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Letter to the Editor

Effectiveness of mucus plug removal by bronchoscopy for typical high-attenuation mucus with allergic bronchopulmonary mycosis

Dear Editor,

Allergic bronchopulmonary mycosis (ABPM) is caused by a hypersensitive reaction to mycosis and leads to worsened pulmonary function with developing airway dysfunction.¹ Systemic corticosteroids (SCSs) are the mainstay of ABPM treatment, but long-term SCS usage causes a number of adverse events.² The use of antifungal drugs in addition to SCSs was reported to aid the treatment of allergic bronchopulmonary aspergillosis (ABPA), but their synergistic use with SCSs in the treatment of ABPM was unclear.³ Recently, biologics originally used for severe asthma were reportedly effective for ABPM,⁴ but the treatment duration and long-term safety are yet to be determined.

To date, it is difficult to prevent the reoccurrence of ABPM. Herein, we report three cases that showed the beneficial effect of removing almost all of the mucus plugs by flexible bronchoscopy during diagnosis, thereby improving symptoms and eliminating the adverse effects of the mucus plug itself.

Flexible bronchoscopy with transbronchial lung biopsy under moderate sedation was planned for each of the three cases. Informed consent was obtained from all patients after explaining the risks associated with the procedure. All three cases met the new 10-component diagnostic criteria for ABPA/ABPM of the Japan ABPM Research Program⁵ and showed typical high-attenuation mucus (HAM) on chest computed tomography (CT). After removing as much of the mucus plugs as possible by flexible bronchoscopy under moderate sedation, which took over 1 h, all three cases showed immediate improvement in HAM and symptoms. Histopathologically, all mucus samples showed allergic mucin with eosinophil infiltration and Charcot–Leyden crystals. No recurrence in the lobe where the mucus plugs were removed by bronchoscopy was observed in any of the cases 2 years after the procedure. Table 1 presents the characteristics of the three cases.

The first case (Case 1) was a 76-year-old with asthma who was diagnosed 3 years previously. Chest CT and bronchoscopy were performed (Fig. 1, Case 1A and 1B, respectively). After removing the mucus plugs by bronchoscopy (Fig. 1, Case 1C), there was an immediate improvement in the symptoms and hypoxia as well as HAM on chest CT and an improved asthma control score (ACT) from 12 to 24 after 4 weeks. Prednisolone treatment was administered for 2 weeks and then changed to inhaled corticosteroids (ICSs). After 1 year without any problems, CT revealed the reoccurrence of HAM in the opposite left lingular lobe on chest CT (Fig. 1, Case 1D).

Case 2 was that of a 56-year-old woman with suspected ABPA who took SCS at another institution. She had no history of other diseases except asthma. Following chest CT (Fig. 1, Case 2A), bronchoscopy was performed to remove the mucus plugs (Fig. 1, Case 2B, 2C), and she was discharged immediately thereafter. She discontinued SCS treatment and was given an ICS only. Her ACT improved from 15 to 22 after 4 weeks, and chest CT revealed the improvement of HAM. The patient has not experienced ABPA reoccurrence for more than 2 years (Fig. 1, Case 2D).

Case 3 was that of a 66-year-old woman with occasional asthma attacks who was treated with SCS. Following chest CT (Fig. 1, Case 3A), bronchoscopy was performed to remove the mucus plugs (Fig. 1, Case 3B, 3C). Thereafter, she was treated with prednisolone for 6 weeks, after which, the treatment was changed to ICS. Her ACT improved from 13 to 23 after 4 weeks, and chest CT revealed the improvement of HAM. HAM reoccurrence was observed on chest CT in the opposite left lingular lobe after 1 year (Fig. 1, Case 3D).

ABPM tends to reoccur in patients where HAM is observed on radiological findings,⁶ and ICS treatment is not considered beneficial for ABPM.⁷ Our cases suggested that removing the mucus plugs by bronchoscopy could be effective because the reoccurrence of HAM observed after 1 year in Cases 1 and 3 was in the lobe on the opposite side to that where the mucus plugs had been previously removed. Additionally, there was no reoccurrence of HAM in the lobe where the mucus plug was removed by bronchoscopy after more than 2 years in all three cases, and none of these patients received treatment with SCSs or antifungal drugs. The primary ABPM lesion was an eosinophilic mucus plug.⁸ Central bronchiectasis occurs when an airway wall that is weakened by eosinophilic inflammation during the growth of the eosinophilic mucus plug is stretched. Thus, mechanically removing the eosinophilic mucus plug potentially prevents bronchiectasis progression. Because the mucus plug of ABPM has a thick consistency and is not easily removed by direct bronchoscopic suction, it takes a long time to perform. At our institution, patients are intubated under moderate sedation with midazolam and inhaled lidocaine. We use flexible bronchoscopes (BF-P290 and BF-1TQ290; Olympus Corporation, Tokyo, Japan) with biopsy forceps (FB233D or FB231; Olympus Corporation) or a curette (CC-4CR-1 or CC-5CR-1; Olympus Corporation) and injected saline to crush and soften the plugs. For those with asthma, 80–125 mg methylprednisolone was injected via a drip before and during the procedure to prevent an acute asthma attack.

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Table 1
Characteristics of our three cases of allergic bronchopulmonary mycosis.

