CASE REPORT

RUPTURED RUDIMENTARY HORN PREGNANCY OF UNICORNUATE UTERUS

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ABSTRACT: Unicornuate uterus can sometimes be associated with rudimentary horn. Pregnancy in a rudimentary horn is rare and usually ends up in rupture. Diagnosis is difficult and can be missed in routine ultrasound scan and is usually detected after rupture. We report a case of G1P1 with rudimentary horn pregnancy which raised suspicion on ultrasound and was later diagnosed by MR imaging. Patient refused termination and presented next day with shock. Laparotomy revealed ruptured right rudimentary horn pregnancy.
KEYWORDS: Ruptured rudimentary horn pregnancy, Unicornuate uterus, Ectopic pregnancy.

INTRODUCTION: Mullerian anomalies were first classified in 1979 by Buttram and Gibbons and revised by American Society of Reproductive Medicine. Unicornuate uterus is type 2 with unilateral hypoplasia or agenesis that can be further subclassified into communicating, non communicating, no cavity, and no horn.[1] A unicornuate uterus accounts for 2.4%–13% Mullerian anomalies.[2] 72–85% of the rudimentary horns are non communicating.[3] Rupture during pregnancy is the most dreaded complication which can be life threatening. We report a case of G1P1 with rudimentary horn pregnancy (RHP) which raised suspicion on ultrasound, later diagnosed by MR imaging and confirmed by emergent laparotomy.

CASE REPORT: A 21 year old recently married female presented with 4 months of amenorrhea for regular check up. On examination patient was healthy, with no signs of anemia and fundus of uterus was palpable corresponding to 16 weeks of gestation. On per vaginal examination uterus was bulky, cervix was normal in length, soft and external Os was closed.

Patient was sent for routine sonographic examination (Figure 1) as it was her first prenatal visit. Sonographic examination revealed gestational sac with single live fetus corresponding to 17 weeks 6 days which was seen separately from the uterus. The gestational sac was surrounded by thick wall and no obvious communication with uterine cavity was seen. Placenta with umbilical cord arising from it was noted in posterior-lateral wall covering the gestational sac. Fetus within the sac showed normal movements and fetal cardiac activity, no obvious anomalies was detected at the time of scan. The empty uterus which was visualised was bulky in size with thickened endometrium suggestive of endometrial reaction. Uterus had single cervix and no obvious communication with the sac. On ultrasound ectopic right adnexal pregnancy was considered and Magnetic resonance imaging was performed for further evaluation.

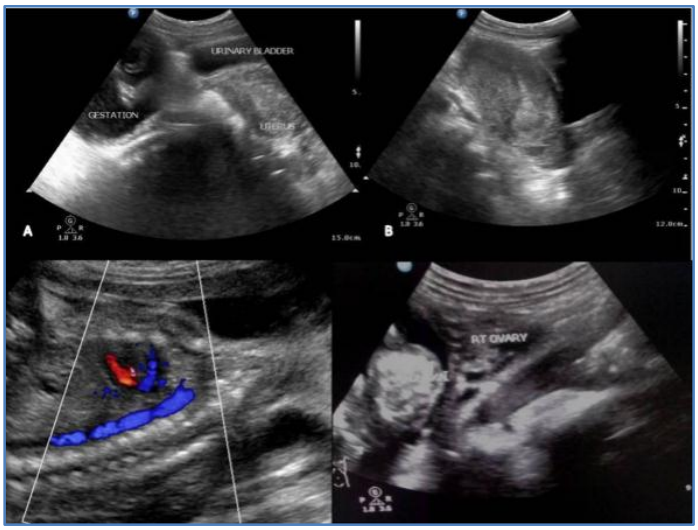

FIGURE 1

(A) Transverse section showing ectopic right adnexal gestation which is separate from the uterus, **(B)** Longitudinal section of empty uterus with thickened endometrium, **(C)** Live fetus with colour uptake of cardia and aorta, **(D)** Figure showing ectopic right adnexal pregnancy with separately visualised normal right ovary.

MR T2 axial, coronal and sagittal sections (Figure 2) were acquired which revealed a gestational sac with fetus in right adnexa separate from the uterus which was bulky with thickened endometrium. The gestational sac had a thin wall which was isointense to the myometrium of the uterus. No obvious communication / fat plane between the sac and uterus or cervix were identified. Bilateral ovaries was visualised separately. With MR findings, differentials of right tubal pregnancy and rudimentary horn pregnancy were considered.

