

Original Research Article

Complete hydatidiform mole with hook effect and severe anemia in 36-week size uterus: A rare case report from India

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Abstract: We present a rare case of a 27-year-old woman with a complete hydatidiform mole, which is an abnormal pregnancy that typically causes vaginal bleeding and elevated serum β -human chorionic gonadotrophin (hCG) levels. However, this patient had a 36-week size uterus, severe anemia, and vaginal bleeding, but with a non-detectable β -hCG level, likely due to the "hook effect." She also had chronic pancreatitis, with a dilated hepatobiliary system and free fluid in the pelvis, caused by the enlarged uterus compressing the abdominal organs. The patient received blood transfusions and was treated with dilation, evacuation, and serial monitoring of serum β -hCG levels. It is important to note that a negative urine pregnancy test or non-detectable β -hCG level should be followed up with a repeat measurement on a diluted sample to avoid the "hook effect."

Keywords: Hydatidiform mole; Anemia; Pregnancy.

1. Introduction

A Hydatidiform mole refers to an abnormal pregnancy where there is trophoblastic proliferation and villous edema in the placenta [1]. There are two types of molar pregnancy, partial and complete. In partial moles, fetal or embryonic tissues are present, and there are chorionic villi of different sizes with focal trophoblastic hyperplasia and swelling, as well as marked villous scalloping. On the other hand, complete moles do not have any fetal or embryonic tissues and are characterized by diffuse trophoblastic hyperplasia and hydropic swelling of villi, along with marked atypia of trophoblast at the implantation site [2].

Most cases of complete molar pregnancy are characterized by vaginal bleeding and significantly elevated levels of β -human chorionic gonadotrophin (hCG) [3,4]. We report a rare case of complete molar pregnancy where the patient had an enlarged uterus that was pressing on nearby organs, severe anemia despite the lack of vaginal bleeding, and a relatively low serum β -hCG level due to the "hook effect."

It is important to be aware of this unique presentation, as the absence of elevated β -hCG levels in complete molar pregnancy can be misleading and delay the diagnosis. In such cases, clinical suspicion should be maintained, and additional diagnostic measures, such as ultrasound examination and histopathological analysis of the evacuated tissue, may be necessary to confirm the diagnosis. Early recognition and appropriate management of complete molar pregnancy are crucial to prevent complications such as persistent gestational trophoblastic neoplasia and its potential for metastasis [5].

2. Case report

A 27-year-old multigravida presented to the emergency department with amenorrhea since last 1-year, abdominal pain, and vaginal bleeding 37 days after her last menstrual period. She also reported right flank pain and urinary frequency that started two days before. She denied having a fever or chills. She had two pregnancies and two live births. She had no medical, surgical, or social history that was relevant, and there was

no family history of uterine, colon, ovarian, or breast cancer. She had a normal blood pressure and temperature but had tachycardia (124 beats per minute). Her abdomen was swollen, and her uterus was 36 weeks in size but not tender. Her initial blood tests showed severe anemia (hemoglobin: 7.0 g/dL), slightly elevated creatinine (1.0 mg/dL), and normal liver function tests. Her β -hCG was not detectable. An abdominopelvic ultrasound and magnetic resonance imaging (MRI) showed an enlarged uterus (23.8 × 19.6 × 15.7 cm) with a large, heterogeneous mass inside it that contained multiple discrete anechoic spaces consistent with a complete mole (Figures 1 and 2).

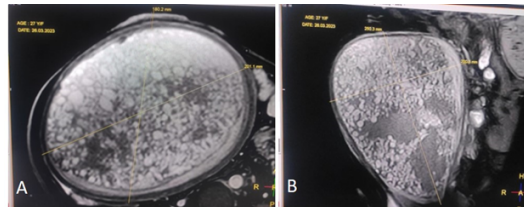


Figure 1. Sagittal (a) and coronal (b) views of an abdominal-pelvic magnetic resonance imaging showing an enlarged uterus (23.8 × 19.6 × 15.7cm) with a large intrauterine heterogeneous mass

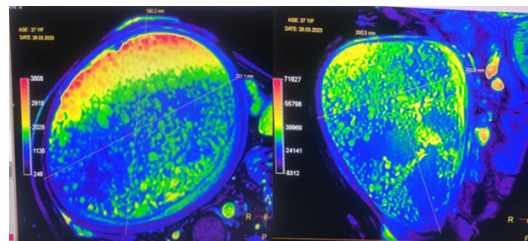


Figure 2. A pelvic 3D MRI showing a large intrauterine heterogeneous mass that includes multiple discrete anechoic spaces consistent with complete mole

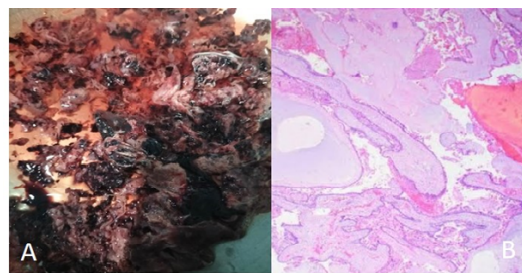


Figure 3. Disproportionate increase in the size of the uterus (a) and histology image (b) showing complete hydatidiform mole

