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PARTIAL HYDATIDIFORM MOLE IN A TRIPLET PREGNANCY FOLLOWING INTRACYTOPLASMIC SPERM INJECTION: CASE REPORT AND REVİEW OF THE LITERATURE

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ABSTRACT

Partial hydatidiform mole occurred in a triplet pregnancy following an intracytoplasmic sperm injection procedure performed for tuboperitoneal and male factor infertility. Massive vaginal bleeding necessitated termination of the pregnancy via hysterotomy at the 17th gestational week. Twins attached with their cords to a normal placental mass were delivered, accompanied by an abnormal placental mass which was a partial hydatidiform mole. The patient's next conception occurred spontaneously but was anembryonic. Histological examination of the curettage specimen was reported as a partial mole.

Key words: *ICSI, multiple pregnancy, partial hydatidiform mole*

ÖZET

İntrasitoplazmik Sperm İnjeksiyonu Sonrası Oluşan Bir Üçüz Gebelikte Parsiyel Molar Gelişim: Olgu Sunumu ve Literatürün Gözden Geçirilmesi

Tuboperitoneal faktör ve erkek faktörüne bağlı infertilite nedeniyle uygulanan intrasitoplazmik sperm injeksiyonu sonrasında, olgumuzda parsiyel molar gelişim gösteren bir üçüz gebelik oluştu. Aşırı vajinal kanama dolayısıyla 17. haftada gebeliğin histerotomi ile sonlandırılması gerekti. İşlem sırasında normal bir plasental doku ve bununla bağlantılı olan ikiz fetusların yanı sıra, parsiyel mol olduğu tespit edilen anormal görünümlü bir plasental doku çıkarıldı. Daha sonra hasta spontan gebe kaldı, ancak gebelik anembriyonikti. Küretaj materyalinin histolojik incelemesi yine parsiyel mol olarak rapor edildi.

Anahtar kelimeler: *çoğul gebelik, intrasitoplazmik sperm injeksiyonu, parsiyel mol.*

INTRODUCTION

Partial hydatidiform mole is a milder version of the complete form, containing both normal and cystic villi with focal trophoblastic hyperplasia. There is a fetus or indication of previous fetal existence. Partial moles are characteristically triploid with 46 chromosomes coming from the father. The pathogenesis is explained by dispermic fertilization of an ovum or monospermic fertilization with duplication of the paternal haploid chromosome[1]. Exceptionally, there are also diploid cases with biparental contribution[2].

CASE

A 25-year-old woman and her husband, who had 4 years of primary infertility, were referred for treatment

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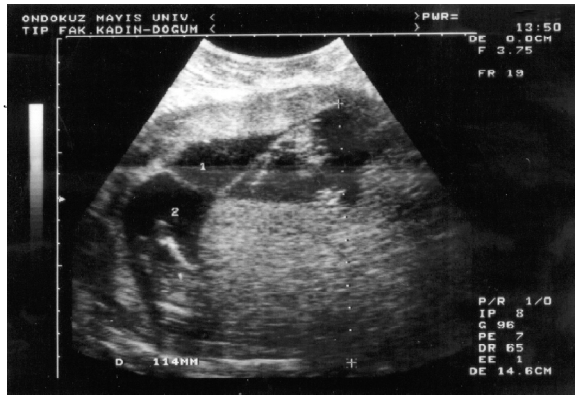
with in vitro fertilization (IVF). At laparoscopy, there were dense periadnexal adhesions and a right proximal tubal obstruction. Semen analysis was subnormal. In June, 2000, in the first controlled ovarian hyperstimulation cycle, six oocytes were retrieved, five of which were metaphase-II. Following intracytoplasmic sperm injection (ICSI) five oocytes fertilized. One grade 2 and two grade 3 embryos were replaced on day 3. Twelve days after the transfer, serum -hCG level was 289 IU/L, and increased to 1050 IU/L after 48 hours. Two weeks later, two gestational sacs with viable embryos and a third suspicious irregular sac were observed at ultrasonography. At the 7th gestational week a repeat ultrasound performed for vaginal bleeding revealed a viable twin pregnancy and a mixed-echoic mass presumed to be a subchorionic hematoma. This hematoma-like mass continued to grow (Figure 1) and the patient required erythrocyte transfusions. At the 17th gestational week, a massive episode of vaginal bleeding necessitated emergency hysterotomy. Two fetuses (100g male and 120g female) were delivered attached with their cords to a normal placental mass. There was another placental mass of about 500 cm3 containing hydropic vesicules. Pathological examination revealed partial trophoblastic proliferation and hydropic degeneration, so the diagnosis was partial hydatidiform mole in a triplet pregnancy. The patient's follow-up was uneventful. She spontaneously conceived after 2.5 years. At the 5th gestational week her serum -hCG level was 12200 IU/L, and an intrauterine gestational sac was visible. Two weeks later, the uterus was evacuated with an ultrasound diagnosis of anembryonic pregnancy. The material was about 8 cm3 and showed vesicular appearance. The histological diagnosis was once again partial hydatidiform mole.

Figure 1: *Transabdominal ultrasonographic appearance at the 17th*



DISCUSSION

Molar tissue coexistent with viable twin fetuses, as observed in the first pregnancy of our case, can be a complete mole in a triplet pregnancy, a partial mole in a twin pregnancy, or a partial mole in a triplet pregnancy. Our diagnosis was the third option according to the macroscopic and microscopic findings. Unfortunately, we weren't 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 increased risk of molar gestation in the following pregnancies[3]. The recurrences are usually of the same histological type[4,5]. This was the case in our patient as well. High incidence of triploid embryos following IVF has been reported in a woman with recurrent gestational trophoblastic disease, indicating a possible oocyte defect[6]. ICSI might overcome dispermic fertilization but is not a guarantee for the prevention of triploidy[7]. There is one case report of a partial molar pregnancy following ICSI and assisted zona hatching in the literature[8]. The molar tissue was diploid and without a fetus in that case. Its pathogenesis was explained by the trophoblastic transformation of the embryonic inner cell mass at a stage just prior to the laying down of the ectoderm. Thus, the the molar vesicles were formed as well as the primitive mesoderm in the villous core. This might also be possible for our case. In fact, the observation of two pronuclei for the confirmation of fertilization in IVF procedures, rules out the presence of triploidy at that stage. Preimplantation genetic diagnosis (PGD) might confirm diploidy at a later stage, and guard against triploid partial moles which result from mechanisms other than dispermic fertilization[9]. However, these procedures probably cannot fully prevent the development of partial hydatidiform mole, especially the ones with a diploid karyotype.

Molar gestation could not be diagnosed ultrasonographically in either pregnancies of our patient. The most common appearance in partial moles, is the presence of translucent areas within the placenta[10]. Serum-hCG titers might be helpful but not absolutely diagnostic[11]. In suspected cases, histological confirmation is possible from the chorionic villus samples. When the molar tissue coexists with chromosomally normal fetus / fetuses, as determined

from the amniotic fluid or the cord blood, conservative follow-up may be tried as long as there are no complications such as preeclampsia, hyperthyroidism, vaginal bleeding or metastatic disease[12]. However, the optimal management is controversial and pregnancy termination might be another choice[13].

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