

Case Report

Molar Pregnancy And Co-Existent Foetus: A Report Of Two Cases

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ABSTRACT

Molar pregnancy with a co-existent foetus will lead to preterm labour, severe preeclampsia or bleeding in most of the cases and may need urgent intervention. However, if it does not become complicated with preeclampsia or preterm labour, the outcome is usually good, with minimal post partum complications and so such pregnancies can be managed with watchful waiting and close observation. The first case was a 29 year-old at 19 weeks of gestation, with hypertension, oedema and severe epigastric pain. Karyotypic assessment of the contents of the uterus revealed a 46-XX foetus with no chromosomal abnormality, as well as the molar placenta also suggesting a complete mole with 46-XX. The second case was a 19 year old woman in labour. A pathological study of the delivered contents of the uterus revealed a complete hydatidiform mole and a normal placenta.

Key Words: Pregnancy, ARTs, Mole, Foetus.

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pregnancies may progress to this condition [2],[3],[4].

It is difficult to determine the true occurrence of this abnormality. Suspicion may arise when an ultrasound scan identifies a foetal pole along an abnormal placenta. Careful clinical evaluation and detailed ultrasonic examination of the placenta, as well as a chromosomal analysis, are needed for definite diagnosis. Differential diagnosis consists of: (a) singleton pregnancy consisting of a partial hydatidiform mole with an abnormal triploid foetus which usually dies in utero during the first half of pregnancy, (b) twin pregnancy of a complete hydatidiform mole and a coexistent foetus (CHCF), and (c) twin pregnancy with a partial hydatidiform mole and a coexistent foetus. The CHCF is extremely rare and its management is challenging because the foetus may be viable. It carries a significant risk of severe complications such as preterm delivery, either spontaneous or induced due to preeclampsia or other pregnancy complications and development of persistent gestational trophoblastic disease (GTD) [5].

Introduction

In the late 1970s, Vassilakos et al. first described two distinct pathological entities, partial and complete hydatidiform moles (CHM), with different mechanisms of origin based on cytogenetic analysis [1]. These may rarely occur in the presence of a foetus. Twin pregnancy with a complete hydatidiform mole and a normal foetus is a truly rare occurrence encountered in clinical obstetrics. It is estimated that 1:22000 to 1:100000 of

In this article, we present two cases of molar pregnancy with co-existent foetus and their outcomes and a brief review on clinical features and previous studies and reports about this condition.

Case Report

The first case was a 29 year-old gravid 1, Para 0 woman presenting at 19 weeks of gestation with hypertension, oedema and severe epigastric pain. She had a 10 year history of infertility because of the male factor (azospermia) and this pregnancy resulted from intracytoplasmic sperm injection (ICSI). At presentation, the patient had a blood pressure of 170/120 mmHg and in the abdominal examination, there was a moderate hypogastric tenderness and foetal height was abnormally higher than the expected height for gestational age. Also a 2⁺ oedema was detected on the face, hands and legs.

An ultrasonic examination revealed a dead foetus and some additional vesicular patterns, suggesting hydatidiform mole. The serum level of β -hCG was 73000 mIU/ml. On the second day of admission, the patient was operated on and the contents of the uterus were delivered. The delivered materials consisted on a dead foetus and a lot of molar tissues weighing approximately 1100 grams [Table/Fig1]. The microscopic examination of the molar tissue revealed a complete hydatidiform mole. The karyotype of the foetus was 46-XX, with no chromosomal abnormality. Genetic analysis of the molar placenta also showed a complete mole with a 46-XX karyotype.



(Table/Fig 1) Delivered Material Consisting Of A Dead Fetus And Molar Tissue Weighing Approx.110 Gms.

After hysterotomy, the patient complained of dyspnoea and so, a chest CT-Scan was done in which a pulmonary oedema was observed, however, although there was no signs of invasion of molar tissues, persistent GTT could

not be ruled out. Therefore, single agent chemotherapy (Methotrexate) was administered to the patient. In the follow up, the level of β -hCG decreased to normal values after 10 weeks and complete remission was diagnosed.

