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PLASTIC BRONCHITIS: A RARE COMPLICATION FOLLOWING A MOTOR VEHICLE COLLISION

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ABSTRACT

Plastic bronchitis, more appropriately termed chyloptysis, is a rare and potentially fatal condition caused by chylous coating of the airways. These cast coating can dislodge and become an obstructive mass in the patient's airway, necessitating rapid intervention. PB is well described to occur following single ventricle physiology heart disease corrective procedures, particularly following Fontan procedures. It is less commonly seen in traumatic settings. We present the youngest known case of a traumatic injury induced plastic bronchitis. A 19-year-old man was involved in a motor vehicle accident with airbag deployment. The airbags struck him in the chest; however, the patient felt well at the time and did not seek medical attention. Several months later the patient began coughing up milky white masses identified as casts. He was initially diagnosed with asthma but did not respond to therapy. He ultimately was found to have evidence of thoracic duct injury. Options for therapy were discussed, including possible thoracic duct ligation. The patient opted to continue a low-fat diet and has remained cast free. This case highlights the importance of considering plastic bronchitis in patients with cast production and a history of trauma to the chest.

Keywords: plastic bronchitis, chyloptysis, casts, trauma, thoracic duct, lymphatic imaging

Plastic bronchitis (PB), more appropriately termed chyloptysis, is a rare, potentially fatal condition caused by formation of airway casts. PB is often diagnosed after expectoration of these branching airway casts created by chylous coating of the airways.

CASE REPORT:

We present an otherwise healthy 19-year-old man who presented with a 14-month history of a chronic productive cough with "lung casts". The patient has provided consent for publication and the institutional IRB board has approved publication. He first experienced coughing up casts a few months after a motor vehicle accident where airbags were deployed, striking him in the chest. He did not seek medical attention at the time. The patient had a productive cough which he thought was phlegm but had produced a milky cast tinged with blood. His cast production was initially infrequent. He established care with a pulmonologist, who diagnosed him with asthma, based on wheezing in his lung fields, and started him on a daily inhaled corticosteroids and

albuterol as needed. Computerized Tomography (CT) of his chest was obtained and only remarkable for two small pulmonary nodules. He then started coughing up casts with increasing frequency and in a cyclic nature, eventually developing an acute worsening of his cough with production of 4-5 casts per day. He also had wheezing and hypoxia with oxygen saturation to 86% at that time. He was evaluated by his primary care physician who prescribed him antibiotics and oral steroids. His cough did not improve, and a chest X-ray was later obtained. The patient was then diagnosed with community acquired pneumonia and antibiotics were switched to amoxicillin at that time. He has no history of congenital heart disease or prior diagnosis of asthma, nor did he have seasonal allergies nor other atopic conditions. Due to continuing productive cough and shortness of breath, he sought further evaluation and was ultimately admitted to the pediatric unit.

Throughout his hospital stay, the patient was able to maintain his oxygen saturation above 95% on room air and he had no signs of respiratory distress. Chest auscultation showed clear breath sounds bilaterally with no crackles or wheezes. Given suspicion of plastic bronchitis, he was immediately started on a low-fat diet. He was also treated with albuterol, inhaled acetylcysteine, and chest physiotherapy; however, no subsequent casts were produced during his admission. Bronchoscopy was performed with bronchoalveolar lavage (BAL). No casts were seen nor obtained to send for tissue pathology. Allergic Bronchopulmonary Aspergillosis work up including galactomannan, fungus culture from BAL, and aspergillus fumigatus IgE were negative. He did not have evidence of tuberculosis with negative AFB culture and QuantiFERON gold. Work up for respiratory infections including legionella and viral upper respiratory infection were negative. Coccidioides IgM and IgG serum were negative. Bipedal lower extremity lymphangioscintigraphy with SPECT-CT was obtained but the radiotracer did not uptake past the inguinal lymph nodes and the thoracic duct was not visualized. Due to the patient's stable clinical condition, he was discharged home with a plan for outpatient follow up with interventional radiology to obtain contrast enhanced magnetic resonance (MR) lymphangiogram.

