

Spontaneous hemoperitoneum due to rupture of uterine arteries in the second trimester of pregnancy – a rare clinical case

Hemoperitoneu espontâneo por rotura das artérias uterinas no segundo trimestre de gravidez – um caso clínico raro

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Abstract

Spontaneous hemoperitoneum during pregnancy due to rupture of uterine artery is an extremely rare event, associated with a high perinatal mortality and maternal morbidity. We report a case of spontaneous nontraumatic hemoperitoneum during a 25 weeks pregnancy, in which the clinical suspicion, diagnostic imaging and rapid intervention led to a favourable maternal outcome.

Keywords: Spontaneous rupture of uterine artery; Spontaneous hemoperitoneum in pregnancy; Hemorrhagic shock.

Resumo

Hemoperitoneu espontâneo durante a gravidez devido a rotura da artéria uterina é um evento extremamente raro, associado a elevada mortalidade perinatal e morbilidade materna. Descreve-se um caso de hemoperitoneu espontâneo não traumático às 25 semanas de gravidez, no qual a suspeição clínica, o diagnóstico imagiológico e a rápida intervenção permitiram um desfecho materno favorável.

Palavras-chave: Rotura espontânea da artéria uterina; Hemoperitoneu espontâneo na gravidez; Choque hemorrágico.

INTRODUCTION

Spontaneous hemoperitoneum during pregnancy (intra-peritoneal bleeding in the absence of trauma) secondary to rupture of uterine artery is a rare but potentially lethal event¹, with less than 50 cases reported in the last two decades. We report a case of spontaneous non-traumatic hemoperitoneum at 25 weeks' gestation,

in which the clinical suspicion, diagnostic imaging and rapid intervention led to a favourable maternal outcome.

Informed consent was obtained from the patient

CASE REPORT

A 29-year-old primigravida at 25 weeks of a singleton spontaneous gestation presented to the obstetrics urgency department with dizziness, general malaise, lipothymia and lower quadrant pain starting two hours before admission. She had no previous medical, gynecological or surgical history and her antenatal course had been uneventful. She denied history of trauma,

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fever, vaginal bleeding, rupture of membranes, uterine contractions, nausea or vomiting and referred normal perception of fetal movements.

On physical examination, the patient was conscious, with slight mucocutaneous paleness, blood pressure (BP) was 100/63 mmHg and heart rate (HR) was 110 bpm. She had a nontender, soft and depressible abdomen, which was painful on lower quadrants palpation, without uterine hypertonus. There was no vaginal bleeding. Transabdominal ultrasound revealed a fetus with normal heart rate and movements, normal amniotic fluid volume and anterior placenta without signs of abruption. Transvaginal ultrasound revealed a 42 mm length cervix. It was not observed free peritoneal fluid.

The patient was admitted for observation and intravenous rehydration. Paracetamol was administered for initial pain management. Laboratory tests were requested, including complete blood count, coagulation tests with fibrinogen, hepatic enzymology and renal function.

One hour after admission, the patient referred worsening of abdominal pain, followed by sudden episode of sweating, lipothymia in decubitus and haemodynamic instability (BP 81/43 mmHg, HR 115 bpm). An ultrasound re-evaluation confirmed fetal wellbeing, placenta without signs of abruption, but revealed extra-uterine free fluid, collected in pelvic and abdominal cavity and flanks extending to the inferior liver border. The previously requested blood test showed a hemoglobin level of 8,7 g/dL. Due to suspected non-traumatic hemoperitoneum of unknown etiology, it was requested collaboration from the general surgery department. After risk-benefit assessment, it was decided to perform an urgent imagiologic exam, with immediate discussion with a radiology specialist, to enable a faster intraoperative identification of the bleeding site, instead of a laborious exploration of the abdominopelvic cavity with a gravid uterus.

