ECRONICON OPEN ACCESS

EC PULMONOLOGY AND RESPIRATORY MEDICINE

Case Report

A Novel Method of Treating Broncho-Pleuro-Cutaneous Fistula with an Endobronchial Valve

Prarthna Chandar Kulandaisamy[1], Michael Wert[1], Harsh Shah[2] and Alberto E Revelo[2]*

[1]Division of Pulmonary, Critical Care and Sleep Medicine, The Ohio State University Wexner Medical Center and James Comprehensive Cancer Center, Columbus, OH, USA

[2]Division of Pulmonary, Critical Care and Sleep Medicine, Interventional Pulmonology Section, The Ohio State University Wexner Medical Center and James Comprehensive Cancer Center, USA

***Corresponding Author:** Alberto E Revelo, Assistant Professor of Medicine, Division of Pulmonary, Critical Care and Sleep Medicine, Interventional Pulmonology Section, The Ohio State University Wexner Medical Center and James Comprehensive Cancer Center, Columbus, OH, USA.

Received: April 01, 2019; **Published:** August 26, 2019

Abstract

The management of prolonged air leaks represents a challenge for surgeons and pulmonologists. The morbidity and costs-due to prolonged hospital stays and extensive use of resources - associated with bronchopleural fistula (BPF) management is high. In the vast majority of cases, a surgical intervention is not an option because often patients are no longer adequate surgical candidates. If a surgical intervention is not feasible, bronchoscopic therapies should be considered. The use of one-way endobronchial valves has gained popularity in recent years, not only in the management of post-surgical, prolonged air leaks (PAL) but also in non-surgical ones. We present a unique case of an infected broncho-pleuro-cutaneous fistula following chemoradiation therapy for lung cancer; successfully managed with an endobronchial valve.

Keywords: *Endobronchial Valves; Persistent Air Leak; Pleuro-Cutaneous Fistula*

Abbreviations

APF: Alveolo-Pleural Fistula; EBV: Endobronchial Valve; BPF: Broncho-Pleural Fistula; BPCF: Broncho-Pleuro-Cutaneous Fistula; PAL: Persistent Air Leak; LUL: Left Upper Lobe

Introduction

An air leak is defined as the escape of air into the pleural space, generally through a defect in the lung parenchyma called alveolo-pleural fistula (APF) which is the most common form; or that, which happens at the sub-segmental airway level or more proximal, known as bronchopleural fistula (BPF) [1]. While these two entities have been described extensively in literature, pleuro cutaneous fistulas [2] -communication between pleural space with subcutaneous tissue- are rare and commonly reported as complications following radiation to the chest or secondary to chronic infections like tuberculosis. Persistent air leak (PAL) due to APF or BPF is that which happens beyond five days [1]. Surgical management is usually the initial approach for the management of such cases. In addition to surgical management, several forms of endoscopic therapies have been reported useful in the literature when surgery is not an option due to patient's poor candidacy. We report a rare case of broncho-pleuro-cutaneous fistula (BPCF), treated innovatively with endobronchial valves (EBV).

Case Report

The patient is a 62 year-old Caucasian male with severe COPD, and coronary artery disease and history of non-small cell lung cancer favoring squamous cell carcinoma, locally advanced of the left upper lobe (LUL) treated with definitive chemoradiation therapy a year prior to this presentation. He received a total radiation therapy of 7600 cGy to the primary tumor and mediastinal lymph nodes. A surveillance chest CT revealed subcutaneous air-fluid collection in the left subpectoral region. Transthoracic needle aspiration of the fluid within the chest wall showed only inflammatory changes. Pathology did not reveal any recurrence of malignancy. He received a course of oral antibiotics, however, despite treatment; he was hospitalized a week later for worsening left sided chest pain and fever. A new chest CT scan showed marked increase in the fluid collection with extensive subcutaneous emphysema and no pneumothorax (Figure 1A). Broad spectrum, intravenous antibiotics were started and he was planned for a CT- guided drain placement to decompress the subcutaneous emphysema and the fluid collection (Figure 1B). Purulent material was aspirated; however, the microbiological culture did not grow a specific organism. We hypothesized; the residual tumor cavity broke through the pleura, creating a fistulous tract directly into the subcutaneous tissue. Local inflammatory changes likely caused local pleurodesis of adjacent visceral and parietal pleura preventing formation of a pneumothorax. A high resolution CT scan with 1.0 mm cuts and airway reconstruction showed the apical-posterior segment of left upper lobe directly communicating with the cavity (Figure 1A). After multidisciplinary discussion at our thoracic tumor board, a decision was made to treat this endoscopically with a valve, as he was deemed not a good surgical candidate.

