Intrauterine adhesions combined with Robert’s uterus: a case report and literature review

Kexin Gao1 · Han Zhang1 · Jihong Zhu1 · Meiling Yu1

Received: 3 July 2021 / Accepted: 8 April 2022 / Published online: 29 April 2022
© The Author(s), under exclusive licence to Springer-Verlag GmbH Germany, part of Springer Nature 2022

Abstract
Purpose To summarize the clinical characteristics and surgical option of Robert’s uterus.
Methods We reported a rare case of Robert’s uterus with severe uterine adhesion with successive laparoscopic and hysteroscopic surgery. To our knowledge, such a case has not been reported previously. We also performed a systematic literature review from the PubMed, Embase, and Cochrane databases.
Results Our patient with Robert’s uterus with severe uterine adhesions was successfully treated with hysteroscopic septal resection and hysteroscopic adhesiolysis, and the intractable dysmenorrhea disappeared after the hysteroscopic septal resection. In our study, we analyzed the selected 22 reported cases, 10/22 cases (45.5%) were diagnosed before age 20; 20/22 cases (90.91%) experienced dysmenorrhea, 19/22 cases (86.36%) were with hematometra. 5/22 cases (22.73%) underwent re-operation or a third surgery before diagnosis and management.
Conclusion Robert’s uterus, a rare congenital abnormality of Mullerian duct development, consists of an oblique septum and non-communicating asymmetrical uterine hemi-cavity. The main symptoms are the presence of hematometra and severe dysmenorrhea. Septal resection is the main surgical procedure; however, the rarity and difficulty obtaining a pre-operative diagnosis lead to a high rate of misdiagnosis and second surgery.

Keywords Robert’s uterus · Mullerian abnormality · Intrauterine adhesions · Hematometra · Dysmenorrhea · Septal resection

Abbreviations
3D-US Three-dimensional ultrasound
MRI Magnetic resonance imaging

Introduction
Robert’s uterus is a rare Mullerian abnormality characterized by an asymmetrical septum, first reported by Robert in 1970. To date, more than 20 cases of Robert’s uterus have been reported. Robert’s uterus is a rare congenital abnormality consisting of a uterine septum asymmetrically separating the uterine cavity from the bottom of the uterus, half of which is a disconnected blind cavity [1]. Gupta et al. suggested that a blind cavity commonly develops on the right side, because the left Mullerian duct develops first, embryologically [2]; however, a blind cavity was more commonly left-sided, in the review. Musset et al. [3] summarized the characteristics of Robert’s uterus as: (1) primary dysmenorrhea; (2) no significant difference in the uterus laparoscopically, and a unicorn uterine cavity found on hysterography; and (3) no deformity of the urinary system. Most patients complain of dysmenorrhea, periodic abdominal pain, abnormal menstruation, miscarriage, or infertility. Ultrasonography, magnetic resonance imaging (MRI), hysteroscopy, and laparoscopy have been performed to diagnose Robert’s uterus. No standard surgical options are recommended recently and different operative approaches on Robert’s uterus have been reported. However, diagnosing Robert’s uterus remains a challenge, especially regarding differentiating serious dysmenorrhea from other acute abdominal diseases, some authors have reported re-operation or a third surgery before diagnosis and management.

In concordance with these findings, the current patient underwent surgical treatment twice and was also...
complicated with severe intrauterine adhesions, which has not been mentioned in the previous literature. Hence, we also performed a systematic literature review to improve pre-operative diagnosis and to avoid inappropriate operation. The literature search was performed in PubMed, Embase, and Cochrane library from inception to March 2021. We used a combination of free terms and MeSH terms as search strategies: the MeSH terms "uterus" and the subheading "abnormalities", combining the free terms of "Roberts uterus" or "genital tract abnormality" or "Mullerian abnormality" or "septate". As shown in the flowchart (Fig. 1), a total of 1410 records were identified using search strategies. Literature with complete clinical features published in English language was included; the forms of brief description or video presentation, as well as other irrelevant literature were excluded. Finally, 18 full-text articles meeting the inclusion criteria were included in the literature review.

