Case Report

Robert's Uterus versus Juvenile Cystic Adenomyoma – Diagnostic and Therapeutic Challenges – Case Report and Review of Literature

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Abstract

This case report highlights the diagnostic dilemma and therapeutic challenges encountered while managing adolescent girls with progressive dysmenorrhoea and management of Robert's uterus. Two girls aged 20 years and 13 years presented with severe progressive dysmenorrhoea. In the first case, laparoscopy revealed juvenile cystic adenomyoma (JCA) of 3 cm × 3 cm on the left side anteroinferior to the round ligament. Laparoscopic resection of the lesion was done, and histopathology revealed features of adenomyosis. In the second case, there was a globular enlargement of the right half of the uterine body with round ligament and adnexa attached to the lesion (Robert's uterus). In view of severe symptoms, complete resection of the lesion and partial resection of hemi-uterus was done, followed by myometrial defect closure. Both cases were initially diagnosed as JCA, and the final diagnosis was made on laparoscopy. Both girls had complete symptomatic relief from the next menstrual cycle and have been under follow-up for 24 months and 18 months, respectively. Due to the rarity of conditions, Robert's uterus and JCA are usually misdiagnosed with each other or with other Mullerian anomalies such as a non-communicating unicornuate uterus. Radiologists and clinicians should be aware of these different pathologies causing similar symptoms. Understanding the pathology, early diagnosis, timely referral and correct surgical procedure are emphasised to improve reproductive outcomes.

Keywords: *Accessory and cavitated uterine mass (ACUM), juvenile cystic adenomyoma, Robert's uterus, severe dysmenorrhoea*

Introduction

The prevalence of congenital uterine malformations is approximately 5.5%–6.7% in the general population and higher in women with infertility and recurrent pregnancy losses.[1] Although most of the anomalies are well-researched, some anomalies are misclassified due to wide variations in clinical presentation and radiological findings. The prevalence of septate uterus in the general population is 2.3%. Robert's uterus, a rare variant of the asymmetric septate uterus, was first reported by Robert in 1970.[2] The number of cases of Robert's uterus reported in the literature are few and not well-known amongst radiologists and clinicians.

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The exact embryological defect in Robert's uterus is debatable with two schools of thought; segmental agenesis of the uterine isthmus with a persistent septum between upper portions of Mullerian ducts. Some authors believe it is due to unilateral cervical aplasia.[3]

Cystic Mullerian anomalies have been termed as juvenile cystic adenomyoma (JCA) by Takeuchi *et al*. in 2010[4] and accessory cavitated uterine malformations (ACUM) by Acién *et al*.[3] JCA/ACUM represent Mullerian variants located at or below the

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level of insertion of round ligament. Although unclear, the pathophysiology is considered to be ectopia or duplication and persistence of ductal Mullerian tissue in a critical area at the level of the attachment of the round ligament.[5] The criteria proposed for diagnosing JCA include age <30 years; cystic lesion >1 cm in diameter, independent of the uterine cavity and covered by hypertrophic myometrium, as seen on radiologic images and association with severe dysmenorrhoea.[4,6]

The latest ASRM Mullerian Anomalies Classification includes Robert's uterus but does not include JCA/accessory cavitated uterine malformations (ACUMs).[7] Due to the similar clinical features of these entities, the diagnosis is complex. It may lead to inappropriate surgeries (haematometra drainage for temporary relief or complete removal of affected hemi-uterus) and adverse reproductive outcomes.[3]

We present two cases of severe progressive dysmenorrhoea diagnosed as JCA on magnetic resonance imaging (MRI). One of these was found to be Robert's uterus on laparoscopy. A review of the literature, diagnostic algorithm, differentiating features of different obstructive Mullerian anomalies and various surgical options has been discussed in this article.

Materials and Methods

For the literature review, we searched PubMed and Google Scholar using the following terms: Robert's uterus, rare Mullerian anomaly and obstructive hemi-uterine anomaly. Case reports and case series of Robert's uterus were included if they met the diagnostic criteria for Robert's uterus as described by Ludwin *et al*.[14] Cases misreported as Robert's uterus were excluded from the study. All articles which fit into the inclusion criteria and were published till December 2021 in the English language were included in the study. Informed written consent was taken for both cases reported in the manuscript.

