

A Case of Partial Hydatidiform Mole Presenting with Ovarian Hyperstimulation and Early Preeclampsia

Ovaryan Hiperstimülasyon ve Erken Dönem Preeklampsi ile Prezente Olan Parsiyel Mol Hidatiform Olgusu

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ABSTRACT Missed or incomplete abortion occurs in cases of partial molar pregnancy, whereas fetal and maternal complications, including fetal anomaly, fetal anemia, and preeclampsia, generally occur during the 20th week of gestation in cases in which the fetus remains viable. To the best of our knowledge the literature does not include any reports of partial molar pregnancy complicated by both preeclampsia and ovarian hyperstimulation. As such, this case presentation aimed to describe a patient with partial molar pregnancy with early preeclampsia and ovarian hyperstimulation, as complications, and a live fetus that was diagnosed during the 20th gestational week.

Key Words: Pre-eclampsia; hydatidiform mole; ovarian hyperstimulation syndrome

ÖZET Parsiyel molar gebelikte eşlik eden embriyoda genellikle missed veya inkomplet abortus gelişirken, gebeliğin ilerleyen haftalarına ulaşan fetüslerde genellikle 20. gebelik haftasında fetal anormali, fetal anemi ve preeklampsi gibi fetal ve maternal komplikasyonlar ortaya çıkabilmektedir. Preeklampsi ve ovaryan hiperstimülasyon komplikasyonlarından her ikisi ile birlikte komplike olmuş parsiyel molar gebelik olgusu ise literatürde henüz bildirilmemiştir. Biz de gebeliğin 20. haftasında saptanmış, preeklampsi ve ovaryan hiperstimülasyon ile komplike olmuş ve ventrikülomegalisi olan canlı fetüsün eşlik ettiği parsiyel molar gebelik olgusunu sunmayı ve parsiyel molar gebeliğe eşlik edebilecek maternal ve fetal komplikasyonları tartışmayı amaçladık.

Anahtar Kelimeler: Pre-eklampsi; hidatiform mol; ovaryan hiperstimülasyon sendromu

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A rare maternal complication of partial molar pregnancy is ovarian hyperstimulation syndrome (OHSS). OHSS generally occurs due to ovulation induction and rarely due to increased β -human chorionadotrophin (β -hCG) without ovulation induction.¹

After the 20th gestational week the frequency of preeclampsia as a maternal complication of partial molar pregnancy is 41.9%, whereas the incidence of OHSS in cases of partial molar pregnancy is not known.² To the best of our knowledge based on a search of Türkiye Citation Index, PubMed, Medline, Embase, ScienceDirect, Cochrane and Google Scholar using the search terms molar pregnancy, hydatidiform mole, partial hydatidiform mole, pre-eclampsia, proteinuria, and ovarian hyperstimulation syndrome,

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the literature does not include any cases of partial mole complicated by preeclampsia and ovarian hyperstimulation. As such, the present case presentation aimed to describe a case of partial molar pregnancy with ovarian hyperstimulation and preeclampsia, as complications, and a live fetus in which ventriculomegaly was ultrasonographically observed during the second trimester.

CASE REPORT

A 28-year-old female with a history of Friedreich's ataxia and a healthy living child born from her second pregnancy was referred due to hypertension diagnosed at a secondary-care healthcare institution during the 20th gestational week. She did not have hypertension during her previous pregnancy or at any time when not pregnant. At presentation her general status was good and her vital findings were as follows: blood pressure: 160/100 mm Hg; pulse rate: 84 bpm; respiratory rate: 22 min⁻¹; body temperature: 36.5 °C. Physical examination showed that the fundus-pubis measurement was compatible with 20 weeks of gestation and there were no other pathological findings. Laboratory findings were as follows: leukocytes: $9.1 \times 10^3 \text{ mm}^{-3}$; hemoglobin: 10.1 g dL⁻¹; platelets: $193 \times 10^3 \text{ mm}^{-3}$; complete urinalysis protein: 300 mg dL⁻¹. Coagulation profile and biochemical parameters were within normal limits. β -hCG was >500,000 mIU mL⁻¹ and 24-h urine protein was 1.8 g.

