



## Paediatric plastic bronchitis in an atopic child; A case report from East Africa

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### ABSTRACT

Paediatric plastic bronchitis (PB) is a rare disease characterized by the presence or expectoration of branching airway casts usually in children with cardiac conditions and allergy. It is thought to be due to obstruction or altered drainage of the lymphatics in those with cardiac conditions. Obstruction can be diffuse and thus fatal if untreated. Less than 600 cases have been described in literature and just one in our region in a patient with sickle cell disease. We present a case of a 7-year-old female with acute symptoms suggestive of a lower respiratory tract infection and left hemithorax opacification and bronchial casts on bronchoscopy.

### 1. Introduction

Plastic bronchitis (PB) is a rare disease, also described in medical literature as fibrinous or pseudo-membranous bronchitis [1]. Described by Galen in the 2nd century (129–200 AD) in which branching casts are formed in the bronchial airways. He described patients that expectorated soft viscous casts that he interpreted as veins and arteries. 1500 years later, Morgagni identified the material as mucus casts originating from the airway rather than pulmonary veins [1].

Patients typically present with symptoms of a lower respiratory tract infection which can include dyspnea, shortness of breath, cough, chest pain, wheezing and occasionally fever. A less common clinical finding is a “flapping” sound like a flag “*bruit de drapau*”, made by the cast during inspiration and exhalation on auscultation and that can sometimes be sensed by patients [2]. It can occur at any age but more so in children with no noted gender predilection. A study on paediatric PB reported an institutional prevalence rate of 6.8 per 100,000 patients with a case fatality rate of 7% [3]. In Africa we do not have any epidemiology figures and the incidence is unknown, with only 1 published case in our region in a patient with sickle cell disease [4].

The exact mechanism with which casts form is not well known and is distinct from the expectoration seen in bronchiectasis from Cystic Fibrosis (CF), non-CF causes or mucoid impaction from asthma [5]. It can be classified based on the lymphangiographic findings or

histopathology [6,7].

Type I–PB associated with high mucin content and airway eosinophilia with breakdown products of eosinophils including Charcot-Leyden crystals as seen in asthma and Allergic Bronchopulmonary Aspergillosis (ABPA).

Type II–PB with abnormal pulmonary lymphatic circulation is seen in patients with congenital heart disease especially post-Fontan procedure where systemic venous flow is directed to the pulmonary arteries increasing blood flow to the lungs. This causes lymphatic drainage imbalance, increase in the pulmonary venous pressure inducing the formation of bronchial casts. Cardiac patients tend to have lymphocytic or monocellular casts with fibrinous content.

Diagnosis for both types is achieved by visualisation of expectorated or bronchoscopically removed bronchial casts. For Type II cases, Dynamic contrast magnetic resonance lymphangiography (DCMRL) a Magnetic Resonance Imaging (MRI) study of the lymphatic system is useful [8]. Injection of contrast dyes into the inguinal nodes with serial imaging of the flow through the thoraco-abdominal region to the thoracic duct to visualize the abnormal pulmonary lymphatic circulation.

### 2. Case presentation

A 7 year old girl presented with a 2 week history of a persistent dry

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cough, nasal congestion and mild rhinorrhea that progressed to acute onset left-sided chest pain and difficulty in breathing with a low-grade fever. She had no present or past history of wheeze, no easy fatigability or dyspnea on exertion, no body or feet swelling. No history of close contact with a patient known to have tuberculosis (TB) or severe acute respiratory syndrome coronavirus 2 disease (COVID-19) no loss of weight or poor growth, no loss of appetite nor night sweats reported. There was no known history of chest trauma.

At the onset of her symptoms she was initially managed at home as nasopharyngitis with a cough syrup with no resolution of symptoms. She was reviewed in the outpatient clinic with a diagnosis of pneumonia a few days later and standard antibiotics were prescribed. 3 days later she was reviewed by the paediatrician due to the persistent cough and difficulty in breathing. Inhaled salbutamol and oral montelukast were added as treatment for possible asthma. A further 3 days later she developed chest pain that necessitated an admission.

The birth and developmental history were unremarkable. She had no previous admission but had allergy symptoms since infancy which included eczema, history of skin eruptions with ingestion of animal proteins and allergic rhinitis with upper airway obstruction symptoms.

Her clinical examination on admission; temperature of 37.5 °C, blood pressure (BP) 98/53(mean BP 63), heart rate (HR) 105, oxygen saturation (SPO<sub>2</sub>) 93% on room air and increased respiratory rate of 33 breaths per minute.

The respiratory examination revealed tracheal shift to the left side, dullness on percussion and reduced air entry on the left lung zone all indicating left lung collapse (Fig. 1). Systemic examination was positive for orbital shiners, enlarged inferior turbinates and lingual tonsils hypertrophy, dental carries and lastly mild eczema patches on her trunk.

