Complete hydatidiform mole with co-existing live fetus: A case report*

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ABSTRACT

The co-existence of a hydatidiform mole with a living fetus during the third trimester is extremely rare. The optimal management of such a case is controversial especially when medical and obstetric complications set in before term. The aim of management is towards avoidance of complications and planning the delivery at the most appropriate time to ensure good maternal and fetal outcome. We report the case of a 27-year-old Gravida 2 Para 1, who was diagnosed with a complete mole with co-existing live fetus at around 12 weeks age of gestation. She was referred to our institution at 31 weeks and 1 day age of gestation due to vaginal bleeding for which an emergency cesarean section was done. She delivered a live baby boy weighing 1.5 kg, with Apgar Score of 4,6,6. Chemoprophylaxis was administered and her serum beta human chorionic gonadotropin was monitored post-partum.

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Keywords: Molar pregnancy, Complete mole, hydatidiform Mole, Twin Pregnancy

INTRODUCTION

The co-existence of a hydatidiform mole with a normal living fetus is a rare obstetric phenomenon. Clinical assessment, detailed ultrasound examination and chromosome analysis are essential for prenatal diagnosis. The condition is a management dilemma due to the associated maternal and fetal complications. The problems in the management include the risks of fetal abnormality, development of severe maternal complications such as preeclampsia, hyperthyroidism, profuse vaginal bleeding, pregnancy failure, and preterm birth and increased risk for gestational trophoblastic neoplasia (GTN) after delivery. Fetal survival should always be weighed against the risk of complications associated with a molar pregnancy. Due to the rarity of the condition, there is no consensus on appropriate management of such pregnancies. We present the case of a complete mole with co-existing live fetus (CMCF) who was delivered at 31 weeks and 1 day age of gestation.

CASE REPORT

A 27-year-old gravida 2 para 1, at 31 weeks and 1 day age of gestation, was referred to our institution due to vaginal bleeding. Past medical and family medical histories were unremarkable. She is a non-smoker and not an alcoholic beverage drinker. She previously delivered to a live term baby girl who died after two weeks due to a congenital heart disease.

For this pregnancy, the patient experienced vaginal spotting at 12 weeks age of gestation. She consulted at a local hospital where ultrasonography showed findings suggestive of a twin pregnancy composed of a live fetus attached to a normal placenta and another placenta with multiple cystic structures suspicious of hydatidiform mole. She was prescribed Dydrogesterone and Nifedipine to control her symptom and was advised to go to a tertiary hospital for further evaluation and management. However, she failed to comply and instead continued to have her prenatal check-ups at their local health center. The vaginal spotting persisted until morning prior to consultation when she had sudden onset of profuse vaginal bleeding soaking one baby diaper. She consulted at their local hospital where a repeat ultrasound showed a complete molar pregnancy at the lower uterine segment with a co-existing fetus with a gestational age of 31 weeks and 4 days, in transverse lie at the upper uterine portion (Figure 1). She was subsequently referred to our institution for further evaluation and management.

On admission, her general and systemic findings were unremarkable. The uterus had a fundic height of 27 cm and the fetal heart rate was 130 beats per minute,

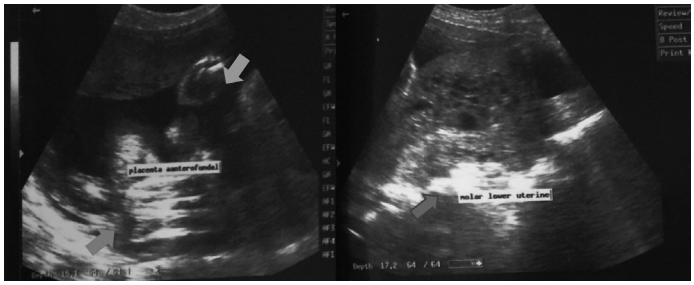

Figure 1. Ultrasound report on admission showing a live fetus (light gray arrow), hydatidiform mole (dark gray arrow) located at the lower uterine with a grape-like appearance, normal placenta (semi light gray arrow) located at the fundal area

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best heard at the left upper quadrant. Leopolds maneuver revealed that the baby was in breech presentation and was floating.

