A 75-year-old woman with severe chronic obstructive pulmonary disease (COPD) and advanced emphysema (ex-smoker, 70 pack-years) was referred to our clinic for endoscopic lung volume reduction. She suffered from increasing dyspnea on exertion (6-minute-walk distance 240 m) despite optimal medical treatment including tiotropium 18 µg qd, salmeterol-fluticasone 50/500 µg bid and long-term oxygen therapy. Pulmonary function tests showed a post-bronchodilator forced expiratory volume in 1s of 0.67 l (32% of predicted) and residual volume of 5.4 l (245% of predicted). Computer tomography (CT) scan of the thorax confirmed heterogeneous emphysema with predominant manifestation in the upper lobes (*Fig. 1a*); left upper lobe was confirmed as the primary target lobe by a perfusion scan showing low perfusion (8%) in this lobe.

After bronchoscopic exclusion of collateral ventilation (Chartis system, Pulmonx Inc., Redwood City, CA, USA), 4 Zephyr endobronchial valves (EBV) (Pulmonx Inc., Redwood City, CA, USA) were placed in the left upper lobe/lingula through a flexible bronchoscope in order to suppress upper lobe ventilation and ultimately to improve lung mechanics, exercise performance and quality of life [1–3]. The procedure including assessment of collateral ventilation and EBV placement lasted 20 min and was uneventful.

However, 24 h post-procedure the patient suffered from a symptomatic pneumothorax which was treated successfully by chest tube insertion. 4 days later a chest x-ray suggested relapsing pneumothorax, and 5 days later, CT showed atelectasis of the left upper lobe (the objective of EBV treatment), the chest tube in situ and, surprisingly, a huge pneumatocele in the lingula (*Fig. 1b*). Pneumatoceles are acquired, thin-walled, air-containing, intraparenchymal lesions of the lungs. In the present case, the localization of the pneumatocele is along the course of the fissure. Therefore, an interlobar pneumothorax could be another differential diagnosis, however, this would be characterized by a more oblong form and by intrafissural restriction due to previous pleural adhesions as a result of pleuodesis or pleural diseases – that are not known in the present case.

In accordance with the standard treatment of prolonged pneumothorax induced by EBV treatment, we removed 1 of the 4 valves one week after the primary procedure. However, the pneumatocele persisted without any significant change in size, while the atelectasis disappeared as expected (*Fig. 1c*). Two of the remaining 3 valves were directly adjacent to the wall of the pneumatocele raising the possibility of bronchial obstruction by the EBV and thereby allowing one-way passage of air into the interstitial space. These 2 valves were therefore removed in a 3rd bronchoscopy whilst
the 4th EBV was left in place; during the same anesthesia, the chest tube was removed. Within the next 14 days, the pneumatocele significantly decreased in size without further invasive treatment (Fig. 1 d–f), and the patient could leave hospital. After 2 months, the patient presented in improved condition (6-minute-walk distance 320 m; FEV1 1.0 l, 45 % of predicted; RV 4.9 l, 217 % of predicted), and the pneumatocele was resolved. Replacement of the removed EBV is not planned.

Clinical studies on 411 individuals revealed that complications of EBV treatment include pneumothorax, COPD exacerbation, pneumonia, hemoptysis and valve migration [1–3]. Whereas pneumothorax, usually appearing within 24 h ipsilateral to the non-treated lobe, is an expected event after EBV therapy in approximately 5–10 % of cases, this is the first documented pneumatocele after EBV therapy. The exact pathogenesis of pulmonary pneumatoceles, commonly seen as a sequela of staphylococcal pneumonia in infants and young children [4] or after blunt pulmonary trauma, is uncertain [5]. In the present case, one may suggest a transient bronchial obstruction by the EBV as a check-valve type. During inspiration, the obstruction may be partly relieved, allowing air to enter the interstitial space but not to leave it. The development of pneumatocele 3–5 days after EBV and pneumothorax favours the possibility that the combined scenario EBV plus distorted anatomy by the pneumothorax (and its resolution) causes the intraluminal obstruction with check-valve formation. Even after removal of 3 of the 4 EBV the pneumatocele persisted suggesting that the subsequently enlarged pneumatocele itself perpetuated the local bronchial obstruction.

Pneumatoceles should be considered and evaluated by CT in patients with relapsing or increasing dyspnea especially a few days after EBV treatment, when the patient normally has left hospital, or after an EBV-related pneumothorax has been treated. In the majority of cases, pneumatoceles resolve spontaneously within few weeks without invasive approaches [4, 5]; invasive procedures should be evaluated only in case of complications such as tension pneumatocele or secondary infection.

Conflict of Interest

The authors have no conflict of interest.

References