Obstetrical ultrasonography (US), showed a fetus with cardiac activity compatible with 20 gestational weeks and severe bilateral ventriculomegaly (Figure 1). The placenta was observed in the anterior wall of the uterus and had an appearance compatible with partial mole (Figure 2). The right ovary was $10.5 \times 7.2 \times 7.8 \text{ cm}$ (313 cm^{-3}) and the left ovary was $9.8 \times 8.6 \times 6.4 \text{ cm}$ (285 cm^{-3}) in size. Both ovaries had a hyperstimulated appearance (Figure 3). Antihypertensive treatment was started because of severe preeclampsia and magnesium sulphate was administered for convulsion prophylaxis.

The patient and her partner were informed about the malformations observed in the fetus, and

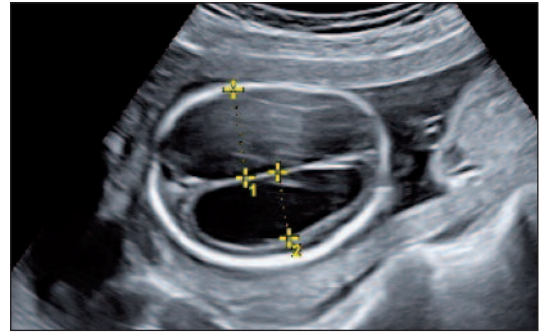


FIGURE 1: Bilateral ventriculomegaly via gray-scale ultrasonography.

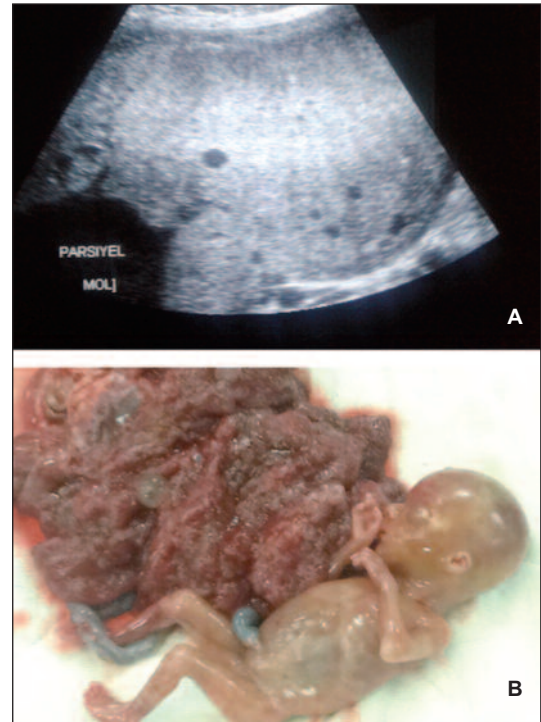


FIGURE 2: A. Gray-scale ultrasonographic view of the placenta compatible with partial mole; B. Gross pathological appearance of placenta indicating partial molar pregnancy.

the maternal and fetal complications associated with partial molar pregnancy. The option of terminating the pregnancy medically using misoprostol was offered to the patient, whose previous delivery was performed via cesarean section. The patient wanted to terminate the pregnancy, but she refused induction with misoprostol. Thus, anterior hysterotomy was performed. The female fetus, which weighed 275 g, had no pathological finding other than low-set ears, and the placenta was sent for pathological examination.

