Case Report

Right middle lobe obstruction associated with synchronous endobronchial carcinoid and aspergillosis

Yoonjoo Kim1*, Dongil Park1* and Chaeuk Chung1,2

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

Date received: 19 October 2020; accepted: 15 March 2021

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

---

Yoonjoo Kim
1 Department of Internal Medicine, College of Medicine, Chungnam National University, Daejeon, Republic of Korea
2 Division of Pulmonology, Department of Internal Medicine, Chungnam National University, Daejeon, Republic of Korea

Dongil Park
1 Department of Internal Medicine, College of Medicine, Chungnam National University, Daejeon, Republic of Korea
2 Division of Pulmonology, Department of Internal Medicine, Chungnam National University, Daejeon, Republic of Korea

Chaeuk Chung
1 Department of Internal Medicine, College of Medicine, Chungnam National University, Daejeon, Republic of Korea
2 Division of Pulmonology, Department of Internal Medicine, Chungnam National University, Daejeon, Republic of Korea

*Corresponding Authors:
Chaeuk Chung, Division of Pulmonology, Department of Internal Medicine, Chungnam National University, 282, Munhwaro, Jung-gu, Daejeon 35015, Republic of Korea.
Email: universe7903@gmail.com

Creative Commons Non Commercial CC BY-NC: This article is distributed under the terms of the Creative Commons Attribution-NonCommercial 4.0 License (https://creativecommons.org/licenses/by-nc/4.0/) which permits non-commercial use, reproduction and distribution of the work without further permission provided the original work is attributed as specified on the SAGE and Open Access pages (https://us.sagepub.com/en-us/nam/open-access-at-sage).
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.9 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. A few cases of carcinoid tumors masked by aspergillosis have been reported. Because these rare causes are sometimes difficult to diagnose 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.

**Funding**

The author(s) disclosed receipt of the following financial support for the research, authorship, and/or publication of this article: This work was supported by the National Research Foundation of Korea (NRF) grant funded by the Korean Government (MSIT) (No. NRF 2017R1A5A2015385).

**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.

**ORCID iD**

Chaeuk Chung  
https://orcid.org/0000-0002-3978-0484

**References**

1. Kousha M, Tadi R and Soubani AO. Pulmonary aspergillosis: a clinical review. *Eur Respir Rev* 2011; 20(121): 156–174.
2. Kumar L, Singh M, Mitra SK, et al. Superadded aspergillosis on carcinoid bronchial adenoma leading to delayed diagnosis. *Postgrad Med J* 1990; 66(781): 938–939.
3. Dieudonné P. [Bronchial carcinoid tumor masked by aspergillosis]. *J Fr Med Chir Thorac* 1969; 23(2): 129–135.
4. Naylor CD, Shkrum MJ, Edmonds MW, et al. Pulmonary aspergillosis and endophthalmitis: complications of Cushing’s syndrome. *CMAJ* 1988; 138(8): 719–720.
5. Homasson JP, Hertzog P and Carnot F. [Vegetating aspergilloma on a carcinoid bronchial obstruction]. *Nouv Presse Med* 1982; 11(43): 3207.
6. Nilsson JR, Restrepo CS and Jagirdar J. Two cases of endobronchial carcinoid masked by superimposed aspergillosis: a review of the literature of primary lung cancers associated with Aspergillus. *Ann Diagn Pathol* 2013; 17(1): 131–136.
7. Krenke R and Grabcezk EM. Tracheobronchial manifestations of *Aspergillus* infections. *Sci World J* 2011; 11: 2310–2329.
8. Bora MK and Vithiavathi S. Primary bronchial carcinoid: a rare differential diagnosis of pulmonary koch in young adult patient. *Lung India* 2012; 29(1): 59–62.
9. Hendifar AE, Marchevsky AM and Tuli R. Neuroendocrine tumors of the lung: current challenges and advances in the diagnosis and management of well-differentiated disease. *J Thorac Oncol* 2017; 12(3): 425–436.
10. Herde RF, Kokeny KE, Reddy CB, et al. Primary pulmonary carcinoid tumor: a long-term single institution experience. *Am J Clin Oncol* 2018; 41(1): 24–29.
11. Gosain R, Mukherjee S, Yendamuri SS, et al. Management of typical and atypical pulmonary carcinoids based on different established guidelines. *Cancer* 2018; 10(12): 510.
12. Reuling E, Dickhoff C, Plaisier PW, et al. Endobronchial and surgical treatment of pulmonary carcinoid tumors: a systematic literature review. *Lung Cancer* 2019; 134: 85–95.
13. Hakami A, Zwartkruis E, Radonic T, et al. Atypical bronchial carcinoid with postobstructive mycobacterial infection: case report and review of literature. *BMC Pulm Med* 2019; 19(1): 41.
14. Nakamura Y, Okada Y, Endo C, et al. Endobronchial carcinoid tumor combined with pulmonary non-tuberculous mycobacterial infection: report of two cases. *Lung Cancer* 2003; 39(2): 227–229.
15. Pelosi G, Sonzogni A, Harari S, et al. Classification of pulmonary neuroendocrine tumors: new insights. *Transl Lung Cancer Res* 2017; 6(5): 513–529.
16. Reuling E, Dickhoff C, Plaisier PW, et al. Endobronchial treatment for bronchial carcinoid: patient selection and predictors of outcome. *Respiration* 2018; 95(4): 220–227.
17. Swarts DR, Ramaekers FC and Speel EJ. Molecular and cellular biology of neuroendocrine lung tumors: evidence for separate biological entities. *Biochim Biophys Acta* 2012; 1826(2): 255–271.