The initial plan at the emergency room was to control the preterm labor using Isoxuprine, which was later shifted to magnesium sulfate for neuroprotection. Dexamethasone was given for fetal lung maturation. Since the patient had an ultrasound result of a twin gestation composed of a live fetus and a co-existing hydatidiform mole, further investigation was warranted to confirm the diagnosis. Doppler and 4D ultrasound was done, which showed an echogenic uterine mass with multiple cystic structures measuring 12.8 x 8.3 x 5.4 cm located at the left posterolateral area overlying the cervical os (Figure 2). The fetus was in breech presentation with an estimated weight of 1,500 grams, which was appropriate for gestational age. The fetus was attached to a normal, grade II placenta implanted at the anterofundal area. There was adequate amniotic fluid volume. Findings were consistent with a complete mole with co-existing live fetus. Doppler flow indices showed normal values for umbilical and uterine arteries. Cervical length was of 3.2cm.

The patient's baseline serum beta human chorionic gonadotropin (β-hCG) was 97,879 mIU/ml. Her other laboratory examination results were essentially normal (Table 1). On the second hospital day, the patient had episodes of elevated blood pressure ranging from 130/90 mmHg to 140/90 mmHg. She also had profuse vaginal bleeding warranting an emergency cesarean section. A normal appearing male baby weighing 1.5 kg was delivered with Apgar scores of 4, 6, and 6 at 1, 3, and 5 minutes, respectively (Figure 3). The placenta and a separate mass with grape-like structures (Figures 4a and 4b) were removed manually. The placenta was disc shaped with a centrally attached, three vessel umbilical cord and looked grossly normal. Intraoperative and postoperative course of the patient were unremarkable. Repeat serum β-hCG on the fourth postoperative day was 7,480 mIU/mL. The infant was admitted to the newborn intensive care unit but expired after three days because of prematurity.

The pathological examination showed that the placental tissue was grossly normal measuring 17 x 14 x 4 cm and weighing 340g. The umbilical cord measured 40cm in length and 2 cm in maximum diameter. Macroscopically, the separate mass with grape-like structures measured 15 x 13 x 5 cm and weighed 300 grams. Close inspection revealed widespread diffuse vesicular formation with maximum diameter of 4 cm. Histological examination of the grape-like mass showed layers of degenerated, attenuated or hyperplastic sheets of trophoblasts with mild to moderate atypia, the cores of the villi showed 'cistern' formation and vessels were absent. These findings were compatible with a complete hydatidiform mole (Figure 5).

The patient received methotrexate as chemoprophylaxis

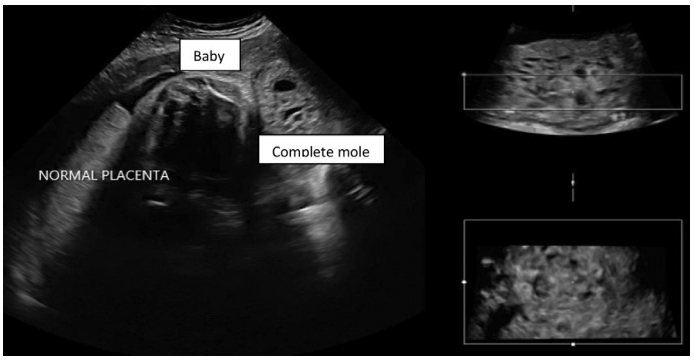

Figure 2. 4D ultrasound showing complete mole with co-existing live fetus. The complete mole occupied the posterior uterus with an echogenic uterine mass located at the left posterolateral area with multiple cystic structure situated over the cervical os

