Breus' Mole Associated with Hereditary Thrombophilia in A Teenage Girl

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Breus' mole is a rare event and it is most often found in the placenta of stillborn fetuses. It may occur in patients with disorders of circulation, complex heart disease, hypertension, diabetes and anticoagulation therapy. A 17 year old women with no previous gravida, was admitted to our clinic with intermittent vaginal bleeding and lower abdominal pain after 2 months amenorrhea. On admission, her general condition was normal and biochemical profiles were within normal ranges. Ultrasonography showed an enlarged uterus with hyper/hypoechogenic vesicular areas. Serum β-human chorionic gonadotropin (HCG) was tested as 33096 U/ml. Suction curettage under was performed due to the diagnosis of molar pregnancy. However, histology of the material was reported as Breus' mole. There was no evidence for complete or partial hydatidiform mole. In the genetic examination of the material, no chromosomal abnormality was detected. Homozygous mutation on factor V leiden gene was determined in the patient. Breus' mole should be considered in differential diagnosis of early gestational disorders, and the patients with Breus'mole should be tested for hereditary thrombophilia.

Key Words: Breus' mole, Hydatidiform mole, Teenage girl, Thrombophilia

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Introduction

Breus' mole is a rare event and it is most often found in the placenta of stillborn fetuses. Since the original description by Breus in 1892, it was believed that massive subchoripnic hematomas beneath the chorionic surface, projecting focally into the amniotic cavity were the result of fetal death.[1] However, Breus' moles in placentas from live-born infants were also reported in the literature.[2,3] Breus' mole is also considered a variant of placental abruption and bleeding from the placental site may cause dissection under the membranes and may form thrombohematomas from this concealed bleeding.

The etiology, pathogenesis, and significance of Breus' mole is not exactly known. Its incidence was reported as 1:1200 placentas and may occur in patients with disorders of circulation, complex heart disease, monosomy, hypertension, diabetes and anticoagulation theraphy.4 Some cases of Breus' mole were identified prenatally in the placenta of living fetuses by ultrasonography.[5,6] In most of these instances, there was intrauterine growth retardation and eventual fetal demise. This condition suggests that Breus' mole is often a premortem event and may cause to fetal death. However, Breus' mole in early pregnancy period mimicking hydatidiform mole have not been reported in the literature.

In the present case report, we reported an unusual form of Breus' mole presenting with signs and complaints of molar pregnancy in a teenage girl during early gestation.

Case Report

A 17 year old women with no previous gravida, was admitted to our clinic with intermittant vaginal bleeding and lower abdominal pain after 2 months amenorrhea. The patient had no any medical or surgical treatment before admission. She had no history of deep vein trombosis, small strokes, and systemic disease such as hypertansion and diabetes mellitus. These complaints were not also recorded in medical history of her first degree relatives. The medical and genetic family history of her husband was also unremarkable. There were no complications such as infection, anemia and trauma in the patient during last two monts.

On admission, her general condition was normal, blood pressure: 120/85 mmHg, pulse rate: 82/min, and body temperature was 36.8 ^{o}C. Rountine blood count, biochemical profiles, and urinanalysis were in normal ranges. Her body mass index (BMI) was 20 kg/m^2 and blood group was 0 positive with no atypical antibodies. At the gynecological examination, no vulvar or vaginal abnormality was observed, cervix apperared nulliparous, and the size of uterus was about 12 weeks of gestation. Ultrasonography showed an enlarged uterus with dimensions of 12 cm x10 cm x9 cm and endometrial thickness

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was 29 mm with hyper/hypoechogenic vesicular areas. There was 44x28mm sized ovaries with crumb view on sonographic examination. Due to the suspicion of molar pregnancy on sonographic examination, serum β-human chorionic gonadotropin (HCG) was tested and determined as 33096 U/ml. After these findings, the patient was diagnosed as having molar pregnancy.

Suction curettage under sedoanalgesia was planned and performed after the diagnosis of molar pregnancy. The patient was placed on the following medications; antibiotic infusion (cefotaksim sodium 2gr/day), intravenous oxcytosin infusion, and hydration theraphy with 1000cc serum phsyologic and 1000cc %5 dextrose. Postoperative vital signs were recorded as normal, and no complication was observed after intervention. The patient was discharged on the postoperative second day with follow-up appointments.

