Two cases of complete hydatidiform mole and coexistent live fetus

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Summary

The aim of this study was to report the clinical features, management, and outcome of complete hydatidiform mole with a coexisting viable fetus. Two cases are reported.
In both cases ultrasound examination demonstrated a normally growing live fetus alongside a normal placenta and an additional intrauterine echogenic mass with features of hydatidiform mole. The hCG levels were significantly increased and fetal karyotypes were normal. A cesarean section performed at 28 weeks' gestation in the first case and at 26 weeks' gestation in the second one resulted in the delivery of live normal infant and two adjoining placentas in both cases. Microscopic examination of the abnormal placentas confirmed complete hydatidiform mole. The babies did well and serial maternal serum hCG levels showed a declining trend and were undetectable by a few months after delivery.
Continuation of a twin pregnancy with complete hydatidiform mole (CHMF) is an acceptable option. There is, although, an increased risk of developing maternal and fetal complications. Close surveillance of an ongoing pregnancy is compulsory to detect potential early signs of complications.

Key words: twin pregnancy, complete hydatidiform mole, gestational trophoblastic disease.

Introduction

Gestational trophoblastic diseases are neoplasias originating from the placenta. Twin pregnancy with a complete hydatidiform mole and a normal fetus is extremely rare, with an estimated incidence of one in 22,000-100,000 pregnancies. The main issue is to differentiate between two diagnoses: dichorionic twin pregnancy with normal fetus (46 chromosomes, 23 maternal and 23 paternal) and complete molar pregnancy (46 chromosomes, all paternal) and singleton pregnancy consisting of a triploid fetus with partial hydatidiform mole placenta (69 chromosomes, 23 maternal and 46 paternal). Twin pregnancy with CHMF (complete hydatidiform mole coexisting with a live twin fetus) resulting in a healthy take-home baby is rare, with only 56 cases documentated in detail in literature. CHMF cases are at high risk of spontaneous abortion, preterm delivery, intrauterine fetal death, bleeding, preeclampsia, persistent trophoblastic disease (PTD). We report two cases encountered at the Department of Obstetrics and Gynaecology, University of Bari, of a twin pregnancy with a CHMF that resulted in live newborns.

Case Report 1

The patient was a 37-year-old woman, Gravida 2, Para 1, with one previous vaginal delivery of a normal female infant. Chorionic villus sampling (CVS) revealed a normal 46 XY karyotype. Ultrasound examination at 13 weeks of gestation was interpreted as a normal pregnancy. Successive examination at 19 weeks of gestation showed a live fetus matching the age of gestation alongside a normal-looking placenta located at the anterior uterine wall and an additional echogenic mass resembling molar placenta located at the posterior uterine wall. Normal ovaries were observed. On admission at 26 week ultrasound examinations confirmed previous ultrasound examination (Fig. 1 A, B). Ultrasound scans were performed with Aloka Prosound α10® device. On biochemical screen elevated levels of β-hCG (231,000 mUI/ml) and AFP (235 ng/ml) were noted. Subsequent investigations confirmed normal thyroid function tests, normal blood pressure, no proteinuria and normal chest X-ray. The patient was stable for the next 5 days after admission, when an emergency cesarean section was performed due to a non-reactive fetal monitoring with regular uterine contractions (cardiotocography was carried out with Philips Series 56 A® device) and maternal bleeding. A live male infant (1025 g) with an Apgar score 6/8 at 1 min and 5 min was delivered. Both normal and molar placentas were separated manually from the inner uterine wall and extracted completely (Fig. 2). On pathological examination, one placenta was 13 x 10 x 2 cm, weighed 230 g with normal basal and chorial plates and an umbilical cord. The second one measured 17 x 15 x 3 cm, weighed 600 g and was made up of large vesicular villi. The microscopic findings consisted of complete mole with trophoblastic atypias, mild hyperplasia, necrosis and spontaneous degeneration areas.
No postoperative complications developed. Two weeks after delivery, the serum β-hCG level was 966 mIU/mL and normalized gradually within 6 months without any citotoxic therapy and with no evidence of persistent or metastatic disease. Careful follow-up showed no sign of PTD. The baby was discharged from hospital weighing 2080 g at 64th postoperative day. Now he is two years old and in good health.

