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Partial regression of a hydatiform mole with coexisting live fetus in a twin gestation: case report

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Summary

Pregnancies resulting in viable fetuses are extremely rare in accompanying a hydatiform mole, often due to the development of maternal complications, including preeclampsia and vaginal bleeding. The risk for gestational trophoblastic neoplasm is another concern because of the delayed evacuation of the molar tissue. In this paper, the authors present a case of complete mole hydatiform with a live co-twin fetus (CHMLF) resulting in the delivery of a healthy male infant with the partial regression of the molar tissue and the decline of serum beta human chorionic gonadotropin (β-hCG) during the pregnancy. In the management of CHMLF, each patient must be considered individually and eligible patients can be followed in the absence of serious maternal complications. Serial ultrasound examinations and close clinical and laboratory surveillance of the mother are certainly indicated.

Key words: Complete mole hydatiform; Twin pregnancy; Gestational trophoblastic neoplasm.

Introduction

Pregnancies consisting of a live fetus accompanying a hydatiform mole are uncommon and comprise one in 22,000–100,000 pregnancies [1]. Among them, the pregnancies resulting in viable fetuses are extremely rare, often due to the development of maternal complications, including preeclampsia and vaginal bleeding. The risk for gestational trophoblastic neoplasm is another concern because of the delayed evacuation of the molar tissue. [2]

In this paper, the authors present a case of complete mole hydatiform (CHM) with a live co-twin fetus (CHMLF) resulting in the delivery of a healthy male infant with the partial regression of the molar tissue and the decline of serum beta human chorionic gonadotropin (β-hCG) during the pregnancy.

Case Report

A 25-year-old woman, gravida 2, para 1, at 22 weeks' gestation was referred to the present institution on suspicion of CHM and CHMLF. Her previous medical history was unremarkable and her obstetric history included one term vaginal birth. She had no problems with the pregnancy except for slight vaginal bleeding and mild hyperemesis by that time. Ultrasound examination revealed a live fetus with normal placenta and adjacent to the placenta a well-defined multi-cystic snowstorm-like mass which was compatible with CMH. This cystic area measured 12.5×7 cm (Figure 1). The amount of amniotic fluid was normal and there were no signs of growth retardation or fetal anomalies. The serum level of β-hCG was > 10,000 IU/ml. CHMLF was strongly suspected. The risk of possible maternal complications, fetal malformations, and subsequent malign transformation were explained and the couple was counseled for termination, but they chose to continue this pregnancy and declined any invasive prenatal testing to confirm the karyotype of the fetus. During the expectant management there were no major maternal complications (thyrotoxiosis, preeclampsia, anemia). Serial ultrasound examinations performed at two-week intervals, demonstrated normal fetal growth and a reduction in size of the molar tissue. Recurrent vaginal spotting continued in the second and third trimester. On the last ultrasound, molar tissue was observed thoroughly small on one side of the uterus. At 36 weeks, a cesarean section was performed because of vaginal bleeding, and a 2,630-gram male infant with an Apgar score of 9 and 10 at one and five minutes was delivered. The placenta and grape-like mass were removed manually. The pathologic examination of the placenta revealed 6.5×4 cm focal area which was compatible with CMH (Figure 2).

Immunohistochemical stains with the P57KIP2 anti-body were helpful in the differential diagnosis of complete hydatiform moles. Expression of P57KİP2, a paternally imprinted gene, is either absent or low in trophoblast in cases of complete moles in contrast to diffuse staining in partial moles and non-molar placentas. In present case, the villous stromal cells and cytotrophoblastic cells did not show P57KIP2 immunoreactivity, while the intermediate trophoblastic cells were positive (Figure 3) [3]. The karyotype in cultured cells from the molar area was diploid. The serum β-hCG level was 3,912 IU/ml at the delivery and patients followed by weekly serum β-hCG measurement. β-hCG levels decreased gradually. At second month of postpartum period the patient's β-hCG persisted at the levels of 56 and 60 IU/ml. Metotrexate with leucovorin rescue treatment was started and β-hCG level returned to normal after third course. No evidence of persistent gestational trophoblastic disease (PTD) was found at first year of postpartum period.

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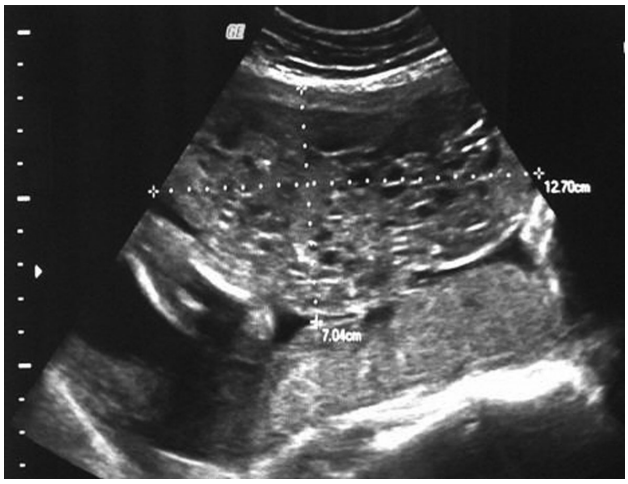

Figure 1. — Ultrasound photograph at 22 weeks' gestation showing complete hydatiform mole (upper part) and normal placenta with a fetus. The molar tissue was 12.7×7 cm.

