

# Spontaneous rupture of utero-ovarian vessels in pregnancy

## Ruptura espontânea dos vasos útero-ováricos na gravidez

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### Abstract

Spontaneous rupture of utero-ovarian vessels during pregnancy is a rare but dramatic and life threatening complication associated with a high maternal-fetal mortality. Rapid recognition together with prompt surgical intervention and supportive medical treatment are essential to achieve a favorable outcome for both mother and child. We report a case of a spontaneous rupture of utero-ovarian vessels with intra and extraperitoneal bleeding in a 30 years old primigravida woman.

**Keywords:** spontaneous rupture of utero-ovarian vessels, hemoperitoneum, extraperitoneal bleeding, pregnancy complication.

### INTRODUCTION

Spontaneous rupture of utero-ovarian vessels is a potentially lethal complication of pregnancy. Despite being described since 1778<sup>1</sup>, only 117 cases have been reported until 1987<sup>2</sup>. Since then, another 17 similar cases were found through a Medline search.

There are three types of utero-ovarian vessels rupture: intraperitoneal (from vessels localized on uterine surface), retroperitoneal (bleeding from venous plexus between the two leaves of broad ligament) and the combination of both<sup>3,4</sup>.

Although it has been described as early as at ten weeks of gestation, spontaneous rupture of utero-ovarian vessels is most frequently observed in the third trimester<sup>3,4</sup>. About 50% of the cases are found in primipara and over 60% are related to labour. The maternal mortality rate ranges from 10% to 40% and the perinatal mortality rate is as high as 30%<sup>5</sup>.

Due to its rarity and unspecific signs and symptoms of clinical presentation, delay in diagnosis may lead to tragic consequences.

### CASE REPORT

A 30-year-old woman, primigravida, came to our

emergency department with a mild lower left abdominal pain at 28 weeks of gestation. Besides hypothyroidism which was well controlled by having a daily dose of levothyroxine, she was otherwise healthy. Her antenatal course had been uneventful. There was no history of vaginal bleeding, rupture of membranes, uterine contractions, recent abdominal trauma or intercourse, previous pelvic surgery, or drug abuse.

On examination, she was hemodynamically stable, with a blood pressure of 117/67mmHg and a heart rate of 66 bpm. She had a soft and depressible abdomen with slight tenderness localized in the left iliac fossa. There were no signs of vaginal discharge or bleeding. The cervix was long and closed.

No noteworthy changes were registered in the initial fetal evaluation. Cardiotocogram during about fifty minutes showed a fetal heart rate baseline of 160 bpm with normal variability and absence of decelerations and uterine contractions. At ultrasound scan, a female fetus with movements and muscle tone was observed. Normal amniotic volume fluid and an anterior placenta, far from internal cervical os and without any signs of abruption were also seen.

An episode of sudden sweating and feeling of faintness was registered three hours later, despite clinical improvement of abdominal pain after a course of analgesic medication. Interpreted as a hypoglycemia episode in a pregnant woman who then still showed vital signs within normal ranges and mentioned feeling fetal movements, an intravenous glucose-saline solution was given and a quick recovery of the patient was veri-

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fied. Shortly after, when the previously requested laboratory analysis came out and showed a hemoglobin level of 8.9 g/dl (last analytical evaluation had been performed two months earlier revealing a hemoglobin level of 12.6g/dl), a second abdominal ultrasound exam was done and revealed absent fetal heartbeat and presence of free intraperitoneal fluid. An emergency laparotomy was performed immediately under the suspicion of hemoperitoneum, which was confirmed. After evacuation of a large quantity of blood and clots, the fetus was delivered through anterior hysterotomy in order to allow an adequate assessment of the intraperitoneal cavity. Careful exploration of the pelvic cavity showed a rupture of left utero-ovarian vessels, a tear in the left latero-posterior uterine wall and marked adhesions between the uterus and left ovary (Figure 1). Neither communication between the tear and uterine cavity was found, nor were signs of obvious underlying condition for the adhesences such as endometriosis observed. After opening the left broad ligament and paravesical space, bleeding along the trajectory of the ureter and an extensive extraperitoneal hematoma extending laterally to the sacrum were seen. Assistance of General Surgery was requested. Despite the effort of applying multiple hemostatic sutures in the lacerated uterine wall and sites of ruptured vessels both in the left ovarian ligament and retroperitoneal space along the left ureter during more than two hours, given the maintained difficulty in hemostasis, a "life-saving" subtotal hysterectomy with left salpingo-oophorectomy as well as abdominal packing was performed to ensure stabilization of the patient.

