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Acute maternal collapse in pregnancy with ruptured rudimentary horn of unicornuate uterus: A case report

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Abstract

Unicornuate uterus with rudimentary horn occurs due to failure of complete development of one of the Mullerian ducts and incomplete fusion with the contralateral side. It is associated with numerous obstetrics and gynecological problems such as Infertility, Endometriosis, Miscarriages, Malpresentations & Intrauterine growth restriction. Pregnancy in the non-communicating rudimentary horn is extremely rare and usually terminates in rupture during first or second trimester of pregnancy. Pregnancy occurs via transperitoneal migration of sperm or zygote. Variable thickness of rudimentary horn musculature, poor distensibility of myometrium lead to rupture. This complication is usually seen in 2nd trimester resulting in shock and haemoperitoneum. Diagnosis of rudimentary horn pregnancy is difficult and can be missed in ultrasound. It requires a high risk of suspicion.

Materials and Methods: We report the case of 24yr primi gravida with 26 weeks of gestation presented with pain abdomen and sudden maternal collapse on arrival, suspected to be intra-abdominal pregnancy in ultrasound and on laparotomy found ruptured rudimentary horn of unicornuate uterus with fetus found intra-abdominally with massive hemoperitoneum.

Conclusion: Pregnancies in rudimentary horn of unicornuate uterus are prone to intra uterine growth restriction and rupture in second trimester so a high index of suspicion is required to save this catastrophic event and associated maternal morbidity and mortality. In our opinion, routine excision of rudimentary horn should be undertaken during non-pregnant state laparoscopically. However, those women who refuse should be adequately counseled regarding potential complications and if pregnancy occurs in rudimentary horn, first trimester laparoscopic excision should be done.

Keywords: Rudimentary horn, unicornuate uterus, hemoperitoneum

Introduction

Anatomical aberrations of the female genital tract result from abnormal fusion of the mullerian ducts and failure of absorption of the female genital tract. The prevalence of congenital uterine anomalies among fertile women is reported as 1:200 to 1:600, whereas that of unicornuate uterus with rudimentary horn is even rare (1 in 100,000) [1]. Many women remain asymptomatic where as some of them are diagnosed by the presence of infertility or obstetrical complications such as recurrent pregnancy loss, malpresentation, and premature labor [2-4]. Unicornuate uterus is caused by the non-development of the mullerian duct; usually associated with various degrees of rudimentary horn which may be communicating or non-communicating with the cavity of the uterus. Pregnancy in the rudimentary horn is rare. The pregnancy outcome in females with unicornuate uterus is poorer compared to general population and live birth rate is as low as 29.2% while premature birth rate is as high as 44% and an ectopic pregnancy rate in females with unicornuate uterus is 4%. Pregnancy wastage rate is also higher among females with unicornuate uterus and account for 24.3% in first trimester, 9.7% in second trimester and 10.5% in subsequent days [5]. Due to variable muscular constitution of the rudimentary horn; pregnancy can be accommodated up to varying gestation in different women. It often presents as rupture of the uterine wall in the second trimester, manifesting as acute abdominal pain with intraperitoneal hemorrhage, with high risk of maternal morbidity and mortality. Despite the recent advances in the ultrasound, diagnosis of cornual pregnancy remains elusive with confirmatory diagnosis usually made during laprotomy. The aim of the present study is to analyze the obstetric implications and review the diagnostic dilemma of rudimentary horn pregnancy.

Case report

A 24-year primigravida un-booked case married since 9 months presently with 26 weeks of gestation came to emergency with severe pain abdomen since last six hours and sudden maternal collapse on arrival. She had no significant medical or surgical history in past. On examination patient was in hypovolemic shock with severe pallor, hypotension, and tachycardia, CPR was started immediately and intubated. The abdomen was tense and symphysiofundal height was 30 weeks. Her bowel sounds were normal. On pelvic examination cervix-os closed, uneffaced, fresh bleeding through os noted, and size of uterus could not be made out due to intense guarding. Immediately two large bore intravenous cannulas were inserted, one liter of fluid was rushed, patient was catheterized and urgent investigations and cross match was sent for four units of blood. Her hemoglobin was 4.6 g%, one unit blood was rushed, and after stabilization urgent emergency ultrasound was done. Her uterus was found to be empty, with a hyper echoic shadow adjacent to it. There was marked free fluid in abdomen. Immediately consultant review was sought, which revealed unicornuate uterus with rudimentary horn (Figure 1). The rudimentary horn was found to be ruptured on posterolateral wall (Figure 2(a)) with moderate free fluid in peritoneal cavity (Figure 2(b)). A dead fetus was found floating in the abdominal cavity (Figures 3(a) and 3(b)). The patient was taken for explorative laparotomy. Intraoperatively, a unicornuate uterus with rupture of rudimentary horn with rent approximately 10-12 cm in length noted with blood oozing from it, A dead Male fetus was found in peritoneal cavity with two liters of hemoperitoneum (Figures 4 and 5). Both the ovaries and tubes were normal (Figure 4) Placenta was found separated in abdominal cavity. Excision of rudimentary horn, ipsilateral salpingectomy done. Patient required massive blood transfusion and developed pulmonary edema with deranged renal function tests hence required dialysis and was treated in critical care unit for 25 days, and was discharged after a month of admission with an advice for hysterosalpingogram and intravenous pyelogram 6 weeks later.

