

A Case Report of Recurrent Type 1 Cast Bronchitis in a Child with Asthma and Literature Review

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Abstract

Background: Cast bronchitis, also known as plastic bronchitis (PB), is an uncommon juvenile pulmonary illness characterized by the formation of branching bronchial casts that partially or completely obstruct the bronchial lumen.

Objective: To present a rare case of recurrent type 1 cast bronchitis in a child with asthma.

Methodology: A retrospective chart review of a patient recurrent type 1 cast bronchitis in a child with Asthma and literature review.

Case Presentation: A 13-year-old boy with persistent respiratory symptoms, including cough, shortness of breath, and exercise intolerance, was frequently hospitalized due to bronchopneumonia. Despite treatment for asthma and cat allergy, his condition showed only partial improvement. At 12 years of age, his symptoms rapidly progressed, prompting hospitalization. Tests revealed elevated eosinophils and a Class 4 RAST result for cat epithelium. Imaging showed obstructive collapse of the right lower lobe. Bronchoscopy identified a cast in the right upper lobe, which was partially extracted. Pathology revealed inflammation, fibrin, and eosinophils. Due to residual casts and inconclusive results, the patient was readmitted a month later, and a second bronchoscopy revealed additional casts, which were again removed. Pathology showed similar findings. After treatment with oral steroids and continued inhalers, the patient improved significantly, although some mucus plugging persisted in the left lung. So, the patient started on benralizumab and showed a good improvement.

Conclusion: Cast bronchitis is a rare, potentially lethal illness in children that requires a high index of suspicion for diagnosis and treatment. While bronchoscopic removal of bronchial casts and medication are primary therapeutic options, patients with recurrent cast formation are at high risk for surgical intervention, such as lobectomy or pneumonectomy.

Keywords: Type 1 Cast Bronchitis; Child; Asthma; Lobectomy; Pneumonectomy; Plastic Bronchitis (PB)

Background

A rare condition known as plastic bronchitis is characterized by the development of cohesive, branched casts that partially or totally obstruct the bronchial tree. Clinical manifestations of PB typically include fever, coughing, dyspnea, wheezing, chest pain, and symptoms of lower respiratory tract infections, during which patients may expectorate the casts. Because the casts move during inspiration and expiration, patients occasionally make a flag-flapping sound (bruit de drapeau) [1].

Based on the components of the casts that form, there are two forms of cast bronchitis: Type 1 is characterized by the formation of highly cellular inflammatory casts, primarily eosinophilic cells, and is typically associated with diseases like asthma, cystic fibrosis, non-cystic fibrosis bronchiectasis, sickle cell disease, and smoke inhalation [2]. Type 2, by contrast, consists of acellular materials made of fibrin and mucin and is typically linked to the post-surgical correction of congenital heart diseases.

A specific variant of cast bronchitis, eosinophilic cast bronchitis, is primarily observed in children and is not well-represented in the literature at this time. It is possible that some people have a medical history of atopy or asthma, although a considerable percentage may not. Cast bronchitis requires a high index of suspicion for diagnosis in order to successfully manage it and prevent future recurrences because the clinical presentation can range from moderate symptoms to total lung collapse that mimics the symptoms of foreign body aspiration.

We describe the first reported case of eosinophilic cast bronchitis in Saudi Arabia and Gulf Cooperation Council countries. Eosinophilic cast bronchitis was diagnosed after examination and investigations, including histological results. The case's treatment and outcome are described.

Case Presentation

We describe the case of a 13-year-old boy who was referred to our institution with recurrent episodes of bronchopneumonia and respiratory symptoms persisting for one year. He sought medical advice multiple times and required hospital admissions. Acute bronchial asthma and cat allergy were identified in the child, and treatment with bronchodilators and inhaled steroids resulted in only partial improvement.

He was admitted to the hospital for intravenous (IV) antibiotics at the age of 12 when he started to have respiratory symptoms in the form of a persistent cough, shortness of breath, and exercise intolerance, sometimes accompanied by fever that necessitated hospitalization for antibiotic treatment.

Initially, his physical examination showed a temperature of 36.8°C, a respiratory rate of 20 breaths/min, a heart rate of 99 beats/min, and a blood pressure of 104/71. Oxygen saturation was 97% in room air. His height and weight were within the median range. An examination of the right side of the chest revealed decreased air entry.

His blood work, including Complete Blood Count with Differential (CBCD), renal profile, hepatic profile, thyroid function test, immunological work-up, inflammatory markers, and allergy testing, were within normal parameters except for high eosinophils and a Class 4 Radioallergosorbent test (RAST) for cat epithelium E1.

