Complete hydatidiform mole coexisting with a live fetus

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Summary

The co-existence of a hydatidiform mole with a living fetus is a rare phenomenon. The condition is a dilemma with respect to the diagnosis and management of associated maternal (a risk of maternal complications, such as preeclampsia, hyperthyrodism, and a risk of malignancy) and fetal (elevated risk of spontaneous abortion, neonatal thyrotoxicosis) complications. A 27-year-old woman was referred to our hospital with a diagnosis of hydatidiform mole and live fetus. The pregnancy was unremarkable except for the complaints of excessive nausea and vomiting. Successive ultrasound examinations demonstrated a normally growing live fetus (14 weeks) alongside a normal placenta and an additional intrauterine echogenic mass with features of hydatidiform mole. Genetic amniocentesis at 18 weeks' of gestation showed normal diploid fetal karyotype. At 20 weeks of pregnancy, a control prenatal visit revealed intrauterine fetal death. The follow-up period for two years was unremarkable. In the case of a normal fetal karyotype and the absence of serious signs of maternal pathology, waiting until fetal viability is achieved can justifiably be proposed, however there is still a risk of prenatal complications such as intrauterine death.

Key words: Mole hydatidiform; Complications; Intrauterine exitus; Twin pregnancy.

Introduction

Gestational trophoblastic disease is a spectrum of cellular proliferations arising from the placental villous trophoblast. Hydatidiform mole (HM) is the most common form of gestational trophoblastic disease. HM refers to an abnormal pregnancy characterized by varying degrees of trophoblastic proliferation and vesicular swelling of placental villi associated with an absent or an abnormal fetus/embryo. Two types are described: partial hydatidiform mole in which there is a triploid karyotype, and complete hydatidiform mole (CHM), in which the karyotype is diploid. CHMs undergo early and uniform hydatid enlargement of villi in the absence of an ascertainable fetus or embryo [1].

The co-existence of hydatidiform mole with a living fetus is a rare phenomenon, with an incidence of one in 22,000 to one in 100,000 [2]. The condition is a dilemma with respect to the diagnosis and management of associated maternal (a risk of maternal complications, such as preeclampsia, hyperthyrodism, and a risk of malignancy) and fetal (elevated risk of spontaneous abortion, neonatal thyrotoxicosis) complications [3]. The aim of this case report was to describe a case of complete hydatidiform mole with a live fetus and uneventful pregnancy course.

Case Report

A 27-year-old woman with gravida 1, para 0, was referred to our hospital with a diagnosis of hydatidiform mole and live fetus. The pregnancy was unremarkable except for the complaints of excessive nausea and vomiting. She had a history of spontaneous conception.

Successive ultrasound (US) examinations demonstrated a normally growing live fetus (14 weeks according to CRL measurement) alongside a normal placenta and an additional intrauterine echogenic mass with features of hydatidiform mole. US evaluation of this mass revealed a central heterogeneous mass with numerous discrete anechoic spaces, which corresponded to diffuse hydatidiform swelling of the hydropic chorionic villi "snowstorm pattern" (Figure 1A-B). Based on these findings, a preliminary diagnosis of hydatidiform mole coexisting with a live fetus was suggested.

Chest X-ray, liver function tests, and 24-hour urine assessment did not suggest any abnormality. Laboratory evaluation revealed high -HCG (> 200,000 mIU/ml), and low TSH (0.01 mIU/ml (range, 0.27-4.2)), elevated levels of free T3 (12.46 mIU/ml, range 2.5-5.8 mIU/ml) and free T4 (38.40 MIU/ml, range 11.5-23.0 mIU/ml). Propylthiouracil (Propicil manufactured by *Dr. F. Frik* 50 mg, 150 mg/day) treatment was instituted to relieve hyperthyroidism symptoms. After three weeks of therapy the thyroid function tests were near normal levels.

Genetic amniocentesis at 18 weeks of gestation showed normal diploid fetal karyotype. Even though the risk of subsequent fetal complications and the potential of malignant transformation were discussed with the patient and her family, she was insistent on continuing the pregnancy.

Two weeks later (at 20 weeks of pregnancy) the control prenatal visit revealed intrauterine fetal death. Induction of abortion was performed with the delivery of a stillborn normal infant (450 g, grossly normal female fetus via normal vaginal route) and two adjoining placentas. The two parts were clearly distinguishable. The normal looking placenta weighed 160 g, while the other placenta which had a multicystic appearance weighed 332 g. On microscopic examination, the appearance of the multicystic placenta was consistent with a complete hydatidiform mole with mild trophoblastic hyperplasia and atypias. Unfortunately, attempted culture of molar tissue was unsuccessful. The follow-up period of two years was unremarkable.

