Peripherally located endobronchial hamartoma mimicking aspergilloma: a case report

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Abstract: We herein report the case of a 75-year-old man with a pulmonary hamartoma that mimicked aspergilloma on chest computed tomography (CT). A CT scan performed to assess an asymptomatic lesion detected on a screening chest radiograph showed a 1.3-cm diameter nodule with an air crescent sign in the left lower lobe. A diagnosis of aspergilloma was made and the patient treated with an antifungal agent for 1 year, following which he underwent radical surgery because of failure of the radiologic lesion to resolve. Pathologic examination of the resected specimen showed an endobronchial hamartoma within the B9 periphery. Peripherally located hamartomas can develop within the peripheral bronchi resulting in an air crescent appearance on radiological images.

Keywords: Air crescent sign; endobronchial pulmonary hamartoma; aspergilloma

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Introduction

Hamartomas, one of the commonest benign pulmonary tumors, are categorized as centrally-located endobronchial and peripherally located pulmonary types. We herein report a case of a hamartoma originating from a peripheral bronchus that resembled an aspergilloma on a computed tomography (CT) scan.

Case presentation

An asymptomatic 75-year-old man was referred to our hospital for assessment of an abnormal shadow detected by screening chest radiography. A chest CT scan revealed a 13-mm diameter nodule with an air crescent sign in the posterior basal segment of the left lung (*Figure 1*). [^{18}F]-2-fluoro-2-deoxy-D-glucose positron emission tomography (FDG-PET)-CT revealed no significant uptake within the nodule. The serum concentration of β-D glucan was within normal limits; however, the serum concentration of aspergillus antigen was 0.5 (cut off index; reference value 0.5). Trans-bronchial biopsy and brushing cytology showed no malignant cells. Cultures for acid-fast bacilli and fungi were negative. The patient underwent antifungal treatment with itraconazole for 1 year, during which the CT appearance of the nodule did not change. Therefore video-assisted radical left lower lobectomy was performed and was successful.

Macroscopically, the target lesion was a well-defined 13-mm diameter white mass within the fifth branch of the dilated posterior basal segmental bronchus of the left lung. A pedicle of the lesion was identified in the dilated bronchial wall (*Figure 2*). Microscopically, the lesion consisted of chondroid tissue covered with bronchial-type epithelium and was diagnosed as a chondromatous hamartoma (*Figure 3*). The dilated bronchi were rich in goblet and smooth muscle cells. No fungal elements were detected within the lesion.

The patient was doing well without any complications 3 years after surgery.

Discussion

Pulmonary hamartomas are the most common benign tumors of the lung, comprising 41.9% of reported lung

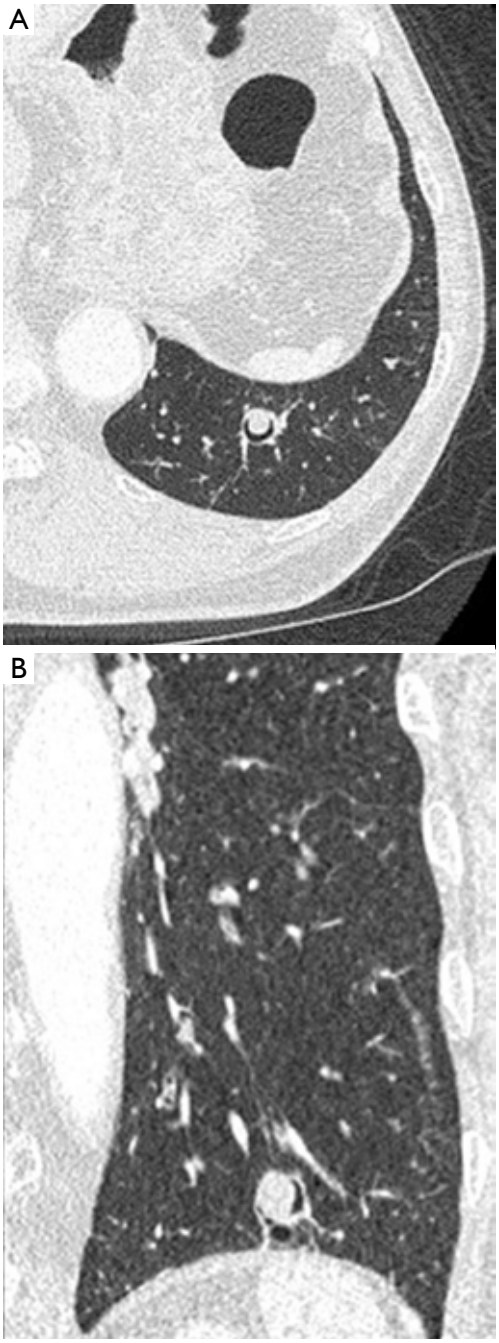

Figure 1 CT image showing a nodule with an air crescent sign is in the left S9. CT, computed tomography.

