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Andressa Oliveira Peixoto[1,2]*, José Dirceu Ribeiro[1,2], Rafael Miranda da Costa[3] and Fernando Augusto Lima Marson[1,2,4]*

[1]Department of Pediatrics, School of Medical Sciences, State University of Campinas, Cidade Universitária Zeferino Vaz, Barão Geraldo, Zip Code: 13083-887, Campinas, São Paulo, Brazil
[2]Centre for Research in Pediatrics (CIPED), School of Medical Sciences, State University of Campinas, Cidade Universitária Zeferino Vaz, Barão Geraldo, Zip Code: 13083-887, Campinas, São Paulo, Brazil
[3]Clinical Hospital of Sumaré, State University of Campinas, Av. da Amizade, 2,400, Jd. Bela Vista, Zip Code: 13175-49, Sumaré, São Paulo, Brazil
[4]Department of Medical Genetics, School of Medical Sciences, State University of Campinas, Cidade Universitária Zeferino Vaz, Barão Geraldo, Zip Code: 13083-887, Campinas, São Paulo, Brazil

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***Corresponding author:** Andressa O Peixoto, Department of Pediatrics, School of Medical Sciences, State University of Campinas, Tessália Vieira de Camargo, 126, Cidade Universitária Zeferino Vaz, Zip Code: 13083-887, Campinas, SP, Brazil, Tel: +55 19 35218983; Fax: +55 19 35218964; E-mail: andressa_op@hotmail.com

Fernando AL Marson, Department of Medical Genetics, School of Medical Sciences, State University of Campinas, Tessália Vieira de Camargo, 126, Cidade Universitária Zeferino Vaz, Zip Code: 13083-887, Campinas, SP, Brazil, E-mail: fernandolimamarson@hotmail.com

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Case Report

Plastic Bronchitis: A Case Report

Abstract

Plastic bronchitis is a rare disease characterized by the formation of bronchial casts that can cause partial or complete obstruction of the airway. In this case report, a patient aged four years, previously healthy, began a chronic cough and underwent repeated courses of antibiotic therapy, inhaled corticosteroids and imaging tests; until there was a spontaneous sputum of a bronchial tree-shaped cast with approximately five cm, making possible the diagnosis of plastic bronchitis, which in our case report has idiopathic etiology. Due to the rarity of the pathology, we believe in the importance of the case report that can help to: (i) better understand the clinical presentation of plastic bronchitis; (ii) understand the difficulty in diagnosing the disease and distinguishing associated pathologies; (iii) disseminate the disease to health professionals; (iv) better understand the available therapies; (v) evaluate the efficacy of the disease treatment.

Introduction

Plastic bronchitis is a rare disease characterized by the formation of extensive bronchial casts, which can be gelatinous or rigid. Bronchial casts represent the exact format of the airway from which they are removed by sputum or bronchoscopy, and they tend to be large and more cohesive than mucous plugs [1-11].

Plastic bronchitis can be commonly associated with cyanotic congenital heart diseases, secondary acute chest syndrome to sickle cell disease and diffuse hypersecretory bronchial disorders, such as asthma, cystic fibrosis, bronchopulmonary aspergillosis, bacterial or viral respiratory infections, in addition to reports on cases of heart failure and pericardial effusion [1-11].

Clinical observation

In our case report, a female patient, aged four years and six months, from the state of Minas Gerais, Brazil, was evaluated. The patient was healthy until the age of four years and three months, when she began a productive cough, accompanied by fever and wheezing, with no dyspnea. At that time, the presence of opacity in the left hemithorax was identified by the simple chest radiography and bronchopneumonia was diagnosed. The treatment of bronchopneumonia was held with the use of amoxicillin for 14 days, associated with N-acetylcysteine [$C_5H_9NO_3S$]. After the treatment, there was partial improvement of the radiological image.

The patient remained symptom free during the two-week period after the treatment, when evolved, with ventilatory-dependent chest pain, with greater intensity in precordial region, associated with coughing and wheezing, but remaining afebrile. At that time, new chest X-rays showed left lobar atelectasis. Due to the clinical pattern, the patient was hospitalized to perform respiratory physical therapy, inhaled therapy with short-acting β_2 agonist and antibiotic therapy for 12 days. Procedures were held at another hospital.

There was no radiological improvement with the interventions described, and bronchoscopy was indicated. The result of the first bronchoscopy showed no abnormalities. However, due to the present and constant radiological alteration, new bronchoscopy was indicated within 72 hours. In the second procedure, the presence of bronchial cast was evidenced, composed of amorphous, necrotic material and fibrinopurulent serous exudate. After the second bronchoscopy,

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the elimination of abundant secretion and bronchial cast (approximately five cm in length) were identified. Thereafter, the patient began to eliminate bronchial tree-shaped material after a cough crisis for a period of approximately one year.

