Case Report

Twin pregnancy with Hydatidiform Mole and Co-existent Live Fetus: Lessons Learnt

Lavanya Rai, Hebbar Shripad, Shyamala Guruvare, Adiga Prashanth, Anjali Mundkur

Submitted: 6 Jan 2014
Accepted: 14 Apr 2014

Department of Obstetrics and Gynaecology, Kasturba Medical College, Manipal University, Manipal – 576 104, Karnataka, India

Abstract

This is a case report of a twin pregnancy with one fetus and a coexistent mole diagnosed at 13 weeks. After thorough counseling, the pregnancy was continued as per the patient's desire. The pregnancy was closely monitored with serial S β hCG, ultrasound for fetal growth, size of molar sac, and theca lutein cysts, which gradually decreased in size during the second trimester of pregnancy. An emergency caesarean delivery was done at 36 weeks due to breech in early labour. A live baby weighing 1.8 kg was delivered in good condition. Her S β hCG reached normal levels at the end of three weeks, and she is now on post-molar surveillance. Though the general trend is to terminate pregnancy in twins with coexistent mole in anticipation of complications, under close surveillance, optimal outcomes can be achieved. Monitoring of S β hCG, serial ultrasound for fetal growth, size of molar component, and theca lutein cysts can help to predict good patient outcomes.

Keywords: *beta subunit, gestational trophoblastic disease, human chorionic gonadotropin, hydatidiform mole, molar pregnancy, twins*

Introduction

A multiple pregnancy with one fetus and a coexisting hydatidiform mole, a rare phenomenon of the past, is showing an upward trend due to the rise in the induction of ovulation. The reported incidence is 1 in 22 000–100 000 pregnancies (1), with most being complete hydatidiform moles (CHM) with a fetus; however, the reported prevalence for a partial mole with a coexisting foetus is 0.005–0.01% of pregnancies (2).

Very few twin pregnancies with a hydatidiform mole and a foetus continue to term as they often have spontaneous or induced terminations for maternal complications. Here, we report a case detected in the first trimester, where pregnancy was continued and patient delivered a live infant.

Case Report

Mrs R, a 25-year-old patient, was referred to our clinic at 13 weeks of gestation with the report of twin gestational sacs: one with a live fetus and the other a suspected hydatidiform mole. She had conceived after ovulation induction with clomiphene citrate, and was diagnosed with twin gestational sacs at six weeks of gestation. At 12 weeks, she was re-evaluated for a risk of abortion, when it was noted that there was one normal foetus and a co-existing hydatidiform mole. The termination of the pregnancy was advised by her doctor, but she refused.

Her general and systemic examinations were unremarkable, and upon evaluation, the uterus was found to be at approximately 14–16 weeks of gestation. No other abnormalities were detected. A transabdominal scan (TAS) at 13 weeks of gestation at our hospital confirmed the findings of a live foetus with a normally appearing placenta in one sac and an adjacent mixed echoic mass with a honeycomb-like pattern, suggestive of a hydatidiform mole (Figure 1). The molar mass measured 10 × 6 cm and was situated over the cervical os, while the foetus with the posterior placenta was situated near the fundus. The bilateral theca lutein cysts measured around 7 × 6 cm each. The patient's S beta human chorionic gonadotropin (S β hCG) was very high, at 374 747 mIU/mL; however, her thyroid function tests were normal. She was counselled regarding the risks involved in continuing the pregnancy, and as per her preference, the pregnancy was continued and she was closely monitored in our high risk pregnancy clinic. Two weeks later, although there was no increase in the S βhCG, the molar mass (11.8 × 6 cm) and theca lutein cysts (11 × 6 cm) showed increases in size. She continued to have vaginal blood spotting, on and off, until approximately 26 weeks of gestation.

We estimated the weekly S β hCG levels and

measured the size of the molar mass and theca lutein cysts. A detailed anomaly scan for the fetus was normal at 20 weeks, and pregnancy continued uneventfully, with a declining trend in the S β hCG beginning in the early second trimester, with a regression of the theca lutein cysts. The size of the molar mass reduced gradually, and at around 30 weeks of gestation, we could no longer see the molar mass or the theca lutein cysts upon ultrasound. Interval foetal growth was maintained around the 10th percentile.

