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【 CASE REPORT 】

Successful Long-term Management of Two Cases of Moderate Hemoptysis Due to Chronic Cavitary Pulmonary Aspergillosis with Bronchial Occlusion Using Silicone Spigots

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Abstract:
Chronic pulmonary aspergillosis is a major cause of life-threatening hemoptysis. In symptomatic patients with simple aspergillomas, surgery is the main therapeutic method for preventing or treating life-threatening hemoptysis. However, the risks of both death and complications are higher in chronic cavitary pulmonary aspergillosis than in simple aspergilloma. We herein report two patients with persistent moderate hemoptysis due to chronic cavitary pulmonary aspergillosis who were not indicated for surgery, but were able to undergo successful long-term management with bronchial occlusion using silicone spigots. In diseases with a high recurrence rate of hemoptysis, the continuous placement of silicone spigots might therefore be effective to prevent rebleeding.

Key words: bronchial occlusion, silicone spigot, endobronchial Watanabe spigot, aspergillosis, hemoptysis

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Introduction

Chronic pulmonary aspergillosis (CPA) is a major cause of hemoptysis, which can be life-threatening. Moreover, from 43-55% of CPA patients suffer from hemoptysis (1, 2). In symptomatic patients with simple aspergillomas, surgery is the main therapeutic method adopted for the prevention and treatment of life-threatening hemoptysis (3, 4). However, the risks of both death and complications, such as pleural space infection, are higher in chronic cavitary pulmonary aspergillosis (CCPA) than in simple aspergilloma (5). The management of hemoptysis due to CCPA is also often difficult.

A silicone spigot, such as the endobronchial Watanabe spigot (EWS), was developed to obtain surer and longer bronchial blockades than those obtained with conventional methods. Bronchial occlusion using EWS can be applied for persistent air leaks in pneumothorax, postoperative or traumatic lung fistula, empyema with fistula, and fistula with other organs (6). Recently, the efficacy of bronchial occlusion using EWS for hemostasis has been reported (7-12). This procedure is usually performed for temporary hemostasis in conjunction with additional treatment methods, such as surgery and bronchial pulmonary embolization (BAE); however, both the long-term efficacy and safety of this method remain unclear.

We herein report the cases of two patients with persistent moderate hemoptysis due to CCPA who were not indicated for surgery, but were able to undergo successful long-term management with bronchial occlusion using EWS.

Case Reports

Case 1

A 62-year-old man with a history of coronary bypass surgery for myocardial infarction was referred to our hospital

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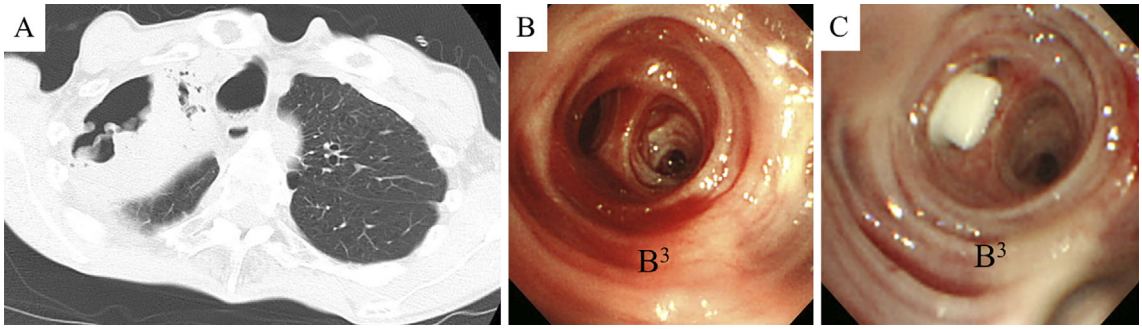

Figure 1. **Chest computed tomography taken at admission. Fluid retention and amorphous matter in the bullous cavity at the right lung apex are shown (A). Bronchoscopic findings on days 7 (B) and 9 of hospitalization (C). Active bleeding from the right B^{3}b is shown (B). A 7-mm spigot was placed in the right B^{3}b (C).**

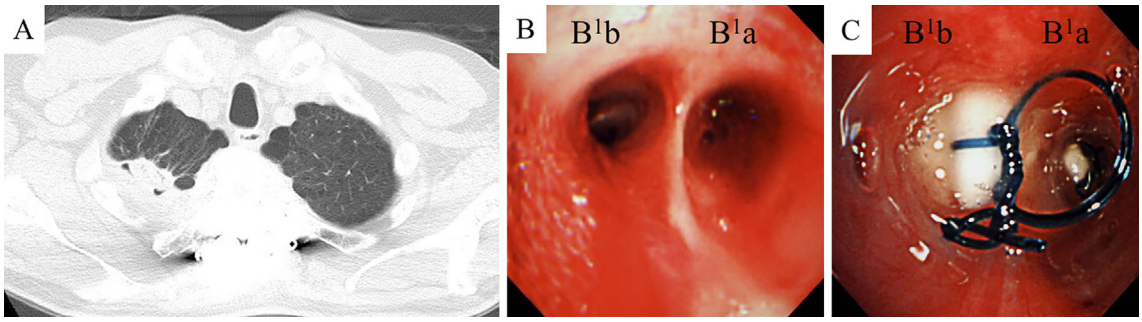

Figure 2. **Chest computed tomography taken at admission. Spheroidal matter in the cavity of the right lung apex is shown (A). Bronchoscopic findings on days 6 (B) and 8 of hospitalization (C). Active bleeding from the right B^{1}a and B^{1}b is shown (B). In each bronchus, 7-mm spigots were placed using sutures for easy removal (C).**

