

Pernicious placenta previa/ placenta percreta complicating active systemic lupus erythematosus resulting in postoperative artery thrombosis

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Abstract

Systemic lupus erythematosus (SLE) increases the risk of adverse pregnancy outcomes and fetal complications. Placenta percreta, involving placental attachment to another organ, is a rare but severe placental abnormality. We report a 26-year-old woman, G2P1, with a 6-year history of SLE with coexisting pernicious placenta previa and placenta percreta detected by second trimester ultrasound. She discontinued prednisone 5 months before admission, without consultation, and active SLE was diagnosed on admission. Because of her progressive condition, the patient underwent infrarenal abdominal aorta balloon occlusion and double J ureteral catheter placement, followed by elective cesarean at 27⁺⁶ weeks gestation. Despite aggressive management, she experienced severe bleeding requiring internal iliac artery ligation and peripartum hysterectomy. The placenta had penetrated the uterus walls and attached to the bladder apex, necessitating bladder repair. Thrombosis was detected in the common iliac artery and common femoral artery in the right leg 1 day postoperatively. Conservative antithrombotic therapy had little effect, and embolectomy by arteriotomy was performed on the 6th post-cesarean day, and an arterial thrombus was removed. Infrarenal abdominal aorta balloon occlusion may increase the risk of postoperative thrombosis in pregnant women with active SLE and coagulation disorders. These patients therefore require close monitoring and timely anticoagulation.

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Keywords

Systemic lupus erythematosus, pernicious placenta previa, placenta percreta, infrarenal abdominal aorta, balloon occlusion, thrombosis

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Introduction

Systemic lupus erythematosus (SLE) is a multisystemic autoimmune disorder with heterogeneous manifestations that is prevalent in females of reproductive age. Pregnant women with SLE are at higher risks of adverse pregnancy outcomes and complications, such as severe preeclampsia, infections, thromboembolic complications, and mortality.¹

Pernicious placenta previa is a specific type of placenta previa that occurs when the placenta attaches to previous cesarean scars.² Placenta implantation is classified into three types, according to the depth of placental invasion of the uterus: placenta accreta, placenta increta, and placenta percreta, respectively.³ Among these, placenta percreta is the least common but most severe type. In cases of placenta percreta, the placenta invades into adjacent organs, such as the bladder, resulting in significantly increased risks of uncontrollable bleeding during delivery, high maternal morbidity, and often the need for extensive life-saving surgical interventions.

Here we describe a rare case of pernicious placenta previa coexisting with placenta percreta in a patient with active SLE, and complicated by postoperative artery embolism.

Case Report

A 26-year-old woman (gravida 2, para 1; body mass index, 28.4) with a history of cesarean section due to pregnancy hypertension 3 years previously, was referred to our hospital with paroxysmal abdominal

pain and irregular uterine contractions at 26⁺³ weeks gestation. She had a 6-year history of SLE and had discontinued prednisone 5 months before admission, without consulting her doctor. The patient had no clear history of menopause and had not received regular antenatal check-ups. Routine blood tests 12 days before admission showed hemoglobin (Hb) 93 g/L and a platelet count of $68 \times 10^9/L$ (reference ranges: 115–150 g/L and $125\text{--}350 \times 10^9/L$, respectively), indicating anemia and thrombocytopenia. Ultrasonography examinations on admission revealed a live intrauterine fetus. The placenta was located directly on the internal cervical os, and the zone between the placenta and myometrium was unclear, with abundant blood flow between the placenta and the bladder (Figure 1). Based on these findings, a diagnosis of pernicious placenta previa coexisting with placenta percreta was made, and was confirmed by pelvic magnetic resonance (MRI) (Figure 2).

On admission, abnormal laboratory results and signs included anemia (Hb, 88 g/L), thrombocytopenia (platelets, $68 \times 10^9/L$), proteinuria, positive autoantibody spectrum (positive anti-SSA/Ro52kD antibody and anti-SSA/Ro60kD antibody, and weakly positive anti-dsDNA antibody), and abnormal cardiac ultrasound findings (moderate aortic incompetence, left ventricular enlargement and reduced left ventricular diastolic function, with an ejection fraction of 55%), and sacrococcygeal pain of unknown origin. Together, these findings indicated active SLE. Anti-SLE treatment was given immediately, including

