Bronchial cast: a case report

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Abstract We present a case of a child who presented with respiratory distress mimicking foreign body aspiration which was treated by bronchoscopic extraction of bronchial cast. Early interventional bronchoscopy in management of plastic bronchitis, though difficult, provides an immediate benefit and good prognosis especially in patients with no underlying cardiopulmonary morbidity.

Keywords Bronchial cast · Bronchoscopy · Plastic bronchitis

Introduction

Plastic bronchitis is an unusual disorder denoting the presence of inspissated bronchial casts that may be coughed up or found at bronchoscopy or in surgical specimen [1]. Bronchial casts are associated with mucus hypersecretion, bronchopulmonary bacterial infection, congenital cardiopathies or pulmonary lymphatic abnormalities [2]. The casts are large pale tan, rubber like causing severe airway obstruction. They could mimic foreign body aspiration or status asthmaticus requiring bronchoscopic extraction [3]. We present a case of one year old child who came with acute onset, respiratory distress mimicking a foreign body aspiration. Bronchoscopic removal of the cast, the histopathological features are described and the literature reviewed.

Case report

An one-year-old male child presented to us with sudden onset, severe respiratory distress for the last twenty four hours. There was progressive increase in the distress. The parents gave no history of fever, upper respiratory tract infection or that suggestive of foreign body aspiration. There was no history of asthma in the child or in the family.

On examination, the child had tachypnoea with decreased chest movements on the left side. Auscultation of the chest showed absent air entry on the left side.

Radiograph of the chest showed shift of mediastinum to the left, haziness in the left upper zone and emphysematous changes on the right side (Fig. 1). We suspected it to be a case of foreign body aspiration.

Accordingly consent for grave risk and tracheostomy was taken from the relatives. Rigid bronchoscopy under general anaesthesia with venturi assisted ventilation, was carried out. The rigid bronchoscope of 3.5 mm was used. On visualizing the left main bronchus, white thick material was seen which could not be sucked out with suction tip. It

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was also difficult to grasp and remove with forceps. After several attempts the thick rubbery material was removed with suction cannula and was put in saline where it revealed itself as a cast of the branching pattern of the left bronchial tree (Fig. 2). The right bronchus was normal. The air entry on left side improved immediately.

In the postoperative period the child was given nebulisation with ambroxol and fluticasone and, oxygen by mask. Intravenous antibiotics, steroids, bronchodilators and mucolytics were included in the postoperative management. The child completely recovered.

Histopathological examination of the cast revealed fibrin with inflammatory exudates. No organisms were isolated from the bronchial aspirate.

Complete roentgenographic resolution was seen in the X-ray of chest taken after 48 hours (Fig. 3). Post operative evaluation of the patient by 2 Dimensional Echocardiography showed that the child had no cardiovascular anomaly or any underlying pulmonary disease that could predispose him to recurrent bronchial cast formation. The child was discharged and followed up to six months. There was no evidence of recurrence during this period.

Discussion

Plastic bronchitis is characterized by the acute and often recurrent development of bronchial casts, which cause obstruction of the tracheobronchial tree and thus, respiratory distress. It is rare and can occur at any age [4]. The casts are of variable sizes and take the shape of the bronchi in which they form. The condition is also called fibrinous bronchitis, pseudomembranous bronchitis [4], bronchitis fibroplastica and bronchitis plastica.

Plastic bronchitis occurs at all levels of the tracheobronchial tree, but is more common in the lower

lobe. Mucoid impaction differs from plastic bronchitis in several ways; the plugs tend to be in the large segmental bronchi of the upper lobe; usually adherent to the wall and are rather retained than expectorated [1]. In most cases, bronchial casts are secondary to underlying diseases of the lung, heart or lymph vessels [2]. These disorders either produce an increase in volume or viscosity of secretions, such as allergic bronchopulmonary aspergillosis, asthma, cystic fibrosis, pneumonia and chronic bronchitis; or structural abnormalities that decrease clearance, such as bronchiectasis. It has been reported in acute chest syndrome of sickle cell disease [5].

Cardiopathies associated with plastic bronchitis include constrictive pericarditis, congenital heart disease [4], patients who have undergone Fontan operation for congenital cardiac anomaly [2]. After Fontan operation for single ventricle anomaly where superior vena cava is joined to pulmonary arteries and the right ventricle is bypassed,

Fig. 2 Bronchial cast showing typical branching pattern of left bronchus.

