European Journal of
Human Health

CASE REPORT

A twin pregnancy with complete hydatidiform mole and coexisting live fetus complicated by severe preeclampsia

Canan SOYER CALISKAN[1], Samettin CELIK[1], Safak HATIRNAZ[2], Mehmet KEFELI[3], Handan CELIK[3], Huri GUVEY[5]

[1] Maternity Hospital, A Branch of Training and Research Hospital, Samsun, TURKEY
[2] IVF Center, Medicana International Hospital, Samsun, TURKEY
[3] Department of Gynecology and Obstetrics, Medical Faculty of Ondokuz Mayis University, Samsun, TURKEY
[3] Obstetrics & Gynecology, Private Office, Kutahya, TURKEY

Objective: A twin pregnancy with associated complete hydatidiform mole is a rare clinical entity which may present in different severe antenatal complications. The aim of this study is to present a case of twinning of a 37 weeks fetus and coexistent complete mole complicated with severe preeclampsia and emergency C-section with a healthy live born fetus.

Case Report: A 32 year old G2 P1 pregnant woman who admitted at the perinatal center of maternity hospital was diagnosed a molar pregnancy coexisting with a healthy growing fetus at 10th weeks of gestation. At 32 weeks of gestation, preeclampsia was developed, the patient was followed until 37 weeks gestation when the clinical status of her turned into severe preeclampsia and cesarean section was carried out with the birth of a healthy fetus.

Conclusions: Twinning with a coexistent complete hydatidiform mole may cause life-threatening complications for both fetus and the mother. Severe preeclampsia may be triggered in patients with coexisting molar pregnancy and such patients are recommended to be evaluated for molar pregnancy associating normal fetus that had a normal antenatal course in their previous pregnancies.

Keywords: Complete hydatidiform mole, Normal fetus, Preeclampsia, Pregnancy complications

Corresponding Author: Canan SOYER CALISKAN canansoyer@hotmail.com

Received: ***July 7, 2021;*** *Accepted:****July 18, 2021;*** *Published Online:* ***August 30, 2021***

Cite this case report as: Soyer Caliskan, C., Samettin, C., Hatirnaz, S., Kefeli, M., Celik, H. & Guvey, H. (2021). A twin pregnancy with complete hydatidiform mole and coexisting live fetus complicated by severe preeclampsia. European journal of Human Health 2(5), 38-41.

Introduction

As the techniques to evaluate pregnancy in detail have advanced in the last 30 years, pregnancies co-twinning with complete hydatidiform mole can easily be diagnosed. Molar pregnancies can be seen in the records of ancient times and are most frequently observed in Asian populations, with decreasing frequencies in Africa, Latin American Countries, Europe and the least frequent in Australia and the USA (1). Twin pregnancy coexisting with a molar pregnancy is a rarity having an incidence of 1/22,000-1/100,000 pregnancies (2). However, exact diagnosis can only be made by histopathological examination (3).Complications due to molar pregnancy include hyperemesis gravidarum, vaginal hemorrhage, preeclampsia, fetal loss in-utero, hyperthyroidism, anemia, preterm delivery, placental abruption and persistent trophoblastic disease (4-7). Early detection of a molar pregnancy as twinning of a normal fetus is crucial in order to apply management strategies for this rare clinical entity. Advanced ultrasound examination, chromosomal evaluation by amniocentesis and a thorough clinical evaluation are essential steps in the diagnosis and management. Not only pregnancy termination or delivery of the fetus is required, but also close follow-up for trophoblastic disease persistence is recommended since the persistence can be seen in one out of two cases (4).

Herein we present twinning of a normal fetus with a molar pregnancy that was complicated with severe preeclampsia in a woman who had a viable fetus delivery without a history of preeclampsia.

