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Fourteen weeks Missed Abortion in Non-communicating Rudimentary Horn of a Unicornuate uterus- A Rare Case of Unruptured Ectopic Pregnancy

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Abstract

Background: Unicornuate uterus with a rudimentary horn is an anomaly of the mullerian duct. This condition results when one of the paired mullerian ducts fails to fuse completely. Pregnancy in the rudimentary horn of the unicornuate uterus is very difficult to diagnose on ultrasound and can be easily missed out. Pregnancy is usually detected after rupture of pregnant horn. Ninety percent of such cases rupture in second trimester.

Case Presentation: Here we report a case of 18-year-old-lady presented with 14 weeks missed abortion. On ultrasonography, unruptured cornual ectopic pregnancy was suspected. Exploratory laparotomy was done. Intra-operatively there was unruptured ectopic pregnancy in non-communicating left horn of a unicornuate uterus. The rudimentary horn with unruptured pregnancy was excised. This case is reported because of its rarity as well as to stress the need for high index of suspicion and role of ultrasonography in the diagnosis of this rare and dreadful entity.

Conclusion: Pregnancy in rudimentary horn is a rare condition. A high index of suspicion and careful ultrasound examination can detect the condition earlier before rupture. If missed, it can be catastrophic.

Keywords: Laparotomy, Rudimentary horn, Unruptured pregnancy, Unicornuate uterus.

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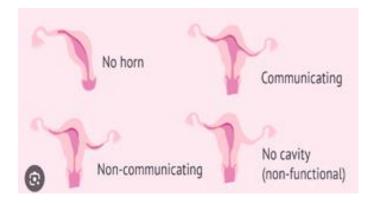
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Introduction:

Two mullerian ducts form the female reproductive system, which includes the fallopian tubes, uterus, cervix, and the upper two-thirds of the vagina. Mullerian malformations result when there is a defective development or fusion of these ducts. A wide range of anomalies can result due to the disruption of the mullerian duct system, ranging from agenesis of the vagina to the duplication of the cervix and vagina. Based on the degree of an anomaly,

mullerian duct anomalies (MDAs) are classified into seven types. A unicornuate uterus is a type 2 MDA and can be further classified into communicating, non-communicating, no cavity, and no horn¹⁶. Rudimentary horn pregnancy in a unicornuate uterus is a rare clinical condition with a reported incidence of 1 in 100,000 to 140,000 pregnancies³. The first case was described by Mauriceau in 1669¹⁰. Rudimentary horn pregnancy can lead to several obstetric complications, most commonly causing rupture, massive hemoperitoneum and shock. According to the available literature, the fetal survival rate is between 0% and 13% in the rudimentary horn pregnancies with only one-third of such gestations reaching term or beyond. More than 50% of the pregnancies end with rupture of the pregnant uterine horn¹⁷. Although diagnosis of rudimentary horn pregnancy remains challenging, few cases of early (first trimester), pre-rupture sonographic diagnosis of this condition have been reported^{18,19}. Once the diagnosis is made, treatment is excision of rudimentary horn. Ipsilateral salpingectomy should also be performed as there is possibility of tubal ectopic pregnancy in future. Success with intracardiac potassium chloride and methotrexate leading to self resorption of conceptus is also reported²⁰. Here we report a case of unruptured rudimentary horn pregnancy.





Types of unicornuate uterus

Schematic diagram representing unicornuate uterus with pregnant rudimentary horn on left side with no connection to the cervix & a small band connecting both

Case Presentation: An 18-year-old young woman consulted a local gynaecologist at her 2 months of pregnancy with mild lower abdominal pain. An ultrasound of pregnancy profile was done which revealed left sided cornual pregnancy (Figure 1) & serum ß hCG was 4000 IU. Medical management was given with inj MTX 2 times. But patient was lost to follow up. Two months later she came to us with irregular scanty vaginal bleeding and mild lower abdominal pain during those 2 months period. She attained menarche at the age of 12 years and her cycle was regular before this pregnancy. She was married for 2 years and the couple practiced barrier method for contraception. This was her first pregnancy. During initial evaluation, her vitals were stable, afebrile and clinically not anaemic. Abdomen was soft, tenderness in left iliac fossaon deep palpation without any rigidity or muscle guarding and bowel sound was present. Speculum examination revealed closed os with minimal bleeding. Uterus was anteverted, 14 weeks pregnancy size, soft, deviated to left and left fornix was full and mildly tender on bimanual examination. Chest and cardiovascular examination findings were normal. Haemoglobin was 11 gm%, liver and renal function tests were within normal range. A colour doppler USG of uterus & adnexae showed that uterus was normal in size with empty cavity. Right ovary & adnexal regions appeared normal. One dead fetus was seen in left adnexal region with BPD: 22 mm, FL: 13 mm corresponding to 14 weeks gestation. Fetal movement & cardiac activity were absent, suggesting left adnexal missed abortion (Figure 2). No collection was seen in the cul-de-sac. Patient & her husband were counselled about the possibility of cornual or rudimentary horn pregnancy and shared decision for laparotomy was taken. Prior to laparotomy hysteroscopy was done at the same setting. Uterine cavity was empty & small, only right ostium was present. After opening peritoneal cavity (Operative findings - Figure 3): no haemoperitoneum was seen. The uterus was unicornuate with right tube and ovary attached to it's cornua, while unruptured d ectopic pregnancy was in rudimentary horn attached to uterus on left side which was non-communicating. The rudimentary horn was excised with pregnancy in situ along with ipsilateral salpingectomy. The post-operative period was uneventful. She was discharged on 4th post-operative day.





