

# HETEROTOPIC OVARIAN PREGNANCY

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Heterotopic pregnancy occurs when an intrauterine pregnancy coexists with an ectopic pregnancy [1,2]. Preoperative diagnosis of this rare event is difficult, and the situation is potentially dangerous for both the mother and the intrauterine fetus. The estimated incidence of heterotopic pregnancy is accepted to be between 1 in 30,000 and 1 in 8,000 pregnancies. The risk of this event has increased owing to the widespread use of ovulation induction therapies and assisted reproductive techniques in modern medicine [3].

A pregnancy implanted within an ovary is termed an ovarian pregnancy. If the primary nidation is within the tube and the ovarian attachment results from tubal abortion, it is termed a "secondary ovarian pregnancy". If implantation only occurs in the ovary, it is a "primary ovarian pregnancy" [4].

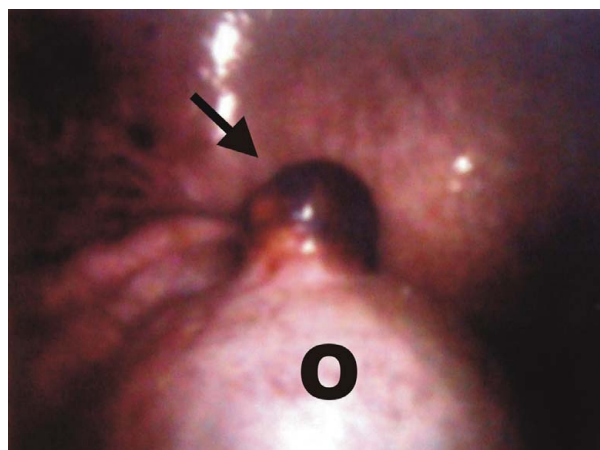
We describe a rare case of primary heterotopic ovarian pregnancy combined with an intrauterine twin pregnancy that was successfully managed using operative laparoscopy.

A 29-year-old, gravida 2, para 0, woman presented at the emergency department of our hospital with a chief complaint of lower abdominal pain for 2 hours. She had a 3-year history of secondary infertility and had conceived after clomiphene and gonadotropin ovulation induction during this cycle. During gonadotropin hyperstimulation, five dominant follicles had been observed on the day of human chorionic gonadotropin injection. On examination in the emergency department, there was generalized abdominal tenderness with rebound and guarding. Speculum examination revealed minimal bleeding from the cervical os. The uterus appeared to be slightly enlarged. The level of the  $\beta$ -subunit of human chorionic gonadotropin of the patient was 81,361 mIU/mL. Her hemoglobin concentration was 7.3 mg/dL. An ultrasound scan detected two viable intrauterine pregnancies of 8 weeks' gestation, but a cystic mass was also present on the left adnexal site. This

included a large amount of fluid in the cul-de-sac and abdominal cavity. Emergency laparoscopy was performed based on a diagnosis of internal bleeding.

The operative findings showed an enlarged uterus consistent with an 8 weeks' gestation intrauterine twin pregnancy, and about 400 mL of blood in the cul-de-sac. The gross appearance of the bilateral oviducts was normal, and there was no bleeding from either fimbria. A left ovarian cyst (6 × 5 cm in diameter) was found, and a dark reddish cystic mass (2 × 2 cm; Figure) was visible over the upper part of the ovarian cyst, suggestive of the products of conception. Blood was seen to be oozing from this ectopic mass. Laparoscopic ovarian cystectomy, including the ectopic mass, was performed, and hemostasis was achieved. Histopathology confirmed an ovarian pregnancy with a trophoblast and villi adjacent to the ovarian cystic wall. The patient was discharged on the second postoperative day after an uneventful recovery. The pregnancy progressed in an uncomplicated manner, and she delivered two healthy babies at 36 weeks' gestation. Delivery was by caesarean section because of malpresentation.

