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Case Report

Spontaneous heterotopic pregnancy with tubal rupture and pregnancy progressing to term

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problem, management can be difficult and may be life threatening even when surgical intervention with laparotomy is performed. We report a case of heterotopic pregnancy in a natural conception cycle that presented with tubal rupture and thereafter the intrauterine pregnancy went to term. An obstetrician should keep in mind the occurrence of an HP while dealing with pregnant females. The ectopic gestation invariably ruptures over a period of time leaving the patient in an emergency situation. A quick assessment and careful handling of the normal gestation can lead the patient to term with gratifying results.

Case report

Introduction

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. It was first reported in the year 1708 as an autopsy finding.¹ It is a rare form of twin pregnancy, with an estimated incidence of 1/7000–1/30,000 in spontaneous pregnancies.² However in the last decades there has been a significant increase of ectopic pregnancy and a subsequent increase of HP. This raised frequency has been attributed to several factors including higher incidence of pelvic inflammatory disease (PID) and the extended use of assisted reproductive technologies (ARTs). For patients who have been treated with ovulation induction medication an incidence of 33/10,000 has been reported,³ while it is extremely rare among women who conceive naturally. Aside from the difficulty of diagnosing the

A 31-year-old primigravida patient a known case of hypothyroidism married for one and a half years was admitted at 9 weeks 3 days period of gestation with severe pain in her abdomen of few hours duration associated with vomiting. It was a spontaneous conception and there was no past history of pelvic inflammatory disease, infertility, abortion, use of contraception or any history of abdominal surgery. In this pregnancy, she underwent ultrasonographic examination at 8 weeks period of gestation (POG) which revealed single live intrauterine pregnancy corresponding to period of amenorrhea. On examination, she was pale with a pulse rate of 96 per minute and blood pressure of 106/60 mmHg. Abdominal examination revealed diffuse, lower abdominal tenderness with significant guarding and rigidity. Pelvic examination revealed an anteverted, enlarged, soft and tender uterus corresponding to 8 weeks of pregnancy. In addition, a tender mass was also palpable in her right adnexa. Cervical movements were

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painful but there was no bleeding. Her hemoglobin was 8.2 gm/dl with a normal white blood count (WBC) and platelet count. Transvaginal sonography revealed an 8 weeks 6 days intrauterine viable pregnancy and a 3.3×2.2 cm echogenic mass near her left ovary with a 1.4×1.0 cm central anechoic area. A moderate amount of fluid ($10 \times 6.5 \times 3$ cm) was present in the cul-de-sac and a diagnosis of heterotopic pregnancy with tubal rupture was made. An emergency laparoscopy on the patient revealed an 8-week gravid uterus and rupture of the left tube near the fimbrial end and the presence of approximately 1.5 L of hemoperitoneum. Laparoscopic left salpingectomy with aspiration of the hemoperitoneum and peritoneal lavage was performed. She was transfused with 2 units of blood during and after the surgery and her postoperative period was uneventful. Post operatively she was put on progesterone support (200 mg/day) which was continued till 16-week POG. The material was sent for definitive histologic examination and was confirmed chorionic villi suggestive of a tubal ectopic. The patient was discharged and followed-up regularly in the antenatal clinic. At 38 weeks gestation she went into spontaneous labor and delivered a healthy female baby weighing 2.8 kg. In the postnatal period the baby develop neonatal jaundice which required phototherapy. Postnatal recovery was uneventful, for mother and they were discharged on the fifth postpartum day.

Discussion

The coexistence of intrauterine and extrauterine pregnancy, also known as heterotopic pregnancy, can occur in different forms: intrauterine pregnancy and tubal, abdominal, corneal, cervical or ovarian pregnancy. A previous review showed that most of extrauterine pregnancies were located in the fallopian tube (72.5%).⁴ The occurrence of an HP is considered rare in natural conception cycles with an incidence of 0.08%, but incidence increases to as high as 1% with assisted reproductive techniques.¹ The increased incidence of multiple pregnancy 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.⁵ The common factors that predispose to occurrence of ectopic pregnancy are tubal surgery and pelvic inflammatory diseases.

The majority of HP cases are diagnosed late. Significant morbidity and occasional mortality have been reported as a result of a delay in diagnosis.⁶ As no single investigation can predict the presence of an HP, it should be suspected in any patient who presents with lower abdominal pain in the early phase of an obvious intrauterine pregnancy (IUP) following fertility treatment.⁷ Reece et al⁸ defined abdominal pain, adnexal mass, peritoneal irritation and an enlarged uterus as signs and symptoms suspicious of HP. Most ultrasonographic reports make no mention of a search for coexistent ectopic pregnancy when evaluating intrauterine gestation, because an HP is still thought to be extremely rare and for this reason, almost all ectopic pregnancies are diagnosed by excluding an IUP. 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.

Spontaneous HP is a potential fatal condition. Clinicians should maintain a high index of suspicion in all patients presenting with amenorrhea, abdominal pain, adnexal mass, peritoneal irritation, and enlarged uterus, even if an IUP has been confirmed. Suspicion should be higher in women with risk factors for an EP and in low risk women who have free fluid with or without an adnexal mass with an IUP.⁹ An HP is difficult to assess as pain and bleeding might be attributed to a threatened abortion. If ART is not involved, the index of suspicion of an HP is usually very low. This fact could cause more serious complications, such as a delayed diagnosis. The presence of an IUP, either viable or not, may actually mask the ectopic component of an HP, resulting in delay of diagnosis. A simultaneous IUP causes difficulties in the interpretation of ultrasound images. The ultrasound visualization of heart activity in both intrauterine and extrauterine gestations is important for diagnosis, but rare. During an ultrasound examination an ovarian pregnancy is easily misdiagnosed with a corpus luteum. Intrauterine gestation with hemorrhagic corpus luteum can simulate heterotopic/ectopic gestation both clinically and on ultrasound. Other surgical conditions of acute abdomen like appendicitis, ruptured ovarian cyst, torsion of hydrosalpinx¹⁰ can also simulate heterotopic gestation clinically and hence the difficulty in clinical diagnosis. Bicornuate uterus with gestation in both cavities may also simulate a heterotopic pregnancy.¹¹

The management of HP remains controversial. Surgical therapy has been the traditional mainstay but involves surgical and anesthetic risks to both the mother and IUP.¹² Studies suggest that laparoscopic management is preferred over laparotomy in patients with a suspected EP, and with a documented IUP because of minimal manipulation of the uterus.⁷ A non-surgical approach can be used safely and effectively to manage patients who are clinically stable and where an HP is recognized relatively early in gestation. The successful non-surgical management of six cases of HP using potassium chloride (KCl) injection into the tubal ectopic pregnancy has been reported.¹² Successful management of heterotopic pregnancy after fetal reduction using potassium chloride and methotrexate was also been reported.¹³

Heterotopic pregnancy can occur in the absence of any predisposing risk factors like in our patient. The detection of an intrauterine pregnancy does not exclude the possibility of the simultaneous existence of an ectopic pregnancy and one should be alert for same at the time of ultrasound. To conclude, one may say that though heterotopic pregnancy is rare, when it does occur, it is potentially dangerous to the mother and the intrauterine conceptus. The diagnosis of a heterotopic pregnancy should always be kept in mind in pregnant women presenting with pain in the abdomen. One should include heterotopic pregnancy as a differential diagnosis in women in the reproductive age group presenting with pelvic pain, even when there are no known risk factors.

Conflicts of interest

All authors have none to declare.

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