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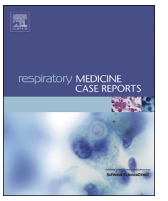

Case Report

Plastic bronchitis secondary to thoracotomy in an adult: A case report

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ABSTRACT

This study presents a rare case of plastic bronchitis (PB) in a 57-year-old adult post-lung lobectomy, a condition predominantly found in post-Fontan children. The patient exhibited progressive dyspnea and complete atelectasis on the surgical side, revealing a gray rubbery bronchial cast obstructing the right main bronchus. Treatment involved repeated bronchoscopies, glucocorticoids, acetylcysteine, physiotherapy, and a low-fat diet, leading to gradual improvement. No similar cases have been reported, highlighting PB's diagnostic challenge. This underscores the need to consider rare conditions like PB in post-lobectomy complications. Timely examinations and bronchoscopies are essential for accurate diagnosis, ensuring prompt treatment and improving patient outcomes.

1. Introduction

Plastic bronchitis (PB) is a rare but life-threatening respiratory condition characterized by the formation of thick, cheesy and bronchial-shaped proteinaceous casts within the airways, leading to obstructive dyspnea. It has been described in populations with respiratory disorders such as asthma, allergies, cystic fibrosis, virus infection, and acute chest syndrome associated with sickle cell disease. However, it is most frequently documented in pediatric patients with congenital heart disease following the Fontan procedure. It is important to identify this type of complication early for timely treatment. Here, we report a 57-yr-old man who had no specific risk factors related to PB who developed bronchial cast formation after a lobectomy procedure. Written informed consent for publication was obtained from the patient.

2. Case presentation

A 57-year-old male patient was admitted to the intensive care unit (ICU) in April 2022 due to progressive dyspnea following an elective lobectomy 6 days ago for pulmonary nodules detected incidentally one year prior. The patient had no history of respiratory symptoms or comorbidities and denied any history of smoking, allergies, or dust exposure. Upon presentation, the patient was noted to have severe respiratory distress with a respiratory rate of 35–40 breaths per minute, tachycardia with a heart rate of 120–140 beats per minute, and hypertension with a blood pressure of 157/106 mmHg. He was also found to have complete absence of respiratory sound in his right lung. A computed tomography (CT) scan revealed complete obstruction of the right mainstem bronchus, resulting in atelectasis of the right lung and moderate pleural effusion (Fig. 1). The patient was immediately initiated on noninvasive mechanical

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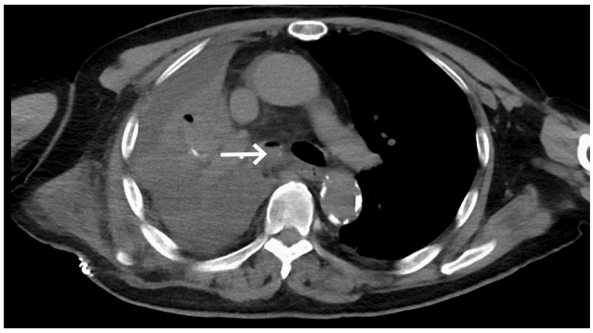

Fig. 1. CT of chest showed totally occlusion of the right mainstem bronchus and atelectasis of the right lung (asterisk).

ventilation and a bronchodilator due to hypoxemia with an arterial oxygen partial pressure to fraction of inspired oxygen ratio (PaO2/FiO2) of 93.

Blood tests showed signs of inflammation, while the patient denied having any foreign bodies stuck in his airway. To identify the underlying cause of the obstruction, a fiberoptic bronchoscopy was performed. This revealed a white-gray, rubbery and thick cast that had completely occluded the right main bronchus (Fig. 2). The cast was difficult to remove, as it was too soft to grasp with forceps but too thick to suction. Subsequently, the patient underwent treatment that involved acetylcysteine nebulization, chest physiotherapy, normal saline lavage, and biopsy forceps, resulting in the extraction of multiple segments of thick mucoid/lipoid-like material and bronchial casts (Fig. 3), which were suggestive of plastic bronchitis. After this treatment, the patient's symptoms improved immediately, and he did not require further treatment. Microscopic examination of the bronchial cast showed the presence of cellulose-like material and a few acute inflammatory cells, while investigations for other possible causes were negative. The chyle test for pleural effusion, however, was positive. The patient was then diagnosed with plastic bronchitis and was treated with systemic and inhaled corticosteroids, acetylcysteine atomization, chest physical therapy, and a low-oil diet. Critical to the treatment were several times of flexible bronchoscopies that were performed to remove the bronchial casts. Thereafter, the patient's dyspnea symptoms were relieved rapidly, and on postoperative day 13, the CT scan showed partial resolution of the atelectasis and consolidation in the right residual lung. Bronchoscopy on the same day revealed a small amount of gray-white thin secretion in the right bronchus, indicating that the treatment was effective. By postoperative day 16, the patient was successfully weaned off noninvasive ventilator support and transferred to a general ward. He was later discharged, and during the follow-up until postoperative day 45, he remained asymptomatic, and no significant abnormality was found on chest CT (Fig. 4).

