Conjunction of a Fungus Ball and a Pulmonary Tumourlet in a Bronchiectatic Cavity

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Herein, we describe the case of a 67-year-old female patient who presented with cough and haemoptysis. Chest computed tomography revealed destruction of the left lower lobe and multiple fungus balls in a bronchiectatic cavity. A left lower lobectomy was performed via thoracotomy. Histopathological examination of the lung showed a concomitant aspergilloma and multiple tumourlets in the large bronchiectatic cavity. Pulmonary intracavitary aspergilloma and concomitant tumourlets are quite rare. Our report presents this interesting case that manifested with haemoptysis.

Key words: 1. Pulmonary aspergilloses 2. Bronchiectasis 3. Fungus 4. Hemoptysis 5. Mycetoma

Case report

A 67-year-old woman with a medical history of tuberculosis was referred to our department for recurrent pulmonary infection and haemoptysis. The patient was a non-smoker. She suffered from hypertension and was taking nebivolol and olmesartan. Auscultation revealed rales in the left lower zone. The results of spirometry and laboratory tests were normal. Computed tomography (CT) demonstrated multiple fungus balls in a large cavity located in the left lower lobe; however, pathologic enlargement of the lymph nodes was not seen in CT images (Fig. 1).

Left lower lobectomy and mediastinal lymph node sampling were performed via thoracotomy. In the pathological examination of the specimen, a cavity measuring 4.5 cm in diameter was found in the superior segment. The material inside the cavity was brown, necrotic, and compatible with aspergilloma. In the samples taken from the cavitated bronchial wall, groups of uniform cells and others scattered throughout a microscopic focus of less than 0.5 cm in diameter were detected. Immunohistochemical staining for CD56 and thyroid transcription factor-1 was positive, synaptophysin and P63 were negative, and the Ki67 level was 2% in these cells (Fig. 2). There was neither atypia nor mitosis. There was no metastasis in the lymph nodes that were examined pathologically. The lesion contained the focal area of a tumourlet and areas of diffuse multiple endocrine neoplasia. The final histopathological diagnosis was aspergilloma, multiple tumourlets, and diffuse idiopathic pul-

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Surgical Treatment of a Pulmonary Tumourlet

Fig. 1. The fungus balls in the large cavity and areas of destruction in the left lower lobe.

Discussion

Also known as intracavitary mycetoma or a fungus ball, aspergilloma is the result of a saprophytic infection of a cavitary diseased lung [1]. The most common symptom is haemoptysis [1]. After a fungus ball has formed in a cavity, antifungal treatment is inefficient because antifungal drugs cannot penetrate through the non-perfused wall of the cavity. Hence, a monary neuroendocrine cell hyperplasia (DIPNECH) in a bronchiectatic lung cavity. No radiologic abnormalities were found on a follow-up CT exam, and 7 months after the operation, the patient remained alive without any notable medical problems.

Fig. 2. (A) Fungus balls, neuroendocrine tumour cell groups, and bronchial wall in the cavitated large bronchial lumen (H&E, ×40). (B) Using immunohistochemistry, Ki67 positivity was found in the neuroendocrine cells (Dako, ×100). (C) Using immunohistochemistry, synaptophysin positivity was found in the neuroendocrine cells (Dako, ×60). (D) Tumourlet (H&E, ×60).
cure can be achieved only by surgical treatment. Additionally, pulmonary tumourlets are a rare pathology, defined as nodular proliferations of neuroendocrine cells less than 0.5 cm in diameter [2]. They are usually detected incidentally in areas of lung destruction that are removed surgically or in cases of bronchiectasis. The coexistence of a fungus ball and a pulmonary neuroendocrine tumourlet has rarely been observed.

