Angular Ectopic Pregnancy Presenting as Rupture of Lateral Wall of the Uterus: Late Presentation in Gestation Week 20

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Abstract

The differential diagnosis of acute abdominal pain in pregnancy is broad and can be complicated by atypical manifestations that are due to the anatomic distortions and physiologic changes of pregnancy. Due to the lack of clinical understanding, angular pregnancy does not appear to be recognized as a clinical entity and many cases are likely to go undiagnosed. This is a case report of a 34-year-old woman who was referred to the obstetrics emergency department with sudden abdominal pain and in a state of hypovolemic shock. She had 20 weeks amenorrhea with a positive blood pregnancy test. She underwent laparatomy with internal hemorrhage diagnosis. During the emergency laparotomy, the authors were surprised to encounter the conceptus of 20 weeks angular pregnancy extruded through the left lateral angulation of uterine cavity. The placenta and amnion were removed and the uterine was repaired. Angular pregnancy is rare, but it should be fully understood since its clinical management, outcomes, and maternal mortality are different.

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Keywords • Ectopic pregnancy • Angular pregnancy • Gestational age

What's Known

Angular pregnancy is rare.
 Diagnosis and treatment of angular pregnancy are yet to be addressed in textbooks. Angular pregnancy is important since it is associated with spontaneous abortion, maternal death, uterine rupture, and abnormal placental implantation.

What's New

• We have introduced a rare case that is unknown with routine pregnancy imaging without a risk factor for ectopic pregnancy. Our finding allows physicians to be aware of the unusual type of ectopic pregnancy and its presentation.

Introduction

Angular pregnancy was first defined in 1898 by the American obstetrician Howard Kelly as "implantation of the embryo just medial to the uterotubal junction, in the lateral angle of the uterine cavity" and medial to the round ligament. In the literature, terms such as "cornual," "interstitial," and "angular" pregnancies are used incoherently. Some sources use "interstitial" and "cornual" synonymously, while others use "cornual" for gestations in bicornuate or septate uteri; others differentiate interstitial from angular pregnancy, while in practice, many physicians are unfamiliar with such classification.

Case Report

A 34-year-old gravida 4, para 1, abortion 2, with previous cesarean section woman was referred to the obstetrics emergency department in June 2015. The patient had sudden abdominal pain an hour prior to admission without any history of spotting and abdominal pain. Her menstrual periods were regular and her gestational age was 20w+2d based on her last menstrual period. She had a history of two curettages. The first sonography

at 6 weeks was normal. Other trimester sonographies reported the fetus in transverse-lie on the left side of uterus. She was referred to a hospital twice (in one week) for abdominal pain, but there were no abnormalities except for a sonography at 14 weeks. It reported moderate free fluid in the pelvic, paracolic, or subhepatic.

Upon arrival, the patient was in hypovolemic shock with intense pallor, filiform peripheral pulses, and cold extremities. Her pulse rate was 120 bpm, blood pressure 80/40 mmHg, and the axillary body temperature was 36.3°C. Generalized abdominal tenderness and rigidity were detected. Vaginal examination showed no bleeding. Bedside transabdominal ultrasound scan showed a live transverse fetus in the uterus with a normal fetal heart rate (FHR) and activity (figure 1). Free abdominal fluid was also detected in the cul-de-sac and Morrison pouch. In addition, laboratory evaluation revealed that hematocrit was 20.7%, hemoglobin 7.2 gr/dl, and white cell blood count 11,900/mL. A live fetus in the uterine ruled out uterine rupture. During the emergency laparotomy, 700 ml blood and nearly 1,300 ml coagulum were drained. The conceptus of 20 weeks angular pregnancy was extruded through the left lateral angulation of uterine cavity (figure 2). Transfusion of packed red cells was started and the fetus, placenta, and amnion were removed and the uterine was repaired in two layers. The patient recovered well after the laparotomy and discharged from the hospital on the third postoperative day. Beta-hCG titration 24 hours after laparatomy was 1,114. Patient's consent was obtained for reporting this case.

