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INTERVENTIONAL BRONCHOSCOPY

Emergency Use of an Endobronchial One-Way Valve in the Management of Severe Air Leak and Massive Subcutaneous Emphysema

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Abstract Bronchopleural (BPF) and alveolar-pleural (APF) fistulas are frequently encountered in clinical practice with persistent air leaks that can lead to significant morbidity, prolonged hospital stay, and potentially increased mortality. BPF and APF are commonly related to pulmonary resections. Other etiologies include minimally invasive procedures (thoracentesis and image-guided biopsies), and spontaneous fistulas related to an underlying structural lung disease (e.g., emphysema) or a necrotizing pulmonary process (e.g., infection or malignancy). Radiofrequency ablation for pulmonary malignancies is an effective modality that can rarely lead to APF with persistent air leak. Surgical intervention remains the standard treatment option for BPF and APF. A variety of minimally invasive bronchoscopic approaches can be considered for selected nonsurgical candidates. The use of one-way endobronchial valves to manage severe and persistent air leaks can be considered a minimally invasive option in selected patients. The valves selectively block inspiratory airflow to a specific segmental or subsegmental airway but allow expiratory flow with drainage of air and secretions from the corresponding distal airways and lung parenchyma.

Keywords Bronchopleural fistula · Alveolar-pleural fistula · One-way endobronchial valve · Lung cancer · Radiofrequency ablation

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Introduction

Bronchopleural fistula (BPF) and alveolar-pleural fistula (APF) are abnormal communications between the tracheobronchial tree and/or the alveolar spaces and the pleural space, with persistent air leak for more than 24-48 hours. BPF and APF are common problems in clinical practice and are associated with a significant morbidity, prolonged hospital stay, and mortality [1]. Air leaks have been described in many pulmonary diseases but remain one of the common complications after pulmonary resection. If air leaks do not resolve within several days, the likelihood of resolution with conservative management that includes chest tube drainage decreases significantly. Historically, the management of persistent air leaks related to BPF and APF had required thoracic surgical repair with video-assisted thoracoscopic surgery or thoracotomy. Recently, however, nonsurgical bronchoscopic approaches have been employed in an attempt to treat BPF and APF without surgical intervention. This would be advantageous for patients with poor pulmonary or cardiovascular reserve and multiple medical problems, where general anesthesia and surgical intervention may incur significant further morbidity and mortality. In this article we describe the use of an endobronchial one-way valve (IBV Valve®, Spiration, Inc., Redmond, WA) to treat an alveolar-pleural fistula with rapidly progressive massive subcutaneous emphysema and respiratory failure.

Case Report

A 63-year-old female patient had a history of partial left-upper-lobe resection and left-lower-lobe wedge resection for non-small-cell lung cancer 3 years prior to this