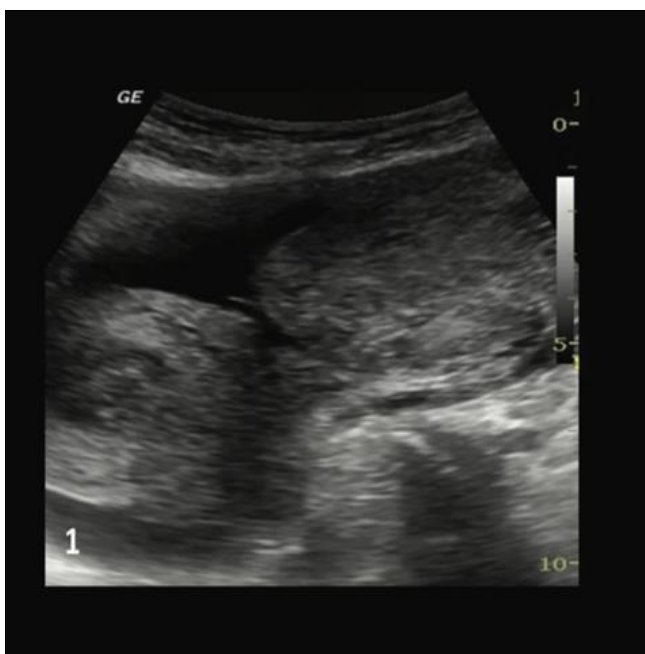


Fig 1: unicornuate uterus with ruptured rudimentary horn (Both are empty with no products of conception seen)

