Secondary Postpartum Hemorrhage due to Placental Site Vascular Subinvolution as a Diagnostic Challenge in Women with Preserved Uterus

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Abstract

Background: Postpartum hemorrhage (PPH) is an important reason for maternal morbidity and mortality. It is directly responsible for one sixth of the mother’s death. The primary postpartum hemorrhage, which starts in the first 24 hours of the puerperium is easy to diagnose and treat on time, because the patient is still hospitalized. But the secondary one, which starts 24 hours after delivery and until the end of puerperium is much more dangerous for the patients and the doctors also, because the patient is at her home already and usually comes to the hospital with profuse bleeding and often in shock statement.

Case report: We present a rare case of placental site vascular subinvolution (VSI) in woman after uncomplicated spontaneous delivery where the uterus preservation was mandatory, the procedure to diagnose it, the management and therapy, also a brief review of the literature about the pathological mechanisms of the disease.

Conclusion: Placental site VSI is the rarest cause of secondary postpartum hemorrhage. The purposed mechanisms for developing VSI as an inadequate interaction between endovascular trophoblast cells and maternal spiral arterioles, the absence of deposition of complement and immunoglobulins in the vessels and overexpression of Bcl-2 antiapoptotic protein do not explain the etiology for developing a VSI. The clinicians misdiagnose it very often because

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of the difficulties of diagnosing VSI in cases when uterus preservation is mandatory. Most often VSI is a diagnosis of exclusion and the therapy is in accordance with the recommendation for treatment of postpartum hemorrhage.

**Key words:** secondary postpartum hemorrhage, puerperium, vascular subinvolution, vaginal bleeding #placental site, uterus
INTRODUCTION

PPH is commonly defined as a blood loss of 500 ml or more within 24 hours after birth. It is associated with high mother’s morbidity and mortality rate, and that is why it represents a serious problem for the mothers and obstetricians. According to WHO the incidence of PPH in general is approximate 5-20% of labors. It’s wide geographic variability shows that in Africa accounts 33.9% of maternal deaths and in Asia 30.8%. The incidence in high developed countries like USA, Canada, Belgium, France and Great Britain rises from 1.9% to 2.8%. There are no data available for the PPH incidence in the developing countries (1). It is directly responsible for one sixth of mother’s death worldwide.

PPH may occur in the first 24 hours of delivery and then is considered as primary, or it may occur 24 hours to 6 weeks after delivery and then it is considered as secondary. Reasons for primary PPH are atonic uterus, retained products of conception, traumatic injuries of the genital tract and coagulopathies, while the secondary one results from infections (often associated with retained products of conception), arteriovenous malformations, gestational trophoblastic disease and placental site VSI as in our case.

The incidence of secondary PPH is 2 % (2). Mostly occurs in first or second week after delivery when the patient is already at home. The patients come to office with heavy and profuse bleeding and very often in shock. Secondary PPH is rare condition, but is life threatening and that is why it deserves special attention among the obstetricians.

CASE REPORT

34 years primiparous woman comes at the Obstetrics and Gynecology Department (PHI General Hospital - Gevgelija) with heavy vaginal bleeding. She gives an anamnestic data for spontaneous delivery of a life female baby 18 days ago in the 39 gestational week. The delivery was uncomplicated and passed normal. The bleeding was paroxysmal, heavy and profuse and she soaked 3 diapers in period of half an hour. As a data from family anamnesis she claims that her mother had same kind of bleeding on the 27-th postpartum day, after spontaneous delivery of her twin brothers 30 years ago.

