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CASE REPORT

Successful Outcome of Spontaneously Conceived Heterotopic Pregnancy masquerading Ovarian Torsion

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ABSTRACT

Aim: To sensitize clinicians regarding possibility of heterotopic pregnancy even in spontaneous conceptions.

Background: Though heterotopic pregnancy is a rare condition, its incidence is rising, which varies from one in 8,000 to 30,000, more in assisted reproductive technique conceptions. It is being commonly misdiagnosed. Here, we present a case of heterotopic pregnancy after spontaneous conception, diagnosis of which was confused with ovarian torsion.

Case Report: A 31-year-old second gravid para one with one living issue at 12 week 6 day period of gestation presented to emergency with acute-onset pain in right lower abdomen for 3 days. On examination, an ovarian tumor with ascitis with probable torsion was suspected along with intrauterine pregnancy. These findings were further supported by similar ultrasound picture. Patient was taken up for laparotomy. Intraoperative finding was suggestive of heterotopic pregnancy with ruptured left tubal ectopic pregnancy. Left salpingectomy was done with minimal uterine manipulation. Histopathology showed blood clots, hyalinized villi, and acute inflammatory cell infiltrate suggestive of ectopic pregnancy. Ultrasonography (USG) done in the postoperative period showed viable intrauterine pregnancy. The patient is currently following with us and is in her second trimester of pregnancy.

Conclusion: Heterotopic pregnancy is a diagnostic and therapeutic challenge; a high index of suspicion is required. It is important for clinicians to see adnexal in detail even if intrauterine pregnancy has been seen clearly during USG, especially in high-risk cases.

Clinical significance: Heterotopic pregnancies can occur even in the absence of high-risk factors. In patients with intrauterine pregnancy, who present with adnexal mass with free fluid in abdomen, clinicians should keep their mind open for possible diagnosis of heterotopic pregnancy. Early diagnosis and surgical management will decrease maternal morbidity and increase chances of survival of intrauterine pregnancy.

Keywords: Ectopic pregnancy, Heterotopic pregnancy, Infertility, *In vitro* fertilization.

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INTRODUCTION

Heterotopic pregnancy is the occurrence of intrauterine and ectopic pregnancies together. The ectopic pregnancy can be tubal, ovarian, cervical, corneal, or abdominal. Though it is a rare condition, its incidence is rising. Its incidence varies from one in 8,000 to 30,000.[1] It is more common after conception with *in vitro* fertilization cycles; reported incidence is around 1 in 100.[2] Diagnosis is often delayed resulting in increased maternal morbidity and poor intrauterine pregnancy outcome. Here, we present a rare case of heterotopic pregnancy in spontaneous conception cycle who presented at 12 week 6 day period of gestation (POG), and was mistaken as torsion of ovarian cyst. There were no risk factors for ectopic pregnancy. These patients generally present early in first trimester, but our case presented comparatively later. A review done by Tal et al[3] reported that 70% of the heterotopic pregnancies were diagnosed between 5 and 8 weeks of gestation, 20% between 9 and 10 weeks, and only 10% after the 11th week.

CASE REPORT

A 31-year-old second gravida para one with one living issue at 12 week 6 day POG presented to the emergency department with acute-onset pain in the left lower abdomen for 3 days. It was a spontaneous conception. There was no history of fainting attack. She also gave history of intermittent episodes of spotting per vaginum throughout first trimester, which was managed conservatively with progesterone supplementation. There was no history of infertility, pelvic inflammatory disease (PID), pelvic surgery, or tubal surgery in the past. On examination, she was pale with pulse rate of 100 beats per minute and normal blood pressure. Abdomen was mildly distended and uterus was 16 weeks pregnant size. On per vaginal and per rectal examination, a bogginess was felt in the pouch of Douglas with fullness in left fornix without any tenderness. Her hemoglobin was 7.7 mg/dL. The Ultrasonography (USG) revealed single live intrauterine