At 36 weeks, she was admitted with labour pains and an emergency caesarean section was done for a breech presentation. A male neonate weighing 1.8 kg with a good Apgar score was delivered, and there was no postpartum haemorrhage. The placenta weighed 586 g, measuring 20 × 14 × 4 cm, and the molar tissue was distinct, with a sheet of vesicles adjacent to the normal placenta (Figure 2). The theca lutein cysts were very small, and histopathology of the placenta was reported as a normal placenta, along with molar tissue containing features suggestive of a partial hydatidiform mole. Karyotyping of the mole and foetus could not be done.

The S β hCG showed rapid regression, and at the end one week it was 111 mIU/mL, reaching normal in another two weeks (Figure 3). The patient is now under post-molar surveillance.

Discussion

Twin pregnancies with one normal foetus and a co-existing molar pregnancy (complete or partial) are fraught with complications for both the mother and the foetus. For the mother, in addition to haemorrhagic complications, there are increased risks of medical problems such as hyperemesis, preeclampsia, thyrotoxicosis and trophoblastic emboli. The risk of persistent trophoblastic disease (PTD) in twin molar pregnancy is more than a single complete mole, and is also increased in partial moles with a diploid foetus, compared to triploidy (3). Preeclampsia has been reported in 34% of these pregnancies (4). There are case reports where patients required hysterotomies and hysterectomies for severe bleeding (5,6).

The risks to the foetus when pregnancy continues in these cases include abortion, intrauterine foetal demise, pre-term labour and foetal growth restrictions. Live births in these pregnancies vary from 16–56% (1,7). Another rare problem that has been reported is the molar placenta previa developing placental abscesses (8). It has been observed that there is a tendency for these molar tissues to be in the lower segment of the uterus, closer to the os, thereby resulting in bleeding (on and off), which was also evident in our case (9). Hysterectomies have recently been reported for molar placenta previa accreta, and also for spontaneous fundal rupture of the uterus in the second trimester (10,11).

The risk of PTD in CHM is said to be as high as 16–50%, and it does not depend on the duration

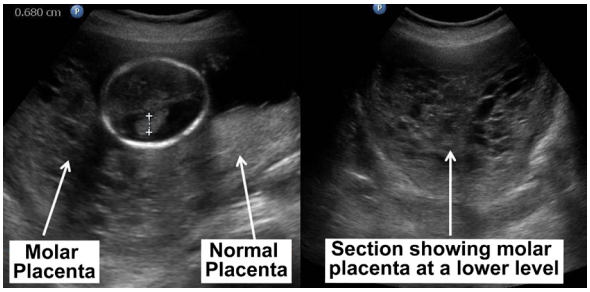

Figure 1: Ultrasound showing the molar placenta.

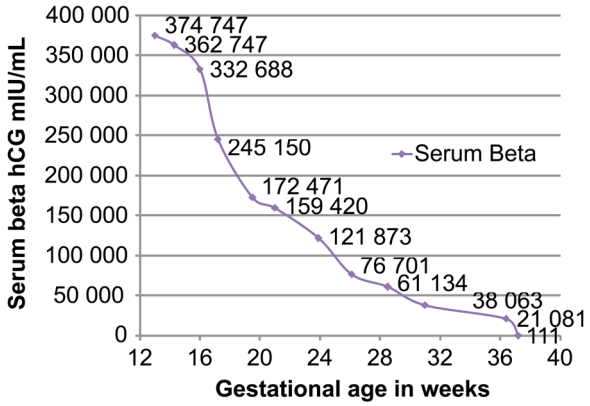

Figure 2: Serum beta human chorionic gonadotropin (HCG) regression curve.

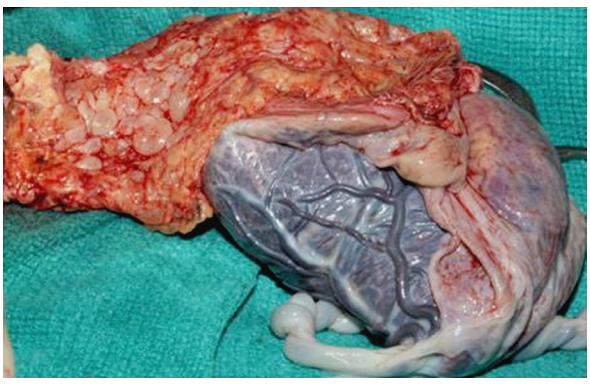

Figure 3: Placenta with attached molar tissue.

of the pregnancy (1,7). The risk of a partial mole developing PTD is 14–33% (12). Stellar et al. noted a higher risk of developing PTD in cases with twin molar pregnancies with a co existent foetus, when compared to singleton molar pregnancies (6). PTD is not only more common, but it requires multiple cycles of combination chemotherapy (13). Another interesting observation made in some studies is that PTD was more often seen in those twin molar pregnancies with maternal complications such as preeclampsia, hyperemesis, etc. (14).

