*} Corresponding author. Department of Obstetrics and Gynecology, Buddhist Tzu Chi General Hospital, Tzu Chi University, 707, Chung-Yang Road, Section 3, Hualien City, Hualien, Taiwan, ROC.

Case Report

A 17-year-old primigravida with a gestational age of 12 weeks presented to our emergency room with right lower abdominal pain. The discomfort had been progressively worsening over the past several hours. The pain was described as severe, and located at the right lower abdominal region. She experienced nausea during every acute abdominal attack. Physical examination revealed an ill-looking woman. Her abdominal examination showed right lower quadrant abdominal tenderness. Her laboratory test results showed a hemoglobin level of 13.0 g/dL and a white blood cell count of 13,500/ μ L. The biochemical test results were within normal limits. Ultrasonography was performed and showed a fetus with a crown rump length of 5 cm (Figure 1A) and fetal heartbeat of 164 beats/min (Figure 1B). A 5-cm complex cystic mass was noted in the right pelvis, located at the anterior aspect of the uterus (Figure 1C). The Doppler study revealed no blood flow in the right ovary.

Because there was a strong likelihood of acute torsion of a right ovarian teratoma, an emergent LESS procedure was performed. We used a homemade wound retractor (Alexis; Applied Medical, Rancho Santa Margarita, CA, USA) and a surgical glove as the single-port device. Maternal vital signs, oxygen saturation, and end-tidal carbon dioxide pressure were continuously monitored during the surgery. Fetal surveillance included ultrasound monitoring of the fetal heart beat prior to and just after the surgery.

Under general anesthesia, the patient was placed lying down on the table without assuming a lithotomy position because no vaginal surgery was needed. A 2-cm vertical incision was made in the umbilicus at the beginning of the surgery. After insertion of the homemade single-port device into the abdominal cavity, two 5-mm sheaths and one 10-mm sheath were inserted through the open fingertip portions of a surgical glove and tied with 3–0 silk ligatures to prevent carbon dioxide leakage. The intra-abdominal pressure was set at 12 mmHg. Intra-abdominal visualization was obtained with a 5-mm 0° laparoscope (Karl Storz, Tuttlingen, Germany) inserted through a 10-mm cannula.

A right ovarian teratoma with concomitant torsion and ischemic change was noted (black color over the ovarian surface) (Figure 2A). Detorsion of the twisted ovary was performed first, and the ovarian surface indicated signs of blood circulation (black turning into red) (Figure 2B). An endobag was placed below the ovary prior to cystectomy. A right ovarian cystectomy was performed without rupturing the cyst (Figure 2C); the tumor was placed into an endobag, and then removed intact from the single port site (Figure 2D). There was minimal estimated blood loss, and surgical time was approximately 45 minutes. The pathological analysis confirmed a mature cystic teratoma, which exhibited skin appendages (Figure 2E). Postoperatively, the fetal heartbeat remained normal and stable. No tocolytic agent was required, and postoperative surgical wounds demonstrated satisfactory cosmetic results. The patient was discharged 4 days after the surgery.

The subsequent prenatal course was uneventful. A normal healthy child was born at the 38th week of gestation by spontaneous vaginal delivery (weight 2940 g, female).

Discussion

The present report added to the knowledge base on LESS procedure performed on pregnant women with adnexal masses. Although reports in the literature have described the feasibility and

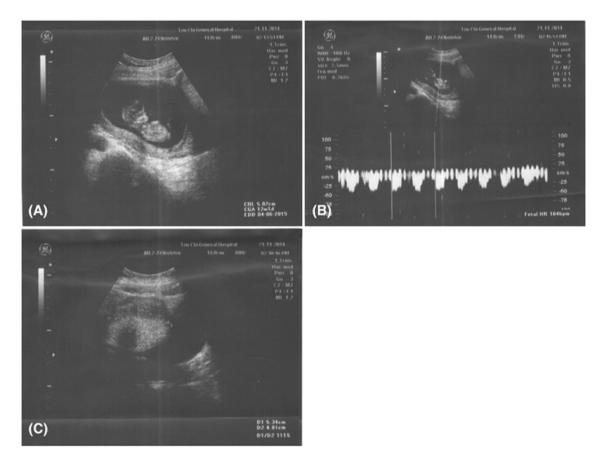


Figure 1. Ultrasonography reveals a fetus (A) with crown—rump length of 5 cm and (B) heartbeat of 164 beats/min. (C) A 5-cm complex cystic mass in the right pelvis located at anterior aspect of uterus.

