Partial Hydatidiform Mole in a Quintuplet Pregnancy Following Intrauterine Sperm Injection: Case Report and Review of the Literature

Elif Agacayak1, Senem Yaman Tunc1, Ali Ozler1, Abdulkadir Turgut1, M.Siddik Evsen1, Neval Yaman Goruk1

1Department of Gynecology and Obstetrics, Dicle University School of Medicine, 21280 Diyarbakir Turkey

Abstract
Partial mole occurs after intrauterine insemination and healthy fetuses are rare in the literature. Partial hydatidiform mole occurred in a quintuplets pregnancy following an intrauterine sperm injection procedure performed for male factor infertility. Both gestational sacs were broken on the eighth week of gestation. Diamniotic dichorionic other twin pregnancies with a gestational sacs presented with two heartbeats on the 17th week of gestation, and fetuses developed normally. The gestational sac evolved from one of the partial molar pregnancies. Massive vaginal bleeding and excessive growth of the uterus necessitated termination of the pregnancy via therapeutic abortion on the 17th gestational week. Twins enclosed with their cords to a normal placental mass were consigned, accompanied by an abnormal placental mass which was a partial hydatidiform mole. Histological inspection of the curettage specimen was reported as a partial mole. The patient was followed-up for one year after abortion with no complications. Our aim in this case, the treatment of multiple pregnancies occurring vaginal bleeding should be considered necessarily be partial mole, and complete mole and early diagnosis. Late diagnosis; both psychological and physical trauma to the mother.

Introduction
Gestational trophoblastic disease is a group fascinating of pregnancy disorders characterized by abnormal proliferation of trophoblasts, ranging from benign to malignant. Because the disease is uncommon, there is a need to formulate treatment methods with the assistance of clinical information. Partial hydatidiform mole is a milder version of the complete form, including both normal and cystic villi with focal trophoblastic hyperplasia; there is fetal development or indication of previous fetal presence. Partial moles are property triploid with 46 chromosomes coming from the father. The pathogenesis is accounted by dispermic fertilization of an ovum or monospermic fertilization with duplication of the paternal haploid chromosome. Partial moles are triploid but a minority are normal diploid; regardless of the karyotype; however, partial moles do not have a high propensity for malignancy (1). Exceptionally, some of the contradictions between cytogenetic and histological classifications of hydatidiform mole may be explained by a diploid biparental partial mole, which seems to constitute a separate subgroup within hydatidiform mole (2). This study reports a case of partial hydatidiform mol following intrauterine inseminasyon (IUI). IUI was used fertilization method because there was male factor infertility due to oligoastenospermia. Possible predisposing...
factor, for partial hydatidiform after IUI are also discussed.

Case
A 27-year-old woman and her husband, who had five years of secondary infertility, were referred for treatment with intrauterine insemination (IUI). Semen analysis was anormal. The patient was treated as an outpatient for intrauterine insemination and, the patient was admitted to our clinic with a rotary units a week. In obstetrical ultrasonography; three gestational sacs were observed. One week after examination revealed five gestational sacs. Three of these were monitored regularly and observed to was one of the sac irregular. One week later, the patient was admitted to hospital with excessive vaginal bleeding. The ultrasound examination of the fetal sacs showed that two of these were not completely broken, however the third pouch was broken. The patient was hospitalized. One unit of red blood cell transfusion was administered. One week later examination of the patient after discharge indicated two healthy fetuses; the remaining three sacs were completely resorbed and subchorionic hematoma was observed to occur. Taking into consideration all of the risks involved, the family decided to continue the pregnancy. During the 17th gestational week the patient was diagnosed with molar pregnancy, with excessive uterine growth and expansion, and the mole was viewed through ultrasound (Figure 1). After excessive vaginal bleeding, and the parents decided to terminate the pregnancy. Two fetuses (170 g male and 180 g female) were delivered attached with their cords to a normal placental mass (Figure 2a). There was another placental mass of about 600 cm³ containing hydropic vesicules. Pathological inspection display partial trophoblastic proliferation and hydropic degeneration, so the diagnosis was partial hydatidiform mole in a quintuplet pregnancy (Figure 2b). Examination after one week showed b-HCG level was 10,600 U/L. The one year follow-up was uneventful.
Discussion
Molar tissue coexistent with viable twin fetuses and dead twin fetuses, as observed in the second pregnancy of our case, can be a complete mole in a triplet pregnancy, a partial mole in a quintuplets pregnancy, or a partial mole in a triplet pregnancy. Our diagnosis was the second option according
to the macroscopic and microscopic findings. Unhappily, we were not able to perform the chromosomal analyses of the abnormal and normal appearing placental tissues and the fetuses under that emergency situation. Women with complete or partial hydatidiform moles have improved risk of molar gestation in the subsequent pregnancies (3). The repetition are generally of the same histological type (4,5).

High incidence of triploid embryos consequent IVF has been recorded in a woman with recurrent gestational trophoblastic disease, representing a possible oocyte defect (6). Intracytoplasmic sperm injection (ICSI) might overcome dispermic fertilization but is not a warranty for the prevention of triploidy (7). There is a few case repord of a partial molar pregnancy following ICSI and assisted zona hatching in the literature (8). The molar tissue was diploid and fetus has in that case. Its pathogenesis was accounted by the trophoblastic transformation of the embryonic inner cell mass at a stage just prior to the laying down of the endoderm. Thus, the molar vesicles were shaped as well as the primitive mesoderm in the villous core. Another feature is that fetal red blood cells can sometimes be observed in the fetal blood vessels. Trophoblastic proliferation is less than in a complete mole. As in our case, sometimes these can be seen as macroscopic vesicles. However, microscopic vesicles are often seen as fetuses reach maturation. This might also be feasible for our case. In reality, the investigation of two pronuclei for the confirmation of fertilization in IVF procedures, rules out the presence of triploidy at that state. Preimplantation genetic diagnosis (PGD) might approve diploidy at a later stage, and preserve against triploid partial moles which result from mechanisms other than dispermic fertilization (9). However, these procedures likely cannot fully prevent the development of partial hydatidiform mole, especially the ones with a diploid karyotype.

The diagnosis of molar pregnancy could not be diagnosed ultrasonographically in the first few weeks of pregnancy of our patient. In our patient management and diagnosis of molar pregnancy week by ultrasonography considered eleventh pregnancy. The most common manifestation in partial moles is the entity of translucent space within the placenta (10). Serum-hCG titers might be useful but not absolutely diagnostic (11). In doubted cases, histological confirmation is possible from the choriocarcinomatous samples. Molar pregnancy follow-up may be tested as long as there are no complications such as preeclampsia, hyperthyroidism, vaginal bleeding or metastatic disease (12). In our patient preeclampsia, hyperthyroidism and metastatic disease did not develop. As such, the optimal administration is contention and pregnancy termination might be another choice (13). Our patient presented with excessive vaginal bleeding and therefore there was an immediate termination of pregnancy after obtaining approval from the family. One week following the b-HCG level was at 10,200 U/L; however, two weeks later this declined to 1,600 U/L. There was no problem at the patient’s one-year follow-up.

References
2. Vejerslev LO, Sunde L, Hansen BF, Larsen JK, Christensen IJ, Larsen G. Hydatidiform mole and fetus with normal