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PREGNANCY IN NON-COMMUNICATING RUDIMENTARY HORN OF A BICORNUATE UTERUS: A CASE REPORT

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ABSTRACT

A rudimentary horn with unicornuate uterus results due to failure of the complete development of one of the Mullerian ducts and incomplete fusion with the contralateral side. Pregnancy in a rudimentary horn of a unicornuate uterus is extremely rare. We report one such rare case of ruptured pregnancy of non-communicating rudimentary horn of bicornuate uterus in a 26-year young female at 20 weeks. This report emphasizes the need for increased awareness of this rare anomaly and having a high index of suspicion, immediate removal of the rudimentary horn is lifesaving.

INTRODUCTION

Unicornuate uterus with a rudimentary horn is a uterine anomaly caused due to failure of complete development of one of the Mullerian ducts and incomplete fusion.[1] The incidence is estimated at 1 per 100,000 to 140,000 pregnancies.[1] It can be communicating or noncommunicating.[1] Pregnancy in a rudimentary horn (RHP) is rare and occurs most commonly in a non-communicating horn.[2] The first case of uterine rupture associated with the rudimentary horn was reported in 1669 by Mauriceau. The timing of rupture varies from 5 to 35 weeks depending on the horn musculature and its ability to hypertrophy and dilate. 70–90% rupture before 20 weeks and can be catastrophic.[1,3] As the uterine wall is thicker and more vascular, bleeding is more severe in rudimentary horn pregnancy rupture.[1] Rupture of the horn is associated with a maternal mortality rate of 5.1%.[1,2] We report a rare case of ruptured rudimentary horn pregnancy, diagnosed as a bicornuate uterus.

CASE HISTORY

A 26 year primigravida with 20 week amenorrhea, presented with bleeding per vaginum since 3 days. She was a known case of epilepsy and rheumatic heart disease, with balloon mitral valvuloplasty done. One month earlier, there was a similar episode of bleeding diagnosed as a threatened abortion for which cervical encirclage was done. On admission her general condition was stable. A sonography done subsequently showed a bicornuate uterus with live intrauterine pregnancy in the right horn. Also the 2 DECHO showed normal biventricular function with ejection fraction of 60%. On 5th day post admission she had pain in lower abdomen and an episode of fainting, the blood pressure was 90 systolic; this was attributed to post diuretic dehydration as she was on diueretics for RHD. On the 6th day she became breathless with pain in abdomen; decreased urine output and rising creatinine were suggestive of acute kidney injury ? sepsis? DIC. Although her condition was worsening, high risk consent for exploratory laparotomy was denied by the relative and patient was being managed conservatively.

On day 8; the patient expired and a complete autopsy was performed. On opening the abdomen, the baby was lying free in the abdominal cavity, surrounded by blood clots approximately weighing 700 gms.(Fig 1a) The umbilical cord was connected to the placenta which was adherent to the ruptured sac like structure that was further connected to the uterus on the right by fibrous tissue. There was no communication between the ruptured sac and the slightly enlarged normal-looking uterus. Only one cervix was seen which was attached to the uterus and was encirclaged. (Fig 1b) Left fallopian tube and ovary were attached to the uterus while the right fallopian tube was attached to the sac like structure. Two differential diagnoses considered at the time of autopsy were ruptured ectopic broad ligament pregnancy and ruptured bicornuate uterus. As there was no communication between the sac and uterus, a broad ligament pregnancy was favored, however the fallopian tube attached to the ruptured sac could not be explained. On microscopy, sections from the uterus showed Arias Stella reaction with sheets of decidua, no trophoblast or chorionic villi were seen. Section from the sac revealed

blood and fibrin with admixed villi infiltrating the peripheral smooth muscles. As the sac had well organized bundles of smooth muscle in the wall,(Fig 2,) broad ligament pregnancy was ruled out and due to lack of communication between the uterus and the pregnant horn, the final diagnosis made was rudimentary horn pregnancy.

DISCUSSION

The European Academy for Gynaecological Surgery has proposed new classification of female genital tract which has 6 classes.[9] Class 0 — normal uterus. Class I— dysmorphic uterus; Ia T-shaped uterus and Ib uterus infantilis. Class II — septate uterus; IIa partial septate uterus and. IIb complete septate uterus Class III — dysfused uterus, IIIa partial disfused uterus and IIIb complete dysfused uterus Class IV — unilaterally formed uterus (formerly unicornuate uterus); IVa horn with cavity (communicating or not), IVb horn without cavity or aplasia Class V — aplastic/dysplastic uterus; Va bilateral or unilateral horn with cavity and Ib bilateral or unilateral horn without cavity or aplasia of both parts

Class VI — for still unclassified cases

A unicornuate uterus accounts for 2.4%–13% of all Mullerian anomalies.[1] Most unicornuate uteri have a rudimentary horn without communication to the uterine cavity, however the attachment of the rudimentary horn to the main uterus varies from a fibro-muscular band to an extensive fusion between the two horns where there is no external separation between them.