The patient is aware, contactable and upset. The skin and mucosa are normal colored and she is afebrile. She is obese with BMI 35.9. She is normotensive 110/70mmHg. The speculum examination shows heavy, profuse bleeding with large coagulums. The cervix is about 2cm long and about 2-3cm dilated. The uterus is subinvoluted and above the symphysis. The ultrasound examination is made with Voluson E8, GE Healthcare, 2009, USA. The uterus was indifferent position, enlarged, with unclear endometrial lining, which in the fundal area showed an echoic thickening. The Doppler flow was negative. The blood count showed WBC 10x10⁹/l, Hb 101g/l, HCT 32% and PLT 484x10⁹/l. We administered uterotonic (Syntocinon 20IE, Methylergometrin 0.2mg/ml
and Prostin 15 (carboprostium trometamoli 0.25mg), rehydration therapy with 2L saline solution and substitution therapy with Gelofusine 4% 500ml. We made an instrumental revision with Boom’s curette and the material was processed by pathologist. It confirmed that the material is necrotized deciduval tissue, in the most part hyalinized. The coagulation tests were normal (prothrombin time 11.8sec.; activated partial time 29.7sec; thrombin time 13.5sec; D dimer 678ngr/ml). βHCG was negative, estradiol 18pg/ml, progesterone 0.10ng/ml. The control blood count immediate after the revision showed reduction of Hb 80g/l, HCT 26%, and 24 hour later Hb70g/l and HCT23%. The patient continued with uterotonic therapy but refused blood transfusion. She was discharged the next day with scarce bleeding and uterotonic therapy and ferrous supplements. One week later the ultrasound examination showed antevrted involuted uterus, clear endometrial line and small echoic coagulum’s in cervix and vagina. The finding correlated with the anamnestic data of scares bleeding, which stopped 3 days later. The patient had her period one month later and all the control findings were normal.
DISCUSSION
Placental site VSI is the rarest reason for secondary postpartum hemorrhage. This diagnosis is complicated to prove, especially when the uterus preservation is mandatory. So, the patient has to be examined of all aspects and to consider all risk factors. According to a cohort-based study in Sweden, which included 1 114 071 women with singleton pregnancies, from 1997 to 2008 year, with high BMI, the grade of obesity is as high as the grade of risk for postpartum hemorrhage (3). Our patient is obese, with high BMI, so the risk of postpartum hemorrhage is double. There are no studies in the literature that can prove the genetic predisposition for VSI, but the anamnestic data for similar event after her mother’s second pregnancy, leads us to think about the genetic connection among the local hemostatic factors disorder and the VSI occurrence. The proposed mechanisms as inadequate interaction among endovascular trophoblasts and mother’s spiral arteries, the absence of sedimentation of complement and immunoglobulins in the vessels and overexpression of antiapoptotic protein Bcl-2, explain the temporary occlusion of the spiral arteries in the placental site and the resorption of

Figure 2. Hyalinized necrotized decidua
the newformed thrombi for 2-3 weeks (4). The local inflammatory processes as endometritis are well known reason for secondary postpartum hemorrhage, but our case exclude this diagnosis because of absence of clinical findings for it and normal count of white blood cells. Negative Doppler flow proved that it is not a case of vascular aneurism and made a suspicion for retained products of conception (5). But the histologic finding showed hyalinized necrotized placenta and absence of trophoblast tissue, suggestive for VSI (6). It correlated with the negative level of βHCG, which excluded a trophoblast disease. We explain the thick endometrium on ultrasound scan with the physiological high level of estradiol in early puerperium. In the 10th to 20th postpartum day they decrease because the absence of placenta, which is additional source of their synthesis in pregnancy (7). Decrease is noted in our case, so we exclude the estrogen hyperstimulation of endometrium as a cause for secondary postpartum hemorrhage. D dimer and coagulation tests were normal so they confirmed that there is no coagulation disorder as consumptive coagulopathy.

The treatment corresponded with the patient’s current state and according to the recommendation for managing postpartum hemorrhage (2,8,9,10). According to hematocrit was estimated that there is about 20% of blood loss of the blood volume, so it seemed logic to start with substitute therapy with isotonic and colloid solutions to compensate the blood volume. The patient refused blood transfusion. The conservative managing with uterotonics resulted with triple uterotonic therapy and we achieved toned uterus and reduction of the vaginal bleeding. Because of the suspicion of residual masses an instrumental revision was made. Antibiotic therapy was administered to prevent superimposed infection. After the discharge home the patient proceeded with uterotonic therapy and iron supplements and resulted with involution of the uterus, absence of bleeding and correction of anemia.

In cases where the fertility sparing is mandatory, diagnosing the VSI is very complicated. It is usually an excluding diagnosis and it can be misdiagnosed by the clinicians. These cases need to be proceeded with consecutively elimination of all the possible causes of secondary postpartum hemorrhage which is very important because of the treatment decision. Very often the conservative management is sufficient for the desired result, which in the women with unfinished reproduction is preservation of the uterus.

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Conflict of interest
None declared.

REFERENCES