



Case Report

A surprising presentation of a partial hydatidiform molar pregnancy in rudimentary horn of unicornuate uterus

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ABSTRACT

Introduction: Partial molar pregnancy in non-communicating rudimentary horn of unicornuate uterus is very rare. It often has catastrophic results. It may be missed as an early diagnosis unless it results in rupture of the horn in the late 1st or early 2nd trimester. Pre rupture diagnosis of rudimentary horn pregnancy with USG is technically difficult with a poor sensitivity.

Case Presentation: We present a case of unruptured non-communicating rudimentary horn pregnancy at 7 weeks with live fetus presenting in our emergency department with pain in abdomen. Laparotomy was carried out and excision of rudimentary horn was done. However, the histopathological reports were more surprising suggesting to be a partial hydatidiform molar pregnancy in rudimentary horn. Beta-hCG report was 50,000 mIU/ml. Serial Beta-hCG was done weekly till its values reached well within normal limits after 10 weeks.

Conclusion: Development of partial molar pregnancy in a rudimentary horn is a very rare phenomenon. High degree of clinical suspicion is required when ultrasound reports are inconclusive regarding intrauterine pregnancy. Further close follow up of patient is required in future pregnancies to avoid recurrence of molar pregnancy.

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1. Introduction

Mullerian duct anomalies are the anomalies associated with the congenital malformation of the upper female genital tract including uterus, cervix and upper part of vagina. Prevalence of these anomalies is 1%.¹ These abnormalities result from incomplete fusion of mesonephric ducts. Unilateral arrested mullerian duct development results in development of unicornuate uterus.² Rarely unicornuate uterus may also have a rudimentary horn. The incidence of unicornuate uterus is estimated to be 1:250 and its occurrence with rudimentary horn is 1:100,000.³

Such anomalies are associated with recurrent miscarriages, recurrent pregnancy losses, preterm labor, infertility and other obstetric complications. Conception in rudimentary horn may occur in case a small communication exists between the horn and uterine cavity. A trans peritoneal migration of the fertilized ovum from the contralateral

side may occur in case of non communicating horn. The proportion of non-communicating rudimentary horns is 70-90%.⁴ The frequency of pregnancy in rudimentary horn is reported to be 1:76000.⁵ Patient presentations may vary from having vague complaints of mild lower abdominal pain to acute abdomen with hemorrhagic shock. The greatest risk of a rudimentary horn pregnancy is the risk of rupture because of poorly developed musculature. In view of the paucity of literature on rare observation of pregnancy in rudimentary horn of uterus, the case reported here is of crucial importance.

2. Case Report

A 28-year old lady, married for 8 years, para 2 with history of two previous full term vaginal delivery presented to emergency department with chief complaint of lower abdominal pain associated with excessive vomiting and amenorrhea of 2 months. Urine pregnancy test was positive. Her vital signs were stable with BP 100/64 mm of Hg and

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pulse rate of 96 per minute. Mild pallor was present. On abdominal examination, there was tenderness over left iliac fossa. No lump was palpable. On per speculum examination cervix was apparently normal and external OS was closed. On bimanual examination uterus was around 8weeks size, right adnexa free and non-tender but there was fullness in left adnexa around 4x5cm mass was felt which was tender.

Transvaginal Ultrasonography done revealed an empty uterine cavity with thickened endometrium. However, live embryo of CRL corresponding to 7 weeks was seen in left adnexa. Left ovary could not be seen separately. No fluid seen in pouch of Douglas. Sonographic diagnosis of left adnexal extra-uterine pregnancy was made with differential diagnosis of tubal and ovarian pregnancy.



Fig. 1: Transvaginal ultrasound showing an embryo in left adnexa with empty uterine cavity

Patient was planned for laparotomy. Per-operatively uterus was of normal size. Left rudimentary horn of size 4x4.5cm was present. Left ovary and fallopian tube were normal. There was no hemoperitoneum. Gestational sac was unruptured. The left rudimentary horn was resected. On cut section of the rudimentary horn gestation sac seen and was sent for histopathology. Patient had a smooth postoperative recovery and was discharged from the hospital on day 5. Histopathology report was suggestive of a partial Hydatidiform molar pregnancy in rudimentary horn of uterus which is very rare phenomenon. Patient's Beta-hCG reports was sent which came out to be 50,000 Miu/ml. Beta-hCG was followed weekly and reached within normal limits in around 10weeks post operatively.

3. Discussion

Rudimentary horn with unicornuate uterus results from incomplete fusion or arrested development of one of the mullerian ducts during fetal life. Rudimentary horn may