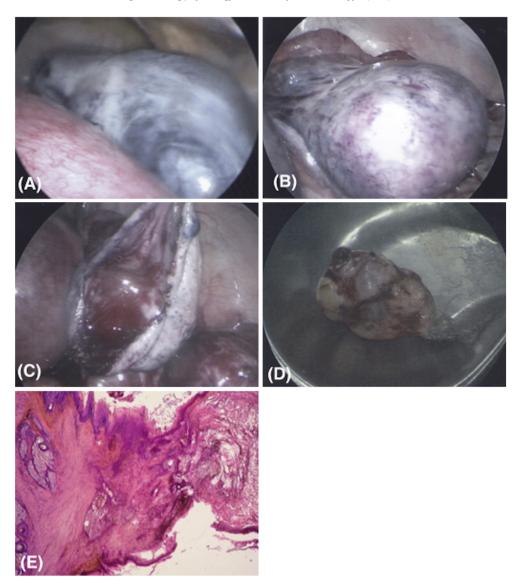


Figure 2. Laparoscopic view of twisted ovarian tumor in early pregnancy. (A) The ischemia twisted ovarian tumor exhibited black color over the ovarian surface. (B) After detorsion, the twisted ovary exhibited red color over the ovarian surface, indicating blood reperfusion. (C) Ovarian inner surface revealed little oozing after removal of teratoma. (D) Teratoma with some hairs in it. (E) Histopathology of the resected teratoma. Skin appendages are shown. (Hematoxylin and eosin, ×200.)

safety of LESS in treatment of adnexal masses, few studies have reported on LESS performed during pregnancy.^{4,8} There are six reports of LESS used in the management of an adnexal mass in pregnancy.^{1,4,9–11} Kim and Kwon⁴ reported the successful treatment of a symptomatic ovarian cyst without torsion in a pregnant woman at 12 weeks of gestational age. The procedure was begun laparoscopically using a homemade single port, after which the cyst was exteriorized through the port; cystectomy was performed extracorporeally, and the ovary was placed back in the peritoneal cavity. In Scheib et al's¹ case series, they demonstrated intracorporeally that LESS ovarian cystectomy or salpingooophorectomy was feasible in pregnant women. Takeda et al 10,11 reported two papers regarding 29 and nine pregnant women, respectively; in these studies, the patients, whose condition was complicated with adnexa mass, received LESS adnexal surgery. Three of these patients had ovarian torsion and were treated with the LESS procedure. Dursun et al⁹ reported two cases of pregnant women with ovarian tumors treated with LESS procedures, one of whom had ovarian torsion. Tsai et al¹² reported the case of a 14week pregnant woman with ovarian teratoma who underwent prophylactic LESS ovarian cystectomy. These abovementioned reports are listed and summarized in Tables 1 and 2.

LESS gynecologic surgery involves the use of a uterine manipulator to provide an adequate countertraction in the pelvis. Nevertheless, the inability to place a uterine manipulator during pregnancy makes the LESS procedure more challenging. We overcame this problem by using longer atraumatic graspers (45 cm in length) and longer suction irrigator (40 cm in length) to gently elevate the adnexa.

The differential diagnoses of abdominal pain during pregnancy include appendicitis, ureteric colic, pelvic inflammatory disease, ectopic pregnancy, and ovarian or adnexal torsion. Acute ovarian torsion is infrequent; timely diagnosis is required to prevent mortality and minimize morbidity. Surgical intervention may be required. Overall, 10–20% of ovarian torsion cases occur during pregnancy. Nevertheless, ovarian torsion is a rare condition with a prevalence rate of 1 in 4725 deliveries. ¹³ It has been identified as the fifth most common surgical emergency in pregnancy. In

Table 1Comparison of the present study with previous reports (LESS, total cases).

