Case report

A case of allergic bronchopulmonary mycosis due to *Schizophyllum commune* with elevated serum carcinoembryonic antigen levels

Mika Yamaguchi*a, Kazushi Yamairi*a, *, Hiroko Fujii*a, Yuzo Miki*a, Takashi Mamoto*a, Kyoko Yagyu*a, Makoto Niki*b, Hiroshi Kakeya*c, Seiichi Shoji*a

*a Department of Respiratory Medicine, Osaka City General Hospital, Osaka, Japan
*b Department of Infection Control and Prevention, Osaka City University Hospital, Osaka, Japan
*c Department of Infection Control Science, Osaka City University Graduate School of Medicine, Osaka, Japan

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**ABSTRACT**

A 78-year-old Japanese woman without any history of asthma or smoking presented prolonged cough. Laboratory data showed elevated serum CEA levels and a chest CT revealed a mass with abnormal uptake in the left lower lobe. One month later, the mass spontaneously regressed, and CEA levels improved. However, the symptoms progressed during the observation period without treatment. Chest radiograph findings revealed collapse of the right middle lobe, and *Schizo phyllum commune* was isolated from the mucous plugs; the patient was diagnosed with allergic bronchopulmonary mycosis (ABPM). Herein, we report the first case of ABPM caused by *S. commune* with elevated CEA levels and mimicking lung cancer.

1. Introduction

Allergic bronchopulmonary mycosis (ABPM) is an immunological disorder caused by a hyperimmune response to endobronchial growth of certain fungi and occurs most frequently in atopic patients with asthma. ABPM due to *Aspergillus* species (allergic bronchopulmonary aspergillosis, ABPA) was first reported in 1952 by Hinson et al. [1], and several clinical diagnostic criteria for ABPA were established. In 1994, Kamei reported the first case of ABPM caused by *Schizophyllum commune* infection [2]. Serum carcinoembryonic antigen (CEA) levels are elevated in some patients with ABPA, and these elevated serum CEA levels decrease as consolidation decreases after treatment [3]. However, to the best of our knowledge, no case of non-Aspergillus ABPM caused by *S. commune* has been reported. Herein, we report the first case of ABPM due to *S. commune* with elevated serum CEA levels and mimicking lung cancer.

2. Case report

A 78-year-old Japanese woman without any history of asthma or smoking presented to another hospital with a prolonged cough in December 2020. Antibiotics and combined inhaled therapy of a long-acting beta-agonist and inhaled corticosteroids did not improve her symptoms; therefore, detailed examinations were performed. Laboratory data revealed an elevated serum CEA of 15.6 ng/ml and cytokeratin-19 fragment (CYFRA 21–1) of 3.6 ng/ml. Chest computed tomography (CT) revealed a mass in the left lower lobe. Fluorine-18-fluorodeoxyglucose (18F-FDG) positron emission tomography (PET)-CT showed fluorodeoxyglucose accumulation in the lungs.
mass (SUVmax 7.6). Bronchoscopy was performed; however, no malignant findings were observed. One month later, the mass spontaneously regressed and CEA levels improved. She was referred to our hospital in June 2021. Although we suspected that the patient had ABPM, the diagnostic criteria were not met. Her serum immunoglobulin E (IgE) levels and eosinophil counts were normal, and specific IgE for Aspergillus species, antibodies, and antigens against Aspergillus species were negative; therefore, we conducted follow-up observations.

The symptoms gradually progressed during the observation period without treatment. She was hospitalized and underwent repeat examinations in September 2021. Serum IgE, carcinoembryonic antigen, and eosinophil counts were elevated. The chest radiograph revealed high attenuation mucous impaction and collapse of the right middle lobe (Fig. 1). Bronchoscopy revealed mucus plugs in the truncus intermedius. Pathologically, infiltration of eosinophils and Charcot-Leyden crystals was observed by hematoxylin and eosin staining (Fig. 2A). Fungal mycelia were detected by Grocott staining (Fig. 2B). Bronchial aspirates and bronchial washing fluid yielded colonies of S. commune. Consequently, 10 months after the symptom appeared, she was diagnosed with ABPM caused by S. commune, and it has not relapsed for 5 months since corticosteroid administration was started. Serum CEA levels decreased after the treatment started.

3. Discussion

Herein, we describe a case of ABPM caused by S. commune that mimics lung cancer with elevated serum CEA levels. Previous studies have shown that elevated serum CEA levels in patients with ABPA are associated with lung consolidation, mucus plugs, and localized inflammation of the lungs. Serum CEA levels were relatively elevated in patients with ABPA, and elevated serum CEA levels can normalize after treatment. Increased CEA levels in patients with ABPA might be positively correlated with eosinophil levels, and eosinophils can serve as CEA-secreting cells in patients with ABPA [4]. No cases of non-Aspergillus ABPM have been reported; however, non-Aspergillus ABPM might have a mechanism similar to ABPA.

Presentations of ABPA as large pulmonary masses have been reported in some studies [5]. FDG-PET has been used to differentiate between benign and malignant lesions. However, numerous reports have demonstrated increased FDG uptake at the site of infection and inflammation [6]. Eosinophilic inflammation caused by ABPM may increase FDG uptake. In this case, it was difficult to differentiate from lung cancer based on CEA levels and FDG accumulation.

Rosenberg et al. [7] proposed the first diagnostic criteria for ABPA in 1977. Subsequently, Greenberger and Patterson [8] added A. fumigatus-specific IgE and IgG to these criteria. The International Society for Human and Animal Mycology (ISHAM) [9] proposed new diagnostic criteria in 2013, which defined asthma or cystic fibrosis as predisposing conditions and included two obligatory criteria: (1) immediate cutaneous hypersensitivity to Aspergillus antigen or elevated IgE levels against A. fumigatus and (2) elevated total IgE levels. The diagnosis of non-Aspergillus ABPM is more challenging, as traditional criteria are specific to ABPA caused by A. fumigatus, and there are no diagnostic criteria available for ABPM. The Japan ABPM Research Program, supported by the Japan Medical Research and Development Organization, developed new 10-component diagnostic criteria for ABPA/ABPM in patients who do not have cystic fibrosis. The new criteria showed better sensitivity than the previous criteria to diagnose pathological and physician diagnosed ABPA/ABPM, with reasonable specificity [10]. However, her ABPM had not met the criteria for 10 months and was initially suspected of lung cancer.

4. Conclusions

Serum CEA levels can be relatively elevated in patients with ABPM. Our case highlights the importance of considering ABPM in patients who may not initially meet the diagnostic criteria and mimic lung cancer. ABPM can be characterized by exacerbations and remission; therefore, close follow-up is needed.

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Declaration of interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to
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