admission. She presented to our institution with a small left pneumothorax and subcutaneous emphysema 8 days after radiofrequency ablation of a left-upper-lobe recurrent lung cancer. The ablation was around the same site of the initial wedge resection in the posterior subsegment of the left upper lobe. She was initially treated with supplemental oxygen and serial radiographs showed stability of the subcutaneous emphysema with resolution of the pneumothorax. Three days after admission, she developed sudden rapidly progressive subcutaneous emphysema and respiratory failure requiring mechanical ventilation and left chest tube placement. On physical examination, she was noted to have massive subcutaneous emphysema involving her entire body from the head to the lower extremities (Fig. 1). She had coarse breath sounds on left anterior chest examination. A large continuous air leak from the chest tube during inspiration and expiration was noted while she was on mechanical ventilation. The air leak was felt to be related to post-ablation tumor necrosis with a subsequent tear in the visceral pleura and surrounding adhesions around the surgical resection lines from the previous left-upper-lobe and left-lower-lobe wedge resections. The patient underwent bronchoscopy to evaluate and potentially treat the persistent air leak as she was deemed to be at high risk for complications related to potential general anesthesia and surgical intervention. Sequential occlusion of the segments of the left-side airways was performed using a balloon catheter. Significant improvement in the air leak was noted after occlusion of the left-upper-lobe apical-posterior segment and the left-lower-lobe superior segment. Fibrin glue was injected in the apical-posterior segment and the superior segment without significant improvement in the air leak. The segmental airways were then measured for bronchial valve placement using a calibrated balloon catheter [2]. A 7-mm endobronchial valve (IBV Valve®, Spiration, Inc.) was placed in the left-upper-lobe apical-posterior segment airway and a 6-mm valve was placed in the left-lower-lobe superior segment airway (Fig. 2). The patient tolerated the procedure well and her vital signs remained stable. The air leak stopped almost completely within 15 min of completion of the procedure. The subcutaneous emphysema gradually improved and resolved completely during follow-up (Fig. 1). The patient was eventually extubated and transferred to a regular ward. Unfortunately, despite the resolution of the air leak and subcutaneous emphysema, she had multiple unrelated complications, including bacteremia, fungemia, sepsis, gastrointestinal bleeding, and recurrent pneumonia that required prolonged mechanical ventilation; the patient eventually died during the same hospitalization.

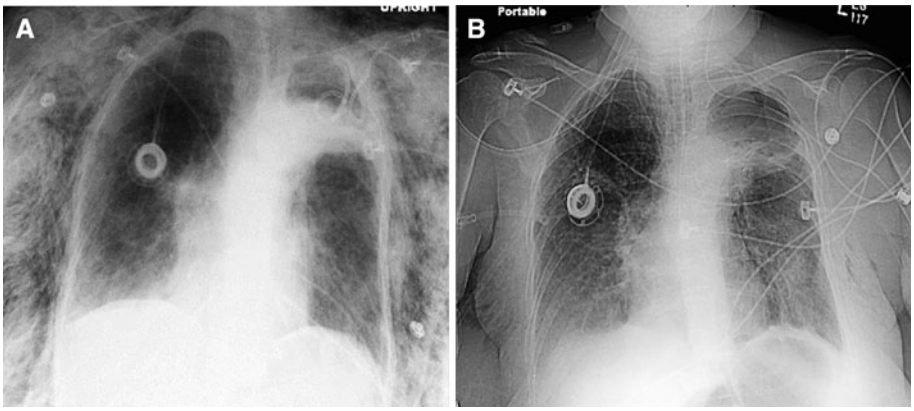

Fig. 1 Chest radiographs of the patient **a** prior to the procedure with massive subcutaneous emphysema and chest tube in the left pleural space, and **b** 2 weeks after the procedure with resolution of the subcutaneous emphysema

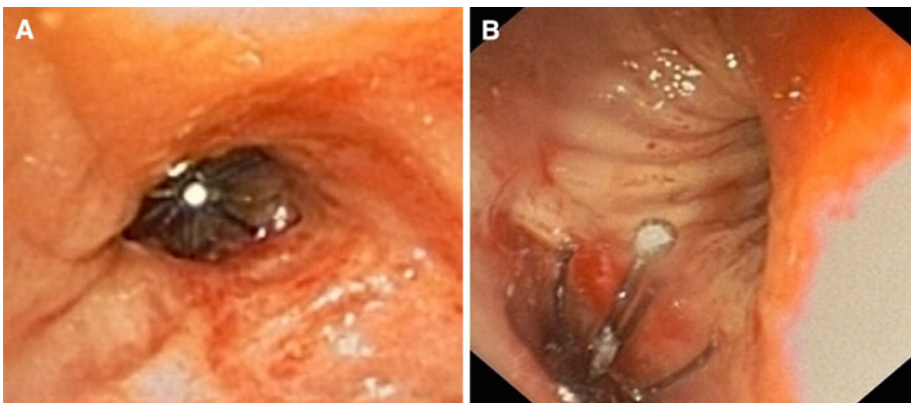

Fig. 2 One-way endobronchial valves (IBV Valve®, Spiration, Inc.) in the airways after placement: **a** 7-mm left-upper-lobe apical-posterior segment valve, and **b** 6-mm left-lower-lobe superior segment valve

