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Case Study

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PARTIAL MOLAR PREGNANCY AT 8 MONTHS WITH A DIZYGOTIC FETUS MISTAKENLY DIAGNOSED AS A RETROPLACENTAL HEMATOMA: A CASE REPORT

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ABSTRACT

Introduction: Partial hydatidiform mole (PHM) is a rare type of gestational trophoblastic disease with an incidence of 3 per 1000 pregnancies. It involves localized trophoblastic hyperplasia and degeneration of chorionic villi, sometimes with identifiable fetal tissue. The genetic cause is typically a triploid conception due to the fertilization of an ovum by two spermatozoa or a duplicated spermatozoon. PHM is very rare when a living fetus with a normal karyotype is present, occurring in 0.005 to 0.01% of pregnancies, and is difficult to diagnose, particularly in the absence of clear clinical signs. **Case presentation:** A 40-year-old woman, with no notable medical history and an unmonitored pregnancy, was admitted for severe preeclampsia, positive bleeding, and contractions and other symptoms. The ultrasound showed a placenta with multiple anechoic areas, revealing a non-viable pregnancy with and a diagnosis of HELLP syndrome with renal failure. An emergency cesarean was performed, resulting in the delivery of a stillborn male fetus and the discovery of a partial mole. Post-operatively, the patient received methotrexate and folinic acid chemotherapy until the beta-HCG levels normalized and was monitored monthly for six months, showing favorable progress. **Discussion: Conclusion**: Partial mole with a diploid fetus is a very rare condition that can be challenging to diagnose due to often lacking clinical evidence. Early suspicion and careful diagnosis are crucial for effective management, with ultrasound being a key diagnostic tool and amniocentesis used for confirmation. In the absence of significant fetal abnormalities and manageable maternal complications, continuing the pregnancy is recommended.

KEYWORDS: Partial mole pregnancy; diploid fetus; cesarean section; methotrexate.

INTRODUCTION

Partial hydatidiform mole (PHM) is a type of gestational trophoblastic disease. It is relatively uncommon, with an incidence of 3 per 1000 pregnancies.[1] When the degenerative process affects one-third or two-thirds of the placenta, it is classified as partial, sometimes with embryonic tissue. This condition is generally benign but can progress to a clinically malignant entity known as gestational trophoblastic tumor.[2] It is characterized by localized and discrete trophoblastic hyperplasia, degeneration of chorionic villi, and the presence of identifiable fetal or embryonic tissue, along with excessive secretion of human chorionic gonadotropin (HCG). The only clearly identified risk factor is maternal age.[2] The genetic origin of PHM involves a triploid conception with an extra set of paternal chromosomes, typically resulting from the fertilization of a normal ovum by two spermatozoa (dispermy) or a duplicated spermatozoon.[4] The occurrence of a living fetus with a normal karyotype in such cases is very rare, happening in 0.005 to 0.01% of all pregnancies, making diagnosis difficult, especially in the absence of indicative clinical signs.[1] In Morocco, research on gestational trophoblastic diseases has been conducted generally, but few studies have reported on the association of a living fetus with a partial hydatidiform mole. The frequency of PHM in Morocco is 0.4 per 1000 pregnancies.[2]

CASE PRESENTATION

The patient is a 40-year-old woman with no significant medical history, G9P8, with an unsupervised pregnancy. She was admitted to the obstetric emergency department for severe preeclampsia, with a blood pressure of 19/12, a heart rate of 90 bpm, a positive: syphilis test, epigastric pain, bleeding, and contractions. Proteinuria was positive at three crosses, fetal heart tones were absent, and the vaginal examination revealed a long closed posterior cervix, with intact membranes and minimal blackish bleeding. An emergency ultrasound showed a non-viable pregnancy with a gestational age of 29-30 weeks and an

