TRIPLET PREGNANCY WITH COMPLETE HYDATIDIFORM MOLE CO-EXISTING WITH TWO FETUSES (HMTCF) – A CASE REPORT

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ABSTRACT

Multiple pregnancies consisting of complete hydatidiform mole and coexisting fetuses are rare. Their incidence is increased due to the widespread use of ovulation induction. Triplet pregnancy with a coexisting hydatidiform mole is an extremely rare clinical condition. We report a case of 19 year old pregnant mother with 5 months amenorrhoea having triplet pregnancy with 2 live intrauterine fetuses and a complete hydatidiform mole.

Keywords: Vesicular mole, triplet pregnancy, complete and partial hydatidiform mole

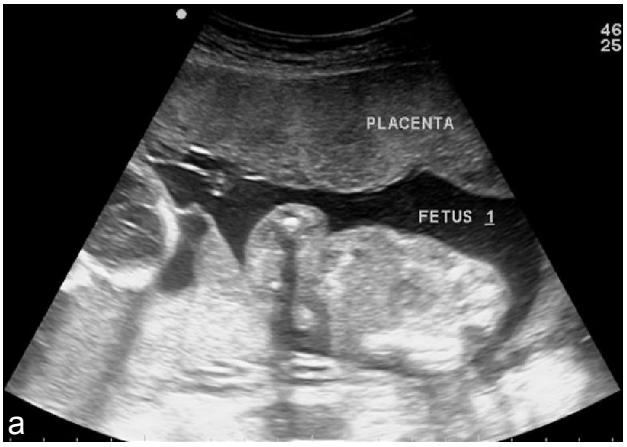
Introduction

Multiple pregnancies consisting of complete hydatidi form mole and coexisting fetuses are rare. Their incidence is increased due to the widespread use of ovulation induction. Triplet pregnancy with a coexisting hydatidiform mole is an extremely rare clinical condition. Previously, after diagnosis termination of pregnancy was the only option. Some authors have suggested continuation of pregnancy in the absence of fetal anomaly or eclampsia, irrespective of the development of persistent gestational trophoblastic disease (pGTD).[1] It is associated with increased risk of persistent gestational trophoblastic tumour.[2] Pregnancy can be continued in the presence of a stable pregnancy, normal sonogram and normal karyotype, however due to maternal complications of the vesicular mole, continuation of the pregnancy until term is impossible.[3] The incidence of complete hydatidiform mole (CHM) in twin pregnancies is 1 in 22,000 - 1,00,000 pregnancies and that in triplet is even rarer.[4] Vesicular mole with twin/ triplet can occur spontaneously or due to assisted reproductive technology.[5]

Case Report

A 19 year old pregnant mother with 5 months amenorrhoea presented with pain in abdomen. There was no history of per vaginal bleeding or leak. Her blood pressure was normal.

Obstetric ultrasound (Fig. 1, 2) revealed two live intrauterine fetuses with normal cardiac activities and movements corresponding to 18-19 weeks. Two separate placentas were noted. One was along anterior wall while other was along posterior wall. A large molar mass of size 10.4 (T) X 8.4 (AP) X 11.5 (CC)



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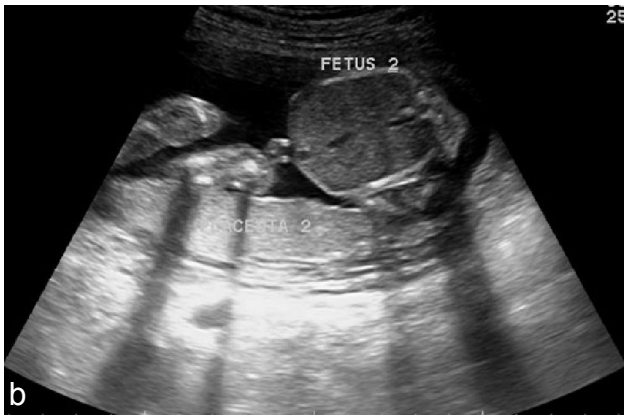


