J. Fetal Med. (December 2015) 2:171–174
DOI 10.1007/s40556-015-0067-6

ORIGINAL ARTICLE

Complete Hydatidiform Mole with Co-existing Live Fetus: A Case Series

Maimoona Ahmed[1] · Geeta Kolar[2] · Suseela Vavilala[3] · Sunil Jaiman[4]

Received: 2 October 2015 / Accepted: 23 November 2015 / Published online: 10 December 2015

Abstract This review was undertaken to evaluate the maternal and fetal risk associated with complete hydatidiform mole with co-existing fetus (CHMF) and to assess the feasibility for continuing such pregnancies. Twin gestations with CHMF were reviewed from the hospital database from 2005 to 2014 at our center. Diagnosis was based on ultrasonography and confirmed post-delivery, histopathologically. Amniocentesis for karyotype was done for the live fetuses. Serum β-hCG levels were followed till they normalized. Six cases of CHMF were salvaged from the archives. Three had live normal babies (50 %), pregnancy was terminated in two cases for excessive bleeding (33.3 %) and one miscarried (16.7 %). One fetus exhibited 47, XXY Klinefelter syndrome while rest showed normal karyotype. Two patients required blood transfusion, one was complicated with severe pre-eclampsia whereas none showed gestational trophoblastic neoplasm. CHMF is a rare condition that can be diagnosed by ultrasonography. Continuation of such a pregnancy is an acceptable option and expectant management instead of therapeutic abortion can be pursued after weighing the possibility of fetal survival against maternal risk. Counseling of the couple and family plays a crucial role. Associated spectrum of maternal and fetal complications mandates close pre- and post-natal surveillance.

Keywords Twin pregnancy · Co-existing hydatidiform mole

Introduction

Twin pregnancy with complete hydatidiform mole and co-existing live fetus is a rare clinical entity. The diagnosis is usually made on obstetric ultrasound examination with the typical appearance of a complete mole with a live fetus in the other sac. The controversy lies in the management of such cases due to the higher incidence of maternal complications like hemorrhage, early onset severe pre-eclampsia, thyrotoxicosis, and the risk of persistent trophoblastic disease and fetal complications such as abnormal karyotype, abortion, and preterm birth [1]. The tumultuous course due to aforementioned complications, coupled with these pregnancies being associated with advanced maternal age and use of assisted reproductive techniques (indicating years of childlessness), would probably seem as the proverbial sword of Damocles for most [2]. However, despite the tribulations, termination of such pregnancies need not be the sole option.

Materials and Methods

The cases of twin gestation with complete hydatidiform mole with co-existing fetus were reviewed over a period of 10 years from 2005 till 2014 at Fernandez hospital, Hyderabad, which is a tertiary referral center. The information was compiled from the patient database. The diagnosis was based on ultrasonography (Fig. 1).

✉ Maimoona Ahmed
azulemontana@gmail.com

[1] High Risk Pregnancy and Perinatology, Fernandez Hospital, Hyderabad, India

[2] Department of Fetal Medicine, Fernandez Hospital, Hyderabad, India

[3] Department of Fetal Medicine, Fernandez Hospital, Hyderabad, India

[4] Department of Perinatology, Fernandez Hospital, Hyderabad, India

Amniocentesis for fetal karyotype was done for the live fetuses. Pregnancy was terminated in cases with severe maternal or fetal complications. Following delivery, the diagnosis was confirmed on histopathological examination. Serum β-hCG levels were followed up till they normalized.

Results

There were total 47,256 deliveries at our hospital in the study period. Of these, six (0.01 %) cases were of twin gestation with complete hydatidiform mole and co-existing fetus. The clinical presentation of the cases and the outcome are summarized in Table 1. The mean maternal age was 27.3 years. Mean gestational age at diagnosis was 20 weeks whereas mean gestational age at delivery was 24.5 weeks. Four mothers had live fetuses with a normal karyotype, one had a fetus with 47, XXY Klinefelter syndrome (Fig. 2) while in one case, which had presented with inevitable miscarriage, karyotype could not be done.

Of the six cases, one (16.7 %) had spontaneous miscarriage; in two cases (33.3 %), pregnancy was terminated for excessive bleeding; and three patients (50 %) delivered live and normal babies but all three had preterm deliveries. Cesarean section rate was 66.7 %. All the six cases had weekly follow-up of serum β-hCG levels and showed falling trend. No case had persistent gestational trophoblastic neoplasm. Histopathological examination confirmed the presence of complete hydatidiform mole along with the normal placenta.