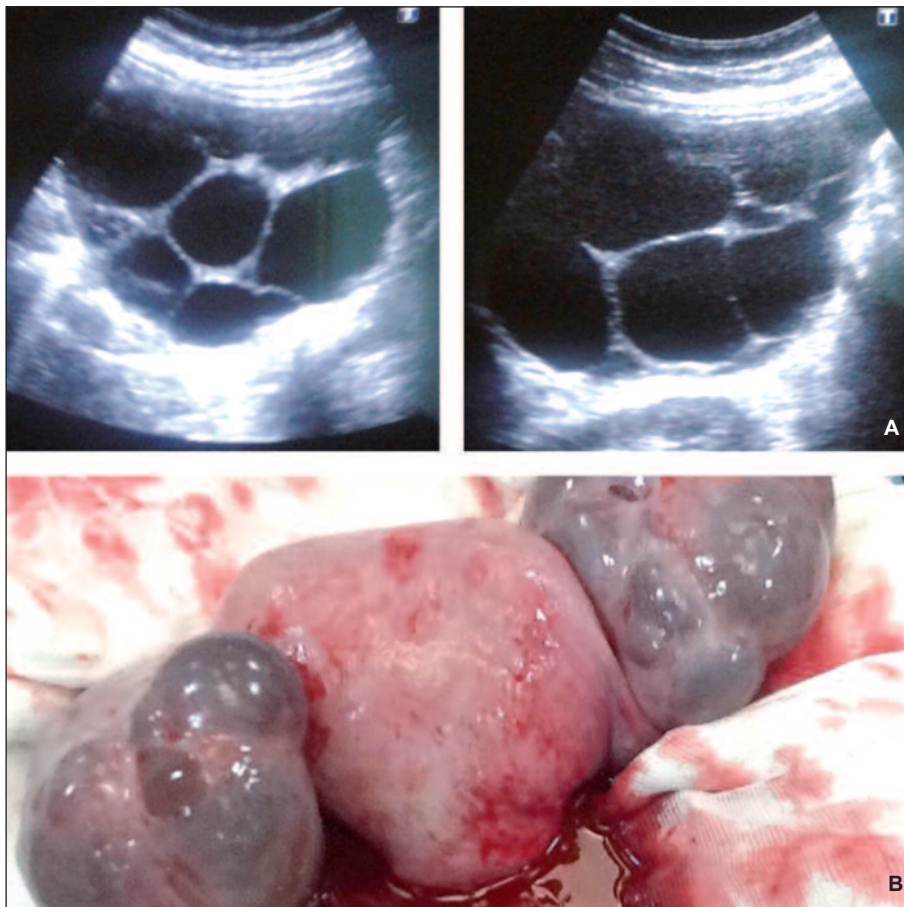


FIGURE 3: A. Gray-scale ultrasonographic view of the ovaries compatible with hyperstimulation. B. Surgical demonstration of hyperstimulated ovaries.

A skin biopsy specimen was obtained from the fetus for genetic analysis. Dilatation in the bilateral ventricles was noted in the fetus based on histopathological examination and the karyotype was 47 XXX (triploidy). Trophoblastic hyperplasia and large villus structures observed in the placenta were reported to be compatible with partial hydatidiform mole. Post-operative follow-up showed no surgical complications; the patient was discharged from hospital 3 d post surgery and was scheduled for weekly β -hCG monitoring.

DISCUSSION

Kirk et al. reported that US identifies 44% of all molar pregnancies, 95% of complete molar pregnancies, and 20% of partial molar pregnancies.³ Lindholm et al. reported that only 40% of partial mole cases are identified via US despite the use of high-resolution US technology.⁴ Partial mole was

not diagnosed in the presented patient during her regular follow-up visits prior to presenting to our tertiary healthcare institution upon referral due to hypertension during the 20th gestational week, at which time ultrasonographic findings indicated partial mole.

The association between partial molar pregnancy and proteinuric hypertension is rare. Preeclampsia varying in severity can accompany partial molar pregnancy. Prasannan et al. reported a partial molar pregnancy accompanied by preeclampsia and fetal growth retardation diagnosed during the 12th gestational week that was followed-up until the 19th gestational week.⁵ The presented case was diagnosed with severe preeclampsia due to hypertension (160/100 mmHg) and proteinuria during the 20th gestational week; therefore, antihypertensive treatment was started and although eclampsia

associated with a hydatidiform mole is extremely rare; magnesium sulphate was administered for convulsion prophylaxis.⁶

OHSS due to elevated β -hCG rarely is observed in molar pregnancies; because the β -hCG level is higher in cases of complete molar pregnancy OHSS is generally observed in patients with complete molar pregnancy.⁷ The present case represents a rare atypical presentation of partial molar pregnancy, as the β -hCG level was $>500,000$ mIU mL⁻¹ and bilateral ovarian hyperstimulation was observed. The most common karyotype accompanying partial molar pregnancy is triploidy. The most common fetal anomalies accompanying triploidy include syndactyly of the 3rd and 4th fin-

gers, ventriculomegaly, atrioventricular cardiac defects, and micrognathia.⁸ In the presented case triploidy (47, XXX) was noted via karyotype analysis of the fetus and bilateral ventriculomegaly observed via obstetrical US during the prenatal period was also noted during autopsy.

Partial molar pregnancy is difficult to diagnose using obstetrical US, especially in cases with a live fetus. Clinicians must consider partial molar pregnancy in patients with preeclampsia before the 20th gestational week that also have hyperstimulated ovaries, so that a diagnosis of partial molar pregnancy will not be missed, and timely administration of appropriate treatment, optimal screening, and follow-up can be performed.

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