Living fetus without congenital malformation in a singleton partial hydatidiform molar pregnancy: a case report and review of the literature

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ABSTRACT
A 36 years old lady gravida 2 para 1, came to our emergency ward at 9:30 pm on 12th June 2010 with complains of bleeding p/v for 2 hours with history of bleeding at 12 weeks. On examination she was anemic and uterus was 32 weeks size with good fetal heart sound. Bleeding was coming through os, on p/s examination. Ultrasonography showed a single live fetus of 1033 grams and thickened placenta (79 mm thickness) showing multiple cystic lesion with peripheral hypervascularity, giving an impression of partial hydatidiform mole. She was managed conservatively and delivered a live preterm male child of 960 grams on 13th June 2010 evening without any congenital malformation. Placental weight was 1800 grams with multiple small vesicles. Now the boy is 4 years old and going to school with normal developmental milestone.

Keywords: Thickened placenta, Partial hydatidiform mole, Congenital malformation, Developmental milestone

INTRODUCTION
Hydatidiform mole is characterized by abnormal fetoplacental development and trophoblastic hyperplasia due to excessive paternally derived genetic material. Partial mole with living fetus without any congenital anomaly or chromosomal stigma is a very rare entity. Singleton living fetus with partial mole is still rarer. Such an occurrence has been found only few times in extensively searched medical literature. Here we are reporting a rare case of partial mole with live fetus and normal developmental milestone over four years of follow up along with its review of literatures.

CASE REPORT
A lady 36 years old G2P1L1 at 29 weeks 4 days of gestational age came to our emergency ward at 9.30 pm on 12th June 2010 with complains of vaginal bleeding for 2 hours. She was having a living issue of 5 year old female child, delivered vaginally at term. She had vaginal bleeding at 12 weeks of gestation for which she was managed conservatively at hospital.

On examination, she was moderately anemic with bilateral pedal edema. Her pulse was 90/min and BP was 130/90 mm Hg. P/A examination - uterus was 32 weeks, relaxed, non-tender and FHR was 142/min. On inspection of vulva, bleeding present and on per speculum examination bleeding was coming through os. Per vaginum examination was not done. She had a sonography report with her, showing a single live fetus of 25 weeks gestation age, placenta anterior and the lowest margin was 30mm away from internal os.

Here we did an USG which showed a single live fetus of 27 weeks 5 days gestational age, AFI - 6.3 cm and EFW 1033 gm. Placenta is anterior with lowest margin is 37 mm away from the closed os. Placental thickness was 79 mm, showing multiple cystic lesions with peripheral hypervascularity giving patches of honeycomb pattern. No placental separation seen.

Doppler study of umbilical artery and middle cerebral artery were within normal limit S/O partial mole with living fetus. Serum beta hCG - 315 miu/ml and hemoglobin was 8.8 gm%. There was dilemma concerning her mode of delivery. She was on conservative management with injection corticosteroid as well as injection tranexamic acid.

After completion of steroid dose, her per abdominal finding was- uterus 32 week size, cephalic presentation, contraction - 1-2/10-20 sec/10min, FHR - 148/min, per vaginal examination finding was - cervix fully effaced, os 4 cm dilated, bag of membrane present and vertex station was (-2).

After augmentation with oxytocin, she delivered a live preterm male child with APGAR score 2/10 and 6/10 at 1 min and 5 min respectively at 6.50 pm on 13th June 2010. Placental weight was 1.8 kg and baby weight was 960 grams. Baby was referred to special neonatal care unit for better management. After 30 days of admission, baby was discharged with weight of 1.5 kg.

Histopathologic reports revealed focal trophoblastic hyperplasia mainly involving syncitiotrophoblast, enlarged villi, and irregular villi with scalloped borders with trophoblast inclusions, suggestive of partial mole. The lady was followed up with serum beta hCG till one year and the baby is being followed up till now (4 years), baby is doing well with normal developmental milestone.
DISCUSSION