**Case presentation**

A 24-year-old married woman suffered reduced menstruation and lower abdominal pain associated with her menstrual period since the age of 22 years, especially before and on the first day of menstruation. The frequency of pain increased afterward (from once every 3 months to once a month) and progressed (from spontaneous remission without the assistance of medications to pain requiring drug assistance). Two months before presenting to our institution, the patient presented to the emergency department of a local hospital owing to severe abdominal pain accompanied by nausea and vomiting. Ultrasonography revealed intrauterine fluid measuring 13 × 20 × 10 mm, and a cystic mass in the right parametrium space measuring 32 × 23 × 20 mm. There was also an isoechoic and hyperechoic cyst on the right ovary measuring 55 × 60 × 57 mm. Emergency laparoscopic surgery was performed for the suspicion of ovarian teratoma torsion, which revealed severe pelvic adhesions, normal uterine size, right hematosalpinx, as well as a cystic mass on 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 unrelieved abdominal pain postoperatively. At the second presentation, the left endometrial thickness was 4 mm, and right uterine cavity fluid measured 42 × 19 mm. The size of the anechoic area in the right uterine horn was 23 × 15 mm, and the area appeared to communicate with the right uterine cavity (Fig. 2a). Three-dimensional ultrasonography (3D-US) confirmed the presence of an oblique septum and fluid in the blind uterine cavity (Fig. 2b). MRI showed a septum in the uterus dividing the endometrial cavity into two cavities of unequal size (Fig. 3a), with hematometra within the right uterine cavity, which was disconnected from the other cavity connecting to the cervix (Fig. 3b); and
hematometra, mainly in the right smaller cavity, mimicking a unicornuate uterus with a communicating rudimentary horn (Fig. 3c). Physical examination and laboratory blood examination findings were normal. Ultrasound-guided hysteroscopic surgery was performed, which revealed a small narrow uterine cavity with one horn communicating with the cervical canal and biased to the left side. The fundus and left wall of the uterine cavity had severe adhesions with a thin endometrium, and the left fallopian ostium was invisible. A fibrous oblique diaphragm was present on the right side of the uterine cavity, with a slight bulge. The right uterine cornu and fallopian ostium were invisible (Fig. 4a). After gently separating the weakest part of the diaphragm along the bulge using a needle electrode, dark-brown hematocoele liquid spilled out, and the oblique septal tissue was gradually removed using band electrodes along the opening. Then, the right-side uterine cavity and the right cornu were exposed, and we separated the intrauterine adhesions with needle electrodes to recover normal uterine morphology. A Foley catheter inflated with 3 ml of saline was placed in the uterine cavity for 7 days to prevent intrauterine adhesion recurrence. The patient was discharged 2 days after the operation. Estrogen and progesterone sequential treatment was prescribed to prevent recurrence. Four weeks after the operation, repeat hysteroscopy showed a normal-appearing cavity and endometrium (Fig. 4b). The intractable dysmenorrhea of the patient was disappeared gradually at follow-up for 2 years after the operation.
Discussion

