



Twin Pregnancy with Complete Hydatidiform Mole and Coexistent Fetus

Embarazo gemelar con mola hidatiforme completa y feto coexistente

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Summary

Included in the spectrum of gestational trophoblastic disease, as one of the least frequent entities, is the coexistence, in a biamniotic twin pregnancy, of a complete hydatidiform mole and a normal viable fetus. The diagnosis of this condition requires a careful and thorough ultrasonographic examination, which will help in excluding the two main differential diagnosis in this scenario: partial hydatidiform mole and mesenchymal placental dysplasia. By following the obstetric examination protocol, and using academic radiologic knowledge, the examiner might reach a precise diagnosis. Here we present a case of a twin pregnancy with complete hydatidiform mole and coexistent fetus, a rare finding. We also present a short literature review on the topic of clinical management, given the fact that this disease might be associated with pregnancy complications and high risk of malignant behavior.

Resumen

Dentro del espectro de enfermedad trofoblástica gestacional, una de las entidades menos frecuente es la coexistencia, en un embarazo gemelar bicorial biamniótico, de una mola hidatiforme completa y un feto viable normal. El diagnóstico de esta condición requiere un cuidadoso examen ecográfico, con el fin de descartar la presencia de dos importantes patologías que componen los principales diagnósticos diferenciales en estos casos: la mola hidatiforme parcial y la displasia mesenquimal placentaria. Siguiendo de forma acuciosa el protocolo de ecografía obstétrica, y utilizando los conocimientos como explorador, se puede realizar el diagnóstico con precisión, diferenciando entre estas dos entidades. A continuación, se presenta el caso de un embarazo gemelar con mola hidatiforme completa y feto normal coexistentes, un hallazgo poco frecuente. Se realiza revisión de la literatura en cuanto a manejo y seguimiento de esta entidad dada su propensión a causar complicaciones en la gestación y a presentar comportamiento maligno.

Introduction

Twin pregnancies with a morphologically normal fetus and hydatidiform mole are a rare finding, with an estimated incidence of 1 in 20,000 to 1 in 100,000 pregnancies (1). They are differentiated from a partial mole by a complete normal-looking live fetus with its respective placenta, which is adjacent to a mass of tissue with changes compatible with hydatidiform mole (2). A case is presented with clear documented ultrasound findings that show the coexistence of a pregnancy with normal fetus and placental tissue with evident molar changes, confirmed by histopathology.

Presentation of the case and radiological findings

19 year old patient, primitive, who goes to the emergency room for moderate genitorragia without other associated normotensive symptoms. On physical examination, only a small amount of genitorragia was found to be positive on vaginal examination. A single live fetus with normal morphology, normal-appearing placenta and fundocorporal location with marginal

cord insertion is documented by ultrasound; adjacent to the placenta, a mass of microcystic composition is described -towards the left side of the uterine body- without Doppler enhancement, occupying the uterine cavity in its left aspect, managing to delimit its contours clearly with respect to the layers of myometrium, the fetus and the normal placental disc previously described (figures 1, 2 and 3). The ecographic study is concluded as a biamniotic bicorial twin pregnancy, with normal gestation of 23 weeks 5 days and complete hydatidiform mole coexisting.

The patient decides to continue the gestation and is hospitalized to receive strict control by the treating specialists. When she reaches 26 weeks of gestation, the patient becomes hypertensive with an altered toxemic profile, for which vaginal delivery is induced; however, due to abundant genitorragia, she must undergo a cesarean section. A single, live fetus is obtained, without external malformations, attached to the placental disc by the umbilical cord. Microcystic material compatible with diagnostic suspicion of hydatidiform mole is extracted from the uterine cavity.

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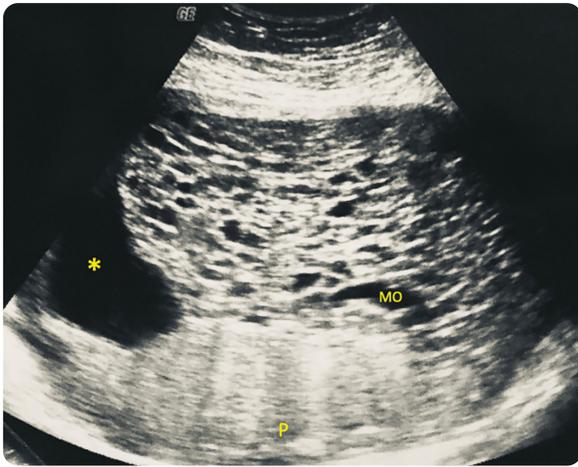


Figure 1. In this image in transversal orientation the following structures are observed: amniotic cavity of viable gestation (asterisk), complete molar gestation (MO) and placental disc of viable gestation (P). Fetus not visible in this cut.



Figure 2. 3D image showing the fetal face without morphological alterations.

