

Hydatidiform mole coexisting with normal foetus: A rare presentation of a case report from University of Maiduguri Teaching Hospital

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Abstract

A twin pregnancy comprising a complete hydatidiform mole coexisting with a foetus is a rare obstetric condition with an incidence of 1 in 22,000 to 1 in 100,000 pregnancies. The management of such case is challenging due to the associated risk of maternal and foetal complications. We report a case of a 25-year-old woman, gravida 2, para 1 with a normal intrauterine pregnancy coexisting with complete hydatidiform mole. An ultrasound scan demonstrated normal foetus and placenta along with coexistent intrauterine echogenic mass with features of hydatidiform mole. The microscopic examination of the abnormal placenta confirmed complete hydatidiform mole. Although twin pregnancy with complete hydatidiform mole and coexistent foetus is associated with increased risk of developing maternal and foetal complications, continuation of pregnancy may be an acceptable option under close monitoring to detect early signs of complications.

Introduction

Gestational trophoblastic diseases (GTDs) are disease condi-

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tions arising from the placenta.¹ Twin pregnancy with a complete hydatidiform mole coexisting with foetus (CHMCF) is very rare with an estimated incidence of 1 in 22,000 to 100,000 pregnancies.¹⁻³ This is in sharp contrast to the occurrence of hydatidiform mole alone which has an incidence of 0.57 to 1.1 per 1000 pregnancies in North America and Europe, and 2.0 per 1000 pregnancies in developing countries of Southeast Asia and Japan.⁴ Several studies from various parts of Nigeria have reported incidence ranging from 1.7 to 6.0 per 1000 births.⁵ The estimated incidence of hydatidiform mole in Northeast Nigeria is 3.8 per 1000 pregnancies.⁶ Complete hydatidiform mole coexisting with foetus cases are at high risk of spontaneous abortion, chromosomal abnormalities, preterm delivery, intrauterine foetal death, intractable vaginal bleeding, preeclampsia, thyrotoxicosis and persistent trophoblastic disease (PTD).^{2,3,7} Although it is still a rare finding, in recent times, there has been a relative increase in the incidence of multiple pregnancies with coexistent hydatidiform moles due to ovulation induction therapy and *in vitro* fertilization.⁸

Case Report

A 25-year-old woman, gravida 2, para 1 with one previous normal delivery, presented to the Emergency Gynaecology Unit of the University of Maiduguri Teaching Hospital with a history of recurrent vaginal bleeding of 1week duration. She was not sure of her last menstrual period but said to be 21 weeks' pregnant. The abnormal vaginal bleeding was profuse with passage of blood clots (estimated at 150 ml) and vesicular materials. It was associated with dizziness, headache and bilateral pedal oedema. There was history of exaggerated pregnancy symptoms characterised by an average of five episodes of vomiting per day.

On general examination, she was in respiratory distress with flaring of ala nasae, pale, and having bilateral pitting pedal oedema up to upper third of the legs. Her temperature was 37.3°C. Cardiovascular examination revealed tachycardia (pulse rate was 120 beats/minute). The blood pressure was 140/80 mmHg. There was a third heart sound with murmurs along with bilateral basal crepitations in the lower lung fields. On abdominal examination, the symphysis-fundal height (SFH) was equivalent to 33 weeks which was higher than the estimated 21-week pregnancy. There was also difficulty in palpating the foetal parts.

The admitting haematocrit level (using microhaematocrit) revealed anaemia with a value of PCV of 22%. Dipstick urine test (urinalysis) showed moderate proteinuria of plus 2 (100 mg/dL) and haematuria. The serum electrolyte, urea and creatinine levels were within normal limit. The liver function test was also within normal limit. The urine pregnancy test (PT) in serial dilution indicated neat positive, 1/50 positive, 1/100 positive and 1/200 negative. A trans-abdominal ultrasound scan (US Scanner Logiq S8, General Electric) done by a senior registrar radiologist revealed a

huge echogenic mass in excess of 15 cm in diameter with *snow-storm* appearance occupying the lower uterine pole, and a coexisting well-formed foetus of approximate gestational age (AGA) of 17 weeks + 3 days, no cardiac activity.

