Italian Journal of
Gynæcology & Obstetrics
June 2024 - Vol. 36 - N. 2 - Quarterly - ISSN 2385 – 0868

Spontaneous hemoperitoneum in pregnancy due to rupture of uterine vessels in woman with endometriosis: a case report

Antonio **Maiorana**, Giulia **Zaccaria**, Giovanni **Parisi**, Antonella **Mercurio**, Francesco **Forlani** *

Department of Obstetrics and Gynecology, Arnas Civico Hospital, Palermo, Italy.

ARTICLE INFO

History
Received: 02 March 2023
Received in revised form: 29 March 2023
Accepted: 05 May 2023
Available online: 03 June 2024

DOI: 10.36129/jog.2023.124

Key words
Spontaneous rupture of uterine vessels; endometriosis; hemoperitoneum; case report.

***Corresponding author**: Francesco **Forlani**, M.D. Department of Obstetrics and Gynecology, Arnas Civico Hospital, Piazza Leotta Nicola 4, 90127 Palermo, Italy.
Email: forlani81@gmail.com.
ORCID: 0000-0002-0583-8633.

ABSTRACT

Background. Spontaneous rupture of uterine vessels is a rare and life-threating event than can rarely occur during spontaneous and low-risk pregnancies. The definitive association between adverse obstetrical events and pelvic endometriosis is still under evaluation. We report a severe case of spontaneous hemoperitoneum in pregnancy (SHiP) due to a rupture of the uterine vessels related to decidualized endometriosis.
Case presentation. A 38-year-old primigravida woman at 33 weeks of pregnancy with an uncomplicated pregnancy and a history of endometriosis was admitted to the emergency room of our Institution, due to a spontaneous rupture of the right uterine vessels. An alive and vital baby was delivered by hysterotomy. In order to achieve haemostasis, a total hysterectomy with bilateral salpingectomy was performed.
Conclusions. In case of spontaneous rupture of uterine vessels and resulting hemoperitoneum, prompt diagnosis and treatment are the crucial points in order to minimize maternal and foetal/neonatal complications. Further studies are necessary in order to identify endometriosis as a possible cause of spontaneous rupture of uterine vessels in pregnancy.

INTRODUCTION

Spontaneous hemoperitoneum in pregnancy (SHiP) is a rare and dramatic complication correlated with a high rate of maternal and foetal/neonatal mortality. Approximately only 100 cases have been reported in the literature, and since 1950 the maternal mortality rate has been 49.3% [1, 2]. However, nowadays, thanks to medical advances, the mortality rate dropped to 3.6% [3, 4]. Although the aetiology of SHiP remains unclear, haemodynamics and hormonals factors have been involved in the pathogenesis [1]. Endometriosis represents a benign chronic disease characterized by the presence of functional endometrial tissue out-side of the uterus. In this sense, the endometriotic lesions undergoing the process of decidualization generate a subsequent inflammatory microenvironment. For all these reasons, endometriosis has been suggested to be involved in the mechanisms of spontaneous rupture of uterine vessels during pregnancy [5]. Because of the increased number of patients with severe endometriosis with the desire for fertility, physicians would need to consider endometriosis-related SHiP among possible causes of hypotension and

acute abdomen during the third trimester of pregnancy. Here, we describe a case of spontaneous hemoperitoneum in a healthy woman at 33 weeks of her first spontaneous pregnancy.

CASE PRESENTATION

A 38-year-old primigravida woman was admitted to the emergency room of our Institution for abdominal pain at week 33 of pregnancy. There was no history of vaginal bleeding, rupture of membranes, abdominal trauma, previous abdominal surgeries, or drug assumptions. The patient had a medical history of deep infiltrating endometriosis treated with progestin-only drugs with good response. The pregnancy was spontaneous, and the antenatal course was uneventful until the admission. On admission, the patient was hypotensive with a blood pressure of 80/50 mmHg, heart rate was 120 beats per minute, respiratory rate was 14 breaths per minute, and body temperature of 36.3 °C. The physical examination detected a gravid abdomen, tender at the superior quadrants. No dysuria, vomiting or diarrhoea was reported. The ultrasound assessment confirmed the presence of a singleton cephalic foetus with a normal biophysical profile, a regular placenta, and normal amniotic fluid volume. Moderate maternal abdominal free liquid was detected. Cervical length was 25 mm, and tocography revealed no uterine contractions. Her laboratory tests resulted in a haemoglobin level of 9.0 g/dL and 18.20 × $10^3/\mu$L leukocytes. Metabolic hepatic panel and urinalysis were negative. Intramuscular steroid therapy was submitted in order to induce foetal lung maturation.