The placement of endobronchial valves for air leaks relies on direct visualization of air leak cessation in the water seal chamber of the collection system as one sequentially occludes each airway with a balloon during bronchoscopy. This helps with localization of an air leak, especially when is not obvious on imaging. Sometimes the air leak becomes challenging to localize it if air bubbles become absent at bronchoscopy or the air leak is intermittent. In this case, we mostly relied on detailed anatomical review of available chest imaging, as it was evident that the airway leading to the cavity was the apico-posterior segment of the left upper lobe (Figure 1A). A five mm Spiration® (Olympus Corp of the Americas, Redmond, WA) endobronchial valve was placed in the apico-posterior segment of the left upper lobe (Figure 1C). The air leak resolved and his clinical condition continued to improve. The Chest tube drain was removed the following day. He was discharged home on antibiotics, completing a total four week course. A repeat chest CT at six weeks showed almost complete resolution of subcutaneous emphysema and minimal subcutaneous fluid collection (Figures 1D). The patient continues to do well and is actively being followed at our clinic.

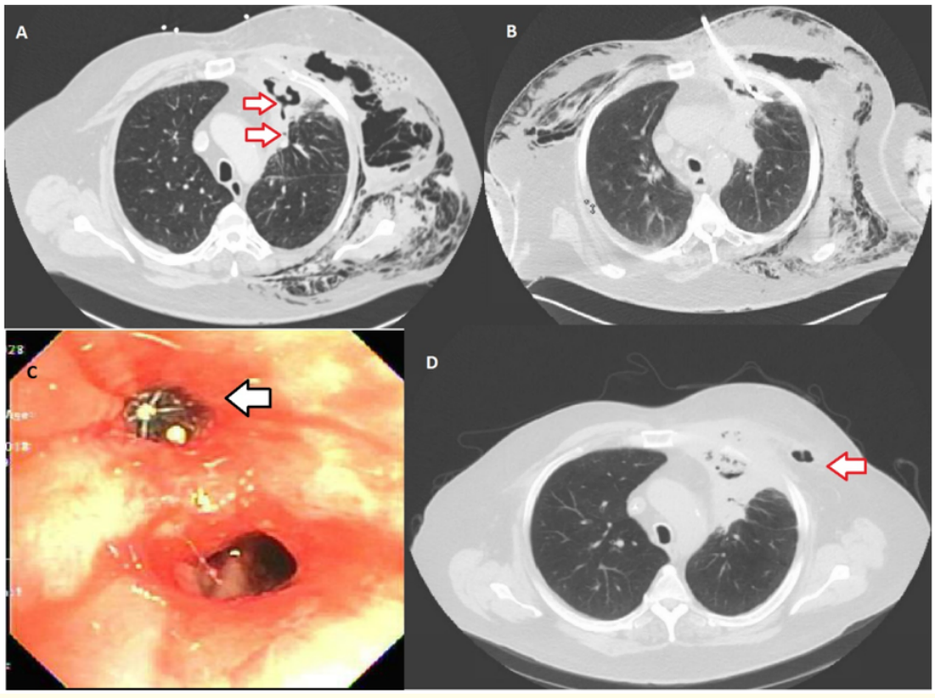

Figure 1: *1A: Axial view of a chest CT showing extensive subcutaneous emphysema and a cavitary lesion with air fluid level in the apical-posterior segment of the left upper lobe (arrows) 1B: A 14Fr chest tube placed in the cavity to drain the purulent material and air, under CT guidance. 1C: Bronchoscopic image showing the endobronchial valve (EBV) placed in satisfactory position at the apical-posterior segment of the left upper lobe. 1D: Repeat chest CT one month following placement of the EBV. Note significant improvement of the fluid collection and subcutaneous emphysema seen in previous images. A chest CT six months later showed near complete resolution of these findings.*