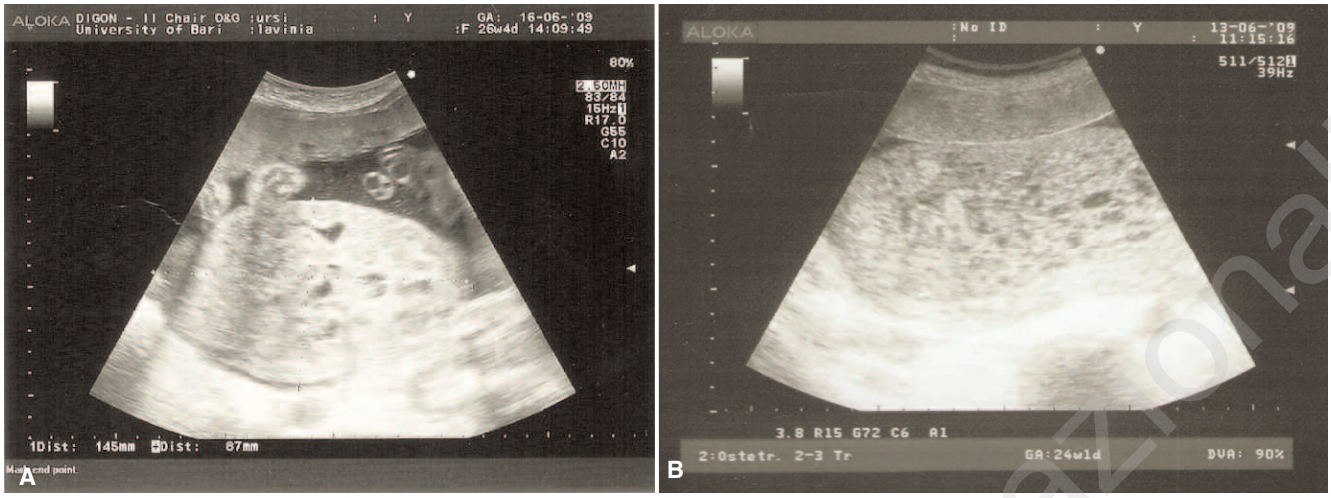

Figure 1 A, B. Ultrasound scan of normal (superior) and molar (inferior) placenta (A); an enlarged view of molar placenta (B).

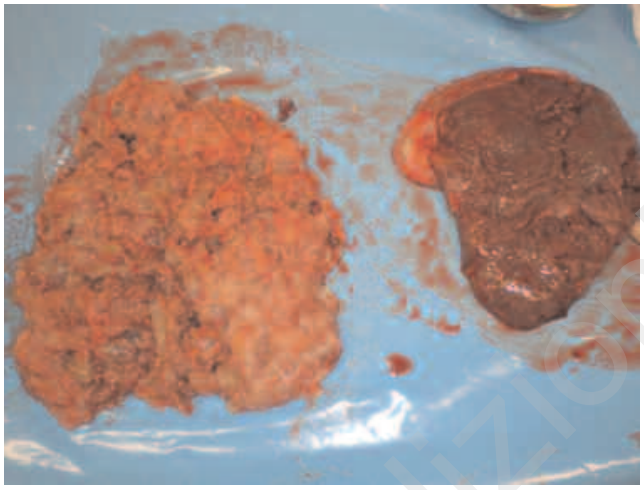

Figure 2. Macroscopic appearance of normal placenta (right) and hydatidiform mole (left).

