Case Report

A Case of Plastic Bronchitis

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Abstract

Plastic bronchitis, causing airway obstruction, is a rare condition, especially in adults. In this paper, an adult male patient with heart failure, viral hepatitis, and a history of previous pneumonia, and expectorating white, plaque-like secretions for the last 6 months is presented along with the review of the relevant literature. The material expectorated by the patient was evaluated; macroscopically, this material was observed to be identical to the shape of bronchial branching. Steroid treatment was commenced after the diagnosis was established, resulting in the nearly total disappearance of the expectorated material.

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Introduction

Plastic bronchitis (PB) is a rare disease, characterized by gelatinous or rigid endobronchial plaque-like formations, identical to the shape of the bronchial airways. This disease was named as Hoffman's bronchitis, cast bronchitis, pseudomembranous bronchitis or fibrinous bronchitis in the past.[1] The material (cast) in the bronchi is generally expectorated spontaneously or with a strong cough.[2]

In this paper, we present an adult patient diagnosed with PB, in the light of the relevant literature.

Case Report

A 47 year-old male patient who was a farmer was admitted to our hospital with complaints of cough and sputum for the last 6 months and white-colored worm-shaped expectoration 2 to 3 times a week. The patient had untreated systolic heart failure and chronic hepatitis B and he was treated with the diagnosis of pneumonia 2 years ago. Cardiopulmonary assessment revealed inspiratory crackles on the right lung base and a systolic murmur, grade 3/6, on the mitral area. Laboratory test results were within normal limits, except an elevation in C-reactive protein [1.7 g/dL (normal range=0–1 g/dL)] levels. When the expectorated white plaque-like secretions were assessed macroscopically, it was observed that they were sticky, gelatinous and their shape was consistent with the anatomy of the bronchi (Figure 1). Chest X-ray revealed cardiomegaly and non-homogeneous patchy infiltration in the right lower zone. Computed tomography of the chest demonstrated ground-glass appearance, patchy areas of consolidation, and peribronchial thickening in the right inferior lobe of the lung. Respiratory function tests were within normal limits. The echocardiogram revealed grade 3 mitral insufficiency and dilatation in the left heart chambers, with ejection fraction at 30%. The patient underwent bronchoscopy. The mucosa was fragile and hyperemic. At the entrance of the right inferior lobe, there was a white plaque-like formation, almost completely obstructing the lumen and separated with difficulty from the bronchial wall by aspiration. Bronchial lavage and the aspirated plaques were evaluated for tuberculosis, fungal and bacterial infections, and no specific finding was observed. Pathological analysis of the aspirated plaques revealed wide fibrin deposits, mature lymphocytes, a few histiocytes, and bronchial epithelial cells (Figure 2). Based on these findings, the patient was diagnosed with PB.

At first, the patient was given treatment for heart failure and clarithromycin at a dose of 1000 mg/day. As there was no decrease in the amount of expectoration after 2 weeks of treatment, 40 mg/day methylprednisolone was commenced in addition to the medications given for heart failure treatment. In the first month of the treatment, the patient stated that the amount of expectoration decreased and disappeared almost completely with time. The patchy infiltration observed on the previous X-ray was completely resolved on the control chest X-ray. Bronchial cast formations were not observed in the control bronchoscopy. Steroid treatment was planned to be tapered and discontinued over 6 months.

Discussion

Plastic bronchitis is a rare obstructive airways disease characterized by coughing and rubber-like, sticky bronchial plaque expectoration obstructing the tracheobronchial tree.[3] The diagnosis is generally established by analysis of the material expectorated or removed by bronchoscopy. Most cases reported in the literature are in the childhood age group and the incidence of the disease in adults is very low. In children, it is generally seen as a complication of congenital heart defect corrective surgery.[4,5] In addition, PB is reported to occur in individuals with lung diseases such as bronchiectasis, tuberculosis, asthma, cystic fibrosis or chronic bronchitis as well as chronic heart disease, rheumatoid arthritis, and amyloidosis.[6,7] Our case had heart failure, chronic hepatitis, and a history of previous pneumonia 2 years ago.

