Laparoscopic treatment of spontaneous ovarian torsion in a twin pregnancy at 16 weeks after in vitro fertilization: a case report and literature review

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Background: Spontaneous ovarian torsion is a rare complication that occurs in fewer than 1 of 5,000 pregnancies but requires emergency treatment. Few reports have described cases of ovarian torsion in multiple pregnancies. According to our literature review, this is the first case report of a successfully laparoscopic management in a twin pregnancy in the second trimester after in vitro fertilization (IVF) in China. More importantly, both the mother and the babies were with a fairly good outcome after surgery.

Case Description: We presented a case of spontaneous ovarian torsion in a 33-year-old Chinese woman at 16 weeks in a twin pregnancy after IVF. The patient was treated through emergency laparoscopic procedure. During the procedure, an enlarged ovary with twisted pedicle was identified, and the ovary had turned purple. After the cyst fluid was punctured and vacuumed, the ovary returned to normal size; while the twist was reversed, the ovary regained its pink color. After surgery, the patient recovered well and no signs of ovary dysfunction or abortion were observed.

Conclusions: We performed a successfully laparoscopic treatment for ovarian torsion in twin pregnancy after IVF. Our experience may help gynecological clinicians better select appropriate therapeutic management in cases similar to ours.

Keywords: Ovarian torsion; twin pregnancy; in vitro fertilization (IVF); laparoscopic treatment; case report

Received: 19 February 2022; Accepted: 19 November 2022; Published online: 22 February 2023.

doi: 10.21037/gpm-22-10

View this article at: https://dx.doi.org/10.21037/gpm-22-10

Introduction

Spontaneous ovarian torsion is a rare complication that occurs in fewer than 1 of 5,000 pregnancies but requires emergency treatment (1). Its incidence may be increasing with the growing use of in vitro fertilization (IVF) (2). Although laparoscopic management of ovarian torsion during pregnancy has been reported in the decades, few reports have described cases of ovarian torsion in multiple pregnancies. And adverse events such as poor gestational outcomes (3) and recurrence within 3 days (4,5) have been reported. Here we describe a twin pregnancy complicated by spontaneous ovarian torsion after IVF. To our best knowledge, this is the first case report of a successfully laparoscopic management in a twin pregnancy after IVF in China. Both the mother and the babies were with fairly good outcomes. We presented this case and conducted a comprehensive literature review to supplement clinical data to management of patients similar to ours. We present this case in accordance with the CARE reporting checklist (available at https://gpm.amegroups.com/article/

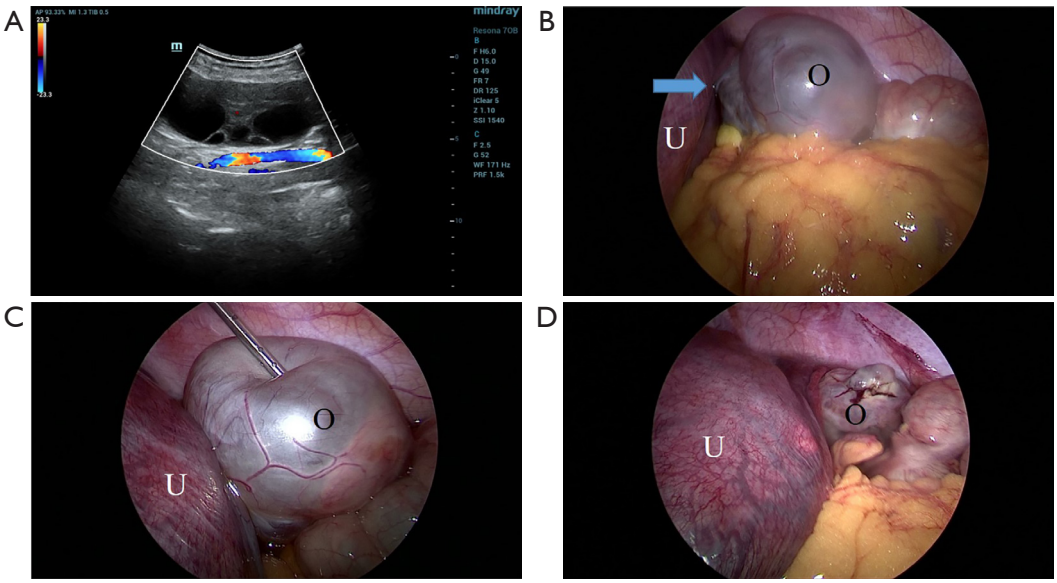

Figure 1 Laparoscopic treatment of ovary torsion in a twin pregnancy at 16 weeks. (A) Color Doppler ultrasonography showing an enlarged ovary with multiple compartments. (B) The enlarged ovary was twisted by one cycle (arrow) and had turned purple. (C) Puncture and vacuuming of the cyst fluid. (D) The recovered ovary after bipolar coagulation hemostasis. O, ovary; U, uterus.

