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Case Note

Heterotopic pregnancy with spontaneous intrauterine conception: A rare clinical entity with diagnostic dilemma

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Abstract
Twenty-one years old primigravida presented to emergency with amenorrhoea for 2 months and complaints of severe abdominal pain for few hours. The pain was associated with 2 episodes of fainting attacks in emergency during the period of observation. Viable intrauterine pregnancy of 8-9 weeks along with collection of fluid in the Pouch of Douglas was detected by ultrasound examination and on laparotomy ectopic pregnancy was confirmed with haemoperitoneum of 2 litres with 500gms of clots. Histopathology report confirmed the tubal ectopic pregnancy and postlaparotomy, transvaginal sonography confirmed the salvage of the intrauterine pregnancy. Despite massive haemoperitoneum, the pregnancy continued till 40^{+6} weeks with uneventful antenatal period. She underwent emergency caesarean section for meconium stained liquor with foetal distress and delivered of an alive healthy female of 2.5 kg with good Apgar score.

Key words: Ectopic, Heterotopic pregnancy

Heterotopic pregnancy is the coexistence of intrauterine and extrauterine pregnancies. It has been traditionally regarded as a very rare event with an estimated incidence of 1 in 30,000 pregnancies[1].

It rarely occurs in spontaneous conception cycles until recently, especially with the advent of assisted reproductive procedures. It is definitely a fatal condition that might even lead to maternal morbidity and even mortality if there is a delay in intervention assuming it to be an intrauterine pregnancy (IUP). IUP diagnosed with ease in this case will enumerate how the source of haemoperitoneum was least thought to be of tubal origin because of the rarity in the incidence of heterotopic pregnancy.

Case report
Twenty-one years old primigravida presented to the emergency with amenorrhoea for two months and complaints of severe abdominal pain for 3-4 hours. The pain was severe pricking in nature and diffuse all over the abdomen, associated with one episode of vomiting, though there was no complaints regarding vaginal bleeding or trauma. She collapsed twice in the emergency where she was resuscitated with 3 litres of intravenous fluids and antispasmodic drugs.

She was regularly menstruating female with cycles of 28 days and bleeding for 3-4 days. She was married for two and half months and had her last menstrual period about a month and half back. As the dates of her marriage was approaching nearer, in order to defer her menstruation she took some pills about 10-12 days prior to her last periods following which she had withdrawal and conceived. Urine pregnancy test done in a missionary hospital confirmed her pregnancy.

On examination she was sick looking, pale, blood pressure was 100/70 mm Hg, pulse 92 beats/min, abdomen was soft with tenderness over the suprapubic region, associated with guarding and rigidity and on auscultation the bowel sound was absent. Per speculum examination revealed closed os with no vaginal discharge or bleeding. Vaginal examination revealed 8 weeks size tender uterus with cervical excitation and fullness in both right and left fornices. Investigations showed haemoglobin of 8.6 gm%. Abdominal ultrasonography done in the emergency showed free fluid in the abdominal cavity and pelvis, along with intrauterine pregnancy and the gestational sac corresponding to 8-9 weeks of gestation with visible cardiac activity.

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Being in dilemma transvaginal sonography was done which also showed single live intrauterine pregnancy with CRL of 1.8 cm corresponding to 8 weeks 4 days ± 7 days (Figure 1a) with doubtful right adenexal mass (Figure 1b) and significant collection in Pouch of Douglas. Haemoperitoneum was confirmed by aspiration of 5 ml of blood from Morison's pouch. The cause of the free fluid in the abdominal cavity and the pelvis was difficult to relate to at that time and because of haemoperitoneum, she was planned for exploratory laparotomy with the differential diagnosis of intrauterine pregnancy with either ruptured corpus luteum cyst or hemorrhagic ovarian cyst, or ruptured ovarian or uterine vessels, thinking least of heterotopic pregnancy. Laparotomy revealed haemoperitoneum of 2 litres with 500gms of clots. Uterus was of 8 weeks size and there was ruptured right-sided isthmic pregnancy with a rent of about 2x2.5 cm and blood was tickling out. Length of the bilateral tubes was approximately 13 cms. The left tube and the bilateral ovaries were normal. Right-sided salpingectomy was done (Figure 2). She received 4 units of blood after surgery raising the postoperative haemoglobin to 10.7 gm%.

Her postoperative period was uneventful. Transvaginal sonography done post laparotomy on the 2nd postoperative day showed a single live intrauterine pregnancy with normal cardiac activity. The histopathology report of the resected right tube was consistent with tubal ectopic pregnancy. She was discharged from the hospital on the 5th postoperative day. There after she was on regular antenatal check up which was uneventful. At 40^{+6} weeks, she was induced with 2 doses of misoprostol for being post-dated but had to under go emergency caesarean section for foetal distress with thick meconium stained liquor. She delivered an alive healthy female baby with birth weight of 2.5 kg with umbilical cord round the neck once loose and with good Apgar score.