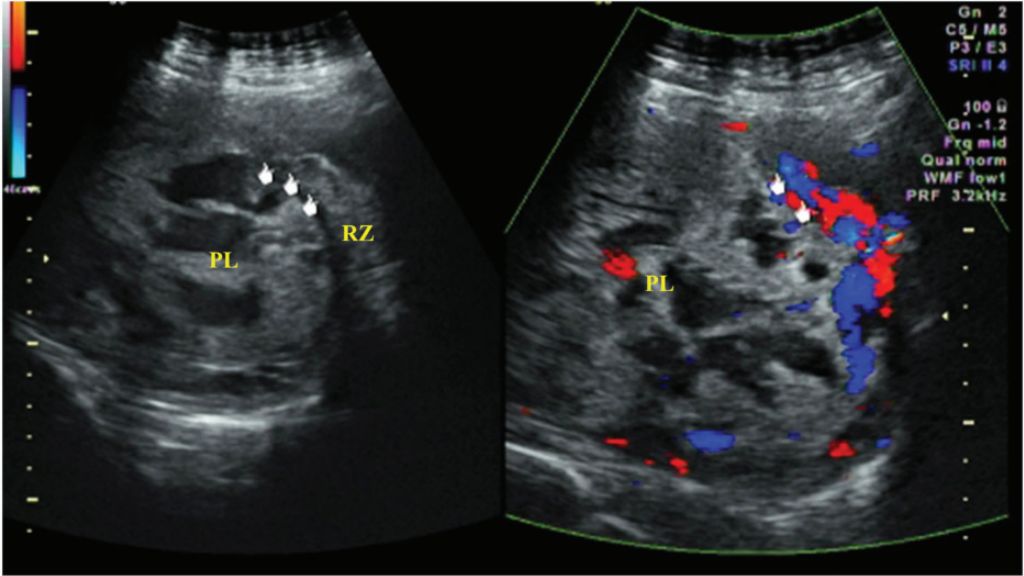


Figure 1. Doppler images. White spots in color Doppler images indicate evidence of placenta percreta (hypervascularity of uteroplacental interface). PL: placenta, RZ: retroplacental zone.

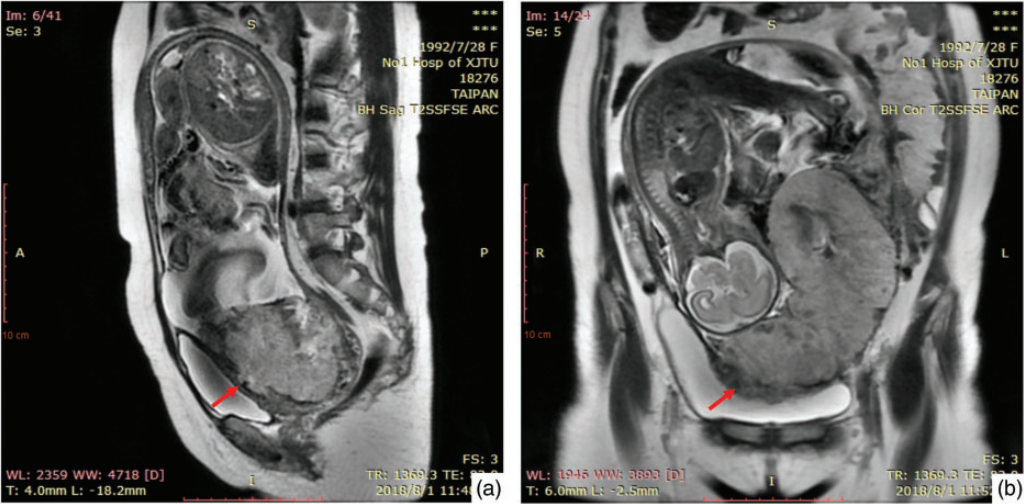


Figure 2. Magnetic resonance images. (a, b) Placenta percreta in the lower uterine segment: the placenta completely covered the lower segment of the anterior and posterior wall of the uterus and was implanted in the uterus cesarean scar of the lower segment (red arrows).

prednisone, hydroxychloroquine, vitamin D calcium, alfacalcidol, and 3-day methylprednisolone shock therapy. Tocolytic agents and hemostatics were also given to

inhibit uterine contractions and vaginal bleeding. Routine blood parameters, coagulation function, and liver and kidney functions were monitored dynamically.

However, the patient's condition became progressively aggravated and laboratory results showed further decreases in Hb (81 g/L) and platelets ($21 \times 10^9/L$). Active SLE-induced secondary fibrinolysis was considered, and recombinant human thrombopoietin, and platelet and plasma infusions were given.

