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Case Report

Dentistry Section

Dual Outcomes: Live Pregnancy Amidst Partial Hydatidiform Mole

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ABSTRACT

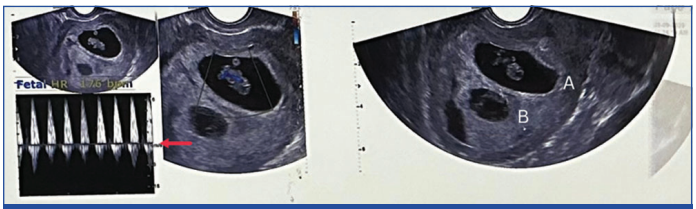
Gestational Trophoblastic Disease (GTD) is a group of cancers that can metastasise to distant sites. Molar pregnancy, a rare and challenging condition, is characterised by Complete Hydatidiform Moles (CHM) occurring when one sperm fertilises an empty ovum, resulting in the absence of maternal chromosomes. Partial Hydatidiform Mole (PHM) is triploid and is often detected early in gestation. In one out of every 20,000 to 100,000 pregnancies, along with a CHM or PHM, a normal co-twin may emerge. Here, we present a similar case of a young primigravida presenting with a live foetus and a concomitant partial mole. In this case, a 22-year-old primigravida female presented with lower abdominal pain, nausea, and vomiting for the last two weeks. She was advised to undergo an antenatal scan at 24 weeks of gestation, which showed a dichorionic diamniotic pregnancy with a single live foetus and a molar placenta. Later, she was put on conservative management and followed up with subsequent delivery of a premature live baby and expulsion of a molar placenta. Twin pregnancies with a normal foetus and a CHM are rare but can lead to live deliveries with no appreciable rise in the probability of malignant transformation of CHM. It is critical to differentiate these pregnancies from other disorders such as Placental Mesenchymal Dysplasia (PMD) and PHM, which also involve a foetus and a cystic placenta. Suction dilation and curettage are the preferred treatments for women with a hydatidiform mole to maintain fertility.

Keywords: Grape-like vesicles, Hydatidiform mole, Prematurity, Twin pregnancy

CASE REPORT

A 22-year-old primigravida female presented with complaints of lower abdominal pain with nausea and vomiting for the last two weeks. On general physical examination, the heart rate, blood pressure, temperature, and other vitals were within normal limits. On per abdominal examination, fundus corresponded to 20 weeks, and foetal heart rate was auscultated.

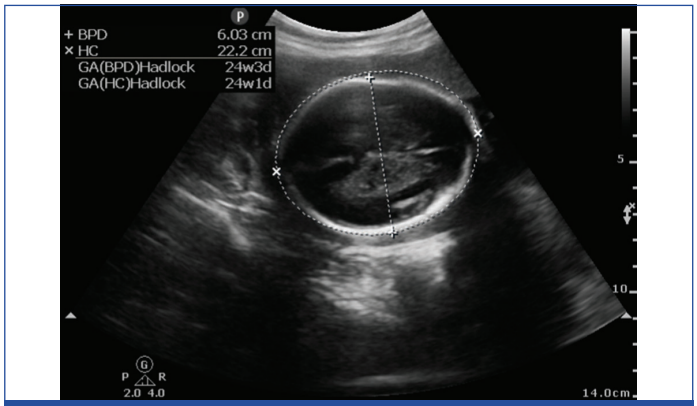
Clinically, she was suspected of having either Intrauterine Growth Restriction (IUGR) or low amniotic fluid volume. She was then referred to our department for an antenatal scan at 24 weeks of gestation. She had an early dating scan done at eight weeks of pregnancy, which showed a dichorionic diamniotic pregnancy with a single live foetus corresponding to eight weeks one day and a blighted ovum corresponding to six weeks four days [Table/Fig-1].



