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Spontaneous dichorionic-diamniotic twins in a noncommunicating uterine horn: A case report

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ABSTRACT

Background: We present a rare case of a dichorionic-diamniotic twin pregnancy in a noncommunicating rudimentary uterine horn diagnosed prior to rupture at 12 weeks of gestation.
Case: A 33-year-old woman with history of two prior spontaneous abortions presented with a spontaneously conceived dichorionic-diamniotic twin pregnancy. Routine first-trimester screening ultrasound detected an extrauterine twin pregnancy. The patient was admitted for observation and treatment planning. Magnetic resonance imaging (MRI) of the pelvis led to the radiologic diagnosis of suspected abdominal ectopic pregnancy. Exploratory laparotomy led to an intraoperative diagnosis of twin pregnancy within a rudimentary uterine horn, which was removed without incident.
Conclusion: This is a rare case of a twin pregnancy contained in a noncommunicating rudimentary uterine horn. The presence of this horn was not detected on ultrasonography or MRI.

1. Introduction

Uterine anomalies are estimated to occur in about 5.5% of the at-large population with increasing prevalence among women with infertility and history of miscarriage, at 8.0% and 13.3%, respectively. [1] The prevalence of unicornuate uterus is estimated at 0.1%. [1] 74–90% of women with a unicornuate uterus have some kind of rudimentary uterine horn. Among these, 55% of rudimentary horns are noncommunicating and contain functional endometrium. [2] Ectopic pregnancy can occur in these noncommunicating uterine horns at an estimated incidence of 1 in 76,000. [2] These pregnancies have a uterine rupture rate of 50% and a maternal mortality rate of 5.7%. [2,3]

2. Case

A 33-year-old woman with history of two prior spontaneous first-trimester abortions presented for routine first-trimester screening ultrasound at 12 weeks of gestation with a spontaneously conceived dichorionic-diamniotic twin pregnancy. At presentation, she was asymptomatic and denied abdominal pain, vaginal bleeding, hematuria, hematochezia, chest pain, dyspnea, and syncope. She denied any history of renal anomaly, dysmenorrhea, endometriosis, or infertility. Past medical and surgical history were noncontributory. Her ultrasound was notable for an empty-appearing uterus and presence of gestational sac, placentas, and viable dichorionic-diamniotic twins in the posterior cul-de-sac. Due to these findings, patient was immediately admitted for urgent MRI of the pelvis and operative planning. The MRI scan showed an empty uterine cavity and a pregnancy in the posterior cul-de-sac ($8.4 \times 8.6 \times 8.3$ cm) with no overlying myometrium (Fig. 1).

The patient was admitted and consultations with gynecologic oncology, reproductive endocrinology/infertility, and vascular surgery services were obtained. Surgical consents were broad and included provisions for possible bowel or bladder resection and reconstruction, vascular reconstruction, and possible local or systemic methotrexate administration in addition to more conventional consent for exploratory laparotomy via vertical midline incision, removal of ectopic pregnancy, fallopian tube, and ovary.

The patient underwent exploratory laparotomy, which revealed dichorionic-diamniotic twin pregnancy entirely contained within a noncommunicating rudimentary right uterine horn. The right fallopian tube was found to be inextricably involved with the rudimentary horn, so the rudimentary horn, right fallopian tube, and ectopic pregnancy were removed en bloc and without incident. The right ovary was left in situ. Inspection of the pelvis showed a left unicornuate uterus and normal left ovary and fallopian tube. There was no evidence of remnant placenta, so methotrexate was not indicated. The patient's

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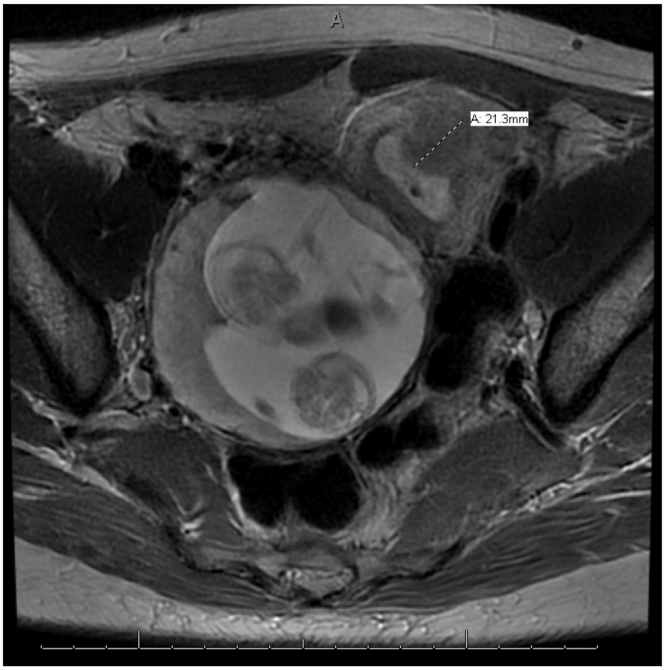

