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# Successful Management of the First Case of a Metastasized Complete Mole in Form of Twin Pregnancy in Jordan

## Authors' Contribution:

Study Design A  
Data Collection B  
Statistical Analysis C  
Data Interpretation D  
Manuscript Preparation E  
Literature Search F  
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**Conflict of interest:** None declared

**Patient:** Female, 33-year-old  
**Final Diagnosis:** Twins molar pregnancy  
**Symptoms:** Vaginal bleeding  
**Medication:** —  
**Clinical Procedure:** Evacuation  
**Specialty:** Obstetrics and Gynecology

**Objective:** Rare co-existence of disease or pathology

**Background:** A complete mole with a living fetus in a form of twin pregnancy that is a rare obstetric event. The development of metastatic gestational trophoblastic disease is the most important and serious complication. It is debatable whether to terminate the pregnancy or to continue. Another point is the validity of the conservative treatment of this type of neoplasm, especially in developing countries.

**Case report:** In this study, we report the first case of a twin pregnancy with complete mole and a living fetus in Jordan, which is a developing country. A 33-year-old woman presented with vaginal bleeding which was revealed to be due to a complete mole in the form of a twin pregnancy. A hysteroscopy was performed with subsequent drop of  $\beta$ -hCG level. However, the  $\beta$ -hCG started to rise and the CT scan revealed multiple metastatic sites. Accordingly, she received 3 cycles of methotrexate, with a subsequent reduction in  $\beta$ -hCG level.

**Conclusions:** This is the first case report of a complete mole in twin pregnancy in Jordan. To overcome the development of metastasis, close follow-up and immediate treatment are crucial when the conservative approach is being considered. This report highlights the importance of considering the diagnosis of complete mole for women with twin and vaginal bleeding.

**MeSH Keywords:** Gestational Trophoblastic Disease • Hydatidiform Mole • Neoplasm Metastasis • Twins

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## Background

Twin pregnancy composed of molar and alive fetus pregnancy is a distinct type of gestational trophoblastic disease (GTD). The estimated incidence of this entity is 1 to 20,000–100,000 [1]. Although the pathogenesis and etiology of this form of pregnancy are unclear, an association of this type of pregnancy with obstetrical complications such as the development of metastasis, which is known to be a feature of late hydatidiform moles, has been reported in several studies. This rare entity increases the risk of serious complications [2–4] and thus requires precise management.

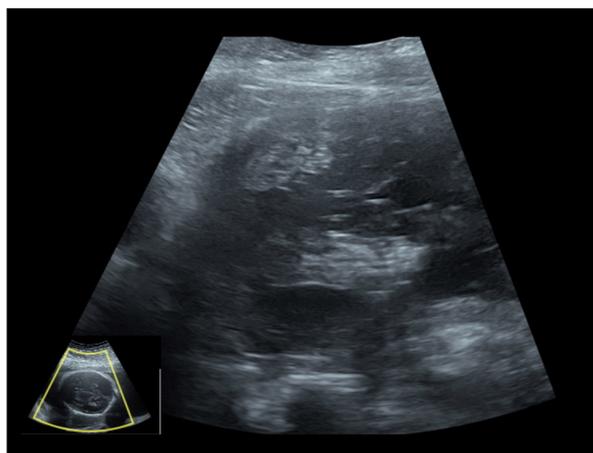
We report the case of a twin pregnancy with complete mole and alive fetus in a 33-year-old woman, which developed into persistent trophoblastic neoplasia after termination of the pregnancy.

## Case report

A 33-year-old woman, married for 6 years, presented to our center complaining of vaginal bleeding of 3-week duration. Her gravidity was 3, with 2 healthy children delivered by cesarean sections. This was the first antenatal visit for the patient, who was in the 13<sup>th</sup> week of her gestation according to her last menstrual period. The vaginal bleeding started 3 weeks prior to her presentation and was associated with passage of clots without abdominal pain. Hyperemesis gravidarum was reported. She reported 6–8 episodes of vomiting daily. She had no known medical illness. Her surgical history was remarkable for 2 prior cesarean sections and no family history of molar pregnancy. On examination, the uterine fundus was above the level of the umbilicus and the pelvic examination revealed that the cervical length was 2 cm. Her blood pressure was 142/93.