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Case presentation

A 33-year-old Chinese woman (gravida 3, para 0) was admitted to our hospital at 16 weeks of gestation with complaints of abdominal pain and vomiting during the previous 6 hours. At 98 days before admission, she had undergone IVF, which led to twin pregnancy.

The patient had unremarkable personal and family medical histories (including ovarian cysts or polycystic ovarian syndrome). The gynaecological ultrasonography before the embryo transferring was negative. Physical examination showed that uterine size was closer to that expected after five months of gestation. Tenderness and rebound pain were detected in the right abdomen. Color Doppler ultrasonography revealed a compartmentalized cyst measuring 8.0 cm × 3.7 cm × 7.4 cm on the right side, behind the uterus (*Figure 1A*). No abnormalities were found in the appendix or urinary tract. Laboratory tests for amylase and lipase were negative. Based on these findings, the patient was diagnosed with ovarian torsion.

Emergency laparoscopic exploration under general anaesthesia was performed using three trocars of 5 mm. The right ovary was found to be enlarged and to have multiple compartments containing clear fluid. The two larger compartments measured 3 cm × 2 cm × 4 cm (*Figure 1B*). The pedicle was twisted by one cycle, and the ovary had turned purple (*Figure 1C*). The cyst fluid was punctured and vacuumed, after which the ovary returned to normal size; after the twist was reversed, the ovary regained its pink color (*Figure 1D*). Bipolar coagulation was used to achieve haemostasis. After surgery, the patient recovered well and no signs of ovary dysfunction or abortion were observed.

Routine prenatal examinations showed that both mother and foetuses were normal until 30 weeks gestation, when the infants were delivered by caesarean section because of foetal distress. One newborn weighed 1,260 g and showed Apgar scores of 8, 9 and 10 at 1, 5 and 10 min after delivery; the other newborn weighed 930 g and showed Apgar scores of 2, 6 and 8. The newborns were transferred to the neonatal intensive care unit and discharged two months later. By the latest follow-up five months after birth, mother and babies were healthy, and the babies showed normal development.

The study was approved by the hospital Ethics Committee. All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013). Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is

available for review by the editorial office of this journal.

Discussion

To place our case in perspective with the literature on women with twin pregnancies who experienced ovarian torsion after IVF, we identified relevant studies in PubMed and EMBASE (*Table 1*). We searched the PubMed and EMBASE databases on studies related to treatment for ovarian torsion in women with multiple pregnancy. Databases were searched from their respective inceptions until February 1, 2022. The predefined search string was the following: (ovarian torsion OR ovary torsion OR adnexal torsion) AND (IVF OR assisted reproductive technology) AND (pregnant OR pregnancy OR gestation). Only publications in English were included. There were no limitations regarding publication date or article type. References within the included publications were also reviewed to identify additional studies. The study selection was described in *Figure 2*.

To be included, studies had to (I) be case report, cohort studies or case-control design; (II) involve subjects who were diagnosed with ovarian torsion through surgical exploration or color Doppler ultrasonography; (III) provide relevant information including gestational outcomes, size of ovaries, management of ovarian torsion; and (IV) report both the mother and baby outcomes. Studies were excluded if (I) they were published in languages other than English; (II) they focused on ovarian torsion in women who were not pregnant or who were singleton pregnant; or (III) relevant information was not available.

Two authors (YF and YFZ) independently screened the titles and abstracts to identify relevant studies based on the eligibility criteria. After initial selection, the full texts of all potential articles were independently read by two authors for further evaluation. Any disagreement was resolved by discussion with the corresponding author.

