

Heterotopic Pregnancy Case Reports

¹Dr. Emrana Rahman,

MS (Obstetrics and Gynaecology), MRCOG (1), Senior Consultant, Advanced maternity fertility centre Janm Ivf, Bhagalpur, Bihar, India

Corresponding Author: Dr. Emrana Rahman

ABSTRACT

Background: Heterotopic pregnancy (HP) is a rare condition where at least two pregnancies are present simultaneously at different implantation sites and one of them located in the uterine cavity. Its prevalence varies from 1 to 30 000 in a natural cycle to around 1 in 100 in an assisted one.

Case presentations: In the current study, we presented four cases of first trimester heterotopic pregnancy diagnosed by ultrasound (US).

Conclusion: However, heterotopic is a rare condition, any pregnant woman presenting with alarming abdominal pain and adnexal abnormality; heterotopic pregnancy should be among the differential diagnosis possibilities. The patient should be thoroughly investigated using 3D ultrasound if needed, to exclude this rare diagnosis and allow on-time proper management.

Keywords: Acute abdomen, Ectopic pregnancy, Heterotopic pregnancy, Second-trimester diagnosis, Case report.

INTRODUCTION

Heterotopic pregnancy (HP) is a rare condition where at least two pregnancies are present simultaneously at different implantation sites and one of them located in the uterine cavity. Its prevalence varies from 1 to 30 000 in a natural cycle to around 1 in 100 in an assisted one.¹ Like in ectopic pregnancy, abdominal pain, vaginal bleeding with positive pregnancy test are the common syndromes, but the diagnosis is more complicated. Even in the era of high-resolution ultrasound imaging and Doppler techniques, most of the time, the diagnosis is based on the presence of acute abdominal symptoms.

In the first trimester, unrecognized heterotopic pregnancy can be a cause of nontraumatic acute abdomen. The increased incidence of multiple pregnancies with ovulation induction and IVF increases the risk of both ectopic and heterotopic gestation. The hydrostatic forces generated during embryo transfer may also contribute to the increased risk.²

There may be an increased risk in patients with previous tubal surgeries.³ Heterotopic pregnancy can have various presentations. It should be considered more likely (a) after assisted reproduction techniques, (b) with persistent or rising chorionic gonadotropin levels after dilatation and curettage for an induced/spontaneous abortion, (c) when the uterine fundus is larger than for menstrual dates, (d) when more than one corpus luteum is present in a natural conception, and (e) when vaginal bleeding is absent in the presence of signs and symptoms of ectopic gestation.⁴

A heterotopic gestation can also present as hematometra and lower quadrant pain in early pregnancy.⁵ Most commonly, the location of ectopic gestation in a heterotopic pregnancy is

the fallopian tube. However, cervical and ovarian heterotopic pregnancies have also been reported.^{6,7} Majority of the reported heterotopic pregnancies are of singleton intrauterine pregnancies. Triplet and quadruplet heterotopic pregnancies have also been reported, though extremely rare.^{8,9} It can be multiple as well.⁴ They can be seen frequently with assisted conceptions. Intrauterine gestation with hemorrhagic corpus luteum can simulate heterotopic/ectopic gestation both clinically and on ultrasound.¹⁰ A heterotopic pregnancy, though extremely rare, can still result from a natural conception; it requires a high index of suspicion for early and timely diagnosis; a timely intervention can result in a successful outcome of the intrauterine fetus.¹¹

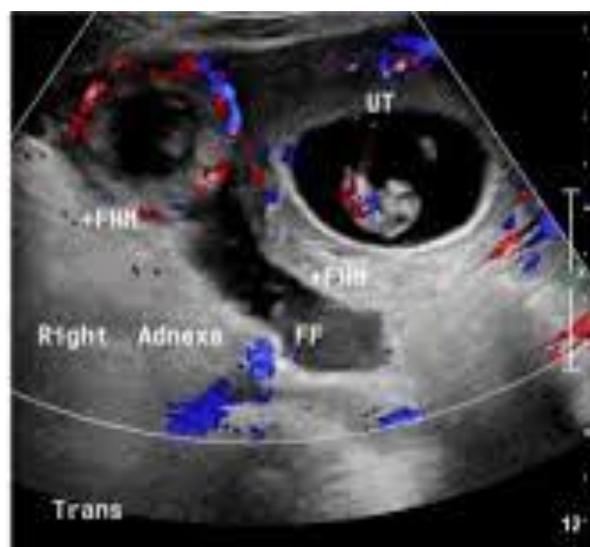
In the current study, we presented four cases of first trimester heterotopic pregnancy diagnosed by ultrasound (US).

