Case Report

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Robert's uterus with delayed diagnosis and potential consequences: a case report

Jing Zhang[1,2,3], Weili Zhou[4], Ying Tang[4], Shiqiao Tan[1,2,3] and Lin Qiao[1,2,3]

Abstract

A 24-year-old woman who wished to become pregnant presented to our hospital with an enlarged ovarian endometrioma and developmental abnormality of the uterus. Robert's uterus complicated by hematosalpinx, ovarian endometrioma, and endometriosis were finally identified 1 year after previously being diagnosed with a cyst and uterine abnormality at a local hospital. The function of the salpinx and the pelvic environment were damaged because of the delayed diagnosis and operation. Gynecologists and sonologists should be aware of and alert to this rare entity while evaluating and managing cases of uterine abnormalities and endometriosis. Prompt early diagnosis and proper management of Robert's uterus are important for avoiding future morbidity because these are major factors in protecting fertility.

Keywords

Endometriosis, fertility, hematosalpinx, ovarian endometrioma, Robert's uterus, hematometra

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Introduction

The prevalence of uterine anomalies is approximately 0.50%. Robert's uterus was

[1]Department of Obstetrics and Gynecology, West China Second University Hospital, Sichuan University, Chengdu, Sichuan, China
[2]Reproductive Endocrinology and Regulation Laboratory, West China Second University Hospital, Sichuan University, Chengdu, Sichuan, China
[3]Key Laboratory of Birth Defects and Related Diseases, Women and Children Ministry of Education, Sichuan University, Chengdu, Sichuan, China
[4]Department of Ultrasound, West China Second University Hospital, Sichuan University, Chengdu, Sichuan, China

Corresponding author:
Lin Qiao, Department of Obstetrics and Gynecology, West China Second University Hospital, Sichuan University, No. 20, 3rd Section, Renmin South Rd., Chengdu 610041, Sichuan, China.
Email: 54266753@qq.com

first reported in 1970 as a rare variant of a complete, but oblique, septate uterus consisting of a non-communicating hemicavity and a contralateral unicornuate uterine cavity in a single uterine body with a normal fundus.[1] We report a case of delayed diagnosis and treatment of Robert's uterus, although ultrasound-guided hysteroscopy and laparoscopy were finally performed.

Case report

A 24-year-old woman presented to the Reproductive Endocrinology Department with the complaint of a gradually enlarging, right ovarian, chocolate cyst. The cyst (3 cm in diameter) and uterine developmental abnormality (complete septate uterus or bicornuate uterus) were found 1 year previously in a local hospital. However, this young woman wanted to attempt pregnancy because of normal levels of tumor biomarkers, without any other examinations or treatments.

The patient attained menarche at 13 years old and had regular menstrual cycles every 30 days with a 7-day duration, accompanied by mild dysmenorrhea that occasionally required medication. A gynecological examination showed an anatomically normal vulva, vagina, and cervix. The uterus was a normal size, but the right corpus was slightly larger than the left.

Three-dimensional ultrasound showed that the uterus was divided by a septum. Therefore, a complete septate or bicornuate uterus was suspected. However, the right uterine hemicavity did not communicate with the single cervix, but was accompanied by hematometra (Figure 1a). Two cysts were located in the right and left adnexa (7 cm and 1.8 cm, respectively), and these were provisionally diagnosed as ovarian endometrioma. Urinary tract ultrasound showed that both kidneys, the bladder, and the ureters were normal.

Laparoscopic oophorocystectomy and hysteroscopic electrotomy of the uterine septum were scheduled to be performed. Laparoscopy showed that the basilar part of the uterus was wide with an obvious bulge on the right fundus due to hematometra (Figure 1b). Scattered violet blue nodules were observed on the outer surface of the uterus (endometriosis). Extensive adhesions were found among the adnexa, pelvic peritoneum, and posterior uterine wall. An ovarian endometrioma (nearly 1 cm) was removed from the left ovary after adhesiolysis. However, a hematosalpinx ($7 \times 5 \times 5$ cm), but not ovarian endometrioma, was evacuated from the right adnexa. The mucosa had disappeared from the inner surface of a dilated, thickened, and stiff right fallopian tube. Hydrotubation with methylene blue was performed. However, the liquid dye did not exit the distal end of the right fallopian tube.