Considering the poor response to conservative treatment, an elective classic midline vertical caesarean section was scheduled after discussion with the patient and with her informed consent. Two days before the scheduled surgery, a double J ureteral catheter was placed to prevent ureteral injury, and infrarenal abdominal aorta balloon occlusion was carried out immediately before surgery to reduce the risk of intraoperative bleeding. However, the patient experienced severe bleeding despite aggressive medical management. A viable male neonate weighing 1110 g was delivered via longitudinal incision of the corpus uteri, with Apgar scores of 7-7-8. The neonate was transferred to the neonatal intensive care unit immediately after birth with a tracheal cannula, which was replaced by non-invasive ventilation 1 day later. The neonate was diagnosed with respiratory distress syndrome and treated with Curosurf and caffeine. There were no signs of intraventricular hemorrhage (irritation symptoms and convulsion) or necrotizing

enterocolitis. Blood products including red blood cell suspension, plasma, albumin, and recombinant human erythropoietin were given for anemia and hypoproteinemia and to improve coagulation, and antibiotics were administered to prevent infection. The neonate was discharged uneventfully 70 days after birth. The placenta was found to have extended from the lower left wall of the myometrium to cover the internal cervical os, penetrating the anterior wall and left side wall of the uterus and the top of the bladder (Figure 3). Emergency internal iliac artery ligation and peripartum hysterectomy were performed. The estimated intraoperative blood loss was 6000 mL, and the patient received blood product transfusions, including 16 U of washed red blood cells, 8 U of suspended red blood cells, 2600 mL of plasma, three doses of platelets, 12 U of cryoprecipitate, and 4 g fibrinogen. The patient was transferred to the intensive care unit directly after surgery for advanced life support.

The patient complained of pain and coldness in her right leg on the first postoperative day, and ultrasonography showed a hypoechoic mass outside the right iliac crest and the common femoral artery, indicating artery thrombosis. Antithrombotic drugs were given immediately but with little effect, and an enlarged artery thrombosis was detected by dynamic ultrasound. On

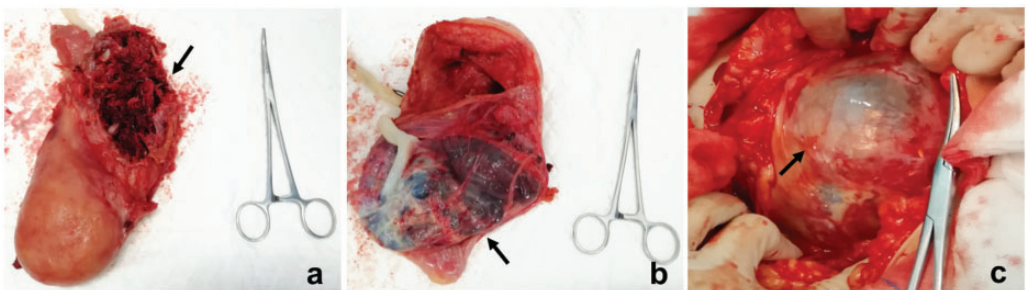


Figure 3. (a, b) The uterus was removed because of uncontrolled bleeding during cesarean section (black arrows indicate placenta percreta). (c) Severe placenta previa with extensive penetrating percreta was found during cesarean section. Thinning or interruption of the uterine interface (black arrow).

the sixth postoperative day, the patient underwent arteriography of the right leg, thrombectomy, and repair of the leg artery (Figure 4a and 4b). A thrombus up to 20 cm in length was removed from the common iliac artery and common femoral artery in the right leg (Figure 4c and 4d). Anti-coagulant treatment including

rivaroxaban, alprostadil, ulinastatin, and heparin sodium was administered after the vascular surgery. The patient's condition gradually improved and stabilized, and she was discharged 5 days after the vascular surgery.