Robert’s uterus is characterized by a non-communicating intrauterine cavity and unilateral hematometra in a blind uterine cavity owing to the oblique septum. Progressively significant dysmenorrhea or severe abdominal pain are the main symptoms [1–3]. Intractable dysmenorrhea appears to be associated with concurrent hematometra. With hormonal stimulation, menstrual blood induced by endometrial shedding could flow retrograde into the abdominal cavity through the ipsilateral fallopian tube during menstruation; thus, initially, dysmenorrhea in Robert’s uterus is periodic and not obvious. Then, with the ipsilateral fallopian tube gradually thickened, the tube becomes blocked, and blood accumulates. As a result, dysmenorrhea gradually worsens owing to the formation of a closed cavity, which may be associated with inflammation or endometriosis. We identified 22 cases in our systematic literature review (Table 1); 20/22 cases (90.91%) experienced dysmenorrhea [1–5, 7–17], 4/22 reported (18.18%) miscarriage [4, 6, 8, 18], and 2/22 cases (9.09%) experienced infertility [14]. 10/22 cases (45.5%) were diagnosed before age 20. 8/22 cases (36.36%) of endometriosis have been documented in patients performed laparoscopy or laparotomy [3–6, 8, 11, 15]. Gupta et al. proposed that endometriosis may be associated with menstrual blood reflux in Robert’s uterus [3]. In our case, after resecting the right fallopian tube, the blind cavity was visibly completely disconnected from outside the uterus. As a result, increased pressure was associated with the retention of menstrual blood in the closed right-sided uterine cavity, which induced worsened abdominal pain.

Ultrasonography, MRI, hysteroscopy, and laparoscopy have been performed to diagnose the Robert’s uterus. The septum, with asymmetrical uterine cavities, is easily discernible, and hematometra and hematosalpinx are easy to identify with noninvasive imaging modalities, such as 3D-US and MRI, which are considered reliable modalities to examine uterine morphology [14, 19]. Even 3D-US are considered better than MRI in diagnosis of Robert’s uterus for its frugality and easy availability [4]. In the literature,

| Authors, (year) | Case | Age (years) | Dysmenorrhea | Other complaints | Hematometra | Other complication |
|----------------|------|-------------|--------------|-----------------|-------------|-------------------|
| Deenadayal M 2021 [4] | 5 | 28.2 (M) (13–39) | Yes (5/5) | Recurrent miscarriage (1/5), infertility (1/5) | Yes (4/5) | Endometriosis (2/5) |
| Zhang J. 2021 [5] | 1 | 24 | Yes | – | Yes | Hematosalpinx, Endometriosis |
| Liu Y. 2021 [6] | 1 | 45 | No | Abnormal menstruation, miscarriage | No | Blind hemi-cavity Pregnancy, Adenomyosis |
| Liu Y. 2020 [7] | 1 | 16 | Yes | Abdominal pain | Yes | Ipsilateral renal agenesis, Hematosalpinx |
| Yang QM. 2019 [8] | 1 | 23 | Yes | Miscarriage | Yes | Blind cavity pregnancy, Ipsilateral renal agenesis, Adenomyosis |
| Shah N. 2019 [1] | 1 | 16 | Yes | – | Yes | – |
| Kiyak H. 2018 [9] | 1 | 15 | Yes | – | Yes | – |
| Biler A. 2017 [10] | 1 | 29 | Yes | Abdominal pain, abnormal menstruation | Yes | – |
| Mittal P. 2017 [11] | 1 | 15 | Yes | – | Yes | Hematosalpinx, Endometriosis |
| John SK. 2017 [12] | 1 | 16 | Yes | Abdominal pain | Yes | – |
| Ludwin A. 2016 [13] | 1 | 22 | Yes | – | Yes | – |
| Di Spiezio SA. 2015 [14] | 1 | 30 | Yes | Infertility | Yes | Uterine myomas, A pseudo-cystic lesion |
| Li J. 2015 [15] | 1 | 26 | Yes | – | Yes | Ovarian endometriotic cyst, Endometriosis |
| Maddukuri SB. 2014 [16] | 1 | 16 | Yes | – | Yes | Hematosalpinx |
| Vural M. 2011 [17] | 1 | 24 | Yes | – | Yes | – |
| Capito C. 2009 [2] | 1 | 15 | Yes | Abdominal pain | Yes | – |
| Gupta N. 2007 [3] | 1 | 19 | Yes | Abdominal pain | Yes | Ovarian endometriotic cyst, Endometriosis |
| Singhal S. 2002 [18] | 1 | 20 | No | Miscarriage | No | Blind cavity pregnancy |
19/22 cases (86.36%) were identified hematometra [1–5, 7–17]. Ludwin et al. classified Robert’s uterus into three types according to the amount of bleeding in the blind cavity: no hematocoele, small hematocoele, and large hematocoele [20]. In addition, we strongly recommend a detailed inspection of the urinary system in similar cases, considering that two cases of ipsilateral renal agenesis have been reported [7, 8]. To the best of our knowledge, our patient is the first case of Robert’s uterus in combination with intrauterine adhesions, which was verified by hysteroscopy. Our patient had no history of intrauterine operation, such as induced abortion, which may be related to obstruction of menstrual blood outflow and inflammation, and there was no evidence of endometritis. In our opinion, early diagnosis and hysteroscopic surgery may reduce the incidence of these complications.

