

Hydatidiform Mole With Coexisting Twin After In-Vitro Fertilization

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Citation

H Kafali, B Cengiz. *Hydatidiform Mole With Coexisting Twin After In-Vitro Fertilization*. The Internet Journal of Gynecology and Obstetrics. 2002 Volume 2 Number 2.

Abstract

We report the case of 38-year-old women with a hydatidiform mole and coexisting fetus (HMCF). HMCF is a rare entity. Two different mechanism of the formation of HMCF are possible: a complete mole coexisting with live fetus and partial mole with an abnormal triploid fetus. In our case, risks of possible fetal malformation and subsequent malignant transformation of the molar pregnancy were explained to parents at the time of diagnosis. They chose to allow the pregnancy to continue.

INTRODUCTION

Hydatidiform mole with coexisting fetus (HMCF) is a rare entity occurring in 0.005-0.01% of all pregnancies (1). With ready availability of antenatal ultrasound, it is likely that number of HMCF diagnosed antenatally will increase, raising clinical dilemmas, particularly when the pregnancy has been long awaited. When the diagnosis has been made antenatally, most patients opted for termination of pregnancy to avoid or reduce the risks of malignant transformation and severe maternal complications such as pre-eclampsia and antepartum hemorrhage. However, it is not so easy for both obstetrician and parents to decide between immediate termination or continuation of pregnancy when HMCF occurs in a much wanted pregnancy such as a woman achieving conception for the first time in their later life or after many attempts of assisted conception. In this report; a case of HMCF occurring after in-vitro fertilization (IVF) is discussed with respect to prenatal diagnosis, basis for continuing the pregnancy, prenatal care, and postpartum management.

CASE REPORT

A 38-year-old women, gravida 0, para 0, was referred for the problem of primary infertility and for assisted conception. She did not conceive despite three courses of clomiphene therapy and six course of gonadotropin therapy. Follicular growth was stimulated with recombinant FSH after achieving of ovarian suppression with gonadotropin releasing (GnRH) analogue. Following the egg collection under ultrasound guidance, oocytes were inseminated in a concentration of 100 000 motile spermatozoa per oocyte.

Normal fertilization of six oocytes was noted 24 hours after egg collection. Three, 4-cell embryos were transferred in to the uterus. Five weeks after embryo transfer, two gestational sacs; one of which poorly defined and the other showing positive fetal pole were identified by transvaginal ultrasound. From this point onwards, the patient was lost to follow-up until 16 weeks of gestation when she experienced the first episode of fresh vaginal bleeding.

Ultrasonographic evaluation revealed a live fetus consistent with 16 weeks of gestation. There was no evidence of growth retardation or fetal anomaly. However, a well-defined and separate multiple cystic mass was found in the low uterine segment and HMCF was suspected (Figure 1,2,3). Prenatal screening for Down's syndrome was performed by triple test which revealed elevated β -HCG level and normal range of AFP and Estriol. Even though there was no significant abnormality noted in serial examination except high β -HCG, amniocentesis was performed regarding to the possible risk of fetal malformation. It revealed normal 46 XY karyotype.

Figure 1

Figure 1, 2: Transabdominal ultrasound image of uterus illustrating the vesicular tissue (M) in upper part of the sac and the normal placenta (P) in the lower part of sac.