After 6 hours, the value of haemoglobin level dropped to 7.9 g/dL, and the free abdomen liquid detected at the ultrasound assessment was significantly increased. Moreover, the patient presented an exacerbation of abdominal pain, despite analgesic infusion.

Ten hours after the arrival at the emergency room, considering the worsening of clinical features and the suspicious diagnosis of hemoperitoneum, the patient underwent urgent laparotomy. About 1.5 L of free blood was aspirated from the abdominal cavity. The surgical exploration revealed a haematoma in the posterior and right uterine walls and active bleeding from the right uterine vessels (**Figure 1**). Moreover, there were several endometriotic foci in the pelvic peritoneum and severe pelvic adhesions. There was no sign of uterine anomalies, such as arteriovenous malformation and uterine rupture. The surgery proceeded with a low-segment caesarean section and extraction of a male foetus, alive and vital, weighed 1.400 kg and with APGAR scores of 8 and 9 at the first and fifth minute, respectively. After uterine closure, persistent bleeding appeared from decidualized endometrial lesions on the posterior surface of the uterus and the right parametrium. Because of the difficulty in achieving safe haemostasis without possible damage to the parametrial structures, we decided to proceed with a total hysterectomy. After intraoperative patient's oral consensus, a total hysterectomy with bilateral salpingectomy and ovarian preservation was performed. Successful haemostasis was finally achieved, and the abdominal wall was closed. Estimated blood loss during the hysterectomy was 700 mL and no intraoperative transfusions were performed. No intraoperative and postoperative blood transfusions were performed. No complications were reported, and the patient was discharged after 6 days. The infant had an uneventful course and was discharged after few weeks in good condition. On histopathology examination, uterine, tubal, and right parametrial specimens have reported elements suggestive of endometriotic foci, such as haemorrhagic infarction, fibrosis and prominent deciduoid changes.

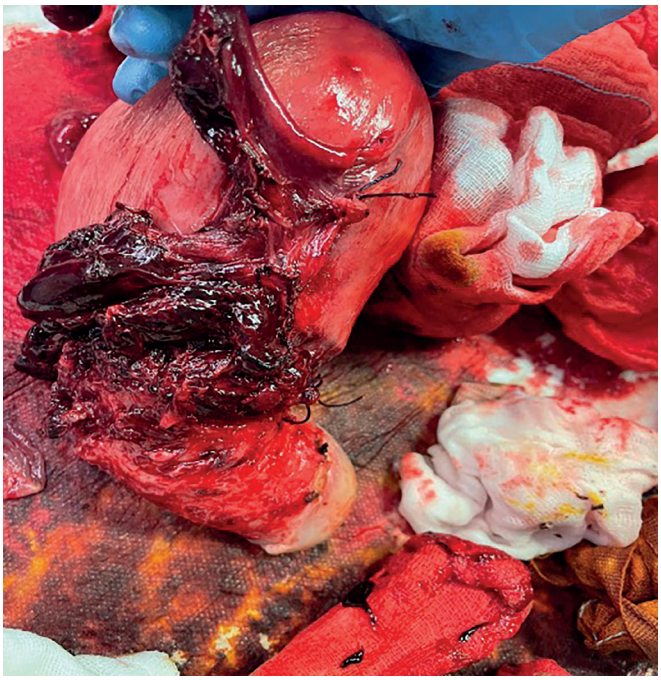

Figure 1. *The right parametrium and the posterior surface of the uterus are covered by blood clots as site of active bleeding.*

DISCUSSION

The current study represents a clear case of spontaneous hemoperitoneum in pregnancy (SHiP) as direct consequence of spontaneous rupture of uterine vessels due to endometriotic lesions in an otherwise uncomplicated pregnancy.

