Early Detection by Ultrasound of Partial Hydatidiform Mole With a Coexistent Live Fetus

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ABSTRACT

Introduction: Twin pregnancy with hydatidiform mole and coexistent live fetus is a rare condition with severe maternal and fetal complications such as preeclampsia, vaginal bleeding, persistent gestational trophoblastic tumor, and fetal death.

Case Presentation: We report a case of a twin pregnancy with histopathologically proven hydatidiform mole and a coexistent live fetus in a 30-year-old Indian woman diagnosed by first trimester ultrasound.

Discussion: Our case emphasizes the role of ultrasound in diagnosing this condition in early pregnancy. A succinct overview of etiological mechanisms, possible complications, and clinical management is provided.

Conclusion: Ultrasound is an effective diagnostic tool to diagnose hydatidiform mole with coexistent live fetus. Early diagnosis of this condition is important for risk stratification and facilitates an informed decision by the patient whether to terminate the pregnancy or to continue until full term with close monitoring after delivery.

INTRODUCTION

Twin pregnancy with hydatidiform mole and a coexistent live fetus is a rare occurrence, with a prevalence of 1 in 20,000 to 100,000 pregnancies.[1,2] A molar pregnancy can be either complete or partial. The majority are a complete hydatidiform mole coexisting with a normal fetus and placenta. Twin pregnancies with partial hydatidiform mole coexisting with a live fetus are rarely reported, since fetuses identified with partial mole are generally triploid and tend to die in the first trimester.[1,3,4] Hydatidiform mole with live fetus is associated with the risk of many maternal and fetal complications such as vaginal bleeding, preeclampsia, thromboembolic disease, hyperemesis, hyperthyroidism, fetal intrauterine demise, and a potentially lethal risk of gestational trophoblastic neoplasia.[5,6] Management of such cases presents a dilemma to both the clinician as well as the patient. The possible risks that may ensue upon continuation of pregnancy must be weighed against the chances of fetal survival so as to facilitate a well-informed decision by the patient to opt between continuation and termination of pregnancy. An early diagnosis of this condition by high resolution ultrasound is important for clinical management and helps the patient in making a decision whether to terminate pregnancy or continue with close feto-maternal monitoring. We report a histopathologically confirmed case of partial hydatidiform mole with coexistent live fetus, diagnosed on a routine first trimester ultrasound.

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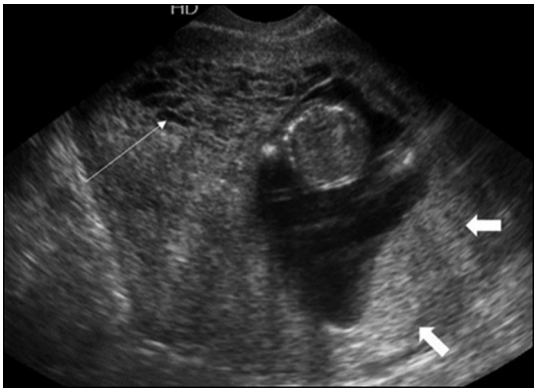
CASE PRESENTATION

A 30-year-old woman (gravida 2, para 1) of Indian origin with a nonconsanguineous conception came for a routine first trimester ultrasound examination at a gestation age of 13 weeks 3 days by last menstrual period. The first child was a healthy 5-year-old girl from an uneventful, normal full-term gestation. The present conception resulted from ovulation induction with a 5-day course of clomiphene citrate. The patient reported 2 episodes of vaginal bleeding and 1 episode of passing white, ball-like structures in the present pregnancy.

Transvaginal ultrasound scanning (Philips HD7; 4-8MHz) revealed a dichorionic diamniotic twin pregnancy. One sac con-

