Singleton Partial Molar Pregnancy Delivered in Third Trimester: A Case Report

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Abstract

Background: A partial mole is the result of fertilization of a haploid ovum by two sperms or duplication of one sperm, resulting in a triploid karyotype (69 XXY, 69 XXX, 69 XYY). To date, there are very few cases of histo-pathologically confirmed partial moles with diploid karyotypes which survived. This case is reported to highlight the rarest variant of partial molar pregnancy.

Case Presentation: Here we present a case of singleton partial molar pregnancy co-existing with a live fetus delivered to an 18 years old primigravida lady at Jimma university medical center (JUMC) after amenorrhea of 8 months. She presented with eclamptic seizure for which she was admitted with a diagnosis of Eclampsia in 3rd trimester pregnancy. Malpresentation (breech) with Abruptio placentae to rule out partial mole was considered initially. Cesarean section (C/S) was done to effect delivery of an alive female neonate weighing 1100 gm with an APGAR score of 4, 6 & 7 at 1st, 5th & the 10th minute of life. The neonate is admitted to neonatal intensive care unit (NICU) where she is complicated by early neonatal death after 65 hours of stay despite many efforts, the possible cause of death being respiratory failure secondary to underlying prematurity. Grossly placenta was single, weighing 1200 gm and invaded by vesicles except its membrane. Histo-pathological finding of placenta was consistent with partial mole.

Conclusion: The optimal management of hydatidiform mole with coexistent live fetus is currently uncertain. Clinicians are recommended to present their individual cases for the establishment of guidelines for the management and prenatal counseling for pregnancies with partial mole with coexistent fetuses.

Keywords: β-hCG; Fetal karyotype; Partial mole; Eclampsia; Case report

Abbreviations: JUMC: Jimma University Medical Center; GTD: Gestational Trophoblastic Diseases; PM: Partial Mole; IUGR: Intra Uterine Growth Restriction; IUFD: Intra Uterine Fetal Death; GTCS: Generalized Tonic Clonic Seizure; NICU: Neonatal Intensive Care Unit; GCS: Glowsom Comma Scale; PTD: Persistent Trophoblastic Disease; PROM: Premature Rupture of Membrane; ENND: Early Neonatal Death

Background

Gestational Trophoblastic diseases (GTD) consist of a broad spectrum of conditions ranging from an uncomplicated partial hydatidiform molar pregnancy to choriocarcinoma with cerebral metastasis. Molar pregnancy is significantly more common in extremes of age [1,2]. Partial molar (PM) pregnancy is a rare entity in which there is a triplo abnormal fetus associated with a large placenta with cystic changes [3]. Pregnancies with normal live fetus coexistent in with partial molar placenta are extremely rare because of numerous maternal and fetal complications. The incidence of hydatidiform mole with a co-existing live fetus varies between 0.005 to 0.01 % of all pregnancies [2].

Partial moles are characteristically triploid with 46 chromosomes coming from the father. The pathogenesis are explained by the dispermic fertilization of an ovum or monospermic fertilization with duplication of the paternal haploid chromosome. Exceptionally, there are also diploid cases with bi-parental contribution [4,5]. Occurrence of PM pregnancy and a coexisting normal fetus presumably follows mitotic abnormalities in the early post-fertilization period and represents placental mosaicism. Survival of a fetus to term in the presence of partial mole remains an extremely uncommon occurrence and such cases represent an extremely rare outcome of molar pregnancy [6]. Here we report one of the rarest presentations of singleton PM pregnancy associated with eclampsia and preterm birth.

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On arrival she was confused with GCS of 14/15 and her vital signs were BP = 170/100 mmHg, PR=96 bpm, RR= 24 Br/min, T=37°C. Her chest was clear and resonant. Abdominal finding was 36 weeks sized gravid uterus with breech presentation. Fetal heart beat was positive. After placenta previa ruled out by sonography, pelvic examination performed. There was minimal dark red blood on examining finger and Bishop Score was 4. Obstetric Ultrasound showed singleton intrauterine pregnancy with positive fetal heartbeat, breech presentation, fundal thick placenta measuring about 11 cm with areas of multiple cystic spaces. Aggregate gestational age was 32 weeks & 2 days and estimated fetal weight was 1190 g, biophysical profile was 8/8.

With the impression of Eclampsia in 3rd trimester pregnancy with breech presentation +? Abruptio placentae to rule out partial mole, she was investigated with blood group & Rh (A~), Urinalysis (protein +3), Blood film (No hemo-parasite seen), RBS (112 gm/dl), CBC, RFT, LFT & TFT were in the normal limit. Abruptio was initially considered due to the presence of vaginal bleeding, increased placental thickness (11 cm) & eclampsia while the diagnosis of molar pregnancy was not made prior to her current presentation. Anticonvulsant (MgSO₄) and antihypertensive (hydralazine) were given to stabilize her. Once BP and convulsion was controlled the patient was taken to operation theatre for emergency caesarean section (C/S) within two hours of arrival. Transverse, lower uterine segment C/S done under general anesthesia to effect the delivery of an olive female neonate weighing 1100 g with an APGAR score of 4, 6 & 7 at 1st, 5th and 10th minutes of life. The neonate was grossly normal and neither visible congenital anomaly nor feature of hydrops was witnessed. The patient's intraoperative condition was smooth. Grossly placenta was invaded by vesicles except its membrane and weighed 1200 gm. The placenta had one cord with its 3 vessels attached to it. But there was no evidence for abruption intra-operatively. The placenta is sent for histo-pathologic examination (Figure 1).

