Letter to the Editor

A case of rudimentary uterine horn pregnancy complicated with placental implantation in later stages of pregnancy

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Submitted Jun 19, 2023. Accepted for publication Oct 16, 2023. Published online Nov 20, 2023.
doi: 10.21037/qims-23-889
View this article at: https://dx.doi.org/10.21037/qims-23-889

Introduction

Rudimentary uterine horn is a kind of congenital uterine malformation, which is rare in clinical practice. According to related literature reports, it accounts for about 5% of the total Müllerian duct malformation, and its incidence rate in infertile women is about 1/100,000 (1-3). Rudimentary uterine horn pregnancy (RHP) is a type of rare ectopic pregnancy. It is mostly found in the childbearing age, has no typical clinical manifestations, and is prone to missed diagnosis or misdiagnosis. It has been reported in the literature that RHP can rarely be maintained to term, is very infrequently discovered in the third trimester of pregnancy, and rarely results in a live fetus. It has been reported that RHP presents a significant high risk in the second and third trimesters of pregnancy. From the perspective of diagnostic techniques, the frequency of use of magnetic resonance imaging (MRI) and laparoscopic surgery is significantly higher in developed countries (4-6). Therefore, correct and timely diagnosis of RHP is very important for early treatment.

Case presentation

The case involved a 37-year-old female at 34^{+2} weeks gestation, who visited the Gansu Provincial Maternity and Child-Care Hospital after the discovery of placenta previa through prenatal ultrasound at another hospital. Birth history: she delivered a baby boy vaginally at 32 weeks of gestation in 2014, and had a spontaneous abortion in 2017. Obstetric examination: fetal movement as usual, fetal heart rate 142 beats/min, regular, no contractions, no broken membranes, and no bloody vaginal secretions. The estimated fetal weight was 2,000 g, fetal position left sacrum transverse (LST), amniotic fluid depth 27.3 cm, and fetal heart monitoring indicated that nonstress test (NST) was responsive and fetal reserve was good. Ultrasound examination in our hospital showed the following: intrauterine single fetus, breech presentation, umbilical cord around the neck, placenta located in the posterior wall of the uterus, with several anechoic areas visible in it, poor sound penetration, and scattered small and dense echo spot floating. Therefore, placental implantation and multiple placental blood pools were considered. Prenatal pelvic MRI showed (*Figure 1*) the following: intrauterine single fetus, breech position, umbilical cord around the neck, placenta mainly located in the posterior inferior wall of the uterus, multiple tortuous blood vessel shadows in the placenta, and accessory placenta signal shadows in the right anterior wall of the uterus. A signal shadow of the uterine body was seen above the posterior bladder and below the pregnant uterus, which communicated with the cervix. Therefore, RHP complicated with placenta