Discussion

The incidence of complete hydatidiform mole with co-existing fetus has been reported as one in 20,000 to one in 1,00,000 pregnancies [3], while the incidence at our institute, over the past 10 years, has been mammoth one in 7692 pregnancies. This disproportionately higher incidence could possibly be explained by our hospital being a tertiary referral center with a specialized fetal medicine unit.

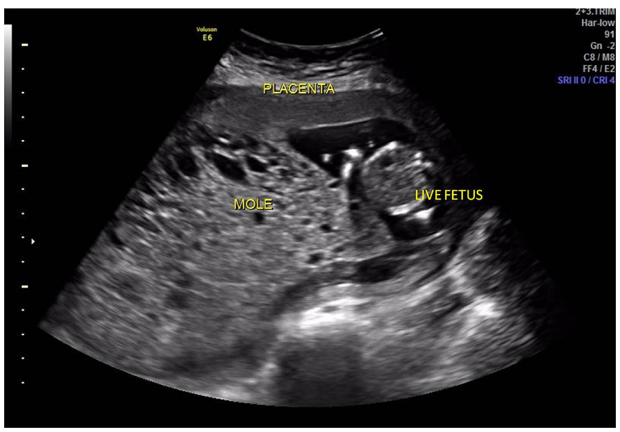

Fig. 1 Ultrasonography image showing the two separate sacs, one with complete mole and other with live fetus

Molar pregnancy can be divided on the basis of cytogenetics, histopathology, and morphology as partial mole or complete mole. A complete mole has a diploid karyotype, no embryo and amnion, and uniform changes of placental villi and trophoblasts. A partial mole, on the other hand, has a triploid karyotype, the presence of an embryo and only focal changes of placental villi and trophoblasts [4, 5]. A complete hydatidiform mole with a co-existing live fetus is a separate special entity and should be differentiated from a partial mole [6]. This is done on the basis of ultrasonography. An early scan showing two gestational sacs or a later scan demonstrating the intertwin membrane, confirms the diagnosis of complete mole with a co-existing fetus. The typical ultrasonographic findings of a molar pregnancy consist of a complex cystic pattern with a 'snowstorm' appearance [7]. Clinically, the patient may present with hyperemesis, hyperthyroidism, vaginal spotting or even heavy bleeding, pregnancy-induced hypertension, and larger-than-gestational age uterus. However, these conditions are not always present. Though ultrasonography is deemed the cornerstone for the diagnosis, confirmation requires histopathological examination.

Prenatal testing of the fetal karyotype is essential in deciding continuation and prognosis of the pregnancy and to differentiate between a partial mole and CHMF. A triploid karyotype indicates a partial mole in which the fetus would be severely malformed and growth restricted. A diploid fetal karyotype indicates a viable fetus with a normal placenta co-existing alongside a twin molar placenta. Most cases in literature have reported the co-existing live normal fetus [8]. In such cases, the pregnancy can be allowed to continue since it has a considerable chance to result in a normal live neonate. Two cases in literature have reported the co-existing live fetus with anencephaly [8]. In the present case series, karyotyping was done in five cases, of which, four were normal and one with 46, XXY karyotype Klinefelter syndrome. None of the cases had any anatomical abnormalities. In the present study, the mean age of diagnosis was 20 weeks. With the introduction of the nuchal scan (11 to 13+6 weeks), early diagnosis is possible in the first trimester itself and the couple can have the option of a safer termination of pregnancy, should that be their choice.

Parents who choose to continue a twin pregnancy with CHMF should be counseled about the risk of possible maternal complications associated with molar pregnancy such as early-onset pre-eclampsia, hyperemesis gravidarum, hyperthyroidism, vaginal bleeding, anemia, development of theca lutein ovarian cysts, respiratory distress

