



Case Report

## **Twin Pregnancy with Hydatidiform Mole and Coexisting Fetus: A Case Report and Review of Literature**

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### **ABSTRACT**

A twin gestation with complete hydatidiform mole and co-existing live fetus is a rare entity. It presents with several dilemmas in management and the woman must be counseled regarding the complications and the risk of persistent trophoblastic disease. We report a case of a 30 year old G<sub>4</sub>P<sub>3</sub>L<sub>3</sub> at 14 weeks gestation came to our Casualty with the complaint of pain abdomen and bleeding PV since 20 days following the intake of abortifacient by self, diagnosed on ultrasound to have twin gestation with a single live fetus of 13 weeks 5 days gestation and complete hydatidiform mole. She expelled a male abortus with a few moles spontaneously and the remaining products were removed by suction evacuation. She was asked to follow up weekly with  $\beta$ -hCG report which decreased remarkably after two weeks and reached normal value two months following the evacuation. There was no evidence of persistent or metastatic disease during the one year follow up period.

**Keywords:** Twin gestation, complete hydatidiform mole, coexisting live fetus

### **INTRODUCTION**

The incidence of a twin gestation consisting of a complete hydatidiform mole (CHM) with a coexisting live fetus (CHMF) is between 1 per 22,000 and 1 per 100,000 pregnancies. <sup>[1]</sup> The clinical entity has aptly been described as Sad Fetus Syndrome. <sup>[2]</sup> The incidence is on the rise with the greater use of assistive reproductive techniques. <sup>[1]</sup> The management of these pregnancies is challenging due to associated complications and high risk to the mother as well as the coexisting fetus. Termination of pregnancy is hence traditionally advised in such cases

though it might not be acceptable to all mothers. In our case, the patient herself opted for termination of pregnancy. This case is presented because of its rarity.

### **CASE REPORT**

A 30 year old G<sub>4</sub>P<sub>3</sub>L<sub>3</sub> at 14 weeks gestation came with the complaint of pain abdomen and bleeding PV since 20 days following the intake of abortifacient by self for the termination of pregnancy. On the day of admission she complained of excessive bleeding per vaginum with passage of clots. She had previous three term vaginal

deliveries. General and systemic examination revealed no abnormality. On Per abdominal examination height of uterus corresponds 18 weeks size. Per vaginal examination revealed os open through which retained products of conception were felt. Ultrasound revealed dichorionic diamniotic twin gestation with a single live fetus of 13 weeks 5 days gestation. Placenta was fundal and left lateral. Another echogenic region of size 10 x 8 cms occupying the right half of uterine cavity filled with multiple cysts like structures suggestive of complete hydatidiform mole was seen(Figure 1). Bilateral ovaries were found to be normal. A diagnosis of twin gestation with complete mole was made. Investigations revealed mild pallor with neutrophilic leukocytosis and serum  $\beta$  human chorionic gonadotropin ( $\beta$ -hCG) 2, 14,900 mIU/ml. Chest X ray and thyroid function tests revealed no abnormality. She expelled a male abortus with few moles spontaneously within an hour of admission (Figure 2). The fetus had no structural abnormality; placenta was separate from the molar tissue and appeared grossly normal. Profuse amount of retained products of conception along with vesicle like structures were removed by suction evacuation and sent for histopathological study. She was asked to follow up weekly with  $\beta$ -hCG report. Serum  $\beta$ -hCG decreased remarkably after two weeks falling to 153 mIU/ml and reached normal value two months following the evacuation. There was no evidence of persistent or metastatic disease during the one year follow up period.