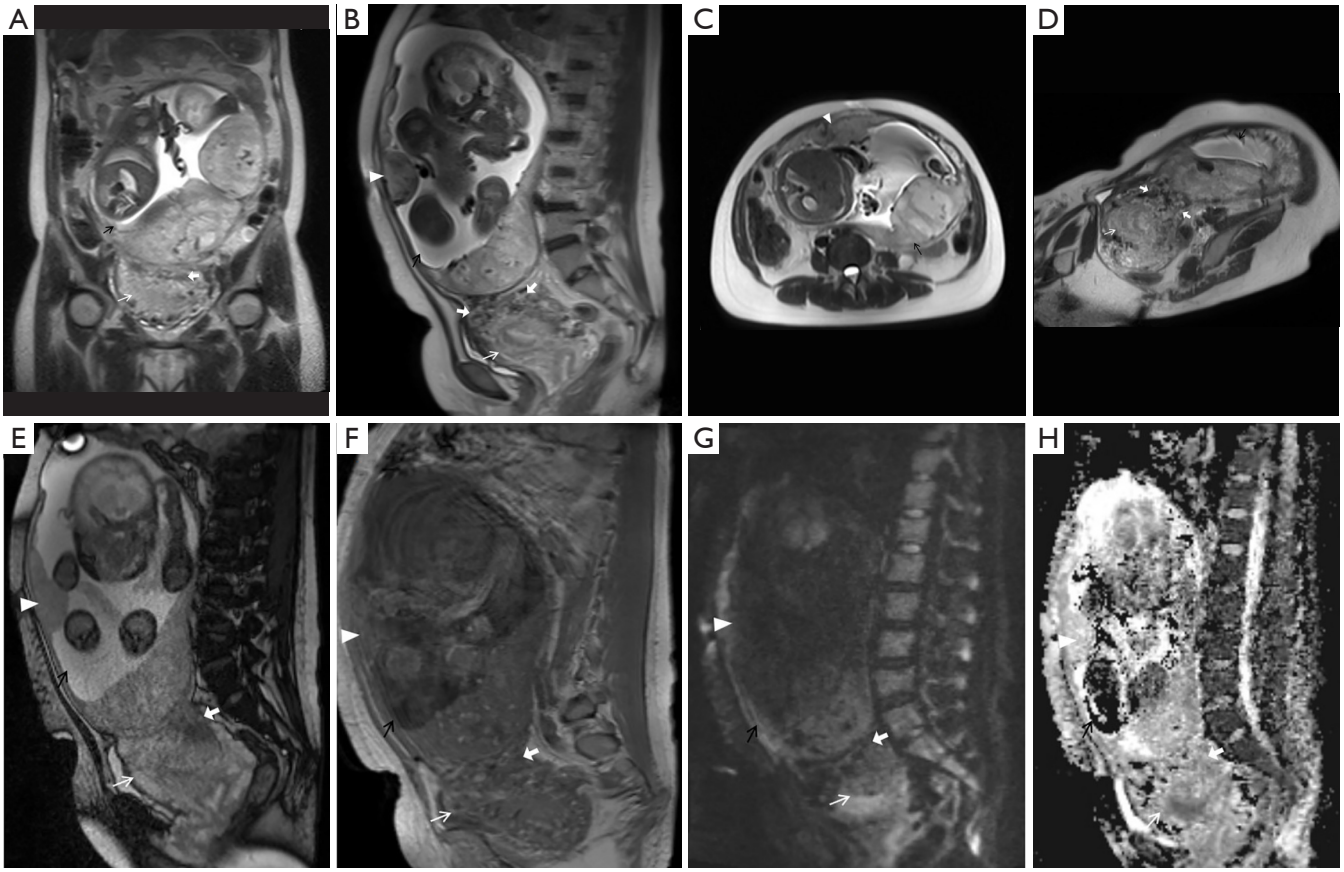

Figure 1 Prenatal pelvic MRI showed RHP complicated with placenta implantation, and accessory placenta. Intrauterine single fetus, breech position, umbilical cord around the neck (A), placenta mainly located in the posterior inferior wall of the uterus, a signal shadow of the uterine body was seen above the posterior bladder and below the pregnant uterus, which communicated with the cervix (B), and accessory placenta signal shadows in the right anterior wall of the uterus (C). The placental parenchyma signals were uneven and multiple tortuous blood vessels were seen in the placenta, T2WI showed low signals (D), T2WI trufi sequences showed equal and slightly lower signals (E), T1WI showed equal and slightly higher signals (F), DWI sequences showed equal and slightly lower signals (G), and ADC images showed slightly lower signals (H). (A) Coronal position on T2WI; (B) sagittal position on T2WI; (C) axial position of T2WI; (D) oblique coronal position on T2WI; (E) T2WI trufi sagittal position; (F) T1WI sagittal position; (G) sagittal position of DWI map; (H) sagittal section of ADC diagram. Normal uterus (fine white arrow), residual horn uterus (fine black arrow), accessory placenta (white triangle); multiple tortuous blood vessels in the placenta (thick white arrow). MRI, magnetic resonance imaging; RHP, rudimentary uterine horn pregnancy; T2WI, T2-weighted imaging; T1WI, T1-weighted imaging; DWI, diffusion-weighted imaging; ADC, apparent diffusion coefficient.

implantation, and accessory placenta was considered. The treatment method was selected according to the gestational age of the RHP. Considering she was in the later stages of pregnancy, rudimentary uterine horn and ipsilateral salpingectomy were performed after caesarean section. Intraoperative observation (*Figure 2*) revealed the following: a uterus was palpable at the lower right side of the pelvic cavity, with the size of about 10 cm × 8 cm; the pregnant uterus was located at the left side of the abdomen connecting with the angle of the lower right side of the uterus; the blood vessels on the surface of the pregnant uterus were dilated. By keeping away from the vascular engorgement, a longitudinal incision was performed on the pregnant uterus, and the baby girl was delivered with foot exposed first. The newborn Apgar score was 9–10, and the newborn baby girl weighed 1,740 g. After delivery of the fetus, hemostatic ligation was conducted on the normal uterus; rudimentary uterine horn and left salpingectomy were performed. The intraoperative examination found no obvious connection between rudimentary uterine horn and

