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Unusual twin pregnancy: complete hydatidiform mole with coexistent normal fetus

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Summary

The authors present a case of complete hydatidiform mole and coexisting fetus (CHMCF) in which mole gestation caused a placenta previa; with a posterior preterm premature rupture of membranes (PPROM) and ending in the 28th week of gestation due to acute chorioamnionitis, obtaining a live preterm newborn.

Key words: Trophoblastic disease; Hydatidiform mole; Twin pregnancy.

Introduction

The presence of a complete hydatidiform mole coexisting with a healthy live fetus (CHMCF) is a rare form of twin pregnancy. Diagnosis is difficult and its management is controversial.

Case Report

The patient, a 30-year- old woman, had become pregnant spontaneously. She presented with hCG levels over ten MoM and hyperemesis at first trimester of gestation. However, an embryo with heartbeat was observed at ultrasound and screening of aneuploidies showed a low risk. A routine ultrasound performed at second trimester of gestation showed a vacuolar image in the posterior uterine wall, with a venous pattern, suggestive of molar degeneration, so a partial mole was suspected.

An amniocentesis was done and result showed a normal karyotype (46, XY), therefore, partial mole was ruled out and the authors faced another possible but rare diagnosis: CHMCF. Fetal and maternal risks related to a possible evolution of trophoblastic disease and adverse outcomes associated with this entity, were explained to the patient, but she decided to continue with the pregnancy. At week 24, the patient had vaginal bleeding and preterm premature rupture of membranes (PPROM) was diagnosed, antibiotic therapy was given, and corticosteroids were administered for accelerating fetal lung maturation. Simultaneously, an ultrasound diagnosis of placenta previa with partial occlusion was performed. Despite the increased risk for this new situation, the patient did not wish to end pregnancy, so an expectant management was kept. Subsequent ultrasounds showed a heterogeneous image with anechoic areas, on the posterior uterine wall, partially covering the internal cervical os, compatible with a molar placenta (Figure 1). Meanwhile, at the anterior uterine wall, a placenta of normal characteristics was present (Figure 2). Fetal growth was adequate and a severe oligoamnios was present.

Maternal physical examination and blood analysis remained under normal parameters up to the 28th week, when uterine contractions, fever, and uterine tenderness were found. Cardiotocography showed an irregular uterine activity and fetal tachycardia. Control hemogram showed leukocytosis.

Due to a clinical setting compatible with acute chorioamnionitis and presence of placenta previa, a cesarean section was performed, obtaining a male newborn of 1,100 grams with an APGAR index of 3/8. Placenta was extracted without complications; observing some vesicles and intraoperative uterine curettage was performed. Histopathology analysis reported a normal placenta without any signs of molar degeneration, and multiple fragments of tissue, of "spongy" aspect, with translucent vesicles up to ten mm in diameter, findings concordant with a complete hydatidiform mole. Follow-up of 24 months showed a satisfactory clinical and analytical evolution of the patient, without any signs of a persistent trophoblastic disease. The newborn remained hospitalized for four months, having complications due to prematurity, but with an adequate evolution.

Discussion

The finding of CHMCF is rare, with an incidence of

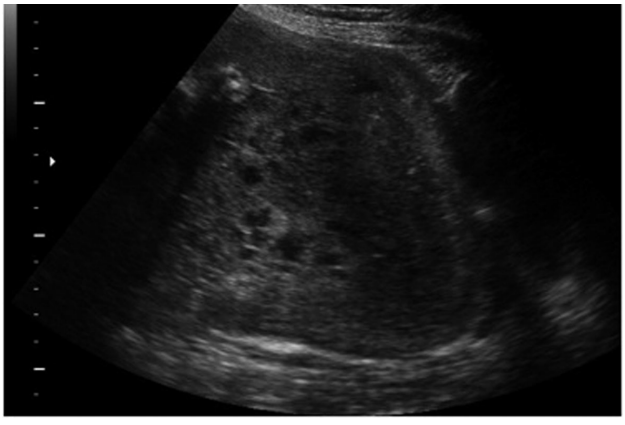

Figure 1. — Ultrasound showing molar placenta.

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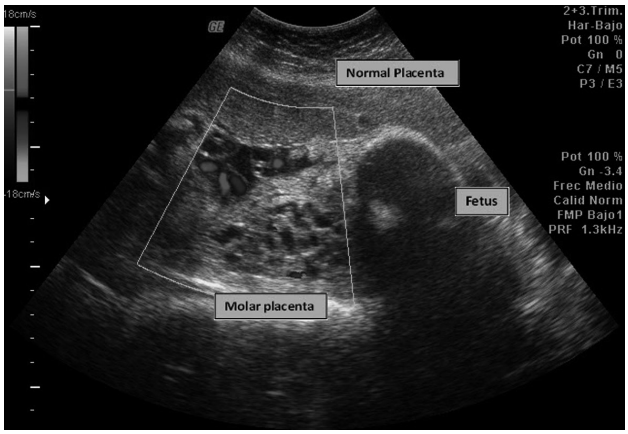

Figure 2. — Ultrasound showing fetus, normal placenta, and molar placenta on the posterior uterine wall.

1/22.000-100.000 pregnancies [1, 2]. Differential diagnosis between partial mole and CHMCF is difficult and a definitive diagnosis is made by histopathology analysis. If there is an ultrasound finding that suggests a CHMCF, an amniocentesis should be performed because the result of diploid karyotype might support the diagnosis of CHMCF; however, a triploidy is concordant with a partial mole. This has a very important clinical relevance because the fetal survival in a CHMCF is reported as high as 40%; as for the partial mole, the termination of the pregnancy, due to a non-viable fetus, is highly encouraged [1, 2].

In the present case, fetus had a diploid karyotype, which supported the decision of continuing the pregnancy under strict surveillance. Definitive diagnosis was a twin pregnancy with independent implantation sites, a complete mole, and a normal fetus.

In spite of the possibility of fetal survival in CHMCF [3]; there is a high percentage of adverse outcomes, such as miscarriage, intrauterine fetal demise, preeclampsia, etc; therefore, its management is controversial. Current evidence indicates that, if fetus has normal karyotype and normal development, expectant management could be a reasonable option with an adequate follow-up of the pregnancy [4-6]. Moreover, a supposed increase in the incidence of persistent trophoblastic disease in these cases, in comparison with a complete mole in single gestation, is uncertain. Some authors described a higher risk of developing persistent trophoblastic disease [7, 8]. However, another article recently concluded that risk in cases of CHMCF is similar [9]. Also, it has been described that the risk of malignancy does not increase with the progression of pregnancy, which supports the decision of expectant management [8]. In the present case, a healthy live newborn was obtained; and, even though obstetrics complications occurred, posterior follow-up was adequate without evidence of persistent trophoblastic disease.

To the present authors' knowledge, although there are several cases reported of CHMCF, none occurred with placenta previa of the molar component and chorioamnionitis, therefore this case provides novel insights and it had a favourable outcome despite complications.

To conclude, in cases with a high suspicion of CHMCF, an amniocentesis should be performed, and if the karyotype is normal, conservative management with a close follow-up could be advised.

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