Case Report

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Spontaneous rupture of the uterus following salpingectomy: a case report and literature review

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Abstract

Laparoscopic salpingectomy (LPSC) is the main treatment for ectopic pregnancy, which leads to spontaneous uterine rupture (UR) during pregnancy. We report the characteristics of a woman who had spontaneous UR during pregnancy with a history of salpingectomy. We experienced a 31-year-old woman with a UR in pregnancy with a history of LPSC twice. The patient had a successful pregnancy. We also performed a literature review including cases with spontaneous UR after LPSC. Twenty-seven case reports of 48 women were included in our review. Thirty-five (83.33%, 35/42) women previously received LPSC and 15 (31.25%) developed interstitial pregnancies. The interval between pregnancy and the last surgery did not affect the frequency of interstitial pregnancy and gestational age. Fetal outcomes in patients with UR at the third trimester were better than those at the first and second trimesters. We suggest that close observation and timely treatment by experienced clinicians lead to good outcomes of pregnant women with suspected UR.

Keywords

Salpingectomy, uterine rupture, cesarean section, ectopic pregnancy, laparoscopy, laparotomy, fetal outcome

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Introduction

Salpingectomy is the main treatment for ectopic pregnancy[1] and a leading cause for uterine rupture (UR).[2] Although the overall incidence of UR is low at a rate of less than

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0.1% in the general population, maternal and fetal outcomes are usually poor once UR occurs.[2–5] Conventional surgical treatment for ectopic pregnancy usually includes laparoscopic salpingectomy (LPSC) and laparotomy (LPT). However, the high incidence of UR after LPSC raises the question of its safety.[3,6,7] A high incidence of cesarean section (CS) also increases the frequency of spontaneous UR during pregnancy.

We report a patient who had two episodes of spontaneous UR during pregnancy after LPSC. A literature review was performed to present the available case reports in women with spontaneous UR after LPSC. English and Chinese publications were included. Maternal and fetal outcomes were recorded. This study could provide further insight into the association between UR during pregnancy with prior LPSC.

Methods

Case presentation

A 31-year-old Chinese woman (gravida 3, para 2) with 34^{+6} gestational weeks of pregnancy was admitted to the Department of Obstetrics, Changning Maternity & Infant Health Hospital of Shanghai in May 2013 for delivery. Her medical history included LPSC in March 2011 for a right tubal pregnancy and a UR at the right corner of the uterus in May 2012 before CS at 34^{+4} gestational weeks.

After LPSC, the patient became pregnant again in September 2011, 6 months after LPSC. She was admitted to our department at 34^{+4} gestational weeks because of persistent lower abdominal pain, nausea, and vomiting for more than 1 day. A physical examination showed stable vital signs and lower tenderness in the right lower quadrant, without rebounding pain. A renal ultrasound showed a small amount of fluid in the right kidney. An ultrasound scan showed a normal fetal heart rate. Hematuria was detected and the hemoglobin (Hb) level was 11.4 g/dL. The patient was admitted to the Urology Department in another hospital and was treated with an injection of progesterone. She reported that the pain was transiently alleviated. On the same night, the woman was admitted to our Emergency Department because of progressive abdominal pain. Her vital signs were as follows: body temperature of 36.6°C; heart rate of 98 beats/minute; blood pressure of 80/40 mmHg; and respiratory rate of 22 times/minute. The heart rate of the fetus was 55 to 69 beats/minute. The patient had severe anemia and hypertonic uterine. Her white blood cell count, neutrophilic granulocyte percentage, and Hb level were 19.4×10^9/L, 91%, and 6.6 g/dL, respectively. A coagulation function examination showed that the prothrombin time was 12.1 seconds, activated partial thromboplastin time was 28.2 seconds, thrombin time was 14.7 seconds, fibrinogen level was 0.48 g/dL, and the D-dimer level was 7.9 μg/mL. A patchy hypoechoic area (anteroposterior diameter: 45 mm) at the attachment of the placenta to the uterus was found and CS was performed immediately. A large amount of hemoperitoneum (approximately 1000 mL) was collected and removed before exposure of the rupture. A rupture (8 cm) was observed in the right corner of the uterus and most of the placenta (3/4) was discharged from it. A total amount of approximately 2000 mL of free blood was collected during the surgery. The amniotic fluid was clear with a total amount of 300 mL. A dead fetus (Apgar score: 0–0) weighing 2460 g during surgery was delivered. The uterus was conservatively repaired in two layers with absorbable sutures. Four units of red blood cells and 200 mL of blood plasma were transfused during the LPT. Postoperative anti-infection treatment was administered. A routine blood examination on the next day showed that her hemoglobin level was 9.2 g/dL. The woman was

discharged on the 6th postoperative day and was advised to have a pregnancy 2 years later.

