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Tubal pregnancy in a case of unicornuate uterus with contralateral streak ovary and renal agenesis: A case report

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ABSTRACT

A 24-year-old woman presented to the emergency department with symptoms consistent with an ectopic pregnancy. Ultrasonographic evaluation reported no evidence of intrauterine pregnancy, a heterogenous mass in the left adnexa, and normal-appearing bilateral ovaries. The right kidney was imaged, though the left kidney was not visualized. Beta-HCG was reported below the discriminatory zone. Two days later, her ultrasound remained unchanged, and beta-HCG had not risen appropriately. Diagnostic laparoscopy revealed a dilated right fallopian tube with products of conception exuding from the fimbriated end. Laparoscopically, the patient's pelvic anatomy was found to be abnormal. There was no left fallopian tube and the uterus appeared unicornuate with absent left horn. A left streak ovary was present superior to the pelvic brim in the pericolic gutter, though the right ovary appeared normal and in the appropriate position. At follow-up, hysterosalpingography showed a right-sided unicornuate uterus without septation, a small tubular endometrial cavity, and a patent right fallopian tube. Defects of unification of the Mullerian ducts, such as unicornuate uterus, do not reduce the ability to achieve pregnancy, but do increase the risk of adverse outcomes and fetal malpresentation.

1. Introduction

During embryologic development, the bilateral mesonephric (Wolffian) ducts serve as scaffolding for the development and fusion of the inferior bilateral paramesonephric (Müllerian) ducts. This fusion and reabsorption of the medial walls of the paramesonephric ducts yield a primordial uterus. Additionally, the mesonephric ducts form ureteric buds, which develop into the collecting tubules, calyces, renal pelvis, and ureters of the adult kidney. The ureteric buds mature secondary to stimulation from the surrounding metanephric mesenchyme. This surrounding tissue develops into the nephrons of the adult kidney. Therefore, unilateral renal agenesis with uterine anomaly can result from injury to or absence of one mesonephric duct, the embryologic structure that links these organ system anomalies [1,2].

Müllerian anomalies, renal agenesis, and ovarian anomalies have been described as co-occurrent in only a few prior case reports [3–5]. The following case describes the diagnosis and clinical management of a tubal ectopic pregnancy in a patient with the rare anatomic constellation of a unicornuate uterus with contralateral renal agenesis and streak ovary. Clinical decision making carried important implications for future spontaneous pregnancies.

2. Case Presentation

A 24-year-old woman presented to the emergency department with vaginal bleeding and pelvic pain. Three days prior, she had experienced a 30-min episode of sharp pain in the right abdomen and flank, radiating down the right leg. While the pain resolved, feelings of pelvic fullness persisted. On the morning of presentation, right-sided pain awoke the patient from sleep, accompanied by dysuria, lightheadedness, and nausea. Physical exam revealed tenderness in the right lower quadrant and suprapubic area. Pelvic and bimanual exam showed blood in the vaginal vault, closed cervical os with active bleeding, and cervical motion tenderness.

The patient reported attempts to conceive for several months. Her history was significant for two miscarriages: one three years prior and another five months prior, thought to be approximately at seven weeks of gestation. Neither required medical or surgical intervention. The patient also reported antibiotic-treated chlamydia three years previously, but no additional medical or surgical history.

In the emergency department, labs showed a positive urine pregnancy test and a serum beta-HCG of 1860 IU/L. Point-of-care retroperitoneal and transvaginal ultrasound showed a heterogenous mass in the

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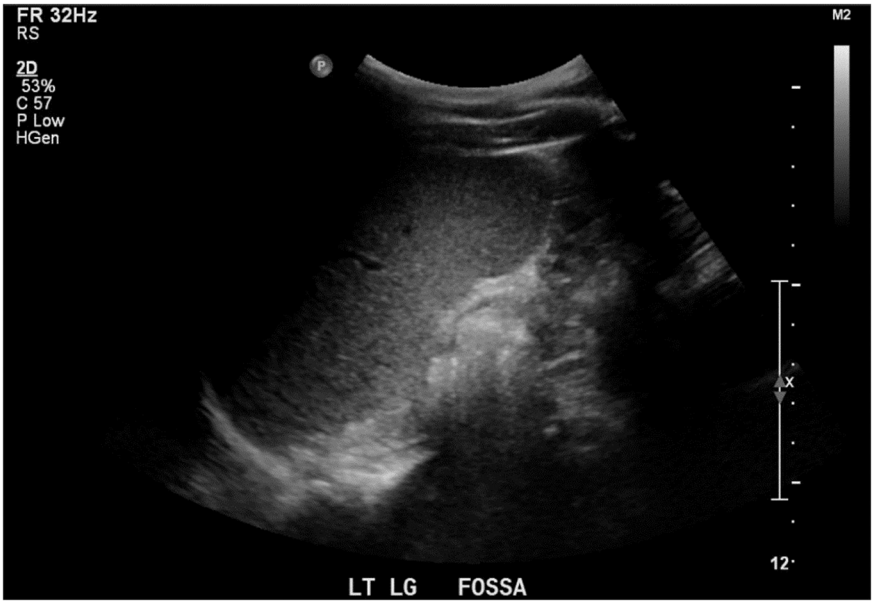

Fig. 1. Ultrasound of the left renal fossa showing the patient's spleen and absent kidney.

