

# Endometriosis-related spontaneous hemoperitoneum in pregnancy (SHiP): report of two cases and review of the literature

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## ABSTRACT

Spontaneous hemoperitoneum in pregnancy (SHiP) is a rare but life-threatening complication, for both the mother and the fetus. Its exact incidence is unknown. Several pathophysiological mechanisms have been suggested. However, the etiology of SHiP remains unknown. Endometriosis, whose incidence is increasing, is currently recognized as a major risk factor in its development.

We report the case of a patient with spontaneous rupture of the right uterine venous plexus revealed by a severe abdominal pain during delivery, and another case with spontaneous rupture of the right uterine artery revealed by hypovolemic shock and fetal distress. In both patients, exploratory laparotomy revealed hemoperitoneum and active bleeding. Hemostasis and hemodynamic stability were obtained after right adnexectomy in the first case, and uterine artery suturing in the second. These two patients with a diagnosis of SHiP had both previously been diagnosed with and treated for endometriotic lesions.

## KEYWORDS

Spontaneous hemoperitoneum, pregnancy, endometriosis, severe hypotension.

## Introduction

Spontaneous hemoperitoneum in pregnancy (SHiP) is non-traumatic intraperitoneal bleeding which, in most cases, is revealed by the onset of acute abdominal pain. It has been described before, during and early after labor in 61%, 18% and 21% of cases, respectively <sup>[1]</sup>. Several etiologies have been described in the literature, some of them related to endometriosis <sup>[2-11]</sup>. The risk of SHiP with endometriosis is currently not predictable. However, this diagnosis must be considered in any pregnant woman with a past medical history of endometriosis and/or suffering from acute abdominal pain, and with signs of hypovolemic shock without external bleeding, and with or without fetal distress. SHiP is a life-threatening condition for both mother and fetus. We report two cases of SHiP recently observed in our department.

## Case 1

A 23-year-old woman, G2P1, was admitted to our delivery ward at 39 weeks of pregnancy (WP), in spontaneous labor. Her medical history was significant for a cesarean section (C/S) performed due to an abnormal fetal heart rate, and surgical laparoscopy for right endometrioma.

At full dilatation, she presented severe abdominal pain radiating from the scapula, associated with an episode of hypotension.

Ultrasound did not demonstrate either hemoperitoneum or

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placental abruption. Therefore, since the patient was hemodynamically stable, a vaginal delivery was allowed and a healthy, 3435-gram female baby was delivered. The baby had Apgar scores of 8 and 9 at 1 and 5 minutes respectively. Manual delivery of the placenta with uterine examination confirmed an intact C/S scar.

Immediately after delivery, the patient complained of chest pain and fresh onset of acute abdominal pain. In view of this new hemodynamic instability, it was decided to perform emergency exploratory laparotomy using the previous C/S (Pfannenstiel) scar.

At incision, a 1-liter hemoperitoneum and blood clots were observed. Abdominal inspection confirmed an intact uterine scar. However, active bleeding was noted next to the right adnexa. In order to facilitate access to the bleeding site, the right round ligament was sectioned and the ipsilateral broad ligament was dissected until visualization of the right ureter. This dissection allowed us to identify several tears in the right uterine venous plexus, indicating the need for a right adnexectomy. The total blood loss during this operation was estimated to be 2 liters.

Anatomopathological analysis of the right adnexa was performed and the report indicated the presence of cytogenic endometriosis of the right ovarian cortex and the mesoovarium, combined with an underlying arteriovenous malformation of the mesoovarium.

## Case 2

A 37-year-old woman, G1P0, was admitted to the labour ward at 41 WP. The pregnancy had been spontaneously conceived following surgical treatment for severe endometriosis followed by 3 months of Gn-RH analog therapy. The endometriosis surgery consisted of laser ablation of ovarian endometriosis and complete resection of deep infiltrating endometrium (DIE), including the right uterosacral ligament up to the right posterolateral parametrium. The latter lesion constricted the right ureter causing right hydronephrosis. During induction of labor, severe hypotension was documented, requiring resuscitation with colloids.

An emergency C/S was performed under general anesthesia due to fetal distress on cardiotocography and hypovolemic shock. During surgery, an abundant hemoperitoneum (2 liters) was noted. A 4-kg baby was delivered. The baby recorded Apgar scores of 2, 6 and 8 at 1, 5 and 10 min respectively. The umbilical arterial pH was 6.8. The placenta showed no signs of abruption and exploration of the uterine cavity was unremarkable.

