

Intrauterine Adhesions Combined with Robert's Uterus : A Case Report and Literature Review

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Case report

Keywords: Robert's uterus, Mullerian abnormality, Intrauterine adhesions, Hematometra, Dysmenorrhea, Septal resection

Posted Date: June 28th, 2021

DOI: <https://doi.org/10.21203/rs.3.rs-620125/v1>

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Abstract

Background: Robert's uterus, a rare congenital abnormality of Mullerian duct development, has oblique septum and non-communicating asymmetric hemi-cavity. Key clinical characterization of the congenital disorder is presence of hematometra and severe dysmenorrhea, some patients experience acute abdominal pain in association with menstruation. Due to the difficulty of preoperative diagnosis and high rate of misdiagnosis, Preoperative assessment matters the choice of emergency surgery and avoid a second surgery. No systematic literature review in detail has been reported previously.

Case presentation: We reported a rare case of Robert's uterus with severe abdominal pain during menstruation, in which the patient initially underwent emergency laparoscopic right ovarian cystectomy, right salpingectomy, and pelvic adhesiolysis on suspicion of ovarian teratoma torsion. However, dysmenorrhea still existed or even aggravated after the operation, hysteroscopic surgery was performed three month later, which revealed two asymmetric uterine cavities, hematometra was located in the right-side blind cavity, and thus diagnosed as Robert's uterus and severe uterine adhesion, which, to our knowledge, has not been reported previously. Hysteroscopic incision of the septum and intrauterine adhesion were performed. Subsequently, Two-year follow-up showed no obvious dysmenorrhea recurrence.

Conclusion: Robert's uterus is uncommon but can mimic other common and acute abdominal disease. Advance imaging technologies, such as three-dimensional ultrasound and magnetic resonance imaging combined with hysteroscopy and laparoscopy are less invasive for diagnosis and treatment of Robert's uterus. Septal resection is main surgical procedure, combining laparoscopy and hysteroscopy is conducive and less invasive treatment.

Background

Robert's uterus is a rare Mullerian abnormality with an asymmetric septum, which was first reported by Robert in 1970. To date, more than 20 cases of Robert's uterus have been reported in previous literature. It is characterized by non-communicating intrauterine cavity and unilateral haematometra in blind uterine cavity attributing to the oblique septum. Progressively significant dysmenorrhea and severe abdominal pain are the main manifestation. We reported and analyzed in detail the clinical features and mechanisms of a case of Robert's uterus with severe abdominal pain. The patient underwent surgical treatment twice and was also complicated with severe adhesions, which was not mentioned in the literature so far. Hence, we simultaneously performed a systematic literature review for purposing of improving preoperative diagnosis and avoiding inappropriate operation performance.

Case Presentation

The patient, a 24-year-old married woman, suffered from reduced menstruation and lower abdominal pain around menstrual period since the age of 22, especially before and on the first day of menstruation. The frequency of pain increased afterward, from once every three months to once a month, from spontaneous remission without the assist of medications to drug assistant. Two months ago, the patient presented to the emergency department of local hospital due to severe abdominal pain, accompanied by nausea and vomiting. Ultrasonography revealed that intrauterine fluid measured 13×20 ×10 mm, cystic mass localized at right parametrium space measured 32×23 ×20 mm, isoechoic and hyperechoic cyst on the right ovary measured 55×60×57 mm. Emergency laparoscopic surgery was performed for suspicion of ovarian teratoma torsion, which has been recorded severe pelvic adhesions, normal size of the uterus, right hematosalpinx, as well as cystic mass of the right ovary without torsion. Laparoscopic right ovarian cystectomy, right salpingectomy, and pelvic adhesiolysis were performed. The pathological diagnosis was ovarian mature cystic

teratoma and right fallopian tube endometriosis. However, the patient presented to our center for the unrelieved abdominal pain after the operation. The left endometrium thickness was 4 mm, and the right uterine cavity fluid measured 42×19 mm. The size of anechoic area in the right uterine horn was 23×15 mm, which seemed to communicate with the right uterine cavity (Fig. 1a). Three-dimensional ultrasound (3D-US) confirmed the presence of oblique septum and fluid in the blind uterine cavity (Fig. 1b). Magnetic resonance imaging (MRI) showed a septum in the uterus divided the endometrial cavity into two cavities of unequal in size, and haematometra was mainly in the right smaller cavity, mimicking the unicornuate uterus with the communicating rudimentary horn (Fig. 2). Physical examination and laboratory blood examination were normal. Hysteroscopic surgery with ultrasound guided was performed, which revealed a small narrow uterine cavity with one horn communicated with cervical canal and biased to the left side. The fundus and left wall of the uterine cavity had severe adhesion with a thin endometrium, and the left fallopian ostium was invisible. A fibrous oblique diaphragm presented on the right-side uterine cavity with a slight bulge. The right corn and fallopian ostium were invisible (Fig. 3a). After gently separating the weak part of the diaphragm along the bulge using a needle electrode, the dark brown hemocele liquid spilled out, and the oblique septal tissue was gradually removed by band electrodes along the opening. Then, the right-side uterine cavity and the right corn were exposed, separating intrauterine adhesions with needle electrode recover uterus normal morphology. A Foley catheter inflated with 3 ml saline was placed in the uterine cavity for 7 days to prevent intrauterine adhesion recurrence. The patient was discharged 2 days after the surgery. Estrogen and progesterone sequential treatment were given for the prevention of recurrence. Four weeks after the operation, the repeat hysteroscopy showed a normal appearing cavity and endometrium (Fig. 3b). The patients were followed up for two-year after the operation, intractable dysmenorrhea disappeared.

