An Unusual Co-existence: Complete Molar Pregnancy Along with a Normal Intrauterine Pregnancy

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Introduction

Gestational Trophoblastic disease (GTD) is a term that includes conditions that are the consequence of an abnormal pregnancy. The modified WHO classification of Gestational Trophoblastic disease includes complete and partial hydatidiform mole, invasive mole, choriocarcinoma, placental site Trophoblastic tumor, epitheloid trophoblastic tumor, exaggerated placental site and placental site nodules.[1] The incidence of gestational trophoblastic disease with normal pregnancy is about 2.5 to 5 percent of the molar pregnancies[2,3] or one in 20,000 – 1,00,000 pregnancies.[4] This clinical entity has pertinently been described as Sad Fetus Syndrome.[5] We report one such case of normal intrauterine pregnancy coexisting with a molar pregnancy.

Case Report

The patient was a 20 yrs old girl who was gravida[2] para[2] but with no alive issue. She had history of two IUDs within 7 months of gestational age. She reported at Combined Military Hospital, Bahawalpur with history of 6 weeks of amenorrhea. Her general physical and gynecological examination was unremarkable. Urine for Pregnancy test was positive. She was referred to the radiology department for routine pelvic scan which revealed minimally enlarged uterus with thickened endometrium and empty endometrial cavity. Follow-up scans were suggested keeping in view the positive pregnancy test. Repeat scan at nine weeks revealed a single gestational sac in fundal region of uterus with small fetal pole showing fetal cardiac activity.

After 17 weeks her pelvic scan showed single alive fetus with average gestational age of 17 weeks 4 days ± 1 week with a posterior low lying placenta. In addition there was multicystic lesion of 6.2 cm along the anterior uterine wall abutting internal os raising the possibility of coexisting molar pregnancy. Patient was then admitted for further investigations. β HCG was advised which was markedly raised to 48400 mlU/ml. Other tests including anticardiolipin IgG antibodies, toxoplasma IgM antibodies, antibodies to CMV, VDRL and IgG antibodies to rubella virus were negative. Chest X-ray was done with abdominal shield which was normal. OGTT was also normal.

Ultrasound was repeated at 19th week which showed single alive fetus of 19 weeks 5 days ± 2 weeks of gestational age. The multicystic lesion had grown in size measuring about 8 × 4.8 cm and was seen projecting inside the amniotic cavity through anterior uterine wall. Her β HCG was repeated which also showed increasing trend with a value of 221000mlU/ml. She was labelled as a case of molar pregnancy along with a viable fetus. It was decided to prolong the pregnancy

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under strict surveillance after an informed consent was obtained from both parents.

The next two serial pelvic scans with the interval of 2 weeks showed insignificant change in the size of the multicystic lesion but a normal fetal growth Fig 1, 2.

At 26 weeks of gestational amenorrhea the abdominal and pelvic scan of patient revealed slight increase in the size of multicystic lesion now measuring about 9.4 × 4.3 cm. The lesion showed increased vascularity on color doppler with some of the vessels traversing into adjacent endometrium along with an alive fetus with gestational age of 26 weeks 5 days ± 2 weeks 5 days. It was also noted that there was a well defined echogenic sub capsular focus measuring about 1.5 × 1.7 cm in the right lobe of liver of mother for which provisional diagnosis of hemangioma was made, however further workup was suggested to rule out the possibility of metastatic focus keeping in view the presence of molar pregnancy. Her repeat chest X-ray with abdominal shield was normal. Other investigations including blood Hb, TLC, LFT, PT, APTT, DRVVT, DRVVT ratio, Protein C were normal, with neither lupus anticoagulant activity nor detection of protein C and protein S. Her vital signs remained in normal limits.

At 27th week, Five days after the last abdominal scan she started having premature uterine contractions and was shifted to the labor room where she initially expelled molar pregnancy followed by preterm alive baby along with normal placenta. The initial APGAR score of the baby was 4/10 which improved to 6/10 and was shifted to the neonatology unit where he expired after two days. The post delivery scan showed no evidence of any residual tissue in uterus.

The multicystic mass and the placenta which was expelled with the fetus were sent for histopathological examination. The multicystic mass had grape like structure on gross examination and histopathology revealed dilated chorionic villi with cistern formation and proliferation of trophoblastic tissue representing molar tissue with absence of any fetal tissue thus confirming the diagnosis of complete hydatidiform mole.

Serial monitoring of β HCG was performed monthly for six months that showed downward trend with level of 0.47 mlU/ml after 6 months. Echogenic focus in the liver was further investigated with liver scan with Tc-99m pertechnetate stannous colloid and in vivo red blood cell labeling which was strongly suggestive of hemangioma. Follow up ultrasound of abdomen and pelvis was unremarkable with no change in the size of this lesion in the liver.

Discussion

We reported a case of complete hydatidiform mole with co-existing alive twin. The literature shows that few such cases have been reported internationally. A case was reported by Zhang et al[6] University of California San Diego where fetus died in utero at 26 weeks. Another study was reported by Bruchim et al[7] in Israel, where one woman delivered at 41 weeks of gestation with partial mole and fetus at 26 weeks. Most of such pregnancies are terminated prematurely either because of persistent hemorrhage or severe pre-eclampsia. Nearly three fourth of the cases do not go beyond 20 weeks of pregnancy.[8]

Usually, when a fetus is present in conjugation with partial mole, it generally exhibits the growth restriction and has multiple congenital malformations.[9] Whereas in this case, fetal growth was normal for the expected age with no obvious anomaly.

The considered absolute indications for terminating the pregnancy are preeclampsia, intractable vaginal bleeding, hyperemesis gravidarum, hyperthyroidism and evidence of trophoblastic embolisation. In a clinically stable patient an additional relative indication for therapeutic intervention is markedly enlarged uterus for dates.[10]

The management of gestational trophoblastic disease is evacuation of uterus but sometimes when molar change is there in the placenta along with an alive fetus then the condition must be managed under strict surveillance.[11] It will be difficult to outline the optimal management in gestational trophoblastic disease co-existent with live twin as most of the suggestions are based on either case reports or retrospective compilation and analysis.

Regular follow up in patients with GTD is required. We performed 09 ultrasound scans over the period of 26 weeks along with regular urine β HCG tests. The question of regular follow up of the patients with molar and coexisting pregnancy by serum β HCG has also been raised by some authors as such patient can also develop choriocarcinoma. In this context, one death has already been reported in the study conducted by Seckel et al.[12]

More studies are required on this topic which might help to formulate appropriate management gui-

delines for this unusual coexistence resulting usually in a loss of a life.

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