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Case Report

Sad fetus syndrome: a rare case report

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ABSTRACT

Gestational trophoblastic disease (GTD) is a condition that develops from abnormal proliferation of placental trophoblasts. A rare division of GTD is a partial mole with a co-existing live fetus-a condition also referred to as "sad fetus syndrome" We report a rare case of a 24-year-old, primigravida with 26 weeks of gestational age with single live intrauterine gestation coexisting with partial molar pregnancy who had a spontaneous preterm delivery at 27 weeks. The post-natal period was uneventful and patient was followed up till βhcg normalised. The diagnostic challenges and dilemma associated with the management of molar pregnancies with an apparently normal fetus, especially in the second trimester, remains challenging.

Keywords: Molar pregnancy, GTD, Partial mole

INTRODUCTION

Molar pregnancies belong to a group of diseases classified as Gestational trophoblastic disease (GTD), which result from an altered fertilization. GTD is a condition that develops from abnormal proliferation of placental trophoblasts. The cases with GTD concurrent with normal intrauterine pregnancy have been reported to being as rare as 2.5 to 5 percent of molar pregnancies. Molar pregnancy have been divided into complete or partial hydatidiform moles on the basis of distinctive histopathological features and genetic abnormalities.[1,2]

Complete moles are typically diploid and tend to cause higher levels of the human chorionic gonadotropin (hCG). In complete moles, the karyotype is 46, XX 90% of the time and 46, XY 10% of the time. It arises when an enucleated egg is fertilized either by two sperms or by a haploid sperm that then duplicates and therefore, only paternal DNA is expressed.

On the contrary partial moles arise when a normal sperm subsequently fertilizes haploid ovum duplicates and or when two sperms fertilize a haploid ovum. In partial moles, both maternal and paternal DNA is expressed and is triploid in 90% cases with the karyotype being either 69, XXX or 69, XXY.[3]

They are usually considered the non-invasive form of GTD.

The clinical presentation of GTD is classically seen with presence of elevated beta-human chorionic gonadotropin (β-hCG) levels and the passage of grape-like vesicles per vaginum. GTD lesion have distinct histological feature of presence of hydropic villi on examination of the specimen and depending on the same can be divided into benign or malignant lesions.[4]

A division of GTD is a partial mole with a co-existing live fetus-a condition also referred to as "sad fetus syndrome" a very rare condition, occurring in 0.005 to 0.01% of all pregnancies.[4] We reported a case of a patient with sad fetus syndrome who presented to our tertiary care hospital.

CASE REPORT

A 24-year-old woman with married life of 8 months, primigravida with 6 months of amenorrhea booked elsewhere was referred in view of obstetric scan

abnormality of aneuploidy with partial mole. It was a spontaneous conception. She had no other obstetric complaints.

She was admitted for further evaluation and management. No family history of similar condition or congenital anomalies. Her vitals on admission were stable and general physical examination did not reveal anything significant.

On abdominal examination fundal height was 28weeks which was more than the period of gestation of 26 weeks. All routine investigations were sent and said to be normal. β HCG was sent and was 152735 IU/L.

Her 13 weeks scan showed a single intrauterine gestation of 13 weeks 4 days ±1 weeks with nuchal translucency of 1.3mm and disparity of one week. Placental imaging revealed multiple cystic areas within, with no colour flow seen on doppler examination. On admission she was sent for a repeat scan which revealed single live intrauterine gestation of 25 weeks 5 days with growth restricted fetus-early onset FGR and no obvious fetal anomalies noted. Placental abnormality was noted with mild placentomegaly noted with small part of placenta showing cystic lesion. The differential diagnosis was given to be as Placental mesenchymal dysplasia and partial molar pregnancy. While admitted, as we were tackling the dilemma about further action for this pregnancy, she set into spontaneous labor after onset of preterm premature rupture of membranes (PPROM) and delivered a preterm baby of birth weight 780 gm with no gross congenital anomalies. Gross examination of placenta revealed multiple cysts within and specks of calcification and was sent for histopathology and reports confirmed partial molar pregnancy. Postnatal period was uneventful.

She was followed up with serial βhcg measurements which was 91IU/L at 2 weeks and undetectable by 6 weeks.