Discussion And Conclusions

Robert's uterus is a rare congenital abnormality, a uterine septum asymmetrically separates the uterine cavity from the bottom of the uterus, half of which is a disconnected blind cavity [1]. Gupta et al. suggested that right side are common for the ahead advancement of left Mullerian duct [2]. However, blind cavity is more commonly left-side in the review. Musset et al [3] have summarized its characteristics include: (1) primary dysmenorrhea; (2) no significant difference of the uterus by laparoscopy and the unicorn uterine cavity found by hystero-graphy; (3) no deformity of the urinary system. Most patients complained of dysmenorrhea, periodic abdominal pain, abnormal menstruation, abortion, or infertility. Twenty-two cases were conducted in our systematic reviewed literature (Table 1), 20/22 cases (81.82%) underwent dysmenorrhea [2, 4–19], 3/22 cases (13.64%) abortion [4, 7, 20], one case endured infertility [14]. It is noted that intractable dysmenorrhea seems to associate with haematometra for the identical existence. Gupta, N proposed that endometriosis may associate with menstrual blood reflux [2]. With hormonal stimulation, menstrual blood induced by the shedding of endometrium could be retrograde into the abdominal cavity through the ipsilateral fallopian tube during menstruation, thus the early dysmenorrhea was periodic but not obvious in initial. Then, with the right fallopian tube gradually thickened, blocked and blood accumulated, dysmenorrhea gradually aggravated due to the formation of a closed cavity, which may associate with inflammation or endometriosis. In our reported case, after the resection of the right fallopian tube, the blind cavity was completely disconnected from the outside world. As a result, increased pressure was associated with the retention of menstrual blood in the closed right-sided uterine cavity, which induced aggravated abdominal pain.

Table 1
Review of Robert's uterus cases reports.

Authors (year)	Case	Age (years)	Dysmenorrhea	Other complaints	Haematometra	Other complication
Deenadayal. 2021	five	28.2(M)	Yes(5/5)	Recurrent abortion(1/5)	Yes(4/4)	Adenomyosis(2/4), Endometrioma(2/4)
Zhang J. 2021	one	24	Yes	-	Yes	Hematosalpinx, Endometriosis
Liu Y. 2021	one	45	No	abnormal menstruation	No	Blind hemicavity Pregnancy, Adenomyosis
Liu Y. 2020	one	16	Yes	Acute abdominal pain	Yes	Ipsilateral renal agenesis, Hematosalpinx
Yang QM. 2019	one	23	Yes	Abortion	Yes	Blind cavity pregnancy, Ipsilateral renal agenesis, Adenomyosis
Shah N. 2019	1	16	Yes	-	Yes	-
Kiyak H. 2018	one	15	Yes	-	Yes	-
Biler A. 2017	one	29	Yes	mild abdominal pain, abnormal menstruation	Yes	-
Mittal P. 2017	one	15	Yes	-	Yes	Haematosalpinx, Endometriosis
John SK. 2017	one	16	Yes	Acute abdominal pain	Yes	-
Ludwin A. 2016	one	22	Yes	-	Yes	-
Di Spiezio Sardo A. 2015	one	30	Yes	infertility	Yes	Uterine myomas, A pseudocystic lesion
Li J. 2015	one	26	Yes	-	Yes	Ovarian endometriotic cyst, Endometriosis
MaddukuriSB.2014	one	16	Yes	-	Yes	Haematosalpinx

Authors (year)	Case	Age (years)	Dysmenorrhea	Other complaints	Haematometra	Other complication
Vural M. 2011	one	24	Yes	-	Yes	-
Capito C. 2009	one	15	Yes	Acute abdominal pain	Yes	-
Gupta N. 2007	one	19	Yes	Acute abdominal pain	Yes	Ovarian endometriotic cyst, Endometriosis
Singhal S. 2002	one	20	No	Abortion	No	Blind cavity pregnancy