Characteristic	Case 1	Case 2	Case 3
Age, years	76	56	66
Sex	Female	Female	Female
SpO ₂ (room air)	91%	97%	98%
Symptoms	Cough, hypoxia, chest pain	Cough	Cough, fever
Eosinophils (/μL)	2206	1418	1548
Serum IgE (IU/ml)	890	2216	2629
Comorbidity	BA, MAC-PD, RA	BA	BA, RA
Fungal species	<i>Aspergillus terreus</i>	<i>Aspergillus fumigatus</i>	<i>Schizophyllum commune</i>
Wheezing	+	+	+
<i>Aspergillus</i> -specific IgE	+	+	+
Precipitating antibodies for <i>Aspergillus</i>	–	+	–
ACT	12	15	13

ACT, asthma control test; BA, bronchial asthma; IgE, immunoglobulin E; MAC-PD, *Mycobacterium avium* complex pulmonary disease; RA, rheumatoid arthritis; SpO₂, peripheral capillary oxygen saturation.

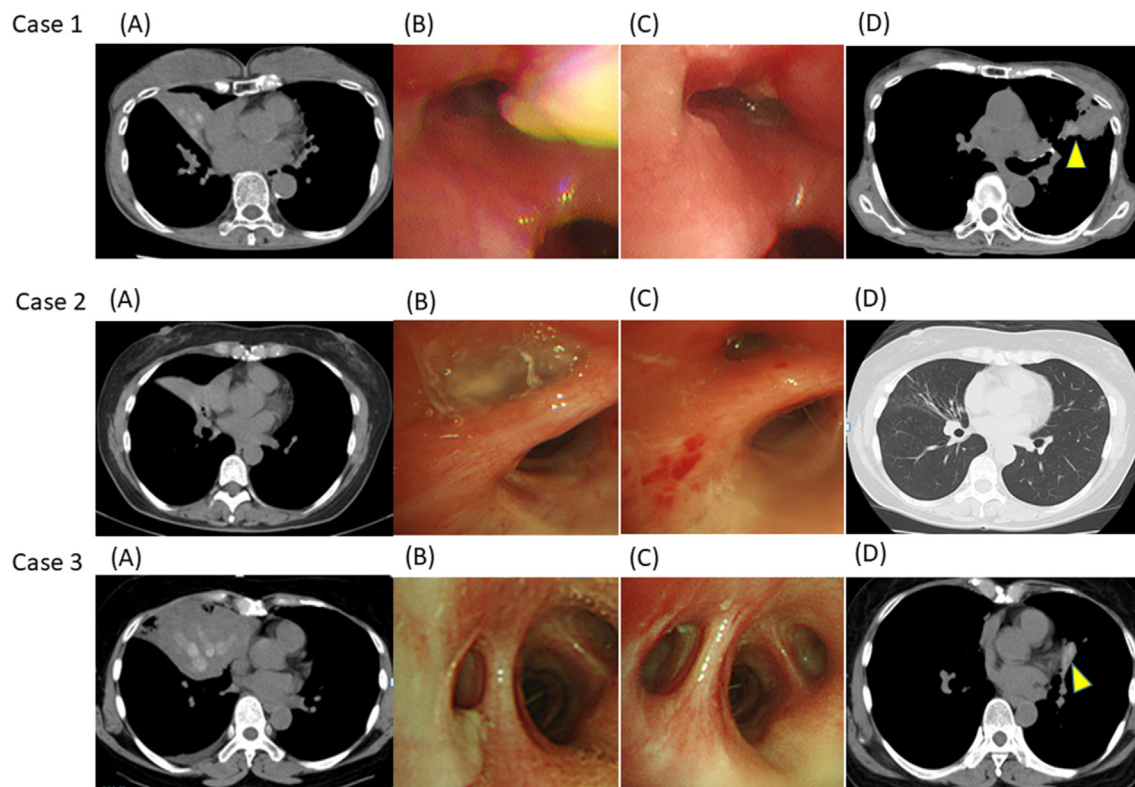


Fig. 1. Chest computed tomography (CT) and bronchoscopy findings in three cases of high-attenuation mucus (HAM) treated using bronchoscopy to remove mucus plugs. (A) CT of allergic bronchopulmonary mycosis (ABPM) showed HAM of the right middle lobe at diagnosis (Case 1–3). (B) Bronchoscopy showed mucous plugs embolized in the right middle lobe bronchus (Case 1–3). (C) After removing the mucus plugs, swelling and reddening of the bronchi were observed. (D) Chest CT upon reoccurrence of ABPM showed HAM of the left lingular lobe 1 year after removing mucus plugs by bronchoscopy (Case 1 and 3, yellow arrows). Chest CT 2 years after removing the mucus plugs by bronchoscopy (Case 2).

Because this report includes only three cases, all with short observation periods, the recurrence of HAM in the opposite side could be part of the natural course of the disease. Future studies with a larger sample size and longer observation periods are essential. Nevertheless, mucous plugs are successfully removed by bronchoscopy at our institution in approximately half of the patients with ABPM who undergo bronchoscopy. No asthma attacks occurred during bronchoscopy in any of the three cases presented here.

Bronchoscopy is not always necessary for ABPM diagnosis, and a careless examination may increase the risk of an asthma attack

during the procedure. Considering the risk of bronchoscopy, it appears to be a good accommodation for patients in whom the mucus plugs are unresponsive to steroid treatment.

The use of bronchoscopy for both diagnosis and concurrent removal of mucus plugs in ABPM would be not only cost-effective but also potentially prevent local inflammation at the early stage of ABPM and irreversible bronchodilation and bronchoalveolar parenchyma damage.

Conflict of interest

The authors have no conflict of interest to declare.

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