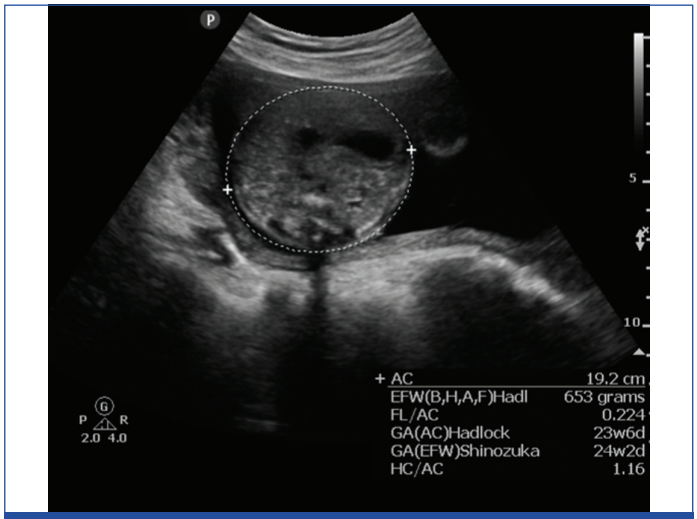
[Table/Fig-1]: Transvaginal (TVS) grey scale B-mode image showing two gestational sacs with one of the sacs (labelled A) showing embryo and yolk sac within, with foetal heart rate of 176 beats per minute (marked by red arrow). The other sac (labelled B) appears irregular and empty, suggestive of a blighted ovum.

Since this was an early dating scan, she was advised to have a Nuchal Translucency (NT) scan later at 11 to 13 weeks, which she missed due to unforeseeable circumstances, and came directly for a level II anomaly scan at our tertiary centre. On Ultrasound (US), there was evidence of a single live intrauterine foetus corresponding to 24 weeks and one day [Table/Fig-2-4].

Furthermore, the placenta was anterior and there was evidence of a heterogeneous lesion at its lateral end containing multiple anechoic cystic spaces within, measuring approximately 11×5 cm [Table/Fig-5].

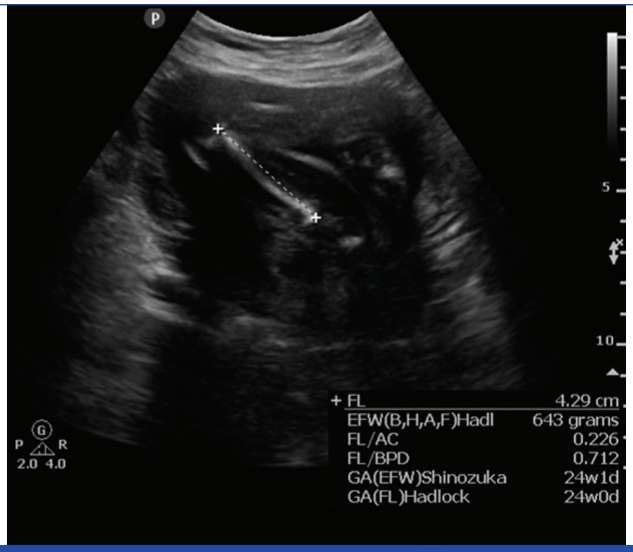


[Table/Fig-2]: USG B-mode image showing Biparietal Diameter (BPD) and Head Circumference (HC) of the live foetus

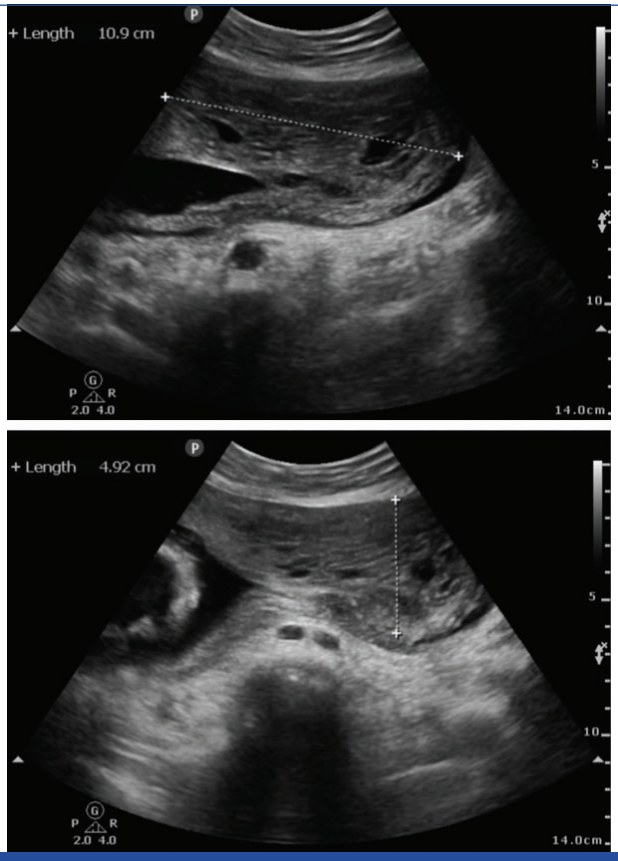


[Table/Fig-3]: USG B-mode image showing Abdominal Circumference (AC) with anechoic (black) stomach bubble.

