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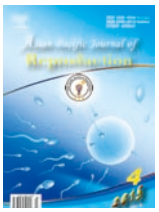

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Twin pregnancy with both complete hydatiform mole and coexistent alive fetus: case report

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ABSTRACT

Twin pregnancy consisting of a complete hydatidiform mole coexisting with a live fetus is a rare condition with an incidence of 1 in 22 000 to 1 in 1 00 000 pregnancies. Clinical information is limited and management is difficult due to the risk of pregnancy complications such as fetal death, vaginal bleeding, preeclampsia, hyperthyroidism, and the risk of persistent gestational trophoblastic disease. Thus, the report described about the change of size and implantation site of the molar mass coexisting with a live fetus is rare especially about sonographic findings such as echo patterns. Recently we experienced a case of complete hydatidiform mole with a healthy infant delivered at term.

1. Introduction

Twin pregnancy with a complete hydatidiform mole and a normal fetus is extremely rare, with an estimated incidence of one in 22 000–100 000 pregnancies[1,2]. Twin pregnancy with CHMF (complete hydatidiform mole coexisting with a live twin fetus) resulting in a healthy take-home baby is rare, with only 56 cases documentated in detail in literature. CHMF cases are at high risk of spontaneous abortion, preterm delivery, intrauterine fetal death, bleeding, preeclampsia, persistent trophoblastic disease (PTD).

This presents the physician and patient with significant clinical dilemma between immediate intervention and expectant management, particularly in the case of highly desired pregnancy. The decision will be guided by the problems that may arise and those that are already present. The patient's parity status will also influence the decisions [3].

We report a case encountered at the emergency Department of Obstetrics and Gynaecology of Maternity and Neonatology Center in Tunisia, a twin pregnancy with a CHMF that resulted in live newborn[1].

2. Case report

A 33-year-old woman, gravida 6, para 3, with two previous normal delivery, of two normal female infant and three miscarriages, was referred from a local clinic with suspected molar pregnancy at 32 weeks' gestation.

It is a poorly monitored pregnancy, the patient has consulted only three times, first time at 8 weeks of amenorrhea and ultrasound

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examination revealed two gestational sacs and only one with fetal pole and cardiac activity and the other with a thickened and vacuolated trophoblast. The second consultation was at SA 16, where Sonography revealed a structurally normal fetus compatible with the gestational age, a normal amount of amniotic fluid and normal-looking placenta in the right posterior portion of the uterus. There was an approximately 8.5 cm × 6.5 cm mixed echoic lesion with a multicystic portion suggestive of molar tissue separately in the right anterior portion of the uterus (Figure 1). the patient was lost to sight and has re-consulted his doctor at 32SA. Detailed targeted sonography at this gestational age revealed a normal-appearing fetus. The maximum length of the molar mass was about 20 cm (Figure 2) for which she was referred to our Emergency Department of Obstetrics and Gynecology. The examination was unremarkable: blood pressure (BP) was 110/80 mm Hg; Urinalysis showed 1+ proteinuria. Her serum -human chorionic gonadotropin (β -hCG) level was greater than 58 000 mIU/mL. At 38 weeks, a healthy female neonate weighing 2 270 g was born by a normal spontaneous vaginaldelivery. The normal-looking placenta weighed 440 g, and an approximately 22 cm 20 cm 4 cm irregularly shaped mass of necrotic tissue with multiple grapelike vesicles was delivered (Figure 3 and 4). Pathologic examination results were consistent with a CHMCF.

One week postpartum, the serum β -hCG level declined to 2700 mIU/mL and was normalized within 12 weeks without any chemotherapy.

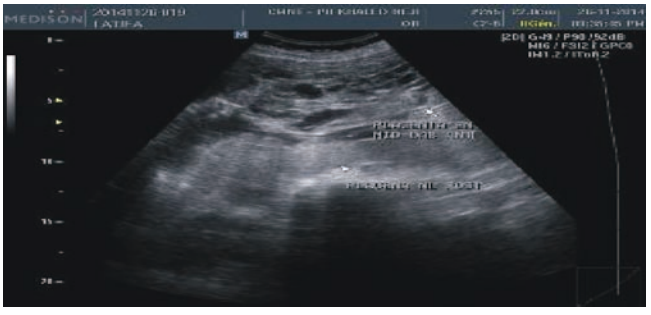

Figure 1. Two placenta normal (posterior) and molar mass(anterior).

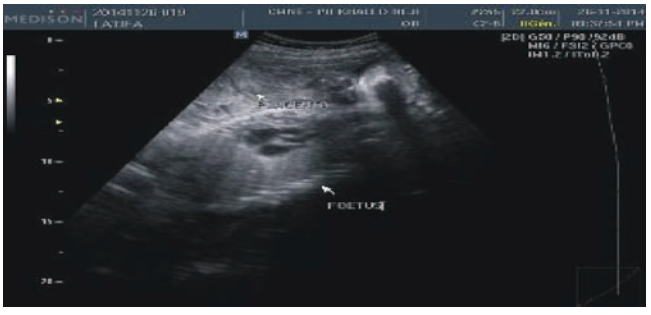

Figure 2. normal-appearing fetus with the molar mass.

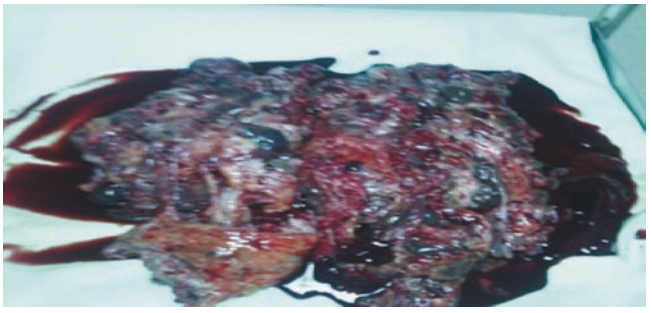

Figure 3. irregularly shaped mass of necrotic tissue with multiple grapelike vesicles.

