



# Complete Hydatidiform Mole Coexisting Two Live Fetuses in Triplet Pregnancy

Won-Kyu Jang, MD,  
Jinkyong Ha, MD,  
Jin-Gon Bae, MD

Department of Obstetrics and  
Gynecology, Dongsan Medical  
Center, Keimyung University School  
of Medicine, Daegu, Korea

We introduce a case of triplet pregnancy with a complete hydatidiform mole coexisting two viable fetuses and important issues for prenatal care in both mothers and fetuses due to a significantly high risk for both mothers and fetuses. A 32-year-old patient conceived a triplet pregnancy after intrauterine insemination. At 14 weeks of gestation, ultrasonography revealed hydatidiform moles that were adjacent to two fetuses. Fetal karyotyping of the two fetuses showed normal results. The second baby developed fetal growth restriction at 24 weeks of gestation and preeclampsia developed at 29 weeks of gestation. At 31 weeks and one day of gestation, cesarean section was performed due to fetal distress of the second baby. Microscopically, the molar tissue was favorably diagnosed as complete hydatidiform mole. This is successful delivery report of a pregnant woman who had triplet pregnancy complicated with one complete hydatidiform mole coexisting two viable fetuses in Korea.

**Key Words:** Hydatidiform mole, Triplet

## Introduction

Triplet pregnancy with a complete hydatidiform mole (CHM) coexisting two viable fetuses rarely occurs and has only been reported in a few cases.<sup>1</sup> Due to its high risk for both mother and fetuses, cases like these are subject to a dilemma whether to keep or terminate pregnancy. In previous cases, only five surviving fetuses were reported.<sup>1-5</sup> We present a case of triplet pregnancy with a complete hydatidiform mole coexisting two viable fetuses in which delivery of both surviving fetuses was performed with expectant management in order to support pregnancy.

## Case

A 32-year-old Korean underwent intrauterine insemination at a local hospital, and she conceived triplets. At 14 weeks of gestation, she was referred to our hospital for triplet pregnancy with one hydatidiform mole. The ultrasonography revealed two fetuses and another honeycomb-like mass, 9×12 cm in size, at the right upper fundus, suggestive of molar tissue (Fig. 1). Quantitative measurements of serum beta-human chorionic gonadotropin ( $\beta$ -hCG) level were >1,000,000 mIU/mL. At 16 weeks of gestation, we confirmed normal karyotypes of both fetuses by amniocentesis (46, XX and 46, XX). At 21 weeks of gestation, we performed a more precise magnetic resonance imaging (MRI), and the results showed the same hydatidiform mole findings as ultrasonography diagnosis without invasiveness (Fig. 2). Serial ultrasonography was performed, and the hydatidiform mole remained stable. The first baby maintained appropriate growth, but the second baby developed fetal growth restriction with three weeks discrepancy at 24 weeks of gestation. The patient was hospitalized for three days due to fetal growth restriction, and she underwent fetal monitoring and doppler tests

**Received:** 6 April 2020

**Revised:** 21 April 2020

**Accepted:** 22 April 2020

### Correspondence to

Jin-Gon Bae, MD  
Department of Obstetrics and  
Gynecology, Dongsan Medical  
Center, Keimyung University School  
of Medicine, 1035 Dalgubeol-daero,  
Dalseo-gu, Daegu 42601, Korea

**Tel:** +82-53-258-4847

**Fax:** +82-53-258-4848

**E-mail:** jgonmd@gmail.com

Copyright© 2020 by The Korean Society  
of Perinatology

This is an Open Access article distributed under the terms of the Creative Commons Attribution Non-Commercial License (<http://creativecommons.org/licenses/by-nc/4.0/>), which permits unrestricted non-commercial use, distribution, and reproduction in any medium, provided that the original work is properly cited.

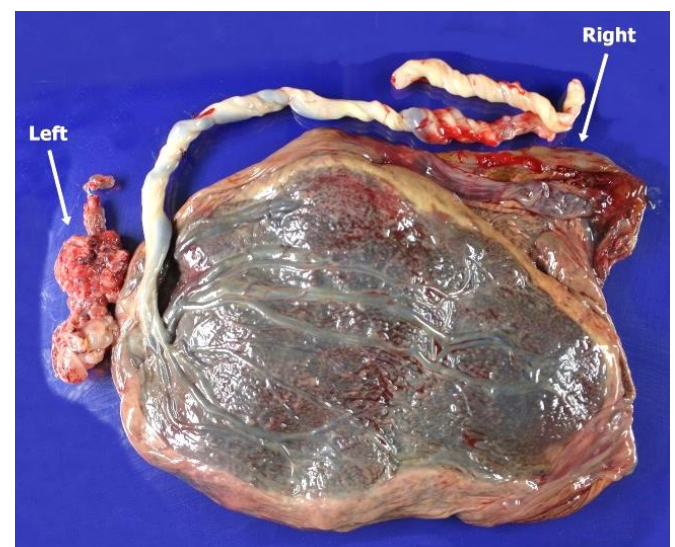


**Fig. 1.** Transabdominal ultrasonography at 14 gestational weeks demonstrating molar tissue with amniotic cavity of two fetuses. Molar tissue is observed with various echogenicity (mainly echogenic) and small vesicles described as “snowstorm” (arrow).