On 4th day of admission, she developed preeclampsia with a blood pressure of 160/100 mmHg along with the pedal oedema and severe proteinuria. She was counselled and had consented for termination of pregnancy by use of misoprostol and subsequent evacuation if need be. She successfully expelled (after 2 doses of misoprostol 600 µg) the well-formed foetus and the molar tissues, with a follow up evacuation with Karman's cannula curettage. The expelled and evacuated products of conception showed grossly normal foetus, intact placenta, abundant fragments of molar tissue and blood clots (estimated blood loss: 300 mL). She was transfused four pints of blood during the period of admission. Subsequent post partum days were uneventful with PCV level of 27%.

Histopathological examination of the products of conception revealed macroscopic features of normal female foetus with biometric and growth parameters consistent with 19 weeks of gestational age. There was accompanying abundant fragments of placental tissue with multiple tiny vesicles having a characteristic *bunch of grapes* appearance (Figure 1). Microscopic examination by a consultant pathologist using LEICA DM750[®] confirmed the diagnosis of complete hydatidiform mole; showing abnormally enlarged and hydropic chorionic villi with circumferential trophoblastic hyperplasia (Figure 2). The other placental fragments showed normal placental parenchyma.

Her general health condition became stable and was discharged home on 11th day of admission (6th day post-evacuation), to come for follow-up after two weeks with results of serial dilution. However, the patient was lost to follow-up.

Discussion

Worldwide, there are only few reported cases of complete hydatidiform mole coexisting with foetus (CHMCF) relative to hydatidiform mole alone due to the rarity of the former.¹⁻³ There are two different pathological entities of this term based on cytogenetic analysis.⁹⁻¹¹ These are: i) twin gestation in which one of the twin is a diploid foetus with a normal placenta and the other twin is a complete hydatidiform mole containing a diploid set of 46 chromosomes, all of paternal origin and no traces of foetal parts; ii) twin gestation in which one of the twin is a diploid foetus with normal placenta and the other twin is a triploid foetus with partial hydatidiform mole placenta that is derived from dispermic fertilization of a haploid normal oocyte.⁹⁻¹¹ Categorization of the case is essential for proper management because complete and partial moles have distinct foetal and maternal complications.⁹⁻¹¹ Partial hydatidiform mole is commonly associated with multiple foetal anomalies and the indication for a termination of pregnancy is evident.^{1,4} In contrast, the foetus may be normal in a twin pregnancy with a CHMCF although continuation of pregnancy is frequently associated with severe maternal complications such as preeclampsia, intractable vaginal bleeding, hyperemesis gravidarum, hyperthyroidism, persistent trophoblastic disease and HELLP (haemolysis, elevated liver enzymes, and low platelet count) syndrome.^{3,7,9-11} In such life-threatening situations, the termination of pregnancy can also be considered.^{4,7,12} It is noteworthy that no foetal anomalies have yet been described and there are several instances in which foetal survival have been reported.¹³ The index case presented to our emergency gynaecology clinic with recurrent vaginal

bleeding complicated by anaemic heart failure. She also developed preeclampsia and deteriorating health condition. There was difficulty in palpating the foetal parts on clinical examination. Sonography revealed absent cardiac activity. Consequently, she was counselled and consented for termination of pregnancy.

A Prenatal diagnosis of hydatidiform mole and coexistent foetus can be made based on the clinical symptoms and signs, physical examination, sonographic findings, and abnormal biochemical data of significantly elevated beta hCG value.^{3,8,10,13} Prenatal diagnosis by chorionic villus sampling, amniocentesis, or foetal cord blood sampling also enables the physician to distinguish cytogenetically between diploid and triploid foetuses.^{3,9,13} In addition, molecular analysis is very important in prediction of the maternal outcome.³ The diagnosis of a CHMCF can be made by first-trimester sonography.^{2,3,8,13} The ultrasound scan shows clearly a

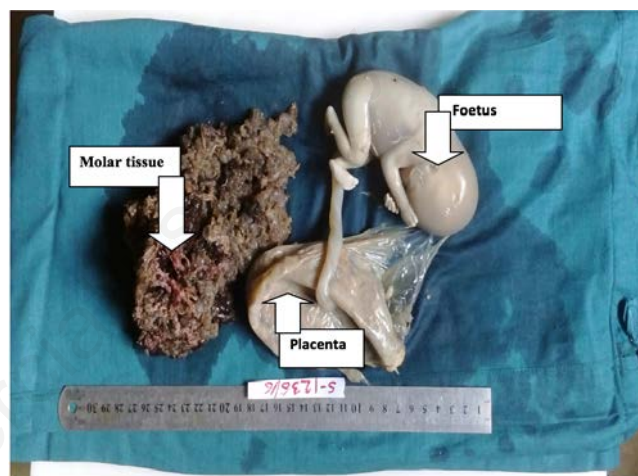


Figure 1. Molar tissue and normal foetus with grossly intact normal placenta.

