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Idiopathic spontaneous hemoperitoneum during pregnancy

A.G. Markou[1], A. Puchar[1], J.M. Muray[1], M. Fysekidis[2]

[1] Department of Obstetrics and Gynecology, René DUBOS Hospital, Cergy-Pontoise cedex
[2] Diabetes, Nutrition and Endocrinology Department, Avicenne Hospital, Bobigny (France)

Summary

Spontaneous hemoperitoneum is defined as bleeding within the peritoneal cavity of non-traumatic and non-iatrogenic etiology. It is a rare and life-threatening condition during pregnancy. Spontaneous hemoperitoneum is considered idiopathic when the source of bleeding is not detected during the exploratory laparotomy. The authors report two cases of spontaneous hemoperitoneum during pregnancy with sudden onset of abdominal pain during the third trimester of their pregnancy. Cesarean section was performed for fetal distress. In both cases, hemoperitoneum with a large quantity of blood was found, but the source of bleeding could not be identified during surgical exploration.

Key words: Hemoperitoneum; Idiopathic; Laparotomy pregnancy; Spontaneous.

Introduction

Spontaneous intraperitoneal hemorrhage (SIH) of non-traumatic etiology during pregnancy is a rare complication and is associated with a high perinatal mortality rate [1]. An accurate diagnosis is rarely reached prior to laparotomy due to a multitude of other more frequent acute surgical and obstetrical diseases that present themselves with a similar clinical picture. Ectopic pregnancy after the first trimester is the most frequent cause of spontaneous hemoperitoneum. SIH is considered idiopathic (ISIH) when the source of bleeding is not identified during exploratory laparotomy and it is a rare condition [2]. The decision for fetal extraction depends on the etiology, the age of pregnancy, and the maternal or fetal condition [1]. The authors present two additional cases of idiopathic spontaneous intraperitoneal bleeding.

Case Report

Case 1

A 32-year-old gravida 3 para 1 with a history of left salpingectomy for ectopic pregnancy and a previous cesarean section. Her prenatal course was unremarkable. She was admitted at 28 weeks of amenorrhea for sudden onset abdominal pain.

During the clinical examination the patient's abdomen was painful with a soft uterus. There was neither vaginal bleeding nor uterine contractions and she was hemodynamically stable. Because there was a cardiotocographic fetal bradycardia, ultrasonagraphic evaluation was not realized. An emergency cesarian section was performed for suspicion of placental abruption or uterine rupture. Upon entering the abdominal cavity 1.3 L of blood was collected with aspiration, with blood clots dispersed throughout the abdomen and pelvis. A girl was delivered with a weight of 1,270 grams and had an Apgar score of 5 and 8 at one and five minutes, respectively, (pH = 7.14. lactates =8 .5), and no blood transfusion was necessary. No active bleeding was found, there was no retroplacental hematoma, the uterus was intact, and the inspection of the abdominal and pelvic cavity did not reveal any source of the bleeding. There were no abdominal adhesions in the site of the previous salpingectomy or in the cesarean section. A visceral surgeon was called and performed an abdominal exploration; the site of bleeding was not found and an abdominal and pelvic drain were left in place. The next day after the surgery, an abdominal intravenous contrast computerised angiography tomography (CT) was performed that revealed a small quantity of intra-abdominal fluid and no active site of bleeding. The drain was removed on the second postoperative day. The laboratory analysis of coagulation parameters was normal. The anatomopathological examination of the placenta did not reveal any lesions that could suggest any active bleeding site. The mother was discharged on the fourth postoperative day and the baby was discharged two months later after hospitalization in Neonatal Intensive Care Unit (NICU).

Case 2

A 32-year-old primigravida with a medical history of hypertension treated by ibesartan, before pregnancy, that was switched to nicardipine at the beginning of pregnancy. Three years before the current pregnancy, she had a laparoscopic surgery with bilateral ovarian kystectomy. The presence of abnormal markers for the detection of Down syndrome led to an amniocentesis with a normal 46, XY karyotype.

At 29 weeks amenorrhea, the patient was admitted for abdominal pain associated with nausea and vomiting that had started one day prior to admission. The patient denied having any prior abdominal trauma and there were no contractions or vaginal bleeding. On the physical examination, the pulse rate was 110 bpm and blood pressure was 110/70 mmHg, with moderate abdominal distension, diffuse rigidity, and rebound tenderness.