Table 1 Clinical presentation and outcomes

No	History	Gestational age at diagnosis (weeks)	Amniocentesis (Kt)	Maternal complication	Gestational age at delivery (weeks)	Outcome
1	Primi, 25 years, spontaneous conception	20	Normal	None	28	Preterm delivery 1.2 kg female, alive and healthy
2	G3A2, 28 years, spontaneous conception	13	Normal	Threatened miscarriage, anemia	17	Spontaneous miscarriage
3	Primi, 34 years, spontaneous conception	23	Normal	Pre-eclampsia, Antepartum hemorrhage, Postpartum hemorrhage	28	Emergency LSCS for APH, 1.6 kg male baby, alive and healthy
4	Primi, 30 years, OI conception	21	Not done	Threatened miscarriage, vaginal bleeding, blood transfusion	21	Termination of pregnancy for inevitable miscarriage
5	G2P1L1, 22 years, previous CS	23	Normal	None	36	Elective LSCS for FGR 1.8 kg male alive and healthy
6	Primi, 25 years, OI conception	14	**47 XXY**	Threatened miscarriage, anemia, blood transfusion	15	Emergency hysterotomy for uncontrolled bleeding

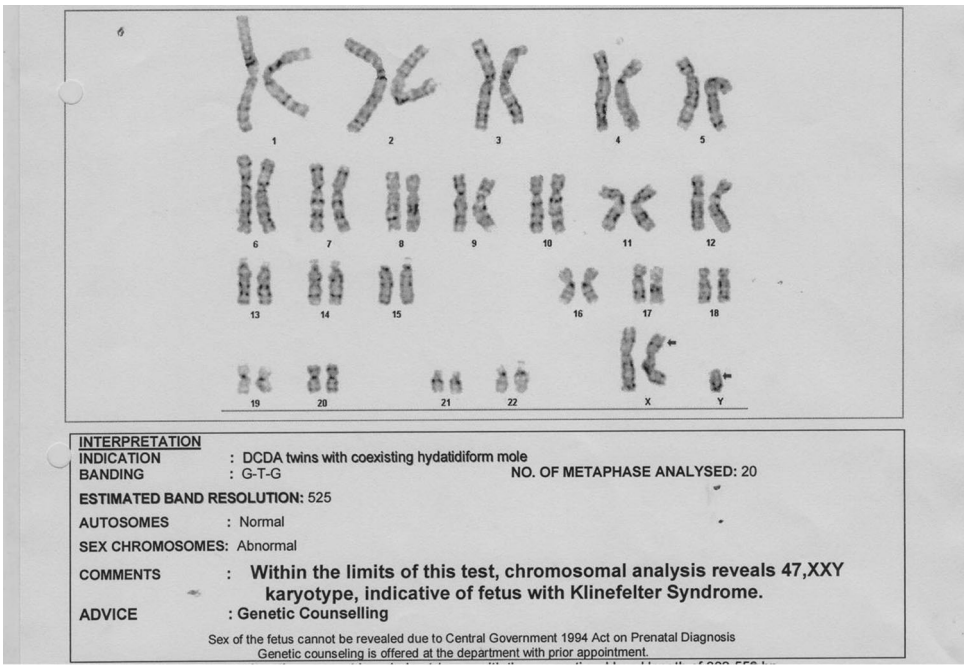

Fig. 2 Karyotype report showing 47 XXY, Klinefelter syndrome

because of trophoblastic embolization to the lungs, and persistent trophoblastic disease. Parents must also be aware that these complications may lead to fetal intrauterine growth restriction, fetal distress, and premature delivery [9].

Vaisbuch et al. [10] reported 130 cases of twins with CHMF pregnancy, of which, 41 % were terminated because of the positive probability of serious maternal complications. Previous studies have reported a greater risk of preterm delivery in women who had a twin pregnancy with CHMF (50–60 %) compared with a singleton molar pregnancy (15 %) [11]. Other studies have also shown an increased risk of persistent trophoblastic disease in such cases [12]. However, a recent study by Neimann et al. [13]

in 2007 revealed that the risk of preterm delivery after a diploid mole with a viable fetus is similar to that after a singleton molar pregnancy, and elective early termination of such pregnancy, because of the risk of preterm delivery alone, should not be recommended. They also concluded that the risk of persistent trophoblastic disease after a diploid mole with co-existing fetus pregnancy is similar to that after a singleton molar pregnancy, and expectant management instead of therapeutic abortion can be pursued.

Sebire et al. [14] reviewed 77 total cases. Of these, 53 decided to continue the pregnancy, 23 spontaneously aborted before 24 weeks, 28 pregnancies lasted more than 28 weeks resulting in 20 livebirths (40 %). There was no statistically significant difference in the occurrence of persistent trophoblastic disease in the women who terminated the pregnancy in the first trimester and those who continued their pregnancy, thus showing that the risk of persistent trophoblastic disease does not increase with advancing gestational age. Piura et al. [15] reviewed 24 studies that reported 30 cases of CHMF resulting in a live birth documented in detail. Cesarean section was reported due to fetal or maternal complications in 14 of 30 cases (46.7 %).

