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Unusual presentation of spontaneous heterotopic pregnancy: A case report

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ABSTRACT: Spontaneous heterotopic pregnancy is the natural co-existence of an intrauterine pregnancy with an ectopic pregnancy. It has a historic incidence of 1 in 30,000. Its clinical presentations can be diverse and can challenge the diagnostic skills of clinicians. I present a case of spontaneous miscarriage of a 6 week intrauterine pregnancy in a patient with spontaneous heterotopic pregnancy. This presentation is an unusual clinical scenario for heterotopic pregnancy. To the best of my knowledge, there are no previous reports of such presentation in the literature. I highlight our diagnostic dilemma with this presentation and reiterate the essential tripod for diagnosis of heterotopic pregnancy.

Key words: heterotopic pregnancy, spontaneous miscarriage, ultrasound, laparoscopy.

Introduction

Spontaneous heterotopic pregnancy is the natural co-existence of an intrauterine pregnancy with an ectopic pregnancy. It was first reported in 1948 and has a historic incidence of 1 in 30,000[1]. In the last few decades there has been an increase in frequency of heterotopic pregnancy. An incidence of 1:4000 is now reported in the general population[2]. This rise in frequency has been attributed to the increasing incidence of pelvic inflammatory disease, endometriosis, tubal surgeries, and intrauterine devise usage as well as the use of assisted reproduction technologies and ovulation induction regimens[3].

The clinical presentations of spontaneous heterotopic pregnancy can be diverse and varied. This can challenge the diagnostic skills of clinicians. It could also have potentially significant medico legal implications. Often, it presents as an acute abdomen with tubal rupture of its ectopic component[4]. It may present as a viable intrauterine pregnancy with unilateral pelvic pain[5] or as acute appendicitis[6]. It can also present as a spontaneous miscarriage of its intrauterine component as was the case with our patient. To the best of our knowledge, there have been no previous reports of spontaneous heterotopic pregnancy presenting as a spontaneous miscarriage in the literature.

In this report, we present a case of spontaneous miscarriage of a 6 week intrauterine pregnancy in a patient with spontaneous heterotopic pregnancy. We highlight our diagnostic dilemma with this presentation, its potential medico legal implications and the important role of laparoscopy in our management of this condition.

CASE REPORT

A 32 year old G7P2^{+4} (3 alive, I set of twins) presented to the Emergency Department following a referral from a general practitioner with a 6/52 history of amenorrhoea and a 2/52 history of right lower abdomen pain associated with intermittent mild PV bleeding. She was in her 7th month postpartum after a vaginal delivery of a set of twins.

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She was not on any contraceptive. She had no history of pelvic inflammatory disease or endometriosis and did not use any ovulation induction agents. Her general condition was stable on admission. She had rebound tenderness and guarding in the right iliac fossa. A speculum examination showed an open cervical Os containing product of conception. These were removed and sent for histopathology. A subsequent bimanual examination revealed a bulky anteverted uterus with a right tender adnexal mass which was separate from the uterus.

A high resolution transvaginal pelvic ultrasound reported a right adnexal heterogeneous mass measuring 9.6 x 4.3 x 7.4cm with no echogenic rim and containing no foetal or yolk sac. The scan also reported coexisting intrauterine products of conception. Results of additional investigations include Haemoglobin of 13.7g% and a serum BHCG of 4630 mIu/ml.

We made a clinical diagnosis of an incomplete spontaneous abortion coexisting with a right ovarian mass (possibly a haemorrhagic corpus luteum cyst). She was subsequently admitted and consented for a suction dilatation and curettage as well as an operative laparoscopy with or without a possible laparotomy.

Laparoscopy revealed a partially ruptured right tubal ectopic pregnancy associated with a hemoperitoneum of 450mls. Because the ectopic mass was difficult to mobilize from the pelvis, we progressed to a laparotomy. At laparotomy, we performed a right salpingectomy and a pelvic lavage. The left fallopian tube and both ovaries appeared normal with no obvious corpus luteum cyst. She had an uneventful post operative recovery and was discharged home on the second day after surgery to her local doctor for follow up.

The Pathology reports of both the intrauterine curetting and the tubal mass subsequently confirmed the diagnosis of heterotopic pregnancy.

