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Review

Individualistic approach to the management of complete hydatidiform mole with coexisting live fetus

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ABSTRACT

Complete hydatidiform mole with a coexisting live fetus (CHMCF) is a rare obstetric occurrence. So far, approximately 177 cases have been documented in the literature with consequent 66 live births. We report a review article along with two cases of CHMCF, one presenting as incomplete abortion and other continued as CHMCF but terminated because of antepartum hemorrhage. Both had histopathologically proven one normal and other complete molar placenta with coexisting normal fetus. No evidence of persistent trophoblastic disease was observed. The dilemma of continuation versus termination of pregnancy is being emphasized in the review of literature. Pregnancy complicated by CHMCF may result in a viable live born infant in approximately one third of the cases. A potentially viable fetus with CHMCF may result in normal live birth with antecedent high risk maternal complications. A decision of termination of pregnancy in all CHMCF will however nullify all the chances of a live birth. An individualistic approach and an informed doctor patient consensus may improve the likely outcome. Appropriate counseling of the mother regarding high incidence of antenatal complications plays an integral part of decision of continuation of such pregnancies.

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Introduction

Twin pregnancy with one complete hydatidiform molar gestation and coexistent live fetus (CHMCF) represents a very rare obstetric crisis. The estimated incidence is about 1 in 22,000–100,000 gestations [1]. Even though successful outcomes in the form of one live birth have been reported, continuation of pregnancy in its natural course till term is usually uncommon. Molar pregnancy may be partial or complete, usually distinguished by gross morphological, histopathological and chromosomal studies. Partial mole is an embryonic and chorionic tissue having a triploid karyotype (usually 69, XXY), due to fertilization of an apparently normal ovum by two sperms. A complete mole (CM) is usually diploid androgenic conceptus due to loss of the maternal nuclear

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genome fertilized by a single haploid sperm cell that duplicates, or by two sperms resulting in a 46, XX or 46, XY karyotype. This results in no embryo development due to lack of maternally derived genes and an excessive trophoblastic growth [1]. Management of these cases poses a clinical dilemma with inherent risk of severe maternal complications like abortion, preterm delivery, preeclampsia (PE), thyrotoxicosis, antepartum hemorrhage (APH), intrauterine fetal death, placenta accreta and gestational trophoblastic disease (GTD). Fetus also carries the significant risk of malformations, chromosomal abnormalities and extreme prematurity [2–4]. Even if fetal aneuploidy and malformations are ruled out with modern prenatal fetal diagnostic techniques, the decision of safe continuation of pregnancy is extremely difficult. We report two cases of CHMCF, both of which had to be terminated due to maternal complications despite efforts to continue the pregnancy. The approach to such cases along with review of literature is discussed.

Case 1

A 21-year-old primigravida presented at 15 weeks' period of gestation (POG) with sonographic features of bilateral theca lutein cysts, an intrauterine fetus, thickened heterogenous placenta and multiple cystic spaces suggestive of twin pregnancy with one live fetus and probable hydatidiform mole. She had repeated bleeding per vaginum since early conception. Despite explaining the fetal and maternal risks associated with continuation of pregnancy, patient decided to continue the pregnancy. Amniocentesis thus performed at 18 weeks POG to know fetal karyotype and Level II sonography with fetal echocardiography was normal. She was hospitalized with massive bout of bleeding per vaginum at 19 weeks POG. Decision of termination of pregnancy was thus taken in view of compromising hemodynamic status of the mother. Suction evacuation was thus performed for molar tissue. She aborted around 1 kg of molar tissue along with a male fetus of 230 g and normal placenta (Fig. 1A and B). She received about 10 units of blood, plasma and platelet concentrate during this period. The gross pathology revealed one specimen showing multiple fragmented tissue pieces composed predominantly of grape like clusters of cysts, the largest being 1 cm without identifiable fetal parts. The other specimen showed a macerated fetus without congenital anomalies, and a normal looking placenta. The microscopic examination of molar tissue showed edematous chorionic villi, conspicuous central cisterns and trophoblastic hyperplasia suggestive of complete hydatidiform mole. The placenta showed second trimester chorionic villi. She was followed up with serial serum beta hCG monitoring which returned to normal level in four months. At two year follow up, she did not reveal any signs of persistent trophoblastic disease.