Considering the possibility of plastic bronchitis diagnosis, the patient was referred to a Tertiary Hospital, for investigation, which included the analysis of underlying diseases.

In the Tertiary Hospital, a High-Resolution Computed Tomography of the chest was held, which showed:

(i) Total atelectasis of the left upper lobe, with volumetric reduction of the lung

(ii) Deviation of mediastinal structures to the left

(iii) Hilar prominence of difficult characterization to the left. For this reason, the radiology team indicated an exam with use of contrast.

The patient was submitted to rigid bronchoscopy in which bronchial casts or anatomical changes to lung segments on the left and right were not observed. However, it was identified high amount of whitish and thick secretion on the left. In the collection of bronchoalveolar lavage, the *Staphylococcus aureus* sensitive to oxacillin was identified in the routine diagnostic culture. In addition to the bacterial examination, analyses were performed for fungi and mycobacteria culture, and all the tests were negative.

After two months, a High-Resolution Computed Tomography of the chest with contrast was held. In the exam, atelectasis of the anterior segment of the left upper lobe and lingula was identified in the pulmonary parenchyma and was associated with bronchiectasis and parenchymal bands.

Since clinical symptoms persisted, the investigation of differential diagnosis or diseases known to underlie plastic bronchitis proceeded, which included the diagnostic analysis for:

I. Cystic fibrosis (determination of chloride ion concentration by the sweat test)

II. Allergic asthma [determination of immunoglobulin E (IgE), eosinophils and lung function performed by spirometry with bronchodilator therapy]

III. Tuberculosis (assessment by Mantoux test)

IV. Cardiopathy (assessed by echocardiography). However, there was normality in the examinations held for the diagnosis of underlying diseases.

In addition to the courses of antibiotic therapy that the patient received during the evolution of the disease, the use of long-acting inhaled corticosteroid and bronchodilator was indicated, due to the apparent possibility of asthma. A year after the procedures were held, the patient stopped presenting cough with mucus, and evolved without symptoms, although the etiology of plastic bronchitis has not been elucidated in the case.

Five years after the first scan, a new image exam was held, and it was noted:

(i) Pulmonary parenchyma with no changes

(ii) topography of the left lower lobe and lingula with discreet dilatation

(iii) Sparse bronchiectasis to the left. In addition, the evidence of pulmonary function remained within the normal range.

After the procedures, the patient continues to be accompanied at the Tertiary Hospital with no further complications and need for drug therapy.

The summary of the tests and its results are described in figure 1.

Discussion

In the scientific literature, the prevalence of plastic bronchitis is unknown, arising mainly from the misdiagnosis in most patients [12,13]. In addition, the knowledge of the disease is based primarily on case reports [3,6-8,12,14-23] and the diagnosis of plastic bronchitis is often held during an autopsy after death by respiratory failure [13]. The disease affects all age groups, with a predominance of cases in women [12], a fact not yet clarified in the literature.

The main characteristic of plastic bronchitis is the formation and expectoration of bronchial casts, which can vary in sizes, from small segments up to larger sizes capable of obstructing the airway of an entire lung. Bronchial casts consist of varying proportions of fibrin, mucin and cellular material. The sputum of bronchial casts varies in frequency and duration, and may occur hemoptysis [12].

In some cases of plastic bronchitis occurs spontaneous elimination of bronchial cast, which can complicate and delay the diagnosis. However, in our case report, the patient showed spontaneous elimination of bronchial cast that lasted for about a year.

The clinical pattern of the disease presents wide variability. However, chronic cough and dyspnea are usually observed, being wheezing the most common finding of physical examination. Radiological examination often identifies the location of the bronchial impaction, with atelectasis or infiltrators, predominantly in lower lobes [12]. Plastic bronchitis can present mild medical conditions with clinical recovery, as in our case report. However, in severe cases, airway obstruction and severe atelectasis can occur, leading to respiratory failure [12,13].

The High-Resolution Computed Tomography allow, in some cases, the visualization of bronchial casts in the airways of larger caliber. However, the diagnosis is usually confirmed by bronchoscopy for demonstrating the airway obstruction by bronchial cast [12].