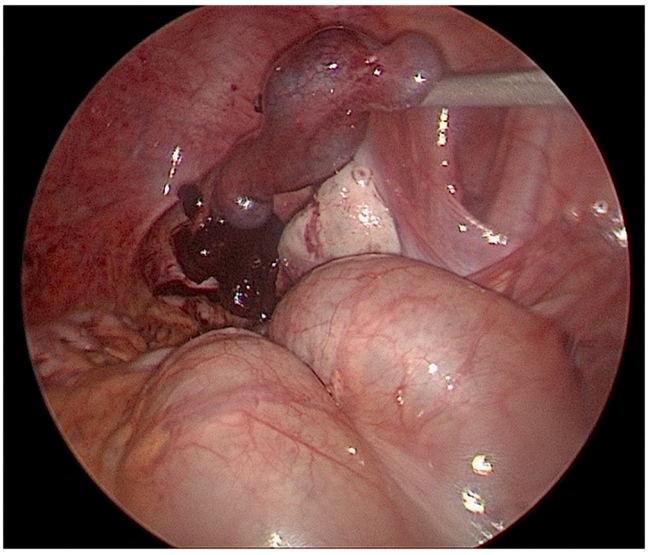

Fig. 2. Laparoscopic image showing dilated right fallopian tube with products exuding from the fimbriated end and normal right ovary.

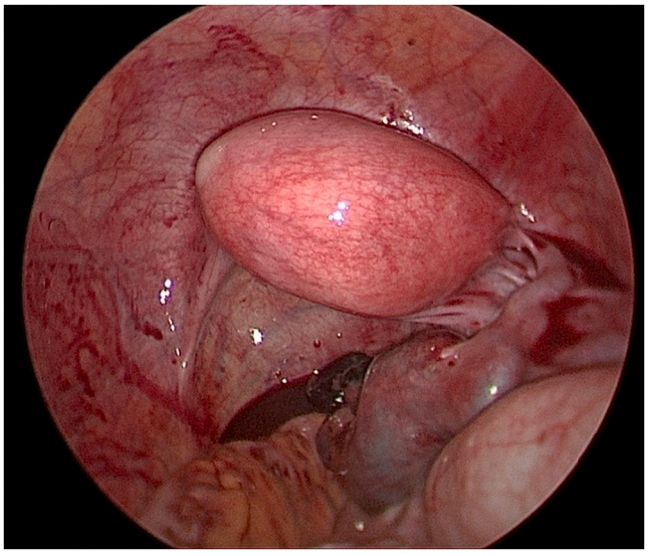

Fig. 3. Laparoscopic image showing unicornuate uterus deviated to the right, absent left horn, and absent left fallopian tube.

left adnexa, concerning for ectopic pregnancy, with a moderate amount of fluid in the pelvic cavity, a small amount of fluid in the endometrial canal, and no evidence of an intrauterine pregnancy. The report noted normal-appearing bilateral ovaries and right kidney, though the left kidney was not visualized [Fig. 1]. The patient was diagnosed with a suspected ectopic pregnancy; however, an intrauterine pregnancy could not be excluded at this time as the beta-HCG level was below the discriminatory zone [6]. She was otherwise stable and discharged with a plan to follow up in two days.

In the clinic two days later, her serum beta-HCG had increased to 2198 IU/L and transvaginal ultrasound was repeated with no change noted. Pregnancy was dated at four weeks and six days based on last menstrual period. Given her persistent pelvic pain and concern for ruptured ectopic pregnancy, medical management was deferred and the patient was taken for diagnostic laparoscopy. Intraoperative findings included: 1) dilated right fallopian tube with products of conception exuding from fimbriated end, contrary to ultrasound findings indicating left-sided pathology [Fig. 2]; 2) normal right ovary [Fig. 2]; 3) suspected unicornuate uterus with absent left horn and absent left fallopian tube [Fig. 3]; 4) left streak ovary superior to the pelvic brim in left pericolic gutter [Fig. 4]; 5) vagina without septum; and 6) mild endometriosis in the anterior/posterior cul de sacs. A salpingostomy with subsequent Floseal application was performed to preserve the patient's only fallopian tube and allow for future spontaneous pregnancy [Fig. 5]. Pathologic analysis of extracted material revealed gestational trophoblastic tissue without chorionic villi.

Post-operatively, the patient received oral contraceptive pills for endometriosis and was scheduled for follow-up. She recovered without complication.

A hysterosalpingogram was scheduled to evaluate the uterine cavity for septate vs. unicornuate uterus and to ensure fallopian tube patency. This revealed a right-sided unicornuate uterus without septation, a small, tubular endometrial cavity, and a patent right fallopian tube.

