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Right middle lobe obstruction associated with synchronous endobronchial carcinoid and aspergillosis

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Abstract

Pulmonary carcinoids originate from neuroendocrine cells of the lung and comprise 0.5%–5% of all lung malignancies. Endobronchial carcinoids are rare, low-grade malignant tumors that occasionally coexist with other infectious diseases, including tuberculosis and nontuberculous mycobacterial infection. We treated a 63-year-old woman who presented with a right middle lobe obstruction. A chest computed tomography scan demonstrated a mass-like lesion in the right middle lobe with mediastinal lymphadenopathy. She underwent an exploratory operation after 2 weeks of antibiotic treatment. The pathology revealed a right middle lobe bronchial carcinoid tumor and aspergillosis. Chest computed tomography scans have revealed no recurrence of the carcinoid or aspergillosis during the 5-year follow-up.

Keywords

Aspergillosis, bronchial neoplasms, carcinoid tumor, oncology, pathology, respiratory medicine

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Introduction

Aspergillus can cause various infectious diseases depending on the host's immunity and pulmonary status, particularly with respect to underlying lung diseases.[1,2] Pulmonary involvement of aspergillosis has been divided into pulmonary aspergilloma, chronic necrotizing pulmonary aspergillosis, allergic bronchopulmonary aspergillosis, and invasive aspergillosis.[2] Endobronchial aspergilloma is an uncommon manifestation of pulmonary aspergillosis, characterized by the growth of *Aspergillus sp.* into the bronchial lumen.[1,3] In patients with an underlying lung cavity, which can have many causes such as sarcoidosis, pulmonary tuberculosis, or pneumoconiosis, *Aspergillus* can colonize and develop in the cavity to form an endobronchial aspergilloma.[1,4,5] Aspergillosis is well known to occur in association with bronchial obstruction and immunocompromised conditions, such as lung transplantation.[6,7]

Pulmonary carcinoid tumors originate from lung neuroendocrine cells. Pulmonary neuroendocrine tumors can be divided into several tumor grades, namely, low-grade neuroendocrine tumors (typical carcinoids), intermediate-grade neuroendocrine tumors (atypical carcinoids), and high-grade neuroendocrine tumors (large cell neuroendocrine and small cell carcinomas).[8,9] Primary carcinoid tumors of the lung are rare, accounting for 0.5%–5% of all lung malignancies and 20%–30% of all carcinoid tumors.[10–12] Patients with pulmonary carcinoid tumors often present with non-specific pulmonary symptoms, such as cough, sputum, and hemoptysis. Carcinoid syndrome, including red flushing and diarrhea, is seen very occasionally.[6,11]

Recurrent pneumonia and bronchial obstruction suggest endobronchial stenosis, which can be associated with many diseases, including lung cancer and endobronchial tuberculosis, as well as a foreign body. Although chest computed tomography (CT) and bronchoscopy are helpful, bronchial obstruction can have an obscure origin. Some case reports have been published on endobronchial carcinoids, which can be confused with other lung diseases, such as tuberculosis and nontuberculous mycobacterial (NTM) infections.[6,13,14]

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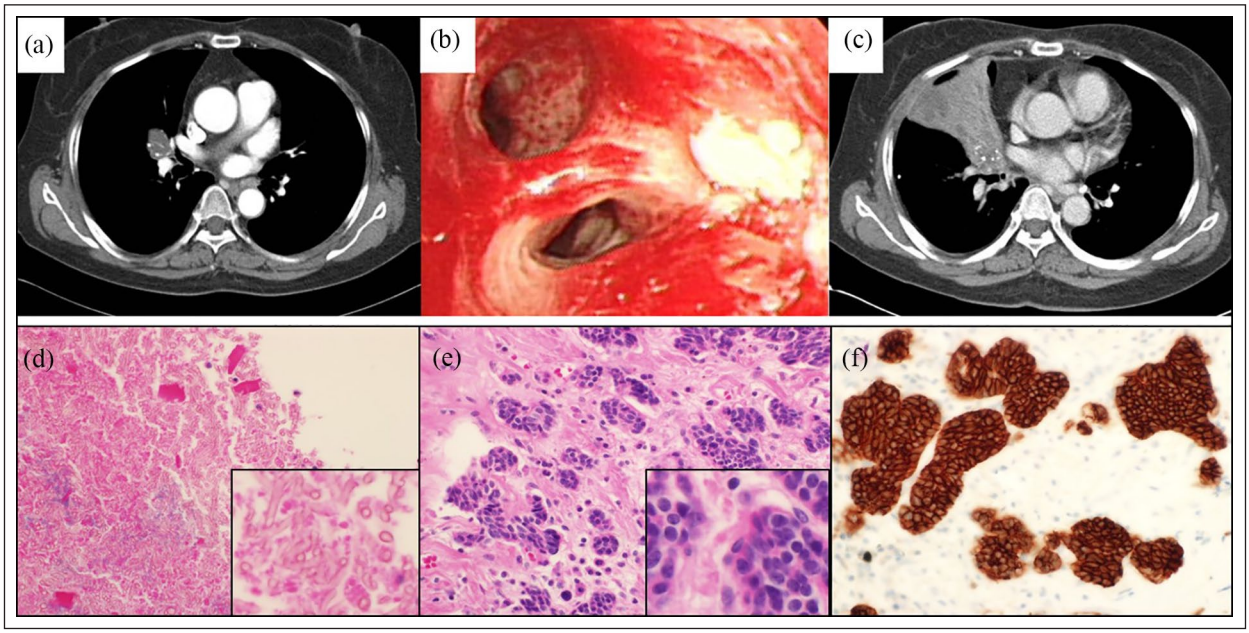