Pulmonary function test (PFT) with pre- and post-bronchodilator spirometry showed a mixed obstructive and restrictive pattern with a partial response post-bronchodilator. The results were as follows: pre-bronchodilator: (FVC: 61%, FEV1: 51%, FEV1/FVC: 75%, FEF25-75%: 33%); post-bronchodilator: (FVC: 65%, FEV1: 56%, FEV1/FVC: 77%, FEF25-75%: 41%).

A chest x-ray showed right lower zone opacity with ipsilateral mediastinal shift (Figure 1a and 1b). A CT scan of the chest showed right lower lobe collapse with a small to moderate-sized intraluminal filling defect after the tracheal bifurcation, prompting our intervention with a flexible bronchoscopy (Figure 1c and 1d).

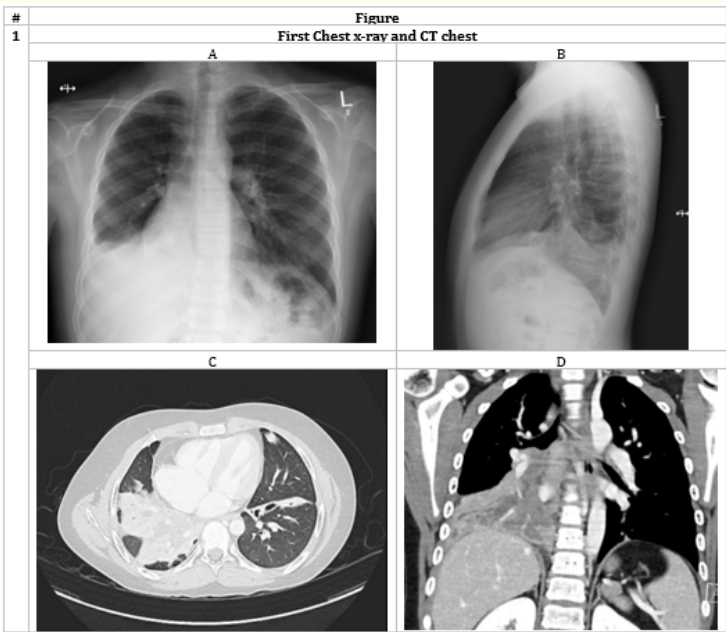


Figure 1: 1a and 1b: Frontal and Lateral view chest x-ray showing Right lower zone opacity is seen with ipsilateral mediastinal shift. 1c and 1d: Axial and coronal view mediastinal and lung window of CT showing Right lower lobe collapse with small to moderate sized intraluminal filling defect in the right lower lobe bronchus.

The bronchoscopy exam showed a gray/yellowish pale protruding cast at the junction of the right upper lobe with the bronchus intermedius. Using a size 1.1 cryoprobe, extraction of the cast in small pieces was performed, yielding variable tissue bulks ranging from less than 0.5 cm in diameter to a maximum of 1.2 cm in length. These appeared to be gray, light brownish, fleshy, and cheesy-like tissue in the endoscopic view (Figure 2a). The collected samples were sent for pathological study (Figure 2b). The patient was discharged home in good condition on Long-Acting Beta Agonists (LABA) inhalers BID and Albuterol PRN.

On follow-up, he mentioned that his symptoms had improved, and we updated him and his family with the new lab results. The pathological report was positive for fibrin, necrosis, and marked acute inflammation with eosinophils (Figure 2c).

He was admitted again, his Physical examination showed a temperature of 36.9c, respiratory rate of 22 breaths/min, heart rate of 97 beats/min, and blood pressure of 97/68. Oxygen saturation was 96% in room air. His height and weight were within the median range. An examination of the right side of the chest revealed decreased air entry. His CBCD showed high eosinophiles.

