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Case Report

Case study of atonic internal postpartum haemorrhage following severe preeclampsia and caesarean section

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ABSTRACT

Postpartum haemorrhage is most often diagnosed by visualizing external vaginal bleeding, however, in some cases the bleeding may be internal, which significantly increases the risk for delayed diagnosis. We present a case report of a puerperal woman with delayed diagnosis of internal postpartum haemorrhage, which is caused by the development of a Couvelair's uterus after caesarean section, resulting in an acute hysterectomy.

Keywords: Intrauterine postpartum haemorrhage, Couvelair's uterus, Caesarean section, Severe preeclampsia, Delayed diagnosis, Postpartum evaluation

INTRODUCTION

Postpartum haemorrhage (PPH) is reported as the cause of death in 19.9-36.2% of all maternal deaths worldwide, and there is a strong relationship between PPH and preeclampsia (PE), the two most important causes of mortality and morbidity of mothers, however PPH is often still not the first condition to be ruled out during the follow-up of a patient with severe PE.[1,2] The case deals with diagnostic difficulties and other circumstances that have led the medical team to false conclusions, and proves the lack of an accurate postpartum follow up period evaluation and management system in cases where several conditions are combined. Based on the underlying condition, as well as typical signs and symptoms, HELLP syndrome was suspected first.

No additional examination was initially done to rule out any other conditions, thus delaying the true diagnosis, which in this case was severe PPH caused by Couvelaire's uterus.

CASE REPORT

A 39 year old woman with no underlying medical problems passed the antenatal period of her first spontaneous pregnancy without any major complaints. The patient had regular gynecologist's appointments, and received antenatal care according to the requirements of the law. Starting from the first trimester, the patient had arterial hypertension (BP up to 140/90 mmHg). Recalculation of FMF at the thirteenth week of pregnancy showed an increased risk of preeclampsia and intrauterine fetal growth restriction, therefore she was started on prophylactic therapy of aspirin 150 mg starting from week fourteen, and received methyldopa 250 mg once every 2 days starting from week twenty-nine. At 33^{+5} weeks during the uterine artery dopplerometry there was a stage II hemodynamic impairment discovered, due to which the patient was referred to the maternity hospital. She had no subjective complaints, however objectively at the admission unit she had arterial hypertension, blood pressure (BP) 220/126 mmHg, and proteinuria. The patient was stationed in the prenatal care department,

where she received antihypertensive and anticonvulsive therapy and prophylactic therapy for newborn respiratory distress syndrome.

On the next day the patient had developed peripheral edema on both legs, and on therapy BP was 143/87 mm Hg. USG at 33^{+6} gestational weeks showed a fetus corresponding to 29 gestational weeks, and the hemodynamic impairment was still persistent. It was decided to resolve the pregnancy at 34^{+0} weeks to promote the health of the mother and the child.

At 34^{+0} gestational weeks the patient had a caesarean section due to fetal distress and preeclampsia. A healthy baby girl was born (Apgar scale assessment 7/8, weight 1520 grams). The surgery was successful. The postoperative period was spent at the ICU.

The early postoperative period didn't show any abnormalities, the uterus was dense, non-bleeding. A fundus uterus was 1 cm below the umbilicus. However, 6 hours after delivery the patient started complaining about image distortion in her left eye, which worsened over time, and horizontal nystagmus was also observed, which can be indicative of cerebral edema. 8 hours after c-section her hourly diuresis was critically low, 44 ml/h, and due to suspected HELLP syndrome the patient was referred to the tertiary level department.

Ten hours after the delivery the patient (G1 in septimae 34^{+0}, partus I praematurus operativus) was transferred by the emergency medical assistance from the maternity hospital to the Riga East clinical university hospital's emergency unit, where she was evaluated as a high risk patient. Emergency registers were critical (Table 1), fibrinogen 0.4 g/l, BP 170/97 mmHg. At the emergency unit the patient complained about having double vision and blurring in both eyes. A head CT without contrast showed only a development variant of the right vertebral artery V4 segment hypoplasia, but no other pathologies. Suspected cerebral edema and renal failure were ruled out. The gynecologist evaluated her general condition as moderately severe, the uterus was at the level of the umbilicus and lochia sanguineous was in moderate volume.

