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Review article

# Complete hydatidiform mole with co-existing fetus: Predictors of live birth

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ABSTRACT

Multiple gestation consisting of complete hydatidiform mole with co-existing fetus (CHMCF) is unusual. From our institution, we reported two cases with unfavorable obstetric consequences. The recommendation for antenatal management is still not distinctly determined. Therefore, the aim of this article was to review the literature according to the predictors of infant survival and to develop a management guidance for pregnancy with CHMCF. Between January 1, 1993 and May 31, 2016, 12 case series and 89 case reports comprising of 204 pregnant women were identified. The pregnancies successfully delivered 78 live births (37.86%). For clinical symptoms, pregnant women with antenatal complications, including pregnancy-induced hypertension (PIH), hyperthyroidism (HTD) and hyperemesis gravidarum (HG), significantly developed adverse perinatal events. Low hCG blood level was the best predictor of fetal survival ($P = 0.006$). We developed a model using logistic regression analysis which was enhanced by including an hCG cut-off level of 400,000 mIU/mL. On the basis of our intensive review, we suggest that the patient with CHMCF without antenatal obstetric problems especially PIH, HTD and HG together with initial serum hCG level less than 400,000 mIU/mL is a good candidate for pregnancy continuation and reaching fetal viability.



**Contents**



## Introduction

Hydatidiform mole is the most common benign lesion of gestational trophoblastic disease (GTD) with an incidence of about

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1 per 1000 pregnancies [1]. It is categorized as complete hydatidiform mole (CHM) or partial hydatidiform mole (PHM) on the basis of gross morphology, histopathology and cytogenetic analysis.

However, twin pregnancy consisting of complete hydatidiform mole with co-existing fetus (CHMCF) is a rare entity, occurring in 1 per 22,000–100,000 pregnancies [2]. The information about prenatal diagnosis and pregnancy outcome are restricted to case reports and small case series. Furthermore, the antenatal care of patients with CHMCF is indeterminate, because they have a risk of severe maternal complication such as massive vaginal bleeding, pre-eclampsia, hyperthyroidism, hyperemesis gravidarum and malignant trophoblastic change as well as perinatal mortality.

Therefore, the objective of the present study was to summarize the clinical details of maternal and fetal conditions affected by CHMCF and to identify the predictors of live-born infant. We expected that this comprehensive information will provide guidance for obstetrical management and counseling.

## Materials and methods

We reported two cases encountered at our institution of twin pregnancy consisting of a CHM with a co-existing fetus. Informed consent was obtained from the patients for being included in this study and the study was approved by the Ethics Committee, Faculty of Medicine, Prince of Songkla University, Songkhla, Thailand.

### *Case 1*

An ultrasound scan identified a normal fetus, without gross anomaly, alongside a normal placenta located at the upper part of the anterior uterine wall and adjacent abundant vesicular tissue without fetus consistent with complete hydatidiform mole (CHM) within the lower uterine segment, completely covering the cervical os (Fig. 1).

Because of several maternal complications including early onset severe pre-eclampsia, hyperthyroidism and massive blood loss, stabilization and termination of pregnancy was performed via hysterotomy under general anesthesia one day after admission (19 $^{5/7}$ weeks' gestation).

The pathological diagnosis confirmed single placental plate consisting of CHM combined with normal placenta (Fig. 2). The fetus was male gender without organ anomaly, the karyotype was normal (46,XY). The cytogenetic analysis from the mole was 46,XX.

The postoperative period was uneventful. Maternal serum hCG level decreased to 28,571 mIU/mL on the 3rd post-operative day, and was within the normal range 15 weeks after termination of pregnancy.

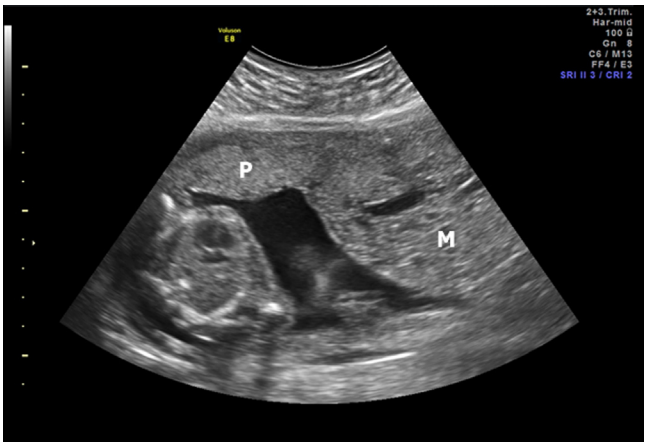


**Fig. 1.** Case 1: Trans abdominal ultrasonography at 19 weeks' gestation demonstrating normal placenta at the upper part of the anterior uterine wall and multiple vesicular tissue consistent with CHM at lower uterine segment (P, normal placenta; M, CHM).

### *Case 2*

Prenatal ultrasound examination showed a live fetus compatible with gestational age and a normal looking placenta at the anterior wall. An additional placenta with multiple cystic appearance was identified at the posterior wall of the uterus (Fig. 3a). Bilateral theca lutein cysts were documented (Fig. 3b).

Pregnancy was terminated due to deteriorating symptoms of thyrotoxicosis at 16 $^{6/7}$ weeks' gestation. Histopathological examination confirmed a normal male fetus with normal placenta combined with CHM (Fig. 4). The cytogenetic analysis from fetus was 46,XY and from part of the mole was 46,XX.

The maternal serum hCG level decreased to a normal range within 18 weeks after termination of pregnancy. However, the subsequent measurements demonstrated a further increase, without evidence of metastasis to any other organs. Chemotherapy with 3 cycles of weekly methotrexate 40 mg/m$^2$ was prescribed. The hCG level normalized 1 week after starting the treatment with no evidence of recurrent disease after one year of follow up.

### *Literature review*

The PubMed database was searched electronically from January 1, 1993 to May 31, 2016 using the terms of 'complete hydatidiform mole' OR 'molar' AND 'coexisting fetuses' OR 'co-existing fetuses' OR 'coexistent fetuses' OR 'co-existent fetuses' OR 'healthy co-

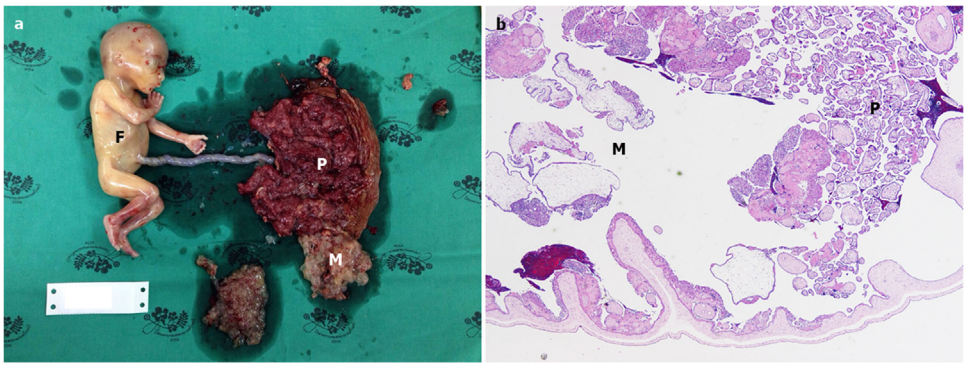


**Fig. 2.** Case 1: **a** Photograph of gross specimen shows a fetus with normal placenta and a part of mole **b** Histological examination demonstrated part of complete hydatidiform mole characterized by diffused swelling of villi with trophoblastic proliferation and normal placenta (F, fetus; P, normal placenta; M, CHM).