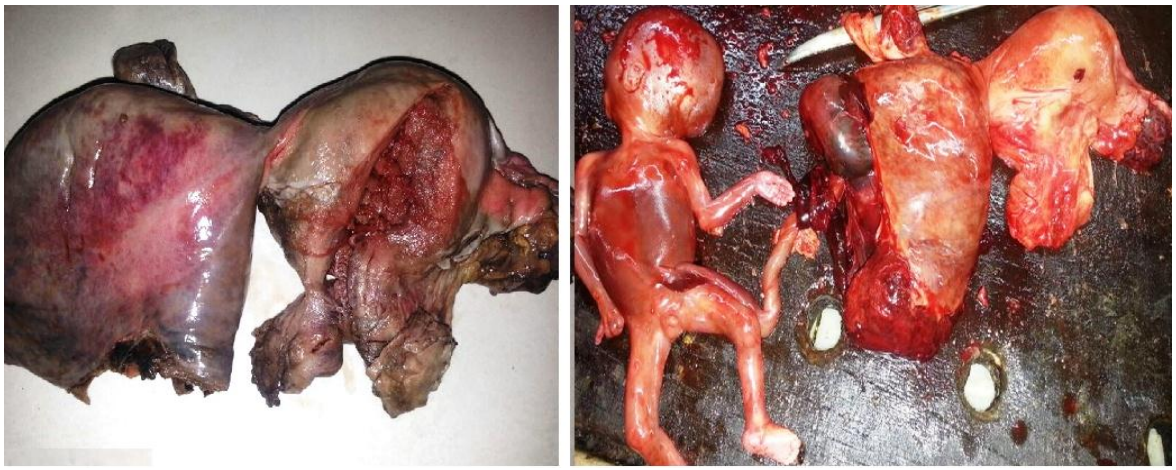

Figure 1: Uterus with cervix & attached non-communicating ruptured horn.
Figure 2: Gross photograph showing uterus with ruptured rudimentary horn with attached placenta & baby lying outside the uterine cavity.

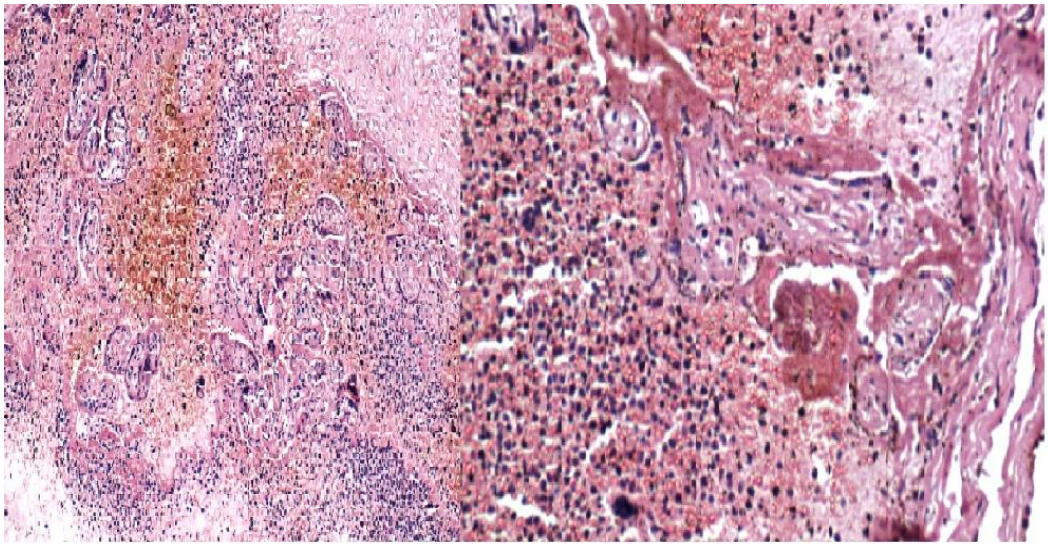

Figure 3 and 4: H & E stained (40X) section from the ruptured horn showing chorionic villi in the haemorrhagic background, smooth muscle layer at the periphery.