The woman became pregnant 5 months after the last surgery. She was admitted to our department at 28^{+6} gestational weeks (in May 2013). Her Hb level was 9.5 g/dL, heart rate was 84 beats/minute, and blood pressure was 110/75 mmHg. Oral iron supplement was advised. At 34 gestational weeks (15 June 2013), the woman complained of abdominal pain at the right lower quadrant. An ultrasound showed that the fetal heart rate was 143 beats/minute. However, CS was immediately implemented according to her medical history. A rupture (4×3 cm) was observed in the right corner of the uterus. A total of 200 mL hemoperitoneum was collected before CS. The woman delivered a boy (Apgar score: 9–9) weighing 2650 g. The rupture was conservatively repaired with two layers of sutures. The woman was discharged on the 3rd postoperative day. The Hb level was 8.4 g/dL on discharge. The patient and fetus had an uneventful follow-up.

Ethics statement

Ethics approval was not applicable because there was no special treatment for the woman. Written informed consent was obtained from the patient before each surgery. Written informed consent for publication was also obtained.

Literature review

English and Chinese literature published during 1996 to 2018 that reported cases of spontaneous UR during pregnancy with prior salpingectomy were screened from PubMed and Wanfang databases. The search terms of "interstitial pregnancy", "salpingectomy" and "uterine rupture" were used. Articles reporting UR during pregnancy with prior salpingectomy (LPSC or LPT) were included in our study. Reports were excluded if they met the following criteria: (1) UR not induced by pregnancy; (2) UR induced by prior CS, and laparoscopic myomectomy or unspecified adnexectomy (ovary or fallopian tube). Literature searches were performed by three authors (Hua ZF, Guo YQ, and Zhang Y). The patients' age, medical history (LPSC, LPT, or others), gestational week, maternal and fetal outcomes, signs for UR on admission, surgical management, and the interval between salpingectomy or the last UR and conception were reported and used for statistical analysis.

Statistical analysis

IBM SPSS 22.0 software (IBM Corp., Armonk, NY, USA) was used for statistical analyses. Differences were analyzed using the χ^2 test. Correlation between pregnancy intervals and gestational age was analyzed using Spearman correlation analysis. A p value <0.05 was used for the threshold of significant difference for all analyses.

Results

Our literature review identified 48 cases of spontaneous UR during pregnancy with prior salpingectomy reported in 27 publications[1–4,6–28] (Table 1). The mean ($\pm$ standard deviation) age of the women was 30.2 ± 5.3 years. LPSC was the primary choice (83.33%, 35/42) for managing previous interstitial pregnancies. Most (66.67%, 32/48) women were admitted to hospital because of abdominal pain. Of these patients, 15 URs (31.25%, 16/48) were induced by interstitial pregnancy. In patients with interstitial pregnancies, all (100%, 15/15) of them had adverse fetal outcomes. Fourteen (42.42%) patients had adverse fetal outcomes among the remaining 33 patients with intrauterine pregnancy.

Table 1. Summary of included studies in the literature review and characteristics of their cases.

