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Successful use of snare electrocautery via flexible fiberoptic bronchoscopy for removal of an endobronchial hamartoma causing chronic lung atelectasis and mimicking malignancy

Chao Liu*, Jia-jia Wang*, Ye-han Zhu and Cheng Chen

***Abstract*:** We present a rare case of giant endobronchial hamartoma coexisting with lung atelectasis for more than 3 years. The small specimen initially biopsied via bronchoscope did not reveal tissue features, but some features were suspicious for squamous cell carcinoma. The lesion was removed completely using snare electrocautery combined with argon plasma coagulation via flexible fiberoptic bronchoscopy. The patient made a satisfactory recovery, and a pathological diagnosis was made. This could be a useful option in selected endobronchial tumors.

Keywords: electrocautery, hamartoma, lung atelectasis, snare

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Introduction

Pulmonary hamartoma represents an unusual and benign neoplasm of the lung in thoracic surgery.[1] Endobronchial hamartoma is a rare form comprising 3–10% of all hamartomas, and may cause airway obstruction, atelectasis and recurrent pneumonia. Most instances of pulmonary hamartoma can be removed by enucleation, wedge resection, or lobectomy.[2–3] It has been reported that benign endobronchial tumors can be successfully treated using bronchoscopy, usually rigid bronchoscopy.[4] We herein present an unusual case of chronic endobronchial hamartoma coexistent with lung atelectasis, which was successfully diagnosed and resected via interventional flexible fiberoptic bronchoscopy.

Case report

A 63-year-old male presented to the outpatient department with complaints of cough with sputum for the past 4 years. His history revealed hospital admission elsewhere for lung disease 3 years ago, when chest computed tomography (CT) examination and fiberoptic bronchoscopy showed an abnormality in the left upper lobe (Figure 1). However, there was no evidence of malignancy. The patient had no history of fever, dyspnea, hemoptysis, dysphagia, loss of appetite or weight loss. On general examination, there was absence of pallor, cyanosis, clubbing, edema and peripheral lymphadenopathy. All laboratory test results were in the normal range except the neuron-specific enolase level, which was increased to 30.27 ng/mL. Furthermore, chest CT was performed and showed a tightly endobronchial soft tissue with calcification located at the left hilum and total atelectasis of the left upper lung, which was enlarged compared with 3 and 4 years ago (Figures 2(a–c)). Also, a large mass was visible blocking the left upper lobe under bronchoscopy. Although biopsy was successful, pathological examination of the small specimen showed only histological inflammation and squamous metaplasia (Figure 2(d)). In view of the cytomorphological features suspicious for

Correspondence to:
Cheng Chen
Respiratory Department, The First Affiliated Hospital of Soochow University, 188 Shizi Street, Suzhou, 215006, China
chenchengatsd@sohu.com

Ye-han Zhu
Respiratory Department, The First Affiliated Hospital of Soochow University, 188 Shizi Street, Suzhou, 215006, China
zhuyehansz@163.com

Chao Liu
Jia-jia Wang
Respiratory Department, The First Affiliated Hospital of Soochow University, Suzhou, China

*These authors contributed equally to this work.

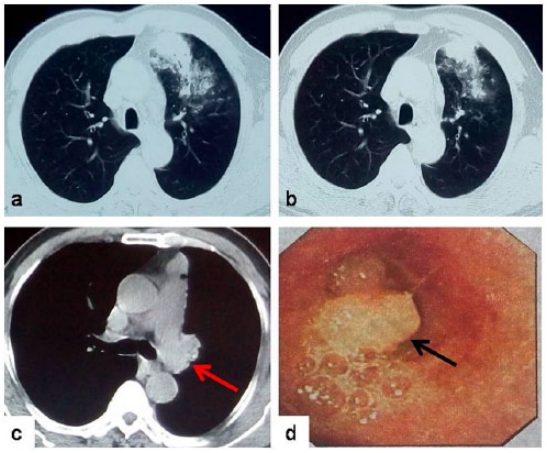

Figure 1. Chest CT examination: (a) June 2012; (b) and (c) June 2013. (d) Fiberoptic bronchoscopy (June 2013, arrow) showed the abnormality in the left upper lobe of the lung 3–4 years ago. In particular, a tightly endobronchial mass was located at the left hilus ((c), arrow).

squamous cell carcinoma and the radiological features, we performed a second, interventional bronchoscopy with snare electrocautery to focus on the protrusive lesion in the left upper bronchial stem. The protrusive lesion was removed completely, despite a little bleeding (Figures 2(e–f)). To prevent recurrence of the neoplasm, the mass root was cauterized with argon plasma coagulation (APC). No complications related to interventional bronchoscopy were observed. Final pathological evaluation of the entire mass was not identical to the initial diagnosis and confirmed the mass as a hamartoma (Figure 2(g)). No additional treatment was required, and the patient was discharged and followed at the outpatient clinic. Most importantly, chest CT examination after 2 months showed complete recovery of the abnormality in the left upper lobe (Figures 2(h–i)). The patient's condition remained stable for at least the following 12 months after interventional therapy.

Discussion

The majority of hamartomas are parenchymal and often located in the periphery of the lung – unlike the present patient, who had a more central lesion known as an endobronchial hamartoma. CT criteria for hamartoma include a diameter of 2.5 cm or less, a smooth edge and focal collections of fat or fat alternating with areas of calcification.[5] Calcification and areas of focal fat in the lesion have been found in some patients, which could be regarded as the diagnostic CT findings of pulmonary hamartoma.[6]

We have presented a case of giant endobronchial hamartoma coexisting with lung atelectasis for more than 3 years. Calcification in the lesion was detected via CT scan. An initial bronchoscopy showed a polypoid lesion within the left upper lobe. However, adequate pathological information could not be obtained via mucosal biopsy. Interventional bronchoscopy with snare electrocautery followed by APC therapy of the base not only resected the polypoid lesion completely but also facilitated pathological diagnosis. The tissue features of the endobronchial hamartoma could not be established from a small specimen; the entire lesion was required to exhibit the total structure of the mass.

