Recurrence of hemoperitoneum in a pregnant woman with endometriosis: A case report

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Abstract
Endometriosis is characterized by the presence of endometrial glands and stroma outside the uterus. This causes dyspareunia, dysmenorrhea and infertility. Endometriosis has been known to increase the rate of complications during pregnancy, such as miscarriage, preterm birth, fetal growth restriction, preeclampsia and antepartum bleeding. The following case describes an exceptional situation in which a patient with known endometriosis suffered a recurrent episode of spontaneous hemoperitoneum within the same pregnancy. This case highlights that while conservative management of spontaneous hemoperitoneum may gain invaluable time for fetal growth, such treatment comes with its own risks.

Key Words:
Endometriosis, pregnancy, hemoperitoneum, acute abdominal pain, uterine embolization

Introduction
Endometriosis is a chronic gynecological disease in which endometrial glands and stroma are found outside the uterus. Patients with the diagnosis often present with symptoms of dyspareunia, dysmenorrhea, dyschezia and infertility [1]. Although endometriosis does not often cause problems during pregnancy, there have been several case reports of patients with endometriosis suffering from complications during pregnancy [2-6]. The following case describes an exceptional situation in which a patient with known endometriosis suffered a recurrent episode of spontaneous hemoperitoneum within the same pregnancy. This case highlights that while conservative management of spontaneous hemoperitoneum may gain invaluable time for fetal growth, such treatment comes with its own risks. Written consent was obtained from the patient in order to present the case.

Case Presentation
A 39-year-old primigravid woman presented at 26 weeks of gestation with an acute abdomen. Her past medical history included an exploratory laparoscopy ten years prior due to peritonitis during which she was diagnosed with endometriosis. Her endometriosis was subsequently managed with oral contraceptive pills until her pregnancy by in vitro fertilization. She also had a prior laparoscopic appendectomy. She was not known for any other medical conditions. The patient received progesterone until week 17 of pregnancy and was followed by an obstetrician every 4 weeks. The risk of fetal trisomy was 1:28. Amniocentesis and fetal echocardiogram were done at weeks 18 and 22 respectively, showing no abnormalities. Her antenatal history was otherwise unremarkable until her presentation at 26 weeks. Following admission for acute abdomen, fetal moni-
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Discussion

Spontaneous hemoperitoneum is a rare complication in the third trimester of pregnancy that can be associated with serious maternal and fetal mortality and morbidity. Endometriosis affects approximately 10% of women and is a known risk factor for spontaneous hemoperitoneum during pregnancy [1, 2, 6, 7]. Ten cases have been reported in the literature [2-4, 7-13]. Bleeding in pregnant patients with endometriosis is hypothesized to be attributed to increased venous pressure in the uterine circulation causing rupture of dilated veins in endometrial lesions [5]. Additionally, the total blood volume increases by 30-40% in pregnancy, leading to a considerable enlargement of many of the blood vessels, especially in the pelvic region [14]. The inflammation caused by endometriosis causes the vessels to be more friable as well. All these factors play a role in the increased risk of bleeding in pregnant patients with endometriosis. Additionally, patients with endometriosis are more likely to undergo in vitro fertilization due to higher rates of infertility [15]. Assisted reproductive technology is an independent factor for an increased risk of bleeding, both intrauterine and extrauterine [16]. What follows is a brief review of the treatments used in this patient to achieve hemostasis. Tranexamic acid is an antifibrinolytic agent which can enhance a patient’s own hemostatic mechanism by blocking plasminogen [17]. A dosage of 1000 mg of this drug was used as an attempt to stop the patient’s second in-trabdominal bleeding episode in order to avoid another laparotomy during pregnancy. Some studies have looked at the administration of tranexamic acid prior to delivery to prevent post-partum bleeding and showed decreased blood loss [18, 19]. However, there has been no study looking at its use during pregnancy to stop hemoperitoneum. In the case of our patient, tranexamic acid was insufficient for hemostasis and the patient still required surgical management. Uterine artery embolization is a minimally-invasive procedure performed by interventional radiologists to control acute and chronic pelvic bleeding [20, 21]. This procedure is ideal in patients who are not candidates for surgery. It has low risks of complications with possibly a small association with decreased fertility in women of child-bearing age [22, 23]. The literature reports very few cases of patients with recurrent bleeding from endometriosis receiving this procedure but in the ones who did, the procedure was successful [24]. There is no report in the current literature as to the outcome of endometriotic