The neonate was referred to NICU with a diagnosis of Preterm + Very low birth weight + Hyaline membrane disease + hypothermia. The neonate has been on CPAP, radiant warmer, maintenance fluid, and antibiotics. Despite the above efforts the neonate was persistently hypothermic and later complicated by early neonatal death after 65 hours of stay at NICU, the possible cause of death being respiratory failure secondary to underlying prematurity. Blood pressure of the mother was dropped to 150/90 soon after birth and she continued MgSO₄ 5 mg IM every 4 hours for first 24 hours after delivery, and overall clinical condition and urine output of the patient was good during postoperative period and discharged on her 7th post-op day with an advice to come for weekly surveillance follow up after provided with Depo-Provera Medroxy Progestrone Acetate injection for contraception as per her choice. Histo- pathologic examination showed mixture of small and large scalloped hydropic villi with central cisternae, mild irregular trophoblastic proliferation, and normal histology of umbilical cord. There are no atypical malignant cells seen. The finding is consistent with partial molar pregnancy (Figures 2 and 3). Serum B-hcG sent 48 hours after operation showed 162.3 mu/ml and her chest X-Ray is normal. During her weekly follow up, her serum B-Hcg level become normal at third week and become undetectable after three months following delivery. No evidence for persistent disease during her surveillance follow up

Discussion

Although triploidy is the most frequent association, a fetus with normal karyotype (diploidy) can survive in cases of partial molar
pregnancy. Singleton Partial mole with a live fetus may occasionally survive into full term[1,2,7] and even to the school age [8,9]. Fetuses with triploidy cannot survive after birth because of multiple malformations and severe intrauterine fetal growth retardation (IUGR) secondary to affected placental circulation. In PM there is partial replacement with hydropic villi and visible abnormal fetal parts mostly leading to termination of pregnancy in the first trimester. It presents with several dilemmas in management during pregnancy and the woman must be counseled regarding the maternal and fetal complications and the risk of persistent trophoblastic disease in later life [10-13].

Hydatidiform mole with coexistent normal fetus is not necessarily considered a partial mole. Cases of such association can be classified into three types. The first and most common is twin pregnancy with one normal fetus having a normal placenta and another complete mole. There have been so far, about 200 such cases fully documented in literature of which only 56 resulted in a term live birth, second type is a twin pregnancy with normal fetus with normal placenta and another partial mole. The third and most uncommon occurrence is a singleton normal fetus with partial molar placenta similar to our case [5,11]. Sometimes the third variant is difficult to diagnose especially when the patient come at third trimester or in labor for the first time as sonography alone can’t tell us everything. In our case the fact that the fetus presented with eclampsia and alive fetus with single large placenta (11 cm thick with multiple hypo echoic area) leads us to consider abruptio placentae while the reality is not. We were in doubt to consider partial mole due to the extreme rarity of this case although there were clues in favor of partial mole.

In the past most molar pregnancies associated with live fetus were terminated in view of adverse maternal and fetal outcome in advanced gestations. Close maternal and fetal surveillance may help in achieving a favorable outcome though termination is required only in cases of gross fetal anomalies or deteriorating maternal condition [9]. Several factors may affect the outcome of the fetus in cases of partial molar pregnancy. These include karyotype of the fetus, the size of the abnormal molar placenta, the speed of molar degeneration, and the occurrence of fetal anemia or other obstetric complications [11].

Molar pregnancy with coexisting fetus carries a significant risk to the mother and the fetus. Maternal risks include abnormal bleeding, hyper emesis gravidarum, preeclampsia, eclampsia, hyperthyroidism, anemia, preterm labor, mal-presentation like transverse lie, Preterm premature rupture of membrane (PROM), persistent trophoblastic disease and abruption. Fetal complications include abortion, congenital anomalies, preterm, severe anemia due to limited placental circulation, IUGR and intra uterine fetal death (IUFD) [6,11-15]. In our case, hyper emesis gravidarum, preeclampsia, eclampsia, preterm delivery, mal-presentation (breech) and fetal anemia were seen Table 1. The serum B-hcG will drop immediately after delivery when compared to the one sent immediately before delivery. This is witnessed in one case report where serum B-Hcg sent before delivery was as high as 16,000 mIU, while sent once again 48 hours of delivery; it was 247 mIU [12]. Immediate post-delivery serum B-hcG value of our case is consistent with this report although difficult to comment on pre-delivery value.