No.	Author, year	Age (years)	Pregnancy method (natural/IVF)	Previous surgery	Interval (months)	Ectopic pregnancy (yes/no)	Gestation (weeks)	Signs and symptoms of UR	Outcomes of mother and fetus	Management	Incisional closure
1	Arbab et al. 1996[2]	25	IVF	NA	<12	Yes	10	HS	UR	LPT+CRU	Two layers
2		34	IVF	NA	<12	Yes	13	AP, HS	UR, PI, fetal death	LPT+ myometrial reduction	Hysterectomy
3		25	IVF	NA	<12	No	20	AP, HS	UR, fetal death	LPT+CRU	Two layers
4		33	IVF	NA	<24	No	18	AP, VB	UR, PT	LPT+CRU	Two layers
5		27	IVF	NA	<72	Yes	26	AP, HS	UR, fetal death	LPT+ myometrial reduction	Hysterectomy
6	Kasprowicz et al. 1996[18]	24	Natural	LPT	3	No	38	AP, VB, HS	UR, PI, fetal death	CS+CRU	Two layers
7	Inovay et al. 1999[19]	31	IVF	LPSC	6	No	14	AP, VB	UR, fetal death	LPT+CRU	Two layers
8	Ayoubi et al. 2003[20]	28	Natural	LPSC	24	No	22	AP	UR, PT	LPT+CRU	Two layers
9	Banaszczyk et al. 2005[21]	27	IVF	LPT	NA	No	23	AP, HS	UR, PI	LPT+CRU	NA
10	Su et al. 2008[5]	30	Natural	LPSC	6	No	40	PR, HS	UR, live birth	LPT+CRU	One layer
11	Chatterjee et al. 2009[23]	29	Natural	LPT	6	No	29	AP, VB	UR, live birth	CS+CRU	Two layers
12	Liao et al. 2009 [24]	29	Natural	LPSC	NA	Yes	13	AP, VB	UR, fetal death	LPT+CRU	Two layers
13	Pluchino et al. 2009[8]	34	Natural	LPSC	<12	Yes	7	AP, HS	UR, fetal death	LPT+CRU	NA
14	Muglu et al. 2012[9]	31	Natural	LPSC	<12	No	24	AP, vomiting	UR, fetal death	LPT+CRU	Two layers
15	Galati et al. 2013[10]	29	Natural	LPSC	<2	No	9	AP, HS	UR, fetal death	LPT+CRU	Hysterectomy
16	Yang 2013[11]	21	Natural	LPSC	<10	No	33+2	AP	UR, live birth	CS+CRU	NA
17	Cai et al. 2014[7]	27	Natural	LPSC	48	No	36+4	AP	UR, fetal death	CS+CRU	NA
18		30	Natural	LPSC	<12	No	30+5	AP	UR, fetal death	CS+CRU	NA
19	Nishijima et al. 2014[12]	45	Natural	LPSC	48	No	26	AP, HS	UR, live birth	CS+CRU	Two layers
20	Gu and	36	Natural	LPSC	19	No	27+2	AP, VB	UR, fetal death	CS+CRU	Two layers
21	Wang 2015[6]	24	Natural	LPSC	12	No	40+1	PR, HS	UR, live birth	LPT+CRU	NA
22	Stanirowski et al. 2015[3]	29	Natural	LPSC	14	No	38	AP	UR, live birth	CS+CRU	Two layers
23	Tan et al. 2015[4]	27	Natural	LPT	13	No	34	AP	UR, live birth	CS+CRU	Three layers
24	Abbas et al. 2015[13]	24	Natural	LPSC	>12	No	39	Antepartum hemorrhage	UR, live birth	CS+CRU	Two layers

(continued)