CASE 1

A 28-year-old woman reported to me with primary infertility of 5 years who was suggested IVFET. She had bilateral cornual block on HSG, was hypothyroid previous folliculogram suggested ovulatory dysfunction. Husband's seminogram was normal.

She was undertaken for Hystero Laparoscopy, Hysteroscopically successful bilateral cornual cannulation and Laparoscopic adhesiolysis was performed. Her tubes appeared to be diseased Hyperaemic congested beaded with tubercies on it. Fimbrial dilatation + combing & milking of tubes was done. There was free spill of dye on right side and dye dripping on left. Tubo ovarian relationship was normal. She was Mantoux positive PCR Endometrium negative.

Antitubercular treatment was initiated on Laparoscopic findings, COHS was given. Patient was in follow up on monthly basis, subsequently in the third post operative month patient landed with 6 weeks amenorrhoea, intermittent spotting and severe acute pain abdomen intermittently her pregnancy test was positive. USG revealed and an alive unruptured Right+ Right ectopic with fetal pole, yolk sac and cardiac activity and one viable intra uterine pregnancy of six weeks with subchorionic bleed. Considering it to be precious pregnancy she was immediately undertaken for emergency Laparoscopy, using the same old ports Linear Salpingostomy on the cystic formation of 2cm on Right tube was done, ectopic tissue was completely retrieved her post-operative recovery was uneventful. She was placed on parenteral progestins aspirin to rescue uterine pregnancy followed on serial USG scan every two weeks, However, patient had spontaneous abortion at 12 weeks with passage of POC.



CASE 2

A 26 years old woman with primary infertility of three years normal HSG with bilateral patent tubes was placed on ovulation induction with clomiphene 50 and FSH injection she was followed with follicular study, 2 months later she presented with acute pain abdomen intermittent spotting and seven weeks amenorrhoea her urine pregnancy test was very weakly positive. TVS revealed complex left TO Mass with small amount of free fluid in peritoneal cavity Doppler study showed low resistance flow.

Uterus was bulky with thick endometrium 1.2 cm, Provisional diagnosis of ruptured ectopic was made in view of clinical history.

Patient was undertaken for emergency Laparoscopy mild amount of Hemoperitoneum around 300 cc of was aspirated ectopic was removed Hemostasis achieved the tube was conserved tissue was sent for histopathology which later confirmed ectopic by presence of chorionic villi. 12 days later patient reported with profuse vomiting nausea giddiness pain abdomen. Ultrasound showed an alive intrauterine pregnancy of five weeks six days with fetal pole YS cardiac activity, her ad nexae were clean with no signs of persistent ectopic as suggested by Doppler flow.

Her intrauterine live gestation was allowed to continue and patient delivered a healthy alive baby 3.5 Kg at term vaginally. She was counselled against termination of pregnancy.

CASE 3

A 32 years old woman gravida 3 para 2 with previous 2 LSCS was under my antenatal care from 12 weeks onwards. Her current pregnancy occurred spontaneously 1st scan showed an alive active fetus of 12 weeks normal NT and left sided complex adnexal mass suggested as corpus luteum cyst of 4.2 x 3.9 cm.

Later after 3 months suddenly patient presented to me with severe pain abdomen clinical features of shock hypotension BP 70/40, thready pulse tachy cardia - 130/mn, Cold clammy skin Blood Biochemistry revealed Hb% 5gm. TLC - 13000 / Platelets 1,20,000, creatinine normal, USG showed intrauterine death of fetus at 24 weeks and huge complex left adnexal mass with heterogenous echoes suggesting ovarian mass, torted ovarian cyst. After resuscitation & some hemodynamic stability patient was undertaken for emergency Laparotomy.

Hysterotomy was performed for extraction of dead fetus. There were severe adhesions/pseudopocketing of omentum walling off an old ruptured ectopic with Moderate haemoperitoneum, dead devitalised tissue debris old clots were removed adhesiolysis done intraperitoneal drain given peritoneal lavage done, left salpingoophorectomy done, patient recovered uneventfully and discharged after 6 days. It was a case ovarian ectopic as lower pole of left ovary had a circular bleeding formation of 5cm rest of the left tube and ovary were near normal except adhesions.

