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Case Report

Obstetrics and Gynaecology Section

Unicornuate Uterus with Non-communicating Rudimentary Horn: A Case Report

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ABSTRACT

Müllerian malformations are structural anomalies of the female genital tract due to failure of fusion or development of the paramesonephric (Müllerian) ducts. Unicornuate uterus is a rare Müllerian malformation that can have a communicating or a non-communicating horn. A unicornuate uterus can present with symptoms ranging from obstructive menstrual flow during earlier puberty to adverse reproductive outcomes later. Obstructive menstrual flow symptoms can mimic dysmenorrhoea during adolescence and is often misdiagnosed and treated symptomatically with analgesics. Further, there is an undue delay in diagnosis and surgical intervention, which can lead to adverse outcomes in the individuals. Here, we present a case of a 13-year-old adolescent girl with a complaint of severe dysmenorrhoea since menarche. Clinical examination was unremarkable except for tenderness in the lower abdomen. MRI imaging showed a right-sided unicornuate uterus with a left-sided non-communicating rudimentary horn associated with haematometra and haematosalpinx. Rudimentary horn resection led to symptomatic relief This case report highlights the need for early diagnosis and prompt management in providing better clinical outcomes.

Keywords: Adolescent gynaecology, Dysmenorrhoea, Müllerian anomaly

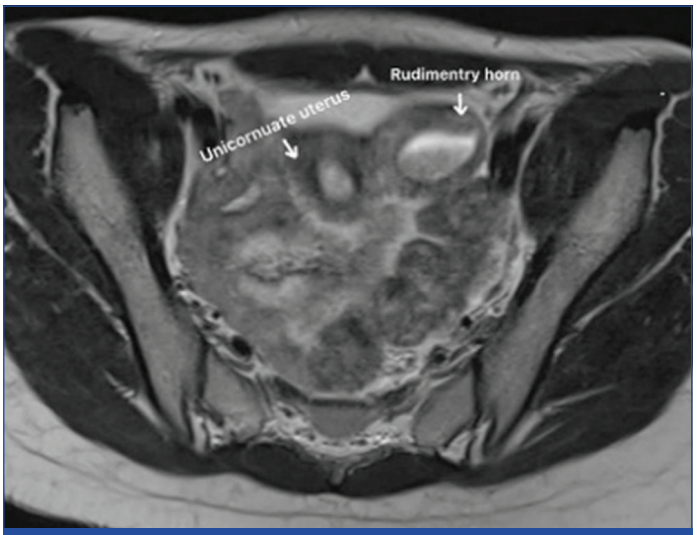
CASE REPORT

A 13-year-old girl presented to the emergency department with complaints of severe lower abdominal pain localised towards the left iliac fossa during her menstrual cycles for the past five months. At the time of presentation, she was on the second day of her menstrual cycle. She had attained menarche five months prior, following which she had two menstrual cycles associated with severe dysmenorrhoea and was managed then with analgesics. The patient experienced cramping pain, with onset a few days before the menses, and persisted even after the bleeding stopped.

She was evaluated at another healthcare facility, where ultrasonography revealed a left adnexal cavitated mass measuring 3.2 × 2.5 cm, isoechoic to the myometrium. The cavity of the lesion contained hyperechoic material measuring approximately 14 mm, suggestive of collected blood. A differential diagnosis of right unicornuate uterus with haematometra in the non-communicating left functional uterine horn or accessory cavitated uterine malformation was made. There were no associated renal abnormalities identified during USG and MRI. However, due to the young age, the parents were counseled to continue with medical management using analgesics and progesterone to suppress menstruation.

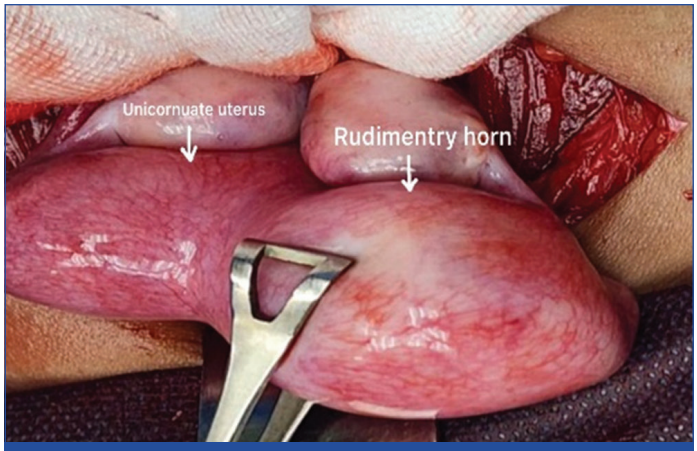
On admission, her vitals were stable her anthropometric measurements were height - 152 cm; weight - 35 kg; body mass index - 15.1 kg/m^2 (underweight); and abdominal examination revealed tenderness in the suprapubic and left iliac fossa region, not associated with any guarding or rigidity. There were no other systemic abnormalities.