Endometriosis has increased in recent decades and is frequently associated with infertility, pelvic pain, and dysmenorrhoea. Endometriosis is a very complex condition that could impact sexuality, quality of life and psychology of affected woman. Although these aspects could not be correlated to the severity of disease, they have an important role on psychological wellbeing and interpersonal relationships [6, 7]. Endometriosis can be correlated with pregnancy complications, such as severe preeclampsia, placental abruption, placental abnormalities, premature rupture of membranes, preterm birth, and retained placenta [8, 9].

SHiP is a rare and potentially life-threatening condition that occurs in pregnant women out-of-labour in 61% of cases, of which 39% happened between 33-37 weeks of gestation [10, 11]. According to the International Network of Obstetric Survey Systems (INOSS), SHiP is defined as a non-traumatic intraperitoneal haemorrhage during pregnancy up to 42 days postpartum, excluding ectopic pregnancy, uterine rupture and caesarean section-associated bleeding [12].

In literature, trends regarding parity, age, and length of gestation in patients with SHiP have not been documented [1].

In a review of 25 cases of SHiP, endometriosis has been recognized as the major risk factor and the spontaneous rupture of uterine vessels or direct bleeding of endometriotic lesions were the most common findings [1].

Moreover, in a recent systematic review by Lier *et al*., the authors reported that the SHiP was associated with rupturing utero-ovarian vessels in 57% of cases, endometriotic implants in 23% of cases, haemorrhagic nodules in 2% of cases, and a combination of these events in 20% of cases [13].

Furthermore, in almost half of the patients reported in the literature, the diagnosis of endometriosis was misunderstood until the laparotomic visualization of endometriotic lesions and the histological confirmation [10]. Conversely, in our case, the diagnosis of endometriosis was already known at the time of clinical presentation, and the histopathologic examination of the samples confirmed the presence of decidualized ectopic endometrial tissue.

It is well known that the phenomenon of decidualization during the first trimester of pregnancy consists of the loss of pigmentation and fibrosis of endometriotic implants [10]. Recently, it has been supposed that the SHiP is linked to an involution of the decidualization process due to the decrease of progesterone levels and a supposed progesterone resistance. This mechanism causes the production of chemokine, proinflammatory cytokine, metalloproteinases, apoptotic factors, cell death, and bleeding [10].

The incidence of SHiP may be influenced by the use of assisted reproductive techniques (ART), as women with endometriosis could overcome subfertility / infertility problems [14]. The use of ART is linked to a high dosage of progesterone, which can facilitate the process of decidualization. In a recent review of 362 pregnancies reported from 2010 to 2018, Benaglia *et al*. documented that the frequency of SHiP in women with endometriosis submitted *in vitro* fertilization is 0.3% [15]. However, in our case, the patient had a spontaneous pregnancy. Besides endometriosis and ART as risk factors, a recent prospective population-based study reported some additional factors associated with SHiP, such as multiple pregnancies, ≥ 35 years of age in mothers, and previous abdominal surgery [11].

Although the recent evidence, the etiopathogenesis of this condition remains unclear. Increased venous pressure in utero-ovarian circulation due to pregnancy status or muscular activity such as defecation and coughing could be possibly implicated in the pathophysiology of SHiP [16].

In the literature, three factors have been described as explanations for spontaneous rupture of uterine vessels: vessels leakage caused by endometriosis-linked chronic inflammation, adhesions between vessels with relative tensions, decidualization of endometrial foci [5, 17, 18]. Our patient had either a diagnosis of deep infiltrating endometriosis or pelvic adhesions. Indeed, during the surgery, adhesiolysis was performed. Moreover, in a few cases, the origin of the bleeding remains unknown, even during laparotomy. During the surgery of our patient, arteries and superficial veins of the posterior surface of the uterus and right parametria have been involved in the bleeding. The surgical visualization of the endometriotic implants' bleeding and the medical history of our patient suggest that the phenomenon of decidualization of endometriotic foci lead to massive and sudden hemoperitoneum in our patient.

