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SAD FOETUS SYNDROME : A PARADOX OF LIFE CONSISTING LIVE FOETUS AND PARTIAL HYDATIDIFORM MOLE

Obstetrics & Gynaecology

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ABSTRACT

It is very rare to have a molar pregnancy in conjugation with a viable foetus at the same time inside the womb. Clinical presentation is bleeding per vagina and uterine overdistension as per the expected size of gravid uterus. Hydatidiform mole with a coexisting live foetus in most cases is complete mole. Partial molar pregnancy with foetus is rare and is almost always ends in miscarriage due to triploid. Cases of partial mole with Co-existing live foetus is known as Sad foetal syndrome. Some of this rare cases we encountered in our clinical practice, that we are sharing here. Molar pregnancy along with a live foetus is a rare and challenging condition. Clinical suspicion along with a good ultrasound report and additional karyotyping preferably by chorionic villous biopsy, if a live foetus is Present, confirms the diagnosis and also helps in the proper management of the pregnancy in the later trimesters. Diagnosis and management of molar pregnancies with an apparently normal foetus, especially in the second trimester, remain a challenging task for budding Obstetrician.

KEYWORDS

Sad Foetus Syndrome, Hydatidiform mole, miscarriage, karyotyping, triploid, molar pregnancy

INTRODUCTION

It is a very sad situation when a foetus co-exists with a molar pregnancy or choriocarcinoma. It has a very bad prognosis, this is why we call such cases as: " Sad Foetus Syndrome".

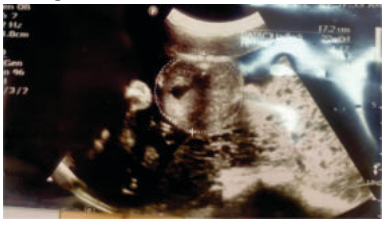
Hydatidiform mole belongs to a group of gestational trophoblastic disease which is characterised by abnormal proliferation of trophoblastic tissue filling up entire uterus. The prevalence is 1 to 2 : 1000 pregnancies in Western countries but 1: 500 pregnancies in Asia countries and 1: 250 pregnancies in Philippines[1]. Gestational trophoblastic disease (GTD) are classified into molar pregnancy which includes a)complete mole and b)partial mole and gestational trophoblastic neoplasia which includes invasive mole, chorio-carcinoma, placental site trophoblastic tumour and epithelioid trophoblastic tumour . GTD can be divided into benign or malignant lesion.

Molar pregnancy are categorised as either complete H mole or partial H mole based on gross morphology, histology and karyotyping[2,3]. Complete H mole is characterised by hydropic degeneration and swelling of villous stroma, proliferation of both cytotrophoblast and syncytiotrophoblast, trophoblastic scalloping absent, inclusion bodies are present and chorionic villi are avascular and foetal tissue absent. Complete mole are mostly with 46 XX karyotype 90% cases and 46 XY in 10% cases. In this fertilization of an empty ovum is either by a haploid sperm duplicating its chromosome or by 2 different sperms. Only one paternal DNA is expressed in this situation. While in partial mole, foetal tissue are seen, chorionic villi are present and vascularity is present in chorionic villi, hydropic degeneration is localised, trophoblastic scalloping are present and inclusion bodies are also present. Partial mole are generally triploid with 69XXY karyotype which results from generally fertilisation of ovum with 2 sperm Or duplication of normal sperm. In this both maternal and paternal DNA is expressed and triploid in 90% cases with 69 XXX or 69 XXY.[4]

These are usually non-invasive form of GTD. Clinical presentations are elevated beta HCG level and bleeding along with presence of grape like vesicles per vaginum.