The second case was a 19 year old gravid 1 woman with labour. The gestational age was 35 weeks according to LMP and the patient was admitted because of preterm labour and cervical dilatation. Magnesium sulfate was administered, but however, the treatment was discontinued because of progression of labour and the delivery was done. A male live foetus with normal placenta and a molar tissue were delivered [Table/Fig 2]. The patient had no signs of hyperemesis gravidarum, hypertension or preeclampsia during pregnancy; but she had severe vaginal bleeding between 12-20 weeks of gestation, when sonographic study showed a normal foetus and a cystic heterogeneous mass that was considered as a subplacental haematoma.



(Table/Fig 2) Molar Tissue Was Delivered Along With Male Live Foetus.

Postpartum, there was no abnormal bleeding. Further evaluation showed normal a chest X-ray and normal β -hCG levels, 3 and 5 weeks after delivery. Pathological studies revealed a complete hydatidiform mole and a normal placenta. However, there were no signs or symptoms of invasion and the patient was discharged without any treatment.

Discussion

The coincidence of a hydatidiform mole with a term foetus is one of the truly rare occurrences encountered in clinical obstetrics. Most presenting symptoms of CHCF are similar to those of a singleton hydatidiform mole and include bleeding, inappropriate uterine growth, and a sudden appearance of hypertension or preeclampsia in the first or second trimester [6],[7]. However, there are several clinical features that distinguish between a singleton

hydatidiform mole and CHCF, i.e., CHCF is diagnosed later and has markedly larger uterine size and higher levels of β -hCG prior to evacuation [3],[5]. When the possibility of CHCF is raised, detailed ultrasonography, both for possible placental pathology and for foetal abnormalities, should be carried out by an expert [7]. Also, prenatal genetic diagnosis by chorionic villous biopsy or amniocentesis is recommended to evaluate the karyotype of the foetus, because it might be important in the strategy of management [8].

Multiple pregnancies and hydatidiform moles have been frequently reported in association with assisted reproductive technologies, i.e. intracytoplasmic sperm injection (ICSI) [9],[10],[11]. Although it was once speculated, there seems to be no relationship between infertility treatment and HM [1]. However, multiple pregnancies resulting from infertility treatment may increase the risk of HM [12]. Hamanoue et al, 2006, reported a case of complete hydatidiform mole and a normal live birth following ICSI [13]. They have recognized a molar pregnancy with a diploid genotype and a viable foetus at the 7th week of gestation [13]. Also, Lin et al, 2005, and Vandenhove et al, 2008, reported other cases of CHM with a coexistent foetus [12],[14]. These studies, in addition to other reports, suggest that most cases of HM following ICSI are of the complete type, with diploid genotype [13], as it was observed in our study.

Until now, four cases of normal live birth following molar pregnancy after ART have been reported [Table/Fig 3][11],[12],[13],[15]. In several previous reports, vaginal bleeding was an important feature of molar pregnancy [11],[16]; however, in the present case, there was no vaginal bleeding. Also, our second case had no signs of hyperemesis gravidarum, preeclampsia or vaginal bleeding.

(Table/Fig 3) Summary of recent reports of hydatidiform mole with co-existent fetus

Method	Presentation	Outcome	Reference
IVF	Vaginal bleeding, preterm labor	Alive	Cheng(1995)
GIFT	Vaginal bleeding	Termination	Shozu(1998)
IVF	Vaginal bleeding	Alive	Monte-de-Oca-Valera(1999)

Marcorettes and colleagues reported four cases of complete hydatidiform mole (CHM), coexisting with a live co-twin [8]. In this study, all pregnancies were spontaneous, two ended with the delivery of a live-born baby and the other two were terminated. They had suggested that 'in the case of a normal foetal karyotype, it is justifiable to await developments in the absence of maternal complications' [8].

In summary, a viable foetus in the presence of molar tissue is a rare condition that may occur in normal conception [17] or ARTs [11],[12],[13]. Although vaginal bleeding is the most common presentation in molar pregnancies, it seems to be less common in molar pregnancies associated with a co-existent foetus and also a persistent mole is uncommon in this condition.

According to our study and similar previous reports, a molar pregnancy with a co-existent foetus will lead to preterm labour, severe preeclampsia or bleeding which is associated with high mortality and morbidity rates in most of cases and may need urgent intervention,. However, if it does not become complicated with preeclampsia or preterm labour, the outcome is usually good, with minimal post partum complications and so such pregnancies could be managed with watchful waiting and close observation.

