

Case Report

# Catastrophic presentation of spontaneous heterotopic pregnancy

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## ABSTRACT

Heterotopic gestation is an uncommon entity with natural conception; however, rising trends have been seen with the use of artificial reproductive techniques. We are reporting a case of 22-year-old G<sub>3</sub>P<sub>1</sub>A<sub>1</sub>L<sub>1</sub> who presented to us at 9 weeks of gestation with complaints of mild pain in the left adnexa from 3 days. Subsequently, her pain was increased in intensity and spread over to whole abdomen. She was referred with the left adnexal mass with persistent pain abdomen. On admission, ultrasound revealed single live intrauterine fetus with the left adnexal mass with fetal node along with hemoperitoneum. She underwent laparotomy and successfully managed with the left side salpingectomy along with blood transfusion. Later, she delivered a healthy baby at term. Ectopic pregnancy should be the first differential diagnosis when no other cause suspected in patients with adnexal mass. Follow-up ultrasound should be done in patients with persistent and localized pain in abdomen even in spontaneous conception. Diagnostic laparoscopy may be the reasonable option to make a final diagnosis as beta-human chorionic gonadotropin is not a reliable test to diagnose ectopic pregnancy in heterotopic pregnancy.

**Keywords:** Heterotopic pregnancy, Assisted reproductive techniques, Ovulation induction, Hemoperitoneum

## INTRODUCTION

Heterotopic pregnancy has been defined as simultaneous intrauterine and extra-uterine implantation of pregnancy. It is seldom reported in spontaneous conception but its incidence has been reported as high as 1:100–1:500 in cohort of patients undergoing treatment with assisted reproductive techniques. Due to high suspicion in these cases, it is diagnosed earlier, hence feasible to manage by medical method. However, it is never in doubt in spontaneous conception, thus diagnosis has been made after rupture only. This case report highlighted the risk factors in spontaneous conception where heterotopic pregnancy should be suspected earlier before rupture so that it can be managed with medical therapy or laparoscopy as risk of failure of medical management and catastrophic hemorrhage will be more with advanced gestation. Hence, patients' complaints, physical examination, and imaging studies all should be carefully considered in patients with intrauterine pregnancy to exclude possibility of heterotopic pregnancy.

## CASE REPORT

A 22-year-old G<sub>3</sub>P<sub>1</sub>A<sub>1</sub>L<sub>1</sub> at 9 weeks gestation came to our Gynaec emergency with the complaint of dull aching, continuous pain in the left side of lower abdomen from the

past 3 days which was increased in intensity and spread to whole abdomen from the past 6 h. She also gave a history of bleeding from vagina 2 weeks back that lasted for 8 days. Obstetric history: She had one normal vaginal delivery 6 years ago and one spontaneous abortion at 24 weeks gestation 4 years back, following which she has been using contraception. This was her spontaneous conception. Her menstrual cycles were regular with average blood flow. Her past and family histories were not significant.

Ultrasound done on same day from outside revealed single live fetus along with the left adnexal, hypoechoic mass of 52 × 41 mm in size with well-defined margins and minimal free fluid seen in cul-de-sac [Figures 1 and 2]. Bilateral ovaries were normal. She was referred in view of adnexal mass with pain abdomen.

On general physical examination: She had severe pallor with cold extremities. Her pulse rate was 120/min and blood pressure was 90/60 mmHg. On abdominal examination, tenderness and guarding were present. There was no bleeding on local examination. Vaginal examination was deferred due to severe pain and tenderness.

Transvaginal ultrasonography done after admission revealed single intrauterine live fetus with crown rump length of 8

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weeks, a well-defined heterogeneous mass seen in the left adnexa with fetal node in it. On transabdominal sonography, echogenic free fluid was there in paracolic gutters, under both the diaphragm and pelvis. Hence, diagnosis of heterotopic pregnancy was made.

