Successful Term Delivery of Twins after Surgical Treatment of a Ruptured Heterotopic Interstitial Pregnancy: A Case Report


A ruptured interstitial pregnancy combined with an intrauterine twin pregnancy is a rare and challenging problem. We present a 34 year-old nulliparous woman with a combined interstitial and intrauterine twin pregnancy after bilateral salpingectomy and in-vitro fertilization with embryo transfer (IVF/ET). An emergency laparotomy was performed because of internal bleeding at 7 weeks of gestation. A cornual resection was performed and the defect was successfully repaired. The intrauterine twin pregnancy progressed to term uneventfully. Obstetricians should be aware of the possibility of heterotopic pregnancy in women who conceive by assisted reproduction techniques. Early diagnosis and surgical treatment of this ruptured interstitial pregnancy permitted the development of the intrauterine pregnancy.

**Key words:** interstitial pregnancy, heterotopic pregnancy, salpingectomy, in vitro fertilization

**Introduction**

Heterotopic pregnancy, defined as the coexistence of an intrauterine pregnancy and an ectopic pregnancy, occurs in approximately 1: 7000 to 1: 30,000 women in the general population. However, the incidence of heterotopic pregnancy has increased to 1% since the development of assisted reproductive technologies over the past two decades. In ectopic gestation in a heterotopic pregnancy, implantation usually occurs within the ampulla of the fallopian tube. When implantation is within the interstitial portion of the fallopian tube, the resulting heterotopic interstitial pregnancy is a therapeutic dilemma because preservation of the intrauterine pregnancy is desired. Few cases of triplet pregnancy combining intrauterine twin pregnancy and interstitial ectopic pregnancy resulting a live twin birth have been described, particularly after a bilateral salpingectomy.

The article reports on a case of combined interstitial and intrauterine twin pregnancies after in vitro fertilization and embryo transfer following bilateral salpingectomy complicated by cornual rupture at 7 weeks of gestation and subsequent term delivery of the intrauterine twin pregnancy.

**Case Report**

This 34-year-old nulliparous woman was a patient at our infertility clinic with secondary infertility of 6 years’ duration. The initial assessment showed bilateral hydrosalpinx. The patient underwent laparoscopic bilateral
salpingectomy due to unrepairable hydrosalpinx and enrolled in our in-vitro fertilization program. Five weeks later, controlled ovarian stimulation was initiated with a multiple-dose gonadotropin releasing hormone antagonist protocol and 11 oocytes were retrieved (day 0). Three 4-cell stage embryos were transferred into the uterine cavity two days after oocyte retrieval (day 2). At 5 weeks’ gestation, a routine transvaginal ultrasound revealed two intrauterine gestational sacs. At 6 weeks’ gestation, ultrasonographic examination showed an in utero twin gestation with active fetal heart motion in both fetuses, with separate gestational sacs (Fig. 1).

One week later, the patient presented to our emergency room with abdominal pain. There was no vaginal bleeding, vomiting or diarrhea. Initially, her vital signs were stable, but later her condition deteriorated, with episodic dizziness and disorientation during attempts at ambulation. Orthostatic hypotension was noted with a blood pressure of 75/45 mmHg and pulse of 122 beats/min. Her abdomen was tender with rebounding pain. The hemoglobin level fell from 11.9 gm/dl to 7.0 gm/dl within four hours. Ultrasonography demonstrated a viable intrauterine twin pregnancy with fluid accumulation in Morison’s pouch (Fig. 2). Under the impression of internal bleeding, possibly related to a heterotopic pregnancy or ovarian corpus luteum cyst rupture, an emergency laparotomy was performed. The operative findings showed hemoperitoneum of about 1500 ml and a ruptured interstitial pregnancy within the left salpingectomy site. A left cornual resection with removal of the interstitial pregnancy was performed carefully. The uterus was then reapproximated with two layers of 1-0 Dexon sutures, and good hemostasis was obtained. The patient received six units of packed red blood cells before and during surgery. Her hemoglobin stabilized at 9.5 gm/dl postoperatively, and she was discharged on her third postoperative day.

The pathologic examination of the left cornual resection revealed chorionic villi. The intrauterine twin pregnancy progressed uneventfully. At 37 weeks’ gestation, a cesarean section was performed resulting in healthy male and female infants weighing 2235 and 2050 gm, respectively. During surgery, the previous left cornual resection site appeared intact.