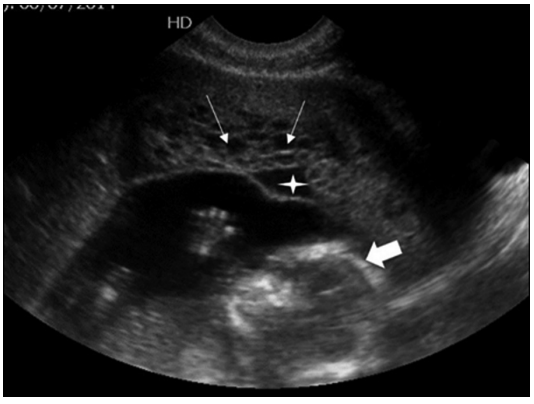
tained sonographically normal live fetus matching the gestational age with unremarkable placenta. The placenta attached to the fetus was examined carefully and found to be normal. Amniotic fluid volume was appropriate. The other sac had an abnormally thickened placenta showing multifocal cystic spaces with no identifiable fetus or umbilical cord (Figures 1 and 2). Doppler evaluation of cystic spaces in the abnormal placenta revealed no significant vascularity. The maternal ovaries were normal. Clinical correlation with serum β-human chorionic gonadotropin (β-hCG) revealed an alarmingly elevated level of 180,000 mIU/ml. A tentative diagnosis of twin gestation with complete molar pregnancy and coexistent fetus was made based on sonographic findings, raised β-hCG level, and clinical history. A fetus in partial mole tends to die early in pregnancy.[3,4] An option of amniocentesis was offered to the patient to confirm the diploid status of the fetus, but the patient refused. The couple was counseled about the possible maternal and fetal risks of this condition, including a potential risk for onset of malignant change termed persistent gestational trophoblastic disease. The patient opted for termination of the pregnancy rather than expectant management. A stillborn male fetus of 90mm length and placenta appropriate for gestation age were delivered upon medical termination of the pregnancy. These were grossly and histopathologically normal. Another placenta with grey-white soft tissue fragments was delivered. On histopathological examination, it demonstrated dilated villi with edema and focal scalloping. The villi were lined by attenuated trophoblasts. These features were compatible with a diagnosis of partial hydatidiform mole (Figure 3). No fetal parts, however, were found along with partial hydatidiform molar placenta. Following termination, close β-hCG surveillance of the mother was done daily, which revealed rapid regression to 7,700 mIU/ml on the third postpartum day. Weekly serum β-hCG surveillance was continued therafter, which showed decreasing levels with insignificant value (0.87mIU/ml) reached in 8 weeks.

Figure 1. Ultrasound Scan of Dichorionic Twin Pregnancy



Transvaginal ultrasound (Philips HD7; 4-8MHz) shows abnormal placenta in 1 sac (thin arrow, left) and twin sac with a live fetus and normal placenta (bold arrows, right) posterior to it.

Figure 2. Ultrasound Scan, Enlarged View



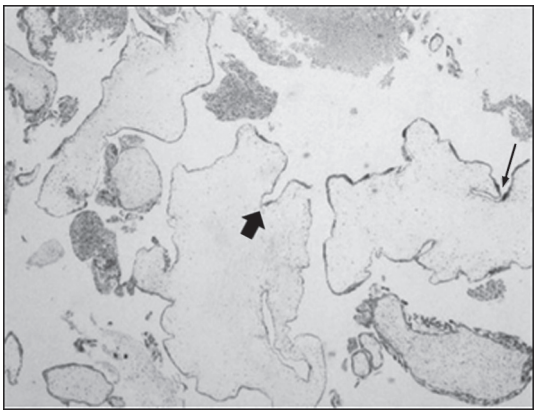
Transvaginal ultrasound scan (Philips HD7; 4-8MHz) with enlarged view of one sac showing abnormal placenta with multifocal sonolucencies (thin arrows) and amniotic fluid (marked by 4-point star). Twin sac revealed a live fetus (bold arrow).

DISCUSSION

Coexistence of molar pregnancy with a twin live fetus is a rare, complex clinical condition. Complete molar pregnancies show complete lack of fetal parts, while partial molar pregnancies show fetus or indication of previous fetal existence. The majority of them are complete hydatidiform mole coexisting with a normal fetus and placenta, with nearly 200 cases documented in literature to 2007.[1,6] Twin pregnancies with partial hydatidiform mole coexisting with a live fetus are rarely reported, since fetuses identified with partial mole are generally triploid and tend to die in the first trimester.[1,3] The possible scenarios of a molar pregnancy coexisting with a fetus in a twin gestation are (1) complete molar gestation with a coexistent normal diploid fetus in a dizygotic twin pregnancy, (2) partial hydatidiform mole with a coexistent normal diploid fetus in a dizygotic twin pregnancy, and (3) partial hydatidiform mole and a coexistent abnormal triploid fetus in a monozygotic twin pregnancy.

In this case, ultrasound demonstrated a normal fetus in one sac and an abnormal, thickened, multicystic placenta without any fetus in the twin sac. Histopathological examination after termination of pregnancy demonstrated a partial molar placenta with no evidence of a fetus or fetal parts noted in the twin sac containing the abnormal placenta. Lack of fetal parts in our case was an interesting finding since a fetus or fetal parts are usually, though not always, present in cases of partial mole.[7] In cases of complete mole, the abnormal placenta has a characteristic "snowstorm" appearance showing multiple sonolucencies sized 1 mm to 30 mm with a normal coexistent twin fetus.[4,8] Detectability of partial mole by ultrasound is only 30%, since partial mole lacks

Figure 3. Molar Placenta Tissue



Microphotograph showing dilated villi lined by attenuated trophoblastic cells. The villi have scalloped borders (bold arrow, left) and stromal inclusions (thin arrow, right). H&E stain, x40 magnification.