enlarged placenta with multiple anechoic areas (the images could not be printed because the device was not working). Laboratory tests revealed HELLP syndrome with renal insufficiency. An emergency cesarean section was performed to save the mother. Intraoperatively, the uterus was of normal size, and a segmental incision was made to extract a fresh stillborn male fetus with normal morphology weighing 950 g, and an enlarged placenta with translucent vesicles. Pathological examination of the placenta revealed a normal umbilical cord, an enormous hematoma, signs of fetal thrombotic vasculopathy, thrombosis of maternal spiral vessels, areas of placental infarction, and large, sinuous chorionic villi with irregular hyperplasia of cytotrophoblast and syncytiotrophoblast, consistent with a partial mole. The karyotype of the stillborn was not performed. Postoperatively, the BHCG level remained stable, and the patient received chemotherapy with methotrexate and folinic acid until the BHCG became negative. She was discharged with monthly BHCG monitoring for 6 months, with a good outcome.

DISCUSSION

The mechanisms of chromosomal abnormalities leading to a partial mole (PM) are most often due to the fertilization of a haploid egg by two haploid sperm (dispermy) or by a diploid sperm (diandry).[3] The distribution of sex chromosomes results in three types of triploidy: 69 XXX, 69 XXY, and 69 XYY, with XXY estimated to be 70% of cases. XYY cases are much rarer as their development beyond four weeks is generally not feasible. A marked predominance of XXX triploidy is observed, with longer gestation periods in XXY triploidy.[4] The triploid population has an increased doubling time, which explains fetal growth restriction, the frequency of malformations, and persistently high hCG levels due to excessive cell proliferation of residual trophoblast.[2]

Clinically, partial moles (PM) often result in spontaneous miscarriages during the first trimester. In these cases, two-thirds of the embryos are typically growth-restricted but otherwise normal, while the remaining third may be malformed.[2-5] By the second trimester, PM can be associated with early pregnancy-related renal issues, including preeclampsia[2] or, less commonly, pseudo-glomerulonephritis, which is more indicative of PM.[6] Late-stage PM can also be linked with preeclampsia or HELLP syndrome, as observed in our patient.[7] Persistent bleeding, severe and rare vomiting, and excessive uterine enlargement—often due to acute hydramnios (with normal or reduced uterine height in cases of fetal growth restriction or fetal death)—are common findings.[7-8] Thyroid hypertrophy has been reported[8], and luteinized ovarian cysts, though rare.[2-6]

Ultrasound is a highly sensitive and specific tool with well-defined diagnostic criteria. The placenta appears large and heterogeneous, with part of the trophoblast having a normal appearance and the other part showing different echogenicity, filling most of the uterine cavity and displaying a "snowflake" pattern. Numerous characteristic cystic spaces are also visible.[5] In the second trimester, severe intrauterine growth restriction (IUGR) and morphological anomalies are observed in 93% of cases. The "Swiss cheese" appearance of the placenta, with thickening greater than 4 cm between 18 and 22 weeks of gestation[8], supports the diagnosis. This appearance can create a differential diagnosis challenge with benign hydropic degeneration of the placental villi, where hCG levels are normal and the fetal karyotype, if performed, is also normal. Ultrasound does not replace histology, which reveals a mixture of molar and non-molar villi. The villous lining shows invaginations and cysts with a double layer of cytotrophoblast and syncytiotrophoblast. Cytogenetic analysis or MHP testing often reveals diandric triploidy, characterized by two sets of chromosomes of paternal origin and one set of maternal origin.[9]

Several factors influence the outcome of the fetus in partial molar pregnancy, with the most important being the karyotype of the fetus. In the present case, there was no obvious congenital anomaly. Other factors include the size of the molar placenta, the speed of molar degeneration, and fetal anemia.[10]

Management of molar changes associated with a normally appearing fetus remains challenging. The serum b-hCG level can be a useful marker; if it remains above 106 mIU/mL, consideration should be given to termination of pregnancy (TOP). In contrast, in cases with successful pregnancy outcomes and viable fetuses, the serum b-hCG level typically begins to decrease from the start of the second trimester, and ultrasound generally shows a reduction in the size of the molar portion of the placenta.[11] Close monitoring of the mother and fetus can help achieve a favorable outcome, and termination of the pregnancy is only required in cases of fetal anomalies or a deterioration in the mother's condition.[12]

