Extensive bronchial occlusion with N-butyl-2-cyanoacrylate for bronchopleural fistula and a destroyed lung

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Bronchopleural fistula, chronic empyema, destroyed lung, endoscopic bronchial occlusion, N-butyl-2-cyanoacrylate.

Abstract
A 72-year-old Japanese man who had undergone resection of a left upper lung carcinoma developed chronic empyema with bronchopleural fistula and destroyed lung 12 years after surgery. Open-window thoracotomy and bronchial occlusion with an endoscopic Watanabe spigot (EWS) were performed to control infection. However, the EWS was easily dislodged due to remarkable bronchial deformation, and he experienced repeated episodes of pneumonia. We performed extensive bronchial filling with N-butyl-2-cyanoacrylate. Stable occlusion was achieved, and there was no recurrence of pneumonia. N-butyl-2-cyanoacrylate was a useful embolic agent because it moulded to the shape of the tracheal lumen and remained in place.

Introduction
Chronic empyema with bronchopleural fistula (BPF) is an intractable complication, in which chronic infection develops, ultimately destroying the lung. Purulent sputum from the empyema cavity and destroyed lung cause severe or recurrent pneumonia in the healthy contralateral lung. [1] In recent years, bronchial occlusion using the endobronchial Watanabe spigot (EWS®, Novatech, La Ciotat, France) and the other materials is performed by flexible bronchoscopy in order to promote fistula closure and prevent pneumonia. However, the ready-moulded product is difficult to insert into the deformed bronchus and is easily dislodged. Here, we report a case of chronic empyema with BPF and destroyed lung in which bronchial occlusion with N-butyl-2-cyanoacrylate (NBCA; Histoacryl®, B. Braun, Melsungen, Germany) and lipiodol (Laboratoire Guerbet, Aulnay-Sous-Bois, France) resulted in the improvement of respiratory distress and recurrent pneumonia.

Case Report
The patient was a 72-year-old man. At 57 years of age, he had undergone left upper lobe and lower superior segment (S6) sleeve resection with pulmonary angioplasty for left upper lobe lung cancer. Since then, there had been no recurrence. At approximately 4 years after surgery, the left lung slowly collapsed and was ultimately destroyed. At 10 years after surgery, he experienced repeated episodes of right pneumonia over a short period of time. Bronchoscopy at 12 years after surgery showed chronic empyema with BPF and destroyed lung. Open-window thoracotomy was performed at 12 years after the operation. Numerous fistulas were observed on the surface of the destroyed lung, and air leakage and purulent sputum flowed out from the fistulas (Fig. 1A,B).

Because the patient was elderly and had a decreased performance status, completion pneumonectomy was not indicated. We therefore attempted to occlude all bronchi in the remaining left lung with a combination of EWS, fibrin glue, and a polyglycolic acid sheet to reduce air leakage. However, the EWS was easily dislodged due to remarkable bronchial deformation, and he experienced repeated episodes of pneumonia. We performed extensive bronchial filling with N-butyl-2-cyanoacrylate. Stable occlusion was achieved, and there was no recurrence of pneumonia. N-butyl-2-cyanoacrylate was a useful embolic agent because it moulded to the shape of the tracheal lumen and remained in place.
leakage and prevent recurrent pneumonia. However, the occlusion was repeatedly dislodged and required reinsertion 13 times in the 2 years after open-window thoracotomy due to bronchial deformation. Every time the embolus was dislodged, the patient developed pneumonia with severe hypoxemia. We therefore decided to perform bronchial occlusion with NBCA and lipiodol.

The operation was started under general anaesthesia. After removing poorly fixed EWS, a 2:1 mixture of NBCA and lipiodol was slowly injected, in order to not form a mist, using a spray tube with a flexible bronchoscope under fluoroscopic monitoring. The mixture was slowly infused from the sub-segmental bronchus of the remaining left basilar segment to the left main bronchial inlet. The mixture was injected at a rate of approximately 0.1 mL/sec; finally, 5.6 mL of the mixture was used (Fig. 2).