Magnetic Resonance Imaging (MRI) revealed a right-sided unicornuate uterus with a left-sided non-communicating rudimentary horn with hematometra and hematosalphinx. The unicornuate uterus measured 5.1×3.1×2.1 cm. The uterus was anteverted, and the standard right horn was in continuity with a single cervix and vagina. The endometrial thickness was normal (7 mm) with a normal junctional layer and myometrium. Further, a non-communicating cavity-like structure with normal endo-myometrial differentiation was noted abutting the left lateral myometrial wall of the right horn with blood fluid level (3.3×2.5×3.1 cm), T1 hyperintense contents pointed out within the tubular tortuous structure, possibly a fallopian tube with hematosalpinx. However, both the ovaries were normal in size and morphology [Table/Fig-1]. Parents were counseled, and with informed consent, the decision to surgically remove the non-communicating functional horn was taken.



[Table/Fig-1]: MRI image revealing right unicornuate uterus with left non-communicating rudimentary. horn

Laparotomy revealed a right unicornuate uterus with an associated distended left rudimentary horn measuring 4×3 cm [Table/Fig-2]. Aspiration of the left horn revealed altered dark coloured blood. The left-side fallopian tube was maldeveloped with a distended isthmic portion having a blind end [Table/Fig-3]. There was no evidence of endometriosis, which was a welcome sign. The right-sided fallopian tube and both ovaries appeared normal. The left rudimentary horn was resected from the right horn and removed. The defect in the unicornuate uterus was closed in two layers with 1-0 vicryl. The resected specimen was sent for histopathological examination,



[Table/Fig-2]: Intraoperative image revealing right unicornuate uterus with left non-communicating rudimentary horn.

[Table/Fig-3]: Intraoperative image revealing the malformed left fallopian tube.

which revealed myometrium containing benign endometrial glands and stroma, thus confirming the diagnosis. There were no intraoperative complications. The reproductive potential was preserved, as the functioning unicornuate uterus with cervix and vagina in continuum was present. The postoperative period was uneventful, and the patient was discharged three days later. She was followed-up on her next menstrual cycle and reported only mild dysmenorrhoea. The patient had an uneventful regular follow-up for six months postsurgery.

DISCUSSION

Congenital developmental malformations of the Uterus, cervix and vagina arise mainly due to fusion defects of the Müllerian or paramesonephric ducts and are prevalent in 4-7% of general population [1]. Various classification systems for Müllerian duct anomalies exist, like American Fertility Society (AFS) criteria (1988), European Society of Human Reproduction and Embryology/ European Society for Gynaecological Endoscopy (ESHRE/ESGE) (2013), Congenital Uterine Malformation by Experts (CUME) criteria (2018), etc., among which the AFS criteria were easy to understand due to the simplified diagrammatic representation of the anomalies [2-4]. In 2021, the American Society of Reproductive Medicine (ASRM) proposed a new classification system called ASRM Müllerian Anomalies Classification 2021 (MAC 2021) to overcome the limitations of the AFS classification and gave nine categories of Müllerian duct anomalies, which were non-numbered but descriptive [5]. They are Müllerian agenesis, cervical agenesis, unicornuate uterus, uterus didelphys, bicornuate uterus, septate uterus, longitudinal vaginal septum, transverse vaginal septum and complex anomalies. The morphological variants of unicornuate uterus according to MAC 2021 include: 1) Unicornuate uterus; 2) Unicornuate uterus with distal atrophic uterine remnant; 3) Unicornuate uterus with distal uterine remnant and functional endometrium 4) Unicornuate uterus with associated atrophic uterine remnant 5) Unicornuate uterus with uterine horn communicating at the level of cervix [Table/Fig-4] [5].

