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Metastatic choriocarcinoma of the lung without a primary: A case report

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ABSTRACT

Delayed presentation of metastatic gestational choriocarcinoma is a very rare phenomenon. We report a case of a 43-year-old female who presented three years after giving birth with a right lung mass and elevated β-human chorionic gonadotropin (HCG) levels. She underwent a right thoracotomy with right lower lobe lobectomy. The final pathology revealed metastatic gestational choriocarcinoma. In these cases, resection is recommended and patients often benefit from adjuvant chemotherapy. The patient was successfully treated with resection and is currently undergoing chemotherapy treatment. A delayed presentation of metastatic gestational choriocarcinoma should be considered as a differential in women presenting with a lung mass and a history of previous pregnancy. Prompt diagnosis and treatment can improve patient outcomes.

Keywords: Choriocarcinoma, Metastatic, Surgery, Thoracic surgery

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INTRODUCTION

Metastatic gestational choriocarcinoma of the lung with delayed presentation is a rare condition with only a handful of cases reported [1]. Gestational choriocarcinoma is the most aggressive trophoblastic disease, affecting approximately 1 in 40,000 pregnancies [2]. The disease has a good prognosis with a cumulated survival of about 80% even in widely disseminated disease [3]. We present a rare case of metastatic gestational choriocarcinoma of the lung in a 43-year-old female, presenting three years after pregnancy.

CASE REPORT

A 43-year-old female with a past medical history of kidney stones presented with complaints of acute onset left-sided chest pain that radiated to her left breast. The pain was sharp and constant, associated with diaphoresis and palpitations. On initial evaluation, she was afebrile with a temperature of 36.6°C. Her vital signs were stable with a heart rate of 70, blood pressure of 121/77, respiratory rate of 15, and an oxygen saturation of 99% on room air. During further evaluation, she revealed that she has been persistently lactating since giving birth via C-section approximately three years before, and was diagnosed with hyperprolactinemia of unknown origin by her endocrinologist. She had regular menstrual periods with her last menstrual period ending two days before her presentation. She was a former smoker, quitting about four years before her presentation.

Given her symptoms of chest pain, an electrocardiogram (ECG) was obtained which showed normal sinus rhythm, normal rate, no acute ischemic changes, and no ST-segment elevation myocardial infarction (STEMI). Initially in the emergency department, a chest X-ray was ordered, but an elevated D-dimer of 623 resulted prior to the chest X-ray

being performed. Therefore, the chest X-ray was canceled and a computed tomography angiography (CTA) of the chest was ordered instead due to concerns of pulmonary embolism. The CT scan performed by the emergency department revealed a lobulated mass in the lower lobe of the right lung measuring 2.9 cm with multiple areas of enhancement suspicious for blood vessels (Figure 1). She was then admitted for further care. Her laboratory work-up was remarkable for elevated troponin at 70, an elevated D-dimer, and a β-HCG level of 21,440 mIU/mL. Due to the elevated β-HCG, OBGYN was consulted and a transvaginal ultrasound was performed which demonstrated a 7-mm endometrium with no evidence of an intrauterine pregnancy. An endometrial biopsy was performed in which Pathology found no evidence of dysplasia or malignancy. A magnetic resonance imaging (MRI) of the brain and CT scan of the abdomen and pelvis did not reveal any abnormalities.

The vascular nature of the mass did not make it amenable to percutaneous biopsy or bronchoscopic biopsy, and after careful discussion with the patient the decision was made to proceed with surgical resection. On day seven after presentation, she was taken to the operating room where a right thoracotomy with right lower lobe mass wedge resection and intraoperative frozen sectioning was performed. Intraoperatively, the frozen sections confirmed the mass to be a non-small cell lung cancer, possibly an adenocarcinoma. The decision was made to proceed with the completion of a right lower lobectomy. She tolerated the operation well, and there were no complications.

Her postoperative course was unremarkable, with a rapid downtrend in serum β-HCG levels following resection. By post-op day 3, her β-HCG was 1,768 mIU/mL. Chest X-rays following lung resection showed no pneumothorax. Her vital signs remained stable throughout her post-operative course. She was discharged home on post-op day 4. Preliminary pathology revealed choriocarcinoma with negative margins. Final pathology revealed poorly differentiated metastatic gestational choriocarcinoma after undergoing short tandem repeat genetic sequencing which revealed paternal alleles and positive HCG staining (Figure 2). She was seen in clinic for follow-up two weeks, one month, and six months after her operation. She also received a positron emission tomography (PET) scan as an outpatient about two months after her resection. The PET scan did not show evidence of active tumor or metastasis. She did not have any issues or concerns during her post-op clinic visits. At this writing, she is currently undergoing chemotherapy treatment.

