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Case Report Open Access

The Unchartered Waters of Obstetrics - Rupture of Noncommunicating Rudimentary Horn Pregnancy (Rnhp) in a Bicornuate Uterus

Tanushree Sandipta Rath[1], Soubhagya Ranjan Tripathy[2]*, Subhashree Rout[3] and Jagannath Mishra[4]

[1]*Senior Resident, Department of O&G, SCB Medical College Hospital, Cuttack, Odisha, India*
[2]*Mch Resident, Department of Neurosurgery, SCB Medical College Hospital, Cuttack, Odisha, India*
[3]*Junior Resident, Department of O&G, SCB Medical College Hospital, Cuttack, Odisha, India*
[4]*Junior Resident, Department of O&G, SCB Medical College Hospital, Cuttack, Odisha, India*

Abstract

Background: Mullerian duct anomalies in female result from failure of complete development of one duct and incomplete fusion of other duct during embryonic life. Bicornuate uterus with rudimentary horn (BURH) is the rarest uterine anomaly. Pregnancy in rudimentary horn is even rarer, i.e. 1:1,40,000 pregnancies.

Case description: A 24-year-old primigravida presented with acute pain abdomen and amenorrhoea for 16 weeks. She was severely pale and was in hypovolemic shock, with diffuse tenderness, guarding and rigidity of abdomen. Per vaginal examination revealed a bulky uterus with motion tenderness and full fornices. On laparotomy there was hemoperitoneum along with rupture of left horn of the bicornuate uterus. The placenta was inside the rudimentary horn, and the horn was non-communicating with the body of the uterus. It was excised completely. An abortus of 16 weeks size was lying within intact membranes, and was found free within the abdominal cavity.

Conclusion: A second trimester pregnancy with features of ruptured ectopic should be screened for the associated uterine anomalies. Rupture of the rudimentary horn of the uterus is one of the important but rare causes of acute abdomen. A high index of suspicion is warranted in cases of advanced gestation presenting with acute abdomen, especially in developing countries like India, where the possibility of early detection before rupture is unlikely, culminating in maternal demise.

Keywords: Non communicating horn; Bicornuate uterus; Ruptured ectopic; Rudimentary horn pregnancy; Laparotomy

Introduction

Mullerian duct anomalies in female result from failure of complete development of one duct or incomplete fusion of other duct [1-3] during embryonic life. The incidence of Mullerian duct anomalies in general population is found to be 3.4%. Bicornuate uterus with rudimentary horn (BURH) is usually non-communicating in 83% cases. Rudimentary horn is the rarest uterine anomaly. The incidence of the bicornuate uterus with rudimentary horn is around 1:1,00,000 and pregnancy in rudimentary horn is even rare i.e. 1:1,40,000 pregnancies [4,5].

Case Description

A 24-year-old primigravida presented with pain abdomen and blood stained mucus discharge per vaginum of one day duration. Her prior menstrual cycles were regular without menorrhagia or dysmennorhoea. She was amenorrhoeic for 16 weeks and her urine pregnancy test was positive at 6 weeks of missed periods. She had no antenatal check-ups or ultrasound scans after detection of pregnancy. She had regular bladder and bowel habits. On examination, she was severely pale with a blood pressure of 80/50 mm Hg and a pulse rate of 108/minute. On abdominal examination, there was diffuse tenderness along with distention, guarding and rigidity; size of the uterus could not be elicited. Per speculum examination findings were not significant and per vaginal examination revealed a bulky uterus with motion tenderness. All the fornices were bulging and full.

After resuscitation with crystalloids and one unit of blood transfusion the patient was shifted to operation theater and planned for immediate exploratory laparotomy under general anesthesia. A transverse suprapubic skin crease incision was given two finger breadths above the symphysis pubis. After separating the subcutaneous tissues and fascia, the peritoneum was opened and there was hemoperitoneum of about 4 liters of blood, mixed with clots; which were evacuated. Rupture of left horn of the bicornuate uterus (Figure 1) was found with the placenta inside the rudimentary horn. The uterus was bulky; bilateral tubes and ovaries were healthy.

