A Case Report of Twin Pregnancy with Hydatidiform Mole and Co-existing Live Fetus

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Abstract

Complete hydatidiform mole with co-existing live fetus (CHMF) is a rare and high-risk pregnancy usually seen with ovulation induction protocols. These pregnancies are complicated with vaginal bleeding, pre-eclampsia, miscarriage, preterm delivery, fetal demise and the risk of gestational trophoblastic neoplasia (GTN). Here, we describe a case of CHMF and a second case of monozygotic twins: partial mole with live fetuses. The pregnancies were conceived after clomiphene citrate ovulation induction. Both cases presented with vaginal bleeding and hyperemesis in the early mid-trimester. The diagnosis was based on history, examination, ultrasound findings and high serum beta-human chorionic gonadotropin (βHCG) levels. A CHMF can be differentiated from a singleton partial molar pregnancy with similar ultrasound appearance by amniocentesis and karyotyping of the live fetus, which is a normal diploid. After adequate counseling, both women refused prenatal karyotyping and underwent the termination of pregnancy. The method of termination needs to be carefully decided. Surgical evacuation maybe difficult due to the well-formed fetus in the second trimester, and uterotonic agents can be associated with the risk of trophoblastic embolization and GTN. Termination with misoprostol followed by ultrasound-guided suction evacuation was successfully done in both cases. Histopathology and karyotyping confirmed the diagnosis of CHMF in the first and partial mole in the second case. βHCG normalized within 7 weeks postevacuation in both, with no increased risk of trophoblastic embolization or GTN. More studies are needed on the methods of termination in such pregnancies. Medical termination with misoprostol appears to be a viable option, though the optimal dosage is yet to be defined.

Keywords: Co-existing live fetus, hydatidiform mole, multiple pregnancy, ovulation induction

INTRODUCTION

Hydatidiform mole with co-existing live fetus is very rare and is considered as a high-risk pregnancy. The incidence of complete hydatidiform mole co-existing live fetus (CHMF) and that of partial hydatidiform mole with co-existing live fetus (PHMF) is 1/20,000–1/100,000[1] and 1/10,000–1/20,000,[2] respectively. To date, about 200 cases of CHMF and PHMF have been reported, and to the best of our knowledge, there was no case of monozygotic twin partial molar pregnancy.

CASE REPORTS

Case 1

A 27-year-old primigravida reported at 13 weeks with...
hyperemesis and vaginal bleeding. She had conceived after ovulation induction with clomiphene citrate (CC). General and systemic examinations were normal. The uterine size was more than the period of gestation (16–18 weeks). Ultrasonography revealed twin gestation wherein one sac had a live fetus of crown-rump length (CRL) 13 weeks 5 days (13W5D) with normal-appearing placenta. The nuchal translucency (NT) and nasal bone (NB) were normal. Adjacent to this, a nonhomogenous placenta with cystic spaces was seen, suggesting molar gestation measuring 6.5 cm × 4 cm [Figure 1]. Bilateral theca lutein ovarian cysts (the left measuring 4.3 cm × 3.5 cm and the right 5.0 cm × 4.2 cm) were noted. Serum beta-human chorionic gonadotropin (βhCG) was 198,880 mIU/ml.

In view of the clinical presentation, ultrasonographic finding and raised serum βhCG, twin pregnancy with hydatidiform mole and a co-existent live fetus or a singleton partial molar pregnancy was suspected. Karyotyping was offered, but on counseling regarding the maternal and fetal risks, the patient opted for termination.

Termination was performed with medical methods followed by surgical evacuation. The patient was advised vaginal misoprostol 800 μg followed by 400 μg 4 hourly (maximum six doses). After the 4th dose, she expelled the products of conception. Thereafter, USG-guided suction evacuation (S/E) was done to safely evacuate the uterus completely. The postoperative period was unremarkable. Histopathology of the molar tissue was in favor of complete hydatidiform mole. Fetal tissue sent for karyotyping was reported as normal diploid of maternal and paternal origin, confirming the diagnosis of CHMF.

