Case Report

Complete hydatidiform mole with a healthy fetus, a twin pregnancy following intrauterine insemination: A case report.

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Abstract

Hydatidiform mole (molar pregnancy) is characterized by abnormal fetoplacental development and trophoblastic hyperplasia. We report an unusual case of a 26 year old woman with the diagnosis of twin pregnancy with a healthy fetus and complete mol hydatidiform following an ovulation induction and subsequent intrauterine insemination (IUI). On 14th weeks of gestation, a fetus with a positive fetal cardiac activity and a second intrauterine ex fetus with a hydropic placenta was seen on ultrasound. A chorionic villus sampling and an amniosynthesis were performed. After the diagnosis of complete mole which was confirmed by histopathology; the pregnancy was terminated with the patient's will on the 17th week of gestation.

Keywords: Intrauterine insemination (IUI); infertility; complete hydatidiform mole; twin pregnancy

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Introduction

Molar pregnancies (hydatidiform mole) are characterized by abnormal fetoplacental development and trophoblastic hyperplasia. A complete mole hydatiform (CMH) is caused by a single (90%) or two (10%) sperm combining with an oocyte which has lost its DNA. In this situation, the sperm reduplicates forming a complete 46 chromosome set (1). The genotype is typically 46,XX due to subsequent mitosis of the fertilizing sperm, but can also be 46,XY (1). A twin pregnancy consisting of a healthy fetus with a CMH is an extremely rare condition with an incidence of 1/20000-100000 (2). Ultrasound and invasive procedures are necessary for exact diagnosis. The major problem in twin pregnancies with a healthy fetus and CMH is the management of the case which is either expectant or termination of the pregnancy. The risk factors for on-going pregnancies are early preeclampsia and thyrotoxicosis as well as occurrence of a persistent trophoblastic disease (2). One of the least common complications of artificial reproductive treatments (ART) is the occurrence of CMH. Here we report; a twin pregnancy consisting of CMH with co-existing fetus after intrauterine insemination (IUI). We wanted to review the literature and management of these cases as well as the Hook effect which can be seen during b-hcg measurements.

Case Report

A 26 year old woman was accepted with secondary infertility to our artificial reproductive therapies department. After ovulation induction with gonadotropins and consequent IUI; a twin

pregnancy was achieved. On the 12th weeks of gestation, she was referred to our center with a pre-diagnosis of a partial mol hydatiform. On the ultrasound two fetal poles; one of which has a positive fetal cardiac activity of a 12 week fetus and a one of which has a negative fetal cardiac activity of a 7 week fetus, were observed (Figure 1,2). The placenta seemed hydropic (Figure 1,2). The patient's ß-HCG level was 222000 IU/ml. To conclude an exact diagnosis and determine the management; a placental biopsy and amniosynthesis (AS) were needed. On the 15th weeks of gestation AS was performed when patient's ß-HCG level was 4.256.000 IU/ml. The chest X-ray and ultrasonographic examination were all normal. The cytologic examination reported the absence of chorionic villi and the cytogenetic assessment revealed a caryotype of 46XX. Given the findings noted, a diagnosis of a twin pregnancy of CMH with a live co-existing fetus was made. The potential risk factors for the on-going pregnancy (abortion, preterm delivery, antepartum bleeding, early preeclampsia, persistent trophoblastic disease) were discussed with the family. With the decision of family, the pregnancy was terminated on 17th weeks of gestation. Six weeks after induced abortion, patient's ß-HCG level was negative and no persistent trophoblastic disease was observed in four months follow-up.