Case Report

A 32 year old G2 P1 pregnant woman was referred to the perinatology department of our maternity hospital with a molar pregnancy coexisting with a healthy growing fetus. At the time of admission, gestational age was 10 weeks according to her last menstrual period. Ultrasound examination revealed a twin pregnancy, including a live fetus and a normally appearing placenta with a co-existing mixed echogenic mass measuring 70x50 mm in diameter with a honeycomb-like pattern suggesting molar pregnancy. The patient was informed about the risks of ongoing pregnancy. She preferred the continuation of pregnancy and she was closely followed by our perinatology department. Ultrasound examination showed an enlarged uterus filled with a heterogeneous pattern and live fetud of a gestational age compatible with 15 weeks (Figure 1). She was offered amniocentesis. Amniocentesis was performed at 16 weeks of gestation for fetal karyotyping and it revealed a normal karyotype. A detailed anomaly scan was normal at 21 weeks of gestation. Pregnancy was continued uneventfully until 32 weeks of gestation. She was admitted to the hospital at 32 weeks of gestation with preeclampsia. She was followed as an outpatient until 37 weeks gestation when the clinical status of the patient turned into severe preeclampsia. A female infant weighting 2300 gram was delivered by cesarean section at 37 weeks of gestation.

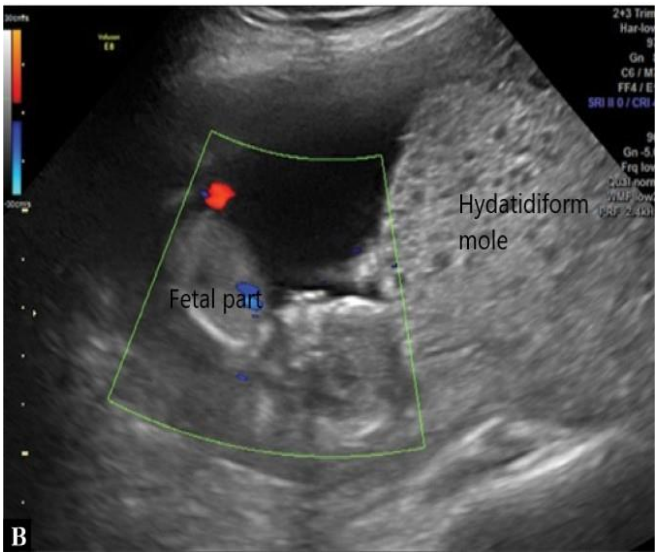

Figure 1. *Ultrasound image with shows a fetus of a gestational age of 14 weeks and a molar site of the placenta*

A molar mass with many vacuoles adherent to the normal appearing placenta and membranes were sent for pathological examination. Histopathologically, a complete hydatidiform mole with large villi and abundant cytotrophoblastic and syncytial trophoblastic proliferation was seen (Figure 2) and the diagnosis of complete hydatidiform mole was confirmed. Blood samples for chromosomal tests of mother, father and born baby were taken and their results were normal. After birth, the patient was strictly followed weekly for β-hCG evaluations until the β -hCG values decreased to zero.

