

# Coexisting complete hydatidiform mole and live fetus: A case report

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## CASE STUDY

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## ABSTRACT

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The authors report a case of twin pregnancy with complete hydatidiform mole coexisting with a live fetus (CHMCF) that developed gestational trophoblastic neoplasia (GTN). The patient with metastatic low risk GTN that developed resistance to single-agent chemotherapy achieved remission with combination chemotherapy. The patient's condition, management of complication and literature review of etoposide, MTX, Act-D, Cyclophosphamide, and vincristine (EMA-CO) are presented in the hope that great caution can be exercised in managing CHMCF.

### Key Words

Gestational trophoblastic neoplasia, EMA-CO protocol, combinational chemotherapy

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### Implications for Practice:

#### 1. What is known about this subject?

For patients with coexisting hydatidiform mole and fetus revealed from ultrasound findings, there are no optimal guidelines for management.

#### 2. What new information is offered in this case study?

Although conservative management is an acceptable trend, we have experienced a case of CHMCF that developed GTN.

#### 3. What are the implications for research, policy, or practice?

The patient with metastatic low risk GTN that developed resistance to single-agent chemotherapy achieved remission with combination chemotherapy and became normal pregnant after treatment.

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## Background

Coexistence of a live fetus and molar changes of placenta is relatively rare condition with an incidence of 1 in 22,000-100,000 pregnancies. Although conservative management is an acceptable trend, a twin pregnancy with complete hydatidiform mole coexisting with a live fetus (CHMCF) may develop gestational trophoblastic neoplasia (GTN). We report a case of CHMCF that developed GTN. The patient with metastatic low risk GTN that developed resistance to single-agent chemotherapy achieved remission with combination chemotherapy. The patient's condition, management of complication and literature review of EMA-CO are presented in the hope that great caution can be exercised in managing CHMCF.

## Case details

A 32-year-old Asian woman, gravida 1, para 0(G2P0) presented at 16 weeks' period of gestation with sonographic findings of a live intrauterine fetus and molar placenta (Figure 1A). She had no medical issue including common clinical signs and symptoms for molar pregnancy such as vaginal bleeding, uterine enlargement greater than expected gestational dates, hyperemesis or pregnancy-induced hypertension. In view of anticipated poor pregnancy outcome and risk of gestational trophoblastic disease, pregnancy was terminated after counselling, as per patient's preference. Suction evacuation was conducted for molar tissues. We did not perform a chromosome test because the patient did not want additional tests. A pathologic examination confirmed fetal tissue and complete molar tissues. The patient's serum level of human Chorionic

Gonadotropin (hCG) declined rapidly after evacuation of molar tissues. The patient was followed up with monitoring of serum hCG measurement every one to two weeks. During six weeks of follow-up, serial hCG gradually declined but serum hCG plateaued at 500-600mIU/mL level for five consecutive weeks. Postmolar gestational trophoblastic neoplasia (GTN) was suspected and computed tomography (CT) scan of chest, abdomen and pelvis (Figure 1B) as well as Magnetic Resonance Imaging (MRI) of brain were conducted for clinical staging. MRI of the brain was within normal limits. On chest CT, a few nodules in the lower lobe were detected (Figure 1C). These indicated high likelihood of metastases. According to the modified World Health Organization (WHO) risk-factor scoring system for GTN,<sup>1-5</sup> score was six. She was categorized as having low-risk stage III GTN. After suction evacuation, initially single agent chemotherapy using Methotrexate was done. However, this treatment failed. We changed single agent regimen into a combination of multi-agent chemotherapy that included etoposide, MTX, Act-D, cyclophosphamide, and vincristine (EMA-CO). Follow-up CT of chest, abdomen and pelvis was conducted after five cycles of chemotherapy to determine effectiveness of therapy. On CT of abdomen and pelvis, previously observed theca lutein cysts were significantly decreased (Figure 2A). When compared with previous CT of chest, a few lung nodules slightly decreased in size (Figure 2B). The patient's serum beta hCG level normalized after five cycles of chemotherapy. The patient was treated for a total period of five months and completed the follow-up for 14 months. There was no additional chemotherapy after five cycles of chemotherapy. Fortunately, a successful pregnancy was confirmed at the last visit.

## Discussion

In the largest study ever published (n=77), risk of GTN does not differ between singleton complete H-mole (CHM) and twin pregnancy with hydatidiform mole coexisting with a healthy foetus (CHMCF). Sebire, et al. found a similar rate of GTN after CHM (16 per cent) and after CHMCF (19 per cent).<sup>1,2</sup> However, results of several reports and reviews (Steller et al., Fishman et al., Matsui et al., Massardier et al.) differ from Sebire's data.<sup>1,3,4</sup> Compared with singleton hydatidiform moles, twin pregnancy with a foetus and a mole brings increased risk for postmolar gestational trophoblastic disease, with a higher proportion of patients having metastatic disease and requiring multi-agent chemotherapy.<sup>1,3,4</sup> Each patient with GTN should be allotted a stage and a risk score. Use of the International Federation of Gynecology and Obstetrics (FIGO) classification and the World Health Organization (WHO) scoring system is essential for determining initial therapy for patients with