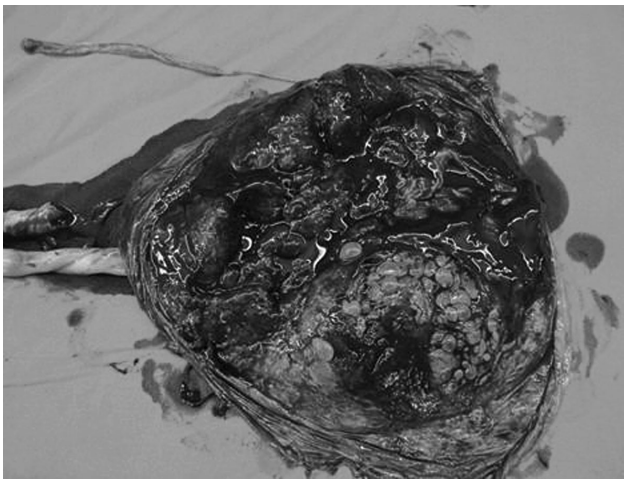

Figure 2. — Macroscopic photograph of the placenta (15×17 cm) and the molar tissue 6.5×4 cm.

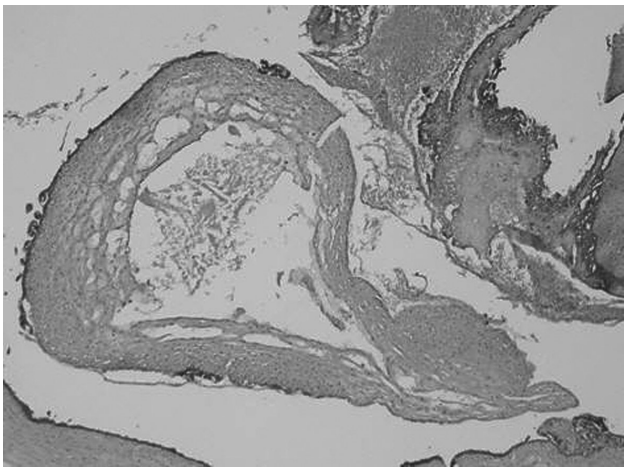

Figure 3. — p57KIP2 immunostaining of a complete hydatiform mole. The villous stromal cells and cytotrophoblastic cells do not show p57KIP2 immunoreactivity, while the intermediate trophoblastic cells were positive for p57.

Discussion

The CHMLF is a relatively uncommon event with a quoted range of incidence from one in 10,000 to one in 100,000 pregnancies [4]. Most of these cases either aborted or resulted in stillbirth and only a few with a living newborn. There are, overall, about 200 cases of twin pregnancy with CHMF documented to date in the literature and only 30 cases of twin pregnancy with CHMF resulting in a live birth documented in detail in the literature. [5]

Clinicians and parents with CHMLF cases encounter a clinical dilemma, as they have to decide between continuation or immediate termination of the pregnancy. The problems in the management of CHMLF involve the risks of fetal abnormality, malignant trophoblastic change, and severe maternal complications [6, 7]. The rate of pregnancy termination due to maternal complications is different in various reports. Fishman *et al.* [8] reported their termination rate as 71% because of maternal complications. This high rate may be due to spontaneous abortions and intrauterine deaths in their series. Conversely Sebire *et al.* [9] reported that only 4% of the pregnancies were terminated because of maternal complications.

As with the singleton molar pregnancies, some clinical criteria of aggressivity for the mother, such as maternal age, parity, uterine size, serum level of β-hCG before evacuation, signs of preeclampsia, and the existence of theca lutein cysts have been studied, but none of them predict progression to PTD [10]. Bristow *et al.* compared the clinical features of CHM in which the fetus remained viable and the pregnancies terminated or ended in stillbirth. In the non-viable group, the peak serum level of β-hCG was higher, and uterine height was greater than expected. In viable molar cases, the trophoblast was less aggressive or progressed to spontaneous degeneration. Spontaneous intrauterine fetal death appears to be very common in CHM with a live cotwin, mostly before 24 weeks [11].

The risk for PTD in the CHMLF cases is not higher than the risk in single molar pregnancies. Bristow *et al.* [11] reported that PTD occurs in 68.4% of CHMLF gestations delivered before fetal viability and 28.6% of those resulting in a living fetus. They concluded that the advanced gestational age required to produce a viable, surviving fetus is not an independent risk factor for the development of PTD. Several studies have shown that the risk level does not change with advanced gestational age [12, 13].

Marcorelles *et al.* [14] suggested that there are two types of spontaneous evolutions during the second trimester of pregnancy: either the molar part becomes quiescent, allow-

ing the pregnancy to continue, or it continues to grow extensively, leading to fetal death and maternal complications.

In the present case during the follow up, no maternal and fetal complications were observed and the pregnancy ended with the delivery of a live baby. The molar tissue regressed and the size of it decreased significantly during the pregnancy (partial regression). After delivery, patients were followed by serial β-hCG measurement; when the β-hCG levels persisted patients were diagnosed as gestational trophoblastic neoplasia (GTN). Because the patients were evaluated as low risk GTN, single-agent methotrexate treatment was administered. Negative β-hCG level was achieved and no recurrence was detected during a one-year period. In the present authors' opinion, in the management of CHMLF each patient must be considered individually and eligible patients can be followed in the absence of serious maternal complications. Serial ultrasound examinations and close clinical and laboratory surveillance of the mother are certainly indicated.

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