Case Reports

Twin pregnancy with a complete hydatidiform mole and a surviving co-existent twin

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Abstract

Hydatidiform molar change, characterized by abnormal fetoplacental development and placental villous trophoblast hyperplasia, results from genetically abnormal conception, which consists of paternally derived genetic material. We report a pregnancy in which molar change is documented in association with a live fetus, with a clear distinction both sonographically and pathologically, between the molar and non-molar regions of the placenta. A greater risk of post molar disease and the literature survey on indications to initiate chemotherapy immediately after delivery, is presented in detail.

Introduction

Hydatidiform mole may by classified as partial (PM) or complete (CM), on the basis of distinctive histopathological features and genetic abnormalities. Complete Hydatidiform mole with coexisting fetus (CHCF) is a rare entity occurring in 0.005-0.01% (1:22000 to 1:100000) of all pregnancies.¹ Pregnancies in which molar change has been reported in association with a live fetus generally represent dizygotic twin pregnancies in which one fertilization results in a complete hydatidiform mole (CM) and the other a normal co-twin.² In such cases, there is usually clear distinction, both sonographically and pathologically, between the molar and non-molar placenta. Exhibiting a different scenario a rare case has also been reported of a monozygotic singleton pregnancy with prenatally detected diffuse placental molar change, resulting in a phenotypically normal female infant at term, with apparent confined placental mosaicism for CM with a coexisting normal fetus.³

Case history

A 27-year old woman in her fourth pregnancy with a past caesarean section and two miscarriages was seen with persistent mild to moderate vaginal bleeding from 16 weeks gestation. Her booking investigations were unremarkable. Ultrasound examination revealed a singleton pregnancy with no fetal structural abnormalities and with confirmed dates. However numerous echoluent cysts were present at the lower edge of the placenta, suggestive of diffuse molar change. Serial ultrasound examinations performed at 3-weekly intervals demonstrated normal fetal growth and biometry and persistence of the apparent molar changes at one end of the placenta (Figure 1).

Of the well-recognised complications of hygroma gravidarum, preeclampsia or vaginal bleeding, only mild to moderate vaginal bleeding was observed from 16 weeks gestation.

At 36 weeks of gestation she was delivered by Caesarean section due to the reduction in fetal growth velocity and fetal heart rate decelerations on cardiotocograph. A phenotypically normal female infant, 1770 g, was delivered with Apgar scores of 6 at 1 min and 9 at 5 min. The placenta weighed 730 g and revealed widespread, diffuse vesicle formation at one end, consistent with molar change, clearly separated from the normal placenta (Figure 2).

Serum β-HCG titre showed a normal regression curve within six weeks postpartum.

Differential diagnosis

1. Singleton pregnancy consisting of a partial hydatidiform mole with an abnormal triploid fetus which usually dies in utero during the first half of pregnancy,
2. Twin pregnancy of a complete hydatidiform mole and a coexistent fetus (CHCF), and
3. Twin pregnancy with a partial hydatidiform mole and a coexistent fetus.

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Histopathology

Placental pathology reported by the Professor of Pathology, Faculty of Medicine Peradeniya, is as follows

Macroscopy:
1. Placenta 15 cm diameter with a 15 cm long umbilical cord. The major part of the specimen appeared unremarkable. Occasional vesicles were seen at the edge
2. Molar tissue 20 cm in diameter

Microscopy:
1. Sections show placenta with no evidence of molar tissue. The cord is normal
2. Sections show a hydatidiform mole. This is likely to be a twin pregnancy with a H. mole, in one.

Discussion

Twin pregnancy with complete hydatidiform mole and co-existent fetus (CHCF) resulting in a healthy take-home baby is rare, with only 30 cases documented in detail in the literature. The natural history is still unclear and complications particularly malignancy are poorly defined. Generally, partial mole is mostly associated with triploid fetuses that tend to die before the end of the first trimester and surviving fetuses after mid pregnancy are rarely encountered. On the other hand the fetus coexisting with complete
mole is usually associated with normal karyotype and has a chance of survival. Where genetic studies have been carried out in these cases, the normal and molar placentas have been genetically distinct.

It was reported that before 28 week of gestation, the chances of survival are minimal and the chance for continuation of pregnancy beyond this point is 60%. Nearly 40% of women who choose to continue their pregnancies have live babies.5

It is unclear whether a greater risk of post molar disease is associated with a more aggressive behavior of the molar tissue or with delayed delivery. Recent reports had pointed out that an advancement of gestational age did not appear to increase the risk of developing a post molar disease. There is no documentation to demonstrate that prolonging pregnancy to term would increase the incidence of invasive mole or choriocarcinoma.4

In a retrospective analysis of the Charing Cross Hospital Trophoblastic Disease Unit database, 126 twin pregnancies with hydatidiform moles and healthy co-twin were identified of which 77 pregnancies were histologically confirmed as CM and co-twin. 28 pregnancies lasted 24 weeks or more, resulting in 20 live births. Chemotherapy to eliminate persistent gestational trophoblastic disease (pGTD) was required in three of 19 women (16%; 95% CI 3-39) who terminated their pregnancies in the first trimester, and in 12 of 58 (21%; 95% CI 11-33%) who continued their pregnancies.7

In the light of current literature we did not see an indication to initiate chemotherapy immediately after delivery.

References