Discussion

The clinical scenarios presented by heterotopic pregnancy can be non specific and widely divergent. It can present as an acute abdomen[4,7], acute appendicitis[6] or spontaneous miscarriage as was the case in our patient. The symptoms and signs of heterotopic pregnancy can also occur in other clinical conditions. For example, abdominal pain, peritoneal irritation, adnexal mass and uterine enlargement can occur in ruptured or hemorrhagic corpus luteum, hyperstimulated ovaries and ectopic gestation[8,9]. This diversity in the clinical presentation of heterotopic pregnancy can pose significant diagnostic dilemmas for clinicians. Further more, the normal rise of BHCG, the failure to demonstrate ectopic gestational sac on ultrasound scan and attributing the unilateral pain to a haemorrhagic corpus luteum or a small degree of ovarian hyperstimulation represent additional diagnostic pitfalls in the management of heterotopic pregnancy[5]. In our patient the presence of visible products of conception at the external Os, an adnexal mass and the ultrasound findings led us to a clinical impression of an incomplete miscarriage with a possible haemorrhagic corpus luteum cyst.

The availability of high resolution sonography using transvaginal probe has improved the diagnostic performance for heterotopic pregnancies[10]. It has been suggested that high resolution ultrasound is the most helpful diagnostic tool in heterotopic pregnancy because of its high sensitivity[11, 12]. The ultrasound diagnosis of heterotopic pregnancy is suggested by the presence of echogenic fluid in the cul de sac in the presence of an intrauterine pregnancy and confirmed by the visualization of both an ectopic and intrauterine pregnancy[10].

Still, there are reports of life threatening delays in detecting the ectopic component of heterotopic pregnancy[13]. Often, the investigations for ectopic pregnancy are terminated if a transvaginal sonogram reveals an intrauterine pregnancy[4,8]. Furthermore, early ultrasound diagnosis of heterotopic pregnancy could be difficult because the identification of heart motion in both intrauterine and extra uterine fetus is rare[10] or in circumstances where there is a "*red herring effect*" like the simultaneous presentation of a spontaneous miscarriage as was the case in our patient. This suggests that clinical assessment and high resolution transvaginal sonography are insufficient for the correct diagnosis of heterotopic pregnancy. Thus, emphasizing the need for laparoscopy in suspicious cases.

In our patient, our clinical assessment and a transvaginal ultrasound were inconclusive for any specific diagnosis hence the need to progress to a laparoscopy. Laparoscopic intervention is the gold standard in the definitive diagnosis and treatment of heterotopic pregnancies[3]. Indeed, early laparoscopic intervention prior to rupture of the ectopic gestation could result in non surgical treatment especially when the intrauterine gestation is viable[14]. In cases of rupture, laparoscopic salpingectomy is the management of choice[15].

The main objective in the management of heterotopic pregnancy is to be as minimally invasive as possible to preserve the intrauterine pregnancy[16]. Although in our patient, her presentation precluded this caution. However, because laparoscopy can be safely performed to aid differential diagnosis in any uncertain condition during early pregnancy[17], missing a diagnosis of heterotopic pregnancy can have potentially significant medico legal implications.

Where the intrauterine gestation is viable and desirable, use of methotrexate injections into the unruptured ectopic sac or excessive handling of the uterus may induce a spontaneous miscarriage of the intrauterine gestation or result in severe congenital anomalies. On the other hand, the non diagnosis of the ectopic component of heterotopic pregnancy may result in life threatening delays with rupture of the ectopic component. Rupture of ectopic

pregnancies in women with heterotopic pregnancies has been shown to have a significantly greater risk for hypovolemic shock and require blood transfusions than those with ectopic gestation alone[9]. It is therefore important to maintain a high index of suspicion especially following assisted reproductive technology, while evaluating a patient presenting with pelvic pain in the face of a documented intrauterine pregnancy[3].

In conclusion, our patient presented an unusual clinical scenario for heterotopic pregnancy. To the best of our knowledge, there are no previous reports of such presentation in the literature. The diversity of presentations of heterotopic pregnancy challenges the diagnostic skills of clinicians and ultrasonographers. Thus, the essential tripod for diagnosis of heterotopic pregnancy would include a high index of suspicion, a detailed high resolution ultrasound and laparoscopy, bearing in mind the potentially significant medico legal implications of a misdiagnosis.

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