Figure 2 Cross-section of left lower lobectomy showing a well-defined white tumor 13 mm in diameter within the dilated peripheral side of left B9 (red arrow).

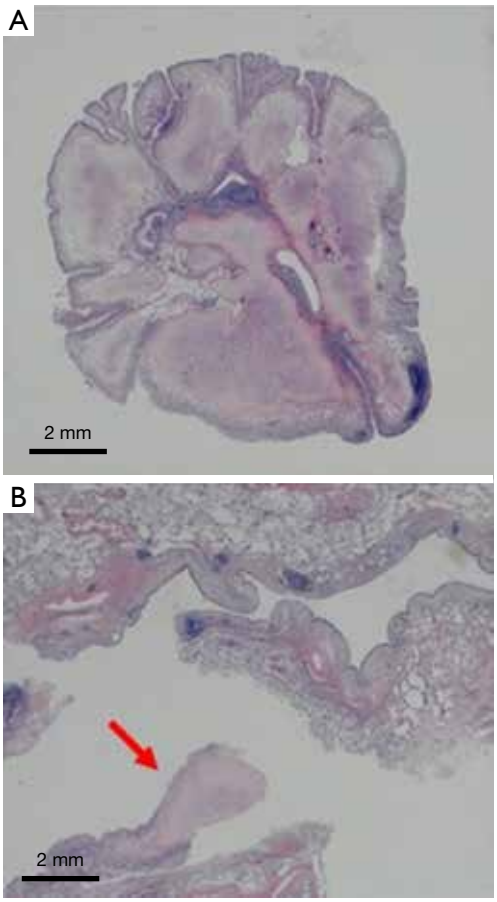

Figure 3 Photomicrographs of hematoxylin-eosin stained section of the resected lesion showing a hamartoma (A) with a pedicle (red arrow) arising from the bronchial wall (B).

tumors (1). Pulmonary hamartomas are classified as peripherally located pulmonary and centrally located endobronchial types. Endobronchial hamartomas occur less frequently, accounting for 2.5–19.5% of hamartomas (2). Peripheral endobronchial hamartomas are very rare. A search of PubMed using the key words of pulmonary hamartoma and/or endobronchial hamartoma identified a few reports (3-5). One of these articles reported a 35-mm diameter endobronchial tumor that had developed in the anterior superior segment of the right lung adjacent to pleura (3) and another a 20-mm diameter endobronchial tumor in the posterior basal segment of the right lung (4).

In the latter case, a radical right upper lobectomy was performed because of a misdiagnosis of lung cancer that was based on enlargement of the hilar lymph nodes.

On CT imaging, pulmonary hamartomas typically appear as well-defined solitary pulmonary nodules in a peripheral location; 34–50% of them contain fat and 15–30% calcification. In our case, a chest CT scan showed an air crescent sign without any of the above features.

An air crescent sign on a radiologic image is the result of collection of air around a pulmonary nodule. Well-demarcated pulmonary tumors such as sclerosing hemangiomas and solitary fibrous tumors can show the air crescent sign, which indicates that air has been trapped around the tumor (6,7). The air crescent sign is typically caused by an aspergilloma located within a preexisting thin-walled cavity with a smooth inner margin, most often formed by a previous tuberculous infection in the upper lobe. Lung cancer, tuberculosis, hydatid cyst, cystic bronchiectasis filled with mucus plugs, and bacterial lung abscess may also cause air crescent signs with or without aspergillus infection (8,9). Pulmonary aspergillomas are typically positive on PET-CT, but may be negative (10). In our case, the lesion was misdiagnosed as an aspergilloma based on these clinical features, leading eventually to radical lobectomy. Such lesions should be excised by radical surgery to obtain a definite diagnosis.

In conclusion, physicians should note that peripherally located hamartomas can develop within the peripheral bronchi resulting in an air crescent appearance on radiological images.

Acknowledgements

None.

Footnote

Conflicts of Interest: The authors have no conflicts of interest to declare.

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