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Case Report

Case Report of a Left Upper Bronchial Cast in A Lebanese Adolescent Patient Diagnosed After Persistent Pneumonia with Obstruction Removed by Dual Rigid and Flexible Bronchoscopy

Ammar El Kheir[1] and Chadi AL Alam[2]*

[1]Pediatrics, Pediatric Pulmonary and Allergic Diseases, Saudi German Hospital, Dammam, KSA and Haykel Hospital, El Koura, Lebanon

[2]Pediatrics and Pediatric Neurology, American Center for Psychiatry and Neurology - ACPN, Abu Dhabi, UAE and Haykel Hospital, El Koura, Lebanon

***Corresponding Author:** Chadi AL Alam, Pediatrics and Pediatric Neurology, American Center for Psychiatry and Neurology - ACPN, Abu Dhabi, UAE and Haykel Hospital, El Koura, Lebanon.

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Abstract

We present in this case an adolescent Lebanese male patient who initially was treated for resistant pneumonia by another physician since 8 months without improvement. Symptoms was consistent with coughing and episodes of fever. During the second hospitalization under intravenous antibiotics, he was not improving, and pulmonology consultation was done. A bronchial cast was suspected due the history of non-treated asthma and the imaging. Patient underwent bronchoscopy for the removal of the cast with direct amelioration of symptoms and imaging follow up.

Keywords: Bronchial Cast; Bronchoscopy; Atelectasis; Resistant Pneumonia; Non-treated Asthma

Introduction

The term bronchial cast or plastic bronchitis is a rare pulmonary condition that refer to the blockage of distal or intermediate airway by the formation of branching intra-bronchial cast. Due to the rarity of cases, the prevalence is not known. The symptoms consists of productive cough to dyspnea, fever, pleuritic pain and even respiratory failure in rare cases. As possible causes, it presents usually in the pediatric population with heart diseases or post open-heart surgery. Other causes could be infectious due to atopy, sickle cell disease, cystic fibrosis, or anatomic/lymphatic malformations. Treatment can vary from pharmacological to bronchoscopy or even surgical intervention.

Case Report

In this study, we present a case of a 13-year-old adolescent male patient who was complaining of persistent right upper lobe pneumonia since 8 months.

As by history patient is a full-term infants born to non-consanguineous parents. He was born by normal vaginal delivery without complications. He had good neonatal adaptation with negative neonatal history.

Upon admission to the hospital, patient was complaining of fever with productive cough and mild respiratory distress with mild tachypnea of 28-30 respiratory rate per minute, associated with mild suprasternal and subcostal retractions. Initial laboratory results showed an elevated CRP with leukocytosis of 16.700 and neutrophils of 81%. Chest X-ray done and showed left upper lobe consolidation with atelectasis. (X ray attached – image 1) Patient was started on intravenous antibiotic (Ceftriaxone 50 mg/kg/day and Vancomycin 60mg/kg/day) and pediatric pulmonary consult was done.

A thorough history showed that our patient was treated once as inpatient and twice as outpatient with antibiotics for the same

Image 1

Image 2

condition. He only had partial and transitory improvement, but the left upper lobe obstruction persisted. He also was found to have history of allergic rhinitis and mild asthma symptoms without proper treatment.

CT scan was ordered and showed consolidation collapse of the left upper lobe with complete obstruction of the proximal main left upper lobe bronchus, underlying obstructive plug or foreign body to be excluded. A sub segmental consolidation collapse of the anterior segment of the left lower lobe with partial filling defect was also found. To note that PPD test was done and was negative.

Foreign body was excluded due to the history, so patient was suspected to have bronchial cast due to even infectious origin or a non-treated asthma with secondary infection.

Flexible bronchoscopy was planned for broncho-alveolar lavage with exploration. Under general anesthesia through a laryngeal mask, a 5mm bronchoscope was introduced. A mucus plug was found over the superior segment of left lower lobe and was removed, but a bronchial cast was found over the upper left lobe (Image 2).

Trial of aspiration post twice broncho-alveolar lavage with 20 ml normal saline failed, and trial of 10% N acetyl cysteine administration (mucolytic agent diluted to 1/2) also failed. Flexible bronchoscope was removed and a 6 mm diameter - 30 mm length rigid bronchoscope was introduced for removal trial. Removal of small part of the bronchial cast was done. Reintroduction of the flexible bronchoscope within the rigid bronchoscope along with large clamp and removal of the complete cast was done (Image 3).

