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Case Report

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Obstetrical and Oncological Outcome of Complete Hydatidiform Mole and Coexisting Fetus: A Case Report

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Abstract

Twin pregnancy consisting of complete hydatidiform mole and coexisting fetus (CHMCF) is a very rare occurrence. We report the case of a patient with a CHMCF diagnosed at 11 weeks gestation where we ended the pregnancy in the second trimester due to maternal complications; following termination of the pregnancy, a persistent trophoblastic disease was diagnosed, and we describe the follow-up, treatment and oncological outcome.

Keywords: *complete hydatidiform mole and coexisting fetus, persistent trophoblastic disease, Gestational trophoblastic disease.*

Introduction

Multiple gestation consisting of a complete hydatidiform mole and coexisting normal foetus is a rare event, occurring in 1 per 22,000-100,000 pregnancies (2). It is associated with an increased risk of maternal complications and unfavourable foetal outcome, therefore close surveillance is essential if it is decided to continue the pregnancy as well as a strict post-partum follow-up in order to exclude a persistent trophoblastic disease (PTD) (9). The association of a complete hydatidiform mole and a coexistent foetus is rare and prenatal care is challenging. We report a case report on the management of a case of complete hydatidiform mole and coexistent fetus with a review of the literature.

Aim

To report a case of complete hydatidiform mole and co-existing foetus

Methods

The case notes of the patient, the laboratory records and radiology images of the patient were retrieved which were utilized to provide a summation of the case. Internet data bases were searched including: Google, Google Scholar, and PUBMED for information on hydatidiform form and molar pregnancy in order to obtain information on the association between Hydatidiform mole and co-existing pregnancy.

Case report

A 36-year-old G IV P II presented to observation with vaginal bleeding at 11 weeks gestation. There was history of caesarean section and operative vaginal delivery four and two years before, and missed abortion with misoprostol evacuation. Ultrasound showed a live and fetus with normal anatomy and normal placenta (Figure 1), and a second amniotic sac without embryo, with a heterogeneous mass 9x10x6 cm with numerous anechoic cystic spaces (Figure 2).

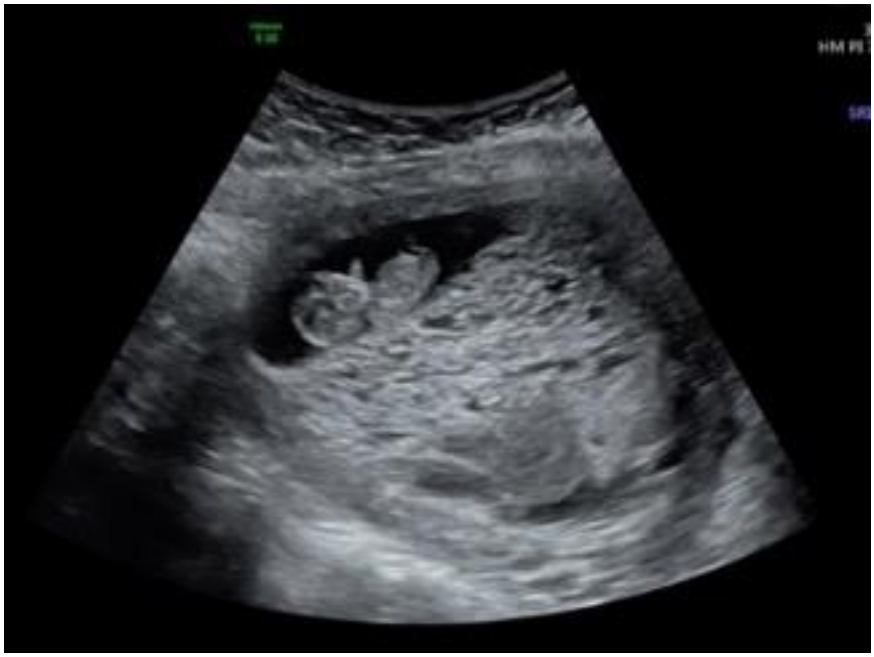

Figure 1: Ultrasound shows complete hydatidiform mole and coexisting live foetus showing.

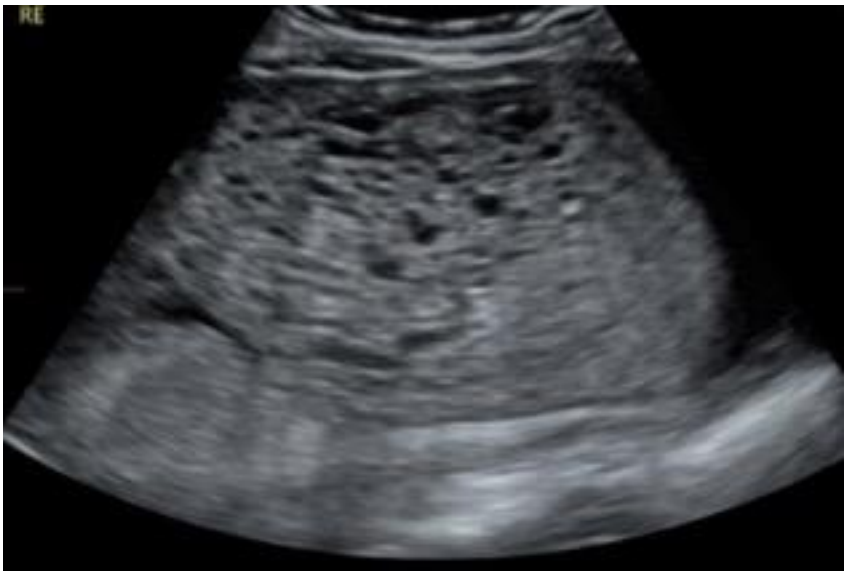

Figure 2: Ultrasound shows the classic "snowstorm" or "granular" lesion with typical multiple echogenic foci.

