

Complete Hydatidiform Mole and Surviving Coexistent Twin – A Case Report

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ABSTRACT

Hydatidiform mole with a coexistent fetus is a rare occurrence with an incidence of 1 per 22,000 – 100,000 pregnancies. It is associated with persistent gestational trophoblastic tumour. Hence an early and correct diagnosis is imperative to plan subsequent management of such patients. We report a case of a primigravida who presented with vaginal bleeding at early second trimester. Expectant management was carried out for her pregnancy which finally ended in an abortion. The pathology, clinical findings and current management of this rare entity is discussed.

Keywords: hydatidiform mole, coexistent fetus

INTRODUCTION

The incidence of twin pregnancy consisting of complete hydatidiform mole and a coexistent fetus is 1 per 22,000 – 100,000 pregnancies⁽¹⁾. This is a condition where the twin pregnancy consists of one twin which is a normal fetus with normal placenta and the other twin a complete hydatidiform mole. This condition is different from a partial molar pregnancy where there is a single conceptus with a singleton fetus and an abnormal placenta. This distinction is important as a partial molar pregnancy generally is associated with triploidy resulting in growth retardation and multiple congenital malformations. They also have a relatively low incidence of producing persistent gestational trophoblastic tumour (GTT) compared to complete moles where the patients have a 20 percent risk of developing persistent GTT⁽²⁾.

Advances in ultrasound and cytogenetics now permit the diagnosis of complete hydatidiform mole and a coexistent fetus (CHAF) at early second trimester. This causes a dilemma in terms of subsequent management of the pregnancies as a decision has to be taken with regards to early intervention (termination of pregnancy) or continuation of pregnancy. Our patient presented to us at 13 weeks of gestation and was diagnosed as CHAF shortly after that. A decision to continue her pregnancy was made after discussion with the patient

and ruling out contraindications to continuation of her pregnancy.

CASE REPORT

A 30-year-old primigravida was admitted to the gynaecological ward on 11 August 1997 at 13 weeks of gestation for vaginal bleeding. She was initially treated as threatened abortion and was thought to have a uterine fibroid on ultrasonography. She was discharged on 13 August 1997. Two weeks later she was admitted with excessive vomiting and recurrence of vaginal bleeding. On examination, a diffusely enlarged goitre was found. The fundal height corresponded to 18 weeks. She was mildly dehydrated. Her thyroxine profiles revealed the TSH levels as 0.1 micrograms/dl and the total T4 levels as 20 micrograms/dl. She was treated with fluids for hyperemesis gravidarum. Her serum electrolytes were normal. Detailed ultrasonography of the pelvis confirmed a hydatidiform mole with a viable coexisting normal pregnancy corresponding to 16 weeks gestation (Fig 1). Fetal survey did not reveal any structural abnormalities. A diagnosis of hydatidiform mole with co-existing viable pregnancy was made. The patient was counselled on the complications of the pregnancy and the modes of management. She opted to continue with her pregnancy.



Fig 1 – Ultrasonography of complete hydatidiform mole and surviving coexistent twin at 16 weeks of gestation.

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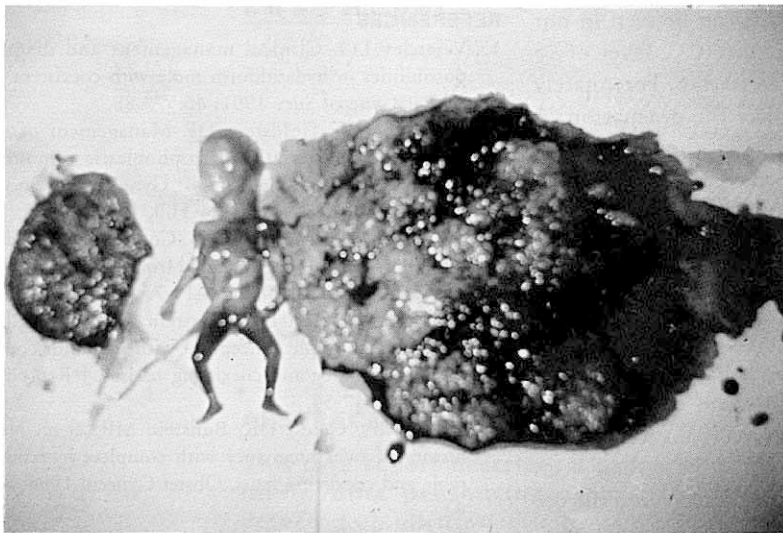


Fig 2 – The delivered fetus with normal placenta on the right and the molar tissue on the left.

Her blood pressure was 120/80 mmHg and haemoglobin was 6.4 g%. She was transfused two pints of packed cells. Her serum beta-HCG levels at the time of diagnosis was > 200,000 mIU/ml. She continued to stay in the ward as she was still having vaginal bleeding. One week later her vaginal bleeding increased in amount. The patient then aborted spontaneously. A normally formed male fetus weighing 235 gm with a normal placenta was confirmed with a separate mass of molar tissue (Fig 2). A dilatation and curettage was done following the abortion. A histopathological diagnosis of gestational trophoblastic disease was confirmed.

She was subsequently discharged two days later after a chest x-ray revealed no abnormalities. She was seen one week later in the gynecological clinic and an ultrasonography of the uterus revealed an empty cavity. The serum beta-HCG levels were monitored fortnightly and it declined very gradually (Table I). It became less than 2 mIU/ml after six months post abortion. Her thyroid function also reverted to normal after three months. She has been advised to use barrier methods for contraception and is still on regular follow-up.