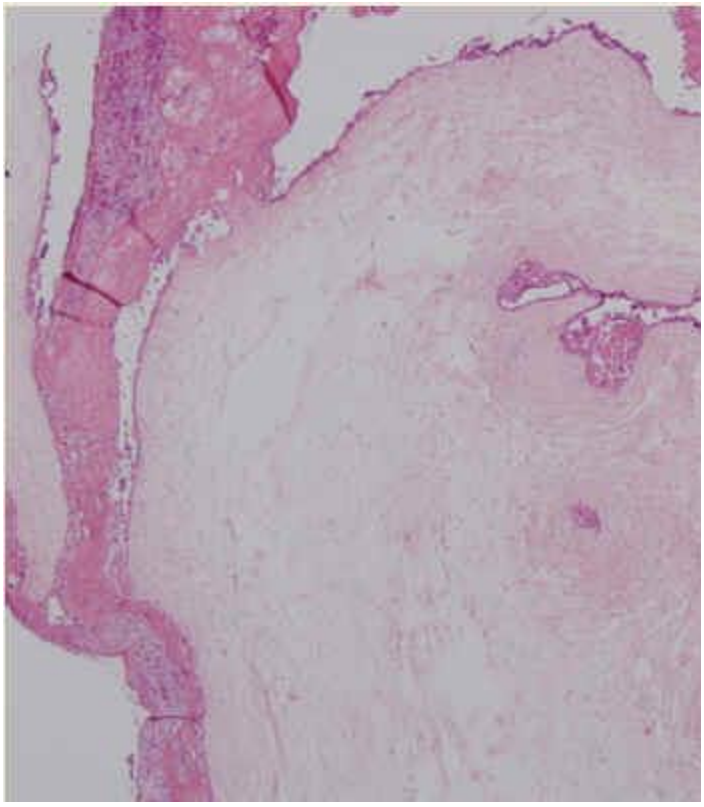

Figure 2. Complete hydatidiform mole with large villi and abundant cytotrophoblastic and syncytial trophoblastic proliferation (H&E, x40).

Discussion

In this rare clinical entity, three subsets of molar pregnancies associated with normal fetuses are present. The first and the most common form is complete hydatidiform mole twinning the normal fetus where a normal placenta and a complete mole are easily distinguished pathologically. The second form is twinning of a partial mole and a normal fetus where a partial mole and a normal placenta can be separately evaluated histopathologically. The third and the rarest form is a normal singleton fetus with partial molar pregnancy where a chromosomally normal fetus is associated with an aneuploid molar pregnancy (8). Prenatal diagnosis by amniocentesis should be recommended to determine the presence of any aneuploidy or triploidy (9). Preterm delivery is of major concern in this clinical situation and pregnancies reaching term or near term, as in our case, are much less frequent than preterm deliveries (2). Forexample, the pregnant women having twin pregnancy of hydatidiform mole and viable fetus delivered at 28th week of gestation with heavy bleeding as repoted by Resende et al. (10) Live birth rates in this clinical entity range from16% to 56% in the literature (11). Sebire et al evaluated 77 cases and reported that the chance of live birth without serious pregnancy complications was around 40% (12).

Two of the three steps in the correct diagnosis for the management of this clinical entity are missing in the antenatal course due to the inconvenience of the patient. For proper management, a complete diagnosis and close follow-up of the patient is crucial. Complete history and anamnesis, detailed ultrasound evaluation and a prenatal chromosomal analysis, preferably by amniocentesis, are three major steps in the correct diagnosis (8). The diagnostic value of serial B-hCG evaluations is important when the patients are under routine antenatal follow-up. B-hCG levels may be misleading in cases where normal hormone levels are very high but laboratory outcomes are very low, which is known as a high-dose hook effect, and in such cases, diluting the sample 1/10-1/1000 may overcome the misdiagnosis (13). Due to the high risk of developing maternal and fetal complications, molar pregnancies were terminated immediately in past years. However, with the advance of medical treatment of trophoblastic diseases and detailed ultrasound evaluations, such high risk patients can be monitored and the chance of delivering term or near term viable fetuses with a complete cure can be obtained (2). With strict follow-up, it would be possible to detect early problems during pregnancy, enabling medical interventions and appropriate guidance to the patient (14). As expected, most complications including preeclampsia can be managed in an appropriate manner in the modern era of medicine (15). The decision of termination or follow-up should be discussed with the patient in detail and an informed consent for the follow-up is recommended in view of the anticipated high risks. It should be kept in mind that the increased rate of twinning of molar pregnancy with a normal fetus is proportional to the increased use of ovulation inductions (11).

In a review of 113 reports, Vejerslev et al reported that almost 30% of patients with molar pregnancies coexistent with normal fetuses develop preeclamptic symptoms (14). Such pregnant women should be monitored strictly in tertiary care settings (3). In the present case, diagnosis of twinning of molar pregnancy with a normal fetus was made at 10 weeks of gestation. The patient refused termination of pregnancy and she was followed until 37 weeks of

gestation when severe preeclampsia developed. Severe preeclampsia imposed on this high risk pregnancy necessitated the termination of pregnancy and removal of placenta and membranes immediately in order to decrease maternal morbidity and mortality.