	Year	No. of patients	Surgery of ovary	Surgical outcome	Pathology	Obstetric outcome
This report Takeda et al ¹¹	2015 2014		Detorsion and cystectomy Unilateral cystectomy (24), bilateral cystectomy (2), USO (2), unilateral salpingectomy (1)	Discharged after 4 d Recovery well	Teratoma Teratoma (13), serous cystadenoma (7), endometriotic cyst (3), mucinous cystadenoma (2), paraovarian cyst (2), corpus luteum (1), paraovarian cystadenoma (1), clear cell carcinoma (1)	NSD NSD: 22, C/S: 6
Takeda et al ¹⁰	2014	11	Detorsion (1), cyst aspiration (2), hemostasis with suture (6), unilateral salpingectomy (1), removal of villous tissue (1)	$\begin{array}{l} Intraoperative \ autologous \\ blood \ transfusion \ (5), \\ hospital \ stay \ (mean = 6 \ d) \end{array}$	Unknown	NSD: 5, C/S: 2
Dursun et al ⁹	2013	2	1 Adnexectomy, 1 cystectomy	Discharge in 24 h (1) and 48 h (1)	Unknown	NSD (1), C/S at 32 wk (1)
Scheib et al ¹	2013	9	LSO (1), cystectomy (5), USO (3), detorsion (1)	Uneventful	Teratoma (3), borderline tumor (1), serous cystadenoma (3), mucinous cystadenoma (1)	NSD: 7, C/S: 1, ongoing pregnancy (1)

C/S = cesarean section; GA = gestational age; LESS = laparoendoscopic single-site surgery; NSD = normal vaginal delivery; USO = unilateral salpingo-oophorectomy.

Table 2 Comparison of the present study with previous reports in LESS (torsion cases).

	Year	Patient number with adnexa torsion	Surgery of ovary	Surgical outcome	Pathology	Obstetric outcome
This report	2015	1	Detorsion and cystectomy	Discharged after 4 d	Teratoma	NSD
Takeda et al ¹¹	2014	1	Unknown	Recovery well	Unknown	Unknown
Takeda et al ¹⁰	2014	2	Detorsion, cyst aspiration	Recovery well	Unknown	Unknown
Dursun et al ⁹	2013	1	Adnexectomy	Uneventful	Unknown	C/S at 32 wk of GA
Scheib et al ¹	2013	2	1 Detorsion, 1 detorsion and cystectomy	Recovery well	1 Corpus luteum cyst, 1 teratoma	2 NSD

C/S = cesarean section; GA = gestational age; LESS = laparoendoscopic single-site surgery; NSD = normal vaginal delivery.

patients with ovarian cystic teratomas smaller than 6 cm, this may not cause torsion throughout pregnancy.¹⁴ Nevertheless, in our patient, a 5-cm cystic teratoma caused ovarian torsion eventually.

Pelvic ultrasound is the initial imaging modality for evaluation of ovarian torsion.¹⁵ Doppler studies have high specificity but low sensitivity and may be normal in 60% of torsion cases.¹⁵ Moreover, the presence of arterial and venous flow does not exclude the diagnosis of torsion. Magnetic resonance imaging with contrast can also aid in the diagnosis of ovarian torsion.⁷

Laparoscopy is considered the procedure of choice for ovarian detorsion. Long-term follow-up of patients after laparoscopic detorsion performed in the setting of ischemic and nonviable appearance of the torsed ovarian revealed that most ovaries regain normal appearance and function. Laparoscopic abdominal entry can be particularly precocious in a woman with gravid uterus. Blind abdominal access techniques such as direct insertion of trocars or use of a Veress needle should be performed carefully to avoid causing injury to the enlarged gravid uterus or displaced viscera. With LESS, abdominal entry is safely accomplished using an open entry technique and no additional incisions or ports required. Also, LESS fascial incision enhances the safety of the open entry technique and facilitates directly visualized fascia closure.

Surgery in pregnancy is considered to carry potential risks to both mother and fetus, including maternal complications, fetal loss, and preterm birth. However, accumulating evidence suggests that laparoscopic treatment of adnexal masses in the 1st trimester of pregnancy is safe and effective. The LESS procedure may be considered for surgery at 10 to 20 weeks of gestational age. The interventions resulted in preservation of ovarian function without adverse effects on the pregnancy. In our case, laparoscopic detorsion was able to preserve ovarian function and was followed by successful pregnancies. Single-port laparoscopy provides convenience during the removal of adnexal tumors. The 2-cm single

port wound enables surgeons to place the tumor in the endobag easily and draw it out without causing rupture.

One concern about the umbilical LESS incision is the increased potential for hernia formation. Pregnant women may be susceptible to this complication because of the laxity of the connective tissue and abdominal stress. Nevertheless, a recent report revealed an overall low risk of umbilical hernia with the LESS procedure with a running mass closure with delayed absorbable suture. ¹⁸ We used 1–0 Vicryl interrupt suture in closing the umbilical wound. In this case, throughout the whole pregnant course, no umbilical hernia was noted.