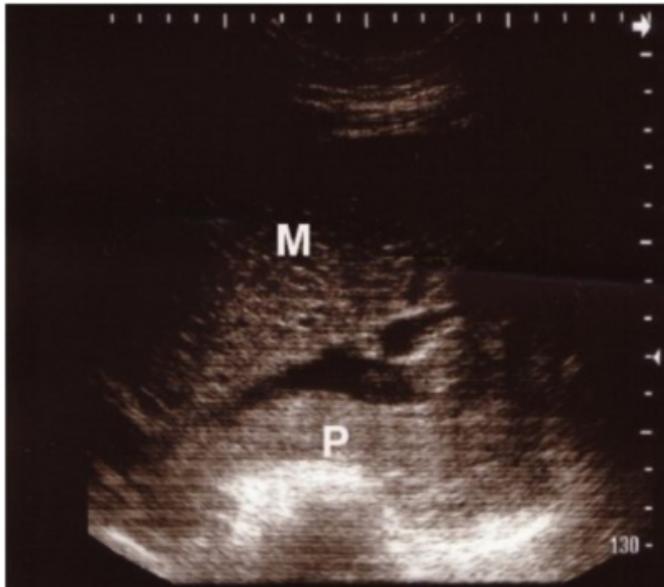


Figure 2

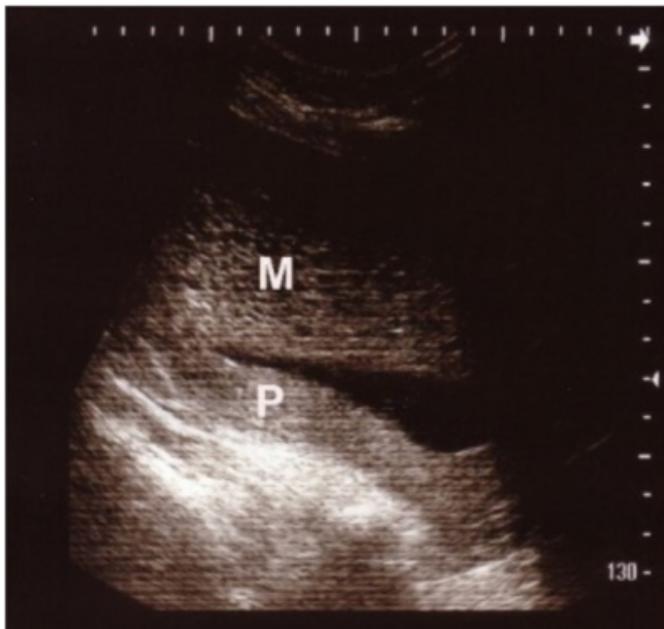
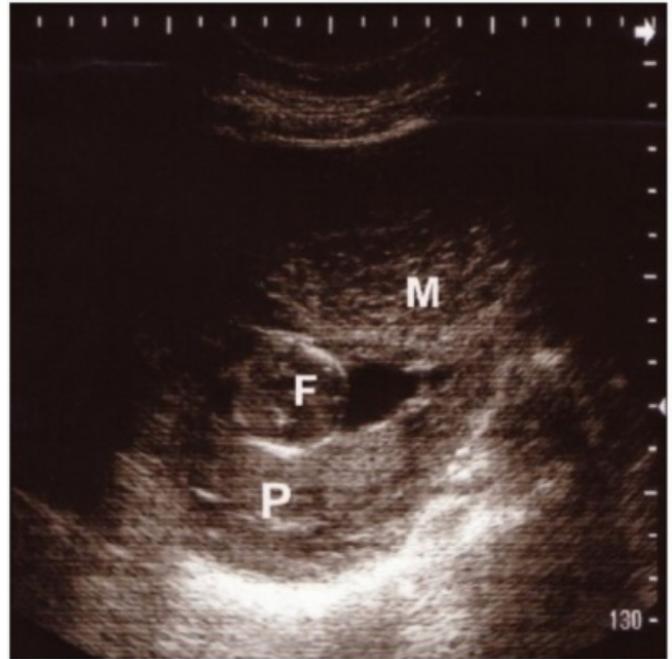


Figure 3

Figure 3: Transabdominal ultrasound image of the mole (M), fetus (F), and normal placenta (P).



During the following 10 weeks patient experienced multiple episodes of antenatal hemorrhage which resolved spontaneously. At 28 weeks of gestation she had a Cesarean section delivery for a reduction in fetal growth velocity and fetal heart rate deceleration. A live male infant (760g) and normal placenta (180g) were delivered. 1050 g of cystic, vesicular mass was removed manually. Histological examination confirmed the clinical impression of one normal placenta and also a complete hydatidiform mole. During the immediate postpartum period, the patient had an uneventful course. Unfortunately, the baby developed 'wet lung' and died at postpartum 7th day. Serum β -HCG titre showed a normal regression curve within a month postpartum. However during the following month a rising tendency was observed.

Ultrasound examination showed a 26x28 mm mass in uterine cavity (Figure 4) but chest and abdominal computer tomography revealed normal finding. The patient received four courses of methotrexate combined with folinic acid after which serum β -HCG fell to a normal level. Patient is now well, using oral contraceptive and has regular follow-up for β -HCG.