Case Report 2

A 22 year old woman, Gravida 0, Para 0 presented at 16 weeks of gestation with mild vaginal bleeding. The pregnancy was achieved following one cicle of ovulation induction with clomiphene citrate. Ultrasound examination performed on admission showed viable fetus with normal anatomy and placenta located at the anterior uterine wall, and a second multicystic molar-appearing placenta located at the posterior wall of the uterus. The ovaries seemed to be normal. Ultrasound scans were performed with Aloka Prosound α10®. Amniocentesis revealed a normal 46 XX karyotype. Laboratory tests showed elevated serum β-hCG level (353,273 mUI/ml) and AFP (274 ng/ml). Thyrotropin (TSH) was below 0,005 IU/ml, with normal free thyroxine (FT 4). Blood pressure and chest X-ray were normal and no proteinuria was found. The patient was discharged home at 17 weeks' gestation and the couple was informed about the possible complications and was invited to a close monitoring of pregnancy. A further ultrasound examination at 21 weeks of gestation confirmed a normal live female fetus alongside a normal looking placenta and co-existent abnormal placenta with definite sonographic characteristics of hydatidiform mole (Fig. 3 A-C).

The patient was stable until 26 weeks' gestation when preterm uterine contractions and vaginal bleedings started so an emergency cesarean section was performed. A 850 g normal female infant was delivered with Apgar Score of 5/8. Both molar and normal placentas were manually extracted. Pathological examination confirmed the diagnosis of complete hydatidiform mole. There were no complications after the delivery and the patient was discharged from the hospital on the fifth postoperative day. The serum hCG level fell to 1598 mUI/ml and normalized totally within 5 weeks and until now there is no evidence of PTD. The baby is now in good health and she is four months old.

Discussion

There have been so far, about 200 cases of twin pregnancy with CHMF fully documented in literature, while only 56 cases result in a live birth (1). In the late 1970s, Vassilakos et al. firstly described two different pathologic entities, partial and complete hydatidiform mole (CHM), with different mechanisms of origin based on cytogenetic analysis (2). Partial moles derive from dispermic fertilization of a haploid normal oocyte and produce a triploid set of chromosomes. A CHM contains a diploid set of 46 chromosomes, all of paternal origin and no traces of fetal parts can be identified. Complete and partial moles have distinct fetal and maternal complications. In the combination of a partial hydatidiform mole, the fetus is almost always triploid and the indication for a termination of pregnancy is evident. In contrast, the fetus may be normal in a twin pregnancy with a CHMF and continuation of pregnancy is frequently associated with severe maternal complications, leading to a mother vs. fetal clinical problem (3).

The management of such pregnancies can be either immediate termination of pregnancy to avoid the potential maternal complications (3, 4). The true incidence of this rare entity is difficult to establish, and some suggest that the modern increased incidence of iatrogenic multiple gestations will cause a higher incidence of CHMF (1).

Ultrasonography has made it possible a diagnosis of a hydatidiform mole and co-existent fetus in the first trimester (5). Prenatal testing of at least fetal karyotype is essential in deciding continuation and prognosis of the pregnancy. A triploid karyotype indicates a triploid fetus which necessarily would be severely malformed and, in this case, termination of pregnancy is recom-