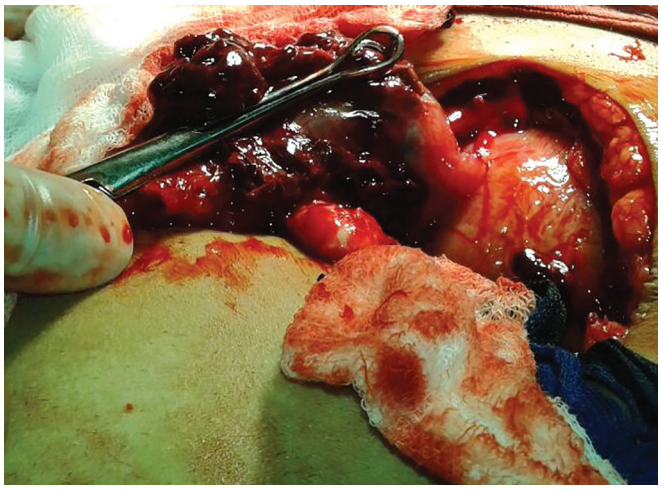

Fig. 1: Intraoperative finding showing ruptured left fallopian tube with enlarged pregnant uterus

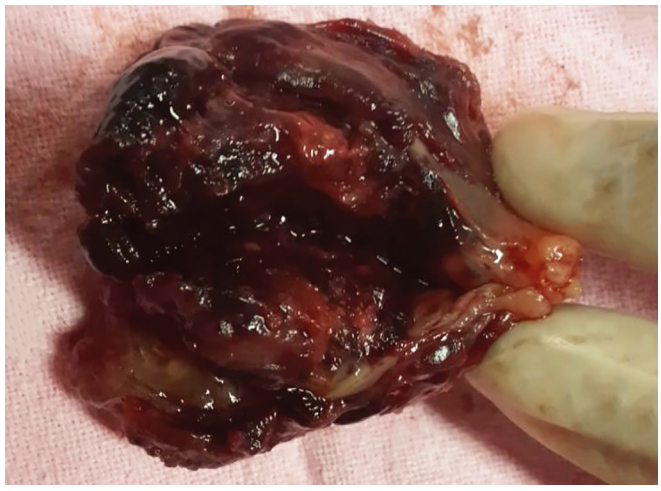

Fig. 2: Cut section of salpingectomy specimen

fetus of 13 week POG along with heteroechoic predominantly solid area of 8.3 × 4.3 cm in pouch of Douglas. Moderate free fluid was also noted in pelvis. Left ovary was not seen separately and right ovary was normal. She carried with her the previous ultrasound report, done at 7 weeks of gestation, which showed a single live intrauterine fetus of 7 weeks 1 day with subchorionic hematoma without any mention of adnexal. Clinically, there was suspicion of ovarian tumor with ascitis with possible torsion; she was taken up for immediate laparotomy. As patient was hemodynamically stable, free fluid in the abdominal cavity was not suspected to be blood. Under general anesthesia, abdomen was opened by midline vertical incision. Intraoperatively, there was around 300 mL of hemoperitoneum along with around 500 mL organized blood clots in the pouch of Douglas adherent to the posterior surface of uterus, left ovary, and omentum. Uterus was of 12 weeks size. Left fallopian tube was ruptured with ongoing bleeding from rupture site (Fig. 1). Contralateral ovary and fallopian tube were normal. Left salpingectomy was done. During entire surgery, care was taken to manipulate uterus minimally. Cut section of salpingectomy specimen showed features similar to product of conception (Fig. 2). She had uneventful recovery in the postoperative period. An USG was done on the 3rd postoperative day, and it showed an intrauterine fetus with normal cardiac activity. The patient was discharged on the 4th postoperative day in stable condition. Histopathology showed blood clots, hyalinized villi, and acute inflammatory cell infiltrates suggestive of ectopic pregnancy. The patient is currently following with us and is in her second trimester of pregnancy.

