

Endometriosis-Related Hemoperitoneum in Pregnancy: A Diagnosis to Keep in Mind

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Background: Endometriosis is an important gynecologic clinical entity, pathologically defined by the ectopic presence of endometrium and frequently associated with pelvic pain, that affects approximately 10% of females of reproductive age. A rare but severe complication of endometriosis during pregnancy is spontaneous hemoperitoneum in pregnancy (SHiP), severe intraabdominal bleeding that can be life threatening.

Case Report: We present the case of a patient with SHiP at 29 weeks of pregnancy. A supraumbilical midline laparotomy was performed, and pelvic exploration revealed a lacerated and bleeding right ovary. Right annessiectomy was performed, and a cesarean section was performed because hemostasis was not achievable. A healthy baby was born, and hemostasis was finally achieved.

Conclusion: We believe that in gravid females with a history of endometriosis, severe abdominal pain, and a reduction of hemoglobin, physicians should always suspect SHiP. Because SHiP is a life-threatening condition for both the mother and the baby, a prompt diagnosis must lead to prompt treatment.

Keywords: *Cesarean section, endometriosis, hemoperitoneum, neonatology, pregnancy, premature birth*

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INTRODUCTION

Endometriosis is an important gynecologic clinical entity, pathologically defined by the ectopic presence of endometrium and clinically associated with pelvic pain. A rare but severe complication of endometriosis during pregnancy is spontaneous hemoperitoneum in pregnancy (SHiP), life-threatening intraabdominal bleeding.

Endometriosis represents one of the main risk factors for SHiP because of the rupture of utero-ovarian vessels or bleeding from endometriosis implants. Because an increased number of patients with severe endometriosis achieve pregnancy thanks to assisted reproduction techniques, physicians should be aware of endometriosis-related SHiP as a cause of acute abdomen during the third trimester of pregnancy. We report the case of a patient with severe abdominal pain caused by SHiP.

CASE REPORT

A 33-year-old gravida 3 para 1 patient was admitted to the hospital for abdominal pain at week 29 of pregnancy. Her medical history was significant for appendectomy at 15 years of age and endometriosis. She underwent laparoscopy in 2007 and 2008 for coagulation and excision of superficial peritoneal endometriosis.

The patient's main symptom was a severe stabbing pelvic pain that started after intercourse. Examination revealed a gravid abdomen of 29 weeks, remarkable for a diffuse tenderness that worsened at the right superior quadrant with some guarding but no rebound and positive bowel sounds. The pain was steady and diffuse. She denied dysuria, constipation, and diarrhea but reported nausea and vomiting.

A fetal assessment confirmed the presence of a normal singleton pregnancy with a regular left-located placenta and a normal amniotic fluid volume. Tocography was negative, and the cervical length was 35 mm. Complete blood count, metabolic profile, and urinalysis were performed. The patient's vital signs were normal: blood pressure of 120/70 mmHg, pulse of 109 beats per minute, and respiratory rate of 15 breaths per minute. Her laboratory results showed hemoglobin of 8.8 g/dL with $18.8 \times 10^9/L$ leukocytes. Metabolic hepatic panel and urinalysis were negative.

As the patient's pain and history were suggestive for acute cholecystitis, acute pyelonephritis, or gastroenteritis, an abdominal and pelvic ultrasound was performed that revealed free fluid in the abdomen; the liver, spleen, kidneys, and cholecyst were regular. Within 7 hours of admission, the patient gradually complained of worsening



Figure. Attempt to repair the excision after bleeding from the lacerated right ovary and from the right-posterior surface of the uterus was observed.

pain. Steroid therapy with betamethasone was administered for fetal lung maturation.

Abdominal and pelvic magnetic resonance imaging was performed but was hampered by the presence of the gravid uterus. Imaging was only able to detect a large quantity of free fluid in the abdomen and pelvis. In a few hours, the patient's hemoglobin dropped to 7.6 g/dL.

We decided to perform surgery because of the patient's worsening clinical features and the diagnosis of acute abdomen and SHiP. The patient underwent a supraumbilical midline laparotomy under general anesthesia.

Almost 1,500 mL of blood was aspirated from the peritoneal cavity. Pelvic exploration revealed a bluish color on the posterior right side of the uterus. The right fallopian tube and ovary had adhesive bands with the right uterine wall. Active bleeding from the lacerated right ovary and from the right-posterior surface of the uterus was observed. Right annessiectomy was performed with an interrupted 2-0 VICRYL (Ethicon US, LLC) single-layer suture to repair the excision (Figure).

Because hemostasis was not achievable with the gravid uterus, a cesarean section was performed. A male baby was extracted who weighed 1.390 kg and whose Apgar scores were 4 and 7 at the first and fifth minute, respectively. After closing the cesarean breach, the uterus reduced its volume, and the bleeding at the site of annessiectomy lessened with few bleeding points left. Hemostasis was achieved with electrocoagulation and a second layer of 2-0 VICRYL interrupted suture.

After careful confirmation of hemostasis, the abdominal wall was closed. Intraoperatively, the patient received 2 units of red blood cells and 2 units of fresh frozen plasma. She recovered well and was discharged on postoperative day 5. The infant had an uneventful course in neonatal intensive care and was discharged 1 month after birth.