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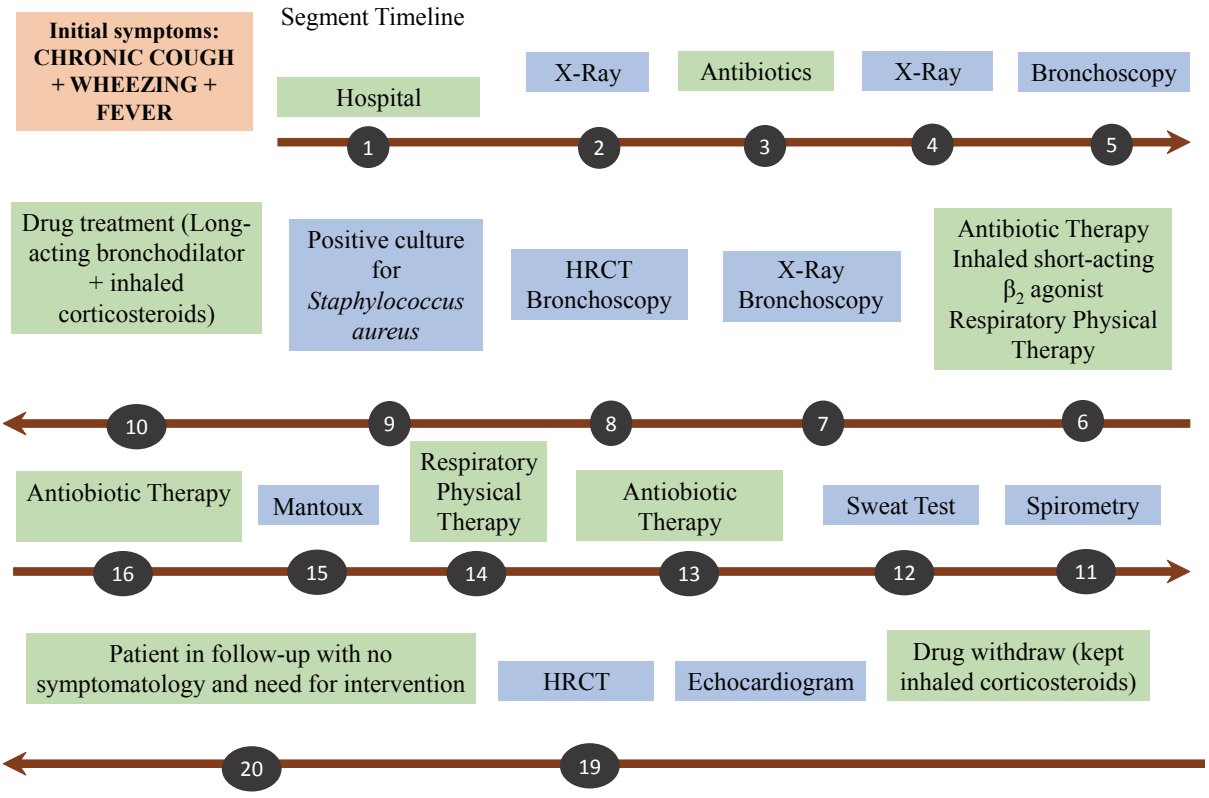

Figure 1: Chronological order in the case report with plastic bronchitis diagnosis segment. Procedures held in the patient segment are presented in blue. Those held on the conduct directed by the tests obtained in the patient segment are presented in green. HRCT, High-Resolution Computed Tomography. Numbers represent the chronological order of the events held in monitoring the patient.

Plastic bronchitis classification in literature is still controversial and previous studies were based on a series of cases, presenting methodological limitations [2,23,24]. Therefore, classifications are based on histological type and associated diseases. Namely:

a) histological classification:

(i) Type 1 – inflammatory (cellular)

(ii) Type 2 (acellular)

b) Classification by underlying disease to plastic bronchitis:

(i) Allergy/Asthma

(ii) Cardiac

(iii) Idiopathic [2,23,24].

Interestingly, on the histological classification, type 1 casts are associated with, primarily, the pathologies affecting the bronchial tubes (i.e., asthma and allergy); whereas type 2 casts are associated with congenital cyanotic heart disease and idiopathic plastic bronchitis cases. The ratings present therapeutic value or limited prognosis [13].

Diseases probably associated with plastic bronchitis are briefly presented in table 1. However, cardiac changes and pulmonary interaction that result in plastic bronchitis are not well known, but they can be associated with abnormalities in the tissue factor [13]. Moreover, it should be considered that, recently, plastic bronchitis can be triggered by common respiratory infections and cause atelectasis, even in healthy children [6].

The assessment of treatment response in plastic bronchitis is restricted, considering that the disease is rare and the patients received different medications, being difficult to determine which therapy was effective [13].

The treatment of plastic bronchitis includes acute therapy to assist the removal and expectoration of bronchial casts and short or long-term treatments that address the hypersecretory process [12]. However, if plastic bronchitis can be a complication of an underlying disease, the underlying condition must be treated to eliminate the formation of bronchial casts.