Concomitant intrauterine and ectopic pregnancies, a so-called heterotopic pregnancy, is a rare event. The incidence of heterotopic pregnancies is reported to range from 1 in 30,000 to 1 in 7,963. [5] However, the incidence is higher in women with a history of pelvic inflammatory disease, tubal surgery or previous ectopic



**Figure.** An ectopic mass (arrow) was found on the surface of the ovarian cyst (O).



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pregnancy. It is associated with assisted fertilization involving ovulation-stimulating drugs or *in vitro* fertilization. An incidence rate as high as 1 in 100 has been reported during assisted reproduction [6]. Very few heterotopic pregnancies reach full term, and these are associated with high rates of maternal and fetal mortality.

Ectopic implantation in the ovarian tissue appears to be rare among heterotopic pregnancies. Reece et al [5] reviewed 66 cases of combined pregnancies, of which only four (6%) were ovarian gestations. There is an increased incidence of heterotopic pregnancy after ovulation induction, but most extrauterine gestations involve implantation in the fallopian tube. Few publications have described simultaneous ovarian and intrauterine pregnancies [7–9].

A final diagnosis of a primary ovarian pregnancy can only be made following histopathologic examination of surgical specimens, on the basis of Spiegelberg's four principles [10]: (1) the gestational sac must occupy the normal position of the ovary; (2) the tube on the side of the pregnancy must be normal; (3) the gestational sac must be connected to the uterus by the utero-ovarian ligament; and (4) ovarian tissue must be histologically determined to be present in the wall of the gestational sac. Our reported case fulfilled all the above criteria. The etiology of ovarian pregnancy is obscure. Fertilization is probably extraovarian, but abundant granulosa cells, which form a sticky cumulus round the corona radiata of the ovum, may adhere to the ruptured follicle. If there are enough sperm with enough hyaluronidase to lyse the radiata and expose the zona pellucida to allow sperm penetration, fertilization occurs and the resulting zygote develops. Our patient had undergone ovulation induction with clomiphene and gonadotropin with resulting multifollicular development. This could have predisposed her to this fertilization process.

The majority of heterotopic pregnancies are diagnosed late. Significant morbidity and occasional mortality have been reported as a result of a delay in diagnosis. Patients frequently present with nonspecific symptoms and signs. Abdominal and vaginal ultrasonography often fails to show the ectopic pregnancy, or else the results are misinterpreted because of the awareness of an existing uterine pregnancy. It is also difficult to make a preoperative diagnosis of an ovarian (and, therefore, also a heterotopic ovarian) pregnancy with certainty, as the clinical history and findings are similar to those of a tubal ectopic pregnancy or a ruptured ovarian cyst. In our case, the patient had a history of secondary infertility, and conception occurred after ovulation induction. Emergency surgical intervention was indicated because of the acute abdominal pain and a hemoperitoneum. Based on our previous clinical experience,

a heterotopic pregnancy or a ruptured ovarian cyst was preoperatively suspected as a possible diagnosis. However, the even more rare condition of heterotopic ovarian pregnancy was finally diagnosed.

Laparoscopic surgery provides a superior method for diagnosis and management of ectopic or ovarian pregnancies owing to its reduced hospital stay, limited postoperative analgesia requirements, lower cost, lower risk of adhesion formation, and better visualization of the operative field. In our case, the ectopic gestation tissues were connected to the left ovarian cystic wall and could easily be removed laparoscopically with only minimal manipulation of the enlarged uterus. In women with heterotopic pregnancies, the prognosis for an intrauterine pregnancy is much better when the event is treated early. After management of the extrauterine pregnancy, 75% of mothers deliver at full term, 16% deliver preterm, and only 9% have a stillbirth or a spontaneous abortion [5]. It is important that the uterus is disturbed as little as possible during the course of surgery and instrumental manipulation of the uterus should be avoided. In this case, there were no apparent ill effects to the fetus caused by the hemoperitoneum.

In conclusion, every physician treating women of reproductive age should be aware of the possibility of ovarian (and heterotopic) pregnancies. The presence of an intrauterine pregnancy cannot rule out the possibility of a heterotopic pregnancy. Conservative management can lead to a successful intrauterine pregnancy outcome.

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