DELAYED UTERINE RUPTURE AFTER FETAL REDUCTION IN A CASE OF CORNUAL HETEROTOPIC PREGNANCY

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SUMMARY

Objective: Assisted reproductive technology has contributed to the rising rate of multiple and ectopic pregnancies. We report a case of heterotopic cornual pregnancy with delayed uterine rupture despite successful fetal reduction. To our knowledge, this has not been previously reported.

Case Report: A 32-year-old woman, gravida 2, para 0, had secondary infertility. She had undergone laparoscopic tuboplasty for bilateral tubal obstruction and laparoscopic bilateral salpingectomy for hydrosalpinx. Successful pregnancy was achieved after transfer of five frozen embryos for this pregnancy. At 7 weeks of gestation, routine pelvic sonography identified three gestational sacs, two in the intrauterine cavity and one in the right cornua. Fetal reduction with potassium chloride injection into the cornual pregnancy was performed at 8 weeks of gestation in a private clinic. At 13 weeks of gestation, she had sudden-onset low abdominal pain and hypovolemic shock. Emergency laparotomy revealed right cornual rupture, with a 3.4-cm translucent sac extruding into the peritoneal cavity. The uterus was repaired by simple closure of the right cornua. The twins survived the operation and were born smoothly at 34 weeks of gestation by cesarean section due to preterm labor and malpresentation. **Conclusion:** Uterine rupture can occur in women who have undergone successful fetal reduction for cornual heterotopic pregnancy. [*Taiwanese J Obstet Gynecol* 2005;44(3):270-272]

Key Words: cornual pregnancy, fetal reduction, uterine rupture

Introduction

Heterotopic pregnancy refers to the co-existence of intrauterine and extrauterine gestation. The incidence of heterotopic pregnancy has risen significantly since the emergence of assisted reproductive technology (ART), and is estimated to be as high as 1 in 100 of ART pregnancies [1–3]. As cornual pregnancy accounts for approximately 3–4% of all ectopic pregnancies [1], the incidence of cornual heterotopic pregnancy after *in vitro* fertilization (IVF) is estimated to be 1 in 3,600

**Correspondence to:* Professor Pao-Lin Kuo, Department of Obstetrics and Gynecology, National Cheng Kung University Medical Center, 138 Sheng Li Road, Tainan 704, Taiwan. E-mail: paolink@mail.ncku.edu.tw Received: November 17, 2004 Revised: November 18, 2004 Accepted: November 18, 2004 pregnancies achieved by ART. To date, heterotopic cornual pregnancy with delayed uterine rupture after successful fetal reduction has not been reported. Herein, we report a case of heterotopic cornual pregnancy with intrauterine twin pregnancy. Although the mother underwent successful fetal reduction of the cornual gestation, she developed delayed uterine rupture at 13 weeks of gestation. After emergency surgery, the remaining twin pregnancy continued successfully to 34 weeks of gestation.

Case Report

A 32-year-old woman, gravida 2, para 0, suffered from secondary infertility without any systemic disease. She had undergone laparoscopic tuboplasty for bilateral tubal obstruction in 2001 and laparoscopic bilateral salpingectomy for hydrosalpinx in 2002. The first cycle of IVF-embryo transfer in August 2002 failed. She underwent the procedure again in November 2002, when five frozen embryos were transferred. At 7 weeks of pregnancy, routine pelvic sonography showed three gestational sacs, one located in the right cornual area and two in the uterine cavity.

Fetal reduction with potassium chloride injection into the right cornual pregnancy was performed at 8 weeks of gestation at a local clinic, and no further fetal development in this sac was observed in the subsequent follow-up. At 13 weeks of gestation, sudden-onset low abdominal pain and hypovolemic shock developed and she was sent to our emergency service. She presented with hypotension (78/48 mmHg) and tachycardia (132 bpm). Pelvic examination revealed an acute abdomen with diffuse rebounding pain. Subsequent pelvic sonography revealed intrauterine viable twin pregnancies (with crown-rump lengths of 6.5 and 6.1 cm, respectively) (Figure 1) and a right cornual sac of $3.4 \times$ 2.4 cm (Figure 2). The cul-de-sac had 4 cm of echo-free space and Morrison's pouch had 2 cm of echo-free space. The patient underwent emergency exploratory laparotomy for suspected twin pregnancies with internal bleeding.

