



Case report

Expect the unexpected: The dilemmas in the diagnosis and management of interstitial ectopic pregnancy—Case report and literature review

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ABSTRACT

Interstitial pregnancy is a rare type of ectopic pregnancy; it tends to present late, is most commonly missed, and is associated with significant maternal morbidity and mortality. Early clinical diagnosis aided by ultrasound and laparoscopy may help contribute towards effective conservative or fertility preserving treatment modalities and thereby reduce potential morbidity and mortality. We present a case of missed interstitial pregnancy in an inner city London hospital, where the ultrasound diagnostic accuracy published is as high as 90%, with the aim of analyzing the difficulties and dilemmas in the diagnosis and the management of interstitial pregnancies.

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Introduction

Interstitial pregnancy is rare, but is one of the most hazardous types of ectopic gestation, accounting for 2–4% of all ectopic pregnancies.¹ The true incidence is probably unknown, as in medical literature the terms *cornual* and *interstitial* pregnancy are used interchangeably.

Interstitial pregnancies often present late due to the distensibility of the surrounding myometrial tissue and rupture at an advanced gestation leading to catastrophic haemorrhage. They pose a significant diagnostic and therapeutic challenge and carry a greater maternal mortality rate in the range of 2.0–2.5%,¹ which is two to five times higher than that of the other tubal ectopic pregnancies. In the 2000–2002 confidential enquiry into maternal and child health report, four out of the 11 deaths from ruptured ectopic pregnancies were due to interstitial pregnancies.²

Despite technological advances in the ultrasound and the availability and easy access to early pregnancy units, interstitial ectopic pregnancies are often missed. Reasons for missing include: lack of suspicion, lack of diagnostic expertise, and late presentation of patients. Advice usually is that patients be educated and seen in dedicated early pregnancy assessment units to ensure early diagnosis and therapy aimed at preserving fertility options. This case demonstrates that even in units where the diagnostic accuracy of cornual/interstitial ectopic pregnancies is high (90%),³ these pregnancies can still be missed.

Case Report

A 37-year-old woman in her second pregnancy presented at 5 weeks' gestation with complaints of vaginal bleeding and abdominal pain. She was scanned by a senior trainee with some scanning experience out of hours and a diagnosis of an incomplete miscarriage was made. Suction evacuation of retained products of conception (ERPC) was performed as opted by the patient. The histology report confirmed products of conception with decidual tissue and chorionic villi in the sample.

Five weeks later, she presented with ongoing symptoms of abdominal pain, vaginal bleeding, and a positive pregnancy test.

Conflicts of interest: All authors declare no conflicts of interest. No resources were received from any third party either directly or indirectly.

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She was thought to have had a new conception, but was unsure of the gestational age as she had no period after the ERPC. Transvaginal ultrasound performed by a consultant with expertise in early pregnancy scanning, revealed a large left-sided interstitial ectopic pregnancy measuring 4.0 cm × 3.6 cm (Figure 1). In view of the patient's symptoms and the large size of the interstitial ectopic pregnancy surgical intervention was deemed safest, although serum β human chorionic gonadotropin level was only 200 IU/L.

At laparoscopy, a complex mass of hemorrhagic tissue was noted in the left uterine cornual region (Figure 2), confirming a left interstitial pregnancy. Laparoscopic wedge resection of interstitial pregnancy with ipsilateral salpingectomy was performed and the uterine interstitial region was repaired in two layers with No. 1 polysorb sutures (Figure 3). Postoperative recovery was uneventful and the patient was discharged the day after the procedure. Histopathology report confirmed interstitial ectopic pregnancy.

Retrospective review of the ultrasound pictures at her initial presentation were thought to be substandard and suggestive of a missed interstitial ectopic pregnancy rather than an incomplete miscarriage or a heterotrophic pregnancy with intrauterine and interstitial pregnancy. Also, the histological confirmation of the products of conception from the suction evacuation misled the clinicians.

Discussion

Interstitial pregnancy is defined as a pregnancy implanted in the proximal portion of the fallopian tube, the interstitium that lies within the muscular wall of the uterus. The interstitial portion of the tube is tortuous, about 1–2 cm long and 0.7 mm wide, surrounded by the myometrium, and has abundant blood supply from the uterine and ovarian vessels.

