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# A rare entity mimicking hydatid cyst: Primary pulmonary choriocarcinoma

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## Abstract:

Primary pulmonary choriocarcinoma is a very rare tumor with a poor prognosis. Due to its non-specific clinical presentation and radiological similarities to infections and other malignancies, it is often misdiagnosed or diagnosed late. Furthermore, there is no standardized treatment protocol. We present the case of a 40-year-old male with a history of tuberculosis who was admitted with hemoptysis and dyspnea. Imaging revealed a large cavitory mass in the right lung, along with nodular and cystic lesions. The initial diagnosis suggested a hydatid cyst; however, further evaluation, including pathological and immunohistochemical analysis of the resected tissue, ultimately identified the cystic lesion as choriocarcinoma. This diagnosis was confirmed by elevated levels of  $\beta$ -human chorionic gonadotropin in the postoperative period. Despite advances in imaging and serologic testing, PPC is frequently misdiagnosed, highlighting the need for a high index of suspicion. Early recognition and appropriate management are essential to improve outcomes in this aggressive tumor.

## Keywords:

Hydatid cyst, lung, primary pulmonary choriocarcinoma, surgery

## Introduction

Extragenital germ cell tumors are a heterogeneous group of lesions that originate at extragenital sites without evidence of a primary tumor in the gonads. Choriocarcinoma is one of the rarest forms of extragenital germ cell tumors and is characterized by the presence of syncytiotrophoblast cells, which secrete  $\beta$ -human chorionic gonadotropin ( $\beta$ -hCG).<sup>[1]</sup> These tumors typically arise in midline structures. In addition to the

lungs, cases involving other internal organs such as the brain, stomach, small intestine, kidney, and adrenal glands have also been reported.<sup>[2]</sup> Although early metastasis to the lungs is well-documented, primary pulmonary choriocarcinoma (PPC) is extremely rare.

## Case Report

A 40-year-old male patient with a history of tuberculosis 20 years prior presented with hemoptysis and dyspnea. Thoracic

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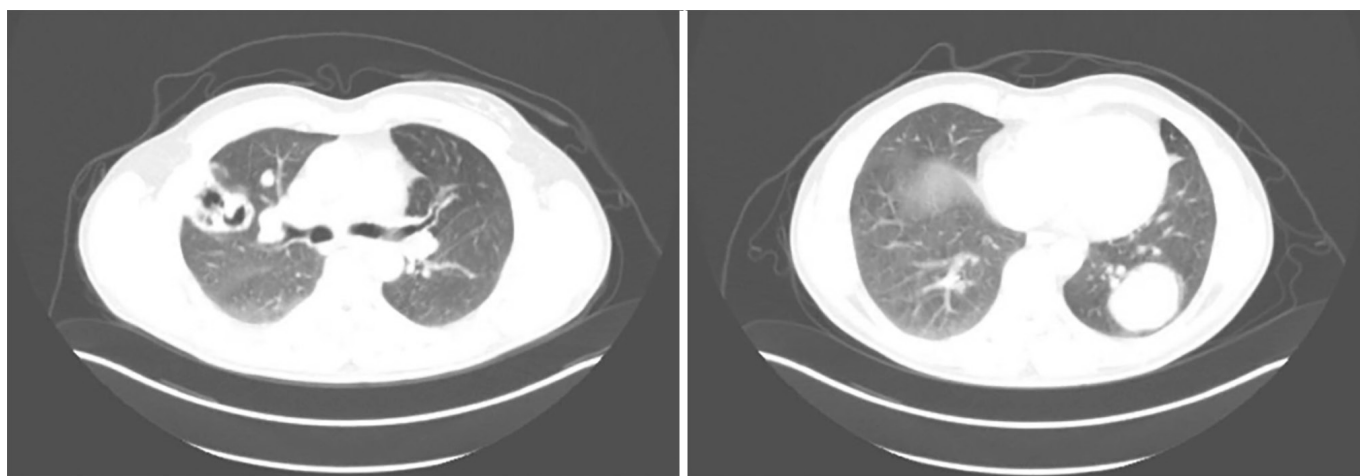