The above case of partial hydatidiform mole with singleton living fetus represents the rarity in obstetric world. Baby’s Apgar score was low due to antepartum hemorrhage which was managed by immediate NICU admission and proper intervention. Hsieh CC, et al.2 classified molar pregnancy into complete mole when there is total replacement of normal placenta by grossly dilated and hydropic villi in the absence of fetus and the incomplete or partial mole showing partial replacement with hydropic villi and visible abnormal fetal parts leading to termination of the pregnancy in first trimester. Dolapcioglu K et al.1 reported that fetus in such cases is rarely alive at the time of diagnosis and often shows gross congenital anomalies associated with triploidy and frequently have a grave prognosis and owing to limited functional placenta circulation and severe intrauterine fetal growth retardation. Coexisting molar pregnancy with single normal live term fetus was seen in some other cases too.1-5 Such an association has been divided into three types. The first and most common is a twin pregnancy with one normal fetus having a normal placenta and another complete mole, second type is a twin pregnancy with normal fetus and placenta and another partial mole and the third and most uncommon occurrence is a singleton normal fetus with partial molar placenta.1 The third type of molar pregnancy has been reported only seven times in extensively searched medical literature. Such a fetus should have a normal karyotype to survive to term, though placenta may have variation, from diploidy of the amnion to triploidy of the chorionic villi.1 However placenta in a partial mole with fetus in a singleton pregnancy results from dispermny and has a triploid karyotype in most cases. The complications of coexisting fetus with molar pregnancy include bleeding, persistent gestational trophoblastic disease, preterm labor, late abortion, and severe anemia in the fetus. The case we presented had preterm labor. The neonate was low for gestational age with low APGAR score.

Zhang P et al.6 from University of California San Diego reported partial molar pregnancy with dead fetus in utero at 26 weeks. Zahida P et al. in Pakistan,7 reported G4P1+2 lady with partial hydatidiform mole along with alive baby managed conservatively 18 weeks onwards and delivered successfully at term. Bruchim I et al.8 reported a case of lady with partial mole at 41 weeks of gestation and another at 26 weeks with complete hydatidiform mole along with twin fetuses. Tamrakar et al. (2011)9 reported a case of preterm gestation along with partial hydatidiform mole and a live fetus. Such patients have risk of developing persistent gestational trophoblastic disease. Though some authors questioned the follow up of patient with partial hydatidiform mole by serum β-hCG, such patients should be followed up at regular interval maximum for one year. Patient with molar pregnancy can develop choriocarcinoma and mortality has been reported by Seckl et al.10 However partial hydatidiform mole rarely requires chemotherapy. Several factors influence the outcome of the fetus in partial molar pregnancy most important being karyotype of the fetus. The present case had no obvious congenital anomaly. Other factors include the size of the molar placenta, the speed of molar degeneration and fetal anemia. The cases we report probably had sufficient placental circulation to sustain through the first and second trimester. The problems in the management of molar pregnancy and a live fetus involve the risks of fetal abnormality, malignant trophoblastic change, and severe maternal complications such as preeclampsia, thyrototoxicosis, heavy bleeding, pregnancy failure, and preterm birth. Termination of pregnancy might be required due to these complications. Amniocentesis should be done for karyotyping. Prognosis of partial mole is usually better than the complete mole as only few cases of partial moles progress to persistent trophoblastic disease. However, the nature and the risks of diploid partial moles are not well established and they seem to be a distinct clinical entity. With close antenatal surveillance, our patient delivered a healthy male infant without any complications. We also performed intensive maternal follow-up with serum beta hCG in the postpartum period for the risk of persistent trophoblastic disease but the patient did not show any evidence of persistent trophoblastic disease within 12th months.

Our case had a focal molar change allowing fetal survival until term. Extensive sampling of the placenta allowed us to visualize the normal villi. The genetic makeup leading to multiple congenital anomalies as well as the compromised blood supply lead to the diminished fetal survival. Normal fetal outcome is therefore barely known in this condition. Antenatal detection of molar pregnancy coexisting with a viable fetus should warrant genetic analysis and search for gross malformation of the fetus by ultrasound. Management of the pregnancy in such rare conditions should be determined on one-to-one basis and the possibility of increased complications should be discussed with the family and prognosis should be explained. In the present case, there must be mitotic abnormalities in the early post-fertilization period and represents placental mosaicism. Since the fetus was normal at birth and the child continues to be growing normally, the abnormal cell population appears to be confined to the placenta.

CONCLUSION

To conclude pregnancies with normal live fetus coexistent and partial molar placenta is extremely rare. In absence of gross fetal abnormalities on sonography, we recommend to continue the pregnancy as long as maternal complications are absent or controllable. Complete evaluation of the placental tissue is important even in cases with normal fetal outcome as focal molar changes which might be unsuspected during antenatal period, may affect the future obstetrical outcome.
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