

Spontaneous Heterotopic Pregnancy with Intrauterine Twin Pregnancy with Tubal Rupture : A Rare Case

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Abstract

Heterotopic pregnancy (HP) is defined as the coexistence of an intrauterine pregnancy and an ectopic pregnancy i.e. the presence of simultaneous gestations at two or more implantation sites [1]. Incidence of heterotopic pregnancy is around in infertility treated patients 33/10,000, but it is very rare among women who conceive spontaneously amounting to 1:30,000 pregnancies [2]. Both diagnosis and management of the disease is difficult and can be sometimes even life threatening. We are reporting a rare case of multiparous woman with a spontaneous conception resulting in a triplet heterotopic pregnancy : a twin intrauterine pregnancy and a single left tubal ectopic pregnancy, that presented with tubal rupture which was successfully surgically treated with preservation of the intrauterine pregnancy.

Keywords: Heterotopic pregnancy; Triplet; Spontaneous conception.

Introduction

Heterotopic pregnancy (HP) is characterized by the presence of coexistent gestations at two or more implantation sites. It is an extremely rare medical condition with an incidence of around 1:30,000 in spontaneous conceptions [2]. It was first time incidentally reported in the year 1708 as an autopsy finding. However in the past couple of decades there

has been a significant increase of ectopic pregnancy as well as heterotopic pregnancy. This increase in incidence is because of several factors including higher incidence of pelvic inflammatory disease (PID) and extensive use of assisted reproductive technologies (ARTs), tubo-ovarian abscess (TOA), previous ectopic pregnancies, or previous tubal surgeries [2]. The incidence is higher in pregnancies resulting from assisted reproductive techniques, ranging from 1/100 to 1/500 and approximately as high as 1% in certain series. Heterotopic triplets are even more rare and very few cases with tubal ectopic and coexisting twin intrauterine pregnancy have been reported in literature. This medical condition can be dangerous to the intrauterine pregnancy and can lead to maternal death as well. An obstetrician as well as radiologist should keep in mind the occurrence of an HP while dealing with pregnant females. The ectopic gestation would eventually rupture over a period of time leaving the patient in an emergency situation. Therefore, a quick assessment and careful handling of the normal gestation can lead the patient to term with good results.

Ultrasound is the main diagnostic modality for heterotopic pregnancy. But it has a low sensitivity because the diagnosis can be missed or overlooked, leading to serious consequences. Surgical intervention is the management of

choice for heterotopic pregnancy. The aim is to remove the ectopic pregnancy without disturbing the intrauterine pregnancy. Laparoscopic salpingectomy is the standard treatment approach for these kinds of pregnancy. Other management options are local injection of potassium chloride, methotrexate or hyperosmolar glucose into the sac under ultrasound guidance followed the ectopic pregnancy aspiration.

Case

A 25-year-old (Gravida 2 para 1) presented to Maharani Laxmibai Medical College (Jhansi, India) OPD with chief complaint of vaginal bleeding and minor abdominal cramping. Her vital parameters were stable. She had no significant past medical or surgical history. She denied taking medications, including oral contraceptive pills, or supplements. Her past obstetric history includes one spontaneous vaginal birth 8 months back which died 12 hours after birth. Her past gynecologic history was unremarkable. There was no history of any

sexually transmitted diseases, intrauterine devices, or any tubal surgery. A physical examination showed stable vital signs, the patient was in no apparent discomfort, mild tenderness to palpation was present in the left lower quadrant and no blood was seen in the per vaginal examination. Transabdominal ultrasonography showed two live intrauterine gestational sac with left adnexal lesion likely ruptured ectopic pregnancy. An exploratory laparotomy was performed revealing an approximately 5 cm × 3 cm left tubal abortion. A left salpingectomy and evacuation of 500 cc of hemoperitoneum was performed. The patient underwent the procedure without complications. Histopathology confirmed the diagnosis of an ectopic pregnancy. The fetal heart rate of both the intrauterine pregnancy was present before and after surgery (confirmed by ultrasonography). The post operative period was uneventful and the patient was discharged.



Fig. 1:



Fig. 2:



Fig. 3:

Table 1:

Uterus	Measures (77x46x43 mm). Two gestation a sacs of size 18 mm and 21mm seen inside uterine cavity. Sacs out line regular with normal decidual reaction. Yolk sac and embryo seen in one sac. CRL of embryo is 3.2 mm. Cardiac activity present in one sac. Gestational age on the basis of CRL is 6 weeks 0 day. Embryonic poles not seen in second sac though yolk sac present in it. Gestational age on the basis of GS is 5 weeks 05 days. Subchorionic hematoma of 14x11 mm present.
Ovary	A oblong isoechoic of size 58x17 mm seen in left adenexa. Internal echoes seen in it, not separable from left ovary. Thick walled cystic lesion of size 17x16 mm seen in left ovary. Right ovary is normal in size and echotexture. Fluid with echoes seen in pelvic cavity.
Interpretation	F/S/O Intrauterine twin pregnancy with left adnexal mass with Mild pelvic fluid. SOL left adenexa... D/d include: inflammatory/?ectopic (heterotopic)



