

Shock caused by spontaneous rupture uterine vessels during puerperal period: a case report and literature review

H.F. WANG, S.R. RONG, A.H. ZHANG, A.L. YANG, Q. LIANG

Tianjin Third Central Hospital, Obstetrics & Gynecology Department, Hedong District, China; Technology Research Central of Public Health Ministry, Tianjin, China

Abstract. – Hemoperitoneum caused by spontaneous rupture of uterine vessels during delivery is relatively rare in obstetric hemorrhage, and even rarer during the puerperal period. It can be life-threatening without timely diagnosis and treatment; therefore, the literature on this topic is very scarce. To explore its etiology and identify its diagnosis and treatment principle, we are reporting a case of shock caused by spontaneous rupture of uterine vessels admitted in our hospital. Its etiology is still unknown, its presenting symptoms are commonly unspecific, and its diagnosis is often made during the surgery. The rupture of uterine vessels during pregnancy should be differentiated from placental abruption, uterine rupture, placenta implantation through the uterus, and abdominal organ rupture. Active and timely operative intervention can prevent the mortality. This case stresses the need for careful post-delivery monitoring for revealed postpartum hemorrhage. We will discuss possible etiologies of uterine vessels rupture during pregnancy, associated imaging findings, and management options.

Key Words:

Hemoperitoneum, Rupture of uterine vessels, Obstetric hemorrhage, Abdominal pain.

Introduction

Spontaneous rupture of uterine vessels during delivery is relatively rare in obstetric hemorrhage, and even rarer during the puerperal period. Its clinical signs and symptoms are usually vague and nonspecific without timely diagnosis, and treatment may result in serious complication of pregnancy. It's a rare OB/GYN (obstetrics/gynecology) emergency with high mortality, especially with the rupture of uterine artery¹. The pathogenesis is still unclear, and the condition is

rarely recognized preoperatively. To explore its etiology and identify its diagnosis and treatment principle, we are reporting a case of shock caused by spontaneous rupture of uterine vessels admitted in our hospital in 2015.

Case Summary

The 32-year-old pregnant woman, gravida 1, para 0, was previously healthy. After 39 weeks of pregnancy at 15:40 on June 7, 2015, the woman delivered a female baby per vagina smoothly, with the Apgar score at 10. There was no history of abdominal pressure increase or curettage. Postpartum uterine contractions are good. After 10 hours of delivery (1:40, 2015-06-08), the patient gave her first urination beside the bed under her families help, which was smooth, the patient went through no abdominal pain and trauma. After that, the patient went to bed for rest. At about 2:00, the patient developed sudden symptoms of shortness of breath, palpitation, sweating, nausea, and started agitating. Body results showed the body temperature of 36.5°C, blood pressure of 85/68 mmHg, pulsing as 88 beats/min, respiratory rate of 20/min, oxygen saturation of 99-100%, clear consciousness, anemia, clammy skin of limbs, lips cyanosis, soft abdomen, complete abdomen tenderness, rebound pain, without muscle tension, keen tenderness of uterine under compression, one finger length under the umbilical and good performance of uterine contraction, with a small amount of vaginal bleeding. In case of amniotic fluid embolism or postpartum circulatory failure, immediate ECG monitoring was given with oxygen mask and two vein channels established, crystalloid, and colloid were supplemented. The blood and plasma provision were urgently prepared, retention catheter was placed and routine checking for blood and blood coagulation function were urgently given. The

symptoms of palpitation and shortness of breath were aggravated, and the blood pressure was progressively decreased, the lowest at 44/27 mmHg. The oxygen saturation was normal, intravenous infusion of 20 mg of dexamethasone was given, and Dopamine and Aminophylline were given for the treatment of hypertension. The monitoring blood pressure was risen to 95/54 mmHg. At 2:30, the ultrasound bedside showed a large amount of abdominal free liquid, and the depth of free liquid around liver, spleen, and pelvis was respectively 6.0 cm, 2.5 cm, and 3.2 cm. At 2:40, abdominal puncture was performed to extract the dark red uncoagulated blood, after that, acute hemorrhagic shock was considered, with the causes unknown. The returned blood test showed that the hemoglobin of the patient was 50 g/l, with blood coagulation dysfunction. Emergency laparotomy and emergency blood transfusion were immediately given.

