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Case Report

A rare cause of bronchial obstruction: Endobronchial hamartoma case report ☆

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ABSTRACT

Most of the pulmonary endobronchial lesions are malignant in origin. In rare instances, benign lesions such as endobronchial hamartoma may be the cause of the endobronchial tree obstruction. We present the case of a 57-year-old male patient from North Africa who presents with a history of a 5-month cough. Imaging, particulary CT scan, showed a mass on the right intermediate bronchus whose radiological characteristics are consistent with hamartoma. A biopsy of the mass obtained via bronchoscopy revealed chronic inflammation with no evidence for malignancy. The patient was treated surgically, and anatomopathology confirmed the diagnosis of hamartoma.

Introduction

Hamartomas represent the most common benign tumors of the lung [1,3,5,11,13,14]. They are usually located in the lung parenchyma and rarely involve the bronchus [1,3,11,12].

In the early stages of its development, hamartoma is usually asymptomatic, and then, depending on its size and location, it manifests signs of irritation such as persistent cough, as in our case, hemoptysis, or signs of obstruction such as dyspnea and recurrent pneumonia.

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Chest CT scans and bronchoscopy can suggest the diagnosis of endobronchial hamartoma [1]. The final diagnosis is made histologically [9].

These are various methods of successful resection of endobronchial hamartomas. The 2 most popular and successful techniques are endoscopic and surgical resection [1,2,15,16]. Other techniques that may be employed include cryotherapy, laser resection, electrocautery snare, argon plasma coagulation, or a combination of these techniques [2,17,18].

We report a rare case of endobronchial hamartoma causing bronchial obstruction.

Case presentation

A 57-year-old man consulted our hospital for a 5-month history of coughing. He had no associated dyspnea, chest pain, or hemoptysis. He had a history of smoking until 14 years ago. There was no familial history of respiratory disease.

There was no weight loss, hematemesis, hematochezia, rash, or mucosal bleeding. He denied a history of recent travel and had no history of tuberculosis exposure.

At that time, he was seen by his primary care physician, vital signs, as well as physical findings, were remarkable. The complete blood count, metabolic panel, and coagulation profile were normal.

A chest radiography was performed showing middle-lobe opacity. The patient received 10 days of antibiotic treatment with a partial remission of symptoms.

15 days after, the follow-up chest radiography showed persistence of the middle lobe opacity.

A CT scan was performed, showing indeed middle lobe consolidation (Fig. 1) and revealing a small, round, lesion in the right intermediate bronchus (Fig. 2). This lesion contained calcification and fat and showed low enhancement (Fig. 3).

The patient had a bronchoscopy the day after, which demonstrated a round lesion with a smooth surface and without hypervascularization, subtotally occluding the intermediate bronchus (Fig. 4). Multiple biopsies were done.

Histopathological examination of the biopsy showed chronic inflammation with no evidence of malignancy.

The patient was treated surgically, a posterolateral thoracotomy was performed, and the mass was removed.

Histological analysis of the removed tumor revealed mature benign tissue with various constituents, concording with the endobronchial hamartoma diagnosis.

The patient made an excellent postoperative recovery. His symptoms eventually resolved, and he continues to follow-up regularly with pulmonology.

Discussion

The most prevalent benign lung tumor is hamartoma [3,19]. The majority are located in the periphery of the lung, and only rarely of lung hamartomas occur in the bronchus [5,11,12]. Within the 154 cases of pulmonary hamartoma that Van den Bosch et al. [20] examined, only 12 of the lesions were endobronchial; the remaining lesions were peripheral.

Notably, malignant endobronchial cancers are the most common endobronchial lesions and surprisingly, the total amount of benign endobronchial tumors accounts for only approximately 2% of all lung tumors [3,21].

These benign tumors consist of different amounts of fibrous tissue, fat, cartilage, and entrapped respiratory epithelium [4].

Most of the pulmonary hamartomas are diagnosed during adulthood, with a peak incidence in the sixth to seventh decade and a male predominance [5,22].

Minalyan [3] who performed a literature search of all case reports and series of patients with endobronchial hamartoma, found that the average age of patients affected was 56. Our patient was 57 years old.

Most of the endobronchial hamartomas are asymptomatic and are discovered incidentally on imaging. However, those that are larger may present with symptoms of airway irritation or obstruction. The symptoms most commonly described by patients include productive cough and shortness of breath. The duration of symptoms before the diagnosis was found to be extremely variable (ranging from 5 days to 15 years) [3,23]. In our case, the patient had 5-month history of coughing.