However, the early diagnosis of uterine malformation by ultrasonography or MRI is significantly affected by the examiner’s skill level. Successful management depends on accurate pre-operative diagnosis. In the literature, 5/22 cases (22.73%) underwent re-operation or a third surgery before diagnosis and management [3, 4, 6, 7, 12]. Some of the Robert’s uterus have normal external uterine contour [12], making it difficult to identify by laparoscopic surgery. We recommend that if the intraoperative findings are inconsistent with the clinical manifestation of severe abdominal pain, uterine inspection via hysteroscopy is needed. Patient trauma can be reduced with adequate evaluation and determining the pre-operative diagnosis.

Surgical treatment is the only recommendation and surgical options should be determined by the patient’s age and fertility desires. The primary surgical goals are draining the hematometra and preventing its recurrence through septal resection [11]. Laparoscopic septal resection and metroplasty are considered, for the minimal invasiveness, especially for adolescent girls. 21 of the 22 Robert’s uterus underwent surgery (Table 2). In 13/21 cases, septal resection was performed by laparoscopy, hysteroscopy, or laparotomy [1, 3–5, 7–10, 12–16]. One case underwent hysterectomy due to lack of fertility requirement [4]. Hysteroscopic septal resection and laparoscopic guidance are the main option for the surgical treatment with the minimal invasive [1, 8, 10, 14], laparoscopy could identify the hematosalpinx, pelvic adhesions, and endometriosis, which cannot be evaluated by auxiliary examination, and, remarkably, hysteroscopic surgery performed by experienced surgeons is essential for the complicated environment in uterus; It has been reported that a patient whose Robert’s uterus underwent hysteroscopic treatment combined with the laparoscopic instrument was successfully pregnant and gave birth [15]. Summary of the statistical characteristics is from the literature review (Table 3). 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 about the

| Table 2 Operative procedures for Robert’s uterus cases reported in the literature |
|----------------------------------|--------|----------------------------------|----------------|
| Authors (year)                   | Side   | Main surgical treatment                      | Number of operations |
| Deenadayal M 2021 [4]            | 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 [5]                | Right  | Hysteroscopic septal resection and laparoscopic oophorectomy | One |
| Liu Y. 2021 [6]                  | Right  | Hysterectomy and right salpingectomy         | Two |
| Liu Y. 2020 [7]                  | Right  | Hysteroscopic septal resection and laparoscopic right salpingectomy | Two |
| Yang QM. 2019 [8]                | Right  | Hysteroscopic septal resection and laparoscopic guidance | One |
| Shah N. 2019 [1]                 | Left   | Hysteroscopic septal resection and laparoscopic guidance | One |
| Kiyak H. 2018 [9]                | Right  | Laparoscopic septal resection               | One |
| Biler A. 2017 [10]               | Right  | Hysteroscopic septal resection and laparoscopic guidance | One |
| Mittal P. 2017 [11]              | Left   | Laparotomy excision of the blind cavity     | One |
| John SK. 2017 [12]               | Right  | Laparotomy septal resection                 | Three |
| Ludwin A. 2016 [13]              | Left   | Hysteroscopic septal resection              | One |
| Di Spiezie SA. 2015 [14]         | Left   | Hysteroscopic septal resection and Laparoscopy guidance | One |
| Li J. 2015 [15]                  | Left   | Laparotomy septal resection and laparoscopic oophorectomy | One |
| Maddukuri SB. 2014 [16]          | Left   | Laparotomy septal resection                 | One |
| Vural M. 2011 [17]               | Left   | Laparotomy endometrectomy of the blind cavity | One |
| Capito C. 2009 [2]               | –      | Laparotomy endometrectomy of the blind cavity | One |
| Gupta N. 2007 [3]                | Right  | Laparotomy septal resection                 | Two |
| Singhal S. 2002 [18]             | Right  | Laparotomy                                 | One |
need of assisted reproduction. Monitoring the situation of pregnancy and appropriate intervention to achieve better pregnancy outcomes if required.

