

Plastic Bronchitis in a Child Presenting as Surgical Emphysema: a Case Report

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Abstract

Plastic bronchitis (PB) refers to the presence of inspissated Bronchial casts (BC) is uncommon in children. Our case presented with subcutaneous emphysema as the dominant manifestation and was suspected to have foreign body in airway, but bronchoscopy revealed a bronchial cast. Subcutaneous emphysema as the initial/sole manifestation of plastic bronchitis is very rare in children.

Key Words: Bronchial casts, Plastic Bronchitis, Surgical Emphysema.

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Introduction

Plastic bronchitis is characterized by the presence of inspissated bronchial casts (1). It was first reported by Galen (131–200 AD), as the expectoration of “arteries and veins” (2). Previously it was also known as fibrinous bronchitis or pseudo-membranous bronchitis and was more prevalent in the 19th and early part of the 20th centuries than it is today (3). Based on the predominance of mucin over fibrin as the cast constituent, the term fibrinous bronchitis now has been replaced by plastic bronchitis.

Though expectoration of large, branching casts is pathognomonic, in children it usually presents with non-specific features like fever, cough and progressive dyspnoea (2, 4). In chest auscultation besides wheeze and decreased breath sounds, sometimes “flag snapping” sign (also called bruit de drapeau) may be heard due to movement of partially obstructing cast in the bronchus (5). Large casts may mimic clinical presentation of foreign body aspiration also. Subcutaneous emphysema as a presentation of foreign body aspiration per se is rare in children (6).

Case Presentation

A 3-year-old male child presented in our emergency department (Dr. SN Medical College, JODHPUR, India) with complaints of progressively increasing swelling over chest, neck and face, associated with distressed breathing for the last 2 days. There was no recent history of cough, cold, fever or choking episode. On examination, patient was a febrile, with heart rate 110 beats per minute, respiratory

rate 58 per minute, and spO₂ 84% on room air. Along with signs of distress, surgical emphysema was noted over chest, neck and face. On chest auscultation there were no added sounds, but air entry on left side was markedly decreased. Rest of the systemic examination was within normal limit. Oxygen supplementation improved saturation up to 93%. Chest x-ray (CXR) revealed collapse on the left side.

Based on clinico-radiological findings a probable diagnosis of foreign body aspiration was entertained, but as there was no history of choking episode, Computed Tomography (CT)- chest was ordered to confirm the diagnosis. CT thorax showed an ill defined soft tissue attenuation mass lesion obliterating the left main bronchus with collapse of left lower lobe and further supported our diagnosis (Figure.1). Child underwent rigid bronchoscopy, which revealed a mass at carina extending towards the left bronchus. Retrieved mass resembled the branching pattern of bronchial tree (Figure.2). After removal of bronchial, gradually child's distress settled and surgical emphysema improved.

Casts histology showed an eosinophilic, fibrinous material with scant amount of mucin at the edges. Most cells in the cast were mononuclear cells. Child was evaluated for the underlying cause of plastic bronchitis, but history, clinical examination, blood counts and Echocardiography (echo) did not suggest of any allergic or infective bronchopulmonary disorder or heart disease. Child was discharged on fifth day of admission and was asymptomatic till 6 months of follow up.

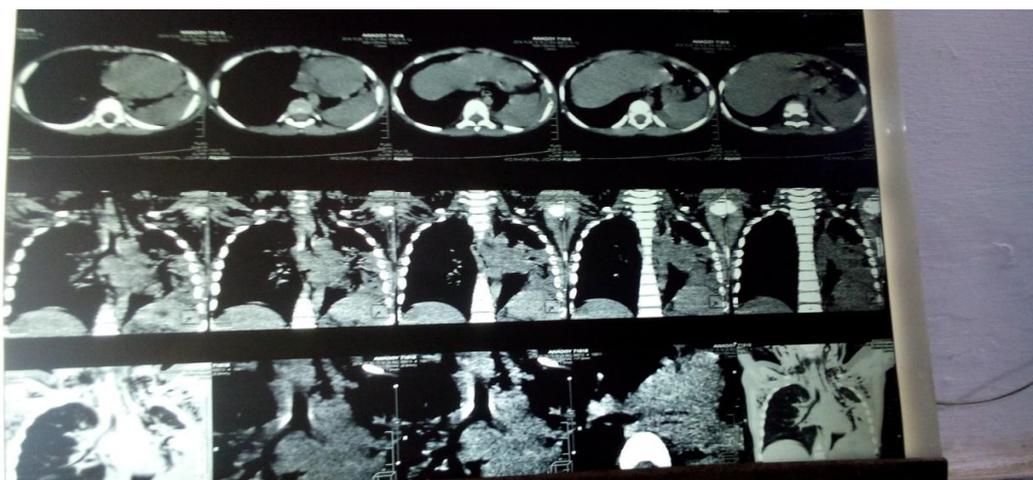


Fig.1: CT SCAN showing collapsed left lung



Fig.2: Bronchial cast removed after bronchoscopy

Discussion

Histological bronchial casts are classified into two categories; type I or cellular casts and type II or hypocellular casts (7). Cellular casts are mainly composed of neutrophils, eosinophils and fibrin and are more commonly associated with allergic and infective bronchopulmonary disorders, whereas hypocellular casts are predominantly composed of mucin and are more commonly associated with congenital heart diseases. Plastic bronchitis has been reported in association with rheumatoid arthritis, amyloidosis, membranous colitis, and large thymuses also (8). In some of the cases despite extensive evaluation, no cause is identified and is labelled as idiopathic, like in our

case. Though exact mechanism of cast formation is not clear, abnormal lymphatic drainage and increased pulmonary venous pressure have been implicated in the pathogenesis. Madsen et al. proposed a 2-step model of cast pathogenesis; susceptible genetic background with superimposed mucus secretion (7, 9, 10).

Pathogenesis of surgical emphysema in plastic bronchitis is not known. A possible explanation is check valve type of bronchial obstruction leading to lobar emphysema. Increased pressure in the alveolus then causes air leak towards the mediastinum through the bronchovascular plane. As the pressure in the mediastinum is increased, decompression occurs in subcutaneous and cervical spaces leading

to subcutaneous emphysema (11). In our case cast behaved like ball valve leading to complete obstruction of left bronchus and subsequent collapse of left lung. Development of compensatory emphysema in contra lateral lung does not explain the subcutaneous emphysema in our case. Probably bronchial cast extending in to carina behaved like check valve for the right lung complicating in to subcutaneous emphysema as described above. Plastic bronchitis is usually a self-limited disease. Medical management varies from case to case and includes bronchial lavage, hydration, mucolytics [acetylcysteine and dornase alpha, fibrinolytics (heparin, urokinase, t-PA)], bronchodilators, corticosteroids, antibiotics, chest physiotherapy and high frequency jet ventilation. Casts are usually coughed up spontaneously, but when they are large and complicating may require bronchoscopic removal also, like in our case it had obstructed whole of the left bronchus leading to air leak syndrome. Rarely surgical interventions like thoracic duct ligation, fenestration of the Fontan circuit in congenital heart diseases and lobectomy may be required (12). If underlying factors are not addressed, bronchial casts may recur also.

Conclusion

Subcutaneous emphysema should be kept in mind as a manifestation of plastic bronchitis when dealing with such type of case.

Conflict of interest: None.

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