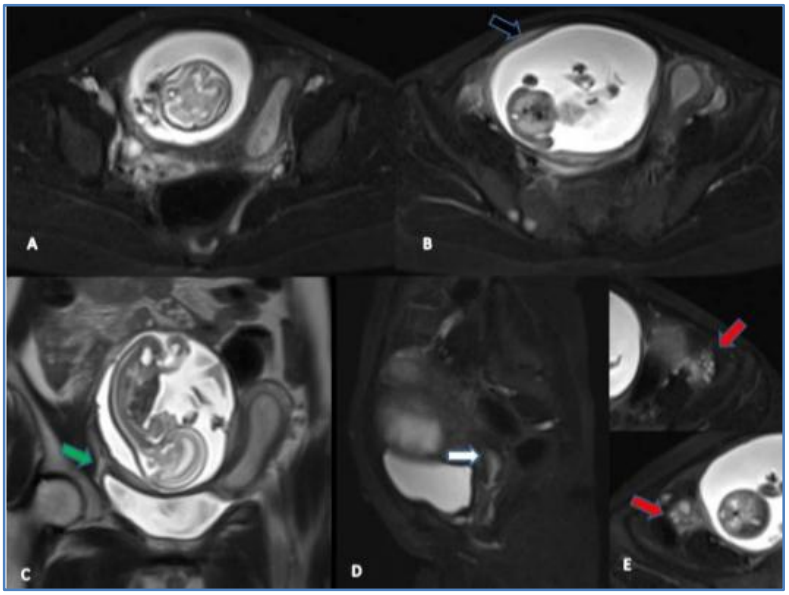

FIGURE 2

(A) Axial T2W sequence showing ectopic right adnexal pregnancy and normal empty uterus to left with thickened endometrium (decidualisation reaction). **(B)** Axial T2W sequence showing ectopic right adnexal pregnancy with well defined thin wall which is iso-intense to myometrium (Blue Arrow). **(C)** Coronal T2W sequence showing ectopic right adnexal pregnancy displacing normal empty uterus to left, A thin tubular iso-intense fallopian tube is seen right lateral to gestation (Green arrow). **(D)** Sagittal T2W sequence showing cervix (White arrow) of empty uterus, no communication was seen with cervix and ectopic gestation. **(E)** Axial T2W Sequence showing both normal ovaries (Red arrow) which are separately visualised from ectopic gestation.

Patient was counselled about the risks of continuing pregnancy and was advised for emergent termination but patient refused termination and hence patient was discharged against medical advice. Next day, patient presented to emergency department with acute pain abdomen, vomiting and abdominal distension. On examination, patient was pale with tachycardia; blood pressure of 150/90mm of Hg. Patient was immediately started with blood transfusion and taken up for emergent laparotomy. On laparotomy (Figure 3) hemoperitoneum and ruptured rudimentary horn with fetus and placenta were identified. The rudimentary horn was excised and post operatively patient was closely monitored. Patient recovered well and was discharged. At present patient is healthy on periodic follow up.

