Complete Hydatidiform Mole with a Coexistent Twin Live Fetus: Analysis of Two Cases

CANLI İKİZ FETUSUN EŞLİK ETTİĞİ KOMPLET MOL HİDATİDİFORM: İKİ OLGUNUN ANALİZİ

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Summary

In hydatidiform mole cases, coexistent live fetus is seen rarely. Twin pregnancies in which live fetus and placenta coexists with complete hydatidiform is an entity encountered even more rarely. These cases are significant in that they are diagnosed either late or mistakenly, fetal anomaly risks, maternal complications and persistent gestational trophoblastic tumor (PGTT) increase and the risk of metastatic development is present. The management of these cases still remains to be debated and larger series are required for better evaluation. In this analysis, we report two cases, in both of whom PGTT has developed and chemotherapy was warranted.

Key Words: Hydatidiform mole, Twin pregnancy

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Özet

Mol hidatidiform olgularında canlı fetusun birlikte bılunması nadir bir durumdur. Komplet mol hidatidiformla birlikte canlı fetus ve normal plasentanın bulunduğu ikiz gebelik olguları daha da nadir bir antitedir. Bu olgular, yanlış veya geç tanı almaları, fetal anomali riski olması, artmış maternal komplikasyonlar, artmış persistan gestasyonel trofoblastik tümör (PGTT) ve metastaz gelişme riski nedeniyle önem taşır. Bu olguların yönetimindeki tartışmalar halen devam etmektedir ve doğru değerlendirmeler için geniş serilere ihtiyaç vardır. Biz de bu sunuda ikisinde de PGTT gelişmiş ve kemoterapi ihtiyacı doğmuş iki olgunun analizini sunduk

Anahtar Kelimeler: Mol hidatidiform, İkiz gebelik

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Although the incidence varies with the geography, it is reported to be around one in thousand pregnancies worldwide and complete hydatidiform mole with a coexistent twin live fetus occurs extremely rarely, with an incidence of one in 22000 to 100000 pregnancies (1). This is different from partial molar pregnancy, because complete molar pregnancy does not involve any fetal tissue. Complete molar pregnancy with co-existent fetus is considered as twin pregnancy with the products of two different pregnancies (abnormal fetus with 46 chromosomes -23 maternal, 23 paternal- and coexistent complete molar pregnancy of completely paternal origin). The definitive diagnosis depends on histopathological criteria and cytogenetic examination. Since partial hydatidiform mole frequently occurs with twin fertilization of a single ovum and malformed fetus with triploid karyotype is formed, abnormal coexistent fetus tends to die in the first trimester while coexistent live fetus with complete hydatidiform mole in dizygotic twin pregnancy has a higher prospect of survival (2). Invasive interventions and the development of diagnostic techniques such as USG makes it possible to diagnose these cases in prenatal period, but the management of these pregnancies is still controversial. As we observed two such cases in such a short period as two months in our hospital, we decided to report these cases and evaluate them in view of recent literature.

Case I

25 years old. Gravida: 3, Parity: 1, Abortion: 1. Patient was 17 weeks pregnant and presented with spotting vaginal bleeding continuing for about one month. Blood pressure varied between 150/90 mmHg and 180/120 mmHg and pulse was 108/min. Cervix was not dilated in examination and uterus size was consistent with 19-20 weeks pregnancy. In the first ultrasound examination, live fetus at 16 weeks was detected and placenta was reported to be far from cervical os, but amniotic index was 25mm and fetal anomaly screening could not be performed optimally due to oligohydramnios. Patient was admitted with the diagnosis of threatened abortion. Vaginal bleeding and high blood pressure continued and in the analysis of spot urine, proteinurea was found to be 100mg/ml, Hb: 10 gr/dl., SGOT:65, SGPT:72. In the second USG carried out