According to the pathological classification commonly used to classify the expectorated casts, the casts are divided into two groups: Type-1 (inflammatory) and Type-2 (acellular). Type-1 casts consist dense fibrin with inflammatory cell infiltration, particularly involving eosinophils, while Type-2 casts are composed of mucin, and sometimes mononuclear cells. The researchers who developed this classification stated that acellular casts develop following surgical treatment of cyanotic congenital heart diseases.

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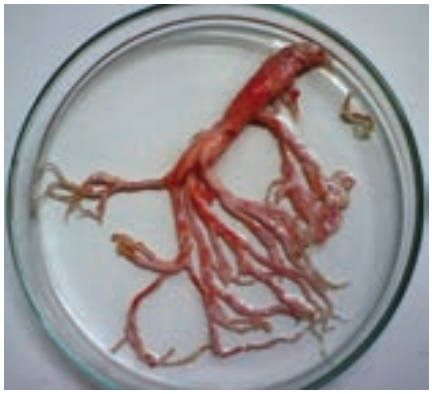

Figure 1. Macroscopic appearance of the expectorated material.

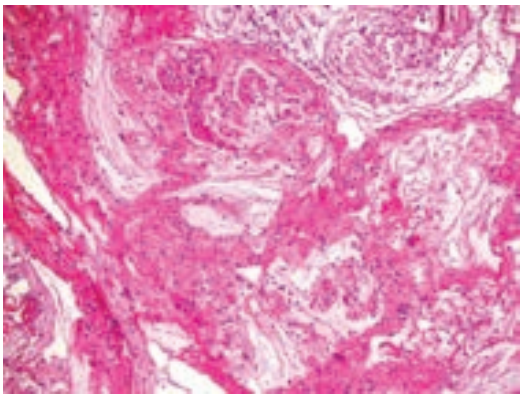

Figure 2. Pathological analysis of the aspirated plaques revealed wide fibrin deposits, mature lymphocytes, a few histiocytes, and bronchial epithelial cells.

They suggested that this condition results from the response of the bronchial epithelium to increased mucin production and elevated pulmonary venous blood pressure.[8] In another study, trauma to the lymphatic vessels around the bronchus, pleural adhesions, and high systemic venous blood pressure caused by this type of corrective cardiac surgery performed in the pediatric age group were determined to be the cause of the casts.[9] In our case, the findings resembled both types of casts. Pneumonia added to the longstanding heart failure might have resulted in the development of casts in our patient. Hence, we observed a pathological appearance involving the characteristics of both cast types according to the classical classification.

Radiologically, PB is generally characterized by patchy atelectasis in the involved segment and compensatory hyperinflation in the adjacent areas.[1,4] However, patients with bilateral patchy consolidation, without findings of volume loss have been reported, as well.[10,11] Patchy consolidations were determined by both chest X-ray and chest tomography in our case.

Traditional treatment of this disease involves bronchodilators, inhaled and oral corticosteroids, mucolytics, antibiotics, and mechanical airway clearance.[1] Apart from these, alternative to bronchoscopy and in addition to medical treatment, inhaled heparin,[12] aerosolized tissue plasminogen activator,[5,13] rhDNase,[10] macrolide antibiotics for mucoregulatory treatment,[14] and high frequency jet ventilation[15] have also been reported in the literature. There is only anecdotal evidence about the benefits of this treatment and this evidence has been generally provided from one or a few patients.

In the largest-scale study evaluating the treatment of PB, the researchers retrospectively reviewed a large series of 32 patients gathered during approximately 10 years. The PB patients who were admitted with hemoptysis on the first attack were divided into two groups; those receiving or not receiving steroids. There was a significant decrease in the amount of hemoptysis and bronchial casts in patients receiving steroids versus those not receiving steroids. However, there was no difference between the groups in terms of mortality and requirement for mechanical ventilation.[6]

In the first month of steroid treatment, bronchial casts in our patient disappeared almost completely. Additionally, radiological and bronchoscopy findings also improved.

We believe that this case report will contribute to the literature due to several facts; first, a patient with heart failure, viral hepatitis, and a history of previous pneumonia as well as PB has not been reported in the literature previously. Secondly, the patient benefited from steroid treatment; and thirdly, PB is a rare disease in adults.

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