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 and a leading cause for uterine rupture (UR). Although the overall incidence of UR is low at a rate of less than...
0.1% in the general population, maternal and fetal outcomes are usually poor once UR occurs. 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. 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 $19.4 \times 10^9$/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 ($4 \times 3$ 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 $\chi^2$ 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 $\pm$ 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. 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 (yes/no) | 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. 1996² | 25          | IVF NA                          | <12                       | Yes              | <12                     | Yes              | 10                      | HS, AP, HS                  | LPT+CRU   | Two layers          |
| 2   | Arbab et al. 1996² | 34          | IVF NA                          | <12                       | Yes              | 13                      | Yes              | 10                      | HS, AP, HS                  | LPT+CRU   | Hysterectomy         |
| 3   | Arbab et al. 1996² | 25          | IVF NA                          | <12                       | No               | 20                      | No               | 20                      | UR, AP, HS                  | LPT+CRU   | Two layers          |
| 4   | Arbab et al. 1996² | 33          | IVF NA                          | <24                       | No               | 18                      | No               | 26                      | AP, HS                      | LPT+CRU   | Two layers          |
| 5   | Arbab et al. 1996² | 27          | IVF NA                          | <72                       | Yes              | 10                      | No               | 10                      | HS, AP, HS                  | LPT+CRU   | Two layers          |
| 6   | Kasprovicz et al. 1996¹⁸ | 24          | Natural LPT                       | 3                         | No               | 38                      | No               | 38                      | AP, VB, HS                  | CS+CRU    | Two layers          |
| 7   | Inovay et al. 1999¹⁹ | 31          | IVF LPSC                         | 6                         | No               | 14                      | No               | 14                      | UR, AP, HS                  | LPT+CRU   | Two layers          |
| 8   | Ayoubi et al. 2003²⁰ | 28          | Natural LPSC                       | 24                        | No               | 22                      | No               | 22                      | AP, HS                      | LPT+CRU   | Two layers          |
| 9   | Banaszczyk et al. 2005²¹ | 27          | IVF LPT NA                       | 23                        | No               | 23                      | No               | 23                      | AP, HS                      | LPT+CRU   | NA                  |
| 10  | Su et al. 2008⁵    | 30          | Natural LPSC                       | 6                         | No               | 40                      | No               | 40                      | PR, HS                      | LPT+CRU   | One layer          |
| 11  | Chatterjee et al. 2009²³ | 29          | Natural LPT                       | 6                         | No               | 29                      | No               | 29                      | AP, VB                      | CS+CRU    | Two layers          |
| 12  | Liao et al. 2009²⁴ | 29          | Natural LPSC                       | NA                        | Yes              | 13                      | No               | 13                      | AP, VB                      | LPT+CRU   | Two layers          |
| 13  | Pluchino et al. 2009¹² | 34          | Natural LPSC                       | <12                       | Yes              | 7                       | No               | 7                       | AP, HS                      | LPT+CRU   | NA                  |
| 14  | Muglu et al. 2012⁷  | 31          | Natural LPSC                       | <12                       | No               | 24                      | No               | 24                      | AP, vomiting                | LPT+CRU   | Two layers          |
| 15  | Galati et al. 2013¹⁰ | 29          | Natural LPSC                       | <2                        | No               | 9                       | No               | 9                       | AP, HS                      | LPT+CRU   | Hysterectomy        |
| 16  | Yang 2013¹¹        | 21          | Natural LPSC                       | <10                       | No               | 33 + 2                  | No               | 33 + 2                  | AP, HS                      | LPT+CRU   | NA                  |
| 17  | Cai et al. 2014⁷   | 27          | Natural LPSC                       | 48                        | No               | 36 + 4                  | No               | 36 + 4                  | AP, HS                      | LPT+CRU   | NA                  |
| 18  | Cai et al. 2014⁷   | 30          | Natural LPSC                       | <12                       | No               | 30 + 5                  | No               | 30 + 5                  | AP, HS                      | LPT+CRU   | NA                  |
| 19  | Nishijima et al. 2014¹² | 45          | Natural LPSC                       | 48                        | No               | 26                      | No               | 26                      | AP, HS                      | LPT+CRU   | Two layers          |
| 20  | Gu and Wu 2015⁶    | 36          | Natural LPSC                       | 19                        | No               | 27 + 2                  | No               | 27 + 2                  | AP, VB                      | UR, live birth             | CS+CRU   | Two layers          |
| 21  | Wang 2015⁶        | 24          | Natural LPSC                       | 12                        | No               | 40 + 1                  | No               | 40 + 1                  | PR, HS                      | UR, live birth             | LPT+CRU   | NA                  |
| 22  | Stanisowski et al. 2015³ | 29          | Natural LPSC                       | 14                        | No               | 38                      | No               | 38                      | AP, HS                      | UR, live birth             | CS+CRU    | Two layers          |
| 23  | Tan et al. 2015⁴   | 27          | Natural LPT                        | 13                        | No               | 34                      | No               | 34                      | AP, HS                      | LPT+CRU   | Three layers        |
| 24  | Abbas et al. 2015¹³ | 24          | Natural LPSC                       | >12                       | No               | 39                      | No               | 39                      | Antepartum hemorrhage       | CS+CRU    | Two layers          |