Among the 11 cases (3-13) reported in those studies and the case reported here, 5 (41.7%) occurred in the first trimester, 6 (50.0%) in the second trimester and 1 in the third trimester. The mean gestational age at which ovarian torsion was diagnosed was 16.4 weeks (range, 5.9–32.9 weeks). Among the 12 patients, 7 were diagnosed with right ovarian torsion, 4 with left ovarian torsion and 1 with bilateral ovarian torsion. And two recurrences were reported after primary surgery. All 12 patients were treated at the time of diagnosis. In the end, 11 patients delivered two healthy infants and 1 patient delivered one infant with lymphoma, while the other infant died on the second day after delivery (*Table 1*).

Seven patients underwent laparoscopic surgery as primary treatment, 3 underwent laparotomy, and 1 each underwent a combination of laparoscopy and laparotomy or a non-surgical procedure. Ovaries were removed from six patients because of necrotic changes in the ovaries, and from one patient on the basis of a decision by the attending physician (9). Five patients underwent detorsion with aspiration, while one patient underwent percutaneous cyst aspiration guided only by ultrasonography (10). No maternal adverse events, abortions, or other foetal adverse events were reported. Two of the five women who experienced ovarian torsion in the first trimester suffered recurrence, while no recurrence was reported in the second or third trimesters.

Our case and literature review suggested that either laparoscopy or laparotomy was effective for treating ovarian torsion in women with twin pregnancies. In fact, laparoscopy is considered to be similarly effective as laparotomy in single pregnancies (2). Moreover, laparoscopy may be safer and more minimally invasive than laparotomy (13,14). Whatever the approach, early diagnosis and treatment are critical for preserving the ovaries and ensuring both foetal and maternal safety.

Our literature review also indicated that ovarian torsion during pregnancy after assisted reproductive technology (ART) were more common in the first and early second trimesters than in the third trimester. It is reported that corpus luteum cysts or ovarian hyperstimulation syndrome (OHSS) are common causes of ovarian torsion during pregnancy (15). And corpus luteum cysts are the most common cause of adnexal masses during pregnancy (16). Moreover, in patients treated with ART, OHSS may occur in 3% to 10% of all ART cycles, and even 20% among high-risk women (17,18). Previous studies also have reported a decreased risk of corpus luteum cysts and OHSS in the second trimester (16,19), which might be a reason for higher incidence of ovarian torsion in early stages of pregnancy after ART.

Our study presents several limitations. First, due to rarity of similar cases, we could only present a case of one patient instead of case series. Second, although a comprehensive literature review was conducted, we could not identify potential risks of ovary torsion recurrence.

Despite these limitations, our study presents important strengths. To our best knowledge, this is the first case report of a successfully laparoscopic management in a twin

Table 1 Characteristics of studies focused on women with twin pregnancies who experienced ovarian torsion after IVF

Study	Country	Age (years)	Approach	Procedure	ART	Gestational age at torsion diagnosis	Side	Max. diameter (cm)	Time to surgery*	Gestational age at delivery (type of delivery)	Foetal outcome	Recurrence?
Present report	China	33	Laparoscopy	Detorsion + aspiration	IVF	16 w	R with 1 rotation	8.0	6 h	30 w (caesarean)	Two healthy infants (NICU)	No
Yu *et al.* (3), 2021	China	32	Laparoscopy	Detorsion	IVF	8 w + 6 d	L with 3 rotations	12.1	7.5 h	30 w (NR)	One died on the 2nd day after delivery, one was born with diffuse large B-cell lymphoma	No
Bernigaud *et al.* (6), 2021	France	36	Laparotomy	Ovariectomy	IVF	32 w + 6 d	R with 3 rotations	7.8	NR	37 w (spontaneous labor)	Two healthy infants	No
Habek *et al.* (7), 2016	Croatia	32	Laparotomy	Ovariectomy	IVF	17 w	R	NR	48 h	37 w (caesarean)	Two healthy infants	No
Aydin *et al.* (8), 2014	Turkey	28	Laparoscopy	Detorsion	IVF	23 w	R	6	NR	35 w (caesarean)	Two healthy infants	No
Dursun *et al.* (9), 2013	Turkey	NR	Laparoscopy	Adnexectomy	IVF	25 w	L	8	NR	32 w (caesarean)	Two healthy infants (NICU)	No
Al Omari *et al.* (4), 2011	United Arab Emirates	30	Laparotomy, laparoscopy (recurrence)	Detorsion + salpingoophorectomy (L) and aspiration (R); detorsion (recurrence)	IVF	10 w	L	12	NR	35 w (caesarean)	Two healthy infants	On the right, 3rd day after surgery
Boswell *et al.* (10), 2010	United States	33	Non-surgical	Percutaneous cyst aspiration	IUI	13 w + 2d	L	5.1	NR	30 w (caesarean)	Two healthy infants	No
Arena *et al.* (11), 2009	Italy	38	Laparoscopy	Adnexectomy	IVF	10 w	R	7.9	NR	NR (caesarean)	Two healthy infants	No
Hasiakos *et al.* (12), 2008	Greece	35	Laparoscopy	Adnexectomy	IVF	10 w	R	10	48 h	37 w (caesarean)	Two healthy infants	No
Bassil *et al.* (13), 1999	Belgium	33	Laparoscopy	Detorsion	IVF	25 w	R, L	6.1	NR	33 w (spontaneous labor)	Two healthy infants	No
Child *et al.* (5), 1997	United Kingdom	35	Laparoscopy + laparotomy, laparotomy (recurrence)	Salpingo-oophorectomy; detorsion (recurrence)	IUI	5 w + 6d	R with 2 rotations	10	Within 4 h, 8 h (recurrence)	37 w (caesarean)	Two healthy infants	On the left, about 60 h after surgery

