Clinical Case Report

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Endobronchial hamartoma mimicking malignant lung tumor contralateral endobronchial metastasis

A case report

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Abstract

Rationale: Endobronchial hamartoma, the most common benign lung tumor, is located in the bronchus, and it easily mimics lung cancer or bronchial metastasis. Endobronchial hamartoma can cause coughing, hemoptysis, and pulmonary infection; thus, it should be treated right away by surgery or fiberoptic bronchoscopy.

Patient concerns: We report a rare case of endobronchial hamartoma in which the clinical symptoms and imaging overlapped strongly with malignant lung tumor contralateral endobronchial metastasis.

Diagnoses: Endobronchial hamartoma coexisting with a malignant lung tumor.

Interventions: Fiberoptic bronchoscopy was conducted, and the pathologic diagnosis was hamartoma. A second fiberoptic bronchoscopy was conducted, and fine-needle aspiration cytology of the enlarged lymph nodes indicated squamous cell carcinoma.

Outcomes: The clinical symptoms were relieved, and the treatment options were docetaxel, cis-dichlorodiamineplatinum, and endostatin.

Lessons: Fiberoptic bronchoscopy needs to be guided by imaging and can be considered an effective method for the diagnosis of endobronchial hamartoma.

Abbreviations: CT = computed tomography, CYFRA21–1 = Cytokeratin 19 fragment, NSE = 2-phospho-D-glycerate hydrolase, PET = positron emission tomography, SUV = standardized uptake value.

Keywords: endobronchial hamartoma, fiberoptic bronchoscopy, malignant lung tumor, PET/CT

1. Introduction

Lung hamartoma is the most common benign tumor.[1,2] Most cases occur in the periphery of the lung, presenting uneven density and a solitary pulmonary nodule in the radiographic images; however, 1.4% of lung hamartomas are located in the bronchus.[1] Pulmonary hamartomas can be classified into the central, peripheral, or mixed types, depending on the invasive site.[1,2] Peripheral pulmonary hamartoma is asymptomatic, but endobronchial hamartoma, belonging to the first type, often causes atypical symptoms, including coughing, hemoptysis, and pulmonary infection.[1,2] Therefore, peripheral pulmonary hamartoma is easily misdiagnosed as the central type carcinoma of the lung or lung metastasis of a malignant tumor, which should urgently be treated by surgery or fiberoptic bronchoscopy.[1,2] Herein, we report a rare case of endobronchial hamartoma that coexisted with a malignant lung tumor and mimicked contralateral endobronchial metastasis, based on radiographic images, laboratory tests, and clinical symptoms. Presenting this case will improve the current understanding of endobronchial hamartoma.

2. Case report

A 63-year-old man with a smoking history of 20 years was admitted to our department, owing to a week-long history of chest tightness with fever, cough, and sputum. He was primarily diagnosed with pulmonary lesions in a local hospital. The physical examination was normal, and the tumor markers 2-phospho-D-glycerate hydrolase (NSE) and Cytokeratin 19 fragment (CYFRA21–1) were present at 17.13 and 4.07 ng/mL, respectively. The chest computed tomography (CT) scan (Fig. 1A–D) revealed a 34 mm × 33 mm crumby-shaped soft tissue density in the upper right lobe of the right lung with a hole-like shadow, short burrs, and pleural depression. Multiple enlarged lymph nodes could be seen in the right hilar and mediastinum. In addition, a nodule could be seen in the upper left bronchus of the left lung. We considered the diagnosis of

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SSC and HZ contributed equally to this work.

Written informed consent was given by the patient.

The authors report no conflicts of interest.