Case Report

Case 1

A 20-year-old female presented to the outpatient department (OPD) with progressive dysmenorrhoea for the past 2 years. The lower abdominal pain affected mainly the left side started 1 day before menses and lasted for 8–10 days. She attained menarche at 13 years and had regular cycles. Initially, there was no dysmenorrhoea, but for the past 2 years, she gradually developed severe congestive dysmenorrhoea requiring parenteral analgesics. On examination, the abdomen was soft with no tenderness or palpable organomegaly. Transabdominal two-dimensional (2D) ultrasonography (USG) revealed a 3 cm × 3 cm adenomyoma present on the left side of the uterus, not communicating with the cavity. MRI revealed a left-sided uterine adenomyoma 3 cm × 3 cm anteroinferior to the left round ligament containing hypodense material [Figure 1a]. With a provisional diagnosis of JCA, a laparoscopic adenomyomectomy was planned. A left-sided adenomyoma was visualised intraoperatively, located anteroinferior to the left round ligament [Figure 1b]. During the dissection, chocolate-coloured fluid was seen coming out of the lesion. Bilateral tubes and ovaries were attached normally [Figure 1c]. The patient is doing well with complete resolution of symptoms for >24 months.

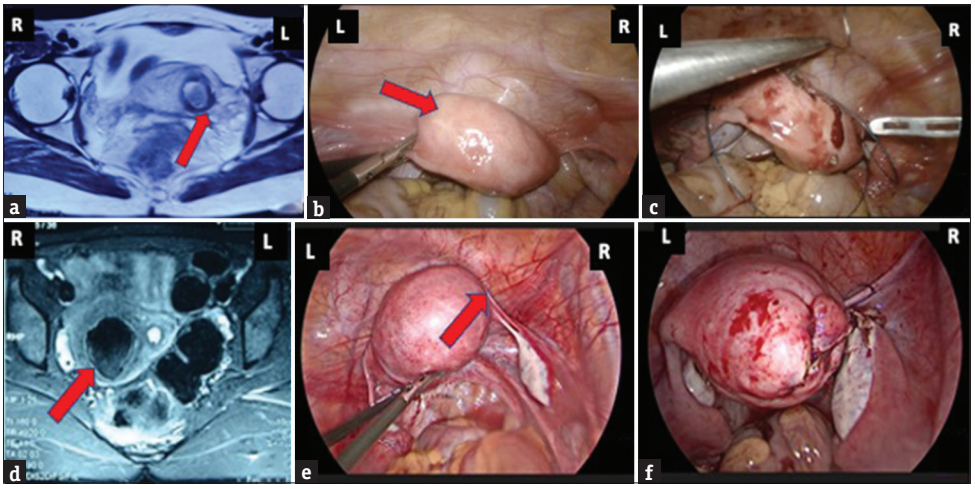

Figure 1: (a) MRI picture showing JCA (marked red arrow), (b) Laparoscopic view showing JCA lesion below left round ligament, (c) Removal of the lesion followed by myometrial defect closure. Normal endometrial cavity is intact with retained both tubal attachments to the uterus, (d) MRI picture showing lesion adjacent to the endometrial cavity, (e) Laparoscopic view showing right-sided blind hemicavity (Robert's uterus), (f) Final picture after removal of the blind uterine horn, myometrial closure, and ovarian plication. JCA = Juvenile cystic adenomyoma, MRI = Magnetic resonance imaging

