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CASE REPORT

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Allergic bronchopulmonary aspergillosis manifested secondary to bacterial pleural empyema

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Abstract

A 54-year-old woman with no history of lung disease including bronchial asthma developed left bacterial pleural empyema due to the perforation of a lung abscess in the left lower lobe. Chest tube drainage and antibiotics improved the pleural empyema. Two months following discharge from the hospital, she developed a cough and left chest pain. Chest computed tomography revealed high-attenuation mucus plugs, atelectasis in the left lower lobe, and an increased peripheral blood eosinophil count. Bronchoscopy revealed a mucoid impaction in B8 of the left lower lobe, confirming the presence of *Aspergillus fumigatus*. A diagnosis of allergic bronchopulmonary aspergillosis was made, and treatment with oral prednisolone was initiated, resulting in the resolution of the mucus plugs and improvement of atelectasis.

KEYWORDS
allergic bronchopulmonary aspergillosis, bacterial pleural empyema, high-attenuation mucus plugs

INTRODUCTION

Allergic bronchopulmonary aspergillosis (ABPA) is a chronic inflammatory airway disease that develops through allergic reactions induced by the ubiquitous fungus *Aspergillus fumigatus* in the airways. A characteristic feature of ABPA is the formation of mucus plugs rich in eosinophils within the bronchi, which are associated with fixed airflow obstruction and usually bilateral bronchiectasis. While usually complicating the course of patients with bronchial asthma and cystic fibrosis, ABPA can occasionally occur in other lung disorders, such as pulmonary tuberculosis.[1] We experienced an unusual case of unilateral high-attenuation mucus plugs and atelectasis following same side bacterial pleural empyema, which led to the diagnosis of ABPA.

CASE REPORT

A 53-year-old woman with a history of hypertension, alcoholic liver disease, and paranasal sinusitis, but no history of lung disease, including bronchial asthma, was urgently admitted with cough, fever, chest pain, and hemoptysis on October 20XX-1. Laboratory test results revealed a white blood cell count of 14,330/μL (neutrophils 74.4%, eosinophils 4.0%), C-reactive protein of 29.44 mg/dL. Chest computed tomography (CT) revealed a mass in the left lower lobe, raising suspicion of a lung abscess (Figure 1A). Although treatment with meropenem 3 g per day was initiated, her dyspnea suddenly worsened 12 h following admission, and she required oxygen therapy at 10 L/min via a reservoir mask. A repeat CT scan revealed left pleural empyema due to perforation of the lung abscess (Figure 1B). Chest tube drainage was initiated, and a reddish-brown turbid pleural fluid was observed. Laboratory test results of pleural fluid showed a cell count of 38,219/μL, a predominant neutrophilic response (91.8%), and decreased glucose levels (4 mg/dL), consistent with pleural empyema. Although bacterial cultures of the pleural fluid did not detect the causative organism, the patient was treated with chest tube drainage for 12 days and antibiotics (meropenem 3 g/day for 12 days, followed by tazobactam/piperacillin 13.5 g/day for 4 days), which improved her symptoms, C-reactive protein levels, and chest CT scan findings (Figure 1C). The patient did not present with symptoms of asthma during hospitalization and was discharged on 23 Nov 20XX-1. Two months following discharge, she developed a cough and left chest pain. She visited a hospital