DISCUSSION

Heterotopic pregnancy carries significant maternal morbidity and mortality due to delay in diagnosis and risk of rupture. Heterotopic pregnancy was reported first in the literature in 1708 by Duverney, in the autopsy of a woman who died of ruptured ectopic pregnancy.[1] The most common location of heterotopic component is fallopian tube, but ovary, cervical, and cesarean scar heterotopic pregnancies have also been reported.[4-6] It is a rare condition, but the incidence has been rising due to tubal surgeries, PID, and, most importantly, due to assisted reproductive techniques (ARTs).[7] In ARTs, transfer of multiple embryos, faulty embryo transfer (ET) which is very high near fundus or cornua, use of excess culture media or force in ET, and Trendelenberg position after ET are all supposed to be responsible factors other than damaged tubes. However, the main factor in ART patients is transfer of multiple embryos. With five or more embryos, the transfer risk rises to 1 in 45 pregnancies.[1]

It is a diagnostic and therapeutic challenge and a high index of suspicion is required. Heterotopic pregnancy should be suspected when there is persistent or rising human chorionic gonadotropin (hCG) after dilation and curettage for abortion, presence of two corpora lutea, and absence of vaginal bleeding with signs and symptoms of ectopic pregnancy.[8] Due to the presence of intact intrauterine pregnancy, bleeding is absent in heterotopic pregnancy, but it was present in our case. It can present as ovarian mass and pain in the lower abdomen as in the present case. However, painful abdomen also occurs in cases of threatened abortion. There are no specific features to make an accurate and early diagnosis other than a high index of suspicion and general awareness of such a possibility. Serial hCG levels are not helpful due to the accompanying intrauterine pregnancy. It is often overlooked in USG due to the presence of intrauterine pregnancy. Further, adnexal sac can be mistaken for hemorrhagic *corpus luteum* or ovarian cyst.[9] Presence of cardiac activity in both intrauterine and ectopic gestation is important for diagnosis, but this is a very rare

finding. In ART cycles, ectopic component of heterotopic pregnancy is often masked by the presence of ovarian hyperstimulation syndrome (OHSS) as some amount of free fluid is there in OHSS.[10] Clinicians, generally, do not try to look for adnexal once intrauterine gestation is seen on USG, leading to delayed diagnosis or misdiagnosis. So, it is important for radiologists to look for adnexal thoroughly, especially in patients who conceive on ART or had a history of PID or pelvic surgery. Preoperative diagnosis is a challenge, and most cases are diagnosed on laparotomy or laparoscopy as in our case. A comparative review done by Barrenetxea et al[11] showed that during the 1971 to 1993 period, the definitive diagnosis of heterotopic pregnancy was performed by laparoscopy or laparotomy in 59% of cases. This proportion increased to 74% from 1994 to 2004. Likewise, the percentage of cases in which an early diagnosis was possible (performed before the 9th week of pregnancy) did not vary in any of the time periods evaluated (71 *vs* 74%). Thus, the probability of early diagnosis has not changed much over the time despite of the advancement in medical knowledge and USG techniques.[11] In one literature, only 1 in 100 heterotopic pregnancies were diagnosed preoperatively.[1] The USG is not very useful in diagnosing heterotopic pregnancy; about half of the cases are being missed. In the presence of two *corpora lutea* on USG, laparoscopy, or laparotomy, one should suspect a heterotopic pregnancy.

Treatment modality can be medical, surgical, or expectant. Surgical removal of ectopic pregnancy is preferred when intrauterine pregnancy is desired. Surgical management can be done by either laparotomy or laparoscopy with earlier recovery with the latter. During surgical treatment, intrauterine instrumentation should be avoided and uterine manipulation should be minimal when intrauterine pregnancy is desired. Rate of abortion is high for intrauterine pregnancy, around 1 in 3.[12] Medical modality is ultrasound- or laparoscopy-guided injection of KCl in the ectopic gestation.[6,13] However, this method is not very effective; risk of continued growth and rupture still exist, so close follow-up is required. In a review of literature, approximately 55% patients required further surgical treatment.[14] Methotrexate has teratogenic effect, so it can be used only when intrauterine pregnancy is not desired.

CONCLUSION

Clinicians should try to look for adnexal during USG of pregnant patients even after confirmation of intrauterine pregnancy. Though rare, it can happen in spontaneous conception as in the present case. In patients with intrauterine pregnancy who present with adnexal mass with free fluid in abdomen, clinicians should keep their mind open for possible diagnosis of heterotopic pregnancy. Early diagnosis will lead to better management of both mother and intrauterine fetus.

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