In conclusion, Robert’s uterus is a congenital abnormality that has an oblique septum and non-communicating asymmetrical uterine hemi-cavity, and the case report and literature review providing detailed clinical features contribute to improving the diagnosis and treatment. Despite the strengths, diagnosing preoperatively remains a challenge for its rarity and the high risk of misdiagnosis, and thus, more studies need to be investigated.

Acknowledgements We thank Jane Charbonneau, DVM, from Liwen Bianji (Edanz) (www.liwenbianji.cn/), for editing the English text of a draft of this manuscript.

Author contributions MY conceived the case report, participated in the surgery, and drafted and edited the manuscript. KG reviewed the literature and drafted the manuscript. ZH participated in the literature review and edited the manuscript. JZ participated in the surgery and drafted and edited the manuscript. All authors read and approved the final manuscript.

Funding Not applicable.

Availability of data and materials The data presented in this study are available within the article.

Declarations

Conflict of interest The authors declare that they have no conflict of interest.

Ethical approval We have obtained informed approval from ethics institutional from Institutional Review Board, First Hospital, Jilin University. We have obtained informed consent from the patient, and this is a noninvasive follow-up observational study.

References

1. Shah N, Changede P (2020) Hysteroscopic management of Robert’s uterus. J Obstet Gynaecol India 70(1):86–88. https://doi.org/10.1007/s13224-019-01208-4
2. Capito C, Sarnacki S (2009) Menstrual retention in a Robert’s uterus. J Pediatr Adolesc Gynecol 22(5):e104-106. https://doi.org/10.1016/j.jpag.2008.03.004
3. Gupta N, Mittal S, Dadhwal V, Misra R (2007) A unique congenital mullerian anomaly: Robert’s uterus. Arch Gynecol Obstet 276(6):641–643. https://doi.org/10.1007/s00404-007-0389-2
4. Deenadayal M, Günther V, Alkatout I, Freytag D, Deenadayal-Mettler A, Deenadayal Tolani A, Sinha R, Mettler L (2021) Critical Role of 3D ultrasound in the diagnosis and management of Robert’s uterus: a single-centre case series and a review. Facts Views Vis Obgyn 13(1):41–49. https://doi.org/10.5205/FVVO.13.1.008
5. Zhang J, Zhou W, Tang Y, Tan S, Qiao L (2021) Robert’s uterus with delayed diagnosis and potential consequences: a case report. J Int Med Res 49(3):300060521999531. https://doi.org/10.1177/0300060521999531
6. Liu Y, Wang S, Hong Y, Wang J, Niu J, Li X, Li H, Wang Y (2021) Pregnancy in the blind hemi-cavity of Robert’s uterus: a case report. Radiol Case Rep 16(5):1085–1088. https://doi.org/10.1016/j.radcr.2021.02.013
7. Liu Y, Yang Y, Duan J, Liu L, Zhang W, Wang Y (2020) Favorable pregnancy outcome for a patient with Robert’s uterus, bicornuate uterus, and ipsilateral renal agenesis. Int J Gynaecol Obstet 151(2):302–303. https://doi.org/10.1007/jogo.13274
8. Yang QM, Li H, He SH, Chen D, Chen L (2019) Pregnancy in a blind hemi-cavity of Robert’s uterus with ipsilateral renal agenesis: a case report and literature review. J Int Med Res 47(7):3427–3434. https://doi.org/10.1177/0300060519850422