Fig. 1. Pelvic MRI of dichorionic diamniotic twins at 12 weeks of gestation with displaced uterus and no evidence of surrounding myometrium.

abdomen was then closed. She had an uncomplicated postoperative course and was discharged home on postoperative day two.

Final pathology showed placenta accreta surrounded by myometrial tissue. There was no placental tissue at the myometrial resection margin. The fetuses did not undergo genetic analysis, so the zygosity and implantation of this pregnancy are unknown. The patient was discharged home with recommendations for outpatient reproductive endocrinology/infertility and maternal-fetal medicine consultations and for renal ultrasound, given the new diagnosis of Mullerian anomaly. Ultimately, her renal ultrasound showed no anomaly.

3. Discussion

While most unicornuate uteri have a rudimentary horn, there are a few reports of twin gestations implanting in these patients. [4,5] We are not aware of any reports prior to this of both embryos of a twin gestation implanting in the rudimentary horn. In the present case, dichorionic diamniotic twins were found in the rudimentary horn in an asymptomatic patient at 12 weeks of gestation. The estimated incidence of twins in the setting of a unicornuate uterus or rudimentary horn is approximately 1 in 10 million gestations. [5]

As most women with Mullerian anomalies conceive without knowledge of that anomaly, it is important that sonographic providers have a low threshold to assess not only fetal viability and early anatomy but also to confirm normal intrauterine implantation and uterine anatomy. In our case, the "normal" (later found to be unicornuate) uterus was clearly seen anteriorly without a gestational sac, and there was a large posterior cul-de-sac mass with the dichorionic diamniotic twins. Initial differential diagnosis included abdominal pregnancy versus uterine anomaly. The suspicion for abdominal pregnancy was highest as there was not clear uterine tissue around the pregnancy, and neither was there a clear connection between uterus and mass in the cul-de-sac. MRI of the pelvis yielded a radiologic diagnosis of abdominal pregnancy due to absence of detectable myometrial tissue surrounding pregnancy. However, on imaging, the pregnancy did appear to be spherical and well-contained, which, in retrospect, was suggestive of the eventually diagnosed rudimentary horn.

Counseling for her surgical care was informed by this diagnosis and included possible bladder or bowel resection, given the concern for invasive placentation. Contingency planning also included possible local or systemic methotrexate administration if portions of placenta were retained. Intraoperatively, the pregnancy was found to be contained entirely within a rudimentary uterine horn, which was removed without incident. If this diagnosis had been made preoperatively, patient counseling would have been considerably different.

The ultrasonographic sensitivity for detecting rudimentary horn pregnancy (RHP) is low, at 29–33%. [6] Criteria have been proposed for sonographic diagnosis of RHP: (1) a pseudopattern of an asymmetrical bicornuate uterus; (2) absent visual continuity tissue surrounding the gestational sac and cervix; and (3) presence of myometrial tissue surrounding the gestational sac. [7,8] In this case, the first and third criteria were absent. We hypothesize that this absence may be due to a twin gestation, which could have caused earlier thinning of myometrium and more substantial imbalance in the size of the uterine horn. Transvaginal imaging was not performed as the diagnosis of abnormal pregnancy was clear, but, in retrospect, this may have allowed for better evaluation of the second criterion.

In summary, we report a case of a patient with dichorionic diamniotic twins in a rudimentary horn. Preoperative diagnosis of abdominal pregnancy led to complex multidisciplinary planning and counseling. Reviewing the literature guiding diagnosis of RHP by ultrasound, we did not have two of the three criteria for this diagnosis but would certainly recommend consideration of transvaginal ultrasound and other views to assess the presence or absence of continuity tissue between the gestational sac and cervix.

Contributors

Megan E. Ross drafted the manuscript
Stephen Scott contributed to review and editing of the manuscript
Kian Behbakht contributed to review and editing of the manuscript
Teresa Harper contributed to review and editing of the manuscript
All authors contributed equally to creation of this manuscript

Conflict of interest

The authors declare that they have no conflict of interest regarding the publication of this case report.

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Patient consent

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