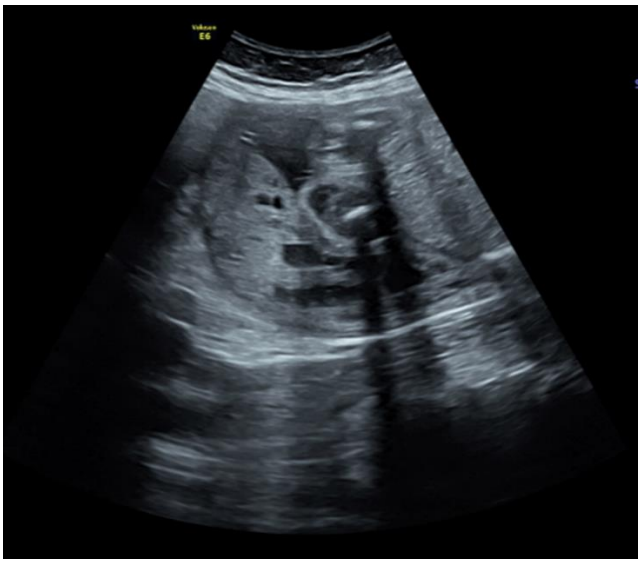

Figure 1: USG finding.

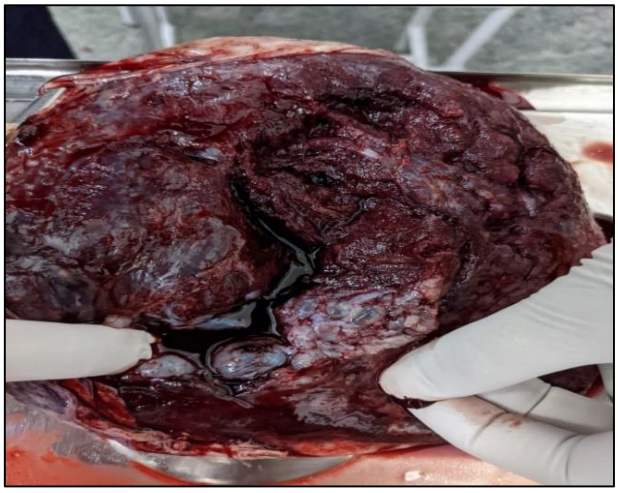

Figure 2: Gross picture of placenta.

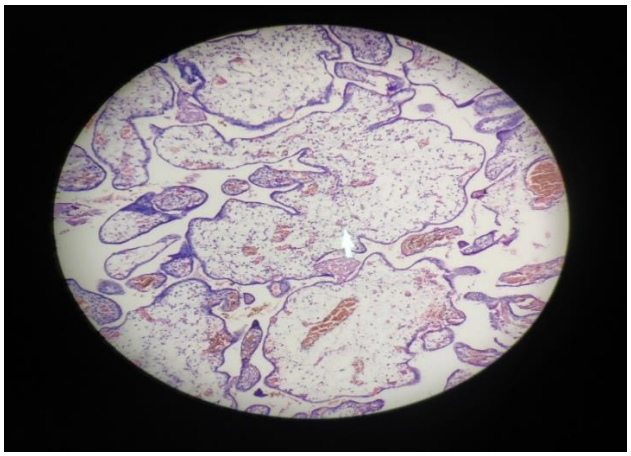

Figure 3: Histopathology of the specimen.

DISCUSSION

Molar pregnancy with coexisting live fetus is a rare occurrence and hence presents with its own challenges in management. The common clinical presentations in this rare condition include bleeding per vaginum, anemia, hyperemesis gravidarum, hypertension, thyrotoxicosis, and uterine size disproportionate to uterine age.[4]

Pregnancy with a partial mole and a single normal fetus evolving to a viable fetus is seen in about 25% of cases and only about 44 cases of partial molar pregnancies with a live fetus have been reported out of which only 36 have progressed up till the second trimester.[5,6]

Ultrasonography is the first choice for screening. The sonographic 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.[7]

Most of the live pregnancies with coexisting molar pregnancy are usually spontaneously lost in the first

trimester due to persistent haemorrhage and high incidences of pre-existing pre-eclampsia. Very few pregnancies continue upto late second trimester as in our case that has been reported. Even after crossing the period of viability there are chances of early termination and poor perinatal outcome owing to the prematurity and associated aneuploidy.

Only about 40% of women who choose to continue their pregnancies have live babies and most delivered beyond 32 weeks' gestation.[8]

Post termination of pregnancy the threat does not end as the incidence of invasive mole after hydatidiform mole has been reported as fluctuating between 5.8 and 3.1% and this poses dangers to the woman in the future and requires adherent follow up.[9]

Since the prognosis for pregnancy outcome is different in each case, a tailored management plan becomes crucial for such rare cases.

CONCLUSION

Molar pregnancy with a coexistent live fetus is a rare and challenging condition. The clinician in concordance with a good sonology report with additional karyotyping preferably by chorionic villous biopsy, if a live fetus is present, confirms the diagnosis and helps in the management of the pregnancy in the later trimesters. Diagnosis and management of molar pregnancies with an apparently normal fetus, especially in the second trimester, remains challenging.

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