



A Rare Case of Spontaneous Uterine Rupture in Second Trimester Pregnancy with Bicornuate Uterus: A Case Report

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ARTICLE INFORMATION

Received: September, 19, 2021

Revised: January, 25, 2022

Available online: February 2022

KEYWORDS

Uterine Rupture, Bicornuate Uterus, Obstetric Emergency, Uterus Anomaly, Supravaginal Hysterectomy

A B S T R A C T

Uterine rupture is a public health problem in developing countries. Uterine rupture in pregnancy is a rare fatal complication with a high fetal and maternal morbidity incidence. In this case report, we present the rare case of a woman with a bicornuate uterus who had a spontaneous uterine rupture in the second trimester of pregnancy. A 34-year-old woman with eighteen weeks of gestation presented abdominal pain due to uterine rupture in the last 12 hours. We discovered abdominal pregnancy by ultrasound examination. Furthermore, an exploratory laparotomy found a bicornuate uterus with uterine rupture and a nonviable fetus. Then, we did a supra-vaginal hysterectomy to control bleeding. Spontaneous uterine rupture in second-trimester pregnancy with a bicornuate uterus can impact fetal and maternal death. This case is rare and requires prompt diagnosis and treatment.

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INTRODUCTION

A bicornuate uterus is a uterine malformation produced due to impairment in the fusion of Mullerian ducts. It is a rare anomaly, but it is associated with worse reproductive outcomes –recurrent pregnancy loss and preterm labor are the most common (Kaur P, 2021). It is characterized by the presence of a cleft > 1 cm in the external contour of the uterine fundus (Narang, Cope, and Teixeira, 2019). In pregnancy, a bicornuate uterus increase the risk of spontaneous abortion, preterm delivery, uterine rupture, cesarean section, breech presentation, and a low live birth rate (Reichman and Laufer, 2010; Fouelifack *et al.*, 2014).

Uterine rupture is a public health problem in developing countries. Spontaneous uterine rupture most often occurs during labor in the context of an injured uterus. Meanwhile, uterine rupture during pregnancy is rare (Guèye *et al.*, 2012). Uterine rupture in pregnancy is a rare and often catastrophic complication with a high fetal and maternal morbidity incidence. Many factors increase the risk of uterine rupture. However, the overall incidence of uterine rupture is low, even in high-risk subgroups. From 1976 to 2012, 25 peer-reviewed publications described the incidence of uterine rupture. There were 2,084 cases

of uterine rupture among 2,951,297 pregnant women. overall, uterine rupture rate was 1 in 1,146 pregnancies (0.07%) (Nahum, 2016).

The diagnosis of uterine rupture is not always precise. In addition, maternal and fetal morbidity and mortality because of uterine rupture are still high (Guèye *et al.*, 2012). Management of this rare pregnancy complication requires several considerations. Cesarean delivery with uterine repair or hysterectomy may be appropriate for fetal viability. However, management decisions are complicated when the fetus is viable or very premature. The traditional approach is to terminate the pregnancy by uterine repair or hysterectomy (Hawkins *et al.*, 2018).

A uterine rupture event begins with an anomaly. Uterine anomalies are variations in the separation between the two uterine cavities, found about 0.1-3% of the female population (Fouelifack *et al.*, 2014). In 6-22 weeks' gestation, bilateral paramesonephric in the Mullerian duct undergoes a process of fusion. In addition, there is a canalization to form the uterus, cervix, and upper-two-thirds vagina (Reichman and Laufer, 2010). One of these abnormalities is the bicornuate uterus, caused by abnormal fusion of ducts. It might be diagnosed before or during pregnancy (Souvizi and Esfehiani, 2016). The European Society of Human Reproduction and Embryology/European Society for Gynecological Endoscopy (ESHRE/ESGE-2013) classified the bicornuate uterus as the uterine anomaly (Grimbizis *et al.*, 2013). The gold standard diagnostics use invasive, such as hysteroscopy, hysterosalpingography, laparoscopy, or laparotomy. In addition, the 2D ultrasound cannot determine the type of uterine anomaly, but the 3D does (Woelfer *et al.*, 2001). Furthermore, MRI can also be an accurate non-invasive diagnostic modality (TAKAGI *et al.*, 2003). Most cases are detected for the first-time during pregnancy and incidentally discovered.

