CASE REPORT

Endobronchial valves in the management of bronchial fistulae caused by bronchopulmonary aspergillosis

Rhys Williams,[1] Rakesh Krishnadas,[2] Bipen Patel,[3] Nabil Jarad[1]

[1]Respiratory Medicine Department, University Hospitals Bristol NHS Foundation Trust, Bristol, UK
[2]Department of Thoracic Surgery, University Hospitals Bristol NHS Foundation Trust, Bristol, UK
[3]Respiratory Medicine Department, Royal Devon & Exeter NHS Foundation Trust, Exeter, UK

Correspondence to
Dr Rhys Williams,
rhys_williams_90@msn.com

Accepted 12 November 2015

SUMMARY

Following an aggressive episode of bronchopulmonary aspergillosis, a 54-year-old man developed a symptomatic air leak via a tunnel between the left upper lobe and an extra chest wall cavity. Following the failure of several surgical procedures to close the tunnel, endobronchial valves normally used in management of emphysema were used to successfully treat the air leak.

BACKGROUND

This case demonstrates the use of emphysema endobronchial valve placement outside its original indication: lung volume reduction in emphysema.

CASE PRESENTATION

A 54-year-old man, 10 years prior, developed aggressive bronchopulmonary aspergillosis in a left upper lobe cavity that originated as a complication of ankylosing spondylosis. Multiple attempts to close the post-aspergilloma cavity were made, including pedicled latissimus dorsi and pectoralis major flaps. A chronic large chest wall cavity remained, with anterior pleural fenestration and a bronchocavity fistula (figure 1). This resulted in a significant air leak causing breathlessness, impairment of phonation and recurrent bacterial infections. An endobronchial valve (EBV) provided good symptomatic relief before becoming displaced after 1 year (figure 2). An additional valve bestowed only short-term benefit. Further multistage surgery was deemed inappropriate due to a lack of potential muscle graft sites, and the risk of pulmonary sepsis posed by an open airway leak.

A large EBV was contemplated to close the tunnel between the lungs and the chest wall cavity.

INVESTIGATIONS

CT imaging illustrated a tennis ball-sized cavity in the left upper lobe, with a bronchocavity fistula (figure 3).

TREATMENT

Fiberoptic bronchoscopy was performed under usual sedation. This showed an air leak around the previous valve placed in the apical segment of the left upper lobe (figure 2). There were also several areas of granulation tissue. Previously unseen accessory bronchi appeared to have formed in the same lobe.

Figure 1 The chest wall cavity before (left) and after (right) valves were inserted. The sealing membrane tests revealed no evidence of air leak.

To cite: Williams R, Krishnadas R, Patel B, *et al*. *BMJ Case Rep* Published online: [*please include* Day Month Year] doi:10.1136/bcr-2015-211955

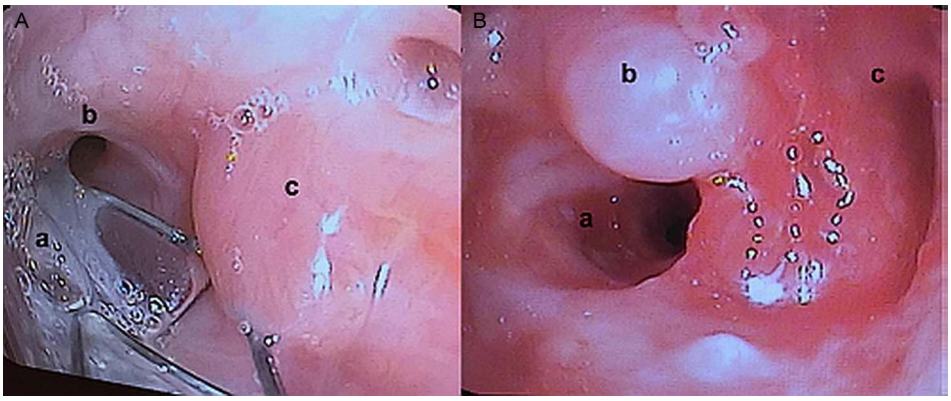

Figure 2 Two bronchoscopic appearances. (A) The old valve (a) is visible in the apical segment of the left upper lobe bronchus. An air leak (b) is seen and an area of granulation tissue is also visible (c). (B) The old valve was removed revealing a tunnel between the left upper lobe and the chest wall (a); the granulation tissue (b) and a new accessory bronchus (c).

