CASE REPORT

Management Considerations for the Treatment of Idiopathic Massive Hemoptysis with Endobronchial Occlusion Combined with Bronchial Artery Embolization

Takashi Adachi¹, Masahide Oki² and Hideo Saka²

Abstract

This paper describes endobronchial embolization using silicone spigots (EESS), which is a potential treatment option for hemoptysis. A 63-year-old man with massive hemoptysis was treated with EESS to the left B³, and bronchial artery embolization (BAE) was subsequently performed. However, the patient’s hemoptysis persisted and we performed another bronchoscopy. Bleeding was found from the left B¹². This was also treated with EESS. Subsequently, the patient achieved complete hemostasis with no complications for four months. EESS can prevent suffocation and can be a definitive treatment for achieving hemostasis in patients with recurrent hemoptysis after BAE.

Key words: Bronchial artery embolization, endobronchial embolization, massive hemoptysis, silicone spigots, Watanabe spigots

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Introduction

Massive hemoptysis should always be considered to be a life-threatening condition that requires a rapid response to prevent suffocation due to blood accumulation. Bronchial artery embolization (BAE) or lobectomy are considered to be the definitive therapies for massive hemoptysis, however, in many facilities, it is not always possible to perform these procedures with the required level of immediacy.

Bronchoscopy is an essential part of the management for hemoptysis because it allows for the localization of the origin of hemoptysis and the endoscopic treatment of the accessible lesions (1). Endobronchial embolization using silicone spigots (EESS) is known to be an effective treatment for fistulous diseases of the lungs (2, 3). More recently, articles have reported EESS as a temporary treatment for hemoptysis, as it can secure the respiratory tract until definitive treatment (4-6). However, there have been no reports on the use of EESS as a definitive treatment for refractory hemoptysis.

We herein report a case of idiopathic massive hemoptysis in which EESS treatment prevented suffocation due to bronchial bleeding. Furthermore, the patient’s recurrent hemoptysis after BAE was definitively treated with an additional EESS procedure.

Case Report

A 63-year-old man was transferred to the emergency room due to the sudden occurrence of massive hemoptysis (estimated to be >200 mL) in the middle of the night. We diagnosed the patient with massive hemoptysis and judged it to be an emergency case. Although a chest X-ray suggested that the left upper lobe was the cause of the bleeding (Fig. 1a), structural destruction of the lung and bronchiectasis lesions were not found on computed tomography (CT) (Fig. 1b). The patient’s medical history included essential hypertension, but he had not been treated with antiplatelet or anticoagulant drugs. After the patient arrived at our hospital, we performed flexible bronchoscopy with an 8-mm diameter endobronchial tube under local anesthesia to prevent mas-

¹Department of Respiratory Medicine, National Hospital Organization, Higashinagoya National Hospital, Japan and ²Department of Respiratory Medicine, Nagoya Medical Center, Japan

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Correspondence to Dr. Takashi Adachi, tadachi@med.nagoya-u.ac.jp
sive bronchial hemorrhage with acute respiratory distress. The bronchoscopic findings showed that the patient’s left main bronchus was completely filled with blood, and that the active bleeding site was in the left upper segment (Fig. 2a). The blood was removed and the original bleeding site was identified on the left B3 by a balloon occlusion test. At this point, bleeding from B1+2 was not apparent. A 6-mm diameter silicone spigot (Endobronchial Watanabe Spigots, EWS®; Novatech, La Ciotat, France) was placed then in the left B3 segment (Fig. 2b), which achieved temporary hemostasis, however, slight hemoptysis remained when the patient coughed.

Four days after admission, the patient underwent bronchial arterial angiography (BAA). Arteriography was also performed in the intercostal artery, internal thoracic artery, and lateral thoracic artery; however, the origin of bleeding could not be clearly identified by BAA. As a result of the findings of chest CT and BAA, he was diagnosed with idiopathic bronchorrhagia, which was assumed to be in the left superior bronchial artery. Gelatin particles and metallic coils were therefore inserted.

Eight days after admission, the patient’s slight hemoptysis persisted. We thus performed another bronchoscopy to identify the other sites of bleeding. After identifying a new bleeding site at the left B1+2 (Fig. 3a), we inserted a 6-mm diameter EWS (Fig. 3b, 4).

Eleven days after admission, hemostasis was achieved and no re-bleeding or other symptoms were observed. The patient was followed on an outpatient basis and was eager to have the spigots removed four months after the onset of hemoptysis. The spigots were therefore carefully removed. Fortunately, there has been no re-bleeding since their removal (Fig. 5).

Discussion

There are few reports on the use of EESS in the treatment of hemoptysis, and there are almost no data on its use as a radical cure. Our study has four major discussion points.