Following the delivery of living fetus and placental materials including molar twin, patients should be strictly followed both for symptoms of preeclampsia and possible risk of developing persistent trophoblastic disease (8).

Conclusions

Term or near term viable fetus delivery in twinning with molar pregnancy is a rare clinical entity requiring close follow-up in high risk pregnancy centers. Antenatal monitoring of the patient with complete diagnostic measures taken is mandatory for proper management of possible complications. Severe preeclampsia is one of the major maternal morbidities secondary to complete hydatidiform mole coexistent with a viable fetus and immediate C-section to deliver the fetus and placental materials is recommended. Molar pregnancy should be included in the differential diagnosis of severe preeclampsia as an underlying reason.

Conflict of interest : The authors declare no conflicts of interest related to this article.

References

1. Bracken MB. Incidence and aetiology of hydatidiform mole: an epidemiological review. Br J Obstet Gynaecol. 1987;94:1123-1135.

2. Peng HH, Huang KG, Chueh HY, Adlan AS, Chang SD, Lee CL. Term delivery of a complete hydatidiform mole with a coexisting living fetus followed by successful treatment of maternal metastatic gestational trophoblastic diseaseTaiwan J Obstet Gynecol. 2014 Sep; 53(3):397-400.

3. Büke B, Topçu HO, Bulgu E, Eminov E, Kazandı M. Complete hydatidiform mole presenting as placenta previa in a twin pregnancy with a coexisting normal fetus: Case report. J Turk Ger Gynecol Assoc. 2014; 15: 256-8

4. Bruchim I, Kidron D, Amiel A, Altaras M, Fejgin MD. Complete hydatidiform mole and a coexistent viable fetus: report of two cases and review of the literature. Gynecol Oncol. 2000; 77:197–202.

5. Vaisbuch E, Ben-Arie A, Dgani R, Perlman S, Sokolovsky N, Hagay Z. Twin pregnancy consisting of a complete hydatidiform mole and co-existent fetus: Report of two cases and review of literature. Gynecol Oncol. 2005;98:19–23.

6. Kashimura Y, Tanaka M, Harada N, et al. Twin pregnancy consisting of 46, XY heterozygous complete mole coexisting with a live fetus. Placenta. 2001;22: 323–327.

7. Aguilera M, Rauk P, Ghebre R, Ramin K. Complete hydatidiform mole presenting as a placenta accreta in a twin pregnancy with a coexisting normal fetus: Case report. Case Rep Obstet Gynecol. 2012; 2012:405085.

8. Atuk AF, Basuni JBM. Molar pregnancy with normal viable fetus presenting with severe pre-eclampsia: a case report. J Med Case Rep. 2018; 12: 140.

9. Guven ESG, Ozturk N, Deveci S, Hizli D, Kandemir O, Dilbaz S. Partial molar pregnancy and coexisting fetus with diploid karyotype. J Matern Fetal Neonatal Med. 2007; 20(2):175-181.

10. Rai L, Shrıpad H, Guruvare S, Prashanth A, Mundkur A. Malays. Twin pregnancy with hydatidiform mole and co-existent live fetus: Lessons Learnt. J med Sci. 2014; 21(6):61-64.

11. Sebire NJ, Foskett M, Paradinas FJ. Outcome of twin pregnancies with complete hydatidiform mole and healthy co-twin. Lancet. 2002;359:2165-2166.

12. Braga A, Obeica B, Werner H, et al. A twin pregnancy wih a hydatidiform mole and coexisting live fetus: prenatal diagnosis, treatment, and follow-up. J Ultrason. 2017;17:299-305.

13. Sheik S, Al-Riyami N, Mathew NR, Al-Sukaiti R, Qureshi A, Mathew M. Twin pregnancy with a complete hydatidiform mole and a coexisting live fetus. Online Case reports. 2015; 11(15): 550-553.

14. Vejerslev LO. Clinical management and diagnostic possibilities in hydatidiform mole with coexistent fetus. Obstetric Gynecol Surv. 1991; 46(9):577-588.