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Antenatal diagnosis of a third trimester interstitial pregnancy: A case report

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Abstract

Interstitial pregnancy cases that advance to term, or near term, are occasionally reported. We present an unusual case of a third trimester interstitial pregnancy with antenatal diagnosis and expectant management. She presented at 20 weeks of pregnancy with an early preterm premature rupture of membranes, and expectant management was initiated. The ultrasound suggested an interstitial location and a posterior magnetic resonance image, obtained at 26 weeks, confirmed the diagnosis. Because of the risk of uterine rupture, an elective cesarean section was performed at 28 weeks. During the laparotomy, the uterine fundus appeared intact with an asymmetric bulge that provided evidence of placenta increta. The baby was delivered, and an obstetric hysterectomy was performed. The newborn was admitted to the neonatal intensive care unit with a severe respiratory distress syndrome. No response to mechanical ventilation was observed, and neonatal death was reported. A uterine pathological examination confirmed the diagnosis.

Key words: ectopic pregnancy, magnetic resonance imaging, prenatal diagnosis, ultrasonography.

Introduction

Interstitial pregnancy (IP) refers to an ectopic gestation located in the proximal portion of the fallopian tube within the muscular wall of the uterus.[1] It accounts for approximately 3% of all tubal gestations and is frequently associated with an increased maternal mortality rate (2.5%), which is 7–15 times higher than other ectopic pregnancies.[2,3] The location in this relatively thick portion of the tube with higher compliance is associated with delayed presentations currently around 7–16 weeks of gestation when catastrophic complications, such as uterine rupture, occur.[2] IP cases that advance to term, or near term, are occasionally reported.[4–6] We present an unusual case of a near-term IP with an antenatal diagnosis and expectant management.

Case Study

A healthy 30-year-old multiparous female was admitted with a 20-week spontaneous pregnancy presenting a second trimester preterm premature rupture of membranes (PPROM). No previous surgical records or tubal pathologies were reported. She had no previous ultrasound during this pregnancy, and no abdominal pain or cervical bleeding was reported.

After admission, broad-spectrum antibiotics were initiated for ten days. The ultrasound found a single fetus of appropriated growth and anatomy. The amniotic fluid was reduced; the placenta was fundal and showed placental lacunae, partial loss of the clear space, and increased color Doppler extending from the placenta to the surrounding tissues (Fig. 1a). The fetus was located in a superior chamber, separated by a

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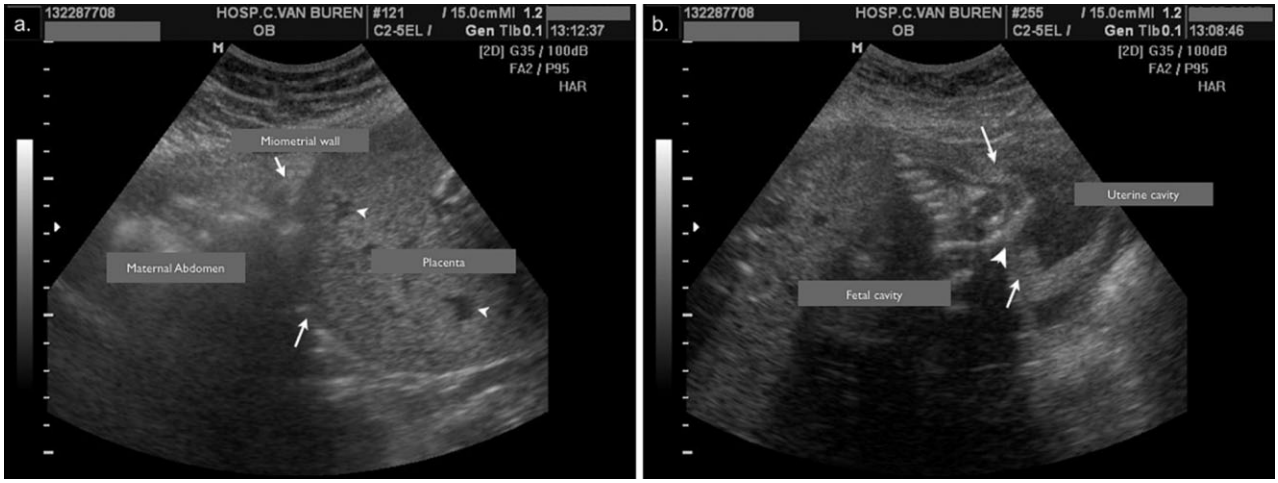

