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BRIEF COMMUNICATION

Twin Pregnancy with Complete Hydatidiform Mole and Co-existent Live Fetus

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Abstract A 14-week pregnant woman had a mixed echogenic mass of size 4x12 cm at placenta portion and normal fetus at ultrasound. The image of the mass was very similar to a huge subchorionic hematoma (SCH). The fetal chromosomes by amniocentesis were normal. The presumed diagnoses were huge SCH or twin pregnancy with complete hydatidiform mole and co-existent fetus (CHMCF). A male fetus weighing 760 g was delivered at 25 gestational weeks. The findings on the placental biopsy showed twin pregnancy with CHCMF. With ultrasound findings of this type, the possibility of a huge SCH and twin pregnancy with CHMCF should be considered.

Keywords Multiple pregnancy · Antepartum bleeding · Hydatidiform mole

Introduction

Twin pregnancy with a complete hydatidiform mole and co-existent fetus (CHMCF) is extremely rare. Its incidence was reported to be one per 22,000–100,000 pregnancies [1, 2]. Although survival of the co-existent fetus in CHMCF cases is important, it is not easy to preserve this type of fetus. The difficulty arises due to the risk of a spontaneous abortion, maternal complications associated with a complete mole, and difficulties of early diagnosis in pregnancy [3, 4]. When a twin pregnancy with CHMCF is suspected, differential diagnosis must consider the possibility of a singleton pregnancy in a partial mole and mesenchymal placental dysplasia (MPD) as well as a huge subchorionic hematoma (SCH) [5]. In particular, women with a huge SCH are accompanied by vaginal spotting during early pregnancy; therefore, diagnosing a twin pregnancy with CHMCF is more difficult.

We report a case of a woman who was presumed, prenatally, to have a huge SCH; however, a twin pregnancy with CHMCF was confirmed after birth. The ultrasound findings of this case were compared with three other cases.

Report of Case

A 31-year-old gravida 4, para 1 patient with a 14-week pregnancy, was referred to our institution with abnormal placental findings on the ultrasound examination. The patient suffered from intermittent vaginal spotting during her pregnancy. There was no massive vaginal bleeding. According to the medical records of the local clinic, there was only one gestational sac at the early stage of pregnancy. The antenatal examinations results including thyroid function tests in the early stage of pregnancy, were normal. The patient had just mild hyperemesis.

The obstetric ultrasound examination showed no abnormal findings in the fetus or amniotic fluid. There was a foam-like mixed echogenic mass of size 4x12 cm in the lower segment of the uterus. The presence of a placenta except abnormal mass could not be clearly confirmed by ultrasound examination. To rule out a single pregnancy in a partial mole, a chromosome analysis via amniocentesis was performed at 16 gestational weeks. Amniocentesis revealed a fetal chromosome complement of 46, XY. The placenta was identified as having a normal ultrasound appearance at

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this time. The patient and her husband received counseling and the information that a normal fetus with a huge SCH was more likely and that a dichorionic twin pregnancy with CHMCF was less likely. The parents decided to continue this pregnancy without other examinations for differential diagnoses because she had no CHM-related abnormal findings.

At 20 weeks of gestation, the fetal size and structure and the amount of amniotic fluid were within the normal ranges. A huge mass was located in the lower uterine section, and a normal placenta was located in the fundal area (Fig. 1). The patient had no abnormal symptoms or signs such as vaginal bleeding, high blood pressure, or proteinuria.

At 25 gestational weeks, the patient complained of recurrence of vaginal spotting with regular abdominal pain. One day later, a cesarean delivery was performed because of severe fetal distress during labor. A male fetus of 760 g was delivered, whose apgar scores were 3 in 1 min and 5 in 5 min. The normal-appearing placenta and foam-like mass were separately delivered from the uterus. The findings of the placental biopsy showed a 240 g immature singleton placenta and another placenta with multiple grape-like cystic vesicles, measuring $16.0 \times 15.0 \times 3.0$ cm in aggregate. The patient was diagnosed as having a dichorionic twin pregnancy with CHMCF according to the results of p57 immunohistochemistry. The maternal serum β-hCG level was 129 mIU/mL at 20th postoperative day. A chest computed tomography showed a small metastatic nodule in the right middle lobe of a lung. Following three administrations of methotrexate by injection, the maternal serum β-hCG level was normalized by 70 days after birth. The infant improved and was discharged at 119 days after birth and is undergoing outpatient observation.