Eleven hours after delivery the patient was transferred to the intensive care unit (ICU), where she was placed on therapy, T. nimodipini 60 mg p/o x6, S. dexamethasoni 8 mg i/v, S. pantoprazoli 40 mg i/v, torasemidi 10 mg i/v, cryoprecipitate 10 masses i/v, erythrocyte 2 masses i/v, S. $MgSO_4$ 25% 10 ml (12 ml/h), S. labetaloli 200 mg (5 ml/h), after which the BP lowered to 135/82 mmHg (Figure 1). Despite this therapy, the patient's overall condition continued to worsen. When the urine catheter was inserted, diuresis was still decreased (<50 ml). 14 hours after delivery the uterus was at the level of umbo, therefore T. misoprostol 400 µg was given.

Table 1: Laboratory values dynamics.

Laboratory values	Date; December 2019						
	11	**13**	**13**	**13**	**14**	**18**	**22**
Er. (millions)	4.2	<3.22	<2.75	<2.59	<2.02	<3.19	<3.63
Hb (g/dl)	13.9	<10.50	<9	<8.4	<7.9 → 6.5	<9.9	<11
Leu. ($x10^9$/l)	9.5	>13.19	>19.43	>19.30	>18.01	9.52	>10.11
Thr. ($x10^9$/l)	206	<127	<100	<94	<42	199	392
Bilirubin (common, µmol/l)	8.74	6.33	13.37	-	-	-	-
Bilirubin (direct, µmol/l)	-	-	-	>6.29	>5,05	>4.30	3.2
ALAT (U/l)	>49	>34	>35	-	>36	>59.2	>56
ASAT (U/l)	>83	>43	>55	-	<61	-	-
Urea (mmol/l)	3.81	5.75	-	-	>10.85	-	-
Creatinine (blood sera, µmol/l)	72.7	>85.5	>144.97	-	>213.54	>110.57	
CRP, mg/L	-	7.47	-	-	>26.77	>37.07	>8.20
Common protein (g/l)	-	<47	-	-	-	-	-
Protein in urine (g/l)	>2.64	>0.97	>1.50	-	-	-	-
Prothrombine (%)	-	86.4	-	<57.30	-	130	130
INR	-	1	-	>1.49	-	<0.91	<0.84
APTL (sec)	-	>41.5	-	>45.60	-	31.3	30.20
Fibrinogen (g/l)	-	<1.52	-	<0.99	<1.61	>5.63	>4.94
D-dimers (µg/ml)	-	-	-	-	>9.86	-	>2.72

21 hours after delivery the patient complained about fatigue, during the examination the uterus was located above the umbilicus.

24 hours after delivery a massage of the uterus was performed which resulted in discharge of 300 ml of blood clots. The patient was immediately put on sol. oxytocini 20 mg with perfusor 12 ml/h and sol. tranexamacidi 1000

mg i/v. S. calcium 8.94 mg/ml was also prescribed for the patient.

15 minutes later, while examining the patient, it was established that the uterus was crammed, dense and had rapidly enlarged to almost reaching the costal arch, 7 cm above the umbo (Figure 2). During uterine massage large amounts of fresh blood were discharged from the vagina.

An ultrasound was performed, which showed a fairly large cervix, an enlarged uterus with hematometra, a bilateral second stage urostasis and thin layers of fluid around the kidneys, as well as free fluid intraperitoneally around the spleen and in the lesser pelvis. Laboratoric values were significantly low; Hb 6.5 g/dl, thr. 42000, leu. 18.01, Er. 2.0 and based on coagulation markers the patient had a significant coagulation disorder. These results were an indication for a hysterectomy. Chest X-ray showed signs of blockage in the small circulatory system. She received a blood transfusion and 2 doses of S. prothrombinum multiplex humanum. Additional 13 CP and 2 TM were ordered, and the patient was prepared for surgery.

25 hours after delivery the beginning of the surgery: laparotomy, total hysterectomy and adnexibus dxtr. due to increasing haematoma lig. suspensorium ovarii dextra. During the surgery 2 masses of thromobocytes were transfused. The blood loss during the operation was approximately 100 ml; the uterus contained approximately 2 l of clotted blood (Figure 3). Urine was light yellow; approximately 350 ml. Final clinical diagnosis was established Couvelaire's uterus, severe preeclampsia, coagulation disorder and uterine bleeding.