**Fig. 2.** Coronal T2 magnetic resonance imaging of the uterus shows similar changes as ultrasonography with complete molar tissue without invasiveness coexisting two amniotic sac of fetuses. Molar tissue appears as a heterogeneous hyperintense and a number of characteristic cystic spaces (white arrow). Myometrium appears more hypoechoic around the molar tissue and is intact (black arrow).

daily. Thereafter, she was discharged without any specific findings and was followed up weekly. Preeclampsia (140/90 mmHg; urine protein, 2+) developed at 29 weeks of gestation, and patient was hospitalized and monitored for fetal and maternal conditions. Laboratory test results at the time of admission were normal, and there were no symptoms such as headache, visual disturbances, or upper abdominal pain. Preeclampsia did not deteriorate and remained stationary, but the fetal umbilical arterial doppler finding suddenly deteriorated, and nonstress test exhibited intermittent pattern of late deceleration in the second baby at 31 weeks and one day of gestation. Therefore, we decided to deliver. The babies were delivered by caesarean section after completing a course of steroid administration to promote fetal lung maturation. Two live babies were born, weighing 1,490 g and 680 g. The Apgar scores of the first newborn at 1 and 5 minutes were 6 and 8, respectively, while the Apgar scores of the second newborn at 1 and 5 minutes were 6 and 8, respectively. The first newborn had no significant events during hospitalization and was discharged in good condition. However, the second newborn expired seven days after birth due to early-onset sepsis. The molar tissue weighed 300 g, and the size was 19.0×15.0×3.0 cm, containing a grape-like tissue which looks the same as the molar change (Fig. 3). Microscopically, the molar tissue was favorably diagnosed as complete hydatidiform mole and no signs of placental infection were found in placenta. Maternal serum  $\beta$ -hCG level decreased to normal range at three months postpartum.



**Fig. 3.** Postpartum gross findings of molar tissue (left) and normal placenta of the first fetus (right).

## Discussion

The incidence of hydatidiform mole coexisting viable fetuses in twin pregnancies varies from 1 in 22,000 to 1 in 100,000 pregnancies.<sup>6</sup> Triplet pregnancy with complete hydatidiform mole occurs even more rarely; most of which have been reported to have been terminated, and only five cases have been reported to have sustained the pregnancy and successfully gave birth to live fetuses.<sup>1-5</sup> To the best of our knowledge, this case is the first successful delivery report of a triplet pregnancy with complete hydatidiform mole coexisting viable fetuses in Korea.

Hydatidiform mole changes occur as a result of excessive paternally derived genetic material, which then results to abnormalities in fetoplacental development and the placenta's villous trophoblast hyperplasia. This is a pathological sign of a genetically abnormal concept. Partial hydatidiform moles occur in about 3 in every 1,000 pregnancies, and pregnancy with a healthy fetus rarely occurs in 1 in 10,000 pregnancies.<sup>7</sup> The incidence of abnormal karyotype is very high in pregnancy with a living fetus and partial mole, which has a high risk of fetal malformation and a low fetal survival rate.<sup>8-10</sup> It is difficult to differentiate the diagnoses between hydropic abortus, partial hydatidiform mole, and complete hydatidiform mole based on histological grounds alone.<sup>11</sup> In addition to histological examination, DNA polymorphic marker analysis can improve the accuracy of diagnosis in molar pregnancy.<sup>12</sup> Therefore, the analysis of zygosity through genetic testing in hydatidiform mole coexisting with normal fetus was required and practical for differential diagnosis of complete hydatidiform mole and partial hydatidiform mole to establish the direction of treatment in these very rare pregnancy complications. Although complete hydatidiform moles have a higher risk of invasive trophoblastic disease in mothers,<sup>13</sup> they provide an opportunity to deliver healthy infants.

