Spontaneous Posterior Uterine Rupture in Twin-Twin Transfusion Syndrome

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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.

Keywords
- uterine rupture
- twin pregnancy
- twin-twin transfusion syndrome

Monochorionic–diamniotic twin pregnancies have the potential for a unique set of complications including twin-twin transfusion syndrome (TTTS), which can develop rapidly and unpredictably. While rare, rapid TTTS has been associated with uterine rupture and subsequent maternal and fetal death.[1] Additional known risk factors for uterine rupture include prior uterine scar most often at the site of prior cesarean scar, maternal age, advanced gestational age, and birth weight exceeding 4,000 g.[2] We present a case of posterior uterine rupture in the setting of rapidly developing TTTS, presenting as acute peritoneal signs in the absence of labor, which we hypothesize occurred at the site of a prior posterior uterine perforation. The purpose of this article is to raise awareness of uterine rupture as a rare complication of TTTS.

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

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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 and in the paracolic gutters. On MRI, the uterine scar was reviewed and appeared to be intact. Chest X-ray and abdominal ultrasound were within normal limits. On hospital day two, she reported leakage of fluid 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 ($\times 10^9$/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, intra-amniotic 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 exploratory laparotomy, with goal to diagnose and treat intra-abdominal complications and continue the pregnancy. The abdomen was entered using a midline vertical incision and upon entry into the peritoneal cavity copious dark old blood was encountered. The lower uterine segment was palpated and found to be intact. The uterus was palpated and noted to be approximately 40 week size with thin edematous serosa. The serosa was noted to be disrupted on the right anterior lower uterine segment with a small amount of bleeding. No obvious source of bleeding was visualized and the abdomen could not be fully explored secondary to uterine size. Intraoperative amnioreduction was performed under ultrasound guidance to access the remainder of the abdomen and 1,200 mL of clear amniotic fluid were evacuated. The uterus was then delivered through the abdominal incision. Visualization remained challenging, and blood was noted to be pooling in the posterior cul-de-sac. Examination revealed a tear in the posterior serosa to the left side of the lower uterine segment with active serosal bleeding. Multiple figures-of-eight sutures were placed in the posterior lower uterine segment to achieve hemostasis. The anterior portion of the uterus was reexamined and a spontaneous bleed was noted on the fundal portion of the uterus. Given inadequate hemostasis, the decision was made to proceed with low transverse hysterotomy and delivery of the two male previable fetuses (fetus A, 468 g and fetus B, 438 g) to prevent maternal morbidity. Following hysterotomy and emptying of the uterus, bleeding was noted in the posterior cul-de-sac and a posterior uterine rupture was confirmed, extending toward the left pelvic sidewall. A small foreign body, a flexible, tan tube approximately 2.4 cm long $\times$ 0.2 cm in diameter (►**Fig. 1**) was identified and removed from the posterior cul-de-sac. The posterior rupture was closed in a running locked fashion, followed by an imbricating layer. An uterine artery ligation suture was placed on the left uterine artery to reduce pulse pressure to the uterus. 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.

Fig. 1 Foreign body located in posterior cul-de-sac.

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

administration[3–5] and in an unscarred uterus[6] or in a remote unrecognized uterine rupture.[7] 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.[8] 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.[1] 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.

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