In all cases of spontaneous hemoperitoneum in pregnancy, the onset symptoms were acute or subacute abdominal pain, free abdominal fluid, hypovolemic shock, and decreasing values of haemoglobin [10]. A prompt differential diagnosis is a crucial requirement. Placental abruption, uterine rupture, placenta percreta, appendix, hepatic, and splenic ruptures are the most common preoperative misdiagnosis. Vascular sources of hemoperitoneum in pregnancy should be considered as a result of the rupture of a visceral abdominal artery aneurysm such as splenic artery [1].

Our patient's symptoms were similar to clinical presentation described in the literature. Lier *et al*. reported the most common signs of presentation of SHiP: subacute abdominal pain (94.9%), a decreased level of haemoglobin (62.7%), imaging showing free peritoneal fluid (62.7%) [13]. The sensibility of contrast enhanced computed tomography in the identification of bleeding is documented, but maternal and foetal potential risks from ionizing radiation have to be considered. Ultrasonography could be helpful for the detection and monitoring of abdominal free fluid, but the real diagnosis is often obtained only by laparotomic exploration. In a preterm pregnancy, the decision making should be balanced between risks related to prematurity, delayed diagnosis, and maternal complications. In our case, the worsening symptoms and the haemoglobin drop level guided our decision on laparotomy.

Several questions remain unknown about the management of endometriosis in pregnancy: whether any medical or surgical treatment of endometriosis in the preconception period would add any benefit and prevent pregnancy complications such as SHiP; whether adopting any particular management in pregnancies with a previous diagnosis of endometriosis (in terms of follow-up and mode of delivery); whether to choose any specific flowchart in case of a pregnant woman with acute abdomen and free blood abdominal liquid. Yet, endometriotic lesions should be considered a possible cause of hemoperitoneum during the third trimester of pregnancy [5]. Rapid diagnosis and prompt intervention are essential to correctly manage such complicated cases.

CONCLUSIONS

In conclusion, our case represents a rare case of SHiP related to endometriosis confirmed by histologic examination. Moreover, a prompt diagnosis of SHiP was crucial in managing this unique clinical scenario without either maternal or fetal complications. Exploring the association between the diffusion of endometriosis and the severity of SHiP could be a new challenge.

In consideration of the risk of spontaneous rupture of uterine vessels and SHiP, physicians should be aware that prompt diagnosis and interventions are crucial to minimize maternal and foetal/neonatal morbidity and mortality. More attention to SHiP, with a particular focus on endometriosis as a cause, would help prevent maternal and foetal adverse events.

COMPLIANCE WITH ETHICAL STANDARDS

Authors contribution

A.M., F.F.: Conceptualization. G.Z.: Writing – original draft. A.M., F.F.: Writing – review & editing. A.M., G.Z., G.P., A.M., F.F.: Data curation, validation.

Funding

None.

Study registration

N/A.

Disclosure of interests

The authors declare that they have no conflict of interests.

Ethical approval

N/A.

Informed consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images.

Data sharing

Data are available under reasonable request to the corresponding author.