due to moderate hemoptysis persisting for 2 days. His body mass index was 14.7 kg/m^2. Chest computed tomography (CT) revealed the retention of fluid containing amorphous matter and blood in a bullous cavity at the right lung apex (Fig. 1A). A tentative diagnosis of pulmonary aspergillosis was made. Surgical resection was considered difficult due to the presence of severe emphysema and a reduced cardiac function (ejection fraction, 30%). BAE was considered difficult because contrast CT suggested complicated collateral vascular channels of the non-bronchial systemic artery. Although tranexamic acid was administered, hemoptysis (100-200 mL/day) persisted. Therefore, the patient's general condition deteriorated. On day 7 of hospitalization, under intubation and mild venous anesthesia with midazolam, bronchoscopy was performed to treat the hemoptysis. As a result, active bleeding from the right B^{3}b and middle lobar bronchus were identified (Fig. 1B). A spigot measuring 7 mm in diameter was placed in the right B^{3}b, and a 6-mm spigot was placed in the middle lobe bronchus, and thereafter the hemoptysis immediately subsided (Fig. 1C). *Aspergillus fumigatus* was detected in a suctioned sputum culture, and anti-aspergillus antibody was positive; thus, CCPA was diagnosed. After bronchial occlusion, the patient was treated with micafungin, followed by maintenance therapy with voriconazole. Although additional treatments, such as surgical intervention and BAE, were not performed to treat pulmonary aspergillosis, hemoptysis did not recur for 34 months after the placement of spigots, until the patient died due to aspiration pneumonia.

Case 2

A 66-year-old man who had been hospitalized elsewhere for the treatment of a femoral fracture was transferred to our hospital due to the persistence of intermittent moderate hemoptysis (50-100 mL/day) for >1 month despite tranexamic acid treatment. He had a history of pulmonary tuberculosis. Chest CT revealed the presence of spheroidal matter in a cavity at the right lung apex (Fig. 2A). The patient had severe emphysema and hemiplegia due to thoracic cord injury. Although pulmonary aspergillosis was suspected, surgical resection was considered difficult because of his poor pulmonary function and performance status. Bronchoscopy was performed under intubation and mild venous anesthesia with midazolam. Active bleeding from the right B^{1}a and B^{1}b was observed (Fig. 2B), and hemoptysis immediately subsided after 7-mm spigots were inserted into each bronchus (Fig. 2C). CCPA was diagnosed and treated with micafungin, followed by maintenance therapy with voriconazole. Thereafter, the patient developed obstructive pneumonia in the peripheral region that had been occluded using EWS;

Table. Literature Review of Hemoptysis Cases Treated by Bronchial Occlusion with EWS and Their Clinical Findings.

Reference No.	Age	Sex	Underlying disease	Amount of hemoptysis	Localization	No. of spigots	Spigot size (mm)	Hemostasis by bronchial occlusion	BAE after bronchial occlusion	Additional treatment	Spigots in place time	Removal of spigots	Follow-up months
(7)	39	F	unknown	massive	RUL	1	6	yes	yes	no	0 days	yes	ND
(8)	48	M	overdose	moderate	LLL	1	6	yes	yes	no	4 days	yes	19.4
(8)	56	F	unknown	moderate	LUL	1	5	yes	yes	no	12 days	yes	14
(8)	83	F	unknown	moderate	LUL	3	5	yes	yes	no	8 days	yes	1.2
(8)	55	F	unknown	moderate	LUL	1	5	yes	yes	lobectomy	4 days	yes	33
(8)	72	M	lung cancer	moderate	RUL	2	6	yes	no	no	210 days	no	8.4
(8)	77	F	lung cancer	moderate	LUL	1	6	no	yes	cyanoacrylate glue	11 days	yes	2
(8)	75	F	bronchiectasis	moderate	RML	1	7	yes	yes	no	4 days	yes	1
(8)	66	M	lung cancer	moderate	RUL	1	7	no	yes	no	6 days	yes	3.5
(8)	61	M	lung cancer	moderate	RUL	2	5, 6	yes	no	lobectomy	3 days	yes	0.4
(9)	65	M	lung cancer	massive	LUL	ND	ND	yes	yes	no	1 days	yes	ND
(10)	57	F	NTM	massive	RML	2	6	no	yes	no	15 days	yes	1.4
(11)	63	M	unknown	massive	LUL	1	6	yes	yes	bronchial occlusion	4 months	yes	4
(12)	78	F	bronchiectasis	massive	LLL	1	7	yes	yes	no	4 days	yes	1
Case 1	62	M	aspergillosis	moderate	RUL, RML	2	6, 7	yes	no	no	34 months	no	34
Case 2	66	M	aspergillosis	moderate	RUL	2	6	yes	no	no	58 months	no	58