During hysteroscopy, a thick muscular septum was found to extend from the fundus to the internal os (Figure 1c). A left uterine hemicavity with a single ostium was identified without any communication with the right hemicavity. A longitudinal incision of the asymmetric septum with a bipolar needle electrode under transabdominal ultrasonic monitoring was performed to enter the right cavity (Figure 1d). The corresponding tubal ostium and endometrium were identified. The methylene blue dye then exited the distal end of the right fallopian tube. Foley's catheter, which was inflated with 7 mL of normal saline, was then placed in the uterine cavity for 7 days. Hyaluronic acid gel was then injected into the cavity after the catheter was pulled out. A follow-up three-dimensional ultrasound 1 month later showed a larger normal uterine cavity (Figure 1e). During a mini-hysteroscopic (2.9 mm in diameter) examination, bilateral tubal ostia were finally observed, with mild postsurgical bulge in the upper part between the two cavities (Figure 1f). The patient experienced regular menstruation postoperatively, with a normal volume,

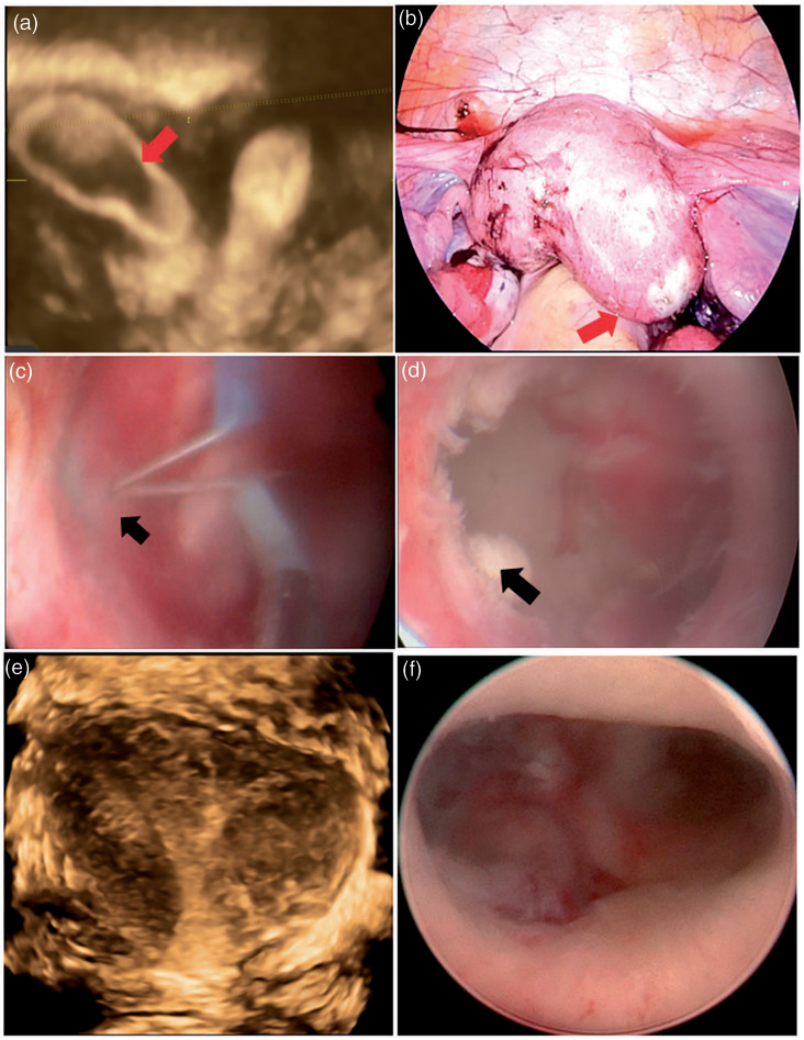

Figure 1. Uterus with hematometra (red arrow) in the right uterine hemicavity on three-dimensional ultrasound before surgery (a). An obvious bulge in the right fundus was observed by laparoscopy (red arrow) (b). Hysteroscopy before (c) and after (d) incision of an asymmetric septum (black arrows). Three-dimensional ultrasound examination 1 month after surgery shows an enlarged cavity (e). A mini-hysteroscopy (2.9 mm in diameter) examination 1 month after surgery shows a uterine cavity with a bulge (f).

but without dysmenorrhea. She has been attempting to become pregnant for 7 months up to December 2020.

Discussion

Robert's uterus is an uncommon congenital Müllerian abnormality, with one blind hemicavity and a contralateral unicornuate uterine cavity. Unilateral cervical aplasia should be present according to classification of the European Society of Human Reproduction and Embryology–European Society for Gynaecological Endoscopy.[2] However, the cervix is normal in some cases, which demonstrates the

embryological/anatomical paradox of the European Society of Human Reproduction and Embryology–European Society for Gynaecological Endoscopy classification for this abnormality. Unilateral hematometra, hematosalpinx, and endometriosis are secondary features of Robert's uterus. Robert's uterus can be divided into three clinical categories by the status of hematometra in the blind hemicavity as follows: with a large hematometra in the blind hemicavity and acute pelvic pain; with an inactive blind hemicavity without hematometra and recurrent miscarriages; and with a small hematometra in the blind hemicavity.[1] Blind horns are more common on the right side because the left Müllerian duct advances slightly ahead of a right Robert's uterus. However, a left Robert's uterus has occasionally been reported.[3]

Rare complications of Robert's uterus, such as pregnancy in the blind cavity (considered as one type of ectopic pregnancy), unique ipsilateral renal agenesis, and a bicornuate uterus, have been reported.[4] In contrast to the classical symptoms, Robert's uterus in the present patient did not show prominent symptoms related to hematometra as previously reported.[5] Therefore, diagnosis and treatment were delayed and the function of the salpinx and pelvic environment were severely damaged. Three-dimensional ultrasound and magnetic resonance imaging are suggested as being useful for evaluating Robert's uterus.[6,7] In the differential diagnosis, a unicornuate uterus with non-communicating rudimentary horn should be considered.