In an ultrasonography performed in the first trimester of pregnancy, hydatidiform moles are most frequently observed with various echogenicity (mainly echogenic) and small vesicles (1–30 m) with enlarged uterus.<sup>14,15</sup> The ultrasonic pattern of these lesions has been described classically as “snowstorm” and “cluster of grapes” because of a number of echogenic foci due to hyperplastic and hydropic villi in pathological findings.<sup>14</sup> In contrast, partial moles are largely associated with growth-delayed or malformed fetuses and show ultrasonography findings of

large and thick placenta with numerous anechoic cystic lesions.<sup>16</sup> This case is observed only in moles of live fetus with CHM, and it needs to be distinguished from partial mole.<sup>17</sup> A theca lutein cyst is found in up to 40% of cases and reflects a significantly elevated hCG level.<sup>14</sup> In partial mole, theca lutein cyst is less observed due to a relatively lower hCG level.<sup>14</sup> Although ultrasonography is a very useful tool for the diagnosis of mole pregnancy, the distinction between complete mole and partial mole is not easy in a limited clinical setting, and the final diagnosis must wait for the results of the pathological findings.

In the first trimester of pregnancy, there may be little to no abnormalities on MRI scans performed. In a T2-weighted imaging performed during the second trimester of pregnancy, the hydatidiform mole appears as a heterogeneous hyperintense tumor that distorts the normal zonal structure and may show a number of characteristic cystic spaces.<sup>18</sup> In the T1-weighted image, molar pregnancy appears isointense or weakly hyperintense compared to myometrium, and a focal hemorrhagic area where signals are hyperintense is observed.<sup>18</sup> The myometrial invasion diffused by molar pregnancy shows hyper-extensive myometrial signal with the disappearance of normal zonal anatomy in the T2-weighted image.<sup>19</sup> Pelvic MRI is superior to ultrasonography in detecting myometrial invasion with extrauterine expansion of molar pregnancy which is also used to diagnose it.

There is a variety of known clinical complications for one or more fetuses and coexisting hydatidiform moles. Complications, such as vaginal bleeding, spontaneous abortion, pelvic pain, hyperemesis gravidarum, anemia, and gestational trophoblastic disease have been reported.<sup>3</sup> The probability of these complications in triplet pregnancy is not entirely known, but as the number of coexisting fetus increases, the probability of complications should increase. For that reason, the ratio of termination of pregnancy in triplet pregnancy with hydatidiform mole and coexisting viable fetuses is known to be quite high.<sup>5</sup> In Korea, Kim et al.<sup>20</sup> published a case on triplet pregnancy with partial hydatidiform mole coexisting in two fetuses in 2008. In this case, termination of pregnancy was performed at the request of the patient. MTX therapy was also administered due to complications, such as persistent gestational trophoblastic disease (pGTD) and pulmonary metastasis. In our case, we decided to preserve pregnancy based on sufficient counselling with the

patient and successfully delivered at 31 weeks without serious maternal complications. Although the second baby expired seven days after birth due to early-onset sepsis, it seems to be a meaningful result overall.

In 2007, Ko et al.<sup>5</sup> reported on pregnancy complications in triplet pregnancy with complete hydatidiform mole. The incidence of early-onset preeclampsia was 23.1% (3/13). In this case, risk factors of preeclampsia, such as nulliparity, multiple pregnancy, and hydatidiform mole, were included. Preeclampsia was developed around 29 weeks, and preterm birth was achieved at 31 weeks due to fetal distress by fetal growth restriction of the second baby. However, live fetuses were successfully delivered. Ko et al.<sup>5</sup> also reported 38.5% (5/13) of persistent trophoblastic tumors. A serial follow-up on  $\beta$ -hCG is crucial and should be confirmed to normalize after delivery. Persistent trophoblastic tumors fortunately did not occur in this case, and follow-up of  $\beta$ -hCG level decreased to normal range within three months after delivery.

A total of seven cases were delivered with viability after 24 weeks, and all had preterm births before 34 weeks for various reasons.<sup>1</sup> Among them, five cases had surviving fetuses. Of these cases, only one case was delivered by maternal complication due to preeclampsia. Of the other cases, five cases were delivered with preterm labor and one case with fetal distress. Our case was not normally delivered due to maternal complication, such as preeclampsia. Although preeclampsia was developed, it was stable and well maintained, and delivery commenced for other reasons, such as fetal distress. In view of this, we would like to point out that if triplet pregnancy with complete hydatidiform mole coexisting two viable fetuses case can sustain well up to the gestational age with viability, you should be less concerned about delivery due to maternal complication. It is believed that this is an important point in deciding to maintain pregnancy without giving up in the early stages of pregnancy.

In conclusion, the treatment policy for triplet pregnancy with hydatidiform mole coexisting viable fetuses is not well established. However, in some cases where live fetus was born, like our case, there is an option for treatment that can keep pregnancy in addition to the termination of pregnancy. Of course, the maternal condition and the requirement of the patient's continuing pregnancy needs to be taken into consideration, and sufficient consultation on pregnancy complications, such as preeclampsia,

preterm birth, and GTD, should be given. We hope that our case will be helpful in making clinical decisions and consultations with patients.