At follow-up in the Interventional Radiology clinic two weeks after discharge, the patient did not have recurrences of expectorating casts while maintaining a low-fat diet. MR lymphangiogram was obtained and revealed evidence of extravasation of contrast surrounding the proximal descending aorta and left posterior paramedian chest wall *(Fig. 1)*. Findings were concerning for injury to the thoracic duct at the level of the proximal descending aorta with lymphatic extravasation into the left hemithorax. Options for conventional lymphangiogram with embolization as treatment was discussed versus the possibility of continuing indefinitely on a low-fat diet alone as treatment. Thus far, he has continued to do well on a low-fat diet.

DISCUSSION:

Plastic bronchitis (PB) is a rare, potentially fatal condition caused by formation of airway casts due to chyle emptying into the airways. PB often presents after expectoration of branching airway casts known as chyloptysis. Other symptoms include cough, fever, shortness of breath, wheezing, or persistent pulmonary obstruction (1). Two types of casts have been described under Seear classification. Type I is classified as inflammatory casts that are made up of fibrin and dense inflammatory infiltrate are usually associated with inflammatory diseases of the lungs such as asthma, pulmonary infections, cystic fibrosis, or sickle cell acute chest syndrome (2-4). Type II is classified as acellular casts formed by mucin is most often associated with congenital heart disease with single ventricle physiology that has been surgically palliated with the Fontan procedure (5). Type 1 inflammatory casts often have an acute presentation while Type II acellular casts often have chronic or recurrent presentation (6).

The underlying mechanism of cast formation is unknown, but it is thought to occur from primary or secondary pulmonary

