

# Spontaneous hemoperitoneum during pregnancy: three case reports and literature review

## *Hemoperitoneo espontáneo durante el embarazo: tres informes de casos y revisión de la literatura*

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### Abstract

**Objectives:** This case report presented cases with spontaneous hemoperitoneum during pregnancy. **Case report:** Case 1 presented with acute abdominal pain with signs of shock. Cases 2 and 3 both presented with stable vital signs and the sudden decline of fetal heart rate. Cesarean section was performed at 27, 36<sup>+</sup>4, and 34 gestational weeks, respectively. Bleeding sites were founded on the surface of the uterus or the parametrium. The perinatal outcome was stillbirth, live birth, and neonatal severe asphyxia. **Conclusion:** Careful physical examination, strict monitoring of vital signs, and timely surgical intervention are critical for improving the prognosis.

**Keywords:** Intra-abdominal hemorrhage. Endometriosis. Uterine surface vascular rupture.

### Resumen

**Objetivo:** Este caso clínico presentó casos con hemoperitoneo espontáneo durante el embarazo. **Reporte del caso:** El caso 1 presentó dolor abdominal agudo con signos de shock, los casos 2 y 3 se presentaron ambos con signos vitales estables y la disminución repentina de la frecuencia cardíaca fetal. La cesárea se realizó a las 27, 36 + 4 y 34 semanas de gestación, respectivamente. Los sitios de sangrado se encontraron en la superficie del útero o el parametrio. **Conclusión:** Un control estricto de los signos vitales y una intervención quirúrgica oportuna son fundamentales para mejorar el pronóstico.

**Palabras clave:** Hemorragia intraabdominal. Endometriosis. Rotura vascular de la superficie uterina.

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## Introduction

Perinatal intra-abdominal hemorrhage due to the rupturing of surface varicose veins on the uterine surface is rare but is associated with a high rate of morbidity and mortality for both the mother and fetus. Afflicted pregnant mothers may present with symptoms of acute abdominal pain and fetal distress among others. Since spontaneous rupture occurs rapidly, diagnosis is difficult without intra-abdominal laparoscopic evaluation, and hemorrhagic shock is frequent and contributes to an overall maternal morbidity and mortality rate of approximately 49%<sup>1</sup>. With the growing awareness and recognition of this disease, recent studies have reported that the maternal mortality rate for intra-abdominal hemorrhage decreased significantly to a rate of 1.7%<sup>2</sup>. However, the perinatal mortality rate was as high as 36%<sup>3</sup>. In this report, we report three rare cases of intra-abdominal hemorrhage resulting from vascular rupture that was treated in our hospital (Table 1).

## Case 1 presentation

A 24-year-old primigravida (singleton pregnancy after embryo transfer on the 15<sup>th</sup> day of menstruation) experienced 2 days of abdominal pain that worsened 4 h before admission at a gestational age of 27 weeks. The patient had a history of infertility and endometriosis, and she presented with progressive inferior abdominal pain with upper gastrointestinal symptoms. Physical examination showed signs of hypovolemic shock, including shortness of breath, pale face, pulse 130 beats/min, her hemoglobin level was 29 g/L, Doppler ultrasound indicated extensive peritoneal effusion, and stillbirth. Immediate emergency cesarean section was performed, and intraoperative findings revealed 3600 mL of hemoperitoneum combined with uterine malformation (left side unicornous uterus and right side rudimentary uterine horn) and stage IV endometriosis. Intraoperatively, bleeding sites including ectopic endometrial vessels and the utero-ovarian vessels were located on the surface of the uterus or the parametrium concurrent with stillbirth. Infusion of blood products and rehydration was to maintain the patient's circulating blood volume, while correcting the abnormal coagulation function, ventilator to maintain breathing, after the operation, the patient was moved to the intensive care unit (ICU), continued supportive



**Figure 1.** Active bleeding was found in the proximal broad ligament of the lower left posterior uterine wall.

treatment. The patient was healed and discharged on the 6<sup>th</sup> day after the operation.

## Case 2 presentation

A 32-year-old multipara presented with fetal ultrasound umbilical blood flow spectrum abnormality at 36<sup>+</sup>2 weeks singleton pregnancy. Two days after admission, the patient suffered sudden acute abdominal pain after defecation accompanied by decreased fetal heart rate (80-90 bpm). Emergency cesarean section was performed due to fetal distress and unexplained abdominal pain. Upon intra-abdominal examination, spontaneous vascular rupture accompanied by active bleeding in the proximal broad ligament was observed (Figure 1). Intraoperative spontaneous vascular rupture in the proximal broad ligament was observed, though no obvious endometriosis was evident. The amount of bleeding in the abdominal cavity was about 2000 mL, intraoperative hemoglobin level was 44 g/L, and neonatal prognosis was good (Apgar score 9 points). The patient was transferred to the ICU after the operation and continued blood transfusion and fluid supplementation, she was discharged four days after cesarean section and her hemoglobin increased to 87 g/L.