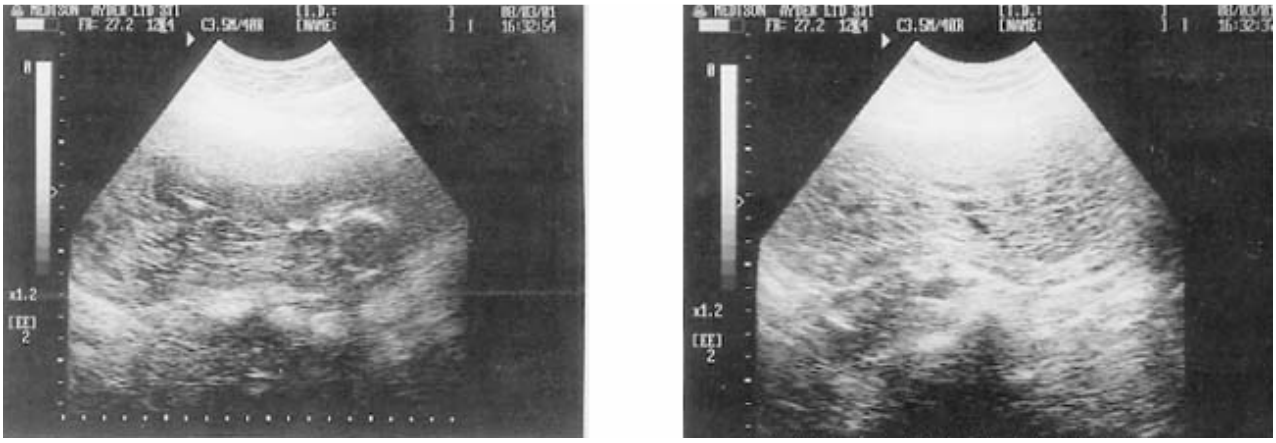

Figure 1, 2. Transverse sonogram obtained at 17 weeks' gestation through head and thorax of fetus and molar pregnancy.

three days later, one live fetus of 16 weeks pregnancy was detected in uterine cavity as well as a mass at 98X34 mm. size, yielding the image of multiple cystic spaces (Figure 1, 2) and due to oligohydramnios fetal anomaly could not be ruled out. This suggested either partial hydatidiform mole or complete hydatidiform mole with a coexistent twin live fetus. ß-hCG level was found to be 148164 IU/ml, freeT3: 11.18pg/ml (↑), freeT4:4.09ng/dl (↑) and TSH: 0.053 IU/lt (↓). Given these findings, patient was diagnosed with hydatidiform mole pregnancy with coexistent twin live fetus and resulting hyperthyroidy and pre-eclampsia. After the family was illuminated about maternal risks, we decided to terminate pregnancy with the approval of the family. In upper abdominal USG and chest radiography, no finding consistent with metastasis was observed. After induction, following the emptying of vesicular material through vagina, male fetus without gross anomaly and its placenta was aborted. The material remaining in the uterine cavity was emptied with sharp curettage. Fetus karyotype was normal, 46,XY. In the histopathological examination of vesicular material, complete hydatidiform mole was diagnosed with moderate trophoblastic hyperplasia in avascular hydropic villi. Four days after the evacuation, blood pressure values and liver function tests were within normal ranges and ß-hCG was 12436 mIU/ml. In the follow-up period, when ß-hCG value rose from 813 mIU/ml at the second week to 2198 mIU/ml at the fourth week, persistent gestational trophoblastic disease was considered and in order to screen for metastasis, liver function tests, chest radiography, pelvic-abdominal USG and cranial CT was carried out, but no metastasis was observed. After four courses of methotrexate administration, ß-hCG values returned back to undetectable levels and remained so for 8 months of follow-up period.