The patient was in the ICU for 4 days, where she received the following therapy; sol. enoxaparini 0.6 s/c, tab. amlodipini 10 mg 2x p/o, tab. enalaprili 10 mg 1x p/o, sol. ceftriaxone 2 g + sol. NaCl 0-9% 250 ml i/v, tab. paracetamoli 500 mg 3x p/o, sol. dexketoprofenum 2.0 i/m, tab. acidum folicum, ferrosi fumaras 1 tab. 2x p/o, microlax per rectum, tab. erythromycini 250 mg 2x p/o, tab. tramadoli 1x p/o.

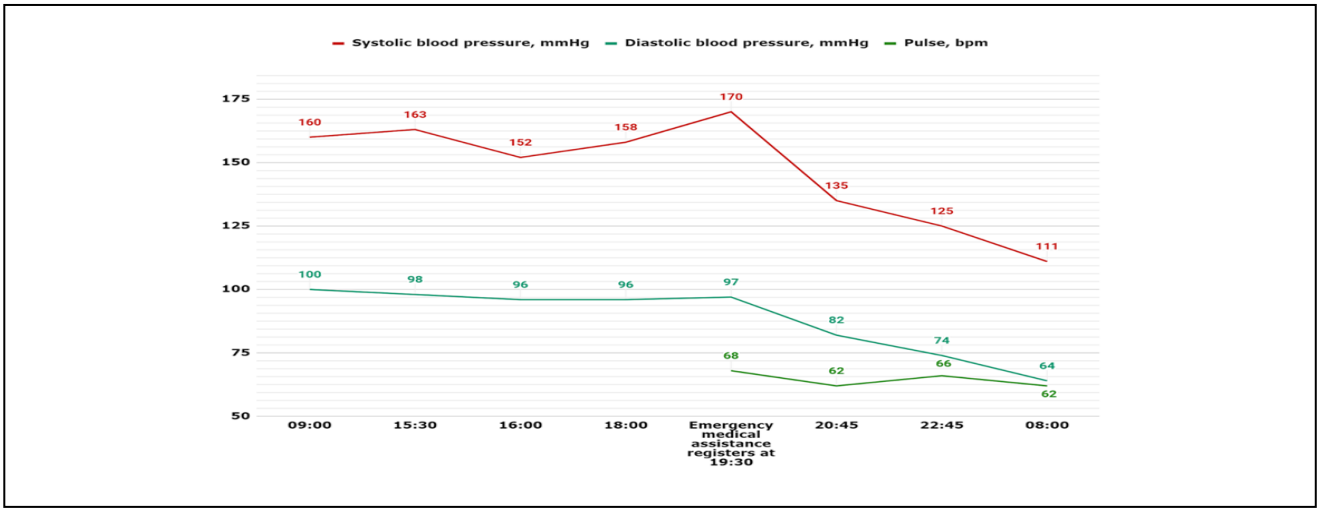

Figure 1: The dynamics of BP and pulse from the 13 December 2019; 9:00 (before c-section) until the 14 December 2019; 8:00 (before acute hysterectomy).