To the best of our knowledge, Intrauterine adhesions are first involved in Robert's uterus which have been verified by the laparoscopy, although no intrauterine operation history such as induced abortion, which may be related to obstruction of menstrual blood outflow and inflammation, although there is no evidence of endometritis. In our opinion, early diagnosis and hysteroscopic surgery may reduce incidence of these complication. Ultrasound, MRI, hysteroscopy, and laparoscopy have been performed for Robert' uterus diagnosis. The septum with asymmetric cavities is easily discernible, hematometra and hematosalpinx are easy to identify by the noninvasive imaging modalities, such as 3D-US and MRI, Which are considered the reliable modality to examine the morphology of the uterus [14, 21]. In addition, we strongly recommended a detailed inspection of urinary system considering that two cases of ipsilateral renal agenesis were reported. Ludwin et al. described three types of Robert's uterus according to the amount of bleeding in the blind cavity: no hematocele, small and large [19]. However, the detection rate of uterine malformation by ultrasound is also significantly affected by the diagnostic level of the examiner. In the literature, 5/22 cases (22.73%) endured re-operation or the third time surgery before diagnosis and management [2, 4, 5, 6, 12]. It is still a challenge for clinicians, especially the differentiation of serious dysmenorrhea of Robert's uterus and other acute abdominal diseases. we recommend that if the intraoperative finding is inconsistent to clinical manifestation of severe abdominal pain, uterine inspection is needed. Trauma would be relatively less if taking adequate evaluation and determining the preoperative diagnosis.

Surgical treatment is recommended as soon as it is highly suspected. Surgical option may determine by the factors of age and fertility desire of patient. The prime targets are drainage of hematometra and prevention of its recurrence through septal resection [11]. Laparoscopic septum resection and metroplasty seems to be considerable for the minimal invasion, especially for the adolescent girl. In twenty-one cases of Robert's uterus involved surgery (Table 2), 13/21 cases (61.90%) were performed septal resection by laparoscopy, hysteroscopy or laparotomy. 11/21 cases (52.38%) of endometriosis have been documented in patients performed laparoscopy or hysteroscopy [7, 8, 10, 11, 13, 14]. There is prevalence in laparoscopy-guided hysteroscopic metroplasty recently for the advantage of security and less invasion, one case underwent hysterectomy due to no fertility requirement. Hysteroscopic surgery performed by experienced surgeons is essential for the complicated environment in uterus. At present, the main surgical methods reported in the literature are hysteroscopy or hysteroscopy combined with laparoscopic surgery [8, 10, 11]. Laparoscopy can deal with hematosalpinx, pelvic adhesions, and endometriosis, which cannot be evaluated by auxiliary examination. Some of the Robert's uterus have normal external uterine contour [12], making it difficult to identify in the laparoscopic surgery. Successful pregnancy and the cesarean section of healthy babies after ultrasound-monitored laparoscopy combined with hysteroscopy in patients with Robert's uterus have been reported [15]. If our patient has

fertility requirements in the future, salpingography can be done first to determine whether the left fallopian tube is unobstructed, and then decide on natural pregnancy or assisted reproductive treatment. Monitoring the situation of pregnancy and appropriate intervention to achieve better pregnancy outcomes if required.

Table 2
Review of Robert's uterus cases reports.

Authors (year)	Side	Main surgical Treatment	Number of operations
Deenadayal. 2021	Left (5/5)	Laparoscopic endometrectomy of the blind cavity (1/5), Hysteroscopic septal resection under laparoscopic control (1/5), Laparoscopic excision of the blind cavity (1/5), Hysterectomy with unilaeral salpingo- oophorectomy (1/5), No treatment (1/5)	one(3/4), three(1/4)
Zhang J. 2021	Right	Hysteroscopic septal resection and laparoscopic oophorectomy	one
Liu Y. 2021	Right	Hysterectomy and right salpingectomy	two
Liu Y. 2020	Right	Hysteroscopic septal resection and Laparoscopic right salpingectomy	two
Yang QM. 2019	Right	Hysteroscopic septal resection and Laparoscopy guidance	one
Shah N. 2019	Left	Hysteroscopic septal resection and Laparoscopy guidance	one
Kiyak H. 2018	Right	Laparoscopic septal resection	one
Biler A. 2017	Right	Hysteroscopic septal resection and Laparoscopy guidance	one
Mittal P. 2017	Left	Laparotomy excision of the blind cavity	one
John SK. 2017	Right	Laparotomy septal resection	three
Ludwin A. 2016	Left	Hysteroscopic septal resection.	one
Di Spiezio Sardo A. 2015	Left	Hysteroscopic septal resection and Laparoscopy guidance	one
Li J. 2015	Left	Laparotomy septal resection and laparoscopic oophorocystectomy	one
MaddukuriSB.2014	Left	Laparotomy septal resection	one
Vural M. 2011	Left	Laparotomy endometrectomy of the blind cavity	one

Authors (year)	Side	Main surgical Treatment	Number of operations
Capito C. 2009	-	Laparotomy endometrectomy of the blind cavity	one
Gupta N. 2007	Right	Laparotomy septal resection	two
Singhal S. 2002	Right	Laparotomy	one

Robert's uterus is rare but can mimic other common acute abdominal disease. Adequate evaluation in each case is crucial for diagnosis and management.