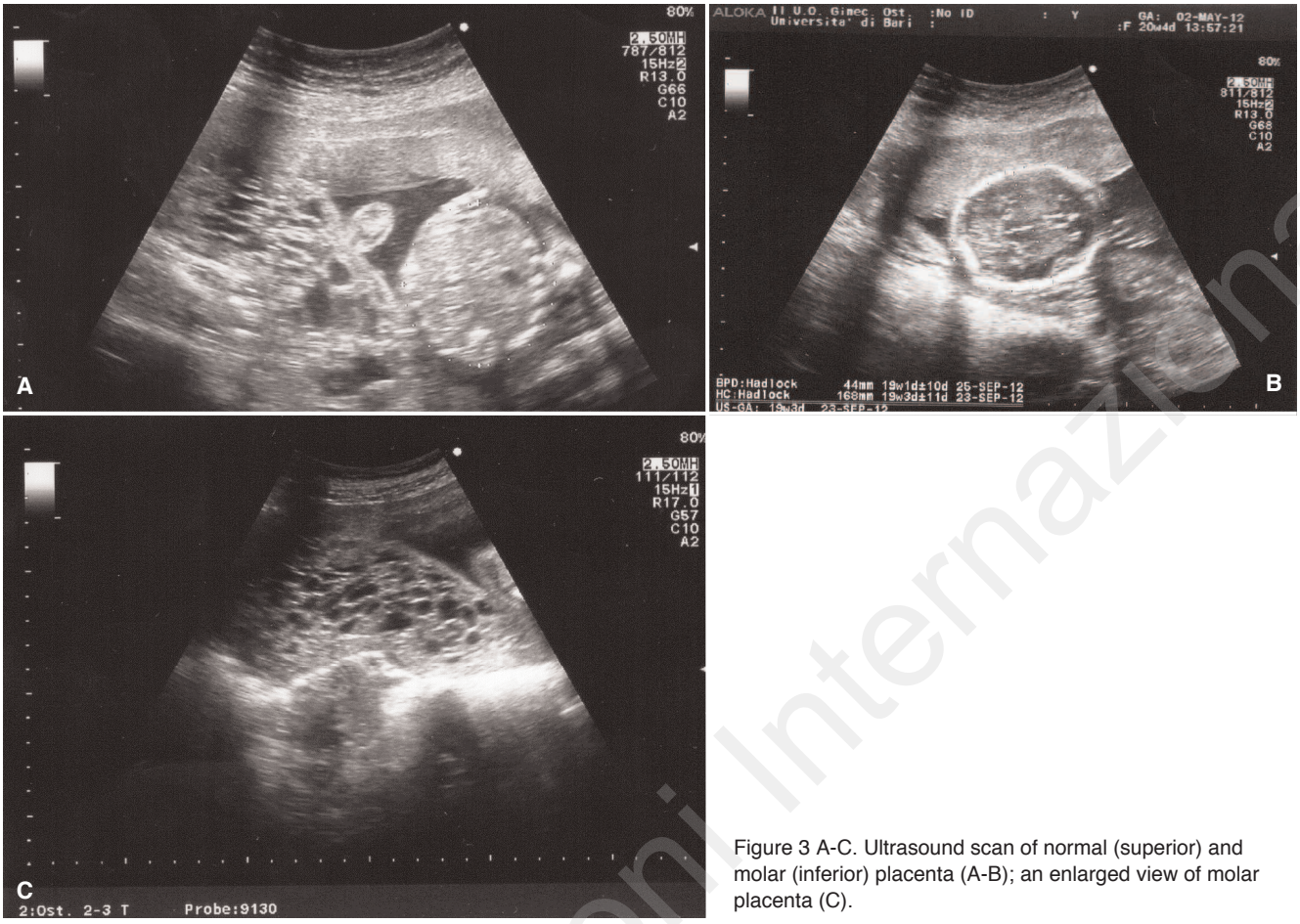

Figure 3 A-C. Ultrasound scan of normal (superior) and molar (inferior) placenta (A-B); an enlarged view of molar placenta (C).

mended. A diploid fetal karyotype (46 chromosomes, 46 XX or 46 XY, 23 maternal and 23 paternal) indicates a viable fetus with a normal placenta co-existing alongside a twin molar placenta, as in our cases. In such a case, the pregnancy can be allowed to continue since it has a considerable chance to result in a normal live neonate. Nevertheless, parents who choose to continue a twin pregnancy with CHMF should agree to take 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 because of trophoblastic embolization to the lungs, and PTD. Parents must also be aware that these complications may lead to fetal intrauterine growth retardation, fetal distress and premature delivery (6). As concerns the risk of PTD, Steller et al. suggest that it is higher in cases of CHMF compared with single molar pregnancies and that, when present, it more commonly progresses to metastatic disease (7). Sebire and Niemann find out that the risk of PTD after CHMF is not significantly higher than in single molar pregnancies (8, 9). A "wait-and-see" approach should be considered rather than immediate termination of pregnancy, because the risk does not increase with advancing gestational age. In our experience no patients developed PTD after one year follow up carried out with montly β-hCG level and ultrasound examinations, so we suppose that the risk is comparable in both single molar pregnancies and CHMF.

