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Ultrasound and MRI Findings of Twin Pregnancies with Complete Hydatidiform Mole and Coexisting Normal Fetus: Two Case Reports

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BACKGROUND: Both twin pregnancies with complete hydatidiform mole and coexisting normal fetus (CHMCF) and partial hydatidiform mole can be found in association with a live fetus and a placenta displaying a molar degeneration. Two cases of CHMCF using magnetic resonance imaging (MRI) for a diagnosis are reported. CASE: In the first, CHMCF was suspected at 12 weeks of gestation. At 18 weeks of gestation, the existence of molar placenta and a sac separating from fetus and normal placenta was clearly depicted on MRI. At 19 weeks of gestations, she had termination of pregnancy because of a development of gestational trophoblastic neoplasia (GTN) and started chemotherapy. In the second case, CHMCF was suspected at 14 weeks of gestation. MRI demonstrated the existence of molar placenta and a sac separating from fetus and normal placenta. She chose induced abortion and there was no evidence of GTN during the 1 year-follow up period. Pathological examination in both cases was consistent with a complete hydration mole and a coexisting normal female fetus. CONCLUSION: MRI was useful for an accurate diagnosis for CHMCF.

INTRODUCTION

Twin pregnancies with complete hydatidiform mole and coexisting normal fetus (CHMCF) are extremely rare with an estimated incidence of 1 case for 20,000 - 100,000 pregnancies [10,17,22]. Twin pregnancies with CHMCF are fraught with several complications for both the mother and the fetus, such as fetal death, vaginal bleeding, preeclampsia, hyperthyroidism, and a development of gestational trophoblastic neoplasia (GTN) which encompasses persistent/invasive mole, choriocarcinoma, placental site trophoblastic tumors and epithelioid trophoblastic tumor [14,17,18,22,23]. Twin pregnancies with CHMCF can be continued if there are no fetal anomalies or severe maternal complications [4,15]. The management of pregnancies with CHMCF has not been standardized. Continuation of CHMCF is an acceptable option, but the chance of a live birth is 7 - 37% [7,9,10,23].

Twin pregnancies with CHMCF are characterized by the coexistence of complete mole (diploid without fetal parts) and a normal live fetus with a normal placenta, while partial mole (triploid with evidence of fetal parts) can be found in association with a live fetus with abnormal placenta [23]. The differential diagnosis is important, because the fetus in CHMCF has a chance to survival, while the fetus of partial mole tends to die [10]. We report two cases of CHMCF in which magnetic resonance imaging (MRI) was useful for an accurate diagnosis during pregnancy. Written informed consent was obtained from the patients for publication of this case report and accompanying images.

CLINICAL CASES

Case 1

A 24-year old woman, gravida 0, para 0, had conceived after an ovulation induction with human menopausal gonadotropin and human chorionic gonadotropin. She had repeated vaginal bleeding since 6 weeks of gestation and got hyperemesis gravidarum at 8 weeks of gestation. At 9 weeks of gestation, transvaginal ultrasound showed a live fetus and multiple cystic lesion in the uterine cavity. At 12 weeks of gestation, she was referred to our hospital with a suspicion of hydatidiform mole and a coexisting live fetus. US images performed with showed a live fetus and adjacent multicystic mass, suggestive of a hydatidiform mole (Fig. 1).

Serum β-human chorionic gonadotropin (β-hCG) levels were 239,100 mIU/mL. Given informed the maternal and fetal risks associated with continuation of pregnancy, she decided to continue the pregnancy. Serial ultrasound examinations identified no apparent fetal abnormalities. Amniocentesis was performed to examine fetal karyotype at 16 weeks of gestation, but amniotic fluid could not be aspirated because of a narrow amniotic cavity. At 17 weeks of gestation, her chest x-ray was normal.

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At 18 weeks of gestation, the existence of molar placenta and a sac separating from fetus and normal placenta was clearly depicted on MRI performed with 1.5-T MR unit (Gyroscan NT; Philips Healthcare, Best, Netherlands) (Fig. 2). At 19 weeks of gestations, a computed tomography (CT) scan of the chest displayed multiple nodules, suggestive of metastatic molar lesions (Fig. 3). For starting the chemotherapy for GTN, a vaginal delivery was conducted due to her desire of an induced abortion at 21 weeks of gestations (Fig. 4). Histopathological findings were consistent with a complete hydration mole and a coexisting normal female fetus. Karyotyping of the mole revealed 46XX and karyotyping of the fetus revealed 46XX.

Four days after performance of artificial abortion, she was started chemotherapy with methotrexate (MTX). Serum β-hCG levels were decreased to normal ranges after 6 cycles of MTX and lung metastases disappeared. She has been without recurrence for three years since an initial chemotherapy.

