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Case Report

Pleural capillary hemangioma mimicking traumatic hematoma: a rare case of massive hemorrhagic effusion

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ABSTRACT

Pleural capillary hemangiomas are exceptionally rare vascular tumors that may present with massive pleural effusion, often mimicking traumatic hematoma or malignancy. Due to nonspecific clinical features and inconclusive imaging, diagnosis is often delayed. We present a 52-year-old woman with progressive dyspnea and left-sided pleural effusion. Imaging revealed a pleural-based lesion, but malignancy was not confirmed by cytology or bronchoscopy. Video-assisted thoracoscopic surgery and histopathology confirmed capillary hemangioma. Surgical excision led to full recovery without recurrence. This case underscores the importance of considering benign vascular tumors in the differential diagnosis of unexplained pleural effusions.

Keywords: capillary hemangioma, pleura, pleural effusion, video-assisted thoracoscopic surgery, differential diagnosis

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Introduction

Pleural capillary hemangioma is a benign vascular neoplasm originating from the pleural surface and is rarely encountered in clinical practice. Hemangiomas are more commonly seen in superficial soft tissues but can occasionally arise within the thoracic cavity, leading to diagnostic challenges [1]. When located in the pleura, these tumors may present with nonspecific respiratory symptoms and imaging findings suggestive of hematoma or malignancy [2,3].

Due to their rarity and ambiguous radiologic appearance, pleural hemangiomas are frequently misdiagnosed or diagnosed late, often requiring histopathological confirmation through surgical excision [2]. In this report, we present a case of pleural capillary hemangioma that mimicked a traumatic hematoma due to hemorrhagic effusion and radiologic features, ultimately diagnosed and successfully treated via video-assisted thoracoscopic surgery (VATS).

Case Report

A 52-year-old woman with hypothyroidism presented with progressive dyspnea lasting two to three months. She had no history of trauma. Physical examination revealed diminished breath sounds in the lower left lung zone. Laboratory findings included leukocytosis (12,500/ μ L), hemoglobin of 12.3 g/dL, and elevated CRP (8.3 mg/L). Pleural fluid analysis revealed an exudate with a leukocyte count of 1,376/ μ L and the growth of *Staphylococcus aureus*. Thoracic CT revealed a 35 \times 20 mm heterogeneously dense lesion at the level of the left 5th–6th ribs with compressive atelectasis in the left lower lobe (Figure 1a). PET-CT demonstrated focal FDG uptake (SUVmax: 10.6) in the lingular segment after chest tube insertion and fluid drainage, likely related to chest tube–associated inflammation, while the pleural-based lesion on thoracic CT showed no FDG avidity (Figure 1b). However, retrospective evaluation suggested that this uptake may have been related to inflammation or the presence of the previously inserted chest tube, rather than the pleural lesion itself. The lesion identified on CT was not FDG-avid. Bronchoscopy and bronchoalveolar lavage cytology showed no abnormalities.

Due to the unresolved effusion and unclear etiology, VATS was planned. Under general anesthesia and sin-

gle-lung ventilation, approximately 800 mL of serohemorrhagic pleural fluid was aspirated. A 2 \times 2 cm cystic lesion was identified on the posterior thoracic wall at the level of the 6th intercostal space and was completely resected. Frozen section analysis indicated lymphoid tissue. Partial decortication was performed, and a 32 Fr chest tube was inserted.

The patient was discharged on the 5th postoperative day without complications. During follow-up, she reported no recurrence of dyspnea. Follow-up thoracic CT showed no pleural effusion and complete resolution of atelectasis. Sections prepared from the thoracic tissue specimen showed a lesion characterized by proliferation of thin-walled vascular structures within soft tissue areas partially covered by mesothelial lining, alongside a fibrinous blood clot. Erythrocytes were observed within some vascular lumens. To support the diagnosis, immunohistochemical analysis was performed using CD31, ERG, Fli-1, CD34, WT-1, pancytokeratin, PHH3, and Ki67. These findings were consistent with a capillary hemangioma. (Figures 2a-c). As of the most recent follow-up in April 2022, the patient remained clinically stable with no evidence of recurrence.

Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

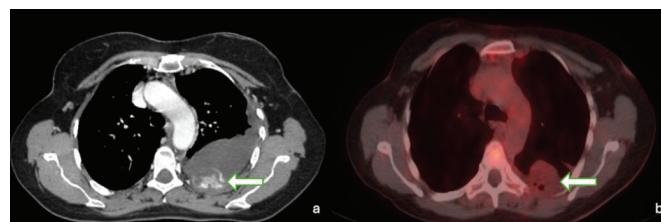


Figure 1. Axial thoracic CT (a), and fused PET-CT (b) images showing a pleural-based lesion (green arrows) adjacent to the left posterior thoracic wall. The lesion is visible on both modalities; however, no significant FDG uptake was detected in PET-CT.