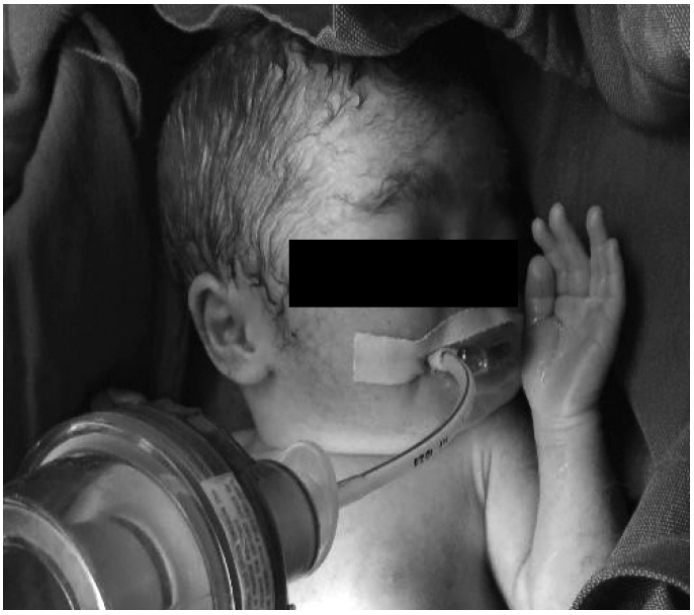

Figure 3. Picture of the normal, live baby

postdelivery. The serum β-hCG titer had a normal regression curve (Figure 6). At present the patient is well, using oral contraception and still has regular follow-up.

CASE DISCUSSION

Twin pregnancy consisting of a complete hydatidiform mole with a co-existing live fetus is rare, with an incidence of 1/22,000 to 1/100,000.[1,2,3,4] Theoretically, there are two possible conditions: a partial mole with an abnormal triploid fetus, and a complete mole co-existing with a normal fetus and placenta.[3,5]

Hydatidiform mole is classified into two entities with respect to cytogenetics, histopathology and morphology: 1) complete, classical mole, which has a diploid karyotype, no embryo and amnion, and uniform changes of the placental villi and trophoblasts; 2) partial mole which usually has a triploid karyotype, presence of an ascertainable embryo, umbilical cord or an amniotic

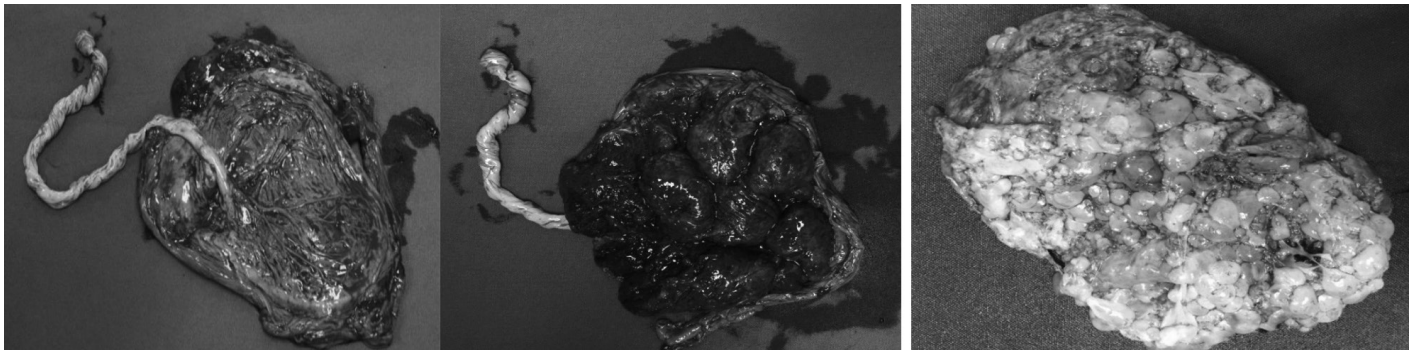

Figure 4. Gross appearance of the normal placenta with umbilical cord (4a) and the hydatidiform mole (4b) with grape-like tissues