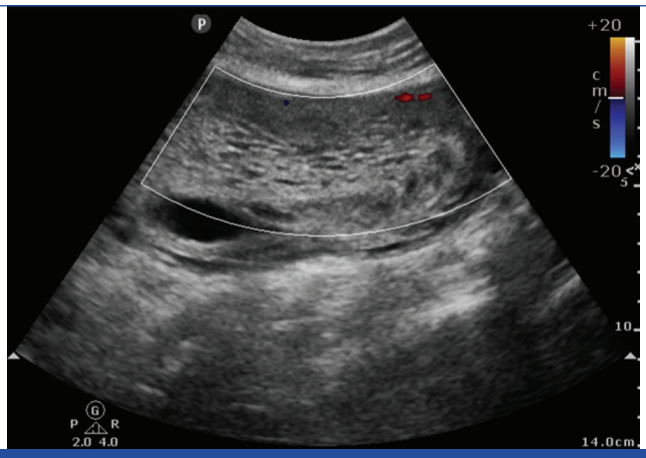
This lesion did not uptake vascularity as revealed using Doppler [Table/Fig-6].



[Table/Fig-4]: USG B-mode image showing Femur Length (FL) as measured with a dashed line.



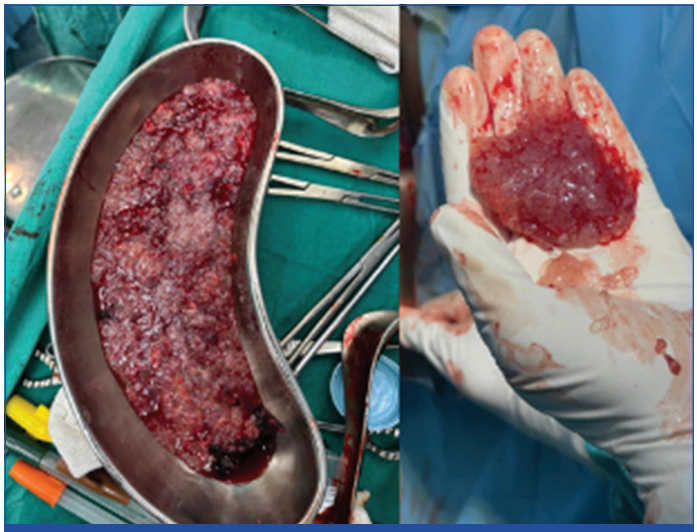
[Table/Fig-5]: USG B-mode image showing a heterogeneous mass in the lateral end of the placenta with multiple variable-sized cystic lesions.



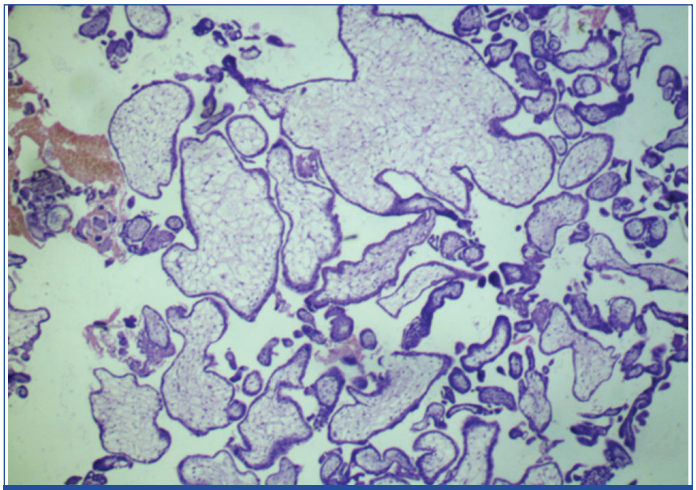
[Table/Fig-6]: Colour Doppler image showing no uptake of colour within the lesion, which can be correlated with the colour scale present in the top right corner.

Radiological differentials included PMD, which has progressive dilatation of chronic arteries and veins that become aneurysmal.