**Discussion**

The accurate diagnosis of abdominal pain in...
a woman during early pregnancy is a challenge for the attending physician. The differential diagnosis of abdominal pain in early pregnancy includes pregnancy-associated conditions and conditions unrelated to pregnancy. Common causes of abdominal pain in early pregnancy are listed in Table 1. Our patient presented to the emergency department with abdominal pain at 7 weeks’ gestation following in vitro fertilization (IVF). There was no vaginal bleeding so a miscarriage was not likely. Ultrasound showed a viable intrauterine twin pregnancy with fluid accumulation in Morison’s pouch. With abdominal pain combined with intra-abdominal fluid in a patient who has had IVF, ovarian hyperstimulation syndrome, heterotopic pregnancy and rupture of enlarged, fragile ovaries should be considered. In our case, the falling hemoglobin and hemodynamic instability indicated a possible hemorrhage. Therefore, early transfusion of packed red blood cells to improve the oxygen-carrying capacity to her vital organs and to her fetus was necessary before surgical intervention. The ruptured interstitial heterotopic pregnancy was diagnosed during emergency laparotomy.

The increase in heterotopic pregnancies is undoubtedly a consequence of modern reproductive medicine, because multiple embryos are transferred in women conceiving by IVF. Therefore, limiting the number of embryos transferred has been suggested to minimize the risks of multiple pregnancies after IVF-embryo transfer. Ectopic gestation usually involves implantation within the fallopian tube in most heterotopic pregnancies. Evidence indicates that fluid in hydrosalpinges adversely affects IVF outcomes. A salpingectomy for hydrosalpinx has been suggested before IVF procedures. Unfortunately, interstitial pregnancies have occurred, because such procedures do not result in excision of the patent cornu and intramural segment of the damaged tube.

The development of a pregnancy in the uterine horn carries a high risk of rupture; often hemorrhage is extensive because the uterine cornu has an abundant blood supply from branches of the ovarian and uterine arteries. Early follow-up evaluation of patients after IVF and high resolution transvaginal ultrasonography are essential to the diagnosis heterotopic pregnancy before rupture. However, ultrasonographic diagnosis of an interstitial heterotopic pregnancy is difficult in the presence of hyperstimulated ovaries and a concurrent intrauterine gestational sac. Therefore, only forty percent of cases reported in the literature were diagnosed before rupture. There is no consensus regarding the optimal management of a heterotopic interstitial pregnancy. Several reports have described treatments such as surgical cornual excision, medical treatment and expectant management. The therapeutic objectives are simple--interrupt the evolution of the ectopic pregnancy and preserve the intrauterine pregnancy.

We decided to use a laparotomy for management in our case. No other therapeutic alternative seemed possible because the uterine horn was ruptured with life-threatening hemoperitoneum and hemorrhagic shock. In addition, a laparotomy could ensure a solid myometrial suture, as well as perfect hemostasis.

Table 1  The common causes of abdominal pain during early pregnancy

<table>
<thead>
<tr>
<th>Pregnancy-associated conditions</th>
<th>Pregnancy-unrelated condition</th>
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<tbody>
<tr>
<td>Threatened, incomplete or miscarriage</td>
<td>Acute appendicitis</td>
</tr>
<tr>
<td>Ectopic pregnancy/ heterotopic pregnancy</td>
<td>Acute cholecystitis</td>
</tr>
<tr>
<td>Corpus luteum cyst rupture</td>
<td>Acute gastroenteritis</td>
</tr>
<tr>
<td>Ovarian torsion</td>
<td>Urinary calculi</td>
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</tbody>
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Table 2  Review of the literature describing interstitial pregnancies, with coexisting intrauterine twins

