Hysterectomy in a malignant molar pregnancy and a healthy baby

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Summary

Hydatidiform mole is a degenerative chorion disturbance of unknown etiology. It is characterized by a permanent whitish enlargement of the chorionic villi with a grape-like appearance and poor vascularization. The symptoms of a molar pregnancy are nausea and vomiting which are present in more than one-third of the patients. Other symptoms include an enlarged uterus and vaginal bleeding. Our patient was a 19-year-old primipara with a twin pregnancy where one embryo was molar. Diagnosis was confirmed by beta-hCG levels.

Key words: Molar pregnancy; Hysterectomy.

Introduction

Hydatidiform mole is a degenerative chorion disturbance of unknown etiology and is a complication in one of 1,500 pregnancies in the USA, almost always during the first 18 weeks of gestation [4]. It is characterized by a permanent whitish enlargement of the villi with grape-like appearance and poor vascularization. Typical symptoms of molar pregnancy are nausea and vomiting which are present in more than one-third of the patients. Other common symptoms include an enlarged uterus and vaginal bleeding [4]. Uterine hemorrhage, present in almost all cases, starts between the 6th and 8th week of gestation and points to a possible threatening miscarriage [1-4]. In at least 40% of the cases the uterus is larger than in normal pregnancies [1]. This is a consequence of the large volume of villi which are vesicularly changed. The lower part of the uterus is thinner. In approximately 15% of the cases, there is bilateral cystic enlargement of the ovaries because of the stimulation by gonadotrophins. Preeclampsia is common in the second trimester. An intact or collapsed vesicle of the mole may be ejected vaginally by hemorrhage because there is no amniotic membrane to prevent ejection. Diagnosis can be established by a 24-hour urine sample and beta-hCG values over 400,000-500,000 units, presenting the highest level in a normal pregnancy at ten weeks of gestation [8-10]. In proliferic trophoblastic disease hCG titre totals more than 1 to 2 million international units over 24 hours. The level of 17-ketosteroid in the urine is often more than doubled compared to the normal level during pregnancy. In some cases hormone production (equal to the thyroid hormone), based on T3 and T4 serum determination, may produce toxic levels because production is similar to TSH by trophoblasts. However, there is no clinical evidence of toxic stroma in these cases [3, 5, 7].

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Case Report

A case of a 19-year-old patient in the 20th week of gestation with no previous deliveries or miscarriages is presented. She complained of bleeding and was examined by ultrasound. Ultrasound findings pointed out a twin pregnancy, where one was molar while the echo anatomy of the other embryo and placenta was normal. Diagnosis was confirmed by beta-hCG levels. With regular observation of this hormone, a very high level and minimal increase was confirmed, which did not endanger the patient or healthy fetus. Lung radiography did not find any trophoblastic disease metastases.

Frequent sonography examinations found molar tissue increasing in the form of tumefaction, and because biometry referred to an older pregnancy (1-2 weeks) for those referent values, amniocentesis was performed in the 33rd week of gestation to establish the age of the healthy fetus. Lecitin and sphingomyelin enzyme level was 1.5:1. In the 34th week of gestation, evident uterine contractions began which demanded tocolytic therapy. Even with maximal intravenous tocolysis, the mother's contractions could not be stopped so an urgent cesarean section was performed. A female with a body weight of 2,000 g and a length of 42 cm was born with little vitality (Apgar score 3 in the first and 4 in the fifth minute of life) thus the neonate needed intense pediatric care. As clinical signs of invasion of the described mole within the uterine tissue were present, surgery needed to be expanded. Histopathology analysis was performed *ex tempore* and indicated an invasive mole. Surgery included hysterectomy with preservation of the ovaries. Both ovaries were analyzed but samples did not show any alterations in ovarian tissue. All the other subsequent tests were unremarkable. Within two weeks there was a decrease in beta-hCG levels. Moreover, X-rays did not show any visible pathological changes.

Discussion

The motive for maintaining the pregnancy was the normal morphological and chromosome condition of the fetus and that the pregnancy was desired (azoospermia of the husband). The mother's condition was under close clinical observation. Beta-hCG findings indicated that

molar tumefaction was not malignant during the pregnancy because the beta-hCG levels, although above normal, were stable.

From 1938 to 1976, 16 cases of coexisting molar and normal pregnancies that ended successfully were published. Only in one case was the diagnosis established during pregnancy. The pregnancies were ended between the 31st and 40th week of gestation. Most newborns were female and hemangiomas were discovered in only two [10]. In more recent publications, sporadic cases with more detail have been described due to modern methods of diagnosis. In two independent cases [1, 2], joined normal and molar pregnancies occurred after ovulation stimulation and embryo transfer (IVF). Both patients had metastasis and hyperthyroidism in the 32nd week of gestation. An unwanted outcome of such diagnosis was noted by Harvard experts [7] who had had eight similar cases in the period from 1973 to 1994. Intensive chemotherapy was applied in five cases and in three cases multiple chemotherapies were necessary. Canadian authors [9] reported four cases of which three patients had unwanted outcomes of pregnancies and diseases that demanded chemotherapy. Only in one of the cases was the pregnancy ended in the 38th week of gestation. Horn *et al.* [3] had a successful outcome of a combined molar and normal pregnancy with the birth of a healthy female newborn and good condition of the mother. Changchien *et al.* [4] urgently ended a pregnancy in the 23rd week of gestation. The newborn was male, 700 g, with an Apgar score of 3 in the first minute and 7 in the fifth minute. The baby survived and the patient had chemotherapy for the first year after delivery. Hoshi et al. [6], after ovulation stimulation, described triplets that developed along side a molar pregnancy. Two babies were born alive and one was stillborn. Authors from Nigeria [5] report a very interesting case of a 56-year-old woman who became pregnant after five years of amenorrhea. A gemellar pregnancy – molar and normal – was diagnosed which ended in miscarriage in the 20th week of gestation; the fetus had vital signs. After curettage, the general condition of the patient was satisfactory.

Regular examination of patients is very important for decision making about further treatment of both mother and fetus. All methods of diagnosis, especially beta-hCG levels and sonography, should be used. The possibility of Hook Syndrome appearance (antibody real value mimicry) should be emphasized which requires an additional dilution technique [8]. Histopathology and genetic analyses of placental tissue demonstrate that DNA samples are of paternal origin in almost all cases [10].

Conclusion

This is a rare case of a successful pregnancy outcome in the case of total hydatidiform mole of one twin and normal development of the other twin. The condition of the patient was preserved and a chromosomally and morphologically healthy and vital fetus was born. The mother's healthy condition enabled us to follow the pregnancy to the end but clinically evident invasion and confirmed ex tempore analysis pointed to the necessity of hysterectomy (malignancy but not spread to the ovaries). Even though the combination of a molar and normal pregnancy is rare, it is necessary to keep in mind even those rare cases in establishing the differential diagnosis.

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