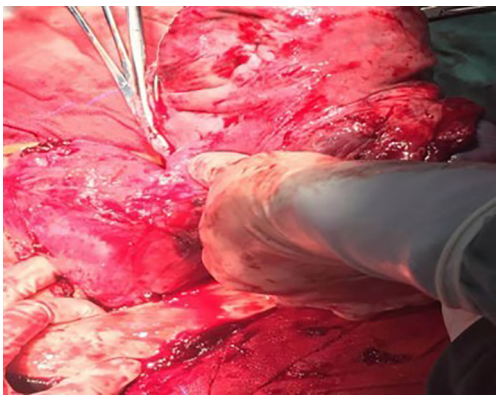

Figure 2 Intraoperative observation: a uterus was palpable at the lower right side of the pelvic cavity, with the size of about 10 cm × 8 cm; the pregnant uterus was located at the left side of the abdomen connecting with the angle of the lower right side of the uterus; the blood vessels on the surface of the pregnant uterus were dilated.

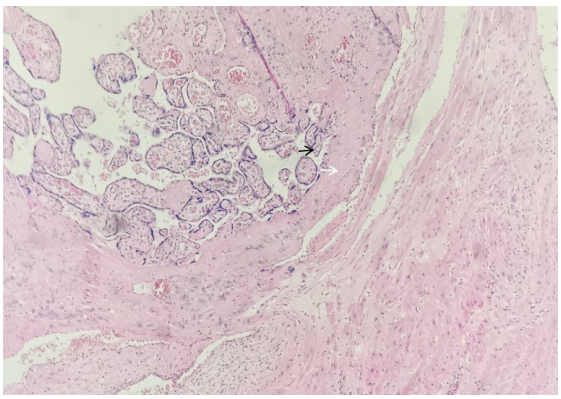

Figure 4 Pathological biopsy (hematoxylin & eosin, ×100): microscopically, the endometrium at placenta attachment lacked the decidua layer and some villi (fine black arrow) invaded the uterine muscle wall (fine white arrow).

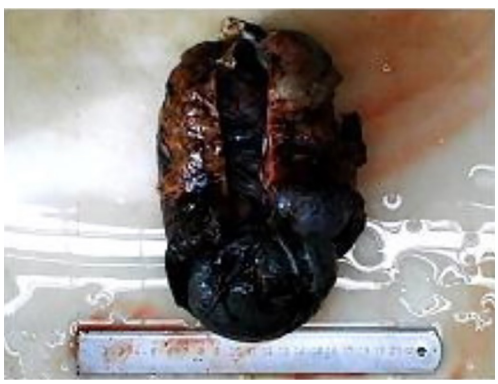

Figure 3 Intraplacental hematoma: a mass of grayish red aplastic tissue is seen, measuring 2.5 cm × 2.0 cm × 1.5 cm.

the normal uterus. Pathology roughly showed that most of the placenta had firmly adhered to the uterine wall, and examination of the intraplacental hematoma revealed a mass of grayish red unplastic tissue measuring 2.5 cm × 2.0 cm × 1.5 cm in size (*Figure 3*). Microscopically, the endometrium at the placental attachment lacked the decidua layer and some villi invaded the uterine muscle wall (*Figure 4*). The final diagnosis was rudimentary uterine horn (type IIb) pregnancy, placental implantation, and accessory placenta. All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013). Written informed consent was provided by the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the editorial office of this journal.

Discussion

This case involved a late-term pregnancy with RHP combined with placental accretion and paraplacenta. The difference between this case and previous reported cases is that late pregnancy with placental accretion has most commonly been reported in older women, and the newborn survival after cesarean section, as reported in this case, is extremely rare. Pregnancy with a residual horn uterus has been reported to carry significant obstetric risks, including miscarriage in the first and second trimesters, fetal growth restriction, preterm delivery, and rupture of the residual horn uterus. Therefore, it is crucial to recognize the RHP at an early stage, and regular prenatal check-ups must be conducted to detect potential complications and implement timely intervention measures.

Rudimentary uterine horn is a kind of congenital uterine malformation that is clinically rare, accounting for about 5% of Müllerian duct malformations according to literature reports (1-3). Different types of rudimentary uterine horn have different clinical characteristics (4-9). Rudimentary uterine horn has an incidence rate of about 1/76,000–1/150,000, and mostly presents as type IIa and

type IIb (10-13). Due to the myometrial maldevelopment of the rudimentary uterine horn, it has been reported that RHP rarely reaches full term, and that the rupture of the rudimentary uterine horn occurs at about 14–20 weeks of gestation. Therefore, in the presence of acute abdominal pain in the second trimester of pregnancy, the rupture possibility of RHP should be considered (13-16). There have also been reports of late-trimester residual horn pregnancy, but the case of RHP complicated with placental implantation in the later stages of pregnancy is very rare. The rudimentary uterine horn is prone to placental adhesion and placental implantation due to its muscular layer hypoplasia. This case detailed a patient with RHP complicated with placental implantation and accessory placenta in the later stages of pregnancy.