Microscopic examination of the endometrial tissue specimens revealed numerous cystic spaces in necrotic decidua, fibrosis on the wall of the small and medium-sized vessels (Figure 1).

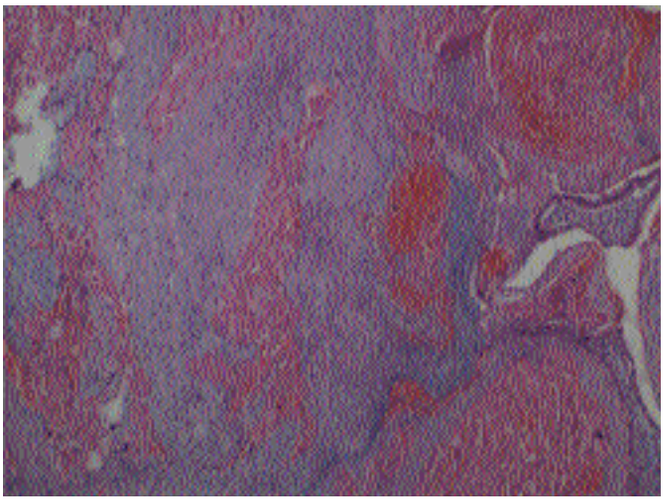

Figure 1: Subchorionic infarction and hematoma (H&E x 40)

Also thrombosis and complete obstruction of the lumina of some vessels with fibrosis were detected. There were colloid-like secretions in these cysts (Figure 2). The clot was formed principally by maternal blood. Histology of the material was reported as hematoma with plasental cysts and appeared to be Breus' mole. There was no evidence for complete or partial hydatidiform mole. In the genetic examination of the material, no chromosomal abnormality was detected. The patients was tested for inherited maternal thrombosis including prothrombin, factor V leiden, methylene tetrahydrofolate reductase (MTHFR) mutations, and protein S, C deficiency, and homozygous mutation on factor V leiden gene was determined. In the postoperative period, β-HCG levels of the patient fell to 16700 IU/ml on postoperative day 2, 165 IU/ml in the 2nd week, and decreased to normal reference ranges in postoperative 4th week.

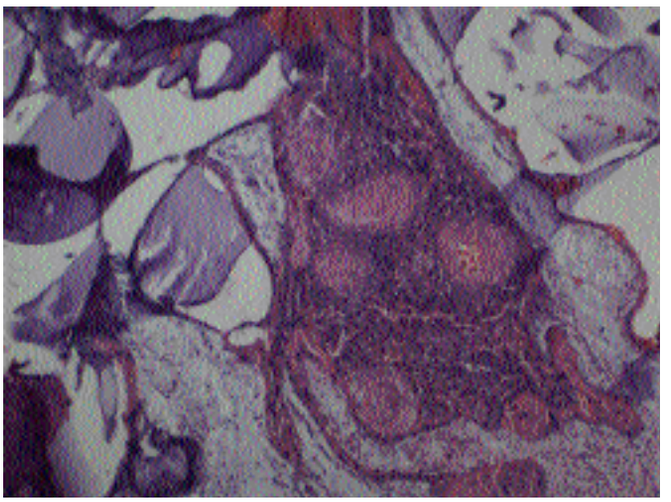

Figure 2: Cystic spaces with colloid-like secretion and fibrosis on the wall of the vessels (H&E x 40).

Discussion

Breus' mole first described by Karl Breus in 1982 is a very rare entity. Breus described a casual relationship between massive thrombohematomas at the preplacental site and fetal demise. The previous reported cases of Breus' mole were usually associated with a living fetus at the second or third trimesters of gestation. The present case is an unusual form of Breus' mole presenting with the findings of hydatidiform mole during early pregnancy period with no gestational sac or fetal pole.

The etiology and pathogenesis of Breus' mole are currently unknown. It has been suggested that a number of possible etiologies including some in which the thrombohematoma is maternal in origin and others in which it is of fetal origin. Fox has claimed that the phenomenon may be secondary to obstruction of the venous channels draining intervillous spaces.[3] Kim et al. showed the presence of both Breus' mole and partial hydatidiform mole on placental examination of a 23-week fetus with triploidy.[7] Differently, in the genetic examination of the material, any chromosomal or fetal abnormalities were not determined in the present case. The clot was formed principally by maternal blood. There was also no evidence for complet or partial hydatidiform mole on histopathologic examination.

