Hemoperitoneum due to spontaneous uterine rupture of varicose veins in a twin pregnancy at term: A clinical case study presentation

Mariétou Thiam, Mouhamadou Mansour Niang, Papa Abdoulaye BA, Lamine Gueye, Marie Edouard Faye Diémé, Mamadou Lamine Cissé

ABSTRACT

Introduction: Spontaneous rupture of uterine vessels during pregnancy is an exceptional situation. Case Report: We report a case of massive hemoperitoneum due to spontaneous rupture of uterine varicose veins, occurring in a patient with twin pregnancy at 38 weeks gestation. The patient presented abdominal pain associated with a sudden drop in blood pressure and severe decompensated anemia. Ultrasound scan revealed in-utero fetal demise of the twins with a peritoneal effusion. An emergency laparotomy followed by a cesarean section confirmed the diagnosis, etiology and management with a favorable maternal outcome.

Conclusion: Spontaneous rupture of uterine varicose veins during pregnancy is a rare condition. Clinicians should be aware with this rare condition as delayed diagnosis can be fatal for mother and fetus. The definitive diagnosis of this condition is often made intraoperatively. However, the clinical presentation with ultrasound scanning is very important in planning the management of this condition.

Keywords: Fetal demise, Hemoperitoneum, Rupture of uterine vessels, Twin pregnancy

INTRODUCTION

Described by Hodgkinson and Christensen in 1950, spontaneous rupture of uterine vessels during pregnancy is an exceptional situation [1]. It is responsible for a significantly high amount of maternal morbidity and fetal mortality. Its incidence is thus far not well known with only a few series found in medical literature. The majority of the publications on the subject are isolated clinical cases. To our knowledge, only one case has been
described occurring in a twin pregnancy [2]. We report a case that occurred in a patient with twin pregnancy at term.

**CASE REPORT**

A prime gravida, aged 23, with no particular medical history, was referred to our hospital with severe anemia and a sudden drop in blood pressure with a twin pregnancy estimated at 38 weeks by ultrasound done in early pregnancy. At the arrival, she was in poor general condition, hypotension at 80/50 mmHg, pale mucous membranes, and restlessness. On cardiovascular auscultation, tachycardia at 140 beats per minute was noted. On the obstetric evaluation, uterine fundal height was at 43 cm, the fetal heart sounds were undetectable using the Pinard fetoscope. Vaginal examination revealed a short posterior cervix, admitting a finger, membranes felt and intact with the first twin in a mobile cephalic presentation. Laboratory tests done 9 hours before showed hemoglobin 5.2 g/dl, MCV 85/mm³, MCH 27.6 pg, MCHC 32.6 g/dl, platelets 350x10⁹/mm³, white blood cell count 14.2x10⁹/mm³. Her blood group type was O Rh negative. A state of shock with severe anemia in a prime gravid woman with intra uterine twin fetal demise was diagnosed. She was resuscitated and transfused with 3 units of type O negative blood. Obstetrical ultrasound confirmed the intra uterine demise of both fetuses without placental abruption and the presence of a intra-peritoneal hemorrhage. Furthermore, blood pressure continued to fall despite resuscitation. A CBC check showed hemoglobin rate of: 5.5 g/dl, MCV 77.7/mm³, MCH 27.6 pg, MCHC 33.5 g/dl, platelets 138x10⁹/mm³, white blood cell count 25.50x10⁹/µl, neutrophils 21.76. The decision for a surgical exploration was made. A midline laparotomy sub umbilical was performed leading finding of a great abundance of hemoperitoneum, aspirating 3 L of blood. A transverse lower section cesarean section was done allowing the extraction of two macerated stillborn babies male and female weighing 2800 g and 2400 g, respectively. Placental removal was without any difficulty. The twin pregnancy was dichorionic, diamniotic with posterior fundal insertion of the two placentas. After uterine closure, exploration of the abdominal-pelvic cavity found a large blood clot in the right flank weighing 1 kg and two bleeding varices on the right lateroposterior wall of the uterine body measuring 2 and 3 cm long. The other abdominal-pelvic viscera were intact. Variceal ligation was performed using vicryl 2.0 in layers. Postoperative period was uneventful apart from an anemia 8 g/dl even after transfusion of 3 units of blood postoperatively. In the immediate postpartum period, she received anti-D serum. She was discharged on day-14 postsurgery.

