

Twin intrauterine and cornual gestation in a case of triplet pregnancy

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ABSTRACT

Ectopic implantation with in-vitro fertilization–embryo transfer may occur in the cornu or tubal stump, which is otherwise rare. Our patient with previous left salpingostomy and right salpingo-oophorectomy had 4 embryos transferred through in vitro fertilization out of which 3 were successfully implanted with twin intrauterine gestation and cornual pregnancy. The cornual pregnancy ruptured at 12 weeks of gestation and the twin intrauterine pregnancy had a successful outcome.

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A heterotopic pregnancy is the coexistence of one or more ectopic and a single or multiple ectopic pregnancy. It occurs in 1% of pregnancies after in-vitro fertilization (IVF) and embryo transfer.¹ The incidence of monozygotic twinning has been estimated to increase after assisted reproductive techniques (ART).² Salpingectomy has been used in an attempt to eliminate the risk of tubal pregnancy. However, many such patients subsequently will undergo in-vitro fertilization–embryo transfer (IVF–ET).

This article reports one such case of triplet IVF pregnancy consisting of an ectopic interstitial pregnancy (cornual pregnancy) and an intrauterine twin pregnancy. The patient had undergone bilateral salpingectomy. After laparotomy for the ruptured cornual pregnancy at 12 weeks and repair of the uterine rent (fetus lying in the abdominal cavity), the intrauterine twin pregnancy achieved a subsequent successful outcome.

Case Report. A 31-year-old lady presented to Advanced Fertility and Genetic Centre in September 2002 with secondary infertility of 7 years duration.

The patient had irregular menstrual cycles, but her first pregnancy was spontaneous and she delivered by cesarean section in 1994. Her next 2 pregnancies were ectopic gestations in 1995 and 1996, when she underwent laparotomy with left salpingostomy in the first and laparotomy with right salpingo-oophorectomy in the second pregnancy. The patient had diagnostic laparoscopy in 2000, which confirmed the presence of a single ovary. Her systemic and vaginal examinations were unremarkable and vaginal scan showed normal sized anteverted uterus and only left ovary. Hormonal profile was normal. Her husband had adequate seminal fluid volume. The couple was counseled for conception by IVF due to tubal factor. The higher chance of repeat ectopic pregnancy was also stressed to the couple.

The first IVF cycle carried out in October 2002 failed and the second IVF attempt was after 2 months, in December 2002. Ovarian stimulation was initiated with ganirelix (gonadotropin releasing hormone antagonist) and recombinant follicular stimulating hormone (Puregon). She showed adequate response and ovulation was triggered by

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Figure 1 - Twin pregnancy.



Figure 2 - Right cornual pregnancy.

human choriongonadotropin (hCG) 10,000 units and 34 hours later transvaginal ultrasound guided follicle aspiration was performed, where 11 oocytes were retrieved. Ten oocytes were injected by intracytoplasmic sperm injection resulting in the fertilization of 8. In January 2003, 4 embryos were transferred using curved cooked catheter. Luteal phase support by natural progesterone (Cyclogest 400mg bid vaginal suppositories) was started.

Her first beta hCG 2 weeks after embryo transfer was 980MIU/ml and was repeated after 48 hours and was 2910MIU/ml, which raised the possibility of multiple pregnancy. First ultrasonography scan (USG) in February 2003 showed 5 weeks and 4 days, 2 intrauterine gestational sacs measuring 10.4 and 11mm. Progesterone support was continued. During her next visits at 7 and 10 weeks gestation, she was asymptomatic and USG scan showed 2 intrauterine viable fetuses. No adnexal masses were seen.

At 12 weeks twin pregnancy, she presented with lower abdominal pain and giddiness. On examination patient was afebrile, vitals stable, uterus 18 weeks size relaxed. There was tenderness in right iliac fossa. First ultrasonography scan revealed intrauterine live twin pregnancy and another sac was seen on the right side with fetal pole and cardiac activity, possibility of cornual pregnancy was put in mind (**Figure 1 & 2**). Patient collapsed soon after admission. She was in hypovolemic shock, resuscitated and shifted to operation theatre for laparotomy. Rescan before laparotomy confirmed the presence of ruptured right cornual pregnancy.

