Uterine rupture in a primigravid patient with an unscarred bicornuate uterus at term

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ABSTRACT

Background: Uterine rupture of an unscarred primigravid uterus is an exceedingly rare event. Cases of spontaneous rupture of an unscarred bicornuate uterus have been reported, but typically occur in the first or second trimester.

Case: A 28-year-old primigravida at 37 weeks gestation with a known bicornuate uterus and no prior surgery underwent an emergent cesarean section after presenting with severe abdominal pain and signs of fetal compromise. She was found to have a uterine rupture with the fetus free in the abdomen accompanied by a large hemoperitoneum. Both mother and baby did well postoperatively.

Conclusion: Bicornuate uterus may be an independent risk factor for uterine rupture, which can occur in primigravid patients and at any gestation.

1. Introduction

Uterine rupture is a rare but catastrophic obstetric event with high fetal and maternal morbidity and mortality. It most commonly occurs in women with prior uterine incisions, but other risk factors include grand multiparity, fetal macrosomia, history of gestational trophoblastic disease, prolonged labor, and labor augmentation with misoprostol or oxytocin [1,2]. Uterine rupture in a primigravid woman with an unscarred uterus is an exceedingly rare event, with incidence of 1:7,643 to 1:16,849 in series from the United States [2]. A recent review of published cases over a 60-year span identified only 27 occurrences of uterine rupture in a primigravid patient without prior uterine surgery, emphasizing just how uncommon it is [3].

Cases of spontaneous rupture of an unscarred bicornuate uterus have been reported, however, the majority involve an essentially ectopic pregnancy in a rudimentary horn that resulted in a spontaneous first or second trimester uterine rupture, although they have been reported up to 34 weeks [4,5]. Here we present the first reported case of uterine rupture in a primigravid patient with an unscarred bicornuate uterus at term.

2. Case

A 28 yr G1P0 at 37w0d with a known bicornuate uterus presented to labor and delivery with complaints of menstrual-like cramping for 2 h and no associated vaginal bleeding or loss of fluid. Her cervix was closed and thick. Fetal heart tracing was category I with accelerations, irregular contractions were seen on tocometer, and urinalysis was negative. She was discharged home to follow up with her primary obstetrician the next day.

1 h later the patient returned to labor and delivery with complaints of acutely worsened abdominal pain that was constant and exacerbated by movement. On exam the patient appeared pale and anxious with abdominal tenderness and rigidity. Her heart rate was 112 and blood pressure was 139/93. No fetal heart tones were initially found per doppler, so a bedside ultrasound was performed that demonstrated the fetus to be in breech presentation high in the abdomen with heart rate in the 90 s. A continuous monitor was placed and fetal heart rate immediately fell into the 60 s. The patient was then taken for an emergent cesarean section under general anesthesia.

Upon entering the abdominal cavity, the fetus was found to be outside the uterus with a hemoperitoneum of approximately two liters. A viable male infant weighing 2590 g was delivered from breech presentation. Apgar scores were 2 and 8 at one and five minutes, and cord gases drawn at delivery showed an arterial pH of 6.84 with a base deficit of 20.8. The abdomen was cleared of clots, and the uterus was inspected and noted to be bicornuate with communication between the two horns. The pregnancy had been in the left horn, which had ruptured on the medial aspect of the lower uterine segment with extension to the medial aspect of the right horn (Fig. 1). A fibrous band was noted overlying the point of fusion of the two horns. The uterus was closed in two layers and replaced into the abdomen.

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The patient required 2 units of packed red blood cells and 1 unit of fresh frozen plasma in the immediately postoperatively but had no other complications. She was discharged home on postoperative day number 3 in stable condition. Her baby was kept in the hospital until day of life number 8 due to periodic episodes of apnea and desaturation, after which he was discharged home in stable condition. The patient was counseled on the high risk of recurrence and was advised to avoid future pregnancies.

3. Discussion

Uterine rupture is most commonly associated with a prior cesarean section, and consequently it occurs very rarely in primigravid patients [6]. Walsh et al. conducted a review encompassing a 60-year period looking at all reported cases of uterine rupture in primigravidas. Of the 36 cases found, 11 had undergone prior uterine surgery, most commonly myomectomy. Twenty-five cases occurred with an unscarred uterus, and, of these, four were bicornuate uteri [3].

The presence of a bicornuate uterus is a rare mullerian duct anomaly caused by incomplete fusion of the mullerian ducts during embryogenesis [7]. There have been several reported cases of rupture in mullerian anomalies where the pregnancy implanted in a rudimentary horn, but these typically occur in the first or second trimester and are essentially ectopic pregnancies which should be considered separately from a pregnancy in the hemicorpus of a bicornuate uterus [5]. Higher rates of uterine rupture have been reported in patients with Mullerian duct abnormalities undergoing a trial of labor after cesarean delivery when compared with patients without Mullerian duct abnormalities, suggesting these anomalies may be an independent risk factor for uterine rupture [8]. The reason for this increased risk is unclear, although a less developed lower uterine segment, as well as uneven strain on the lower uterine segment from the irregular shape, have been proposed [8]. Four cases, including ours, reported a fibrous band in the form of a rectovesical ligament that runs between the two hemocorpora [4,7,9]. It is conceivable that this band has a restrictive effect on the expansion of the pregnant horn of a bicornuate uterus, thereby weakening the medial aspect of the horn.

Apart from the four cases reported by Walsh et al., our review of the literature found only two cases of uterine rupture of a bicornuate uterus in a primigravid patient. In a case from India in 2011, a primigravid patient was induced with misoprostol after a presumed eclamptic seizure at 30 weeks gestation. After labor failed to progress, ultrasound revealed an extrauterine fetal demise. The patient was transferred to a higher level of care where she underwent laparotomy with repair of a ruptured bicornuate uterus [7]. A case from Nigeria in 2012 described the spontaneous rupture of a bicornuate uterus at 20 weeks gestation in a patient who had undergone unspecified termination attempts at 8 and 12 weeks. On laparotomy the ruptured left hemicorpus was removed and the resulting defect from the right hemicorpus repaired [4]. When considering all six published cases of uterine rupture in a primigravid bicornuate uterus, the average gestational age at the time of rupture was 26.5 weeks, with a range of 20–32 weeks (Table 1). It is unclear why these events tend to occur prior to term, but our case demonstrates that uterine rupture is possible even at term in a patient with no other risk factors.

4. Conclusion

While rare, uterine rupture can occur with devastating consequences in a primigravid patient, and the presence of a bicornuate uterus may be an independent risk factor. As the first report of a ruptured primigravid bicornuate uterus at term, this case contributes to the knowledge of this rare event and emphasizes the importance of maintaining a higher index of suspicion for uterine rupture in cases of a known bicornuate uterus regardless of parity or gestation.

Disclosures

None.

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Table 1

| Author         | Year | Gestation | Patient age | Rupture site | Outcome       |
|----------------|------|-----------|-------------|--------------|---------------|
| Donnelly (3)   | 1951 | 28 weeks  | NA          | Right horn   | N/A           |
| Boogd (3)      | 1956 | 28 weeks  | 23          | Right horn   | Fetal demise  |
| Schrinsky (3)  | 1978 | 32 weeks  | 36          | NA           | Live birth    |
| Jones (3)      | 1978 | 21 weeks  | 25          | Left horn    | Preivable birth|
| Jayaprakash (7)| 1999 | 30 weeks  | 20          | Posterior right horn | Fetal demise |
| Agu (4)        | 2012 | 20 weeks  | 25          | Left horn    | Preivable birth|

Fig. 1. Ruptured bicornuate uterus after repair.