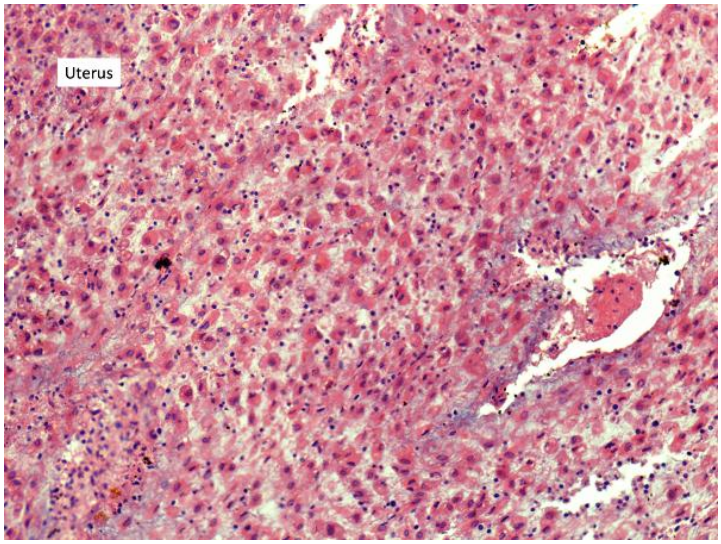

Figure 5: H & E stained (40X) Section from communicating horn showing decasualized endometrium.

Rudimentary horn of unicornuate uterus with may be associated with gynecological and obstetric complications like infertility, endometriosis, hematometra, urinary tract anomalies, abortions, and preterm deliveries.[1]

Pregnancy in a rudimentary horn is not as rare as thought earlier and in a review Nahum found 588 such cases with rupture occurring in 80%.[2] Few published cases of pre rupture diagnosis are also available in literature. The likely explanation for pregnancy in the non communicating rudimentary horn is by transperitoneal migration of spermatozoa through the contralateral tube.[2] Rupture during pregnancy is the most dreaded complication which can be life threatening to the mother. Early diagnosis is crucial however, there are no definite clinical criteria to diagnose this and diagnosis is often difficult as the enlarging horn can obscure adjacent anatomic structures.[2]

Ultrasound, hysterosalpingogram, hysteroscopy, laparoscopy, and MRI are diagnostic tools.[1] The differential diagnosis of RHP is a tubal pregnancy, cornual pregnancy and an intrauterine pregnancy in a bicornuate uterus.[2] Ectopic pregnancy beyond 12 week is unlikely to be tubal. An interstitial line extending from uterine cavity to the gestational sac can be seen on sonography in corneal pregnancy.[6] Variation in the thickness of the myometrium between the two horns and a large gap between the two horns favour Rudimentory horn over bicornuate uterus.[2]

Tsafrir et al has developed the following radiologic criteria for diagnosis of rudimentary horn *(1)* a pseudopattern of an asymmetrical bicornuate uterus, *(2)* absent visual continuity of tissue surrounding the gestational sac and the uterine cervix, and *(3)* the presence of myometrial tissue surrounding the gestational sac.[2] Sensitivity of ultrasound is only26% in diagnosing this condition and the sensitivity decreases with the advancement of pregnancy[1] The possibility of missing this on ultrasonography is higher in unexperienced hand.[1] Our case was misdiagnosed on sonography as bicornuate uterus. Similar cases are described in the literature.[5] MRI provides images with detailed internal and external uterine structure and is very useful in detection and preoperative planning in suspected cases.[2,6]

Renal anomalies are found in 36 % of cases and hence further workup is required.[1]

Rupture of rudimentary horn pregnancy in cases with previous normal pregnancies, are also described.[4] Rudimentary horn pregnancy is commonly associated with placenta accrete.[2] The thin muscular wall of the pregnant horn along with the presence of placenta accreta, further increases the risk of rupture [2] Rare cases of live births from RHP are described by using cesarean sections.[5,7] Primary policy of management of rudimentary horn is surgical removal, even in a non ruptured uterine horn pregnancy.,[1,8]

The usual outcome in second trimester in more than 90% cases is fetal demise.[8]

Our case emphasizes the importance of a high index of suspicion, differentiation from other more common conditions mimicking rudimentary horn pregnancy and the need for early radiologic evaluation and more sensitive modalities like MRI in suspected cases to ensure a favorable outcome.

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