

A Partial Hydatidiform Mole with a Rarely Normal Karyotype; Differential Diagnosis and Management

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1. Abstract

A hydatiform mole is a gestational trophoblastic disease, which originates from the placenta, it can be complete or partial with a viable fetus. The karyotype of the fetus is mostly triploid with rarely seen normal karyotypes. Case. We present a case of partial molar preeclampsia diagnosed with preeclampsia and fetal anomalies on sonography, genetic studies were obtained normal, but termination of pregnancy at 21-22 weeks were done because of worsening of disease. After termination the follow up continued one year. No complications were seen. In differential diagnosis, placental mesenchyme disease must be taken in consideration which goes with milder symptoms but similar placental cystic spaces.

2. Introduction

A hydatiform mole is a gestational trophoblastic disease, which originates from the placenta and can metastasize [1].

In developed countries, the incidence of complete moles is approximately 1–3 per 1000 pregnancies and those of the partial moles about 3 per 1000 pregnancies [2]. There are 2 types of hydatidiform moles: complete and partial. Complete Hydatidiform Mole (CHM) is a placental pathology with androgenetic diploid origin (chromosomes only from paternal origin). Placental villi is characterised with an abnormal hyperproliferation and hydropic degeneration forming a mass of multiple vesicles associated with the absence of embryo. The karyotype is 46, XX 90% of the time and 46, XY 10% of the time [1].

On the other hand, in partial moles, the karyotype is 90% triploid and either 69, XXX or 69, XXY [1, 2]. The partial hydatidiform mole is usually triploid, with one maternal and two paternal hap-

loid sets. There is usually a fetus and a large placenta. The hydropic villi show a less florid appearance than is seen with a complete hydatidiform mole and are interspersed with normal chorionic villi [2]. The fetus usually dies within a few weeks of conception, and a recent review did not identify any case in which a fetus of paternal (diandric) origin survived to term. Very rarely, a partial molar pregnancy develops with two maternal and one paternal haploid set (digynic). In these cases, the placenta is small, the villi show minimal hydropic changes, and the fetus is growth-restricted. Some of these pregnancies have been reported to result in live births, with subsequent early neonatal death [3, 5].

3. Case

A 28 year old patient was referred us at 17-18. Weeks of pregnancy because of her high AFP (3.97) MoM, T-hCG MoM value was 23 MoM and inhibin MoM value was 43.7MoM at quadruple screening test. Open NTD risk was above the cutoff with 1/6.

On ultrasonography; biometry was concordant with LMP, hyperechogenic fetal bowel, pericardial effusion and thickened vacuolar cystic placenta was observed on fetal echo right ventricular dominance was recorded with hypertrophy of ventricular walls (Figure 1, 2). At the beginning molar pregnancy or placental mesenchymal dysplasia were thought in differential diagnosis and because of fetal sonographic anomalies, chromosome, FISH and microarray analysis were performed with amniocentesis and FISH analysis was normal with karyotype analysis was found to be 46XX and array report was likely benign.

We also sent TORCH panel; ICT (indirect coombs test) from mother and but found no specific finding.

While we were gathering the results the mother started to have hypertension resistant to medical therapy and proteinuria ensued, blood urea and creatinine levels started to rise, growth restriction of the fetus was observed with progressive deteriorating doppler changes. We decided to terminate the pregnancy with the diagnosis of severe preeclampsia and IUGR at 22 weeks of gestation. A 510 gr, female fetus was aborted. Postpartum, placenta was sent for histopathologic examination and final diagnosis was concordant with partial mole hydatiform (Figure 3 and 4).



Figure 1: Sonography of fetus



Figure 2: Sonography of placenta



Figure 3: View of molar placenta



Figure 4: View of molar vesicles

4. Discussion

The diagnosis of a molar pregnancy might be suspected based on a number of clinical features: abnormal vaginal bleeding in early pregnancy is the most common presentation; uterus large for dates (25%); large benign theca-lutein cysts (20%); exaggerated pregnancy symptoms including hyperemesis (10%), hyperthyroidism (5%), early preeclampsia (5%) like our patient in neglected cases vaginal passage of grape-like vesicles (10%). Sonography is used to diagnose molar pregnancy before 12 weeks, showing a fine vascular or honeycomb appearance. In advanced pregnancy a complete mole is characteristically described as snowstorm appearance, representing hydropic villi and intrauterine hemorrhage. The ovaries usually contain multiple large theca-lutein cysts due to increased ovarian stimulation by increased beta-hCG production [2, 4].

Ultrasound diagnosis of partial mole is more difficult: the fetus is still viable, but may show signs of chromosomal anomaly consistent of triploidy, such as early growth restriction or developmental or congenital abnormalities. This is in accordance with our case which presented with IUGR, pericardial effusion in the fetus, hypertension in the mother. There is a scattered cystic space within the placenta, and ovarian cystic changes could be seen lesser than complete mole. There were theca lutein cysts in both ovaries in our case which we recorded when looking for a differential diagnosis.