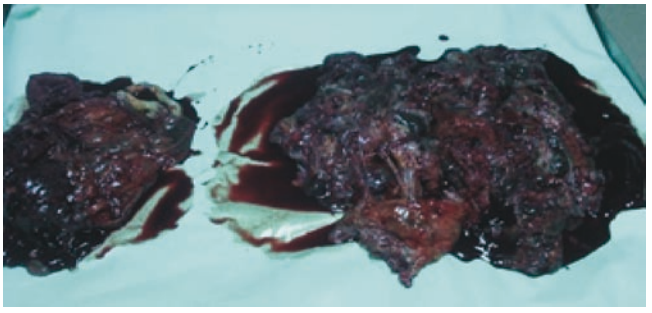

Figure 4. Two placenta normal and molar mass.

3. Discussion

In the late 1970s, Vassilakos et al. firstly described two different pathologic entities, partial and complete hydatidiform mole (CHM), with different mechanisms of origin based on cytogenetic analysis[1].

Complete and partial moles have distinct fetal and maternal complications. In the combination of a partial hydatidiform mole, the fetus is almost always triploid and the indication for a termination of pregnancy is evident. In contrast, the fetus may be normal in a twin pregnancy with a CHMF and continuation of pregnancy is frequently associated with severe maternal complications, leading to a mother vs. fetal clinical problem[4]. The true incidence of this rare entity is difficult to establish, and some suggest that the modern increased incidence of iatrogenic multiple gestations will cause a higher incidence of CHMF[5].

Natural course of these pregnancies is unpredictable, with 60% going beyond 28 weeks. Length of pregnancy has no impact on complications like persistent trophoblastic disease3 or malignant sequelae. Favourable response to chemotherapy in gestational trophoblastic disease is another factor favouring conservative approach[6].

Nevertheless, parents who choose to continue a twin pregnancy with CHMF should agree to take the risk of possible maternal complications associated with molar pregnancy such as early-onset pre-eclampsia, hyperemesis gravidarum, hyperthyroidism, vaginal bleeding, anemia, development of theca lutein ovarian cysts, respiratory distress because of trophoblastic embolization to the lungs, and PTD. Parents must also be aware that these complications

may lead to fetal intrauterine growth retardation, fetal distress and premature delivery[7]. As concerns the risk of PTD, Steller et al. suggest that it is higher in cases of CHMF compared with single molar pregnancies and that, when present, it more commonly progresses to metastatic disease[8]. Niemann and al find out that the risk of PTD after CHMF is not significantly higher than in single molar pregnancies [9]. A 'wait-and-see' approach should be considered rather than immediate termination of pregnancy, because the risk does not increase with advancing gestational age. In our experience our patient has't developed PTD after one year follow up carried out with montly β-hCG level and ultrasound examinations, so we suppose that the risk is comparable in both single molar pregnancies and CHMF[1].

Ultrasound examination is helpful in making a pre-evacuation diagnosis but the definitive diagnosis is made by histological examination of the products of conception. The use of ultrasound in early pregnancy has probably led to the earlier diagnosis of molar pregnancy. The majority of histologically proven complete moles are associated with an ultrasound diagnosis of delayed miscarriage or anembryonic pregnancy [3,4].

Ultrasonography has made it possible a diagnosis of a hydatidiform mole and co-existent fetus in the first trimester[10]. Prenatal testing of at least fetal karyotype is essential in deciding continuation and prognosis of the pregnancy[1].

The serum β-hCG level can be a helpful marker in the management of a CHMCF because it is usually highest at the beginning of the second trimester of pregnancy. When the serum β-hCG level remains greater than 106 mIU/mL, TOP(termination of pregnancy) should be considered. In contrast, in cases of successful pregnancy outcomes with viable fetuses, the serum β-hCG level usually starts to decline from the beginning of the second trimester, and sonography usually reveals a decrease in the size of the molar portion of the placenta.13,14 In our case, we had a fall in the serum β-hCG level and a decrease in the size of the molar mass, as well as disappearance of the patient's symptoms (eg, vaginal bleeding)[2].

Diagnosis should also include molar placental karyotype[11]. Although not available for our patient, as in most documented cases[4,5]. We are convinced that our case was CHMF due to diploid karyotipe, normal newborn, ultrasound demarcation between the normal and molar placenta and histopatological examinations[1].

In most cases when diagnosis was made in early preg nancy, termination of pregnancy was recommended. The maternal complication and the necessity of termi nation of pregnancy is an important matter in clinical management. Although the data of both studies come through oncologic reports and not exactly through gynecologic and obstetrics reports[9].

Therefore, management of molar pregnancy with an alive fetus is optional, although accurate and great care is required to find early signs of maternal or fetal com plications and in the presence of a stable pregnancy, nor mal karyotype and a normal sonogram, it is reasonable to allow the pregnancy to continue[12].

In conclusion, management of multiple pregnancy with a CHMCF still remains uncertain, In the past, most CHMF gestations were terminated immediately following diagnosis because of poor information concerning clinical features and natural history. But from the results of our case, we carefully suggest that in a CHMCF with a normal karyotype and no gross abnormalities on sonography, pregnancy may be continued as long as maternal complications are absent or, if present, controllable. However, treatment criteria still need improvement, and intensive maternal follow-up with serial radiologic examinations and β-hCG level monitoring is necessary during antenatal care and in the postpartum period and a detailed discussion (and informed consent) with the couple is necessary.

Conflict of interest statement

We declare that we have no conflict of interest.

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