Primary Pulmonary Choriocarcinoma Presenting with a Hemothorax

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Abstract: Primary pulmonary choriocarcinoma is an extremely rare condition that is difficult to diagnose. In this report, we describe a case of primary pulmonary choriocarcinoma that presented with a hemothorax, initially diagnosed as an ectopic pregnancy. The patient was treated successfully with surgery followed by combination chemotherapy. In rare instances such as this, choriocarcinoma can originate in sites outside the genital tract; a thorough and thoughtful evaluation will ensure an accurate diagnosis and prompt treatment.

Key Words: Choriocarcinoma, Primary pulmonary Choriocarcinoma, Hemothorax.

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Choriocarcinoma usually arises in the uterine cavity. Although this may metastasizes to the lung, primary pulmonary choriocarcinoma is extremely rare. The clinical manifestations of pulmonary choriocarcinoma vary from an asymptomatic state to hemoptysis, chest pain, or dyspnea, according to the site or pattern of the tumor. A few cases of choriocarcinoma presenting with a hemothorax have been reported, but most of these cases involved metastatic tumors. We report a highly unusual case of primary pulmonary choriocarcinoma that presented with a hemothorax.

CASE REPORT

A 19-year-old woman was admitted to the emergency room for chest pain that had developed suddenly 2 hours before admission. She was hemodynamically stable and had decreased breath sounds, particularly in the right lower lung field. Her last menstrual period had occurred 5 months before presentation and a qualitative urine beta human chorionic gonadotrophin (β-hCG) test was positive. However, a transvaginal ultrasound could detect neither an intrauterine nor extrauterine gestational sac. Chest radiograph and computed tomography showed a substantial effusion in the right pleural cavity and chest tube insertion yielded a bloody effusion, which confirmed the diagnosis of hemothorax. Follow-up computed tomography demonstrated a thin-walled mass measuring 2.5 cm in the right lower lobe of the lung (Figure 1). The serum level of β-hCG was 4247 mIU/ml. Endometrial curettage was performed and no gestational tissue was found. The patient was preoperatively diagnosed with an ectopic gestation in the abdominal cavity involving the diaphragm. During thoracoscopy, a mass was found in the right lower lung near the diaphragm. Frozen pathologic examination suggested that it was a choriocarcinoma. A thoracotomy was performed for complete removal of the tumor because its extensive adhesion to the pleura impaired thoracoscopic resection of the mass. The final pathologic examination confirmed the lesion as a primary pulmonary choriocarcinoma (Figure 2) and the patient received three courses of the multiagent etoposide, methotrexate, actinomycin, cyclophosphamide, vincristine chemotherapy regimen (etoposide 200 mg/m²/d on days 1 and 2, methotrexate 300 mg/m²/d on day 1, actinomycin 0.5 mg/d on days 1 and 2, cyclophosphamide 600 mg/m²/d on day 8, and vincristine 1.0 mg/m²/d on day 8). After a year, her serum β-hCG levels were within the normal range.

DISCUSSION

Primary pulmonary choriocarcinoma is extremely rare, with only 23 cases reported in the literature. To our knowledge, a primary pulmonary choriocarcinoma presenting with a hemothorax has never been reported. Although the origin of primary pulmonary choriocarcinomas is controversial, several theories about the pathogenesis of this tumor have been proposed, such as differentiation of the pulmonary epithelium into trophoblastic structures or metastatic emboli of gestational trophoblastic tissue undergoing spontaneous regression. In our case, we excluded metastatic gestational choriocarcinoma because the pelviscopy revealed grossly normal pelvic organs and endometrial tissues. The present case is an unusual type of primary pulmonary choriocarcinoma that manifested as a hemothorax. Although it was originally erroneously diagnosed as an ectopic pregnancy, the patient...
FIGURE 1.  A, A chest radiograph showing a large right pleural effusion.  B, Transvaginal ultrasound reveals no specific findings in the pelvic organs.  C, A contrast-enhanced chest computed tomography (CT) showing a large pleural effusion and a thin-walled cystic mass in the right lower lobe (arrow).  D, After drainage of the hemothorax, a cystic mass is still visible on CT (arrow).

FIGURE 2.  A, The cut surface shows a circumscribed hemorrhagic mass involving the pleura.  B, Low-magnification microscopy of the choriocarcinoma shows a sheet-like proliferation of atypical cytotrophoblasts (arrow) and scattered syncytiotrophoblasts (arrow head) with hemorrhage (hematoxylin-eosin, original magnification ×100).  C, Tumor cells stain strongly immunopositive for β-hCG (original magnification ×100).  D, High magnification of the tumor cells shows marked nuclear and cellular atypia and increased mitotic activity (arrow) (hematoxylin-eosin, original magnification ×400).
was treated successfully by surgical resection and combination chemotherapy.

REFERENCES
