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Pneumatocele triggered by continuous positive airway pressure after lung resection

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A B S T R A C T

A pneumatocele is a cystic change of the lung that is caused by a check valve in the bronchiole due to infection, trauma and positive-pressure ventilation. We herein report a case of pneumatocele triggered by using of continuous positive airway pressure (CPAP) for sleep apnea syndrome (SAS) after pulmonary resection. A 69-year-old man underwent right upper lobectomy for lung cancer and developed interstitial pneumonia (IP) 10th postoperative day (POD). He was treated with steroid pulse therapy (solmedrol 500 mg × 3 days), and thereafter with oral steroid therapy (predonin 30mg/day). Well responded to the steroid therapy, IP was improved. However, he noticed bloody sputum 29th POD, and chest computed tomography showed a giant cystic lesion on the dorsal right lower lobe. We resected the cyst and the pathological findings revealed that the cystic lesion was pneumatocele, and CPAP was strongly suspected of triggering this disease.

1. Introduction

A pneumatocele is a cystic change that occurs in the lung and is caused by damage to the bronchiole due to inflammation, infection, trauma and positive-pressure ventilation. It causes air to be stored in the lung parenchyma by a mechanism involving the check valve [1]. Most cases disappear spontaneously after resolution of the causative event and are usually treated conservatively [2]. It is rare for a pneumatocele to develop after pulmonary resection [3]. We herein report a case of pneumatocele triggered by the use of continuous positive airway pressure (CPAP) for sleep apnea syndrome (SAS) after pulmonary resection.

2. Case report

A 69-year-old man was referred to our hospital with abnormal chest shadow. He had a history of asthma and sleep apnea syndrome (SAS), and he used CPAP for the treatment of SAS. He had smoked 10 cigarettes per day for 16 years.

No abnormality was found in the respiratory function test. Chest computed tomography (CT) revealed a 14-mm tumor in the right upper lobe with no lymph node swelling. Positron emission tomography (PET)-CT showed a strong fluorodeoxyglucose (FDG) accumulation in the tumor (SUV_{max} 9.2). We performed video-assisted thoracoscopic right upper lobectomy with suspicion of lung cancer (cT1bN0M0 cStageIA2). The pathological diagnosis was pT1bN2M0, stage IIIA and adenocarcinoma. Lymph node metastasis was found in #2R, #4R, #11S, and #12. Interstitial pneumonia developed on the 10th postoperative day (POD). He was treated with steroid pulse therapy (solmedrol 500 mg × 3 days) and thereafter treated with oral steroid (predonin 30 mg/day). The infiltrative shadow improved, and he was discharged on the 22nd POD.

One week after discharge (29th POD), he noticed bloody sputum and visited the hospital. Chest X-ray showed abnormal fluid retention in the right thoracic cavity (Fig. 1a). Chest CT showed a cystic lesion on the dorsal right lower lobe (segment 10) (Fig. 1b). As the air pressure due to CPAP was considered to be the trigger of the cystic lesion, we discontinued the use of CPAP after monitoring to ensure that the SpO_2 did not drop below 90% while sleeping. After placement of a chest drain in the thoracic cavity, we inserted a pigtail catheter in the cystic lesion (Fig. 1c). After these treatments, the cystic lesion shrinked a little, but pneumothorax appeared (Fig. 1d). Endobronchial Watanabe spigot was performed to shut off the air flow into the cystic lesion, but it did not shrink. We therefore performed video-assisted thoracoscopic surgery for the cystic lesion 42 days after the first operation. During the operation, we found a cystic lung lesion, the wall of which was very thick, on the dorsal right lower lobe (Fig. 2a). We resected the wall of the cyst and found an exposed bronchiole in the cyst in segment 10 of right lower lobe. We closed it by suturing with fascia obtained from intercostal muscle (Fig. 2b). The postoperative course was uneventful, and he was discharged on the 9th POD after the second operation.