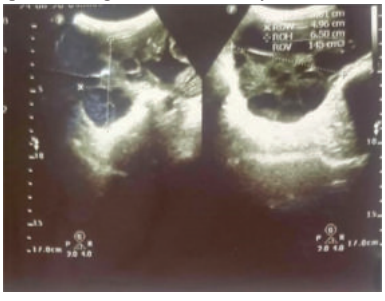
Case :1

18 year old primigravida came to labour room emergency with complain of bleeding per vagina and pain in lower abdomen. On examination, the patient was conscious, pulse rate : 102 beats/min., BP : 90 / 60 mm Hg, SPO_2 : 90 on room air. On per abdominal examination, uterus was 26 weeks size that is larger than the period of gestation and foetal heart rate was absent . On pelvic examination, external OS was fully dilated . On blood examination, Hb : 6.2 gm/dl, WBC : 12.82 x 10^3/l, platelet count : 172 x 10^3/l, her liver function test and renal function test were within normal limit. She was carrying an ultrasound report of 2 days back showing single live intrauterine foetus of 21 week 6 days in breech presentation, placenta was upper posterior, an intra uterine mass of size 185 x 80 mm with a multiple tiny cystic structure that resembles a snow storm appearance or bunch of grapes noted in the lower posterior uterine cavity, the mass is completely covering the internal OS and filled the posterior or lower uterine cavity, this mass is separated from the placenta.



Ultrasound Report Showing Multiple Tiny Cystic Structure That Resembles A Snow Storm Appearance.

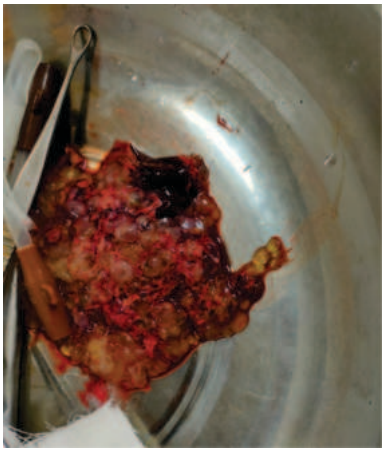
Her previous USG report of 18 weeks 5 days showed lower posterior placenta touching the internal OS and various cystic lesion and hematoma in placenta, right sided adnexal cyst of size 64 x 62 mm size.



Ultrasound Report Showing Theca Luteal Cyst

She delivered a dead foetus along with placenta and abundant of grape like tissue spontaneously. She was transfused with 2 unit packed RBC. Tissue sent for histological examination. Histopathological examination reports showed was a partial mole. The patient was

followed up with beta HCG reports weekly. Pre-expulsion hCG was 606889 mIU/ml, 4 weeks after the expulsion hCG level was 486 mIU/ml. But after 6 weeks it became undetectable.

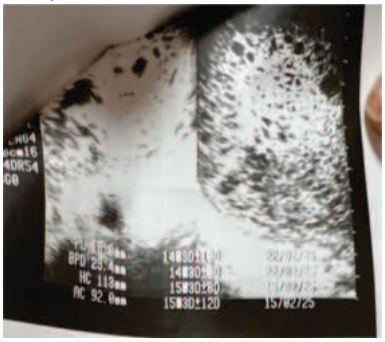


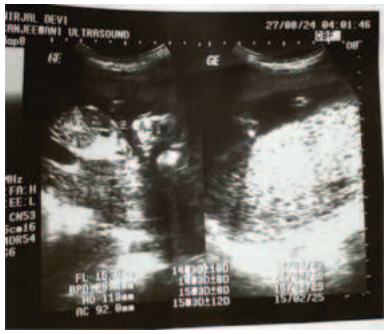
Picture Showing Multiple Grape Like Tissue

Picture Showing Dead Foetus

Case : 2

A 19 year old primi-gravida came to emergency labour room of DMCH with 4 month pregnancy, having lower abdominal pain and vaginal bleeding since one day. She doesn't remember her last menstrual period (LMP). This was an unplanned pregnancy after 8 month of marriage. The pregnancy was confirmed by urinary pregnancy test (UPT) at home. Her previous menstrual cycle were normal of 4 to 5 days and occurring at 28 to 30 days interval. Her past medical and surgical history was unremarkable. On physical examination, she was conscious cooperative but she was pale, her pulse was 122 beats/min, BP was 90/50 mm Hg, abdomen non tender with a 28 to 30 weeks fundal height. Speculum examination revealed active bleeding and on per vaginal examination OS was open introducing only one finger and 2 fists of clots was taken out. She was carrying an ultrasound report one week back which was showing bulky uterus with hydatidiform mole with one live foetus and gestation age was 13 weeks 6 days.