This study explains the case reports of second-trimester pregnancy with a bicornuate uterus. This case discussion can prevent pregnancy complications through an active role in antenatal care and appropriate supporting examinations.

CASE PRESENTATION

Mrs. DH, 34 years old, second pregnancy at 18 weeks gestation, was referred to the emergency room of a district referral hospital in East Java, Indonesia. Her chief complaint was abdominal pain in the last 12 hours. Abdominal pain started in the lower abdomen then extended throughout the abdomen without radiating pain. Before referral, the patient was diagnosed with an abdominal colic. The patient was given oral pain medication but no response. The patient's age of menarche is 12 years, with the regular menstrual cycle for 5-6 days every 28-30 days. The patient was currently using intrauterine contraception for five years. The patient had her first term pregnancy eight years earlier with the spontaneous birth of a 3400-gram baby boy. The patient had amenorrhea in 4 months. She did a home pregnancy test two months earlier, and it was positive. The patient has never had prenatal care in this pregnancy.

On clinical examination found abdominal pain, nausea, vomiting, and weakness. Blood pressure 124/84mmHg, pulse 118bpm, respiration rate 20rpm, and temperature 36.6oC. There was no pale conjunctiva and no abnormalities on lung examination. Her stomach was a bit bloated. The vagina was normal and had no discharge. However, there was pain on palpation of the posterior cervical fornix and left adnexa with a pain scale of 6 (visual analog score-VAS). 2D ultrasound examination showed free fluid and the fetus in the peritoneal cavity. Preoperative laboratory examinations were within normal limits. In addition, the SARS-CoV-2 PCR swab test was negative.

Initial management of the patient was oxygenation, rehydration, and injection of 30 mg ketorolac. We did laparotomy 2 hours after the patient came into the hospital. During exploration, the uterus was found with two horns. The left side was more extensive and contained a nonviable fetus in the peritoneum. However, the right horn was still intact. Macroscopically, the right and left ovaries and fallopian tubes were within normal limits. We performed a supra-vaginal hysterectomy on the left uterine horn. In addition, we removed the fetus to control bleeding (see figure 1).



Figure 1. Nonviable fetus and partial left uterine horn

DISCUSSION

Spontaneous uterine rupture in second-trimester pregnancy with the bicornuate uterus is rare. However, a pregnant mother with a bicornuate uterus must deal with uterine rupture. Bicornuate uterus (BU) occurs because of incomplete fusion of the two Müllerian ducts during embryogenesis. It rarely can lead to rupture of the uterus during the early pregnancy. Still, it has high mortality and morbidity rates (Hefny *et al.*, 2015). A study also reported a ruptured left horn in the bicornuate uterus. The rupture in bicornuate uterus cases occurs because of the inability of the malformed uterus to expand like a normal uterus. The rudimentary horn rupture is possible in the late first trimester or even in the second trimester (Kore S, Pandole A, Akolekar R, Vaidya N, 2000).

Fetal death can occur due to spontaneous uterine rupture. Uterine rupture can also occur in the third trimester of pregnancy. A previous case report found a 33-year-old primigravida with severe acute abdominal pain and signs and symptoms of hemorrhagic shock. Still, the intrauterine fetus was alive at 28 weeks. Unfortunately, the fetal heart rate was inconclusive with variable decelerations and fetal bradycardia (Nikolaou *et al.*, 2013). In this case, the uterine anomaly was just identified when the uterine rupture occurred in the second pregnancy. Severe abdominal pain was initially diagnosed as abdominal colic. The early signs and symptoms of uterine rupture are usually nonspecific, making diagnosis difficult and delaying definitive therapy. Generally, fetal morbidity becomes clinically significant inevitable. It occurs only 10-37 minutes From diagnosis time to delivery (Nahum, 2016).

