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Case report

Laparoscopic resection of cornual heterotopic pregnancy after in vitro fertilization and embryo transfer leading to successful live birth



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ABSTRACT

Heterotopic cornual pregnancy refers to the co-existence of intrauterine and extrauterine cornual pregnancies. This is rare, but the incidence increases in pregnancies associated with in vitro fertilization and embryo transfer (IVF-ET). A 36 year old gravida 2 para 0 woman with a history of bilateral salphingectomy underwent IVF-ET and was diagnosed with a heterotopic cornual pregnancy. She underwent diagnostic laparoscopy and wedge resection of the right cornual ectopic pregnancy. The intrauterine pregnancy progressed uneventfully and was delivered via caesarean section at 36 + 6 weeks. This case report illustrates the importance of early diagnosis of the condition, and how meticulous surgical technique is effective in removing the cornual ectopic pregnancy while preserving the intrauterine pregnancy.

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Introduction

Heterotopic pregnancy refers to the coexistence of intrauterine and extrauterine pregnancies. The incidence of heterotopic pregnancy ranges from 1:7963 to 1:30,000 natural pregnancies. However, the incidence increases to 1:100 in pregnancies associated with in vitro fertilization and embryo transfer (IVF-ET). There may also be other predisposing factors in these gestations that increase the chance of heterotopic pregnancies, such as previously damaged tubes.

The clinical picture proves to be even more challenging in cornual ectopic pregnancies. Its incidence is estimated to be 1:3600 in pregnancies associated with IVF-ET. Early diagnosis is essential but difficult in cornual pregnancies, and significant hemorrhage as well as rupture can result if diagnosed late. The Eighth Report of the Confidential Enquiries into Maternal Deaths in the United Kingdom 2006–2008, reported that six deaths occurred due to ruptured ectopics, and most of them were due to late diagnosis.

We present a case report of a woman who underwent a successful laparoscopic resection of a heterotopic cornual pregnancy that developed after IVF-ET, leading ultimately to the birth of a healthy baby boy.

Case report

A 36-year-old Chinese woman, gravida 2 para 0, had a known history of bilateral hydrosalphinges. She conceived naturally but it resulted in a left ectopic pregnancy, for which she underwent bilateral salphingectomy. After this treatment, she underwent IVF-ET to conceive again. She had two embryos transferred during the procedure.

At a routine follow-up on Day 31 after IVF-ET, ultrasound showed a heterotopic pregnancy, with two gestational sacs and two fetal hearts. Twin A was an intrauterine pregnancy and had a crown-rump length of 52 mm. Twin B was a right cornual pregnancy, with a crown-rump length of 32 mm. There was no sign of any intra-abdominal free fluid.

The patient was stable and asymptomatic upon ultrasound diagnosis. However, she developed sudden onset of lower abdominal pain half a day later. The intensity of the pain increased rapidly. The patient became pale. The blood pressure was 90/ 50 mmHg and pulse rate was 88 beats/minute. The abdomen was slightly distended, with lower abdominal tenderness, as well as guarding and rebound tenderness.

The decision was made for emergent diagnostic laparoscopy with a view to proceed to wedge resection of the right cornual pregnancy. Informed consent was taken. A four-port laparoscopy was performed. Intraoperatively, a 3 cm partially ruptured right cornual ectopic pregnancy was seen (Fig. 1). Both fallopian tubes were absent due to previous salphingectomies. The left ovary was

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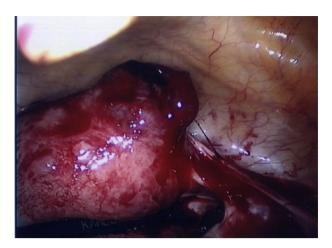


Fig. 1. Ruptured right cornual ectopic pregnancy with active bleeding.

normal. There were 5 L of hemoperitoneum with active bleeding. A wedge resection of the right cornual ectopic pregnancy was performed (Fig. 2). Instruments used were the Richard Wolf Bipolar Coagulating Forceps 3-mm Blade and Ethicon Endo-Surgery Endopath 5-mm Curved Scissors. The pregnancy bed was diathermised with short pulses of energy. The uterine defect was repaired in two layers with monocryl suture (Fig. 3). There was no uterine manipulation intraoperatively. Hemostasis was successfully secured.