Discussion

Bronchopleural fistulas are more common after resectional thoracic surgeries [3,4]. They can also develop after chemoradiation therapy, or a delayed consequence of chronic lung infections like tuberculosis. Surgical repair is the standard of treatment. However, majority of these patients are not good surgical candidates and have poor cardio-pulmonary reserve [5]. In patients who cannot undergo surgery, bronchoscopic approach for temporary or definitive treatment must be considered. Some of the non-surgical options available are endoscopic fistula closure using endobronchial valves, biological glues, coils, stents, spigots, sealants and blood patch [1,6]. The Spiration Valve System is FDA-approved under a Humanitarian Device Exemption (HDE) for BPF occurring post-lobectomy, segmentectomy and lung volume reduction surgery. Many authors have demonstrated the utility of EBV placement and other anecdotal approaches for non-operative BPF closure. While EBVs are now frequently used in treatment for BPF, the placement depends on the presence of an air leak. The technique involves sequential balloon occlusion of various bronchial segments to localize the site of air leak. Sometimes, localization of an air leak cannot be accomplished by conventional methods and alternative ones have been described and should be considered [7,8]. Sometimes more than one segment is involved and deployment of multiple EBV may be required.

In our case, we were not able to apply this technique due to absence of air bubbles seen in the water collection system. Other methods to help localize air leaks have been summarized [1]. High-resolution chest CT with thin cuts is an invaluable tool in the assessment of persistent air leaks prior to bronchoscopy or surgical intervention.

Another important aspect of treating these patients is the concomitant treatment of any pleural space infection, if such exists, that could delay the healing of a fistula. The optimal duration of antibiotic therapy in these cases is not known although four to six weeks of therapy have traditionally been given, and a longer course may be necessary unless there is prompt resolution of symptoms. Fungal diseases may require longer period of therapy [9,10].

Conclusion

Our case provides an innovative, non-surgical treatment option for double fistulas with EBV's. It also illustrates how useful is a chest CT to help localize culprit airway defects. A thin-cut and high resolution scan should always be obtained prior to any therapy. Finally, despite the lack of established guidelines for endoscopic management of prolonged air leaks, there is a good amount of reports and retrospective studies showing successful treatment approaches; further supporting the use endoscopic, minimally invasive techniques. Adequate patient selection remains key to success.

Acknowledgement

None.

Conflict of Interest

No conflict of interest to disclose.

Bibliography

1. Keshishyan Sevak., *et al*. "Bronchoscopic management of prolonged air leak". *Journal of Thoracic Disease* 9.10 (2017): S1034-S1046.
2. Aiyappan Senthil Kumar., *et al*. "Double fistula: Bronchopleural and pleurocutaneous". *Lung India* 32.1 (2015): 88-89.
3. Darling GE. "Risk of a right pneumonectomy: role of bronchopleural fistula". *Annals of Thoracic Surgery* 79.2 (2005): 433-437.
4. DeCamp MM. "Patient and surgical factors influencing air leak after lung volume reduction surgery: lessons learned from the National Emphysema Treatment Trial". *Annals of Thoracic Surgery* 82.1 (2006): 197-206.
5. Dugan Karen C., *et al*. "Management of Persistent Air Leaks". *Chest* 152.2 (2017): 417-423.
6. Ding Mei., *et al*. "Endobronchial one-way valves for treatment of persistent air leaks: a systematic review". *Respiratory Research* 18.1 (2017): 186.

7. Revelo Alberto., *et al*. "Diagnostic and therapeutic challenges in managing persistent air leaks". *Journal of Thoracic Disease* 10.1 (2018): 522-528.

8. Mahajan AK and Khandhar SJ. "Bronchoscopic valves for prolonged air leak: current status and technique". *Journal of Thoracic Disease* 9.2 (2017): S110-S115.

9. Davies HE., *et al*. "Management of pleural infection in adults: British Thoracic Society pleural disease guideline 2010". *Thorax* 65.2 (2010): ii41-ii53.

10. Rosenstengel A. "Pleural infection: current diagnosis and management". *Journal of Thoracic Disease* 4.2 (2012): 186-193.

Volume 8 Issue 9 September 2019