9. Kiyak H, Karacan T, Wetherilt LS, Seckin KD, Ozuyrek ES (2018) Laparoscopic blinded endometrial cavity resection for Robert’s uterus. J Minim Invasive Gynecol 25(2):340. https://doi.org/10.1016/j.jmig.2017.09.003
10. Biler A, Akdemir A, Peker N, Sendag F (2018) A rare uterine malformation: asymmetric septate uterus. J Minim Invasive Gynecol 25(1):28–29. https://doi.org/10.1016/j.jmig.2017.06.015
11. Mittal P, Gupta R, Mittal A, Taneja A, Sekhon PS, Gupta S (2017) Magnetic resonance imaging (MRI) depiction of Robert’s uterus: a rare mullerian duct anomaly presenting with cyclical pain in young menstruating woman. Pol J Radiol 82:134–136. https://doi.org/10.12659/PJR.900436
12. John SK, Prabhu PS, Virmani S, Kumar V, Thotan SP (2017) Misdiagnosed Roberts uterus leading to surgical misadventures. J Pediatr Adolesc Gynecol 30(4):508–510. https://doi.org/10.1016/j.jpag.2017.01.005
13. Ludwin A, Ludwin I, Martins WP (2016) Robert’s uterus: modern imaging techniques and ultrasound-guided hysteroscopic treatment without laparoscopy or laparotomy. Ultrasound Obstet Gynecol 48(4):526–529. https://doi.org/10.1002/uog.15976
14. Di Spiezio SA, Giampaolino P, Scognamiglio M, Varelli C, Nazzaro G, Mansueto G, Nappi C, Grimbizis GF (2016) An exceptional case of complete septate uterus with unilateral cervical aplasia (Class U2bC3V0/ESHRE/ESGE classification) and isolated mullerian remnants: combined hysteroscopic and laparoscopic treatment. J Minim Invasive Gynecol 23(1):16–17. https://doi.org/10.1016/j.jmig.2015.09.006

15. Li J, Yu W, Wang M, Feng LM (2015) Hysteroscopic treatment of Robert’s uterus with laparoscopy. J Obstet Gynaecol Res 41(9):1491–1494. https://doi.org/10.1111/jog.12735

16. Maddukuri SB, Karegowda LH, Prakashini K, Kantipudi S (2014) Robert’s uterus: a rare congenital mullerian duct anomaly causing haematometra. BMJ Case Rep 10:16. https://doi.org/10.1136/bcr-2014-204489

17. Vural M, Yildiz S, Cece H, Camuzcuoglu H (2011) Favourable pregnancy outcome after endometrectomy for a Robert’s uterus. J Obstet Gynaecol 31(7):668–669. https://doi.org/10.3109/01443615.2011.593653

18. Singhal S, Agarwal U, Sharma D, Sirohiwal D (2003) Pregnancy in asymmetric blind hemicavity of Robert’s uterus—a previously unreported phenomenon. Eur J Obstet Gynecol Reprod Biol 107(1):93–95. https://doi.org/10.1016/s0301-2115(02)00238-5

19. Ludwin A, Martins WP, Ludwin I (2017) Uterine cavity imaging, volume estimation and quantification of degree of deformity using automatic volume calculation: description of technique. Ultrasound Obstet Gynecol 50(1):138–140. https://doi.org/10.1002/uog.15890

20. Ludwin A, Ludwin I, Bhagavath B, Lindheim SR (2018) Pre-, intra-, and postoperative management of Robert’s uterus. Fertil Steril 110(4):778–779. https://doi.org/10.1016/j.fertnstert.2018.05.033

Publisher’s Note Springer Nature remains neutral with regard to jurisdictional claims in published maps and institutional affiliations.