CEOG Clinical and Experimental Obstetrics & Gynecology
Case Report

Endometriosis-related spontaneous haemoperitoneum in pregnancy – case report and literature review

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Summary

Spontaneous haemoperitoneum in pregnancy (SHiP) is a rare disease that is associated with adverse pregnancy outcome. The authors present a case of a 35-year-old pregnant woman who developed spontaneous haemoperitoneum at 33 weeks of gestation. An emergency laparotomy was performed, which revealed massive haemoperitoneum with active bleeding from the endometriotic lesions at the right back of uterus. The authors wish to highlight this uncommon but potentially life-threatening condition, which requires early recognition and prompt surgical intervention to reduce morbidity and mortality.

Key words: Hemoperitoneum; Endometriosis; Pregnancy; Spontaneous; Bleeding.

Introduction

Spontaneous haemoperitoneum in pregnancy (SHiP) is a rare but potentially life-threatening complication, which occurs predominantly during the third trimester of pregnancy [1]. SHiP is associated with adverse pregnancy outcomes for both mother and child [2\3]. Endometriosis is a major risk factor causing spontaneous haemoperitoneum [4]. The authors present a case of a 35-year-old pregnant woman who developed spontaneous haemoperitoneum at 33 weeks of gestation. An emergency laparotomy was performed, which revealed the source of bleeding from an endometriotic cysts at posterior surface of right parametrium.

Case Report

A 35-year-old patient (gravida 0, para 0) was admitted at the 33rd week of pregnancy, with complaints of lower abdominal pain attributed to premature contractions lasting longer than three hours. Before admission into the hospital, she was treated with dexamethasone 6 mg in another hospital. The patient had no vaginal bleeding and rupture of membranes. Her medical record showed that at seven weeks of gestation, she had a diameter of 3 cm right ovarian corpus luteum cyst identified by transvaginal sonography. At nine weeks of gestation, the ultrasound confirmed that the cyst had disappeared. No other disease, surgery or drug allergy were shown in the medical records. Vital signs of the patient were stable. The abdomen of the patient was soft with irregular uterine contractions. Other metric included haemoglobin 11.4 g/dl, total white cell count 18.62×109/L, platelets 263×109/L, and NEUT 91.8%. On admission, the patient was treated with tocolytic agents because of irregular uterine contractions. Due to high leucocyte counts, to prevent potential infection, antibiotics were administered. Dexamethasone was also administered for fetal lung maturation. The patient showed signs of recovery. On the third day after admission, the patient suffered with sudden abdominal pain, bloating, and vomiting, without fever. Abdominal examination revealed abdominal distension, left abdominal tenderness (+), and significant pain at the left upper quadrant. Her haemoglobin level decreased to 8.7 g/dl.

Liver, gallbladder, pancreas, spleen, and kidneys were normal based on abdominopelvic ultrasound. Small amount of free fluid was noted in the perisplenic and perihepatic areas. Abdominal CT suggested free fluid of 1.8 cm in hepatic peritoneal and 2.6 cm in spleen perihepatic areas. Fetal heart rate decreased to 80-120 beats /minute. Pulse increased to 122-130 beats/minute. An emergency midline incision for laparotomy was performed because of suspicion of placental abruption. The operation revealed 2,500 mL of free blood and clots in the abdomen. The active bleeding was from a purplish lesion's break, with length of 3-4 cm.The lesion had dimension of 4×4×2cm, on the right posterior surface of the uterus near lower uterine segment. (Figure 1) Abdominal incision was extended to the umbilicus. Liver, spleen, bowels, and their mesentries were examined and found to be intact. After careful confirmation of hemostasis, the abdominal wall was closed. During the surgical operation, the patient received four units of red blood cells and two units of fresh frozen plasma.

The patient recovered well and was discharged on postoperative day six with her baby. A female baby was extracted who weighed 2.40 kg and with an Apgar score seven at the first minute, and nine at the fifth and tenth minutes. Microscopic sections showed haemorrhage, haemosiderin laden macrophages, and some decidua (Figure 2)

Discussion

Approximately 25 cases of SHiP have been described in the literature during the past 20 years [5]. This condition has been described out of labor (61%), during labor (18%), and in the early postpartum period (21%) [4, 6]. The major

Revised manuscript accepted for publication January 9, 2018

Clin. Exp. Obstet. Gynecol. - ISSN: 0390-6663
XLVI, n. 2, 2019
doi: 10.12891/ceog4558.2019

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Published: 10 April 2019

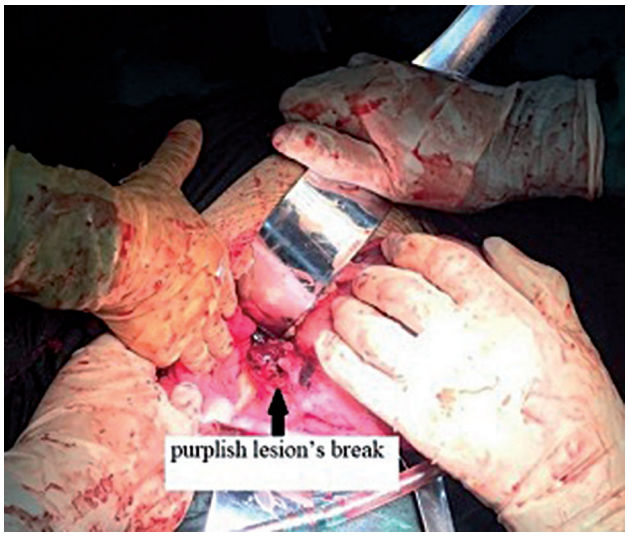

Figure 1. — The right lesion is broken.