computed tomography (CT) revealed a cavitory mass lesion measuring 55×53 mm in the right upper lobe, along with a 13 mm nodular lesion in the same lobe. A cystic lesion measuring 50×46 mm was also observed in the left lower lobe [Fig. 1]. The indirect hemagglutination (IHA) test for hydatid cyst returned positive. Acid-fast bacilli were negative in three sputum samples, and no growth was observed in mycobacterial culture. A transthoracic fine-needle aspiration biopsy was performed on the cavitory lesion in the right lung, with an initial clinical suspicion of malignancy. Pathology revealed primary lung carcinoma, but subtyping could not be determined. A positron emission tomography-computed tomography (PET-CT) scan showed a maximum standardized uptake value (SUVmax) of 6.8 in the cavitory lesion in the right upper lobe, 1.4 in the adjacent nodule, and 4.0 around the cystic lesion in the left lower lobe, all indicating fluorodeoxyglucose (FDG) uptake. Mediastinal lymphadenopathy was also noted, with an SUVmax of 4.2 in the right hilar and right lower paratracheal regions. No distant organ metastasis was detected. Bronchoscopy revealed no endobronchial lesion. However, endobronchial ultrasound performed for mediastinal staging confirmed carcinoma metastasis in the right lower paratracheal and subcarinal lymph nodes. Due to the cystic appearance of the lesion in the left lung on radiological imaging and the positive IHA test, a hydatid cyst was initially included in the differential diagnosis. Therefore, surgical resection was planned. During perioperative exploration, the lesion was aspirated, revealing necrotic and hemorrhagic pus. Frozen section analysis confirmed malignancy. A left lower lobectomy was performed due to the exposed cavitory lesion and diagnostic uncertainty. Microscopic examination revealed

