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Case Report

Prenatal diagnosis and management of pregnancy complicated by a coexisting mole: A case report ☆

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ABSTRACT

Twin pregnancies with a complete hydatidiform mole and a coexisting live fetus are rare. The incidence is estimated to be 1 in 20,000-100,000 pregnancies. Prenatal diagnosis can be made with ultrasound findings, abnormally elevated β-hCG levels, and fetal karyotype. There are various complications following these pregnancies which include hyperemesis gravidarum, vaginal bleeding, spontaneous abortion, pre-eclampsia, intrauterine growth retardation, preterm delivery, and persistent trophoblastic disease. We report an interesting case of twin pregnancy consisting of a complete hydatidiform mole and a normal fetus achieved with in-vitro fertilization in a primary infertile couple. Suspicion of molar pregnancy was made on ultrasound examination, but the couple refused other prenatal testing and wanted to continue the pregnancy. Although the pregnancy was at high risk because of the patient's age and complications associated with a molar pregnancy, a vigorous female baby was delivered at term. The purpose of this report is to present a case of a rare obstetric condition, give evidence that gestational trophoblastic disease is occurring more commonly in multiple gestations and in-vitro fertilization pregnancies, and highlight the importance of ultrasound in prenatal diagnostics and monitoring of high-risk pregnancies.

Introduction

Twin pregnancies with a complete hydatidiform mole and a viable live fetus are very rare, with an estimated incidence of 1 in 20,000-100,000 pregnancies [1,2]. Asian countries have the highest incidence. There is also an increased risk in women over the age of 35 [3]. These pregnancies are commonly associated with various complications including vaginal bleeding, anemia, spontaneous abortion, pre-eclampsia, preterm delivery, intrauterine fetal death, and persistent gestational trophoblastic disease. Therefore, it was usually recommended to

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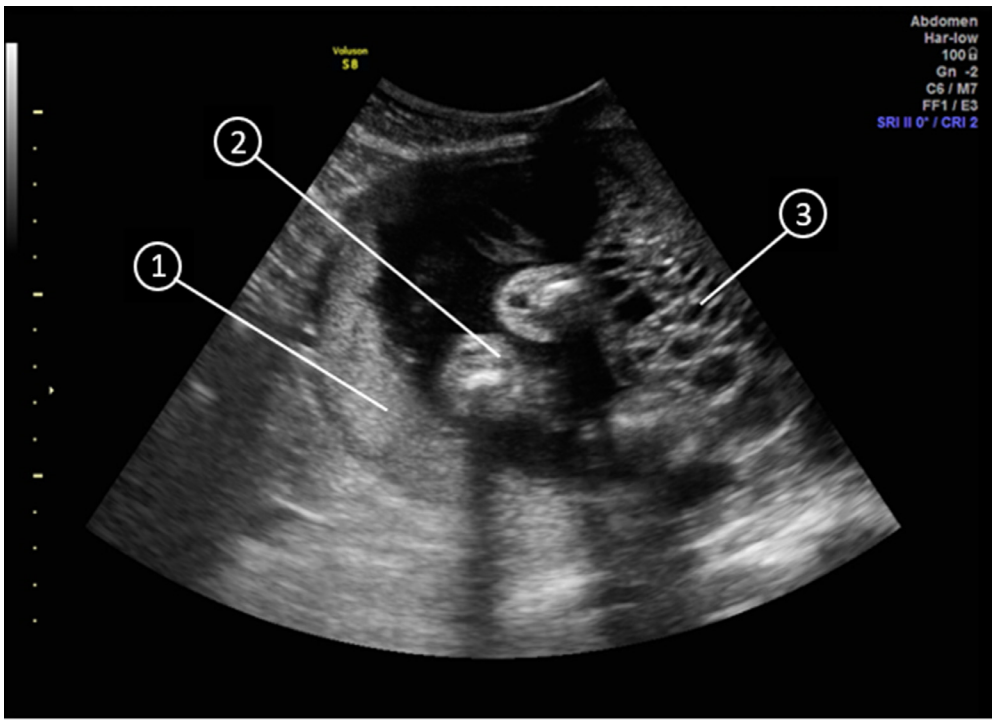

Fig. 1 – Ultrasound findings at 20 weeks gestation demonstrating the placenta (1), normal fetal parts (2), and characteristic "snowstorm" pattern of hydatidiform mole (3).

terminate such pregnancies in the past. Although there is a trend in the last decades to continue molar pregnancies if reasonable, less than 40% have a favorable outcome and result in live births [4]. We report the case of a twin pregnancy consisting of a complete hydatidiform mole and a coexisting normal fetus (CHMCF) achieved after in-vitro fertilization (IVF) in a primary infertile couple.

