

Partial Hydatidiform Mole with Alive Term IUGR Foetus

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Summary:

Gestational Trophoblastic diseases consist of a broad spectrum of conditions ranging from an uncomplicated partial hydatidiform molar pregnancy to stage-IV choriocarcinoma with cerebral metastasis. Incidence of hydatidiform mole with a co-existing live fetus varies between 0.005 to 0.01 % of all pregnancies. We report a case of partial molar pregnancy with alive term IUGR (intrauterine growth retardation) foetus. Diagnosis was made by sonographic findings of molar changes at her 28 weeks of gestation.

Introduction:

Gestational Trophoblastic diseases encompass a diverse group of lesion with specific pathogenesis, morphological characteristics and clinical features. The modified world health organization classification of Gestational Trophoblastic disease includes complete and partial hydatidiform mole, invasive mole, choriocarcinoma, placental site trophoblastic tumor, epitheloid, trophoblastic tumor, exaggerated placental site and placental site nodules¹ The Hydatidiform mole is characterized histologically by abnormalities of chorionic villi, trophoblastic proliferation and edema of villous stroma. Molar pregnancy is significantly more common in extreme age. The usual management of gestational trophoblastic diseases is evacuation of the uterus and follow up because of higher chances to develop choriocarcinoma². But some time when molar changes are in placenta along with alive foetus, expectant management can be performed under strict

Anomaly scan showed no fetal abnormality. At 40 weeks of pregnancy patient went to spontaneous labour and delivered a severely IUGR baby weighing 1.5 kg and it was morphologically normal. There was a single placenta; one third of it was replaced by molar tissue. As the singleton alive pregnancy with partial molar changes is extremely rare occurrence we reported the case here.

Keywords: Partial mole, IUGR, alive foetus.

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surveillance³. In each visit, history should be taken properly like hyperemesis, irregular per vaginal bleeding, sign of preeclampsia, features of thyrotoxicosis or any sign suggestive of metastasis. Patient is also advised to do serum β -hcg & USG for pregnancy profile. We described a case of singleton pregnancy, in which partial molar change was detected, however, the pregnancy ended in phenotypically normal term IUGR fetus.

Case Report

A 26 years old patient who was P-1+0, G-2nd admitted in the gynecological emergency at 40 weeks gestation with labour pain. Previously she had one supervised uncomplicated pregnancy. In this pregnancy she had antenatal care in primary health care facility. She didn't give any history of vaginal bleeding, excessive vomiting and other features suggestive of molar pregnancy. Obstetric USG at her 28 weeks of pregnancy showed single live intrauterine gestation with multiple cysts involving the part of the placenta suggestive of molar changes. The risk of continuation of pregnancy was explained to the patient and the relatives. Subsequently anomaly scan was done at 29 weeks of pregnancy which showed no obvious gross foetal anomaly but growth retarded foetus and oligohydramnios and molar changes in the placenta. As there was no preeclampsia, hyperthyroidism and any other complications and the no obvious gross anomaly so pregnancy was continued with advice to regular follow up. Serum β -hcg was not done antenatally due to lack of facility. Patient was on followed up upto 34 weeks of gestation thereafter she missed follow up. Later she came in hospital at 40 weeks pregnancy with labour pain. There was no specific past and family history of molar pregnancy. Labour was

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monitored with maintenance of partograph. After 6 hrs of labour pain, she delivered a male baby of 1.5 kg with good APGAR score. Active management of third stage of labour was done. Weight of the placenta was 600 gram and almost 1/3 of the placenta was replaced by molar tissue. Umbilical cord was normal, having 3 vessels.

The baby was examined by paediatrician and no gross congenital abnormality was detected. The baby was on breast feeding. Maternal serum β -hcg 24 hours after delivery was 300mIU/ml. Baby and the mother were discharged from the hospital on 2nd postnatal day. Histopathology of the placenta showed molar changes but chromosomal study of the placenta was not done.

The patient was followed up with serum β -hcg and chest x-ray after 15 days of delivery, both were normal. Again patient came after 6 weeks for postnatal check up, serum β -hcg was normal and both the mother and the baby were fine. Her repeated USG didn't show any features of myometrial invasion. Karyotype of the baby was done after 3 months and that was 46XY.

Discussion:

Hydatidiform mole can be separated into two entities with respect to cytogenetics, histopathology and morphology: firstly, complete, classical mole has diploid karyotype, no embryo and amnion and uniform changes of the placental villi and trophoblasts; secondly, partial mole usually has a triploid karyotype, resulting from the fertilization of a normal ovum with two sperms, the presence of ascertainable embryo, umbilical cord or an amniotic membrane and only focal changes of the placental villi and trophoblasts⁴. Histologically molar pregnancy is characterized by 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 absent⁵. Incidence of molar pregnancy varies from 1 in 2000 pregnancies in USA to 1 in 200 in certain part of Asia^(2,6). Partial molar pregnancy with coexisting normal live fetus is extremely rare complication of pregnancy with the incidence of .005% to .01% of all pregnancy⁷. Partial molar pregnancy resulting in a live and healthy child is a rare and estimated to occur in 1 in 20,000 to 1 in 100,000 pregnancies⁸. It is even rarer for the resulting child to be born with a normal karyotype. There are few cases reported in the literature, however some study do report

on these rare condition.^(9,10-13)