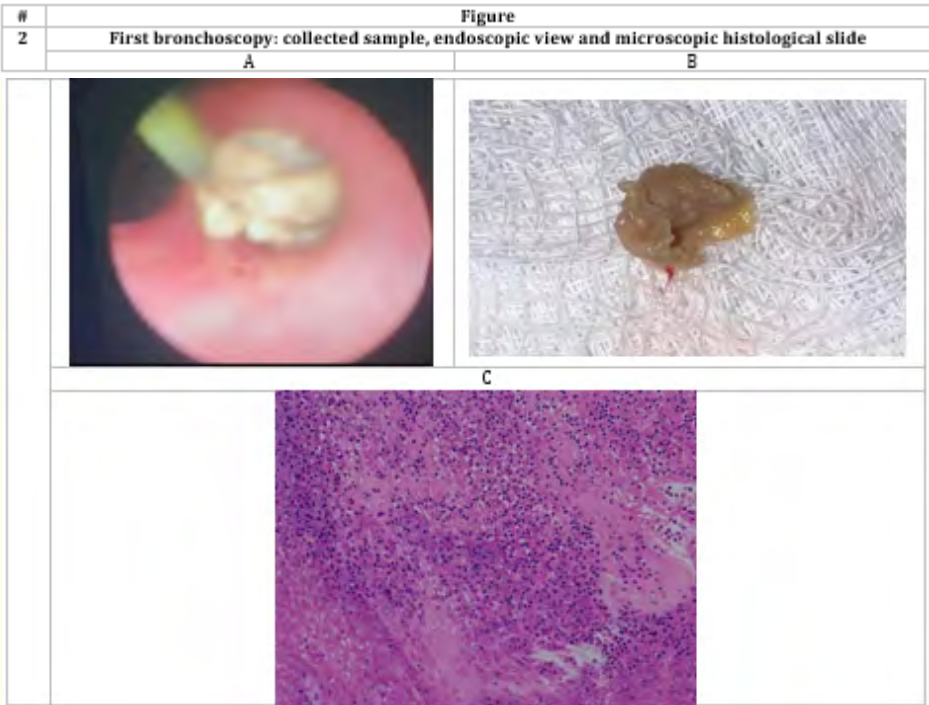


Figure 2: 2a: Showing a sample of the collected tissue from the debulked endobronchial lesion by flexible bronchoscope. 2b: Showing a removal of the intraluminal tissue by basket during flexible bronchoscopy procedure. 2c: The microscopic appearance of the cast which is showing fibrin, necrosis, marked acute inflammation with eosinophils. Hematoxylin-eosin stain.

PFT (spirometry) done for the second time which showed a moderate restrictive pattern with no evidence of obstructive. The results as following: FVC: 58%, FEV1: 61%, FEV1/FVC: 93%, FEF25-75%: 88%.

Chest x-ray Interval slight improvement of the upper pole right lower lobe collapse with persistent residual segmental collapse (Figure 3a and 3b).

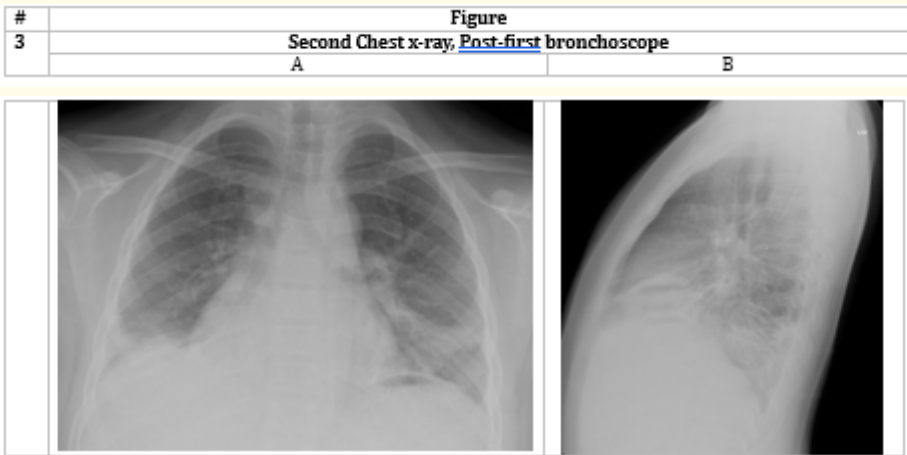


Figure 3: 3a and 3b: Frontal and Lateral view chest x-ray showing Redemonstration of collapse right lower lobe with mild atel-ectasis seen within the right middle lobe. Interval slight progression of the patchy left lower lobe air space opacity/atelectasis.

Second bronchoscopy was considered which revealed again yellowish cheesy casts attached to the mucosal wall at the takeoff of the right upper lobe and bronchus intermedius. cryoprobe size 1.7 was used to remove these, Series of extraction attempts using cryoprobe with variable tissue obtained ranging from 0.3 cm to maximum of 3 cm in length; all soft, fleshy, brownish in appearance with minimal bleeding at the site (Figure 4a). At the end three sets of bronchoalveolar lavage from the left lower lobe which sent for laboratory test. He tolerated the procedure well and the collected samples were (Figure 4b) sent for Pathological study.

