CASE REPORT

Complete Hydatidiform Mole with Coexisting Twin Fetus: Usefulness of MRI in Management Planning

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Introduction

Complete hydatidiform mole with coexistent twin fetus is a rare condition. Only one out of a total reported 16 cases before the 1980s was diagnosed antenatally; hence, there was really no need to take a clinical decision. The number of cases of CHMCTF has since gone up dramatically with the availability of advanced ultrasound and emerging IVF techniques including newer ovulation drugs [1]. The optimal management is a challenge that remains a clinical dilemma. The present viewpoint is tilted toward expectant management with strict surveillance to prevent/treat complications early, so as to be able to deliver a normal baby in many of these patients. No difference in the risk of developing persistent gestational trophoblastic disease (PGTD) and gestational trophoblastic neoplasia (GTN) in patients undergoing termination of pregnancy as well as patients allowed to continue pregnancy has permitted the

clinician to delay intervention till after delivery, while successfully managing the complications associated with trophoblastic disease. The role of MRI in addition to ultrasound in correctly distinguishing this condition from the partial hydatidiform mole is discussed, thus assisting in the management plan.

Case History

The patient, a 25-year-old gravid 3, para 1, live 0 woman, with 4 months amenorrhea, was referred by a local clinic where she was diagnosed on ultrasound to have “Partial mole.” The patient had history of a missed abortion at 2 months, 7 years ago. She had delivered a live baby at home 4 years ago who died at the age of 3 years following prolonged fever.

An ultrasound at time of admission revealed a 16-week gestation with EFW 142 g. The liquor amount was normal with no fetal anomalies. The placenta was posterior grade 0. An abutting well-defined multicystic snowstorm-like mass (11.0 × 5.3 cm) was seen in the anterior and fundal location (Fig. 1a, b). The Doppler showed negligible vascularity in this mass. The cord was seen to be inserting normally to a normal-appearing posterior placenta. Bilateral theca lutein cysts were present. A diagnosis of complete hydatidiform mole with coexisting twin fetus was made.

An MRI scan done at the time of admission confirmed the sonographic diagnosis. The existence of molar placenta and a sac separate from embryo and normal placenta was clearly and unambiguously made out on MRI (Fig. 2a–c).

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Fig. 1 **a** Sonographic image showing a multicystic mass showing snowstorm appearance anteriorly within the gravid uterus. The second sac with the normal-appearing placenta and fetal part is also seen posteriorly. **b** BPD and FL are measured in the fetus corresponding to a gestational age of 20 weeks, 5 days

The β hcG was 811,780 units. TSH < 0.01, FT3 was 4.16, and FT4 was 1.96 (suggestive of thyrotoxicosis). BP was normal between 116/80 and 110/70 mmHg. There was no history of bleeding or passage of vesicles per vaginally. Other biochemical tests were normal.

The patient was put on expectant treatment, but unfortunately aborted 6 weeks later spontaneously. Serial β hcG were measured and fall monitored on a weekly basis till it returned to normal levels in about 12 weeks' time. Subsequent β hcG at fortnightly intervals showed no rise.

Discussion

A new life takes shape when one of zillions of sperms successfully attaches itself and fertilizes an ovum, resulting in zygote formation which subsequently divides and subdivides to result in an embryo (Fig. 3).

In aberration of this normal process of fertilization, at times, one or even two sperms fertilize an empty ovum. The fertilized egg (zygote) therefore contains only paternal DNA along with maternal mitochondria. This divides and subdivides to form a complete hydatidiform mole (CHM) pregnancy with no embryo development (46XX or 46XY-diploidy) (Fig. 3). This earlier view of the absence of embryo has now been challenged by the reported demonstration of androgenic CHM with embryonic tissue, which dies early in pregnancy itself [2, 3].

Sometimes, more than one sperm successfully fertilizes an ovum, resulting in a zygote which may be 69XXY (triploidy) or even 92 XXYY (quadripleidy) and which evolves into "partial hydatidiform mole" (PHM). Here, a small abnormal embryo develops, which is severely growth retarded along with focal molar changes in the placenta. This does not progress and soon aborts (Fig. 4).

