One-way endobronchial valve for bronchopleural fistula after necrotizing pneumonia
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Bronchopleural fistula (BPF) formation is a serious complication following pulmonary infections or pulmonary resections. Traditionally, complex BPF has been treated with surgical closure, however, morbidity is significant and many patients are poor surgical candidates. We successfully treated a longstanding BPF following necrotizing pneumonia, which drained into an infected pleural cavity, using a bronchoscopically placed one-way endobronchial valve, originally designed for controlling pulmonary emphysema.

A Caucasian man in his twenties with a history of Charcot-Marie-Tooth disease was travelling in Thailand when he acquired severe pneumonia and sepsis. Computed tomography (CT) showed extensive left upper lobe necrotizing pneumonia and empyema caused by bacterial infection with *Burkholderia pseudomallei*. Despite optimal treatment with ceftazidime and insertion of chest tubes, his condition deteriorated. Five weeks later he was transported by air ambulance to Iceland in a septic state. An emergency thoracotomy was performed. Most of the left upper lobe of the lung was necrotic and it was removed (sublobar resection) together with decortication of the left lower lobe. The patient’s condition improved but postoperative treatment was complicated by a productive cough and continuous air leakage that was treated with a Heimlich valve connected to a chest tube. He was discharged and followed up by air ambulance to Iceland in a septic state. An emergency thoracotomy was performed. Most of the left upper lobe of the lung was necrotic and it was removed (sublobar resection) together with decortication of the left lower lobe. The patient’s condition improved but postoperative treatment was complicated by a productive cough and continuous air leakage that was treated with a Heimlich valve connected to a chest tube. He was discharged and followed up by CT at 3 months postoperatively, which revealed a 4-mm BPF between the left apicoposterior segmental bronchus and an air- and pus-filled pleural cavity in the left upper hemithorax (Figure 1a). After 6 months of unsuccessful treatment with chest tubes and antibiotics, he still had air leakage and a productive cough with positive sputum cultures. He was referred to Skane University Hospital in Sweden where a one-way valve was placed bronchoscopically in an attempt to close the BPF: a treatment option used for lung resection in patients with severe pulmonary emphysema. Using flexible bronchoscopy, 4- and 5.5-mm Zephyr valves (Pulmonx, Redwood City, CA, USA) were inserted in the B1 and B3 segmental branches, respectively, whereupon air leakage immediately ceased. Infectious parameters normalized in the next weeks, and a follow-up CT one month later showed that the cavity in the left hemithorax had subsided (Figure 1b). Two years after initial presentation, both valves were removed bronchoscopically, without any difficulties. A CT scan after removal of the stents showed only a minor apical pleural cavity and an almost normal appearance of the inferior lobe (Figure 1c). The patient has had no signs of infection, is free of cough, and reports decreasing dyspnea and fatigue.

Complex BPF, especially those related to empyema and extensive necrosis of the lung, present a clinical challenge. Historically, invasive procedures with muscle flaps, thoracoplasty, or omentoplasty have been used to close the fistula; however, these procedures are often technically challenging with significant long-term morbidity. We decided to place an endobronchial one-way valve to close the fistula, and both the air leakage and cough ceased immediately. A week later, the chest tubes could be removed and there were no signs of recurrent infection in the left pleural cavity. We used the Zephyr cylinder-shaped valve that is composed of nitinol covered with silicon. The valve allows the retrograde passage of air and infected secretions, but blocks anterograde air flow to the distal airways. The use of endobronchial one-way valves to close a BPF has been described previously, but reported cases are few, and most have involved high-risk patients such as those with diffuse lung metastases or aspergillosis sepsis. Our patient was a previously healthy young man who presented with a large and persistent BPF that drained into an infected pleural cavity. He was deemed to be a surgical candidate, but other minimally invasive treatment options that have been described for smaller BPF (sclerosant, tissue glue, coils, or endobronchial spigots via bronchoscopy) were not deemed feasible due to the chronically infected cavity.

The BPF in this case was caused by necrotizing pneumonia and empyema secondary to *Burkholderia pseudomallei* infection, a relatively low-virulent soil bacteria found in SE Asia, and NE Australia. These infections, also called pulmonary melioidosis, can be
life-threatening and often require surgical drainage or resection together with antibiotics. To the best of our knowledge, this is the first reported case of treatment for BPF related to pulmonary melioidosis. This case shows that one-way endobronchial valves can be used for a complex BPF, even in cases following extensive pulmonary infections with infected intrathoracic cavities. Although the indication for the procedure in question would be regarded as “off-label”, the treatment was successful and spared this young patient extensive surgery.

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References

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Figure 1. (a) Computed tomography showing necrotizing pneumonia and abscess formation in the left superior lung lobe. (b) Two one-way valves in the left upper segmental branches 2 months after endobronchial placement with a bronchoscope. (c) Computed tomography 2 years later, after the valves had been removed.