Figure 1 Antenatal diagnosis of interstitial pregnancy. Abdominal ultrasound showing: (a) a fundal placenta with placental lacunae (head of arrows) and a reduced myometrial thickness (arrows). (b) Fetal chamber was separated by a myometrial layer (arrows) from the main uterine cavity. Both cavities are connected by a gap in the myometrium (head of arrow).

myometrial layer from the main uterine cavity. Both cavities were connected by a gap in the myometrium (Fig. 1b). Differential diagnosis between a cornual ectopic pregnancy with placenta accreta and IP was postulated.

After completing antibiotics therapy, an amniocentesis ruled out intra-amniotic infection. Our legislation does not allow feticide or abortion, so expectant management was offered after a detailed explanation to both parents.

At 25 weeks of pregnancy antenatal corticosteroids were initiated. The ultrasound revealed a reduced fetal thoracic circumference and pulmonary pulsatility index suggesting pulmonary hypoplasia. A magnetic resonance image performed at 26 + 1 weeks presented the following: ruled out any uterine Müllerian malformation; showed a myometrial infiltrating placenta; showed that the uterine cavity was divided in two, but communicated by a small gap between both chambers (in the laterosuperior cavity, the fetus held an eccentric position surrounded by myometrium); showed a severe oligohydramnios; and revealed a marked reduction in signal intensities and lung volumes (Fig. 2). These findings match the diagnosis of placenta accreta, IP and pulmonary hypoplasia.

At 27 + 5 weeks of gestation, the patient reported abdominal discomfort and minimal vaginal bleeding. After ruling out fetal distress, the patient was informed of the risks associated with uterine rupture. Informed consent was obtained for an elective cesarean section with an eventual obstetric hysterectomy. After a new course of corticosteroids, an elective cesarean section was performed at 28 weeks. During the laparotomy, the uterus fundus appeared intact with an asymmetric bulge in the right cornual region, showing evidence of placenta increta (Fig. 3a). A hysterotomy was made avoiding the placental insertion (Fig. 3b). A male infant weighting 1000 g was delivered with a 5-minute Apgar score of 9 (Fig. 3c). The placenta was left *in situ* (Fig. 3d), and the obstetric hysterectomy was performed. The newborn was admitted into our neonatal intensive care unit with a severe respiratory distress syndrome. No response to intubation, surfactant therapy and mechanical ventilation was observed, and neonatal death was reported 12 h after delivery. The clinical and radiological examination suggested pulmonary hypoplasia.

The pathological examination of the uterus revealed an ectopic gestation in an artificial 12-cm cavity located in the right tubal interstitial segment with an 11-cm placenta invading the myometrium up to the serosa (Fig. 3e–f). The distal section of the right tube was intact, and the normal uterine cavity was empty, only revealing decidual transformation. The postoperative period was uneventful, and patient was discharged 5 days after surgery.

Discussion

Interstitial pregnancy (IP) refers to an ectopic gestation located in the proximal portion of the fallopian tube within the muscular wall of the uterus.[1] Although it