Laboratory investigation showed lymphocytosis with raised Erythrocyte Sedimentation Rate (ESR) and beta-Human Chorionic Gonadotrophin hormone (b-hCG) levels measured 2,46,441 mIU/mL. The patient was admitted and put on conservative management, including fluid resuscitation, blood transfusion, and steroids for lung maturation, following which she delivered at 30 weeks and three days of gestation, a live male, premature, and very low birth weight baby weighing 1.2 kg. The placenta was expelled consecutively with a large cluster of grape-like mass measuring about 600 grams [Table/Fig-7], with its histopathological image in [Table/Fig-8] confirming the diagnosis of PHM.



[Table/Fig-7]: Gross specimen of the expelled conceptus products.



[Table/Fig-8]: Histopathological image under Haematoxylin and Eosin (H&E) stain in 40x magnification showing oedematous and hydropic villi.

Follow-up b-hCG levels were 1,00,57 and 40,657 mIU/mL on postnatal day 2 and 4, respectively. She was discharged on day 7 when suction and evacuation were done and b-hCG levels were 300 mIU/mL.

DISCUSSION

The term "GTD" describes a broad category of tumours that display a variety of biological activities and the capacity to spread to distant places. The range of benign and malignant conditions known as GTD includes choriocarcinomas, Placental Site Trophoblastic Tumours (PSTT), Epithelioid Trophoblastic Tumours (ETT), partial and CHMs, and invasive moles [1]. The final four are known as Gestational Trophoblastic Neoplasia (GTN); if left untreated, they can all spread and be lethal.

According to reports, the prevalence of molar pregnancy varies greatly worldwide, with North America and Europe reporting 0.5

to 1 per 1000 pregnancies, Japan and China reporting 1 to 2 per 1000 pregnancies, and Indonesia, India, and Turkey reporting 12 per 1000 pregnancies [2].

When one sperm fertilises a missing maternal chromosome in an empty ovum, CHM happens 90% of the time. After that, the sperm replicates its own DNA to create the "complete" 46-chromosome set. PHMs are almost always triploid, with the genotypes 69, XXX,69, XXY, or 69, XYY, when a healthy ovum is fertilised by two sperm [3] or by a single self-replicating sperm. Patients with CHM are often diagnosed asymptomatically and are identified early in pregnancy due to the common use of US and β-hCG tests [4]. Vaginal bleeding, which typically occurs between weeks 6 and 16 of pregnancy (46%), large-for-date uterine size (24%), and hyperemesis (14%) are common presenting symptoms [5].

Histologic examination of curettage specimens following an incomplete or missed abortion is typically used to diagnose PHM patients, who are less likely to receive a diagnosis before uterine evacuation [3]. In one out of every 20,000 to 100,000 pregnancies, along with a CHM or PHM, a normal co-twin may emerge [6]. Here, we present a similar case of a young primigravida presenting with a live foetus and a concomitant partial mole.

Most often, molar pregnancies take place inside the uterus. Nevertheless, an uncommon circumstance could arise that could make the clinical picture more complicated: twin pregnancies with CHM and a normal foetus. Although there is a large chance of spontaneous abortion in CHM and healthy co-twin pregnancies, 40% of these pregnancies end in live births, where there is no appreciable rise in the likelihood of transformation into malignancy of CHM [6].

It is critical to distinguish twin pregnancies with a CHM and a healthy foetus from other disorders, such as PMD and PHM, which also involve a foetus with a cystic placenta. In PHM, an abnormal or dead foetus is observed alongside cystic areas within the placenta. Therefore, a CHM with a normal foetus should raise suspicions of a twin pregnancy if there is an aberrant cystic placenta along with a normal, alive foetus that is the right size for its gestational age [7].

There are three types of molar pregnancies in which a living foetus co-exists [8]. A twin pregnancy, in which one foetus is healthy and the other is a full mole, is the most prevalent variation. Healthy single foetuses with partial molar placentas [8], like our patient, are the rarest variety.

As highlighted by [Table/Fig-9] [9-13], Singh M et al., used MRI as an imaging modality to look for possible local invasion of the molar placenta, as PHM can further complicate hydatidiform disease, which can turn malignant and metastasise [11]. Thus, modern imaging tools such as MRI can provide us with essential details such as progression towards malignancy and should be conducted in patients who have very high values of b-hCG, pointing towards the possibility of malignant changes.