Case 2

A 13-year-old girl presented to OPD with complaints of severe progressive dysmenorrhoea since menarche. She attained menarche at the age of 11 years and had regular menstrual cycles. Following an episode of acute abdomen 1 year back, she was clinically diagnosed with acute appendicitis at a local hospital. Emergency laparotomy was performed for the presumed diagnosis of appendicitis, but the appendix was normal in appearance. However, her symptoms did not improve, and she dropped out of school due to the increasing severity of dysmenorrhoea. On examination, the abdomen was soft with no tenderness or organomegaly. Transabdominal 2D USG revealed a 4.5 cm × 3.3 cm lesion in the right uterine myometrium indenting the endometrial cavity. Bilateral ovaries were normal. MRI showed a heterogeneous lesion of size 5.4 cm × 4.5 cm in the right lateral wall of the uterus abutting the adjacent junctional zone medially, not communicating with the endometrial cavity [Figure 1d]. A provisional diagnosis of JCA was made, and the patient was posted for surgery. However, on laparoscopy, a bulge was noted on the right side of the uterus with the right round ligament, the fallopian tube and the ovary attached to the lesion [Figure 1e]. An intraoperative diagnosis of the blind uterine horn/ Robert's uterus was made. Hysteroscopy revealed a tubular cavity with a single ostium seen on the left side. In view of the severe symptoms affecting her quality of life, it was decided to excise the mass completely. The incision was given at the junction of the normal uterus and lesion with utmost care to leave a thick myometrial margin. Myometrial closure was done using 1-0 V-Loc suture, and ovarian plication was done using a number 1 Vicryl suture [Figure 1f]. The patient had an uneventful post-operative recovery. On post-operative follow-up, the patient is doing well and has been having regular periods with no dysmenorrhoea for the last 18 months and has started going to school again.

Discussion

The present cases describe the challenges in the clinical diagnosis and management of Robert's uterus and its differentiation from other obstructuive uterine anomalies. Although MRI being the gold standard imaging modality, the misdiagnosis of Robert's uterus as JCA in the present case may be due to low awareness about both pathologies.

Amongst most of Robert's uterus cases reported in the literature so far [Table 1], it has been misdiagnosed as a unicornuate uterus with a non-communicating horn with haematometra due to a similar clinical presentation in most of the cases. Few cases have been misdiagnosed as JCA/ACUM as in our case,[13,22-24] and most of the cases have been managed as resection of blind horn either due to lack of awareness about the entity or due to severe symptoms or large lesions. The anomaly may go either unrecognised till the patient presents with some pregnancy-related complications[8] or undergo unindicated surgeries such as appendectomy as in our case.[17]

Table 2 describes the salient differentiating features of obstructive uterine anomalies. Figure 2 illustrates the evaluation algorithm for girls presenting with severe progressive dysmenorrhoea. Surgical management is recommended for complete cure in these obstructive anomalies, and laparoscopy gives the advantage of confirmation of diagnosis and surgical therapeutic intervention in the same sitting. As described in Figure 2, findings on imaging and laparoscopy are usually clear in cases of rudimentary functional horn, and difficulty may arise while differentiating JCA and Robert's uterus. Hysteroscopy may be combined with

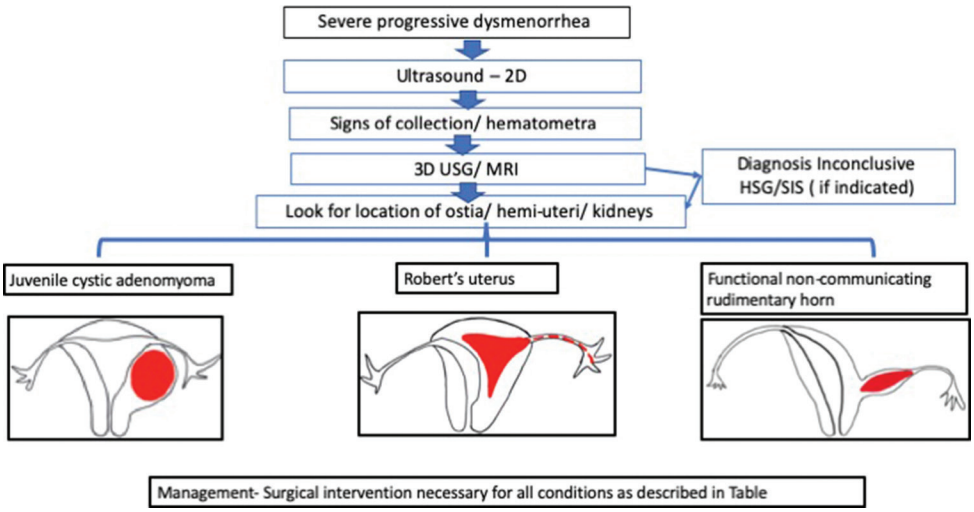

Figure 2: Evaluation algorithm and schematic diagram showing different obstructive uterine anomalies in women with progressive dysmenorrhoea