The estimated blood loss was 3 L. Six units of red cell and 4 units of plasma were transfused perioperatively. The patient was transferred to an intensive care unit. After 36 hours, a second laparotomy was con-



FIGURE 1.

ducted for packing removal. Adequate hemostasis was achieved by additional suturing and hemostatic patch placement. The recovery after the second surgical intervention was favourable and the patient was discharged on the 9<sup>th</sup> day after admission.

Histopathology revealed a pale and complete placenta with normal tissue. Marked hemorrhagic infiltration was found in uterus, left ovary and fallopian tube. No signs of vasculitis or other vascular disorders were observed. Autopsy of the stillborn showed a female newborn weighing 1266g with signs suggestive of acute anoxia. No malformation was detected.

## DISCUSSION

Spontaneous rupture of utero-ovarian vessels during pregnancy is such a rare complication that is not mentioned in the standard text books. Most obstetricians would not even see a single case during their practice<sup>6</sup>.

The etiology of spontaneous rupture of utero-ovarian vessels during pregnancy remains poorly understood. However, several etiologic hypothesis have been postulated, basing on the previously reported cases: arteriovenous malformation, uterine artery aneurysm<sup>7</sup>, endometriosis<sup>8</sup>, increase in venous pressure (specially during labour), free anastomosis of uterine and ovarian vessels within broad ligament, and absence of valves of ovarian veins and weakness of vessel<sup>9-11</sup>. On the other hand, macrosomia associated with prolonged labour does not seem to be a predisposing factor for rupture<sup>2</sup>.

Presenting symptoms include acute-onset abdominal pain and maternal hypovolemic shock<sup>12</sup>. In the majority of cases, there is no revealing bleeding and a marked decrease in hemoglobin level is a common finding<sup>13</sup>. The diagnosis can be particularly difficult during labour, because pain can be subjective and hypotension can be attributed erroneously to epidural analgesia<sup>6</sup>.

Placental abruption has been the most common differential diagnosis before laparotomy, according to previously reported cases. Ultrasound can be a useful tool to exclude it, detect free fluid in the abdominal cavity and perform adequate fetal assessment<sup>13,14</sup>. Other differential diagnosis may include uterine rupture, abdominal pregnancy, spontaneous rupture of maternal umbilical vein or aneurysmal vessels, rupture of liver or spleen or their vasculature, appendix rupture and HELLP syndrome<sup>6,13,15-20</sup>.

The diagnosis of the condition is usually made during laparotomy. Nevertheless, in some cases the site of rupture is not found at surgery or autopsy<sup>5</sup>. Preservation of an intact preterm pregnancy is highly desirable, however, sometimes it is necessary to perform cesarean delivery to identify and deal with the source of the bleeding and ensure the best maternal prognosis<sup>15</sup>.

Unlike most of the reported cases in which acute-onset abdominal pain, signs of maternal hypovolemic shock and abnormal fetal heart rate tracing were the most common findings at the presentation, in our case, a mild abdominal pain in a hemodynamically stable pregnant woman with normal findings in her physical examination (besides slight tenderness localized in the left iliac fossa) and in the initial fetal assessment had misled us to assume a muscular origin of her complaints. The gravity of the situation had been realised after encountering a low hemoglobin level, an absent fetal heartbeat in the subsequent assessment and the presence of intraperitoneal fluid in the ultrasound scan. Even then, the patient showed vital signs within normal ranges. The immediate laparotomy confirmed hemoperitoneum, revealing both utero-ovarian and retroperitoneal bleeding. Although hemostatic sutures were applied to the bleeding sites, the extensive intra and extraperitoneal hemorrhage had made hemostasis extremely difficult. Despite all the efforts to preserve fertility in a primigravida, an ultimate decision of subtotal hysterectomy was undertaken and an abdominal packing performed, in the attempt of achieving hemodynamic stabilization and bleeding control. Fortunately, a rapid and favourable recovery was registered after the second surgery for packing removal and further hemostasis control.

Although only speculative, the utero-ovarian adhesions could have contributed to the vascular rupture and uterine laceration through traction on parametrial structures. Nevertheless, the underlying cause remains unknown, as no signs of endometriosis, vascular abnormalities or other obvious causes were observed in the histopathological findings.

This is a case of hemoperitoneum in pregnancy resulted from spontaneously ruptured utero-ovarian vessels. After having encountered such a rare condition with an atypical presentation, we feel the need to consider a more rigorous fetal monitoring in cases of un-specific maternal abdominal pain (even with normal initial fetal assessment) as one of the keys for an earlier recognition of complications and to prevent dramatic maternal and fetal outcomes.

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