The older valve was removed with ordinary forceps via fiberoptic bronchoscopy. Two new valves were inserted in the two accessory bronchi. The most challenging stage of the procedure was closing the tunnel surrounding the older valve. An initial attempt led to the valve dislodging distally and having to be retrieved manually from the chest wall cavity, in an unprecedented turn of events.

A second valve of the largest size available was chosen so that the stronger stenting mechanism would provide greater stability and prevent future displacement. Following insertion, an airtight dressing was used to confirm that the three valves had successfully closed the leak (figure 1).

OUTCOME AND FOLLOW-UP

Plain imaging following the procedure indicated good overall volume reduction in the left lung, as manifested by elevation of the left hemi-diaphragm (figure 3). The exposed chest cavity was packed with ribbon postoperatively, to promote granulation over exposed airways distal to the valve. Unlike in emphysema patients, postprocedural lung function tests were not an outcome measure relevant to the ultimate aim of valve placement, namely plugging the fistula. The patient has, however, benefitted from improved exercise tolerance since the procedure, and is able to phonate (speak) normally.

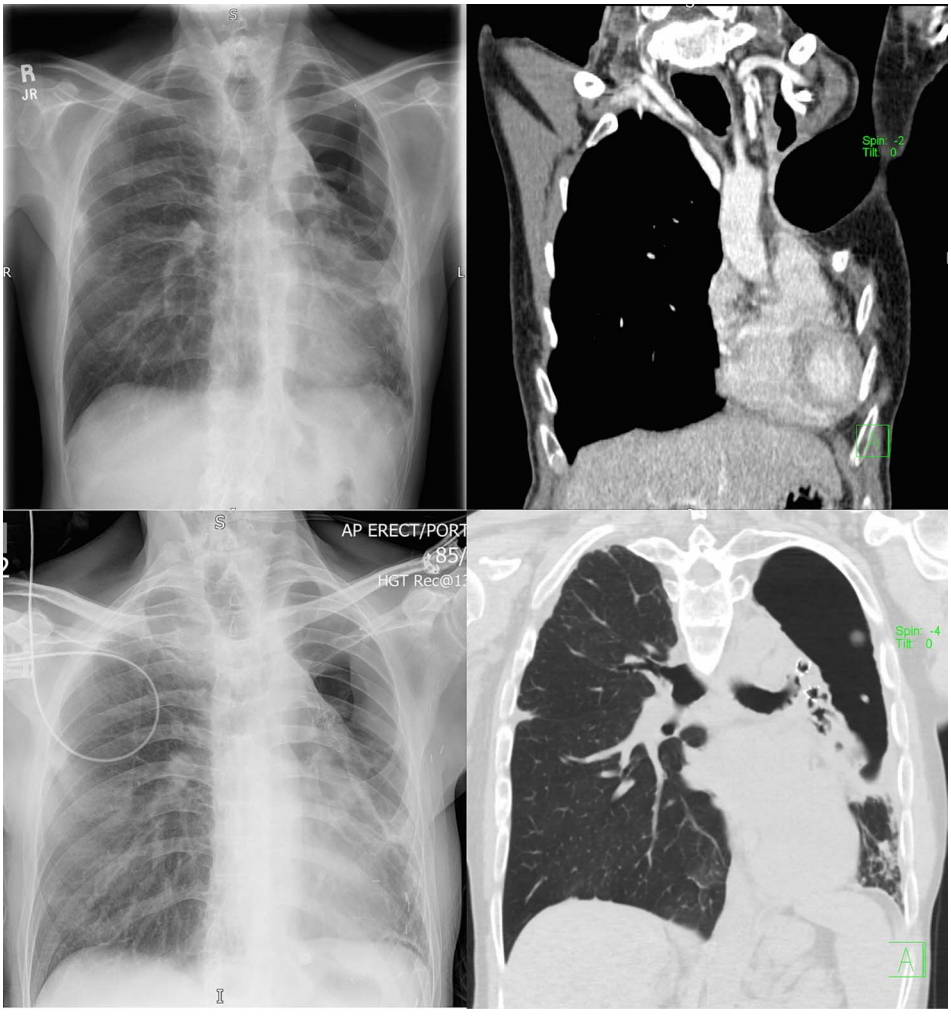

Figure 3 Upper panel: A chest X-ray and coronal CT imaging before valve insertion. The lower panel shows images following the insertion of valves. Note the elevation of the left hemi-diaphragm, demonstrating the intended loss of volume as a result of valve insertion.