The patient was followed up weekly with serum βhCG, which returned to normal 6 weeks after S/E. βhCG remained normal during the surveillance for 6 months.

Case 2
A 20-year-old, second gravida, with one abortion, reported at 14W4D, with irregular vaginal bleeding. She had conceived after CC induction. General and systemic examinations were normal. The uterine size was more than the period of gestation (18–20 weeks).

Ultrasonography revealed a monochorionic monoamniotic twin with CRL1 13W5D and CRL2 13W4D. Placenta was partly cystic, consistent with molar gestation [Figure 2]. The NT/NB scan was abnormal, suggesting aneuploidy. Serum βhCG was 232,518 mIU/ml. In view of these findings, monozygotic twin partial molar pregnancy was suspected. The patient underwent termination at 15W2D. We used the same method of termination as that in the first case. The patient expelled dysmorphic twin fetuses with the placenta and molar tissue after the third dose of misoprostol.

The histopathology was in favor of a partial molar gestation. On weekly follow-up, serum βhCG returned to normal at 7th week post-evacuation and remained normal throughout the surveillance.

DISCUSSION
An increasing trend of CHMF and PHMF has been reported in recent years due to the liberal use of ovulation induction methods. These pregnancies are associated with several maternal and fetal complications. Maternal complications include hyperemesis, pre-eclampsia, thyrotoxicosis, uterine rupture, refractory vaginal bleeding, trophoblastic emboli and gestational trophoblastic neoplasia (GTN). Rare complications include molar placenta previa/accreta and placental abcess. Fetal complications include spontaneous abortion, fetal growth restriction, preterm labor and intra-uterine fetal demise.

There are two distinct entities in the ultrasound finding of a live fetus with molar changes in the placenta. It could be either a dizygotic gestation with one complete molar or partial molar pregnancy along with a co-existing live fetus having a normal karyotype attached to a normal placenta, or it could be a singleton partial mole with a dysmorphic fetus and abnormal karyotype (mostly triploid). These two conditions have different genetic etiologies and outcomes, and thus need to be identified and differentiated at the time of primary ultrasound detection. Prenatal diagnosis can be done by amniocentesis to rule out triploidy or any other genetic abnormality before advising the continuation of pregnancy.

Diagnosis by ultrasonography is usually between 12 and 14 weeks, with a detection rate of 68%. When detected early in pregnancy, most women are either advised or opt for termination of pregnancy. However, as many of these pregnancies arise due to infertility treatment, some women may wish to continue pregnancy with added risk. Both our patients opted for termination.

Histopathology of the second case was reported as a partial mole. We added this case in our report to impress on the fact that it is extremely important to ascertain the karyotype and normalcy of the live fetus before embarking on the continuation of the pregnancy. A partial mole with similar
ultrasound appearance needs to be differentiated from a CHMF/PHMF, which has a co-existing fetus with normal karyotype. Whereas one might consider the continuation of pregnancy in CHMF/PHMF, the partial molar pregnancy should be terminated at diagnosis.

The method of termination of these pregnancies needs to be carefully decided. The diagnosis is usually confirmed in the second trimester, where the surgical evacuation has increased risk due to the well-formed fetus. Although the use of uterotonics is best avoided because of the risk of trophoblastic embolization and GTN,[9,11] medical termination with misoprostol is a viable alternative to surgical methods.[13] The optimal dosage of misoprostol is not defined. We used vaginal misoprostol, as described earlier, followed by the ultrasound-guided S/E.