After delivery serial follow up of mother for early detection of persistent trophoblastic disease (PTD) should be done as it is done in our case. Fetal karyotype is mandatory to know whether the partial mole is of normal or abnormal karyotype. Immuno-histochemical evaluation for p57 protein is occasionally needed to differentiate between complete or partial molar pregnancy. Early target scanning and early prophylactic antenatal steroids are recommended [15]. In our case emergent presentation of patient necessitating immediate delivery hinders us not to have targeted sonography and to provide corticosteroids for fetal lung maturity. Unfortunately, we don’t have karyotyping and Immuno-histochemical evaluation for p57 protein services in our medical center. From review of literatures all partial moles survived beyond late 2nd trimester were all having normal karyotype.

Although we don’t have karyotype and Immuno-histochemical evaluation for p57 protein, the fact that the fetus is apparently normal and alive with single placenta and histo-pathologic finding consistent with partial mole favors diagnosis of singleton partial molar pregnancy likely of normal karyotype than diagnosing complete molar pregnancy with co-twin fetus.
Conclusion

The optimal management of Hydatidiform mole with coexistent alive fetus is currently uncertain. Clinicians are recommended to present their individual cases for the establishment of guidelines for the management and prenatal counseling for pregnancies with partial mole with coexistent fetuses. From review of the literatures only partial mole with coexistent fetuses.  From review of the literatures only partial

References

13. Koregol M, Parviz S [14] 23 G2P1 11.5/26 46XY 64750 mIU/mL M, dead Placenta =100 g -PROM, IUFD

Table 1: Review of cases of partial mole co-existing with live fetus in case of singleton pregnancy.

<table>
<thead>
<tr>
<th>Maternal age (years)</th>
<th>Reproductive History</th>
<th>GA at Dx/ Delivery</th>
<th>Fetal karyotype</th>
<th>Serum B-Hcg</th>
<th>Outcome</th>
<th>Maternal &amp; fetal complications</th>
</tr>
</thead>
<tbody>
<tr>
<td>Parveen et al. [1]</td>
<td>23</td>
<td>G4P1A2</td>
<td>18/39</td>
<td>NA</td>
<td>F=2700 g, Alive</td>
<td>-Bleeding, anemia, IUGR</td>
</tr>
<tr>
<td>Ara et al. [2]</td>
<td>26</td>
<td>G2P1</td>
<td>28/40</td>
<td>46XY</td>
<td>M=1500 g, alive</td>
<td>- IUGR</td>
</tr>
<tr>
<td>Singh et al. [3]</td>
<td>24</td>
<td>G1</td>
<td>20.4/20.4</td>
<td>Trisomy 21</td>
<td>4, 24, 249 miU/ml</td>
<td>Alive, 200 g -Vaginal bleeding, anemia, IUGR</td>
</tr>
<tr>
<td>Deveer et al. [5]</td>
<td>35</td>
<td>G5P4</td>
<td>14/36</td>
<td>46XY</td>
<td>Not mentioned</td>
<td>Placenta =700 g, -pre eclampsia, but No PTD</td>
</tr>
<tr>
<td>Rathod et al. [6]</td>
<td>23</td>
<td>G1</td>
<td>22/34</td>
<td>46XX</td>
<td>23500 miU/ml</td>
<td>F=1400 g, alive - Pre eclampsia, anemia, PROM, Placenta =1700 g, - preterm labor, IUGR, breech, -microphaly but no PTD</td>
</tr>
<tr>
<td>Ramani B et al. [9]</td>
<td>36</td>
<td>G2P1</td>
<td>29.5/34</td>
<td>NA</td>
<td>M=960 g, alive</td>
<td>-Vaginal bleeding, anemia, preterm Placenta =1800 g, -but no PTD</td>
</tr>
<tr>
<td>Hsieh CC et al. [11]</td>
<td>30</td>
<td>G1</td>
<td>18/32.6</td>
<td>46XX</td>
<td>167, 596 miU/ml</td>
<td>F=1651 g alive -maternal anemia, fetal anemia - but no PTD</td>
</tr>
<tr>
<td>Shobha et al. [12]</td>
<td>21</td>
<td>G1</td>
<td>20/37</td>
<td>NA</td>
<td>16,000, 247 miU/ml</td>
<td>M=1500 g, dead - IUGR, IUFD Placenta =600 g, -No PTD</td>
</tr>
<tr>
<td>Rahamni M, Parviz S</td>
<td>23</td>
<td>G2P1</td>
<td>11.5/26</td>
<td>46XY</td>
<td>64750 miU/mL</td>
<td>M, dead Placenta =100 g -PROM, IUFD</td>
</tr>
<tr>
<td>Koregol M et al. [13]</td>
<td>22</td>
<td>G2P1</td>
<td>31/31</td>
<td>NA</td>
<td>M=2000 g, alive</td>
<td>Placenta =700 g, -bilateral Tallipes equinovarus, -ENND, No PTD</td>
</tr>
<tr>
<td>This case</td>
<td>18</td>
<td>G1</td>
<td>32.3/32.3</td>
<td>NA</td>
<td>162.3 miU/ml</td>
<td>F=1100 g, alive -Preeclampsia, eclampsia, preterm delivery, mal-presentation(breech) and fetal anemia, ENND, No PTD</td>
</tr>
</tbody>
</table>