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Fertility after treatment of a noncommunicating rudimentary horn pregnancy: A case report

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ABSTRACT

Rudimentary horn pregnancies are rare but are associated with high mortality and morbidity. The diagnosis can be difficult as it may be challenging to distinguish a rudimentary horn pregnancy from an intrauterine pregnancy on ultrasound. Magnetic resonance imaging can often be used to confirm a rudimentary horn pregnancy. When a second-trimester rudimentary horn pregnancy is diagnosed, surgical intervention should be performed to avoid uterine rupture and hemoperitoneum. The correct diagnosis and management of rudimentary horn pregnancies help to preserve the fertility of younger patients.

This case report describes a second-trimester rudimentary horn pregnancy that was diagnosed by ultrasound and magnetic resonance imaging. It was then surgically resected via laparotomy. This patient maintained her fertility and was able to conceive naturally, leading to an uncomplicated term pregnancy.

1. Introduction

Ectopic pregnancy occurring in the rudimentary horn of a unicornuate uterus is extremely rare, ranging from 1 in 75,000 to 150,000 pregnacies [1–3]. However, there is significant mortality associated with rudimentary horn pregnancies, including a 50% risk of uterine rupture, which can result in hemoperitoneum and even maternal death [4]. Recent innovations in diagnostic imaging with 3D ultrasound and magnetic resource imaging (MRI) have improved the diagnosis of rudimentary horn pregnancy. Diagnosis prior to rupture can be critical in facilitating timely treatment of the rudimentary horn. There are only a handful of reported cases involving repeat pregnancies and deliveries following surgical resection of a rudimentary horn [3,5,6]. Here we present a case where the patient had two pregnancies and one term delivery following resection of a non-communicating rudimentary horn pregnancy.

2. Case Presentation

A 21-year-old gravida 5 para 1 (term vaginal home birth) woman with a history of a bicornuate uterus secondary to possible exposure to diethylstilbestrol in utero presented to the emergency room as a transfer from an outside facility due to concern for ectopic pregnancy. She had an obstetrical ultrasound performed at an outside facility the day prior due to her history of pregnancy in the setting of a bicornuate uterus. The ultrasound showed a gestational sac posterior to the uterus with a live pregnancy, with measurements indicating 16 weeks 2 days of gestation. The patient was referred to a maternal fetal medicine obstetrician and ultrasound was repeated and suspicious for a 16-week left cornual pregnancy (Fig. 1). She was referred to the emergency room for additional evaluation and imaging. Upon arrival at the emergency room, the patient was immediately evaluated and voiced no complaints. She specifically denied any abdominal pain or vaginal bleeding.

On physical examination, she appeared stable and all of her vital signs were within normal limits. Her abdomen was soft, nontender, nondistended, and negative for rebound.

A stat MRI of the pelvis without contrast was obtained given her ultrasound findings. The MRI showed a left adnexal ectopic pregnancy appearing to be in the second trimester. The gestational sac was surrounded by a wall that had the same signal intensity on MRI as the myometrium (Fig. 2). Upon further questioning, the patient reported a history of bicornuate uterus. However, the endometrium that lined the gestational sac did not appear to be continuous with the uterine endometrium.

Due to concern for eventual rupture of the uterus leading to intra-abdominal hemorrhage, operative intervention with diagnostic

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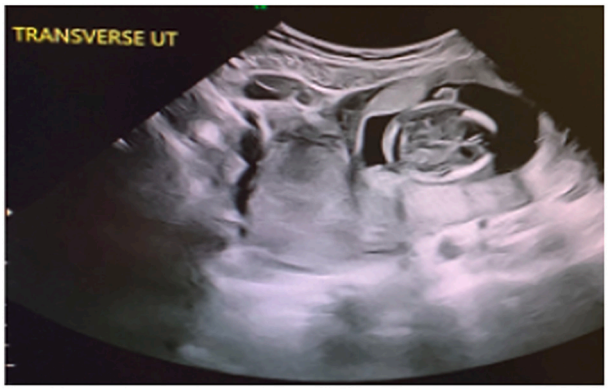

Fig. 1. Transabdominal ultrasound image demonstrating gestational sac outside of the uterine body.

laparoscopy and possible laparotomy was discussed. Upon laparoscopic survey of the pelvis, a non-ruptured ectopic pregnancy was noted in a left rudimentary uterine horn. It was difficult to visualize the true uterus via the laparoscopic approach. At that time, the procedure was converted to a laparotomy via a Pfannenstiel skin incision. With more thorough visualization of the uterus and ectopic pregnancy, a right-sided unicornuate uterus attached to a normal fallopian tube and ovary was noted (Fig. 3). On the left side, there was a small intact noncommunicating rudimentary uterine horn containing an approximately 16-week ectopic pregnancy attached to the left round ligament and normal-appearing left fallopian tube and ovary (Fig. 3, Fig. 4).