Appropriate management of uterine rupture depends on early detection. In the past, health providers should evaluate classic signs of uterine ruptures, such as sudden abdominal pain that begins with a "ripping" sensation, vaginal bleeding, termination of uterine contractions, and fetal regression. However, recent experience has shown that these signs are unreliable and often absent. Nowadays, fetal distress is the most reliable clinical symptom (Toppenberg and Block, 2002). In this paper, the authors discovered free fluid and fetus in the peritoneum by 2D ultrasound. Then, we diagnosed the patient had an abdominal pregnancy. When we did exploratory laparotomy, we found that the uterus had an indentation in the fundus, thus forming two uterine horns without septa. The left side was more extensive and contained a nonviable fetus in the peritoneum. However, the right horn was still intact. In this case, the type of anomaly was challenging to determine because of her uterine break.

In this case report, a patient has been pregnant and had a vaginal delivery in the previous pregnancy history. We suspected that she had one normal uterine horn and one rudimentary horn. In this pregnancy, implantation in the rudimentary horn caused the uterine rupture. So, we concluded as a Hemi-uterus with a rudimentary communicating cavity. A prior case reported a possible successful pregnancy in a complete bicornuate uterus (Souvizi and Esfehiani, 2016). In addition, A study reported a bicornuate uterine rupture that resulted in a live birth at 30 weeks gestation. Four hours after rupture, the laparotomy was performed (Nitzsche, Dwiggins, and Catt, 2017). On the other hand, there was a delayed operation in this case, and the fetus was not viable. In addition, the patient never had antenatal care and any history of previous section cesarean surgery. A Case report revealed a correlation between the history of uterine rupture and spontaneous uterine rupture in 27-28 weeks of gestation (M.D. and Dewi, 2018). However, there was no history of previous uterine rupture in this case.

Asymptomatic and low prevalence make uterine screening unpopular. Another study showed a 30 weeks pregnant woman who did not respond to induction on misoprostol – 200 mcg weekly 4 hours every week. The woman was diagnosed with bicornuate uterine rupture. In this case, the patient had never had labor induction. It increases the risk of uterine rupture in uterine anomalies. Spontaneous uterine rupture in

early pregnancy is an infrequent complication and usually occurs in uterine scar tissue. Uterine anomalies are a risk factor for spontaneous uterine rupture in early pregnancy. In early pregnancy, the clinical signs of uterine rupture are nonspecific and must be distinguished from an acute abdominal emergency (Tola, 2014).

According to previous studies, many of these abnormalities might be asymptomatic. It remains undiagnosed until abdominal surgeries, such as hysterectomy. In this regard, one of the first diagnostic clues is the occurrence of obstetrical complications. Studies have shown that uterine rupture might occur during pregnancy because of a thin wall and the inability of the malformed uterus to expand like a normal one (Souvizi and Esfehiani, 2016). uterine rupture in the rudimentary horn frequently occurs in the second and third trimesters. It is an obstetric emergency. The recommended treatment is surgical resection of the horn rupture or total hysterectomy (Heinonen, 2000).

Uterine rupture during pregnancy is rare; however, the incidence increases. Critical steps for successfully managing uterine rupture include prompt diagnosis and definitive surgical management with concomitant maternal hemodynamic stabilization (Sutton *et al.*, 2016). In addition, the patient feels difficult to accept her situation. Thus, psychological assistance should support patients after a pregnancy failure.

CONCLUSION

Spontaneous uterine rupture in second-trimester pregnancy with a bicornuate uterus can impact fetal and maternal death. This case is rare and requires prompt diagnosis and treatment. Uterine anomalies are rare, but they have a high risk of uterine rupture. However, a quick response can prevent it from getting worse. Early diagnosis of uterine abnormalities before pregnancy will increase antenatal care and birth plan awareness. Premarital programs should include screening for uterine anomalies to prevent maternal death. In addition, patients with pregnancy failure should receive psychological support, especially when they can't get pregnant again.

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