The risk of GTN in smaller series has been reported to be higher in CHMF pregnancies (50%) compared to singleton molar pregnancy (15%).[14] Thus, after termination, serum βhCG should be monitored. However, a larger case series concluded that the rates of GTN requiring chemotherapy in CHMF who opted to continue with the pregnancy were not different from those who terminated their pregnancy or those with singleton molar gestation.[13] There was no increase in the risk of GTN with the second-trimester medical termination in our cases, unlike previous reports.[9]

CONCLUSION

Normal karyotype of the live fetus should be ascertained before the continuation of pregnancy in CHMF/PHMF. When indicated, medical termination with misoprostol is a viable option in these cases; however, the optimal dose schedule is not yet defined. After the completion of the pregnancy event, surveillance is required to rule out GTN.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patients have given their consent for their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published, and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Peer review

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Conflicts of interest

There are no conflicts of interest.

REFERENCES

1. Rai L, Shripad H, Guruvare S, Prashanth A, Mundkur A. Twin pregnancy with Hydatidiform Mole and Co-existent Live Fetus: Lessons Learnt. Malays J Med Sci 2014;21:61-4.
2. Rahamni M, Parviz S. A Case report of partial molar pregnancy associated with a normal appearing dizygotic fetus. Asian Pac J Reprod 2016;5:171-3.
3. Shazly SA, Ali MK, Abdel Badee AY, Alsokkary AB, Khodary MM, Mostafa NA. Twin pregnancy with complete hydatidiform mole and coexisting fetus following ovulation induction with a non-prescribed clomiphene citrate regimen: A case report. J Med Case Rep 2012;6:95.
4. Moini A, Ahmadi F, Eslami B, Zafarani F. Dizygotic twin pregnancy with a complete hydatidiform mole and a coexisting viable fetus. Iran J Radiol 2011;8:249-52.
5. Lee SW, Kim MY, Chung JH, Yang JH, Lee YH, Chun YK. Clinical findings of multiple pregnancy with a complete hydatidiform mole.
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and coexisting fetus. J Ultrasound Med 2010;29:271-80.
6. Atuk FA, Basuni JBM. Molar pregnancy with normal viable fetus presenting with severe pre-eclampsia: A case report. J Med Case Rep 2018;12:140.
7. Suri S, Davies M, Jauniaux E. Twin pregnancy presenting as a praevia complete hydatidiform mole and coexisting fetus complicated by a placental abscess. Fetal Diagn Ther 2009;26:181-4.
8. Surendran S, Thomas T, Vishnupriya VS. Partial Molar pregnancy with a normal fetus with complete placenta previa. J Obstet Gynecol India 2018;68:227.
9. Peng M, Li L, Zheng J, Ding Y, Yu I, Huang J. Termination of twin pregnancies with hydatidiform moles: A case series of four patients. Iran J Public Health 2014;43:1000-6.
10. Ahmed M, Kolar G, Vavilala S, Jaiman S. Complete hydatidiform mole with co-existing live fetus: A case series. J Fetal Med 2015;2:171.
11. Braga A, Obeica B, Werner H, Sun SY, Amim Júniors J, Filho JR, et al. A twin pregnancy with a hydatidiform mole and a coexisting live fetus: Prenatal diagnosis, treatment, and follow-up. J Ultrasound 2017;17:299-305.
12. Imafu H, Miyahara Y, Ebina Y, Yamada H. Ultrasound and MRI findings of twin pregnancies with complete hydatidiform mole and coexisting normal fetus: Two case reports. Kobe J Med Sci 2018;64:E1-E5.
13. Ferraz TJ, Bartosch CM, Ramalho CM, Carvalho FA, Carvalho BC, Brandão OG, et al. Complete mole in a dichorionic twin pregnancy after intracytoplasmic sperm injection. Rev Bras Ginecol Obstet 2013;35:39-43.
14. Massardier J, Golfier F, Journet D, Frappart L, Zalaquett M, Schott AM, et al. Twin pregnancy with complete hydatidiform mole and coexistent fetus: Obstetrical and oncological outcomes in a series of 14 cases. Eur J Obstet Gynecol Reprod Biol 2009;143:84-7.
15. Sebire NJ, Foskett M, Paradinas FJ, Fisher RA, Francis RJ, Short D, et al. Outcome of twin pregnancies with complete hydatidiform mole and healthy co-twin. Lancet 2002;359:2165-6.