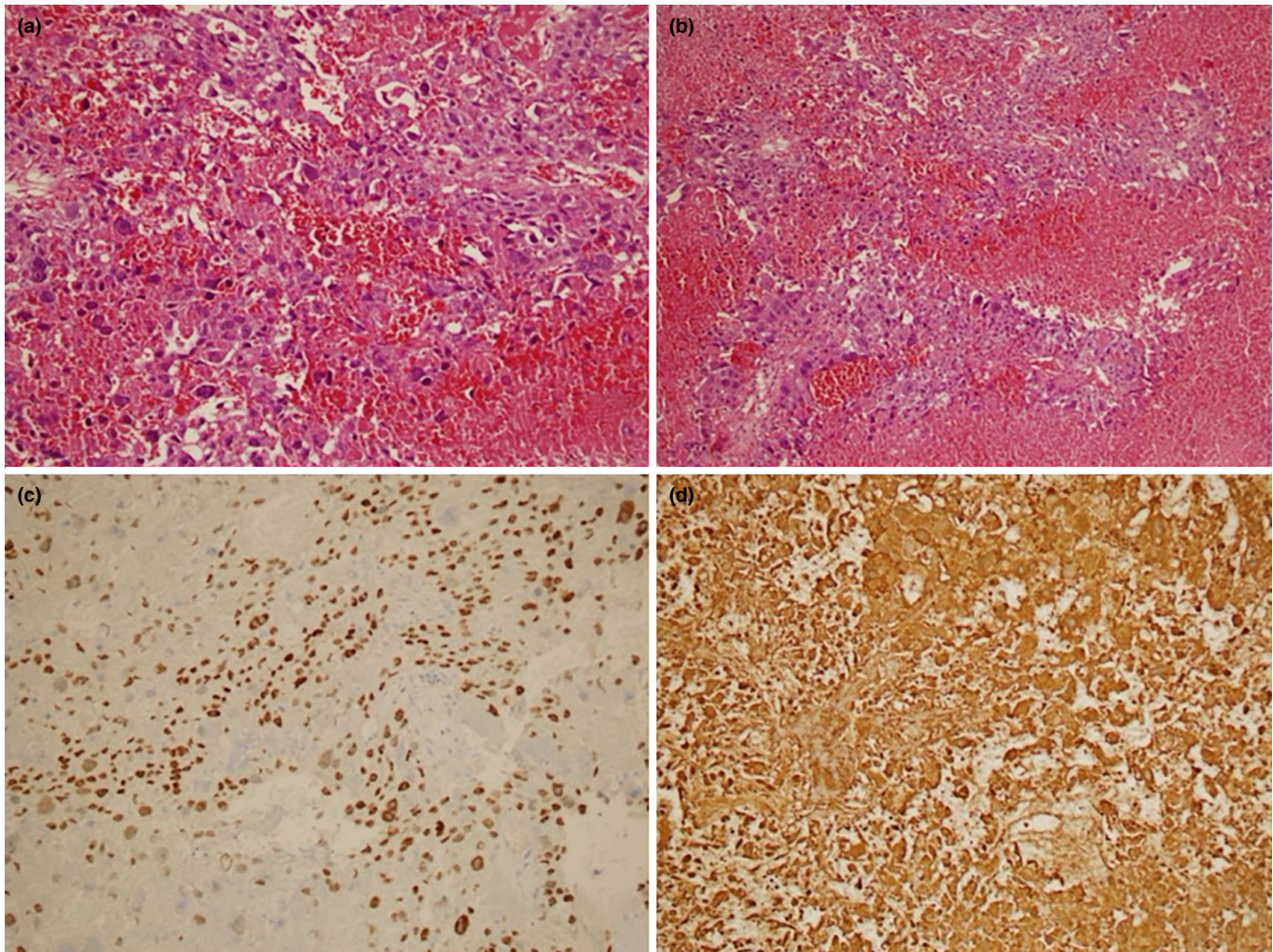
extensive hemorrhagic and necrotic areas containing multiple cell types. Large cells with abundant cytoplasm and degenerative morphology, characterized by broad, pleomorphic, hyperchromatic nuclei, were observed [Fig. 2]. After excluding lung adenocarcinoma (negative for thyroid transcription factor-1 [TTF-1] and napsin A), squamous cell carcinoma (negative for p40), and neuroendocrine tumors (negative for neuroendocrine markers), choriocarcinoma was suspected due to the presence of solid islands, syncytial structures, and trophoblastic cells. Immunohistochemical staining was therefore performed accordingly. The diagnosis of choriocarcinoma was confirmed by positive immunohistochemical staining with GATA-binding protein 3 (GATA3), Cytokeratin 7 (CK7),  $\beta$ -HCG, and Sal-like protein 4 (SALL4) [Fig. 2]. In the postoperative period, the patient's  $\beta$ -HCG and alpha-fetoprotein levels were elevated above the normal range. Histological examination of fine-needle aspiration samples from the right upper lobe and lymph nodes revealed features similar to the tumor found in the left lower lobectomy specimen. As no lesions suggestive of choriocarcinoma were identified on genital examination, the patient was diagnosed with metastatic primary pulmonary choriocarcinoma (PPC), and systemic chemotherapy was initiated by the medical oncology team.

## Discussion

Primary pulmonary choriocarcinoma is an extremely rare and aggressive tumor. In the literature, Cao *et al.*<sup>[3]</sup> analyzed 68 patients with PPC and reported a mean age of  $44.5 \pm 16.8$  years. We present the case of a 40-year-old male diagnosed with PPC. In a meta-analysis of 55 pa-