Discussion

The terms bronchopleural fistula (BPF) and alveolar-pleural fistula (APF) describe an abnormal communication between the tracheobronchial tree and/or the alveolar spaces and the pleural space, with persistent air leak for more than 24–48 hours. The most common etiology of BPF and APF is pulmonary resection. The incidence of persistent air leaks after pulmonary resections is in the range of 1–46% [1, 3, 4] depending on the extent of the resection and the specific definition of this condition. Air leaks are thought to be more common after larger anatomical pulmonary resection (i.e., lobectomy vs VATS wedge resection). Less common etiologies of persistent air leaks include minimally invasive procedures such as thoracentesis, image-guided parenchymal lung biopsies, and radiofrequency ablation. Spontaneous APF has also been described with structural lung disease (e.g., bullous emphysema and other cystic parenchymal disorders), lung infections, and structural lung diseases that can lead to necrosis or direct destruction of the lung parenchyma and the visceral pleural surface (e.g., pneumonia, lung cancer).

The diagnosis of BPF or APF requires identification of the cause of the air leak and the location of the injury to the bronchial tree, lung parenchyma, and visceral pleura. In one study, computed tomography (CT) was used to identify the specific location of persistent air leaks with a success rate of approximately 55% [5]. Other techniques such as inhalation of labeled gaseous mixtures (O_2 and N_2O) and detection in the chest drainage container have also been described [6]. Bronchoscopy is one way to detect the location of the injury and the airway or airways feeding this location by direct visualization (BPF), in addition to other techniques that can be used to detect the specific location and severity of the air leak. Methylene blue can be instilled through the bronchoscope while observing the change in color of the chest tube drainage. Sequential bronchoscopic balloon catheter occlusion while observing the severity of the air leak is the most widely used approach [7]. Bronchoscopy also allows direct evaluation of the stump in the case of lung resection and can provide microbiological and cytological samples to exclude infection and malignancy as potential etiologies.

Image-guided radiofrequency ablation (RFA) is a minimally invasive procedure that is used to treat both primary lung malignancy and metastatic disease to the lung. Originally described in 2000 by Dupuy et al. [8], RFA may offer potential cure for patients with stage I non-small-cell lung cancer less than 3 cm in diameter and palliation for patients with larger tumors. The incidence of pneumothorax after RFA is in the range of 15-45% and 20% require chest tube placement [9]. In one study, the rate of intractable pneumothoraces from RFA was 0.6%, which was thought to be related to necrosis of the lung tissue between the bronchus and the pleural space, leading to APF and potentially BPF [10]. While the diagnosis of pneumothorax is often made immediately after RFA, delayed pneumothoraces up to 5 days after the procedure have been reported [11]. Our patient had a delayed presentation of a small pneumothorax and then rapidly progressive subcutaneous emphysema probably related to delayed tissue necrosis after the ablation procedure.

Treatment of BPF and APF with persistent air leak had been limited to surgical repair, pleurodesis, or continued tube thoracostomy drainage. Surgical repair remains the standard of care for the management of persistent air leaks. However, the management of patients with poor cardiopulmonary reserve and who are at risk for further morbidity and mortality related to general anesthesia and surgical interventions always presents a challenge. Specific surgical approaches include video-assisted thoracoscopic surgery (VATS) with repair and mechanical or chemical pleurodesis, and thoracotomy with a variety of techniques that include repair with mechanical or chemical pleurodesis, the use of muscle or omentum flaps, bronchial resection, aggressive debridement, closure of a dysfunctional stump, and antiseptic packing of the pleural space. Surgical approaches are usually effective and generally well tolerated and safe in good surgical candidates. Success rates of these techniques for the management of BPF and APF vary between 84 and 95%, but these techniques have been limited by infectious complications and persistent air leaks [7, 12, 13]. Minimally invasive bronchoscopic approaches were attempted as an alternative to surgical approaches, especially in patients who are poor surgical candidates because surgery and general anesthesia carry the risk of further significant morbidity and mortality. These approaches include bronchoscopic instillation of ethanol [14], fibrin glue [15], coiling [16], and occluding devices [17]. More recently, several reports have described the use of one-way endobronchial valves to treat persistent air leaks [18–21]. The valves selectively block inspiratory airflow to a specific segmental or subsegmental airway in the tracheobronchial tree but allow expiratory flow with drainage of air and secretions from the corresponding distal airways and lung parenchyma.