Initial blood gas analysis done showed compensatory respiratory alkalosis and hypoxia: (pH) 7.426, partial pressure of carbon dioxide (PCO<sub>2</sub>)25.4 mmHg, partial pressure of oxygen (PO<sub>2</sub>)55 mmHg, base excess (BE)-8mmol/L, serum bicarbonate (HCO<sub>3</sub>)16.7mmol/L, total carbon dioxide (TCO<sub>2</sub>)17 mmHg, oxygen saturation (SPO<sub>2</sub>)90% and lactate 0.65mmol/L. Normal values for Abbott® blood gas analysis machine i-STAT CG8+: pH (7.35–7.45), PaO<sub>2</sub> (80–105 mmHg), PaCO<sub>2</sub> (35–45 mmHg), BE (–2 to +3 mmol/L), HCO<sub>3</sub> (22–26 mmol/L), TCO<sub>2</sub> (23–27 mmHg), SPO<sub>2</sub> (95–98%), lactate 0.36–1.25mmol/L.

The patient was admitted to the high dependency unit (HDU) due to features of compensated respiratory failure for monitoring, oxygen supplementation, intravenous fluids and intravenous antibiotics.

A working diagnosis of features of lung collapse/consolidation, pneumonia and possible foreign body aspiration.



**Fig. 1.** Chest x-ray (CXR) before bronchoscopy showed right lung hyperinflation, total opacification of the left hemithorax with tracheal shift to the left suggestive of left lung collapse.

Initial investigations were as follows: Haemogram and serum biochemistry were normal, the inflammatory/infective markers C-reactive protein (CRP) and Pro-calcitonin (PCT) were low and blood culture was negative. Multiplex respiratory panel polymerase chain reaction test (PCR) was negative for viruses and atypical bacteria tested, tuberculosis microbiology and PCR was also negative. COVID-19 test was also negative. Vitamin D levels were low at 22.9 (30–100) U/L.

Based on her clinical and laboratory findings, an additional diagnosis of; upper airway obstruction, due to adenotonsillar hypertrophy, dental carries, eczema and Vitamin D deficiency was made. She was started on intravenous ceftriaxone and flucloxacillin, skin emollients and Vitamin D supplementation. By the second day she had increased work of breathing and was started on continuous positive airway pressure (CPAP) non-invasive ventilation by mask alternating with a non-rebreather high flow oxygen mask (NRM).

A multidisciplinary consult was sought that included the ear, nose and throat specialist (ENT), pulmonologist and the cardiothoracic surgeon. A plan was made for a computerized tomography (CT) scan of the chest, upper airway endoscopy and bronchoscopy with a possible open surgical approach if needed (Fig. 2).

Rigid bronchoscopy was done on day 3 of admission and removal of a small fat like consistency foreign body from the left main bronchus. It was noted that there was minimal mucus in the trachea and no features of major lower airway inflammation of the visualized segment. Grade 3 fibrotic tonsils and enlarged adenoids were confirmed.

Further history revealed that she had choked on chewing gum 2 months prior to presentation but had not experienced any significant respiratory symptoms warranting a review.

She was maintained on oxygen supplementation by NRM on that day, which was being weaned off gradually as tolerated and remained clinically and vitally stable.

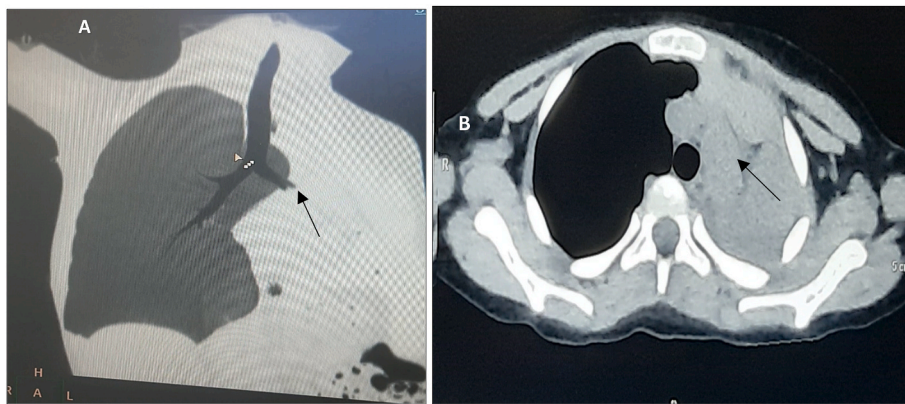
By the second day post-bronchoscopy, we managed to wean off oxygen to 1Litre/min via nasal prongs with no respiratory distress. However, there was still minimal air entry on the left upper and mid lung zones. A repeat CXR done 24 hours after the procedure showed a persistent lung collapse with no interval change. She was initiated on aggressive chest physiotherapy and incentive spirometry to recruit the lung for up to 3 days and continued on antimicrobials despite no sign of obvious infection, with no clinical improvement.

