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Twin Dizygotic Pregnancy Associating Partial Mole and a Normal Appearing Fetus: A Case Report and Review of Literature

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Abstract: Pregnancies with a hydatidiform mole and a live fetus are rare form of trophoblastic gestational disease, this situation occurs in about 1 in 20 000 to 1 in 100 000 gestations. The symptomatology is poor and the diagnosis can be made by ultrasound assessment. Management is still problematic seen that fetal survival is constrained by complications of molar pregnancy. The risk of persistent trophoblastic disease in twin molar pregnancies is higher compared to single mole .We report here a case of a woman who consulted for vaginal spotting and vomiting in the first trimester twin pregnancy with partial hydatidiform mole detected by ultrasound. Following spontaneous miscarriage, histopathological examination findings were compatible with a partial mole pregnancy coexisting with normal diploid fetus.
Keywords: Partial mole, dizygotic pregnancy, ultrasound, karyotype, β-hCG.

INTRODUCTION

Twin molar pregnancy with a partial mole and a coexisting live fetus is a particular type of gestational trophoblastic disease with the prevalence of 0.005%-0.01% of all pregnancies [1], that keeps increasing with the use of assistive reproductive techniques [2]. The partial mole can coexist with a normal diploid fetus or with a triploid abnormal fetal structure.

The management consists generally to offer termination of pregnancy however normal fetal karyotype allows pregnancy to proceed with the consent of the patient under strict follow-up and control of maternal complications in particular persistent trophoblastic disease.

With this case and through a review of literature, we will try to understand mechanisms of this rare entity, diagnosis modalities and management issues of partial hydatidiform mole and coexisting normal fetus.

CASE PRESENTATION

A 26 year-old married woman, gravida 3 para 1, with one vaginal normal delivery was admitted to our unit at 11 weeks of gestation for pelvic pain associate to incoercible vomiting and mild bleeding from the vagina. She has a history of one early pregnancy loss; she does no smoke or drink, and no personal or familial genetic disorders.

Examination on admission finds a closed cervix, with spotting from cervical os, uterine size corresponding to 14 weeks, her Heart rate was 100 beat/min and blood pressure was measured as 120/80 mmHg.

Transvaginal ultrasonography examination showed a gravid uterus containing a single live fetus with crown rump length of 46 mm corresponding to a gestational age of 11 weeks and 4 days (Fig. 1), the placenta in the anterior region of uterus contained a focal area of enlargement with numerous cysts suggestive of molar pregnancy (Fig.2); the rest of the placenta seems to be normal. Moreover the right ovary was enlarged and measured 8,89x 7, 97 cm likewise the left ovary measured 10, 5 x5, 5 cm (Fig.3).

Laboratory findings included hemoglobin at 10 g/dl, normal platelet count, the serum titer of β-human chorionic gonadotropin level was 72 000 mIU/mL, and thyroid function tests were correct.

Pregnancy termination has been proposed, the patient was counseled regarding abnormal ultrasound images and informed about the risks and complications associated with this type of pregnancy .the decision to maintain the pregnancy was made provided closely monitoring.

3 weeks later the patient was readmitted presenting heavy vaginal bleeding, she went in a spontaneous abortion, emergency aspiration was carried out to get hemostasis the pregnancy products was sent for histopathologic examination.

Macroscopic observation of the aspired product showed a normal looking placenta measuring 18 x 12x 5 cm and most cotyledons appear without abnormalities, the fetus and umbilical cord appear normal (Fig.4), however on the side a small adjacent area was covered with multiple large vesicles associated to hemorrhagic fragments and necrotic tissue (Fig.5). Closer examination has identified a dichorionic diamniotic membrane between the two structures.

Microscopic study revealed normal chorionic villi on the placenta and molar villi on the small area. Fluorescence in situ hybridization confirmed the diagnosis of partial mole pregnancy composed by triploid cell line XXY coexisting with a normal diploid fetus.

The patient was discharged and placed on post molar follow-up, β-HCG showed normal regression curve, the levels reached normal within 5 weeks. Ultrasound assessment after 6 months shows no persistent trophoblastic tissue.