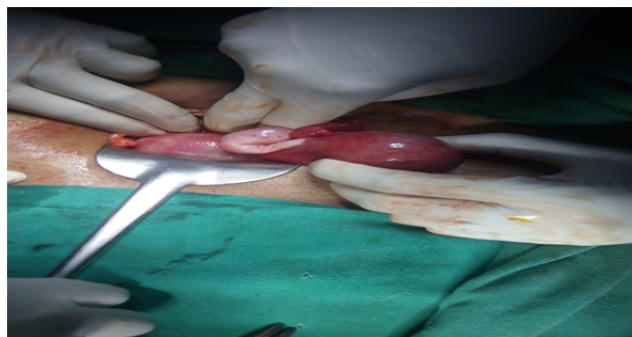


Fig. 2: Peroperative finding of a gravid rudimentary horn of unicornuate uterus

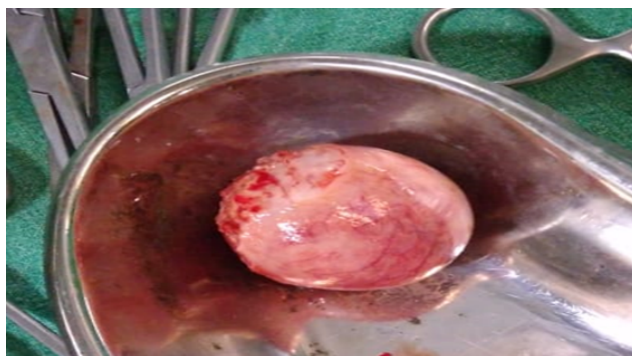


Fig. 3: Showing excised distended gravid rudimentary horn

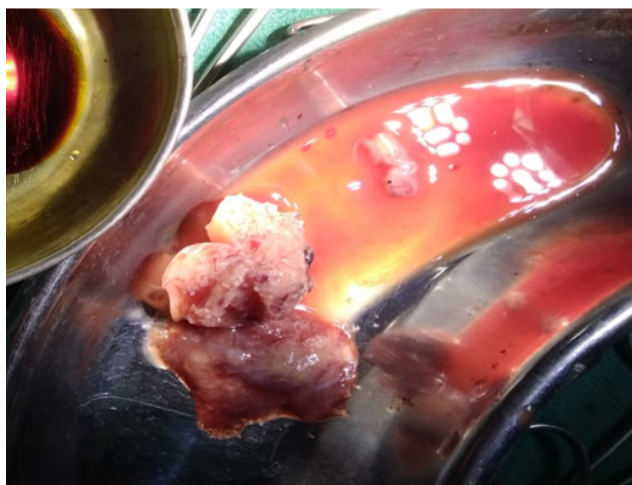


Fig. 4: Cut section of rudimentary horn with embryonic tissue

be communicating or non-communicating. Trans peritoneal migration of sperm or fertilized ovum lead to pregnancy in rudimentary horn.⁶ As the fetus grows in the rudimentary horn, the chances of rupture in the first or second trimester are increased due to poor distensibility of horn. Massive hemorrhage may occur increasing maternal morbidity and mortality. The key for diagnosis is high index of clinical suspicion. Patient may give history of severe dysmenorrhea, previous recurrent abortions, infertility or history of preterm vaginal deliveries. On examination a palpable adnexal mass should provide suspicion of müllerian anomaly.

Earlier cases were diagnosed after rupture of the rudimentary horn. However, with the advancement in diagnostic modalities such as transvaginal USG, CT Scan, MRI scan and laparoscopy diagnosis can be made before rupture.

Tsafir et al. suggests following criteria for diagnosing a pregnancy in rudimentary horn.⁷

1. A pseudo pattern of asymmetrical bicornuate uterus.
2. Absent visual continuity between cervical canal and lumen of pregnant horn.
3. Presence of myometrial tissue surrounding gestational sac.

The sensitivity of USG decreases with increasing gestation age as the enlarging gravid rudimentary horn causes difficulty in visualization of surrounding structures. In the present case scenario, the patient presented to the emergency with intense abdominal pain at 7 weeks 5 days of pregnancy. Trans vaginal sonography was useful in early diagnosis of ectopic pregnancy. On laparotomy it was revealed that the pregnancy was in rudimentary horn of the uterus. During laparotomy there was no hemorrhage as the horn was unruptured. Hence due to early diagnosis and immediate laparotomy with excision of the rudimentary horn the patient was saved and any catastrophic event was avoided. The histopathology report of the ectopic pregnancy came as a surprise suggesting partial molar pregnancy in rudimentary horn.

Partial hydatidiform mole was first diagnosed in the late 1970s and term refers to the combination of a fetus with localized placental molar degenerations. Histologically it is characterized by focal swelling of the villous tissue, focal trophoblastic hyperplasia, and embryonic or fetal tissue.⁸ The abnormal villi are present within macroscopically normal placental tissue. Patients with partial hydatidiform mole generally present with signs and symptoms consistent with missed abortion. Estimated incidence of partial mole is 1 per 700 intrauterine pregnancies.⁹ Several ultrasound features have been proposed that might increase the ultrasound detection of molar change in missed miscarriages in the first trimester. These include gestational sac diameter ratios, cystic changes in the placenta, and the increased echogenicity of placental tissue. Abnormally high levels of β -HCG support the diagnosis. Close follow up is

required for level of β -HCG to return to normal. The excision of rudimentary horn and ipsilateral salpingectomy is the recommended treatment for patient for preserving future fertility.

4. Conclusion

Partial molar pregnancy in rudimentary horn is rare entity. High degree of clinical suspicion is required. Removal of rudimentary horn is essential to avoid future risk. Close monitoring of the patient is required in future pregnancies with early TVS & β -HCG level to avoid recurrence of molar pregnancy. Patients attending gynecological clinic and women in general population must be educated on the gynecological and obstetric complication of extra uterine pregnancy. Early diagnosis and confirmation of pregnancy by ultrasonography is must.

5. Source of funding

None.

6. Conflict of interest

None.

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