REFERENCES

1. Hardin N, Delozier A, Alireza Torabi A, Laks S. Spontaneous Rupture of the Uterine Artery in an Otherwise Normal Pregnancy. J Radiol Case Rep. 2017;11(7):7-13. doi: 10.3941/jrcr.v11i1.2946.
2. Hodgkinson CP, Christensen RC. Hemorrhage from ruptured utero-ovarian veins during pregnancy; report of 3 cases and review of the literature. Am J Obstet Gynecol. 1950;59(5):112-7. doi: 10.1016/s0002-9378(16)39178-5.
3. Jang JH, Kyeong KS, Lee S, Hong SH, Ji I, Jeong EH. A case of spontaneous hemoperitoneum by uterine vessel rupture in pregnancy. Obstet Gynecol Sci. 2016;59(6):530-4. doi: 10.5468/ogs.2016.59.6.530.
4. Simonetto C, Garzon S, Laganà AS, Raffaelli R, Cromi A, Uccella S, et al. Maternal sepsis: a comprehensive review from definition to treatment. Ital J Gynaecol Obstet. 2020;32(3):166-81. doi: 10.36129/jog.32.03.03
5. Cozzolino M, Corioni S, Maggio L, Sorbi F, Guaschino S, Fambrini M. Endometriosis-Related Hemoperitoneum in Pregnancy: A Diagnosis to Keep in Mind. Ochsner J. 2015;15(3):262-4.
6. La Rosa VL, De Franciscis P, Barra F, Schiattarella A, Török P, Shah M, et al. Quality of life in women with endometriosis: a narrative overview. Minerva Med. 2020;111(1):68-78. doi: 10.23736/S0026-4806.19.06298-0.
7. La Rosa VL, De Franciscis P, Barra F, Schiattarella A, Tropea A, Tesarik J, et al. Sexuality in women with endometriosis: a critical narrative review. Minerva Med. 2020;111(1):79-89. doi: 10.23736/S0026-4806.19.06299-2.
8. Gruber TM, Ortlieb L, Henrich W, Mechsner S. Deep Infiltrating Endometriosis and Adenomyosis: Implications on Pregnancy and Outcome. J Clin Med. 2021;11(1):157. doi: 10.3390/jcm11010157.
9. Vannuccini S, La Torre F, Gallucci E, Toscano F, Ruotolo A, Capezzuoli T, et al. Previous surgery for endometriosis: a further risk for obstetric complications? Ital J Gynaecol Obstet. 2023;35(Suppl 1):81. doi: 10.36129/jog.2022.S80.
10. Brosens IA, Fusi L, Brosens JJ. Endometriosis is a risk factor for spontaneous hemoperitoneum during pregnancy. Fertil Steril. 2009;92(4):1243-5. doi: 10.1016/j.fertnstert.2009.03.091.
11. Mazzocco MI, Donati S, Maraschini A, Corsi E, Colciago E, Guelfi F, et al. Spontaneous hemoperitoneum in pregnancy: Italian prospective population-based cohort study. Acta Obstet Gynecol Scand. 2022;101(11):1220-6. doi: 10.1111/aogs.14431.
12. Say L, Souza JP, Pattinson RC; WHO working group on Maternal Mortality and Morbidity classifications. Maternal near miss--towards a standard tool for monitoring quality of maternal health care. Best Pract Res Clin Obstet Gynaecol. 2009;23(3):287-96. doi: 10.1016/j.bpobgyn.2009.01.007.
13. Lier MCI, Malik RF, Ket JCF, Lambalk CB, Brosens IA, Mijatovic V. Spontaneous hemoperitoneum in pregnancy (SHiP) and endometriosis - A systematic review of the recent literature. Eur J Obstet Gynecol Reprod Biol. 2017;219:57-65. doi: 10.1016/j.ejogrb.2017.10.012.
14. Gao FM, Liu GL. Four Case Reports of Endometriosis-Related Hemoperitoneum in Pregnancy. Chin Med J (Engl). 2018;131(4):502-4. doi: 10.4103/0366-6999.225048.
15. Benaglia L, Reschini M, La Vecchia I, Candotti G, Somigliana E, Vercellini P. Endometriosis and spontaneous hemoperitoneum in pregnancy: evaluation of the magnitude of the risk in women becoming pregnant via in vitro fertilization. Fertil Steril. 2021;115(4):1023-8. doi: 10.1016/j.fertnstert.2020.10.030.
16. Katorza E, Soriano D, Stockheim D, Mashiach R, Zolti M, Seidman DS, Schiff E, Goldenberg M. Severe intraabdominal bleeding caused by endometriotic lesions during the third trimester of pregnancy. Am J Obstet Gynecol. 2007;197(5):501.e1-4. doi: 10.1016/j.ajog.2007.04.030.
17. Inoue T, Moriwaki T, Niki I. Endometriosis and spontaneous rupture of utero-ovarian vessels during pregnancy. Lancet. 1992;340(8813):240-1. doi: 10.1016/0140-6736(92)90506-x.
18. Passos F, Calhaz-Jorge C, Graça LM. Endometriosis is a possible risk factor for spontaneous hemoperitoneum in the third trimester of pregnancy. Fertil Steril. 2008;89(1):251-2. doi: 10.1016/j.fertnstert.2007.02.009.