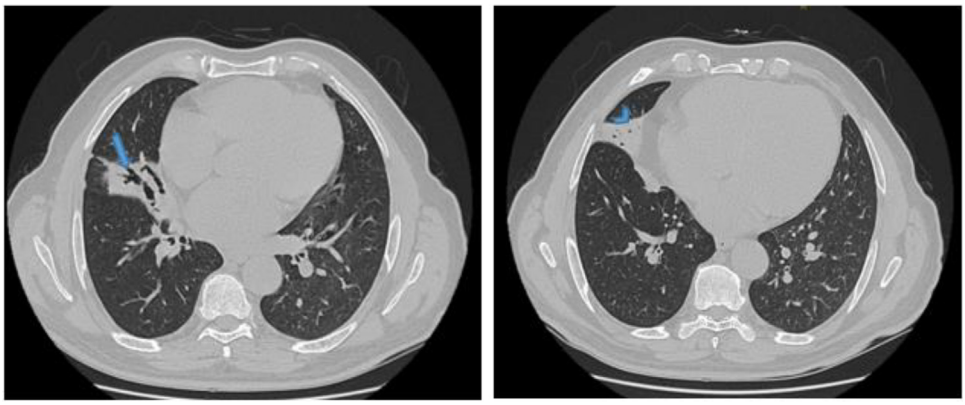

Fig. 1 – Computed tomography of the chest-lung window: Right middle lobe consolidation (Arrow head) associated with bronchiectasis (Arrow).

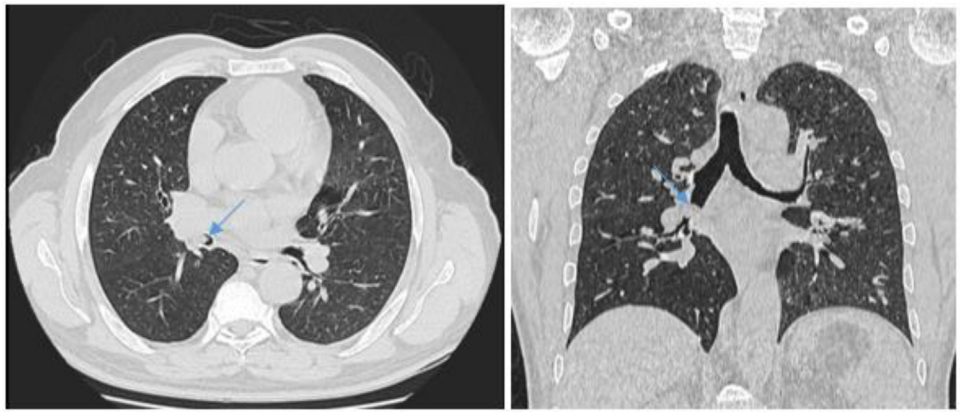

Fig. 2 – Computed tomography of the chest- lung window: Endobronchial lesion in the right middle lobar bronchus (arrow) causing atelectasis.

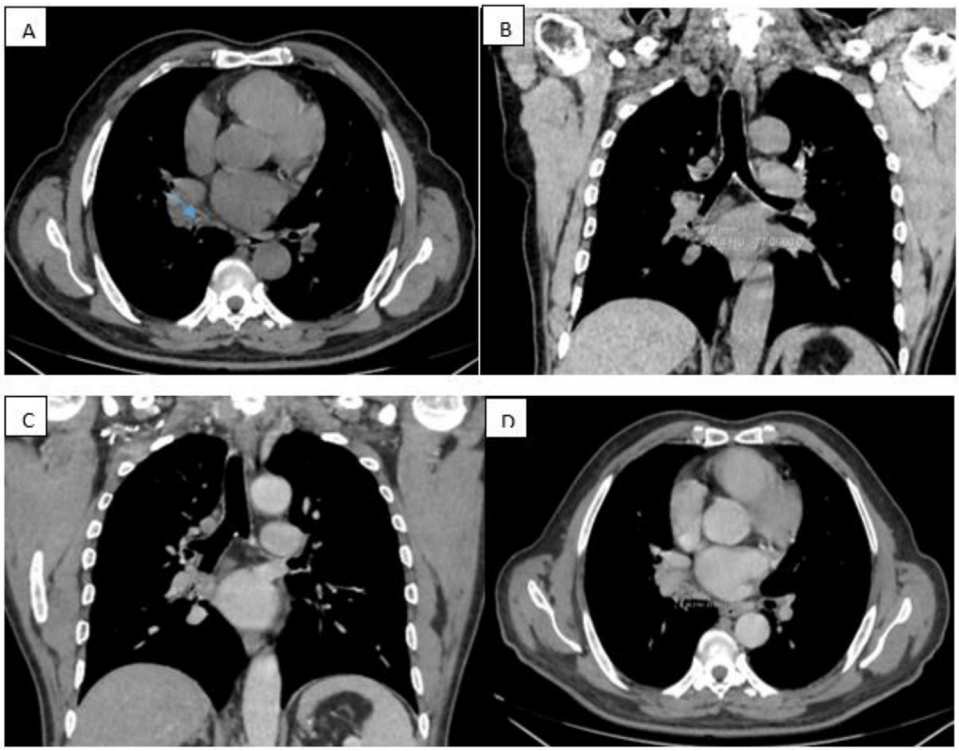

Fig. 3 – Computed tomography of the chest- mediastinal window: The endobronchial mass contain calcification (arrow, A), and fat (B) with low enhancement (C-D).