All procedures were performed in accordance with the Declaration of Helsinki and

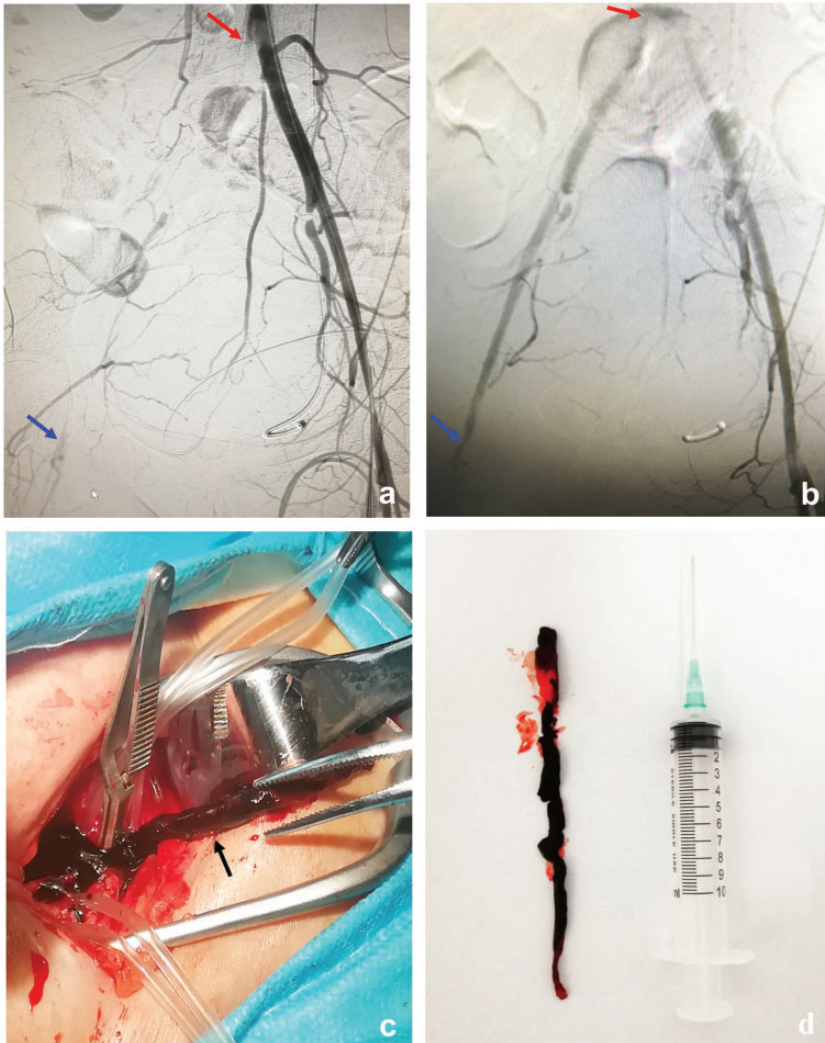


Figure 4. (a) Preoperative and (b) postoperative arteriography of right leg arteries (red arrows indicate opening of right common iliac artery; blue arrows indicate 0.5 cm superior to the branching of the right common femoral artery). (c) Embolectomy by arteriotomy in the right leg arteries. (d) Arterial thrombi removed from the right common iliac artery and right common femoral artery.

were approved by the ethics committee of the First Affiliated Hospital of Xi'an Jiaotong University. The patient was treated solely according to standard treatments. Written informed consent was obtained from the patient for the publication of this case report and any accompanying images.

Discussion

Pernicious placenta previa coexisting with placenta percreta in a patient with active SLE has rarely been reported, but each of these conditions could increase the risk of complicated outcomes.^{1,3-5} A retrospective multi-center study suggested that lupus patients who underwent planned pregnancy with inactive or stable disease achieved better outcomes than those with unplanned pregnancies, demonstrated by a significantly decreased rate of fetal loss, more favorable outcomes for preterm infants, and a reduced occurrence of severe disease flares.⁶⁻⁸ It is therefore important for women with SLE to receive appropriate pre-pregnancy counseling and medication adjustments, as well as strict disease control before pregnancy and intensive surveillance during and after pregnancy.⁷⁻⁹ In contrast, discontinuing anti-rheumatic drugs that are compatible with pregnancy is likely to have adverse outcomes.¹⁰ In the current case, the patient discontinued prednisone during her first trimester without consulting her doctor, which might have contributed to the adverse pregnancy events and maternal outcome.⁴ The birth weight of the neonate was 1110 g, which was compatible with the gestational age of 27⁺⁶ weeks, but the Apgar scores were 7-7-8, which were much lower than average for neonates born at a similar gestational age, indicating unfavorable intrauterine conditions for the fetus.¹¹

