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CASE REPORT

Pediatric plastic bronchitis: A case report and literature review

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Summary Plastic bronchitis is a rare condition characterized by inspissated endobronchial casts that can lead to respiratory distress and even death. We present a case of plastic bronchitis in a previously healthy two-year-old girl. Bronchoscopy revealed the characteristic inspissated endobronchial cast. With removal of the obstructive airway debris, her clinical condition rapidly improved. A review of the literature was performed to elucidate the etiology, pathogenesis, presentation, diagnosis, classification, and management of plastic bronchitis. Underlying etiologies include congenital heart disease, pulmonary disorders, and a few other conditions. Treatment includes rigid bronchoscopy with cast removal and treatment of the underlying condition.

1. Introduction

Plastic bronchitis is an unusual, potentially fatal condition characterized by the formation of extensive inspissated mucofibrinous endobronchial casts that mimic the three-dimensional architecture of the tracheobronchial tree [1,2]. It is the cohesiveness, consistency, and notoriously difficult bronchoscopic removal that delineates this condition from ordinary mucus plugging [3].

Patients may present with expectoration of these casts or they may be found at bronchoscopy or in a surgical specimen. Conditions associated with plastic bronchitis include congenital heart disease, especially patients with Fontan physiology (diversion of systemic venous return to the pulmonary circulation), various pulmonary disorders including asthma and cystic fibrosis, as well as other conditions such as sickle cell anemia [1]. The pathogenesis is unclear and may represent a final common pathway to the disorder.

The treatment for plastic bronchitis varies from medical treatment with corticosteroids to various inhaled lytic agents, bronchoscopy, and potentially other surgical treatments.

2. Case

A two-year-old previously healthy girl presented with a three-day history of barky cough, low-grade fevers, and progressive respiratory distress. She had a history of subclinical asthma, but no other underlying disorders. On physical examination she was in moderate respiratory distress requiring supplemen-

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tal oxygen by facemask, was tachypnic, had sternal retractions, and decreased breath sounds in the right upper and middle lobes. A chest X-ray showed a hazy right upper lobe lung field with suggestion of obstruction at the area of the bronchus intermedius (Fig. 1).

The decision was made to take the patient to the operating room for bronchoscopy to rule out foreign body. Instead, tenacious bronchial mucus casts were removed from the right upper lobe bronchus and bronchus intermedius. Histopathology demonstrated that the casts were composed of dense fibrinous debris with an eosinophil-rich, mixed inflammatory infiltrate (Fig. 2).

Postoperatively, the patient's clinical condition rapidly improved and she was discharged to home on postoperative day two. The patient has not had any recurrences to date and remains on as needed treatment with albuterol for mild asthma. A sweat choride test was negative.

3. Discussion

Historically, the first decriptions of plastic bronchitis date back to Galen (AD 131-200) in which he described ''venae arteriosae expectorantii,'' literally ''expectorated arteries and veins.'' Morgagni later postulated that these structures consisted of inspissated bronchial mucus [4]. The characteristic branching mucoid bronchial casts once referred to as fibrinous bronchitis or pseudomembranous bronchitis came to be uniformly known as plastic bronchitis in the 20th century [5].

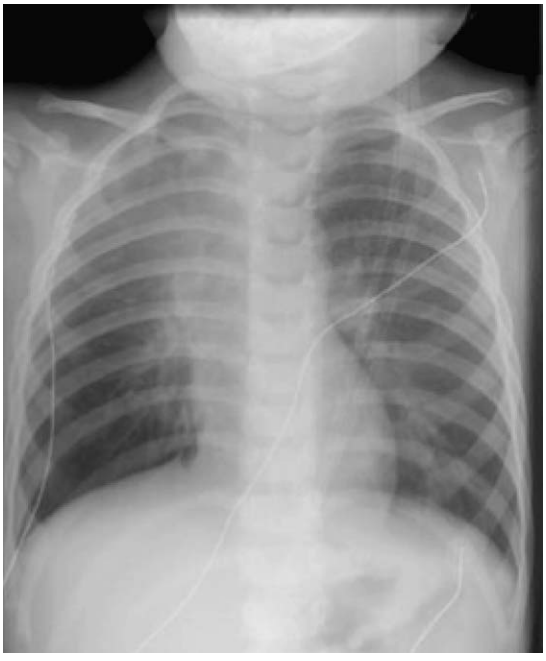

Fig. 1 PA CXR showing a hazy right upper lobe lung field with suggestion of obstruction at the area of the bronchus intermedius.

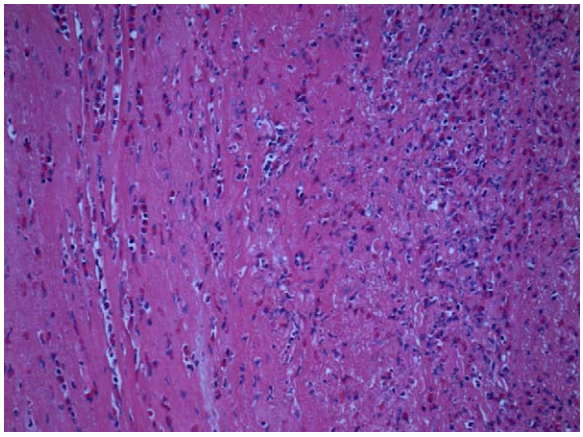

Fig. 2 Histopathology demonstrating that the casts were composed of dense fibrinous debris with an eosinophil-rich, mixed inflammatory infiltrate.

Demographically, plastic bronchitis affects all age groups but is more freqeuntly reported in children, average age of six years in a relatively large review [1]. Males and females tend to be equally affected according to most reports, but Eberlein et al. suggested a female predominance [1,5,6]. The true prevalence of plastic bronchitis in unkown.

Clinical presentation varies from mild symptoms to life threatening disease. The hallmark is expectoration of branching bronchial casts or discovery of these on bronchoscopy. Typically patients present with dyspnea, coughing, wheezing, and respiratory distress, occasionally with fevers or chest pain [3,7]. Radiographic evaluation typically shows atelectasis and infiltrates on the ipsilateral side, often with hyperinflation of the contralateral side. CT scanning may allow for visualization of impacted casts within the major airways [8]. Diagnosis may be made from previously expectorated casts or on bronchoscopy.

The pathogenesis of plastic bronchitis is unclear. Several theories have been proposed including increased pulmonary venous pressure, increased inflammatory response, trauma to bronchial lymphatic channels, and ischemia of the bronchial tree [3,7,9]. Plastic bronchitis may represent a final common pathway to a variety of insults to the bronchial tree.

Seear et al. have proposed a two-type classification scheme based on cast histology as shown in Table 1. Type I or inflammatory casts are composed of fibrin with a dense eosinophilic infiltrate. These casts tend to be associated with broncho-pulmonary disorders. Patients with type I casts often present with a more acute clinical presentation. Type II or acellular casts consist maily of mucin. Type II casts