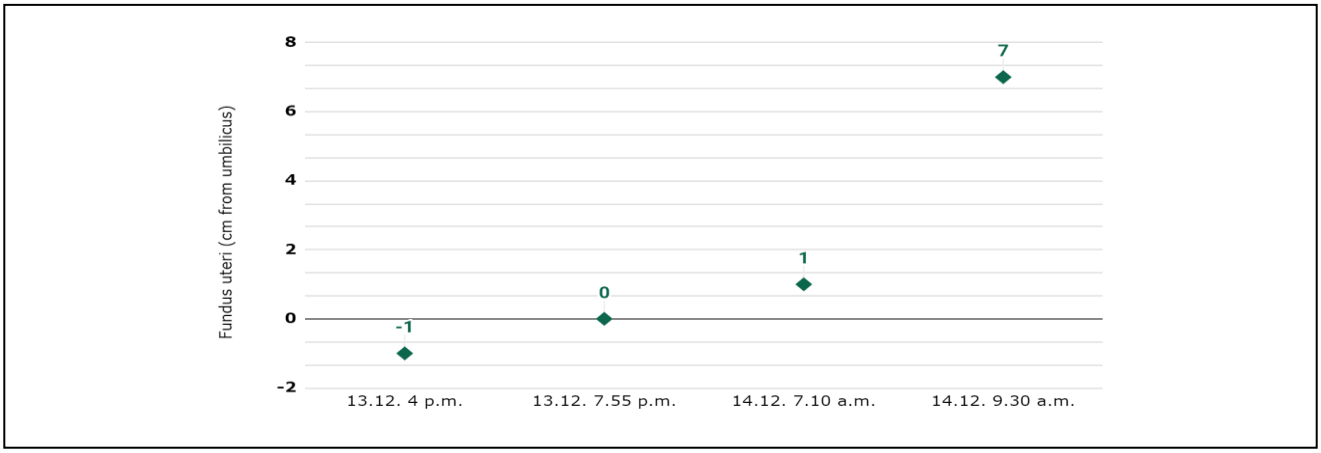

Figure 2: The position of fundus uteri in dynamics.

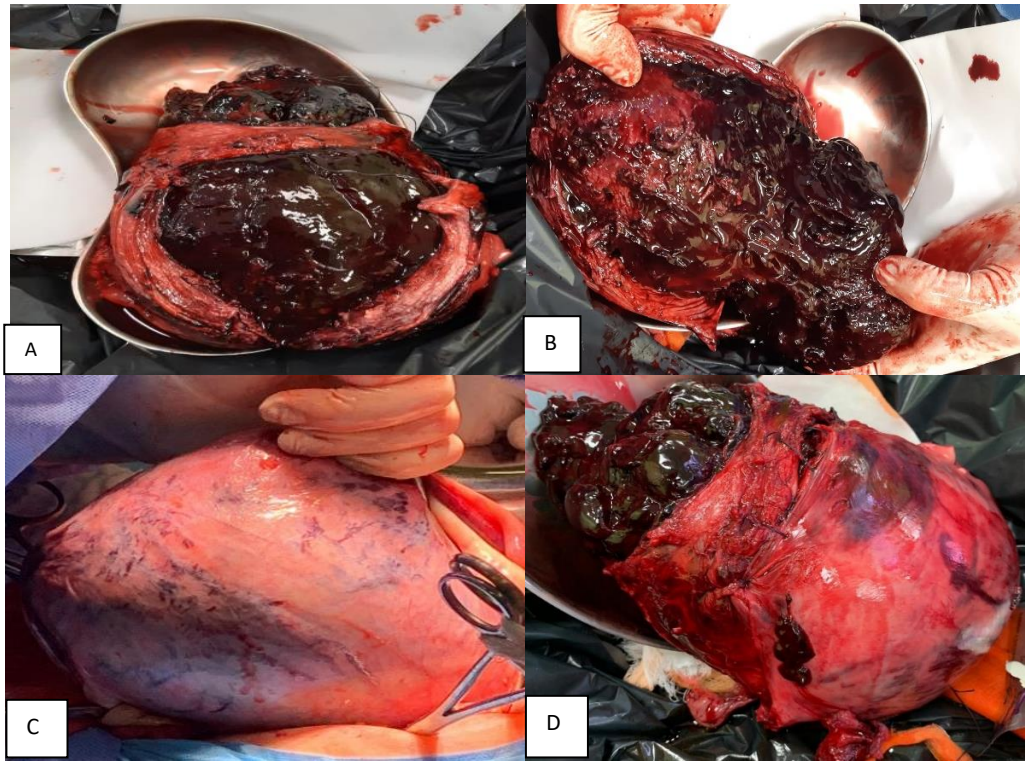

Figure 3 (A-D): A specimen of hysterectomy showing an enlarged uterus, corresponding to 30 gestational weeks, extremely cyanotic with haemorrhaging on the wall of the uterus and cavity full of blood clots.

