

Spontaneous rupture of uterine varices in third trimester pregnancy: an unexpected cause of hemoperitoneum.

A case report and literature review

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Summary

The authors report a case of spontaneous rupture of uterine varices occurring in the third trimester of pregnancy. It was responsible for a collapse in breast hemoperitoneum. The patient, whose pregnancy was a simple, suddenly felt symptoms of preterm labor. There was a rapid onset of maternal collapse with acute fetal distress. Only emergency exploratory laparotomy allowed for etiological diagnosis and treatment.

Key words: Uterine varices; Hemoperitoneum; Intraperitoneal hemorrhage.

Introduction

Intraperitoneal hemorrhage from spontaneous rupture of uterine varices during pregnancy is uncommon [1]. Its diagnosis is not easy because of its rarity, but also due to the lack of specific signs [2]. It should be considered in any unexplained collapse in pregnant women. The clinical symptoms are stereotypes, however it is associated with acute abdominal pain and shock. Other "classic" causes of hemoperitoneum should of course be mentioned [1]. Exploratory laparotomy is immediately needed to reduce fetal mortality and high risk of maternal morbidity [2]. Through this observation, the authors wanted to emphasize the need to perform Doppler of uterine veins during monitoring of pregnant women to detect uterine varicose veins.

Case Report

The patient was primiparous and 33-years-old. She was at 35 weeks of a twin pregnancy when consulted for remarkable pelvic pain in the last 24 hours. At admission she also complained of fatigue and a feeling of nausea. Conjunctivas were stained and hemodynamic was regular (BP: 130 / 80 mmHg, Pulse: 96 pul / min).

The clinical review found her with uterine contractions and the fetal heart rate recording was normal. On examination, the patient did not describe any bleeding, or uterine trauma. She reported only a throbbing pain in the right flank, as well as several episodes of vaginal malaise. The authors then talked about a premature birth and the inevitable removal of the uterine cerclage wire was performed. Minutes after admission there was sudden bradycardia. An emergency cesarean section was then decided associated with in utero-fetal reanimation. Upon admission to the operating room, there was a sudden and unexplained collapse with severe maternal hypotension (70/40 mmHg) and profuse sweating and agitation. Resuscitation was then performed with blood transfusion and macromolecules. Laparotomy revealed an abundant hemoperitoneum (1,100 ml) associated with many clots (Figures 1 and 2). After aspiration, exploration denoted active bleeding from a ruptured varicose package on the right-hand edge of the uterus. Cross-segmental hysterotomy allowed for the extraction of viable twins in apparent good health. Afterbirth and uterine contractions were immediate. No abruptio placentae or uterine rupture were found. After hysterorhaphy, the authors provided hemostasis of the ruptured varicose veins with several "X" stitches and a hemostatic dressing was applied. Blood pressure was good (140/90 mmHg) at the end of the intervention. The patient was admitted to intensive care for 12 hours. The postoperative course was simple and the patient was discharged on the fifth postoperative day. The newborn male showed signs of brain suffering with diffuse seizures during hospitalization. The evolution was rapidly favorable and electroencephalogram monitoring on the fourth day was satisfactory. The newborn female by comparison had a rather favorable course. Psychomotor development of these children is to be assessed.

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Discussion

Frequency

Hemoperitoneum during pregnancy by rupture of uterine varices is an unclear complication because rare and is infrequently described [1]. The present is the only recorded case in decades of the authors' practice. In the literature, very few authors make reference to it and the first cases were described by Hodgkinson *et al.* [3]. It concerned mostly primiparous women (55%), apart from any labor in 60% of cases [1]. Closer to home, only nine cases were reported of which seven heifers in the case described by Pittion *et al.* in 2000 [2]. The predominance of primiparae seems fairly clear such as described in the present case.

Pathophysiology

The pathophysiology of such vascular rupture is still obscure [1]. Several hypotheses have been put forward to try and explain it. According to Hodgkinson *et al.* [3], the

Fig. 1



Figure 1. — Ruptured uterine varices.

Figure 2. — Ruptured uterine varices.



Fig. 2

rupture would occur under the combined effect of hypertension in the uterine veins and their physiological distension in the third trimester of pregnancy, particularly related to their role as a reservoir [2]. It would then be linked to several contributing factors or triggers including postural changes, uterine contractions [3], as well as the posterior insertion of the placenta [2]. In addition, the fineness of vein walls in late pregnancy, unlike those of the arteries, are associated to physiological hypertension and are at risk of rupture [4].

