CASE REPORT

Spontaneous rupture of unscarred uterus in a primigravida with preterm prelabour rupture of membranes

Wael Sayed Mourad,¹ Debbra J Bersano,² Peter B Greenspan,³ Diane Medved Harper⁴

¹Department of Community and Family Medicine, Obstetrics Fellowship Division and Family Medicine Residency Program, University of Missouri Kansas City, Kansas City, Missouri, USA

²Department of Obstetrics and Gynecology, Truman Medical Center Lakewood, Kansas City, Missouri, USA

³Department of Obstetrics and Gynecology, University of Missouri Kansas City School of Medicine, Kansas City, Missouri, USA

⁴Department of Family and Geriatric Medicine, University of Louisville School of Medicine, Louisville, Kentucky, USA

Correspondence to

Dr Wael Sayed Mourad,
wael.mourad@tmcmcd.org

Accepted 7 May 2015

SUMMARY

Intrapartum uterine rupture is a true obstetrical emergency. Uterine rupture is associated with severe maternal and fetal morbidity and mortality. It is rare in the unscarred uterus of a primigravida. A 23-year-old primigravida with an unscarred uterus was admitted with preterm prelabour rupture of membranes at 36⁺4 weeks of gestation. Abnormal fetal heart monitoring, associated with acute onset of severe abdominopelvic pain, developed on admission. Rupture occurred prior to onset of regular uterine contractions and in the absence of any interventional oxytocin. The neonate had evidence of severe acidosis despite emergency caesarean delivery. This case highlights the importance of maintaining a high index of suspicion for uterine rupture, even in the unlikely setting of a primigravida with an unscarred uterus.

BACKGROUND

Uterine rupture during pregnancy is a rare and often catastrophic event, with an incidence of 1/1536 pregnancies. Associated maternal and fetal morbidity and mortality is high.¹ We report a case of spontaneous uterine rupture in a primigravida with an unscarred uterus, an especially rare event.

CASE PRESENTATION

A 23-year-old primigravida was admitted with preterm prelabour rupture of membranes at 36⁺4 weeks of gestation. The cardiotocograph classification was normal. Contractions were irregular and infrequent. On speculum examination, there was no advanced cervical dilation. The amniotic fluid was clear. The antenatal course, and medical and gynaecological history were complicated by cigarette smoking, low-grade squamous intraepithelial lesion on cervical smear that did not result in any cervical therapies and a urinary tract infection, which was treated. There was no history of gynaecological surgery or uterine instrumentation. There was no history of a connective tissue disorder or adenomyosis. Antenatal investigations were unremarkable. A mid-trimester antenatal ultrasound showed a normal appearing anterior placenta. Within minutes of admission, the patient reported severe abdominopelvic pain causing her to clutch her abdomen. A prolonged fetal bradycardia to 65 bpm was noted, consistent with a pathological pattern. Owing to the rapidly deteriorating state of the fetus, an obstetrical examination was deferred in favour of emergency caesarean delivery.

Emergent laparotomy following refractory fetal bradycardia revealed a vertical stellate midline anterior uterine rupture with partial fetal extrusion. During delivery, meconium stained amniotic fluid was noted. Cord gasses were abnormal. A grossly normal appearing placenta was sent for histopathological examination, which later revealed infarction of 2% of the placental surface, and no retroplacental clot. Intraoperative inspection did not yield evidence of placental abruption. The rupture site extended from the maternal left anterior fundus to the maternal right corner of the lower uterine segment. The uterus had a Couvelaire appearance, but no structural anomalies were noted. After repair of the rupture site, excellent haemostasis was noted, and thus a hysterectomy was not performed. The ovaries and fallopian tubes were normal. An estimated blood loss of 1.2 L was noted, and no blood products were administered at that time. The patient had a complicated postoperative course. On postoperative day 1, her haemoglobin dropped to a low of 47 g/L from 113 g/L preoperatively, and 81 g/L immediately postoperatively. Two units of packed red blood cells were transfused, and the haemoglobin increased to 80 g/L later that day. Urine output was 157 mL during this time interval. Owing to concern of a possible occult intra-abdominal haemorrhage, the patient was monitored more frequently. Also on postoperative day 1, the patient's maximum temperature was 37.9°C and white blood cell count was 13.8 thousand per mm³. Blood cultures were obtained and were negative. The patient became mildly hypertensive and persistently tachycardic, with values up to 150/78 mm Hg and 133 bpm, respectively. The heart rate improved to 105 bpm after the transfusion of packed red blood cells. Urine output had subsequently improved. Physical examination revealed that the patient was well perfused. Maternal toxicology screening was positive only for morphine, which was accounted for by postoperative administration of morphine via patient controlled analgesia. On postoperative day 2, the patient was found to be unresponsive, and due to concern for intra-abdominal haemorrhage, an emergency repeat laparotomy was performed, revealing a 100 mL haemoperitoneum. The repaired uterine rupture site remained intact. The Couvelaire appearance of the uterus was improved. The left fallopian tube was noted to be newly avulsed, and was subsequently repaired. Examination of the left broad ligament revealed signs of a large stable retroperitoneal

