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Case Report

Pregnancy in rudimentary horn of unicornuate uterus: a rare case

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ABSTRACT

Congenital malformations of the female genital tract result from embryological maldevelopment of Mullerian or paramesonephric ducts. Mullerian duct anomalies (MDAs) are due to agenesis, defective fusion or resorption during embryological development. Unicornuate uterus results due to defective lateral fusion of Mullerian duct. This report discussed a case of pregnancy in rudimentary horn of unicornuate uterus which resulted in rupture of the horn. A patient 35 year old G2A1 with spontaneous conception with 4 months pregnancy came to emergency room with complaints of pain in abdomen and giddiness. On examination her general condition was moderate with pulse rate of 128 bpm, blood pressure of 90/60 mmHg, pallor was present. On per abdomen examination guarding, rigidity and diffuse tenderness was present. On per vaginum examination, uterus size could not be appreciated. Her haemoglobin level was 6 gm%. Ultrasonography of abdomen showed presence of unicornuate uterus with ruptured right horn with fetus in the abdominal cavity and presence of hemoperitoneum. Immediate resuscitative measures were performed, blood transfusion was started and simultaneously patient was shifted to operation theatre for exploratory laparotomy. Intraoperatively hemoperitoneum with unicornuate uterus with non communicating ruptured accessory horn on right side was present and foetus in peritoneal cavity was seen. Ruptured horn was excised and uterus was repaired. The accessory horn and foetus were sent for histopathology examination which was suggestive of placenta increta with gestational hypertrophy and hyperplasia of myometrium with normal tube and ovary. Patient tolerated the procedure well.

Keywords: Unicornuate uterus, Ruptured rudimentory horn, Pregnancy with mullerian anomalies

INTRODUCTION

MDAs are a broad and complex spectrum of defects resulting from maldevelopment of Mullerian or paramesonephric ducts that are often associated with primary amenorrhea, infertility, endometriosis, obstetric complications. Women with MDAs experience a higher incidence of complications during pregnancy and delivery.

The actual prevalence of MDAs is difficult to ascertain because many of the cases are asymptomatic and thus remain undiagnosed. According to most estimates, these anomalies affect between 0.5 and 5.0 percent of the general population. $^{\rm 1}$

In a literature review regarding infertile and fertile women with MDAs, it was found that frequency of specific anomalies were septate uterus (35%), bicornuate uterus (26%), arcuate uterus (18%), unicornuate uterus (10%), didelphys (8%) and agenesis (3%).²

Although some uterine anomalies can cause infertility, most patients with uterine anomalies are able to conceive without difficulty. Uterine anomalies can be associated with perfectly normal reproductive performance, however, the incidences of spontaneous abortion, premature birth, fetal loss, malpresentation, caesarean section, placenta previa, increased need of manual removal of placenta, subinvolution, decreased live births are increased when a uterine anomaly is present.¹⁻³

The pregnancy outcome of patients with bicornuate and septate uterus is poor, with a term delivery rate of only 40% while for unicornuate uterus live birth rate is only 29.2%.

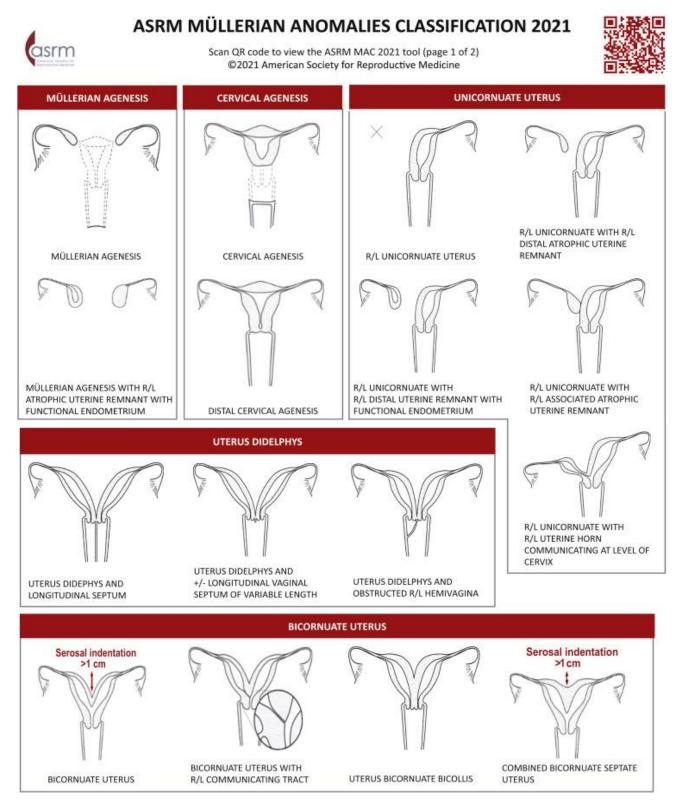


Figure 1: The classification of Mullerian anomalies by American Society of Reproductive Medicine 2021 (ASRM).⁴

In this case report, we discussed a case of unicornuate uterus with rupture of gravid rudimentary horn.

CASE REPORT

The patient, 35 year old G2A1 with 4 months pregnancy came to emergency room with complaints of pain in abdomen and giddiness since morning.



Figure 2: Ultrasonography image.

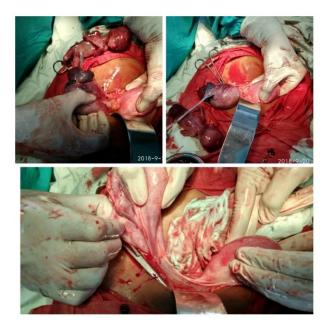


Figure 3: Intra operative images.