BAL Lab results came and showed: presence of 7% of eosinophils in Fluid cell count, all others tests were normal. The second pathological report was positive for necrosis, fibrin and mixed acute and chronic inflammation, no viable tissue identified, no foreign body material identified (Figure 4c). The patient discharge home with a good condition on Oral steroid for 2 weeks, LABA inhalers BID and Albuterol PRN.

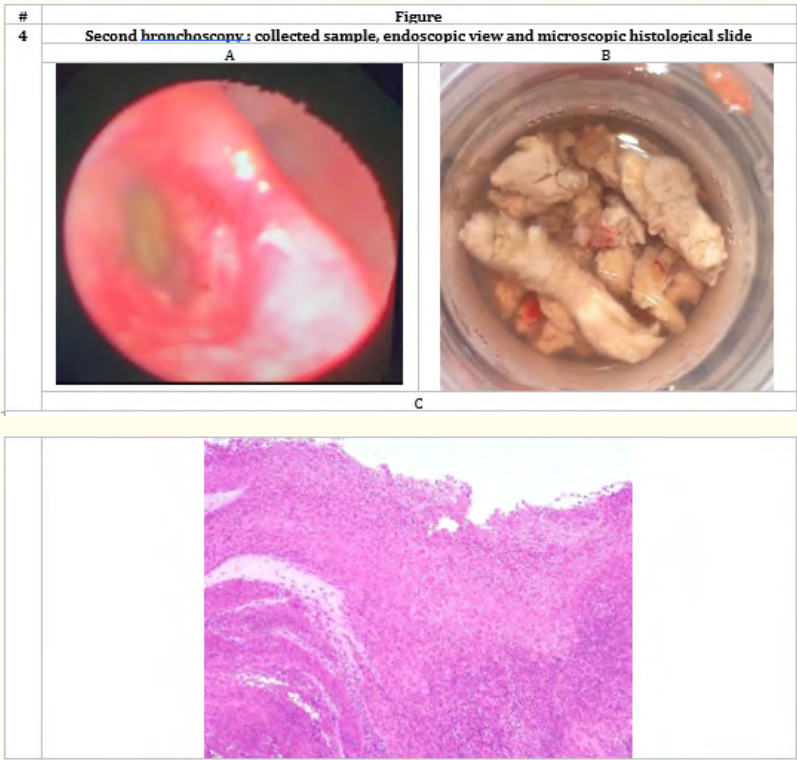


Figure 4: 4a: Showing a sample of the collected tissue from the debulked endobronchial lesion by flexible bronchoscope in the second bronchoscopy procedure. 4b: Showing residual of intraluminal tissue in the medial segment of the right lower lobe. 4c: The microscopic appearance of the collected cast from the second bronchoscopy procedure which is showing fibrin, necrosis, marked acute inflammation with eosinophils. Hematoxylin-eosin stain.

Two weeks later, he had completed the oral steroid course and he had good improvement with resolution of his respiratory symptoms and exercise intolerance.

The second CT chest showed: Interval resolution of right lower and mid bronchial tree endobronchial filling defects, right lower lobe consolidation with reversed halo sign identified, Interval development of left upper lobe and lingular mucus plugging/inspissated secretions (similar appearing to a prior CT filling defects) (Figure 5a and 5b).

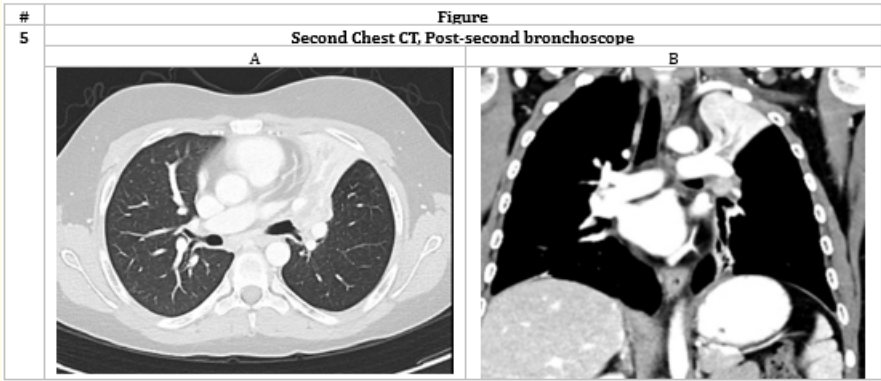


Figure 5: 5a and 5b: Frontal and lateral view chest x-ray showing Interval resolution of right lower and mid bronchial tree endobronchial filling defects, Interval development of left upper lobe and lingular mucus plugging/inspissated secretions.