Figure 1(a,b): USG showing twins with both anterior and posterior placenta

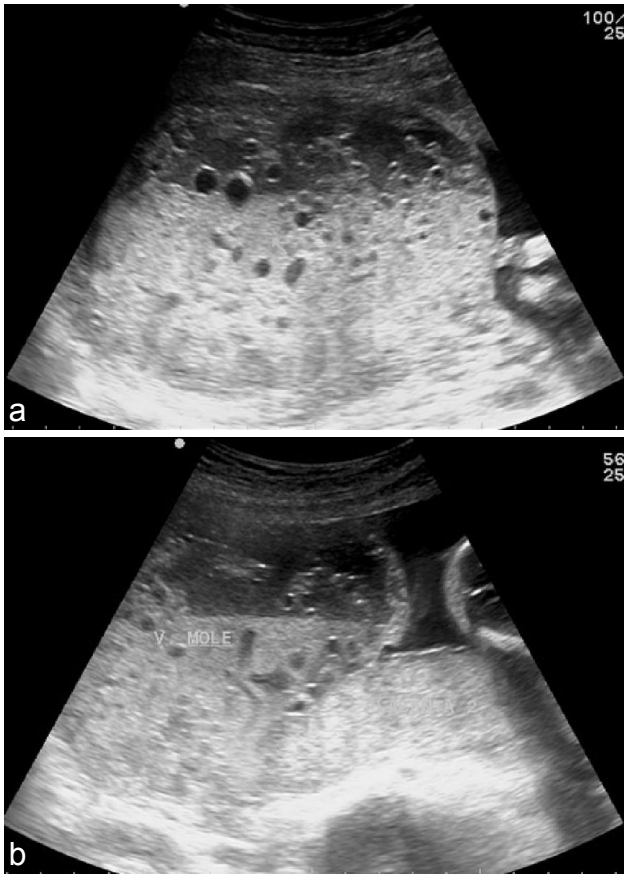
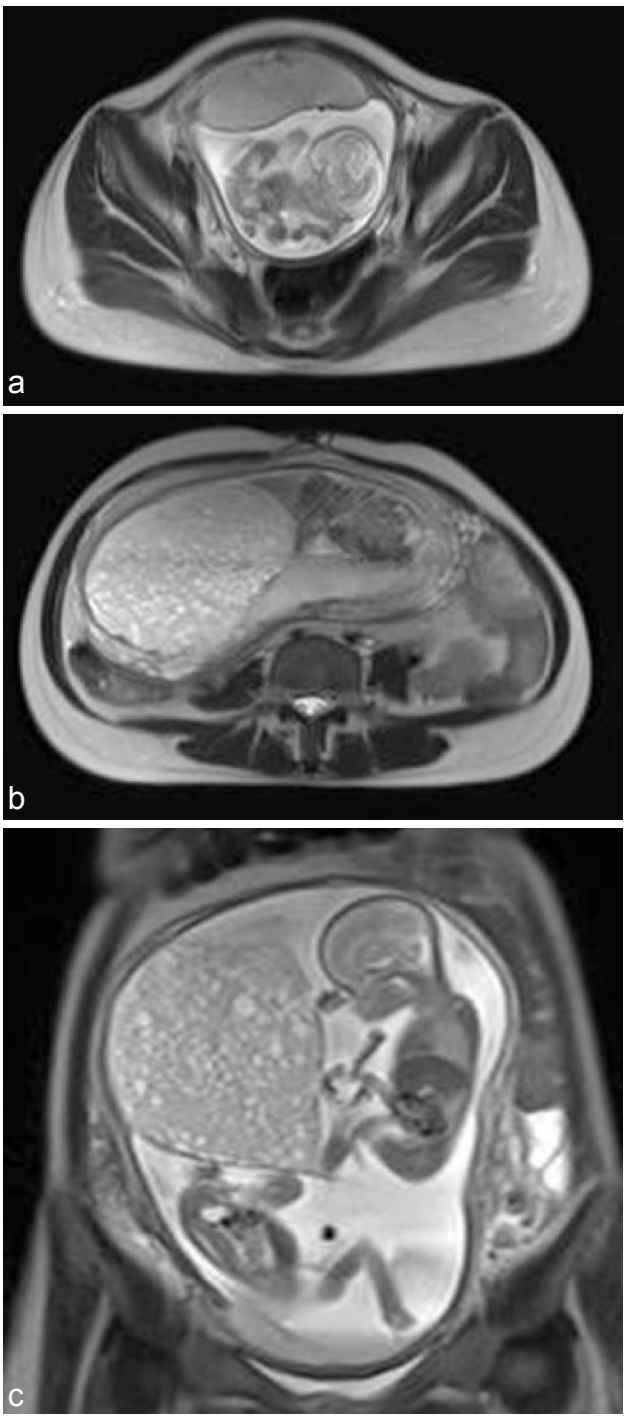


