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Spontaneous hemoperitoneum in pregnancy (SHiP) complicated by endometriosis: A case report

Yukyoung Sim[1*], Jonghyun Kim[1,2], Youngju Jeong[1,2], Chulhee Rheu[1,2] and Heesuk Chae[1,2]

[1]Department of Obstetrics and Gynecology, Chonbuk National University Hospital, Jeonju, South Korea

[2]Research Institutue of Clinical Medicine, Chonbuk National University Hospital, Jeonju, South Korea

Abstract

Background: Spontaneous hemoperitoneum in pregnancy (SHiP) is a rare but life-threatening condition associated with maternal and perinatal mortality. Herein, we report a case of intra-abdominal hemorrhage caused by spontaneous utero-ovarian vessel rupture at 27 gestational weeks following surgery two years earlier for severe endometriosis.

Case: A 31-year-old woman primigravida presented to the hospital at 27 weeks' gestation with acute abdominal pain. She had a history of laparoscopy for severe endometriosis and bilateral ovarian endometriomas. Although the initial ultrasound done was normal, there was a clinical suspicion of hemoperitoneum in pregnancy, which was confirmed by MRI. An emergency laparotomy was performed.

Results: About 2000 mL of blood in the abdominal cavity was evacuated. Cesarean section was performed at the same time due to difficulty in hemostasis and difficulty in securing the field of vision. Extensive adhesions including both adnexa tightly adhered to the posterior surface of the uterus and completely obliterated pouch of Douglas behind the uterus were noted. Active bleeding from the left-posterior uterine-ovarian vessels was found and hemostasis was achieved.

Conclusions: If pregnant women with a history of previous endometriosis treatment present with acute abdominal pain, a fall in hemoglobin, and the absence of vaginal bleeding, intraabdominal hemorrhage should be considered.

Introduction

Spontaneous hemoperitoneum in pregnancy (SHiP) is a rare but almost always life-threatening condition associated with high maternal and perinatal mortality [1,2]. This situation occurs mainly in the second and third trimester of pregnancy [3]. The etiology of this condition is unknown, but recently, it is believed to be related to endometriosis [1,3,4]. We report a case of SHiP complicated by endometriosis.

Case

A 31-year-old woman primigravida presented to the hospital at 27 weeks' gestation with acute abdominal pain. Besides previous pelvic surgery which she had received laparoscopy because of severe endometriosis and bilateral ovarian endometriomas two years previously, she was otherwise healthy. Her antenatal course had been uneventful. The patient's main symptom started during a defecation attempt.

On admission, her blood pressure and heart rate were 120/70 mmHg and 77 beats/min, respectively. Her circulating hemoglobin level was 8.6 g/dL. On examination, she had a soft abdomen with slight tenderness on gentle palpation. There were no signs of vaginal bleeding or vaginal discharge. The cervix was closed. Cardiotocogram demonstrated no uterine contractions and showed fetal heart rate with normal variability. Ultrasound imaging showed normal amniotic fluid volume and a placenta without any signs of abruption.

The following day the patient's clinical condition abruptly worsened when she tried to stool again. Blood pressure was 80/50 mmHg and the hemoglobin level decreased to 6.0 g/dL. Magnetic resonance image (MRI) revealed massive intra-abdominal fluid collection (Figure 1A and 1B).

An emergency laparotomy was performed. Intraoperatively, almost 2,000 mL of blood and clots were noted. However, because of the gravid uterus, it was difficult to find the bleeding site, so caesarean section was decided. A female baby weighing 905 g was extracted. The Apgar scores were 1 and 3 at 1 and 5 minutes, respectively. Both adnexa were tightly adhered to the posterior surface of the uterus and completely