Diagnosis should also include molar placental karyotype (10). Although not available for our patients, as in most documented cases (1, 3, 5, 10), we are convinced that both our cases were CHMF due to diploid karyotipe, normal newborn, ultrasound demarcation between the normal and molar placenta and histopatological examinations. The prenatal diagnosis of a twin pregnancy with CHMF was reinforced in our cases by demonstrating in the first one a normal fetal 46 XY karyotype by CVS at the first trimester of the pregnancy, and in the second one a normal fetal 46 XX karyotype by amniocentesis at the second trimester. The diagnosis was definitively confirmed after delivery, by histological examination of the molar placenta in both cases.

Conclusions

The prenatal diagnosis of twin pregnancy with CHMF was based in these patients on ultrasound findings, abnormally elevated β-hCG levels and normal fetal karyotype. In the past, most CHMF gestations were terminated immediately following diagnosis because of poor information concerning clinical features and natural history. This circumstance has changed in recent years and that pregnancy may be continued when fetal anomalies and abnormal karyotype are excluded (10-12). However, close surveillance to detect potential early signs of maternal and fetal complications is compulsory and a detailed discussion (and informed consent) with the couple is necessary.

Parallel to the literature, our opinion is that these pregnancies should continue under close follow-up during gestational time and after to exclude PTD.

References

1. Dolapcioglu K, Gungoren A, Hakverdi S, Hakverdi AU, Egilmez E. Twin pregnancy with a complete hydatidiform mole and co-existent live fetus: two case reports and review of the literature. Arch Gynecol Obstet 2009; 279:431-436.
2. Moini A, Riazi K. Molar pregnancy with a coexisting fetus progressing to a viable infant. Int J Gynecol Obstet 2003; 82:63-64.
3. 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.
4. Bruchim I, Kidron D, Amiel A, Altaras M, Fejgin MD. Complete hydatidiform mole and a coexistent viable fetus: report to two cases and review of the literature. Gynecol Oncol 2000; 77:197-202.
5. Montes-de-Oca-Valero F, Macara L, Shaker A. Twin pregnancy with a complete mole and co-existing fetus following in vitro fertilization. Hum Reprod 1999; 14:2905-2907.
6. Matsui H, Sekiya S, Hando T, Wake N, Tomoda Y. Hydatidiform mole coexisting with a twin live fetus: a national collaborative study in Japan. Hum Reprod 2000; 15:608-611.
7. Steller MA, Genest DR, Bernstein MR, et al. Natural history of twin pregnancy with complete hydatidiform mole and coexisting fetus. Obstet Gynaecol 1994; 83:35-42.
8. Sebire NJ. Prenatal diagnosis and management of twin pregnancies complicated by a co-existing molar pregnancy. Prenat Diagn 2006; 26(4):373.
9. Niemann I, Sunde L, Peterson 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; 197:451-455.
10. Piura B, Rabinovich A, Hershkovitz R, Maor E, Mazor M. Twin pregnancy with a complete hydatidiform mole and surviving co-existent fetus. Arch Gynecol Obstet 2008; 278:377-382.
11. Klatt TE, Franciosi RA, Cruikshank DP. Normal fetus with a twin presenting as both a complete hydatidiform mole and placenta previa. Obstet Gynecol 2006; 107:527-530.
12. Shahabi S, Naome G, Cobin L, Veroughstruele A, Musters L, Zengbe V, Noel JC, Avni EF, Donner C, Vershet A, Foulon W, Rodesch F. Complete hydatidiform mole and coexisting fetus. A case report of two cases with contrasting outcomes. J Reprod Med 1977; 42:756-760.