

Marsupialisation of a pneumatocele: a novel intervention for a rare post-COVID-19 sequelae

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Introduction

Pneumatoceles, also referred to as pseudo-cysts, are thin-walled air-filled cysts that form within the lung interstitium. They are generally post-infectious complications, and are often reported in children and less commonly in adults. A pneumatocele developing following COVID-19 may go undetected if it is asymptomatic. The exact mechanism of formation of spontaneous pneumatoceles in the absence of positive pressure ventilation and barotrauma

Case report

A 43-year-old man, who was a non-smoker with no known comorbidities, presented to the hospital with progressive breathlessness on routine activities of 2 months duration. He had been hospitalised for 10 days for moderate COVID-19 pneumonia 4 months earlier. He had been treated with steroids, remdesivir and low flow oxygen through nasal prongs at 2 litres per minute. His condition improved and he was discharged, but 2 months after discharge, he developed worsening breathlessness and was re-admitted to a tertiary centre. A high resolution computed tomography scan of the chest revealed a pneumatocele in the right lower lobe of the right lung, along with minimal pleural effusion and post-COVID changes. He was managed conservatively until he was stabilised and was discharged on domiciliary oxygen, which he used for over 2 months for around 18 hours per day. Re-evaluation with high resolution computed tomography showed persistence of the right lower lobe pneumatocele (**Figure 1**). Respiratory examination revealed absent breath sounds in the right lower lobe with a hyper-resonant percussion note. Inflammatory markers and white blood cell count were within normal limits.

Owing to the non-resolution of the pneumatocele after 2 months of observation, an opinion was sought from cardiothoracic surgeons and the patient was offered surgery. Excision of the pneumatocele was planned. As the wall of the lesion could be dissected and was discrete from the bronchus, the pneumatocele was marsupialised by partly running a prolene suture all around the cut edge of the cyst wall, almost up to the functioning parenchyma (**Figure 2**). Microscopic examination of the cyst wall showed fibrocollagenous tissue without epithelial lining. It revealed mixed inflammatory infiltrate with numerous congested blood vessels, and foci of air spaces lined by pneumocytes surrounded by inflammatory cells (**Figures 3a and b**).

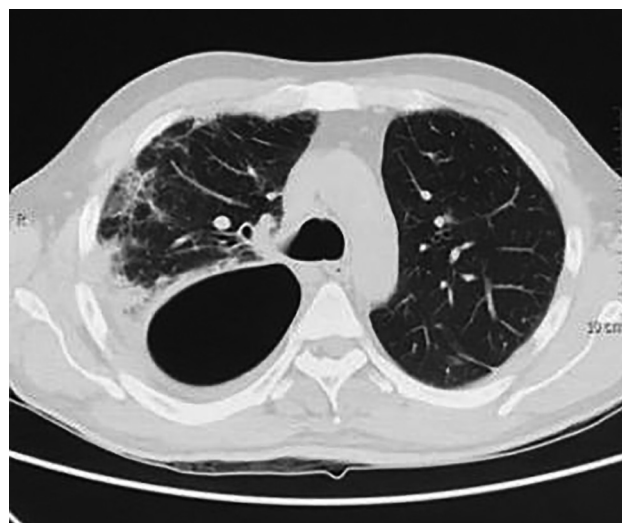


Figure 1. High resolution computed tomography scan of the chest showing a 12 x 8.6 cm pneumatocele in the lower lobe of the left lung.

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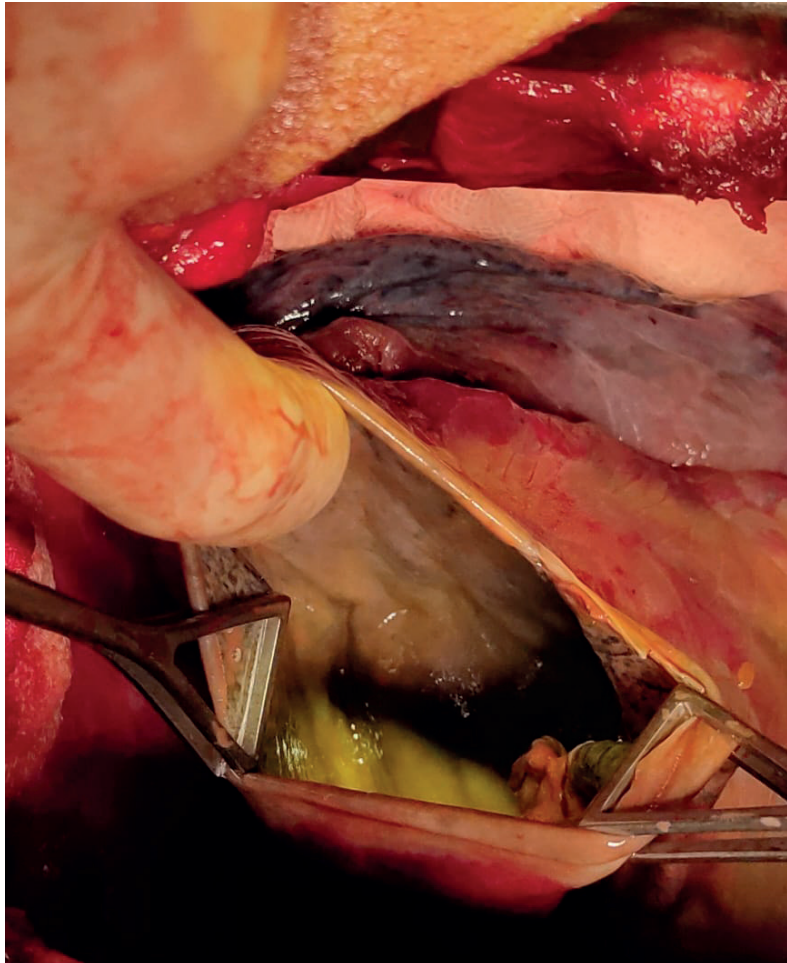


Figure 2. Intraoperative image of pneumatocele excision.

