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Non-communicated rudimentary uterine horn pregnancy and uterine rupture: A case report

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Abstract

Rudimentary uterine horn pregnancy is a rare and serious type of ectopic pregnancy and is hard to diagnose due to a lack of typical clinical symptoms at the early stage. A 35-year-old woman who was17weeks pregnant and had a complicated history of infertility came to our hospital complaining of abdominal pain without vaginal bleeding. Computed tomography scan after 12 hours showed that her pregnancy was in the small cavity of a rudimentary uterine horn, which had ruptured. Noncommunicating rudimentary uterine horn pregnancy is an extremely rare form of ectopic gestation, and its diagnosis and management remain challenging. Nevertheless, physician awareness of various forms of unicornuate uteri and rudimentary uterine horn can save lives.

Keywords

Rudimentary uterine horn pregnancy, uterine rupture, laparotomy, risk management, case report

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Introduction

Rudimentary uterine horn (RUH) pregnancy is a rare and serious type of ectopic pregnancy and is very hard to diagnose due to a lack of typical clinical symptoms at the early stage.[1] A "rudimentary" uterine horn can exist in relation to a rare genetic condition resulting in a unicornuate uterus, in which normal differentiation of only one Müllerian duct occurs, and formation of a rudimentary horn, which does not communicate with the uterus.[2] This condition is related to higher rates of infertility, recurrent pregnancy loss, prematurity, and other obstetric complications, although in many patients they may remain asymptomatic.[3–5] A pregnancy within a noncommunicating rudimentary horn is an extremely rare form of ectopic gestation; however, the natural course of an RUH pregnancy is rupture which can threaten a woman's life. This case study aims to describe an occurrence of this rare pregnancy within a complicated infertility history, to discuss the clinical management in an emergency situation.

Case report

Chief complaint

A 35-year-old woman who was 17 weeks pregnant presented at the Emergency Department of our hospital complaining of an abdominal pain.

History of present illness

Her pregnancy had been confirmed 4 weeks ago when she went to a doctor with uncomfortable digestion that is a reflection of morning sickness. The patient's symptoms on the day of her Emergency Department visit had started suddenly 2 h after defecating without vaginal bleeding.

History of this patient

The patient had a history of infrequent menstruation of 4–5 days duration, approximately every 6 months, and a complicated infertility history. She reports having undergone a left salpingectomy for tubal ectopic pregnancy 12 years ago in another clinical center. In the next few years, because of secondary infertility, as well as diagnosed poly cystic

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ovarian syndromes (PCOS), she tried in vitro fertilization–embryo transfer (IVF-ET) treatments. The first 6 treatments failed, but at the seventh treatment, 4 years ago, she successfully became pregnant. However, the patient had a spontaneous abortion at 12 weeks' gestation. At that point, the patient stopped IVE-ET efforts and did not use any contraceptives afterward.

Physical and laboratory examination

The patient's abdomen was rigid with rebound tenderness, especially on the left, where a tender mass was palpated. The mass seemed to be connected with the uterus. Differential diagnoses we considered included a threatened abortion or a second lower placenta. Blood analysis revealed a white blood cell (WBC) count of 15.22×10^9/L ($3.5–9.8 \times 10^9$/L), Hb of 116 g/L (115–150 g/L), and serum amylase of 41.9 IU/L (35–135 IU/L).

Imaging examinations

An abdominal ultrasound examination showed a viable 17-week-sized fetus who has a fetal heart rate (FHR) of 155/min with an maximum vertical pocket (MVP)[6] of 6.8 cm. There was no free fluid in the abdominal or pelvic cavity. The placenta seemed low lying with signs of its edge near the cervical internal orifice.

Further diagnostic work-up

During the following 12 h, the patient had aggravated abdominal distention. Blood analysis showed a WBC count of 13.23×10^9/L ($3.5–9.8 \times 10^9$/L), Hb of 98 g/L (115–150 g/L), and C-reactive protein (CRP) of 33.91 mg/L. An abdominal ultrasound examination was repeated, which then showed that the FHR was 163/min, but there was no amniotic fluid for the fetus and pelvic placenta. Low echoic area of approximately 2.0 cm was noted between the patient's liver and kidney.

Evolving differential diagnosis

At this point, the patient's symptoms did not fit with those of threatened abortion, which most often include vaginal bleeding and signs of inflammation, which were not present or supported by the amylase or other relevant lab results. Although there was no imaging evidence to support an ovarian cyst, we could not entirely exclude ovarian cyst or tumor rupture based on the physical examination. New questions had arisen from comparative ultrasound examination 12 h apart that is the amniotic fluid reduction and appearance of ascites. Uterine rupture was suspected but seeing an emergency situation developing this night, a computed tomography (CT) scan was ordered to provide further clarification.