Although a partial mole often coexists with a triploid foetus, there are case reports of twin pregnancies with foetuses of normal karyotype. Triploid foetuses have asymmetrical intra uterine growth restriction (IUGR) and multiple malformations (12). In a case report of twin foetuses with a hydatidiform mole, macro and microscopic findings diagnosed a partial hydatidiform mole. However, DNA polymorphic analysis demonstrated foetal DNA of biparental origin, and the molar DNA was of paternal origin (15). The DNA polymorphic analysis could not be done in our case, where histopathology showed a partial mole, while clinically we believed it was a complete mole. Very often, a foetus coexisting with molar tissue is diagnosed as a partial mole. To prognosticate, it is important to differentiate between partial and complete moles, with DNA polymorphic analysis helping in the genetic characterization of molar pregnancies, and it should be used for accurate diagnosis.

Seiji et al. showed in their case report that conventional diagnostic methods are inadequate for diagnosis (15). However, paternally imprinted gene product p57 and flow cytometry used for DNA analyses are useful for discriminating complete and partial moles (7). Other differential diagnoses for molar changes in the placenta, such as mesenchymal dysplasia and hydropic changes in the placenta, were ruled out by clinical features as well as the histopathology of the placenta after delivery.

Prenatal diagnosis by amniocentesis has been advocated to rule out triploidy or any other genetic abnormalities, before advising the continuation of a pregnancy (4,12). Chorionic villi sampling for karyotype is not recommended, as it may vary from that of the foetus because of confined placental mosaicism. Most often, the triploid foetus co-existing with a partial mole tends to die, while a foetus with a complete mole tends to survive (14). A foetus with a partial mole may survive when it occurs in a dizygotic twin, with one foetus and the other oocyte giving rise to a partial diploid mole, however, a monozygotic twin with triploidy gives rise to a partial mole with an abnormal foetus (16).

The incidence of twins with one molar pregnancy is expected to rise with aggressive treatment for infertility and early diagnosis with ultrasound. The lessons that have been learned from this case are that serial monitoring for the regression of the size of the molar mass, theca lutein cysts and a declining serial S β hCG are good prognostic indicators. It has been reported that very high S β hCG (> 10^6 IU/L) and the presence of medical complications portend a poor outcome, and may serve as an indication for the termination of pregnancy because they are suggestive of aggressive trophoblastic growth (13). Fortunately, our patient did not develop any medical complications and the pregnancy continued uneventfully.

When detected early in the pregnancy, the management approach is generally to offer termination in view of the anticipated complications. Nevertheless, it seems reasonable to opt for conservative management with the consent of the patient, after thorough counselling. As many of these molar pregnancies occur following the induction of ovulation, the desire to continue pregnancy often outweighs that of termination. However, one must be prepared for termination in the event of maternal complications like severe preeclampsia or bleeding, and conservative management in these pregnancies is for the foetus' sake, with the mother being at risk of serious complications. Thus, a twin pregnancy with a hydatidiform mole and a coexisting live foetus requires a thorough evaluation, and pregnancy may be continued under close surveillance for an optimal outcome.

Acknowledgement

None.

Conflict of interest

None.

Funds

None.

Authors' Contributions

Conception, design and drafting of article: LR, SH
Critical revision of articles for content: SG, HS, PA, AM
Provision of study materials: LR, SG

Management of the case: HS, PA, AM, LR
Administrative & technical Support: LR

Correspondence

Dr Lavanya Rai
MD, DGO (India)
Department of Obstetrics and Gynaecology
Kasturba Medical College
Manipal University
Manipal – 576 104
India
Tel: 0820 2922176
Fax: 91-820-2571934, 2570061/62
Email: railavanya@yahoo.com
lavanya.rai@manipal.edu

References

1. Sebire NJ, Foskett M, Paradinas FJ. Outcome of twin pregnancies with complete hydatidiform mole and healthy co-twin. *Lancet*. 2002;**359**:2165–2166. doi: 10.1016/S0140-6736(02)09085-2.