Anomaly	AFS 1988 Classification	ASRM MAC 2021 Classification
Uterine agenesis / hypoplasia	Class I – MRKH syndrome (uterine/vaginal agenesis) only; no subtypes	Includes complete agenesis, hypoplastic uterus, ± functional remnants; details cervico-vaginal status
Unicornuate uterus	Class II – Unicornuate ± rudimentary horn (no standardised subtypes)	Laterality specified; subtypes: communicating horn, non-communicating functional horn, non-functional horn, isolated
Uterus didelphys	Class III – Complete uterine duplication ± double cervix; vaginal status not considered	Distinguishes associated longitudinal vaginal septum ± obstruction; unicollis vs bicollis noted
Bicornuate uterus	Class IV – External contour abnormality; no quantitative criteria	Fundal indentation >1 cm; unicollis or bicollis; ± partial septum; can combine with vaginal/cervical anomalies
Septate uterus	Class V – Defined by internal septum; no measurement criteria; arcuate confusion	Septum >1 cm, angle <90°; subtypes: partial, complete ± cervical/vaginal septa; includes Robert's uterus
Arcuate uterus	Class VI – Considered a separate anomaly	Considered a normal variant; no longer pathologic
DES-related (T-shaped) Uterus	Class VII – T-shaped uterus due to in-utero DES exposure	Not separately classified (historical/iatrogenic)
Cervical anomalies	Not included	New category: agenesis, dysgenesis, duplication, atresia
Vaginal anomalies	Not included	New categories: longitudinal and transverse septa, obstructed/ non-obstructed
Complex / combined anomalies	Not included	New category for rare mixed anomalies (e.g., bicornuate with unilateral cervical/vaginal agenesis)

[Table/Fig-4]: Comparison of Müllerian anomalies as classified by AFS 1988 system and ASRM MAC 2021.

However, our case was of a unicornuate uterus with an associated remnant, which was functional and non-communicating, but we were unable to place the unicornuate uterus variant of our patient into any of the above-given categories of the MAC 2021 classification. This discrepancy was highlighted in a critical review of the MAC 2021 classification by Ludwin A et al., who commented upon the imprecise definition of rudimentary horns and confusion regarding the variants of unicornuate uterus. In their opinion, the rare morphology of unicornuate uterus with a distal functional remnant is listed in MAC 2021, whereas the more common and symptomatic variety of associated functional remnant has not been mentioned [5].

The clinical presentation of a young female with pelvic pain warrants consideration of the following clinical conditions as differential diagnosis: primary dysmenorrhoea, leiomyoma, endometriosis, and adenomyosis apart from Müllerian anomalies [6]. The above clinical conditions were ruled out by history taking, clinical examination, and imaging in the current case being reported.

Unicornuate uterus is commonly associated with urinary tract abnormalities in up to 40% of individuals, of which unilateral renal agenesis is most common [7]. Contemporary case reports on unicornuate uterus tend to be associated with VACTERL and other urogenital abnormalities [8,9]. However, in the current case being reported, no urinary tract abnormalities were associated with Müllerian malformation.

The incomplete unification of the Müllerian ducts during the sixth week of gestation results in the development of a unicornuate uterus. The development of unicornuate uterus can be multifactorial, involving extra and intrauterine factors like infections, exposure to radiation, and teratogenic agents during early pregnancy [10]. In this case, such factors were ruled out through detailed history, and the etiology was considered congenital.

The gynaecological issues faced by individuals with unicornuate uterus include haematometra, haematosalpinx, endometriosis, chronic pelvic pain, dysmenorrhoea, and primary infertility [11]. Further, such individuals experience obstetric complications such as ectopic pregnancy, miscarriage, fetal growth restriction, rupture of the uterus, recurrent fetal malpresentations and preterm labour [11-13]. In our case, the individual has presented with severe pelvic pain and dysmenorrhoea within a few months of attaining menarche.

The reproductive outcomes in unicornuate uterus are plagued by lower live birth rates (47%) and higher rates of miscarriage (42%) [14]. Fortunately, in the case reported, the functioning unicornuate uterus is in continuum with cervix and vagina ensuring reproductive potential. Further, the excision of the rudimentary horn reduces the future possibility of ectopic pregnancy in the horn and the risk of endometriosis.

For precise assessment of the morphology of the unicornuate uterus, 3-dimensional Transvaginal Ultrasound (TVS) and MRI is usually carried out [15]. The evaluation of this individual was carried out by MRI and transabdominal sonography before the procedure.

The surgical management of the non-communicating rudimentary horn involves drainage and medical suppression till hemiresection [16]. However, other surgical approaches, such as unifying the obstructed horn with unicornuate uterus have fallen from usage due to fear of infections and inadequate improvement in obstetric outcomes [17]. In the current case, resection of the rudimentary horn via laparotomy was done, which improved the complaints of severe dysmenorrhoea.

CONCLUSION(S)

Delay in decision making can be debilitating for individuals in the form of dysmenorrhoea and its consequences. Prompt diagnosis and early treatment of Müllerian anomalies improve the reproductive outcomes and quality of life.

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