Breus'mole is also found in the placenta macerated stillborn fetuses and may be the preceeding event before fetal demise.7 Breus' mole when massive can be associated with flow disturbance in spiral arteries and may lead to growth restriction, fetal distress, spontaneous abortion and perinatal death by interferring with blood supply to the developing fetus. The site of hematoma is the critical factor for the prognosis of pregnancy and it can be diagnosed on prenatal imaging.[8] If it was determined antenatally, serial scans to monitor the size of

the haematoma would have been indicated. It was shown that the presence of Breus' mole in early pregnancy was associated with adverse pregnancy outcome.[9] Madu reported a Breus' mole case presented for antenatal care at 12 weeks' gestation.[10] He followed the pregnancy throughout the 33 weeks' of gestation. Preeclampsia and intrauterine growth retardation (IUGR) were developed as adverse pregnancy outcomes.

Koçak et al. reported a rare form of Breus' mole presenting with abruptio placenta and preterm labor at 27 weeks of gestation.[11] The case had also resembled a large abdominal wall defect with appearance of multiple coiled masses in the amniotic cavity. Nishida et al. reported a massive subchorionic hematoma complicated by IUGR and oligohydramnios diagnosed at 22 weeks of gestation.[12] The patient was managed until delivery at 33 weeks of gestation. All previous cases with Breus' mole were detected in ongoing pregnancies which were diagnosed on ultrasonography. However, the present case was admitted in early pregnacy weeks with no fetal or gestational structures. The present case has suggested that early development of Breus' mole lead to embryonic demise. After fetal demise, only hematoma was observed on ultrasonographic examination, and this appearance was easly confused with hydatidiform mole. In addition, homozygous mutation of factor V leiden may be a risk factors associated with Breus' mole in the present case.

Some authors describe Breus mole as a morphologic "variant of missed abortion". The β-hCG will probably be higher with Breus' mole as the blood supply of the chorionic tissue is completely severed in missed abortion.[1] Similarly, β-hCG level was higher, and no fetal pole or gestational sac was determined on ultrasonography. Therefore, we first diagnosed the case as hydatidiform mole instead missed abortion.

In conclusion, Breus'mole is usually identified in the placeta of living fetuses. However, it may also occur during early period of pregnancy, and it may be easly confused with some disorders such as hydatidiform mole or missed abortion. Therefore, Breus' mole should be considered in differential diagnosis of early gestational disorders, and the patients with Breus'mole should be tested for hereditary thrombophilia.

Teenage Bir Kızda Herediter Trombofilinin Eşlik Ettiği Bir Breus' Mole Vakası

Breus mole çoğunlukla ölü doğumların plasentasında rastlanan oldukça nadir bir durumdur. Genellikle dolaşım bozukluğu, kardiyak hastalığı, hipertansiyonu, diyabeti olan ve antikoagulan tedavi alan hastalarda gözlenir. Onyedi yaşında daha önce gebeliği olmayan hasta 2 aylık amenore sonrası oluşan intermittan vajinal kanama ve alt abdomen ağrısı şikayetiyle kliniğe başvurdu. Başvuru esnasında, genel durumu normal ve biyokimyasal bulguları normal sınırlar içindeydi. Yapılan ultrasonografide içerisinde hiper/hipoekojenik alanlar içeren oldukça büyük bir uterus saptandı. Serum β-HCG 33096 U/ml olarak tespit edildi ve hastaya molar gebelik ön tanısıyla suction küretaj yapıldı. Bununla birlikte histoloji Breus' mole olarak rapor edildi. Komplet veya parsiyel hidatiform mol bulgularına rastlanmadı. Materyalin genetik incelemesinde herhangi bir kromozomal bozukluk saptanmadı. Hastanın trombofili incelemesinde homozigot factor V leiden mutasyonu tespit edildi. Breus' mole erken gebelik patolojilerinde göz önünde bulundurulmalı ve bu bozukluğun saptandığı hastalar herediter trombofili açısından taranmalıdır.

Anahtar Kelimeler: Breus' mole, Hidatiform mol, Teenage kız, Trombofili

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