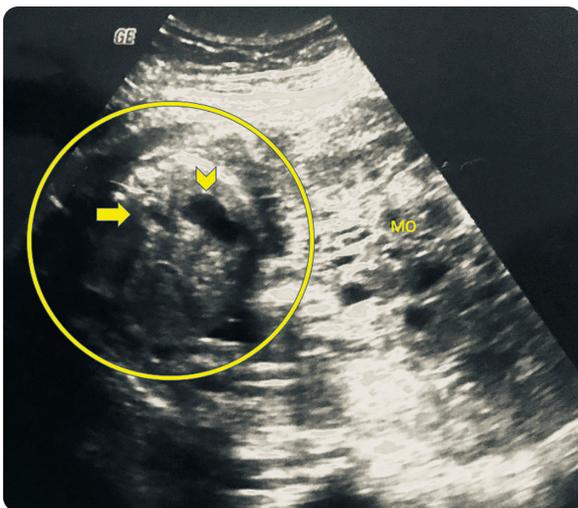


Figure 3. In this image in transversal orientation the following structures are identified: the fetal abdomen is shown inside the circle; the arrow points to the fetal abdominal aorta, while the gastric cavity is indicated by the arrow head. Adjacent to these structures, on the right, is the molar gestation (MO).

The placental disc, cord and suspected mass of molar gestation are sent for anatomopathological study, which confirms that it is a bi-cortical, biamniotic gestation, with one of the products consisting of complete hydatidiform mole.

Discussion

L Gestational trophoblastic disease consists of a spectrum of malignant and benign tumors that include complete or partial hydatidiform mole, infiltrating mole, choriocarcinoma, trophoblastic tumor of the placental insertion site and epithelioid trophoblastic tumor (3).

High variation in the prevalence of trophoblastic disease has been documented depending on the reporting country: 1 per 1,000 pregnancies in the United States and Europe. Asian countries have the highest incidence of these cases, which is attributed to severe vitamin A deficiency (4). A healthy twin can develop in coexistence with a complete hydatidiform mole in up to 1 in 20,000 pregnancies, and up to 40% end in live birth (3). The management of these cases is a challenge, due to the high risk of prenatal and perinatal complications, which include antepartum hemorrhage and fetal death; it is important to emphasize that up to 63% of pregnant women will develop persistent disease after delivery, with metastasis in half of the cases (4).

The coexistence of hydatidiform mole and normal fetus can be divided into three types. The first, corresponding to that described in this case, consists of a twin pregnancy in which a normal fetus is presented - with a diploid karyotype (46 chromosomes, 23 maternal and 23 paternal) and with a placenta of usual appearance - accompanied by a twin consisting of a complete hydatidiform mole (46 chromosomes of paternal origin) (4). The second type results in a single fetus of triploid karyotype (69 chromosomes, 23 maternal and 46 paternal) accompanied by a placenta with partial hydatidiform changes (4). Finally, there may be a twin pregnancy in which one of the products is diploid with normal placenta (46 chromosomes, 23 maternal and 23 paternal) and the other is triploid with partial hydatidiform mole in the placenta (69 chromosomes, 23 maternal and 46 paternal) (4).

Ultrasound remains the main diagnostic tool in partial or complete molar gestations (3). In this context, the molar component will be observed as multiple small spaces of anechoic content, which vary in size from 1 to 30 mm, which has been described as “grape cluster” and correspond to hydrophic chorionic villi (3). Two differential diagnoses of twin gestation with complete mole and coexisting normal fetus consist of partial hydatidiform mole and placental mesenchymal dysplasia. In the first case, cystic spaces are presented together with a morphologically altered deceased fetus, while the second entity is characterized by the vascularization of the cystic spaces; furthermore, the latter consists of a monochorial gestation, while twin pregnancy with coexisting complete hydatidiform mole is dizygotic, in which a normal placenta and fetus are identified, separated from a completely molar placenta, as is the case under discussion (3).

As mentioned, there is an increased risk of maternal and fetal complications in twin pregnancies with a hydatidiform mole, including vaginal bleeding, preeclampsia, hyperthyroidism, preterm delivery, and spontaneous fetal death (5). There is also the probability of persistent trophoblastic/metastatic disease requiring chemotherapy, with the lungs being the most frequent site of metastasis, followed by the vagina (5).

Previously, the management of twin gestation composed of complete hydatidiform mole and live fetus consisted of early termination of pregnancy (5, 6). However, continuing gestation has become a viable option in the 21st century, supported by a series of Japanese cases in which researchers noted that the risk of malignancy does not change with advanced gestational age, but adequate monitoring of complications is important (7).

In the context of malignant gestational trophoblastic disease, it has been described that it is highly sensitive to chemotherapy and there are several management schemes (5, 8). In the early stage, this entity is frequently cured using a single chemotherapeutic agent, while in advanced stages several agents are required to achieve such an outcome, among them the combination of etoposide, methotrexate, actinomycin D, cyclophosphamide and vincristine is the most used, with complete remission in 78% of the cases (5, 8).

The described case becomes important due to the richness of its imaging findings and its low frequency; it is important the adequate visualization of all the fetal parts, the presence of the umbilical cord together with a placenta of usual ultrasound aspect and its coexistence with a complete mole that, ultrasound wise, manages to separate from the viable gestation.

From the clinical point of view, it is to note that these gestations can reach term depending on the maternal comorbidities, the fetal welfare and the quality of monitoring that can be offered. Clinical follow-up is also important at the end of the gestation and in subsequent pregnancies, given the risk of metastatic disease and recurrence of molar gestation.

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