The skin-to-skin operating time was 70 minutes. In total, 1500 mL of crystalloids (Ringer's lactate), 500 mL of colloids (Voluven), and one unit of packed red cells were infused. There were no intra- or postoperative complications. An ultrasound scan on the 1st postoperative day revealed a positive fetal heart in the intrauterine gestational sac. The patient was discharged home on the 3rd postoperative day. The pathological report of the surgical specimen concurred with the diagnosis.

The rest of the pregnancy proceeded uneventfully. Antenatal ultrasound scans were unremarkable and there was no antenatal finding of uterine dehiscence. An elective cesarean delivery was planned at 37+3 weeks of gestation. However, the woman went into labor at 36+6 weeks and underwent an emergency cesarean delivery. Intraoperatively, the right cornual edge was noted to be thinned out and bulging, and a defect of about $2 \, \mathrm{cm}$ in diameter was felt in the myometrium (Fig. 4). The defect was repaired in two layers with monocryl suture (Fig. 5).



Fig. 2. Right cornual ectopic pregnancy resected.



Fig. 3. Uterine defect repaired with two-layer closure.



Fig. 4. Bulging defect at cornual edge noted at cesarean delivery.

A healthy baby boy weighing 2784 g was delivered, with Apgar scores of 9 and 9. The mother had an uneventful *postpartum* course and was discharged with her infant after 3 days.

Discussion

The risk factors of heterotopic pregnancy in IVF-ET are an increased number of transferred embryos, excessive pressure on



Fig. 5. Uterine defect repaired with two-layer closure after cesarean delivery.

the syringe during the transfer, transfer near the uterine horn, or difficulties during the IVF-ET procedure. Other risk factors include tubal surgeries, peri- and intratubular adhesions and endometriosis. In our case, the history of bilateral salphingectomy increased the woman's risk of cornual pregnancy after IVF. A review by Habana et al¹ noted a 48.6% incidence of rupture of cornual ectopic pregnancies, which has the potential to lead to massive bleeding, often requiring hysterectomy. In addition, the previous salpingectomies weakened the myometrium of the uterine cornua, thus predisposing to cornual rupture.

Our patient was diagnosed with heterotopic pregnancy on Day 31 after embryo transfer, before she developed any symptoms. Because of its location, early detection of cornual pregnancies is difficult. Upon ultrasound, it is also difficult to differentiate it from an eccentric intrauterine pregnancy. However, in experienced hands, Tulandi and Al-Jaroudi² reported that the diagnosis was established in 71.4% of 32 cases of cornual pregnancies.

When multiple embryos have been transferred, searching for heterotopic pregnancies should be considered even though an obvious intrauterine gestational sac is seen. In addition, the National Institute for Health and Clinical Excellence guidelines for Fertility (2013) recommends transfer of not more than two embryos in each IVF cycle, thus reducing the incidence of multiple pregnancies and heterotopic pregnancies. Certainly, it has been reported that the rate of heterotopic pregnancies for embryo transfers of less than four embryos was 1:119, as opposed to the rate of 1:45 for transfers of more than four embryos.³

The options for treatment of a cornual heterotopic pregnancy include surgery, medical treatment with methotrexate or potassium chloride injection, and expectant management. In a review of the literature of the different modalities of management of cornual heterotopic pregnancies, Chin et al⁴ found that the live-birth rate for the women who underwent surgical management was 57.9%. They concluded that surgical management is the best course of action in the presence of significant symptoms, cornual rupture or large cornual ectopic pregnancies.

Medical and expectant management is associated with an increased risk of continued growth and subsequent rupture. Medical management, with its toxic effects, may also compromise an intrauterine fetus, but can be considered in selected cases in the first trimester in asymptomatic patients. Expectant management has been reported by Fernandez et al,⁵ and was chosen because the patient was clinically asymptomatic and ultrasonography demonstrated a nonviable ectopic pregnancy.

In our case report, the woman quickly developed clinical symptoms of abdominal pain. Thus, surgical management without delay was essential. The uterine cornua has a rich vascularization from branches of the ovarian and uterine arteries, and massive bleeding can result if diagnosis and treatment is delayed. In our patient, there was early recognition of the severity of her clinical condition, allowing us to resect the ruptured ectopic early, preventing any complications from massive hemorrhage.

There is a myriad of surgical techniques described in the treatment of cornual ectopic pregnancies. They include hysteroscopic removal of the ectopic pregnancy, laparoscopic cornuostomy, laparoscopic cornual resection, laparoscopic and ultrasound guided transcervical evacuation of the cornual pregnancy, and laparotomy. Of all these different techniques, laparoscopic resection is most commonly performed.