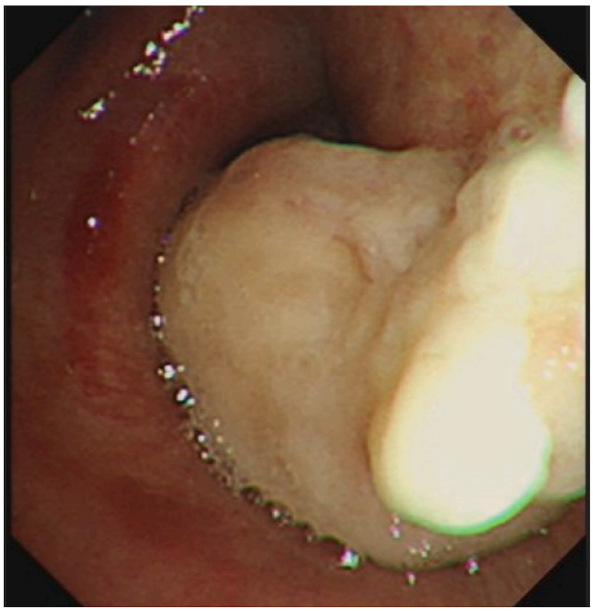

Fig. 2. Fiberoptic bronchoscopy showed a white-gray, rubbery cast completely occluding the right main bronchus.

Fig. 3. Segments of bronchial casts removed from the right main bronchi.

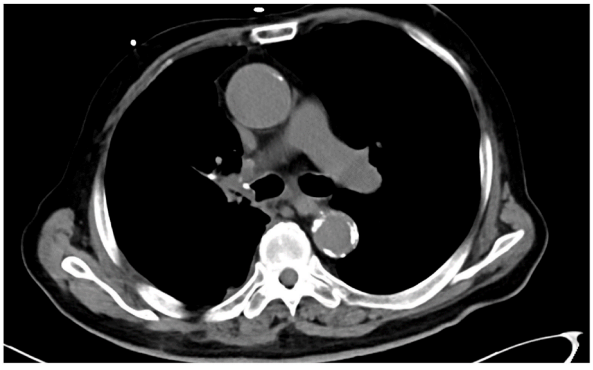

Fig. 4. Chest CT on postoperative day 45.

3. Discussion

Plastic bronchitis is a rare condition characterized by the formation of thick, rubbery mucus plugs that cause airway obstruction, which can be mistaken for foreign body aspiration [1]. In this case, the diagnosis of PB was made based on fiberoptic bronchoscopy and pathology, ruling out other causes of dyspnea post-surgery. To our knowledge, this is the first reported case of plastic bronchitis occurring following routine pulmonary lobectomy.

Although the first case illustrating PB was reported half a century ago [2], the underlying causes and pathogenesis remain unclear. It is believed to be associated with diseases that increase airway secretion volume or viscosity [3]. Typically, PB is classified into 2 types based on histology [4]. Type I is characterized by numerous inflammatory cells (mainly eosinophils and neutrophils) and cellulose, namely, the inflammatory cell type. Type II is a noninflammatory cell type, characterized by a noncellular substance (mainly mucin and little cellulose), which is more common after congenital heart disease surgery with associated abnormal lymphatic drainage. In this case, the cast was classified as type I, consisting of cellulose and inflammatory cells. The formation of casts may be related to abnormal lymphatic circulation, as evidenced by the improvement of symptoms after a low-fat diet. However, the exact causes of cast formation remain unknown.

The optimal treatment for PB is controversial, but expelling casts from the airway is crucial to avoid life-threatening complications [5,6]. Various therapies have been reported to facilitate cast expectoration, including chest physiotherapy, bronchodilators, fibrinolytics, and mucolytics [7–10]. For chronic PB, systemic or inhaled glucocorticoids, antibiotics, hypertonic saline, pulmonary circulation decompression, and lymphatic embolization have been used [11,12]. However, evidence-based research is lacking to evaluate the effectiveness of these measures [13].

In a literature review, only one case of PB after thoracic surgery has been reported [14]. In that case, a 55-year-old man who developed an extensive occlusive bronchial cast after tracheal esophageal fistula repair and died of secondary severe ARDS and uncontrollable infection despite treatment with bronchoscopic extraction, steroids, antibiotics, and antifungals. In contrast, our patient's symptoms rapidly improved with comprehensive bronchoscopy-based treatment and did not recur after discharge.

4. Conclusion

This case highlights the importance of considering PB as a potential cause of post-lobectomy pulmonary atelectasis, particularly when worm-like, rubbery, and thick casts are identified during bronchoscopy. Early diagnosis and treatment with a combination of strategies are crucial to prevent progression to a life-threatening condition. Bronchoscopy plays a critical role in the diagnosis and

management of PB. Additionally, prompt and comprehensive treatment should be initiated, even if the underlying causes of PB remain unclear.

CRediT authorship contribution statement

Jia Wang: Investigation, Writing – original draft, Writing – review & editing. **Long Tian:** Writing – original draft, Writing – review & editing. **Bin Liu:** Supervision, Writing – review & editing.

Declaration of competing interest

No conflict.

List of abbreviations

PB	Plastic bronchitis
ICU	Intensive care unit
PaO_2	Partial Pressure of Oxygen in Arterial Blood
FiO_2	Fraction of inspiration O_2
CT	Computed tomography

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