Case report

The patient is a 38-year-old Caucasian female (gravida 0, para 0) who was in fertility treatment with her husband for 10 years without success. The patient is healthy and takes no chronic therapy. Due to azoospermia in her husband, testicular sperm extraction and intracytoplasmic sperm injection were performed. The pregnancy was achieved through IVF followed by the transfer of 2 embryos which contained 2 apparently normal pronuclei into the uterus. The first prenatal ultrasound (US) at 7 weeks showed a dichorionic, diamniotic twin gestation. The first gestational sac was normal, measuring 24 mm in diameter and crown-rump length (CRL) of 12.1 mm, and the second gestational sac seemed empty. The second US at 11 weeks gestation showed a vital embryo (CRL 38 mm) in the first gestational sac and a snowstorm-like multicystic formation in the second gestational sac measuring 61 × 35 mm. The patient was referred to our clinic by her primary gynecologist because of high serum β-hCG (373,423.00 IU/L) and an unclear ultrasound finding suggestive of a complete hydatidiform mole. The couple was informed about the finding, and they decided to maintain the gestation but refused any kind of prenatal screening. US at 13 weeks showed a vital fetus (CRL 67 mm) and a molar mass measuring 97 × 64 mm. The pregnancy was closely monitored with serial β-hCG, thyroid function tests, and ultrasound for fetal growth and size of the molar formation. Serum β-hCG levels and size of the hydatidiform mole were at maximum at 20 weeks of gestation (Fig. 1). The patient's thyrotropin (TSH) was 0.01 mIU/L , followed by normal free thyroxine (FT4), but no therapy was introduced since she was asymptomatic. The patient was readmitted at 26 weeks with gestational hypertension, but no proteinuria was found. She received antihypertensive therapy until her blood pressure stabilized. At 30 weeks the patient presented again with vaginal bleeding containing vacuolar tissue. Vaginal bleeding stopped spontaneously after a few days of bed rest during which the pregnancy was continuously monitored. A rapid fall in β-hCG levels was noticed during observation. The pregnancy was managed conservatively until 38 weeks gestation when an elective Caesarean section was performed because of suspect intrauterine growth restriction (IUGR). A female baby with birth weight and length of 2570 g (fifth percentile) and 49 cm (50th percentile) respectively, was delivered in good condition with an Apgar score of 10/10. The placenta and molar tissue (Fig. 2) were sent for pathologic and cytogenetic analysis after delivery and the diagnosis of a complete hydatidiform mole was confirmed (Fig. 3). The patient recovered well during the follow-up and her serum β-hCG level normalized gradually within 12 weeks without any cytotoxic therapy.

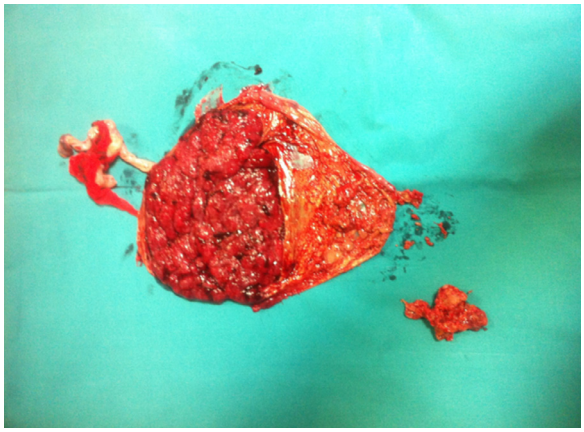

Fig. 2 – Postoperative findings-placenta and molar tissue.