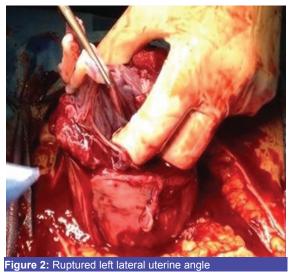
Discussion

Interstitial pregnancy is rare, but account for 2-4% of ectopic pregnancies.³ Specific criteria for diagnosing an angular pregnancy were proposed by Jansen and Elliott in 1981 as:¹

- Clinical presentation with painful asymmetric enlargement of the uterus
- Directly observed lateral distension of the uterus, with or without rupture, accompanied by displacement of the round ligament reflection laterally
- Retention of the placenta in the uterine angle. In over 75% of cases, the evolution involves abortion between 12 and 20 weeks of gestation, fetal death, or preterm birth of a child who does not survive. Spontaneous rupture of the uterus is at times possible in the absence of contractions as a result of overdistension of the walls of the angular sides, particularly thin at this level. Sometimes this condition forces



Figure 1: Bedside transabdominal ultrasound scan shows alive transverse fetus in the uterus with normal fetal heart rate and activity.



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hysterectomy, as it is life threatening to the patient.⁴ Considering the intrauterine location of angular pregnancies and the enveloping myometrium, these patients would possibly present the symptoms later than those with other types of ectopic pregnancies.

Considering the similarities between three-dimensional sonography and MRI in the diagnosis of uterine malformations, the relationship between the cavity and fundus being visualized with both techniques would be equivalent. With MRI, angular pregnancies appear as gestational sacs implanted in the lateral angle of the uterus and can be mistaken with normal pregnancies. These gestational sacs will be completely surrounded by uterine myometrium, although focal thinning is seen at times. Considering the tendency to rupture, it is also important to look for myometrial discontinuity and hemoperitoneum whenever angular pregnancy is suspected.

Three-dimensional sonography is preferred because of more rapid access and lower cost. In addition, 3D sonography can be used to visualize uterine malformations as well as the location of the gestational sac.¹

Reports have suggested that conservative management by transvaginal route is an effective and safe option for some angular pregnancies.5 A conservative approach is possible when the diagnosis is made early by medical therapy with methotrexate. Treatment with methotrexate started via intramuscular injection (dose of 1 mg/ kg on days 1 and 3) in combination with folic acid IM (dose of 0.1 mg/kg on days 2 and 4). An operative hysteroscopy was done after two days, which appeared almost completely collapsed.6 The patient was first treated with methotrexate IM (dose of 1 mg/kg of body weight on days 0, 2, 4, and 6) in combination with folic acid IM (dose of 0.1 mg/kg of body weight on days 1, 3, 5, and 7). The following day, after the end of the medical treatment, a diagnostic laparascopy and hysteroscopy were performed.7 For the other management, 20 mg of MTX in 10 ml of saline was injected into the sac using a 22-gauge needle. Three months later, the gestational sac was measured and it was undetectable by ultrasound 6 months later (Dei Malatesta et al. 2007). Laparotomy is needed to confirm if the ectopic pregnancy has ruptured. The portion of the uterus was sutured in a double layer because its integrity seems to be very important to prevent uterine rupture in subsequent pregnancies, and often, hysterectomy.8

Baldawa et al. had a similar patient as in our case who was admitted to the emergency obstetrical room in a state of hypovolemic shock. In exploratory laparatomy, they found right lateral wall rupture uterus and ~14 weeks fetus with the placenta lying freely in the peritoneal cavity. Alves et al. had a patient where interstitial pregnancy was diagnosed at 6 weeks of pregnancy, located close to the right cornual portion of the uterus. Prenatal monitoring was carried out until birth at 36 weeks gestation with uterine conservation. Ultrasound scan and magnetic resonance imaging were realized to confirm the diagnosis and to monitor the evolution.

Conclusion

Angular pregnancy is a rare form of ectopic pregnancy with different clinical management, outcomes, and maternal mortality. Angular pregnancy corresponds to a viable intrauterine pregnancy that is implanted in one of the lateral angles of the uterine cavity, medial to the

uterotubal junction, by endovaginal sonography at 6 weeks gestation. The options to treat angular pregnancy are multiple and vary depending on the time of diagnosis, risk factors, choice of patient, and desire for future pregnancies. A conservative approach is possible when the diagnosis is made early.

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Conflict of Interest: None declared.

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