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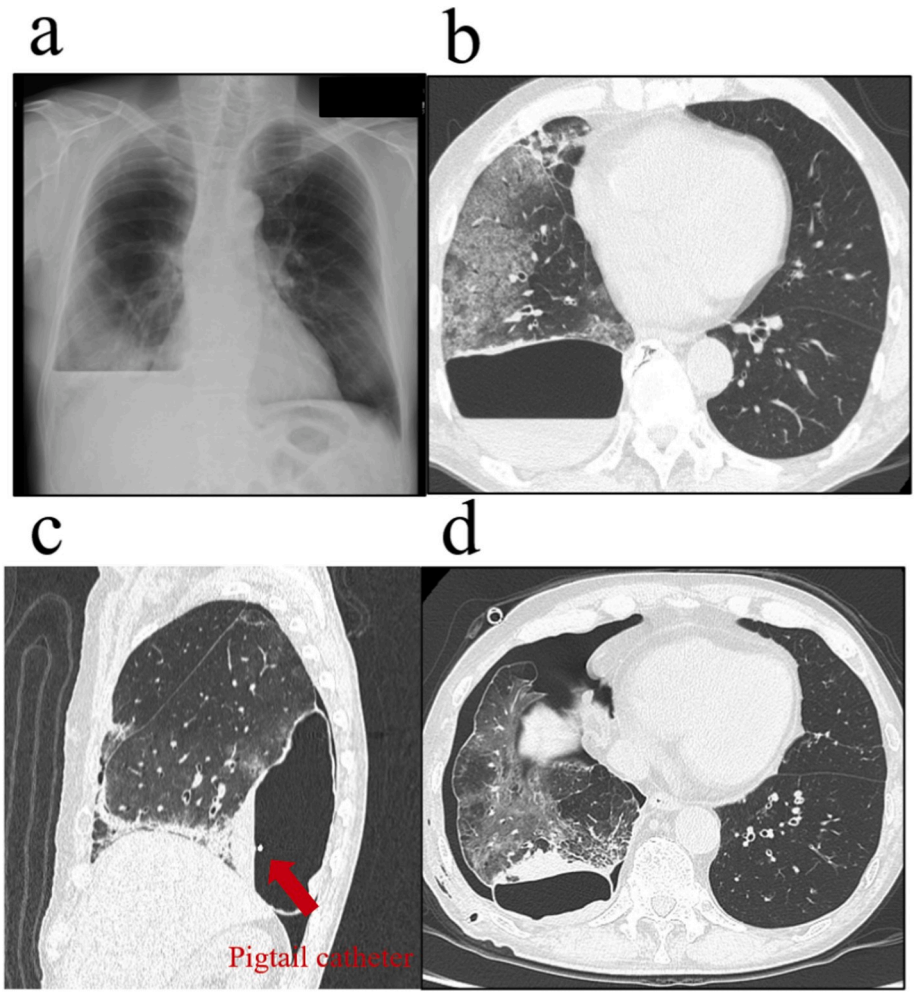

Fig. 1. a) b) Chest X-ray and CT show the cystic lesion was found on the dorsal right lower lobe. c) Pigtail catheter was inserted in the cystic lesion. d) After pigtail catheter insertion, pneumothorax was detected on CT.

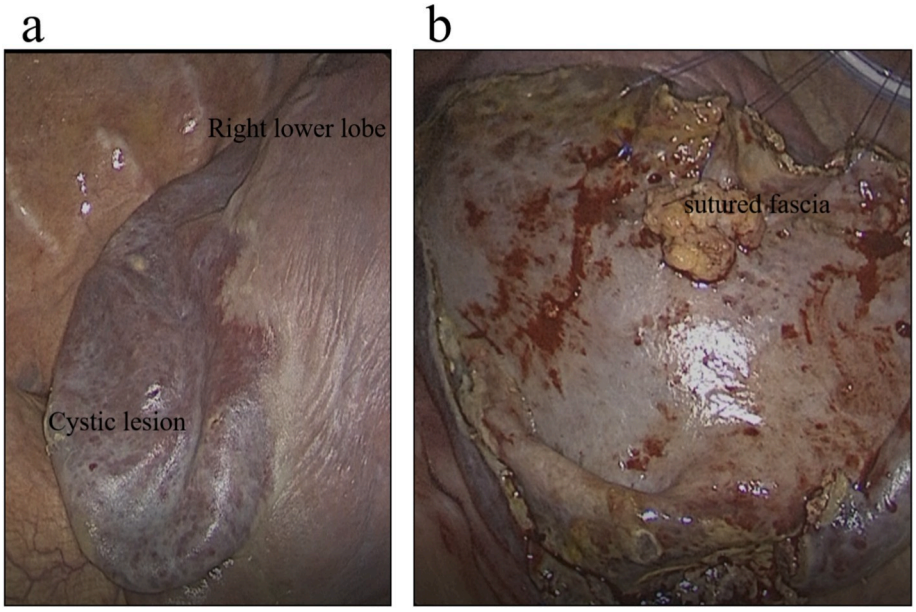

Fig. 2. a) A cystic lung lesion with hemorrhaging was found during the operative. b) We excised the cystic wall and removed the hematoma.