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To cite: Mourad WS, Bersano DJ, Greenspan PB, et al. *BMJ Case Rep* Published online: [please include Day Month Year] doi:10.1136/bcr-2014-207321

Figure 1 Arteriogram of the left uterine artery with active extravasation. Gelfoam slurry and vascular coil embolisation is shown. Note the points of vasoconstriction along the artery. Surgical skin staples from the patient's recent vertical midline laparotomy can be seen on the left.

haematoma, which extended to the level of the left kidney. Intraoperatively, the patient was administered 3 units of packed red blood cells, 2 units of fresh frozen plasma and 1 unit of platelets. The patient was transferred as an emergency to a tertiary care centre on the second postoperative day, where an abdominal and pelvic arteriogram demonstrated active extravasation from the left uterine artery, which was successfully treated by means of vascular embolisation (figure 1). Respiratory failure and bilateral pleural effusions, requiring a thoracentesis, complicated the patient's subsequent clinical course. Her condition gradually improved and she was discharged home in stable condition on hospital day 11.

The infant's weight was 2637 g. Apgar scores were 1, 3 and 4 at 1, 5 and 10 min, respectively. The infant's right deltoid area was lacerated during the delivery. The infant required cardiopulmonary resuscitation, but no intubation or mechanical ventilation. An initial blood gas study from a cord segment obtained during the delivery showed a severe mixed metabolic and respiratory acidosis with an initial umbilical artery pH of 6.525, pCO₂ of 151.0 mm Hg, pO₂ of 10.3 mm Hg, bicarbonate level of 12.2 mEq/L, and base excess of -30.7 mmol/L. The infant underwent hypothermia treatment protocol within 1 h of life. The infant was transferred to a tertiary care centre, and survived a complicated neonatal course. Intravenous pressure support was administered during the first 2 days of life for hypotension. Discharge from the neonatal intensive care unit occurred on the 25th day of life.

OUTCOME AND FOLLOW-UP

Twenty months after the spontaneous uterine rupture, the patient underwent a laparoscopic-assisted vaginal hysterectomy for severe chronic pelvic pain. The uterus and cervix were histologically normal. The child was assessed as having borderline development of language and motor skills at age 12 months, but cognitive skills had developed normally.

DISCUSSION

Uterine rupture of the primigravid unscarred uterus is an extremely rare event. The estimated incidence of spontaneous