Pernicious placenta previa coexisting with placenta percreta is a rare but life-threatening complication of pregnancy,

leading to serious consequences such as severe bleeding and bladder injury during surgery.⁵ Several preoperative preventive techniques have recently been developed to reduce intraoperative bleeding, including double J ureteral catheter, infrarenal abdominal aorta balloon occlusion, prior embolization of the uterine arteries, and balloon occlusion of the hypogastric arteries.¹²⁻¹⁵ The condition is frequently diagnosed only after failed manual removal of the placenta, and a precise preoperative diagnosis is thus crucial to allow appropriate preparation.^{2,3} In the current case, both pelvic ultrasonography and MRI indicated a diagnosis of pernicious placenta previa coexisting with placenta percreta, allowing comprehensive preoperative preparations to be made, including correcting anemia, improving coagulation function, and assessing the feasibility and safety of different preoperative preventive techniques. Finally, both double J ureteral catheter and infrarenal abdominal aorta balloon occlusion were placed before the planned cesarean section, to minimize the risks of bladder injury and severe bleeding. However, despite this aggressive preoperative management, the patient still experienced massive intraoperative bleeding. There are several possible explanations for this profuse bleeding even after aggressive preoperative management. First, the complexity and long duration of the surgery may have increased the risk of profuse bleeding; the total operation time was >6 hours and the operative procedures were complex, including midline vertical cesarean section, manual removal of the placenta, bilateral hypogastric artery ligation, bladder repair, and peripartum hysterectomy. Second, coagulation disorders caused by active SLE and massive blood transfusion could have aggravated the bleeding tendency.¹⁶ Third, the abdominal aorta occlusion did not completely block the uterine supply, to avoid possible ischemic

necrosis of the legs. We set the occlusion limit at 10 minutes, while others have reported continuous aortic occlusion times of 20–45 minutes.^{2,17} Finally, there were other sources of blood supply in addition to the bilateral uterine arteries.¹⁸

Some of the complications, including the maternal thromboembolic events, could have been associated with the pelvic balloon occlusion. Possible reasons include acute limb ischemia, ischemia reperfusion injury, arterial pseudoaneurysms, dissection, and arterial rupture.^{19,20} The early onset of arterial thrombosis was a notable feature in this patient, with the patient complaining of pain and coldness in her right leg just 1 day after surgery, and an arterial thrombosis was detected by ultrasonography. This early occurrence of postoperative thrombosis might have been caused by abnormal coagulation function associated with active SLE, leading to a higher risk of postoperative thrombosis.^{21,22} Furthermore, heparin should be supplemented regularly to prevent anticoagulation during prolonged infrarenal abdominal aorta balloon occlusion, given that interventional procedures such as balloon insertion are normally considered as intravascular foreign bodies, and have been shown to be involved in blood vessel injury.^{2,21,23} However, heparin was not supplemented in the present case because of the severe intraoperative bleeding, and anticoagulation and hemostasis seemed contradictory in this patient.

There were several possible mistakes during the treatment of this patient. First, the patient should have been assessed for venous thromboembolism risk at the beginning of her pregnancy, given that her long history of SLE and pregnancy both marked her as a high-risk individual for venous thromboembolism. However, the patient had no clear history of amenorrhea and experienced irregular vaginal bleeding during the first trimester, and she was

therefore unaware of the pregnancy until fetal movements started in the second trimester. In addition, the patient unfortunately did not receive any antenatal risk assessment for venous thromboembolism, because such risk assessment is not applied in our hospital. Third, the patient should have received venous thromboprophylaxis directly after cessation of the bleeding risk. The most common venous thromboprophylactic agents are heparin and low-molecular-weight heparin, which mainly act by antagonizing coagulation factor Xa. Notably, the best way to monitor the efficacy of heparin or LMWH therapy is by dynamically monitoring changes in activated partial thromboplastin time for heparin, and the activity of coagulation factor Xa for LMWH. These indices can help to monitor the efficacy of venous thromboprophylaxis and indicate when to stop the therapy. However, detection of coagulation factor Xa activity is not widely applied in obstetric centers in Chinese hospitals, but we expect it to be available in the near future.

In conclusion, we report a rare case of a pregnant woman diagnosed with pernicious placenta previa/placenta percreta complicating active SLE. This case highlights the fact that infrarenal abdominal aorta balloon occlusion may increase the risk of postoperative thrombosis in patients with active SLE and coagulation disorders. It is therefore necessary to monitor such patients closely and provide timely anticoagulation treatment, to establish a balance between anticoagulation and hemostasis.

Declaration of conflicting interest


The authors declare that there is no conflict of interest.

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