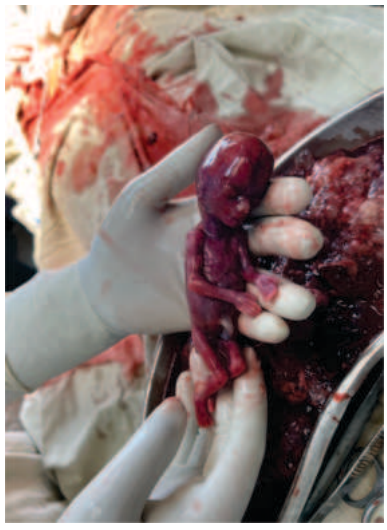




Ultrasound Plate Showing Snowstorm Appearance

Also having another ultrasound report of one day back which shows bulky uterus with one hydatidiform mole and another single fetus of 15 weeks 1 day, placenta was upper anterior and liquor was adequate and foetal heart rate was 156 beats/min. She was having blood investigation which shows Hb : 6.3 gm/dl, WBC count 13,000 mm^3 and platelet 1,35,000 per l, blood group was O positive. Liver function test and kidney function test were within normal limit .As OS was admitting only one finger and she was bleeding profusely, emergency LSCS was done. Foetus was around 14-16weeks along with grape like structure and placenta was taken out. Post operative period was uneventful. She was transfused with 4 unit PRBC and she was followed up with hCG report weekly and after 6 weeks her hCG report were within normal limit. Tissue sent for histological examination. Histopathological examination reports showed was a partial mole.

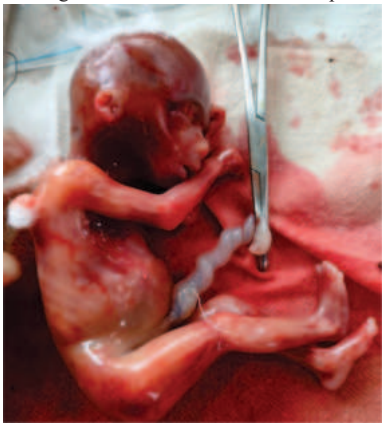
Picture Showing Foetus Along With Grape Like Vesicles And Placental Tissue



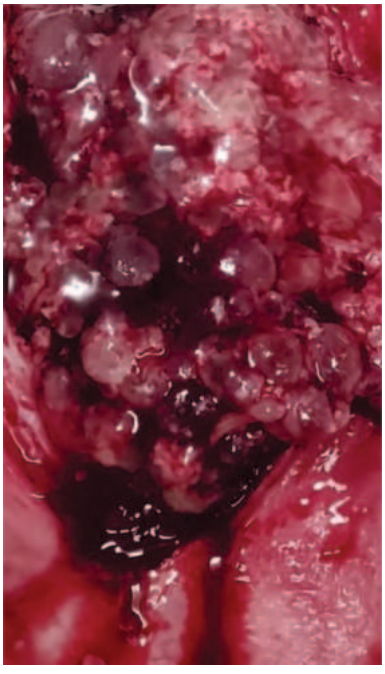
Picture Showing Dead Foetus

Case:3

A 24 years female, primigravida came to the emergency with complain of with 5 months pregnancy and vaginal spotting. This was an unplanned pregnancy and she doesn't have any ante-natal check-up till now. On examination, patient was conscious and oriented, having tachycardia with lower range of blood pressure. She was looking pale. On per-abdominal examination, uterus was 20-22 weeks size. On perineal examination, active bleeding was present. On per-vaginal examination OS was 8-9 cm dilated, cervix was fully effaced. Grapes like vesicle was seen on fingertip. Patient spontaneously expelled foetus along with multiple grapes like vesicles. Foetus died immediately after the birth. Under anaesthesia curettage was done. All blood investigation along with hCG sent. Patient was resuscitated and blood was transfused. Post operative period was uneventful .Patient was followed up with beta hCG . Patient beta HCG before expulsion was 705009 mIU/ml, 48 hrs after the expulsion hCG level was 6500mIU/ml. But patient was loss to follow up after that.. Tissue that was sent for histological examination showed was a partial mole.



Picture Showing Dead Foetus



Picture Showing Grape Like Vesicles

DISCUSSION

Molar pregnancy along with live Foetus is known as sad foetus syndrome. It is a very rare phenomena and it incidence is 0.005% to 0.01% of all pregnancies[5]. There are 3 varieties in which molar pregnancy coexist with live fetus[6] .The most common variant includes

1) Twin pregnancy with one being a live foetus and another being a complete mole,
2) Twin pregnancy with a healthy foetus and a partial mole and
3) Rarest variant is healthy single foetus with a partial mole[6].