## Conflict of interest

No potential conflict of interest relevant to this article was reported.

## References

- 1) Suganya U, Kanchana MP, Revathy M. Complete hydatidiform mole in a triplet pregnancy with coexisting two live fetuses-a case report and review of literature. *J Evolution Med Dent Sci* 2017;6:1056-8.
- 2) Amr MF, Fisher RA, Foskett MA, Paradinas FJ. Triplet pregnancy with hydatidiform mole. *Int J Gynecol Cancer* 2000;10:76-81.
- 3) Takagi K, Unno N, Hyodo HE, Hyodo H, Kashima H, Kubota N, et al. Complete hydatidiform mole in a triplet pregnancy coexisting two viable fetuses: case report and review of the literature. *J Obstet Gynaecol Res* 2003;29:330-8.
- 4) Bovicelli L, Ghi T, Pilu G, Farina A, Savelli L, Simonazzi G, et al. Prenatal diagnosis of a complete mole coexisting with a dichorionic twin pregnancy: case report. *Hum Reprod* 2004;19:1231-4.
- 5) Ko PC, Peng HH, Soong YK, Chang SD. Triplet pregnancy complicated with one hydatidiform mole and preeclampsia in a 46,XY female with gonadal dysgenesis. *Taiwan J Obstet Gynecol* 2007;46:276-80.
- 6) Jones WB, Lauersen NH. Hydatidiform mole with coexistent fetus. *Am J Obstet Gynecol* 1975;122:267-72.
- 7) Steller MA, Genest DR, Bernstein MR, Lage JM, Goldstein DP, Berkowitz RS. Natural history of twin pregnancy with complete hydatidiform mole and coexisting fetus. *Obstet Gynecol* 1994;83:35-42.
- 8) Shahabi S, Naome G, Cobin L, Verougstraete A, Masters L, Zengbe V, et al. Complete hydatidiform mole and coexisting normal fetuses. A report of two cases with contrasting outcomes. *J Reprod Med* 1997;42:756-60.
- 9) Jauniaux E, Elkazen N, Leroy F, Wilkin P, Rodesch F, Hustin J. Clinical and morphologic aspects of the vanishing twin phenomenon. *Obstet Gynecol* 1988;72:577-81.
- 10) Lawler SD, Fisher RA, Dent J. A prospective genetic study of complete and partial hydatidiform moles. *Am J Obstet Gynecol* 1991;164:1270-7.
- 11) Gschwendtner A, Neher A, Kreczy A, Müller-Holzner E, Volgger B, Mairinger T. DNA ploidy determination of early molar pregnancies by image analysis: comparison to histologic classification. *Arch Pathol Lab Med* 1998;122:1000-4.
- 12) Harada I, Tsutsumi O, Takai Y, Iida T, Sakai M, Yoshikawa H, et al. DNA polymorphism analysis of a case of complete hydatidiform mole coexisting with a fetus. *Hum Reprod* 1997;12:2563-6.



- 13) Sun SY, Melamed A, Joseph NT, Gockley AA, Goldstein DP, Bernstein MR, et al. Clinical presentation of complete hydatidiform mole and partial hydatidiform mole at a regional trophoblastic disease center in the United States over the past 2 decades. *Int J Gynecol Cancer* 2016;26:367-70.
- 14) Wagner BJ, Woodward PJ, Dickey GE. From the archives of the AFIP. Gestational trophoblastic disease: radiologic-pathologic correlation. *Radiographics* 1996;16:131-48.
- 15) Allen SD, Lim AK, Seckl MJ, Blunt DM, Mitchell AW. Radiology of gestational trophoblastic neoplasia. *Clin Radiol* 2006;61:301-13.
- 16) DeBaz BP, Lewis TJ. Imaging of gestational trophoblastic disease. *Semin Oncol* 1995;22:130-41.
- 17) Vejerslev LO. Clinical management and diagnostic possibilities in hydatidiform mole with coexistent fetus. *Obstet Gynecol Surv* 1991;46:577-88.
- 18) Kohorn EI, McCarthy SM, Barton JW. Is magnetic resonance imaging a useful aid in confirming the diagnosis of nonmetastatic gestational trophoblastic neoplasia? *Int J Gynecol Cancer* 1996;6:128-34.
- 19) Nagayama M, Watanabe Y, Okumura A, Amoh Y, Nakashita S, Dodo Y. Fast MR imaging in obstetrics. *Radiographics* 2002;22:563-80; discussion 80-2.
- 20) Kim CH, Kim YH, Kim JW, Kim KM, Cho MK, Kim SM, et al. Triplet pregnancy with partial hydatidiform mole coexisting with two fetuses: a case report. *J Obstet Gynaecol Res* 2008;34(4 Pt 2):641-4.