Serum hCG was 833.000 IU/L, clinical examination revealed a soft and very enlarged uterus. Ovaries were normal and not enlarged. Due to the hCG level and the poor perfusion a mesenchymal dysplasia was excluded. After comprehensive counselling about the risk of severe complications, the patient chose to continue the pregnancy under close observation. Coagulation was intact with normal fibrinogen and D-dimer, there was no vaginal bleeding more, no signs of preeclampsia or thyrotoxicosis. At 16 weeks gestation the patient had a severe haemorrhage with haemorrhagic shock and beginning expulsion of the mole, so it was decided to end the pregnancy per laparotomy and hysterotomy.

Weekly hCG monitoring showed decreasing levels to a minimum of 5700 IU/L six weeks after delivery, then a rise to 12500 IU/L. A computed tomography scan showed pulmonary metastases; there was no evidence of cerebral involvement by a magnetic resonance. Therefore, a gestational trophoblastic neoplasia stage T1 M1a was diagnosed, low risk (WHO prognostic score 3-4). A single-agent chemotherapy was begun with Methotrexate. After a short response, the patient presented a plateau and then an increase in the hCG levels, so was administered a second-line single-agent regimen with Actinomycin D, but this proved to be unsuccessful too. A hysteroscopy was performed to remove residual trophoblastic tissue, but had to be broken off due to acute bleeding. A laparoscopic hysterectomy was thus indicated and performed without complications. Histologic diagnosis proved a choriocarcinoma. Following the hysterectomy the serum hCG levels fell until undetectable 4 weeks postoperatively without any other chemotherapy.

Discussion

Gestational trophoblastic disease (GTD) defines a spectrum of proliferative disorders of the placental trophoblastic epithelium (1).

The incidence of gestational trophoblastic disease varies according to geographic location, and is higher among women of Asian, West African and South American origin (2,3). Genetic factors, race, maternal age and previous molar pregnancy are risk factors for molar pregnancy (2).

GTD is histologically classified as benign forms of complete and partial hydatidiform moles and malignant forms of invasive moles, gestational choriocarcinoma, placental site trophoblastic tumours (PSTT) and epithelioid trophoblastic tumours (6).
Molar twin pregnancy is a rare obstetric event, characterised by the coexistence of a hydatidiform mole and a potentially viable foetus with an associated normal placenta (1). The most common symptom is vaginal bleeding, as well as symptoms associated with high HCG levels such as hyperemesis gravidarum and hyperthyroidism (4,5).

CHMCF is associated with an increased risk of obstetrical complications which include abortion, preterm birth, intrauterine death, preeclampsia and haemorragae (7,8). In our clinical case a conservative approach was considered possible, as close perinatal surveillance was crucial for an adequate management of possible complications. The pregnancy had to be terminated due to haemorrhage.

Regarding the possibly increased risk of CHMCF progressing to GTN compared with a single complete mole, the literature is not conclusive. It was reported a significantly higher rate of GTN in CHMCF (9-13) so that elective termination of pregnancy was traditionally advised, but these findings were based on case reports and small series. However, Sebire et al found similar rate of GTN after CHM (16%) and after CHMCF (19%) (8). He conducted the largest study ever published, so we think that his conclusions should be considered the most reliable, even though a small but significant increase in the risk of GTN cannot be excluded. Similar results were found by Niemann, who reported a similar risk of persistent trophoblastic disease after a diploid mole with coexisting fetus pregnancy and after a singleton molar pregnancy (25% vs 17%). Moreover, the severity of persistent trophoblastic disease after CHMCF pregnancy did not seem to be aggravated compared with PTD after singleton diploid mole (14).

Elective termination of the gestation did not influence the risk for GTN; but the need for termination due to complications and higher hCG levels were associated with development of GTN in CHMCF (7,8,14).

A number of studies estimated the probability to obtain a live birth among patients carrying on a CHMCF pregnancy between 29 and 40% (7,8,14). Suksai attempted to identify possible predictors of favorable obstetric outcome: CHMCF without antenatal maternal complications mainly pregnancy-induced hypertension, hyperthyroidism and hyperemesis gravidarum especially together with initial serum hCG levels lower than 400,000 mIU/ml may possibly achieve a favorable outcome for the fetus (7).

Conclusions

- A CHMCF is a high-risk pregnancy.
- Continuation of the pregnancy in cases of CHMCF is an acceptable option, but a comprehensive and detailed discussion with the parents is needed. They should be aware of an increased risk of maternal complications and a possibly poor outcome for the fetus. They should also be aware of a potentially high risk of developing GTN, even though termination of pregnancy does not decrease the risk of GTN.
- The probability of having a child in a pregnancy with complete hydatidiform mole with coexisting living fetus is low, even though several cases are reported in the literature.
- A close monitoring during gestational time is mandatory to detect early signs of maternal and fetal complications, as well as a careful postpartum follow-up is critical to exclude PTD and a progression to GTN.

Contributors
All authors made a substantive contribution to the information and material submitted for publication.

Conflict of interest
The authors declare that they have no conflict of interest regarding the publication of this case report.

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Patient consent
Informed consent was obtained from the patient for the publication of this case report and the accompanying images.

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