A case conference held with the multi-disciplinary team suggested a second CT scan or a repeat rigid and flexible bronchoscopy to investigate cause of the airway obstruction or endobronchial lesion with a possibility of a thoracotomy and open surgical approach.

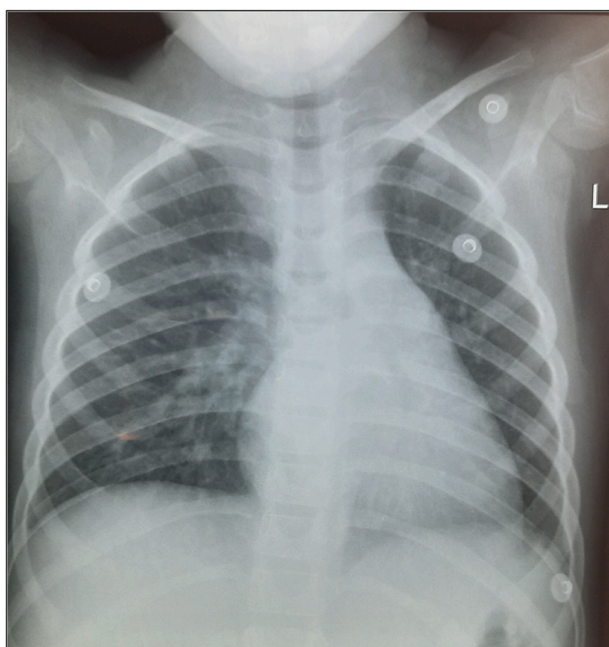
Flexible bronchoscopy done on day 8, found an endobronchial lesion completely occluding the left main bronchus slightly below the carina that could not be suctioned. The right bronchial tree was normal. Rigid bronchoscopy was done and we found a tenacious mass, soft to firm in consistency extending to the segmental branches with attendant bronchial wall inflammation throughout. Mucoïd discharge was noted from the left basal inferior segment. Multiple segments of thick mucoïd/lipoid like material and bronchial casts suggestive of PB were extracted by use of forceps through the rigid bronchoscope and eventual bronchial toilet to conclude the procedure.

Immediate post bronchoscopy period there was minimal oxygen requirements, no respiratory distress, marked improvement in air entry on the left lung zone and we managed to wean off oxygen within 24 hours (Fig. 3).

The team began a regime of combined inhaled corticosteroid with a long-acting beta-agonist (ICS/LABA) and a 3-day course of intravenous methylprednisolone, continuation of chest physiotherapy and incentive spirometry as a deep breathing exercise to improve her lung capacity after lung collapse, antimicrobials and other supportive management. She remained hemodynamically stable with no respiratory distress, vitally stable off oxygen, alert and ambulant. Cardiac sonography was done to also rule out complications from the upper airway obstruction and was normal.



**Fig. 2.** CT scan chest before bronchoscopy. (A) Coronal section shows a soft tissue density lesion obstructing the left main bronchus (arrow). (B) Resultant left lung collapse on the axial view (arrow).



**Fig. 3.** Chest x-ray 48 hours post-procedure shows normal bilateral aeration of the lung fields.

**2.1. Histopathology**

Microscopy showed a suppurative and mixed inflammatory cellular debris with mainly neutrophils and eosinophils. Necrotic debris and flakes of squamous epithelium were also noted. No foreign body

material could be discerned (Fig. 4).

Further tests for Immunoglobulin E (IgE) 0.53Ku/l (<90) and Aspergillus Fumigatus IgE were negative.

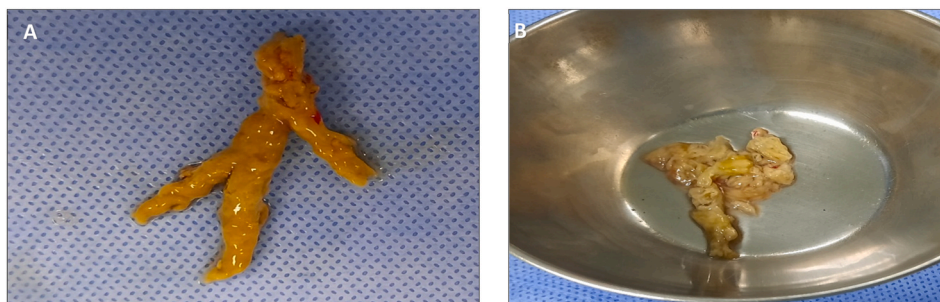
She was discharged on day 12 on the ICS/LABA and a plan to complete 3 more courses of monthly pulsed methylprednisolone with possibility of adeno-tonsillectomy if upper airway symptoms of obstruction persists. Post discharge she's doing well, no further symptom and normal chest exam and x-ray after the 3rd course of methylprednisolone, her dental carries were filled and no further upper or lower airway symptoms persist.

