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Yoav Dori, MD PhD, Maxim Itkin, MD

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Etiology and new treatment options for patients with plastic bronchitis

Yoav Dori MD PhD

Corresponding author

The Children's Hospital of Philadelphia, Cardiology

doriy@email.chop.edu

Maxim Itkin MD

Hospital of the University of Pennsylvania, Radiology

Maxim.Itkin@uphs.upenn.edu

In this issue of the journal, Ugaki and colleagues describe a case of a 4-year-old child diagnosed with hypoplastic left heart syndrome and plastic bronchitis (PB) that developed shortly after his Fontan operation. The patient failed conventional management including nebulized tPA and Dornase Alpha, and Fontan fenestration. Due to persistent symptoms of PB bilateral groin intranodal lymphangiography was performed. Access to the cisterna chyli was obtained via the transhepatic approach and contrast injection demonstrated an accessory right duct perfusing the right hilar and peribronchial lymphatics. The thoracic duct (TD) could not be selectively cannulated so disruption of the cisterna chyli was performed. The patient had an improvement in his symptoms but ultimately succumbed to hypoxia and other comorbidities.

Until recently the etiology of PB was poorly understood although lymphatic involvement has been considered to be part of the pathological process.¹⁻³ Poor understanding of the disease process has led to variability in therapies across centers. Therapies have been aimed at either lowering central venous pressure using medications such as sildenafil or catheter techniques by creation of a fenestration or they have focused on prevention of cast formation using inhaled therapies such as inhaled tPA. These therapies have led to symptomatic improvement in some cases but usually do not lead to resolution of symptoms.⁴ Heart transplantation has been reported in some cases to result in long term resolution of symptoms but has a high mortality risks.⁵ Recent development of new lymphatic imaging techniques has demonstrated abnormal pulmonary lymphatic perfusion in most patients who have developed the disease after single ventricle palliation.⁶⁻⁸ Furthermore, embolization of the ducts responsible for this flow has been shown to be safe and effective and can result in short term and potentially long-term resolution of symptoms alleviating the need for heart transplant. As a result, percutaneous lymphatic based interventions are now becoming the main therapeutic approach to patient's with this disorder.^{7,8}

The cause of plastic bronchitis in this report is consistent with type I plastic bronchitis due to a single lymphatic channel perfusing the right sided peribronchial lymphatic network.⁸ Selective catheterization and embolization of these abnormal lymphatic ducts has been demonstrated to resolved PB while maintaining central lymphatic flow.^{7,8} In this case cannulation of the TD was not successful so disruption of the cisterna chyli was performed. Disruption of the cisterna chyli has been reported as an alternative to TD embolization in treatment of chylothorax, however with lower clinical success.⁹⁻¹¹

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