The patient was reviewed in the clinic after 6 months and reported a slight worsening of symptoms. Chest X-ray was showed Linear atelectasis-scarring, left perihilar hazy opacities (Figure 6a and 6b). Laboratory evaluation revealed an eosinophil count of 8.3%. Bronchoscopy was performed, which showed inflamed bronchial mucosa. Bronchial washings were carried out from both right and left subsegments to clear thick purulent secretions.

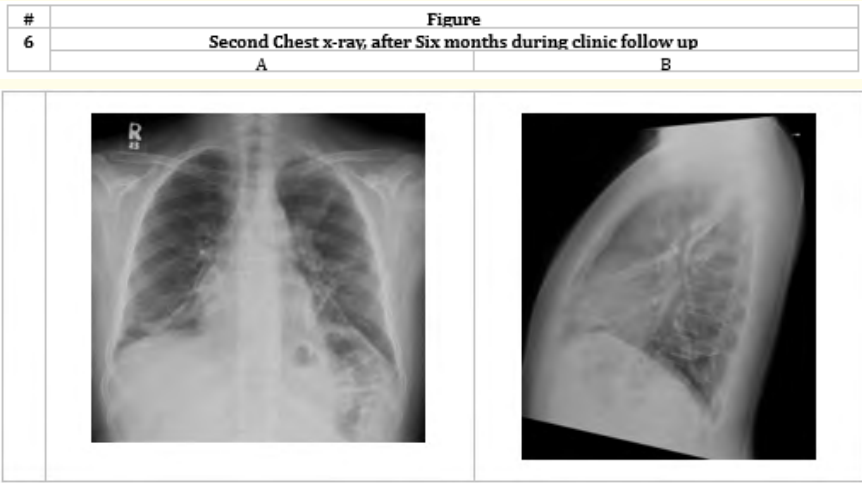


Figure 6: 6a and 6b: Frontal and lateral view chest x-ray showing A cardiomediastinal silhouette is comparable and unchanged like our loop. Linear atelectasis-scarring, left perihilar hazy opacities, no pleural effusion or pneumothorax.

Bronchoalveolar lavage (BAL) analysis differential cell count: neutrophils 12%, lymphocytes 42%, and monocytes 3%. Microbiological culture was negative.

The patient was subsequently initiated on benralizumab, with the first dose administered in the hospital. He reports improvement in his symptoms after 3 weeks after first dose, will continue following him up in clinic.

Discussion

Bronchial casts are categorized as type I when composed of inflammatory cells and accompanying disorders, such as asthma and pneumonia, and type II when acellular and related to congenital heart abnormalities following operations, such as Fontan procedure [3].

Plastic bronchitis and cast formation have a variety of causes. This includes lymphatic abnormalities, infections, hematological diseases, atopic diseases, cancer, and other ailments. Improper lymphatic drainage may result in casts associated with congenital heart disease. The genesis of the cast in patients with asthma is thought to be persistent inflammation and cellular infiltration with neutrophils and eosinophils, which is consistent with our case.[4].

In pediatric patients with type 1 cast bronchitis, both inhaled and systemic corticosteroids have been demonstrated to be effective in reducing inflammatory cast development and managing cast bronchitis symptoms. Long-term usage of low-dose corticosteroids and macrolides has shown a favorable anti-inflammatory impact in reducing cast formation, particularly in type 1 cast bronchitis [5].

In addition to multiple drug regimens, flexible or rigid bronchoscopic removal of the casts has emerged as the gold standard for treating plastic bronchitis since it can both diagnose the underlying cause and relieve symptoms [6].

Treatment of the underlying condition is useful in preventing the production of new casts. Lymphatic embolization, thoracic duct ligation, and thoracic duct stent grafting may be successful in type 2 cast bronchitis, however in rare cases of type 1 cast bronchitis not responding to appropriate medicinal treatment may require a surgical intervention such as lobectomy [7].

Conclusion

Cast bronchitis is a rare, potentially lethal illness in children that requires a high index of suspicion for diagnosis and treatment. While bronchoscopic removal of bronchial casts and medication are primary therapeutic options, patients with recurrent cast formation are at high risk for surgical intervention, such as lobectomy or pneumonectomy.

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