Interstitial pregnancies tend to present relatively late at 7–12 weeks' gestation due to myometrial distensibility and the specific symptoms and signs are often missing leading to significant diagnostic and therapeutic challenges. Rupture can lead to massive hemorrhage leading to hypovolemic shock and often death.

Despite the well-known fact that the interstitial and cornual ectopics are two different entities, they are often used as synonyms and the medical literature includes references that use the terms *cornual pregnancy* and *interstitial pregnancy* interchangeably. It is important to differentiate and report these two entities separately as the clinical course and management differ markedly between cornual gestations and interstitial ectopic gestations.⁴

Historically, the diagnosis of interstitial pregnancy has been difficult due to its location. The diagnostic accuracy can be



Figure 2. Laparoscopic view of left interstitial ectopic pregnancy.

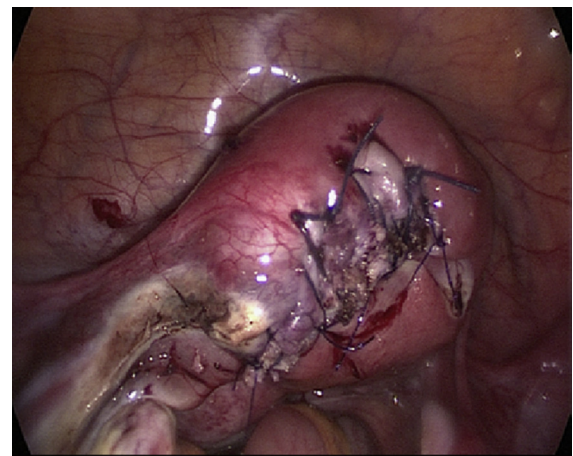


Figure 3. Uterus postlaparoscopic wedge resection and repair.

improved with transabdominal and transvaginal ultrasound using the following criteria⁵: (1) an empty uterine cavity; (2) gestational sac seen separately and > 1 cm from the most lateral edge of the uterine cavity or products of conception located outside of the endometrial cavity; and (3) a thin (< 5 mm) myometrial layer surrounding the gestational sac or products of conception.

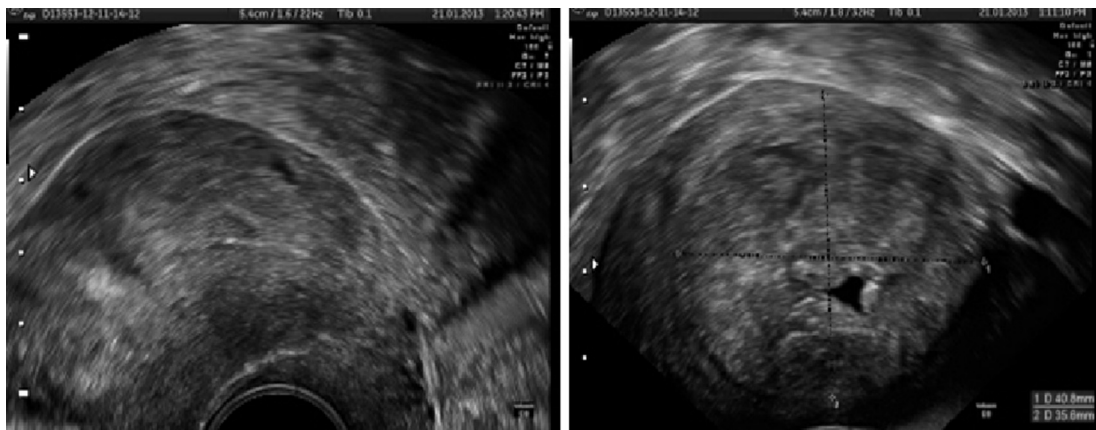


Figure 1. Sonographic pictures: interstitial pregnancy.

Another useful sign is the *interstitial line sign*, which is the presence of an uninterrupted, thin echogenic line extending between the gestational sac and the endometrium, suggesting that the pregnancy is outside the endometrial cavity. The gestational sac is usually in the lateral portion of the uterus early in gestation, but in advanced interstitial pregnancy, it can be located above the uterine fundus and can be confused with an eccentric intrauterine pregnancy.