## DISCUSSION

Complete hydatidiform mole consists of generalized swelling of the villous tissue, diffuse trophoblastic hyperplasia and no embryonic or fetal tissue. The complete mole is diploid and chromosomes are totally derived from the

paternal genome. The identification is difficult in early stages of gestation and can be definitively diagnosed only by identifying two different genetic origins by using analytical techniques involving DNA polymorphisms. [3] Accurate differentiation between partial mole and complete mole with a coexisting fetus is crucial because of the chance of survival of coexisting fetus in instances that include a complete mole.  $\beta$ -hCG is normally raised in twin gestations and a relationship between  $\beta$ -hCG and coexisting molar in twin gestation has not been reported. Bilateral theca lutein cysts of ovary are also seen in a quarter of such cases after the end of first trimester under the influence of high maternal  $\beta$ -hCG values. [4]



Figure 1: Ultrasound showing complete hydatidiform mole (CHM) with a coexisting live fetus.



Figure 2: Expelled male abortus with complete hydatidiform mole.

Complications associated with a molar pregnancy, such as hypertensive disorders of pregnancy, hyperthyroidism, hemorrhage, pulmonary edema and thromboembolic phenomena, appear to be increased among CHMs coexistent with a live twin fetus. Also, there may be a higher risk of persistent gestational trophoblastic disease in these patients than in patients with an isolated CHM pregnancy. The estimated risk has been reported to be as high as 50%.<sup>[5]</sup> Fetal complications such as spontaneous miscarriages, intrauterine death, malpresentations and preterm labour may also occur. A woman who decides to continue with such pregnancies must be aware that she has 20% chance of early onset preeclampsia and less than a 50% chance of live term birth.<sup>[6]</sup> After detailed discussion with the couple about the risks involved, pregnancy may be followed with regular ultrasound assessment of fetal anatomy and growth.

There have been so far, about 200 cases of twin pregnancy with CHMF fully documented in literature, while only 56 cases result in a live birth.<sup>[7]</sup> Makrydimas et al reported a case of singleton pregnancy with a phenotypically normal fetus in which diffuse changes of complete mole were present in the placenta. The fetus was normal at birth and developed normally at 15 months.<sup>[8]</sup> Marijo Aguilera et al reported a case of CHMF with the molar component presenting antenatally as a placenta previa, which ultimately resulted in placenta accreta at the time of delivery. A live male infant was delivered at 34 weeks' gestation via planned cesarean section, and a hysterectomy was performed following unsuccessful removal of the molar component.<sup>[9]</sup> In a large study by Vaisbuch et al., they reported 130 cases of twins with CHMF pregnancy of which 41% were terminated because of the positive

probability of serious maternal complications.<sup>[10]</sup>

Shazly et al reported the case of a twin pregnancy with complete hydatidiform mole and coexisting fetus following ovulation induction with a non-prescribed clomiphene citrate regimen for a seven year period of primary infertility. Hysterotomy was done at 16 weeks due to vaginal bleeding and the 680 g male fetus expired with half an hour in neonatal intensive care unit.<sup>[11]</sup> F.Montes-de-Oca-Valero et al reported a case of twin pregnancy with a complete hydatidiform mole and co-existing fetus following in-vitro fertilization which required caesarean section at 28 weeks due to severe antepartum haemorrhage.<sup>[12]</sup> Sarah A et al reported the sonographic findings of metastatic gestational trophoblastic disease following a twin gestation with a complete mole and a live anencephalic fetus.<sup>[13]</sup> Papoutsis et al reported a partial mole pregnancy with a chromosomically and phenotypically normal embryo.<sup>[14]</sup> Copeland et al also reported dizygotic twin pregnancy with a normal fetus and partial hydatidiform mole.<sup>[15]</sup> One of the noticeable facts in studies reviewed was the gender of the fetus which was female. Studies revealed a connection of partial moles with male fetus.

## CONCLUSION

In the past, most CHMF gestations were terminated immediately following diagnosis because of poor information concerning clinical features and natural history. This circumstance has changed in recent years and that pregnancy may be continued when fetal anomalies and abnormal karyotype are excluded. However, close surveillance of risk factors is required for a favorable maternal and fetal outcome.

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