the typical "snowstorm" appearance. Partial mole shows heterogenous placenta with multicystic spaces and interspersed solid areas. Color Doppler may show the presence of high-velocity, low impedance flow in the multicystic placental mass.[4,9] The differential diagnoses of such a sonographic appearance are placental mesenchymal dysplasia, choriocarcinoma, and placental hematoma. Placental mesenchymal dysplasia is a recently recognized, rare placental vascular anomaly characterized by placentomegaly and grape-like vesicles appearing as hypoechoic areas on sonography.[9] Confirmation of diagnosis is based on histopathological examination. Choriocarcinoma is differentiated on sonography with increased vascularity or a large feeding vessel inside the tumor.[9] Placenta hematoma may sometimes resemble partial mole with its partially solid, partially cystic sonographic appearance. Because of their similar sonographic appearances, the differentiation between these diagnoses may be difficult to make antenatally and often can be made successfully only in the postpartum/termination period, as was in our case. Confirmation of partial molar placenta is done histopathologically depicting organization of villous tissue with hyperplastic surface trophoblasts and irregular outline of villi leading to stromal inclusions.[5] A prior molar pregnancy increases the risk of recurrence up to 2%.[10]

Other etiological risk factors for partial mole are repeated past abortions, irregular cycles, and maternal age of more than 40 years.[5,11] Incidence of partial mole is higher in Asia than North America and Europe, being highest in India.[5,10,11] Common clinical presentations of molar pregnancies are vaginal bleeding in the first or second trimester, unusually elevated serum β-hCG levels, and early development of preeclampsia.[4] There is a suggestion in recent literature of increased incidence of twin pregnancies with mole and coexistent fetus in patients using drugs for ovulation induction for fertility, as in our case. However, ovulation induction drugs are yet to be established as a potential risk factor due to a limited number of reports available up to date.[12] In view of associated high complications, molar pregnancy with live fetus has complex clinical considerations in a strongly desired pregnancy. Termination of pregnancy is generally recommended in a severely malformed fetus.[5,8] In sonographically normal fetuses, the prenatal diagnosis of fetal karyotype can be attempted by chorionic villus sampling, amniocentesis, or fetal cord blood sampling.[1] Women should be counseled about the risk of both the maternal complications, including preeclampsia, thromboembolic disease, hyperemesis, and hyperthyroidism and fetal complications including spontaneous miscarriage, intrauterine death, and preterm labor.[5,6,8] A potentially fatal complication of molar pregnancies is the risk of persistent gestational trophoblastic disease (pGTD).[2,6,10] The incidence of pGTD after surgical evacuation in complete hydatidiform mole with fetus is considerably higher as compared to partial hydatidiform mole with fetus (10-28% vs 3-5%).[13] The risk of pGTD remains unchanged whether the patient opts for early termination of pregnancy or chooses to continue the pregnancy. Thus, expectant management may be permitted in cases of a complete molar pregnancy with coexisting fetus, which has up to a 40% chance of a viable live birth.[2,6,8] If the fetus is euploid and sonographically normal and the mother is clinically well, the pregnancy can be continued under careful perinatal monitoring.

Expectant mothers with a twin and partial mole are difficult to counsel due to the paucity of available literature. In our case, the patient did not consent to prenatal karyotyping and chose to terminate the pregnancy, in view of the risk of severe maternal and fetal complications. In cases where continuation of pregnancy is desired, guidelines for patient management should include serial serum β-hCG levels and a chest x-ray to screen for metastases for gestational trophoblastic neoplasia.[14] Post evacuation, intense maternal follow-up is required by (1) β-hCG assays to confirm its return to baseline values (<5 mIU/mL), and (2) sonographic evaluation and measurement of endometrial thickness.[10,13-15] Serial quantitative serum β-hCG assay is recommended within 48 hours of evacuation, every 1 to 2 weeks while elevated, and monthly for 6 months after the β-hCG levels return to normal.[14,15] Treatment for pGTD with chemotherapy is indicated in cases of persistently raised β-hCG at 6 months after evacuation or with histopathological diagnosis of choriocarcinoma.[10]

CONCLUSION

It is clinically challenging to diagnose a molar placenta in an ongoing pregnancy. Early diagnosis depends on a combination of detecting an unusually high level of β-hCG and abnormalities detected on ultrasound examination. This case emphasizes the importance of ultrasound as a good diagnostic tool in cases of twin pregnancy with a molar and a coexistent live fetus. Ultrasound

alone is insufficient for diagnosing such cases but serves to raise the possibility of this condition. Partial molar pregnancies generally present in the second trimester and manifest with complications such as vaginal bleeding or spontaneous abortion. In our case, however, the patient was relatively asymptomatic at presentation and the condition was diagnosed much earlier on a routine first trimester ultrasound scan. The radiologist should be aware of the sonographic manifestations and differential diagnosis of this condition to ensure appropriate management. Careful monitoring of these patients in the postpartum period and close monitoring of subsequent pregnancies to rule out the recurrence of gestational trophoblastic disease is of utmost importance.

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