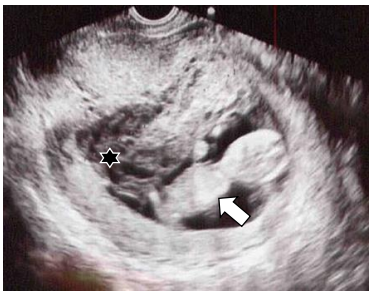

Fig. 1
Sonographic image at 12 weeks of gestation showing a multicystic mass (asterisk) and a live fetus (arrow).

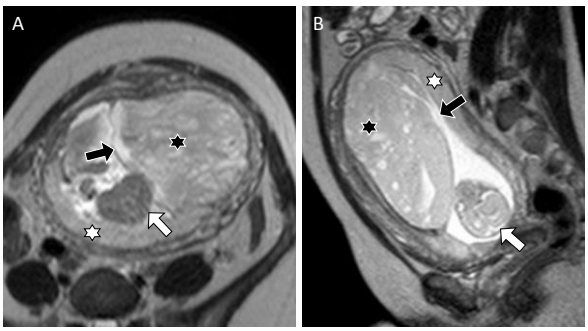

Fig. 2
MR images at 18 weeks of gestation
A. a transverse plane T2W image showing two sacs was separated by amniotic membrane (black arrow). The multisystic mass (black asterisk) and a normal placenta (white asterisk) and a live fetus abdomen (white arrow) were seen in the second sac. B. a sagittal T2W image showing two sacs was separated by amniotic membrane (black arrow). The multicystic mass (black asterisk) was within the uterine cavity anteriorly. The second sac is seen posteriorly with a fetal head (white arrow) and a normal placenta (white asterisk).

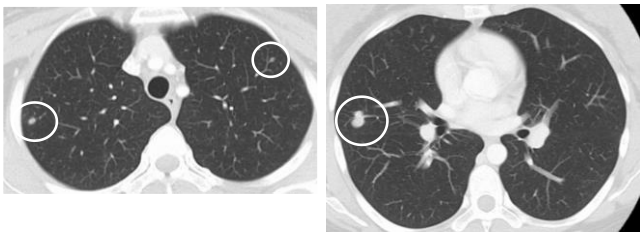

Fig. 3
CT scan of the chest at 19 weeks of gestation showing the multiple nodules, suggestive of pulmonary metastases.

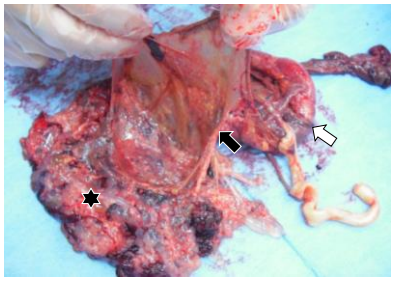

Fig. 4
Macroscopic appearance of normal placenta with umbilical cord (white arrow), amniotic membrane (black arrow) and hydatidiform mole (black asterisk).

Case 2

A 27-year old woman gravida, 0, para 0, had conceived naturally. At 14 weeks of gestation, she was referred to our hospital with a suspicion of CHMCF. Serum β-hCG levels were 296,052 mIU/mL. US images performed with Voluson E10 showed a live fetus and adjacent multicystic mass, suggestive of a hydatidiform mole (Fig. 5). Her chest x-ray was normal. MR images performed with 1.5-T MR unit (Achieva; Philips Healthcare, Best, Netherlands) demonstrated the existence of molar placenta and a sac separating from fetus and normal placenta (Fig. 6). At 15 weeks of gestations, she chose induced abortion (Fig. 7). Two days after performance of artificial abortion, a CT scan of the chest was normal. Histopathological findings were consistent with a complete hydration

mole and a coexisting normal female fetus. Karyotyping of the mole revealed 46XX. At 4 months-follow-up, serum β-hCG levels were decreased to normal ranges. At one year-follow-up, she was free of any sings of GTN.

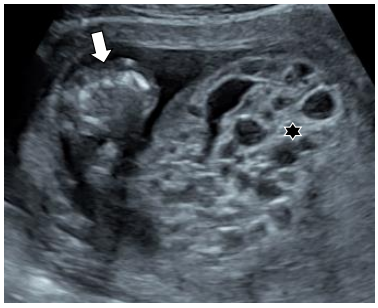

Fig. 5
Sonographic image 14 weeks of gestation showing a multicystic mass (asterisk) and a live fetus (arrow).

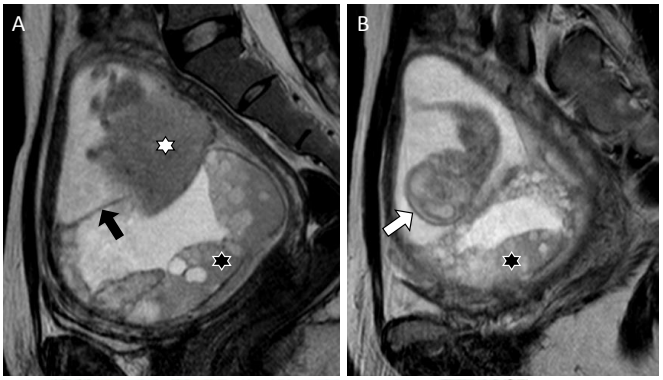

Fig. 6
MR images at 14 weeks of gestation
A. sagittal T2W image showing two sacs was separated by amniotic membrane (black arrow). The multicystic mass (black asterisk) was within the uterine cavity and a normal placenta (white arrow) was seen in the second sac. B. sagittal T2W image showing the multicystic mass (black asterisk) within the uterine cavity. A fetal head (white arrow) was seen in the second sac.