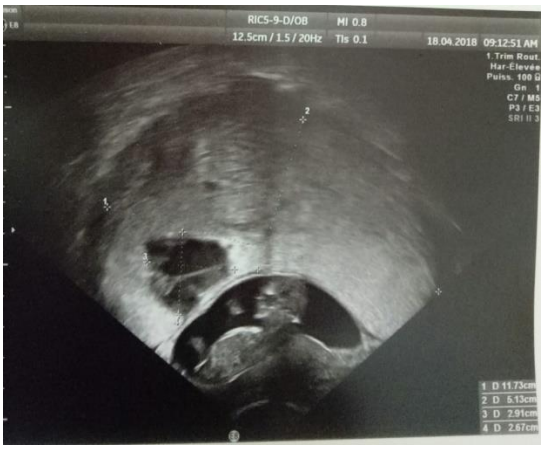

Fig-1: Ultrasound showing enlarged focal area suggestive of partial mole coexisting with live fetus at 11 weeks of gestation

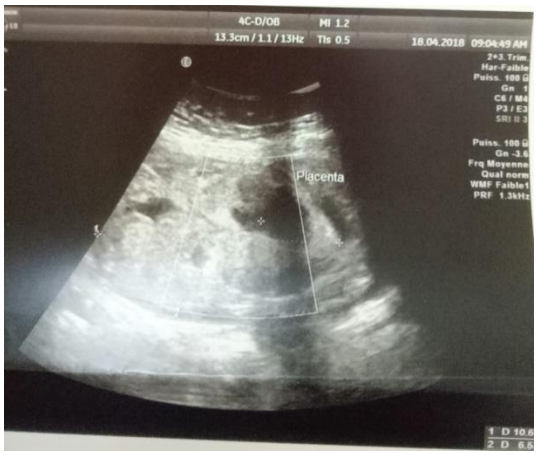

Fig-2: Section showing molar changes in placenta

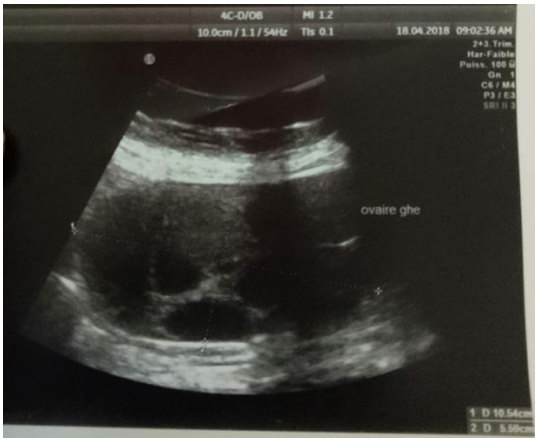

Fig-3: The enlarged left ovary with multiple theca lutein cysts

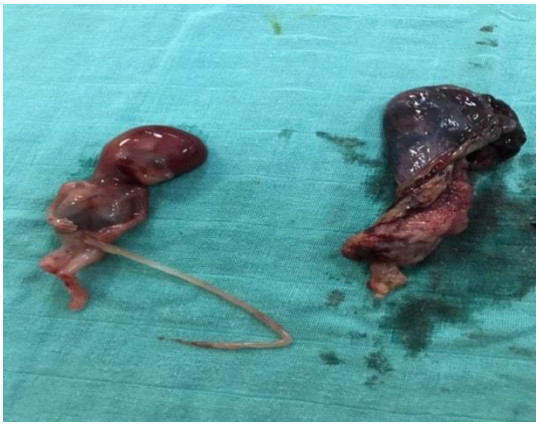

Fig-4: Picture of the normal fetus with umbilical cord and placenta with molar tissue