Intraoperative findings suggested that the volume of intraperitoneal free blood and gore was about 3,500 ml, and the size of uterine was similar to that of 16 weeks pregnant women, and the uterine surface was smooth; moreover, tortuous engorgement was seen in the vein on the near broad ligament of the left rear wall, with a vein rupture with the diameter of 0.3 cm (Figure 1) and active hemorrhage could be seen. The suture was given to the uterine vascular rupture for hemostasis after clearing up the he-

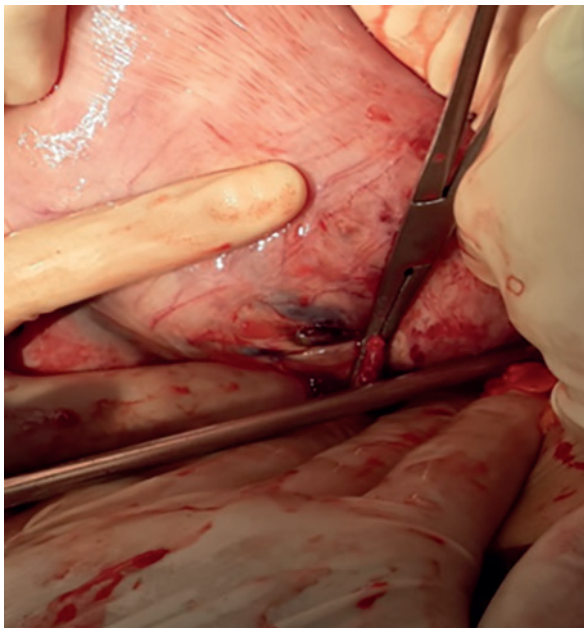


Figure 1. The bleeding site of uterine.

matocele and blood clot. The lowest hemoglobin during the surgery was 32 g/l, therefore, four units of Packed Red Blood Cells (PRBC) were transfused during the operation. During the abdomen-closing process, disseminated bleeding could be seen on the wound surface in the abdominal incision of the patient, with symptoms of disseminated intravascular coagulation (DIC). Four units of frozen fresh plasma and 750 ml autologous blood transfusion were transfused immediately, and other four units of PRBC were transfused after the surgery. A little rotten brittle tissue taken from the ruptured blood vessels was sent for pathological examination. The pathologic results showed a little granulation tissue, part of the blood vessel was seen with cystic dilatation complicated with organization and inflammatory exudation, and bleeding. The patient was rehabilitated and discharged from our hospital seven days after surgery.

Discussion

Hemoperitoneum caused by pregnancy uterine vascular spontaneous rupture is a rare but it is a potentially lethal clinical disease with prevalence during pregnancy at 1/10000 and the perinatal maternal mortality is very high (31%)². The etiology is still unknown, and the disease can occur on any period of pregnancy, but more common on third trimester. The most frequent site of the rupture is the broad ligament³, just as this case of patient. The statistical studies recorded in the foreign literatures say that the prevalence of this disease is 61% before childbirth, 18% during childbirth, and 21% in early puerperium. It has been reported one case of rupture bleeding of uterine vessels during puerperal period⁴. Chinese studies have no such report about uterine vascular spontaneous rupture during puerperal period.

The presenting symptoms are commonly unspecific. Clinical presentations are usually hypovolemic shock signs with severe abdominal pain and reduction in hemoglobin levels. The pathogenesis is unclear, and the possible reasons are: (1) markedly increase of blood flow in the uterine during late pregnancy, and the blood during pregnancy is in hypercoagulable state, with slow blood flow susceptible to venous engorgement and vein varicosity which could be clearly seen in this case of patient; (2) under uterine compression on the post cava, the backflow of the blood in pelvic cavity was obstructed, which