Heterotopic Pregnancy



CASE 4

A patient X came to me with early fetal demise 8 weeks under ANC care by other doctor with proper USG reports from 2 places confirmed on ultrasound by me also

I performed D & E on her. Two days later she again approached me with severe acute pain abdomen.

I undertook the patient for immediate USG, to my shock. I could find a tubal ring in her left tube with mild haemoperitoneum and empty uterus with thin echogenic lining. Her urine pregnancy test was strongly positive.

Presuming it to be ectopic, I took her for laparoscopy - for retrieval of unruptured ectopic. On laparoscopy bulging ectopic sac was found in ampullary region, linear salpingostomy was done ectopic removed however tube had to be sacrificed owing to persistent bleeding.

The diagnosis was missed in this case as I totally focussed on intrauterine early fetal demise.

DISCUSSION

Heterotopic pregnancy describes the occurrence of two pregnancies in different implantation sites simultaneously, mostly manifested as intrauterine and ectopic pregnancies (ampullary in 80%). It is a challenge to diagnose such a problem due to complex clinical and laboratory findings.¹²⁻¹⁵ Most HPs were diagnosed during surgery, either laparoscopy or laparotomy (59–74%), the rest by ultrasonography. Barrenetxea's report of HP term delivery occurred in 62.5%, preterm in 6%, and 31% of gestations ended with a miscarriage of intrauterine pregnancy.¹⁶

The diagnosis of a heterotopic pregnancy poses unique therapeutic challenges. Clinically, it manifests non-specifically with abdominal pain, vaginal bleeding, and spotting which presents similarly to both normal pregnancies and abnormal obstetrical complications. While ectopic pregnancy can be early diagnosed during the screening of the serum b-HCG and endovaginal

US in routine prenatal assessment, heterotopic pregnancy diagnosis can be a challenge as it may be delayed in the concurrence of intrauterine pregnancies. This can be due to the differential diagnosis between ectopic pregnancy and other situations that may be associated with normal pregnancy as hemorrhagic corpus luteum or adnexal torsion.¹⁷⁻²⁰

In normal pregnancies, serum B-HCG above 1500–2000 mIU/mL should be associated with ultrasound visualized intrauterine (IU) pregnancy; however, this IU pregnancy will not necessarily exclude the chance of having a heterotopic pregnancy especially in cases receiving conception-assisted techniques. Ultrasound picture of heterotopic pregnancy may be adnexal complex cysts or mass which can be explained by being hematosalpinx, tubal ring, or embryo. Free intra-peritoneal fluid can be seen also.^{14,18-20}

In equivocal cases, MRI can be helpful; it can show an adnexal lesion that may be cystic or looks like a gestational sac. Tubal cystic dilatation with a thickened wall can also be seen, while tubal rupture will cause hematoma.²⁰

Heterotopic pregnancy treatment needs laparoscopy and, most often, a salpingectomy or salpingostomy. However, in hemodynamically unstable cases, laparotomy may be needed. Systemic methotrexate has no role in the management of heterotopic pregnancy due to the presence of a viable intrauterine pregnancy. Some literature described the use of local injection of potassium chloride and methotrexate, but the success rate is controversial.^{21,22}

Although heterotopic pregnancy in natural conception is a rare event than one as a consequence of assisted reproduction techniques, but the outcomes are same if heterotopic pregnancy occurs. Practitioners should carry a high index of suspicion for heterotopic pregnancy in patients with intrauterine pregnancy presenting with acute abdominal pain, abdominal tenderness, and or free fluid in the abdominal cavity.

CONCLUSION

However, heterotopic is a rare condition, any pregnant woman presenting with alarming abdominal pain and adnexal abnormality; heterotopic pregnancy should be among the differential diagnosis possibilities. The patient should be thoroughly investigated using 3D ultrasound if needed, to exclude this rare diagnosis and allow on-time proper management.

REFERENCES

1. Wallach EE, Tal J, Haddad S, Gordon N, Timor-Tritsch I. Heterotopic pregnancy after ovulation induction and assisted reproductive technologies: a literature review from 1971 to 1993. *Fertility and sterility*. 1996 Jul 1;66(1):1-2.
2. Lyons EA, Levi CS, Sidney M. Dashefsky in diagnostic ultrasound. In: Rumak CM, Wilson SR, Charboneau WK, editors. 2nd ed. Volume 2, Mosby: 1998. p. 999.
3. Gruber I, Lahodny J, Illmensee K, Löscher A. Heterotopic pregnancy: report of three cases. *Wiener klinische Wochenschrift*. 2002 Mar 1;114(5-6):229-32.
4. Multifetal Ectopic Pregnancy, in Chapter 34. Ectopic Pregnancy, text book of - Williams Obstetrics, 21st ed. p. 888-9.
5. Cheng PJ, Chueh HY, Qiu JT. Heterotopic pregnancy in a natural conception cycle presenting as hematometra. *Obstetrics & Gynecology*. 2004 Nov 1;104(5):1195-8.
6. Hirose M, Nomura T, Wakuda K, Ishiguro T, Yoshida Y. Combined intrauterine and ovarian pregnancy: a case report. *Asia-Oceania Journal of Obstetrics and Gynaecology*. 1994 Mar;20(1):25-9.

