Preeclamptic pregnancy complicated by huge retroplacental hematoma and Couvelaire uterus – a case report

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Abstract

The paper presents a case of 28-year old pregnant woman, gravida 2 para 1, referred to the hospital at 33 weeks of gestation because of pregnancy induced hypertension and IUGR. The ultrasound screening, Doppler blood flow velocimetry, cardiotocography and 24-hour blood pressure monitoring was performed. Shortly after hospital admission the BP suddenly increased up to 196/137 and the patient reported a solid abdominal pain. Immediately performed ultrasound revealed a marginal placental abruption and a hematoma of 10 cm in the diameter located on the posterior uterine wall. During urgent cesarean section a Couvelaire uterus syndrome was diagnosed. Approximately 700 ml of blood clots from the uterine cavity and 700 ml of sanguineous fluid from peritoneal cavity were evacuated. Prematurely delivered neonate weighed 1580 grams, the Apgar score 3, 3 and 6 points in the 1st, 5th and 10th minute and was admitted to Neonatal Intensive Care Unit. The consequences and complication linked to retroplacental hematoma and Couvelaire uterus are also described.

Key words: preeclampsia, IUGR, placental abruption, Couvelaire uterus, uteroplacental apoplexy

Introduction

Ten percent of women worldwide experience high blood pressure during pregnancy [1]. One of its leading causes is pregnancy induced hypertension, defined as systolic blood pressure ≥ 140 and/or diastolic blood pressure ≥ 90 appearing after 20 weeks of gestation in a previously normotensive patient. Pregnancy induced hypertension (PIH) complicates about 6% of pregnancies and its prevalence is constantly rising [2]. Not only PIH, but every hypertension disorder in pregnancy increases the risk of both maternal and fetal morbidity and mortality. It concerns notably those patients who develop its severe forms – preeclampsia and eclampsia [2]. Preeclampsia, affecting 4.6% of pregnancies is diagnosed on the basis of PIH concurrent with proteinuria and/or one of the following: low platelet count (< 100 000/ml), high serum creatinine, doubling of hepatic transaminases’ level, pulmonary edema, visual or cerebral symptoms [3, 4]. Main fetal complications are intrauterine growth restriction (IUGR) and prematurity, both resulting in low birth weight and numerous diseases [5]. Possible maternal after-effects concern hepatic, renal, central nervous system and coagulation disorders. Preeclampsia and eclampsia cause 10-15% of direct maternal deaths worldwide [1].

One of the significant life-threatening consequence of preeclampsia is placental abruption, defined as partial or total separation of the placenta occurring before delivery of the fetus. The separated part of the placenta ceases to exchange gas and nutrients, resulting in fetal compromise. The risk of severe placental abruption in pregnancies complicated by hypertension is five-fold higher comparing to normotensive gravidas. What’s more, many studies suggest that antihypertensive treatment of chronic hypertension in pregnancy do not reduce this risk [6]. Placental abruption may be caused by maternal abdominal trauma, but mostly it is related to the underlying process of chronic placental pathology. Typical clinical features are abdominal pain, vaginal bleeding, tenseness of the uterus, often concurrent with hypertonic uterine contractions and non-reassuring FHR tracings. Possible maternal consequences are: obstetrical haemorrhage, emergency cesarean section, emergency hysterectomy, DIC and renal failure. A very rare complication is bleeding infiltrating the myometrium that results in condition called Couvelaire’s uterus [42]. In case of placental abruption, the frequency of maternal death increases seven fold. The adverse effects for the fetus include preterm delivery, low birth weight, low Apgar scores, perinatal asphyxia and death [7, 8]. Recently decreasing perinatal mortality in placental abruption nowadays amounts to approximately 12%. Almost 80% of perinatal deaths are intrauterine [9]. Although obstetricians have wide and detailed knowledge on the risk factors of placental abruption, still in most of the cases it is very hard to establish its cause.