Endobronchial hamartoma typically contains fat, calcification, or a combination of both [6]. A chest X-ray's sensitivity is limited, and it frequently yields normal results. However, in individuals who are symptomatic, it may occasionally reveal post-obstructive alterations such as atelectasis and pleural effusion [10,13]. The presence of calcifications (typically popcorn type) can suggest the diagnosis [7]. In the Van den Bosch et al. [20] series segmental atelectasis was the most common finding. Our patient initially had a chest radiograph, which showed middle-lobe atelectasis.

CT scan has far superior sensitivity than a chest X-ray for detecting endobronchial hamartoma. Usually, it demonstrates rounded soft tissue masses that frequently exhibit calcification that clump throughout the lesion in the popcorn configuration and fat density [7,24]. In our case, it revealed a small, round, lesion containing calcification and fat (Figs. 2 and 3).

On endoscopy, this tumor appears as a central, well-circumscribed, polypoid, sessile, or pedunculated lesion with a smooth yellowish surface and no signs of submucosal infiltration [8,28,24]. A bronchoscopy was performed on our patient

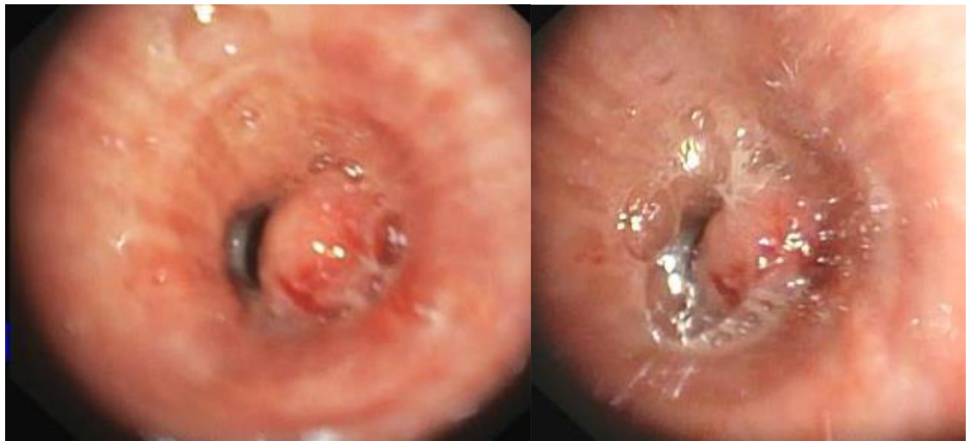

Fig. 4 – Bronchoscopic view of a well-circumscribed, yellowish polypoid mass obstructing the right intermediate bronchus lumen.

which revealed, a smooth mass occluding the right intermediate bronchus lumen.

Histological examination is necessary for a definitive diagnosis [9]. In our case, the biopsy obtained via bronchoscopy showed chronic non-specific inflammation, and the histological analysis of the removed tumor revealed mature benign tissue with various components that concorded with the endobronchial hamartoma diagnosis.

The main differential diagnoses are other benign lesions such as bronchial tuberculosis, bronchial lipoma, leiomyoma, fibroma, chondroma and neurogenic tumors but also malignant airway tumors such as a carcinoid tumor and bronchial metastasis [11,5,25].

Treatment options for endobronchial hamartomas are numerous. Historically, the primary method of treating these lesions has been surgical resection using either wedge resection, lobectomy, or pneumonectomy [1,15,26].

Thanks to advancements in bronchoscopy, the gold standard for treatment has become rigid bronchoscopy with laser therapy, as it provides an excellent outcome. Cryotherapy and endobronchial laser resection are particularly useful for individuals who are not good candidates for surgery or who will not consent to surgery [2,17,18].

The features of each patient and each hamartoma, including the lesion's size, activity level, and hardness, must be taken into account while determining the appropriate course of treatment [2]. We report a surgically removed endobronchial hamartoma.

Endobronchial hamartoma has a usually good prognosis. Following removal, endobronchial hamartoma has extremely low chances of recurrence [3].

Conclusion

Although pulmonary hamartoma is the most frequent benign lung tumor, it is unusual for it to be localized in the endobronchi. Unlike the peripheral subtype, endobronchial hamartomas usually present with recurrent respiratory symptoms [3]. It cause bronchus obstruction resulting in symptoms of fever, cough, expectoration, wheezing, and dyspnea [4,27].

Although endobronchial hamartoma is benign tumor, it can cause irreversible post-obstructive pulmonary destruction, so early diagnosis and treatment are very important.

The first test performed is a chest X-ray. In general, it is unable to identify the endobronchial lesion; thereby, advanced diagnostic tests particularly CT scans are needed for further evaluation. A thorough examination of the endobronchial lesion is necessary, and the probability of endobronchial hamartochondroma must be taken into account, particularly in the event of endolesional fat and calcification. The preferred diagnostic technique for suspected endobronchial masses is bronchoscopy [4].

Endoscopic excision, wedge resection, or sleeve excision are conservative therapies for endobronchial hamartomas in the absence of chronic post-obstructive lung injury; lobectomy may be necessary in situations of recurrent pneumonia. The prognosis for endobronchial hamartoma is typically favorable. There is no consensus regarding the follow-up interval and its frequency [4].

Patient consent

Informed written consent was obtained from the patient for publication of the case report and all imaging studies.

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