Imaging examination plays a significant role in the diagnosis of female genital tract malformation. Currently, in the opinion of most scholars, ultrasound is the preferred examination method for the diagnosis of rudimentary uterine horn (17,18). However, by virtue of multisequencing, multiparameters, and multi-dimensional imaging, MRI has higher soft tissue and spatial resolution, can clearly display the anatomical structure of uterus and vagina, as well as the external form and contour of the uterus. Thus, it can more effectively evaluate the morphology change and classification of uterine malformation, and improve the diagnostic accuracy and detection rate of the female genital tract abnormalities (19-22). In March 2020, the European Society of Urogenital Radiology (ESUR) issued guidelines to standardize and guide MRI examination of female congenital reproductive tract abnormalities.

According to the classification of the American Fertility Society (AFS), the MRI of type IIa is manifested as rudimentary uterine horn with uterine cavity yet no cervix uteri, with functional endometrial endometrium, and connected with unicornuate uterus. The MRI of type IIb is manifested as rudimentary uterine horn with uterine cavity yet no cervix uteri, with functional endometrial endometrium, unconnected with unicornuate uterus, and hemoperitoneum on the side of rudimentary uterine horn. The MRI of type IIc is mainly manifested as the soft tissue signal consistent with the rudimentary uterine horn and muscle layer signal, which is connected with the single horn uterus by fibrous band, but not connected with the unicornuate uterus. The MRI of type IId mainly shows that the uterus is tilted to one side of the pelvic cavity, presenting “spindle shape” or “banana shape”, without the normal inverted triangle shape of the uterine cavity, with abnormal signal, and normal or small volume. Although prenatal MRI is generally not recommended before the 18th week of pregnancy, timely selection of MRI based on prenatal ultrasound and evaluation can be a great aid in preoperative evaluation and guidance of surgery. For those who are suspected to have a residual horn uterus by ultrasound examination in the second and third trimester, MRI examination can be used to objectively evaluate the different types of the residual horn uterus, prenatal placenta, and fetus.

The treatment method of RHP is selected according to the gestational weeks (23-25). Once a definite diagnosis is made in the first and second trimesters, rudimentary uterine horn and ipsilateral salpingectomy should be performed. If a definite diagnosis is made in the late stage of pregnancy, it is suggested to perform caesarean section first, then rudimentary uterine horn and ipsilateral salpingectomy. This case was treated in the third trimester. This case was treated in the third trimester of pregnancy by cesarean section first, followed by resection of the residual horn uterus and ipsilateral salpingectomy. Timely preoperative evaluation and treatment ensured the safety of pregnant women and newborns and avoided misdiagnosis and missed diagnosis. With the deepening of the understanding of rudimentary uterine horn and the improvement of diagnosis and treatment technology, great progress has been made in the diagnosis and treatment of rudimentary uterine horn.

Conclusions

RHP has low incidence rate and presents no typical clinical symptoms. In order to avoid missed diagnosis and misdiagnosis as well as the occurrence of serious complications, it is suggested to enhance the clinical awareness of such diseases and improve the diagnosis rate of preoperative examination. Pelvic MRI examination can accurately and objectively evaluate the different types and related complications of rudimentary uterine horn. Therefore, it is the best and most effective examination method for preoperative imaging evaluation and can provide considerable help for preoperative clinical evaluation and operation guidance.

Acknowledgments

We would like to thank the whole study team at the Gansu Provincial Maternity and Child-Care Hospital and the NMR Room of Lanzhou University Second Hospital, for

their continued support.
Funding: None.

Footnote

Conflicts of Interest: All authors have completed the ICMJE uniform disclosure form (available at https://qims.amegroups.com/article/view/10.21037/qims-23-889/coif). The authors have no conflicts of interest to declare.

Ethical Statement: The authors are accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013). Written informed consent was provided by the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the editorial office of this journal.

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Cite this article as: Xu S, Zhang J, Yue S, Qian J, Yang L, Xu YZ, Zhang J. A case of rudimentary uterine horn pregnancy complicated with placental implantation in later stages of pregnancy. Quant Imaging Med Surg 2024;14(1):1167-1172. doi: 10.21037/qims-23-889

