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Case Report

Ruptured rudimentary horn pregnancy: a case report

Gopika Venugopal, Soumya Patil*, Nandish S. Manoli, Pratap T., Hemapriya, Madhuri N.

Department of Obstetrics and Gynecology, JSS Medical College, Mysuru, Karnataka, India

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***Correspondence:**
Dr. Soumya Patil,
E-mail: soumyarpatil999@gmail.com

ABSTRACT

Uterine anomalies are congenital malformations arising due to embryological mal-development of mullerian ducts. The European society of Human Reproduction and Embryology (ESHRE) classifies these anomalies into 6 classes. Hemi-uterus is one such class of formation defect of mullerian duct with unilateral uterine development; the contralateral part could be either incompletely formed or absent. In a rare form of ectopic pregnancy, implantation can occur in the cavity of a rudimentary horn of the hemi-uterus. Authors report a case of 22-year G2P1L1 with 12 weeks gestation who presented with acute abdomen. Ultrasound showed hemoperitoneum with suspicion of ectopic pregnancy. Laparotomy confirmed the diagnosis of ruptured right rudimentary horn with fetus and placenta in the peritoneal cavity. Immediate laparotomy and excision of the horn with transfusion of blood and blood products saved the patient in the nick of time.

Keywords: Ectopic pregnancy, Hemi-uterus, Ruptured rudimentary horn pregnancy, Unicornuate uterus

INTRODUCTION

Uterine anomalies are congenital malformations as a result of embryological mal-development of mullerian or para-mesonephric ducts. These anomalies resulting from abnormal formation, fusion or resorption of the Müllerian ducts is broadly classified by ESHRE into 6 classes. Hemi-uterus is one such class (Class U4) of formation defect of mullerian duct with unilateral uterine development; the contralateral part could be either incompletely formed or absent.It is further divided into two sub-classes depending on the presence or not of a functional rudimentary cavity.[1,2] The presence of functional rudimentary cavity, either communicating or non-communicating makes it a clinically important factor for complications such as hematometra or ectopic pregnancy.

Mullerian duct anomalies (MDA) represent a common benign condition with a prevalence of 4-7%.[1] Incidence of unicornuate uterus with rudimentary horn is approximately 0.4%.[3] Pregnancy in a rudimentary horn is a rare condition reported with incidence of 1 in 100,000 to 140,000 pregnancies.[3] The thin muscular wall of the pregnant uterus ruptures early because of under-development and poor distensibility of the myometrium and ruptured rudimentary horn pregnancy is a life-threatening condition. Incidence of uterine rupture is observed in 90% of cases mostly in second trimester.[4] Authors report a case of ruptured non-communicating rudimentary horn with Hemi-uterus.

CASE REPORT

A 22-year-old was referred from a local PHC to the emergency department in view of severe anemia, PV bleeding and non-availability of blood. Patient gave history of pain abdomen and giddiness since 1 day. Pain was insidious in onset, progressive, confined to lower abdomen and non-radiating. Her LMP was 3 months

back. She had bleeding per vagina for 5 days not associated with pain or passage of clots. Patient also gave history of vomiting 3 episodes on the day of admission.

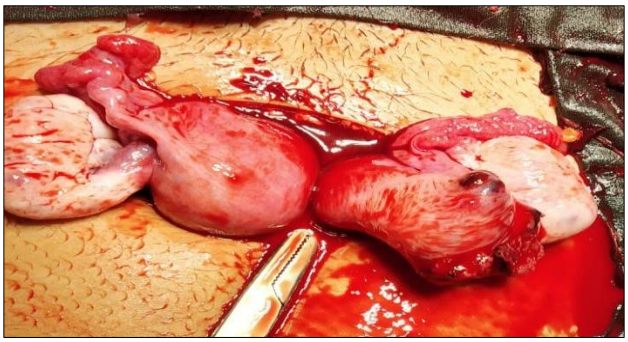

Figure 1: Uterus with right side ruptured rudimentary horn with left side normal tube and normal bilateral ovaries.

