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CASE REPORT

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Complete hydatidiform mole with a coexisting twin live fetus (CHMTF): the uncommon diagnostic enigma—simplified

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Abstract

Background Twin pregnancy comprising of a complete hydatidiform mole with a coexisting twin live fetus is an uncommon condition with an incidence rate of 1 in 20,000 to 1 in 100,000 pregnancies, more so in assisted reproductive technologies. The primary diagnosis is made on ultrasound and adjunct fetal MRI helps in unequivocally differentiating it from other disorder.

Case presentation We present a case report of a twin pregnancy consisting of complete hydatidiform mole with a coexisting twin live fetus in a 27-year-old primigravida conception, primarily focusing on its diagnostic algorithm and related clinical aspects, adding to the paucity of existing literature.

Conclusions Following the correct diagnostic algorithm with imaging studies like USG and more importantly MRI, combined with the bioclinical picture helps in reaching the accurate diagnosis.

Keywords Twin pregnancy, Complete hydatidiform mole with a coexisting twin live fetus, CHMTF, Fetal ultrasound, Fetal MRI

Background

Twin pregnancy comprising of a complete hydatidiform mole with a coexisting twin live fetus (CHMTF) is an uncommon condition with an incidence rate of 1 in 20,000 to 1 in 100,000 pregnancies, more so in assisted reproductive technologies [1]. Patients with such a condition usually present with vaginal bleeding in the late first trimester. These are mostly diagnosed between 12 and 14 weeks using ultrasound with a surprisingly low detection rate of approximately 68%, as reported in the literature [2]. Diagnosis of this condition on ultrasound can be challenging; if unfamiliar with the characteristic appearance and their variations; as it can be easily confused with other conditions like singleton pregnancy with partial mole and live fetus or singleton pregnancy with a large intrauterine clot [3]. MRI has the added advantage of providing better soft tissue resolution, better tissue characterization and a larger field of view, allowing clear demonstration of two separate amniotic cavities, one containing a normal fetus and placenta, and the other containing a complete mole, thereby unequivocally differentiating it from the above-mentioned differentials [4].

We present a case report of a twin pregnancy consisting of a complete hydatidiform mole with live fetus, primarily focusing on its diagnoses and related clinical aspects.

Case presentation

A 27-year-old primigravida with intrauterine insemination conception in non-consanguineous marriage, presented in the emergency department of apex tertiary care referral center in North India at 12 weeks four days of pregnancy with complaints of vaginal bleeding for two weeks. Clinical per abdominal examination revealed the

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uterine size of ~ 16 weeks, which was large for gestation age. Multiple ultrasounds performed during this period at other places reported the presence of a live fetus along with a large intrauterine clot. The emergency transabdominal USG scan done by a convex probe in radiology department revealed a live intrauterine fetus with its normal appearing posteriorly located placenta in the superior uterine cavity and a large heterogeneously hyperechoic lesion with multiple interspersed internal variable-sized anechoic spaces giving snowstorm or bunch of grapes appearance in the inferior uterine cavity (Fig. 1a–c). A suspicious membrane was seen separating them (Fig. 1a). Both ovaries were bulky with multiple hemorrhagic cysts within them. Dedicated and targeted fetal MRI (Fig. 2a–d) including the T2W HASTE sequences in all planes, DWI and Axial T2* images was done to further characterize the etiology confirmed the ultrasound findings. It clearly demonstrated the inter-twin membrane (Fig. 2c)

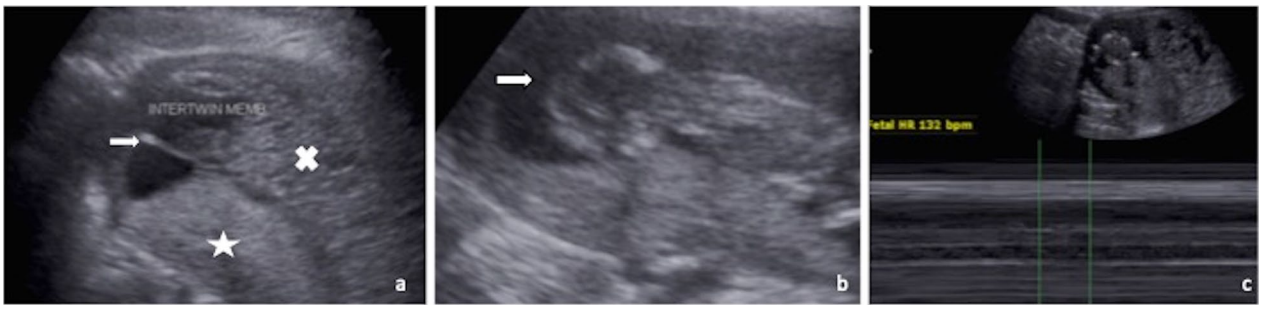

Fig. 1 Emergency 2D USG in longitudinal view **a** shows a possible intertwin echogenic membrane (arrow) with echogenic mass with multiple internal cystic spaces in the inferior aspect (cross) and normal placenta in the superior aspect (star). **b** and **c** reveal a normal fetus along (arrow) with its placenta with normal fetal cardiac activity (M Mode)