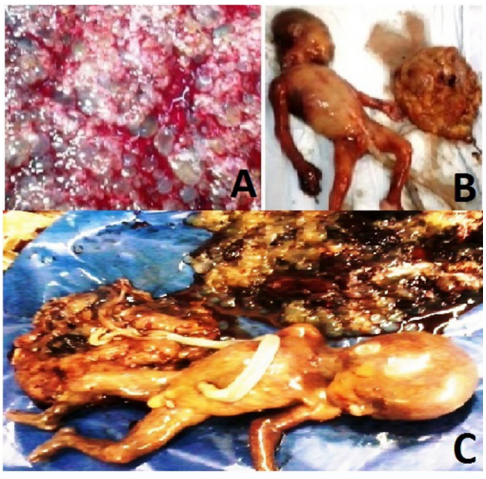

Fig. 1. (A) Case 1, molar tissues; (B) Case 1, abortus with normal placenta; and (C) Case 2, abortus with molar tissues and normal placenta.

Case 2

A 25-year-old 4th gravida, hospitalized at 19 weeks POG for excessive bleeding per vaginum. She was in the process of abortion and expelled plenty of molar tissue along with a live abortus of 200 g. Her pregnancy was completely unsupervised, without any obstetric ultrasound. She received 3 units of blood and plasma concentrates and underwent a complete suction evacuation of molar tissues. The gross pathology was suggestive of two placentas with one fetus. Fetus was attached with normal looking placenta through 20 cm long umbilical cord and grossly normal vasculature. The crown rump length of fetus was 15 cm and there was no facial dysmorphism or limb deformity, the other placenta showed multiple grapes like vesicles (Fig. 1C). The microscopic examination showed numerous edematous chorionic villi, conspicuous central cisterns and trophoblastic hyperplasia suggestive of complete hydatidiform mole. Section from normal looking placenta had second trimester chorionic villi. The fetus did not show any congenital malformation. She recovered well in 3 days of hospitalization. At six months follow up her beta HCG levels were normal. She was healthy at one year follow up without any evidence of persistent GTD (Gestational trophoblastic disease).

Methodology

We searched electronic medical databases using keywords twin pregnancy, complete mole, co-existent live fetus. Bibliographies of included articles reviewed and relevant article were included. All case reports and series published after 2009 were reviewed. Numbers of cases reported prior to 2009 were added to results. Eligible studies provided antenatal complications, pregnancy outcome and follow up of molar pregnancy. All antenatal complications in terms of abortion, antepartum hemorrhage, preeclampsia, preterm birth, postpartum hemorrhage and need of cesarean hysterectomy were reviewed. Pregnancy outcome in terms of abortion, normal delivery, cesarean section, live birth or neonatal death were recorded. Follow-up of complete molar pregnancy as gestational trophoblastic disease and need of chemotherapy were also evaluated. We collected all descriptive and outcome data.

Results

One hundred and fifty-nine cases and 56 live births were reported till 2009. Searched databases revealed 18 more case reports including 10 live births from 2009 till date. Thus approximately 177 such cases have been documented in the literature with only 66 live births (Table 1). All information about antenatal complications, pregnancy outcome and follow up of molar pregnancy was analyzed from the studies. Incidence of occurrence of antenatal complication like spontaneous abortion (7/18), antepartum hemorrhage (6/18), preeclampsia (2/18), preterm birth (11/18), postpartum hemorrhage (2/18) and gestational trophoblastic disease (3/18) was noted. Antepartum hemorrhage (No. 6/18) was the most common antenatal complication requiring induction of abortion, and termination of pregnancy. Two patients had to undergo total abdominal hysterectomy in view of postpartum hemorrhage. Follow up of molar pregnancy as gestational trophoblastic disease (18%) was also recorded.