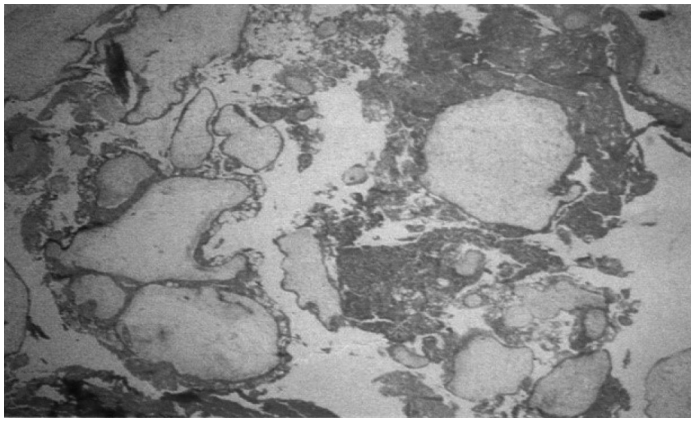

Figure 5. Photomicrograph of the complete hydatidiform mole. This is a low power view showing numerous edematous villi with frequent cistern formation and severe trophoblastic proliferation

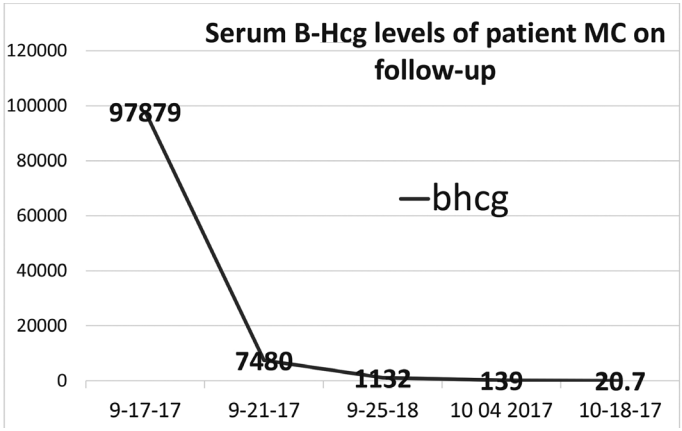

Figure 6. hCG regression curve of the patient postpartum

membrane, and only focal changes of placental villi and trophoblasts.[4,6] A hydatidiform mole with a co-existing fetus can be explained by the partial mole syndrome or by a twin pregnancy where the other conceptus has degenerated into a mole.[5] In the present case, although fetal karyotyping was not done, the histopathological and morphological findings of a normal placenta and separate molar tissues with marked hydropic, avascular villi support the possibility that it resulted from a dizygotic pregnancy in which one of the twins is a normal fetus while the other developed into a complete mole.

Table 1. Laboratory result of the patient

	RESULT	REFERENCE RANGE
SGPT	16.34 U/L	14-63.0
Creatinine	50.5umol/L	39-113.0
Potassium	3.7umol/L	3.5-5.1L
Sodium	135.7umol/L	136-144

Complete blood count	9/17/18	9/21/18
WBC	27.69 x 10^3 /uL	15.20
Hemoglobin	124.0 g/dL	112.0
Hematocrit	0.37	0.34
RBC	4.42 x 10 ^3 /uL	3.88
DIFFERENTIAL		
NEUTROPHIL	88	74
LYMPHOCYTE	8.0	18
MONOCYTE	3	5.0
EOSINOPHIL	0	2
BASOPHIL	1	1
PLATELET	374 x 10 ^3 /uL	331
MCV	83.9 fl	28.9
MCH	28.1 pg	32.9
MCHC	33.4g/dL	87.6

Prenatal diagnosis of a co-existent mole and fetus depend upon the clinical signs and symptoms, physical examination, sonographic findings, and abnormal biochemical data.[1] Clinically, the patient may present with hyperemesis, hyperthyroidism, vaginal spotting or bleeding, pregnancy-induced hypertension and larger-than-gestational age uterus.[1,4,5] Vaginal bleeding remains to be the most common reason for admission to the hospital,[4,7] as in the case of our patient. A variety of ancillary techniques such as Immunohistochemical assessment with p57 and molecular genotyping have been developed to improve the diagnosis.[5] However, we do not have these techniques in our institution.