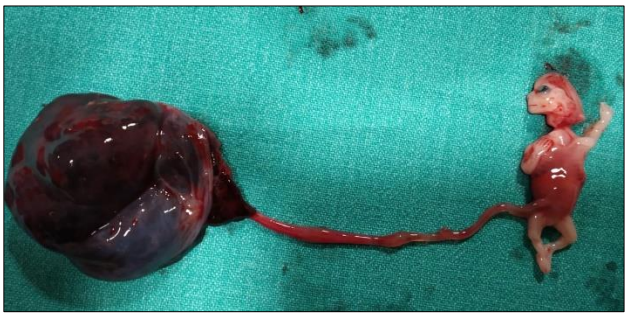

Figure 2: Fetus of around 10-12 weeks with umbilical cord and the placenta.

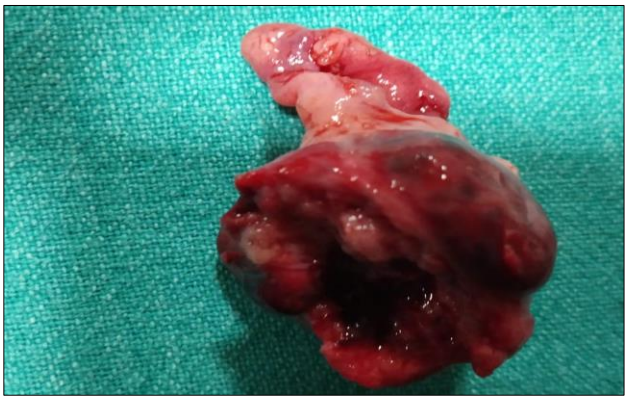

Figure 3: Specimen of right rudimentary horn excision and right salpingectomy.

The patient was G2P1L1 with previous uneventful vaginal delivery at term, one and half years back. She had normal menstrual cycles with moderate flow and mild dysmenorrhea. LMP was 3 months back with 12 weeks of gestation. Her current pregnancy was detected by UPT done at home; no scan was done to confirm the site of pregnancy. Over the counter MTP kit was taken by the patient following which she had bleeding. There was no significant past medical or surgical history.

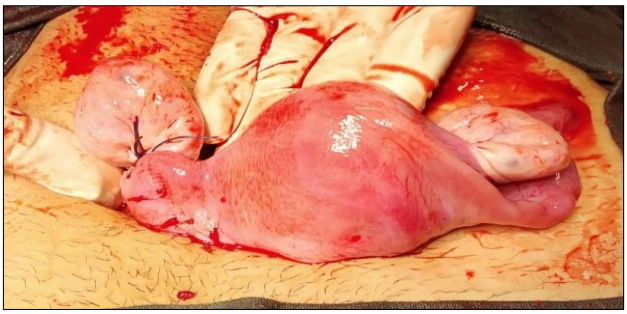

Figure 4: Final picture.

On examination her general condition was fair with stable vitals. Severe pallor was present. Physical examination of the abdomen revealed generalized tenderness. Per speculum examination showed minimal bleeding from OS and on bimanual examination cervix appeared deviated to left, fullness present in POD and mild cervical motion tenderness. Hemoglobin estimation was done, which was 6 gm/dl. An abdomino-pelvic ultrasound showed a well-defined hyperechoic mass lesion seen in the right adnexa measuring 5 × 5 cm with increased vascularity and moderate hemoperitoneum, a working diagnosis of ruptured right tubal ectopic pregnancy was made. After emergency transfusion of adequate blood and blood products emergency laparotomy was done which revealed hemoperitoneum consisting of 250 gm clots, rupture of the non-communicating right rudimentary horn, bulky uterus, normal left adnexa with fetus and placenta in the peritoneal cavity. Right salpingectomy with resection of right rudimentary horn was performed after extraction of all the fetal parts and placental components from the peritoneal cavity. Right ovary, left fallopian tube, left ovary and the uterus appeared normal and was left intact. Right side - ruptured rudimentary horn with tube and ovary seen, on left side uterus with intact tubes and ovaries seen (Figure 1).

