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ORIGINAL ARTICLE

A Case of a Complete Hydatidiform Molar Pregnancy with a Co-existent Live Fetus

Aradhana Aggarwal[1] · Chanchal Singh[2] · Shreyasi Sharma[3] · Apoorva Reddy[4] · Anita Kaul[5]

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Abstract Complete hydatidiform mole with a co-existent healthy fetus (CHMCF) is a rare occurrence. The estimated incidence is about 1/22,000 to 1/100,000 pregnancies. As CHMCF is often associated with a high risk of pre-eclampsia, hyperthyroidism, bilateral ovarian cysts, intrauterine fetal demise and malignant neoplasia, fetal survival should always be weighed against the risk of complications of molar pregnancy. We report a case of a 35 years-old patient with raised β-hCG levels of 10.2 MoM and sonographic findings suggestive of a molar pregnancy at 15 weeks, who chose to continue the pregnancy after extensive counselling. With close surveillance, we managed to continue the pregnancy till 36 weeks, when the pregnancy was terminated via an elective Caesarean in view of fetal growth restriction with oligohydramnios, cerebral redistribution and previous 2 LSCS. A baby boy weighing 1600 grams was born with an Apgar score of 8, 9. Placental histopathology and microscopy showing large distended and cystic dilated villi with odematous cores was suggestive of a hydatidiform mole confirming our diagnosis of CHMCF.

Keywords Complete hydatidiform mole with a co-exsistent healthy fetus · Pre-eclampsia · High risk pregnancy · Molar pregnancy · Dichorionic twin pregnancy · Triploid fetus · Partial hydatidiform mole

✉ Aradhana Aggarwal
dr.aradhanabhalla@gmail.com

Chanchal Singh
chanchalsngh@gmail.com

Shreyasi Sharma
shreyasi2003@gmail.com

Apoorva Reddy
apoorvagalaxy@gmail.com

Anita Kaul
anitakaul@gmail.com

[1] Apollo Cradle Hospital, Amritsar, Punjab, India

[2] Madhukar Rainbow Children's Hospital, New Delhi, India

[3] CK Birla Hospital for Women, Gurugram, India

[4] Maharaja Agrasen Hospital, New Delhi, India

[5] Apollo Hospital for Fetal Medicine, Indraprastha Apollo Hospital, New Delhi, India

Introduction

Molar pregnancy is characterised histologically by abnormalities of chorionic villi that consist of trophoblastic proliferation and oedema of villious stroma. The absence or presence of a fetus or embryonic elements has been used to describe them as complete or partial mole [1]. We describe a case of complete hydatidiform mole with a healthy viable fetus with a diagnostic dilemma whether it comprises a dichorionic twin pregnancy with normal fetus and complete molar pregnancy or singleton pregnancy consisting of a triploid fetus with partial hydatidiform mole placenta. The incidence of a CHMCF is a rare occurrence of 1/22,000 to 1/100,000.

Case Report

A 35 year-old, G5P2A2, previous 2 LSCS, presented to the Fetal Medicine Centre at 15 weeks and 2 days gestation for a second opinion in view of suspected partial mole. The patient gave history of recurrent episodes of vaginal bleeding since conception. Her last pregnancy was 5 years ago. She did not give any history of intermenstrual