The interstitial line had better sensitivity (80%) and specificity (98%) than eccentric gestational sac location (sensitivity, 40%; specificity, 88%) and myometrial thinning (sensitivity, 40%; specificity, 93%) for the diagnosis of interstitial pregnancy.⁶

Three-dimensional sonography is an excellent imaging modality in the diagnosis of interstitial pregnancy and the features that are useful include presence of gestational sac surrounded by myometrium below the uterine cornu lying outside the endometrium.^{7,8}

Abbott et al⁹ in 1990 illustrated 10 common pitfalls in the diagnosis of ectopic pregnancy and, so far, the majority of these factors contribute to the misdiagnosis of ectopic gestations. In our case, there was atypical presentation, misdiagnosis on ultrasound due to misinterpretation, and tissue *diagnosis* following ERPC.

Similar cases have been reported in the literature since 1992, when there was a rupture of an interstitial pregnancy during a mid-trimester (18 weeks' gestation) prostaglandin-induced medical termination of pregnancy for presumed intrauterine pregnancy.¹⁰

In 2003, Chan et al¹¹ reported 36 cases of interstitial ectopic pregnancies with an emphasis on the pitfalls in the diagnosis and treatment of these cases. In this series, 41.7% of interstitial ectopics were misdiagnosed at the first presentation, where all but one were mistaken as intrauterine pregnancies. Rupture of interstitial pregnancy occurred in 40% of these women and in two cases, at an advanced gestation of 18–20 weeks. There was a median delay in the diagnosis of 13 days (4–70 days). The most common diagnostic pitfall was intrauterine pregnancy, either viable or nonviable, where a third (33%) of the women underwent suction evacuation and a further 10% were either referred for or planned to have termination of pregnancy or suction evacuation.¹¹

MacRae et al³ reported a case series of 11 women with interstitial pregnancy in our unit, where the diagnostic accuracy was reported to be 90% at an initial ultrasound. In our database with a pool of 998 women with ectopic pregnancies from 2000–2013, 26 women had interstitial (cornual) pregnancies, a prevalence of 2.6%. Even though 24 out of 26 women had a correct diagnosis at initial scan, every missed ectopic should be an incident and these cases should be reviewed in multidisciplinary or morbidity meetings and regular teaching sessions with a view to minimize and prevent the occurrence.³

Where symptoms and signs are taken into consideration, the most common types of ectopic pregnancies missed are those that either present with atypical symptoms or those that are in atypical extratubal locations, such as interstitial^{3,10,11} or ovarian ectopics.¹² Occasionally, an assumption that the diagnosis of ectopic gestation has been excluded by others has led to the delayed diagnoses and adverse outcomes including death, as in the confidential enquiry into maternal and child health report¹² (1994–1996), where a maternal death at 9 weeks' gestation occurred from a ruptured cornual ectopic pregnancy, in a woman who underwent surgical curettage for termination of pregnancy twice.

The recommendations from the above case reports and series are that the possibility of ectopic pregnancy should be considered and ruled out in women presenting with abdominal pain after therapeutic termination in the first or second trimester or after suction evacuation for an incomplete miscarriage of a presumed intrauterine pregnancy. It is important for an experienced sonologist to exclude atypical extratubal ectopics, as the risk of mortality among women with ectopic pregnancy undergoing termination of pregnancy is 1.3 times higher.¹³

Conclusion

Lack of suspicion and lack of expertise are the common causes of missing these rare ectopic pregnancies. There should be established early pregnancy units with systems in place for a regular review of cases performed out of hours or by personnel with limited scanning experience in an emergency and also to provide regular training opportunities in order to mitigate complications and enhance the best possible outcomes for women. Early diagnosis is the key to the management of interstitial pregnancies and expert opinion by a sonologist and/or by a multidisciplinary team should be considered in case of atypical presentations or when there is inappropriate response to treatment to avoid maternal deaths associated with rupture of interstitial ectopic pregnancies.

It is important to differentiate and document interstitial and cornual pregnancies as two separate entities and where possible to offer conservative fertility preserving procedures and to have good patient selection for successful medical management.

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