GTN to assure best possible outcomes with least morbidity.<sup>5,6</sup> Low-risk GTN includes patients with non-metastatic (stage I) and metastatic (stages II and III) GTN whose prognostic risk score is less than 7. These patients may be treated with single-agent chemotherapy resulting in a survival rate approaching 100 percent.<sup>5,7</sup> High-risk metastatic GTN (FIGO stage IV and stages II-III, score $\geq$ 7) requires initial multi-agent chemotherapy with or without adjuvant radiation and surgery to achieve a survival rate of 80–90 percent.<sup>5</sup> Patient we reported was classified as low-risk metastatic GTN because she had lung metastasis and a six-point risk score. Patients with low risk GTN that develop resistance to single-agent chemotherapy may usually achieve remission with combination chemotherapy, with either MAC (Methotrexate, Actinomycin D, and cyclophosphamide), or EMA-CO.<sup>7</sup> Patients that fail single-agent treatment with higher levels of hCG should be treated with combination EMA-CO chemotherapy.<sup>8</sup> EMA-CO chemotherapy is a highly effective and well-tolerated treatment for metastatic, high-risk gestational trophoblastic tumours. Schink et al. reported 83 per cent complete response rate, 100 per cent overall survival and minimal toxicity using this protocol.<sup>9,10</sup> In this case, the patient had a high initial hCG level and a lung metastatic lesion, that was resistant to monotherapy and treated with multiple chemotherapy. In almost all reports about experiences using EMA-CO, treatment with EMA-CO was generally well tolerated and toxicity was mild. In our case, there was no life-threatening toxicity, but hair loss, vomiting and neutropenia occurred. If pre-treatment white blood cell count was less than 3000/mL, we used Granulocyte colony-stimulating factor (G-CSF) to prevent delayed chemotherapy as much as possible. Much like the above case, as Matsui et al. argue, if CHMCF progresses to gestational trophoblastic neoplasia (GTN), it is likely to have metastatic lesions or require a combination chemotherapeutic agent.<sup>3</sup>

## Conclusion

Although conservative management is an acceptable trend, a twin pregnancy with complete hydatidiform mole coexisting with a live foetus (CHMCF) may develop gestational trophoblastic neoplasia. This fact should be fully explained to the patient who wishes to remain pregnant. Further clinical trials and cost-effective studies are needed to determine a more effective choice of treatment in the case of twin pregnancy with complete hydatidiform mole coexisting with a live foetus (CHMCF) that developed metastatic gestational trophoblastic neoplasia (GTN).

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### PEER REVIEW

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### CONFLICTS OF INTEREST

The authors declare that they have no competing interests.

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None

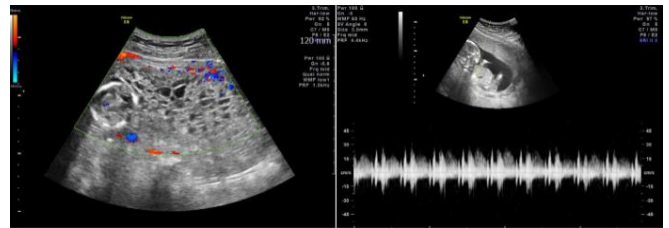
### PATIENT CONSENT

The authors, *Han JS, Kim JS*, declare that:

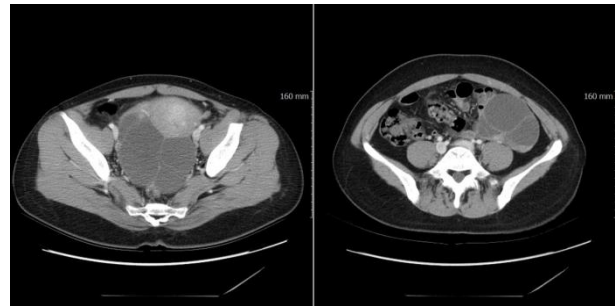
1. They have obtained written, informed consent for the publication of the details relating to the patient(s) in this report.
2. All possible steps have been taken to safeguard the identity of the patient(s).
3. This submission is compliant with the requirements of local research ethics committees.

**Figure 1: The images of the ultrasound (Figure 1A) and CT images (Figure 1B, Figure 1C) before treatment**

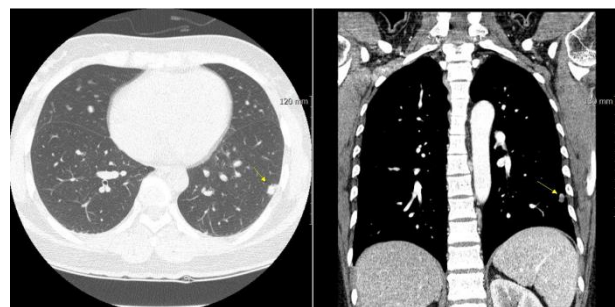
**Figure 1A Ultrasound shows a live intrauterine fetus and thickened molar placenta containing multiple focal cystic spaces**



**Figure 1B Abdominopelvis CT shows bilateral theca lutein cysts measuring 8.7×8.2cm on the left and 10×6.8cm on the right**

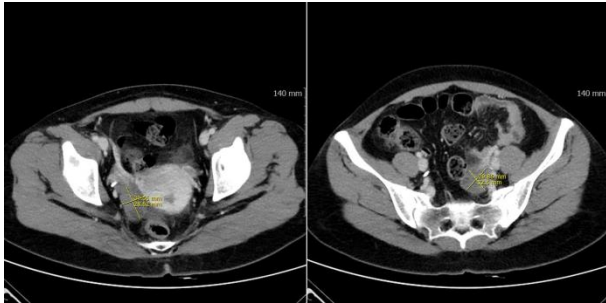


**Figure 1C: Chest CT shows a few nodules in left lower lobe including a largest one (arrow) measuring 1.3×0.8cm**



**Figure 2: The changes in CT images (Figure 2A, Figure 2B) after treatment**

**Figure 2A: On the follow-up CT conducted after five cycles of chemotherapy, previously seen theca lutein cysts were significantly decreased**



**Figure 2B: When compared with previous CT of chest, a few lung nodules slightly decreased in size**

