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CASE REPORT

Rudimentary Horn Rupture: A Case Report

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ABSTRACT

Rudimentary horn is a developmental anomaly of the uterus, and pregnancy in a non-communicating rudimentary horn is very difficult to diagnose before it ruptures. As the fetus enlarges in the rudimentary horn, the chances of rupture in the first or second trimester are increased, since the uterine cavity is the only place properly designed to expand and accommodate the developing fetus. Catastrophic hemorrhage results in increased maternal and perinatal mortality and morbidity. To date, management of such cases remains a challenge due to diagnostic dilemma. Expertise in ultrasonography and early resort to surgical management are lifesaving in such cases. A case of undiagnosed rudimentary horn pregnancy presented to the Nizwa regional referral hospital in shock with features of acute abdomen, and the diagnosis was confirmed at laparotomy.

Key words: *Acute abdomen, Collapse, Hemoperitoneum, Müllerian anomalies, Rudimentary horn*

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ÖZET

Rudimenter Horn Rüptürü: Olgu Sunumu

Rudimenter horn uterusun gelişimsel anomalisidir ve gebelikle ilişkisi olmayan rudimenter hornların rüptüre olmadan tanısının koyulması çok zordur. Uterin kavite, gelişmekte olan fetüse genişlemesi ve barınması için tasarlanmış en uygun yerdir ve fetüs rudimenter horn içerisinde genişledikçe birinci veya ikinci trimestırda rüptür ihtimali artmıştır. Katastrofik kanama maternal ve perinatal mortalite ve morbiditenin artmasıyla sonuçlanacaktır. Böyle olguların yönetimi tanısal ikilem nedeniyle günümüze kadar sorun olmuştur. Bu olgularda ultrasonografide deneyim ve hastanın erken cerrahi yonetiminin tesisi hayat kurtarıcıdır. Tanısı konmamış rudimenter horn gebelik olgusu şok ve akut batın bulgularıyla Nizwa bölgesel sevk hastanemize başvurdu. Rudimenter horn gebelik olgusu laparotomide teyit edildi.

Anahtar kelimeler: *Akut abdomen, Kollaps, Hemoperitoneum, Müllerian anomaliler, Rudimenter horn*

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INTRODUCTION

Ruptured rudimentary horn is a life-threatening obstetrical emergency encountered frequently in the emergency department, where the diagnosis is either missed or delayed. Unicornuate uterus results from abnormal development and fusion of the müllerian ducts usually associated with various degrees of rudimentary horn, which may be communicating or non-communicating with the uterine cavity. The connection of the horn with the uterus may be fibrous or fibromuscular. There is no communication between the two cavities in 75-90% of the cases, and the incidence of pregnancy in a non-communicating horn is as high as 83%, with incidence of uterine rupture observed in 90% of cases, mostly in the second trimester, as observed in our case[1,2]. The thin muscular wall of the pregnant uterus ruptures early because of under development and poor distensibility of the myometrium.