bleeding in the interpregnancy period and none of her previous pregnancies was a molar pregnancy. Ultrasound was done using a Voluson E8 scanner (GE Healthcare, Milwaukee, WI) equipped with a convex 4–8 MHz abdominal transducer and 6–12 MHz endovaginal probe. 2D ultrasound showed a single placental mass with one-fourth of its substance consisting of focal, cystic spaces which were avascular on Colour Doppler (Fig. 1). The adjoining fetus appeared structurally normal. There was no retroplacental collection of blood. Good fetal movements were seen on the ultrasound. The cervical length on TVS was 36 mm. The placenta was not low lying. Bilaterally normal ovaries were observed. The couple was counselled regarding the strong suspicion of partial mole and advised amniocentesis to rule out triploidy. However, the couple declined invasive testing and wanted to continue with expectant management. They were counselled regarding the high risk of gestational hypertension, preeclampsia, HELLP syndrome, intrauterine fetal growth restriction, intrauterine fetal demise and preterm delivery, both spontaneous and iatrogenic. Her first trimester biochemical screening was suggestive of raised β hCG levels of 10.2 MoMs. The high β hCG also poses a risk of hyperthyroidism. The fetus was followed up with serial growth scans which showed satisfactory interval growth, though all growth parameters were below the 5th centile for gestation. Amniotic fluid volume was normal. Maternal surveillance included monitoring for the complications mentioned above. The patient did not develop hypertension or hyperthyroidism. The patient presented at 36 weeks with decreased fetal movements and was delivered by lower segment caesarean section (LSCS) in view of fetal growth restriction, oligohydramnios, cerebral redistribution of fetal blood flow and previous 2 LSCS. A baby boy weighing 1600 grams was born with an Apgar score of 8, 9, and both mother and baby were discharged well after 4 days of hospital stay. Maternal β hCG began to show a declining trend and the baby was doing well 6 weeks postnatally. The placenta was sent for gross histopathology and confirmed complete mole (Fig. 2a, b). The microscopic evaluation was suggestive of abnormal placental part showing large distended and cystic dilated villi with odematous cores suggestive of hydatidiform mole.

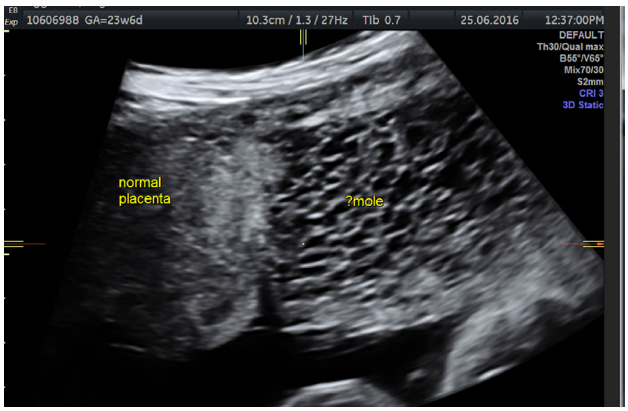

Fig. 1 A 2D USG image of a single placental mass with one-fourth of its substance consisting of focal and cystic spaces

Discussion

Twin pregnancy with an apparently healthy fetus and a hydatidiform mole are a rare obstetric finding. CHMCF (complete hydatidiform mole coexisting with a live twin fetus) has an estimated incidence of about 1/22,000 to 1/100,000 pregnancies [2]. Twin pregnancy with CHMCF resulting in a healthy take-home baby is rare, with the largest observational study of 177 cases documented in detail in literature state a live birth rate of 37% [3]. The approach to this case presented with a diagnostic dilemma to differentiate between two diagnoses: dichorionic twin pregnancy with normal fetus (46 chromosomes, 23 maternal and 23 paternal) and complete molar pregnancy (46 chromosomes, all paternal) and singleton pregnancy consisting of a triploid fetus with partial hydatidiform mole placenta (69 chromosomes, 23 maternal and 46 paternal). Non-specific symptoms associated with pregnancy such as vaginal bleeding raised the suspicion of a co-existing molar pregnancy. Others being hyperemesis gravidarum and hypertension or thyrotoxicosis which were not present.

Ultrasonography has made it possible to diagnose a hydatidiform mole and co-existent fetus in the first trimester however, there are limitations in sonographic diagnosis of a partial hydatidiform mole as the typical findings of a complete hydatidiform mole, such as a snowstorm appearance, are usually absent in a partial mole. Hence, only about 30% of partial moles are reported to be detected by sonography [4].

Serial monitoring of serum β-hCG level is a useful marker in the management of a CHMCF with its peak at the beginning of the second trimester of pregnancy, a decline thereafter is reassuring with successful pregnancy outcomes with a viable fetus.