Table 1. Continued

No.	Author, year	Age (years)	Pregnancy method (natural/IVF)	Previous surgery	Interval (months)	Ectopic pregnancy (yes/no)	Gestation (weeks)	Signs and symptoms of UR	Outcomes of mother and fetus	Management	Incisional closure
25	Xu et al. 2016[14]	28	IVF	LPSC	>12	Yes	5	Internal bleeding	UR, PT	LPT+CRU	NA
26		28	IVF	LPSC	<12	No	4	AP	UR, PT	LPT+CRU	NA
27	Marciniak et al. 2016[1]	29	Natural	LPSC	6	Yes	8	AP, VB	UR, PT	LPT+CRU	NA
28	Paradise et al. 2016[15]	38	IVF	LPSC	120	No	26	AP	UR, live birth	CS+CRU	NA
29	Xu et al. 2016[16]	25	IVF	LPSC	11	No	36+1	AP	UR, live birth	CS+CRU	NA
30	Lin 2017[28]	42	Natural	LPSC	27	Yes	23	NA	UR, PT	CS+CRU	NA
31		24	IVF	LPSC	18	No	35	AP	UR, live birth	CS+CRU	NA
32		38	IVF	LPSC	10	No	36	FE	UR, live birth	CS+CRU	NA
33		28	IVF	LPSC	9	No	20	AP, vomiting	UR, PT	CS+CRU	NA
34	Wu et al. 2018[17]	NA	NA	LPSC	11	No	29	NA	UR	CRU	One layer
35		NA	NA	LPSC	4	No	31	NA	UR	NA	NA
36	Lin et al. 2018[25]	~30	IVF	LPSC	>36	No	32+6	AP, vomiting	UR, live birth	CS+CRU	NA
37		~30	IVF	LPSC	>36	No	35	AP	UR, live birth	CS+CRU	NA
38		~30	IVF	LPSC	>36	No	33	AP, VB	UR, live birth	CS+CRU	NA
39		~30	IVF	LPSC	>36	No	35+1	AP	UR, live birth	CS+CRU	NA
40		~30	Natural	LPSC	>36	No	35+4	AP	UR, live birth	CS+CRU	NA
41	Yuan and Peng 2018[26]	30	IVF	LPSC	5	No	34	AP	UR, live birth	CS+CRU	NA
42	Jiang and Zhao 2018[27]	29	IVF	LPSC	9	Yes	7	NA	UR, PT	LP+CRU	NA
43		35	Natural	LPSC	24	Yes	5+4	NA	UR, PT	LP+CRU	NA
44		30	IVF	LPT	24	Yes	7+2	NA	UR, PT	LP+CRU	NA
45		35	IVF	NA	24	Yes	7	NA	UR, PT	LP+CRU	NA
46		39	IVF	LPT	12	Yes	8+2	NA	UR, PT	LP+CRU	NA
47		34	IVF	LPT	24	Yes	6	NA	UR, PT	LP+CRU	NA
48		27	IVF	LPSC	7	Yes	8	NA	UR, fetal death	LP+CRU	NA

AP, abdominal pain; CRU, conservative repair of the uterus; CS, cesarean section; FE, fetal embarrassment; HS, hemorrhagic shock; interval, interval between salpingectomy and conception. IVF, *in-vitro* fertilization; LP, laparoscopy; LPSC, laparoscopy-assisted salpingectomy; LPT, laparotomy; NA, not applicable; natural, natural conception; NL, natural labor; PI, placenta increta; PR, placental retention; PT, pregnancy termination; UR, uterine rupture; VB, vaginal bleeding.

Of these, 25 (54.35%, 25/46 reported) women received *in-vitro* fertilization (IVF).[2,14–16,19,21,25–28] There was no significant difference in the frequency of interstitial pregnancy between patients who received IVF (40.00%, 10/25) and those who did not (23.81%, 5/21; $\chi^2 = 1.361$, $p = 0.243$). Among patients who were conceived at ≤ 6 ($n = 8$, 17.02%), 7 to 12 ($n = 16$, 34.03%), 13 to 24 ($n = 12$, 25.53%), and >24 months ($n = 10$, 21.28%) after surgery (46 reported), one (12.50%), six (37.50%), five (41.67%), and two (20.00%) patients had interstitial pregnancies, respectively. There was no significant difference in the frequency of interstitial pregnancy among the four groups ($\chi^2 = 2.822$, p=0.420).

UR at ≤ 27 gestational weeks during the first (13 weeks) and second trimesters (14–27 weeks) ($n = 27$) usually suggested pregnancy termination or fetal death (96.27%, 26/27). For UR at the third trimester (≥ 28 weeks, $n = 21$), a few (14.29%, 3/21) patients reported adverse fetal outcomes. Fetal outcome was significantly worse in women who had UR during the first and second trimesters compared with those who had UR at the third trimester ($\chi^2 = 33.221$, $p < 0.001$). CS (89.47%, 17/19 reported) was the first management for UR at the third trimester and LPT (81.47%, 22/27) was the first management for UR at the first and second trimesters (Table 1). Of the 48 cases of UR, 22 (81.47%, 22/27) cases of UR were treated with LPT during the first and second trimesters. Long gestational weeks (>30 weeks) resulted in good fetal outcomes (86.67%, 26/30). Spearman correlation analysis showed there was no correlation between the pregnancy interval and gestational age ($\beta = 0.138$, 95% CI −0.147–0.432, $p = 0.356$).