The difference between a partial and complete mole cannot be firmly established by ultrasound because both

present with the same vesicular pattern.[4,6] Commonly, a partial mole, presents with a malformed fetus attached to a cystic placenta However, in our case the ultrasound, both the 2-dimentsional and 4-dimentional, clearly revealed not only a normal appearing fetus connected to a normal placenta but also the presence of a well delineated endometrial mass with cystic spaces indicative of a hydatidiform mole. Because of this, a molar pregnancy with a concurrent normal, live fetus was diagnosed prenatally.

Ultrasonography can also aid in differentiating a complete mole with a coexisting fetus from other pathologies. One consideration is a chorangioma, which is a focal vascular placental lesion associated with a normal diploid fetus. Differentiation is made by its sonographic feature which shows a well circumscribed lesion with different echo patterns from the rest of the placenta. The chorangiona can be located on the fetal placental surface or protruding into the amniotic cavity. Increased vascularity or a large feeding vessel inside the tumor with the same pulsation rate as in the umbilical cord is likewise seen, a finding that is absent in molar gestation.[6]

Serum levels of β-hCG are usually high at the time of admission, although it should be kept in mind that β-hCG level is expected to be higher in multiple gestations. A high level of β-hCG at the time of admission may be an indication of poor prognosis of the disease.[4] The β-hCG of the patient at the time of admission was 97,879 miU/ml, which was relatively low for a complete hydatidiform mole that has reached 31 weeks age of gestation.

It is important to distinguish between a partial mole and a complete mole with a co-existent fetus because management differs. In partial hydatidiform mole, the fetus has multiple congenital anomalies that are not compatible with life. As such, the fetus commonly dies early in gestation and termination of pregnancy is done. On the other hand, a twin pregnancy with a complete hydatidiform mole and a normal live fetus is associated with increased risk for maternal complications. In most of these cases, the pregnancy is terminated due to complications such as vaginal bleeding, pre-eclampsia and hyperthyroidism. Some recommend immediate termination even in the absence of any complications.[2,3] Others recommend expectant management and delivery done only in the presence of medical and obstetric indications. In this case, close maternal and fetal surveillance is imperative for an optimal outcome. Counselling is likewise important since continuing the pregnancy poses an increased risk to both the mother and the fetus. A complete discussion of maternal and fetal risks, the need for chemoprophylaxis after delivery and even the possibility of a hysterectomy needs to be done. For our index patient, termination of pregnancy was due to vaginal bleeding.

Chemoprophylaxis is advised among patients with a complete mole with a twin live fetus due to an increased tendency to progress to gestational trophoblastic neoplasia.[1,4,5,7] It is also necessary to have regular β-hCG surveillance during the postnatal period for the early detection of post molar GTN.[2]

SUMMARY

Molar pregnancy with a co-existing normal, live fetus is a rare occurrence. In such cases, delivery of a viable, healthy infant is an even more rare occurrence. Diagnosis is achieved with the help of a thorough ultrasound examination coupled with chromosomal evaluation. Prenatal consultation should include discussion of maternal and fetal risks, the need for chemoprophylaxis, the possibility of a hysterectomy and the need for regular post-operative monitoring to detect malignant degeneration. In most of these cases, the pregnancy is terminated due profuse vaginal bleeding or associated medical complications. Due to the rarity of the condition, the management of complete mole with a co-existing live fetus remains controversial. In the present case the decision to terminate the pregnancy was due to the profuse vaginal bleeding while she was admitted at the high-risk unit. ■

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