The left round ligament and utero-ovarian ligament were isolated and ligated. Dilute vasopressin was then injected along the border of the fibrous connection between the unicornuate uterus and rudimentary horn. A straight zeppelin clamp was placed across the connection between the uterus and rudimentary horn. The rudimentary horn containing the pregnancy along with the left fallopian tube was resected from the left side of the main unicornuate uterus using a scalpel. No communication was noted between the uterus and rudimentary horn. The defect on the left side of the main uterus was closed in two layers. The first layer consisted of multiple figure-of-eight 0-Vicryl sutures; the Zeppelin clamp was removed prior to securing the sutures. A second imbricating layer was then completed with 2–0 Monocryl. Meticulous hemostasis was achieved prior to closure (Fig. 5). The pelvis was irrigated and the peritoneum, fascia, and skin were closed. Total

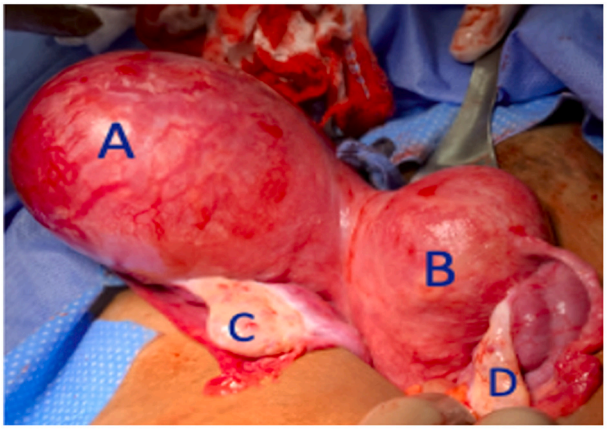

Fig. 3. Cranial view of pelvic anatomy. Left non-communicating uterine horn pregnancy (A), unicornuate uterine horn (B), normal-appearing left ovary (C) and normal-appearing right ovary (D).

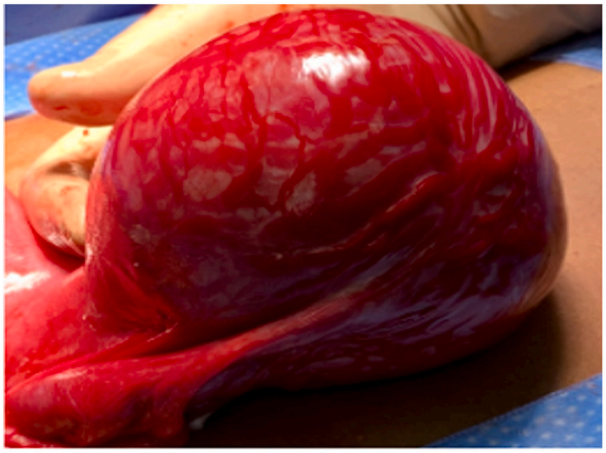

Fig. 4. Detailed view of pregnancy within rudimentary horn with thin and vascular myometrial outpouching noted.

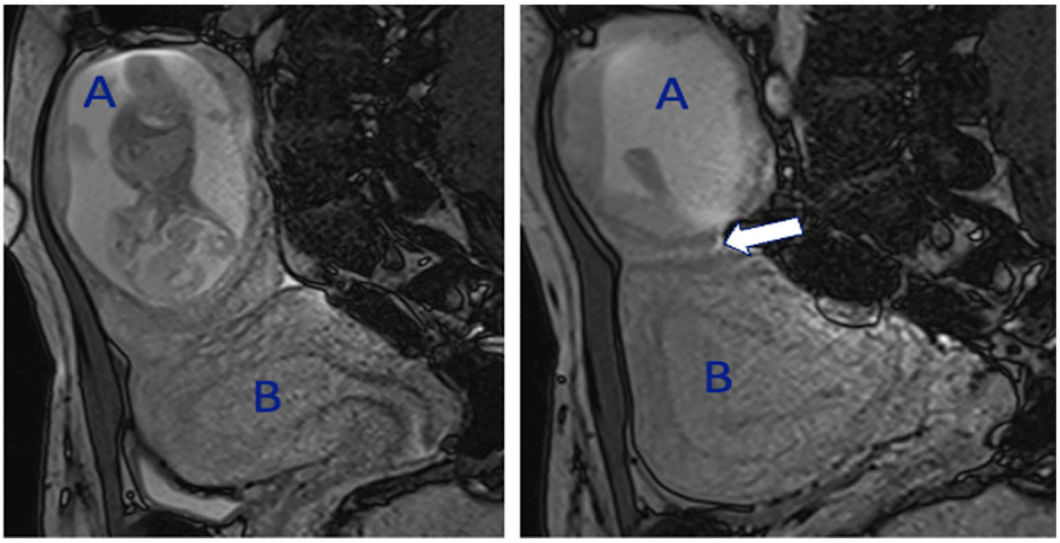

Fig. 2. Sagittal MRI demonstrating pregnancy within a noncommunicating horn (A), along with a unicornuate uterine body (B). Arrow demarks myometrial separation between two cavities.