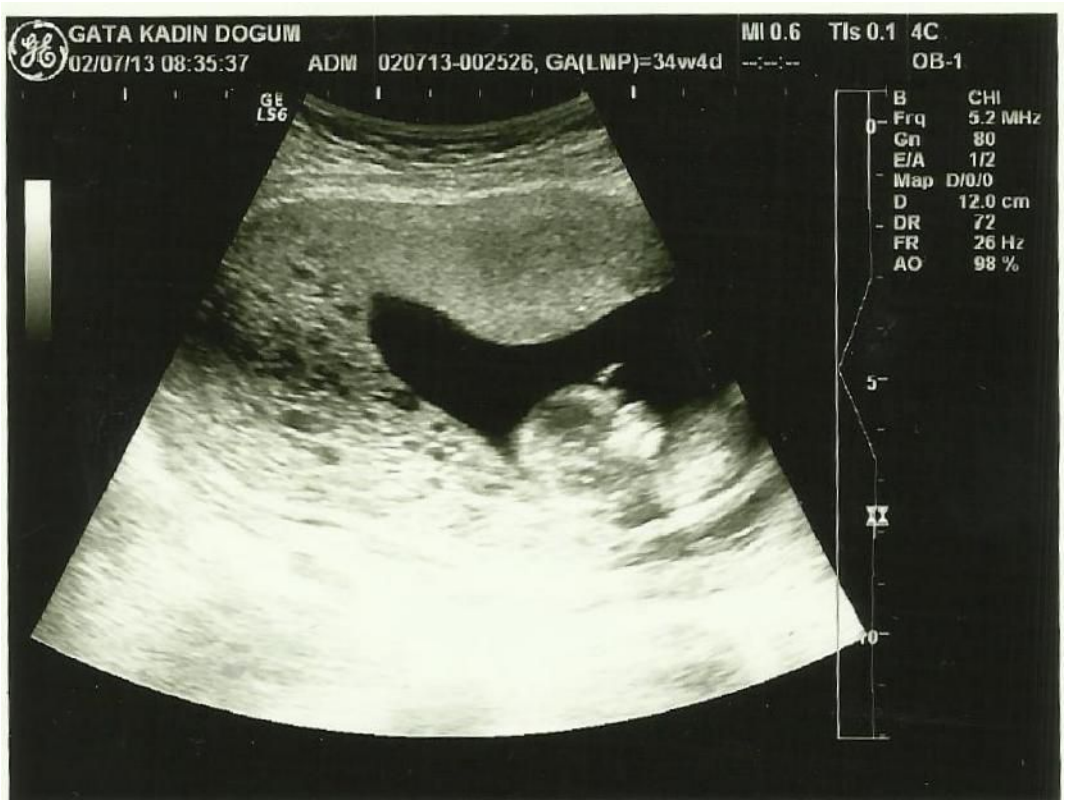

Figure 1: The 12th weeks fetus and the hydropic placenta

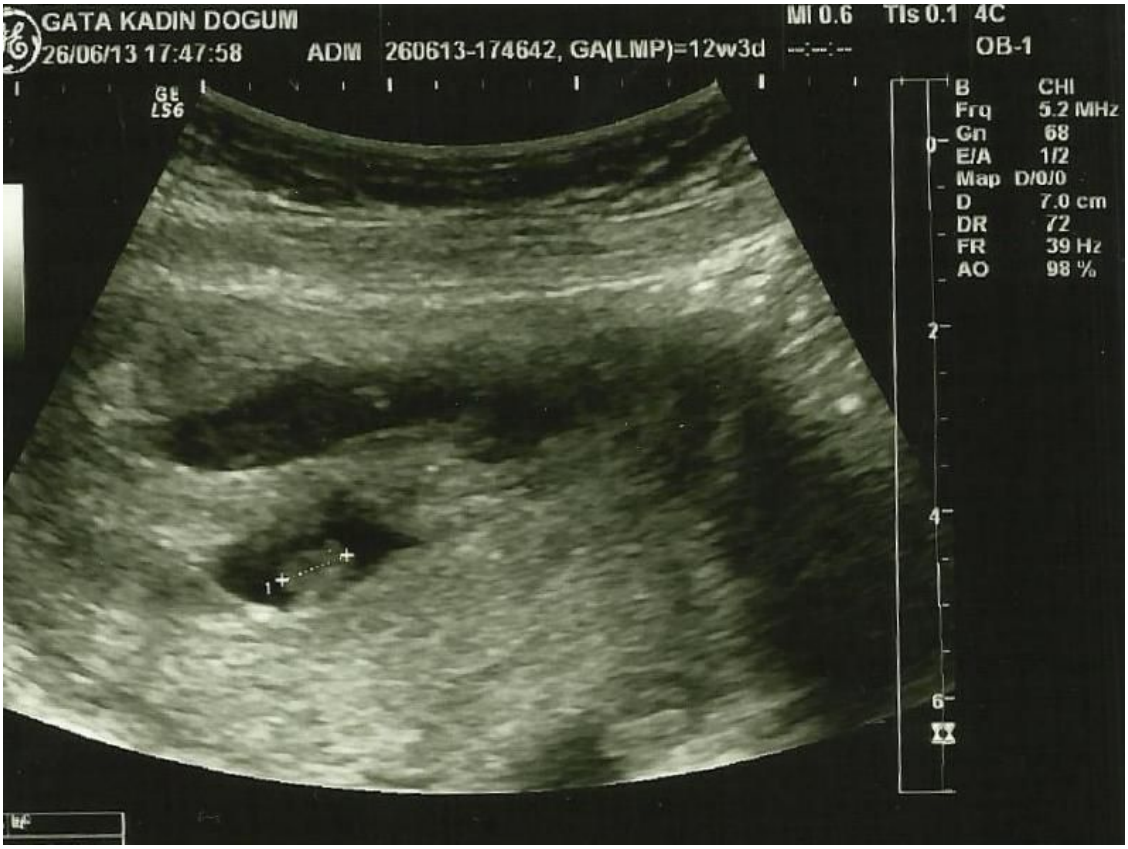

Figure 2: The second fetal pole with a negative fetal cardiac activity.

Discussion

The number of births arising by means of fertility treatment (especially ART) has risen dramatically in the last decade. Despite the widespread use of these treatments, gestational trophoblastic disease (GTD) after artificial reproductive therapies seems to be a rare complication. But it is a well-known fact that, during fertility treatments, the normal products of conception can be altered which can also be related to the development of GTD. Several case reports have documented GTD in pregnancies arising as a consequence of assisted conception (3,4). A variety of risk factors have been implicated in GTD, but no mechanism of causation has been established yet. Viral infections, poor nutrition, defective germ cell, prior pregnancies, maternal and paternal age, genetic and environmental factors and high daily progesterone supplementations were blamed (3). Twin pregnancies with one CMH and co-existing fetus are extremely rare (2) but as the incidence of multiple gestations increase with ARTs it is seen more common and reported as several case reports in the literature (5,6). As the data is poor, the management of these pregnancies remains as a debate. Nearly 70% of these pregnancies are diagnosed in the first trimester (7); but abnormal placentation, hematoma or existence of a placental tumor can interfere with the diagnosis. On the ultrasound; it is important to define the type (complete or partial) of molar pregnancy, because partial molar pregnancies are usually with multiple congenital anomalies. On the other hand, complete molar pregnancies have a higher incidence of maternal complications. The most helpful diagnostic approaches are ultrasound, chorionic villus sampling or AS. The most important challenge in these pregnancies is the management of the case. Most of the cases prefer the termination of the pregnancy because of the possible maternal risks. On the other hand, AS may constitute an option for the patient; if the