To our knowledge, this is the first published case of an emergency use of a one-way endobronchial valve to manage severe air leak with rapidly progressive massive subcutaneous emphysema and respiratory failure. Our patient likely developed the severe persistent air leak as a consequence of recent RFA therapy to manage lung cancer, with subsequent tissue necrosis and injury of the visceral pleura, lung parenchyma, and possibly the distal tracheobronchial tree in that area. Her history of surgical intervention that involved the left upper lobe and left lower

lobe, with adhesions involving the visceral pleural surface, previous surgical staple lines within the treated area, and positive-pressure ventilation probably all contributed to the subsequent development of a persistent, severe, and rapidly progressive air leak. The response of the air leak to this minimally invasive management approach was both rapid and impressive. The air leak stopped almost completely within 15 min after placement of the two valves in the left-upper-lobe apical-posterior segment and the left-lower-lobe superior segment and resolved completely within a few days. The IBV valve is deployed using a plastic catheter through the working channel of the flexible bronchoscope. It limits the airflow to the distal bronchopulmonary segments while allowing secretions and air to escape to the more proximal central airways. The valves can be safely removed after deployment. The IBV valve system was developed as a minimally invasive bronchoscopic approach to manage severe upper-lobe-predominant emphysema. The pilot data regarding the safety and effectiveness of the IBV system in the management of severe upper-lobe-predominant emphysema were presented in two recent reports [2, 22]. The system is undergoing further evaluation in patients with severe upper-lobe-predominant emphysema with significant functional limitations despite maximum medical management in a pivotal prospective, randomized, blinded, placebo-controlled trial that involves more than 30 sites at this point. The device has received humanitarian device exemption (HDE) approval from the United States Food and Drug Administration (FDA) to control prolonged air leaks or air leaks likely to become prolonged from the lung after lobectomy, segmentectomy, or lung volume reduction surgery. However, the effectiveness of this device for the management of prolonged air leaks has not been demonstrated. This case report demonstrates the potential role of this one-way endobronchial valve system in the management of persistent air leaks, especially in patients with limited pulmonary and cardiovascular reserve where general anesthesia and thoracic surgical interventions are potentially associated with additional morbidity and mortality.

Conclusion

Persistent air leaks related to BPF and APF are frequently encountered in clinical practice and can be associated with significant morbidity and mortality. Thoracic surgical management remains the standard treatment option for this condition. Minimally invasive bronchoscopic approaches are additional management options for persistent air leaks, especially in poor surgical candidates where thoracic surgical intervention under general anesthesia poses significant additional risks of morbidity and mortality. One-way endobronchial valve systems such as the IBV system used in this patient (IBV Valve®, Spiration, Inc.) were designed as a nonsurgical management option for severe upper-lobe-predominant emphysema and significant functional limitation despite maximum medical management. This system can be considered for treating persistent air leaks, other air leaks that are likely to become prolonged, or rapidly progressive air leaks. Further studies are necessary to establish a more definitive role of one-way endobronchial valves in the management of persistent air leaks related to BPF and APF.