rupture of unscarred uteri in developed countries is 12/100 000 pregnancies.¹ The most common risk factor for uterine rupture is a scarred uterus from previous uterine surgery, with most cases occurring in women with a previous caesarean section. Based on data from the UK Obstetric Surveillance System (UKOSS) from April 2009 to April 2010, researchers determined that uterine rupture is a rare occurrence, with an estimated incidence of 0.2/1000 maternities within the UK. Of the 159 women identified as having a uterine rupture, 139 had a history of at least one previous caesarean section.² In a review by Walsh and Baxi in 2007, 36 published cases of rupture of the primigravid uterus since 1946 were identified. Eleven of those cases involved a history of previous uterine surgery. Of the remaining 25 cases, only 2 had no known risk factors. Risk factors for the remaining cases included labour, congenital uterine anomaly, a morbidly adherent placenta, oxytocin use, prostaglandin analogues, connective tissue disorder and adenomyosis.³ In a more recent review by Uccella and colleagues in 2011, 24 cases of prelabour uterine rupture in a primigravida were identified over the previous 60 years. Of 23 cases in which specific clinical data were available, 16 cases had a history of previous uterine surgery or instrumentation, which were mostly myomectomies. Other identified risk factors included placenta percreta, a congenitally abnormal uterus, Ehlers-Danlos syndrome, a thin uterine wall and adenomyosis. Only one case had no clear risk factor.⁴ The case presented involves the extremely rare occurrence of spontaneous uterine rupture in an unscarred primigravid uterus that was congenitally normal, was without any history of uterine instrumentation and was not exposed to oxytocin or prostaglandin use. There was no known placental abnormality, amniotic fluid volume abnormality, fetal malpresentation, personal history of connective tissue disorder or adenomyosis, nor trauma or illicit drug use.

Given the rarity of an idiopathic spontaneous uterine rupture in an unscarred primigravid uterus, the authors speculate that, based on the patient's presentation and risk factors, a placental abruption is a more likely cause of the uterine rupture in the case described. A review of the literature did not yield a direct pathophysiological link between preterm prelabour rupture of membranes (pPROM) and myometrial compromise resulting in uterine rupture. It did, however, reveal that pPROM is a predisposing factor for placental abruption, which cannot be ruled out as a cause of uterine rupture in this case. A large population based study by Markhus *et al*⁵ in 2011 showed an OR of 2.6 for the occurrence of placental abruption in patients with pPROM compared to the general obstetrical population, confirming the results of previous retrospective studies. A large retrospective cohort study by Boisrime *et al*⁶ in 2014 demonstrated an OR of 9.5 for pPROM in patients with placental abruption, further strengthening this association. And although a formal diagnosis of gestational hypertension or pre-eclampsia was not assigned in the described case, isolated mildly elevated blood pressures were noted in the postpartum period, raising the possibility of occult gestational hypertensive disease. Along with a known history of tobacco use disorder, these serve as additional risk factors for placental abruption.⁷ The presence of a Couvelaire uterus, which is often associated with placental abruption, as well as evidence of placental infarction on histopathological examination, effectively rule in the possibility of placental abruption.^{7 8} Although a review of the literature did not identify a causative link between placental abruption and uterine rupture, there is some evidence of the association between these two entities. A 14-year retrospective analysis of uterine rupture cases by Turgut *et al*⁹ in 2013 identified

placental abruption as the most common coexistent obstetrical pathology, occurring in nearly 1 in 20 cases. For these reasons, the authors speculate that, as compared to a truly idiopathic spontaneous rupture of an unscarred primigravid uterus, a more likely cause of the uterine rupture described is that of a placental abruption, a predisposition to which is pPROM.

In a case series by Felmus *et al*, the case of a multiparous gravida at term with no known risk factors who presented with PROM and reassuring fetal well-being before suffering a catastrophic uterine rupture was reported. In this case, pathological evaluation of the uterus removed by emergency caesarean hysterectomy revealed extensive myometrial infarction and necrosis adjacent to the rupture site secondary to thrombosis of a uterine artery. Felmus *et al*¹⁰ also cite other cases of spontaneous uterine rupture associated with myometrial infarction and uterine artery thrombosis. Whether a uterine artery thrombosis contributed to the uterine rupture in our case will remain unknown.

An additional finding during the second laparotomy was the avulsed left fallopian tube. The authors speculate that an acute rupture of the left uterine artery, possibly secondary to a previously unrecognised injury to the uterus or broad ligament, which subsequently progressed, caused a significant mass effect resulting in an acute dextrorotation of the uterus, and an accompanying avulsion of the left fallopian tube.

