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Twin Pregnancies Combining A Molar Pregnancy And A Normal Fetus: A Rare Case Report And Review Of The Literature.

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Abstract*: The association of a twin pregnancy and a complete hydatidiform mole is an extremely rare obstetrical situation. All authors agree on the poor prognosis of such an association, with an increased potential for evolution towards a gestational trophoblastic tumour with sometimes a molar invasion engaging the maternal and foetal prognosis. We report the case of a patient admitted for metrorrhagia at 16 weeks of amenorrhoea, and whose obstetrical ultrasound showed a twin pregnancy associating a molar pregnancy and an evolving singleton pregnancy, and whose evolution was marked by a late abortion at 20 weeks of amenorrhoea, with an early post-abortal evolution towards a non-invasive gestational trophoblastic tumour.*

Keywords : gemellar pregnancy, hydatidiform mole, invasive mole

INTRODUCTION :

Multiple pregnancies with one or more apparently normal embryos in a molar pregnancy are exceptional. This entity can pose diagnostic and therapeutic difficulties. Once the diagnosis has been made, continuing the pregnancy or waiting is a clinical dilemma, given the risk of immediate serious maternal complications or distant complications. In this context, we report a case of a twin pregnancy with a complete mole and a live fetus diagnosed prenatally, and whose evolution was marked by the occurrence of a late abortion at 20 weeks of amenorrhea, with an early post-abortal evolution towards a non-invasive gestational trophoblastic tumour.

CASE REPORT :

A 25 year old primigravida patient, followed for 2 years for hyperthyroidism on Carbimazole, admitted for minimal metrorrhagia at 16 SA of her spontaneous pregnancy.

The clinical examination found a patient with slightly discoloured conjunctiva, with a gynaecological examination with a uterine height greater than the gestational age estimated at 18 SA, with a macroscopically normal, closed cervix and bleeding of endo-uterine origin.

A practical biological work-up was in favour of a microcytic hypochromic anaemia with a haemoglobin level of 10.10/dl, and a quantitative B-hCG level of 186,000 UI

An obstetric ultrasound was performed showing a heterogeneous echogenic image suggestive of hydatidiform mole associated with an apparently normal fetus with its placenta apart (figure 1). The decision was expectant with close clinical, biological and ultrasound monitoring.

At 20 days' gestation, the patient was readmitted to the obstetric emergency room for heavy metrorrhagia. After conditioning, and during monitoring, the patient spontaneously ruptured her water bag with expulsion of a 300g fetus with its normal placenta, and an associated vesicular product (figure 2).

The patient subsequently underwent an ultrasound-guided aspiration.

The pelvic ultrasound examination to check the vacuity was satisfactory.

Pathological examination was consistent with complete mole.

Subsequent weekly monitoring of the serum B-hCG level was marked by a stagnation of values over 4 consecutive weeks, suggesting a diagnosis of gestational trophoblastic tumour. The extension work-up concluded that there was no evidence of invasion or molar metastasis, and the patient was scored as a 2 according to the FIGO 2000 prognostic classification. The patient received 4 courses of methotrexate-based mono-chemotherapy 1mg/kg/d with a good clinical and biological evolution and tolerance. The patient received a total of 5 courses, divided into 3 initial courses and 2 courses after negativation.

One year's monitoring was satisfactory, with a subsequent spontaneous pregnancy after 3 years, with no obstetrical features.

DISCUSSION :

Twin pregnancies with hydatidiform mole and a normal live fetus are rare and complicated. Its incidence is estimated to be between 1/22,000 and 1/100,000 pregnancies (1).

The diagnosis of molar pregnancies is most often made in the first trimester, whereas the diagnosis of twin pregnancies with a live fetus and hydatidiform mole is classically made later in the second trimester, in a picture of gestational trophoblastic tumour (GTT) [2]. The ultrasound appearance is that of a normal fetus, with its normal placenta associated with a molar mass next to it that may take on the characteristic appearance of a complete hydatidiform mole in the honeycomb. [2] The b-hCG level is not reliable enough to make the diagnosis, although it may guide it. Only definitive pathological examination, aided by immunohistochemistry (anti-p57 antibodies), confirms the diagnosis and predicts the prognosis [3-5].

Two distinct types are described, based on morphological, histological and cytogenetic criteria: complete mole with diploid fetus and diploid placenta and partial mole with live triploid fetus associated with lethal malformations, 16

Early recognition of such a situation is a challenge for the practitioner to indicate termination of pregnancy or to opt for a wait-and-see attitude while running the risk of likely fetal complications, such as in-utero fetal death and abortion, and/or maternal complications such as massive bleeding, severe pre-eclampsia, hyperthyroidism, and the risk of tumour transformation or invasion, (7,8).

The risk of tumour transformation is much higher in this pathological association, and the incidence according to most authors is estimated to be between 50 and 57% of cases. [5, 7, 9]

The management of such a situation is difficult and not consensual due to the rarity of cases published in the literature, but the general trend is to resort to therapeutic termination of pregnancy when the diagnosis is made sometimes early in the first trimester. [10]

With the development of monitoring methods, notably ultrasound and Doppler, most authors opt for continuing the pregnancy with the possibility of even allowing it to progress to term, despite a viability rate of no more than 52% of cases [11].

The risk of tumour transformation of a gestational trophoblastic tumour is debatable:

In the literature; Sebire et al. report a rate despite continued pregnancy close to that observed in complete hydatidiform mole. In the literature, Sebire et al. report a rate close to that observed in complete hydatidiform mole despite continuing the pregnancy [12], while other authors indicate a higher rate in the region of 50% of cases [15,7,9], which justifies armed biological and ultrasound surveillance after delivery.

