CASE REPORT

Persistent air leak after pulmonary transplantation

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Accepted 28 June 2017

SUMMARY

A 59-year-old man with bilateral apical emphysema underwent a double lung transplant for end-stage chronic obstructive pulmonary disease leaving remnant right apical native tissue due to pleural adhesions. Initial postoperative course was uneventful until the chest drains were removed. This revealed a small pneumomediastinum, which progressively increased in size causing gross surgical emphysema. Re-insertion of the chest drain stabilised the patient so that the cause could be identified and corrected. Two bronchoscopies excluded anastomotic dehiscence as a cause. Therefore the subcostal wound was refashioned under video-assisted thoracoscopic surgery in case there was a defect. Unfortunately this also failed to halt the air leak; therefore another cause was sought. A multidisciplinary team meeting review of the radiology revealed that the patient's native bullous tissue was still inflated. Subsequent bronchoscopy revealed a native bronchial communication, due to variant anatomy, proximal to the surgical anastomosis. This was subsequently occluded using a bronchial valve allowing the patient to make a swift recovery.

BACKGROUND

This case demonstrates the clinical importance of aberrant anatomy and how it can affect a patient's health. It also demonstrates that novel use of an endobronchial valve can prevent invasive and complicated surgery.

CASE PRESENTATION

A 59-year-old man underwent a double lung transplant. His pretransplant CT scan showed significant bilateral apical emphysema, consistent with a diagnosis of chronic obstructive pulmonary disease. A bilateral lung transplant operation was performed through a standard clamshell incision without complications: the lungs were removed using an Echelon stapler to divide vessels, followed by incision of the respective main bronchi. Significant pleural adhesions caused small islands of native lung to be left behind following parenchymal stapling, as attempting to remove these would significantly increase the bleeding risk. Donor lungs were then transplanted in a sequential manner and the patient was transferred uneventfully to the cardiothoracic intensive care unit.

The patient made a good initial postoperative recovery. Since there was minimal chest tube drainage, the chest drains were removed on the fourth and fifth days after transplantation. After removal of the right apical chest drain, the chest

radiograph revealed minimal surgical emphysema and a small pneumomediastinum. Over the next 2 weeks this surgical emphysema got significantly worse, suggesting a persistent air leak.

INVESTIGATIONS

The most likely cause was anastomotic dehiscence, therefore two separate bronchoscopies were performed, each confirming that the anastomoses were intact. Since the aetiology of the surgical emphysema remained unclear a right apical chest drain was re-inserted as a temporary measure. Subcostal leakage of air from the surgical wound could be another cause of the surgical emphysema, therefore the wound was examined and refashioned under video-assisted thoracoscopic surgery (VATS); however this failed to stop the air leak necessitating further investigation. Review of the case at the local multidisciplinary team meeting revealed that the patient's native bullous tissue in the right apex was still inflated (figure 1A) despite there being no obvious endobronchial communication.

DIFFERENTIAL DIAGNOSIS

Persistent surgical emphysema occurs when there is direct communication between the lung and subcutaneous space. In the transplant setting this often occurs due to dehiscence at the bronchial anastomosis creating a bronchopleural fistula.¹ In our patient however repeated bronchoscopy and VATS excluded this diagnosis. Less common causes for persistent air leak include focal defects of the surgical wound in the chest wall or aberrant placement of a chest drain.

In our case the CT scan showed the patient's native bullae were inflated (figure 1A), despite the right bronchus being connected to the allograft. This is surprising as the surgeon only staples the right main bronchus proximal to any division removing any communication to the native lung. Subsequent flexible bronchoscopy revealed a barely perceptible aberrant bronchus communicating between the trachea and the patient's native lung (figure 2).

TREATMENT

Resection of the native lung would be risky due to the extensive pleural disease therefore an endobronchial solution was attempted. The accessory bronchus was occluded with a Chartis balloon causing the air leak to immediately cease; this demonstrated that this was the only communication. A one-way endobronchial Zephyr valve was therefore placed to occlude the accessory bronchus (figure 3) causing the native remnant to collapse and the air leak to stop.²

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To cite: Pearmain L, Krysiak P, Blaikley J, et al. *BMJ Case Rep* Published Online First: [please include Day Month Year]. doi:10.1136/bcr-2017-220176

Figure 1 CT scan of the thorax showing an air leak around the right lung (A), which resolved (B) after an endobronchial intervention.

OUTCOME AND FOLLOW-UP

After the procedure the chest drains were removed and the patient was subsequently discharged without any reoccurrence of the surgical emphysema. A 2-month follow-up CT scan confirmed complete atelectasis of the native tissue (figure 1B).

DISCUSSION

Aberrant bronchial anatomy occurs in roughly 2% of the population, with the right upper lobe being the most common region of the bronchial tree to exhibit congenital abnormalities, as with our patient.^{3 4} The nomenclature of aberrant anatomy has some variation, but the term ‘tracheal bronchus’ may include any anomalous bronchial connection originating from the trachea, or in some sources also the main bronchus.³ An accessory bronchus is any ‘tracheal bronchus’ that originates from aforementioned locations directed towards the upper lobe; if it is directed towards the pericardium then this is known as an accessory cardiac bronchus. If the accessory bronchus is in addition to normal bronchial anatomy then it is termed supernumerary. The clinical significance of accessory bronchi varies from being completely asymptomatic, to those revealed as causing pathology only when involved in procedures or surgery (as in this case), to patients presenting with recurrent pneumonias or other regional pathologies which is much more common in paediatric patients than adults.^{3 4}

Figure 2 Endoscopic image showing a right accessory bronchus (A) coming off proximally to the anastomotic right main bronchial anastomosis (B).

Figure 3 Endoscopic image demonstrating Zephyr endobronchial valve in the accessory bronchus (A) with the right main bronchus (B) seen at the bottom of the image.

The management of persistent postoperative air leaks depends on the underlying cause. Endobronchial valves may be effective in treating persistent air leaks,⁵ with up to 29 reported cases so far. We believe however this is the first reported case where it has been used in an accessory native bronchus not recognised at the time of double lung transplantation, although it has been used to occlude the native left upper lobe of a patient with lymphangiomatosis after a right single-transplant.^{5 6} In selected cases this can be superior to the traditional approach of removing the native lung or surgical decortication/pleurodesis, avoiding the postoperative risks of bleeding and infection. Certainly our patient was discharged home within a week and had no complications from the endobronchial valve.

Endobronchial valves were originally developed for the purpose of lung volume reduction of areas of severe emphysema in patients with heterogeneous emphysema.⁷ The rationale was the functional and symptomatic improvement that emphysematous patients derive without having complex lung reduction surgery. Results from trials have clearly shown that endobronchial valves are an effective minimally invasive option in this group of patients. As demonstrated in this case report endobronchial valves have recently been used in

Learning points

- Knowledge and understanding of thoracic anatomy, including aberrancy, can be key in diagnosing unusual clinical presentations.
- Appreciation of underlying anatomy and pathology guides management and helps provide individualised solutions to patients with intrathoracic air leaks.
- Common causes of persistent postoperative air leaks are a wrongly placed chest drain and surgical wound dehiscence.

various cases where a minimally invasive option is required for treatment of broncho-pleural communications; this is still an emerging area but appears to be an effective treatment option.⁵

Contributors MA, JB, LP conceived the case report. PK and MA designed the intervention. All authors drafted and approved the manuscript before publication.

Competing interests None declared.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

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