Therefore, ultrasound (US) was performed and revealed 2 sacs; the first sac had a positive beating fetal heart with normal placenta, while the second one showed a sac filled with a complete molar pregnancy (Figure 1). A urine dipstick result was +1. Laboratory investigations disclosed a serum  $\beta$ -human chorionic gonadotropin ( $\beta$ -hCG) level of 171,820 IU/L, hemoglobin level of 9.30 g/dl, low thyroid stimulating hormone, elevated T4 and T3, and elevated D-dimer. The other laboratory results were within normal range. Accordingly, she was admitted as a twin pregnancy-molar and alive. She received 2 units of blood and 2 units of fresh frozen plasma. A chest X-ray was negative (Figure 2A).

Subsequently, the decision was made to perform a hysterotomy through a Pfannenstiel incision. A uterus consistent with 26–28 weeks of gestation and bilateral theca lutein cyst 4×5 cm were detected. In addition, multiple pieces of brown

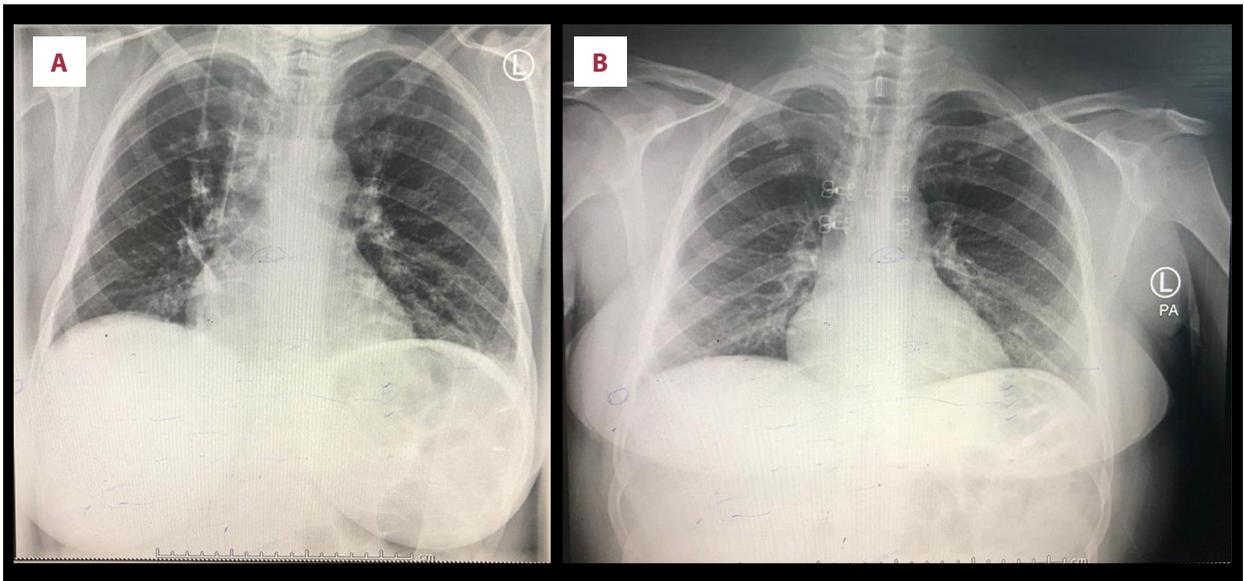


**Figure 1.** Ultrasound image indicating a suspected complete mole (indicated by arrows).

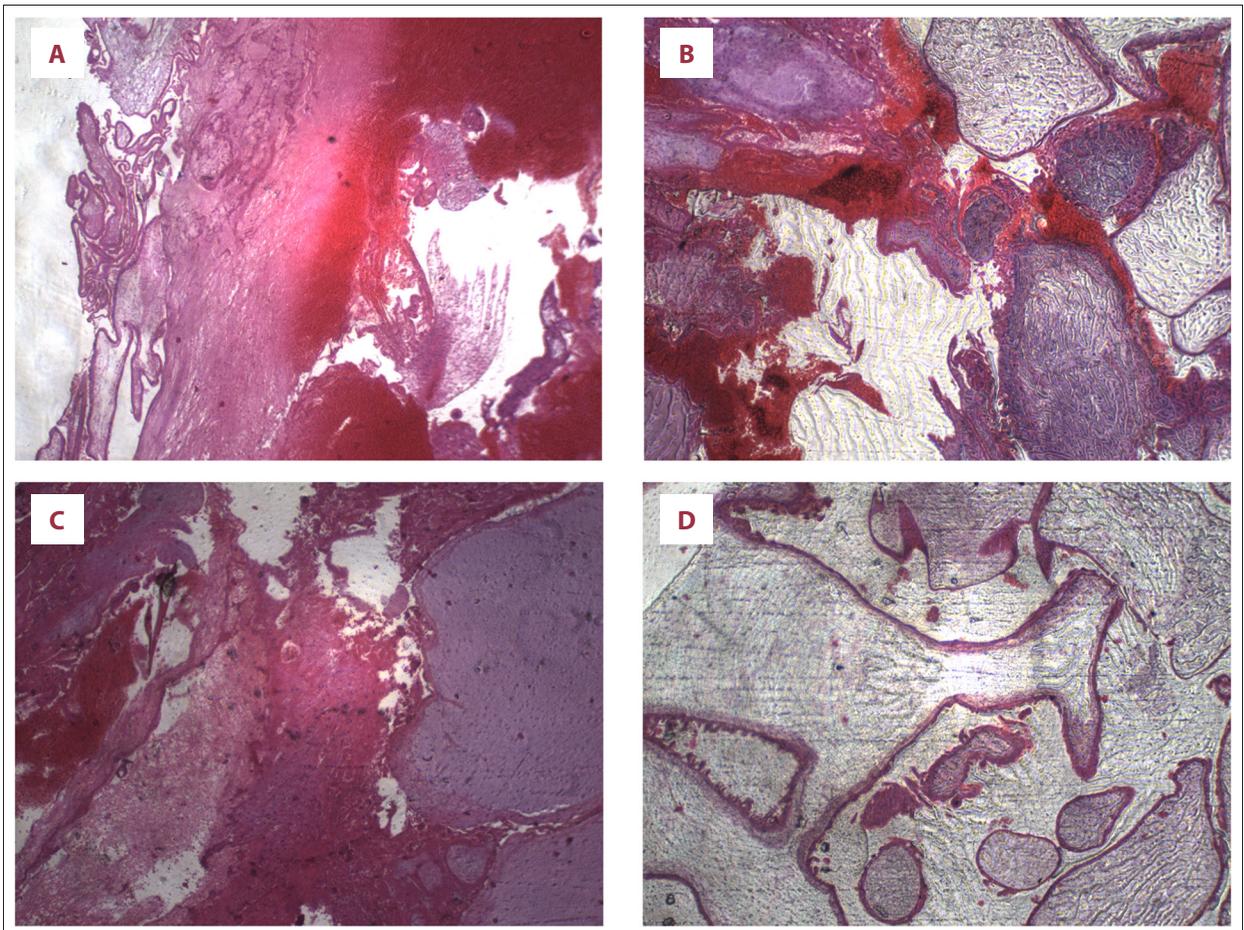
soft tissue with grape-like villi measuring 21×16×4.5 cm in aggregate were seen. A fetus with attached umbilical cord measuring 22×0.3 cm with a crown rump length of 9.0 cm, thigh length of 2.0 cm, and head circumference of 8.4 cm was found. The histopathological microscopic examination showed decidua and chorionic villi of irregular shapes with scalloped borders and central cisterns along with diffuse circumferential trophoblastic proliferation (Figure 3). There were normal chorionic villi also seen, along with an umbilical cord with 3 normal vessels. The diagnosis was consistent with molar pregnancy, in favor of complete mole with alive fetus.