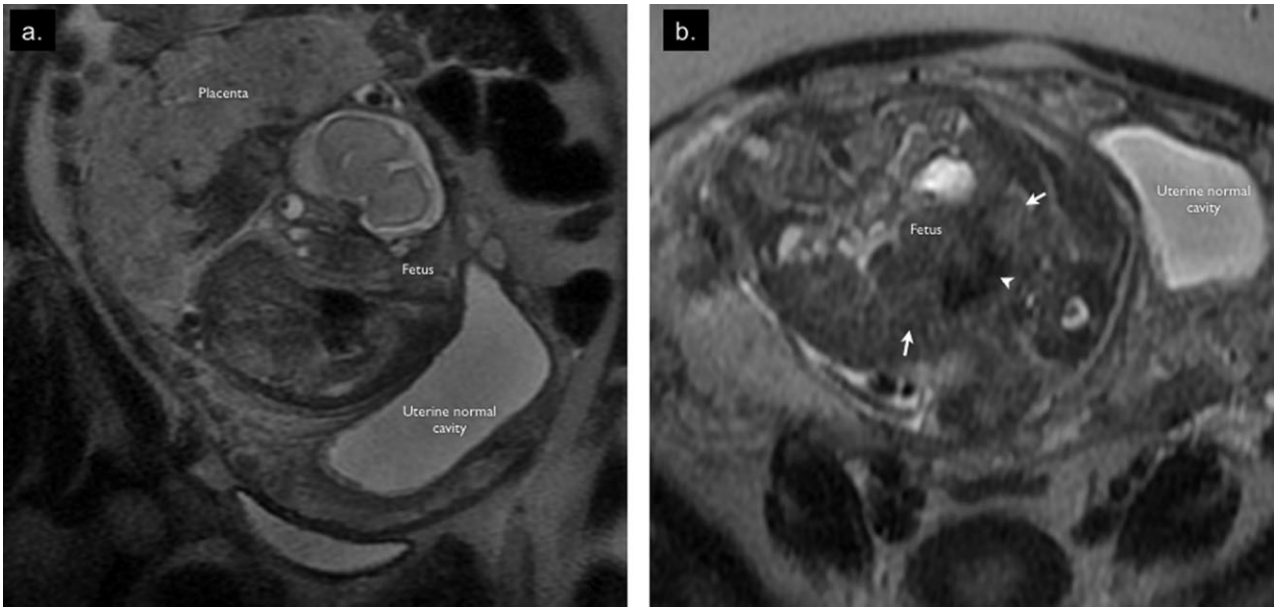

Figure 2 Interstitial pregnancy magnetic resonance imaging showing: (a) sagittal fetal image showing the fetus in a right lateral fundal chamber separated by a myometrial layer from the main uterine cavity with amniotic fluid. The fetus shows a severe oligohydramnios and an infiltrating placenta. (b) Fetal axial section showing with severe oligohydramnios separated from the normal uterine cavity. Lungs show a reduced volume (arrows) next to the fetal heart (head of arrow).