Figure 4

Figure 4: Ultrasonographic examination after evacuation revealed an intrauterine lesion suggesting post molar disease.



DISCUSSION

It is difficult to explain how a HMCF developed during IVF despite replacement of apparently normal 4-cell embryos. However, some of clinical characters such as advanced maternal/paternal age, poor oocyte quality might predispose to this problem. Development of complete mole needs either fertilization of empty ovum by diploid sperm or duplication of haploid sperm in empty ovum. We speculated that empty ovum taking place in this process may be the large second polar body. Second polar body is small and extruded at the time of fertilization, but if retained, it can appear as large as normal pronucleus. The fertilizing spermatozoon, if diploid, could have lead to appropriate cleavage on day 2, giving a normal appearance to the embryo. It is well-known that spermatozoon may remain diploid especially as paternal age increase. Alternatively the two pronuclei seen may be as a result of dispermy.

The natural history of pregnancy affected by HMCF is still unclear. Complications particularly malignancy are poorly defined. Although HMCF detected antenatally is terminated in most cases, optimal management protocol remains controversial. Some authors recommended immediate termination of pregnancy when the HMCF has been diagnosed because HMCF carries a higher risk of severe maternal complications and post molar trophoblastic disease^(1,2). However, others has suggested that, in the presence of a normal karyotype demonstrated by amniocentesis, stable clinical course, pregnancy may be continued pending fetal maturation⁽³⁾.

Two different mechanism of the formation of HMCF are possible; a complete mole coexisting with live fetus and partial mole with an abnormal triploid fetus. Although recent advances in ultrasonography make it possible to diagnose a twin molar pregnancy from the end of first trimester, differentiation of HMCF from partial mole based on morphological observation is sometimes difficult and inaccurate, particularly in cases where a coexisting twin is aborted at an early stage of gestation. However in our case, ultrasound scan clearly revealed a normal placenta and sharply defined molar tissue.

It is important to distinguish between complete and partial mole when a fetus coexists because these clinical entities are different with different perinatal outcomes and complications of pregnancy. Generally, partial mole is mostly associated with triploid fetuses that tend to die before the end of the first trimester and surviving fetuses after mid pregnancy are rarely encountered. On the other hand the fetus coexisting with complete mole is usually associated with normal karyotype and has a chance of survival. It was reported that before 28 week of gestation, the chances of survival are minimal and the chance for continuation of pregnancy beyond this point is 60 %. Of pregnancies which continue beyond the 28th week, a surviving child may be expected in 70% of pregnancies⁽⁴⁾. In this present case, risks of possible fetal malformation and subsequent malignant transformation of the molar pregnancy were explained to parents at the time of diagnosis. They chose to allow the pregnancy to continue. It is prudent that the decision to allow such a pregnancy to continue should be taken with the couple. If fetus is chromosomally normal and clinical course of pregnancy is benign, an expectant management until infant viability must be considered⁽⁵⁾. Such a management may be encouraged in those patients who had fertility therapy. At the same time the malignant potential of disease should be taken in consideration as it had been reported that MHCF had a more aggressive post evacuation behavior with a risk of post molar disease higher than a singleton mole⁽⁵⁾.

It is unclear whether the greater risk of post molar disease is associated with a more aggressive behavior of the molar tissue or with delayed delivery. Recent reports had pointed out that an advancement of gestational age did not appear to increase the risk of developing a post molar disease⁽³⁾. No reports had demonstrated that prolonging pregnancy to term would increase the incidence of invasive mole or choriocarcinoma. Some reports showed that chemotherapy was ultimately required even when termination of pregnancy

was performed during early gestation of pregnancy (6). In our case, post molar disease was controlled with fairly non-aggressive protocol but in the light of current literature it may be better to initiate more aggressive chemotherapy immediately after termination.

It is our opinion that patients with HMCF may be allowed to continue pregnancy, provided that the fetal karyotype is normal and maternal complications can be controlled. However, it is necessary to have detail counseling for the couples that include complete discussion of maternal and fetal risk, particularly the possible requirement of chemotherapy or even hysterectomy .

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