Several studies^(12,14) have reported cases of a single normal foetus associated with partial molar placenta and stated that the foetus must have a normal karyotype to survive in utero, although the placenta can show some variation from diploidy of the amnion to triploidy of the chorionic villi¹⁴ such an occurrence is presumably follows mitotic abnormalities in the early postfertilization period and represents placental mosaicism. Survival of a foetus to term in the presence of partial mole remains an extremely uncommon occurrence which is similar to this case. From this clinical perspective there are two different type of gross pathology that can affect the placenta, focal degeneration and diffuse partial degeneration.^(12,15,16,17)

Molar pregnancy with co-existing normal live foetus has been divided into three types. The first and most common is twin pregnancy with one normal fetus having a normal placenta and another complete mole, second type is a twin pregnancy with normal fetus and the placenta and another partial mole. The third and most uncommon occurrence is a singleton normal fetus with partial molar placenta¹³ similar to our case. The diagnosis of molar pregnancy with co-existing fetus is difficult. An elevated serum β -hcg with low serum placental lactogen and snowstorm appearance of placenta in USG help to diagnosis the case. This condition co-existing with viable foetus warrant for genetic analysis and search for gross congenital anomaly^{3,15}. Histopathology and cytogenetics help in final diagnosis.

Molar pregnancy with coexisting fetus carries a significant risk to mother and the fetus. Maternal risks include abnormal bleeding, preeclampsia, eclampsia, hyperthyroidism, anemia, persistent trophoblastic disease and abortion. Fetal complications include abortion, congenital anomalies, preterm, severe anemia, IUGR and IUFD^(3,13). Several factors influence the outcome in partial molar pregnancy, most important being the foetal karyotype. Other factor includes the size of the molar placenta, the speed of molar degeneration and foetal anaemia¹³

Previously most partial molar pregnancies identified early were terminated^(15,18,19) with or without medical complications but especially when preeclampsia was also present²⁰. However termination of pregnancy is not always the only option as the pregnancy can be

managed conservatively if the foetus appears normal and healthy on ultrasound and if there are no maternal complication. Patients who develop partial molar placenta may find the pregnancy complicated by intrauterine growth restriction and oligohydraamnios which were both observed in the case discussed here²¹. Jones and Lauresen recommend immediate termination after the diagnosis of molar pregnancy with co-existing foetus²². Suzuki et al, however, state that in the absence of preeclampsia or fetal abnormality the pregnancy can be allowed to continue till term⁷. In our case though there was partial molar changes in placenta, baby was grossly IUGR and there wasn't any significant symptoms or signs, so pregnancy was advised to continue. In our case prenatal karyotyping of the foetus was not possible but the postnatal karyotype of the foetus was normal 46XY, though the placental karyotyping was not done. However placenta in a partial mole with foetus in a single tone pregnancy results from dispermia and diploid karyotype in most cases.²³ Hydatidiform mole with co-existing fetus can be established by partial mole syndrome or by a twin pregnancy where the other conceptus has degenerated into a mole⁵. It is important to distinguish between a complete and partial mole when a foetus co-exists because it has been reported that a complete mole has a 20% tendency to become an invasive mole or even a choriocarcinoma, while the risk was far less for partial moles²⁴. Some Studies^{18,25,26} have questioned whether patients with partial hydatidiform mole require follow up observation and assessment of serum β -hcg concentration. However patients can develop choriocarcinoma if not followed up correctly and one study by Seckl et al reported the death of one such patient.

Szulman and Surti²⁷ reviewed 86 cases of partial hydatidiform mole and reported that the overall prevalence of the disease was 4%, however Berkowitz et al²⁵ reported a higher prevalence (9.9%) among patients who had developed persistent gestational trophoblastic disease¹³. The patient in the case discussed here did not show any persistent trophoblastic disease after the birth and her serum β -hcg concentration returned to normal within 6 weeks.

Post delivery follow up includes measurement of β -hcg value at delivery and weekly values plotted on a standard regression curve, adjusted for local reference

standard. This is followed by weekly values until three values are obtained below the limit. Then every second weekly for two months and then monthly for one year²⁸.

A single assessment of a patient's serum β -hcg concentration after the termination of pregnancy is sufficient to confirm remission in these patients²⁴

The case we reported probably had sufficient placental circulation to sustain through the 1st and 2nd trimester, however had severe IUGR due to limited placental circulation. Grossly two types of placental pathology have been described previously, focal and diffuse partial degeneration¹³. Our case had focal molar changes allowing fetal survival until term. 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 condition³⁰. The case discussed here the baby is running two years, milestones are normal and both the baby and mother are in good health.

Conclusion:

Pregnancies with normal live fetus coexistent in with partial molar placenta are extremely rare because of numerous maternal and fetal complications. In our case the foetus was born with severe IUGR but having no gross congenital abnormality, karyotyping was normal on chromosomal study and the child was continued to grow normally, the abnormal cell population appears to be confined to the placenta. The management of such rare condition should be determined on one to one basis and the possibility of increased complications and prognosis should be discussed with the family. Complete evaluation of the placental tissue is important even in cases with normal fetal outcome as molar changes which might be unsuspected antenatally might affect the future obstetrical outcome.

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