Cardiotocography revealed persistent fetal tachycardia with variable decelerations and reactivity loss. The ultrasound examination revealed the presence of a live fetus, normal placenta, and

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Table 1. — *Most common causes of abdominal hemorrhage (HELLP: hemolysis, elevated liver enzymes, low platelet count).*

Gynecologic-obstetric causes	Non-obstetric-gynecologic causes
Uterine rupture	Rupture of splenic artery
Ectopic pregnancy	Aneurysm or vein
HELLP-syndrome with liver rupture	Rupture of hepatic artery
Hematoma	Aneurysm or vein
Placenta percreta	Rupture of a maternal
Ectopic deciduosis	umbilical vein
Endometriosis	Coagulopathic hemorrhage
Rupture of hemorrhagic ovarian cyst	Aortic aneurysm
Rupture of a pelvic vessel:	Visceral malignancy
uterine artery or vein	
uterine varicose veins	
utero-ovarian vessel	
Varix of broad ligament.	
Idiopathic	

normal amniotic fluid volume; the existence of intraperitoneal free fluid evoked the possibility of hemoperitoneum. Hemoglobin level decreased to 8.6 g/dL. An emergency cesarean section was performed with the suspicion of uterine rupture.

When the authors incised the parietal peritoneum, a considerable amount of blood and clots was present within the peritoneal cavity, measuring at 1.4 L. The uterus was intact with no signs of rupture. A boy with a weight of 1,485 grams was delivered with an Apgar score of 3 and 8 at one and five minutes, respectively, (pH = 7.20, lactates = 8.26). The placenta was fundal and there were no other abnormalities. There was no active bleeding during surgical pelvic exploration. The ovaries did not have any adhesions in the site of the previous kystectomy. When a gynaecological source of bleeding was not found, the visceral surgeon was called. After performing a detailed abdominal exploration, the origin of the hemorrhage was not detected. The patient was transfused three units of whole blood and three of plasma during the operation and a surgical drain was left in place.

In the first day of the post-surgical period, an intravenous contrast computerised angiography (CT) scan was performed but it did not revealed the site of bleeding. The drain was removed on the second postoperative day with a total drainage of less than 30 ml of sero-sanguin fluid. Complementary laboratory studies did not reveal any coagulation disorders. The mother was discharged on the fifth postoperative day and the baby discharged healthy after a seven-week stay in the NICU.

Discussion

SHI is a rare, but life-threatening condition that is defined as blood within the peritoneal cavity of non-traumatic and not-iatrogenic aetiology [3] and was described for the first time by Barber in a pregnant woman during labor in 1909 [4]. The mortality rate is 8.6% in patients with arterial bleeding of defined origin, but when the bleeding site is not detected, the mortality rate can rise up to 50% [5].

There are various reasons for hemoperitoneum in pregnancy [6]; the most common cause except from ectopic pregnancy and the uterine rupture are summarised in Table 1.

The presence of an undetected source of haemorrhage is very rare and until now, only four similar cases were reported [7-10] and are summarized in Table 2. All these patients were referred for spontaneous hemoperitoneum without any history of traumatic event, without haematological disease, and no source of active bleeding in exploratory laparotomy.

Usually patients are referred to the emergency department with atypical symptoms such as acute abdominal pain, nausea, vomiting, and discomfort with a normal gynaecologic examination. Given the non-specific symptomatology of the abdominal pain during pregnancy, the diagnosis remains unsuspected until the patient develops hypovolemic shock. However the predominant symptom is the sudden onset of abdominal pain and ultrasound scans may be helpful in identifying the presence of free peritoneal fluid during bleeding. The diagnosis of this condition and the identification of the source of bleeding can be made with exploratory surgery. The use of other radiological imaging such as CT may be very helpful in localizing the site of bleeding and establishing diagnosis [3], but this depends

Table 2. — *Six cases of reported idiopathic spontaneous bleeding during pregnancy with maternal mortality.*

Author	Age (years)	Parity	Gyneco-logical history	Current history of pregnancy	Principal symptom	Fetal distress	Term of diagnosis	Term of delivery	Differential diagnosis	Blood volume (ml)	Fetal outcome
Soffer (1975)	Uknown	Uknown	Uknown	Uknown	Uknown	Uknown	Uknown	Aborted	Uknown	Large quantity of blood	
Kofman (2006)	24	0	Absent	Absent	Abdominal pain	Yes	37	37	Placenta abruption	700	Alive
Maya (2012)	30	1	Absent	Cerclage	Abdominal pain	No	29	37	Uterine rupture	3,500	Alive
Shi (2014)	33	1	Absent	Cholestasis	Abdominal pain	No	32	33	Appendicitis	1,500	Alive
Markou (2015)	31	2	Ectopic pregnancy, cesarian	Absent	Abdominal pain	Yes	28	28	Uterine rupture	1,400	Alive
Markou (2015)	32	1	Bilateral ovarian cystectomy	Absent	Abdominal pain	Yes	29	29	Placenta abruption	1,300	Alive

on the presence of good maternal hemodynamics, fetal status, and degree of emergency.