Table I – Trend of fall of beta-HCG levels post abortion

Date	Values mIU/ml
26.09.97	> 200,000
08.09.97	86,922
15.09.97	3,917
22.09.97	777
07.10.97	663
23.10.97	523
09.11.97	404
06.12.97	126
06.01.98	75
08.02.98	1.5
09.03.98	1

DISCUSSION

Twin conception consisting of a hydatidiform mole and coexisting fetus (CHAF) is a rare entity. Hence detailed information on the clinical features of the natural history of CHAF is limited in the literature. Many of the patients with CHAF present with vaginal bleeding. In one review, up to 55% of the patients presented in this manner⁽¹⁾. They can also present with inappropriate uterine growth and pre-eclampsia in the first and second trimester. Our patient presented to us with vaginal bleeding and a uterus which was larger than her dates.

Establishment of an early diagnosis of CHAF is important to avoid fatal complications like severe pre-eclampsia and uncontrolled vaginal bleeding. Ultrasonographic diagnosis can be made as early as 11 weeks⁽³⁾. In our patient, the diagnosis was only made at 15 weeks of gestation. Misinterpretation of ultrasonographic findings of CHAF can be made in up to 30% of cases⁽⁴⁾. Most of the time they are diagnosed to have placental haematomas or degenerating fibroids.

The dilemma in the management of patients with CHAF after the antenatal diagnosis is whether to manage these patients expectantly or to terminate the pregnancy immediately. The optimal management is still uncertain as there are limited data to guide clinical decisions because most published reports consist of case reports in which the distinction between a partial mole and CHAF has not been established. There is a fear that CHAF pregnancies may end up as persistent GTT. It has been documented that these pregnancies are at high risk of developing persistent GTT compared with the singleton counterpart⁽⁵⁾. Others have reported cases where the pregnancies have been managed expectantly with the delivery of a live fetus and these patients didn't develop persistent GTT⁽⁶⁾. Our patient was counselled regarding persistent GTT but she was keen to continue her pregnancy.

Apart from persistent GTT one has to anticipate other associated complications of a CHAF. These include hyperemesis gravidarum, hyperthyroidism and trophoblastic embolization. The presence of any one of the above may be an indication for immediate evacuation of the pregnancy. In a clinically stable patient, an additional relative indication for therapeutic intervention is uterine enlargement markedly greater than that predicted by dates. Our patient was managed conservatively up to 16 weeks gestation when she started to have intractable vaginal bleeding. She also had hyperemesis gravidarum and hyperthyroidism. Our decision then was to evacuate her pregnancy but fortunately for her, she aborted spontaneously.

Whatever the antenatal management approach for patients with CHAF pregnancies, close monitoring of the serum beta-HCG levels are warranted both antenatally and postnatally. A high level of pre-evacuation serum beta-HCG level is not an indication for termination of pregnancy but it may predict the development of persistent GTT as this may be a reflection of the aggressiveness of

the trophoblastic growth. This can be seen in our patient who had a serum beta-HCG level of > 200,000 mIU/ml prior to evacuation. Fortunately for her the levels declined after the evacuation. A point of note in this patient is that the serum beta-HCG levels took almost 6 months to return to normal levels (< 2 mIU/ml). In a 'singleton' complete hydatidiform mole, the serum beta-HCG levels usually attain normal levels within 2 months of evacuation.

The management of patients with CHAF pregnancies remains clinically challenging because of their propensity to develop medical complications and persistent GTT. It is expected that such patients will be encountered more frequently as there is an association between CHAF pregnancies and the use of ovulation induction and assisted reproduction⁽⁷⁾. As such our understanding of CHAF pregnancies can only be advanced by reporting the management experience of such patients.

REFERENCES

1. Vejerslev LO. Clinical management and diagnostic possibilities in hydatidiform mole with coexistent fetus. *Obstet Gynecol Surv* 1991; 46:577-88.
2. Berkowitz RS, Goldstein DP. Management of molar pregnancy and gestational trophoblastic tumours. In: Knapp RC, Berkowitz RS, eds, *Gynecologic Oncology*, 2nd Ed. New York: Mc Graw Hill, 1992:328-38.
3. Sauerhes EE, Salem S, Fayle B. Coexistent hydatidiform mole and live fetus in the second trimester: An ultrasound study. *Radiology* 1980; 135:415-7.
4. Stellar MA, Gerest DR, Bonstein MR, et al. Clinical features of multiple conception with partial or complete molar pregnancy and coexisting fetuses. *J Reprod Med* 1994; 39:147-54.
5. Stellar MA, Gerest DR, Bonstein MR, et al. Natural history of twin pregnancy with complete hydatidiform mole and coexisting fetus. *Obstet Gynecol* 1994; 83:35-42.
6. Thomas EI, Pryces WI, Maltby FL, Duncan SLB. The prespective management of a coexistent hydatidiform mole and fetus. *Aust NZ J Obstet Gynecol* 1987; 27:343-5.
7. Grenman SE, Salmi T, Meurman L. Post molar trophoblastic disease following coexisting molar pregnancy and living fetus subsequent to clomiphene citrate therapy. *Int J Gynecol Obstet* 1990; 32:381-5.