Fig. 2 **a** Axial gradient image (BTfE) showing the multicystic mass within the uterine cavity anteriorly. A thin layer of hypointense collection is seen posteriorly. **b** Axial T1W image shows that the collection has a bright rim suggestive of hemorrhage. **c** Sagittal gradient (BTfE) image showing the relationship of intrauterine

structures with the multicystic mass lying anteriorly. The hemorrhagic collection (*solid black arrow*) is seen posterosuperiorly. The second sac is seen posteroinferior to the mass with fetal head and a normal placenta (*asterisk*)

In yet another situation, a dizygotic pregnancy occurs where a CHM develops side by side with a normal twin embryo. Here, the embryo is single, but the placentae are two—one molar and the other normal (Fig. 4).

The reported incidence of CHM in the USA is one in 1,000 pregnancies [4]. Its incidence is much higher in certain Asian races such as Japanese and in South East Asian countries. Indonesia has reported an incidence as high as one in 100 pregnancies.

The incidence of PHM is much higher, but is difficult to fathom as many of these are not noticed and simply labeled as “missed abortion” (non-molar).

The incidence of CHMCTF is quite rare and has been reported to be one in 22,000–100,000 pregnancies [5].

While management for PHM and CHM is early diagnosis and immediate evacuation with follow-up till a year after β hCG has returned to normal, the management of CHMCTF is more complex and challenging and has still

not been standardized. The earlier prevalent view was to let the pregnancy progress to delivery, despite the supposed increased risk of complications such as conversion into GTN, Preeclampsia, high likelihood of abortion, Thyrotoxicosis, vaginal bleeding, etc [6]. Only 40 % of these pregnancies result in live baby, with others normally aborting by the second trimester [7]. Since the fetus is viable and many of these patients have conceived late in age after many IVF attempts, the pregnancy is extremely precious. The current suggested management of these is counseling the patient of the risks involved and close monitoring till delivery, followed by management of CHM. The babies, thus born, have been reported to be normal [7].

In a large series of 77 CHMCTF patients, Sebire et al. [8] reported successful delivery of 31 live babies. In contrast, in a recent study by Lee in 2010, only one out of six patients delivered a live baby, while five ended in pregnancy termination. The successful CHMCTF pregnancy had few complications during

Fig. 3 Line diagram showing the genetic basis for development of normal pregnancy and complete hydatidiform mole

Fig. 4 Line diagram showing genetic basis for development of partial hydatidiform mole and complete H mole with coexistent twin fetus

pregnancy, no theca lutein cysts, with β hCG of around 250,000 units compared to over 500,000 units in a failed CHMCTF.

A large number (50 %) of these patients of CHMCTF develop PGTD [9]. This risk is higher than in CHM patients (20 %) [10] or PHM patients (4–11 %) [11, 12]. It has now been demonstrated that the risk of PGTD is not reduced even if pregnancy is terminated early instead of being allowed to continue to term.

Since the prognosis and management of PHM and CHMCTF differ, it is important to diagnose accurately and unambiguously. Since 1970, advances in ultrasound have revolutionized the diagnosis of CHM. It has been found to be extremely sensitive and specific in diagnosing molar pregnancy early. However, it is not as specific in the detection of PHM. Identification of a normal placenta separate from molar placenta suggests the presence of a twin and not a PHM. The role of MRI in demonstrating myometrial invasion by trophoblastic tissue and diagnosis of PGTD is well recognized and already proven. In addition, MRI was helpful in demonstrating the existence of molar placenta and a sac separate from the embryo and normal placenta in our case, making the diagnosis of

Fig. 5 Photograph of the abortus showing a dead fetus with the normal cord and placenta. The complete hydatidiform molar mass with bunch of grapes appearance is seen alongside

CHMCTF and conclusively ruling out PHM. A suspected diagnosis of PHM without MRI would have entailed CVS/ amniocentesis, which are invasive procedures and have consequent risks to pregnancy.

Hence, by using MRI, we could accurately diagnose CHMCTF and exclude PHM, and thereby avoid invasive procedures. The patient was put through expectant treatment with close monitoring and surveillance, and the pregnancy carried through till the patient unfortunately aborted spontaneously 6 weeks later (Fig. 5).

Key Statement

- In the case of complete hydatidiform mole with twin fetus, an earnest attempt must be made to manage expectantly till delivery.
- In a significant number (40 %) of such cases, it is possible to achieve the goal of delivering a healthy normal baby.
- The risks of complications such as GTN and PTD are not increased if the intervention is delayed till after delivery.

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