caryotype is normal with absence of a congenital abnormality, the pregnancy can be followed up after the informed consents are taken from the parents for the possible maternal and fetal risks. The biggest concern for the decision of on-going pregnancy is the risk of persistant trophoblastic disease. The risk of gestational trophoblastic neoplasia (GTN) is more (56% vs 20%) in twin pregnancies with mol hydtiform than singleton pregnancies (8). Moreover, the risk of GTN is more (68.4% vs 28.6%) after the co-existing fetus gains the viability (9). In the largest case series by Sebire et al. live birth rate was 27% and the incidence of GTN was 19% in the on-going pregnancies (10). On the other hand, persistent GTD incidence was 50% in another study (11). In the research by Stellar et al, the authors reported the occurrence of persistent GTD in 12 of 22 (52%) twin pregnancies of mol hydatiform and co-existing fetus. One of these patients had vaginal metastasis and eight of these patients (66%) needed combined chemo-therapeutic regiments (12). In the diagnosis of molar pregnancies, ßhcg level is one of the most helpful laboratory measurements. But sometimes, the levels of ßhcg may not be elevated as much as expected. In our case, a similar situation, a falsely low level of ßhcg, which is called as "Hook Effect", was encountered (13). This effect is caused by incomplete antigen-antibody complex formation; but can easily be solved by dilution of the blood sample. Thus, in suspicion of molar pregnancies, the laboratory should be informed about this and the measurement of diluted ßhcg level should be taken into account. The benefits of AS are the diagnosis of a triploid fetus and providing the opportunity of termination. In our case, the caryotype was normal but the parents decided for a termination because of possible maternal risks. In conclusion, a twin pregnancy with CMH and co-existing fetus is a very rare condition which has various maternal and fetal risks. In management of these cases, ultrasono graphy and cytogenetic analysis takes an important place. The decision for the follow up should be based on multiple factors like viability of the fetus and the preference of the family

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