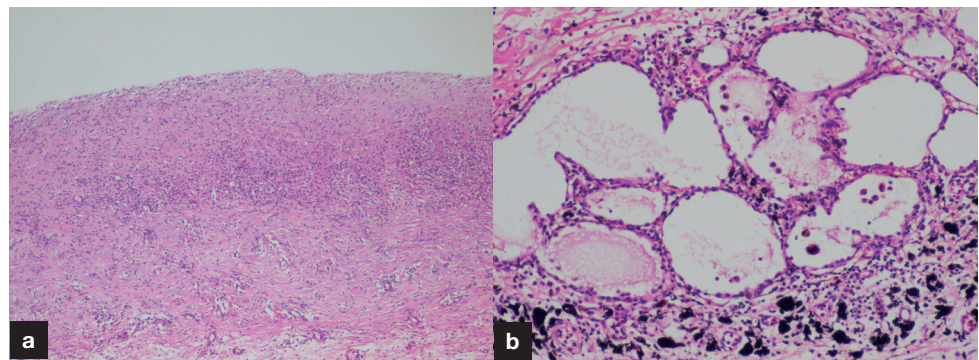


Figure 3. a. Pseudocyst wall with fibrocollagenous tissue and inflammatory cells (4X, haematoxylin and eosin). b. Air spaces lined by pneumocytes and surrounded by inflammatory cells (10X, haematoxylin and eosin).

remains largely unknown. This article reports a rare presentation of a pneumatocele that developed following COVID-19 pneumonia, in the absence of positive pressure ventilation, and was diagnosed only in the post-recovery phase of the disease. It was a huge cyst that required surgical intervention.

Discussion

Pneumatocèles are thin-walled air-filled cystic lesions that develop within the lung parenchyma. They often occur as a complication of pneumonia, commonly caused by *Staphylococcus aureus*, *Streptococcus pneumoniae* or *Haemophilus influenzae*.

Learning points

- Patients with COVID-19 pneumonia and those who have recovered from COVID-19 should be carefully monitored for complications such as pneumatoceles, as these can lead to sudden onset of pneumothorax with fatal results.
- Pneumatoceles and pneumothoraces can develop spontaneously with COVID-19 pneumonia even in the absence of positive pressure ventilation.
- Patients with COVID-19 pneumonia should be followed up as complications such as pneumatocele can be a delayed silent manifestation with catastrophic results.
- Large pneumatoceles seldom resolve spontaneously and often need surgical intervention.

Since pulmonary lesions caused by severe acute respiratory syndrome coronavirus 2 infection are predominantly peripheral and sub-pleural in distribution (Qi et al, 2020; Werberich et al, 2020), they are often described as ‘emphysema’ or ‘cystic air spaces’, terms that are used interchangeably with pneumatocele (Shi et al, 2020).

Pneumatocele in the lungs can occur in up to 10% of patients with COVID-19 (Shi et al, 2020). Sato et al (2021), Sugimoto et al (2021) and Odackal et al (2021) mention that while complications such as pneumo-mediastinum and pneumothorax are fairly widely reported, either resulting from mechanical or positive pressure ventilation or directly related to COVID-19 pneumonitis, the development of pneumatoceles during recovery appears less common.

Odackal et al (2021) outlined some theories for the formation of spontaneous pneumatoceles in the absence of positive pressure ventilation and barotrauma, including the respiratory valve being obstructed by inflammatory exudate within the airway lumen leading to distal dilatation of the bronchi and alveoli. Conway (1951) suggested that the focal collection of air in the pleura dissects down to end in pneumatoceles.

Barotrauma in patients who underwent prolonged ventilation with high lung volumes could result in alveolar and intra-acinar ruptured septa; conversely, ventilation with low lung volumes may cause atelectasis especially in patients with acute respiratory distress syndrome (DiBardino et al, 2003; Beitler et al, 2016). This patient received only high flow oxygen therapy during hospitalisation for COVID-19 pneumonia, hence the pneumatocele may have been caused by COVID-19 pneumonia without barotrauma.

Pneumatocele following COVID-19 may have a clinically indolent course and is often undetected. Presence of a pneumatocele is an independent risk factor for pneumothorax, and sudden pneumothorax as a result of a ruptured pneumatocele has resulted in death (Beck, 1998). Cardiopulmonary collapse can also result from rapid enlargement of a pneumatocele. An air–fluid level within the pneumatocele is a sign of infection.

Multiple and small-sized pneumatoceles can be observed periodically as they often regress spontaneously. However, pneumatoceles larger than 6 cm and those that persist often require elective intervention. The authors advocate early percutaneous drainage as a first-line strategy; if this approach fails, surgical resection should be considered, especially in critical patients where it can be life saving.

In the current case, as the lesion was not densely adherent, it could be dissected away from the sleeve of the bronchus and a less radical but novel approach in the form of marsupialisation was carried out. The patient had an uneventful recovery and was stable on follow up.

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