An urgent abdominopelvic computed tomography angiography was performed, which revealed a large hemoperitoneum, collected mainly in pelvic cavity around the uterus. In the arterial phase, there was accumulation of contrast in the pelvis, on the left side, next to the lower segment of the uterus, suggesting active bleeding from the left uterine artery. Placenta had

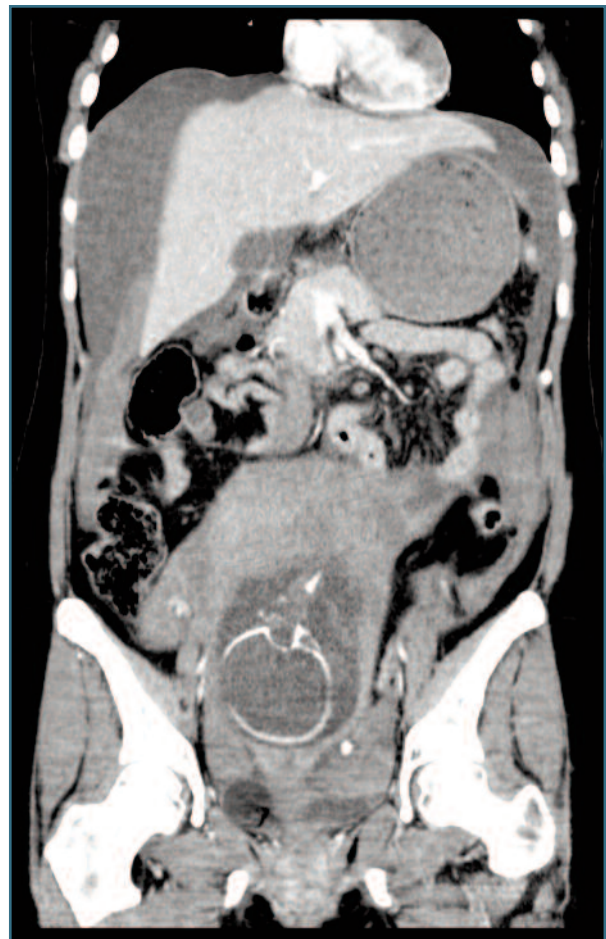


FIGURE 1. Coronal view of Computed tomography (CT) angiography showing extensive hemoperitoneum.

abnormal contrast enhancement suggesting ischemia (Figures 1, 2 and 3).

The gynecological and obstetric team decided an emergency exploratory laparotomy. Under general anesthesia, an infraumbilical midline incision was performed, the suspicion of hemoperitoneum was confirmed and 2600 cc of blood and clots were suctioned from the abdominopelvic cavity. Due to difficulty in accessing the uterine artery, intraoperative maternal hemodynamic instability and unknown fetal status, a lower segment caesarean section was performed. After rupture of membranes with release of clear amniotic fluid, a 910 g female stillborn was delivered. Umbilical cord blood samples were not collected due to maternal haemodynamic instability. The placenta was extracted and the hysterotomy was temporally packed.