Table 1: Review of literature related to Robert's uterus

Author, year	*n*	Age	Presenting complaints	Associated	Imaging	Diagnosis	Side of lesion	Interventions done	Outcomes
Singhal *et al.*, 2003[8]	1	18	Pregnancy in blind hemicavity with failed MTP attempts	No	USG	Non-communicating horn pregnancy	Right	Hysterotomy + CT + ipsilateral tubal ligation	Not complete
Gupta *et al.*, 2007[9]	1	19	Intractable dysmenorrhea with past history of laparoscopy	Endometriosis	USG, MRI	Unicornuate uterus with functional non-communicating rudimentary horn	Right	Laparotomy + adhesiolysis + metroplasty + endometriotic cystectomy	Complete
Rani *et al.*, 2009[10]	1	21	Pregnancy with failed attempts of MTP	No	2D US	Non-communicating rudimentary horn	Left	Hysterotomy + hemi-uterine excision	Complete
Vural *et al.*, 2011[11]	1	24	Dysmenorrhoea with miscarriages	Endometriosis	MRI	Non-communicating rudimentary horn	Left	Hysterotomy + endometrectomy	Complete
Bhuyar and Deshmukh 2014[12]	1	18	Dysmenorrhoea	Endometriosis	2D US	Functional non-communicating rudimentary horn; previous history of laparotomy	Left	Laparotomy + hemi-uterine excision + ipsilateral salpingo-oophorectomy	Complete
Li, 2015[13]	1	26	Dysmenorrhoea with 1 miscarriage	Endometriosis	US	Cystic uterine intramural collection	Left	Hysteroscopic metroplasty	Complete
Ludwin *et al.*, 2016[14]	1	22	Dysmenorrhoea progressed to chronic pelvic pain	No	3D US, sonohystero AVC	Robert's uterus-correct	Left	USG-guided hysteroscopic metroplasty twice with weekly balloon adhesiolysis till 8 weeks	Correct
Ramesh *et al.*, 2016[15]	2	17, 19	Dysmenorrhoea	Endometriosis	USG, MRI	Non-communicatng rudimentary horn	Left	Laparoscopic hemi-uterine excision in one case and hysteroscopic metroplasty in another case	Complete
Kiyak *et al.*, 2018[16]	1	15	Dysmenorrhoea	Not mentioned	MRI	Robert's uterus	Right	Laparoscopic endometrectomy	Complete
John *et al.*, 2017[17]	1	16	Cyclical dysmenorrhoea		US, MRI	Robert's uterus (history of appendicectomy and diagnostic laparoscopy)	Right	Exploratory laparotomy + metroplasty	Complete
Mittal *et al.*, 2017[18]	1	15	Dysmenorrhoea	Endometriosis	MRI	Robert's uterus	Left	Laparotomy + left hemi-uterine excision + left endometriotic cystectomy	Complete
Yang *et al.*, 2019[19]	1	23	Pregnancy in blind cavity	Ipsilateral renal agenesis	USG, MRI	Robert's uterus	Not mentioned	Hysteroscopic metroplasty under lap + USG guidance	Complete
Shah and Changede 2020[20]	1	16	Dysmenorrhoea	No	MRI	Robert's uterus	Left	Laparoscopy + hysteroscopic metroplasty	Complete
Kisu *et al.*, 2021[5]	1	16	Lower abdominal pain, progressive dysmenorrhoea	Nothing significant	US, MRI, HSG	Non-communicating rudimentary horn	Left	Laparoscopic excision of hemi-uterus + salpingectomy	Complete
Deenadayal *et al.*, 2021[21]	5	13, 25, 28, 36, 39	Dysmenorrhoea, infertility	Endometriosis	2D-US, 3D-US, MRI	Robert's uterus	Left in all	Endometrectomy, hysteroscopic septal resection, laparoscopic horn excision, laparotomy and hysterectomy, lost to follow up	Complete

Contd...