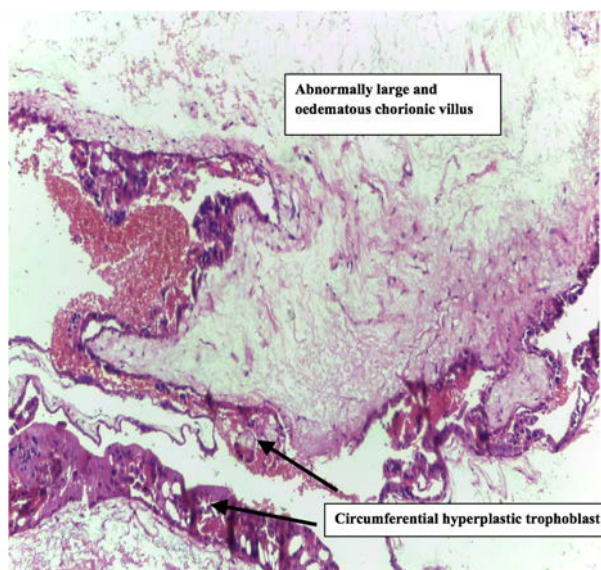


Figure 2. Complete hydatidiform mole showing abnormally large and oedematous chorionic villi with circumferential hyperplastic trophoblast. (H&E x 100)

normal-appearing foetus and a normal placenta connecting with a sharply defined molar tissue; with its typical ultrasonographic findings of abnormal placental echoes showing complex cystic pattern with a 'snowstorm' appearance.^{3,10} The serum beta hCG titre in hydatidiform mole is generally higher than is seen in non-molar gestations.² The beta hCG titre can be a helpful marker in the management of a CHMCF because it is usually highest at the beginning of the second trimester of pregnancy.^{2,3,8,11} It has been observed that the excessive production of beta hCG may identify gestational trophoblastic disease in patients during pregnancy and also identifies patients with a high risk of gestational trophoblastic disease.^{2,3,8,10,12} The index case was 21-week pregnant and had presented with clinical features consistent with molar gestation. The ultrasonography done showed a huge echogenic mass in excess of 15cm in diameter with *snowstorm* appearance occupying the lower uterine pole, and a coexisting foetus (Figure 3). The beta hCG titre was also elevated as indicated by Pregnancy Test in serial dilution. A histopathological diagnosis of complete hydatidiform mole was made coexisting with normal placenta and foetus.

The diagnosis of CHMCF also includes ancillary investigations such as foetal karyotyping,^{1-4,9,12} cytogenetic analyses of the normal placental sample and molar tissue by DNA flow cytometry,³ and fluorescence in situ hybridization (FISH) specific for X and Y chromosomes.³ Other investigations are the polymerase chain reaction (PCR) and microsatellite genotyping of DNA preparations obtained from blood samples of the woman and her partner and from formalin-fixed paraffin embedded placental and molar tissue for determination of zygosity,¹⁴ and use of immunohistochemical stain p57 on molar tissue to distinguish between partial and complete hydatidiform moles.⁸ Although these ancillary investigations were not available for our patient, as in some documented cases,^{2,7,8,11,12} a diagnosis of CHMCF was made based on the characteristic clinical features, typical ultrasonographic findings, elevated beta hCG and confirmatory histopathological examinations.

The management of patients with CHMCF still carries a risk but there are reported cases of successful deliveries and survivors.^{1,2,3,7,8,9,11,13,14} Therefore, patients with CHMCF may be allowed to continue pregnancy under strict observation and mater-

nal follow-up with regular ultrasonography and beta hCG level monitoring during antenatal care and in the postpartum period.

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Figure 3. Trans-abdominal sonogram at 17 weeks' gestation showing a normal-appearing foetus and a normal placenta connecting with a sharply defined huge echogenic mass with *snowstorm* appearance occupying the lower uterine pole.