Abbreviations

3D-US

Three-dimensional ultrasound;

MRI

Magnetic resonance imaging.

Declarations

Ethics approval and consent to participate

We have obtained informed approval from ethics institutional from Institutional Review Board, First Hospital, Jilin University.

Consent for publication

The details/images/videos will be freely available on the internet and may be seen by the general public. The consent form is provided.

Availability of data and materials

The datasets used and/or analysed during the current study are available from the corresponding author on reasonable request.

Competing interests

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

Funding

This work was supported by the National Natural Science Foundation of China (Grant Nos. 81701436).

Authors' contributions

Meiling Yu: conceived the case report, participated in the surgery, drafted and edited the manuscript.

KeXin Gao: reviewed the literature and drafted the manuscript.

Zhang Han: participated in the literature review and edited the manuscript.

Jihong Zhu: participated in the surgery and literature review.

All authors read and approved the final manuscript.

Acknowledgements

Not applicable.

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Figures

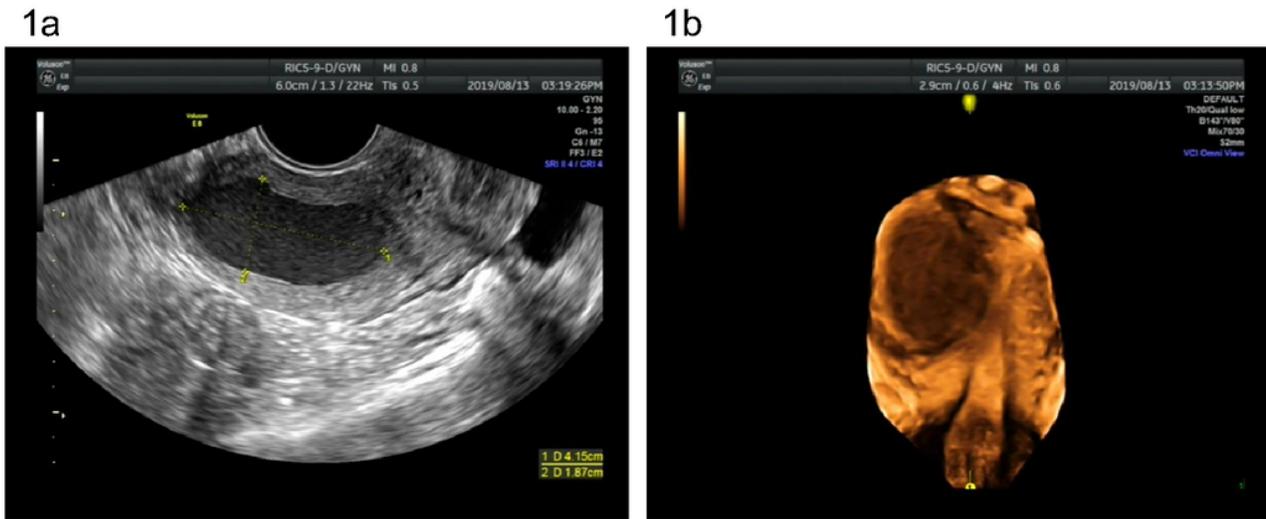


Figure 1

(a) Ultrasound revealed that anechoic zone of hematometra in the right uterine cavity. (b) Three-dimensional ultrasound shows a uterine malformation, the presence of oblique septum and hematometra in the blind uterine cavity.

2



Figure 2

Magnetic resonance imaging shows that a septum in the uterus divided the endometrial cavity into two cavities, hematometra within the right uterine cavity was disconnected from the other cavity which connected to the cervix.

3a



3b



Figure 3

(a) The preoperative image shows a fibrous oblique septum in the uterine cavity, a single fusiform cavity connecting to the cervix was biased to the left side of uterine cavity. Severe adhesions located in the fundus and left wall of uterus cavity, and the left uterine horn and bilateral fallopian ostium were invisible. (b) The follow-up hysteroscopic image showed that the morphology of the uterine cavity is close to normal at four weeks postoperatively.