Further diagnostic work-up

There showed an increase in density on CT for pelvic cavity as well as the uterine rupture location (Figure 1).

Treatment and final diagnosis

The patient underwent an emergency laparotomy, revealing about 400 mL of blood and blood clots in the pelvic cavity, predominantly on the left. We identified the source of the bleeding to be a thin RUH which has ruptured and which was still bleeding heavily. Besides the crevasse, there was little dead fetus and its affiliations (Figure 2). The rudimentary horn did not communicate with the unicornuate uterus. We did residual horn hysterectomy and hysteroplasty for this patient and she received 4 U erythrocyte suspension and 600 mL plasma during the surgery.

Final diagnosis was confirmed during the surgery: noncommunicated rudimentary uterine horn pregnancy and uterine horn rupture. This case is also shown in a schematic figure (Figure 3).

Outcome

Her physical course was uneventful, although she was experiencing severe acute grief, and she was discharged 7 days after surgery.

Discussion

A unicornuate uterus results from the normal differentiation of only one Müllerian duct and is observed in 0.4% of women. Müllerian abnormalities are found in 0.17% of fertile women and 3.5% of infertile women.[7–9] At the same time, approximately 84% of unicornuate uteri have a contralateral rudimentary horn.[10] Partial development of the other duct results in a rudimentary horn that is either with cavity, communicating with the uterus (type A1a) or with cavity, not communicating (type A1b), or is without a cavity (type A2).[11,12]

Unicornuate uteri are related to higher rates of infertility, recurrent pregnancy loss, prematurity, and other obstetric complications, although in many patients they may remain asymptomatic.[3] As one type of ectopic pregnancy, the incidence of rudimentary horn pregnancy is only 1/140,000 to 1/75,000, which is about 10 times lower than the incidence of celiac pregnancy.[10] The true statistics of these conditions remains elusive, varying from 1 in 10 to 1 in 1600 in various reports.[3] The estimated 1 in 76,000 pregnancies that occur within a noncommunicating rudimentary horn, however, confirms that it is an extremely rare form of ectopic gestation.[1] Pregnancy in a noncommunicating uterine horn can be explained by the phenomenon of transperitoneal migration of either sperm or the fertilized ovum from the patent contralateral fallopian tube to the rudimentary horn.[13] In our

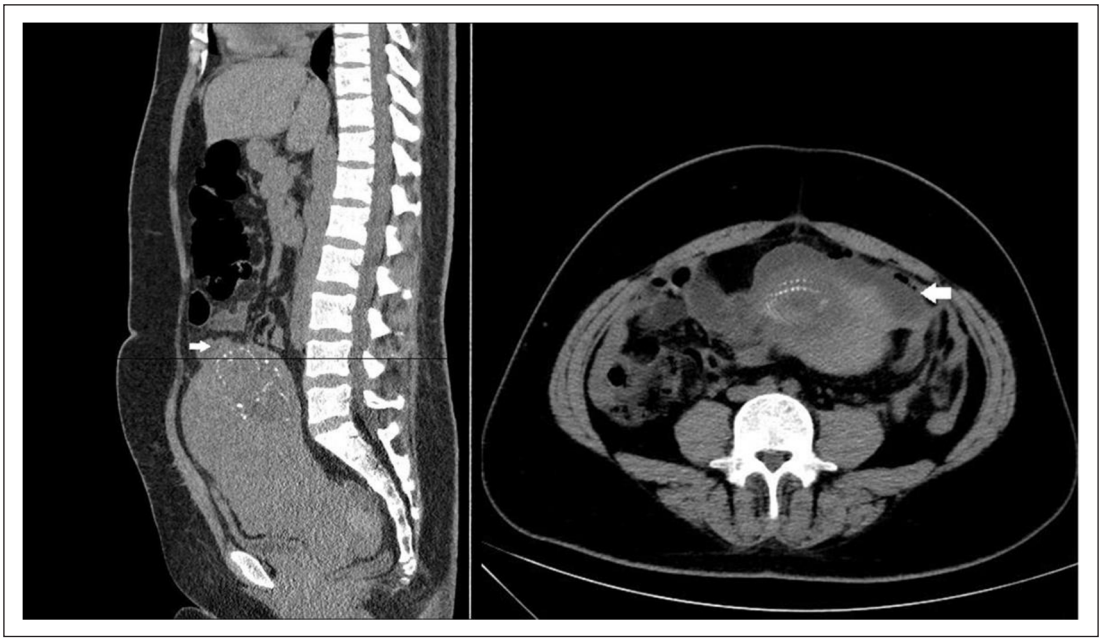

Figure 1. Vertical section and transverse section image of abdominal CT scan. CT scan detected the fetus in uterus and increased density in the pelvic cavity, as well as the uterine rupture location (marked by white arrow).