In women with a complete mole, the quantitative serum beta-hCG level is higher than expected, often exceeding 100,000 IU/L. In partial mole, the level of beta-hCG is less pronounced. For these reasons the diagnosis of a partial mole can be missed (4).

In case of a mole, systemic examination must be done, early preeclampsia is common as in our case, systolic and diastolic arterial tension was around 140-90. /160-100. Laboratory investigations must include a complete blood count, creatinine and electrolytes, kidney, liver and thyroid function tests and urinary examination for proteinuria must be checked.

Management is by suction curettage with vacuum, the preferred method of evacuation regardless of uterine size in patients who desire to preserve fertility. It is best to avoid prior cervical prepara-

tion, oxytocic drugs and sharp curettage or medical evacuation, to minimize the risk of dissemination of tissue leading to metastatic disease [4,6]. Oxytocic agents and prostaglandin analogues are best used only after uterine evacuations when there is significant hemorrhage. In partial mole, because there was fetus inside, we started induction protocol with PGE₂, the induction procedure was without any complication, vacuolar placenta was expelled spontaneously and sent for histopathology. In this case the sonographic examination of the placenta was not so typical for partial mole and we thought of placental mesenchymal dysplasia in differential diagnosis. Karyotype was diploid not triploid as expected, so we thought in favor of placental mesenchymal dysplasia with enlarged placenta. In that case also placenta is enlarged with cystic villi and dilated thick vascular structures and grapelike vesicles resembling partial mole but there is no trophoblastic proliferation and there is aneurysmally dilated vessels on the placenta in placental mesenchymal dysplasia also in this state, fetus is phenotypically normal and pregnancy outcome was more favorable with a rate of 33% IUGR, 30% preterm labor and 13% IUFD rates [7, 9].

In a partial mole, there typically is the finding of a fetus which may be viable, the presence of amniotic fluid and the placenta appears to have enlarged with cystic spaces and trophoblastic proliferation but karyotype is mostly triploid. For partial moles, fertilization takes place between the oocyte and 2 sperms. Therefore the zygote is triploid with extra chromosome of paternal origin. In rare cases the partial hydatidiform mole have also been reported with normal karyotypes [3, 8]. Mitotic abnormalities in the early post-fertilization period and placental mosaicism can be a potential cause for this event. Besides reaching to term in 4-5 cases in literature review, Guven reported 17 cases of partial hydatidiform mole associated with normal fetal karyotype. They concluded that despite the high rate of adverse perinatal outcomes, continuing the pregnancy with close follow-up in a tertiary center is feasible if amniocentesis or fetal blood sampling reveals a normal karyotype [10]. We could not let the pregnancy to continue because of severe preeclampsia symptoms and early IUGR with umbilical artery doppler changes.

The outcome of a partial hydatidiform mole after uterine evacuation is almost always benign. Persistent disease occurs in 1.2% to 4% of cases; metastasis occurs only in 0.1% of cases. In complete moles, these risks are approximately 5 times greater after treatment with uterine evacuation and 2-3 times greater after hysterectomy [2, 3]. The risk of persistent or recurrent GTD is greatest in the first 12 months after evacuation, with most cases presenting within 6 months. Family planning should be done at least one year with contraceptive pills. A careful pelvic and abdominal ultrasound scan should be done to look for evidence of an invasive mole, and check for possible metastatic disease. Computed tomography or magnetic resonance imaging may provide further information. Chest radiography or computed tomography should be considered if there are symptoms that suggest pulmonary metastases [1, 2, 6]. In pregnancy these tests are kept after pregnancy termination.

Total abdominal hysterectomy is a reasonable option for patients who do not wish to preserve their fertility. Though hysterectomy eliminates the risk of locally invasive disease, it does not prevent metastases and reduces the subsequent risk of persistent trophoblastic disease by up to 50% [3, 6].

Patients with prior partial or complete moles have a 10-fold increased risk (1-2% incidence) of a second hydatidiform mole in a subsequent pregnancy [2]. Therefore, all future pregnancies should be evaluated by early obstetric ultrasonography.

In case of life-threatening complications in such patients like intractable hyperemesis or preeclampsia or eclampsia, termination of the pregnancy is the treatment of choice as in our case. Our case was healthy after 1 year follow up hCG without progression of the disease.

5. Conclusion

Differential diagnosis should be done in cystic appearing placentas on antenatal sonography, in presence of a normal karyotype and absence of notable fetal/maternal complications, pregnancy can be continued with discussing the risk both antenatal and postnatal complications in molar pregnancy like progression to persistent trophoblastic disease.

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