Learning points

- ▶ The most common risk factor for uterine rupture is a scarred uterus from previous uterine surgery, with most cases occurring in women with a previous caesarean delivery. Other risk factors include history of connective tissue disorder, trauma, fetal malpresentation, labour, congenital uterine anomaly, adenomyosis, a morbidly adherent placenta, amniotic fluid volume abnormality, oxytocin use, prostaglandin analogues and a thin uterine wall.
- ▶ The estimated incidence of spontaneous rupture of unscarred uteri in developed countries is 12/100 000 pregnancies. Most of these cases will have one or more identifiable risk factors.
- ▶ Placental abruption may be a cause of spontaneous uterine rupture in some cases.
- ▶ There is growing evidence of the association between preterm prelabour rupture of membranes and placental abruption.
- ▶ Given the potentially catastrophic nature of uterine rupture, a heightened clinical index of suspicion is required, regardless of the presence of risk factors.

The patient may have been reticent to disclose medical or surgical history that could have included previous uterine instrumentation such as curettage. This, as well as illegal drug exposures, may play a role in some cases of spontaneous uterine rupture.

Our case report adds to a limited number of reported occurrences involving spontaneous rupture of an unscarred uterus in a primigravid patient. This case highlights the association between pPROM and placental abruption, and suggests an association between placental abruption and uterine rupture. This case also reinforces the need for a heightened clinical index of suspicion for the diagnosis of uterine rupture, regardless of the presence of risk factors.

Acknowledgements The authors would like to acknowledge Gwen E Sprague, MLS, our Clinical Medical Librarian at Truman Medical Center Lakewood, for critical assistance, as well as her tireless passion for the enhancement of scholarship and contribution of knowledge. The authors would also like to acknowledge Dr Stephanie Reid, MD, our staff radiologist, for assisting in identifying a suitable arteriogram image for use in our manuscript.

Contributors WSM contributed as follows: review of manuscript with contributions to its writing; verification of clinical facts presented; acquisition and review of medical records. DJB was instrumental in the following: review of manuscript with contributions to its writing; verification of clinical facts presented; acquisition and review of medical records. PBG was instrumental in the following: review of manuscript with contributions to its writing; verification of clinical facts presented. DMH was instrumental in the following: review of manuscript with contributions to its writing.

Competing interests None declared.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

REFERENCES

- 1 Kaur J, Goel B, Sehgal A. Rupture uterus following blunt trauma at 16 weeks gestation. *Int J Reprod Contracept Obstet Gynecol* 2012;1:64–6.
- 2 Fitzpatrick KE, Kurinczuk JJ, Alfirevic Z, *et al*. Uterine rupture by intended mode of delivery in the UK: a national case-control study. *PLoS Med* 2012;9:e1001184.
- 3 Walsh CA, Baxi LV. Rupture of the primigravid uterus: a review of the literature. *Obstet Gynecol Surv* 2007;62:327–34.
- 4 Uccella S, Cromi A, Bogani G, *et al*. Spontaneous prelabour uterine rupture in a primigravida: a case report and review of the literature. *Am J Obstet Gynecol* 2011;205:e6–8.
- 5 Markhus VH, Rasmussen S, Lie SA, *et al*. Placental abruption and premature rupture of membranes. *Acta Obstet Gynecol Scand* 2011;90:1024–9.
- 6 Boisrame T, Sananes N, Fritz G, *et al*. Placental Abruption: risk factors, management, and maternal-fetal prognosis. Cohort study over 10 years. *Eur J Obstet Gynecol Reprod Biol* 2014;179:100–4.
- 7 Francois KE, Foley MR. Antepartum and Postpartum Hemorrhage. In: Gabbe SG, Niebyl JR, Simpson JL, eds. *Obstetrics: normal and problem pregnancies*. 5th edn. Philadelphia: Churchill Livingstone Elsevier, 2007:458–62.
- 8 Elsasser DA, Ananth CV, Prasad V, *et al*. New Jersey-Placental Abruption Study Investigators. Diagnosis of placental abruption: relationship between clinical and histopathological findings. *Eur J Obstet Gynecol Reprod Biol* 2010;148:125–30.
- 9 Turgut A, Ozler A, Siddik Evsen M, *et al*. Uterine rupture revisited: predisposing factors, clinical features, management and outcomes from a tertiary care center in Turkey. *Pak J Med Sci* 2013;29:753–7.
- 10 Felmus LB, Pedowitz P, Nassberg S. Spontaneous rupture of the apparently normal uterus during pregnancy; a review. *Obstet Gynecol Surv* 1953;8:155–72.

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