Laparoscopic resection of a cornual ectopic poses more challenges compared to resection of an ectopic pregnancy located at other areas of the fallopian tube. These include difficulty at achieving hemostasis, preserving cornual integrity for the intrauterine pregnancy and for future pregnancies, as well as the possibility of persistent trophoblastic activity.

Because of the special circumstance of having a coexistent intrauterine pregnancy to preserve while resecting the cornual ectopic pregnancy, there were a few factors that made the surgery more challenging than a usual resection of an ectopic pregnancy.

Firstly, we were unable to antevert the uterus during the resection due to the obvious fact of needing to preserve the intrauterine pregnancy. This would make the surgery more challenging, especially in an obese patient. Secondly, we took pains not to excessively manipulate the uterus externally during the surgery. Thirdly, we were careful not to irrigate and suction copiously so as not to overstimulate the uterus unnecessarily.

Fourthly, intramyometrial vasopressin was not used due to the coexistent intrauterine pregnancy. Herein lies the importance of meticulous suturing postexcision of the ectopic and good laparoscopic technique. Fifthly, there should be adequate resection of the ectopic pregnancy, at the same time avoiding myometrial resection, which would predispose to a higher risk of uterine rupture subsequently. Lastly, there should be minimal electrocautery diathermy to prevent ischemic necrosis, which would harm the growing intrauterine fetus. Thus we were careful to only use short pulses of bipolar diathermy to secure hemostasis for small bleeders, and the importance of suturing for adequate hemostasis is further highlighted. We used a double layer repair to close the uterine defect after resection

Table 1 is a literature review of all reported cornual heterotopic pregnancies that have been treated surgically.

Monitoring of the resolution of the cornual pregnancy is difficult, because the presence of the ongoing pregnancy precludes the trending of human chorionic gonadotropin levels. Thus the patient

 Table 1

 Review of literature describing cornual heterotopic pregnancies treated surgically.

Case no.	Reference	Year	Tubal risk factor	Method of conception	Treatment modality	Outcome of intrauterine pregnancy
1	Lukásová et al ⁶	2012	Tubal factor	IVF-ET	Laparoscopic cornual resection	Prostin induced vaginal delivery at 40 weeks
2	Sherer et al ⁷	1995	Not reported	IVF-ET	Laparoscopic cornual resection at 8 weeks	Cesarean delivery of triplets at 33 weeks
3	Lee et al ⁸	2012	Bilateral salphingectomy	IVF-ET	Laparoscopic resection at 7 weeks	Cesarean delivery at term
4	Vilos ⁹	1995	Left salphingectomy	Ovulation induction	Laparoscopic right salphingectomy and cornual resectionat 7.5 weeks	Normal spontaneous delivery at 37.5 weeks
5	Chachan et al ¹⁰	2009	Nil	Clomiphene induced	Laparoscopic resection at 6.5 weeks	Elective cesarean delivery at 38 weeks
6	Divry et al ¹¹	2007	Bilateral salphingectomy	IVF-ET	Laparoscopy converted to laparotomy and cornual resection	Cesarean delivery of twins at 31 weeks

was monitored very closely throughout the pregnancy via ultrasound scans.

We advocate a planned elective cesarean delivery at term due to the high risk of uterine rupture in labor, and this was scheduled in advance for our patient. In fact, the finding of a thinned out and bulging corneal edge during the cesarean delivery reinforces this point. Furthermore, such pregnancies tend to be precious pregnancies conceived via IVF.

With the rise in IVF-ET, the incidence of cornual heterotopic pregnancies is set to be on the rise. This case report, together with the previous above-mentioned reports, demonstrates that laparoscopic resection is a reproducible technique in such a scenario with reproducible favorable results.

In conclusion, the early diagnosis and management of cornual ectopic pregnancies remains a challenge. The possible diagnosis should always be borne in mind especially for those with risk factors. The visualization of an intrauterine pregnancy in a patient who has undergone IVF-ET does not rule out the possibility of a simultaneous ectopic pregnancy. Because of the need to consider the preservation of the intrauterine pregnancy, treatment options are often not straightforward, and have to be tailored individually to each case. Laparoscopic surgery, if carried out by experienced hands, is safe and effective, a reliable diagnostic tool, and averts the need for laparotomy or further treatment.

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