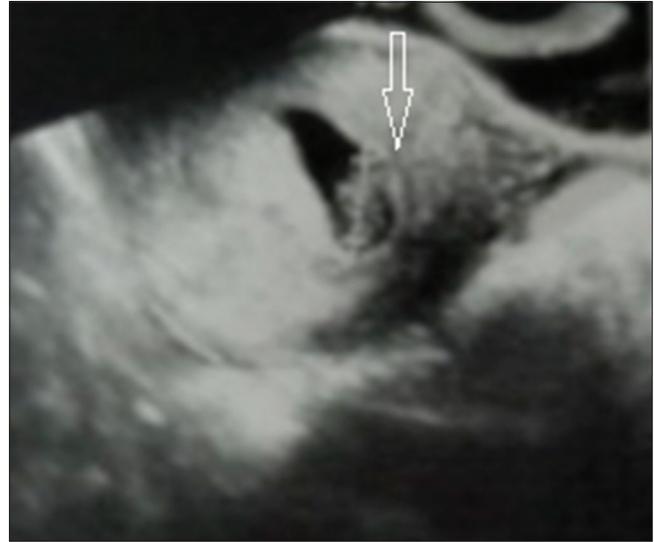
Investigations on admission revealed hemoglobin of 5.8 g%, platelet count was 2.6 lacs, and PTI was 82%. She underwent resuscitation with intravenous fluid and blood, simultaneously taken up for emergency laparotomy after informed consent. She was given hydroxyprogesterone caproate 250 mg intramuscular injection in perioperative period. There was unruptured distended, left-sided tubal pregnancy, about 4 × 3 cm in size at isthmic-ampullary junction within the process of abortion with active bleeding from the fimbrial end along with hemoperitoneum [Figure 3]. There were approximately 1 L hemoperitoneum and 200 g clots which were drained. Peritoneal lavage and left-sided salpingectomy were done. She received 3 units of blood transfusions. Hemoglobin on discharge was 6.9 g/dl, platelets counts were 1.77 lac, and blood urea and creatinine were 21 mmol/L/0.7 μmol/L. She was discharged on day 5 of surgery with advice of rest and antenatal follow-up. Subsequently, her antenatal period was uncomplicated. She gave birth to a healthy baby at 39 weeks.

## DISCUSSION

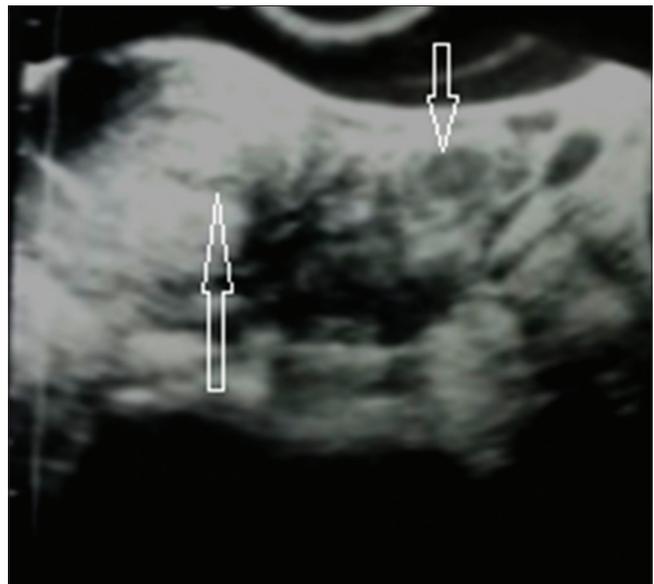
Spontaneous heterotopic pregnancy is an extremely rare condition occurring in only 1 in 30,000 pregnancies.<sup>[1]</sup> However, with the advent of ovulation induction and *in vitro* fertilization conception, overall incidence of heterotopic pregnancy has been raised to approximately 1 in 3900 pregnancies.<sup>[2]</sup>