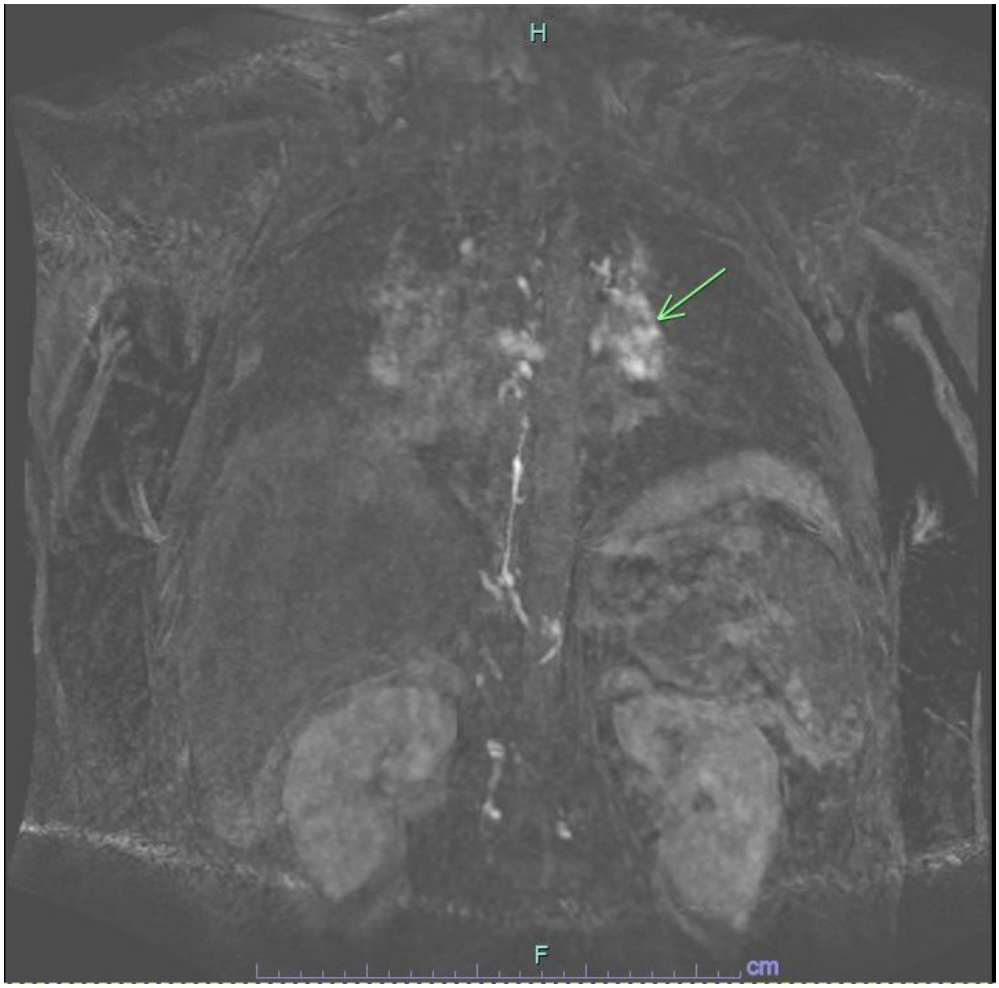

Fig. 1. *MR lymphangiogram image of the chest demonstrating extravasation of tracer (arrow) into the left hemithorax at the level of the descending aorta.*

lymphatic abnormalities (7). Cases of PB as a complication after Fontan procedure are thought to be due to hemodynamic alterations contributing to a break in mucosal integrity and injury to the alveolar-capillary barrier or operative trauma (2-4,6,8). Previously reported cases are often associated with post-operative injury either due to changes in hemodynamics as a mechanism for lymphatic damage and/or trauma. We present the youngest known case of a previously healthy patient developing PB following a traumatic impact to his chest. The motor vehicle collision led to injury of the thoracic duct with lymphatic extravasation into the left hemithorax.

Diagnosis can be made using invasive or non-invasive techniques. Invasive techniques include bronchoscopy with visualization of casts in the airways. Non-invasive techniques include MR lymphography, MR lymphangiography, and lymphangioscintigraphy with SPECT-CT. Noncontrast MR lymphography and MR lymphangiography use very long T2 weighted sequences that image the lymphatic system and can detect lymphatic abnormalities (9). These scans are helpful for pre-surgical planning for ligation if abnormalities that are found. Ligation can be simultaneously

done with conventional lymphangiogram. Lymphangioscintigraphy is a nuclear medicine scan where radiotracer is injected into the lower and/or upper extremities to identify areas of abnormal tracer accumulation or leakage. This allows assessment of the status of the central lymphatic channels for sites of disruption, obstruction, and tracer leakage but does not produce specific mapping of the lymphatic vessels as lymphogram studies can.

There is no uniform treatment plan for PB given the variability in presentations and underlying conditions it can be associated with and known therapeutic options have only been supported by anecdotal evidence. Generally, treatment would include both therapies directed at expelling or removing casts as well as reducing secretion of lymph. If found to be associated with an underlying condition, then treatment of the underlying disorder often leads to resolution of bronchial cast formation (6). Mechanical mucolysis and retrieval with bronchoscopy can be lifesaving in patients who present with casts obstructing major airways. Chest physiotherapy, aerosolized bronchodilators, corticosteroids, and mucolytics may also be useful in improving cast clearance by disrupting casts and hopefully thinning them to ease expectoration (6). Efforts to reduce hypersecretion can include systemic corticosteroids, particularly in those with underlying atopic, asthmatic, or infectious conditions (6). Steroids are generally ineffective in type II acellular cast production (6). Low dose azithromycin has also been anecdotally useful. In post-Fontan patients, bosentan, sildenafil, and thoracic duct ligation have been used (6). Patients with cyanotic congenital heart disease can benefit from optimizing cardiac rhythm and output. Our patient has had no further episodes of cast production by following a low-fat diet, which has previously been demonstrated to be an effective approach (10).

The prognosis for PB is generally favorable especially when secondary to an underlying disease process that can be treated. PB secondary to cyanotic congenital heart disease however is often more associated with respiratory failure due to central airway obstruction (6). Acellular casts also generally have a poorer prognosis compared to inflammatory casts (6).

Our case highlights the importance of considering plastic bronchitis in those patients who present with wheezing and cast formation following trauma to the chest. While asthma is a known cause of cast production, our patient had no history suggesting an atopic condition. Due to the potentially life-threatening nature of cast production, it is important to diagnose PB and begin therapy. An awareness of the disease can assist physicians in identifying plastic bronchitis amongst the other causes of cast production.

CONFLICT OF INTEREST AND DISCLOSURE

The authors declare no competing financial interests exist.

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