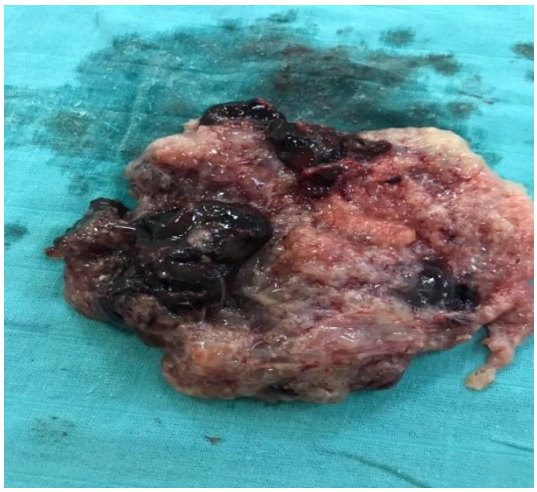

Fig-5: Molar tissue attached to placenta with hemorrhagic fragments

DISCUSSION

Coexisting molar pregnancy with normal fetus is extremely rare; this condition is even exceptional when the co-existing normal live fetus is associated with a partial mole than a complete hydatidiform mole [3]. Live births in this pregnancies vary from 16 to 56% [4], effectively very few fetus reached term as they often have spontaneous or induced terminations owing to innumerable maternal complications.

Complete mole is diploid and all chromosomes have a paternal origin, it's the commonest type of molar degeneration, differently majority of partial moles are triploid containing two lots of paternal chromosomes and one lot of maternal haploid genes[3]. Fetus coexisting with mole has chances of survival in dizygotic pregnancies; on the contrary when the twin pregnancy is monozygotic the coexisting fetus with partial mole is triploid and tends to die in the first trimester [5]. Our case matches with the situation where

the partial mole is associated with a diploid normal fetus in a context of dizygotic pregnancy.

Many complications have been described in the case of co-existing fetus with molar pregnancy like miscarriage, preterm labour, heavy bleeding, and severe anemia in the fetus, in addition to preeclampsia, trophoblastic hyperthyroidism, pulmonary oedema and thromboembolic phenomena [6]. Maternal complications also include persistent gestational trophoblastic disease that is associated with partial molar in 14-33% of cases [7]. This risk seems not to increase with gestational age [8]. Stellar *et al.* reported a higher risk of developing persistent trophoblastic disease in cases with twin molar pregnancy with a coexisting fetus compared to singleton molar pregnancies [9].

Ultrasound examination is helpful in making diagnosis; two scenarios present themselves focal and diffuse molar changes, most of the time the differentiation between partial mole and complete mole associated to twin pregnancy is challenging mostly in first trimester (sensitivity of 18% to 49%) [10], the trick is to follow the fetus umbilical cord, if it connects to the molar placenta one could exclude the twin pregnancy contrariwise when it is inserted on the normal placenta [11].

A lot of differential diagnosis can also disrupt the vision, mesenchymal dysplasia, hydropic changes in the placenta and focal vascular lesion as chorioangioma, MRI evaluation according to Himoto *et al*. can distinguish between placental mesenchymal dysplasia and twin pregnancy with mole coexisting with live fetus [12]. Concerning the choriangioma velocity blood flow analysis by Doppler has a value in differentiating partial molar pregnancy from chorioangioma [11].

The rule of prenatal diagnosis by amniocentesis to predict genetic abnormalities is indisputable before advising the continuation of pregnancy. Fetal karyotype is the most important predictor of fetal outcome in partial molar pregnancy [6]; In fact the karyotype can determine the different genetic origin of conceptual product which can help to differentiate partial molar pregnancy from complete hydatidiform mole [13]. This technique can be limited in early stages of gestation, an alternative using DNA polymorphic analysis give more accuracy [14].

Serum β-human chorionic gonadotropin level is also a helpful indicator to determine the prognosis; some authors have fixed a rate of 10^6 mIU/ ml beyond which the termination of pregnancy is privileged .High level may signify aggressive trophoblastic growth and it's suggestive of pregnancy termination [15].

Management of twin gestation with partial mole and coexisting live fetus is challenging, generally obstetricians propose the termination of pregnancy in order to anticipate maternal complications [1], currently conservative attitude is possible in practice and the pregnancy may be continued with the consent of couple as long as complications are absent, this approach should always be considered whenever tertiary care services and strict follow-up are available [16].

Postpartum period surveillance is based on the evolution curve of β-human chorionic gonadotropin and serial sonographic examination in order to detect tissue retention or eventual recurrence.

CONCLUSION

Twin gestation with partial mole and coexisting diploid fetus is a rare condition that can be diagnosed by ultrasound assessment and cytogenetic analysis; the chances of delivering a healthy baby are very low in addition to high risk of maternal complications that can be fatal for the mother. Treatment criteria still need improvement the reason for which clinicians must weigh challenges in management approach for an optimal outcome.

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