EWS: Endobronchial Watanabe Spigot, No.: number, F: female, M: male, NTM: nontuberculous mycobacteriosis, RUL: right upper lobe, RML: right middle lobe, LUL: left upper lobe, LLL: left lower lobe, ND: not described, BAE: bronchial artery embolization

however, he rapidly recovered after the administration of antibacterial agents. There has been no recurrence of hemoptysis since the placement of the spigots 58 months ago.

Discussion

In the two cases of CCPA, our findings demonstrated that bronchial occlusion using EWS was effective not only for

obtaining temporal hemostasis, but also for the long-term management of hemoptysis.

Bronchoscopy plays an essential role in the management of hemoptysis because it helps identify the origin of hemoptysis and thereby endoscopically treat accessible lesions (13). Bronchial occlusion using EWS is performed to control any hemorrhaging from peripheral lung lesions. The data and clinical findings of previous reports and the present two cases of hemoptysis treated with bronchial occlusion using EWS are described in Table (7-12). In 2006, Dutau et al. first reported a case of massive hemoptysis due to idiopathic bronchial hemorrhaging that was successfully treated with bronchial occlusion using EWS (7). Subsequently, Bylicki et al. performed a retrospective study of bronchial occlusion using EWS for moderate hemoptysis and reported that rapid hemostasis was achieved in seven of nine cases (8). Adachi et al. reported a case of massive hemoptysis wherein complete hemostasis was not obtained through bronchial occlusion using EWS, but the respiratory condition was stabilized by reducing the amount of bleeding, and BAE could thereafter be performed (10). In these reports, bronchial occlusion using EWS was performed for temporary hemostasis in conjunction with definitive surgery and BAE, and in most cases, silicone spigots were removed within 2 weeks of BAE. Meanwhile, in one case, bronchial occlusion using EWS was effective for the treatment of massive hemoptysis that could not be stopped with BAE (11). Additionally, bronchial occlusion using EWS can be applied in combination with BAE or as a definitive treatment in some situations.

The highlight of this report is that the continuous placement of silicone spigots was effective for the long-term management of hemoptysis due to CCPA. Although BAE is the standard conservative hemostatic method for hemoptysis due to CPA, the success and recurrence rates are reported to be approximately 50-90% and 30-50%, respectively (14-16). These treatment outcomes were generally poorer than those observed in hemoptysis due to other causes or because of the involvement of complex collateral vascular channels in CPA. If performing BAE is considered difficult due to the involvement of complex collateral vascular channels on contrast CT and/or CT angiography, then bronchial occlusion with EWS might be performed prior to BAE. Rebleeding is often difficult to treat and fatal. Hence, in diseases with high recurrence rates, once bronchial occlusion with EWS is successful, then the long-term placement of silicone spigots might prevent rebleeding (as seen in the present cases).

A previous study evaluated the safety of the long-term placement of silicone spigots in 21 patients with pneumothorax. The median follow-up period was 19 months, and the incidence of major complications was only 5% (obstructive pneumonia in one case) (17). In another study of 24 patients with pneumothorax, only 4 patients required the removal of silicone spigots-one due to hypoxic atelectasis, one due to lung abscess, and two due to the patient's request. In the remaining 20 patients, silicone spigots were permanently placed without any late-phase complications during the follow-up period (18). In one of the two cases discussed in this report, obstructive pneumonia was noted and it was rapidly treated with an antibacterial agent without removing the silicone spigots. Therefore, the long-term placement of silicone spigots is considered to be tolerable even in patients with hemoptysis. However, the long-term safety of bronchial occlusion using silicone spigots for patients with hemoptysis should be further investigated because most of the previous reports discussed only the short-term placement of silicone spigots.

For CCPA, long-term, perhaps lifelong, antifungal treatment is required. In CPA, the response to systemically administered voriconazole is favorable, with an improvement in the symptoms and stabilization or improvement in anti-aspergillus antibody titers and radiologic findings (19). Recently, micafungin has been reported to be as effective as and significantly safer than voriconazole for the initial treatment of CPA (20). In our two cases, effective antifungal treatment might have contributed to the successful long-term hemostasis obtained in both cases.

In conclusion, we herein described two cases of moderate hemoptysis due to CCPA, in which successful long-term management was achieved through bronchial occlusion using EWS. In diseases with a high recurrence rate of hemoptysis, the continuous placement of silicone spigots might be effective to prevent rebleeding. Additional cases are required to clarify the long-term efficacy and safety of this method.

The authors state that they have no Conflict of Interest (COI).

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