Figure 2(a,b): USG showing large complete vesicular mole with multiple anechoic cystic spaces at edge of posterior placenta

cm was noted near fundus on right side adjacent to posterior placenta but well demarcated from it. No obvious congenital anomaly was noted in both fetuses. The molar mass showed multiple small anechoic cystic spaces of size 2-5 mm. No significant increase in vascularity was noted. Diagnosis of triplets with one complete vesicular mole was made.

Obstetric MRI (Fig. 3 a-f) was done for confirmation which revealed two normal fetii with separate placenta with intervening membrane. One of the placenta was



anterior while other was posterior. A well-defined lesion measuring approx. 10.4 (T) X 8.4 (AP) X 11.5 (CC) cm was noted adjacent to right lateral edge of posterior placenta. It showed multiple small cystic

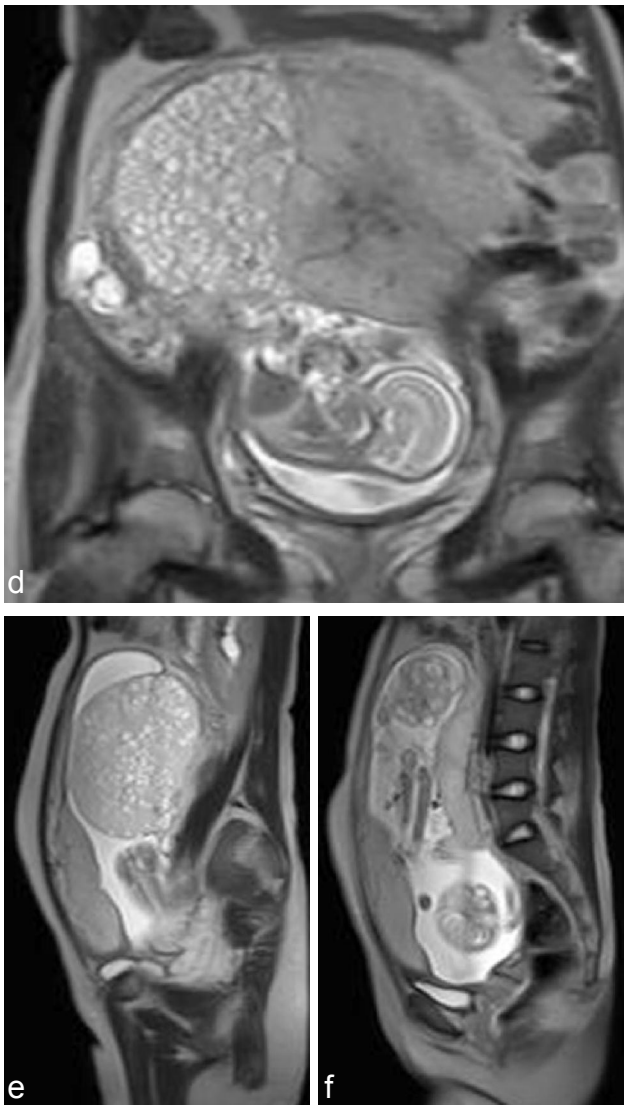


Figure 3: (Axial – **a,b;** coronal- **c,d;** sagittal- **e,f**): Obstetric MRI (T2 haste) showing two fetii with anterior and posterior placenta and complete vesicular mole at edge of posterior placenta