Management of hamartoma should be individualized according to tumor size, anatomical distribution, growth rate and the performance status of the patient. For asymptomatic and slowly growing lesions less than 2.5 cm in diameter, recommended treatment is usually conservative with regular chest roentgenograms during follow up. By contrast, tumor resection is advocated for endobronchial, large and fast-growing lesions. Certain benign endobronchial tumors can be successfully treated via bronchoscopy, usually rigid bronchoscopy. Flexible fiberoptic bronchoscopy requires only local anesthesia and provides access to the more distal airways and the upper pulmonary bronchi. For these reasons, many diagnostic and therapeutic procedures have recently been performed using flexible bronchoscopy.[7]

In the present case, the tumor was local and slowly growing, but had protruded into the airway. Complete resection of the endobronchial hamartoma was achieved via snare electrocautery. Of note, to prevent recurrence of the neoplasm, the mass root was cauterized with APC. The patient's condition remained stable for at least the following 12 months after interventional therapy. Therefore, when a firm diagnosis is made preoperatively, surgical removal may not be necessary unless symptoms are present or tumor expansion is noted. In addition, electrocautery followed by APC therapy

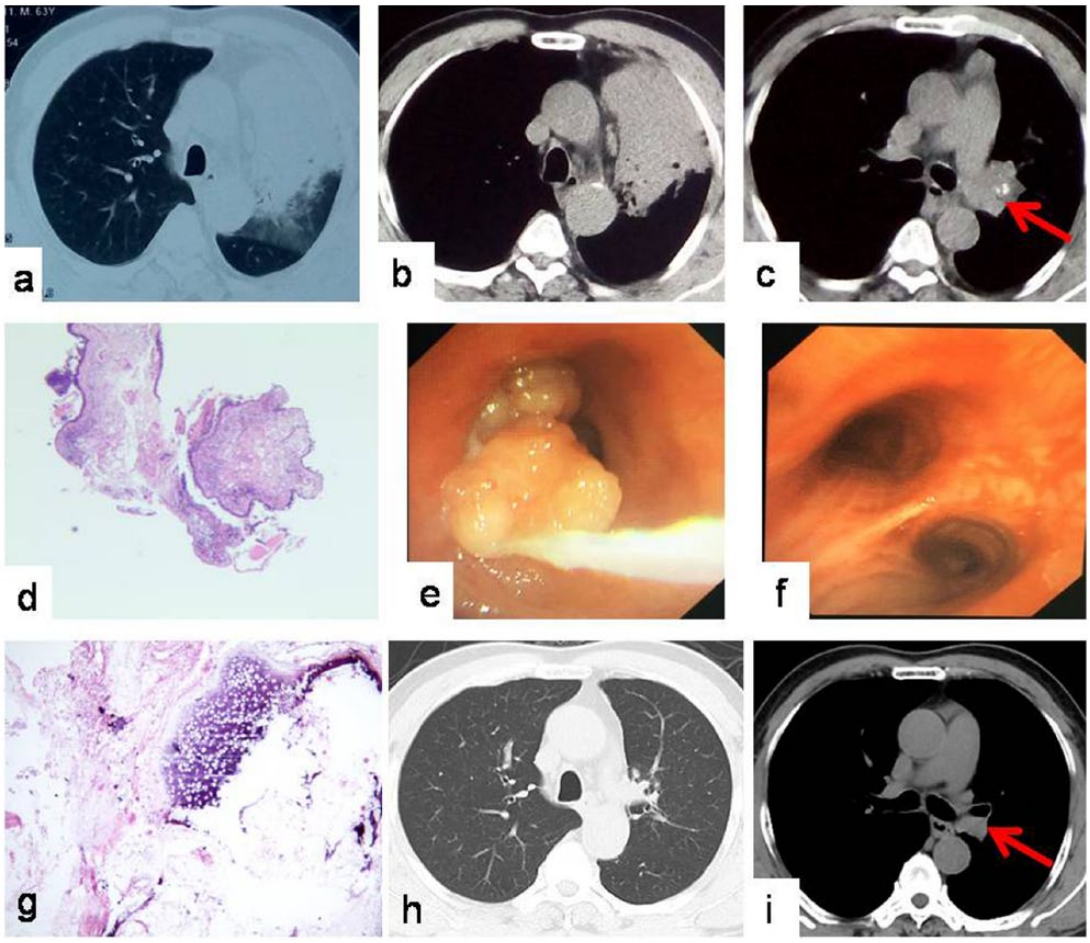

Figure 2. Chest CT examination (April 2016) showed the abnormality in the left upper lobe (a, b) and endobronchial mass containing calcifications (c). The small specimen biopsied by bronchoscope revealed histological inflammation and squamous metaplasia (d). The protrusive lesion located at the left upper lobe (e) was removed completely by snare electrocautery and APC (f). Pathological examination revealed a hamartoma (g). Chest CT examination after 2 months showed complete recovery of the abnormality in the left upper lobe of the lung (h, i).

may present an alternative therapy for resection of selected endobronchial hamartoma. However, in patients at high risk of hemorrhage or asphyxia, the snare procedure should be performed in an operating theater to allow for rigid bronchoscopy or surgical intervention if needed.

Consent

The patient and his legally authorized representative provided written informed consent for this case report to be published in a medical journal. The ethics and review committee of the First Affiliated Hospital of Soochow University approved the reporting of this case.

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Conflict of interest statement

The authors declare that there is no conflict of interest.

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