<table>
<thead>
<tr>
<th>Author</th>
<th>Year</th>
<th>Prior tubal surgery</th>
<th>Fertilization methods</th>
<th>Site, performance and management of cornual pregnancy</th>
<th>Outcome of intrauterine twins</th>
</tr>
</thead>
<tbody>
<tr>
<td>Loret de Mola et al.(^{11})</td>
<td>1995</td>
<td>Tuboplasty Right salpingectomy</td>
<td>IVF/ET 5 embryos transferred</td>
<td>Left, ruptured Laparotomy cornual resection at 9 wks</td>
<td>Tocolysis since 26 weeks C-S at 28 wks Live twin: 1172g/ 1111g</td>
</tr>
<tr>
<td>Gaudier et al.(^{12})</td>
<td>1995</td>
<td>Not available</td>
<td>IVF</td>
<td>Right, ruptured Laparotomy cornual resection at 15 wks</td>
<td>Tocolysis since 20 weeks C-S at 34 wks Live twin: 2100g/1500g</td>
</tr>
<tr>
<td>Kasum et al.(^{13})</td>
<td>1998</td>
<td>Bilateral salpingectomy</td>
<td>IVF/ET 4 embryos transferred</td>
<td>Left, ruptured Laparotomy cornual repair at 14 wks</td>
<td>C-S at 36 wks Live twin: 2470g/2490g</td>
</tr>
<tr>
<td>Divry et al.(^{14})</td>
<td>2007</td>
<td>Bilateral salpingectomy</td>
<td>IVF/ET 3 embryos transferred</td>
<td>Right, preruptured Laparotomy cornual resection at 6 wks</td>
<td>Hospitalization at 20 weeks C-S at 31 wks Live twin: 1360g/1590g</td>
</tr>
</tbody>
</table>

Abbreviations: IVF/ET, in vitro fertilization/embryo transfer; C-S: cesarean section

cesarean section was later performed before labor, to avoid the possibility of uterine rupture during delivery.

Most heterotopic interstitial pregnancies have cornual rupture when the gestational age is over 8 weeks\(^{9}\). In our case, the ovulation induction program was started five weeks after the bilateral salpingectomy and the ruptured interstitial pregnancy occurred earlier at 7 weeks’ gestation. This may have been due to poor healing at the salpingectomy site, because the interval between the salpingectomy procedure and the in vitro fertilization program was too short. Blazar et al. described a case of heterotopic interstitial pregnancy following bilateral salpingectomy with cornual rupture at 6 weeks’ gestation, but there was no mention of the interval between the salpingectomy and conception\(^{10}\).

Only a few cases of triplet pregnancy combining intrauterine twin pregnancy and interstitial pregnancy have been described in the literature (Table 2). All four reported women had IVF and two had received a bilateral salpingectomy. The three women with cornual rupture, all received a laparotomy with cornual resection to maintain better control of hemostasis. No uterine rupture was reported and all women delivered live infants by cesarean section before term. To our knowledge, this is the first case of ruptured interstitial pregnancy, with coexisting intrauterine twins after IVF embryo transfer progressing until term delivery.

With the increasing use of assisted reproduction techniques, obstetricians should be aware of the possibility of heterotopic pregnancies in order to diagnose this condition as soon as possible. Interstitial heterotopic pregnancy can
occur in women conceiving by IVF after bilateral salpingectomy, and the uterine cornua should be checked carefully with high-resolution transvaginal ultrasonography. When a diagnosis is reached in time and the site of rupture is carefully repaired, the rate of pregnancies that reach term after treatment is encouraging. Finally, it seems important to limit the number of embryos transferred to prevent heterotopic pregnancy.

References


成功治療破裂間質部異位妊娠之
雙胞胎足月生產：病例報告

陳恒如1,2  黃建榮1,3  林禹宏1,2
蕭國明1  謝碧純1,2

破裂之間質部妊娠合併子宮雙胞胎懷孕是一種少見且具有挑戰性的問題。我們報告一位已接受雙側
輸卵管切除手術之不孕症患者，於接受試管嬰兒及胚胎植入後，仍不幸發生間質部妊娠合併子宮內雙胞
胎懷孕之個案。在懷孕七週時，間質部妊娠破裂引發大量出血，在緊急剖腹手術及子宮角修補止血後，
病人子宮內雙胞胎繼續成長直到足月生產。產科醫師須對接受人工生殖技術懷孕的婦女，保有異位妊娠
的警覺心。唯有早期診斷，早期介入治療，才能保有子宮腔內胚胎繼續生長的可能。

關鍵詞：間質部妊娠，異位妊娠，輸卵管切除，試管嬰兒