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Case

A 28-year old female, gravida 2 para 1, was admitted to the Department of Perinatology and Gynaecology, University of Medical Sciences in Poznań because of pregnancy induced hypertension and IUGR. Her previous pregnancy 8 years before was uncomplicated and she gave vaginal birth at 41 week of gestation. The female infant weighed 3100 g and received 9 points in the Apgar score. The patient had no relevant medical history. Her family history was positive for hypertension and diabetes. Her partner was 33 years old and healthy.

Besides mild urinary tract infection treated with Furagin, the first half of current gestation was asymptomatic and uncomplicated. She denied smoking tobacco, drinking alcohol or using drugs, diet supplements or herbs during this pregnancy. She also denied abdominal trauma in the course of gestation. At 21 weeks of gestation, she noticed asymptomatic elevation of blood pressure (BP). The maximum BP was 140/100 mm Hg and no medication was administered. At 28 weeks of gestation, the BP increased up to 150/100 mm Hg and the ambulatory ultrasound revealed signs of fetal growth restriction (IUGR). The Doppler examination demonstrated elevated resistance and notching in both uterine arteries. Fetal anatomy was normal. At 29 weeks of pregnancy the patient was hospitalized in our Department for the first time, because of mild hypertension and lower limbs and face edema. She didn’t report any other symptoms. Obstetrical examination did not indicate any pathologies and the CTG tracings were reassuring. Results of blood laboratory tests suggested no liver or kidney pathology. The total protein in urine was an equivalent of 1+ on dipstick test. On ultrasound, the estimated fetal weight (EFW) using Hadlock formula was 931 g, which was below the 3rd percentile for gestational age. There was elevated resistance to blood flow and periodic notching in both uterine arteries. The biophysical profile was 8/8 points and the fetus’ presentation was cephalic. On 24-hour BP monitoring the mean BP values were 134 and 87.5 mm Hg for systolic and diastolic BP, respectively. The maximum BP was 180/136 mm Hg. The BP was normalized with methyldopa and patient was discharged from the hospital. The therapy with methyldopa was recommended, as well as BP monitoring 4 times a day, counting fetal body movements and ambulatory follow up in 7 days.

The patient was re-admitted at 33 weeks of gestation. She did not report any abnormal clinical symptoms or elevated BP. However, on admission the value of BP was 150/105 mm Hg. The vaginal discharge was normal, there was 0.5 cm external dilation and there were no signs of membranes rupture. The fundus of the uterus appeared to be only 5 cm above the umbilicus. On ultrasound, the fetus was in cephalic position and EFW was 1488 g, below the 3rd percentile for gestational age.

Fig. 1. Marginal placental abruption resulting in huge retroplacental hematoma

Fig. 2. Maternal surface of the placenta – notice huge placental hematoma on the upper ring of the placenta