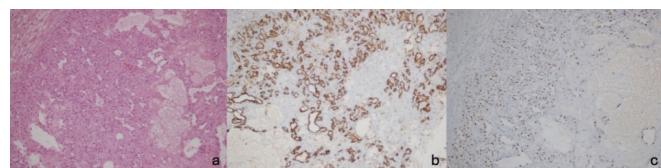


Figure 2. Histopathological appearance of the excised lesion. Lobular proliferation of thin-walled capillary vessels in fibrous stroma (H&E, $\times 200$) (a), immunohistochemical positivity for CD31 (b), and ERG (c) confirms vascular endothelial origin ($\times 200$ each).

Discussion

Capillary hemangiomas are benign vascular neoplasms composed of capillary-sized blood vessels lined by a single layer of endothelial cells. While commonly seen in the skin and mucosa, their presentation within the thoracic cavity, especially from the pleural surface, is exceedingly rare. The pathogenesis of pleural hemangiomas remains unclear but may involve congenital vascular malformations or acquired proliferative stimuli in the pleural microvasculature [1,2].

Clinical manifestations are often nonspecific. Patients may present with symptoms such as dyspnea, cough, or pleuritic chest pain due to unilateral pleural effusion, which can be serous or hemorrhagic. The etiology of such effusions is frequently misattributed to malignancy, infection, or trauma. In our case, despite the hemorrhagic nature of the effusion and hypermetabolic findings on PET-CT, repeated cytological examinations and bronchoscopic evaluations failed to establish a diagnosis, consistent with other reported cases where non-invasive modalities proved inconclusive [2,3].

Radiological findings also pose a challenge. CT may show pleural thickening or nodularity, while PET-CT can demonstrate increased metabolic activity. In our patient, the lingular lesion demonstrated intense FDG uptake, which typically raises suspicion for malignancy but can also be observed in benign vascular tumors due to high endothelial turnover [4]. Such overlap necessitates histopathological confirmation. In our case, although PET-CT showed increased FDG uptake, this did not correspond to the excised lesion. The uptake was likely attributable to reactive changes near the chest tube site, rather than the capillary hemangioma itself, which was not FDG-avid. This highlights a potential pitfall in interpreting PET-CT findings in the context of inflammation or recent interventions.

Surgical exploration remains both diagnostic and therapeutic. In this case, VATS allowed direct visualization and excision of the lesion, which appeared cystic and hemorrhagic. Frozen section analysis suggested lymphoid tissue, but definitive diagnosis required permanent sections. Histopathology demonstrated tightly packed capillary vessels, confirming a capillary hemangioma. In our case, immunohistochemical stain-

ing including CD31, CD34, ERG, Fli-1, WT-1, pancytokeratin, PHH3, and Ki67 supported the diagnosis of capillary hemangioma and excluded malignancy [3,5].

Embryologically, pleural capillary hemangiomas are thought to arise from congenital vascular malformations of mesodermal origin, although acquired proliferative changes in the pleural microvasculature have also been suggested [1,3]. Their true incidence is extremely low; to date, only a handful of cases have been published, mostly as single case reports or small series [6,7]. Because of this rarity and nonspecific presentation, pleural hemangiomas are often misdiagnosed as traumatic hematoma, empyema, or malignant pleural disease, resulting in delayed treatment [1,3].

Differential diagnosis of vascular tumors in the pleura includes more aggressive entities such as hemangiendothelioma, angiosarcoma, or even metastatic disease, especially when imaging features are ambiguous. In particular, epithelioid hemangiendothelioma can present similarly but carries a worse prognosis and typically requires systemic therapy [8].

Management depends on tumor resectability and clinical symptoms. Surgical excision is curative in most reported cases, with low recurrence and excellent prognosis [2,4].

The current case adds to the limited but growing body of literature describing pleural capillary hemangiomas as an important, albeit rare, cause of unexplained or recurrent pleural effusion. Our findings reinforce the value of early surgical intervention when noninvasive methods fail and highlight the importance of considering benign vascular tumors in the differential diagnosis of PET-positive pleural lesions.

Intraoperative VATS and macroscopic/wound photographs could not be obtained due to equipment malfunction at the time of surgery; this absence is a limitation of the present report.

In conclusion, pleural capillary hemangioma should be considered in the differential diagnosis of recurrent or massive pleural effusion of unclear etiology, particularly when malignancy or trauma are excluded. Surgical excision is both diagnostic and therapeutic, with favorable outcomes. Early recognition and multidisciplinary evaluation are essential for appropriate management.

Declaration of conflicting interests

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Authors' contribution

CI: Conceptualization, Methodology, Supervision, Writing – original draft, Writing – review & editing. MGP: Writing – original draft, Visualization, Writing – review & editing. FBD: Data curation, Investigation, Writing – original draft. YS: Histopathological analysis, Immunohistochemical evaluation, Validation. BC: Validation, Supervision, Writing – review & editing.

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