Complete Molar Cervical Previal Pregnancy with a Viable Co-Twin and Placental Percreta Following Corporal Hysterotomy: A Case Report

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Abstract

Background: Cases of coexistence of a twin molar pregnancy with a living second fetus are known in the literature, with different outcomes and treatment options, from uterine preservation procedures and cesarean section to hysterectomy due to the vital threat to the pregnant woman. Later pregnancies are dependent on uterine preservation - evacuation procedures in the first pregnancy.

Aim: Obstetrics forensic commentaries on the management of two different trophoblastic diseases.

Methods: This extremely rare case of previal cervical molar pregnancy in a twin pregnancy and primary completion by laparotomy in the first pregnancy is directly related to the occurrence of placental percretism with hematoperitoneum at 30 weeks of pregnancy due to distension and rupture of a placental blood vessel and supracervical hysterectomy. Conclusion: Thus, these problems became professional failures with an irreversible "domino effect". After such decisions and treatment, menstruation and reproduction were prevented by hysterectomy, and the possible treatment options for such conditions in the first and second pregnancies are discussed in the text with a forensic perspective.

Keywords: molar pregnancy, cervical pregnancy, placenta accreta spectrum, hysterectomy, treatment, medicolegality

Introduction

Cases of coexistence of a twin molar pregnancy with a living second fetus are known in the literature, with different outcomes and treatment options, from uterine preservation procedures and cesarean section to hysterectomy due to the vital threat to the pregnant woman. Later pregnancies are dependent on uterine preservation - evacuation procedures in the first pregnancy (1-4). I present an example from my own clinical and forensic practice, but with a completely different trophoblastic entity (cervical previal gestational trophoblastic disease and morbid placentation – placental percretism) in a second pregnancy with a bad perinatal outcome, along with a comment on the procedures in the treatment of both pathological pregnancies.

Case report

A 33-year-old healthy nulliparous woman with a history of one artificial abortion was treated for infertility: hysteroscopic polypectomy and laparoscopic chromopertubation were performed five months before successful one blastocyst artificial homologous insemination. Early combined screening indicated the absence of significance for chromosomal abnormalities with extremely high MoM free BHCG, and in the 14th week ultrasound showed inhomogeneous chorial tissue cervicoisthmically with a vital fetus in a separate gestational sac intracavitary, so it was decided to perform early amniocentesis. So, it was a monozygotic biamniotic pregnancy with trophoblastic degeneration of the first pregnancy and a second healthy fetus. In the 17th week of pregnancy, the pregnant woman bled profusely and was admitted to the clinic, where ultrasound found a dilated cervix filled with a mass similar to inhomogeneous molar tissue with numerous anechoic inclusions with an unclear border towards the wall of the cervix (Figure 1.) with an intact second gestational sac and healthy fetus. The ultrasound findings suggested a suspected complete mole probably after a missed abortion of the first twin with a normal pregnancy of the second twin. Due to laboratory findings of hyperthyroidism, propylthiouracil 3x200 mg and propranolol 2x20 mg were prescribed. Other laboratory findings and blood pressure were normal. Considering the above, the gynaecologists, in agreement with the patient, decided on a laparotomy approach to evacuate both pregnancies, so they performed a corporeal hysterotomy and evacuation of the uterine cavity under general anaesthesia at the 19th week of pregnancy. The surgical procedure and the postoperative course went well with the prescribed 3 doses of concentrated erythrocytes (KE) without bleeding from the cervix. The pathohistological findings indicated complete molar degeneration of the first conceptus and the normal anatomy of the second fetus. The karyotype of the healthy twin was normal.

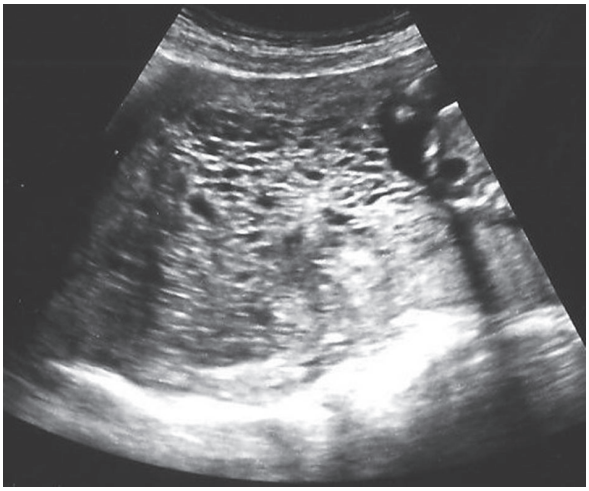

Figure 1. *Ultrasound pictures of cervical previal molar pregnancy: inhomogeneous mass with multiple anechogenic areas in the dilated cervix. Above the cervix, an intact gestational sac with a healthy fetus.*