The management of complete mole after evacuation or delivery is similar whether there is a co-existing fetus or not. Uterine evacuation is followed by β-hCG monitoring. Chest radiography is mandatory. Additional imaging investigations may be needed as directed by symptoms. β-hCG is performed weekly until ascertainment of normal values for two consecutive weeks; then monthly up to one year. Effective means of contraception are recommended to avoid pregnancy for at least 6–12 months. Chemotherapy is indicated if β-hCG levels are persistent or rising, or metastasis to lungs or other sites appear. Hysterectomy may be needed for life-threatening hemorrhage or in a patient with rising β-hCG titres, but no evidence of metastasis, who does not desire fertility or refuses chemotherapy [16].

Conclusion

Twin pregnancy with complete hydatidiform mole and co-existent live fetus is a rare condition that can be diagnosed by obstetric ultrasound. Termination versus expectant management should be decided after karyotype and detailed anatomical survey of the live fetus. Decision to continue the pregnancy should be taken after weighing the possibility of fetal survival against maternal risk. Counseling of the couple and family plays a crucial role in such cases. Those who choose to continue the pregnancy, the management has to be in a tertiary care hospital with strict vigilance on maternal, fetal, and neonatal condition. Long term follow-up with serum β-hCG levels is mandatory.

Compliance with ethical standards

Conflict of interest None.

References

1. Matsui H, Sekiya S, Hando T, Wake N, Tomoda Y. Hydatidiform mole coexistent with a twin live fetus: a national collaborative study in Japan. Hum Reprod. 2000;15:608–11.
2. Montes-de-Oca-Valero F, Macara L, Shaker A. Twin pregnancy with a complete hydatidiform mole and co-existing fetus following in-vitro fertilization. Hum Reprod. 1999;14:2905–7.
3. Malhotra N, Deka D, Takkar D, Kochar S, Goel S, Sharma MC. Hydatiform mole with coexisting live fetus in dichorionic twin gestation. Eur J Obstet Gynecol Reprod Biol. 2001;94:301–3.
4. Szulman AE, Surti U. The syndromes of hydatidiform mole. (I) Cytogenetic and morphologic correlations. Am J Obstet Gynecol. 1978;131:665–71.
5. Szulman AE, Surti U. The syndromes of hydatidiform mole. (II) Morphologic evolution of the complete and partial mole. Am J Obstet Gynecol. 1978;132:20–7.
6. Chen Fang-Ping. Molar pregnancy and living normal fetus coexisting until term: prenatal biochemical and sonographic diagnosis. Hum Reprod. 1997;12(4):853–6.
7. Ferraz TJ, Bartosch CM, Ramalho CM, Carvalho FA, Carvalho BC, Brandao OG, Montenegro NA. Complete mole in a dichorionic twin pregnancy after intracytoplasmic sperm injection. Revista Brasileira de Ginecologia e Obstetricia. 2013;35(1):39–43.
8. Sarah A, et al. Metastatic gestational trophobalstic disease following a complete hydatidiform mole coexistent with an anencehalic fetus diagnosed at 10 weeks gestation. J Ultrasound Med. 2008;27:1533–6.
9. RCOG. The management of gestational trophoblastic disease. Green-top guideline no. 38. London: RCOG; 2010.
10. 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(1):19–23.
11. 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(1):35–42.
12. 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(1):197–202.
13. Niemann I, Sunde L, Petersen LK. Evaluation of the risk of persistent trophoblastic disease after twin pregnancy with diploid hydatidiform mole and coexisting normal fetus. Am J Obstet Gynecol. 2007;197(1):45-e1–5.
14. 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(9324):2165–6.
15. Piura B, Rabinovich A, Hershkovitz R, Maor E, Mazor M. Twin pregnancy with a complete hydatidiform mole and surviving coexistent fetus. Arch Gynecol Obstet. 2008;278(4):377–82.
16. Makary Raafat, et al. Twin gestation with complete hydatidiform mole and a coexisting live fetus: case report and brief review of literature. Obstet Med. 2010;3:30–2.