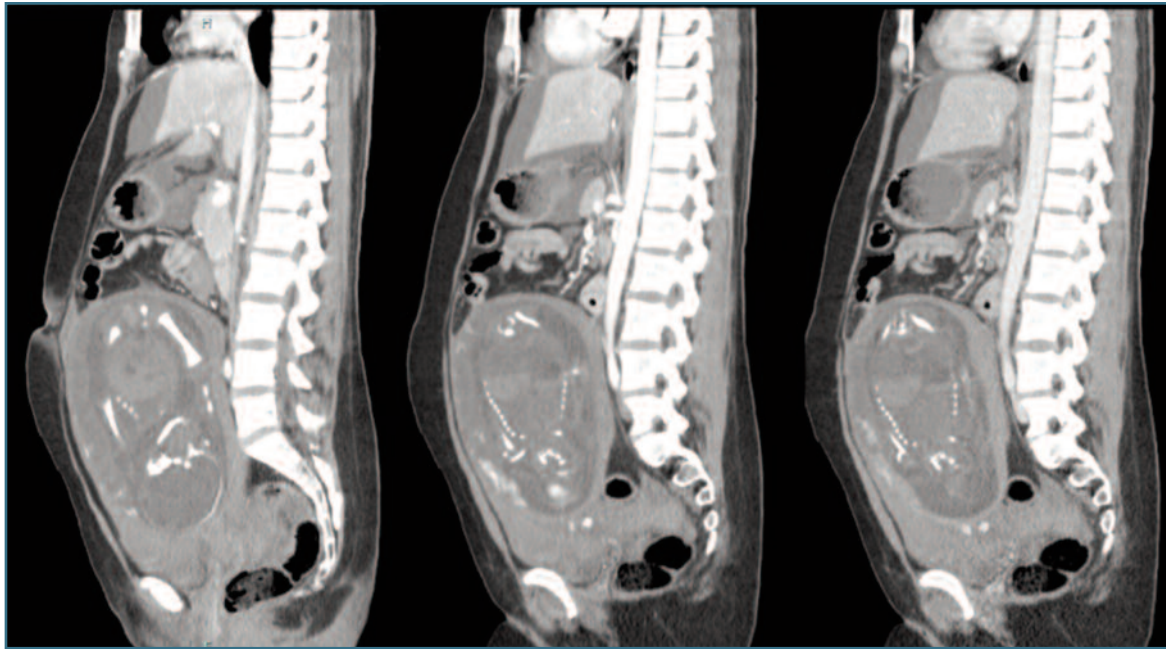


FIGURE 2. Sagittal view of Computed tomography (CT) angiography during the non-contrast, arterial and venous phase, showing haemorrhage from the left uterine artery.

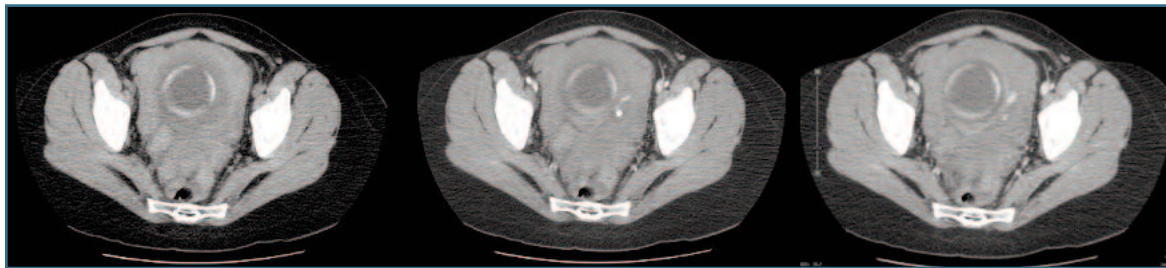


FIGURE 3. Axial view of Computed tomography (CT) angiography during the non-contrast, arterial and venous phase, showing haemorrhage from the left uterine artery.

The left uterine artery was promptly identified. There was an active haemorrhage in its ascending branch, close to the lateral uterine border. The vessel was immediately clamped. An extensive dissection of the broad ligament and parietal peritoneum was also found, with exposure of the iliac vessels and ureters. Large calibre uterine arteries were identified, along with dissection and active bleeding from the contralateral (right) uterine artery, which was also clamped. After laborious haemostasis, bleeding control was accomplished with conservation of the uterus and adnexa.

In the end of the procedure, the estimated blood loss was 3500 cc. Intraoperatively, the patient was resusci-

tated with 4000 cc of crystalloids, 3 units of erythrocyte concentrate, 2 units of fresh frozen plasma, 2 g of fibrinogen and 1 g of tranexamic acid.

The recovery was favourable, with the need of one more unit of erythrocyte concentrate transfusion in the immediate postpartum. Psychological support was offered. On the 5th day after surgery, the patient was asymptomatic, with hemoglobin level of 10,3 g/dL and was discharged from the hospital.

The autopsy showed a female stillborn without malformations, with signs suggestive of anoxia due to acute anemia.

In postpartum consultation, she had no relevant

complaints. An abdominopelvic computed tomography angiography was repeated after 6 months revealing no vascular malformations. Rheumatologic tests showed positive antinuclear antibodies titer, positive rheumatoid factor and negative anti-double-stranded DNA antibody test. Thrombophilia study did not detect antiphospholipid antibodies and the activity of antithrombin, protein C and protein S was normal, and factor V Leiden test was negative. Genetic tests showed a compound heterozygosity for variants of the MTHFR and PAI genes with uncertain association with venous thrombosis. Follow-up was kept in rheumatology and obstetrics specialities. Eight months after the surgery, the patient is clinically well, with regular menstrual cycles and wishing for a future pregnancy.