Twin pregnancy with one foetus and another coexisting mole was previously uncommon but due to increase in ovulation induction, its incidence is increasing progressively. Ultrasonography helps in diagnosis. Ultrasonographic appearance of H mole has been described as an echogenic tissue with multicystic area in the first trimester and as snow storm appearance in the second trimester. And one can see a molar placenta which is significantly enlarged along with the cystic area . Although complete hydatidiform moles can be easily diagnosed using routine ultrasound assessments early in pregnancy by snow-storm appearance of the placenta; Partial H. mole may mimic missed or incomplete abortion. It is associated with higher risk of hyperemesis gravidarum(28.9%), bleeding per vagina(90%), enlarged uterus, anaemia(51.1%), pregnancy associated hypertension (12.2%) and thyrotoxicosis (3%)[7]. Type of molar pregnancy is diagnosed on placental Histology. The beta HCG titre return back to normal in most cases. Generally due to bleeding termination occurs before 20 weeks of gestation, very few pregnancy continue up to late second trimester. foetal survival depends on various factors such as normal karyotype of the foetus, smaller molar placenta ,rate of molar degeneration, the absence of anaemia and coexisting maternal complication such as pre-eclampsia, thyrotoxicosis, vaginal bleeding ,if the size of molar placenta is big it is a poor prognosis. Amniocentesis remains the diagnostic tool. It can be done for twin pregnancy associated with molar pregnancy mostly for complete mole because partial mole pregnancy has higher chances of abnormal malformation for foetus with defective karyotyping. Only about 40% of women choose to continue their pregnancy and have live babies and most delivered beyond 32 weeks. There is a risk of invasive mole and poses danger for the female in future. Parveen et al. reported a case of partial mole with a live foetus with term gestation that resulted in a live, healthy fetus[8].

There are 2 main differential diagnosis first is placental mesenchymal dysplasia (PMD) and twin pregnancy.

PMD is a rare placental vascular anomaly with an ultrasound appearance of a large placenta with grape like vesicles mimicking a molar pregnancy .The serum beta HCG level in PMD is normal or slightly elevated. PMD has a poor prognosis associated with foetal growth restriction, absence of foetal development, foetal or neonatal death. This is secondary to foetal vascular obstruction causing a prolonged severe foetal hypoxia[9]. So It should be differentiated from mesenchymal dysplasia by morphologic features and immuno-histochemistry.[10]

Histopathological finding of a partial mole are focal swelling of chorionic villi and trophoblastic hyperplasia. moreover scalloping of chorionic villi and trophoblastic stromal inclusions is present. The use of P 57 immunostaining improves diagnostic accuracy for complete H.mole which are almost always P 57 negative. Very rarely do complete mole show aberrant expression and this is secondary to the retention of maternal copy of chromosome 11. Conversely, in rare occasion P 57 immunostaining may show negative staining in partial mole when there is a loss of maternal chromosome. In a systemic review and meta-analysis by Marie et al comparing the accuracy of P 53 immunostaining for the diagnosis complete hydatidiform mole, it is found out that out that occasional partial h. moles can demonstrate negative staining secondary to the loss of maternal chromosome[11]. A risk of persistent gestational trophoblastic disease has been observed in some studies. In one series, 12 of 22 (55%) patients with a complete mole and a coexisting viable foetus developed persistent gestational trophoblastic disease and required chemotherapy, five of these patients developed metastatic disease requiring multiple cycles of chemotherapy.[12] The most common metastatic site of gestational trophoblastic disease is the lungs, which are affected in over 80% of patients. The vagina is the second most common site of metastasis, accounting for 30% of cases. Malignant gestational trophoblastic disease is very sensitive to chemotherapy and a number of chemotherapy regimens are used to treat the disease .[13]

CONCLUSION

Common clinical presentation should alert the clinician to this rare condition includes bleeding per vaginum, anaemia, hyperemesis gravidarum, hypertension, thyrotoxicosis and uterine size disproportion. Ultrasound examination and beta HCG level measurement are the cost effective way to diagnose this in first trimester. Amniocentesis plays an important role when there is molar pregnancy coexisting with life normal foetus to this side further continuation or termination of pregnancy based on maternal and foetal prognosis. after providing appropriate counselling to the pregnant patient regarding continuation of pregnancy, pregnancy can be

continued but higher risk of complications remains example preterm labour and trophoblastic cancers.

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