Case II

The patient was 44 years old, Gravida: 7, Para: 5, Abortion: 1. The patient, who did not know the date of her last menstruation, presented to the emergency service with complaints of vaginal bleeding and inguinal pain. In physical examination, cervical os was 1 cm. dilated and uterus was at the 16-17th week of pregnancy. After the patient was hospitalized with the diagnosis of threatened abortion, in USG, one live fetus at 15 weeks of pregnancy and a mass at the size of 137x75 mm. with the appearance of 'snow storm' pathognomonic of hydatidiform mole. Theca lutein cyst was found in neither case. Blood pressure and pulse were normal, Hb was found to be 9.0 gr/dl., liver function tests were normal and ß-hCG was 391792 mIU/ml. But, on the first day after hospitalization, a fetus at 110 gr. was aborted with vesicular material and placenta at normal appearance. Material remaining in the cavity was drained with sharp curettage. Fetus was morphologically and placenta histopathologically normal and vesicular structures were between 2mm-2cm and hydropic villi contained trophoblastic proliferation, but not fetal tissues. Seven days after evacuation, ß-hCG value was 20087 mIU/ml. Because of the increase starting from third week on-7809 and 14000 mIU/ml respectively- persistent gestational trophoblastic tumor was considered. In order to detect metastases, chest radiography, liver function tests, cranial CT, abdominal-pelvic USG was carried out, but no metastasis was found. After administration of six courses of methotrexate, ß-hCG levels fell to undetectable levels and remained so in six months of follow-up.

Discussion

Clinicians and families find it difficult to reach a decision on whether to terminate the pregnancy in multiple

pregnancies in complete hydatidiform mole cases with a coexistent twin live fetus. Although these cases are reported to increase with common employment of ovulation induction, they also occur after the application of assisted reproductive techniques (3,4). The most frequent complaints are vaginal bleeding and they are usually diagnosed as threatened abortion. Due to the presence of fetal heart motion, diagnosis and treatment is delayed. Although there are cases diagnosed as late as 38th week of pregnancy, mean pregnancy duration at diagnosis is usually 20,1 week and ß-hCG level is mean 839563 mIU/ml. USG in these cases is diagnostic at 68% of the cases whilst abnormal placental appearances may be misinterpreted as hematoma, placental tumor or marginal placenta previa and in a study, theca lutein cyst were found in six out of twenty two cases (1). The main problems in the management of these cases are the risk of fetal anomaly, malignant trophoblastic changes and severe maternal complications such as hyperemesis gravidarum, hemorrhage, thyrotoxicosis, early pre-eclampsia and preterm delivery. Although controversies continue on this issue, the criteria for the continuation of pregnancy are lack of massive bleeding, lack or control of pre-eclampsia, satisfactory development of fetus and lack of invasive trophoblastic disease provided that fetus has normal karyotype and family desires the pregnancy to be continued. The majority of these pregnancies are terminated after prenatal diagnosis, as in our cases since the risk of the development of persistent gestational trophoblastic disease is as high as 19.4% in addition to pregnancy induced hypertension, premature birth, or hemorrhage. (5). However, there are also cases followed with conservative approach and live fetus is delivered or cases terminated due to the development of metastasis during expectant approach (6-9).

The incidence of PGTT (the plateau level or increase in ß-hCG level at least for three consecutive weeks) is 20% in complete hydatidiform cases and 4% in partial hydatidiform cases (10). In another study, PGTT was found to be 14% in single complete hydatidiform mole cases, with no metastatic development. While in all cases of hydatidiform mole with a coexistent live fetus the incidence of PGTT was %30.6, in complete hydatidiform cases with a coexistent live fetus this rate was found to be 50% (9/18) and 55% (12/22) and lung metastasis have developed in 66% and 42% of these cases respectively, but following chemotherapy they have gone into remission (1,5). In a case in the literature, vaginal metastasis has been detected apart from lung metastasis. In both of our cases, PGTT has developed but without metastasis. It is not certain if the increased risk of persistent PGTT is because of a delay in diagnosis or treatment or of the greater aggressiveness of multiple pregnancy with complete hydatidiform mole and a coexisting fetus (1). The incidence of maternal complications is higher in cases developing PGTT than those who do not do so.

It is our conclusion that, management should be adjusted to individual patient, the decision must be based on factors such as fetal viability, gestational age, desire to continue pregnancy and clinical condition of the case and in cases with maternal complications, pregnancy must be terminated (11) and in those conservative approach is preferred, the karyotype of the fetus must be determined and whether it is euploid or not should be ascertained. The ß-hCG values of the patients should be monitorized whether expectant approach or termination is preferred and screening for metastasis should be carried out in all cases.

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