A year later, the patient became pregnant spontaneously with an orderly course of pregnancy until the 30th week, when she was admitted to the clinic due to peracute and continuous abdominal pain without vaginal hemorrhage. On admission, signs of an acute abdomen are evident, and free fluid in the abdomen is found on ultrasound, along with an orderly biophysical profile of the fetus and cardiotocography and the finding of anterior invasive malplacentation – placental percretism. Due to the acute abdomen, an

emergency cesarean section is indicated, and after the Pfannenstiel relaparotomy, 1000 mL of fresh blood and clots are found in the abdomen with a 5 cm zone of percretism in the scar from the previous corporeal hysterotomy, which is actively bleeding. A fundal hysterotomy was performed on a freshly dead male newborn 1530 g/ 44 cm, without the effect of resuscitation. With regard to cicatricial placental percretism, the gynaecologist on duty decides on a supracervical hysterectomy, which is performed with the transfusion of 4 KE, 2 fresh frozen plasm and 2 cryoprecipitates. The pathohistological findings of the uterus indicated a placenta percreta through the scar, and the autopsy findings of the stillborn showed a premature morphological finding of the organs with signs of asphyxia.

In the case of these two different trophoblast entities that became directly conditioned: molar cervical pregnancy in the first pregnancy and morbid (invasive) placentation (placenta accreta spectrum - PAS) in the next pregnancy from the clinical and forensic aspects. According to previous works, about 300 cases of coexisting complete molar twin pregnancy with a living second fetus have been published as an extremely rare obstetric phenomenon. Obstetrical complications such as bleeding and spontaneous abortions, hyperthyroidism, preeclampsia and fetal death are not rare in such cases and are related to the metabolic-hormonal disorder of such pathological pregnancies (placental tissue). Molar trophoblast degeneration can be partial or complete, as described in rare case reports, but also the development of persistent intermediate or malignant forms of gestational trophoblastic disease. Ultrasonography is the basic method of early diagnosis and monitoring of pregnancies along with biochemical monitoring of HCG (5-7).

Wang et al. have recently published a case report of a coexisting molar pregnancy with a healthy fetus and several episodes of bleeding during pregnancy. A live eutrophic newborn with a normal karyotype was born by cesarean section, and the diagnosis of a complete placental hydatid tumor was confirmed pathohistologically (6). Rodriguez et al. presented a case of complete hydatidiform mole and coexisting fetus with premature delivery at 28 weeks due to chorioamnionitis (7). Gupta presented a similar case but with termination of pregnancy in the 1st trimester due to persistent gestational trophoblastic disease (8). The same case discovered in the second trimester with the birth of a healthy child was presented by Lin et al. (9) but with suspected choriocarcinoma that was treated with chemotherapy and relapsed with suspected intermediate trophoblastic tumor.

In modern literature, cervical ectopic pregnancies, even if they are rare or molar, are solved very successfully with preservation procedures, such as cerclage with evacuation curettage, evacuation curettage with gauze or balloon tamponade (9-10), and hysterectomy is reserved only for severe refractory hemorrhage and invasive cervical malplacentation with obstetric shock development (11-12). PAS is today an iatrogenically conditioned modern disease of the 21st century in direct correlation with the extremely high incidence of cesarean sections and other uterine procedures and thus increased maternal morbidity and mortality due to hemorrhage and peripartum hysterectomies (12-16). Although the outcomes of such pregnancies in more than 50 cases are completed with the birth of healthy children, the outcomes of such bizarre pregnancies depend on the place of placentation, the type of molar degeneration, the comorbidity that developed during the pregnancy, and possible PAS (16). Although there were no consequences in terms of litigation, as a clinician and gynaecological-obstetrical forensic expert, I am of the opinion that a transvaginal procedure to evacuate the cervical molar pregnancy should have been performed after placement of the cerclage with local anemization and ligation of the cervical branches, when the integrity of the second twin would have been preserved with a high probability. Furthermore, even when percretism was noticed during relaparotomy and cesarean section with, unfortunately, a recently deceased child due to most likely asphyxia, resection of the uterine wall and sutures with preservation of the uterus should have been performed.

This extremely rare case of previal cervical molar pregnancy in a twin pregnancy and primary completion by laparotomy in the first pregnancy is directly related to the occurrence of placental percretism with hematoperitoneum at 30 weeks of pregnancy due to distension and rupture of a placental blood vessel and hysterectomy. Thus, these problems became professional failures with an irreversible "domino effect". Because of the above, I consider sharing this presentation as a contribution to the importance of a collegial approach and the forgotten Hippocrates' "primum nil nocere".

Declarations

Authors' contributions: Dubravko Habek designed the study, wrote the main manuscript and critically reviewed the manuscript.

Ethics: Ethical approval and informed consent statements: Ethics Committee of Clinical Hospital Sveti Duh Zagreb. Nbr. 01-03-2089/4 from May 12, 2022. The patient gave verbal consent to the publication of data from her case report.

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