Fig 1a: IUGS (Intrauterine gestational sac) with live foetus shown

Fig 1b: Heterotopic pregnancy seen along with the IUGS

Fig 2: Resected ruptured tube: a seat of heterotopic tubal pregnancy

Discussion

Very few cases of heterotopic pregnancies have been reported till date. The incidence is increasing mainly due to increase in the development of medical procreation techniques, and has increased to 1 in every 7000 pregnancies in general population and 1 in 100 pregnancies among the women who have assisted conception [2.]

A rare case of simultaneous tubal-splenic pregnancy after assisted reproductive technology as well as a case of heterotopic cervical and intrauterine pregnancy in a spontaneous cycle has also been reported [3,4]. A large number of heterotopic pregnancies following artificial reproductive technique have been reported but heterotopic pregnancy following a spontaneous conception along with the salvage of a live healthy baby following such massive haemoperitoneum and anaesthetic exposure is rare and not written about.

Heterotopic pregnancy in this case could be attributed to the longer length of the tubes (12-13 cms), which hampers the propulsion of fertilized ovum towards the uterine cavity, as much longer distance has to be travelled by it. Pills probably progesterone taken by the patient to defer her menstruation could have been responsible as they decrease the motility of the tube. Transperitoneal migration of sperm and the oocyte or the fertilized ovum remains uncertain. A transperitoneal migration appears plausible if contralateral corpus luteum is found as usually seen in 16% of ectopic pregnancies, which was not seen in this case [5]. It could be due to some chemotactic factors that attract the sperm or the fertilized ovum across the peritoneum in direction of the contra lateral tube [5.] These two points are rather theoretical.
The abdominal pain could be attributed to ruptured corpus luteum or hemorrhagic ovarian cyst, though the most frequent symptom in heterotopic pregnancy is pain abdomen, which is seen in about 82.7% of cases [6]. Patient with pain abdomen and an intrauterine pregnancy should always heighten the possibility of heterotopic pregnancy.

The urine pregnancy test that was done did suggest of pregnancy but it did not rule out the possibility of a simultaneous intrauterine with the extrauterine pregnancy.

So we opted for ultrasonography. Whenever confusion arises regarding the intra or an extrauterine pregnancy it is always a wise decision to approach for transvaginal sonography (TVS) that has a specificity of 73.7% and positive predictive value of 89.8%, though there are studies reporting, that abdominal sonographic diagnosis of heterotopic pregnancy is sometimes possible but not always so[7,8]. The identification of a live embryo within a gestational sac outside the uterus is the gold standard for the sonographic diagnosis of ectopic pregnancy [9]. However an over whelming diagnostic difficulty was faced in this case as both transabdominal and transvaginal sonography were not of much help because the ectopic pregnancy was easily missed on transabdominal and though a mass was seen transvaginally it was misdiagnosed as an adenexal mass which could have been the ruptured corpus luteum or a hemorrhagic ovarian cyst, along with the presence of a live intrauterine pregnancy of 8-9 weeks, thinking least of heterotopic pregnancy .The echogenecity of an adenexal mass may help distinguish the tubal ring of an ectopic pregnancy from corpus luteum. The tubal ring of an ectopic pregnancy is usually more echogenic than ovarian parenchyma, and the corpus luteum is usually equal to or less echogenic than ovary [10].

Though the history, the clinical examination as well as fluid aspirated from the Morison's pouch did give a clue regarding the heterotopic pregnancy, it was far from thought as it was such a rare event that we hardly come across.

Laparoscopy on the other hand could have been helpful in establishing the diagnosis. These days' large numbers of cases are being diagnosed as well as treated simultaneously with laparoscopy. Some authors recommend magnetic resonance imaging (MRI) when there is high suspicion of ectopic or heterotopic pregnancy, where transvaginal sonography does not point to the accurate location of pregnancy and in situations where laparoscopy is contraindicated or not available [11].

Laparotomy is the choice of treatment reserved for cases of internal bleeding as in our case.

The main challenge in case of heterotopic pregnancy is to preserve the development of the intrauterine pregnancy, with subsequent normal pregnancy course and outcome, which we were able to do by stabilizing the hemodynamic compromise with intravenous fluid infusion and blood transfusion that sub served to promote progression of intrauterine pregnancy with subsequent good foetal outcome, which is really rewarding, though there was delay in detecting the ectopic pregnancy component.

Conclusion

This case suggests that the clinician should maintain a reasonable index of suspicion while evaluating a patient with pain abdomen and amenorrhoea even in face of documented intrauterine pregnancy. An early and prompt intervention at the very first sight of heterotopic pregnancy is a must to salvage the intrauterine pregnancy and to avoid missing this potentially life-threatening condition that can lead to maternal morbidity and mortality.

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