After the occlusion, purulent sputum was reduced, and respiratory distress was improved. Outflow of purulent sputum was observed from the fistula on the surface of the destroyed lung, as it had been before occlusion, and post-operative computed tomography (CT) showed no abscess formation in the lung (Fig. 1C). In the 6 months since bronchial occlusion, there has been no recurrence of pneumonia. If sterilization of the empyema cavity and the destroyed lung is achieved in the future, we will consider a closed-window surgery.

Discussion

Chronic empyema with BPF is refractory, and open-window thoracotomy is performed to control infection. Permanent fenestration may be required when it is difficult
to purify the empyema cavity and residual lung. In order to prevent pneumonia of the healthy contralateral lung due to purulent sputum from the destroyed lung, we administered only macrolide antibiotics for a long time in proportion to the treatment of chronic lower respiratory tract inflammation. We did not use broad-spectrum antibiotics for intrathoracic purification because long-term use of broad-spectrum antibiotics promotes the emergence of resistant bacteria in the destroyed lung. In this situation, when the sputum derived from the destroyed lung flows into the healthy contralateral lung, and pneumonia develops, the antibacterial drug may be ineffective, and the patient becomes fatal. In a previous study, pneumonectomy of the affected lung was associated with an overall mortality rate of 2.4–5.9% [2,3]. In this case, we judged surgery to have a high risk due to the presence of advanced age and poor performance status.

In recent years, endoscopic bronchial occlusion is often performed for the treatment of empyema with BPF. In this case, the EWS was repeatedly dislodged and reinserted, and other embolic materials were ineffective. In consideration of the clinical course and because there was no need for removal, bronchial occlusion with NBCA was performed. It has been reported that occlusion with an EWS can allow for long-term indwelling for 5 years or more without complications. With sufficient patient selection, permanent embolization of the bronchi is acceptable regardless of the embolic material [4].

NBCA is originally a liquid that polymerizes and hardens when it comes into contact with the anions of blood or body fluid. NBCA and lipiodol are often mixed and used together to adjust the polymerization time of NBCA and confirm the site after embolization. NBCA has a very short polymerization time, but the time until polymerization can be extended by mixing with lipiodol. No reports have examined the mixing ratio of NBCA and lipiodol for bronchial embolization. Thus, we used a 2:1 mixture of NBCA and lipiodol because the medical package insert for NBCA recommends diluting NBCA to 62.5–75.0% with lipiodol for general usage. NBCA is not carcinogenic and is an inexpensive embolus [5].

The most unique point of our report in comparison to previous studies was not only the blocking of the distal end of the fistula with “a certain amount” of NBCA but also the planning of the bronchial embolization range required for occluding all fistulas without NBCA dropping out and the variation of the amount of NBCA used according to the planned bronchial occlusion range. As a result, we performed extensive embolization with NBCA and were able to stably block all the large and multiple fistulas in this case. Scappaticci et al. reported that the successful closure of a single fistula was obtained in 70% of cases after BPF with a small amount of NBCA (0.5 mL of NBCA) [6]. It has been reported that bronchial embolization using NBCA alone often fails. We believed that this was because the amount of NBCA was insufficient and that the embolus did not form a stable shape with the shape of the bronchi. In this case, it was possible to perform stable embolization by continuously filling with NBCA from a distal sub-segment bronchus to a main bronchial inlet and then creating a “bridge” of NBCA between the bronchi of these subsegments. However, it is necessary to carefully plan the range of embolization for the following reasons: (1) if a functional lung remains, a wide embolus causes obstructive pneumonia; (2) in the destroyed lung, embolization of the peripheral bronchus may cause retention of purulent sputum in the destroyed lung and exacerbate infection.

NBCA moulds to the shape of the tracheal lumen and resists dislodgement. Furthermore, a more stable embolus can be obtained by straddling the bronchial branch. In particular, NBCA is highly useful in cases where removal of the embolus is unnecessary or where another embolization with removable materials is ineffective.

 Disclosure Statement

Appropriate written informed consent was obtained for publication of this case report and accompanying images.

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