spaces of size 2-5 mm appearing slightly hypointense with respect to muscle on T1WI and slightly hyperintense on T2WI. No flow voids or hemorrhagic foci were noted. Interface between the lesion and underlying myometrium was well maintained. Both ovaries appeared slightly bulky and showed multiple small cysts. Right ovary measured approx. 3.6 X 2.0 cm. Left ovary measured approx 3.9 X 2.7 cm. No ascites was noted in maternal abdomen. Diagnosis of triplets with one complete vesicular mole and two normal fetii was made with bilateral small theca lutein cysts. Both parents decided to continue with the pregnancy after counselling as the other two fetii are normal. Presently, it is 29-30 weeks gestation with normal two fetii and complete vesicular mole. Patient has been referred to higher center for further management. Differential diagnosis of placental mesenchymal dysplasia was given. β-hCG levels were 1,16,301 mIU/ml i.e. markedly raised (normal range <5 and 13,500-80,000 in 2nd trimester).

Discussion

12 cases of triplet pregnancy with coexistent complete hydatidiform mole have been reported till 2007.[5]

Case Number and Year	Hormone therapy	Gestational age at delivery (wk)	Number of surviving fetuses	Maternal complications	Maternal chromosome
1-1980	Clomiphene	22	0	Vaginal bleeding, preeclampsia at 22 weeks	46,XX
2-1986	hMG+ hCG	17	0	Vaginal bleeding, persistent trophoblastic tumor	46,XX
3-1992	hMG+hCG	19	0	Vaginal bleeding	46,XX
4-1997	Clomiphene	17	0	Persistent trophoblastic tumor, choriocarcinoma, lung metastasis, hyperthyroidism	46,XX
5-1998	IVF-ET	15	0	Vaginal bleeding, persistent trophoblastic tumor	46,XX
6-1999	Clomiphene + FSH + hCG	15	0	Preeclampsia, secondary hyperthyroidism, persistent trophoblastic tumor	46,XX
7-2000	Clomiphene +hCG	30	1	None	46,XX
8-2000	None	24	0	Vaginal bleeding	46,XX
9-2001	None	21	0	Vaginal bleeding	46,XX
10-2003	hMG+hCG	28	2	Persistent trophoblastic tumor, lung metastasis	46,XX
11-2004	IVF-ET	31	1	None	46,XX
12-2007	IVF-ET	33	2	Preeclampsia	46,XY
Present Case	Clomiphene	Pregnancy in progress	0	None till yet	Patient refused

IVF-ET Invitro fertilization – embryo transfer

Cases of triplet pregnancy with two fetuses and a complete hydatidiform mole with maternal and fetal outcomes[5]

Hydatidiform mole with a coexistent fetus (HMCF) can be complete (CHMCF) or partial (PHMCF). Both CHMCF and PHMCF are also known as SAD FETUS SYNDROME.[6]

On the basis of pathology and genetic studies, hydatidiform mole can be classified as complete (CHM)

or partial (PHM). CHM is characterized by conspicuous trophoblastic hyperplasia, cystic villi and absence of fetal development. Genetically they are diploid but androgenic (Fig. 4). Their entire nuclear genome is paternal in origin. PHM shows normal to cystic villi with focal hyperplasia and evidence of fetal development or fetus. They are generally triploid, occasionally tetraploid and result due to fertilization of an egg by two or more sperms.[7] Differentiation between the two is done on ultrasound. Complete vesicular mole on ultrasound shows a complex cystic pattern with a snow storm appearance.[8] Cystic changes are less in partial vesicular mole.