Fig. 3. The appearance of the uterus immediately after delivery. Notice uteroplacental apoplexy characterized by bleeding permeating myometrium casing its characteristic appearance – a bluish and purple colour mottled by ecchymoses
Doppler ultrasound was normal. The gestational age was verified on the basis of ultrasound measurement in the I trimester. 4-hour BP monitoring was applied. Then, suddenly the BP increased up to 199/137 mm Hg and the patient complained of sharp, increasing abdominal pain and mild scotomata. On CTG the basal FHR was 160 bpm with no decelerations and good variability and regular, low-amplitude uterine contractions. The ultrasound performed rapidly revealed marginal placental abruption and a 10 cm in the diameter retroplacental hematoma. The emergency cesarean section was performed. During surgery, approximately 700 ml of sanguineous fluid was discovered in peritoneal cavity. The uterus appeared cyanosed with ecchymoses seen mainly in both uterine horns. Signs of placental abruption occurred on placental surface. Approximately 700 ml of blood clots were evacuated from the uterine cavity after placental extraction. On the basis of uterus’ appearance, the Couvelaire syndrome was diagnosed. Intravenous carbetocin and misoprostol per rectum were administered in order to constrict the uterus. During puerperium patient suffered from anemia, however it did not require blood transfusion. The delivered male infant weighed 1580 g and the Apgar score was 3, 3, 6. The pH was 7.17 in the umbilical artery and 7.21 in the umbilical vein. The newborn was admitted to the Neonatal Intensive Care Unit due to prematurity, anemia, neonatal respiratory distress syndrome, low birth weight and signs of perinatal hypoxia. Further-
more, the patient complained of sharp, increasing abdominal pain and mild scotomata. On CTG the basal FHR was 160 bpm with no decelerations and good variability and regular, low-amplitude uterine contractions. The ultrasound performed rapidly revealed marginal placental abruption and a 10 cm in the diameter retroplacental hematoma. The emergency cesarean section was performed. During surgery, approximately 700 ml of sanguineous fluid was discovered in peritoneal cavity. The uterus appeared cyanosed with ecchymoses seen mainly in both uterine horns. Signs of placental abruption occurred on placental surface. Approximately 700 ml of blood clots were evacuated from the uterine cavity after placental extraction. On the basis of uterus’ appearance, the Couvelaire syndrome was diagnosed. Intravenous carbetocin and misoprostol per rectum were administered in order to constrict the uterus.

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Discussion

Placental abruption, being the most common pathological cause of vaginal bleeding in late pregnancy, is a life-threatening incident complicating 0.4-1% of pregnancies [7, 8]. The ethiopathogenesis is multifactorial and still a subject of extensive research [7]. The incidence of this complication is increasing, most likely due to augmentation of risk factors’ prevalence. Risk factors of placental abruption include those of acute etiology, such as abdominal trauma or drug abuse. Others comprise of hypertension disorders, especially severe preeclampsia and eclampsia, polyhydramnios, prolonged rupture of membranes, short umbilical cord, uterine tumors and anomalies, chorioamnionitis, anemia, renal disorders, intrapartum fever and prior cesarean delivery. Sociodemographic risk factors enclose maternal age, parity, tobacco use and male infant sex. Also a history of previous ischemic placental disease, preeclampsia, IUGR or SGA and placental abruption increase the risk [7, 8, 11-14]. Hypertension in pregnancy appears to be the most frequent pathology concurring with placental abruption. Particularly a sudden increase of BP may result in acute placental abruption [14].

Every pregnancy complicated by preeclampsia should be considered high-risk. Early-onset preeclampsia (commencing before 34th week of gestation) is more dangerous than late-onset and increases the risk of fetal death almost 6 times. The risk of perinatal death or severe morbidity in this disorder is 16-fold higher than in normal pregnancy [15]. What’s interesting, preeclampsia occurs only in simultaneous presence of the placenta. There is growing evidence that pathological vasculatory development in the placental bed that begins early in pregnancy leads to preeclampsia. These abnormalities particularly concern remodeling and trophoblast invasion of the uteroplacental spiral arteries. Altered development lead to lack of arterial dilation resulting in increased resistance in uteroplacental vasculature [16, 17]. Following abnormal deep placentation and hypoperfusion lead to placental insufficiency, a known cause of fetal hypoxia and ischemia. These events finally may result in a wide range of pathologies – preeclampsia, IUGR, preterm labor, preterm premature rupture of membranes, spontaneous abortion in late pregnancy and placental abruption [17, 18]. Excessive antiangiogenic and inflammatory factors release following placental abnormal perfusion affect the function of the endothelium leading to multi-system response [20]. The organs that are exceptionally vulnerable to endothelial injury and inflammation are the brain, lungs, kidneys, liver, heart and vessels [23]. The daily urine protein excretion rising above or equal 0.3 g is a first sign of glomerulonephritis [26]. There are also data that altered immunologic response in the HLA system, recalling graft versus host disease underlie preeclampsia [19]. Likewise, oxidative stress occurring in ischemic conditions plays a significant role in preeclampsia, as it destroys the structure of the syncytium [21, 22]. Genetic basis of this pathology, suggesting the contribution of modifier genes and environmental factors is a subject of intensive research as well [25].
A patient with preeclampsia should be hospitalized and considering the often unusual symptoms of preeclampsia, every pregnant women who feels “poorly” should be screened for this disorder [27]. In preeclamptic patients it is crucial to maintain the correct blood pressure and avoid aggravation to eclampsia [23]. Magnesium sulfate is the medication of choice to counteract eclampsia and it is proven to decrease maternal mortality [2, 24]. Moreover, a course of betamethasone for fetal lung maturation should be administered in every case of early-onset preeclampsia. Both medications were administered to our patient.