Postoperatively, the patient was vitally stable, she had no abnormal vaginal bleeding, and the uterus was well contracted. Chest X-ray and  $\beta$ -hCG were repeated; the chest X-ray was normal (Figure 2B) and the level of  $\beta$ -hCG had started to decrease. The patient was discharged on postoperative day 4.

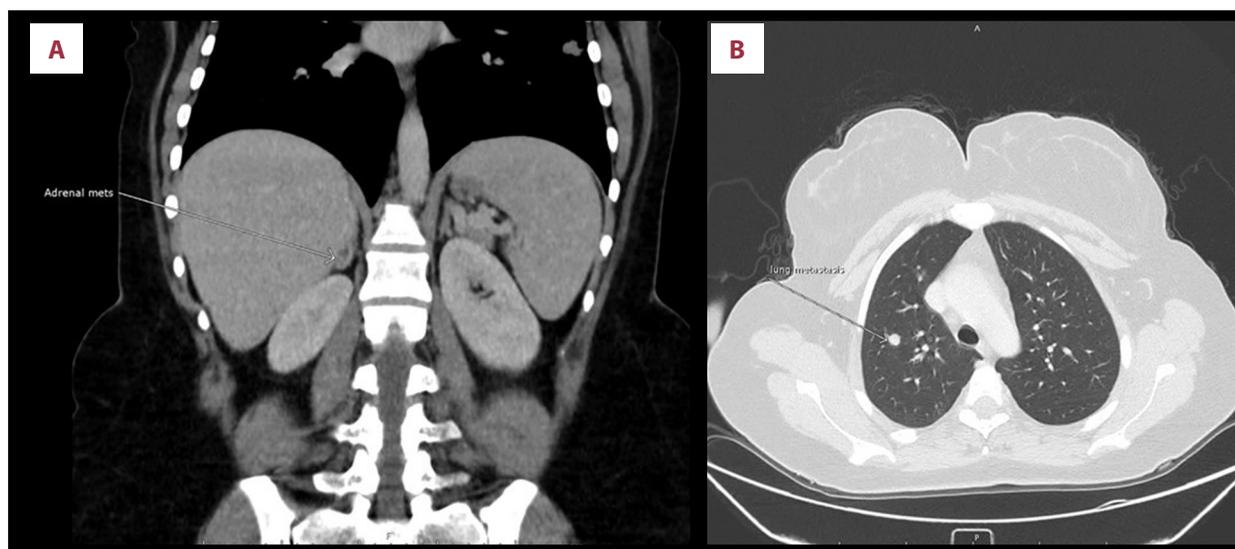
On follow-up, the  $\beta$ -hCG readings increased from 3,349 to 3,706 IU/L and continued to increase, reaching 7,334 IU/L over 10 days. Thus, she was admitted again, as a case of persistent trophoblastic neoplasia, for chemotherapy and chest-abdominopelvis computed tomography (CT). The CT scan was done with IV and oral contrast and showed chest multiple bilateral intraparenchymal and pleural-based nodules; the largest one measured about 1.2 cm in the posterior segment of the left lower lung lobe (Figure 4A) and prominent left paratracheal lymph node measuring about 0.8 cm representing metastasis. In the abdomen and pelvis, there was a well-defined hypodense lesion arising from the right side of the uterus, measuring about 5.8×4.7 cm, consistent with the patient's history of hydatidiform mole (Figure 4B). A hypodense lesion in segment VIII of the liver and a heterogeneously enhancing lesion in the medial limb of the right adrenal gland were detected, representing metastases.



**Figure 2.** Chest X-ray of the patient preoperatively (A) and immediately postoperatively (B) without evidence of metastasis.



