# Twin Pregnancy With A Partial Hydatidiform Mole After In Vitro Fertilization: A Case Report

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**Abstract**

Twin pregnancy with a hydatidiform mole after in vitro fertilization (IVF) is less common than after natural conception. Such pregnancies are associated with a high risk of maternal and fetal complications, and fetal anomalies; many women discontinue their pregnancies. We report the case of a 33-year-old primipara who developed massive vaginal bleeding and abdominal pain at 10 + 6 weeks of gestation. She had undergone IVF. At 8 weeks of gestation, a hydatidiform mole was diagnosed but the patient decided to continue her pregnancy. On arrival in the emergency room, ultrasonography revealed a fetus without a heartbeat and focal cystic spaces in the placenta, suggesting a missed twin abortion. Emergency dilatation and curettage were performed, and revealed a normal placenta with a male fetus and partial mole. After operation, the patient's serum β-hCG level began to drop rapidly, but had returned to normal 3 months later. In the past, termination has traditionally been recommended for twin pregnancies with hydatidiform moles. However, pregnancy can be continued if highly desired, as in our case. Such patients require intensive counseling on complications, the risk of a persistent gestational trophoblastic tumor, and the poor fetal outcome rate. Fetal growth and maternal complications must be closely monitored during the entire pregnancy.

## INTRODUCTION

A hydatidiform mole is a benign, gestational trophoblastic disease classified as complete or partial depending on the histological characteristics and embryo status. A partial mole pregnancy with a fetus is very rare; the incidence is 1/10,000–1/20,000.1 When assistive reproductive techniques (ARTs) are used, the incidence is lower than after natural conception, but is not zero.2 A partial mole pregnancy with a fetus is associated with a high risk of various complications, progression to a persistent gestational trophoblastic tumor, and a poor fetal outcome. Thus, termination is often recommended.3 However, continuation is possible if the pregnancy is highly desired, i.e., after in vitro fertilization (IVF).

## CASE REPORT

A 33-year-old primipara at 10 + 6 weeks of gestation presented to our emergency room with massive vaginal bleeding and mild abdominal pain. Pregnancy had been accomplished via IVF; vaginal spotting had commenced very early. Initially, we considered that she had a twin pregnancy. However, at 8 weeks of gestation, only one fetus (with a heartbeat) combined with a hydatidiform mole were ultrasonographically observed in another hospital. She was under close weekly monitoring when sudden, massive vaginal bleeding and abdominal pain developed. On arrival at our emergency room, the initial vital signs were 130/80-120-20-36.5, and old clots and placenta-like material were discharged through the vagina. She was anemic (Hb 9.4 g/dL) and the serum β-hCG level was elevated to 3,862,000 IU/mL. Ultrasonography revealed a fetus without a heartbeat and focal cystic spaces in the uterine placenta, suggesting twin pregnancy with a hydatidiform mole (Figures 1 and 2). The thyroid function test and chest X-ray were normal. Under a diagnosis of a missed abortion, emergency dilatation and curettage were immediately performed and tissue samples were sent to the Department of Pathology (Figure 3). The histopathological report confirmed a normal placenta with a male fetus and partial hydatidiform mole. The patient was treated conservatively (with antibiotics) and recovered quickly without any complications. The serum β-hCG levels were 60,037 and 18,990 IU/mL on days 1 and 3 after treatment. The patient was discharged and scheduled for regular serum β-hCG checkups while taking a contraceptive. Three months later, her serum β-hCG level was completely normal and no relapse

was noted during the 1-year follow up.