Discussion

Twin pregnancy with a complete hydatidiform mole and a coexisting fetus is a rare obstetric condition. Previous studies observed that the incidences of both iatrogenic multiple gestations and molar pregnancies increased with improvement of medically assisted reproduction treatments [3,5], which is in accordance with our case where the patient conceived after IVF-ET. On the other hand, Massardier et al. [6] claimed that the stimulation of ovulation increases the rate of twin pregnancies, but does not seem to raise the risk of complete hydatidiform mole or gestational trophoblastic neoplasia. However, an increased availability of ultrasound examination in early pregnancy enables diagnosis of a molar pregnancy within the first trimester and later monitoring of these high-risk pregnancies [7]. Moreover, it is usual to do amniocentesis to obtain genetic material from the fetus with intention to determine the fetal karyotype [8]. In addition to the ultrasound finding, genetic analysis is helpful in the decision of continuation of the pregnancy and its prognosis. In our case, the molar formation was recognized during the first trimester, but the couple refused prenatal testing and decided to continue the pregnancy regardless high risk of complications and an unfavorable outcome. Since a CHMCF was determined, the pregnancy was closely monitored using ultrasound and serial serum β-hCG. Other authors also recommend using serum β-hCG level as a marker in the management of such pregnancies, because it is usually highest at the beginning of the second trimester and then gradually decreases [8,9]. A recent study on the predictors of fetal survival found that β-hCG levels up to 400,000 IU/L are the best indicators for a favorable outcome [4]. Nevertheless, molar pregnancies are often associated with severe complications, which can be a cause to terminate the pregnancy. Some studies observed a tendency for molar tissue to be in the lower segment of the uterus, consequently resulting in bleeding [10] which was also evident in our case. Except vaginal bleeding, other possible maternal complications associated with molar pregnancies are hyperemesis gravidarum, development of theca lutein ovarian cysts, hyperthyroidism, early-onset preeclampsia, respiratory distress because of trophoblastic embolization to the lungs, persistent trophoblastic disease [10,11]. In our case, hyperthyroidism was found during routine monitoring of the patient's thyroid function, but since she was asymptomatic and in euthyreosis, no medical treatment was introduced. Hyperthyroidism is usually a result of high levels of hCG which lead to TSH suppression, but stimulate the release of thyroid hormones, since the alpha subunit of hCG is homologous to the alpha subunit of TSH [12]. During a check-up at 26 weeks gestation, high blood pressure was measured in our patient, but in the absence of proteinuria and rapid stabilization after antihypertensive therapy, preeclampsia could be ruled out. The risk of persistent trophoblastic disease in a complete hydatidiform mole is established at about 16%-50%, and according to most studies, it does not depend on the duration of pregnancy [5,13]. Fetal complications in molar pregnancy include IUGR, fetal distress, and premature delivery. Although our patient showed a number of complications, they were recognized early and managed accurately so the pregnancy was successfully continued to term. At 38 weeks gestation, after thorough insight into the patient's condition, suspicion of fetal IUGR, and ultrasound finding of the molar formation localized in the lower uterine segment, a decision was made to perform a Caesarean section. Regular follow-up was carried out on the patient in the postpartum period

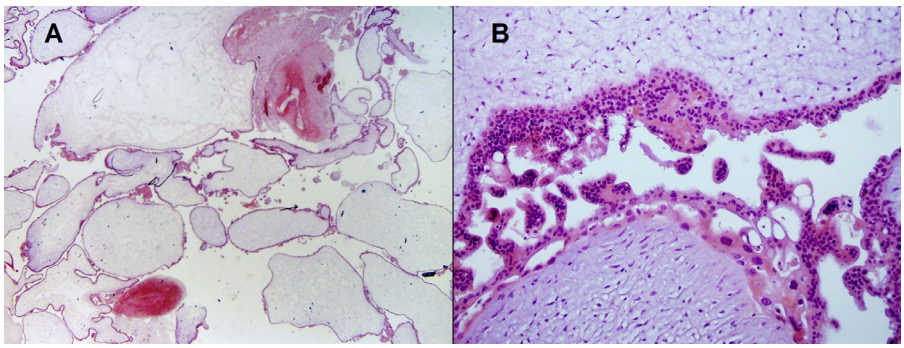

Fig. 3 – Microscopic findings of the complete hydatidiform mole using HE staining. (A) Molar tissue showing dilated avascular villi (magnification: 2x). (B) Molar tissue showing proliferation of syncytiotrophoblasts on its surface (magnification: 20x).

because of the risk of developing postnatal gestational trophoblastic neoplasia [14]. The patient recovered well and her serum β-hCG showed rapid regression which indicated that there are no signs of developing gestational trophoblastic neoplasia.

The prenatal diagnosis of twin pregnancy with CHMCF in our case was based on ultrasound findings and abnormally elevated β-hCG levels. In the past, termination of these pregnancies was recommended considering possible life-threatening maternal and fetal complications. However, as it is shown in our case, the general trend today is to continue such pregnancies, regarding the viable fetus and the fact some patients conceived late in age after multiple IVF attempts. In the presence of a normal fetus and stable pregnancy, an optimal outcome can be achieved even in high-risk patients. Prenatally, close monitoring through ultrasound and laboratory tests is important to find early signs of complications and introduce adequate treatment, and accurate care and follow-up are essential in the postnatal period.

Patient consent

The authors confirm that written, informed consent for the publication of their case was obtained from the patient.

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