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Complete hydatidiform mole and a coexistent viable fetus: a case report and review of the literature

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Summary

A complete hydatidiform mole with a co-existing viable fetus (CMCF) is a rare clinical presentation. It is difficult to diagnose and there is no standard protocol for its management. The risks of pregnancy continuation in cases of CMCF still remain uncertain due to rarity of this situation and the possible complications (persistent gestational trophoblastic disease (GTD), preterm labour, and pre-eclampsia) must be assessed in relation to CMCF with mother. In such cases, it should be explained to family that a healthy fetus may be born together with/despite pregnancy related complications. Furthermore, due to GTD development risk after birth, the patient should be monitored until beta-hCG decreases to an undetectable level.

Key words: Mole pregnancy; Viable fetus; Twin pregnancy.

Introduction

A complete hydatidiform mole with a co-existing viable fetus (CMCF) is a rare clinical presentation. The main issue regarding this rare entity is its diagnosis. Its incidence is around 1:22,000 to 1:100,000, but its exact incidence is unknown because of the diagnose issues [1]. Careful clinical evaluation and detailed ultrasonic examination of the placenta are needed for the diagnosis of CMCF. There is no standard protocol for its management and only a few case reports have shared clinical management of this rare entity. Vaginal bleeding, hyperthyroidism, preterm delivery, and pre-eclampsia are the main complications of this condition [2].

The authors present an unusual case in light of the literature, where the patient was diagnosed with CMCF on ultrasound at 17 weeks of gestation. The diagnosis was confirmed on histopathologic examination of molar tissue after birth with a surviving neonate. Transabdominal ultrasound examination revealed a dichorionic twin.

Case Report

A 33-year-old, gravida 6, para 2 presented with three months' amenorrhea and vaginal bleeding. Her last pregnancy resulted uneventfully in a full-term vaginal birth four years prior. She had no prior obstetrical examination. Transabdominal ultrasound examination confirmed a dichorionic, twin pregnancy of nine weeks gestation with live fetuses. The two placentae and two gestational sacs were separate with sonolucent areas within them, interpreted as hematomas (Figure 1). After four days of progesterone supplementation, she was discharged from hospital.

Ultrasound examination was repeated at 17 weeks of gestation which revealed structurally normal fetus. However the placentae of both sacs were separate and one of them adjacent to the dividing membrane had cystic changes suggestive of hydatiform mole (Figure 2). Serum beta-hCG level was 236,532 mUI/ml and eight mean of medians (MoM). Amniocentesis and chorion villus biopsy from both placentae was advised. The patient however declined any intervention. The authors opted for expectant management. The patient showed no signs of preeclampsia, vaginal bleeding or any other complication while she was on expectant management.

At 33 weeks of gestation, she was admitted to the hospital in preterm labor and had a normal spontaneous delivery of a 2,240-gram normal female infant with Apgar scores of 9 and 10. The placenta delivered spontaneously. A grape-like mass with a diameter of 77×87 mm consisting of multiple cysts was noticed (Figure 3). The placenta was sent for pathological examination. On microscopic examination, part of the placenta consisted of

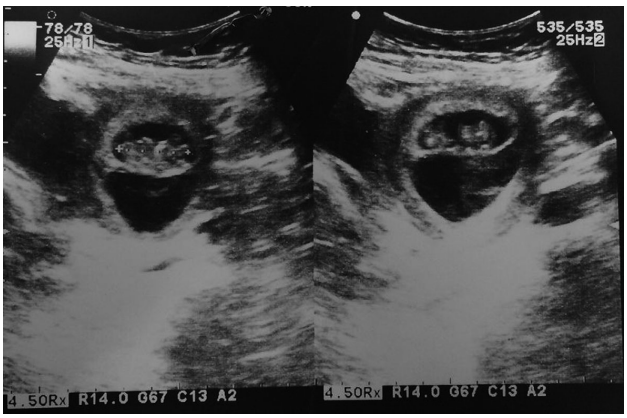

Figure 1. — Ultrasound image of fetuses at nine gestational weeks. Two gestational sacs separated with sonoluscent areas within them are interpreted as hematomas