CASE REPORT

An unusual case of a primigravida, who was married four months previously with a pregnancy of 22 weeks, reported to the Nizwa Regional Referral Hospital with acute abdominal pain of two hours. On admission, she was in shock with pale, cold, clammy extremities, a feeble thready pulse of 120 beats/minute, blood pressure of 78/45 mmHg, and a respiratory rate of 18/minute. The patient was known as hypothyroid and was on 50 mcg of Eltroxin. Her abdomen was enlarged to 28 weeks in size, and was tense with generalized acute tenderness all over. A speculum examination did not reveal any cervical or vaginal pathology. The cervical os was tightly closed with no active vaginal bleeding. She was resuscitated with intravenous fluids and blood transfusion. Abdominopelvic ultrasound showed a fetus of 22 weeks with absent cardiac activity with increased free fluid in the abdominal cavity. The uterus with cervix separate from the gestational sac was seen clearly lower down in the pelvis. She was taken for emergency laparotomy with the provisional diagnosis of abdominal pregnancy with fetal death. During laparotomy, hemoperitoneum of around 3 L of blood with clots was noted. There was complete rupture of the left rudimentary horn of the uterus with the dead fetus lying in the intact amniotic sac covered with 1000 g of clots (Figure 1). The placenta and cord were attached to the uterine horn. No evidence of placental adherence to the rudimentary horn was observed. The left fallopian tube and left ovary appeared normal, and both were attached to the left rudimentary horn (Figure 2). The fetus within the amniotic sac along with the placenta and membranes were removed from the abdominal cavity (Figure 3). A fibromuscular band was seen attached between the unicornuate uterus and rudimentary horn. There was no communication between the rudimentary horn and the main unicornuate uterine cavity, which was confirmed with a probe. The uterus, lying separate in the pelvis, was soft in consistency, globular and enlarged to a size consistent with eight weeks. The right fallopian tube and ovary were found healthy and were attached normally to the unicornuate uterus. Excision of the rudimentary horn and left fallopian tube with conservation of the left ovary was done. The specimen was sent for histopathological examination, which was reported as "sections from the uterine horn show areas of hemorrhage and necrosis. Section from the fallopian tube was morphologically normal. Sections

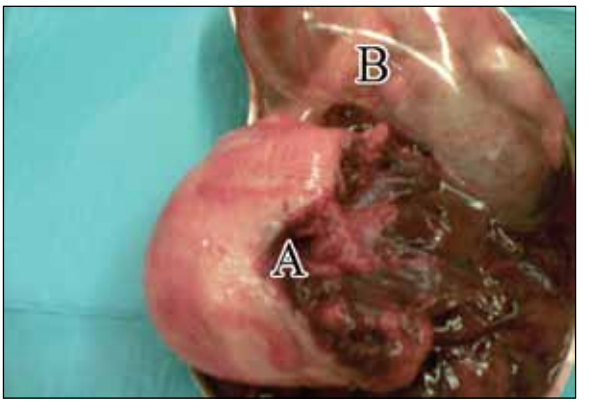

Figure 1. (A) Left ruptured rudimentary horn, (B) Fetus in intact amniotic sac.

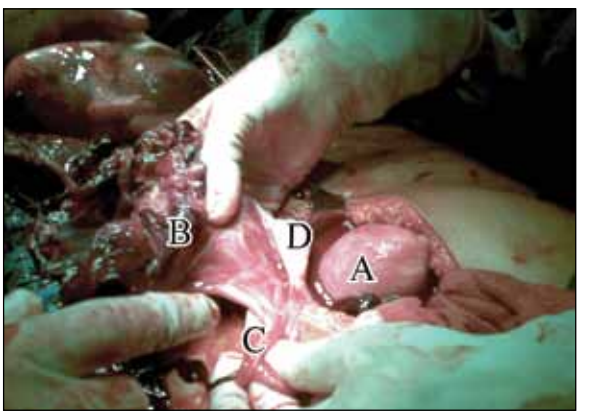

Figure 2. (A) Right unicornuate uterus, (B) Left ruptured rudimentary horn, (C) Left fallopian tube, (D) Left ovary.

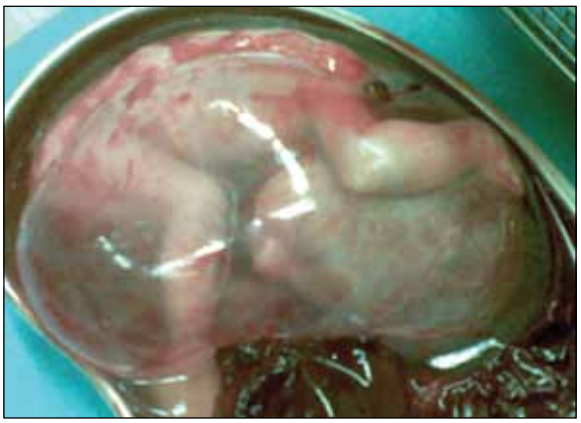

Figure 3. Fetus in intact amniotic sac.

of the placenta show fibrosed chorionic villi with syncytial knots. No villitis seen.". Histopathology confirmed ruptured rudimentary horn of the uterus. Follow-up appointment was planned for the patient for intravenous urogram to rule out any associated renal anomalies. She reported after three weeks with complaints of abdominal pain, which subsided with mild analgesics. A repeat abdominopelvic ultrasonography followed by computed tomography of the abdomen and pelvis was found to be completely normal. No associated renal anomaly was diagnosed.