In the case of our patient, we opted for expectant care despite her young age, hyperthyroidism and anaemic syndrome, while running the risk of complications. The evolution was towards a gestational trophoblastic tumour scored at low risk according to the FIGO 2000 prognostic classification [10] and methotrexate-based mono-chemotherapy was instituted with a good evolution, without repercussion on the subsequent obstetrical outcome.

CONCLUSION :

The evolutionary potential of molar pathologies requires an armed clinical and biological follow-up. Ultrasound is not always suggestive and macroscopic examination does not always show a typical grape cluster appearance. Only the histological aspect of the villi, helped by an immuno-histo-chemical study (anti p57 antibodies), can confirm the diagnosis.

The course of action is generally to favour an expectant attitude, given the absence of any relevant argument for the need to terminate these pregnancies, and to introduce surveillance despite the maternal risks.

ICONOGRAPHY :

Figure 1: Ultrasound appearance showing an embryo with normal placenta, associated with an adjacent heterogeneous echogenic honeycomb image.

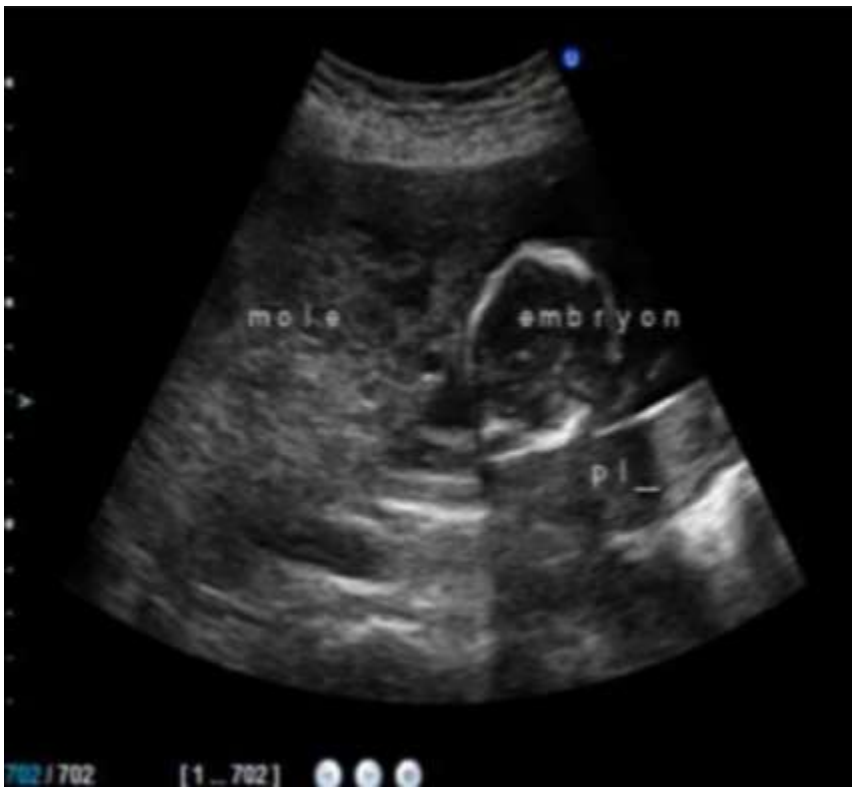
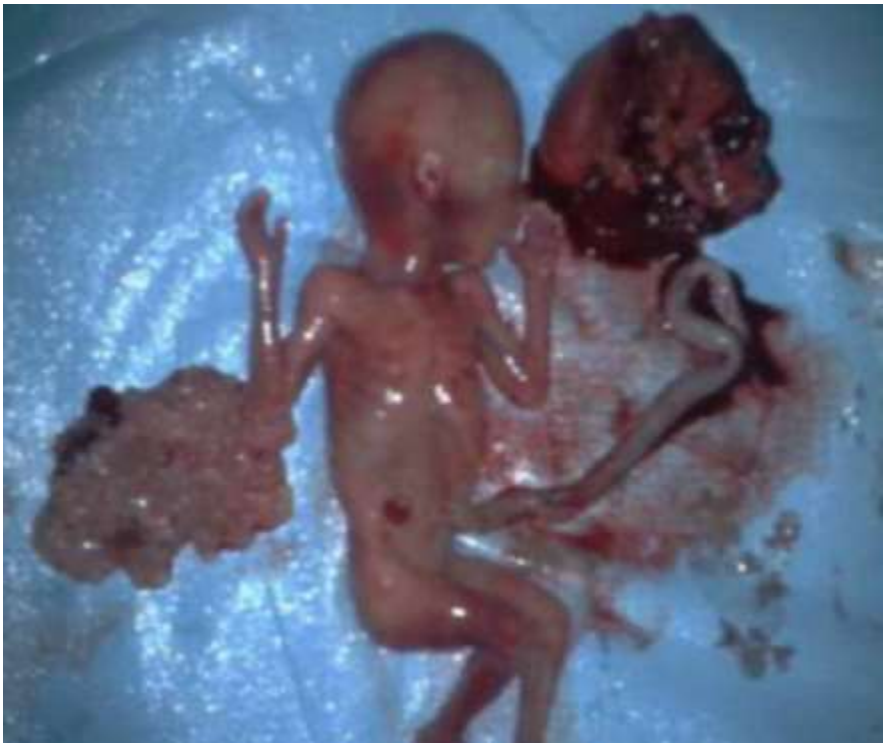


Figure 2: Normal fetus with associated adnexa and a second grape-like placental mass after a late abortion



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