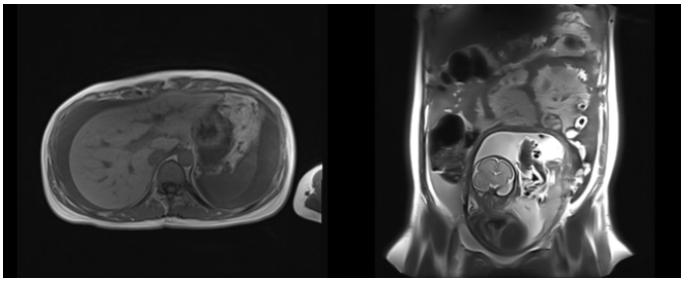

Figure 1(A). Transverse MR image shows perihepatic hemoperitoneum. **(B)** Coronal MR image shows fluid collection in the abdomen

Correspondence to: Heesuk Chae, Department of Obstetrics and Gynecology, Chonbuk National University Hospital, Jeonju, South Korea, Tel: 821062644762; E-mail: jb0429@hanmail.net

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obliterated pouch of Douglas were observed. Active bleeding was found in the fragile veins at the left-posterior uterine-ovarian vessels, which was then sutured for hemostasis. During surgery, the patient received 3 units of red blood cells and 2 units of cryoprecipitate. The patient had an uneventful course and was discharged 6 days after the operation. The infant was transferred to an intensive care unit and was discharged 10 weeks after birth.

Discussion

Spontaneous hemoperitoneum in pregnancy (SHiP) is a relatively rare condition associated with maternal and perinatal mortality [1,5]. Furthermore, as in the first case, hemoperitoneum in pregnancy resulting from spontaneous rupture of uterine-ovarian vessels is extremely rare. During the past 20 years over 25 cases of SHiP have been reported, occurring during the antepartum (61%), labor (18%), and early postpartum periods (21%) [6,7].

The cause of the condition remains poorly understood. However, several etiologic factors have been proposed: increase in venous pressure in the uteroovarian circulation, resulting from the physiologic demands of pregnancy and during muscular activity and straining, such as coughing, defecation, coitus, or during the pushing phase of second stage labor [2]. However, a series of recent reports have been suggested that endometriosis may cause SHiP. Hemoperitoneum in pregnant women with endometriosis can be caused by spontaneous rupture of utero-ovarian vessels or direct bleeding of endometriosis implants. Inoue, *et al.* suggested first that endometriosis might be a possible risk factor for SHiP [4]. Friable uterine vessels resulting from chronic inflammation accompanied by endometriosis or vessel tension resulting from adhesion presumably contributes to vessel rupture [4,8]. Brosens, *et al.* noticed that SHiP was associated with endometriosis in >50% of cases [1]. The pathologic examination of the site of bleeding during pregnancy confirmed endometriosis, characterized by prominent vascularization and decidualization of the lesion [1,9]. We agree with Inoue's hypothesis based on the facts that severe endometriosis was observed at the time of surgery and hemoperitoneum due to spontaneous rupture of the uterine vessels rather than endometriosis lesions occurred. The uterine vessels weakened by endometriosis cause rupture when intraabdominal pressure during physical activity such as defecation.

The diagnosis of SHiP is rarely made before laparotomy due to their extreme rarity and it is frequently misdiagnosed as placental abruption. Other differential diagnoses include uterine rupture, ruptured appendix, abdominal pregnancy, ruptured vasculature of the liver or spleen, and HELLP syndrome [10]. In addition, a correct preoperative diagnosis based on a clinical entity is difficult because SHiP is usually related to non-specific symptoms of acute abdominal pain, vomiting and maternal anemia [3]. This explains a higher rate of maternal and perinatal mortalities of SHiP [11,12].

Conclusion

In conclusion, as shown in our case, the presence of endometriosis may be a risk factor for SHiP. If pregnant women with a history of endometriosis had development of conditions of sudden abdominal pain, a reduction of hemoglobin, and the absence of vaginal bleeding, even if rare, obstetricians should bear in mind the possibility of SHiP.

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Authors disclosure statement

The authors have no conflicts of interest to declare.

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