Clinical, ultrasound, and biological monitoring is essential after the evacuation of a molar pregnancy to detect early progression to gestational trophoblastic tumor (GTT). The risk of developing GTT after a partial hydatidiform mole (PHM) is 2% to 4%, and this risk increases with the duration of the pregnancy and the promptness of the intervention.[13] The literature reports a favorable outcome in over 96% of PHM cases. Recurrence of a PHM is rare, with the risk of recurrence ranging from 0.6% to 2% depending on the studies.[5] The risk of recurrence is much higher in patients with a mutation in the NLRP7 gene.[14] Oral contraception should be started immediately after evacuation and maintained for six months to a year after β-hCG levels have normalized. Monochemotherapy with methotrexate may be recommended depending on clinical evolution.[15]

Figures

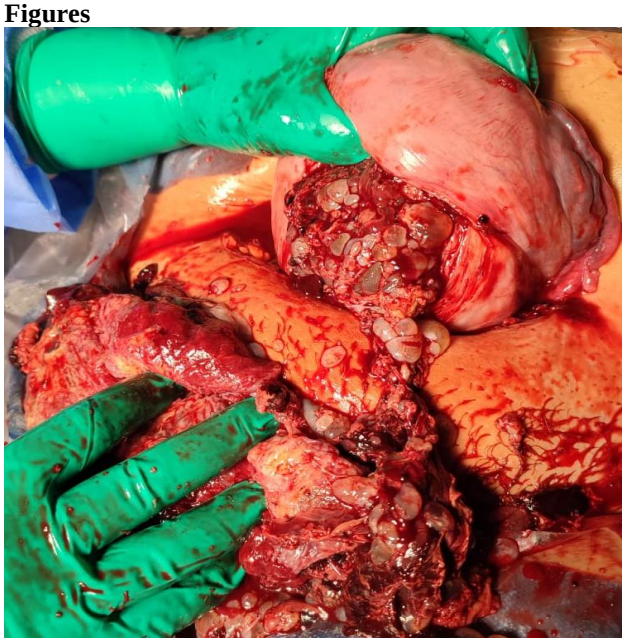

Figure 1: Large placenta with multiple vesicles.

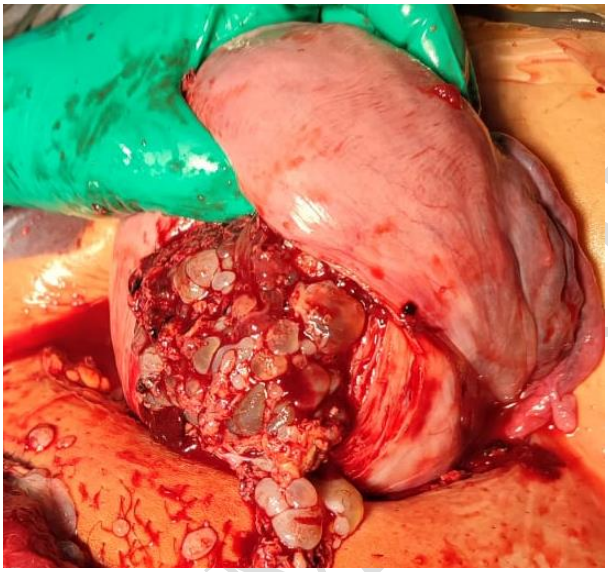

Figure 2: Intrauterine vacuoles after delivery of the placenta.

CONCLUSION

Partial mole with a diploid fetus remains a very rare condition and can be confusing due to the often lack of clinical evidence supporting the diagnosis. High vigilance is necessary to suspect it early and to achieve a reliable diagnosis for optimal management. Ultrasound plays a crucial role in diagnosis, which is strongly suspected based on clinical and ultrasound criteria and confirmed by the rapid acquisition of a fetal karyotype through amniocentesis. In absence of gross fetal abnormalities on sonography, we recommend to continue the pregnancy as long as maternal complications are absent or controllable.

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