DISCUSSION

Spontaneous hemoperitoneum during pregnancy (intra-peritoneal bleeding in the absence of trauma) secondary to the rupture of uterine artery is a rare but potentially lethal event, with less than 50 cases reported in the last five decades. The real incidence of this condition is unknown^{1,2}. Most of the cases occurred in the second and third trimester of pregnancy, but there are also reported cases up to three weeks after delivery³.

Spontaneous vascular rupture during pregnancy is described in different vessels, such as hepatic artery (secondary to HELLP syndrome), renal, splenic, uterine or ovarian vessels^{4,5}. The exact etiology of spontaneous rupture of the uterine artery related to pregnancy remains unclear.

Some known causes are congenital malformations, vascular degenerative processes or aneurysms (as it occurs in Marfan syndrome, Ehlers Danlos syndrome or Turner syndrome), adhesions or endometriosis^{1,6,7}. During pregnancy, hemodynamic alterations and estrogen, progesterone and relaxin action may contribute to increased diameter and vascular fragility. Additionally, the compression by the gravid uterus on pelvic vessels and inferior vena cava, increase in the venous pressure in pelvic vessels, which may predispose their rupture. In this case, the underlying cause remained uncertain, as there was no history of trauma or previous surgeries, no signs of endometriosis, vascular

abnormalities or inherited connective tissue disorders. The rheumatologic tests alone did not allow the establishment of a definitive diagnosis.

In most cases, the clinical presentation includes non-specific symptoms, as abdominal pain, general malaise, dizziness, nausea, mucocutaneous paleness and hypotension that progressively worsens and may evolve to hypovolemic shock.

Laboratory findings include reduction of hemoglobin and hematocrit. Ultrasound allows the identification of intra-peritoneal and extra-uterine fluid, the evaluation of fetal viability and may help to exclude uterine rupture or placental detachment^{1,3}.

Abdominopelvic computed tomography angiography allows the diagnosis in most cases. The risks of delayed diagnosis and clinical deterioration outweigh the potential risks of ionizing radiation and contrast use. Magnetic resonance angiography, computed tomography angiography and arteriography use for selective vascular embolization is described in selected hemodynamically stable patients, when available in the institution^{8,9}. In a situation of hemodynamic instability, it may be necessary an immediate exploratory laparotomy without having an imagiologic exam. However, in this case the diagnostic imaging allowed the determination of the hemorrhage source and a fast intra-operative identification of the bleeding vessels.

The preferential approach is diagnostic laparoscopy. However, the presence of hemodynamic instability and hypovolemic shock are considered as contraindications. Additionally, the gestational age and the size of the gravid uterus may difficult this approach³. In most reported cases, laparotomy was described as the primary approach, being the main objective the identification and clamping of the bleeding vessel, while ensuring the patient's hemodynamic stability^{4,6}. Depending on the gestational age, caesarean section is often required in order to locate the source of haemorrhage and to deliver the fetus, which also has high risk of hypoxia and intrauterine death.

The authors reviewed 35 similar published case reports, seven diagnosed during the second trimester, 12 during the third trimester and 16 in the postpartum. During the second trimester, exploratory laparotomy with vessel ligation was the preferred option. In the third trimester, an emergent cesarean section was

performed in most cases due to fetal distress. In the postpartum, vascular embolization was the preferred approach. Poor perinatal outcomes included stillbirth, hypoxic-ischemic encephalopathy and neonatal death. Adverse maternal morbidities included severe anemia requiring transfusion of blood products in most cases. There was one case with need for hysterectomy and maternal death.

A multidisciplinary approach is essential for a favourable maternal outcome. In this case, it was possible to preserve fertility. The risk of recurrence of spontaneous uterine artery rupture is unknown. Although it is well reported the occurrence of spontaneous uneventful pregnancies after bilateral uterine artery ligation, there is no evidence in the literature of case reports regarding subsequent pregnancies after spontaneous rupture of uterine arteries in the second trimester of pregnancy⁶.

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RECEBIDO EM: 01/06/2021

ACEITE PARA PUBLICAÇÃO: 14/02/2022