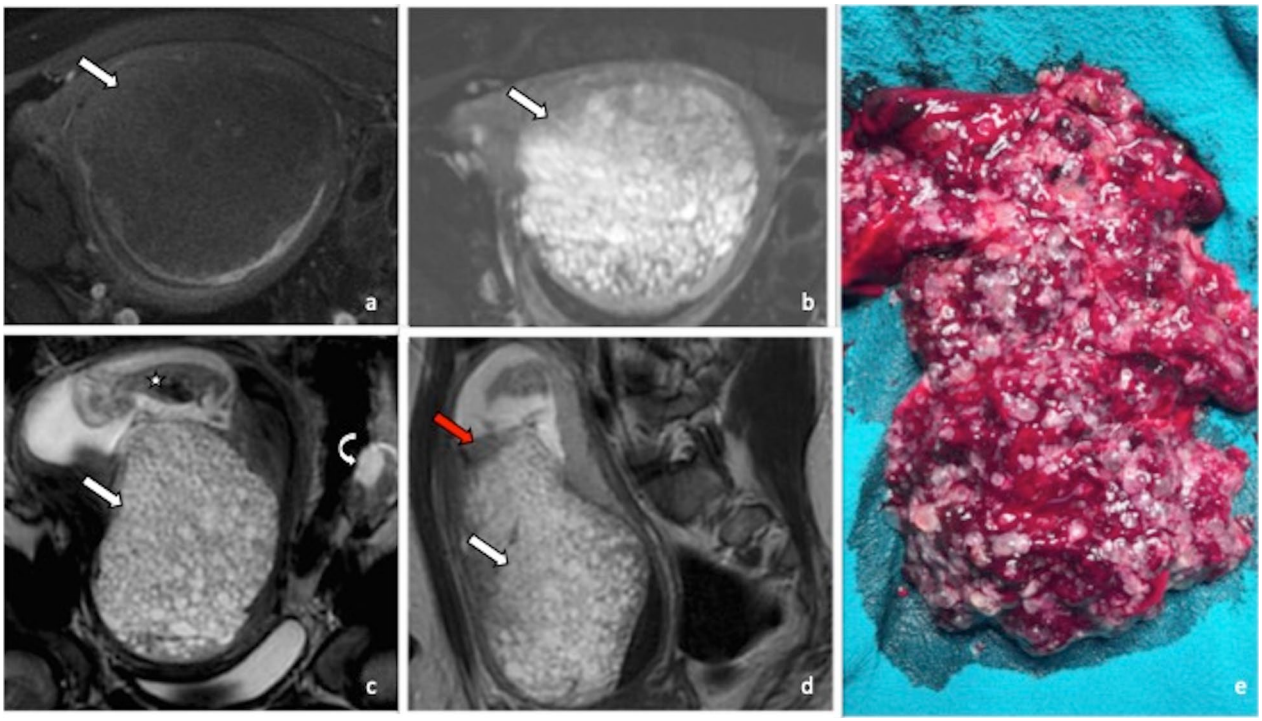

Fig. 2 Axial T1FS **a** and axial T2FS **b** show molar degeneration in the inferior uterine cavity (arrow). Coronal T2FS image **c** shows molar degeneration in the inferior amniotic cavity (arrow) with fetus in the superior amniotic cavity (star) separated by a thin hypointense intertwin membrane and bulky left ovary with hemorrhagic cysts within (curved arrow). Sagittal T2FS **d** image shows molar degeneration in the inferior amniotic cavity and normal placenta in the superior amniotic cavity with fluid hemorrhagic levels in the superior cavity (red arrow). Gross specimen **e** showing bunch of grapes appearance—suggestive of complete hydatidiform mole

suggestive of diamniotic conception, thereby acting as the problem-solving tool. Additionally, sub-chorionic hematoma (Fig. 2d) was also seen in both amniotic sacs. One day prior serum beta-HCG levels were quite high, measuring > 200,000 mIU/ml. The thyroid hormonal profile revealed mild thyrotoxicosis with serum TSH- 0.05 mIU/ml, serum T3 3.45 mIU/ml and serum T4 10.7 mIU/ml. Considering the clinical presentation, ultrasound findings and biochemical profile, a final radiological diagnosis of CHMTF, associated sub-chorionic hemorrhage in both sacs and bilateral ovarian hyperstimulation syndrome was made.