Tubal ectopic is the most common site; although various other sites of extra-uterine pregnancy have been reported including interstitial, cervical, ovarian, scar site, and abdominal pregnancy.<sup>[2-4]</sup> Heterotopic pregnancy with multiple gestations such as twins, triplet, and even quadruplet in number has been reported after spontaneous conception<sup>[5]</sup> but predominantly occurs after the use of artificial reproductive techniques.<sup>[6]</sup> It has been diagnosed in each trimester of pregnancy and remain misdiagnosed as twin pregnancy till term in abdominal heterotopic pregnancy.<sup>[7,8]</sup>

Regardless of the site of implantation, endometrium always shows some changes due to effects of pregnancy-related hormones, so bleeding from vagina will be the more frequent complaints in all ectopic pregnancies followed by pain in adnexa due to stretching, separation, and rupture of ectopic site. However, pain will be the predominant symptoms although vaginal bleeding has also been reported even in heterotopic pregnancy like in the present case.<sup>[9]</sup> Site of pain, its nature, and associated clinical findings can help in differentiate it from threatened abortion, incomplete



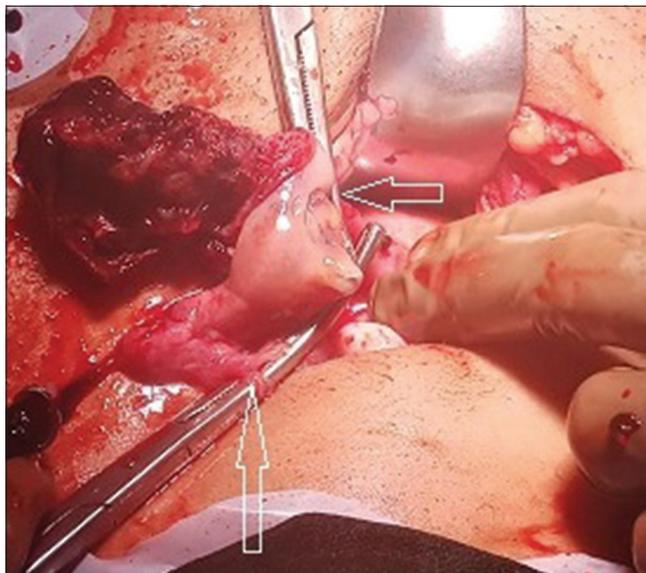
**Figure 1:** Ultrasound revealed intrauterine pregnancy (arrow head).



**Figure 2:** Transvaginal sonography revealed intrauterine pregnancy (long arrow) and left adnexal mass (small arrow).

abortion, torsion of ovarian cyst, and acute surgical conditions.

In general, it should be suspected in patients conceived through assisted reproductive techniques. Complaints of pain in lower abdomen and bleeding were overlooked in the present case due to spontaneous conception and confirmed intrauterine pregnancy, therefore, early diagnosis of heterotopic pregnancy could have avoided life-threatening hemorrhage in the present case. In addition, uterine rupture with or without hypovolemic shock may be the first



**Figure 3:** Intraoperative findings: left unruptured tubal ectopic pregnancy (small arrow) within the process of tubal abortion (long arrow).

presenting feature in scar pregnancy.<sup>[4]</sup> Trends of serum beta-human chorionic gonadotropin (B-HCG) level may not be helpful due to the presence of intrauterine pregnancy in these cases. However, rise in B-HCG may be useful when it rises after salpingectomy in tubal ectopic pregnancy.

Ultrasound is a first imaging modality which can diagnose > 90% all types ectopic pregnancy before rupture. Tubal ectopic pregnancy usually reveals an inhomogeneous adnexal mass with yolk sac and fetal nodes in addition to intrauterine pregnancy as revealed in the present case. Corpus luteal cyst may be misdiagnosed as tubal pregnancy or conversely tubal pregnancy may be under diagnosed as corpus luteal cyst due to similarity in appearance and presence of ring of fire in both of these entities.<sup>[10-12]</sup> Free fluid may be a sign of ovarian hyperstimulation syndrome, ruptured ectopic pregnancy, and corpus luteal hemorrhagic cyst. It should also be suspected in acute surgical conditions like ruptured appendicular mass when aspiration of fluid may be considered. However, management of patients should not be delayed for diagnostic test and imaging studies in acute conditions.