The ruptured horn was non-communicating with the body of the uterus (RNHP) and was excised completely with placement of double clamps; the lateral wall of uterus was repaired (Figure 2, 3 and 5). A transfusion of one unit of whole blood was given. An abortus of 16 weeks size was lying within intact membranes, and was found free within the abdominal cavity (Figure 4), which was taken out. Hemostasis achieved and the wound was closed in layers. Postoperative course was uneventful.

Discussion

Pregnancy in a non-communicating rudimentary horn is very difficult to diagnose before it ruptures. Pregnancy in a rudimentary horn of a bicornuate uterus is rare. It is difficult to truly estimate the incidence of these complications as the data available are in the form

***Corresponding author:** Soubhagya Ranjan Tripathy, Department of Neurosurgery, SCB Medical College Hospital, Cuttack, Odisha, India, Tel: +91-9861008487; E-mail: sourav.tripathy@gmail.com

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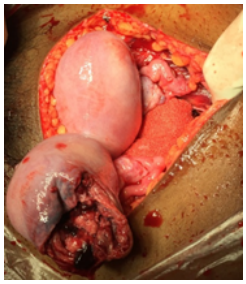

Figure 1: Rupture of left rudimentary horn.

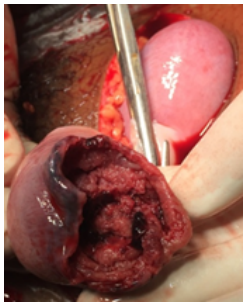

Figure 2: Rupture of left horn with placenta *in situ*.

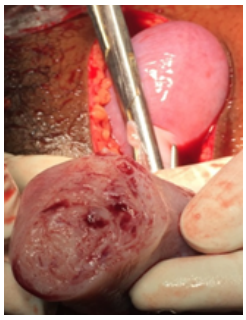

Figure 3: Excised non-communicating horn.

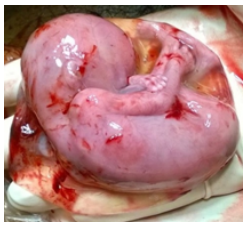

Figure 4: 16 weeks fetus lying within intact membranes, free in abdomen.

of case reports and surveys collected from the literature that are usually unintentional inherent bias towards the more severe cases requiring surgery.

A fibrous or fibro-muscular band usually connects the horns of the ducts but in 80 - 90% of cases there is no communication. The rudimentary horn may consist of a functional endometrial cavity or it may be a small solid lump of uterine muscle with no functional endometrium. This malformation is rare and it can be associated with many complications throughout a woman's reproductive life beginning from menarche when hormonal stimulation may gradually activate the endometrium of the rudimentary horn. The resulting obstruction of the menstrual flow may cause hematometra, leading to endometriosis and infertility. Because of functioning endometrium and hematometra, teenagers may give a history of spasmodic dysmenorrhoea; married women may present with history of infertility, recurrent second trimester abortion and threatened abortion.

Pregnancy in a non-communicating rudimentary horn is a rare form of ectopic gestation. Bicornuate uterus with rudimentary horn is usually associated with miscellaneous other obstetrical complications including miscarriage, cervical incompetence, ectopic pregnancy, uterine rupture, preterm labour, malpresentations, intrauterine growth retardation, and caesarean delivery. Conception in rudimentary horn arises from a small communication with the uterine cavity (communicating) or by transperitoneal migration of the fertilised ovum via the contralateral side (non-communicating). The zygote then enters the tube of rudimentary horn [1].

The most significant threat of a BURH pregnancy is the risk of rupture (usually in the second trimester) because of the poorly developed musculature and this commonly presents with abdominal pain which may occur before or after rupture [6]. Variable thickness of rudimentary horn musculature, dysfunctional endometrium and poor distensibility of the myometrium lead to rupture of the rudimentary horn. Diagnosis is only on suspicion by ectopic minded people.

Nonetheless most cases remain undiagnosed until it ruptures and presents as an emergency. The pregnancy usually overcomes the first trimester period uneventfully as the rudimentary horn is thicker than the fallopian tube and 80 - 90% of the ruptures occur in the second trimester.

Tsafrir et al outlined a set of criteria for diagnosing pregnancy in the rudimentary horn. They are: 1) A pseudo pattern of asymmetrical bicornuate uterus; 2) Absent visual continuity of the tissue surrounding the gestational sac and the uterine cervix; 3) Presence of myometrial tissue surrounding the gestation sac [7]. Sensitivity of USG is 26%, which decreases with advancing age of pregnancy. Three-dimensional USG may be used for diagnosis of uterine anomalies.

