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A Rare Case of Multiple Endobronchial Hamartoma

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Abstract
Pulmonary hamartomas are rare benign tumors, with a prevalence of 0.25% in the general population, accounting for approximately 8% of all benign lung neoplasms. Only 10% of pulmonary hamartomas are endobronchial in location, while the rest are peripheral. We report the case of a 50-year-old woman presenting with right basal thoracic pain lasting four months, associated with exortional dyspnea and dry cough, all within a context of general health deterioration. Thoracic CT revealed a calcified mediastinal-pulmonary lesion in the right lower lobe, causing lobar atelectasis. Flexible and bronchoscopy revealed a tumor bud bridging the orifice of the right lower lobe and the middle lobe bronchus (fowler), associated with infiltrative stenosis. The diagnosis of endobronchialhamartoma was confirmed after pneumonectomy. The postoperative course was favorable, with significant clinical improvement after one year.

Introduction

Pulmonary hamartomas are benign tumors arising from peribronchialmesenchymal tissue and composed of variable proportions of cartilage, adipose tissue, connective tissue, respiratory epithelium, and smooth muscle.Clinical manifestations are non-specific and depend on tumor size and location.Hamartoma is the most common benign tumour of the lung. However, endobronchialhamartoma are rare. Definitive diagnosis requires histopathological confirmation, and surgical intervention remains the standard of care.[1]

Case Report

A 50-year-old woman, chronically exposed to wood smoke, presented with a four-month history of right basal thoracic pain, exortional dyspnea, and dry cough. Her symptoms were accompanied by febrile sensations, night sweats, and general fatigue. Physical examination revealed signs of pleural effusion in the lower third of the right hemithorax. Chest X-ray showed a triangular right paracardiac opacity with elevation of the right diaphragmatic dome, on a retracted right hemithorax, along with a hyperdense image within the right bronchial tree. **(Figure 1)**

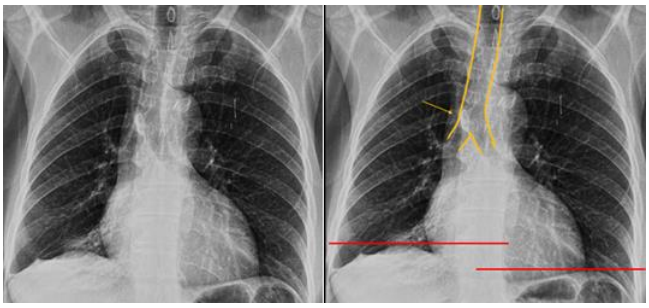

Figure 1: Chest X-ray showing triangular right paracardiac opacity with elevation of the right diaphragmatic dome, on a retracted right hemithorax, along with a hyperdense image within the right bronchial tree

CT scan identified a calcified mediastino-pulmonary mass involving the right lower lobe, associated with post-obstructive atelectasis.**(Figure 2)**

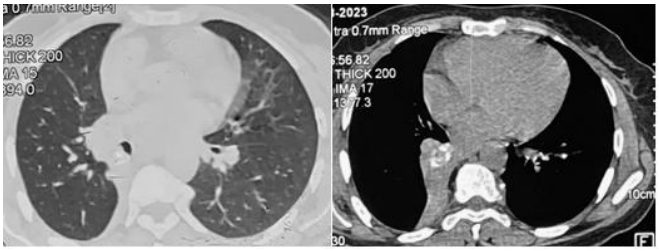

Figure 2: CT scan identified a calcified mediastino-pulmonary mass involving the right

lower lobe, associated with post-obstructive atelectasis

Flexible bronchoscopy revealed a smooth mucosal surface with a highly vascularized tumor bud extending across the orifices of the right middle and lower lobe bronchi, along with an infiltrative stenosis and an additional tumoral bud at the anterior segmental bronchial orifice of the right upper lobe. Bronchial biopsies showed non-specific fibro-inflammatory remodeling, and microbiological cultures were sterile. **(Figure 3)**

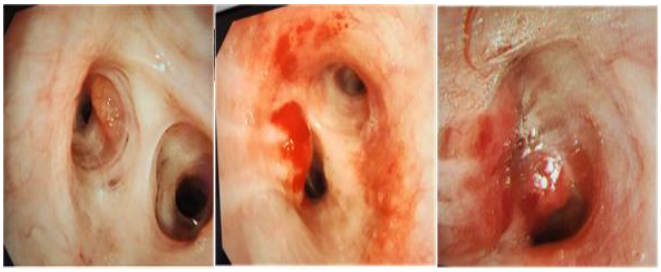

Figure 3: Flexible bronchoscopy revealed the presence of multiple tumoral buds, including one at the ventral opening of the right upper lobe bronchus with a smooth, vascularized surface, and another straddling the opening of the right lower lobe and the middle lobe (Fowler). An infiltrative stenosis of the right lower lobe bronchus was also noted, topped by a few smooth-surfaced buds, surrounded by spontaneously bleeding infiltrated mucosa.

Rigid bronchoscopy confirmed the same endoscopic findings. Histological examination of repeat biopsies again showed non-specific inflammation.
18F-FDG PET/CT revealed a hypermetabolic tissue mass in the posterobasal region of the right lower lobe with pleural adherence and associated of hypermetabolic lymphadenopathy involving the right upper and lower paratracheal, prevascular, right hilar, and subcarinal regions. **(Figure 4)** These findings are suggestive of active nodal involvement, possibly of malignant origin in the appropriate clinical context.