**Figure 1**

Partial mole of uterus on TAUSG

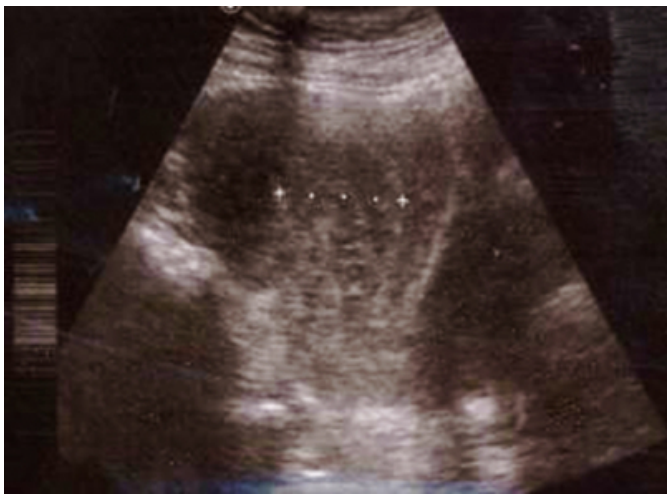

**Figure 2**

Co-existing fetus on TAUSG

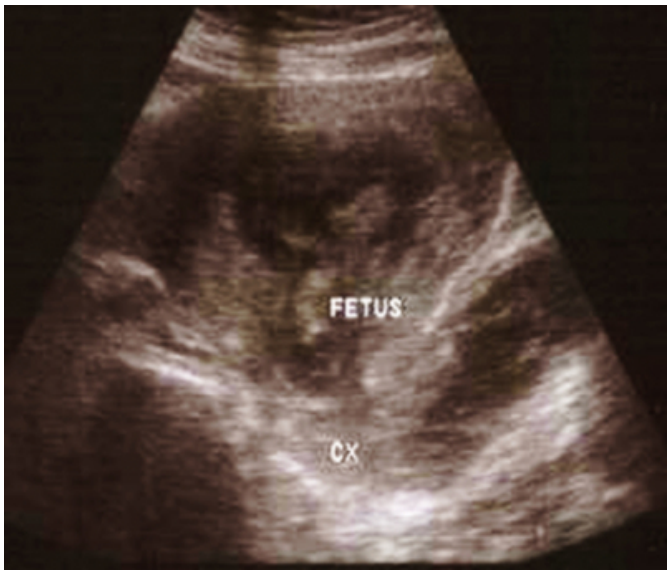


**Figure 3**

Gross picture of partial mole and fetus

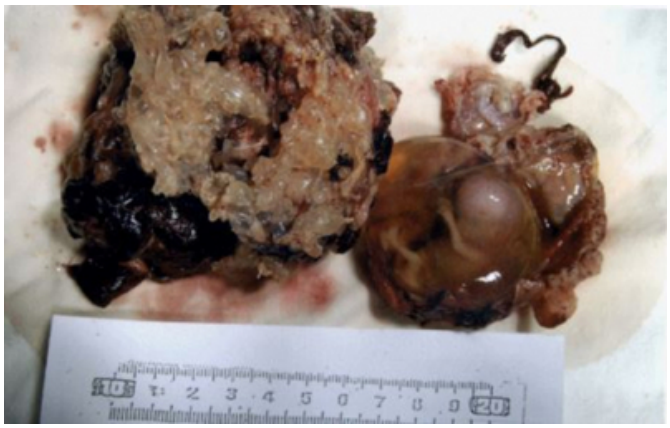

**Table 1**

Twin pregnancies after ART

| Authors [Reference] | Year | ART type | Maternal age | complications | Delivery weeks | Fetal outcome | GTD |
|---|---|---|---|---|---|---|---|
| Jinno et al.[18] | 1994 | IVF | 35 | Hyperthyroidism, Lung metastasis | 31 | Dead for RDS | yes |
| Cheng et al.[14] | 1995 | | 29 | Preterm labor | 29 | N-S | |
| Montes et al.[15] | 1999 | | 41 | Preeclampsia, Vaginal bleeding | 27 | N-S | |
| Lin et al.[20] | 2005 | | 39 | Vaginal bleeding | 36 | Small for GA | yes |
| Hamanoue et al.[38] | 2006 | ICSI | 40 | Preterm labor | 33 | N-S | |
| Dedes et al.[39] | 2008 | | 32 | Hyperthyroidism, Preterm labor | 26 | Dead for extreme prematurity | |
| Dolapcioglu et al.[21] | 2009 | | 34 | Preeclampsia, Vaginal bleeding | 29 | Small for GA | |
| Fatima et al.[17] | 2014 | | 29 | no | 37 | N-S | |