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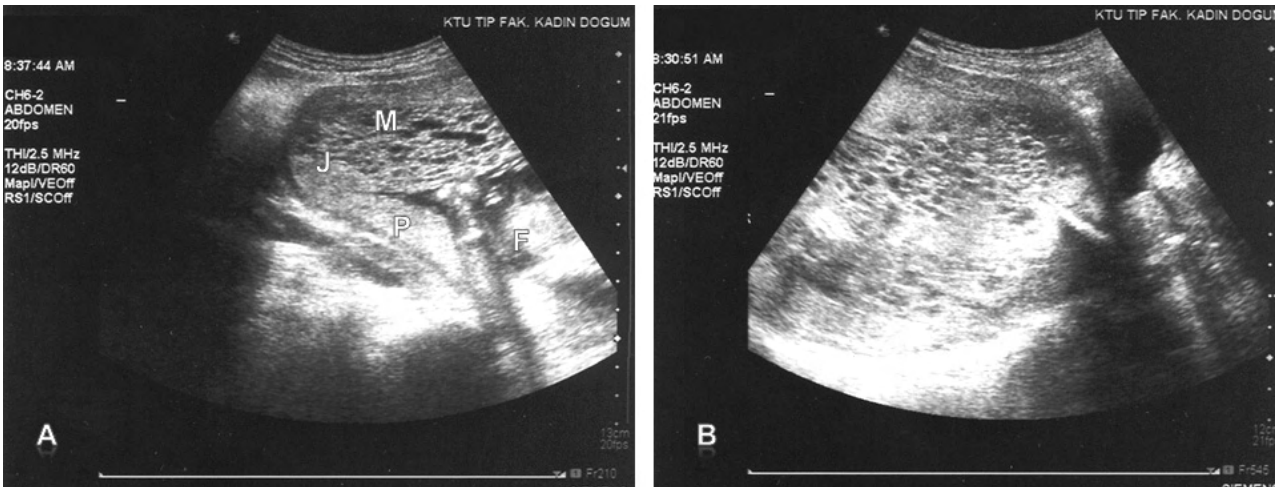

Figure 1. — (A) Sonographic appearance at 14 weeks of a twin pregnancy with co-existing hydatiform mole. P, normal placenta, F, fetus, M, area of hydatiform mole, J, junction of normal abnormal placeta. (B) Central heterogeneous mass with numerous discrete anechoic spaces which correspond to diffuse hydatidiform swelling of the hydropic chorionic villi "snowstorm pattern".

Discussion

Hydatidiform mole with a live co-twin fetus is a rare obstetric occurrence. When clinicians suspect a hydatidiform mole and a live co-twin fetus, they should keep in mind the following four possible associations: (a) dizygotic twin pregnancy having a normal fetus and hydatidiform mole; (b) dizygotic twin pregnancy having a normal fetus and a blighted ovum with normal constitutional karyotype and microscopic findings compatible with hydatidiform mole; (c) partial hydatidiform mole; and (d) mesenchymal dysplasia of the placenta [4]. Herein, we have reported a case of complete hydatidiform mole with an initially live fetus which was highly correlated with category a above.

A hydatidiform mole coexistenting with a twin live fetus is a different entity from a partial mole in that there is independent fertilization of two eggs as the origin of mole and normal fetuses. This rare condition also named as twin gestation syndrome (discordant diandric diploidy) in which it is theorized that one gestation develops normally while the other undergoes molar degeneration [5]. There are two types of spontaneous evolution during the second trimester of pregnancy: either the molar part becomes quiescent, allowing the pregnancy to continue, or it goes on growing extensively, leading to fetal death and maternal complications [3].

Based on recent guidelines, women with a complete hydatidiform mole and live fetus could be followed if certain conditions are made. When the fetus is sonographically normal, an amniocentesis or a fetal blood sampling should be carried out after 15 weeks. If the fetus is euploid and the mother is clinically well, she should be followed fortnightly with US assessment of fetal anatomy and growth, cervical length to assess risk of preterm labor. Women should be counseled about the risk of both maternal complications such as pre-eclampsia, hyperthyroidism and theca lutein cysts and fetal ones such as spontaneous miscarriage, intrauterine death and preterm labor [6]. Malignant trophoblastic disease may also possible in 55% of complete hydatidiform mole and fetus cases [7].

According to Sebire et al. data, viable fetuses can be expected in 40% of hydatidiform mole pregnancies that continue beyond 14 weeks of gestational age, while 60% of complete hydatidiform mole and healthy co-twin pregnancies, which are not electively terminated, will result in either intrauterine death of the co-twin or spontaneous pregnancy loss. Most of these events happen in the second trimester, before fetal viability (mostly before 24 weeks of gestational age) [8]. About 50 cases of viable fetuses have been reported up to now, although an increased risk of preterm delivery and of cesarean section is associated with molar complications [3, 8]. Complete hydatidiform mole and healthy co-twin pregnancies can also be associated with potentially life threatening maternal complications such as thromboembolic disease and severe pre-eclampsia. The risk of persistent gestational trophoblastic disease is similar to that after a singleton complete hydatidiform mole (16%) [8].

In conclusion the management of each such pregnancy must be discussed individually with the mother, who must be made aware of the risks of medical complications such as severe vaginal bleeding, preeclampsia and hyperthyroidism. In the case of a normal fetal karyotype and the absence of serious signs of maternal pathology, waiting until fetal viability is achieved can justifiably be proposed, however there is still a risk of prenatal complications such as intrauterine death.

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