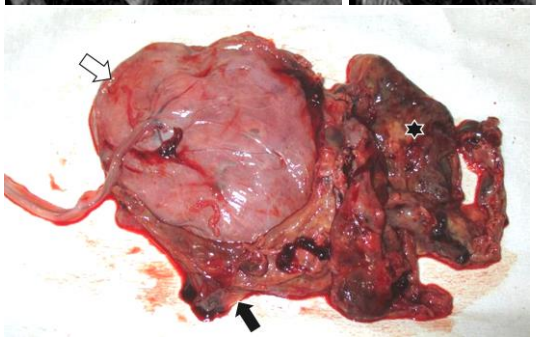

Fig. 7
Macroscopic appearance of normal placenta with umbilical cord (white arrow), amniotic membrane (black arrow) and hydatidiform mole (black asterisk).

DISCUSSION

Both CHMCF and partial hydatidiform mole can be found in association with a live fetus and a placenta displaying a molar degeneration. The differential diagnosis is important because the fetus in CHMCF has a chance to survival, while the fetus of partial mole tends to die [10]. US is a firstchoice screening modality. The US appearance of a hydatidiform mole has been described as an echogenic tissue with multicystic areas in the first trimester and as a 'snowstorm' appearance in the second trimester [2]. In CHMCF, the complete hydatidiform mole was depicted outside the fetal sac [6]. It may be the most important factor to obtain a whole image of the uterus and to assess the relation between the hydatidiform mole, fetal sac and the placenta [6]. In some reports, only 43-68% of patients with CHMCF were diagnosed correctly by US [5,20,21].

Relative to US, MR imaging has the advantages of nonionizing radiation, a large field of view, good tissue contrast and relative operator independence [3,11]. A recent study showed that exposure to MR imaging during pregnancy was not associated with increased risk of harm to the fetus [16]. The hydatidiform mole usually appears as a heterogeneous markedly hyperintense mass on T2-weighted images that distends the endometrial cavity [11]. MR imaging demonstrates myometrial invasion and extension into the parametrium clearly for diagnosing invasive gestational trophoblastic tumor [11]. There are no studies which have investigated the diagnostic accuracy of MR imaging in CHMCF. Some reports showed that MR imaging was useful in demonstrating the existence of molar placenta and a sac separate from the embryo and normal placenta, thereby leading to making a precise diagnosis of CHMCH and conclusively ruling out partial mole [1,6,13,24]. In current cases, the mole tissue appeared as multicystic lesions by US, but their location in relation to the fetal sac and the normal placenta was difficult to determine. The wide field of view and the excellent tissue contrast of MR imaging enable us to obtain a whole image of the uterus and to assess the relation between the mole, the normal placenta and a live fetus. We could diagnose CHMCF and exclude partial mole at 14 weeks and 18 weeks by using MR imaging. Amniocentesis is

available for prenatal diagnosis of the karyotype of the coexistent fetus [23]. An amniocentesis is adaptable after 15 weeks and it takes time to obtain 2 - 3 weeks the result, while MR imaging is adaptable before 15 weeks and images of placenta and fetus can be obtained immediately. MR imaging may be a possible alternative for a differential diagnosis during pregnancy.

In a series of Japanese patients, the rate of GTN was shown to be considerably higher in patients with CHMCF than those with simple complete mole [10]. Some authors reported that continuation of CHMCF pregnancy did not increase the risk of GTN [9,12]. A mainstay of management for complete and partial hydatidiform mole consists in an early diagnosis and immediate evacuation with a careful follow-up of patients until a normalizaton of serum hCG levels. However, the management of pregnancies with CHMCF has not been established. When GTN is suspected, chest x-ray is essential. Even if the chest x-ray is negative, a CT scan of the chest is recommended [8]. Micro-pulmonary metastases are shown to be seen on chest CT scan in 30-40% of patients with normal chest x-rays [8]. On the other hand, in the UK, a CT of the chest is not needed when chest x-ray is normal. Because small pulmonary metastatic lesions can spontaneously regress, discovery of micro-metastases does not affect prognosis of the patients [19]. In our case, micro-pulmonary metastases were detected on chest CT scan at 19 weeks of gestations, and those lesions increased 4 weeks later. CT scan may be acceptable for the diagnosis of GTN during pregnancies with CHMCF.

MR imaging might provide important information for a diagnosis for CHMCF rather than ultrasound examination and CT may be necessary during pregnancies with CHMCF occasionally.

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