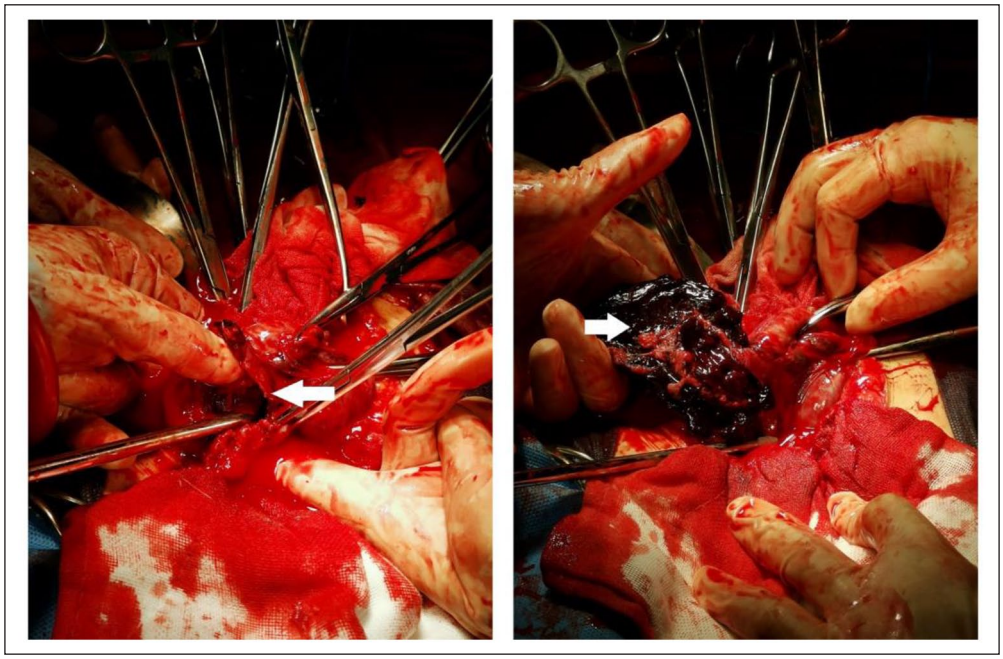

Figure 2. The patient underwent an emergency laparotomy. We identified the source of the bleeding to be a thin rudimentary uterine horn which has ruptured and which was still bleeding heavily. Besides the crevasse, there was little dead fetus and its affiliations.

case, the horn cavity seemed to have been completely separate from the unicornuate uterus and leaves to mystery how exactly the fertilized ovum got into the rudimentary cavity. We hypothesize that the oocyte retrieval for IVF several years ago was done in part from the rudimentary horn and may have damaged the isolated cavity, creating a perforation that later provided a narrow alley for the sperm or fertilized ovum to enter.

The natural course of a rudimentary horn pregnancy is rupture as the fetus "outgrows" the rudimentary horn. Mysteries still exist for diagnosing rudimentary horn pregnancy because more than 45% of patients have no clinical symptoms, thus