Operative findings revealed right-ruptured cornual pregnancy with fetus lying in the abdominal cavity, uterine size 18 weeks with absent right tube and right ovary. Left tube with previous salpingostomy scar was seen along with left ovary. The site of uterine rupture was repaired. Patient

received 3 units of blood pre and intraoperatively. Postoperative period was uneventful and scan at the time of discharge revealed diamniotic dichorionic live twin pregnancy. Patient was discharged on hematinics and hydroxyprogesterone caproate (Proluton depot) 500mg intramuscularly weekly. Patient had regular antenatal visits and had an elective lower segment cesarean section at 36 weeks pregnancy with successful outcome of 2 healthy female babies, 2.5 and 2.8kg in weight.

Discussion. Child bearing with ART is associated with higher rates of interstitial and heterotopic pregnancy.³ Heterotopic pregnancy is defined as the coexistence of an intrauterine pregnancy and an ectopic pregnancy. The unique anatomic location of an interstitial pregnancy commonly leads to a delay in diagnosis ensuing chances of rupture of uterus beyond 12 weeks of amenorrhea.⁴ Salpingectomy or cornual occlusion has been suggested as a preventive measure for ectopic pregnancy before IVF cycle. A 7-year study on 515 pregnancies resulting after transfer of either fresh or frozen embryos had 1.3% pregnancies located in the cornual or the tubal stump even after prior salpingectomy.²

The combination of high index of suspicion, sensitive serum beta hCG assays and transvaginal sonography has revolutionized the diagnosis of tubal pregnancy. Interstitial pregnancy, however, remains the most difficult ectopic pregnancy to diagnose preoperatively.^{4,5} The main issue in the treatment of heterotopic pregnancy is to be minimally invasive as possible to preserve the intrauterine pregnancy. Our patient is unique, not only did she have a heterotopic pregnancy after left salpingostomy and right salpingo-oophorectomy but with rupture of cornual pregnancy, later had a successful twin pregnancy. The patient underwent laparotomy at 12 weeks of pregnancy for ruptured

cornual pregnancy with hemoperitoneum, and repair of the site of rupture in uterus. Minimal handling of the uterus with proper repair of the rent was carried out to achieve successful outcome of the viable intrauterine twin pregnancy.

Current recommendations of the American Society of Reproductive Medicine suggest that 2 good quality embryos should be replaced in young women in view of multiple embryo transfer being considered as a predisposing factor for heterotopic pregnancy.^{1,4} Replacement of only 2 good quality embryos could have theoretically prevented the occurrence of this rare complication.^{6,7}

In conclusion, heterotopic interstitial ectopic pregnancy is a rare type of ectopic pregnancy whose rate will escalate secondary to the worldwide use of ART even after bilateral salpingectomy. Bilateral salpingectomy cannot avoid subsequent heterotopic pregnancy completely. Catastrophic conditions may occur, as the ectopic gestation is always located within the interstitial, rather than ampullary portion of the fallopian tube. Women undergoing IVF-ET must be informed of the risk of ectopic pregnancy. Irrespective of the availability of newer diagnostic modalities, obstetrician has to be cautious for symptoms and signs of abdominal pain or vaginal

spotting, keeping in view the probability of undetected interstitial pregnancy that may rupture.

References

1. Dor J, Seidman DS, Levran D, Ben-Shlomo I, Mashiach S. The incidence of combined intrauterine and extrauterine pregnancy after in vitro fertilization and embryo transfer. *Fertil Steril* 1991; 55: 833-834.
2. Agarwal SK, Wisot AL, Garzo G. Cornual pregnancies in patients with prior salpingectomy undergoing in-vitro fertilization and embryo transfer. *Fertil Steril* 1996; 65: 659-660.
3. Wenstrom KD, Syrop CH, Hammit DG, Van Voorhis BJ. Increased risk of monozygotic twinning associated with assisted reproduction. *Fertil Steril* 1993; 60: 510-514.
4. Lau S, Tulandi T. Conservative medical and surgical management of interstitial ectopic pregnancy. *Fertil Steril* 1999; 72: 207-215.
5. Ackerman TE, Levi CS, Dashefsky SM, Holt SC, Lindsay DJ. Interstitial Line: Sonographic Finding in Interstitial (Cornual) Ectopic Pregnancy. *Radiology* 1993; 189: 83-87.
6. Chang Y, Lee JN, Yang CH, Hsu SC, Tsai EM. An unexpected quadruplet heterotopic pregnancy after bilateral salpingectomy and replacement of 3 embryos. *Fertil Steril* 2002; 80: 218-220.
7. Hu Y, Maxson WS, Hoffman DI, Ory SJ, Eager S, Dupre J, et al. Maximising pregnancy rates and limiting higher-order multiple conceptions by determining the optimal number of embryos to transfer based on quality. *Fertil Steril* 1998; 69: 650-657.