It is widely known that delivery leads to recovery from preeclampsia and eclampsia. Considering the mode of labor in preeclamptic women, Steegers and colleagues suggest vaginal birth after 30 weeks of pregnancy, if fetal and cervical status allows it. However, when the fetus is growth restricted, cesarean delivery appears to be more safe [23]. Nassar et al. recommend labor induction for the patients with severe preeclampsia at ≥34 weeks of gestation, proving that almost 50% of them successfully deliver vaginally after induction [28]. Physicians agree that preeclampsia after 37 weeks of gestation is an unquestioned indication to deliver the fetus. Considering the onset of preeclampsia prior to 36 weeks of gestation, the management is still a subject of discussion. Commonly, expectant management with close surveillance is recommended, unless the disease has severe features. CTG non stress test, repeated ultrasound and Doppler velocimetry assessments are the methods of choice to monitor fetal well-being. Typical Doppler ultrasound findings are: increased pulsatility index or and notching in the uterine arteries and alterations in umbilical artery flows, such as AEDF and REDF [27].

According to Walker, placental insufficiency leads to intrauterine growth restriction in 30% of preeclamptic pregnancies. IUGR is referred as inability of the fetus to reach its predetermined growth potential, resulting in a SGA fetus (small for gestational age). Affected fetuses are at higher risk of fetal, neonatal and perinatal morbidity and mortality [30-32]. Furthermore, the risk of intrapartum death rises 5 to 10-fold [29]. Sometimes poor fetal growth in early pregnancy is the first symptom preluding preeclampsia. This less frequent form, referred as early-onset IUGR is associated with preeclampsia and pathological Doppler uterine artery pattern [33]. Severe fetal growth restriction, hypertensive disorder and male fetal sex – all found in our patient are proven to be significant risk factors of placental abruption [34]. There is enough research to state that most of the cases are the consequence of alterations in placental development resulting in chronic placental pathology [35, 36]. Large retrospective cohort studies by Rasmussen et al. prove that women whose first pregnancy was complicated by IUGR are at higher risk of placental abruption in subsequent pregnancies [40].

Diagnosis of placental abruption is made upon clinical symptoms confirmed by ultrasound evaluation. Unfortunately, often there are no ultrasound findings despite placental abruption – the sensitivity of USG in diagnosing this pathology is 25-50%, approximately. However, a presence of adverse ultrasound findings has a predictive value up to 88% [39]. Typical ultrasound finding is a retroplacental hematoma. Its visualisation and large diameter is prognostically unpropitious, likewise the percentage of the surface of placental detachment [37]. Other potential ultrasound findings are: subchorionic fluid accumulation, echogenic debris in the amniotic fluid and a so-called Jello sign. Jello sign is diagnosed when a thickened layer of placenta shimmers as the patient moves during examination [39].

Severe placental abruption, referred as an abruption concurrent with unstable maternal status and/or non-reassuring CTG tracing, is an indication for immediate cesarean delivery. Because of the high risk of coagulopathy, blood, platelets, fresh frozen plasma and cryoprecipitate should be present at the operating room. Patients with severe abruption, DIC or hypovolemic shock are at high risk of multiorgan failure and death. Close surveillance of the mother and neonate is necessary also postpartum. Prevention of uterine atony, monitoring blood pressure, blood loss and laboratory tests are crucial. Uterine atony is managed by oxytocin, prostaglandine analogues and methylergonovine. It is important to remember that methylergonovine is contraindicated in preeclampsia as it increases the blood pressure [41].