2. Elizabeth A, Danemond S, Hull A. Placental pathology casebook. Complete hydatidiform mole with coexistent term twin pregnancy. *J Perinatol*. 2001;**21(1)**:72–75.

3. Sánchez-Ferrer ML, Machado-Linde F, Martínez-Espejo Cerezo A, Peñalver Parres C, Ferri B, López-Expósito I, et al. Management of dichorionic twin pregnancy with a normal fetus and a androgenetic diploid complete hydatidiform mole. *Fetal Diagn Ther*. 2013;**33(3)**:194–200. doi: 10.1159/000338926.

4. Vejerslev LO. Clinical Management and diagnostic possibilities in Hydatidiform Mole with coexistent fetus. *Obstetric Gynecol Surv*. 1991;**46(9)**:577–588

5. Lee SW, Kim MY, Chung JH, Yang JH , Lee YH, Chun YK. Clinical Findings of Multiple Pregnancy With a Complete Hydatidiform Mole and Coexisting Fetus. *J Ultrasound Med*. 2010;**29(2)**:271–280.

6. Stellar MA, Genest DR, Bernstein M, Lage JM, Goldstein DP, Berkowitz RS. Natural history of twin pregnancy with complete hydatidiform mole and coexisting fetus. *Obstet Gynecol*. 1994;**83(1)**:35–42.

7. Massardier J, Golfier F, Journet D, Frappart L, Zalaquett M, Schott AM, et al. Twin pregnancy with complete hydatidiform mole and co existent fetus: Obstetrical and oncological outcomes in a series of 14 cases. *Euro J Obstet Gynecol Reprod Biol*. 2009; **143(2)**:84–87. doi: 10.1016/j.ejogrb.2008.12.006.

8. Suri S, Davies M, Jauniaux. Twin pregnancy presenting as a previa complete hydatidiform mole and co existing fetus complicated by a placental abscess. *Fetal Diagn Ther*. 2009;**26(4)**:181–184. doi: 10.1159/000253272.

9. Marcorelles P, Audrezet MP, Le Bris MJ, Laurent Y, Chabaud JJ, Ferec C, et al. Diagnosis and outcome of complete hydatidiform mole coexisting with a live twin fetus. *Eur J Obstet Gynecol Reprod Biol*. 2005;**118(1)**:21–27. doi: 10.1016/j.ejogrb.2004.02.042.

10. 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 Reports Obstetr Gynecol*. 2012; **2012(2012)**:4. doi: 10.1155/2012/405085.

11. Sánchez- Ferrer ML, Hernández-Martíne F, Machado-Linde F, Ferri B, Carbonel P, Nieto-Diaz A. Uterine rupture in twin pregnancy with normal fetus and complete hydatidiform mole. *Gynecol Obstet Invest*. 2014;**77(2)**:127–133. doi: 10.1159/000355566.

12. Guven ESG, Ozturk N, Deveci S, Hizli D, Kandemir O, Dilbaz S. Partial Molar Pregnancy and coexisting fetus with Diploid Karyotype. *J Matern FetalNeonatal Med*. 2007;**20(2)**:175–181. doi: 10.1080/14767050601134991.

13. Bristow RE, Shumway JB, Khouzami AN, Witter FR. Complete hydatidiform mole and surviving coexistent twin. *Obstet Gynecol Surv*. 1996;**51(12)**:705–709.

14. Matsui H, Sekiya S, Hando T, Wake N, Tomoda Y. Hydatidiform mole a coexistent with a twin live fetus - A national Collaborative study in Japan. *Hum Reprod*. 2000;**15(3)**:608–611. doi: 10.1093/humrep/15.3.608.

15. Seiji S, Itakura A , Yamamoto T, Nagasaka T, Yamamoto E, Ino K, Kikkawa F. Genetically identified complete hydatidiform mole coexisting with a live twin fetus: Comparison with conventional diagnosis. *Gynecol Obstet Invest*. 2007;**64**:228–231. doi: 10.1159/000106496.

16. Chu W, Chapman J, Persons DL, Fan F. Twin pregnancy with partial hydatidiform mole and coexistent fetus. *Arch Pathol Lab Med*. 2004;**128(11)**:1305–1306. doi: 10.1043/1543-2165%282004%29128