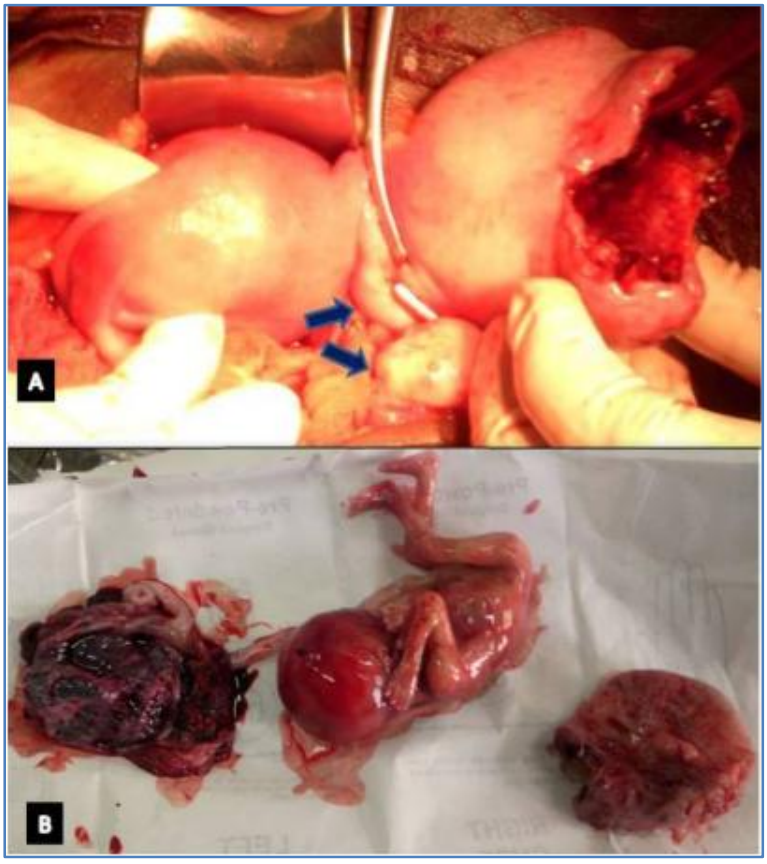

FIGURE 3

(A) Intra operative photograph showing ruptured rudimentary horn pregnancy. Normal right ovary and right fallopian tube are visualised (Blue Arrow). **(B)** Post operative specimen showing excised ruptured rudim.

DISCUSSION: Pregnancy in a non communicating rudimentary horn is uncommon, estimated to occur in 1 per 100000 to 140000 pregnancies. The first described pregnancy in a rudimentary uterine horn was made in 1669 by Mauriceau[4] and, worldwide, it have been described up to now in about 700 cases. Besides, it is rare for such pregnancy to result in a viable fetus: only 10 percent reach term, and the newborn survival rate is about 2%[4] enhancing the importance of this case report.

The only possible explanation for pregnancy to occur in this case is by transperitoneal migration of spermatozoa through the contralateral tube. The natural history of RHP is usually rupture of the pregnant horn during the second or third trimester, resulting in life-threatening heavy bleeding. Therefore, early, prerupture diagnosis is of major importance. But prerupture diagnosis of RHP is difficult and requires high index of suspicion. This has led to late and false diagnosis leading to subsequent uterine rupture which has been reported repeatedly in the recent literature.[5]

Some cases were diagnosed during an attempt to evacuate the uterus for termination of pregnancy which were initially incorrectly diagnosed as intrauterine pregnancy,[6] proving that early diagnosis of RHP remains challenging. The endometrium of rudimentary horn is usually thinner and non functional.[7] Pathologic placentation leading to placenta accrete have been described in literature[8,9,10] in RHP probably due to thin myometrial wall surrounding the endometrial cavity and thus allowing creeping of placentation into the myometrium. The thin muscular wall of the pregnant horn and associated placenta accrete further increases the risk of rupture.

Sonographic imaging features which should raise high index of suspicion for diagnosis of RHP are: (1) Gestational sac is seen separately from the empty uterus which may be bulky in size with thickened endometrium due to decidualisation reaction and mimicking false pattern of an asymmetric bicornuate uterus, (2) Absence of definable continuity between the cervical canal and the lumen of pregnant horn and (3) Presence of thick wall with echogenicity similar to myometrium in ultrasound and isointense to myometrium in MR imaging. Rarely focal hypervascularisation extending into the myometrium of rudimentary horn suggestive of placenta accrete may support the diagnosis of RHP.

The differential diagnoses which have to be considered for suspected RHP is tubal pregnancy, cornual pregnancy and intrauterine pregnancy in bicornuate uterus. A tubal pregnancy will not show myometrium surrounding the gestational sac. In a pregnancy in one of horn of bicornuate uterus usually myometrium will be thicker and clearly delineable, continuity between the endometrial lining of pregnant horn and other horn will be visualised. In contrast, In RHP there will be variation in thickness of myometrium between the two horns with thin myometrium in rudimentary horn and marked distance between the two horns.[11]

Magnetic Resonance Imaging (MRI) is a problem solving tool in cases of dilemma on sonographic imaging, in cases for confirmation of RHP and cases of mullerian anomalies.[12,13] MRI

increases the spatial resolution and offers multiplanar images which reveals both internal and external uterine structure. In cases of RHP, coronal sagittal and axial planes are used for accurate assessment of uterine connection to horn and either confirm or rule out communication with endometrial cavity of endocervical canal. MR imaging can also demonstrate thin endometrium, thus help to differentiate from tubal pregnancy and can also demonstrate ovaries separately from gestational sac thus ruling out ovarian ectopic pregnancy.

Primary strategy in ruptured horn is surgical removal and repair of the defect.[14] Immediate surgery is recommended by most obstetricians even in a non-ruptured uterine horn pregnancy if diagnosed before catastrophe.[15]

CONCLUSION: We report a rare case of RHP in an unicornuate uterus with subsequent rupture. In this report we also suggest salient sonographic features which should raise suspicion of RHP and MR imaging features which acts like a problem solving tool for confirmation. Thus cases of suspected sonographic RHP, MRI should be done to prevent rupture and catastrophe to the patient and also to aid in surgical planning in prerupture cases.

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