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Case Report

Rupture of non-communicating rudimentary horn ectopic pregnancy of a unicornuate uterus in first trimester

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ABSTRACT

Pregnancy in rudimentary horn of a unicornuate uterus is a very rare type of ectopic pregnancy. Ninety percent of cases eventually rupture in the second trimester. Only few cases of rupture of the pregnant rudimentary horn occur during first trimester. A 23 year old primigravida presented with 8 weeks of amenorrhea with acute pain abdomen and vomiting. On examination, the patient was in shock. On ultrasound, unicornuate uterus with ruptured rudimentary horn ectopic pregnancy was suspected. On laparotomy, the diagnosis was confirmed. The rudimentary horn was excised and ipsilateral salpingectomy was done. This case is presented because of rarity of rupture of rudimentary horn ectopic in first trimester and also to emphasize on the importance of recognizing subtle clinical findings which are suggestive of this rare condition.

Keywords: Pregnancy, Ectopic pregnancy trimester, First urogenital abnormalities

INTRODUCTION

Unicornuate uterus is a rare type of mullerian anomaly. Pregnancy in rudimentary horn of the unicornuate uterus is still rarer. Majority of these ectopic pregnancies eventually rupture during the second trimester. Rupture of the pregnant rudimentary horn in first trimester is uncommon. For diagnosis, a high index of suspicion is required. We report a case of a primigravida patient with rupture of rudimentary horn ectopic pregnancy in first trimester.

CASE REPORT

A 23 year old female, married for 6 months came to the emergency department with two months of amenorrhea with pain lower abdomen and vomiting for one day. The pain was for moderate intensity and intermittent. She had few episodes of vomiting which was non projectile. The patient was conscious and oriented. She looked pale; her pulse rate was 120 per minute and blood pressure was

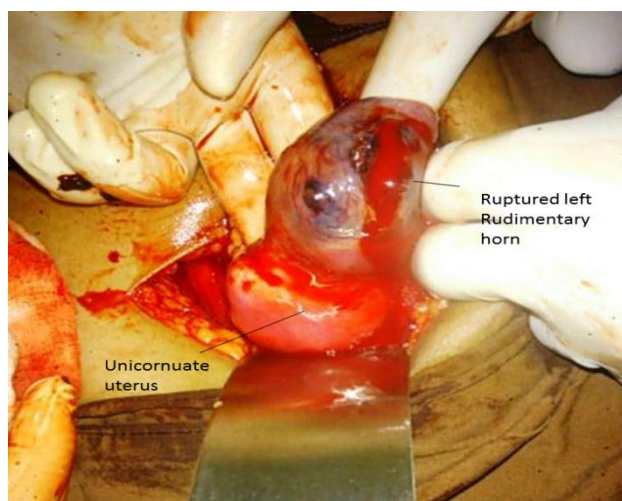
90/60 mm Hg. On abdominal examination, guarding was present and tenderness was localised to the suprapubic region. On vaginal examination, cervix was soft and smooth with no motion tenderness, uterus was normal size, and no mass was palpable through the fornices. Trans abdominal ultrasonography was done which revealed an empty uterine cavity. An irregular gestational sac of 2.1 cm, surrounded by a rim myometrium tissue, was seen adjacent to the uterus (Figure 1). A nonviable foetus with crown rump length of 0.93 cm was seen. There was no adnexal mass bilaterally. Free fluid was seen in the pouch of douglas.

The localization of pain and tenderness in the suprapubic region, absence of significant tenderness on vaginal examination along with the ultrasonographic findings led us to consider the possibility of ruptured rudimentary horn ectopic pregnancy. The patient and relatives were counselled about the possibility of both rudimentary horn and tubal ectopic pregnancy and the need for either

rudimentary horn excision and/or salpingectomy according to the intraoperative findings.



On laparotomy, around 800 ml of haemoperitoneum was present. The uterus was unicornuate with a ruptured rudimentary horn attached along its left wall with active bleeding from the rupture site (Figure 2).



The rudimentary horn was non communicating. The left fallopian tube was oedematous and swollen. Bilateral ovaries were normal. The rudimentary horn was excised followed by left salpingectomy. The uterine wall was repaired in layers with polyglactin sutures (Figure 3). Few endometriotic spots were present in the pouch of douglas which was partially obliterated by bowel adhesions. These spots were fulgurated. Abdomen was closed in layers after ensuring haemostasis. She was transfused two unit of blood, one intra-operatively and the other post-operatively. The post-operative recovery was uneventful. She was discharged on fourth postoperative day and advised combined hormonal pill for contraception as well as endometriosis. The histopathology report showed rudimentary horn ectopic

with left salpingitis. She was counselled about possible adverse outcomes in future pregnancies due to unicornuate uterus. She is presently scheduled to undergo an intravenous pyelogram due to the possibility of associated renal anomalies..