**Figure 1:** Blood clot due to massive hemoperitoneum in the abdominal cavity.

**Figure 2:** Raptured varicose veins on the posterior uterine body.

**Figure 3:** Simple hemostatic sutures using 2.0 vicryl.
DISCUSSION

The spontaneous rupture of uterine vessels during pregnancy is an exceptional and unknown situation. Its frequency has never been evaluated, most publications are clinical cases. Hodgkinson was the first who reported 75 cases in 1950 [1]. This is an obstetric emergency that can endanger maternal and fetal life. With advances in surgical management and resuscitation, maternal mortality has improved from 49.3% in 1950 [1] to 4% in 1985 for Ginsburg et al. [3] and zero deaths in 2011 for Girard [4], while fetal and perinatal mortality remain high at 31% [4].

This is a condition that can occur during pregnancy from 10 weeks amenorrhea with a predilection for the third trimester and in the postpartum up to three weeks after delivery. It occurs most often in the primigravidas women, without any labor as is the case in our patient.

The pathophysiology is poorly elucidated up to now. Several hypotheses have been proposed and are based on physiological changes of the utero-ovarian arteries and veins during pregnancy. Indeed, we observe an increase in the vascular flow during pregnancy up to 10–15 times normal, and even more so in a twin pregnancy. To this is added mechanical phenomenon of compression of the external iliac vessels, and the inferior vena cava vein by the gravid uterus becoming a source of an increase in the pressure and venous distensibility giving a serpentine path appearance. Added to this is a slowing of the blood flow favoring the occurrence of venous aneurysms in the upstream network which weaken the veins that are devoid of valves. This weakness is also related to histological changes in type of finesse by atrophy of the muscular layer favoring the rupture [5] The predisposing factors implicated in the occurrence of ruptures are: coughing, defecation, sexual intercourse, endometriosis, and pelvic adhesions [4, 6].

The diagnosis of the rupture is difficult, most often seen only during surgery, clinical signs are not specific. Abdominal pain and signs of hypovolemia such as hypotension, tachycardia and anemia leading to hemorrhagic shock are classic but not specific and have been described by most authors [6–8]. The state of hypotension and severe anemia of acute onset, without externalized bleeding, caught our attention in our patient. Added to this is the lack of rise in hemoglobin externalized bleeding, caught our attention in our patient. Added to this is the lack of rise in hemoglobin despite repeated transfusions, and peritoneal effusion discovered by pelvic ultrasound which leads to the decision for exploratory laparotomy by which the definitive diagnosis was made. Pelvic ultrasound is the complementary examination of choice according to the authors [4, 6] because of its accessibility in emergency, allowing valuable diagnostic orientation by revealing the peritoneal effusion without specifying its nature. It helped eliminate the possibility of a retro placental hematoma or uterine rupture in our patient. Spontaneous rupture of full viscera such as the liver and spleen are also to evoke especially in cases of associated preeclampsia.

Detrich et al. [6] proposed MRI which allows to specify the hemorrhagic nature of the effusion. However, this is a test that cannot be recommended because of the time required for its completion given the urgent nature of this pathology.

In any case, laparotomy is indicated urgently given the clinical picture of abdominal pain associated with hemoperitoneum in a pregnant patient. The surgical approach can be done by laparoscopy in early pregnancy and in the postpartum period or by laparotomy. In our case, laparotomy helped to make the diagnosis of spontaneous rupture of uterine varicose veins. Hemostasis was obtained quickly after extraction of the fetuses due to uterine retraction reinforced by the intravenous injection of oxytocin. Simple sutures in layers on varicose veins helped complete hemostasis.

The literature review shows that hemostasis can be achieved by several means:
- Compression hemostasis [9]
- Vascular ligatures using simple ties described in the majority of cases [7]
- arterial embolization [10] in patients desiring a subsequent pregnancy and when the hemodynamic status is stable
- and finally hemostasis hysterectomy [11] in extreme cases if bleeding is not controlled

Maternal prognosis was good. However, there was significant morbidity dominated by severe anemia needing repeated transfusions. In addition, due to the delay in diagnosis, fetal prognosis was bad in our patient marked by intrauterine fetal death of both fetuses due to maternal collapse. Rare cases of vaginal delivery of a healthy child after surgical management during pregnancy, have been described [5].