DISCUSSION

Endometriosis is a common gynecologic condition that affects approximately 10% of females of reproductive age. It is a chronic inflammatory disease defined as the presence of endometrial-like tissue outside the uterine cavity. Endo-

metriosis generally involves the peritoneum, ovaries, and rectovaginal septum but may also involve other abdominal or extraabdominal sites. Clinically, the symptoms include dysmenorrhea, pelvic pain, deep dyspareunia, and infertility.¹⁻³

Endometriosis is associated with an increased risk of early pregnancy complications, such as ectopic pregnancy and miscarriage, but the literature contains few reports of complications linked to severe endometriosis during the third trimester of pregnancy.² A rare but potentially catastrophic complication occurring in advanced pregnancy is hemoperitoneum as a result of spontaneous bleeding from blood vessels.¹⁻⁴

During pregnancy, hemoperitoneum caused by spontaneous rupture of uterine vessels is an extremely rare condition but is almost always a life-threatening complication associated with maternal and perinatal mortality.^{2,5} Published series report maternal mortality ranging from 3.6%-49%.² Maternal mortality associated with SHiP dropped dramatically during the second half of the 20th century to approximately 4%, but fetal mortality remains high (approximately 31%), with 44% of the deaths attributable to maternal shock.^{1,6} The diagnosis of this condition is difficult and rarely reached without surgery. Laparotomy is often needed to manage these patients. Typical presenting symptoms are sudden intense abdominal pain and signs of hypovolemic shock; a marked drop of hemoglobin level is a frequent finding. The pitfall of SHiP is differential diagnosis. Placental abruption is the most common preoperative misdiagnosis. Other differential diagnoses include uterine rupture; abdominal pregnancy; ruptured appendix; severe preeclampsia; HELLP (hemolysis, elevated liver enzymes, low platelet count) syndrome; acute fatty liver disease; chorioamnionitis; and hepatic or splenic rupture.²

Approximately 25 cases of SHiP have been described in the literature during the past 20 years. This condition has been described out of labor (61%), during labor (18%), and in the early postpartum period (21%).^{6,7} Twenty-nine percent of out-of-labor SHiP cases have been reported to occur before 33 weeks of gestation, 39% of cases occurred between 33-37 weeks of gestation, and 32% of cases occurred at term. Endometriosis is recognized as the major risk factor for SHiP.^{1,2}

Hemoperitoneum in pregnant women with endometriosis can be caused by spontaneous rupture of utero-ovarian vessels or bleeding endometriosis implants. The spontaneous endometriosis-linked rupture of utero-ovarian vessels may be attributed to 3 factors: chronic inflammation caused by endometriosis may cause utero-ovarian vessel leakage;² adhesions may create tension on these vessels as the uterus becomes enlarged in pregnancy;¹ decidualization of endometrial foci during pregnancy may cause utero-ovarian vessel perforation.^{2,3,6} Moreover, venous pressure in the utero-ovarian circulation increases during pregnancy. The venous pressure can be accentuated by physical effort, such as muscular activity, coughing, defecation, coitus, or the pushing phase of labor.² Our patient reported intercourse as the trigger of abdominal pain. Although in a few cases the origin of bleeding remains unknown even during laparotomy, arteries and superficial veins or varicosities on the posterior surface of the uterus or parametria are often the site of bleeding.⁷

The 2009 study by Brosens et al reviewed all cases of SHiP described since 1987.⁷ The authors noticed that SHiP was associated with endometriosis in >50% of cases; the diagnosis of endometriosis was unknown until then in almost half of the affected women. Biopsy samples of the site of bleeding during pregnancy were obtained in 5 cases that revealed endometriosis, characterized by prominent vascularization and decidualization of the lesion. In our patient, the diagnosis of endometriosis was already known, and the pathologic examination of the specimen confirmed the presence of decidualized ectopic endometrial tissue.

Several aspects of treating endometriosis remain controversial: whether surgical treatment for endometriosis should be adopted not only to improve fertility but also to prevent significant pregnancy complications such as SHiP; whether a tailored prenatal follow-up for pregnant women with endometriosis should be implemented and how it should be performed (obstetrics and gynecology department visit, ultrasound, magnetic resonance imaging); and whether cesarean section should be performed routinely to prevent irreversible fetal damage when laparotomy is required.²

Lesions caused by severe endometriosis can lead to significant abdominal bleeding during the third trimester of pregnancy.² Bleeding from endometrial implants is an established cause of acute hemoperitoneum in pregnancy, and physicians should keep it in mind in cases of abdominal pain during pregnancy.¹ Prompt intervention may be required in such cases, and an expert surgical team should be involved with adequate availability of blood products.²

CONCLUSION

We believe that in gravid females with a history of endometriosis, severe abdominal pain, and a reduction of hemoglobin, physicians should always suspect SHiP.

Because SHiP is a life-threatening condition for both the mother and the baby, a prompt diagnosis must lead to prompt treatment. A greater awareness of SHiP and its associated risk factors, such as pelvic endometriosis, may facilitate the diagnosis of this condition and expedite the intervention to improve maternal and fetal outcomes.

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