A careful pelvic examination in the 1st trimester showing deviated uterus with palpable contra-lateral pelvic adnexa should arouse suspicion of uterine anomaly. Bimanual palpation of a mass extending outside the uterine angle or displacement of fundus to contralateral side with rotation of uterus and elevation of the affected horn or deviation of uterus to one side with additional mass in pregnancy may indicate BURH.

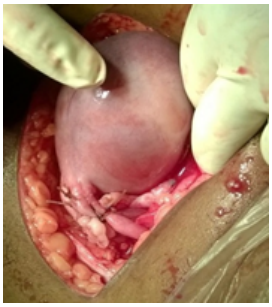

Figure 5: Non-communicating pregnant rudimentary horn excised and lateral wall of uterus repaired.

In patients presenting with infertility, hysterosalpingography shows the uterus to be deviated to one side and there is unilateral tubal block. MRI and CT scan are also gaining popularity for diagnosing uterine malformations. Both clinically and radiologically the diagnosis is more accurate in the early first trimester when the two horns are separate in the pelvis. Once the diagnosis is strongly suspected these patients should be taken up for laparoscopy or laparotomy, depending upon the general condition of the patient, and the rudimentary horn should be excised ,although subtotal or total hysterectomy may be necessary to save the life of the woman. Laparoscopy is most accurate for diagnosis.

Recently different methods of treatment have been described. Cases were treated by laparoscopy using various techniques or administration of methotrexate for termination of an early pregnancy in a rudimentary horn followed by elective laparoscopic resection [8,9].

31% patients with Mullerian anomalies will have urinary anomalies with associated urological anomalies; i.e. congenital absence of a kidney, pelvic kidney and duplication of renal system on one side [2]. In these cases, it is mandatory to have future assessment as it was advised in our patient.

90% of rudimentary horn pregnancies usually end with rupture and fetal demise. However, live birth cases have been reported after caesarean, for pregnancies which have progressed to third trimester [4]. Neonatal survival in BURH pregnancy is poor, occurring in only 11% cases during the last half century.

In the present case, the diagnosis of extrauterine pregnancy was established as she was 16 weeks amenorrhoeic with positive urine pregnancy test and presented in hypovolemic shock along with acute abdomen. The non communicating rudimentary horn rupture was confirmed intraoperatively and removal of rudimentary horn was done.

Conclusion

Any patient in the second trimester of pregnancy who comes with features of ruptured ectopic with a gestational age more than 12 weeks should be screened for the associated uterine anomalies. Rupture of the rudimentary horn of the uterus is one of the remote causes of acute abdomen in pregnancy. However, missing the diagnosis can lead to fatal complications, while early detection can save the life of the mother. It should preferably be diagnosed before the rupture occurs by routine antenatal ultrasound and should be treated by immediate surgery. A high index of suspicion is warranted in the teenagers presenting with dysmenorrhoea to detect this rare and very important complication in future pregnancy; especially in developing countries like India, where the possibility of early detection before rupture is unlikely before uterine rupture culminating in maternal demise.

Consent

Written informed consent was obtained from the patient for publication of this case report and all accompanying images. A copy of the written consent is available for review by the author of this journal.

Standards of Reporting

We submit that we have adhered to the standards of reporting, as is deemed necessary by this journal.

Data Availability

Submission of a manuscript implies that readily reproducible materials described in the manuscript, including all relevant raw data, will be freely available to any scientist wishing to use them for non-commercial purposes, without breaching participant confidentiality.

Competing Interests

There is no competing interest of any of the author either separately or jointly; academically or financially.

Author's Contribution

1) TSR, SRT- made substantial contributions to conception, design, analysis and interpretation of data.

2) SRT, TSR- have been involved in drafting the manuscript or revising it critically for important intellectual content.

3) TSR, SR- involved in the pre and postoperative care of the patient.

3) TSR, SRT, SR, JM - acquisition of data, critical analysis of published literature.

All authors read and approved the final manuscript.

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Staff of department of O & G, SCB MC, Cuttack, Odisha, India.

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