Discussion

The clinical manifestations of UR are complex and varied. The most common manifestations of UR are sudden abdominal pain and hemorrhagic shock with frequent disappearance of fetal heart rate.[29,30] UR often occurs at the late stage of intrauterine pregnancy and in the early and middle stages of interstitial pregnancy. UR is mainly diagnosed intraoperatively.[2,29]

Our patient with her first UR was misdiagnosed because of atypical clinical symptoms, which led to untimely surgical management for her. Clinicians may suspect the possibility of internal and surgical acute abdomen. UR may be diagnosed through a careful gynecological examination and detailed ultrasound examination in most susceptible patients.[2,4,29] Some scholars have pointed out that when pregnant women show abdominal pain, vomiting, and peritoneal irritation symptoms, especially when pelvic effusion is indicated, emergency obstetric services should be scheduled. This service should be scheduled even if the pregnant woman has intrauterine pregnancy, stable vital signs, and a normal range of fetal heart rate.[2] Close observation and priority should be provided to pregnant women who have predisposing factors, including a medical history of CS, salpingectomy, embryo transfer (i.e., IVF), laparoscopic myomectomy, and other laparoscopic uterine surgery.[2,17,31–33]

Early diagnosis and timely treatment can significantly improve maternal and fetal outcomes. In the present case, the risk factors for UR were not taken into consideration at her first admission to our hospital, which led to untimely treatment and an adverse fetal outcome. To avoid an adverse fetal outcome during the second pregnancy, the pregnant woman was closely observed during the last month before delivery and immediately treated with CS at the time of abdominal pain, even if there were no abnormal vital signs in her most recent admission. UR was observed in the right corner of the uterus with a total amount of 200 mL hemoperitoneum. The timely

treatment led to good maternal and fetal (alive, Apgar score: 9–9) outcomes.

Some researchers have shown that IVF may increase the occurrence of UR.[14] Patients who receive IVF show a 2.5 to 5 times higher UR.[2,14,34] However, our literature analysis showed that the incidence of UR in patients who received IVF embryo transfer was 40.00% (10/25). Most of these cases were mainly from assisted reproductive centers,[2,14,25,27,28] while UR in pregnant women without IVF embryo transfer was sporadic.

Transabdominal salpingectomy and hysterectomy are the primary surgical treatments for interstitial pregnancy. Of the cases reported in our literature search, 35 (83.33%, 35/42) women underwent LPSC and seven (16.67%) underwent LPT before UR occurred. A stratified suture is adopted for uterine wounds in LPT, while unipolar or bipolar electrocoagulation hemostasis is usually used for laparoscopy. UR repair is mainly mediated by connective tissue hyperplasia or proliferation, followed by scar fibrosis and muscle cell regeneration. Application of electrocoagulation damages local tissue around the scar,[35–37] and then delayed muscularization of the local tissue and elasticity are poor. Additionally, insufficient suture needles may lead to small hematoma in the myometrium of the uterus, resulting in poor healing of the scar.[3,6,7,12] More studies are required to confirm this hypothesis. For patients with fertility requirements, the advantages and disadvantages of LPT and laparoscopic surgery must be discussed with the patients.

In addition to suture techniques, we suspect that the time interval between conception and the last salpingectomy may affect the incidence of UR. In the literature, the shortest time between conception and the last salpingectomy was 2 months[10] and the longest time was 10 years,[7,15] and 78.26% (36/46) of patients were pregnant within 2 years after surgery. A 2-year period after an operation is considered sufficient for wound healing and scar maturing.[38,39] The present patient had two pregnancies within 1 year, which might be a risk factor for secondary UR. Therefore, patients need to be informed of the risk factors of UR to prevent its occurrence.

Conclusions

Close observation and timely treatment can achieve good outcomes of pregnant women with a risk of UR. Careful review of the patient's medical history and clinicians' experience are important factors for a good prognosis of patients with UR.

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Declaration of conflicting interest

The authors declare that there is no conflict of interest.

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