*, time from symptom onset to surgery. IVF, in vitro fertilization; ART, assisted reproductive technology; w, weeks; R, right; h, hours; NICU, neonatal intensive care unit; d, days; L, left; NR, not reported; IUI, intra-uterine insemination.

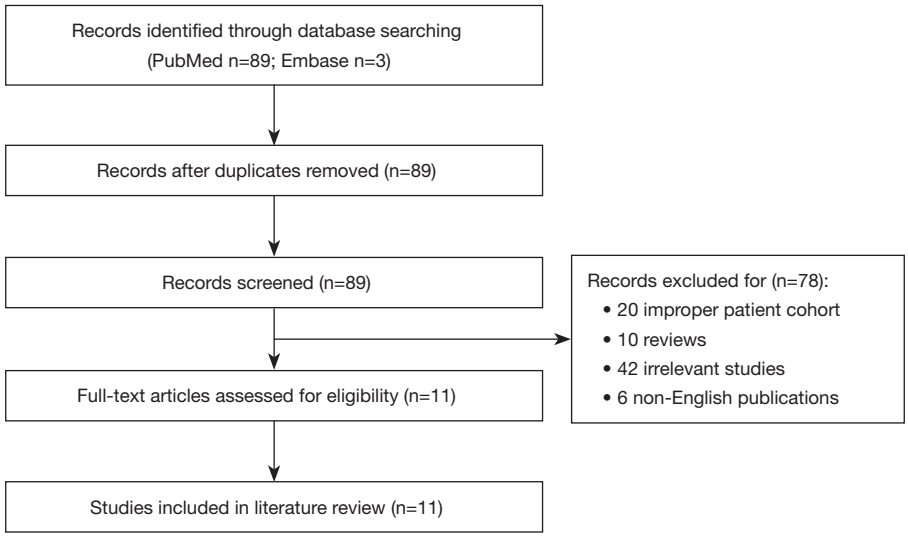

Figure 2 The flow diagram of literature searching and selection.

pregnancy in the second trimester after IVF in China. Our study fully demonstrated the effectiveness and safety of laparoscopic treatment for ovarian torsion in multiple pregnancy.

Conclusions

We performed a successful laparoscopic treatment for ovarian torsion in twin pregnancy after IVF. Our experience may help gynecological clinicians better select appropriate therapeutic management in cases similar to ours.

Acknowledgments

Funding: This work was supported by the Science and Technology Department of Sichuan Province under Grant 2017SZ0118. The funding organisation did not play any role in the design or conduct of the study.

Footnote

Reporting Checklist: The authors have completed the CARE reporting checklist. Available at https://gpm.amegroups.com/article/view/10.21037/gpm-22-10/rc

Conflicts of Interest: All authors have completed the ICMJE uniform disclosure form (available at https://gpm.amegroups.com/article/view/10.21037/gpm-22-10/coif). The authors have no conflicts of interest to declare.

Ethical Statement: The authors are accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. The study was approved by the hospital Ethics Committee. All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013). Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the editorial office of this journal.

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doi: 10.21037/gpm-22-10
Cite this article as: Fan Y, Zhang YF, Li JK. Laparoscopic treatment of spontaneous ovarian torsion in a twin pregnancy at 16 weeks after in vitro fertilization: a case report and literature review. Gynecol Pelvic Med 2023;6:7.