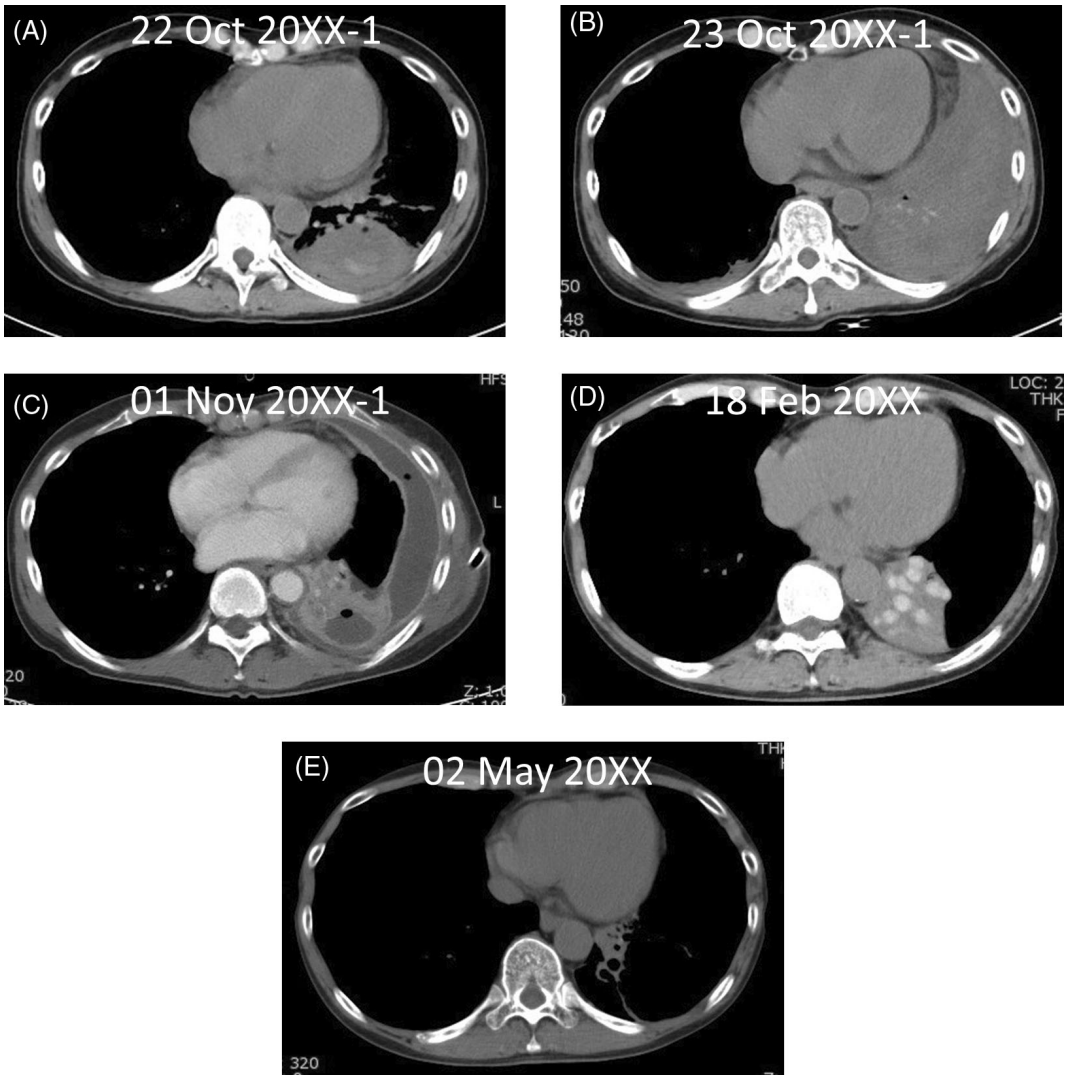

FIGURE 1 Changes in findings of chest computed tomography scan. (A) Suspected left lung abscess on initial admission. (B) Suspected left pleural empyema due to perforation of lung abscess 12 h after initial admission. (C) Improvement in left pleural empyema by treatment with chest tube drainage and antibiotics. (D) High-attenuation mucus plugs and atelectasis in the left lower lobe. (E) Disappearance of mucus plugs and the improvement in atelectasis after oral prednisolone treatment.

because of increasing chest pain in February 20XX, and a chest CT scan showed high-attenuation mucus plugs and atelectasis in the left lower lobe (Figure 1D). Blood tests revealed elevated peripheral eosinophil counts (2535 /μL) and Aspergillus-specific IgG antibodies (>80 AU/mL), but not serum total IgE (53 IU/mL) and Aspergillus-specific IgE antibodies levels. Asp f 1-specific IgE antibody measurements and skin test reactivity to *A. fumigatus* were not performed. Pulmonary function tests revealed reversible airway obstruction, with forced expiratory volume in 1 s (FEV_1)/forced vital capacity 1.92/3.27 L, FEV_1% predicted 89.7%, FEV_1 reversibility after inhalation of albuterol +0.32 L and +16.7%. The levels of fractional exhaled nitric oxide were 6 ppb. Bronchoscopy revealed a mucoid impaction in B8 of the left lower lobe (Figure 2A). A lung biopsy of the left lower lobe revealed mucus and eosinophilic infiltration. Samples from the bronchial lavage and sputum were positive for Grocott staining, and cultures of mucus plugs confirmed the presence of *A. fumigatus* (Figure 2B). Based on these findings, she was diagnosed with ABPA and treated with oral prednisolone at 20 mg/day, in addition to inhalation therapy with fluticasone furoate 100 μg/vilanterol 25 μg. These treatments resulted in the rapid disappearance of mucus plugs and an improvement in atelectasis in the left lower lobe (Figure 1E).

DISCUSSION

Here, we present a case of unilateral high-attenuation mucus plugs and atelectasis that developed after bacterial pleural empyema on the same side, leading to a diagnosis of ABPA.