Twin pregnancies in which the placenta and foetus are healthy but have a partial mole are less common. The "twin peak" sign, which confirms the existence of a dichorionic twin gestation by separating from the molar pregnancy's usual twin sac through the formation of a triangle echogenic structure formed by chorionic tissue extending into the intertwin membrane, is another useful US sign that may be observed in this circumstance [14].

Case (author, year)	Patient details	Radiologic findings	Modality	Other details
Rasuli B, 2020 [9]	30-year-old primigravida	Enlarged placenta with internal cystic spaces; "snowstorm" appearance; small nonviable embryo displaced by molar tissue	Ultrasound, Doppler	Mild peripheral vascularity noted
Maher MA et al., 2017 [10]	30-year-old female	Cystic intrauterine mass with snowstorm appearance; compressed amniotic sac	Ultrasound, Color Doppler	β-hCG ~225,000 mIU/mL; confirmed on histopathology
Singh M, et al., 2018 [11]	Not specified	Cystic placenta with hypervascularity; MRI: heterogeneous placental lesion with early enhancement and thinning	US, Doppler, MRI	MRI indicated possible local invasion
Sharma R et al., 2019 [12]	29-year-old at 14 weeks	Thickened placenta with honeycomb pattern; foetus present; no Doppler flow	Ultrasound, Doppler	Elevated β-hCG; confirmed PHM after termination
Almasi A, 2016 [13]	Low-risk primigravida	Thickened placenta, small gestational sac, no cysts	Ultrasound	Confirmed on curettage and histology

[Table/Fig-9]: Summarising previous case reports and their imaging findings [9-13].

It can be difficult to distinguish twin pregnancies with a normal foetus from PMD and a CHM because both disorders are typified by an enlarged placenta that appears cystic at US. Kuwata T et al., have referred to this enhanced vascularity as the "stained-glass" indication [15]. On the other hand, hydropic swelling of villi is the cause of the cysts in molar pregnancy, which do not exhibit blood flow on colour Doppler pictures [15].

Foetal survival when a single healthy foetus has a partial mole is affected by the foetus's normal karyotype, the placenta's smaller molar, the extent of molar degeneration, the lack of anaemia, and any maternal diseases that may be present at the same time, such as vaginal haemorrhage, thyrotoxicosis, or pre-eclampsia [16].

Twin pregnancies with CHMs are linked to an increased risk of maternal complications, including antepartum haemorrhage, severe early-onset pre-eclampsia or eclampsia, placenta previa, preterm premature rupture of membranes, and preterm labour [17]. A case of a partial mole that resulted in a healthy, viable foetus at full term, with 800 gm of placental tissue in which one-third had molar tissue, was described by Wang Y et al., [18]. The termination of pregnancy in women with HM is primarily influenced by gestational duration and disease status. In early pregnancy, termination is typically achieved through complete uterine curettage. However, there remains significant debate concerning the use of intra-amniotic Rivanol injection, intravenous oxytocin, and caesarean section in the second trimester [18]. Caesarean section is advised for women with hydatidiform mole pregnancies due to the elevated risk of pulmonary embolism, which arises from recurrent uterine contractions that may facilitate the displacement of hydatidiform mole tissue into the abdominal cavity. US-Guided (USG) suction dilation and curettage can be carried out to prevent uterine perforation, and are the preferred treatments for patients with hydatidiform mole who want to maintain fertility [1].

Because of the extremely small number of occurrences, managing pregnancy in the event of PHM while allowing for the presence of a normal foetus can be quite difficult, particularly when twins are involved. A multidisciplinary team comprising an obstetrician, maternal foetal medicine specialist, gynaecologic oncologist, and neonatologist should be included in the patient's treatment when the woman chooses to proceed with the pregnancy, as recommended by a prior review on PHM co-existing with a living foetus [19]. Additionally, to ascertain the foetal karyotype, an amniocentesis must be made available at 16 weeks. Serial scans for the high risk of foetal growth restriction and oligohydramnios must be scheduled at least every two to three weeks in conjunction with US to rule out congenital abnormalities in the living foetus [19].

CONCLUSION(S)

Prenatal consultation should cover all maternal and foetal risks, particularly the potential need for chemotherapy or even a hysterectomy, in cases of partial molar pregnancy with a co-existing live foetus. This is in addition to chromosomal evaluation and a thorough US examination to rule out foetal abnormalities before deciding to postpone intervention. Close monitoring and assessment of the patient's status during the prenatal and postnatal phases are equally essential.

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