Image 3

Ceftriaxone was continued and Vancomycin was stopped, and a follow up chest X-ray showed major amelioration of the lung aeration and of the consolidation collapse (Image 4).

Broncho-alveolar lavage showed pleiocytosis with white blood cells of 174/ul consisting mainly of neutrophils (51%) followed

Image 4

by monocytes and macrophages (33%) and lymphocytes (16%). Lavage culture turned out to be sterile.

48 hours after the procedure, patient showed clinical amelioration and significant improvement, and he was discharged on oral antibiotic to continue the course of 10 days, with asthma and allergic rhinitis medications pending ulterior spirometry.

Discussion

We reported in this article one case of bronchial cast diagnosed with pneumonia rebel to treatment until endoscopy was done with removal of the cast and net amelioration was found on follow up.

Bronchial cast or plastic bronchitis is a rare pulmonary condition that refers to the blockage of distal or intermediate airway by the formation of branching intra-bronchial cast. The blockage can be partial or complete [1].

The symptoms consist of productive cough, dyspnea, fever, pleuritic pain, and even respiratory failure in rare cases. It can also present similarly to other chronic pulmonary diseases such as asthma, cystic fibrosis, or bronchiectasis.

It is more prevalent in pediatric patient than adults with most causes of plastic bronchitis are more common in males, except for allergic causes, which has a female predominance [2].

As for diagnosis, there is no specific lab test nor diagnostic criteria. It should be diagnosed based on clinical presentation, in addition to imaging investigations and by endoscopy.

By physical examination, usually we can find basal decrease air entry with wheezes [3] associated sometimes with history of cast expectoration, which might persist for years before diagnosis. In addition, on chest X-ray we can find bronchial obstruction with lobar atelectasis [4].

The pathway behind cast formation is still unknown but casts are usually divided into 2 types: type I and type II.

Type I is an inflammatory cast which consists of neutrophils, eosinophils, or Charcot Leyden crystals (as seen in cystic fibrosis and asthma). It is supposed to be due to decreased muco-ciliary clearance [5]. However, type II casts are usually acellular but composed mainly of proteinaceous lymph material, and are associated to post cardiac surgeries mainly Fontan procedure due to insufficient lymphatic drainage.

Secondary to cast formation and to airway obstruction, we have an increase in the pulmonary venous pressure, which further propagates cast formation [5].

Despite the absence of clear explanation of the pathophysiology of cast formation, the treatment has evolved well since it started on 1960 with manual removal by Johson., *et al*. [5]. Treatment nowadays consists of even pharmacologic treatment or surgical removal.

While type II cast usually requires treatment of underlying causes for prevention, type I usually responds better to pharmacological treatment [3]. Inhaled or systemic steroids, as anti-inflammatory agents, are usually effective in reducing inflammatory cast formation and reduce symptoms in pediatric and adult patients [6] but have no significant role in decreasing plastic bronchitis complications and mortality. If steroids are ineffective or contraindicated, other anti-inflammatory agents can be used, such as macrolide [7] or immunosuppressants like Sirolimus, by the reduction of chyle formation, which plays a role in cast formation [8]. The patients with infectious cast confirmed by positive cultures, antimicrobial therapy is also added to the treatment strategy [3].

In addition, other pharmacologic treatment consists of using N acetyl cysteine, a mucolytic agent, as it is safe and can be used as inhaled agent prior to bronchoscopy [9]. N acetyl cysteine has

showed superiority over other agents, such as DNAse (enzyme used for DNA degradation in cystic fibrosis), and plasminogen activators, which can be used in cast with fibrin formation [3].

If pharmacologic treatment was insufficient, surgical therapy has emerged also as effective treatment. Surgical option can even remove the existing cast or can also be used as preventive therapy by lymphatic ductal intervention. Finally, Bronchial cast removal via flexible bronchoscopy has elicited successful outcomes in patients with plastic bronchitis. It can be used after respiratory kinesiotherapy to facilitate cast removal and can be coupled to cryotherapy [10].

Conclusion

Plastic bronchitis is a rare respiratory condition which affect all ages even more common in pediatric group. Causes differ from cardiac to infectious and may be associated to diseases like sickle cell disease and cystic fibrosis. The symptoms vary also from simple productive cough to dyspnea with fever and even respiratory distress. Due to the different etiologies, we have different managements, ranging from pharmacological treatment, to bronchoscopy, or even to surgery. Till now, there is no clear explanation how the cast is created and even if it is rare, we should keep it in our differential diagnosis to prevent complications including cardio-pulmonary compromise.

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