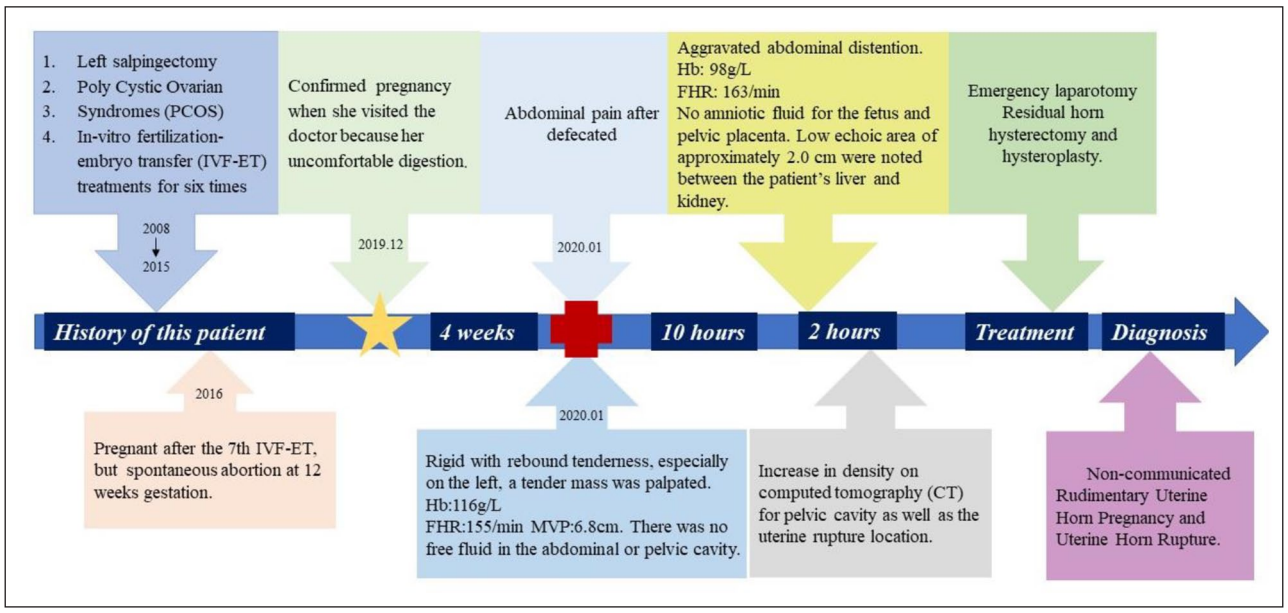

Figure 3. Schematic figure of this case.

only about 8% of them could be recognized.[14] The most common symptom is abdominal pain and it may happen at any time during the pregnancy, dependent on period due to the structural defects of the horned uterus itself, and at what point it can no longer match the growth of embryo. The swelling and stretching of the cavity initially cause increased intermittent pain or tension in the lower abdomen; when this progress to severe, persistent pain with abdominal tension, uterine rupture should be immediately considered among differential diagnoses within the clinical work-up. Vaginal bleeding during pregnancy can be an indication of a threatened pregnancy for both patients and doctors, but this symptom is not common in rudimentary horn pregnancy. On rare occasion, vaginal bleeding may be seen in a naturally terminating early A1b unicornuate pregnancy, when the endometrial lining is shed (an A1b rudimentary horn contains endometrium), and the rudimentary horn communicate with the cervix. Otherwise, lack of specific symptoms in patients with RUH pregnancies can lead to misdiagnosis and mistreatments for these rare but serious events which can be easily missed by doctors.

Many women with an RUH do not conceive, but those who do may with acute uterine rupture during pregnancy. Each examination has limited effect for diagnosis of RUH. In our case, the woman is infertile and had a complicated history of pregnancy. Clinical doctors should consider history, but be vigilant for combinations such as difficulty conceiving or assumed infertility, recurrent miscarriages, fetal growth restriction, and/or placental abnormalities, as these are women at higher risk to fall in this dilemma.[15]

Ultrasound examination is widely used and plays an important role in obstetrics, but the diagnostic sensitivity for rudimentary horn pregnancy is only about 26%, and it will be decreased by the process of gestation, only 8% of rudimentary horn pregnancies without symptoms can be diagnosed by ultrasound examination.[10] These data indicate that RUH pregnancy should always be considered within the differential diagnosis, as well as tubal pregnancy and intrauterine pregnancy in a bicornuate uterus. Lawhon et al.[16] report a case in which magnetic resonance imaging (MRI) was used to successfully diagnose a rudimentary horn pregnancy, permitting timely surgical intervention and avoiding catastrophic rupture. MRI proved most valuable in making the diagnosis of rudimentary horn pregnancy after ultrasonography was unable to make a clear location. However, it is not suitable to use MRI as a regular prenatal examination for normal pregnant women.

As in our case, which quickly gained emergency status, the use of CT and a prompt diagnosis allowed for successful residual horn hysterectomy and hysteroplasty rather than a more tragic outcome. They are the keys of successful risk management. Medical and radiological personnel must maintain a high degree of alertness to prevent the morbidity associated with this condition.

Conclusion

Noncommunicating RUH pregnancy is an extremely rare form of ectopic pregnancy, and its diagnosis and management remain challenging. CT scan can be used in an emergency circumstance and is considered key tool for correct diagnosis and risk management.

Author contributions

W.S., W.L., and Y.Z. performed the surgery. L.W. organized the discussion and updated the references. S.R. provided help to output

the CT images. L.W. and S.R. wrote the manuscript. All authors contributed to editorial changes in the manuscript. All authors read and approved the final manuscript.

Declaration of conflicting interests

The author(s) declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

Ethical approval

Ethical approval to report this case was obtained from Liaocheng People's Hospital Ethical Review of Medical Research of Human Being (APPROVAL NUMBER: 2019056).

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Informed consent

Written informed consent was obtained from the patient(s) for their anonymized information to be published in this article.

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