Fig. 4:

Discussion

Heterotopic pregnancy is the presence of multiple pregnancies with one or more intrauterine pregnancies co-existing with an ectopic pregnancy. The ectopic pregnancy can be tubal, ovarian, cervical, cornual or abdominal. Tubal pregnancies are the most common. The incidence of a heterotopic pregnancy is very rare in spontaneous conception cycles around 0.08%, but incidence increases to as high as 1% with assisted reproductive techniques. The causative factors that attribute to occurrence of ectopic pregnancy are tubal surgeries and pelvic inflammatory diseases [3].

Usually early diagnosis of heterotopic pregnancy is difficult because of the absence of clinical symptoms. Reece *et al.* [4] defined abdominal pain, adnexal mass, peritoneal irritation and an enlarged uterus as signs and symptoms suspicious of HP. Transvaginal ultrasound and assessment of the whole pelvis, even in the presence of intrauterine pregnancy, can be an important aid in the diagnosis of HP. Further, visualization of heart activity in both intrauterine and extrauterine gestation confirms the diagnosis of heterotopic pregnancy.

Undiagnosed heterotopic pregnancy can lead to serious complications, to include loss of the intrauterine pregnancy, hemoperitoneum, tubal rupture, acute abdomen syndrome, oophorectomy, salpingectomy, hysterectomy, and death. Therefore, HP should always be included on a differential in a woman of reproductive age, regardless of risk factors.

The most important diagnostic modality is transvaginal sonography (TVS), with a recent study by Li *et al.* [5] with sensitivity of 92.4% and 100% specificity for the detection of heterotopic

pregnancy. Some studies state detection rates between 41% and 84%. Serial TVS might increase the detection rate. Ultrasonographic findings that might increase detection of heterotopic pregnancy include an extrauterine gestational sac with fetal cardiac activity, fetal node, hyperechogenic ring surrounding the gestational sac, and an adnexal mass. Obstetricians should routinely consider early TVS in women with known risk factors for heterotopic pregnancy to confirm the pregnancy location.

In the present case, the intrauterine pregnancy and ectopic pregnancy were discovered simultaneously via ultrasound. Left salpingectomy was done to remove the extrauterine nonviable product of conception which allowed the viable intrauterine pregnancy to develop to term, ultimately leading to a spontaneous vaginal delivery. Thus, all surgeons operating for ruptured ectopic must bear possibility of heterotopic pregnancy in mind and must handle uterus with care.

Treatment should be as minimally invasive as possible to conserve the developing intrauterine pregnancy. With early diagnosis and treatment, majority of the intrauterine pregnancies will reach viability. All operated patients with ruptured ectopic must be followed up with clinical examination, and subsequent ultrasonography and β hCG levels on clinical suspicion of on-going intrauterine pregnancy.

For heterotopic pregnancy management is primarily surgical. Other modalities are also available. However there is always an option to wait for spontaneous resolution but this approach is dangerous due to the unstable nature of extrauterine gestations with the potential of rupture resulting in maternal death and jeopardy of intrauterine gestation(s). Adding to this is the lack of clear guidelines as to which patients are able to be safely managed expectantly and how they are best assessed for interval gestational growth and heterotopic pregnancy resolution. Medical management includes laparoscopic or TVS-guided injection of potassium chloride or hyperosmolar glucose into an intact ectopic or heterotopic gestational sac. But more than half of those with tubal heterotopic gestations require subsequent salpingectomy [3].

The management of choice for heterotopic pregnancies is surgery via laparoscopy or laparotomy. Laparoscopic approaches better compared to open procedures except in cases

of hemorrhagic shock with intra-abdominal hemorrhage where laparotomy may be preferred. In our case, we opted for laparotomy - left salpingectomy as the tube was ruptured as evidenced by TVS.

Current evidence shows that despite our best efforts, the intrauterine component of a heterotopic pregnancy has a higher likelihood of miscarriage than sole intrauterine pregnancies, although survival rates of intrauterine heterotopic pregnancies have improved over the past few decades with those that proceed to live birth demonstrating no significantly different rates of adverse birth outcomes.

Conclusion

Heterotopic pregnancy can be potentially life-threatening condition that, though rare, but potentially has grave consequences for both the mother and fetus. High-risk groups should get early pregnancy ultrasound as a part of routine antenatal care for timely management. As highlighted in our case above, it must remain at the forefront of a clinician's diagnostic algorithm in all women as it may occur in the absence of risk factors in a natural conception cycle. Despite it being a challenging diagnosis, clinical acumen along with skilled TVS and timely management is able to achieve optimal clinical outcomes.

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