During surgery, around 2,000 mL hemoperitoneum and right cornual rupture with active bleeding was noted. In addition, a translucent gestational sac protruding into the peritoneal cavity from the right cornual area was detected. After removing the blood clot and protruding gestational tissue, the right cornual area was repaired using 2-0 Dexon interrupted sutures. After surgery, both intrauterine twins survived until 31 weeks of gestation, when preterm labor started. With tocolysis, the preterm labor was controlled for 3 weeks. At 34 weeks of gestation, tocolysis failed to control preterm labor and an emergency cesarean section was



Figure 1. Viable twin pregnancies with crown-rump lengths of 6.5 cm and 6.1 cm, respectively.



Figure 2. Right cornual gestation which had shrunk to a sac of 3.4×2.4 cm after selective fetal reduction.

performed due to malpresentation. Two healthy twin babies, one male and one female, were born, with birth weights of 2,200 g and 2,375 g, respectively. Their Apgar scores were 8 at 1 minute and 9 at 5 minutes. At the time of writing, the twins were live and well after a check-up at 1 year.

Discussion

Patients undergoing IVF are at a relatively increased risk of cornual heterotopic pregnancy because of the high multiple and ectopic pregnancy rates. The incidence of heterotopic pregnancy with the extrauterine gestation located in the cornual area after IVF is not known, but is calculated at around 1 in 3,600 IVF pregnancies [4]. However, no case of ruptured heterotopic cornual pregnancy after IVF and fetal reduction has been reported in Taiwan or in the international medical literature. Although bilateral total salpingectomy is generally considered to eliminate the risk of ectopic pregnancy, it does not eliminate the risk of cornual pregnancy. Infertile women with a history of salpingectomy may be at increased risk of cornual pregnancy after IVF [5], with an underlying mechanism of previous salpingectomy weakening the myometrium of the uterine cornua and, thus, predisposing to cornual rupture.

Diagnosis of cornual pregnancy is often delayed, with only 10% of cases in the literature diagnosed before surgical intervention [6]. Such delays in diagnosis may result in a catastrophic outcome, because the uterine cornua has an abundant blood supply from branches of the ovarian and uterine arteries. To date, only one cornual pregnancy has been reported to survive beyond 26 weeks of gestation [7]. Careful monitoring with serial measurement of β -human chorionic gonadotropin, pelvic examination and ultrasonography is very important in pregnancies resulting from ART, allowing early detection of abnormal pregnancies.

Treatment options for cornual pregnancy include surgical, medical, and expectant treatment. Surgical management includes cornual resection, linear incision, or hysterectomy via either laparoscopy or laparotomy. The choice of operation depends on the lesion and the patient's condition, especially regarding whether she would like to have further pregnancies. Medical treatment with potassium chloride [6,8,9], with or without methotrexate [10], has been used for reduction of cornual gestations in heterotopic pregnancies, especially in early gestation before rupture. In a series described by Habana et al [4], the gestational age at diagnosis $(7.1 \pm 0.8 \text{ weeks})$ was younger than that in the surgical group. Of 11 patients, 10 conceived after ART and heterotopic pregnancy was diagnosed early on serial sonar evaluation [4]. All 11 patients had successful resolution of the cornual gestation and did not need surgical intervention [4]. Expectant management was reported by Fernandez et al [6], and was chosen mainly because the patient was free of symptoms and ultrasonography documented fetal death for the cornual gestation. Generally speaking, medical treatment seems to be preferred over surgical treatment and expectant management for cornual heterotopic pregnancy when the cornual pregnancy is diagnosed early in gestation [4]. In our case, however, uterine rupture occurred despite successful fetal reduction.

Usually, heterotopic cornual pregnancy has a smooth course after successful fetal reduction. There are no previous reports of a heterotopic pregnancy rupturing after successful fetal reduction. To the best of our knowledge, this is the first case report regarding uterine rupture after successful fetal reduction of a cornual heterotopic IVF pregnancy, either in Taiwan or worldwide. From this case, we recommend careful surveillance of patients who undergo successful fetal reduction for heterotopic cornual pregnancy.

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