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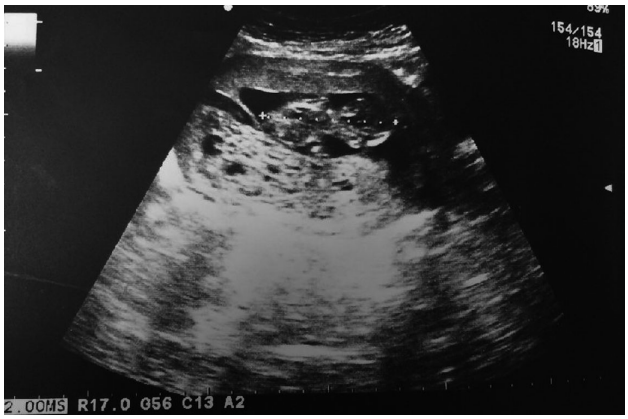

Figure 2. — Ultrasound image of fetuses at 17 gestational weeks. Alive fetus and the mass have cystic changes suggestive of hydatiform mole.

Figure 3. — Placenta and cyctic mass after birth. A grape-like mass with a diameter of 77×87 mm and the placenta belong to live fetus.

grape-like villi up to two cm in diameter, with a complete hydatidiform mole with mild trophoblastic hyperplasia. Serial serum beta-hCG were undetectable by three weeks post-delivery.

Discussion

Gestational trophoblastic disease (GTD) includes, partial hydatiform mole, complete mole, and choriocarcinoma. The coexistence of a complete hydatiform mole and a healthy fetus as a twin pregnancy is rare type of GTD and its incidence may be increasing with the greater use of assisted reproductive techniques [3]. The risks regarding CMCF pregnancy include significant antepartum haemorrhage, pre-eclampsia, preterm labour, and persistent trophoblastic disease [2].

The diagnosis of CMCF in the first trimester is difficult due to its rarity. Ultrasonography may miss molar changes in placenta in approximately 40% of cases [4], as in the present case with the first ultrasound.

Three possible types of pregnancy resulting in CMCF are described. First possible mechanism is a singleton pregnancy consisting of a partial mole and a live fetus. Second possible mechanism is a twin pregnancy with one placenta exhibiting a complete mole (no fetus), and the other placenta (sometimes in close approximation with other) sustaining a normal twin. The third possible mechanism is a combination of partial mole and a twin in one sac and a normal twin in the other [5].

Continuation of pregnancy in CMCF is associated with severe maternal complications. The most dangerous one o is persistent GTD. Most reports concerning CMCF revealed a very high risk (50–57%) for persistent GTD, however, in the largest reported series, only a 19% risk for developing persistent GTD was found [6]. When the CMCF is diagnosed, termination of pregnancy should be considered to prevent persistent GTD.

Most cases of molar pregnancies are diagnosed during the first trimester by ultrasonography. CMCF pregnancies are usually diagnosed in second trimester [5], as in the present case. Elevated hCG levels may suggest ultrasound findings in CMCF, but this is not reliable enough in twin pregnancies. A diagnosis of CMCF can be reached only by invasive methods. However, amniocentesis was not performed in the present case.

In a review, Vaisbuch *et al.* showed that live birth rate in CMCF pregnancies was only 33% and persistent GTD rate was 33% [5]. There is no optimal management strategy for CMCF pregnancies. In the past, termination of pregnancy was the main management method for CMCF, but recently published data suggest the continuation of pregnancy in CMCF [5, 7]. In the presence of ultrasonic-diagnosed malformation, termination of pregnancy would seem to be appropriate. Continuation of the pregnancy appears to be an acceptable option if the diagnosis is made near the fetal viability.

The risks of pregnancy continuation in cases of CMCF still remain uncertain due to rarity of this situation and possible complications (persistent GTD, preterm labour, preeclampsia) must be assessed in relation to CMCF with mother.

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