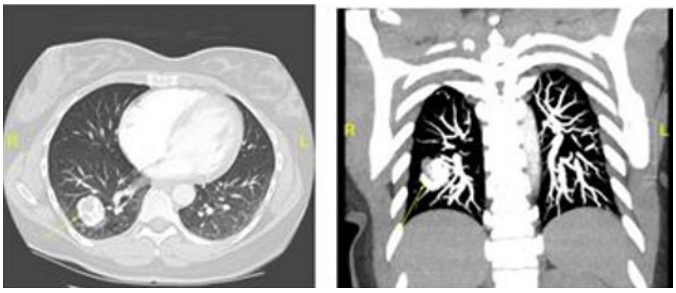

Figure 1: CTA chest in axial and coronal views demonstrating a mass in the right lower lobe measuring up to 2.9 cm with multiple areas of enhancement suspicious for vessels/internal pseudoaneurysms.

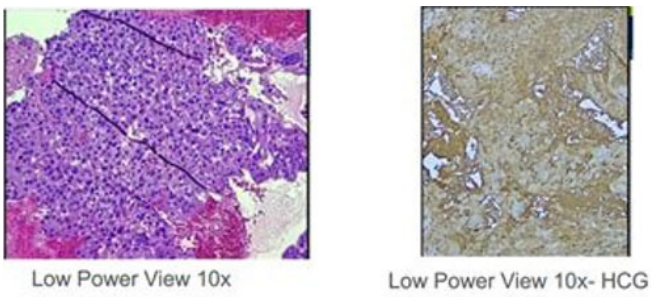

Figure 2: Histology slides revealing pathology of choriocarcinoma with right image showing positive staining for β-HCG.

DISCUSSION

We presented the case of a patient with metastatic gestational choriocarcinoma with presentation three years after a live birth by cesarean section. In metastatic choriocarcinoma, 80% of cases metastasize to the lung and 30% metastasize to the vagina. Patients may present with chest pain, shortness of breath, or vaginal bleeding [4]. Timely diagnosis is key to increase survival. Diagnosis is most often made with a combination of detailed history, CT imaging, pelvic exams, tissue biopsy, and advanced genetic sequencing technology [5]. Initially, the frozen pathology showed likely a non-small cell lung cancer, possibly an adenocarcinoma. We believe that the discordance of frozen and final pathology likely is due to lung adenocarcinoma's ability to mimic choriocarcinoma histologically as they share similar features. These shared histologic features make differentiating between lung adenocarcinoma and choriocarcinoma very difficult, especially before special staining for cytotrophoblasts and β-HCG are performed [6]. In our patient's situation, special staining for β-HCG was not performed until permanent pathology. The short tandem repeat genetic sequencing which confirmed metastatic gestational choriocarcinoma is also not something that is performed on frozen pathology, but rather permanent pathology. Therefore, it is not surprising that there was discordance between frozen pathology and final pathology.

What makes our case unique is the substantial delay in the onset of the choriocarcinoma. In this case, the choriocarcinoma of the lung represented metastases that arose from unknown trophoblastic disease from a pregnancy. We believe the delay was a result of spontaneous regression of the choriocarcinoma after metastasis. This

unique phenomenon has been cited in literature as the "burned out" hypothesis, which reflects the unique and specific feature of choriocarcinoma. That is, it is known to metastasize and become dormant before a primary lesion is detected [6, 7]. Due to this phenomenon, diagnosis is often delayed until tissue diagnosis is obtained and short tandem repeat genetic sequencing is performed to differentiate primary choriocarcinoma from metastatic gestational choriocarcinoma [8, 9]. If short tandem repeat genetic sequencing reveals paternal alleles, it indicates that the choriocarcinoma's origin is gestational, as the paternal alleles can only be from gestational origin. If the short tandem repeat genetic sequencing reveals no paternal alleles, it shows the choriocarcinoma is a primary lesion [8, 9].

While our patient presented three years after giving birth with chest pain, it is important to note other presentations of gestational choriocarcinoma so the diagnosis can be made as quickly as possible. In a case presented by Tian et al., the patient presented during her second trimester of pregnancy [10]. Another case, by Parajuli et al., describes a presentation three years after delivery with anemia and bilateral kidney and lung metastasis [11]. In a systematic review on choriocarcinoma by Mangla et al., only 50 out of their 121 patients with choriocarcinoma had an antecedent pregnancy [12]. These different studies illustrate the varying ways in which gestational choriocarcinoma can present.

Treatment of metastatic gestational choriocarcinoma has been well documented to involve resection of the mass as well as chemotherapy in the form of methotrexate, or a combination of methotrexate, etoposide, and actinomycin D [13]. This case illustrates the need for high suspicion of metastatic gestational choriocarcinoma in young females with lung masses.

CONCLUSION

This is a case of metastatic gestational choriocarcinoma of the lung presenting three years after pregnancy. This rare occurrence was successfully treated with surgical resection and chemotherapy. Although rare, suspicion of metastatic gestational choriocarcinoma in female patients with chest pain may help lead to faster diagnosis and treatment.

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Trina Capelli – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all

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Authors declare no conflict of interest.

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