The MRI revealed moderate chronic pancreatitis with distal obstruction from the enlarged uterus. Her ovaries and urinary bladder were normal, and her chest X-ray was also normal. She received 2 units of packed red blood cells but developed heavy vaginal bleeding (estimated blood loss: 1500βmL) and required dilation and evacuation, along with 2 more units of whole blood transfusion. The histologic review confirmed the presence of a complete hydatidiform mole (Figure 3). She had minimal vaginal bleeding in the postoperative period, and her hemoglobin was 7.8g/dL. Her serum β -hCG level was high (642,763mIU/L) on the first postoperative day but dropped gradually (Day 3: 59749mIU/mL) until it reached undetectable levels after 120 days. The patient's consent was obtained for publishing this information.

3. Discussion

Hydatidiform moles are rare, with a prevalence of approximately 1 in 1,000 pregnancies in Europe and North and South America, but 5- to 15-fold higher in East Asia [5]. Complete moles have a unique karyotype

of 46,XX, with all chromosomes derived from the father [6,7]. The classical clinical signs of hydatidiform mole include vaginal bleeding, disproportionate increase in the size of the uterus, high levels of β -hCG for gestational age, hyperemesis gravidarum, cystic enlargement of the ovaries, and eventually hyperthyroidism and pregnancy-induced hypertension [8]. However, earlier diagnosis with ultrasound and β -hCG assays has changed the typical clinical presentation [9,10].

According to a study of 113 cases of hydatidiform mole in China from 1989 to 2006, vaginal bleeding was the primary symptom in 83.2% of cases, followed by an enlarged uterus in 46% of cases. However, the overall occurrence of vaginal bleeding and preeclampsia was lower than in historical data [9]. Similarly, a review of 189 cases in Italy from 1992 to 2004 found that while vaginal bleeding remained the most common symptom, its frequency had decreased from 74% in cases diagnosed between 1977 and 1985 to 51%. This decrease was attributed to advances in technology and earlier diagnosis. The incidence of other symptoms such as a disproportionately large uterus and bilateral ovarian cysts had also decreased [10].

Our patient presented with a 36-week size uterus with abdominal pain and vaginal bleeding. After removing the molar pregnancy, the abdominal pain and distension subsided, and the creatinine level normalized. Notably, the patient was severely anemic, had a negative urine pregnancy test, and her β -hCG levels were undetectable, despite having an enlarged uterus. This can be explained by the accumulation of blood inside the uterus, causing it to enlarge quickly and leading to the sudden onset of heavy vaginal bleeding. This possibility is supported by the rapid enlargement of the uterus and the absence of any other causes of acute severe anemia. Therefore, in similar cases of molar pregnancy, acute anemia should be interpreted with caution, as heavy vaginal bleeding is likely to occur spontaneously or during the evacuation procedure.

Complete molar pregnancies are often correctly diagnosed by ultrasound, especially with the presence of characteristic placental features such as cystic changes and overt masses [11]. In our patient, the diagnosis was aided by an ultrasound that revealed a large, heterogeneous mass containing multiple anechoic spaces within the uterus. The diagnosis was later confirmed by histologic evaluation of the evacuated tissues. MRI imaging, which provides 3D images, is now readily available and can be even more helpful than ultrasound. Patients with complete molar pregnancies typically have an abnormally high level of β -hCG, which can exceed 100,000 β mIU/L in approximately half of cases [3]. Our patient, however, had a non-detectable β -hCG level at the initial assessment, which can sometimes cause a delay in diagnosis. However, the fact that the β -hCG level increased significantly after the evacuation suggests that the initial measurement was falsely low due to the "hook effect." This effect occurs when extremely high β -hCG levels saturate both the immobilized capture antibodies and the free tracer antibodies, preventing the formation of the necessary sandwich structure for a positive test result. This can lead to a falsely low serum β -hCG test or falsely negative urine pregnancy test [12,13]. Diluting the sample to reduce the amount of β -hCG tested can overcome this limitation. In our patient, the evacuation of the molar pregnancy reduced serum β -hCG concentrations, leading to a more accurate measurement.

The preferred treatment for molar pregnancy is suction dilation and evacuation, which involves dilating the cervix and using suction to remove the abnormal tissue. To reduce the risk of uterine atony, intravenous oxytocin is usually given following cervical dilation. One potential complication of molar pregnancy is gestational trophoblastic neoplasia, which can develop in 6-32% of cases of complete moles. Because of this risk, it is recommended to monitor serum β -hCG levels frequently until they reach undetectable levels, and then continue with monthly measurements for an additional 6 months [2]. It is also important for patients to use reliable hormonal contraception during this monitoring period.

4. Conclusions

A complete molar pregnancy can manifest as an enlarged uterus that presses on nearby organs and severe anemia with negative urine pregnancy test and non-detectable β -hCG, with or without vaginal bleeding due to the accumulation of blood in the uterine cavity. If a complete mole pregnancy is suspected and the β -hCG level is low or non-detectable, it is recommended to repeat the measurement on a diluted sample to prevent the "hook effect" from leading to a false-negative result.

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