The pregnancy was terminated at 13 weeks four days, considering the patient's worsening clinical condition. The gross specimen (Fig. 2e) revealed a single fully formed fetus and a large mass with multiple grape-like vesicles, which on histopathology was confirmed to be complete hydatidiform mole.

Discussion

Complete hydatidiform mole with a coexisting twin live fetus is a type of dichorionic diamniotic twin pregnancy, in which one of the amniotic sacs contains a complete hydatidiform mole, while the other sac contains a normal fetus with its normal placenta. Complications that may arise in such a pregnancy are due to the molar component, which include heavy vaginal bleeding, hyperemesis gravidarum, hyperthyroidism and pre-eclampsia [3].

Gestational trophoblastic disease (GTD) is a group of pregnancy-related abnormalities which consist of hydatidiform moles and gestational trophoblastic neoplasia (GTN) [5]. Hydatidiform moles are of two types—partial and complete. GTN includes invasive mole, choriocarcinoma, placental site trophoblastic tumor and epithelioid trophoblastic tumor. All the GTDs originate from the trophoblasts which later form the fetal portion of placenta. These are made of the cytotrophoblasts, syncytiotrophoblasts and intermediate trophoblasts. Hydatidiform moles and choriocarcinoma originate from cytotrophoblasts and syncytiotrophoblasts, while placental site trophoblastic tumor and epithelioid trophoblastic tumor arise from intermediate trophoblasts. A complete hydatidiform mole occurs when an empty ovum is fertilized by either one sperm that duplicates its DNA, resulting in a diploid 46,XX karyotype (seen in 90% of cases), or by two different sperms, resulting in a diploid 46,XY karyotype (seen in 10% of cases). On the other hand, a partial hydatidiform mole develops when a normal ovum is fertilized by either two sperms or a single sperm that duplicates itself, resulting in almost always triploid 69,XXX or 69,XXY genotypes [5].

Braga A et al [6] also reported a similar case in a 37-year-old female diagnosed at 10 weeks of gestation where clinico-radiological picture and histopathological examination confirmed the diagnosis of CHMTF as was done in our case. Pregnancy with one CHMTF presents a diagnostic challenge as it is an uncommon occurrence that requires consideration of several possible differentials on ultrasound like partial mole with coexisting live fetus, placental mesenchymal dysplasia and intrauterine clot [3]. It is extremely important to differentiate it from singleton pregnancy with partial mole, and a systematic approach can help in reaching the diagnosis as depicted in Fig. 3. It is because partial mole means the fetus is definitely abnormal and the pregnancy needs to be immediately terminated. On the other hand, in CHMTF, the fetus is normal, and the pregnancy can be continued till delivery [4, 6].

We suggest that USG should be followed by MRI as it completely rules out the above-mentioned differential diagnoses, thus serving as a problem-solving tool. MRI, due to its higher soft tissue resolution and larger field of view, clearly shows the presence of an intertwin membrane, normal appearing placenta of the live fetus separated from complete mole, indicating the presence of two amniotic cavities—which suggests CHMTF. Also, it helps in ruling out myometrial invasion by the mole as well as the differential of placental mesenchymal dysplasia. [4]

Assisted reproductive technique is one of the risk factors associated with this pregnancy, with an approximate frequency of 22% [1, 2]. This is consistent with our case. Other risk factors have not been well documented in the literature.

A CHMTF pregnancy is on average terminated by 16 weeks [1]. It is because the patient develops complications such as heavy vaginal bleeding, hyperemesis, hyperthyroidism, pre-eclampsia and placental abruption, which may necessitate the termination of pregnancy or lead to premature birth. The most common complication of the said condition is the formation of gestational trophoblastic neoplasia, reported to develop in 5–33% of cases and the most dreaded being choriocarcinoma—for which proper follow-up with beta-HCG has to be done [1, 2]. Suksai M et al [7] reported the live birth rate of ~ 37% when such a pregnancy could be continued beyond the first trimester.

Beta-HCG values are in lacs, peaking in early 2nd trimester. In our case, two beta-HCG values were obtained—one as > 200,000 and other as 499.2 obtained 6 days later. Such a discrepancy can be explained by the Hook effect [8], which states that large amounts of beta-HCG (> 100,000) can result in falsely low beta-HCG values in the presently commercially available two-site non-competitive beta-HCG immunometric assays.