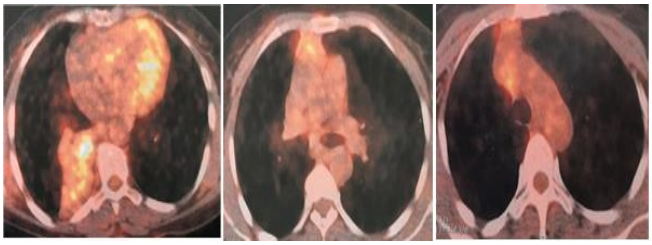

Figure 4: 18F-FDG PET/CT revealed a hypermetabolic tissue mass in the posterobasal region of the right lower lobe with pleural adherence and associated of hypermetabolic lymphadenopathy involving the right upper and lower paratracheal, prevascular, right hilar, and subcarinal regions

Given the diagnostic uncertainty and obstructive symptoms, the multidisciplinary team (MDT) meetingrecommended surgical management if mediastinoscopy fails to provide a definitive diagnosisis.
Mediastinoscopy demonstrated reactive adenitis, with no histological signs of malignancy.The patient underwent a right pneumonectomy via video-assisted thoracoscopic surgery (VATS) with right posterolateral thoracotomy, given that the pulmonary process extends beyond the major fissure, is proximal in location, and is in close contact with the right pulmonary artery and pulmonary veins, with apparent invasion of the inferior vena cava. Macroscopic examination revealed a 4-cm firm, whitish-beige tumor with calcific remodeling, located 0.5 cm from the bronchial margin**(Figure 5).**

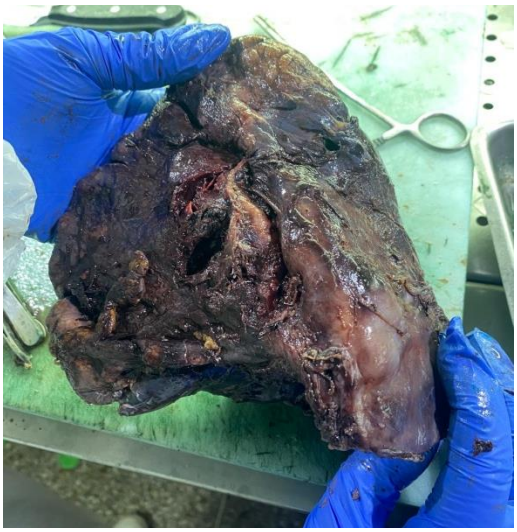

Figure 5: Right pneumonectomy specimen – macroscopic examination

Histological analysis demonstrated a mixture of hyaline cartilage, bone trabeculae with marrow

spaces, fibroadipose tissue, and striated muscle fibers—consistent with hamartoma. Multiple white nodules and a focus of bronchial dilatation measuring 8.5 cm were noted on the diaphragmatic surface. Lymph node dissection (stations 4R, 9R, 10R) confirmed reactive adenitis.

The final diagnosis was multiple endobronchialhamartomas. The uniqueness of this case lies in the exceptional central location of the lesion and its radioclinical mimicry of a bronchogenic carcinoma. Postoperative recovery was uneventful, with marked respiratory improvement.

Discussion

Pulmonary hamartomas are the most common benign pulmonary neoplasms. Most of these are, however, intra-parenchymal, with endobronchialhamartomas accounting for only 1.5% of all cases **[2]**. Endobrachialhamartomas frequently present with obstructive symptoms, including cough, dyspnea, wheezing, hemoptysis, recurrent infections, atelectasis, or localized bronchiectasis. **[3]**

Radiographic abnormalities include alveolar infiltrates (30%), atelectasis (25%) as a case of our patient , or both (22%). Chest radiographs may be normal in up to 14% of cases. On CT, an endobronchialhamartoma appears as an intraluminal mass, sometimes with calcifications and an adipose component, but without significant contrast enhancement. These features can aid differentiation from malignancy. **[4]**

However, PET/CT may show increased FDG uptake, making it challenging to differentiate hamartomas from malignancies. **[5]**

Bronchoscopic findings often suggest a benign lesion. Typically, the tumor is well-circumscribed, polypoid or pedunculated, with a smooth mucosal surface and no submucosal infiltration, usually located in proximal bronchi. Despite these features, biopsies are essential to rule out malignancy or other benign tumors, including carcinoid tumors. **[6]**

Treatment options include endoscopic modalities (laser photocoagulation, electrocoagulation, cryotherapy, argon plasma coagulation) or surgical resection (bronchotomy or pulmonary lobectomy/pneumonectomy).**[7]**Bronchoscopic resection might require multiple sessions, and has a relapse rate of 0% to 26.7%.Patients may require multiple sessions for complete removal as a significant recurrence rate is present, but is usually managed effectively by repeated bronchoscopic management. **[8]**

Surgery remains the only definitive curative option available. In the event of surgery, preservation of functional lung tissue is the primary goal. Therefore, enucleation and wedge resections are the most common surgical choices, with more radical lobectomy or total pneumonectomy reserved for intense lesions, multiple or large lesions that make wedge resection impossible, or lesions adhering severely to the hilum of the lung. **[9]**

The prognosis for patients with lung hamartoma is generally excellent. Lesions are slow-growing, and in cases where symptoms are present and persistent, surgery is curative. Malignant transformation or subsequent malignancy are rare occurrences, and if patients adhere to a conservative observation schedule, malignant growths are likely to be diagnosed early on. **[10, 11]**

Conclusion

Pulmonary hamartomas are benign lesions that may pose diagnostic challenges when located endobronchially. Early diagnosis using bronchoscopy and histological examination is essential to avoid unnecessary major surgery. While endoscopic management is preferred, surgical resection remains a valid option in symptomatic or diagnostically uncertain cases. The prognosisremains favorable, especiallywithcompleteresection.

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