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Unusual Case of Plastic Bronchitis Presenting with Pneumopericardium

Authors' Contribution:
Study Design A
Data Collection B
Statistical Analysis C
Data Interpretation D
Manuscript Preparation E
Literature Search F
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Patient: Female, 24
Final Diagnosis: Plastic bronchitis
Symptoms: Crepitus • dyspnea • neck fullness
Medication: —
Clinical Procedure: —
Specialty: Pulnonology





Objective: Rare co-existence of disease or pathology
Background: Lymphatic circulation in the thorax enters the systemic blood flow at the subclavian vein. Instances where diversion occurs leads to complications such as pleural effusion. A rare complication of lymphatic diversion results in fluid accumulation in the bronchial tree, causing plastic bronchitis. The following case is the first ever report of plastic bronchitis presenting with pneumopericardium.

Case Report: A 24-year-old female presented to our Emergency Department with an asthma exacerbation. After initiating bronchodilators, a chest radiograph (CXR) showed extensive subcutaneous emphysema, pneumomediastinum, and pneumothorax with atelectasis of the left lung. Chest tomography supported the CXR findings, as well as a finding of pneumopericardium. A thoracostomy tube was placed and a mediastinal window and pericardial window procedure were performed in an attempt to relieve the pressure upon the collapsed lung. Despite these invasive procedures, there was minimal improvement of lung volume with further respiratory deterioration; the patient eventually required mechanical ventilation. Bronchoscopy was performed, which evacuated a white chalky and rubbery substance that created a mold of the bronchial airways. Following the bronchoscopy, the patient's respiratory status improved, requiring less ventilator support, and that patient was successfully extubated.

Conclusions: This case highlights the most crucial management of a patient presenting with pneumomediastinum, pneumothorax, and pneumopericardium due to plastic bronchitis. Plastic bronchitis should be high on the list of differential diagnoses. The management of plastic bronchitis with bronchoscopy is supported by the fact that no invasive therapy such as thoracostomy tube or mechanical ventilator alleviated the problem, however, bronchoscopy removed the worm-like cast lodged within the lumen of the bronchial tree.

MeSH Keywords: Bronchitis • Bronchoscopy • Pneumopericardium

Full-text PDF: <https://www.amjcaserep.com/abstract/index/idArt/911311>

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Background

Lymphatic circulation in the thorax traverses from the lymphatic system to the subclavian vein, returning into systemic circulation. Abnormal flow is often retrograde from the thoracic duct to the lung parenchyma [1]. Instances where the drainage is diverted leads to complications such as pleural effusion, or even more rarely, bronchitis within the spectrum of pulmonary lymphatic perfusion syndrome [2]. Patients prone to diversion of lymph fluid are those with congenital cardiac anomalies or pulmonary diseases such as asthma. Plastic bronchitis is a rare disorder in which the lymphatic fluid collects in the bronchial airway forming a rubbery substance obstructing airflow characterized by large, rigid airway plugs, often more cohesive than ordinary mucus plugs [3]. In a large university center, researchers discovered 14 cases in a pool of over 205 000 patients, portraying the rarity of the disease [4]. We present the first ever reported case of plastic bronchitis in a patient who was found to have a pneumopericardium.

Case Report

This is a case of a 24-year-old previously healthy female presenting to the hospital with complaints of shortness of breath with neck fullness that acutely worsened prior to arrival. She began feeling ill approximately 1 week prior to her visit, with upper respiratory tract infection with occasional chest tightness. In the Emergency Department, a chest radiograph showed extensive pneumomediastinum, pneumothorax, and subcutaneous emphysema. Follow-up chest tomography confirmed the pneumomediastinum, emphysema, as well as pneumopericardium (Figure 1). The patient received a chest tube, and pericardial window and mediastinal window procedures. Despite chest tube placement, the left upper and lower lobes remained collapsed. Fiber optic bronchoscopy was performed which revealed extensive bilateral mucous plugging with findings described as plastic worm-like substance following the tracks of the airway mostly in the left upper lobe, left lower lobe, and right upper lobe (Figure 2).

Discussion

Mucosal or alveolar capillary barrier injury causes cast formation by protein-rich material, and endobronchial lymph or exudative airway inflammation [5]. These casts were composed of material with proteins, mucus, neutrophils, and eosinophils depending on the type of group [6,7]. Type 1 inflammatory casts consist of fibrin and inflammatory cells such as eosinophils, whereas type 2 acellular casts consist primarily of mucin. Type 2 generally occurs in children with congenital heart disease [8,9]. Type 1 is often associated with an acute

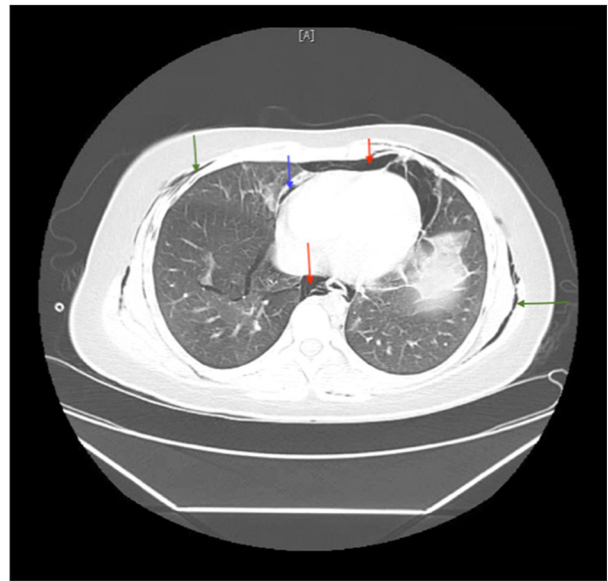


Figure 1. Chest computed tomography: green arrow, Subcutaneous emphysema; red arrows, Pneumomediastinum; blue arrow, Pneumopericardium.



Figure 2. Worm-like mucous plug.

presentation versus the chronic or recurrent presentation of type 2 [10]. One case reported in the literature demonstrated plastic bronchitis secondary to silicosis, and the casts consisted of silica laden macrophages [11].

The management of plastic bronchitis consists largely of mechanical evacuation of the casts acutely with bronchoscopy

and preventing recurrence. A large case series of plastic bronchitis patients found that extraction via bronchoscopy is the only effective treatment [5]. Prior to extraction, the treatment includes bronchodilators, corticosteroids, mucolytics, and macrolide antibiotics [9]. One case report found that steroid treatment dissolved the secretions [12]. In cases where clots cannot be mobilized, chest physiotherapy might be an option. Dissolving the clots can be achieved by either inhaled mucolytic or fibrinolytic agents such as tissue plasminogen activator (tPA) [13]. Another study utilized thoracic duct stents to prevent abnormal or retrograde flow back to the pulmonary circulation [1]. One group found that utilization of complete parenteral low-fat nutrition decreased the amount of cast formation [14].

Conclusions

This case presents a rare cause of dyspnea in a young healthy patient without pre-existing comorbidities, and illustrates the severe complications of plastic bronchitis. In the event a

dyspneic patient is found to have extensive pneumomediastinum, pneumopericardium, pneumothorax, or subcutaneous emphysema, unexplained by usual causes such as trauma, then plastic bronchitis should be included as a differential diagnosis. This is especially true if there is no improvement of lobar atelectasis after chest tube insertion. Asthma can cause plastic bronchitis with type 1 cast formation. It is important to distinguish classic asthma exacerbations from exacerbations with cast formation as the treatment will vary considerably. Although there were many possible treatment options discussed in this case report, patients rarely improve without the extraction of casts by bronchoscopy.

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Conflict of interest

None.

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