Table 1: Contd...

Author, year	*n*	Age	Presenting complaints	Associated	Imaging	Diagnosis	Side of lesion	Interventions done	Outcomes
Zhang *et al*., 2021[22]	1	24	Dysmenorrhoea	Endometriosis	2D US, 3D US, MRI	Robert's uterus	Right	Laparoscopy + hysteroscopic septal resection	Complete
Nigam *et al*., 2021[23]	1	30	Dysmenorrhoea (3 laparotomies for the same complaint)	Endometriosis	MRI	Cystic adenomyosis/obstructed rudimentary horn	Right	Laparotomy + hemi-uterine excision	Complete
Protopapas *et al*.[24]	1	14	Severe progressive dysmenorrhoea	No	MRI	JCA/blind uterine horn	Left	Laparoscopy + hemi-uterine excision	Complete
Zhang *et al*., 2021[22]	1	17	Dysmenorrhoea	Endometriosis with hematosalpinx	B mode USG	Non-communicating rudimentary horn	Right	Hysteroscopic metroplasty	Complete
Ballabh *et al*., 2021[25]	4	44, 18, 36, 21	Dysmenorrhoea	Endometriosis	MRI	Robert's uterus	Left	Not mentioned	
Liu *et al*., 2021[26]	1	45	Pregnancy in blind horn with failed MTP	No	MRI Abandoned laparoscopy	Non-communicating rudimentary horn	Right	Laparotomy + hysterectomy + right salpingectomy	Complete
Present case	1	13	History of minilaparotomy and appendectomy	No	MRI	JCA	Right	Laparoscopic partial hemihysterectomy	Complete

MTP=Medical termination of pregnancy, USG=Ultrasonography, MRI=Magnetic resonance imaging, US=Ultrasound, AVC=Automated volume calculation, HSG=Hysterosalpingography, CT=Computed tomography, JCA=Juvenile cystic adenomyoma, 2D=Two-dimensional, 3D=Three-dimensional

Table 2: Salient features of obstructive uterine anomalies presenting with progressive dysmenorrhea

Traits	JCA/ACUM	Robert's uterus	Unicornuate uterus with functional non-communicating rudimentary horn
Age at presentation	Adolescent to reproductive age group depending upon functionality of the endometrium		
Clinical features	Progressive dysmenorrhea	Progressive dysmenorrhea	Progressive dysmenorrhea
Site of defect/etiology			
Etiology	Persistence of Mullerian remnant	Resorption defect	Fusion defect
Uterine cavity	Accessory cystic lesion in myometrium inferior to round ligament	Asymmetrically separated by an oblique septum	2 separate uterine bodies of different sizes
Isthmus	Well developed	Well developed	Absent on the rudimentary half
Cervix	Well developed	Ipsilateral cervical aplasia	Absent on the rudimentary half
Vagina	Normal	Septum may or may not be present	Normal
Imaging			
HSG/SSG	Normal uterine cavity Bilateral fallopian tubes will be delineated	Only one uterine horn and ipsilateral fallopian tube will be opacified/delineated Affected side of hemiuterus and fallopian tube will not be delineated	Only one uterine horn will be opacified/delineated along with ipsilateral fallopian tube. Affected side of hemiuterus and fallopian tube will not be delineated
Intra-operative findings			
Laparoscopy	Normal uterine fundus and cornua, normal tubes and ovaries; separate thick walled cystic lesion in myometrium below and lateral to round ligament	Broad fundus (external indentation<1 cm) with bulge on obstructed side at the level of tubal attachment with/without ipsilateral hemato-salpinx and endometriosis	Two different fundi with separate cavities and ostia (external indentation>1cm) with/without ipsilateral hematosalpinx and/or endometriosis
Chromopertubation	Bilateral fallopian tubes will be communicating with endometrial cavity	Only one fallopian tube will be communicating with endometrial cavity	Only one fallopian tube will be communicating with endometrial cavity
Hysteroscopy	Both fallopian tube ostia will be visible	Only one fallopian tube ostium will be visible	Only one fallopian tube ostium will be visible
Treatment	Adeno-myomectomy	Hysteroscopic metroplasty – first and ideal choice Endometrectomy Tompkin's metroplasty Hemi-uterine excision – last option	Rudimentary horn excision with ipsilateral salpingectomy