**Figure 1:** Thoracic computed tomography image showing bilateral lesions in the lungs



**Figure 2:** (a) Hematoxylin and eosin, x20: Solid nests and sheets of syncytial cells. Syncytiotrophoblasts appear as large cells with abundant dense eosinophilic cytoplasm. Their nuclei often exhibit a degenerated and smudged appearance. (b) Hematoxylin and eosin, x40: Hemorrhage is commonly observed. (c) Immunohistochemistry (IHC), x200: Positive staining for Sal-like protein 4 (SALL4). (d) Immunohistochemistry (IHC), x200:  $\beta$  Positive staining for beta-human chorionic gonadotropin ( $\beta$ -HCG)

tients with PPC, 47 were symptomatic at diagnosis, presenting with symptoms such as persistent cough, dyspnea, and hemoptysis.<sup>[2]</sup> These findings highlight that PPC often manifests with vague clinical signs, leading to frequent misdiagnosis or delayed diagnosis. In a case report by Wang *et al.*,<sup>[1]</sup> treatment was initiated based on the presence of a cystic lesion on thoracic CT that was initially presumed to be infectious in origin but was later confirmed pathologically as PPC. Similarly, in our case, imaging revealed a cavitory lesion with an adjacent nodule in the right lung and a cystic lesion in the left lung. Due to the patient's history of tuberculosis and the presence of a cavitory lesion, tuberculosis was initially considered. Additionally, the cystic lesion and a positive IHA test led to the inclusion of hydatid cyst in the differential diagnosis. Given the patient's hemoptysis and

history of smoking, malignancy was also considered. The presence of a cystic lesion in the left lung, together with a positive IHA result, strongly supported the suspicion of a hydatid cyst in the differential diagnosis. However, one of the major limitations in the serological diagnosis of hydatid disease is false positivity, which can occur due to cross-reactions with other conditions. Parasitic diseases, cirrhosis, and liver or lung malignancies are among the conditions known to cause false-positive results.<sup>[4]</sup> In our case, the positive IHA result was likely a false positive secondary to the presence of a primary malignancy. In a meta-analysis, it was statistically shown that patients diagnosed before the age of 40, with smaller, non-metastatic PPC, had higher survival rates.<sup>[2]</sup> Therefore, PPC should be considered in the differential diagnosis of nodules, cavities, or cystic lesions in the

lung. Early diagnosis is crucial for enabling diagnosis at an early stage and improving survival outcomes. The diagnosis of PPC primarily relies on immunohistochemical marker positivity in pathological specimens, elevated  $\beta$ -HCG levels, and the exclusion of other potential primary sites through systemic examination. In a case similar to ours, a patient with a lung mass was initially diagnosed with lung carcinoma through fine-needle aspiration biopsy; however, subtype classification was not possible. A definitive diagnosis of choriocarcinoma was made following histopathological examination of the resected specimen.<sup>[5]</sup> Both the literature and our case suggest that immunohistochemical markers play a critical role in the diagnosis of PPC.

Due to its nonspecific clinical presentation, radiological resemblance to infections and other malignancies, and the frequent need for resection material for diagnosis, PPC is often misdiagnosed or diagnosed at a late stage.

### **Ethics Committee Approval**

This is a single case report, and therefore ethics committee approval was not required in accordance with institutional policies.

### **Informed Consent**

This study was conducted with the informed consent of the patient, who was fully informed about the nature of the research, as well as the potential risks and benefits involved. The patient's anonymity and confidentiality were strictly maintained throughout the study.

### **Conflict of Interest Statement**

The authors have no conflicts of interest to declare.

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We affirm that no artificial intelligence (AI)-assisted technologies, including large language models (LLMs), chatbots, or image generators, were used in the preparation of this submitted work.

### **Author Contributions**

Concept – R.D., S.F.E.A.; Design – S.F.E.A.; Supervision – R.D.; Resource – R.D.; Materials – R.D.; Data Collection and/or Processing – G.G.G.; Analysis and/or Interpretation – R.D., S.F.E.A.; Literature Review – S.F.E.A.; Writing – S.F.E.A.; Critical Review – R.D., G.G.G.

### **Peer-review**

Externally peer-reviewed.

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