## DISCUSSION

Hydatidiform mole is the only benign gestational trophoblastic disease, and is divided into complete (classical; mole with diffuse trophoblastic hyperplasia and a diploid karyotype [usually 46, XX]) and incomplete (partial; mole with focal trophoblastic hyperplasia and a triploid karyotype) types. Complete mole associated with twin pregnancy is very rare, but the incidence is higher in Asia than elsewhere.4 The condition can be divided into three major types: twin gestation in which one twin is a diploid fetus with a normal placenta (46 chromosomes; 23 maternal and 23 paternal) and the other is a complete hydatidiform mole (46 chromosomes of paternal origin; thus, there is no fetus); singleton gestation of a triploid fetus with a partial hydatidiform mole placenta (69 chromosomes; 23 maternal and 46 paternal); and twin gestation in which one twin is a diploid fetus with a normal placenta (46 chromosomes; 23 maternal and 23 paternal) and the other is a triploid fetus with a partial, hydatidiform mole placenta (69 chromosomes; 23 maternal and 46 paternal).5 Ultrasonography is optimal for diagnosis of a molar pregnancy. The ultrasonographic

features of a hydatidiform mole are cystic changes, and irregularity and increased echogenicity of the decidual reaction of the placenta or myometrium.6 Also, the transverse:anteroposterior ratio of the sac usually exceeds 1.5.7 It is difficult to distinguish complete and partial moles in the absence of biopsy, but ultrasonography may help. In a complete mole, the tissues appear heterogeneous and sometimes present as a polypoid mass before acquiring the typical shape. A partial mole usually presents as an enlarged placenta with an anomalous fetus. Hydropic villi are often absent early in gestation, explaining why a partial mole is sometimes misdiagnosed as an abortion with a cystic change of the placenta. Magnetic resonance imaging (MRI) is also diagnostic, and exerts no adverse effect on the fetus. The field of view is larger than that of ultrasonography and the results are objective, unlike ultrasonography. Hydatidiform mole presents as a heterogeneous hyperintense mass on T2-weighted MRI images.8 If an invasive gestational trophoblastic tumor is present, MRI reveals myometrial invasion and extension into the parametrium.9

In most women with twin pregnancies and hydatidiform moles, termination is recommended because of the various potential maternal and fetal complications. The maternal complications include vaginal bleeding, pre-eclampsia, hyperthyroidism, pulmonary edema, thromboembolic disease, preterm labor, and persistently poor glucose tolerance test results.10-12 The fetus is at high risk of anomalies, growth restriction, preterm birth, and abortion.11-13 However, pregnancy continuation is possible. Although our case exhibited spontaneous abortion during observation, some reports on live births from twin molar pregnancies developing after IVF have appeared (Table 1). Almost all women experienced complications that triggered preterm delivery, but in four of the cases listed in Table 1 the fetal outcomes were good (healthy infants).14-17 Two preterm infants died: one because of prematurity and the other from respiratory distress syndrome.18,19 Two more infants were small for gestational age, suggesting possible intrauterine growth restriction.20,21 In one case, the mother developed a lung metastasis during pregnancy, which was terminated at 31 weeks.

Given the several potential complications and risks, women who wish to continue pregnancy require intensive consultation. They must understand that even if they are healthy and the fetal karyotype is normal, risks remain at all times. Appropriate management of maternal complications is key for achieving term delivery. The risk of fetal growth restriction and prematurity is high. Maintenance of a twin pregnancy with a mole must involve close follow-up of both the mother and fetus, up to delivery. After pregnancy termination, the risk of a persistent gestational trophoblastic tumor remains. All patients require 1 year of out-patient follow-up, preferably weekly for the first 3 weeks and monthly thereafter.22 Serum □-hCG regression must also be assessed. Contraception should be maintained for 1 year.23 If the □-hCG level does not normalize within 3 months, or a metastasis is detected, chemotherapy with methotrexate must be added.23

## CONCLUSION

When a twin pregnancy with a hydatidiform mole is diagnosed, observation is possible if the pregnancy is highly desired. In women who wish to continue pregnancy, intensive consultation on the complications and risks is required. Fetal growth and maternal complications must be closely monitored throughout the entire pregnancy. To ensure complete disease remission, postpartum follow-up for 1 year is recommended.

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