In our patient, the ultrasound diagnosis of a retroplacental hematoma was confirmed during surgery. What’s more, during c-section a Couvelaire uterus with blood that reached the peritoneal cavity was diagnosed. Couvelaire uterus, also described as uteroplacental apoplexy, is a rare, life threatening condition characterized by bleeding permeating myometrium, rarely crossing to the peritoneal cavity or infiltrating broad ligaments and ovaries [42, 44, 47]. It was first described in 1911 by French obstetrician, Alexandre Couvelaire. The diagnosis of Couvelaire uterus can only be established on the basis of biopsy or visual examination during surgery [42, 43]. Its characteristic appearance is a bluish or purple colour, mottled by ecchymoses. In such case, the
risk of uterine rupture during constriction is very high, as the myometrium is extremely weak. On the other hand, the patients with a Couvelaire uterus are at higher risk of uterine atony and haemorrhage, because the blood-infiltrated myometrium is less able to contract and exsanguination. In our patient carbetocin and misoprostol with satisfactory effect was administered. Surgical methods of managing uterine atony and haemorrhage, applied when pharmaceutical methods fail, are uterine vessels ligation, uterine compression sutures, embolization and obstetrical hysterectomy as last resort. The suture technique particularly recommended and effective in managing haemorrhages in patients with hypertensive disorders, placental abruption and Couvelaire uterus is the B-Lynch technique [52]. Hysterectomy was the management of choice for many decades [44]. Nowadays the recommended treatment is conservative [42].

The etiology of Couvelaire uterus remains unidentified and few available reports link it with placental abruption, placenta previa, preeclampsia, coagulopathy, amniotic fluid embolism and uterine rupture [42-45, 47-48]. It is estimated that Couvelaire uterus develops in 5% of patients with placental abruption [51]. It is important to emphasise that Couvelaire uterus is not always a result of placental abruption. A very recent case described by Shreedevi and colleagues presents a diagnosis of Couvelaire uterus in the absence of placental abruption and any other pathologies [49]. A different recent case report describes a Couvelaire uterus that developed after a dilation and curettage in missed abortion of 13 weeks old pregnancy. The mechanism suggested by authors is iatrogenic perforation of the uterine wall during curettage, letting the blood infiltrate the myometrium. Hysterectomy was indispensable [46]. In the case reported by Gogola et al. the debris and air bubbles in the uterine veins were concurrent with the Couvelaire uterus and the authors suggest the blood-permeated area of uterus to be the “portal” for the debris to reach maternal vessels and cause amniotic fluid embolism [48]. Avery and Wells retrospectively describe the ultrasound findings in Couvelaire syndrome confirmed during cesarean section. Those findings are large retroplacental blood clot and blood dissecting into the myometrium. They state that ultrasound diagnosis of Couvelaire uterus is limited, but possible [50].

One should be aware that a patient diagnosed with preeclampsia should control her blood pressure after discharge from the hospital. Sometimes hypertensives are essential during puerperium. There are also reports that these patients should avoid non-steroid anti-inflammatory drugs in treating post-cesarean pain. The meta-analysis from 2015 reports that among women who experienced preeclampsia, 20% will develop hypertension and 16% will have preeclampsia in subsequent pregnancy [10]. Low doses of ASA [53] and calcium supplementation [2] appear to be useful in prevention of preeclampsia among high-risk patients. What is important, women with a history of preeclampsia have higher risk of cardiovascular disorders in the future, most probably as a result of systemic endothelial dysfunction [3, 54]. There is no scientific data on the recurrence of Couvelaire uterus.

References


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