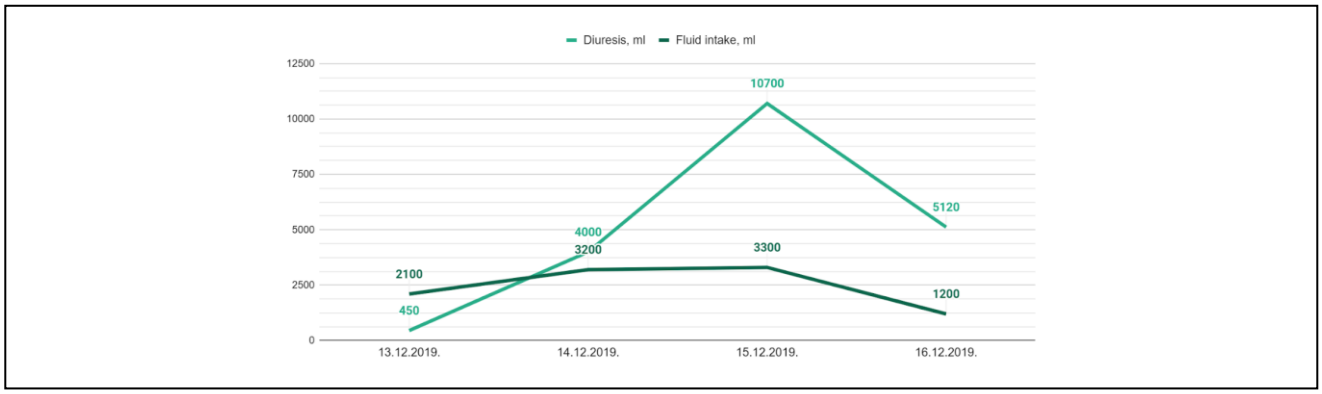

Figure 4: The dynamics of fluid intake and diuresis.

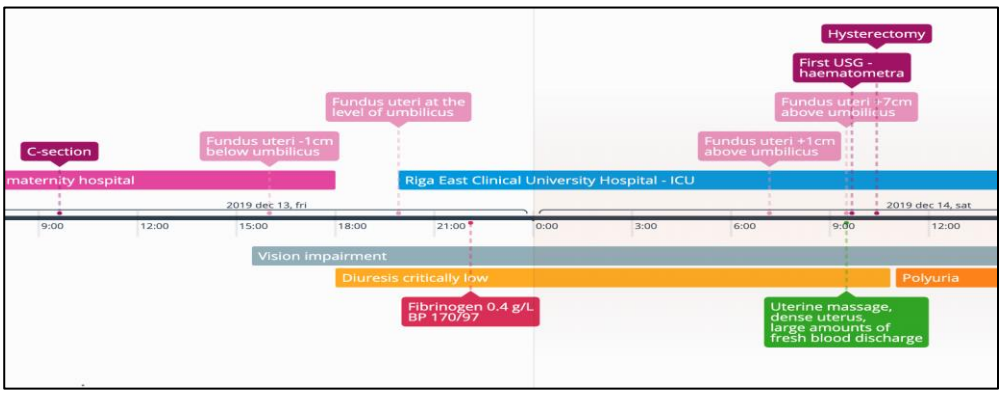

Figure 5: Timeline of the case.

After surgery the patient developed an increasing polyuria (Figure 4) and her vision was still impaired. She received 1 more EM transfusion, and over the next couple of days the polyuria subsided, she regained her vision and the peripheral edema decreased. During her time in the ICU her BP remained increased, approximately 140/70 mmHg.

4 days later the patient was transferred to the gynaecology department with positive dynamics of her general condition. The patient was discharged from the hospital 5 days later in a stable condition, the timeline showing the course of the patient's condition is shown in (Figure 5).

DISCUSSION

Postpartum hemorrhage is defined as >500 ml blood loss after vaginal birth and >1000 ml blood loss after caesarean section. Causes of postpartum hemorrhage can be obstetric lacerations, uterine atony, coagulation dysfunction and retained placental tissue.[3] PPH has been reported in approximately 5% of all deliveries.[4]

Preeclampsia is a progressive multisystemic syndrome, featuring gestational hypertension with proteinuria, renal insufficiency, thrombocytopenia, evidence of liver damage (e.g., elevated liver enzymes, epigastric pain), pulmonary edema, and/or cerebral edema (headache, visual blurring, vomiting, an altered mental state).[5] It occurs in approximately 4-6% of all pregnancies.[6] In 10 to 20% of severe preeclampsia cases, it progresses to HELLP syndrome (hemolysis elevated liver enzymes, and low platelet count).[7]

Couvelaire uterus is a blood infiltration of the uterine myometrium usually due to a massive retroplacental hematoma formation, the uterus becomes atonic, which leads to a high risk of PPH.[8] It is frequently associated with one or more of obstetrical complications such as placental abruption, uterine atony, bleeding from placenta previa, coagulopathy, amniotic fluid embolism, uterine rupture, iatrogenic perforation during dilation and curettage, as the blood invade the myometrium.[9,10] This leads to the conclusion that any uterine lesion can be the cause of the development of the Couvelaire's uterus, which in our case might have been the caesarean section.

