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Castleman Disease: Atypical Cause of Pneumonectomy

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Figure 1. (A) Preoperative chest radiography revealed a left perihilar lesion, (B) chest computed tomography showed a rounded left perihilar lung mass, well-defined, with 45 mm × 40 mm and slight contrast uptake, (C) hematoxylin and eosin staining showed regressed germinal centers (arrows) and (D) Follicular dendritic cell prominence, surrounded by mantle zones containing small lymphocytes arranged in a concentric pattern.

Case Description

A previously healthy 24-year-old male, smoker (4 pack-year), presented with a traumatic fracture of the right humerus. Preoperative chest radiography revealed a left perihilar lesion (Figure 1A). He had no respiratory or constitutional symptoms. The physical examination was unremarkable. Laboratory data, including neuron-specific enolase and chromogranin, were normal. Chest computed tomography showed a rounded left perihilar lung mass, well-defined, with 45 mm × 40 mm and slight contrast uptake (Figure 1B). Flexible bronchoscopy revealed signs of extrinsic compression of the left main bronchus, with hypervascularization and widening of the dividing spurs. Endobronchial biopsies and brushing were negative for malignant cells.

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Positron emission tomography revealed increased fluorodeoxyglucose-F18 uptake (SUVmax: 5, 4) in the left hilar lesion. 68Ga-DOTA-NOC PET-CT also showed an abnormal uptake from the nodular formation in left pulmonary hilum, suggesting a neuroendocrine tumor. Endobronchial ultrasound-guided transbronchial needle aspiration (EBUS-TBNA) was performed with punction of left mass; TBNA samples were negative for malignancy. Lung function was normal. He was therefore referred for evaluation of thoracic surgery.

Given the tumor central location with major pulmonary vessels and main bronchus involvement, he underwent left pneumonectomy. Hematoxylin and eosin staining showed regressed germinal centers (Figure 1C, (arrows)) with follicular dendritic cell prominence, surrounded by mantle zones containing small lymphocytes arranged in a concentric pattern (Figure 1D). Microscopic features and immunostaining were consistent with Castleman Disease– Hyaline Vascular Variant. The patient received no further therapy, maintaining regular surveillance.

Unicentric Castleman Disease (UCD) frequently presents as an incidental solitary mediastinal mass, however, intrapulmonary location with hilum involvement is rare. The preoperative diagnosis can be challenging as clinical and radiological findings are nonspecific [1,2]. The standard treatment for UCD is complete surgical resectio [3]. This case emphasizes that although hilar-presentation UCD is a rare and benign condition, anatomic resection and even a pneumonectomy may be required for diagnostic and therapeutic purposes.

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