DIPNECH, pulmonary tumourlets, and typical carcinoid tumours are distinct subsets of neuroendocrine tumours that have several characteristics in common [3]. Tumourlets are nodular proliferations of neuroendocrine cells (Kulchitsky) that are normally present in the airways. For unknown reasons, these cells sometimes become hyperplastic in certain lung diseases. Tumourlets are usually nodular proliferations smaller than 5 mm in diameter and are often multiple. Larger tumours are called carcinoids [2-5]. They are asymptomatic and common incidental findings in scarred lungs, but they may also be found in normal lungs [4]. They also lack mitotic activity; necrosis and cellular atypia are not observed. In recent studies, DIPNECH and tumourlets have been regarded as preneoplastic lesions that rarely convert to carcinoid tumours [5,6]. Lymph node metastases have been noted in several case reports [7]. Nodal metastasis generally occurs in the hilar or peribronchial lymph nodes. Tumourlets are generally encountered in patients between 60 and 70 years of age, and are more common in female patients than in male patients (male to female ratio, 1: >4) [2]. Tumourlets should be included in the differential diagnosis when a patient complains of dyspnoea and small nodules are identified on a chest CT scan; however, positron emission tomography/CT is not the best method to diagnose this entity [2]. There are no well-established guidelines for the treatment and management of DIPNECH. Currently, there is no known method to prevent DIPNECH, but various steps can be taken to help decrease the risk of its occurrence, such as complete smoking cessation, physical activity, avoiding exposure to certain materials and chemicals (including asbestos, arsenic, chromium, nickel, and tar), and limiting alcohol consumption. Several medications have been studied as potential treatments for DIPNECH, such as octreotide, 18F-DOPA amino acid analogues, inhaled cortico-steroids, and short-acting beta agonists [2,6].

Some previous case reports have reported pulmonary tumourlets associated with diffuse bronchiectasis and intralobar sequestration, in pulmonary sequestration with bronchiectasis after breast cancer, or associated with marginal zone B-cell lymphoma [8]. Pulmonary tumourlets often occur in patients with chronic lung damage, such as pulmonary fibrosis, chronic or granulomatous inflammation, bronchiectasis, or giant cell pneumonia [3]. However, the association of aspergilloma with tumourlets or DIPNECH in a bronchiectatic cavity has not yet been reported. In this report, we present a case where both a tumourlet and aspergilloma were detected in the left lower lobe of a previous tuberculosis patient with a bronchiectatic cavity in a destroyed lobe. We think that there may be a pathological relationship between aspergilloma and DIPNECH. Although the patient did not receive any postoperative medical treatment, recurrence or metastasis has not occurred during the follow-up period.

There is also no consensus regarding the timing or type of surgery needed for aspergilloma. Surgical resection is risky and difficult due to the presence of an underlying chronic disease and the negative effects of infection on the lung. However, the coexistence of a large bronchiectatic cavity, aspergilloma, and tumourlet in our case suggests that such lung diseases also provide a basis for premalignant lesions, as well as for complications such as haemoptysis and recurrent infections.

In conclusion, surgical treatment can therefore prevent both the recurrence of symptoms and the development of premalignant lesions.

Conflict of interest

No potential conflict of interest relevant to this article was reported.

References

1. Jheon SH, Lee JT, Kim KT. Surgical treatment of pulmonary aspergillosis. Korean J Thorac Cardiovasc Surg 1989;22:170-5.
2. Kallianos A, Velentza L, Zarogoulidis P, et al. Progressive dyspnea due to pulmonary carcinoid tumourlets. Respir Med Case Rep 2017;21:84-5.
3. He P, Gu X, Wu Q, Lin Y, Gu Y, He J. Pulmonary carcinoid tumorlet without underlying lung disease: analysis of its relationship to fibrosis. J Thorac Dis 2012;4:655-8.

4. Churg A, Warnock ML. Pulmonary tumorlet: a form of peripheral carcinoid. Cancer 1976;37:1469-77.

5. Hendifar AE, Marchevsky AM, Tuli R. Neuroendocrine tumors of the lung: current challenges and advances in the diagnosis and management of well-differentiated disease. J Thorac Oncol 2017;12:425-36.

6. Wirtschafter E, Walts AE, Liu ST, Marchevsky AM. Diffuse idiopathic pulmonary neuroendocrine cell hyperplasia of the lung (DIPNECH): current best evidence. Lung 2015;193:659-67.

7. D’Agati VD, Perzin KH. Carcinoid tumorlets of the lung with metastasis to a peribronchial lymph node: report of a case and review of the literature. Cancer 1985;55:2472-6.

8. Dewan M, Malatani TS, Osinowo O, al-Nour M, Zahrani ME. Carcinoid tumorlets associated with diffuse bronchiectasis and intralobar sequestration. J R Soc Promot Health 2000;120:192-5.