

CASE REPORT

Heterotopic Quadruplet Pregnancy After ICSI Conception

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About the Author



Nupur Tamhane was a resident at KEM Hospital, Pune, in the Department of Obstetrics and Gynecology. She completed her M.B.B.S from B.J. Govt. Medical College, Pune. She and her co-author Aditi Parikh followed this case throughout the gestational period and also assisted in patient's surgical intervention and postoperative period. They were involved in drafting the manuscript and revising it critically for important intellectual content and acquisition of data under the guidance of Dr. Vivek Joshi.

Abstract

Background Heterotopic pregnancy (HP) is a condition characterized by the coexistence of multiple fetuses at two or more implantation sites. It occurs in 1% of pregnancies after assisted reproductive techniques (ART). Presence of triplet intrauterine pregnancy with ectopic gestational sac is

one of the rarest forms of HP. Ectopic pregnancy is implanted in the ampullary segment of the fallopian tube in 80% of cases. Most of the patients present with acute abdominal symptoms due to rupture of the tube.

Case Presentation This article reports a case of quadruplet heterotopic pregnancy after intracytoplasmic sperm injection (ICSI) with an ampullary ectopic pregnancy and intrauterine triplet pregnancies. The ruptured ampullary pregnancy was emergently managed by right salpingectomy. This was followed by embryo reduction at 12 + 6 weeks and successful outcome of intrauterine twin pregnancy.

Nupur A. Tamhane and Aditi Parikh have contributed equally.

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Case Presentation

A 32-year-old woman at 10 + 4-week gestation by dates (an ICSI conception) presented with chief complaints of severe pain in abdomen since 24 h and per vaginal spotting since 1 month. She had a spontaneous ectopic tubal pregnancy 2 years back, which was medically managed. On admission, her pulse was 110/min and blood pressure 110/70 mm of Hg and on per abdominal examination there was tenderness in right iliac fossa. Laboratory values showed elevated leukocyte count $11,300/\text{mm}^3$ and Hb 8.4 mg/dl. Ultrasound examination was suggestive of triplet live intrauterine gestation with fetus 1 CRL 3.7 cm (10 + 5 weeks), fetus 2 CRL 4 cm (10 + 6 weeks), fetus 3 CRL 3.4 cm (10 + 6 weeks) and another sac in the right iliac fossa with viable fetus of CRL 4.5 cm (11 + 2 weeks) (Fig. 1). Minimal free fluid was seen in paracolic space and hepatorenal fossa. Blood grouping and cross-matching were done. Patient was shifted to operation room, and laparotomy was performed. Right tubular ampullary ruptured ectopic pregnancy with blood, and blood clots in the cul-de-sac were found intraoperatively. Flimsy adhesions were present between bowel and posterior wall of uterus. While manipulating the right fallopian tube, fetus got extruded through the rent in the ampulla (Fig. 2). Right salpingectomy and evacuation of blood from the pelvis were performed with minimal manipulation of the uterus. One-unit PCV was transfused intraoperatively. The histopathological report confirmed tubal pregnancy with the presence of chorionic villi and trophoblastic tissue in the fallopian tube. Postoperatively, patient was given intravenous antibiotics and nifedipine sustained release 20 mg BD was given for tocolysis for a week. Also, progesterone support was given with hydroxyprogesterone caproate 250-mg single injection and dydrogesterone 20-mg tablets BD for 1 week. A repeat ultrasound on postoperative day seven revealed live triplet intrauterine gestation. At 12 + 6 weeks, patient underwent embryo reduction and pregnancy was reduced to twin gestation. At 16 weeks, McDonald's cervical cerclage was performed as the cervical length was less than 2.5 cm. At 30 + 6 weeks, she had preterm labor. Two doses of injection betamethasone were given for fetal lung maturity, and tocolysis was achieved with tocolytics and she was discharged.

At 34 + 2 weeks, the patient was admitted for preterm labor with first fetus with breech presentation confirmed by ultrasound. On per vaginal examination, cervical dilatation was 3 cm with 70% effacement and station 1. Patient was taken for lower segment cesarean section, and McDonald's suture removed. Intraop and post-op courses were uneventful. Both babies weighed more than 2 kg and did not require NICU services and care.