JCA=Juvenile cystic adenomyoma, HSG=Hysterosalpingography, SSG=Sonosalpingography, ACUM=Accessory cavitated uterine mass, JCA=Juvenile cystic adenomyoma

laparoscopy, confirming the diagnosis as a single ostium will be visualised in Robert's uterus while both ostia will be visualised in JCA.

The triad of Robert's uterus comprises (1) Unicornuate uterus, (2) contralateral blind hemicavity with ± haematometra and (3) normal uterine fundus ± slight external indentation [Figure 1]. Three types of Robert's uterus have been described by Ludwin *et al*.[27] based on the functionality of the contralateral cavity and its size. In Type 1 Robert's uterus (typical Robert's uterus), hemi-uterus is fully functional and compresses on the adjacent cavity. Type 1 usually presents early in an adolescent age group with severe progressive dysmenorrhoea. Type 2 Roberts uterus has a non-functional hemi-uterus and presents relatively late with recurrent pregnancy loss or infertility and is usually diagnosed as a unicornuate uterus. Type 3 Robert's uterus has a small collection because of residual functioning endometrial tissue in the hemi-uterus. Type 3 might present in the early reproductive age group with progressive dysmenorrhoea and endometriosis. Despite this clear theoretical classification, misdiagnosis is common due to low awareness about this Mullerian anomaly.[28]

Surgical options for Robert's uterus include hysteroscopic resection of the intervening septum with accompanying dilation, endometrectomy and myometrial closure and hysterectomy.[11,16] The choice of surgery for Robert's uterus depends upon age, parity, the severity of symptoms, previous obstetric history, future fertility

desires, willingness for follow-up (for repeat intervention, if needed) and surgical expertise of the surgeon.[27] In cases of severe symptoms with a large adenomyotic lesion and thick muscular septum, excision of the blind hemicavity and haematometra (endometrectomy) followed by myometrial closure is appropriate.[28] Care should be taken to leave sufficient myometrial tissue to support the remaining hemi-uterus. The ipsilateral fallopian tube should be removed if the haematometra has extended to the fallopian tube and/or the tube is damaged/swollen. Hence, if it is decided for ipsilateral salpingectomy, judicious use of cautery is emphasised to avoid any thermal insult to the ovary. Hysteroscopic metroplasty has been used by some authors as a cavity expansion procedure that involves resection of the septum[13,15,19,20,26,29] under USG guidance, followed by anti-adhesiolysis therapy such as hormone replacement therapy, intrauterine contraceptive device or sequential balloon therapy.[14] Septal thickness might play a crucial role in hysteroscopic metroplasty as thick and muscular septa are difficult to cut and may cause a recurrence of symptoms. MRI and/or three-dimensional USG might help surgeons regarding septal thickness and the location of the weakest point for choosing the right approach to metroplasty. Further long-term data on follow-up are still lacking after hysteroscopic metroplasty, and its benefit in terms of cavity expansion and reproductive performance and regular follow-up is needed. Furthermore, the procedure is technically challenging, requiring surgical expertise, which might not be available in every set-up.

Conclusion

Diagnosis of Robert's uterus is challenging because of the rarity of the condition and lack of awareness which leads to a significant delay in diagnosis and suboptimal/overaggressive surgical interventions in most cases. Understanding the anatomical defect on MRI and selecting the best surgical procedure should be opted for in every case. Hysteroscopic metroplasty with cavity expansion needs long-term follow-up in terms of symptom relief and future reproductive performance.

Consent

Informed written consent has been taken from both patients for the publication of case details.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patients have given their consent for their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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