2D ultrasound showing uterus is bicornuate & gravid. Two uterine cavities are seen. Right uterine cavity is empty. Left uterine cavity shows a gestational sac with fetal pole inside. Fetal cardiac movement is seen. Fig 1a- CRL = 16 mm, fig 1b- CRL = 20 mm, correspond to 8 ± 1 weeks & 9 ± 1 weeks respectively.

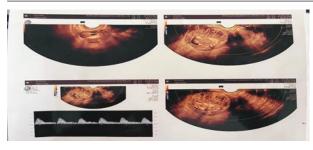


Figure 2- Colour Doppler USG of uterus & adnexae A distorted dead fetus is seen in the left adnexal region (BPD: 22 mm, FL: 13 mm, corresponds to 14 weeks of gestation) – suggestive of left adnexal missed abortion.

Figure 3
Per-operative findings
Left horn with 14 wks pregnancy



Right non-pregnant horn Fibrous connecting band Unruptured left rudimentary non-communicating horn of a unicornuate uterus



Excised left horn with 14 weeks pregnancy & left fallopian tube



After reconstruction of attachment site of left non-communicating horn



Macroscopic picture of excised rudimentary horn with embryo

Preoperative Diagnosis: 14 weeks missed abortion in left horn of unicornuate uterus

Surgical treatment: Laparotomy followed by excision of left horn.

Discussion:

Among mullerian anomalies, unicornuate uterus accounts for 2.4%-13% 1,2. A unicornuate uterus with a rudimentary horn develops due to the failure of the development of one mullerian duct and incomplete fusion with the contralateral side. A rudimentary horn of the unicornuate uterus may be of communicating or non-communicating type⁴. In 83% of the cases the rudimentary horn is non-communicating⁵. commonest associated anomaly is renal (36%) most common being ipsilateral renal agenesis followed by pelvic kidney¹¹. The possible explanation of ectopic pregnancy in rudimentary horn cases is transperitoneal migration of the spermatozoa or fertilized ovum from the contralateral tube. Unlike tubal ectopic pregnancy, which usually ruptures in first trimester, about 90% of these pregnancies culminate in rupture mostly in the second trimester⁶. Diagnosis prior to rupture is unusual, requires high suspicion index⁷. Diagnosis could be made on 2D ultrasound with accuracy being 29%-33%. It should be supplemented with 3D ultrasound, which improves accuracy rates. MRI also confirms the diagnosis, but it is expensive and not available globally. It can be done when ultrasound imaging is inconclusive 8. Tsafrir suggested a criterion to diagnose early pregnancy in the rudimentary horn via ultrasound: pseudo pattern of asymmetrical bicornuate uterus, absent visual continuity between cervical canal and lumen of pregnant horn and presence of myometrial tissue around the gestational sac, hyper-vascularization typical of placenta accrete9. None-the-less most of cases remain undiagnosed until it ruptures and presents as an emergency. Here, we report pregnancy in ASRM classification type IIb of unicornuate uterus/ ESHRE-ESGE classification U4a ¹².

Our patient was subjected to 2D & doppler USG, left cornual ectopic pregnancy was confirmed. There was one cervix, which was communicating with the right uterine horn, the left horn had no communication with the cervix or with the right horn. There was a dead fetus and gestational sac was surrounded by myometrium. Differential diagnosis of ectopic in fallopian tube and pregnancy in an anomalous uterus should be ruled out, because treatment differs. 3D ultrasound configurations help in differentiating when in doubt. Transverse sections of the normal uterus and anomalous uterus with ectopic pregnancy may look the same, but with 3D configurations, they can be easily differentiated. A table to help differentiate between them by ultrasound has been made (Table 1). Management will depend upon the hemodynamic condition of the woman, her gestational age. Essentially, removal of uterine horn is the line of management. Earlier days, or in places where access to health care is difficult, when diagnosis is a problem, women often come with rupture of rudimentary horn with unstable hemodynamic condition. Emergency open surgery with multiple blood transfusions is the only option^{13,14}. But with advent of better diagnostics, and more women being diagnosed in first trimester scans, medical line of management with surgical removal being done on a later date has become possible¹⁵. There are no fixed guidelines to manage such ectopic pregnancy, but like management of other ectopic pregnancy, in early hemodynamically stable pregnancy, intrauterine or intramuscular methotrexate, or intrauterine KCL can be injected. The patient is followed up with ßhCG. Once pregnancy completely resolves, she is advised to get the rudimentary horn and ipsilateral fallopian tube removed to prevent further ectopic pregnancy [15]. This method improves operative morbidity and chances of intraoperative hemorrhage but delays definitive management.

Reproductive outcome after excision of unicornuate horn is similar to reproductive outcomes of mullerian anomalies [21]. Every patient should be screened carefully. Based on patients' history and surgical outcome, plan for delivery should be made. Vaginal delivery is possible if patient is carefully selected, monitored, and counseled well [21, 22].

Table 1 Ultrasound features to help differentiate between fallopian tube ectopic pregnancy, bicornuate uterus pregnancy & rudimentary horn pregnancy

USG features	Fallopian tube ectopic pregnancy	Bicornuate uterine pregnancy	Unicornuate uterus with rudimentary horn pregnancy
Two uterine horns	Absent	Present	Present
Ring of myometrium around G-sac	Absent, present in interstitial ectopic pregnancy <5mm	Present	Present
Visual continuity between G-sac & cervix	Absent	Present	Absent

Conclusion:

Pregnancy in rudimentary horn of unicornuate uterus is a rare but life-threatening condition when present with rupture of horn. High index of suspicion with prior knowledge and early diagnosis with a good ultrasound scan followed by early intervention can save many lives.

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- Guarantor accuracy and integrity of the work: Ansary SA1, Tabasum S2, Jahan MS3, Israt S4

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