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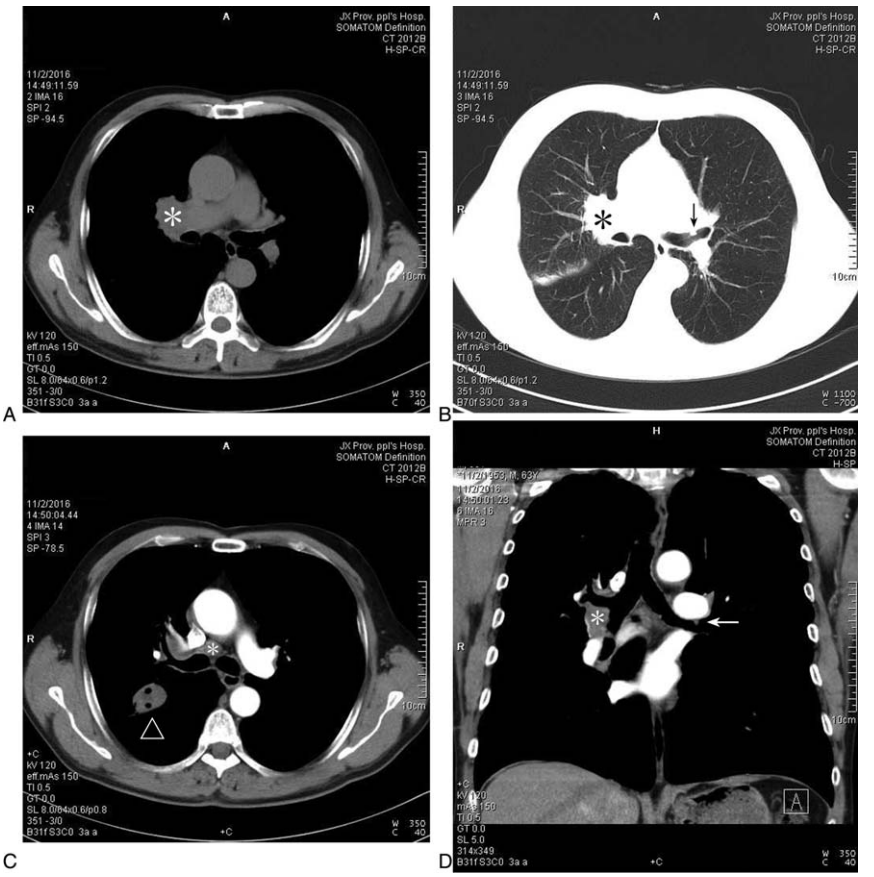

Figure 1. Chest CT revealed a crumby-shaped soft tissue density in the upper right lobe of the right lung (arrows), swollen lymph nodes in the hilar and mediastinum (asterisk), and a "neoplasm," with a smooth surface and normal mucosa breaking into the upper left bronchus in the left lung (arrowheads) (shown in A, B, C and D).

malignant lung tumor with ipsilateral hilar, mediastinal lymph node, and contralateral bronchial metastasis. Next, a fiberoptic bronchoscopy was conducted, which revealed a smooth "neoplasm" in the upper left bronchial (Fig. 2A). Interestingly, the whole "neoplasm" was clamped at once during the process of bronchoscopic biopsy (Fig. 2B, C). The pathologic diagnosis of the "neoplasm" was hamartoma (Figure 2D). To further clarify the diagnosis, a PET/CT examination was conducted that showed a 45 mm × 25 mm mass, high-density shadow, and multiple enlarged lymph nodes, with high uptake in the right lung (Fig. 3A–D). To identify the pathological type, a second fiberoptic bronchoscopy (Fig. 4A) was conducted, and fine-needle aspiration cytology of the right middle bronchus (Fig. 4B) indicated squamous cell carcinoma (cytology) (Fig. 5). The final diagnosis was endobronchial hamartoma coexisting with lung squamous cell carcinoma. The treatment options were docetaxel, cis-dichlorodiamineplatinum, and endostatin.