Drugs used for treating plastic bronchitis present low scientific evidence. In short, for the different drugs already used, we have the following conclusions, mainly, obtained in the consensus of the European Respiratory Society (ERS) (2013) [13] and in the international registry data of plastic bronchitis (Table 2) [12,13,15,16,25-27]:

I. Antibiotics: Plastic bronchitis is not associated with bacterial infection and, in general, antibiotics are not recommended in the treatment. However, low-dose macrolides can decrease the amount of mucin by inhibiting the production of extracellular signal-regulated kinases (ERKs). The low-dose macrolides can

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Table 1: Diseases probably associated with plastic bronchitis.

Probable	Congenital heart disease with Fontan physiology*
	Sickle cell disease with acute chest syndrome
	Pulmonary lymphatic abnormalities
	Pulmonary infection by Influenza A virus
	Toxic inhalation (chemicals or thermals)
Possible	Other congenital cyanotic heart diseases
	Non-pulmonary lymphatic abnormalities
	Hypersecretion and severe asthma (eosinophilic casts)
	Allergic bronchopulmonary aspergillosis
Improbable	Cystic fibrosis
	Chronic obstructive pulmonary disease
	Bronchiectasis
	Bacterial pneumonia

* associated with patients with a lower age. Adapted from Rubin BK, Moskowitz WB. 1st European Respiratory Society Handbook: Paediatric Respiratory Medicine. Plastic bronchitis. 2013; 577-81.

Table 2: Evidence of the use of different drugs in plastic bronchitis.

Good evidence	Airway debugging including physical therapy and devices such as High-Frequency Chest Compression vest.
	Aerosol heparin
	Inhalation of tissue plasminogen activator (tPA)
	Heart transplant
Anecdotal or based on a case study	Hypertonic saline
	Low-dose oral macrolide (clarithromycin or azithromycin)
	Oral or inhaled corticosteroids (only for eosinophilic casts)
	Ligation of the thoracic duct
	Fontan modifications (Fenestration or withdrawal)
No evidence	β-agonist aerosol
	Dornase alfa (Pulmozyme)
	Mucolytics such as N-acetylcysteine
	Expectorants such as guaifenesin
	Non-macrolide antibiotics

Adapted from Rubin BK, Moskowitz WB. 1st European Respiratory Society Handbook: Paediatric Respiratory Medicine. Plastic bronchitis. 2013; 577-81.

mitigate the severity of plastic bronchitis, similar to the use in cystic fibrosis and diffuse panbronchiolitis.

II. Expectorants: Expectorants, such as guaifenesin, hypertonic saline solution or mucolytics (N-acetylcysteine), may induce mucus secretion and/or increase the airway inflammation and must be used with caution, since the clinical benefit is still limited.

III. Short-acting β_2 agonists and inhaled corticosteroids: no apparent benefit.

IV. Dornase alpha: No apparent benefit, since bronchial cast does not contain polymeric DNA.

V. Inhalation of tissue plasminogen activator (tPA): Can ease symptoms through fibrin depolymerization. However, the drug presents a high cost and can cause airway irritability.

VI. Inhaled heparin: It acts by reducing mucin secretion and prevents the activation of the fibrin tissue factor, besides presenting anti-inflammatory action, has lower cost and causes less irritation than tPA.

VII. Additional Factors: In the diagnosis of plastic bronchitis, the routine with the daily use of a High-Frequency Chest Compression (HFCC) vest in patients with effective cough must be initiated, or the use of the Cough Assist device (Philips Respironics®, France) in patients with ineffective cough.

From the physiological and genetic point of view, plastic bronchitis is little understood and studies have been conducted to understand these factors and allow a better characterization of the pathology [13]. However, as evidenced by Rüegger et al. (2013), in monozygotic twins, the genetic contribution must be considered and, in the future, the genetics in response to the environmental factor may show the real etiology in the formation of the bronchial casts [6].

Conclusion

Plastic bronchitis is not part of the routine care, even in specialty centers for pneumology, being a challenge in clinical practice the diagnosis and follow-up of patients with the disease. This is a phenotype unknown by many doctors, even in the specialty of pneumology. Hence, case reports become important to gradually promote the understanding of the disease and make a proper management of the patient, with improvement in the quality of life. The management of the most severe cases of the disease, which can evolve with great worsening of pulmonary function, can be evaluated and carried out regarding the knowledge achieved in the previous published case reports.

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Contributorship

AOP, JDR, RMC and FALM made substantial contributions to data acquisition; were involved in drafting the manuscript and revising it critically for important intellectual content; gave final approval of the manuscript version to be published; and agreed to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work were appropriately investigated and resolved.

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