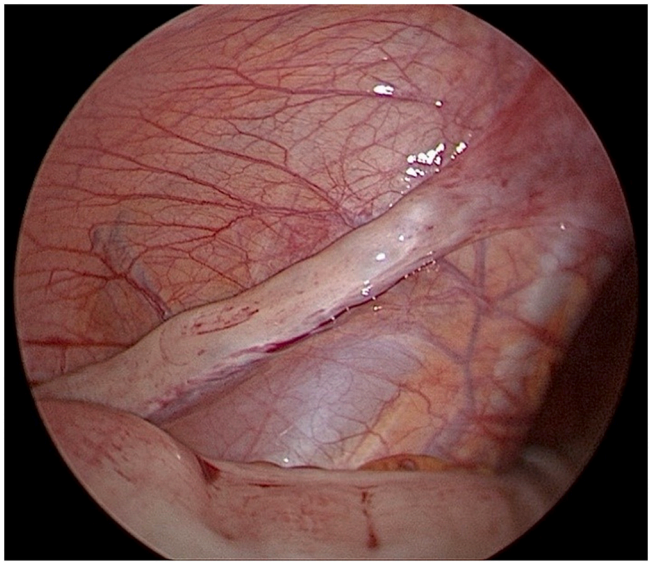

Fig. 4. Laparoscopic image showing left streak ovary superior to the pelvic brim in left pericolic gutter.

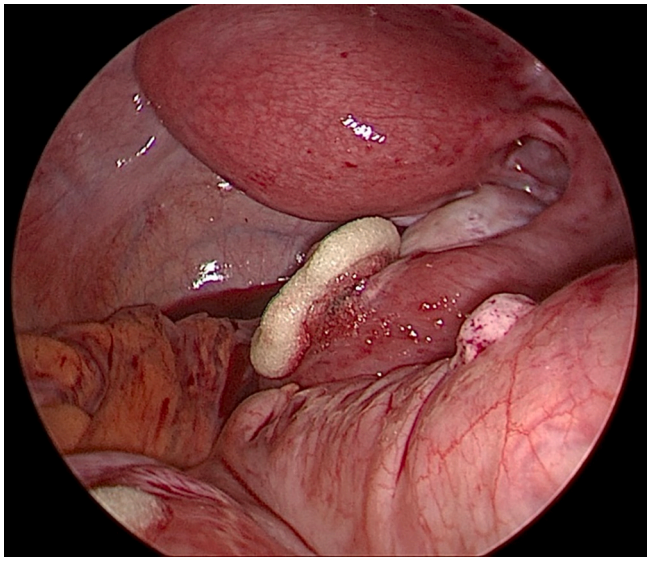

Fig. 5. Laparoscopic image showing completed right salpingostomy and subsequent Floseal application.

3. Discussion

The presence of uterine malformation is estimated to be 5.5% in the general population and 8% in women with infertility [7]. Of all uterine malformations, unicornuate uterus accounts for 2.5–13.2% [7]. According to a review by Chan et al. [8], defects of unification of the Mullerian ducts, such as unicornuate uterus, do not reduce the ability to achieve pregnancy, but do increase the risk of adverse outcomes such as miscarriage and preterm delivery. The patient in this case experienced both ectopic pregnancy and miscarriages. According to a review by Reichman et al. [9], pregnant patients with unicornuate uterus had the following pregnancy outcomes:

- 49.9% live-birth
- 24.3% first-trimester miscarriage
- 20.1% preterm delivery
- 10.5% intrauterine fetal demise
- 9.7% second-trimester miscarriage
- 2.7% ectopic pregnancy

The most common reasons for adverse outcomes included decreased uterine muscle mass, abnormal uterine blood flow, and cervical insufficiency [9]. Additionally, all uterine anomalies are associated with increased risk of fetal malpresentations [8]. With future pregnancies, this patient should be considered high risk, given her history of adverse outcomes and genitourinary anomalies.

4. Conclusion

Uterine malformations are not uncommon and this case highlights several points to consider for clinical practice. Before proceeding with salpingectomy for ectopic pregnancy, it is important to examine pelvic anatomy for other abnormalities. In cases of tubal pregnancies where the contralateral fallopian tube is abnormal or absent, clinicians should consider salpingostomy rather than salpingectomy in an attempt to preserve the patient's ability to achieve a future spontaneous pregnancy. Additionally, as seen in this case, structures in the pelvis can cross the midline and be mislabeled on imaging. Therefore, when consenting for surgery, leave flexibility in the consent as the laterality may be different than anticipated.

Contributors

Lauren H. Barnts was involved in this patient's follow-up outpatient care and contributed to the writing of this case report.

Taylor J. Kratochvil was involved in this patient's acute surgical care and contributed to the writing of this case report.

Sylvia J. Ziegenbein was involved in this patient's acute surgical care, the patient's follow-up outpatient care, and contributed to the editing of this case report.

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Patient consent

The patient consented to the publication of this report and accompanying images.

Provenance and peer review

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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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