On the right side foetus of 10-12 weeks with umbilical cord, and left side the placenta is seen (Figure 2).

Excised right rudimentary horn with right side fallopian tube. (Figure 3).

Post-operative picture after excision of the right rudimentary horn and right salphingectomy showing uterus in the hand of the operating surgeon, bilateral ovaries and left tube seen (Figure 4).

DISCUSSION

MDAs occur due to failure in organogenesis, fusion and/or septal resorption of the mullerian ducts. Failure in organogenesis of one or both ducts results in uterine agenesis/hypoplasia or a unicornuate uterus. Failure in fusion of both ducts results in a bicornuate or didelphys uterus. Once the ducts have fused, septal resorption occurs. Defects in this septal resorption results in a septate or arcuate uterus.[1] A unicornuate uterus with non-

communicating rudimentary horn remains a very rare MDA in comparison to other anomalies as described in the Buttram and Gibbons classification.

Most women with a unicornuate uterus with rudimentary horn are asymptomatic, but may present with unilateral dysmenorrhea in the presence of a functioning endometrial cavity in the horn. Pregnancy in a non-communicating horn is possible by intraperitoneal sperm and ovum transmigration or contralateral tubal pick up of the fertilized ovum within the peritoneal cavity.[4] It is extremely uncommon for such cases to continue beyond 2nd trimester of pregnancy and result in a viable baby. Rupture of the horn in the second trimester typically between the 10th and 20th week of gestation is the most common outcome in these cases, although a rupture has been reported even at 34 weeks.[3] The rupture occurs because of the underdevelopment of the myometrium and a dysfunctional endometrium. As the uterine wall more vascular, bleeding is more severe in rudimentary horn pregnancy rupture, resulting in a life threatening obstetric emergency.[5,6] Only 10% of cases such as these reach term, and the fetal salvage rate is only 2%.[5] The first case of uterine rupture associated with rudimentary horn was reported in 1669 by Mauriceau.[7]

The key for diagnosis prior to the rupture is a high index of clinical suspicion. A history of severe unilateral dysmenorrhea may be a clue for diagnosis in a functional endometrial cavity in the horn. A careful pelvic examination in the first trimester showing a deviated uterus with a palpable adnexal mass should provoke suspicion of a Müllerian anomaly. An ultrasound or MRI can confirm it. Ultrasound, CT scan, hysterosalpingogram, hysteroscopy, laparoscopy, 3D ultrasound and MRI are few useful diagnostic tools.

Tsafrir et al, reported 2 cases of rudimentary horn pregnancy found in the first trimester by sonography and confirmed by MRI.[8] For diagnosing the rudimentary horn pregnancy a set of criteria was created, which are as follows.[8] (a) a pseudo pattern of asymmetrical bicornuate uterus, (b) absence in continuity of tissue surrounding the gestation sac and the uterine cervix; (c) Gestational sac with surrounding myometrial tissue.[8] Most of the cases remain undiagnosed due to silent rupture or delayed diagnosis and present to us as emergency. Use of labor induction agents for termination of pregnancy in a rudimentary horn is unsuccessful and can in-turn lead to rupture of the horn. Pregnant women not responding to induced abortion should be investigated with a high index of suspicion.

Primary and chief mode of management of rudimentary horn is surgical removal. Immediate surgery is recommended even in unruptured cases. Early diagnosis and laparotomy/laparoscopic excision of the rudimentary horn have been the gold standard method to save the patient with a ruptured horn. Medical management with methotrexate and its resection by laparoscopy has also been reported. Renal anomalies are often found in association with the uterine anomalies due to the close association of embryological development between mesonephric and paramesonephric ducts. Hence, it is mandatory to further assess these women for renal anomalies and skeletal abnormalities.

CONCLUSION

Ruptured rudimentary horn pregnancy can result in catastrophic hemorrhage leading to high rates of mortality and morbidity. High clinical suspicion, early referral, timely diagnosis, resuscitation with blood and blood products and immediate surgery remain the cornerstones of therapy while managing such cases.

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