However, the patient presented a profuse active hemorrhage from the right uterine artery in the parametrial region. The posterior wall of the uterine artery was ripped and stretched by a suspected endometriotic adhesion. Ligation of the uterine artery was performed and successful hemostasis was achieved.

Recovery was uneventful and the patient was discharged five days after delivery.

## Discussion

Spontaneous hemoperitoneum in pregnancy (SHiP) is a rare complication that most often occurs during the third trimester<sup>[3, 12-17]</sup>. It consists of intraperitoneal bleeding that manifests itself, in the absence of trauma, during pregnancy and up to 42 days after delivery<sup>[18]</sup>. It has been described before, during and early after labor in 61%, 18% and 21% of cases, respectively<sup>[1]</sup>.

In 89.5% of cases described in the literature, SHiP presented as acute or subacute abdominal pain. In almost all patients, the suspicion of hemoperitoneum arose during assessment following a rapid drop in the hemoglobin level and/or because of signs of hypovolemic shock and/or fetal distress on cardiotocography.

The amount of hemoperitoneum is variable, ranging from 150 mL to 4000 mL (median: 2125 mL).

Fifty-seven clinical cases of SHiP have been reported in the literature between 2000 and 2018 and the findings of this literature review can be summarized as follows.

Forty-two patients underwent an imaging examination, which diagnosed hemoperitoneum in 34 of them (80.9%). Ultrasonography examination revealed free fluids in the perito-

neal cavity in 29 patients, in seven cases confirmed by MRI or CT scan. Five of the 42 patients were diagnosed with hemoperitoneum by CT scan only.

The nature of the fluid could be identified only during the operative management in all the cases, with the exception of one who was hemodynamically stable and underwent conservative treatment but died during the transfer to a hospital by a MIC (maternal intensive care) service<sup>[19]</sup>. Eight of the 57 (14%) patients presented a spontaneous hemoperitoneum in the post-partum, up to Eight days after the delivery, occurring at intervals of 35 to 40 weeks.

Fifty of the 57 patients were treated by laparotomy to manage the haemorrhage, and only 31 of them underwent emergency C/S.

In both the cases encountered in our department, the spontaneous hemoperitoneum was observed during labor. In the first case, a salpingo-oophorectomy was needed in order to achieve hemostasis, while in the second, uterine artery suturing enabled us to achieve correct hemostasis. The hemodynamic status of the second patient prompted us to perform an emergency C/S, and this allowed us to make the unsuspected diagnosis of severe hemoperitoneum.

Both of these patients had a history of surgical laparoscopy for endometriosis, and histological analysis of the right adnexa in the first patient demonstrated the presence of cytogenic endometriosis of the ovarian cortex.

The hypothesis that endometriosis may be an etiological factor of SHiP was first raised in 1992<sup>[2]</sup>. Under this hypothesis, the pathophysiological mechanisms of endometriosis as a risk factor for SHiP would be multiple:

- 1) the chronic inflammation caused by endometriosis would make the blood vessels more prone to rupture;
- 2) the combined presence of pelvic adhesions and an increased uterine volume would put the vessels at increased risk of rupture<sup>[9]</sup>;
- 3) the increased size of endometriosis lesions during pregnancy, due to the phenomenon of first-trimester decidualization, combined with their resistance to progesterone, could also be responsible for the appearance of SHiP<sup>[5]</sup>.

No correlation between the stage of endometriosis and incidence of SHiP has been demonstrated<sup>[5]</sup>.

The vessels responsible for SHiP are mostly venous, but in rare cases, including our second patient, uterine artery ruptures are described. In 90% of cases the bleeding is located on the posterior surface of the uterus or in the parametrial region<sup>[20]</sup>.

Other causes of spontaneous hemoperitoneum during pregnancy, such as uterine rupture, rupture of the liver and spleen and of their vessels, bowel perforation, placenta percreta and ovarian cyst rupture, have to be excluded.

## Conclusion

Physicians should be informed about the risk of obstetric complications associated with a medical history of endometriosis.

In patients with acute abdominal pain, signs of hypovolemia, which may or may not be associated with abnormal

fetal heart rate, should raise the suspicion of hemoperitoneum, especially in women with a history of endometriosis. Appropriate management is required in order to reduce the morbidity of both mother and fetus.

The frequency of spontaneous hemoperitoneum in pregnancy is probably underestimated due to the lack of case reports. To date, the exact incidence of SHiP remains unknown, however several countries have recently decided to establish a common SHiP database for prospective purposes [21]. This initiative would increase our knowledge about the impact and causes of this event. It might also lead to recommendations for primary prevention and optimal management.

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**Conflict of interest:** The authors declare that there is no conflict of interest