Fig. 1 Radiograph of chest showing collapse, consolidation on left with emphysema on right..

Fig. 3 Radiograph of chest taken 48 hours post- op showing normal findings.

the resultant increased central venous pressure being responsible for cast formation.

The formation of bronchial casts has been also attributed to lymphatic leakage into the bronchi. In patients with no congenital lymphatic anomalies, the principal factors resulting in formation of casts may be surgical trauma to the lymphatic channels surrounding the bronchi, pleural adhesions and high systemic venous pressure [2].

No underlying disease has been found in some cases rendering them, 'idiopathic' [4, 6].

Our patient had no history suggestive of any respiratory or cardiac disease nor did the investigations done post operatively reveal such findings, hence we can say that it was idiopathic. He presented with acute severe respiratory distress mimicking foreign body aspiration. It has been reported that plastic bronchitis is an infrequent cause of acute life threatening respiratory failure that can mimic foreign body aspiration or status asthmaticus making urgent bronchoscopy necessary [3, 6].

It has been proposed that the bronchial casts may be divided into two clinicopathological groups [4]. Type I casts are inflammatory, consisting mainly of fibrin with eosinophilic cellular infiltrates, and occur in inflammatory diseases of the lung. Type II, or acellular casts, consist mainly of mucin with a few cells, and usually occur following surgery for congenital cardiac defects. Our patient's cast had histopathological features of inflammatory type.

A high degree of suspicion is necessary to make the diagnosis of plastic bronchitis. Though computerized tomography of thorax is more informative a preliminary radiograph of the chest helps, but the diagnosis is made by bronchoscopy and removal of the casts. Radiograph of the chest in our patient was suggestive of foreign body aspiration on the left side with compensatory emphysema on the right. Radiological findings during periods of bronchial cast formation include atelectasis, obstructive emphysema, bronchiectasis, pleural effusion and pneumomediastinum [1].

Treatment of plastic bronchitis includes specific measures to treat the underlying pulmonary condition as well as maneuvers designed to remove or facilitate the expectoration of bronchial casts [1]. In cases like ours wherein patients present with acute onset severe respiratory distress, urgent bronchoscopy for diagnosis and removal of casts is recommended [3, 6]. This entity is probably underestimated, as the casts with their specific ramifications are difficult to recognize. It has been recommended to immerse all mucus plugs discovered in patients with predisposing diseases, in normal saline [3]. Removal of casts by bronchoscopy is technically challenging. This is because the material is too soft to grab with forceps and too thick to be suctioned out [6]. In our case we succeeded in removing the thick white rubbery cast from the left bronchus, bronchoscopically after several attempts in the same sitting by gentle pulling with forceps. On immersing the material in saline, it showed the typical branching pattern of the left lung.

Various treatment modalities for facilitating the expectoration or removal of casts are mucolytics, chest physiotherapy, use of intratracheal rhDNase [5], aerosolized acetylcysteine which breaks disulfide bonds in mucoproteins, subcutaneous high molecular weight heparin, inhalation of heparin, aerosolized urokinase, aerosolized tissue plasminogen activator that causes fibrin enhanced conversion of plasminogen to plasmin initializing local fibrinolysis, oral corticosteroids and saline lavage. Low dose azithromycin has been also advocated. Surgical intervention is in form of endoscopic removal but in extreme cases lobectomy has been also described [1].

Postoperatively our patient was given intravenous antibiotics, bronchodilators, steroids, mucolytics and nebulisations. Investigations revealed no cardiopulmonary anomalies as predisposing factors. Recovery was complete with no recurrence. In patients with underlying cardiopulmonary diseases recurrences are common, and are high risk for serious complications especially those with cardiac disease.

Conclusions

Plastic bronchitis is a rare condition and an unusual cause for sudden onset respiratory distress mimicking foreign body in bronchus or status asthmaticus. Thus all that wheezes is not asthma. Early interventional bronchoscopy is justified. Management of plastic bronchitis itself presents an unusual and interesting bronchoscopic challenge. Though difficult it provides immediate benefit and good prognosis especially in patients with no underlying cardiopulmonary morbidity.

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