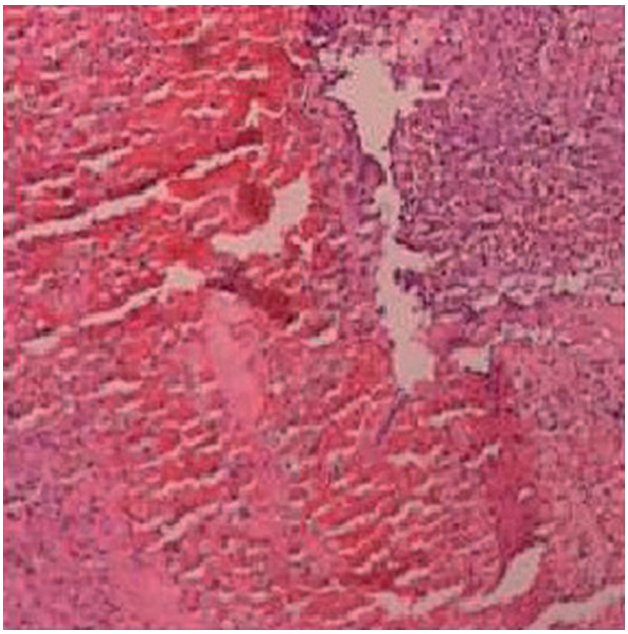

Figure 2. — The broken lesion see microscopically.

symptoms were acute or subacute abdominal pain, followed by hypovolemic shock and fetal distress [4]. Despite this reduction in maternal mortality in the last centuries, perinatal mortality remains high (36%) until recently [4]. The rate of emergency caesarean sections at preterm age still reached 54.5% [7]. Twenty-nine percent of out-of-labor SHiP cases have been reported to occurred before 33 weeks of gestation, 39% of cases occurred between 33 to 37 weeks of gestation, and 32% of cases occurred at term. Endometriosis is recognized as the major risk factor for SHiP [2\8].

Endometriosis is a chronic disease that affects 4–30% of all women of child-bearing age. The prevalence is even higher among women with infertility (up to 50%) [9]. With the development of assisted reproductive techniques, subfertile women with endometriosis are able to become pregnant easily. Symptoms of endometriosis often disappear during pregnancy. Yet patients with severe endometriosis undergoing IVF treatment may still be at increased risk for intra-abdominal bleeding in third trimester of pregnancy. The bleeding can occur from spontaneous rupture of utero-ovarian vessels, or from endometrial lesions [8]. The most common site of bleeding was noted to be posterior surface of uterus or parametria [4].

The exact pathophysiological mechanism is still unclear. Various theories have been proposed ranging from progesterone resistance in eutopic endometrium to decidualisation of ectopic endometrium [2]. The suspicion that endometriosis is a possible risk factor for SHiP was first suggested by Inoue *et al.* [10] in 1992. They explained the involvement of endometriosis in SHiP by two mechanisms: (1) utero-ovarian vessels are more friable due to chronic inflammations associated with endometriosis, and (2) resultant pelvic adhesions due to endometriosis in combination with enlargement of the uterus during pregnancy can place these vessels under greater tension, increasing the risk of rupture and bleeding. Because decidualised endometrium is dependent upon sustained progesterone signalling, failing progesterone levels or devascularisation can cause a weakening of the tissue. In endometriotic tissue, characterised by dysregulation of gene expression leads to progesterone resistance [11]. This can result in increased physiological demands of pregnancy and various muscular activities, such as coughing, defaecation, coitus or straining during the second stage of labour, which all exclusively result in a sudden rise in venous pressure [12].

Preoperative diagnosis of SHiP can be challenging. Placental abruption is the most common prelaparotomy diagnosis in reported cases, just as the present case. In the third trimester of pregnancy, adequate imaging, especially by transvaginal ultrasound, can be difficult due to the position of the uterus and inability to get an overview of the abdominal cavity. Imaging by abdominal ultrasound or MRI proved to be a good diagnostic tool when free peritoneal fluid was detected. Observations of intra-abdominal fluid collections can enhance the suspicion of a haemoperitoneum, although the exact origin and quantity of the bleeding usually remains unknown until surgery [7]. MRI can help characterise the abdominopelvic anatomy better, helping to rule out a surgical cause of the acute abdomen, for example, perforated appendix [13].

It was reported that a case of a 30-year-old pregnant woman who at 29 weeks of gestation developed spontaneous haemoperitoneum. The exploratory laparotomy did not find a clear bleeding point. After surgery at 37 completed weeks of gestation, she had an elective caesarean section [14]. As shown by Brouckaert *et al.* during the ob-

servation process, an excessive haemoperitoneum in an endometriosis patient at 17 weeks of gestation with bleeding at multiple sites, necessitated a hysterectomy with the fetus in situ [15]. However, laparotomy becomes necessary in most of these cases, either after maternal collapse or due to fetal distress. A midline incision for laparotomy can allow adequate access to explore the entire abdomen and pelvis [13]**.**

Conclusion

In pregnant women with the triad of a history of previous endometriosis, severe abdominal pain and a fall in haemoglobin, intra-abdominal haemorrhage must be considered [6]. Even if the patient has no history of endometriosis, this diagnosis still can not be ignored. Weighing the mother's gestational, fetal prognosis, and other factors, laparotomy is conducive to diagnosis and treatment. Active multi-disciplinary collaboration could be crucial in diagnosing and treatment of the disease, and could effectively reduce maternal and child mortality.

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