DISCUSSION

Obstructive genitourinary malformations may be diagnosed in the prepregnancy workup for complaints of dysmenorrhea, endometriosis, and infertility, and for various pregnancy complications like recurrent miscarriages, preterm labor and malpresentations. However, if the rudimentary horn is underdeveloped with a non-functional endometrium, dysmenorrhea may be absent. The use of ultrasonography, computed tomography scan, magnetic resonance imaging, 3D ultrasound, and laparoscopy may be helpful for diagnosing such abnormalities. Buntugu used placement of a Foley catheter into the uterine cavity prior to performing a transabdominal ultrasound for diagnosing an extrauterine pregnancy, although it is not accepted as a preferred method[3]. The associated urologic anomalies are reported to be as high as 50-80% due to close approximation of the two systems, and must be diagnosed either at laparotomy, by palpation, or postnatally by magnetic resonance imaging or intravenous urogram[4].

Pregnancy in a non-communicating uterine horn is possible by intraperitoneal sperm and ovum transmigration or contralateral tubal pick-up of the fertilized ovum within the peritoneal cavity. The reported incidence of pregnancy in the rudimentary horn is 1 in 100.000-140.000, being a rare form of ectopic pregnancy[5]. Rupture of a pregnancy in the rudimentary horn by the second trimester is the most common outcome, but silent rupture with continuation of pregnancy as a secondary abdominal pregnancy was reported in some studies. Cases of pregnancy progressing to the third trimester and resulting in a live birth after cesarean section have also been documented. Pregnancy continued till term abdominally after ruptured rudimentary horn of a unicornuate uterus, and the placenta was attached in part to the myometrium of the horn, deriving the blood supply for the live fetus[6-8]. A very unusual case of twin pregnancy in a unicornuate uterus with one fetus in the non-communicating rudimentary horn has been reported, where the outcome was successful birth of twins by cesarean section[4].

Chances of placental adherence are increased due to poorly developed musculature, scant decidualization and small size of the horn. Magnetic resonance imaging has been a useful preoperative tool for both diagnosing pregnancy in the rudimentary horn and any abnormal placentation. Ultrasound has a sensitivity of 33.3% for diagnosing this anomaly, and sensitivity reduces with advancing pregnancy, adding to the diagnostic dilemma[1]. Sonographic diagnostic criteria suggested by Tsafrir are presence of pseudopattern of an asymmetrical bicornuate uterus, absent visual continuity between the cervical canal and the lumen of the pregnant horn, and the presence of myometrial tissue surrounding the gestational sac[9].

Immediate surgery is recommended whenever rudimentary horn pregnancy is diagnosed, but conservative management until viability is achieved has been advocated in very select cases with larger myometrial mass, where facilities are available for emergency surgery at any time. A rudimentary horn pregnancy can never be delivered vaginally, and the mode of delivery is always a laparotomy, both with eventuality of ruptured horn or if the pregnancy continues as abdominal post-rupture. Surgical removal of the rudimentary horn is mandatory to avoid risk of recurrence of rupture with increased maternal morbidity. However, laparoscopic excision of an unruptured rudimentary horn pregnancy has been increasingly carried out with safe and favorable outcome in many expert centers now[10].

In conclusion, a careful examination of the uterus by an experienced obstetrician in every case suspected

as mullerian anomaly may help to avoid misdiagnosis and catastrophic hemorrhage. High clinical suspicion, early diagnosis and timely laparotomy can reduce maternal and perinatal mortality and morbidity.

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