Uncommon presentations of choriocarcinoma

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Abstract
Choriocarcinoma is a rare disease in the young population. It uncommonly occurs after a normal full term pregnancy or in men. In the report, we presented two uncommon manifestations of choriocarcinoma. In the first case, a young woman developed choriocarcinoma metastasis after a normal full term pregnancy. She was treated with chemotherapy after the diagnosis and finally recovered. In the second case, a young man suffered from severe respiratory failure due to lung pathology and finally succumbed. The diagnosis of pulmonary choriocarcinoma could only be made after postmortem examination. Both cases illustrated atypical presentations of this malignant but curable disease.

Key words: Lung, choriocarcinoma, respiratory failure.

Introduction
Choriocarcinoma is a malignant proliferation of the Langerhans cell and of syncytial cells of trophoblastic origin. (1) It is normally situated in the female genital tract and the common presentation is vaginal bleeding. It uncommonly occurs following a normal term live birth (2) and rarely occurs in male. In men, choriocarcinoma may locate in the testis in combination with other tumours such as seminoma, teratoma, or embryonal carcinoma. (3) We presented two uncommon presentations of choriocarcinoma. The first case was a woman who developed respiratory failure after spontaneous normal delivery. She was subsequently diagnosed to have metastatic choriocarcinoma. The other case was a gentleman who presented with lung consolidation and respiratory failure. He deteriorated rapidly and invasive operation could not be performed. He finally succumbed before the cause of his lung pathology was identified. The diagnosis of choriocarcinoma of lung was made after postmortem examination.

Case 1
A 35-year-old lady, a known HbsAg carrier, was admitted to the hospital for term pregnancy. She had a molar pregnancy in China several years before without any adjuvant treatment. She had spontaneous delivery of a baby. After delivery, she suffered from breathlessness and her condition gradually deteriorated. She developed severe respiratory failure and was put on ventilator support 2 weeks after her delivery. She had mild anemia with hemoglobin level of 8.9 g/dL, leucocytosis (12.4x10^9/L), and normal platelet count. Tracheal aspirate grew MRSA and Acinetobacter. Chest X-ray showed bilateral lung infiltrates. Her liver and renal functions were unremarkable. CT thorax (Figure 1) showed multiple centrilobular ground-glass densities and patchy consolidations in bilateral lungs. It was present in both upper and lower zones but more extensive in posterior dependent areas. Some small nodular densities were also noted. Pulmonary trunk and arteries are dilated but no definite filling defect was detected in the pulmonary trunk and arteries and their major branches. CT scan of brain showed no gross abnormality. Her serum HCG level was grossly elevated to a maximum of 978732 IU/L. Her serum alfa fetoprotein level was mildly elevated to 30 IU/mL. She was suspected to have choriocarcinoma. Left thoracotomy was performed with wedge resection of left lung. She was found to have multiple firm consolidations over the whole left lung. Frozen section examination confirmed the diagnosis of choriocarcinoma of the lung. She was put on chemotherapy, with a total of 4 cycles of methotrexate. Her condition gradually improved and her HCG level gradually decreased after chemotherapy (Figure 2).
**Case 2**
A 41-year-old gentleman was admitted to the hospital because of breathlessness, pleuritic chest pain and hemoptysis. He also had fever of 38 degrees Celsius. On examination, his blood pressure was normal. He has no lymphadenopathy. His cardiovascular system was unremarkable. There were crepitations over both lung fields. Chest X-ray showed cardiomegaly and patchy nodular consolidation at right middle, lower, anterior segment of upper lobe and left lower lobe. His complete blood picture was essentially normal except mild leucocytosis. There was no significant derangement of liver and renal function. His blood culture yielded no growth. No acid-fast bacilli were detected in his sputum smear and no positive culture was grown from his sputum. Also, there were no malignant cells in his sputum. Autoimmune markers, including anti-nuclear antibodies and ANCA were negative.

Echocardiogram showed significant pericardial effusion with cardiac tamponade. Pericardiectomy was performed and there was heavy bloodstained pericardial effusion under mild tension. No malignant cells were detected in the pericardial fluid. Biopsy of the pericardium was done. Microscopic examination showed a piece of fibrous tissue with small amount of chronic inflammatory infiltrate. Some fibrin deposits were present on the surface. Focal reactive epithelial cell changes were seen. There was no granuloma or malignancy. Ziehl-Neelsen stain revealed no acid-fast bacilli. Bronchoscopy was performed. Sputum and bronchoscopic lavage were sent for tuberculosis PCR but all results were negative. His condition was critical so CT scan of thorax and thoracotomy could not be performed. He gradually deteriorated and finally succumbed. He was referred for postmortem examination.

The autopsy showed multiple tumour nodules in both lobes of lung with bilateral pleural effusion. The right lower and right middle lobes were replaced by tumour and the right upper lobe nodule invaded pericardium. There were multiple metastatic choriocarcinoma in the left frontal lobe, the liver and spleen, involving portal vein. The testis was atrophic without any germ cell tumour. Microscopically, it showed a mixed uni and multinucleated giant cell with extensive hemorrhage and necrosis and vascular permeation. Tumour cells were reactive toward HCG. The features were in favour of primary choriocarcinoma of lung.

**Discussion**
Choriocarcinoma is a highly malignant tumour arising most commonly from a complete hydatid mole. One-third of patients present with metastases. Metastases occur frequently in the lungs (80%). (4) It may occur following a variable time period between the molar pregnancy and the onset of symptoms. Choriocarcinomas following term normal delivery are very rare. It was reported to have an incidence of 1:50,000 live births. (5) Choriocarcinoma occurring during pregnancy is very rare and choriocarcinoma associated with a viable pregnancy is even rarer. The mother usually dies before the diagnosis of choriocarcinoma is even suspected. Only 5 cases of gestational choriocarcinoma coexisting with intrauterine pregnancy have been reported in which both mother and child survived. (4)

Primary choriocarcinoma of the lung have been reported and it may occur in combination with adenocarcinoma. (6-8) To the best of our knowledge, less than 30 cases were reported. (8)

In the first case, metastatic choriocarcinoma was not suspected initially as she had a normal full term delivery. Moreover, there were no evidence of choriocarcinoma involvement of the placenta and genital tract. It is likely that the metastatic choriocarcinoma coexisted with the pregnancy, as she was symptomatic immediately after the delivery. She has history of molar pregnancy. Whether the present episode of choriocarcinoma was related to her present pregnancy or the previous molar pregnancy could not be determined. It is important maintain a high index of suspicion of metastatic choriocarcinoma, even after a normal term pregnancy.

In the second case, the gentleman suffered from choriocarcinoma of the lung and his condition was too critical before any invasive surgery could be performed. Early histological examination might be useful if lung biopsy was performed before his deterioration. We did not check his HCG level, which might have provided a hint to his lung pathology. Such diagnosis was made only after postmortem examination.
**Figure 1.** Contrast CT scan of thorax of patient 1

![Contrast CT scan of thorax of patient 1](image)

**Figure 2.** Change of HCG level of patient 1

![Change of HCG level of patient 1](image)
References