caused the uterine venous pressure to increase, which rupture bleeding of uterine vessels before delivery or childbirth may be associated with. But this factor could be ruled out in this case, since the patient has been delivered successfully; (3) the surface of veins under perimetrium was superficial, with thin walls and no outer sheath. When the uterine contracts or given external impact or sudden increase of abdominal pressure, the superficial yet turgor vessels may be torn or damaged to rupture and bleeding. The rupture of blood vessels on the surface of the uterine after urination may be associated with this reason. The pressure of the inferior vena cava and iliac vessels by the gravid uterus leads to an overall increase in the venous pressure⁵. If endometriosis or inflammation occurred, based on the anatomic and hemodynamic changes, the veins under perimetrium become more superficial, blood vessel more brittle, more susceptible to engorgement, and even bareness, thus more vulnerable to bleeding. The uterine fibroid is a high-risk factor of spontaneous rupture of uterine vessels. The bleeding site of the surface of the fundal myoma was often derived from a superficial, tortuous, and dilated vein⁶. This patient was healthy previously. After urination, the surface vessel of the uterine was ruptured, which was thought to be associated with the sudden increase of the abdominal pressure during urination. In retrospective studies it was found that 90% of the rupture site of vessels was located in the posterior wall of the uterus and parametrial tissue, and 52% of the patients were complicated with endometriosis⁷. Further, a case of hemoperitoneum is reported resulting from uterine vein rupture at a delivery as a delayed consequence of laparoscopic resection of DIE (deep infiltrating endometriosis) and suggested that the operation for DIE included a risk of uterine vessel rupture during pregnancy⁸. Some scholars claim that endometriosis is the main risk factor of spontaneous rupture of uterine blood vessels. In this case of patient, a little rotten brittle tissue taken from the ruptured blood vessels was sent for pathological examination, and the pathologic results showed a little granulation tissue, part of the blood vessel was seen with cystic dilatation complicated with organization, inflammatory exudation, and bleeding. Malformation of the uterine or blood vessels may also be one of the factors to explain the spontaneous rupture. It was reported that a patient pregnant with twin fetuses for 28+1 weeks

experienced abdominal hemorrhage as a result of spontaneous rupture of uterine vessels and rupture of rudimentary horn⁹. Moreover, oversize of the fetuses or excessively high tension of the uterine can also induce spontaneous rupture of veins under perimetrium¹⁰.

The rupture of uterine vessels during pregnancy should be differentiated from placental abruption, uterine rupture, placenta implantation through the uterus, and abdominal organ rupture. For persistent abdominal pain during pregnancy for unknown reasons, intra-abdominal hemorrhage should be taken into account, and close monitoring of vital signs and hemoglobin should be given based on the patient's medical history. Ultrasound examination is difficult to find patients with abdominal bleeding during pregnancy. Furthermore, vaginal ultrasound or CT or MRI examination is needed. Unfortunately, abdominal bleeding still cannot be found. The posterior fornix or abdominal puncture can help to confirm the diagnosis. Hemorrhage caused by puerperal uterine vascular rupture is rare yet difficult to diagnose and needs to be differentiated from uterine pseudotumor or uterine rupture of aneurysms. Literatures reported that, the manifestations of acute abdominal pain and shock one hour after vaginal delivery of mature baby has been confirmed by abdominal puncture as intra-abdominal hemorrhage, and laparotomy found a piece of solid lump at the bottom of the uterus, measuring 6 cm-5 cm, with rupture and bleeding of the surface blood vessel, which the postoperative pathology indicated as granuloma and calcification of collagen and fibrous tissue¹¹. In addition, it still needs to be differentiated from postpartum atypical amniotic fluid embolism and pulmonary embolism. The symptoms of amniotic fluid embolism or pulmonary embolism are shock, dyspnea, reduction of oxygen saturation and heart rate, and bilateral pulmonary diffuse patchy infiltrates shown in bedside chest X-ray. On the other hand, however, hypovolemic shock caused by intra-abdominal hemorrhage was mainly manifested as normal oxygen saturation and abdominal effusion shown by the ultrasound. When the symptoms of progressive decrease of the blood pressure, cyanosis of lips, complete abdomen tenderness, and rebound tenderness occurred, acute abdominal disease was not taken into account. In the very beginning, the patient was misdiagnosed as amniotic fluid embolism and was given dexamethasone for treatment; only after given ultrasound

for reexamination, the patient was immediately treated with intraperitoneal puncture to extract uncoagulated blood and was given treatment based on hypovolemic shock. The misdiagnosis is the reason for the occurrence of disseminated intravascular coagulation (DIC), which has put the patient's life into risk.