**Table 1. Summary of clinical manifestations of the three cases presented in this study**

Characteristic	Case 1	Case 2	Case 3
Age (years)	24	32	32
Gravidity (G, P)	G <sub>1</sub> P <sub>0</sub>	G <sub>3</sub> P <sub>1</sub>	G <sub>1</sub> P <sub>0</sub>
Complications/comorbidities	Pre-eclampsia	Fetal distress	Fetal distress; moderate anemia
Termination of pregnancy (gestational weeks)	27	36 <sup>+4</sup>	34
Termination of pregnancy (method)	Cesarean section	Cesarean section	Cesarean section
Pregnancy outcome	Stillbirth (weight = 1360 g)	Live baby boy (weight = 2760 g), Apgar rsated 9 points	Neonatal severe asphyxia, neonatal death
Assisted reproduction and pregnancy	IVF-ET	No	No
Pre-pregnancy diagnosis of endometriosis	No	No	No
Uterine malformation	Left side unicornous uterus; right side rudimentary horn of uterine	No	No
Clinical manifestations	Progressively exacerbated abdominal pain with dizziness, palpitations, and tenesmus	Sudden, severe lower abdominal pain and persistent pain under the xiphoid process, with fetal heart rate decline	After torso rotation, there was pain in the pubic symphysis, which transferred to the upper abdomen; paroxysmal, with nausea and vomiting
Shock	Yes	No	No
DIC	Yes	No	No
Intraoperative observations of pelvic adhesions	Intestine, omentum and the posterior uterine wall were densely adhered and sealed in recto-uterine fossa; Bilateral uterine appendages were twisted and adhered to the posterior lobe of the ipsilateral broad ligament	No obvious pelvic adhesions	The posterior wall of the uterus and the sacral ligament were moderately adhered
Intraoperative uterine condition	The inner surface of the uterine wall was found to be intact. The rudimentary uterine horn cavity was not connected to the pregnant uterus	Vascular breach in the proximal broad ligament of the left posterior uterine wall	The left posterior wall and the left uterine horn showed extensive abnormal vasculature
Bleeding position	Active heterovascular bleeding on the surface of uterine anterior wall and left utero-ovarian vessels	Severe bleeding in the proximal broad ligament of the lower left posterior uterine wall	Active hemorrhage ~8×2 cm on the serosal surface of the left posterior wall near the oviduct
Hemostasis	Double loop transfixion in the uterine anterior wall	Transfixion of ruptured blood vessels to stop bleeding	Interrupted vascular suture at the bleeding site
Estimated bleeding volume	Abdominal and pelvic hemorrhage (including blood clots) (~3.6 L)	Abdominal hemorrhage and blood clots (~2.0 L)	Abdominal hemorrhage and blood clots (~3.0 L)

### Case 3 presentation

A 32-year-old primigravida presented to our hospital with sudden onset of acute abdominal pain at a gestational age of 34 weeks. The patient's vital signs were stable, and no progressive decrease in hemoglobin was documented. In addition, ultrasonography

did not indicate the presence of ascites. Initial pain started above the pubic symphysis and expanded to the mid-upper abdomen with nausea and vomiting. Examination revealed abdominal muscle tension and marked tenderness under the xiphoid process. Meanwhile, the fetal heart rate decreased to 80 bpm at 8-h post-admission. Abdominal pain with decreased fetal

heart rate, the patient was considered the possibility of placental abruption and an emergency cesarean section was performed. The neonatal 1-min, 5-min, and 10-min Apgar scores were 1, 1, and 0, respectively. In addition, massive abdominal hemorrhage (about 3000 mL) was observed, and uterine surface vein rupture occurred on the posterior uterine wall. An active hemorrhage with an area of  $8 \times 2$  cm on the serosal surface of the left posterior wall near the oviduct and endometriosis-like adhesions in the pelvic cavity were present, other organs in the abdomen were explored during the operation, and no other bleeding foci were found. Interrupted vascular suture stopped the hemorrhage at the bleeding site. She recovered well after the operation and was discharged on schedule.