3. Discussion

Pulmonary hamartoma, accounting for 75% of benign lung tumors, can occur anywhere in the lung at any age, but especially between 40 and 60 years old, and the male-to-female ratio is 2 to 4:1.[1,2] Congenital malformation, normal hyperplasia, tumorigenesis, and inflammatory changes are hypothesized to underlie the pathogeny, but the cause remains to be elucidated.[2]

Pulmonary hamartomas originate from undifferentiated mesenchymal cells in the bronchial submucosa tissue.[2] They can arise from cartilage, connective tissue, smooth muscle, and other tissue types.[2] The imaging characteristics depend on the content and distribution of cartilage, fatty, and fibrous tissue in the hamartoma.[1,2] The typical imaging of a pulmonary hamartoma shows calcification and/or adipose tissue in the lesion, and the diagnosis depends on pathological examination because it is difficult to make a definitive diagnosis by imaging alone.[1,2]

Endobronchial hamartomas are located in the primary bronchi, lobar, or segmental bronchus.[1,2] The clinical symptoms of endobronchial hamartoma depend on the location and the severity of the obstruction.[1,2] When the tumor breaks into the lumen in a polyp-like manner, it may stimulate mucous membranes and block a secondary infection, which can cause atypical symptoms, including coughing, sputum, fever, chest pain, hemoptysis, and other symptoms exhibited in this case report.[1–3] The differential diagnosis, including bronchial tuberculosis, bronchial lipoma, bronchial chondroma, and even central lung cancer and bronchial metastasis, could all show the same symptoms as endobronchial hamartoma.[1–4] As reported in this case, endobronchial hamartoma with a malignant lung tumor is rarely reported, and it mimics bronchial metastasis, based on radiographic images, laboratory tests, and clinical symptoms.

The traditional treatment is surgery, including tracheotomy, bronchialectomy, lobe bronchoplasty, and lobectomia pulmonalis, and the postoperative recovery is affected by surgical trauma and high cost.[5,6] In recent years, transbronchoscopic

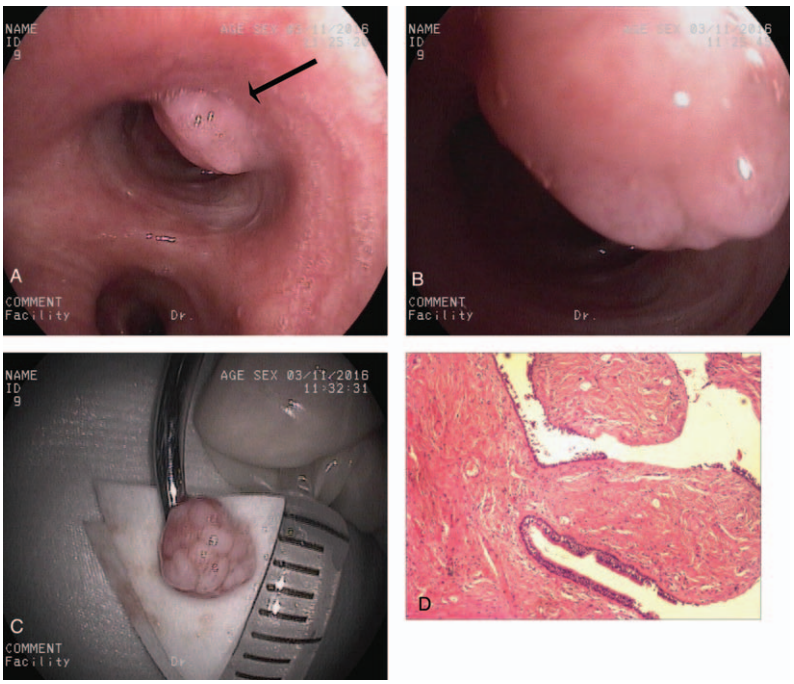

Figure 2. Endobronchial hamartoma (arrows) was moved by fiberoptic bronchoscopy (shown in A, B, and C). The pathologic diagnosis of the "neoplasm" was hamartoma, presenting with irregular arrangement of bone tissue, fibrous tissue, and adipose tissue, viewed under a microscope (shown in D) (hematoxylin and eosin stain, original magnification ×10).