Fig. 1 Ultrasound showing quadruplet heterotopic pregnancy with triplet intrauterine gestation and right tubal ectopic pregnancy

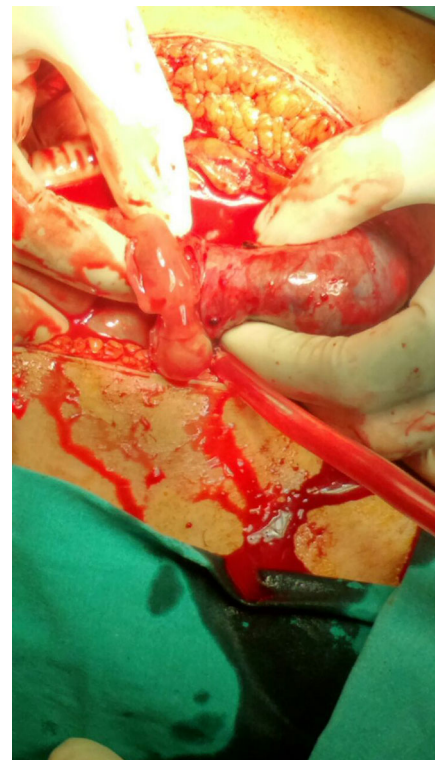


Fig. 2 Fetus being extruded out through ampulla of right ruptured fallopian tube

Discussion and Conclusions

The incidence of heterotopic pregnancy in general population is estimated to be as low as 1.25:10,000 [1]. However, after the advent of ART, there has been an increase in incidence to 33:10,000 after ovulation induction and 100:10,000 after IVF [1]. The risk of heterotopic pregnancy

in a woman undergoing infertility treatment further increases due to the presence of additional factors such as ovulation induction, higher incidence of tubal malformation and damage and factors associated with embryo transfer. Additionally, ART by itself predisposes woman to multiple gestation. Most of the reported heterotopic pregnancies have single intrauterine pregnancy. However, triplet, quadruplet and sextuplet intrauterine gestations have rarely been reported [2]. While performing ART, certain factors should be considered which could influence the incidence of ectopic pregnancy. Day 5 embryo transfer compared to day 3 embryo transfer has significantly lowered the rate of ectopic pregnancy [3]. Additionally, the likelihood of ectopic pregnancy was lower for frozen blastocyst transfer as compared with fresh blastocyst transfer [4]. Recent studies have shown no correlation between number of transferred embryos and risk of heterotopic pregnancy [1].

Diagnosing heterotopic pregnancy in a woman presenting with acute abdomen is a challenge in itself. Differential diagnosis could be ovarian torsion, appendicitis and ovarian hyper-stimulation syndrome (OHSS). Moreover, if the first trimester scan has already confirmed intrauterine pregnancy, the diagnosis of a simultaneous ectopic pregnancy is often missed. Physicians should have a high index of suspicion to diagnose heterotopic pregnancy especially if the patient has undergone ART. The most important tool in diagnosing heterotopic pregnancy is transvaginal sonography, which can detect cardiac activity in both intrauterine and ectopic gestational sacs. In case, if patient presents after rupture, blood in pouch of Douglas and paracolic gutters can be detected by sonography. Our patient was referred to us with heterotopic quadruplet pregnancy with live triplet intrauterine gestation and right ruptured tubal ectopic gestational sac detected by sonography. (Details of number of embryos transferred were not available). Such cases have to be managed in an emergent manner as it is a life-threatening condition. Laparotomy with salpingectomy and minimal uterine handling is the

primary management. Further, the intrauterine pregnancies should be managed appropriately for a successful outcome.

Multiple gestation incurs the risk of preterm delivery with its inherent complications such as low birth weight, respiratory distress syndrome and intraventricular hemorrhage to name a few. Hence, we decided to perform embryo reduction and reduce the intrauterine triplets to twin gestation sacs. To our best knowledge, in the past 5 years, only a single case has been reported in the English literature where embryo reduction was done as an additional management procedure for higher-order heterotopic pregnancy with successful outcome [2].

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Compliance with Ethical Standards

Conflict of interest The authors declare that they have no competing interests.

Research Involving Human Participants and/or Animals Not applicable.

Informed Consent Informed consent was obtained from all individual participants included in the study.

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