Complete hydatidiform mole and coexisting normal fetus (CHMCF) pregnancies are rare and can be life-threatening to the mother. Definitive diagnosis can be made with chorionic villus sampling or amniocentesis. However, invasive procedures carry a risk of bleeding. We present the case of a twin molar pregnancy where a cell-free DNA screening test was utilized to evaluate for CHMCF pregnancy.

**Case** A patient presented at 15-week gestational age with suspected CHMCF pregnancy. Ultrasound revealed a normal-appearing pregnancy abutting a multicystic lesion concerning for a complete mole. Cell-free DNA was obtained and was suggestive of complete paternal uniparental disomy. Pathological evaluation of the products of conception confirmed the diagnosis of CHMCF.

**Conclusion** In atypical cases, cell-free DNA may be useful in evaluation of molar pregnancy.
for complete paternal uniparental disomy, most consistent with a complete molar pregnancy.

Findings of the cell-free DNA in combination with the typical ultrasound findings and high β-HCG were communicated to the patient as highly suspicious for a complete molar pregnancy with coexisting normal pregnancy. An approximate 30% chance of a live birth with the possibility of major morbidity and mortality were discussed as concerns with continuing the pregnancy. The patient chose to terminate the pregnancy at 17 weeks of gestation with dilation and evacuation (D&E). She underwent cervical preparation which included a paracervical block of 20-cc 1% lidocaine prior to laminaria placement. She was admitted for overnight observation to monitor vaginal bleeding. A D&E was performed in standard fashion under ultrasound guidance. She received a paracervical block with 20-cc 1% lidocaine with 8 units of vasopressin prior to the procedure and prophylactic oxytocin following the procedure. Estimated blood loss was 75 cc. Histologic evaluation revealed hydropic chorionic villi with trophoblastic hyperplasia consistent with complete molar pregnancy and fragments of well-developed fetal tissue, consistent with CHMCF.

**Discussion**

CHMCF pregnancies occur in 1:22,000 to 1:100,000 pregnancies. The diagnosis of CHMCF is suspected by identifying two separate concepts: (1) a fetus with normal placentation and (2) an adjacent molar gestation separated by a membrane. The molar component has a characteristic vesicular sonographic pattern. Complete moles are diploid and the chromosomes are derived only from paternal genome. Differential diagnosis of CHMCF includes partial hydatidiform mole, twin pregnancy with a partial hydatidiform mole and coexisting normal fetus, and placentals mesenchymal dysplasia. Placental mesenchymal dysplasia is a benign placental vascular anomaly that is difficult to distinguish from CHMCF and is not an indication for termination of pregnancy. Differentiating these entities using ultrasound alone can be difficult. In addition, molar change is a progressive phenomenon and hydatidiform changes are often less prominent in the first trimester.

We employed cell-free DNA in this case of CHMCF to assist in patient counseling and management. Cell-free DNA screening was performed with single-nucleotide polymorphism (SNP) sequencing of maternal blood. The test is widely utilized for screening for single chromosomal aneuploidy. A case report demonstrated the use of cell-free DNA to determine the origin of the genome in a choriocarcinoma in a woman with a choriocarcinoma with coexisting normal fetus. And now, we suggest that cell-free DNA may be useful for screening in cases where CHMCF is suspected. The potential for cell-free DNA screening to differentiate a complete hydatidiform mole from a partial hydatidiform mole or placentals mesenchymal dysplasia deserves investigation in a larger patient cohort to assess accuracy of the test for this purpose. Its use may complement findings noted on ultrasound.

Complications of CHMCF are similar to molar pregnancy alone, and include but are not limited to thyrotoxicosis, hyperemesis gravidarum, preeclampsia, intrauterine fetal demise, preivable premature rupture of membranes, preterm delivery, obstetric hemorrhage, coagulopathy, gestational trophoblastic neoplasia (GTN), and trophoblastic embolization. GTN occurs in a significant number of women. The high risk of maternal complications without accurate predictors of which patients will progress to a live birth makes the decision regarding termination or continuation of pregnancy a difficult one for patients. Termination of pregnancy should be offered and it minimizes maternal risk. Careful continuation of pregnancy with close follow-up is possible in some patients.

In this case, the patient presented with an abnormal ultrasound. Though there is a high sensitivity and specificity of ultrasound for the diagnosis of molar pregnancy, laboratory adjuncts are potentially of value in differentiating molar pregnancies from placental mesenchymal dysplasia. In this case, the pregnancy's appearance on ultrasound suggested CHMCF and the results of Cell-free DNA screening were used as part of the risk assessment. The overwhelming amount of paternal DNA detected in the maternal circulation was identified by a Cell-free DNA screening test and suggested the
presence of a complete molar pregnancy. The patient experi-
enced hyperemesis gravidarum and bleeding complications
and ultimately decided not to continue the pregnancy because
of the associated risks. The patient had an uneventful dilation
and evacuation and continues surveillance for persistent
 trophoblastic disease. We found the use of cell-free DNA was
helpful in her care and believe that test validation in more
pregnancies would be beneficial in the management of
CHMCF. Additionally, the use of cell-free DNA for follow-up
surveillance would be of interest.

Key Points

1. Complete hydatidiform mole and coexisting normal fetus
(CHMCF) is a rare and life-threatening condition.
2. Cell-free DNA testing may be of use to increase the
certainty of molar pregnancy diagnoses.

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Conflict of Interest
None declared.

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