References

- [1]. Vassilakos P, Riotton G, Kajii T. Hydatidiform mole: two entities. *Am J Obstet Gynecol* 1977; 127:167- 70.
- [2]. Azuma C, Saji F, Takemura M, Ohashi K, Kimura T, Miyake A, Tagaki T, Tanizama O: Triplete pregnancy involving complete hydatidiform mole and two fetuses: genetic analysis by deoxyribonucleic acid fingerprint. *Am J Obstet Gynecol.* 1992; 166:664-67.
- [3]. Steller MA, Genest DR, Bernstein MR, Lage JM, Goldstein DP, Berkowits RS: Natural history of twin pregnancy with complete hydatidiform mole and coexisting fetus. *Obstet Gynecol.* 1994; 83:35- 42.
- [4]. Choi-Hung SR, Genest DR, Crum PR, Berkowits R, Goldstein DR, Schofield DE: Twin pregnancies with complete hydatidiform mole and coexistent fetus. *Hum Pathol* 1995; 26:1175-80,
- [5]. Bristow RE, Shumway JB, Khouzami AN, Witter FR. Complete hydatidiform mole and surviving coexistent twin. *Obstet Gynecol Surv* 1996; 51:705-9.
- [6]. Miller D, Jackson R, Ehlen T, McMurtie E: Complete hydatidiform mole coexistent with a twin live fetus. Clinical course of four cases with

- complete cytogenetic analysis. *Gynecol Oncol* 1993; 50:119-23,
- [7]. Block MF, Merill JA: Hydatidiform mole with coexistent fetus. *Obstet Gynecol* 1982; 60:129-34.
- [8]. Marcocelles P, Audrezet MP, Le Bris MJ, et al. Diagnosis and outcome of complete hydatidiform mole coexisting with a live twin fetus. *Eur J Obstet Gynecol Reprod Biol* 2005; 118(1):21-7.
- [9]. Petignat P, Senn A, Hohlfeld P, Blant SA, Laurini R, Germond M. Molar pregnancy with a coexistent fetus after intracytoplasmic sperm injection. A case report. *J Reprod Med* 2001; 46(3):270-4.
- [10]. Kwon HE, Park EJ, Kim SH, Chae HD, Won HS, Kim CH, Kang BM. A case of twin pregnancy with complete hydatidiform mole and coexisting fetus following IVF-ET. *J Assist Reprod Genet* 2002; 19(3):144-8.
- [11]. Cheng PJ, Chang FH, Liang CC, Chang TC, Soong YK, Hsueh C. A twin pregnancy with a hydatidiform mole and an alive, coexistent baby after in vitro fertilization and embryo transfer. *J Assist Reprod Genet* 1995; 12(6):389-92.
- [12]. Lin YH, Huang LW, Hwang JL. Complete hydatidiform mole with a coexisting fetus resulting from in vitro fertilization. *Int J Gynaecol Obstet* 2005; 90(1):74-5.
- [13]. Hamanoue H, Umezumi N, Okuda M, et al. Complete hydatidiform mole and normal live birth following intracytoplasmic sperm injection. *J Hum Genet* 2006; 51(5):477-9.
- [14]. Vandenhove M, Amant F, van Schoubroeck D, et al. Complete hydatidiform mole with co-existing healthy fetus: a case report. *J Matern Fetal Neonatal Med* 2008; 21(5):341-4.
- [15]. Montes-de-Oca-Valero F, Macara L, Shaker A. Twin pregnancy with a complete hydatidiform mole and co-existing fetus following in-vitro fertilization: case report. *Hum Reprod* 1999; 14(11):2905-7.
- [16]. Shozu M, Akimoto K, Kasai T, et al. Hydatidiform moles associated with multiple gestations after assisted reproduction: diagnosis by analysis of DNA fingerprint. *Mol Hum Reprod* 1998; 4(9):877-80.
- [17]. Ogura T, Katoh H, Satoh S, et al. Complete mole coexistent with a twin fetus. *J Obstet Gynaecol Res* 2006; 32(6):593-601.