The subsequent obstetric follow-up of these patients is not well described in literature. This is most often an isolated accident, no cases of recurrence have been described. For Pitton et al. [12], a Doppler ultrasound may help localize recurrent uterine varicose veins while Girardi [4] recommends a scheduled cesarean between 38–39 weeks of gestation for the next pregnancy.

CONCLUSION

Spontaneous rupture of uterine varicose veins during pregnancy is a rare condition. Clinicians should be aware with this rare condition as delayed diagnosis can be fatal for mother and fetus. The definitive diagnosis of this condition is often made intraoperatively. However, the clinical presentation with ultrasound scanning is very important in planning the management of this condition.

**********

Author Contributions
Mariétou Thiam – Substantial contributions to conception and design, Acquisition of data, Analysis
and interpretation of data. Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Mouhamadou Mansour Niang – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Papa Abdoulaye BA – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Lamine Gueye – Acquisition of data, Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Marie Edouard Faye Diémé – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Mamadou Lamine Cissé – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor
The corresponding author is the guarantor of submission.

Conflict of Interest
Authors declare no conflict of interest.

Copyright
© 2016 Mariétou Thiam et al. This article is distributed under the terms of Creative Commons Attribution License which permits unrestricted use, distribution and reproduction in any medium provided the original author(s) and original publisher are properly credited. Please see the copyright policy on the journal website for more information.

REFERENCES

1. Hodgkinson PC, Christensen RC. Hemorrhage from ruptured utero-ovarian veins during pregnancy: report of 3 cases and review of the literature. Am J Obstet Gynecol 1950 May;59(5):1112–7.

2. Berisavac M, Sparic R, Pervulov M, et al. Spontaneous intra-abdominal bleeding in twin pregnancy--case report. [Article in Serbian]. Srp Arh Celok Lek 2008 May-Jun;136(5-6):299–301.

3. Ginsburg KA, Valdes C, Schneider G. Spontaneous utero-ovarian vessel rupture during pregnancy: three case reports and a review of the literature. Obstet Gynecol 1987 Mar;69(3 Pt 2):474–6.

4. Girard C, Chatrian A, Veran C, Hoffmann P, Pons JC, Sergent F. Spontaneous rupture of uterine vessels during pregnancy, about three cases. [Article in French]. J Gynecol Obstet Biol Reprod (Paris) 2012 Jun;41(4):374–7.

5. Roger N, Chirat Y, Souhaid A, Rezig K, St. Leger S. haemoperitoneum by Varicose Uterine rupture during Case report and literature review. J Obstet Gynecol 2005;34:495–500.

6. Detriche O, Vaesen S, Carlier C, Dutranoy JC, Givron O, Bosschaert P. Spontaneous rupture of varicose veins in the third trimester of pregnancy: diagnosis achieved by MRI. [Article in French]. J Gynecol Obstet Biol Reprod (Paris) 2012 Jun;41(4):370–3.

7. Nguessan KL, Mian DB, Assis GA, Oussou C, Boni S. Spontaneous rupture of uterine varices in third trimester pregnancy: an unexpected cause of hemoperitoneum. A case report and literature review. Clin Exp Obstet Gynecol 2013;40(1):175–7.

8. Lim PS, Ng SP, Shafiee MN, Kampan N, Jamil MA. Spontaneous rupture of uterine varicose veins: a rare cause for obstetric shock. J Obstet Gynaecol Res 2014 Jun;40(6):1791–4.

9. Nakaya Y, Itoh H, Muramatsu K, et al. A case of spontaneous rupture of a uterine superficial varicose vein in midgestation. J Obstet Gynaecol Res 2011 Aug;37(8):1149–53.

10. Díaz-Murillo R, Tobías-González P, López-Magallón S, Magdaleno-Dans F, Bartha JL. Spontaneous Hemoperitoneum due to Rupture of Uterine Varicose Veins during Labor Successfully Treated by Percutaneous Embolization. Case Rep Obstet Gynecol 2014;2014:580384.

11. Hartmann G, Altermann M. Spontaneous rupture of an uterine vein during pregnancy. A case report (author's transl). [Article in German]. Geburtshilfe Frauenheilkd 1981 Jul;41(7):315–6.

12. Pittion S, Refahi N, Barjot P, Von Theobald P, Dreyfus M. Spontaneous rupture of uterine varices in the third trimester of pregnancy. [Article in French]. J Gynecol Obstet Biol Reprod (Paris) 2000 Dec;29(8):801–2.