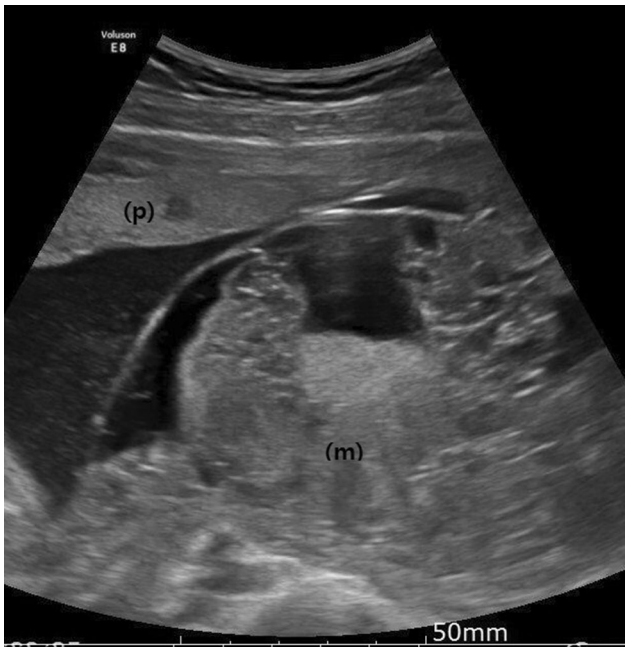

Fig. 1 The ultrasound image of normal placenta (*p*) and complete mole (*m*) of presented case at 21 weeks of gestation

Discussion

The occurrence of a twin pregnancy with CHMCF is extremely rare [1, 2]. Although cases have been reported, the natural history of the condition has been poorly defined until recently. The optimal management remains challenging. Early diagnosis is important because of the risk of maternal and fetal complications. However, there are many cases diagnosed retrospectively after birth or abortions. When a foam-like mass is seen on an ultrasound, physicians may suspect a twin pregnancy with CHMCF. To reach an accurate diagnosis, other conditions such as a singleton pregnancy consisting of a partial hydatidiform mole with a fetus, MPD, and a huge SCH should be excluded. Classification of the condition with only an ultrasound examination is challenging. In a twin pregnancy with CHMCF, a "snow-storm" appearance typically occurs in addition to the normal placenta due to a complex, echogenic, intrauterine mass containing small cystic space on the ultrasound [6]. For a single pregnancy in partial mole, a normal placenta is not visible, and only a snow-storm-like placental mass is visible. MPD, which is a rare placental vascular anomaly characterized by placentomegaly and grape-like vesicles, is present in 0.02 % of all pregnancies. It is difficult to distinguish between a molar pregnancy and MPD with ultrasound alone. The most important factor in the differential diagnosis is the location of the abnormal mass. A CHM mass is located outside the fetal sac, and an MPD mass is within the fetal sac. Magnetic resonance imaging (MRI) could help to identify the location of an abnormal mass based in a fetal sac [7]. In the event of a huge SCH, both of hematoma and normal placenta are visible. The appearance of a hematoma is similar to that of a molar pregnancy mass. In these cases, there is no significant difference between a twin pregnancy with CHMCF and a singleton pregnancy with a huge SCH on a prenatal ultrasound. The ultrasound findings of the three other cases are presented. Figure 2a–c are images from the three cases of second trimester pregnancies presenting with a huge SCH. All of the three images in Fig. 2 and the image of present case in Fig. 1 show normal placenta and foam-like mass that are mixed, echogenic, and irregular.

In women who have suspicious findings on ultrasound accompanied by vaginal spotting, early onset pre-eclampsia, severe morning sickness, or abnormal thyroid