**3. Discussion**

The prevalence of PB is unknown with only an institutional prevalence reported [3], and it is likely that many patients go undiagnosed. PB affects patients of all age groups, the clinical presentation is similar to that of pneumonia or asthma with cough, difficulty in breathing chest pain or wheeze with or without fever but can present in respiratory failure if total atelectasis occurs [1,9]. Duration of symptoms is usually over a more chronic period.

Our initial diagnosis was foreign body aspiration with secondary atelectasis and pneumonia as she had symptoms of lung collapse that includes; difficulty in breathing, tracheal shift, ipsilateral dullness on percussion and reduced air entry with suggestive radiological findings. PB diagnosis was only entertained at the time of bronchoscopy. Current literature does not support foreign body aspiration as a cause but a mimic of PB. Suspicion of foreign body or radiologic suggestion of endobronchial occlusion should lead to diagnostic and therapeutic bronchoscopy [1,10].

In cases of asthma and allergies, mucoid impaction accompanied by other clinical symptoms like wheeze, chest tightness or heaviness can mimic or overlap PB with similar features [5,10]. It occurs due to



**Fig. 4.** (A) Gross histopathology of the specimens obtained on bronchoscopy. A tan coloured bronchial cast. (B) Tenacious mucopolipoid material removed by forceps within the main bronchus.

hypersecretion of the mucus glands in response to a trigger. This usually causes plugs affecting the upper lobes and responds to bronchodilators, mucolytics and airway clearance exercises unlike PB where the removal of the cast has immediate relief as in our patient [9].

Our patient did not have preceding symptoms of recurrent wheeze or chest tightness but more of upper airway symptoms and eczema putting her at risk of developing asthma. Furthermore, on flexible bronchoscopy the right bronchial tree had no signs of inflammation and her serum eosinophil count and total IgE were normal. Bronchial casts have been reported in a child preceding their asthma diagnosis [10,11].

The chest radiograph findings corresponded to our clinical findings suggestive of left lung collapse/consolidation, CT scan was sensitive enough to detect a luminal obstruction of the left main bronchus by a soft tissue density lesion and was useful in guiding the decision to perform bronchoscopy [12]. The evaluation of the cardiovascular system both clinically and with a cardiac sonogram was normal and neither did she have features or symptoms suggestive of sickle cell disease [4].

Histopathology of our case was more in keeping with type I inflammatory casts, seen mainly in patients with asthma or allergy [6,9]. The bronchial casts appear as pale, tan, or gray-green masses with varying consistencies [3,9,10]. The bronchial cast can be mistaken for a mucus plug, aspirated food or other foreign bodies [10]. Lack of an observed aspiration episode does not rule out an FB.

In view of the unilateral nature of the pathology and the normal right lung on flexible bronchoscopy, we postulate that the preceding aspiration of chewing gum a non-degradable foreign material and the respiratory infection may have triggered the inflammatory reaction and formation of the cast [13].

An initial rigid bronchoscopy by the ENT specialist is the common practice in our region to manage suspected foreign bodies as there is limited access to flexible bronchoscopy. Any complications are handled by the thoracic surgeon [1,14].

After successful removal of the PB our patient was prescribed both inhaled and systemic steroids as prevention of further development. This decision was based on evidence from just a few studies on management of type I PB which is seen in inflammatory and allergic conditions [7,9,15].

Methylprednisolone was used over a 3-month period and stopped at this time with no further events at the time of publication of this case report. Our institution did not have tissue plasminogen activator (TPA) so this was not considered as a treatment option, heparin a cheaper and alternative therapy is available but only useful in those with cardiac related PB [3,15].

PB although rarely reported in Africa does exist. It is hoped that the case report will sensitise clinicians to this diagnosis in atopic patients who present with unilateral total absence of breath sounds or features of lung collapse and a negative history of foreign body aspiration.

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## Author contributions

Anne Irungu: conceptualisation, acquisition of data, writing of the original draft, review and editing. Cynthia Achola: acquisition of data, writing of the original draft, review.

Adil Waris: acquisition of data, writing of the original draft, review and editing.

Mark Awori: acquisition of data, writing of the original draft, review and editing.

Barrack Ongulo: acquisition of data and review.

All authors were involved in the clinical management of the case, all read and approved the final manuscript.

## Declaration of competing interest

The authors declare no competing interest.

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