The latest point of view on managing such patients is to closely monitor them for any complications, and

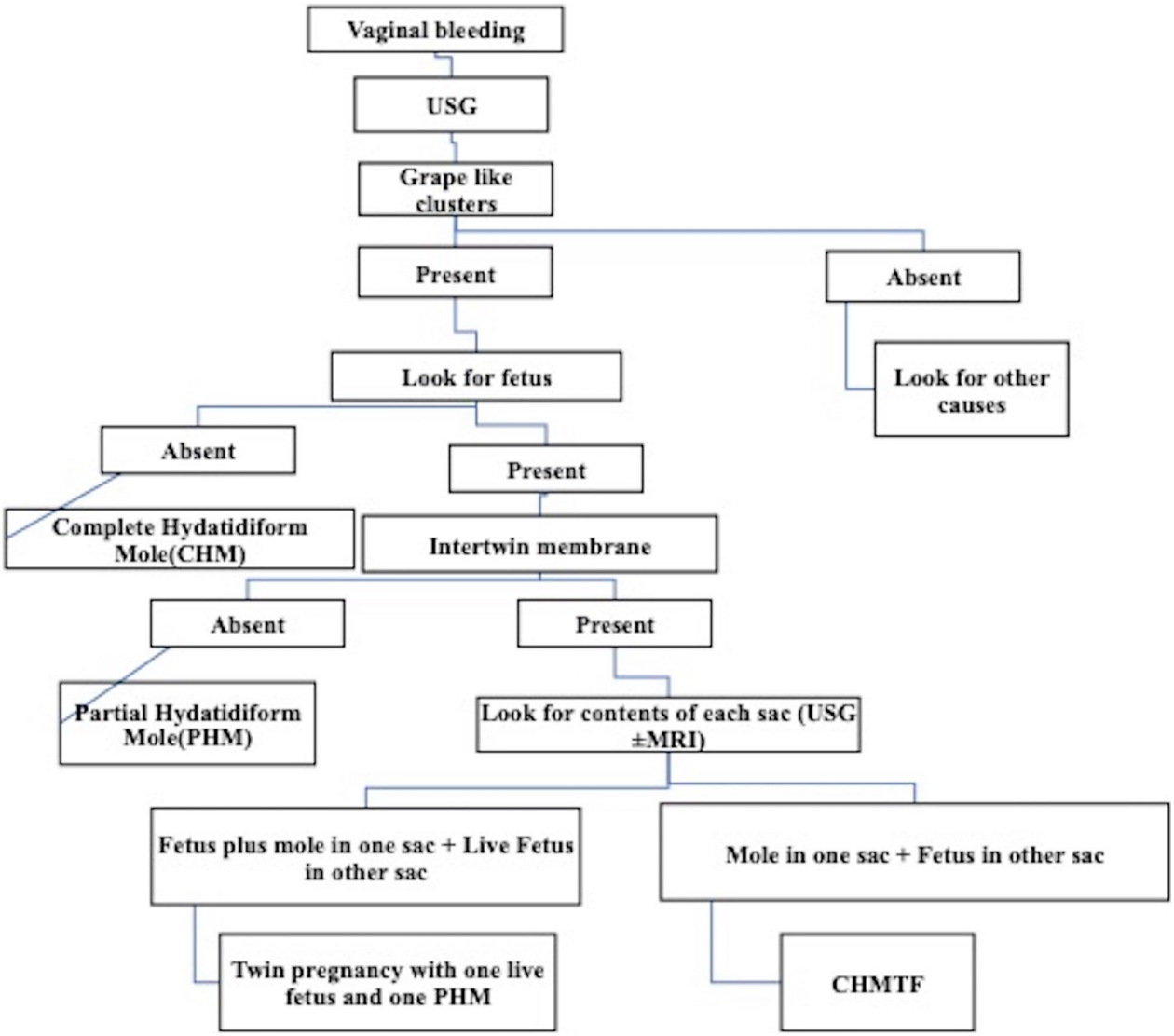

Fig. 3 Diagnostic algorithm for grape-like structures in uterine cavity

only terminate the pregnancy, if necessary, followed by management of the complete hydatidiform mole. This approach is recommended for two reasons. Firstly, many of such patients have conceived using assisted reproductive technologies making the pregnancy as very precious to the patient. Secondly, there is no increased risk of developing persistent GTD whether a pregnancy is terminated or continued to term [6].

In our case, termination of pregnancy was justified at 13 week 4 days due to heavy vaginal bleeding.

Conclusions

A pregnancy involving both a complete mole and a coexisting fetus is a rare occurrence that poses a diagnostic challenge to radiologists due to very close differential diagnoses. USG and more importantly MRI, combining with the clinical manifestations and beta-HCG values, help in its accurate diagnosis.

Abbreviations

CHMTF Complete hydatidiform mole with a coexisting twin live fetus
USG Ultrasonography

MRI Magnetic resonance imaging
GTD Gestational trophoblastic diseases
GTN Gestational trophoblastic neoplasia

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Author contributions
Dr PS and Dr R prepared the initial draft, performed the primary ultrasound and read the initial MRI. Dr JG edited the manuscript and read the MRI images. All authors read and approved the final manuscript.

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Departmental permission was taken for case publication.

Consent for publication
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Competing interests
The authors declare that they have no competing interests.

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