DISCUSSION

EBVs are self-expanding one-way valves that can be placed in segmental or lobar bronchi via bronchoscopy to create unidirectional flow of air and secretions in aid of expiration only. The main purpose of these valves is volume reduction in patients with emphysema.

EBVs reduce dynamic hyperinflation, facilitating ventilation in healthier areas of lung and improving chest cavity mechanics. EBV insertion achieves the best results in patients with heterogeneous emphysema with an absence of cross-ventilation between the target lobe and the adjacent lobe. Marked improvements in forced expiratory volume in 1 s, St George's Respiratory Questionnaire score and 6-min walk distance have been illustrated when atelectasis has occurred in the target lobe after valve insertion.[1 2]

Valves are also used in the management of pneumothorax-inducing bronchopleural fistula in patients who are unable to tolerate surgical interventions.[3] The procedure can cause instant occlusion of the lobe in which the fistula is present. In addition to providing immediate improvement in lung ventilation, it is thought that cessation of airflow through fistulae may benefit healing and closure. Evidence of efficacy in this application is illustrated in a recent case series of 40 patients where EBV insertion achieved complete cessation of leakage in 48% of patients.[1] We predict that, as seen in therapeutic atelectasis in emphysema, outcome is likely to be better in patients with an absence of cross-ventilation.

Collapsing target lobes may also result in occlusion of cavities. This was found to be useful in cavities induced by multidrug-resistant tuberculosis. Collapse of tuberculous cavities is thought to facilitate disease regression by starving the aerophilic mycobacteria of oxygen, preventing bacterial seeding and aiding cavity closure.[4]

To the best of our knowledge, this is the first case in which features of EBVs have been applied to close a tunnel induced by *Aspergillus* infection.

Patient's perspective

- Despite the fact that I had significant breathing difficulties, my local health authority was offering no further treatment because of the complexity of my case. I had to challenge the extraordinary treatments panel in order to be referred elsewhere. This was a very stressful and unpleasant experience leaving me at times feeling isolated and rejected.
- I have had the insertion of endobronchial valve procedure carried out a few times now and each time it has been successful in blocking the targeted airways. However, my chest wound is still changing and new air leaks have subsequently tended to appear. I hope that by blocking the relevant airways and promoting my own healing we will be able to stop the leaks entirely.
- Before the last procedure, I was venting so much air through my chest wound that talking for more than a few minutes made me breathless. I could not read a complete story to my granddaughter or hold a telephone conversation. Immediately following the procedure, I could feel the benefit and talk without difficulty. I can now walk long distances instead of pausing for rest every 200 m and I can even ride my push bike once again (although not uphill!).
- The endobronchial valve procedure is the least invasive and simplest of the surgical procedures that I have experienced. The benefits are immediate and although I am very grateful for this I still resent the fact that I had to fight my local health authority for this treatment.

Learning points

- Endobronchial valves originally designed to cause lung volume reduction in emphysema can be used to treat persistent air leaks not amenable to surgical intervention.
- Misplaced valves may be removed distally under direct manual manipulation or by using a bronchoscope inserted via the distal cavity.
- The procedure is minimally invasive and usually well-tolerated.

Contributors BP was responsible for the referral to undergo the procedure. NJ and RK carried out the procedure, with RW observing. RW researched the literature and wrote the case report. All the authors approved the final manuscript. NJ advised on the formation of the manuscript and provided the images and the figures.

Competing interests None declared.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

REFERENCES

1 Herth FJ, Noppen M, Valipour A. Efficacy predictors of lung volume reduction with Zephyr valves in a European Cohort. *Eur Respir J* 2012;39:1334–42.
2 Shah P, Herth FJF. Current status of bronchoscopic lung volume reduction with endobronchial valves. *Thorax* 2014;69:280–6.
3 Travaline JM, McKenna RJ, De Giacomo T, *et al*. Treatment of persistent pulmonary air leaks using endo-bronchial valves. *Chest* 2009;136:355–60.
4 Corbetta L, Montinaro F, Rogasi PG, *et al*. Improvement in tubercular cavities following adjuvant treatment with endobronchial valves: a case report. *Int J Tuberc Lung Dis* 2013;17:850–1.