The diagnosis of PPH is primarily based on the visualisation of excessive external bleeding and signs and symptoms of hypovolemia. It's important to note that up to 25-30% of a patient's blood volume (≥1500 ml) can be lost before BP falls and heart rate rises.[11,12] The diagnosis may be delayed in symptomatic women when bleeding is not observed. To avoid this, a symptomatic patient without severe vaginal bleeding should undergo additional examination, such as an abdominal CT, to determine the cause of the symptoms.[13] A USG examination of the uterus can be used as an alternative as it allows to evaluate a number of postpartum complications, including internal bleeding.[14] Lousquy et al suggests that ultrasound is more effective for the detection of intrauterine blood accumulation or retained products of conception than manual palpation, and therefore should be integrated into the evaluation of women with a high risk of severe PPH.[15] It is also confirmed that ultrasonographic diagnosis of Couvelaire's uterus may be restricted, but however possible.[16]

In the case of oliguria, it is important to exclude the cause of it, which can be unrecognized blood loss.[17,18] The hematological changes of pregnancy can hide the typical signs of hypovolemia, but evidence has shown that a fall of fibrinogen is a predictive value of a massive bleeding.[19,20]

In current case the patient didn't have significant external bleeding, however, she showed multiple signs indicative of PPH, including oliguria, low fibrinogen (0.4 g/l), gradual decrease in Hb (10.5 to 6.5 g/dl), impaired vision, light-headedness, confusion, fatigue, pallor, and a dense and rising uterus. Additionally, she had multiple serious risk factors for PPH, such as PE, c-section, and (severe) coagulopathy, therefore an imaging examination, such as abdominal USG or CT, was indicated to rule out internal bleeding.[21-24] Unfortunately, when arriving at the emergency unit, no additional examination of the uterus was done until the next morning, because based on the underlying condition, as well as typical signs and symptoms, HELLP syndrome was suspected first, and taking into consideration the potentially deadly complications of this syndrome, other conditions were overlooked, thus delaying the true diagnosis, which in this case was Couvelaire's uterus and severe PPH.

Routine prophylactic use of uterotonic drugs, as combination of oxytocin with misoprostol, significantly lessens the risk of PPH.[25] Some studies show that oxytocin reduces PPH and the need for therapeutic uterotonic agents by over 40%, and that adding mysoprostol is more effective than using oxytocin alone.[26,27]

Once the PPH is recognized, the conservative first line initial therapy is gaining uterine tonicity (up to 40 IU of oxytocin should be administered over 30 minutes), setting up a monitoring system and identifying the cause of bleeding (tone, tissue, trauma, and thrombin). Blood volume and coagulation must be restored, ensuring hemodynamic stability. Further options include bakri balloon tamponade, B-lynch suture, uterine artery embolization and ligation of uterine or uterine-ovarian arteries, which are aimed to decrease the need of hysterectomy.[28-30]

Scientific authors claim that Couvelaire uterus is managed conservatively, and hysterectomy is not required and should even be avoided.[31] For example, Shikanova et al presented a case of successful management of major complications of severe preeclampsia, where intraoperatively the placenta was totally separated, and Couvelaire's uterus and atonic postpartum haemorrhage

were managed by B-lynch suturing, intravenous carbetocin (100 μg), tranexamic acid (1 gram) and misoprostol (800 μg).[32] So in our case an earlier diagnosis may have changed the plan of management of the patient, and her fertility could have been preserved.

CONCLUSION

We have found very few studies that focus primarily on the connection between preeclampsia, postpartum hemorrhage and hypovolemic signs of the patient. We are concerned that although maternity health-care programs provide different algorithms, evaluation systems and follow-up criteria of the postpartum period, in cases where there are several overlying conditions, it is difficult to distinguish the right course of action. We believe that for patients with a high risk of PPH development improved diagnostic approach guidelines for hidden postpartum hemorrhage must be developed.

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