References

1. Cefolio RJ (2005) Recent advances in the treatment of air leaks. Curr Opin Pulm Med 11:319–323
2. Wood DE, McKenna RJ, Yusen RD et al (2007) A multicenter trial of an intrabronchial valve for treatment of severe emphysema. J Thorac Cardiovasc Surg 133:65–73
3. Lois M, Noppen M (2005) Bronchopleural fistulas: an overview of the problem with special focus on endoscopic management. Chest 128:3955–3965
4. Mitchell KM, Boley TM, Hazelrigg SR (2006) Endobronchial valves for treatment of bronchopleural fistula. Ann Thorac Surg 81:1129–1131
5. Ricci ZJ, Haramati LB, Rosenbaum AT et al (2002) Role of computed tomography in guiding the management of peripheral bronchopleural fistula. J Thorac Imaging 17:214–218
6. Alifano M, Sepulveda S, Mulot A et al (2003) A new method for detection of post-pneumonectomy broncho-pleural fistulas. Ann Thorac Surg 75:1662–1664
7. Lois M, Noppen M (2005) Bronchopleural fistulas: an overview of the problem with special focus on endoscopic management. Chest 128(6):3955–3965
8. Dupuy DE, Zagoria RJ, Akerley W, Mayo-Smith WW, Kavanagh PV, Safran H (2000) Percutaneous radiofrequency ablation of malignancies in the lung. AJR Am J Roentgenol 174(1):57–59
9. Matsuoka T, Okuma T (2007) CT-guided radiofrequency ablation for lung cancer. Int J Clin Oncol 12:71–78
10. Sakurai J, Hiraki T, Mukai T et al (2007) Intractable pneumothorax due to bronchopleural fistula after radiofrequency ablation of lung tumors. J Vasc Interv Radiol 18:141–145
11. Clasen S, Kettenbach J, Kosan B et al (2009) Delayed development of pneumothorax after pulmonary radiofrequency ablation. Cardiovasc Intervent Radiol 32:484–490
12. Snell GI, Holsworth L, Fowler S et al (2005) Occlusion of a broncho-cutaneous fistula with endobronchial one-way valves. Ann Thorac Surg 80:1930–1932
13. Singh SS, Pyragius MD, Shah PJ et al (2007) Management of a large bronchopleural fistula using a tracheobronchial stent. Heart Lung Circ 16:57–59
14. Takaoka K, Inoue S, Ohira S (2002) Central bronchopleural fistulas closed by bronchoscopic injection of absolute ethanol. Chest 122:374–378
15. Onotera RT, Unruh HW (1988) Closure of post-pneumonectomy bronchopleural fistula with fibrin sealant (Tisseel). Thorax 43: 1015–1016
16. Ponn RB, D'Agostino RS, Stern H et al (1993) Treatment of peripheral bronchopleural fistulas with endobronchial occlusion coils. Ann Thorac Surg 56:1343–1347

17. Weinreb N, Riker D, Beamis J et al (2009) Ease of use of watanabe spigot for alveolopleural fistulas. J Bronchol Intervent Pulmonol 16:130–132
18. Feller-Kopman D, Bechara R, Garland R et al (2006) Use of removable endobronchial valve for the treatment of bronchopleural fistula. Chest 130:273–275
19. Anile M, Venuta F, De Giacomo T et al (2006) Treatment of persistent air leakage with endobronchial one-way valves. J Thorac Cardiovasc Surg 132:711–712
20. Travaline JM, McKenna RJ Jr, DeGiacomo T et al (2009) Treatment of persistent pulmonary air leaks using endobronchial valves. Chest 136(2):355–360
21. Fann JI, Berry GJ, Burdon TA (2006) The use of endobronchial valve device to eliminate air leak. Respir Med 100:1402–1406
22. Springmeyer SC, Bolliger CT, Waddell TK, Gonzalez X, Wood DE, IBV Valve Pilot Trials Research Teams (2009) Treatment of heterogeneous emphysema using the Spiration IBV Valves. Thorac Surg Clin 19(2):247–253