Figure 1. Right middle lobe (RML) obstruction associated with a synchronous endobronchial carcinoid and aspergillosis. (a) 27 × 21 mm lung mass was found in the RML on the initial chest computed tomography scan. (b) Initial bronchoscopy showed fibrosis and stenosis of the lateral segment of the RML with abundant mucoid material. (c) The mass had increased in size to 66 × 39 mm 3 months later, and caused obstructive pneumonia in the RML. (d) Acute-angle branching, septate, and narrow *Aspergillus* hyphae indicate aspergillosis. (e) Hematoxylin and eosin staining showing the carcinoid tumor. (f) CD56 immunostaining of the carcinoid tumor.

Here, we present a rare case of synchronous aspergillosis and a carcinoid, which caused bronchial obstruction.

Case

A 63-year-old woman with a history of rheumatoid arthritis for 15 years consulted our pulmonology department for evaluation of a lung mass. She was a non-smoker and presented with complaints of cough and sputum. The anti-inflammatory drug for rheumatoid arthritis included methotrexate, but based on complete blood count, she was not immunocompromised. Chest CT revealed a 27 × 21 mm lung mass on the right middle lobe (RML) (Figure 1(a)). No fungal organisms or acid-fast bacilli were found in sputum cultures. A bronchoscopic examination revealed fibrosis and stenosis of the lateral segment of the RML, with abundant mucoid material (Figure 1(b)). The bronchoscopic biopsy results demonstrated non-specific findings except inflammatory cells. An exploratory operation was recommended, but the patient refused further evaluation. Three months later, she presented to the emergency medicine department with fever, cough, and right pleuritic pain. A chest CT showed RML atelectasis and enlargement of the perivascular area lymph node, with a 66 × 39 mm mass-like lesion (Figure 1(c)). A double sleeve lobectomy was performed for obstructive pneumonia after 2 weeks of antibiotic treatment. Fortunately, most of the lesions that looked like mass on chest CT were chronic inflammation and atelectatic parenchyma, not actual mass. The pathological examination showed bronchial obstruction with distal bronchiectasis and peripheral parenchymal consolidation. Unexpectedly, a microscopic examination of the biopsy revealed an RML bronchial carcinoid tumor and aspergillosis (Figure 1(d) and (e)). The size of carcinoid tumor and aspergillosis was less than 10 mm in diameter. There was no invasion of bronchial wall and lymph nodes. The tumor cells stained positive for neuroendocrine markers, including CD56 (Figure 1(f)), synaptophysin, and chromogranin. Other tissue fragments were chronically inflamed. Chest CT revealed no recurrence of the carcinoid or aspergillosis during a 5-year follow-up.

Discussion

Bronchial carcinoids are unusual, slow growing, low-grade malignant tumors comprising 0.5%–5% of all primary lung cancers.[9,13,15] While typical carcinoid tumors are generally found in the central main bronchi, atypical carcinoid tumors occur in the lung periphery.[16] The relationship between bronchial carcinoid tumor and smoking was not confirmed and its exact cause is unknown.[17] The prognosis of a patient with a typical carcinoid is better than that of one with an atypical carcinoid.[16] Bronchial obstruction and pneumonia can occur due to bronchial carcinoids.[8] Unfortunately, the diagnosis may be delayed in those cases because the patients are treated with antibiotics for recurrent pneumonia.

Aspergillus contributes to a broad spectrum of pulmonary diseases, depending on the patient's immune status and underlying lung condition.[1] Bronchial obstruction due to

synchronous endobronchial aspergilloma and a carcinoid is very rare, but can cause severe complications.[6] A few cases of carcinoid tumors masked by aspergillosis have been reported.[6] Because these rare causes are sometimes difficult to diagnosis via routine examinations, such as chest CT and bronchoscopy, physicians should consider this rare condition and perform an exploratory operation. In this case, the diagnosis was delayed but the carcinoid tumor and aspergillosis were cured by surgery.

Conclusion

Bronchial carcinoids can occur in association with bronchial obstruction and pneumonia and can coexist with aspergillosis, tuberculosis, or an NTM infection. Although rare, the possibility of a bronchial carcinoid and aspergillosis should be considered if the patient has recurrent pneumonia or bronchial obstruction of uncertain cause.

Author contributions

C.C. contributed to conceptualization, supervision, and writing. Y.K. and D.P. contributed to reviewing and editing.

Declaration of conflicting interests

The author(s) declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

Ethical approval

Ethical approval to report this case was obtained from Clinical Research Ethics Committee of Chungnam National University Hospital. Institutional review board (IRB). IRB file number is 2015-07-001-002.

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Informed consent

Written informed consent was obtained from the patient for her anonymized information to be published in this article.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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