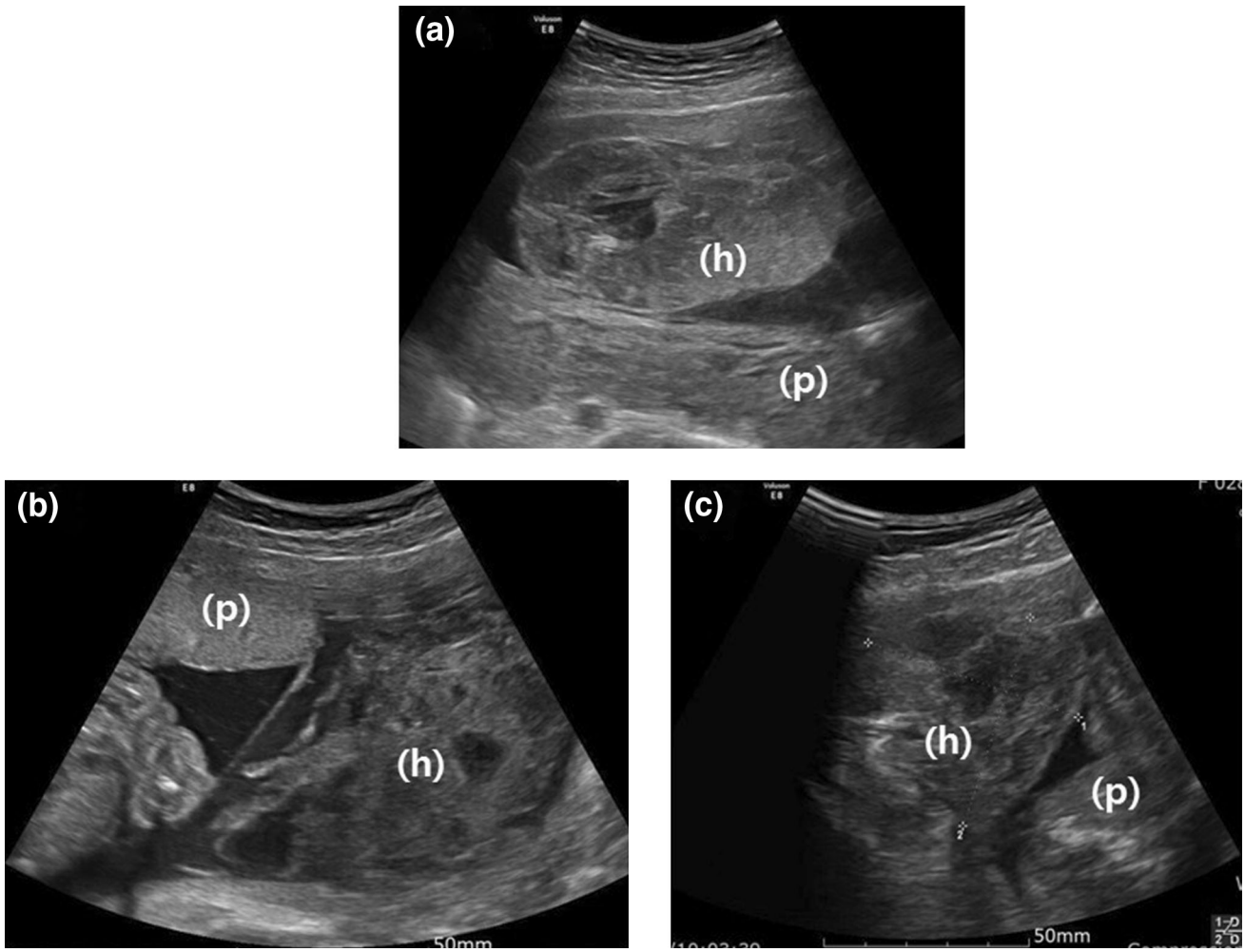

Fig. 2 The ultrasound images of normal placentas (*p*) and huge subchorionic hematoma (*h*) of three different pregnant women, at 23 weeks of gestation (**a**), 21 weeks of gestation (**b**), and 18 weeks of gestation (**c**)

functioning, the physician could suspect a twin pregnancy with CHMCF. However, those symptoms are not specific. A confirmation of their chromosomes via chorionic villus sampling of abnormal foam-like tissue might facilitate reaching a differential diagnosis; the sampling procedure carries a risk of hemorrhage. Serial follow-up of the maternal serum β-hCG levels might be of assistance in diagnosis during pregnancy.

When a twin pregnancy with CHMCF is diagnosed during pregnancy, detailed counseling with the patient regarding further treatment is needed because complications might arise, including pre-eclampsia, fetal demise, thyrotoxicosis, hemorrhage, trophoblastic embolism, and persistent trophoblastic disease [8]. According to previous reports, the fetal survival rate is <50 %, and 33 % of the women progress to a persistent gestational trophoblastic state after delivery [5]. Preterm deliveries might frequently arise because of vaginal bleeding, preterm labor, and fetal distress, as in the present case [9, 10]. There are cases in which termination of the pregnancy is necessary because of serious maternal complications including hemorrhage and pre-eclampsia [9]. There is a possibility of a full-term delivery without complications, and it might be reasonable to continue the pregnancy [11].

In this case, the patient was referred at the 14th gestational week, and, it was too late in the gestation to undergo chorionic villous sampling. Intermittent vaginal spotting was the only symptom suggesting the diagnosis of twin pregnancy with CHMCF, which frequently arises in cases of pregnancies with SCH. Amniocentesis, instead of a sequential test, was performed to rule out a partial mole. We could not monitor the maternal serum β-hCG levels. The condition, in this case, was not diagnosed exactly before birth. Although it was a very rare twin pregnancy with CHMCF, the pregnancy was continued without serious maternal complications, and the neonate has grown well. The natural progress of a twin pregnancy with CHMCF remains unknown. Reporting this case and the

ultrasound findings might be helpful for the diagnosis and treatment in future cases.

Compliance with ethical standards

Conflict of interest None.

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