The previous one abortion was spontaneous first trimester abortion, wherein no check curettage and ultrasonography were done. This pregnancy was spontaneous conception and was unregistered, unimmunised.

On examination patient's general condition was moderate, pulse rate was 128 bpm, blood pressure 90/60 mmHg, pallor was present. On per abdomen examination, guarding, rigidity and diffuse tenderness was present. On per vaginum examination, cervix os was closed, no bleeding per vaginum and uterus size could not be appreciated.

Her haemoglobin level was 6 gm%, WBC 17000, platelets count was 2.3 lacs.

Diagnosis of acute abdomen was made and initial stabilisation done with volume replacement with IV fluids and blood transfusion.

Urgent ultrasonography of abdomen-pelvis was done which showed presence of unicornuate uterus with ruptured right horn and fetus in the abdominal cavity with presence of hemoperitoneum.

After initial resuscitative measures, simultaneously the patient was shifted to operation theatre for exploratory laparotomy in view of ruptured gravid rudimentary horn of unicornuate uterus. Two pint PCV were issued and abdomen was opened in midline vertical incision. Intraoperatively one litre of hemoperitoneum with unicornuate uterus with non-communicating ruptured accessory horn on right side with foetus and placenta in peritoneal cavity was seen. Fallopian tubes and ovaries appeared normal.

Ruptured horn was excised and uterus was repaired. Ipsilateral ovary was preserved. The accessory horn and foetus were sent for histopathology examination which was suggestive of placenta increta with gestational hypertrophy and hyperplasia of myometrium with normal tube and ovary. Patient tolerated the procedure well. Postoperatively ultrasound of abdomen+pelvis was done which showed no renal anomalies.

DISCUSSION

Maldevelopment of the Mullerian ducts occur in a variety of forms and each anomaly is distinctive. No classification of Mullerian development can focus entirely on the uterus, the vagina is often involved and sometimes the tubes are involved as well. Disorders of lateral fusion of the two Mullerian ducts can be symmetric-unobstructed or asymmetric-obstructed. The bicornuate uterus which is a symmetric-unobstructed disorder of lateral fusion can have a partial or almost complete separation of the uterine cavities. For diagnosis, invasive methods such as hysteroscopy, hysterosalpingography, or laparoscopy have been used.⁶ The first sort of imaging that was routinely done was a 2D ultrasound. It was, however, insufficient for diagnosis because it cannot consistently distinguish between kinds of abnormalities. The diagnostic method was 3D/4D ultrasonography, which was non-invasive and allowed assessment of both the endometrial cavity and the uterine fundus in coronal view. For the diagnosis of MDAs, magnetic resonance imaging was as accurate. It was non-invasive and can also detect associated renal anomalies at the same time. Despite this, subjectivity, slight changes in morphology and shifting classification systems made it difficult to discriminate between these distinct anomalies on imaging modalities.⁴

Despite these challenges, Grimbizis et al found that the septate uterus was the most prevalent, accounting for 35% of all uterine abnormalities, followed by bicornuate at 25%, arcuate at 20%, unicornuate at 9.6% and complete agenesis at 3%. Didelphys uterus was shown to be the second least frequent MDA, accounting for 8.3% of all MDAs.⁴

A standard speculum exam, when observation of anatomical abnormalities demands further study, usually led to initial suspicion of the disorder, followed by diagnosis. Furthermore, because Mullerian ducts frequently formed in combination with Wolffian ducts, renal anomalies may be discovered alongside uterine abnormalities.^{1,2}

Most of the patients with unicornuate uterus remained asymptomatic and diagnosed incidentally during ultrasound or caesarean section.

There was a higher risk of spontaneous abortion, foetal growth retardation and preterm labour as well as a decreased likelihood of carrying a pregnancy to term (45 percent). The results were equivalent to bicornuate uterus, however they were poor when compared to the more common MDAs of septate and bicornuate uterus.^{1,2,5}

Surgical repair of a unidelphys uterus with rudimentary horn was usually not recommended unless all other options for improving fertility have been exhausted.^{4,5} If the patient was symptomatic, with dyspareunia or pain from hematometrocolpos due to blockage, longitudinal vaginal septum excision may be explored. The presence of a unicornuate uterus was not an indication for caesarean section but many times it was associated with malpresentations requiring operative interference, thus vaginal delivery should be considered first.¹⁻³ Unicornuate uterus was rarely associated with cervical incompetence, so encerclage was done only when there was a history of cervical incompetence or previous preterm labour.^{2,5}

Herlyn-Werner-Wunderlich (HWW) syndrome, also called as obstructed hemivagina and ipsilateral renal anomaly (OHVIRA) can be associated with Mullerian anomalies like unicornuate uterus. This involved triad of Mullerian and Wolfian duct abnormalities with blocked hemivagina and ipsilateral renal agenesis.⁴ These can result in hematocolpos or hematometrocolpos on the side of an obstructed hemivagina, resulting in abdominal lump and lower abdominal pain.⁶

CONCLUSION

Unicornuate uterus with rudimentary, non-communicating horn is rare Mullerian anomaly. Pregnancy in noncommunicating horn can occur due to transperitoneal migration of sperms from contralateral fallopian tubes. Rudimentary horn must have functional endometrium for implantation of such zygote and rupture of rudimentary horn occurs as it fails to accommodate the growing fetus. Surgical correction (metroplasty) is usually not recommended. Presence of renal anomalies should be looked for in every case of MDAs. There are increased chances of obstetric complications in unicornuate uterus as compared to other anomalies.

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