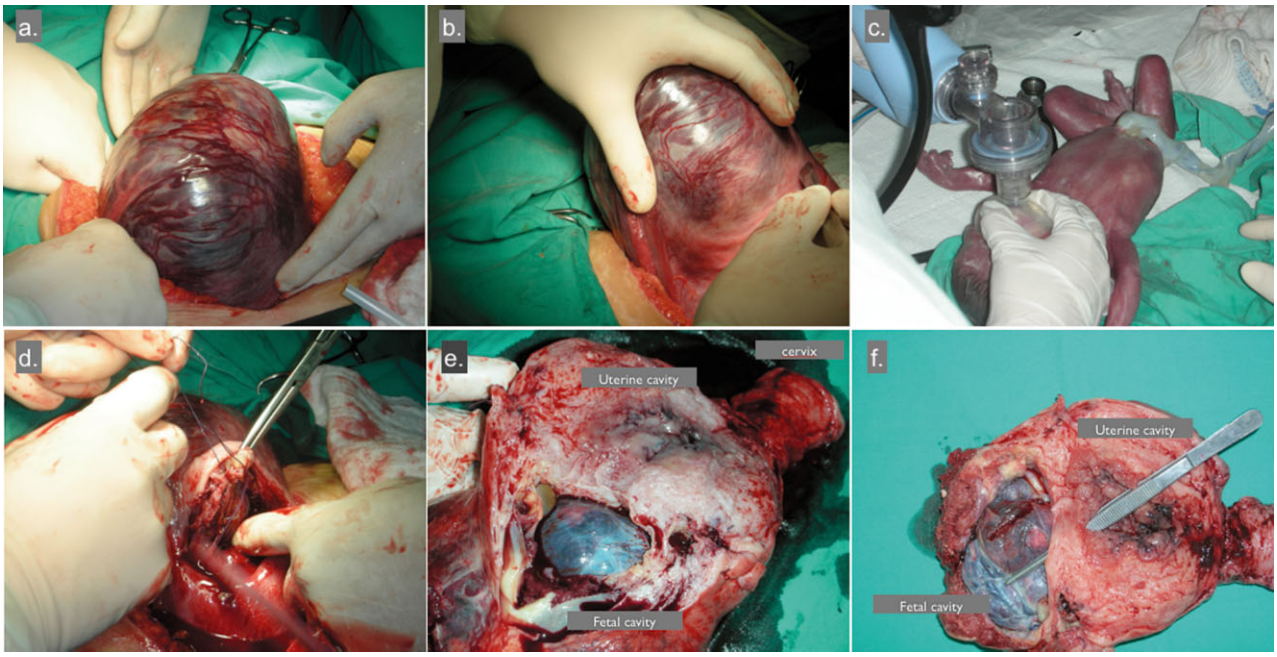

Figure 3 Elective cesarean section performed at 28 weeks. (a) Unruptered uterus with evidence of placenta increta. (b) Anterolateral hysterotomy avoiding the placental insertion. (c) Male neonate. (d) Cord clamping with the placenta *in situ* before hysterectomy. (e) Pathological examination of the dissected uterus with two uterine cavities divided by myometrium. The placenta is located in an artificial cavity located in the right tubal interstitial segment. (f) Forceps showing a myometrial gap communicating both uterine cavities.

accounts for 3% of all ectopic locations, it represents more than 30% of all maternal deaths associated to ectopic pregnancies.[7] Because of the intramyometrial location, between the uterine and ovarian vasculature, the hemorrhage may be massive and life threatening. Furthermore, the bleeding is 2.5–5 times greater than other ectopic pregnancy locations.[8] Contrary to previous reports, it appears that IP can present relatively early in the pregnancy, which ranges from 6.9 to 8.2 weeks.[9,10] Term or near-term IPs are unusual with a few cases reported in the literature.[4–6] To our knowledge, this is the first third trimester IP with unruptured uterus with an antenatal diagnosis reported.

Common findings in IP include abdominal pain and vaginal bleeding during first trimester of pregnancy.[1] During the third trimester, the most characteristic form of presentation is uterine rupture with or without hemodynamic complications. In fact, of the nine term or near-term IP reported in the literature, 6 were manifested with uterine rupture.[4–6,11–16] Interestingly, this patient was asymptomatic until PPROM presented at 20 weeks of gestation.

Early diagnosis of IP is difficult to archive. It must be clearly differentiated from cornual, angular and intrauterine pregnancies because the behavior, management and clinical outcomes of these pregnancies are different. The combination of a high index suspicion, the development of a high resolution ultrasound and access to β-hCG assays are necessary for an early and accurate diagnosis.[1] When ultrasound is inconclusive, magnetic resonance image has been used as a useful tool to differentiate IP from other ectopic pregnancies. Several criteria for the diagnoses of IP have been established; unfortunately, they only apply to early pregnancies.[1]

The incidence of previable PPROM is as low as 0.37%, but it has significant complications for the mother and fetus.[17] Pulmonary hypoplasia is a serious complication of previable PPROM, and mortality ranges between 50–100%.[17] Our patient presented with previable PPROM and was managed expectantly, with latency to delivery that was higher than 8 weeks. Even though pulmonary hypoplasia was antenatally diagnosed, the fetal outcome was unwelcome.

As we discussed in this report, for an IP without clinical symptoms, a supervised expectant management can be offered as an option to patients. No guidelines have been postulated to manage third trimester IPs, nevertheless, it must be highlighted that efforts should be made to establish an early diagnosis of IPs before rupture and symptom develop, when more conservative and non-invasive treatments can be introduced.

Disclosure

The authors have no potential conflicts of interest to disclose. Patient approval: written consent was obtained from the patient.

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