DISCUSSION

Mullerian anomalies result from maldevelopment of the mullerian ducts which first appear as invaginations of the dorsal coelomic epithelium on each side of the Wolffian duct during 7th week of intrauterine life. The incidence rate of mullerian anomaly is 1 in 250.¹ Unicornuate uterus is a type of mullerian anomaly which is still rarer, with an incidence of 1 in 1,00,000. It results from partial or complete failure in development of one of the mullerian ducts. Hence, a unicornuate uterus may or may not be associated with a rudimentary horn. Most of the rudimentary horns are non-communicating and may be in continuity with the uterus proper or connected to it through a fibrous band. A unicornuate uterus with a non-functioning non-communicating rudimentary horn connected through a fibrous band is most common variety.² Our patient had non communicating rudimentary horn, which is supported by the fact that she also had endometriosis. According to the new ESHRE classification, our patient can be classified as UIVa C0 V0, as the patient's cervix (C0) and vagina (V0) were normal.³

The commonest associated anomaly is renal (36%)⁴ most common being ipsilateral renal agenesis followed by pelvic kidney.⁵ Therefore, an intravenous pyelogram should be performed in all cases of mullerian anomaly.

An ectopic pregnancy in rudimentary horn is very rare. The first case was described by Mauriceau in 1669.⁶ The reported incidence ranges from 1 in 76,000 to 1 in 1,40,000 pregnancies.⁷ The possible explanation of ectopic pregnancy in rudimentary horn cases is trans peritoneal 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 2nd trimester.⁸ This is because the myometrium supporting and surrounding the gestational sac can expand with the growing foetus, but only upto a certain extent. Few cases of rudimentary horn pregnancy continuing till late 3rd trimester resulting in rupture⁹ or live birth by caesarean have been reported.¹⁰

Rudimentary horn ectopic pregnancy in first trimester may present acutely with rupture, which is rare, or may be diagnosed on routine first trimester scan. The latter is possible in patients who have been previously diagnosed to have unicornuate uterus with rudimentary uterus. In other patients, a high index of suspicion is required especially when there is a history suggestive of mullerian anomaly such as infertility, chronic pelvic pain, endometriosis, recurrent abortion, preterm birth,

intrauterine growth restriction and cyclical pain abdomen. A history of repeated failed attempts at surgical termination of pregnancy should also raise suspicion. There are only few case reports on prerule diagnosis of rudimentary horn ectopic.^{11,12} Tsafrir et al. suggested the ultrasound criteria for diagnosis of rudimentary horn pregnancy: (1) pseudo pattern of an asymmetrical bicornuate uterus, (2) absent visual continuity between the cervical canal and the lumen of the pregnant horn, and (3) the presence of myometrial tissue surrounding the gestational sac. Presence of hyper vascularization typical to placenta accrete may additionally support the diagnosis of rudimentary horn pregnancy.¹³ The reported sensitivity of ultrasonography in detecting such an anomaly is only 26%.¹⁴ Magnetic resonance imaging (MRI) provides confirmatory diagnosis when ultrasonography result is ambiguous. MRI provides multiplanar images with detailed uterine structure, both internal and external, without any risk of ionising radiation. However, it is not feasible in cases with acute presentation where laparotomy has to be done immediately. The other concern is uniform availability and affordability of MRI, especially in developing countries.

Once the diagnosis is made, treatment is excision of rudimentary horn. Some authors have reported success with intracardiac potassium chloride and methotrexate leading to self resorption of the conceptus.¹⁵ Ipsilateral salpingectomy should also be performed as there is possibility of tubal ectopic pregnancy in future. The approach may be laparotomy or laparoscopy depending upon haemodynamic stability of the patient and expertise in laparoscopic surgery.

Diagnosis of rudimentary horn pregnancy in early pregnancy prior to rupture is important so that surgical excision can be performed in an elective setting. This assumes more significance with low resource setting in developing countries where such a mishap may lead to maternal death. In patients with history suggestive of possible mullerian anomaly, a careful and targeted ultrasonographic examination should be done to look for presence of rudimentary horn. A high index of suspicion is the key to the diagnosis.

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