Management of Robert's uterus is not fully established because of the few reports on this condition. Drainage and prevention of recurrence of hematometra, and excision of adnexal endometriomas and hematosalpinx are the main therapies. Robert's uterus was typically previously managed via laparotomy (hysterotomy incision, horn resection, and repair of the myometrium) or endometrectomy of the blind cavity.[8] However, in a previous report, a scar formed around the operation site, the cavity shape and volume of the uterus were not improved, and placenta accreta had the possibility of occurring during pregnancy (amputation and ablative surgical therapy).[9] All of these factors greatly postponed the following attempt at pregnancy.

Currently, ultrasound combined with hysteroscopy is considered as a practical, minimally invasive, safe choice for Robert's uterus, and this could be an option in women without visible signs of endometriosis in ultrasound (https: //www.youtube.com/watch?v = 4DWwCrwEeF4).[1,10] Use of laparoscopy may be helpful for some conditions, such as infertility with hematosalpinx.[11] These methods significantly reduce surgical trauma, result in prompt postoperative recovery, and preserve the integrity of the uterus, which are good for protecting fertility. However, if doctors do not have sufficient expertise in treatment or the patient is young and without any major symptoms, conservative management with long-term follow-up, but delayed therapy, could be considered.[12]

To protect fertility and avoid inappropriate management, gynecologists and sonologists should be aware of and alert to Robert's uterus while evaluating and managing cases of uterine abnormality and endometriosis. Prompt early diagnosis and proper management of Robert's uterus are important for avoiding future morbidity, and these are major factors for protecting fertility.

Ethics statement

The patient gave informed consent for treatment and publication of this case report. Approval by the Medical Ethics Committee of West China Second University Hospital of Sichuan University (No. 128) was received for

publication of this case report. The CARE guidelines were followed for this case report.

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Declaration of conflicting interest

The authors declare that there is no conflict of interest.

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ORCID iD

Jing Zhang https://orcid.org/0000-0002-1978-0030

References

1. Ludwin A, Ludwin I, Bhagavath B, et al. Pre-, intra-, and postoperative management of Robert's uterus. *Fertil Steril* 2018; 110: 778–779.
2. di Spiezio Sardo A, Campo R, Gordts S, et al. The comprehensiveness of the ESHRE/ESGE classification of female genital tract congenital anomalies: a systematic review of cases not classified by the AFS system. *Hum Reprod* 2015; 30: 1046–1058.
3. Shah N and Changede P. Hysteroscopic Management of Robert's Uterus. *J Obstet Gynaecol India* 2020; 70: 86–88.
4. Liu Y, Yang Y, Duan J, et al. Favorable pregnancy outcome for a patient with Robert's uterus, bicornuate uterus, and ipsilateral renal agenesis. *Int J Gynaecol Obstet* 2020. doi: 10.1002/ijgo.13274. (Online ahead of print)
5. Singhal S, Agarwal U, Sharma, D, et al. Pregnancy in asymmetric blind hemicavity of Robert's uterus – a previously unreported phenomenon. *Eur J Obstet Gynecol Reprod Biol* 2003; 107: 93–95.
6. Ludwin A, Martins WP and Ludwin I. Uterine cavity imaging, volume estimation and quantification of degree of deformity using automatic volume calculation: description of technique. *Ultrasound Obstet Gynecol* 2017; 50: 138–140.
7. Mittal P, Gupta R, Mittal A, et al. Magnetic Resonance Imaging (MRI) Depiction of Robert's Uterus: A Rare Müllerian Duct Anomaly Presenting with Cyclical Pain in Young Menstruating Woman. *Pol J Radiol* 2017; 82: 134–136.
8. Kiyak H, Karacan T, Wetherilt LS, et al. Laparoscopic blinded endometrial cavity resection for Robert's uterus. *J Minim Invasive Gynecol* 2018; 25: 340.
9. Vural M, Yildiz S, Cece H, et al. Favourable pregnancy outcome after endometrectomy for a Robert's uterus. *J Obstet Gynaecol* 2011; 31: 668–669.
10. Ludwin A, Ludwin I and Martins WP. Robert's uterus: modern imaging techniques and ultrasound-guided hysteroscopic treatment without laparoscopy or laparotomy. *Ultrasound Obstet Gynecol* 2016; 48: 526–529.
11. Biler A, Akdemir A, Peker N, et al. A Rare Uterine Malformation: Asymmetric Septate Uterus. *J Minim Invasive Gynecol* 2018; 25: 28–29.
12. Ludwin A and Pfeifer SM. Reproductive surgery for müllerian anomalies: a review of progress in the last decade. *Fertil Steril* 2019; 112: 408–416.