In summary, LESS is a relatively new technique, and much remains to be learned about its applicability and usefulness. Use of LESS in pregnancy has several advantages: this technology is safe for both the mother and the fetus, and it allows easy removal of specimens. Its drawbacks are its technical complexity and limitation of the working space. In conclusion, LESS seems to be a viable alternative to multiport laparoscopic surgery for the treatment of adnexal masses in pregnancy.

Acknowledgments

The authors are grateful to Y.-H. Hsu for providing the histopathological pictures.

References

- Scheib SA, Jones HH, Boruta DM, et al. Laparoendoscopic single-site surgery for management of adnexal masses in pregnancy: case series. J Minim Invasive Gynecol. 2013;20:701–707.
- Kang HJ, Davis OK, Rosenwaks Z. Simultaneous bilateral ovarian torsion in the follicular phase after gonadotropin stimulation. Fertil Steril. 2006;86: 462e413–462e464.
- 3. Koo YJ, Park JY, Kim DY, et al. Laparoscopic versus open surgery for adnexal tumor in pregnant women. *Gynecol Minim Invasive Ther*. 2013;2:57–60.

- Kim WC, Kwon YS. Laparoendoscopic single-site surgery for exteriorization and cystectomy of an ovarian tumor during pregnancy. J Minim Invasive Gynecol. 2010;17:386–389.
- Hong MK, Wang JH, Chu TY, Ding DC. Laparoendoscopic single-site hysterectomy with Ligasure is better than conventional laparoscopic assisted vaginal hysterectomy. *Gynecol Minim Invasive Ther*. 2014;3:78–81.
- **6.** Ding DC, Chen SS. Conservative laparoscopic management of ovarian teratoma torsion in a young woman. *J Chin Med Assoc.* 2005;68:37–39.
- Tan KH, Chen KC, Wang TL, Chong CF, Chen CC. Ovarian cystic teratoma torsion in pregnancy. *Emerg Med J.* 2010;27:879

 –880.
- 8. Topgul K, Yuruker SS, Kuru B. Single-incision laparoscopic cholecystectomy in a 6-month pregnant woman: a report of a case. Surg Laparosc Endosc Percutan Tech. 2011;21:e100—e103.
- Dursun P, Gulumser C, Caglar M, Araz C, Zeyneloglu H, Haberal A. Laparoendoscopic single-site surgery for acute adnexal pathology during pregnancy: preliminary experience. *J Matern Fetal Neonatal Med.* 2013;26: 1282–1286.
- Takeda A, Hayashi S, Imoto S, Sugiyama C, Nakamura H. Pregnancy outcomes after emergent laparoscopic surgery for acute adnexal disorders at less than 10 weeks of gestation. J Obstet Gynaecol Res. 2014;40:1281–1287.
- Takeda A, Imoto S, Nakamura H. Gasless laparoendoscopic single-site surgery for management of adnexal masses during pregnancy. Eur J Obstet Gynecol Reprod Biol. 2014;180:28–34.

- 12. Tsai HW, Chen CY, Wang PH, et al. Single-port laparoscopic ovarian cystectomy of teratoma during pregnancy. *Gynecol Minim Invasive Ther*. 2013;2: 137–139.
- Child TJ, Watson NR, Ledger WL. Sequential bilateral adnexal torsion after a single cycle of gonadotropin ovulation induction with intrauterine insemination. Fertil Steril. 1997;67:573

 –575.
- Caspi B, Levi R, Appelman Z, Rabinerson D, Goldman G, Hagay Z. Conservative management of ovarian cystic teratoma during pregnancy and labor. Am J Obstet Gynecol. 2000:182:503

 –505.
- Pena JE, Ufberg D, Cooney N, Denis AL. Usefulness of Doppler sonography in the diagnosis of ovarian torsion. Fertil Steril. 2000;73:1047–1050.
- Oelsner G, Cohen SB, Soriano D, Admon D, Mashiach S, Carp H. Minimal surgery for the twisted ischaemic adnexa can preserve ovarian function. *Hum Reprod*. 2003:18:2599–2602.
- Oh NJ, Kim WY. Laparoendoscopic single-site surgery (LESS) for large benign adnexal tumors: one surgeon's experience over one-year period. Clin Exp Obstet Gynecol. 2014:41:319–322.
- Lowry PS, Moon TD, D'Alessandro A, Nakada SY. Symptomatic port-site hernia associated with a non-bladed trocar after laparoscopic live-donor nephrectomy. J Endourol. 2003;17:493

 –494.