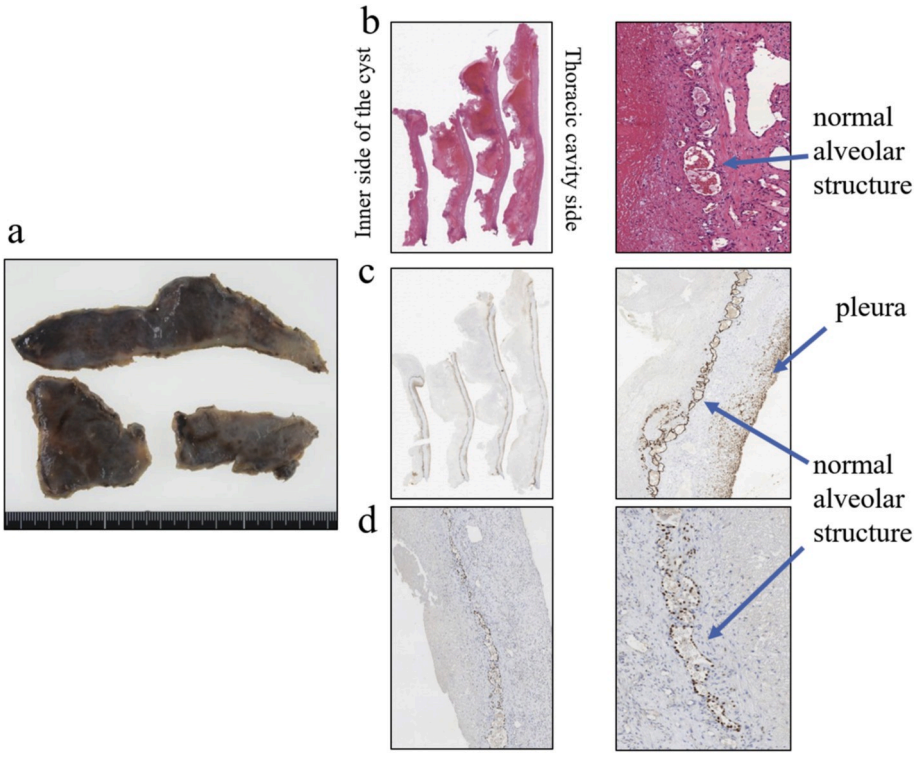

Fig. 3. a) Macroscopic findings of the resected cyst. b) HE staining (left × 40, right × 100), c) AE1/AE3 staining (left × 40, right × 100), and d) TFF-1 staining (left × 100, right × 400) of the resected wall of the cyst.

A pathological examination showed that the wall of the cyst was thickened (Fig. 3a), and it was mainly composed of visceral pleura and a normal lung parenchymal layer (Fig. 3b), which was confirmed by an immunohistochemical analysis of epithelial marker AE1/AE3 (Fig. 3c) and lung epithelial marker TTF-1 (Fig. 3d). Remnants of hematoma and fibrin were found in the cystic cavity. These findings showed that the cystic lung lesion was a pneumatocele that had developed due to mechanical air pressure generated at the inner side of the alveola, and CPAP was strongly suspected as the cause of this disease.

3. Discussion

A pneumatocele is a cystic lung lesion generated by the check-valve mechanism that can cause bloody sputum and infection. A pneumatocele caused by nasal CPAP has been reported in children [5], and invasive positive pressure ventilation has been reported as the cause in adults [4], but CPAP used in the treatment of SAS as the cause is not reported in adults.

In the present case, positive-pressure ventilation with CPAP was considered to have been the trigger of pneumatocele, as the lung tissue becomes fragile after interstitial pneumonia and steroid use. CPAP is an effective treatment for SAS and is commonly used worldwide. CPAP opens the obstructed airway by applying pressure to it. The pressure required for treatment is determined by various factors, such as the sleep pattern as evaluated by polysomnography, but a pressure of at least 4–5 cmH_2O and at most 25–30 cmH_2O airway pressure is required. An invasive ventilator is often controlled to roughly 30 cmH_2O airway pressure. Although there have been no cases of pneumatocele with CPAP reported in adults, several cases of pneumatocele due to invasive ventilatory management have been reported [4]. Therefore, we presume that CPAP triggered the generation of pneumatocele.

A pneumatocele often disappears spontaneously and is typically treated conservatively [1,2]. Conservative treatments include only follow-up, the administration of antibiotics to prevent infection caused by atelectasis and necrosis [4] and CT-guided catheter drainage of the cyst to remove air and hematoma [6]. Although surgical resection has also been reported in severe case [3,7], there are few cases that require surgical resection, and it is extremely rare for a pneumatocele to occur after lung resection [3]. In the present case, we performed surgical resection for pneumatocele because conservative treatments were not effective. During the operation, we found an exposed bronchiole inner side of the pneumatocele, which might have made conservative treatment difficult.

4. Conclusion

We experienced a rare case of pneumatocele triggered by the use of CPAP during steroid therapy for interstitial pneumonia after pulmonary resection. Must be careful when using CPAP for patients with interstitial pneumonia and steroid use.

Declaration of competing interest

The authors have declared no conflict of interest.

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