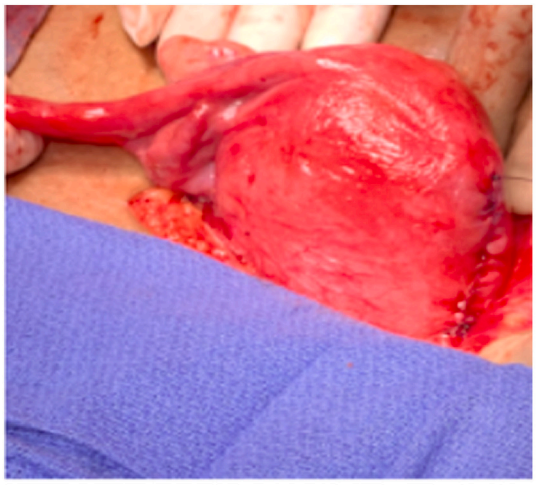

Fig. 5. Postoperative view of uterus with removal of pregnancy and rudimentary horn. Unicornuate uterus with normal right fallopian tube and ovary.

estimated blood loss from the procedure was 150 cc. The patient had an uncomplicated post-operative course and was discharged to home on post-operative day two.

The final histopathology report of the demonstrated a fetus with a crown-rump length of 12 cm; the resected rudimentary horn measured 13x10x4.5 cm with a highly vascularized serosal surface; and placenta firmly adherent to the inner aspect.

Three months after laparotomy, the patient proceeded to conceive naturally. She had an uncomplicated pregnancy with an uncomplicated low transverse cesarean section at 38 weeks of gestation. 15 months after rudimentary horn resection, and 12 months after the cesarean section, she had conceived again naturally and was currently 24 weeks pregnant.

3. Discussion

Approximately 1 in 4000 females is found to have a unicornuate uterus, an anomaly caused by either partial or complete failure of development of one of the Mullerian ducts [7]. A partial development of the Mullerian ducts results in a rudimentary horn [4]. Pregnancy occurring in the rudimentary horn of a unicornuate uterus is extremely rare, ranging from 1 in 75,000 to 150,000 [1–3]. The cavity of the rudimentary horn can be communicating or non-communicating (about 85% are non-communicating). For pregnancy to occur within a non-communicating rudimentary horn, there must be transperitoneal migration of sperm from the contralateral fallopian tube [3,4]. Rudimentary horn pregnancies are associated with significant maternal morbidity and mortality; they carry a 50% risk of uterine rupture, which can result in devastating hemoperitoneum and even death [4]. Given the significant risk of life-threatening complications, early diagnosis and management of a rudimentary horn pregnancy are critical.

Historically, diagnosis of rudimentary horn pregnancy is difficult and often occurs incidentally during laparotomy when the gestational horn ruptures [8]. With recent advances in diagnostic imaging modalities, ultrasound and magnetic resource imaging (MRI) can be used to diagnose rudimentary horn pregnancies prior to rupture. The sensitivity of ultrasound alone has been noted to be low, at around 26%, with only 14% of cases diagnosed prior to clinical symptoms [9,10]. The addition of MRI and 3D reconstruction of the ultrasound can improve the diagnostic sensitivity by utilizing multiplanar images to visualize the exact Mullerian anomalies and confirm the diagnosis of rudimentary horn diagnosis prior to rupture [3,10].

Management of non-communicating rudimentary horn after diagnosis is the excision of the pregnant horn [8]. Laparoscopy is the standard treatment in hemodynamically stable patients; however, the threshold to convert to laparotomy should be low, given the typical hypervascularity of rudimentary horn pregnancies [3,5,10]. Recently, there have been case reports of combination medical and surgical management for rudimentary horn pregnancies [5]. Ueda et al. demonstrated methotrexate, intracardiac potassium chloride, or lidocaine injections as effective medical treatment options prior to surgery in order to decrease hypervascularity and overall complications [5].

In this case, resection of the rudimentary horn pregnancy in the second trimester was not feasible with laparoscopy. The conversion to laparotomy allowed for resection of the uterine horn but also preservation the remaining unicornuate uterus. The patient was able to conceive natural shortly after the resection and delivery via cesarean section at term. She was further able to conceive for a second time, with an ongoing pregnancy.

There are few case reports of pregnancies and deliveries after treatment of rudimentary horn pregnancy [3,5,6]. A majority of these cases involved pregnancy after laparoscopic resection of uterine horn. Our case demonstrates that early diagnosis and timely surgical management of a second-trimester rudimentary horn pregnancy are crucial steps in supporting future fertility for women of reproductive age.

Contributors

Salina Zhang drafted the case report.
Alessandra Lamari revised the case report.
Edward Ferris acquired the key data used in the case report and was involved in care of the patient as the primary surgeon.
Priya Maseelall was responsible for the conception of the case report.
All authors approved the final article to be submitted.

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Patient consent

Obtained.

Provenance and peer review

This article was not commissioned and was peer reviewed.

Conflict of interest statement

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

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