Although spontaneous rupture of uterine vessels during puerperal period is rare, its treatment principle is similar to the hemorrhage during pregnancy caused by other causes, for which transfusion of crystalloid, colloid, and blood products is very crucial to maintain blood volume. Treatment is based on the systemic correction of hypovolemia and immediate surgery *via* laparotomy or laparoscopy in cases in the first trimester of pregnancy for hemostatic purposes¹. Emergency exploratory laparotomy allowed for etiological diagnosis and treatment. Once diagnosed, the patients need to be given immediately open abdominal operation regardless of the conditions of the fetus. During the operation, identifying the bleeding site is the priority, after which suture, and hemostasis can be given. Racing the time to perform laparotomy, hemostasis or cesarean section to realize bleeding control is the only effective treatment to ensure maternal and child safety. The key to the successful rescue of this patient is early diagnosis, with only 40 minutes taken from the onset to diagnosis, and timely laparotomy and effective correction of hypovolemic shock and DIC are crucial for the success of the treatment. Pelvic arterial embolization is a good option in the management of puerperal hematomas, if the patient is hemodynamically stable and the necessary equipment and staff are available¹². Xu¹³ reported that the uterus was preserved in 20 patients after a single embolization of internal iliac artery or uterine artery in the treatment of massive obstetrical and gynecological hemorrhages, including postpartum hemorrhages. The main advantage of this technique is the low rate of serious complications and the preservation of reproductive function. If both of these conditions are not met, as in this case, then laparotomy is indicated.

Conclusions

The postpartum uterine vessels rupture is very rare in clinical treatment, which shows no typical symptoms, thus very easy to be misdiagnosed.

Once it happens, the progression of the disease is very rapid, with high maternal mortality. Although very rare, hemoperitoneum should be included in the differential diagnosis when a pregnant woman experiences acute-onset, severe abdominal pain, even without an episode of abdominal trauma.

Conflict of Interest

The Authors declare that they have no conflict of interests.

References

- 1) Hardin N, Delozier A, Torabi A, Laks S. Spontaneous rupture of the uterine artery in an otherwise normal pregnancy. *J Radiol Case Rep* 2017; 11: 7-13.
- 2) Díaz-Murillo R, Tobías-González P, López-Magallón S, Magdaleno-Dans F, Bartha JL. Spontaneous hemoperitoneum due to rupture of uterine varicose veins during labor successfully treated by percutaneous embolization. *Case Rep Obstet Gynecol* 2014; 2014: 580384.
- 3) Giulini S, Zanin R, Volpe A. Hemoperitoneum in pregnancy from a ruptured varix of broad ligament. *Arch Gynecol Obstet* 2010; 282: 459-461.
- 4) Ziereisen V, Bellens B, Gérard C, Baeyens L. Spontaneous rupture of utero-ovarian vessels in postpartal period: a case report and review of the literature. *J Gynecol Obstet Biol Reprod (Paris)* 2003; 32: 51-54.
- 5) Thain S, Rajeswari K. A rare case of spontaneous rupture of uterine surface vessels in pregnancy mimicking acute appendicitis. *J Obstet Gynaecol Res* 2019; 45: 1197-1200.
- 6) Jenayah AA, Saoudi S, Sferi N, Skander R, Marzouk SB, Cherni A, Sfar E, Chelli D, Boudaya F. Spontaneous subserosal venous rupture overlying a uterine leiomyoma in a young woman. Spontaneous subserosal venous rupture overlying a uterine leiomyoma in a young woman. *Pan Afr Med J* 2017; 28: 205.
- 7) Brosens IA, Fusi L, Brosens JJ. Endometriosis is a risk factor for spontaneous hemoperitoneum during pregnancy. *Fertil Steril* 2009; 92: 1243-1245.
- 8) Wada S, Yoshiyuki F, Fujino T, Sato C. Uterine vein rupture at delivery as a delayed consequence of laparoscopic surgery for endometriosis: a case report. *J Minim Invasive Gynecol* 2009; 16: 510-512.
- 9) Cai XH, Lin L. Hemoperitoneum in third trimester pregnancy: 2 cases report and literature review. *Chinese Journal of Clinical Obstetrics and Gynecology* 2015; 16: 181-183.

- 10) Doger E, Cakiroglu Y, Yildirim Kopuk S, Akar B, Caliskan E, Yucesoy G. Spontaneous rupture of uterine vein in twin pregnancy. *Case Rep Obstet Gynecol* 2013; 2013: 596707.
- 11) Ekane GH, Tebeu PM, Obinchemti TE. Postpartum hemoperitoneum due to rupture of a blood vessel on a uterine pseudo tumor: a case report. *BMC Res Notes* 2014; 25: 106-109.
- 12) Chughtai NG, Rizvi RM. A rare case: rupture of internal pudendal and uterine artery in a vaginal delivery. *J Coll Physicians Surg Pak* 2018; 28: 49-50.
- 13) Xu JQ. Effectiveness of embolization of the internal iliac or uterine arteries in the treatment of massive obstetrical and gynecological hemorrhages. *Eur Rev Med Pharmacol Sci* 2015; 19: 372-374.