toring showed normal tracing. Urgent abdominal ultrasound showed hemoperitoneum with a clots surrounding the right ovary. Ruptured ovarian cyst was suspected and she was brought immediately to the operating room. One dose of betamethasone and indomethacin were given pre-operatively. There was approximately 1 liter (L) of fresh blood in her abdomen. The source of bleeding could not be found by laparoscopic exploration. Conversion to a laparotomy was then performed. The source of bleeding was noted to originate from ruptured posterior cul-de-sac uterine adhesions. The adhesions were suspicious of endometriotic implants. Hemostatic matrices (Gelfoam and FloSeal) were applied at the lesion site to stop the bleeding, after a failed attempt with vascular clips. The patient recovered well after the operation and was discharged on post-operative day 5 with oral progesterone with no signs of preterm labor or rebleeding. Betamethasone for fetal lung maturation was completed prior to discharge. She was seen for weekly follow-up and was treated for staphylococcus aureus wound infection. Three weeks later, at 29 weeks of gestation, the patient returned with recurrence of acute abdomen. Urgent ultrasound showed half a litre of blood in the abdomen. There was no sign of fetal distress on monitoring. Decision was made by the obstetrical team to start the patient on intravenous tranexamic acid 1000 milligrams (mg) every 6 hours for medical management. Overnight, the patient experienced increasingly severe abdominal pain and fetal monitor began to show fetal distress. The patient was brought in for an emergency Cae-sarean-section. A live baby girl was delivered at 29 weeks and 2 days of gestational age and admitted to the neonatal intensive care unit. The patient had 1L of blood in her abdomen, originating again from the posterior cul-de-sac. The bleeding was controlled prior to closure of the incision. Postoperatively, the patient was transfused 7 units of packed red blood cells until she was stabilized, but still continuously complained of diffuse abdominal pain. Four days after her surgery, prophylactic bilateral uterine embolization for prevention of rebleeding was performed successfully by interventional radiology. She continued to complain of abdominal pain, but did not have another episode of bleeding. The baby was admitted to the neonatal intensive care unit immediately after birth and suffered from respiratory problems and feeding difficulties. The baby also developed a grade 1 intraventricular hemorrhage which remains stable. The baby continues to improve in the neonatal intensive care 2 months after birth.

Conclusion

The patient recovered well after the operation and was discharged on post-operative day 5 with oral progesterone with no signs of preterm labor or rebleeding. Betamethasone for fetal lung maturation was completed prior to discharge. She was seen for weekly follow-up and was treated for staphylococcus aureus wound infection. Three weeks later, at 29 weeks of gestation, the patient returned with recurrence of acute abdomen. Urgent ultrasound showed half a litre of blood in the abdomen. There was no sign of fetal distress on monitoring. Decision was made by the obstetrical team to start the patient on intravenous tranexamic acid 1000 milligrams (mg) every 6 hours for medical management. Overnight, the patient experienced increasingly severe abdominal pain and fetal monitor began to show fetal distress. The patient was brought in for an emergency Cae-sarean-section. A live baby girl was delivered at 29 weeks and 2 days of gestational age and admitted to the neonatal intensive care unit. The patient had 1L of blood in her abdomen, originating again from the posterior cul-de-sac. The bleeding was controlled prior to closure of the incision. Postoperatively, the patient was transfused 7 units of packed red blood cells until she was stabilized, but still continuously complained of diffuse abdominal pain. Four days after her surgery, prophylactic bilateral uterine embolization for prevention of rebleeding was performed successfully by interventional radiology. She continued to complain of abdominal pain, but did not have another episode of bleeding. The baby was admitted to the neonatal intensive care unit immediately after birth and suffered from respiratory problems and feeding difficulties. The baby also developed a grade 1 intraventricular hemorrhage which remains stable. The baby continues to improve in the neonatal intensive care 2 months after birth.

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implants following uterine artery embolization. This patient received bilateral uterine artery embolization under fluoroscopic guidance during the postpartum period. Following the procedure, she had no recurrent episode of bleeding. In conclusion, we report the case of a pregnant 39-year-old patient known for endometriosis, who presented twice to the delivery unit for severe abdominal pain and hemoperitoneum. Spontaneous bleeding from endometriosis lesions was noted on both occasions. By proceeding with a more conservative management of not delivering the baby at 26 weeks of gestation by cesarean during the first surgery, the patient was able to gain three invaluable weeks of pregnancy, which increased the survival of her baby significantly. Despite the 3 additional weeks, the newborn still required admission to the NICU for more than 2 months. The downside of the conservative approach is that the patient required two laparotomies. Postpartum prophylactic uterine artery embolization was performed to ensure that no rebleeding occurred. Use of tranexamic acid for intrapartum hemostasis was attempted unsuccessfully. More studies are required to look at the safety or usefulness of tranexamic acid use in pregnancy.

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Conflict of Interest Statement
The authors declare no conflict of interest

References