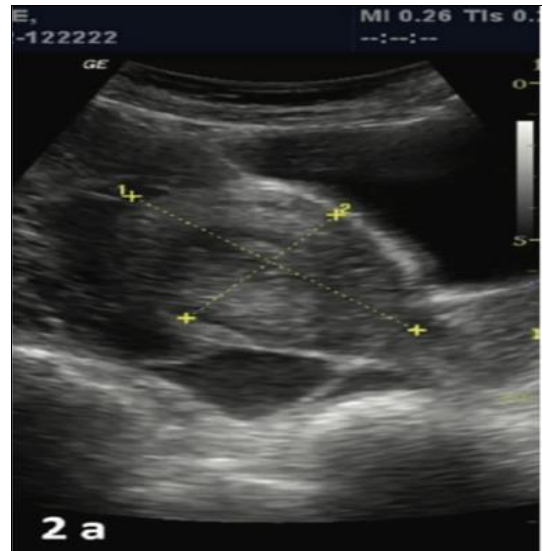


Fig 2a: rudimentary horn with rupture posterior wall and with pelvic collection,



Fig 2b: moderate free fluid in morrison's pouch

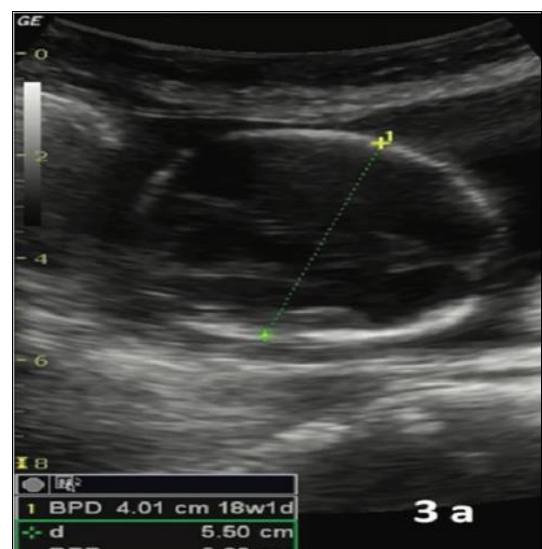


Fig 3a: fetal head lying in peritoneal cavity

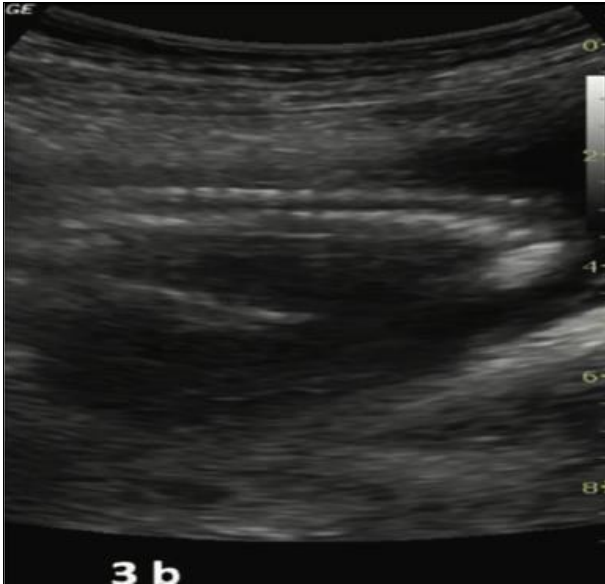


Fig 3b: Fetal spine in peritoneal cavity



Fig 4: Unicornuate uterus with rupture rudimentary horn



Fig 5: Unicornuate uterus with rupture pregnant rudimentary horn with fetus lying outside the uterus (Intraoperative)

Discussion

Pregnancies occur in both communicating and non-communicating horns in proportion to their relative incidence

and are equally likely to rupture [6]. Neonatal mortality is very high as most cases are emergency laparotomies after uterine rupture at premature gestational age [7, 8]. Maternal mortality is low (0.5%) [2] But morbidity is very high in view of massive blood loss and morbidly adherent placentation [9, 10]. This is witnessed in a study long ago by Mariceau and Vassal, who published the first description of a rudimentary horn pregnancy in 1669, and 600 cases have since been described [11].

The prerule diagnosis of pregnancy in rudimentary horn has drastically reduced maternal mortality [12]. But the sensitivity of ultrasound to detect prerule rudimentary horn pregnancy is very poor (30%) [13, 14], probably because of rarity of the diagnosis and nonfamiliarity of the radiologists about this potentially lethal condition. Early diagnosis before rupture can be managed laparoscopically by immediate excision of the horn, pregnancy, and the ipsilateral fallopian tube [15]. Tsafirir *et al.* proposed the following criteria for ultrasonographic diagnosis: (1) a pseudo pattern of an asymmetrical bicornuate uterus, (2) absent visual continuity tissue surrounding the gestation sac and the uterine cervix, and (3) the presence of myometrial tissue surrounding the gestation sac [19]. In any doubtful case three-dimensional ultrasound or magnetic resonance imaging should be done to avoid the potential complications. This case highlights the fact that despite having risk factor for suspected uterine anomaly, that is, previous cesarean section for fetal malpresentation, this patient was missed on routine malformation scan one week prior to the catastrophic rupture of the rudimentary horn. Her previous operative records were not reviewed, which when subsequently reviewed clearly stated the presence of unicornuate uterus with noncommunicating rudimentary horn. This woman was also not warned regarding potential complications. Hence this case is being reported to familiarize the radiologists regarding this rare but potentially lethal presentation which if diagnosed safely in prerule state can be managed laparoscopically without the associated sequelae of rupture uterus. Three-dimensional ultrasound imaging and MRI are useful tools in the improvement of diagnostic accuracy, guiding both counseling and surgical planning [20]. This case further raises the question of whether routine excision of rudimentary horn be undertaken in women with the potential complications and if pregnancy occurs in rudimentary horn first trimester laparoscopic excision should be done.

Conclusion

Despite advances in ultrasound and other diagnostic modalities, prenatal diagnosis remains elusive, with confirmatory diagnosis being laparotomy. The diagnosis can be missed in ultrasound especially in inexperienced hands [21]. Precious time may be lost due to delay in diagnosis or misdiagnosis and the general condition of the person may worsen. Timely resuscitation, surgery, and blood transfusion are needed to save the patient. Proper diagnostic methods and early referral from the peripheral hospitals is needed to reduce the morbidity and mortality of the patients. There is a need for an increased awareness of this condition especially in developing countries where the possibility of detection before pregnancy or before the rupture is unlikely, and precious time is lost in shifting these women to the referral hospital.

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Author's Contribution

Not available

Conflict of Interest

Not available

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