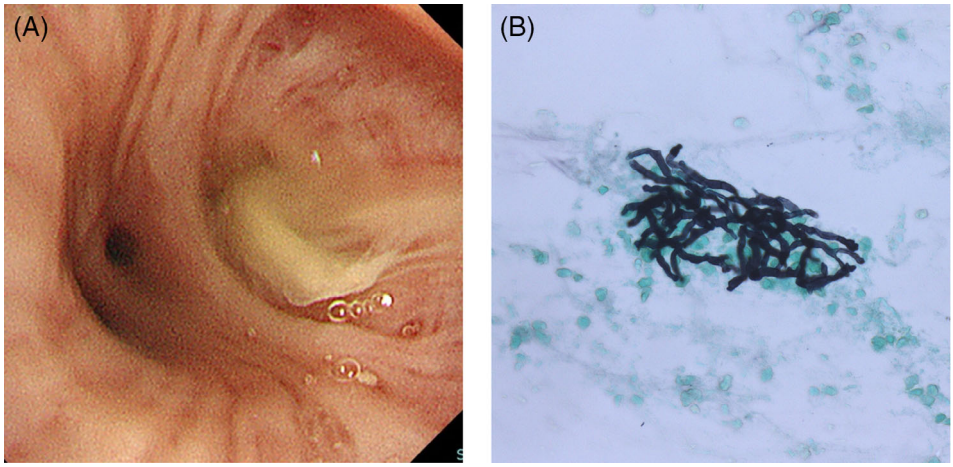

FIGURE 2 (A) Mucoid impaction in B8 of the left lower lobe. (B) Hyphae of fungi found in sputum (Grocott staining, ×400).

Rosenberg's diagnostic criteria are non-specific to ABPA and include the presence of bronchial asthma, peripheral blood eosinophilia, immediate skin test reactivity to *A. fumigatus*, presence of serum precipitins against *A. fumigatus* antigen, elevated serum IgE levels, transient or fixed pulmonary infiltrates, and central bronchiectasis.

This case satisfied three diagnostic criteria of six: the presence of bronchial asthma, peripheral blood eosinophilia, and central bronchiectasis. No atopic predisposition or fungal-related environmental problems were found in the medical interview. Although mucus plugs in ABPA are generally hypodense, high-attenuation mucus is a characteristic radiologic finding in 18.7%–41% of ABPA.[2–4] In this case, despite poor asthma symptoms, ABPA was strongly suspected due to the presence of high-attenuation mucus plugs secondary to bacterial pleural empyema.

The Japan Allergic Bronchopulmonary Mycosis (ABPM) Research Program developed new 10-component diagnostic criteria proposed by Asano et al., which showed better sensitivity and specificity for diagnosing ABPA/ABPM compared with existing criteria.[5] This case satisfied 7 items of 10 and was diagnosed with ABPA: current history of asthma, peripheral blood eosinophilia, presence of specific IgG for *A. fumigatus*, positive culture of *A. fumigatus*, central bronchiectasis on CT, presence of mucus plugs in central bronchi, HAM in the bronchi on CT. Okada et al. recently identified three components (allergic, eosinophilic, and fungal) by factor analysis that determined the clinical characteristics of ABPA.[6] This case was classified with ABPA sans asthma (no preceding asthma) dominant in eosinophilic component. Okada et al. also reported that 24% of the cases were ABPA sans asthma, and the eosinophilic component of ABPA is considered as the cardinal feature of ABPA regardless of the presence of preceding asthma or atopic predisposition. After the ABPA diagnosis, the patient's sinusitis, which had affected the patient for many years, was diagnosed as fungal sinusitis.

This case suggested that pleural empyema may have contributed to eosinophilic mucus plug formation, possibly due to airflow restriction and mucus retention in the bronchi, allowing inhaled fungi to settle and multiply in the lower respiratory tract. As there have been reports of bacterial lung abscesses secondary to ABPA, the possibility that this patient had ABPA prior to the onset of the lung abscess cannot be denied.[7] However, this case had no respiratory symptoms prior to the lung abscess, and no abnormalities had been noted on chest x-ray during a previous physical examination. Previous pathological findings suggested that the primary lesions of ABPA are eosinophilic mucus plugs, but not central bronchiectasis or pulmonary infiltration.[4,8] The expansion of eosinophilic mucus plugs and the transluminal spreading of fungal components from the mucus plugs causes central bronchiectasis and peripheral lung lesions. As early diagnosis and therapeutic intervention of ABPA are important to prevent disease progression, this case informs us about the possible development of mucus plugs in ABPA following conditions such as pleural empyema, which can interfere with airway clearance.

AUTHOR CONTRIBUTIONS

Hiromu Kawano and Ayaka Egashira contributed to the literature review and the preparation of the initial manuscript. Keiko Kan-o and Isamu Okamoto contributed to the conceptualization and revision of the final manuscript.

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CONFLICT OF INTEREST STATEMENT

Keiko Kan-o is the Editor in chief of Respirology Case Reports, she was excluded from all editorial decision-making related to the acceptance of this article for publication. The acting Editor in chief for this publication was Deputy Editor Philip Bardin.

DATA AVAILABILITY STATEMENT

Data available on request from the authors.

ETHICS STATEMENT

The authors declare that appropriate written informed consent was obtained for the publication of this manuscript and accompanying images.

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