Spontaneous Posterior Uterine Rupture in Twin-Twin Transfusion Syndrome

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- uterine rupture
- twin pregnancy
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Abstract
Background  The maternal and fetal risks of uterine distension in rapidly progressive twin-twin transfusion syndrome (TTTS) in the setting of prior uterine scar are poorly characterized.

Case  We present the case of a 42-year-old woman, G4P1201, at 21 weeks gestation with stage-1 TTTS who developed a spontaneous posterior uterine rupture necessitating emergent laparotomy and delivery of previable fetuses, possibly due to prior uterine scar from a displaced intrauterine device.

Conclusion  TTTS may be a risk factor for uterine rupture, including uterine rupture in atypical anatomic locations. Prior unrecognized uterine scars, including perforations, may magnify the risk for atypical uterine rupture in the setting of excessive uterine distension.

Case Study
A 42-year-old woman, G4P1021, presented at 21 weeks and 3 days estimated gestational age with a monochorionic-diamniotic twin pregnancy, complicated by Quinterro stage-1 TTTS diagnosed 5 days before admission (twin A maximum vertical pocket [MVP] 1.7 cm, twin B MVP 13.3 cm, normal multivessel Doppler and bladders visualized for both twins), with several days of constant right upper quadrant pain and emesis. Before 2 weeks of admission, amniotic fluid of the twins was normal (twin A MVP 3.1 cm; twin B MVP 4.1 cm). Her obstetrical history was significant for two first trimester miscarriages followed by one term cesarean section for breech presentation. After the second miscarriage, she underwent a dilation and curettage (D&C) and hysteroscopy, which revealed products of conception and a normal appearing uterine cavity. After 6 weeks of her cesarean section, she underwent levonorgestrel intrauterine device placement. Removal of the device several years later was noted to be difficult, due to embedment into the uterine wall or possible perforation.

On presentation, she was afebrile with normal vital signs and there were no contractions on tocometer with normal fetal heart rate pattern for both twins. Her white blood count (WBC) was 8.9 \times 10^9/L, hematocrit (HCT) 29.7%, with a normal urine analysis. Initially her symptoms were attributed...
to gastroenteritis with differential diagnosis including subclinical chorioamnionitis, placental abruption, appendicitis, urinary tract infection, and nephrolithiasis. She was treated supportively with intravenous fluids and antiemetics. Given persistent abdominal pain, she underwent a magnetic resonance imaging (MRI), which revealed a normal appendix, bladder, and kidneys with moderate amount of free fluid around the liver and spleen in the paracolic gutters. On MRI, the uterus was reexamined and a spontaneous bleed was noted from the vagina. Sterile speculum examination and ultrasound were both consistent with premature rupture of membranes (PPROM) of twin A. There was again no evidence of preterm labor. She was counseled, elected for expectant management, and started on latency antibiotics. An amniocentesis for gram stain, glucose, culture, therapeutic amnioreduction, and amnio instillation of indigo carmine to confirm PPROM was offered and declined. On day of admission two, she became tachycardic to 130 beats per minute and WBC increased to 18.8 (× 10^9/L) and HCT decreased to 26.7%. Overnight, her urine output decreased to < 30 mL/h and her hematocrit decreased to 21%. Examination revealed tenderness to palpation with peritoneal signs and the decision was made to proceed with exploratory laparotomy for possible uterine rupture, or other intra-abdominal processes that would be amenable to treatment with continuation of the pregnancy. The differential diagnosis also included abruption, intraamniotic infection, or medical complications from the TTTS, given paracolic fluid seen on the MRI and the evolving clinical picture; we were highly suspicious for uterine rupture with leakage of amniotic fluid and blood into the peritoneal cavity. Fetal heart tones were present for both twins before transfer to the operating room. She was transfused two units of packed red blood cells immediately and taken for emergent laparotomy, with goal to diagnose and treat peritoneal signs and the decision was made to proceed with exploratory laparotomy for possible uterine rupture. Although uterine rupture was included in the differential diagnosis of the acute abdominal examination, the presence of an intact cesarean scar, the more likely site of uterine rupture, on MRI, initial stable maternal vital signs, and fetal cardiotocography may have delayed the initial diagnosis. Once hemostasis was achieved, the uterus was returned to the abdomen and abdominal cavity was closed. Postoperatively, she recovered quickly and was discharged on postoperative day five.

The foreign body was reviewed with the pathologist and did not appear to be part of any known IUD or IUD placement device and the exact identity remains unclear.

**Discussion**

This case highlights the rare occurrence of posterior uterine rupture in the setting of rapid TTTS in a monochorionic-diamniotic pregnancy. We postulate that there may have been an unidentified prior uterine perforation at the time of hysteroscopy, D&C, IUD placement, and/or removal. We hypothesize that the rapid distension of the uterus due to twin pregnancy and TTTS on an unripened cervix in the presence of prior posterior uterine defect may have predisposed to this posterior uterine rupture. Although uterine rupture was included in the differential diagnosis of the acute abdominal examination, the presence of an intact cesarean scar, the more likely site of uterine rupture, on MRI, initial stable maternal vital signs, and fetal cardiotocography may have delayed the initial diagnosis. Once dropping hematocrit developed, the need for emergent laparotomy became clear, however, our suspicion for a posterior rupture remained low.

Posterior uterine ruptures are rare and have been reported in trial of labor after cesarean with prostaglandin

![Fig. 1 Foreign body located in posterior cul-de-sac.](image-url)
administration and in an unscarred uterus or in a remote unrecognized uterine rupture. Rapid development of polyhydramnios of the uterus, resulting in overdistension of the uterus, has been reported as the only known risk factor for a lateral uterine rupture in a singleton gestation. Tutschek et al, report a case of midtrimester uterine rupture with rapidly developing TTTS in a woman with a prior cesarean section, which led to maternal and fetal death. Our case and this previously reported case highlight both the difficulty of diagnosis and the risk of maternal and fetal mortality in rapidly developing TTTS. In this case, the acute maternal instability suggested an intra-abdominal process in addition to symptomatic polyhydramnios and one that precluded usual treatment options for TTTS. The goal for the exploratory laparotomy was to treat the suspected abdominal process to allow continuation of the pregnancy with standard treatment for TTTS if progression to higher stage warranted treatment. The combination of risk factors including monochorionic twin pregnancy with TTTS and polyhydramnios resulting in uterine distension, with the highly likely posterior uterine scar from complications of IUD placement and removal likely resulted in an atypical uterine rupture, despite the early gestational age and absence of labor.

This case highlights the potential for increased risk of atypical uterine rupture when multiple risk factors are present, including some not typically associated with uterine rupture risk such as IUD placement particularly with a history of difficult removal. The presence of the foreign body without other abdominal surgery, suggests potentially more extensive complication with hysteroscopy, IUD placement or IUD removal than suspected before exploratory laparotomy. While such clinical circumstances may not preclude trial of labor after cesarean section or indicate a need for delivery in the late-preterm or early-term period, the presence of severe abdominal pain in the setting of rapidly developing uterine distension should heighten the suspicion for uterine rupture.

Disclosures
We have no financial disclosures.

References
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