In the present first case, the patient had an acute abdominal pain one day prior to admission and the diagnosis of hemoperitoneum was revealed by ultrasound examination. In most cases, when patients present hypovolemic shock or fetal distress, hemoperitoneum is detected during emergency cesarean section, as was performed in the present second case [3].

Brosens *et al.* [11] noted that in 52% of all cases of spontaneous hemoperitoneum in pregnancy reported in the last 20 years, sub-clinical or clinical endometriosis was present. In the present two cases, neither patient had previous endometriosis or any clinical symptoms for suspecting endometriosis, and even after thorough abdominal exploration during cesarean, no lesions were seen.

Another gynecologic cause of spontaneous intra-abdominal hemorrhage is ectopic deciduosis [12] and rupture of pelvic vessels [1, 12]. This is probably due to a pathologic intrusion of the decidualized stroma into the vessel wall resulting in fragilisation and eventual rupture of the vessel [12]. In the present two cases, neither patient presented macroscopic lesions of deciduosis.

In the present authors' opinion, idiopathic hemorrhage could be stopped as a result in the drop of the blood pressure, an increase in the intra-abdominal pressure, and clot formation, while the clot could eventually dislodge afterwards, leaving no trace of previous bleeding.

Harbour *et al.* reported two cases where the source of bleeding had not been found during macroscopic autopsy but only after careful dissection and microscopic study [13]. Dedouit *et al.* [2] reported another similar case in which at autopsy, the source of bleeding was not macroscopically or microscopically identified.

Certain authors speculate that spontaneous rupture of a visceral artery with no identifiable cause is thought to be related to common vascular disease, such as arteriosclerosis and hypertension [14]. In the present second case, the patient had medical history of chronic hypertension treated by calcium channel blocker. The CT angiogram that was performed the first postoperative day for both patients did not show any focal aneurysm or calcified plaques signs, suggestive of moderate atherosclerotic changes.

Differential diagnosis included uterine rupture and placental abruption. Although the obstetrical examination is frequently normal, most of the time the obstetrician would suspect one of these complications when a similar clinical situation with abdominal pain, hypovolemic shock, and fetal distress, is present. Shi *et al.* in a similar case suspected an acute appendicitis because the patient presented an acute abdominal pain in the right middle abdomen with laboratory results suggesting an infection [10].

The management of spontaneous haemoperitoneum in pregnancy depends on the clinical situation. In the presence of hemodynamic instability, an emergency exploratory laparotomy must be performed in the presence of both an obstetrician and a general surgeon. The choice of an emergency cesarian section is based on the age of pregnancy, etiology of bleeding, and stability of both mother and fetus.

In the present case the authors performed an emergency cesarean section due to fetal distress. Furthermore in cases 3 and 4 (Table 2), the authors reported a conservative management after exploratory laparotomy. The patient was hemodynamically stable with a normal fetal heart rate, they treated the mother with dexamethasone in order to accelerate fetal lung maturation, and a cesarian section was performed. The same approach has already been described by other authors for similar situations [15]; when the pregnancy was complicated by hemoperitoneum in the second trimester and hemostasis had been successful, pregnancies continued normally to term.

Idiopathic spontaneous haemorrhage during pregnancy is a rare event but can be life-threatening, for the mother and the fetus. Given the potentially high perinatal mortality, emergency exploratory laparotomy must be performed by a multidisciplinary team. The present authors reported two new cases of idiopathic spontaneous hemorrhage in the second half of the pregnancy. To their knowledge, only six reported cases of ISIH have been reported and the results were favorable for the mother in all cases and for the fetus in five out of six cases. When hemoperitoneum appears in early pregnancy with a high risk of exposing the fetus to extreme prematurity, in the absence of fetal distress, the pregnancy can be continued if there is no active bleeding and good hemostasis.

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Corresponding Author:
G.A. MARKOU, M.D.
Department of Obstetrics and Gynecology
René DUBOS Hospital
6, avenue de l'Ile-de-France
95303 Cergy-Pontoise cedex (France)
e-mail: georgesmarkou@hotmail.com