7. Peleg D, Bar-Hava I, Neuman-Levin M, Ashkenazi J, Ben-Rafael Z. Early diagnosis and successful nonsurgical treatment of viable combined intrauterine and cervical pregnancy. *Fertility and sterility*. 1994 Aug 1;62(2):405-8.
8. Alsunaidi MI. An unexpected spontaneous triplet heterotopic pregnancy. *Saudi medical journal*. 2005 Jan 1;26(1):136-8.
9. Sherer DM, Scibetta JJ, Sanko SR. Heterotopic quadruplet gestation with laparoscopic resection of ruptured interstitial pregnancy and subsequent successful outcome of triplets. *American journal of obstetrics and gynecology*. 1995 Jan 1;172(1):216-7.
10. Sohail S. Hemorrhagic corpus luteum mimicking heterotopic pregnancy.
11. Espinosaa Picazo M, Alcántara Mendoza MA. Embarazo heterotópico: informe de un caso y revisión bibliográfica. *Ginecol. obstet. Méx.* 1997;482-6.
12. Fernandez H, Gervaise A. Ectopic pregnancies after infertility treatment: modern diagnosis and therapeutic strategy. *Human reproduction update*. 2004 Nov;10(6):503-13.
13. Xiao HM, Gong F, Mao ZH, Zhang H, Lu GX. Analysis of 92 ectopic pregnancy patients after in vitro fertilization and embryo transfer. *Zhong nan da xue xue bao. Yi xue ban= Journal of Central South University. Medical Sciences*. 2006 Aug 1;31(4):584-7.
14. Louis-Sylvestre C, Morice P, Chapron C, Dubuisson JB. The role of laparoscopy in the diagnosis and management of heterotopic pregnancies. *Human reproduction (Oxford, England)*. 1997 May 1;12(5):1100-2.
15. Jeon HS, Shin HJ, Kim IH, Chung DY. A case of spontaneous heterotopic pregnancy presenting with heart activity of both embryos. *Korean Journal of Obstetrics & Gynecology*. 2012 May 16;55(5):339-42.
16. Barrenetxea G, Barinaga-Rementeria L, de Larruzea AL, Agirregoikoa JA, Mandiola M, Carbonero K. Heterotopic pregnancy: two cases and a comparative review. *Fertility and sterility*. 2007 Feb 1;87(2):417-e9.
17. Callen PW Ultrasonography in obstetrics and gynecology. In: Levine D (ed) *Ectopic Pregnancy*, 5th edn. Saunders Elsevier, Philadelphia, pp 1020–1047
18. Dundar O, Tutuncu L, Mungen E, Muhcu M, Yergok YZ. Heterotopic pregnancy: Tubal ectopic pregnancy and monochorionic monoamniotic twin pregnancy: A case report. *Perinatal Journal*. 2006;14(2):96-100.
19. Tamai K, Koyama T, Togashi K. MR features of ectopic pregnancy. *European radiology*. 2007 Dec;17(12):3236-46.
20. Li XH, Ouyang Y, Lu GX. Value of transvaginal sonography in diagnosing heterotopic pregnancy after in-vitro fertilization with embryo transfer. *Ultrasound in Obstetrics & Gynecology*. 2013 May;41(5):563-9.
21. Li JB, Kong LZ, Yang JB, Niu G, Fan L, Huang JZ, Chen SQ. Management of heterotopic pregnancy: experience from 1 tertiary medical center. *Medicine*. 2016 Feb;95(5).
22. Baxi A, Kaushal M, Karmalkar HK, Sahu P, Kadhi P, Daval B. Successful expectant management of tubal heterotopic pregnancy. *Journal of Human Reproductive Sciences*. 2010 May 1;3(2):108.

Received:12-08-2022 Revised:23-08-2022. Accepted:11-09-2022