Scar pregnancy should be suspected in a low lying gestational sac at the level of the internal os where scar site's area appears to bulge beyond the anterior contour of uterus.<sup>[11]</sup> Along with the presence of trophoblastic tissue between anterior uterine wall and the bladder, increased peritrophoblastic vasculature and invasion into bladder can be seen in the absence of myometrium between gestational sac and bladder. Whereas, cervical pregnancy implants below the level of internal os and within the dilated cervical canal which can be easily

diagnosed with the help of ultrasonography in addition to per speculum examination of cervix.<sup>[11]</sup>

Interstitial pregnancy is defined when gestational sac implants in intramural portion of fallopian tube.<sup>[11]</sup> Ultrasonographically, it is diagnosed as eccentrically placed gestational sac >1 cm away from endometrial cavity with an interstitial line sign and myometrium thickness of < 5 mm. Angular pregnancy is also eccentrically placed but implants within the endometrial cavity and well enveloped by more than 5 mm thickness of myometrium.<sup>[11]</sup> It should be followed up regularly to ensure development of fetus toward the cavity rather than eccentrically. Ovarian pregnancy is usually diagnosed during laparotomy; however, Spiegelberg criteria should be fulfilled on ultrasonography including cystic structure with a wide echogenic ring covered by ovarian tissue which must be confirmed by histopathology.<sup>[11]</sup> MRI may be found useful if ultrasound is not able to find out location of pregnancy like in abdominal heterotopic pregnancy.

Management of heterotopic pregnancy depends on the site of pregnancy and clinical condition of the patient. As most of the heterotopic pregnancies following spontaneous conception are usually diagnosed after rupture which requires urgent laparotomy to stop massive intraabdominal hemorrhage. Otherwise, tubal and cornual pregnancy can be resected by laparoscopy in stable patient. Laparoscopic management could have been possible in the present case if diagnosis could be suspected before tubal abortion and massive hemoperitoneum.

Wedge resection of scar site pregnancy can also be tried through laparotomy or laparoscopy.<sup>[4]</sup> Heterotopic cervical pregnancy has been managed with extraction and ultrasound-guided curettage. However, failed medical management may require hysterectomy or uterine artery embolization.

Live intrauterine pregnancy is an absolute contraindication for systemic methotrexate therapy. Potassium chloride, methotrexate injections, and hyperosmolar glucose injections can be used for selective reduction of live ectopic pregnancy, especially in scar site, cervical, cornual, and interstitial ectopic to retain the intrauterine pregnancy.<sup>[9]</sup> Besides selective reduction, successful selective embryo aspiration has been done under ultrasound guidance in these cases.<sup>[9]</sup>

Long-term follow-up is required as medical treatment takes long time for product of conception to resolve besides risk of rupture and hemorrhage. Whereas, surgical treatment is completely devoid of these complications. However, risk of abortion and maternal mortality and morbidity in heterotopic pregnancy are higher than that of singleton pregnancy.<sup>[13]</sup> Risk of complications will be more in ongoing intrauterine pregnancy, especially in scar site heterotopic

pregnancy such as vaginal bleeding, hemorrhage, and risk of peripartum hysterectomy.<sup>[4]</sup>

## CONCLUSION

Pregnancy should be regularly followed in patients with pain abdomen and bleeding from vagina even in spontaneous conception to exclude heterotopic pregnancy. Diagnostic laparoscopy should be considered in patient with persistent localized pain with adnexal mass to avoid laparotomy and massive hemorrhage.

## Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Nil.

## Conflicts of interest

There are no conflicts of interest.

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