First, we considered how to access the bleeding point.
The use of EESS in the treatment of massive hemoptysis was first described by Dutau et al. (4). Bylicki et al. reported nine moderate hemoptysis cases treated with EESS in 2012 (5). The reported pulmonary intervention was performed under general anesthesia and all patients were intubated with a rigid bronchoscope. In their study, EESS was performed with a flexible bronchoscope through the rigid bronchoscope. This procedure was commonly used by the group in the treatment of hemoptysis. The method has clear advantages over other methods for controlling massive bleeding. Using a rigid bronchoscope and deeper sedation, it is possible to secure the respiratory tract, enabling adequate ventilation. The rigid bronchoscope also provides a wider range of interventional options, such as stent placement or laser coagulation based on situational demands. This approach, although the best to date, is not expected to become a mainstream treatment worldwide (7). Most institutions in Japan are yet to rigid bronchoscopy unit. In the present case, flexible bronchoscopy was performed through an endobronchial tube under sedation with midazolam. Despite the patient’s massive hemoptysis, we achieved hemostasis using EESS with a flexible bronchoscope through an endobronchial tube. In facilities with a rigid bronchoscope, this approach could be selected depending on the amount of bleeding and the status of the patient.

Second, we considered how to insert the EWS into the target bronchus. Initially, to identify the origin, an occlusion balloon is useful (8), and inserting a balloon catheter into the bleeding bronchus may serve as a bronchial tamponade (9). In the present case, we only used a forceps to insert the EWS into target lesions, but we were prepared to use a curette. A curette enables the placement of the EWS into the difficult superior segment and B, however, releasing the EWS requires skill. To prevent migration, the full insertion of the EWS is needed to counteract the pressure of peripheral bleeding; however, it is unclear how an EWS inserted using a curette would be able to tolerate the pressure of bronchial bleeding. There is also a risk that an EWS inserted with a curette may be impossible to remove. If an infection develops the difficult withdrawal of EWS may cause complications.

Third, we investigated the duration of EWS placement. The length of time for which an EWS should remain in place after hemostasis has been achieved remains controversial. Sasada et al. reported that severe infections were not observed in most patients with a permanent EESS (2). However, Lee et al. insisted that temporary insertion is preferable in patients with a current lung infection or patients who have risk factors that predispose them to infection (10). These considerations are for lung fistulous diseases, and no specific duration has been universally accepted for bronchial bleeding. In this case we considered that the spigots should be left in the bronchus for as long as possible, because the EWS was highly effective for this patient; the cause of bronchial bleeding could not be clearly pinpointed, and he was immunocompetent. The spigots were finally removed in accordance with the patient’s wishes. Fortunately, re-
bleeding, migration, and pneumonia did not occur. However, secretion outflow was observed after the removal of the EWS (Fig. 5a). The Long-term placement of an EWS may be undesirable in cases of hemoptysis.

Because the primary purpose of EESS is to prevent suffocation due to hemoptysis, a bleeding bronchus that is identified visually should be occluded to the smallest extent that is possible. In addition, the EWS should be removed promptly to avoid the risk of infection after the responsible bronchus is identified on BAA, and arterial embolization is confirmed. It is important to decide the period of EWS placement according to the status of the patient and the cause of hemoptysis. The incidence of bronchorrhagia caused by chronic respiratory infectious diseases such as nontuberculous acid-fast bacterial disease or pneumomycosis is increasing in Japan, thus we can expect to see more data relating to EESS treatment.

Finally, we considered the use of EESS as a radical treatment for idiopathic hemoptysis. Preventing suffocation using a bronchoscope and then proceeding to a definitive therapy, such as BAE, is the normal procedure of hemoptysis treatment. A high percentage of hemoptysis cases have been resolved successfully with BAE (11) and, as a result, this is the primary therapy for hemoptysis; however, some institutions have no BAE facilities or specialists. In addition, although BAE is a superior treatment, it is not without issues. A few articles have reported the risk of hemoptysis recurrence and various complications of BAE. Shao et al. reported that 21.5% of patients had recurrent hemoptysis within one month after BAE, and they mentioned complications such as subintimal dissection, arterial perforation by a guide wire, fever, chest pain, and dyspnea (12). In our case, we were not able to identify the responsible arteries in BAA, but we hypothesized that the left superior bronchial artery was the cause and performed artery embolization because we initially judged that BAE would be advantageous. As a result, the BAE procedure was incomplete and several days later, another EWS had to be placed at B1+2. The effect of BAE was therefore unclear. In addition, we could have rejected BAE because of the above noted risk of re-hemoptysis and complications of artery embolization. If abnormal vessels or the destruction of the lung structure are not identified as the cause of the hemoptysis on CT and/or arteriography, then EESS could enable BAE or lobectomy to be avoided.

We performed EESS not only to prevent suffocation but also to resolve sustained hemoptysis after BAE. There are a few previous reports concerning EESS as a temporary treatment for hemoptysis (5). To the best of our knowledge, this is a rare case in which radical treatment of intractable idiopathic hemoptysis was achieved through the complementary use of BAE. It is unclear whether this method can be generalized to cases of severe bronchial bleeding due to other causes, but clinical investigation is warranted.

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

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