**Figure 3.** (A-D) Microscopic images of the complete mole.



**Figure 4.** CT scan indicated the presence of adrenal metastasis (indicated by arrow) (A) and lung nodule (indicated by arrow) (B).

Therefore, she was started on methotrexate administered in 3 cycles, each cycle composed of 0.4 mg/kg/day of methotrexate for 5 days. After the first cycle, the  $\beta$ -hCG level was 219, after the second cycle it was 23, and after the third cycle it was 1.9. She was then counseled against pregnancy and was prescribed combined oral contraceptive pills and monthly  $\beta$ -hCG level monitoring. The  $\beta$ -hCG was repeated monthly for 6 months, with negative results (it plateaued at 0.10 IU/L). Her  $\beta$ -hCG stayed negative for the entire period of follow-up and she has been doing well since then. The total period of follow-up from the initiation of chemotherapy until the last follow-up was 8 months.

## Discussion

To the best of our knowledge, this is the first case report in Jordan of a complete mole in the form of a twin pregnancy with another alive fetus. Twin pregnancy with a complete mole and a coexisting live fetus is also known as sad fetus syndrome because the coexisting fetus is usually chromosomally normal and can be alive; however, in most cases, it is terminated [5,6].

Complete moles are a division of the neoplastic subdivision of GTD. Molar pregnancies have 2 distinct types, the complete or partial types based on genetic, clinical, and histological characteristics. The complete mole consists of diploid paternal genetic material as a result of the duplication of a single sperm fertilizing an empty ovum. The complete mole consists of edematous villous tissue and trophoblastic hyperplasia, without fetal tissue [6,7]. The clinical diagnosis of complete moles has now become more incidental because of advances in ultrasound. However, vaginal bleeding is the most common symptom of a complete mole. Also, the uterus may be distended

and large for gestational age. In addition, patients may complain of hyperemesis gravidarum. Moreover, signs and symptoms of hyperthyroidism can be present [8,9].

Many risk factors are associated with the occurrence of molar pregnancy. It has been reported that the incidence of GTD is much higher among women from Asia, West African, and South America. Also, very young and old maternal ages are risk factors. In addition, a previous history of molar pregnancy increases the risk of another molar pregnancy by 1–3%, and the risk rises to 15% with 2 previous molar pregnancies [6,10].

A 2015 review by Rohilla et al. found only 18 articles reporting 177 cases of twin pregnancy with one molar and one alive fetus; they found that preterm labor was the most common reported complication, antepartum hemorrhage was reported in 6 out of 18 studies [11], the reported incidence of persistent GTD was 33%, and only 66 fetuses out of 177 were alive [11].

Sebire et al. investigated the risk of pregnancy completion versus pregnancy termination in 77 twin pregnancies with one complete mole and 1 alive fetus, reporting that the chance of developing a viable fetus was 40%, with a very low incidence of fatal complications, including persistent GTD, if the pregnancy was continued [6,12].

Conservative management is indicated only in tertiary centers if close surveillance is present. On the other hand, termination of the pregnancy is indicated only if the patient develops complications such as severe pre-eclampsia, trophoblastic embolization, or significant vaginal bleeding [6,13], and it should also be considered in developing countries where there are fewer tertiary hospitals and where close follow-up is difficult. In our case, the pregnancy was terminated due to significant

vaginal bleeding and due to the possibility that the patient would be lost to follow-up.

GTD is classified by the FIGO system [14] into 4 stages: "Stage I: Disease is only in the uterus; Stage II: GTD extends outside the uterus but is limited to the genital structures; Stage III: GTD extends to the lungs and may or may not involve the genital tract; and Stage IV: GTD has extended to other distant sites."

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## Conclusions

We report the first case of a complete mole in twin pregnancy in Jordan. In twin pregnancies with one complete mole, it is unlikely that a healthy neonate will be delivered. Development of persistent GTD is a serious complication that needs comprehensive counseling, close follow-up, and precise intervention during the conservative management of this rare disease.

## Conflict of interest

None.