(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 | 28          | IVF                            | LPSC              | >12               | Yes                         | 5               | Internal bleeding         | UR, PT                       | NA         |                  |
| 26  |               | 28          | IVF                            | LPSC              | <12               | No                          | 4               |                           | UR, PT                       | LPT+CRU    | NA               |
| 27  | Marcinak et al. 2016 | 29 | Natural                        | LPSC              | 6                 | Yes                         | 8               |                           | AP, VB                       | UR, PT     | LPT+CRU          |
| 28  | Paradise et al. 2016 | 38 | IVF                            | LPSC              | 120               | No                          | 26              |                           | AP                           | UR, live birth | CS+CRU          |
| 29  | Xu et al. 2016 16 | 25          | IVF                            | LPSC              | 11                | No                          | 36+1            |                           | AP                           | UR, live birth | CS+CRU          |
| 30  | Lin 2017 28     | 42          | Natural                        | LPSC              | 27                | Yes                         | 23              |                           | NA                           | UR, PT     | CS+CRU           |
| 31  |               | 24          | IVF                            | LPSC              | 18                | No                          | 35              |                           | AP                           | UR, live birth | CS+CRU          |
| 32  |               | 38          | IVF                            | LPSC              | 10                | No                          | 36              |                           | FE                           | UR, live birth | CS+CRU          |
| 33  |               | 28          | IVF                            | LPSC              | 9                 | No                          | 20              |                           | AP, vomiting                 | UR, PT     | CS+CRU           |
| 34  | Wu et al. 2018 17 | NA          | NA                             | LPSC              | 11                | No                          | 29              |                           | NA                           | UR         | CRU              |
| 35  | Lin 2018 25    | NA          | NA                             | LPSC              | 4                 | No                          | 31              |                           | NA                           | NA         |                  |
| 36  | Lin et al. 2018 | -30         | IVF                            | LPSC              | >36               | No                          | 32+6            |                           | AP, vomiting                 | UR, live birth | CS+CRU          |
| 37  |               | -30         | IVF                            | LPSC              | >36               | No                          | 35              |                           | AP                           | UR, live birth | CS+CRU          |
| 38  |               | -30         | IVF                            | LPSC              | >36               | No                          | 33              |                           | AP, VB                       | UR, live birth | CS+CRU          |
| 39  |               | -30         | IVF                            | LPSC              | >36               | No                          | 35+1            |                           | AP                           | UR, live birth | CS+CRU          |
| 40  |               | -30         | Natural                        | LPSC              | >36               | No                          | 35+4            |                           | AP                           | UR, live birth | CS+CRU          |
| 41  | Yuan and Peng 2018 | 30 | IVF                            | LPSC              | 5                 | No                          | 34              |                           | AP                           | UR, live birth | CS+CRU          |
| 42  | Jiang and Zhao 2018 | 29 | IVF                            | LPSC              | 9                 | Yes                         | 7               |                           | NA                           | UR         | PT               |
| 43  |               | 35          | Natural                        | LPSC              | 24                | Yes                         | 5+4             |                           | NA                           | UR         | PT               |
| 44  |               | 30          | IVF                            | LPT               | 24                | Yes                         | 7+2             |                           | NA                           | UR         | PT               |
| 45  |               | 35          | IVF                            | NA                | 24                | Yes                         | 7               |                           | NA                           | NA         | PT               |
| 46  |               | 39          | IVF                            | LPT               | 12                | Yes                         | 8+2             |                           | NA                           | UR         | PT               |
| 47  |               | 34          | IVF                            | LPT               | 24                | Yes                         | 6               |                           | NA                           | NA         | PT               |
| 48  |               | 27          | IVF                            | LPSC              | 7                 | Yes                         | 8               |                           | NA                           | UR, fetal death | LPT+CRU      |

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).\textsuperscript{2,14–16,21,25–28} 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 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.\textsuperscript{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.\textsuperscript{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.\textsuperscript{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.\textsuperscript{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.\textsuperscript{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.\textsuperscript{14} Patients who receive IVF show a 2.5 to 5 times higher UR.\textsuperscript{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,\textsuperscript{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,\textsuperscript{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.\textsuperscript{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\textsuperscript{10} and the longest time was 10 years,\textsuperscript{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.\textsuperscript{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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