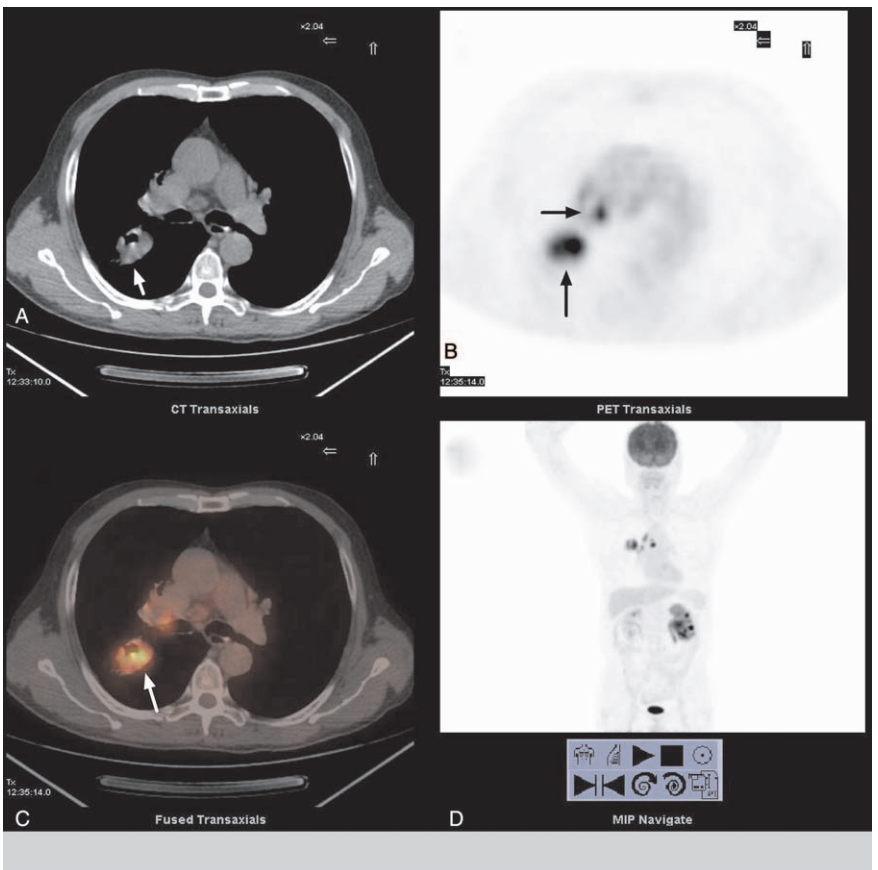

Figure 3. PET/CT scan showed a 45 mm × 25 mm mass, high-density shadow, behaving as a cavity and burrs with high uptake in the right lung (SUVmax10.6) (arrows). Multiple enlarged lymph nodes could be seen in the right pulmonary hilum and mediastinum with high uptake (SUVmax11.6) (arrows) (shown in A, B, C, and D).

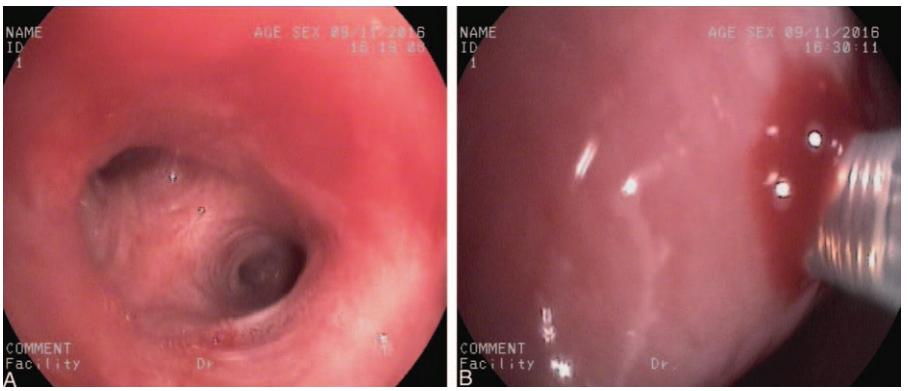

Figure 4. The second fiberoptic bronchoscopy image (left upper bronchus) (A) and fine-needle aspiration cytology (B).