## Discussion

Uterine surface blood vessel rupture and bleeding, characterized by diverse clinical manifestations, difficult diagnosis, and poor mother and child outcomes, is a rare event in obstetrics. The cause of spontaneous rupture of blood vessels on the surface of the uterus or in the parametrium remains unclear. Numerous studies have reported that endometriosis may be involved in the onset of this rare obstetric emergency<sup>4,5</sup>. Endometriosis occurs in more than 50% of all patients presenting with intra-abdominal hemorrhage during pregnancy and increases the risk of hemorrhage during gestation<sup>6</sup>.

In this study, cases 1 and 3 were accompanied by typical pelvic endometriosis. Presentation of endometriosis during pregnancy is atypical and can be a result of decidualization of ectopic endometrium. Leeners et al. found that pregnancy does not correlate with overall reduction in the size and number of endometriotic lesions<sup>7</sup>. Changes in hormone levels during pregnancy can greatly impact the vascularization of endometriotic tissue and cause degeneration of ectopic endometrial tissue<sup>8,9</sup>. Mechanisms that increase the risk of bleeding and intra-abdominal hemorrhage during pregnancy with endometriosis include fragility of decidualized ectopic endometrium and chronic ectopic endometrial inflammation<sup>7,10</sup>. In addition, pelvic adhesions caused by endometriosis, accompanied by uterine enlargement during pregnancy, can increase ectopic endometrial vascular tension and elevate the risk of rupture and bleeding<sup>4</sup>.

The prognosis of this disease varies significantly due to the timing of treatment. Case 1 progressed

rapidly with massive intra-abdominal hemorrhage, hemorrhagic shock, and intrauterine fetal death when the patient was referred to our hospital. The patient in Case 2 was fortunate and properly underwent cesarean section because of decreased fetal heart rate without hemorrhagic shock. The patient in Case 3 presented initially with symptoms of digestive tract discomfort and normal vital signs, and ultrasonography did not identify ascites in the pelvic and abdominal cavity. As such, she was misdiagnosed until the fetal heart rate recovered. When cesarean section was performed, a large amount of intra-abdominal hemorrhage was found during the operation with severe neonatal asphyxia and neonatal death.

The timing of intervention depends on early diagnosis and the health status of the pregnant patient. Once intra-abdominal hemorrhage is considered, vital signs in pregnancy are often unstable, regardless of gestational age. Therefore, exploratory laparotomy should be performed immediately. In addition, the necessity of cesarean section at the same time as the operation is dependent on the fetal health status. Timely diagnosis of this disease is challenging, but a number of clinical manifestations may hint at the diagnosis of this rare disease. Non-specific symptoms, including abdominal pain, nausea, and vomiting, should be taken seriously, and the possibility of intra-abdominal hemorrhage should be noted, especially if uterine rupture occurs and abruption of the placenta, HELLP syndrome, rupture of hepatic/splenic aneurysm, and appendicular perforation are excluded. Ectopic decidual hemorrhage needs to be considered, especially when there is a history of endometriosis. During laparotomy, it is necessary to determine whether there is a rupture in the ectopic blood vessels on the surface of the uterus. Although ultrasound has certain limitations for the diagnosis of intra-abdominal hemorrhage<sup>11</sup>, especially at the early stage of intraperitoneal hemorrhage, changes in the patient's vital signs and a decline in hemoglobin necessitate the repeated use of ultrasonography and even diagnostic puncture of the abdominal cavity. Due to the decreased blood supply to the placenta, changes in fetal heart rate often occur earlier than alterations in maternal hemodynamics. As such, decreased fetal heart rate can also be a sign of insufficient blood supply to the uterus.

## Conclusion

In summary, the clinical manifestations of uterine vascular rupture during pregnancy are non-specific,

and the disease progresses rapidly, seriously endangering both mother and child. For patients with confirmed intra-abdominal hemorrhage during pregnancy, identifying and monitoring for uterine vascular rupture are highly recommended. Strict monitoring, timely diagnosis, and implementing the necessary interventions are critical for improving outcomes for both mother and child.

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## Conflicts of interest

There are no conflicts of interest by any author.

## Ethical disclosures

**Protection of human and animal subjects.** The authors declare that the procedures followed were in accordance with the regulations of the relevant clinical research ethics committee and with those of the Code of Ethics of the World Medical Association (Declaration of Helsinki).

**Confidentiality of data.** The authors declare that they have followed the protocols of their work center on the publication of patient data.

**Right to privacy and informed consent.** The authors have obtained the written informed consent of the patients or subjects mentioned in the article. The corresponding author is in possession of this document.

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