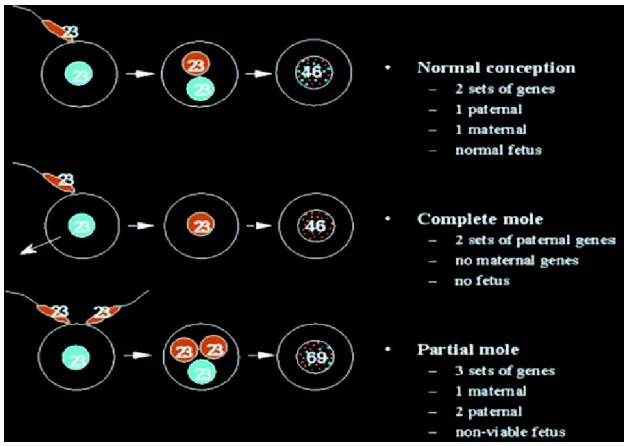


Figure 4: Genetic events in normal conceptions, complete and partial vesicular mole

Courtesy: Routhu M, Chouhan S. Role of Ultrasonography in Molar Pregnancy Coexisting with Viable Fetus: A Prospective Study. Int J Sci Stud[9]

Majority are CHMCF with significant risk of persistent trophoblastic disease (PTD) in 19.5-62.5% cases. PHMCF is rare with risk of PTD in 1.5-6% cases.[10] A complete hydatidiform mole (CHM) coexisting with normal fetus carries significant risk to both fetuses and mother and needs close surveillance. The fetal loss rate is nearly 60%. Maternal complications occur in 10% of the cases. Risk of tumor persistence is about 20% which is not influenced by the duration of pregnancy.[11]

It is associated with increased risk of preeclampsia, vaginal bleeding, hyperemesis gravidarum, premature delivery, persistent gestational trophoblastic disease (pGTD) and hyperthyroidism.[12] Complete vesicular mole has higher risk of invasive trophoblastic disease to the mother but offers a chance of delivering a healthy newborn infant. Whereas partial mole carries small risk of persistent trophoblastic tumours to the mother and is lethal for the fetus.[5] (Fig. 5)

Features	Partial Mole	Complete Mole
Karyotype	Most commonly 69, XXX or XXY	Most commonly 46 XX, XY
Pathology		
Fetus	Often present	Absent
Amnion, fetal RBC	Usually present	Absent
Villous edema	Variable, focal	Diffuse
Trophoblastic proliferation	Focal, slight-moderate	Diffuse, slight-severe
Clinical presentation		
Diagnosis	Missed abortion	Molar gestation
Uterine size	Smaller for dates	50% large for dates
Theca lutein cyst	Rare	25-30%
Medical complications	Rare	10-25%
Post molar GTN	2.5-7.5%	6.8-20%

GTN: Gestational trophoblastic neoplasia, RBC: Red blood cells

Figure 5: Differences between complete mole and partial mole

Courtesy: Routhu M, Chouhan S. Role of Ultrasonography in Molar Pregnancy Coexisting with Viable Fetus: A Prospective Study. Int J Sci Stud[9]

Theca lutein cysts are seen in upto 40% cases and occur due to hyperstimulation from the high circulating levels of β- hCG. These levels do not rise until early in the 2nd trimester and hence are not seen in early molar pregnancies. They are usually bilateral and multilocular. Doppler shows high vascularity with high diastolic flow due to reduced vessel tone.

Placental mesenchymal dysplasia is a differential diagnosis of vesicular mole. It is a benign entity which is associated with fetal Beckwith Wiedemann syndrome.

Conclusion

Triplet pregnancy with a coexisting hydatidiform mole is an extremely rare clinical condition. A complete hydatidiform mole (CHM) coexisting with normal fetus carries significant risk to both fetuses and mother and needs close surveillance. Differentiation between complete and partial vesicular mole associated with normal fetus can be done on ultrasound.

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