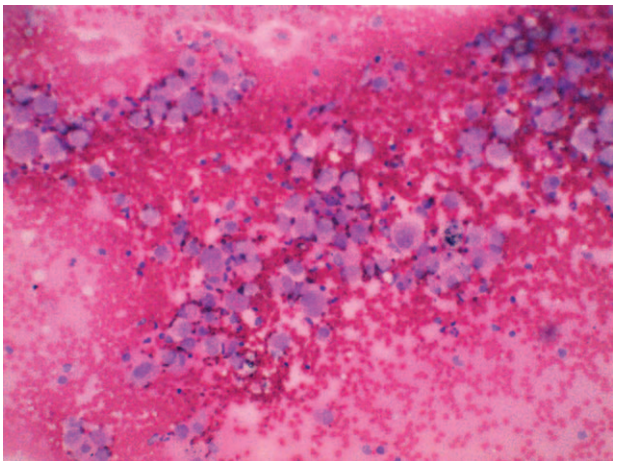

Figure 5. Fine-needle aspiration cytology in right hilar lymph nodes indicated squamous cell carcinoma (tendency) (original magnification ×20).

interventions, including snare excisions, laser therapy, cryotherapy, and argon plasma coagulation, have become the major therapeutic options in view of their high safety, reduced trauma, low cost, and fewer complications.[7,8] These are the best choices for patients with poor pulmonary function and intolerance to surgery, despite pneumothorax complications.[7,8] Interestingly, recurrence and malignancy after resection are rarely reported for endobronchial hamartoma, but close follow-up is still necessary.

4. Conclusion

We reported a very rare endobronchial hamartoma coexisting with a malignant lung tumor. Given that imaging and clinical symptoms cannot always clearly differentiate between endobronchial hamartoma and bronchial metastatic nodules, fiberoptic bronchoscopy needs to be guided by imaging and can be considered an effective method for the diagnosis of endobronchial hamartoma.

References

[1] Gjevre JA, Myers JL, Prakash UB. Pulmonary hamartomas. Mayo Clin Proc 1996;71:14–20.
[2] Cosio BG, Villena V, Echave-Sustaeta J, et al. Endobronchial hamartoma. Chest 2002;122:202–5.
[3] El-Kersh K, Perez RL, Gauhar U. A 63-year-old man with a chronic cough and an endobronchial lesion. Diagnosis: endobronchial hamartoma. Chest 2014;145:919–22.
[4] Nussbaumer-Ochsner Y, Rassouli F, Uhlmann F, et al. Endobronchial lipoma mimicking bronchial carcinoid tumour. Thorax 2015;70:809.
[5] Wang T, Liu Y. Outcomes of surgical treatments of pulmonary hamartoma. J Cancer Res Ther 2016;12:116–9.
[6] Abdel HS, Elbastawisy SE, Hassaballa AS, et al. Is surgical resection superior to bronchoscopic resection in patients with symptomatic endobronchial hamartoma? Interact Cardiovasc Thorac Surg 2017;24:778–82.
[7] Wang JW, Huang M, Zha WJ, et al. [Flexible bronchoscopic intervention for endobronchial hamartoma]. Zhonghua Jie He He Hu Xi Za Zhi 2013;36:963–7.
[8] Kim JH, Jeong GM, Park KH, et al. Large endobronchial hamartoma successfully resected by snare through flexible bronchoscopy. Ann Thorac Surg 2015;100:1107–9.