The authors are in agreement with Pittion *et al.* [2] that the rupture of varicose veins is related to a localized vascular ectasia, which is under the combined effect of hypertension and physiological triggers such as abdominal hypertension (uterine overdistension of the twins).

The formation of uterine varices appears to fall within the general framework of venous pathology in pregnancy and in particular of venous insufficiency. Changes in venous hemodynamics in pregnant women are partly due to compression of the inferior vena cava and iliac vessels by the gravid uterus. This results in an increase in pressure and venous distensibility associated with a slowing of blood flow. The result is the delicacy and fragility of the walls through atrophy of the muscular layer [1].

Diagnosis

The differential diagnosis of ruptured uterine varices is for most authors not easy due to the absence of specific signs. However it must be suspected in front of unexplained hypovolemic shock in pregnancy. If the disease is exceptional, the fact remains that many similar cases have been described that highlight the stereotyped nature of the clinical picture. The patient complains of abdominal pain of sudden onset, spontaneous, well-localized in the first instance and then more diffuse. Peristalsis may be present but not evident. There is no reported abdominal trauma, but often a syncopal episode or vagal syndrome [1] as illustrated by this observation. Sudden and unexplained state of shock with no vaginal externalized bleeding is to be expected. The clinical symptoms suggest a uterine rupture in cases of scarred uterus or placental abruption in front of a uterine contraction [2]. Faced with an array of pre-eclampsia, capsular rupture of

the liver can be suspected as well a traumatic rupture of the splenic vein.

In front of maternal collapse and fetal distress, it is necessary to refer briefly to such a diagnosis due to hemoperitoneum and / or shock in order to preserve both maternal and fetal prognoses. Any delay in diagnosis and treatment can be of serious consequence especially in an African limited medical context.

Also among the observations reported to date, only intraoperative findings allowed for the diagnosis of uterine varices, as in the observation described. Often diagnosis is only made intraoperatively, as intervention in this case, during the cesarean section performed for shock and / or acute fetal distress that is generally attributed to a placental abruption or uterine rupture.

In addition to the varices, there are several other diagnoses that coincide with hemoperitoneum during pregnancy aside from traumatic causes:

- the obstetric causes are uterine rupture, placenta increta or percreta, retrouterine hematoma, characterized by acute fetal distress and abdominal pain;
- spontaneous rupture of the liver [5] that occurs in an array of pre-eclampsia and is accompanied by sudden pain shifting to the right upper quadrant and epigastrium;
- spontaneous rupture of the spleen [6] for toxemia, disease (malaria, hemangioma) or post-traumatic.

Treatment

Whatever the diagnostic hypothesis mentioned, exploratory laparotomy is immediately necessary due to maternal collapse and fetal distress and will establish diagnosis and proper treatment. Conservative treatment is most often recommended, but caution must be taken to improve the vital prognosis of the patient. Because of the gravity of the hemorrhagic response, some authors have even resorted to hysterectomy [1].

In the presented case, after cesarean section, the authors ensured hemostasis of the ruptures of varicose veins with many “X” stitches. For other authors, hemostasis was achieved by simple points [2, 7, 8]. In the current case, the conservative approach was largely due to stable hemodynamics during surgery and to the speed with which hemostasis was achieved as experienced by

Pittion *et al.* [2] in 2000. However, cases of hysterectomy were reported in the literature [1]. This shows the seriousness of the unpredictable situation for the prognosis.

Prognosis

Most cases described are far more dramatic than the present, with many examples of in utero fetal death [1]. The maternal mortality rate is high (49.3%) for women not in labor and 76.3% for parturients which have been reported in the literature by Hodgkinson *et al.* [3]. Other authors have found a maternal death in 28 cases registered and nine perinatal deaths due to breast collapse itself [1]. The maternal and fetal prognosis is better in the presented patient because of early surgery and maternal and neonatal reanimation.

The obstetrical future of these patients is indeed not easier to identify compared to Western literature [2] as sub-Saharan Africa context is less medicalized where patients are lost at discharge. However, the authors believe that patients should be provided with the same advice regarding healthy living rather than suffering from venous insufficiency of the lower limbs: avoid prolonged standing, elevate the legs when lying down, resting in lateral recumbency on the left, the fight against obesity, and regular physical activity [1]. Furthermore, the use of color Doppler could help diagnose the presence of uterine varices in these patients with or without a history of uterine varices. This practice is not widespread in the context of Africa, where its use should be more popularized.

Conclusion

Uterine varices in the third trimester of pregnancy are a cause of hemoperitoneum and is rarely mentioned in the literature. It is therefore necessary to keep this complica-

tion in mind because the diagnosis and prompt management determine the maternal and fetal prognosis.

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