Prenatal diagnosis by chorionic villus sampling, amniocentesis, or fetal cord blood sampling enables one to distinguish between diploid and triploid foetuses and DNA polymorphism analysis distinguishes between a partial mole and CHMCF [5]. Postnatal histopathology, as in our case confirmed the diagnosis of an androgenic complete mole with a characteristic appearance of a bunch of grapes with generalised trophoblastic proliferation constituting large distended and cystically dilated villi with odematous cores. Hence, this was a classical mole co-existing with a normal fetus and placenta with molar transformation of one ovum in a dizygotic dichorionic pregnancy.

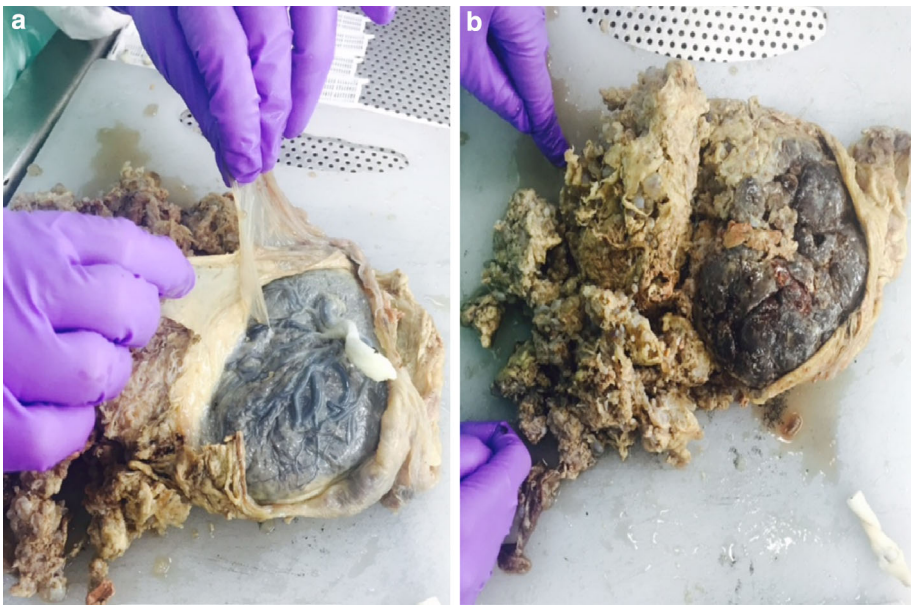

Fig. 2 **a**, **b** Gross histopathology demonstrating separate membranes suggestive of dichorionicity and demarcation between a normal looking quarter of the placenta with molar changes observed in the remaining part

As 25% of gestational trophoblastic neoplasia develop following an apparently normal pregnancy a weekly follow-up of the serum β-hCG level is recommended as the main indicator. Also, initial serum levels less than 400,000 mIU/mL is associated with a good outcome [6]. The management is not only to monitor for but to also optimise the increased risks of gestational hypertension, preeclampsia, HELLP syndrome, maternal hyperthyroidism, intrauterine fetal growth restriction, intrauterine fetal demise, preterm delivery, uterine rupture and the development of malignant neoplasia (trophoblastic tumor) which pose a grim prognosis and serve as a useful predictor of live birth.

As mentioned earlier, prenatal testing of at least fetal karyotype is essential in deciding continuation and prognosis of the pregnancy, however, it was declined by our patient. Nevertheless, the parents who choose to continue a pregnancy with suspected CHMCF were asked to agree to take the risk of possible maternal complications associated with molar pregnancy.

Conclusions

Twin pregnancy with a healthy fetus and a hydatidiform mole remains a challenging prenatal diagnosis as molar changes are progressive and less marked in the first trimester. Though our limitations remained due to decline of prenatal invasive testing, early ultrasonography and serial β hCG levels aided in the diagnosis and management of the case. Prenatal consultation included a thorough discussion of maternal and fetal risks while explaining a 40% chance of successful outcome [2]. Post-partum histopathology was confirmative of the diagnosis with β hCG surveillance to rule out persistent trophoblastic disease.

For this type of study formal consent is not required.

Compliance with Ethical Standards

Conflict of interest The authors declare that they have no conflict of interest.

Ethical Approval This article does not contain any studies with human participants or animals performed by any of the authors.

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