palliative intervention for Fontan patients with plastic bronchitis in order to simplify management.

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References

1. Dori Y, Keller MS, Rychik J, Itkin M. Successful treatment of plastic bronchitis by lymphatic embolization in a Fontan patient. *Pediatrics*. 2014;134:e590-5.
2. Dori Y, Keller MS, Fogel MA, Rome JJ, Whitehead KK, Harris MA. MRI of lymphatic abnormalities after functional single-ventricle palliation surgery. *AJR Am J Roentgenol*. 2014;203:426-31.
3. Avitabile CM, Goldberg DJ, Dodds K, Dori Y, Ravishanker C, Rychik J. A multi-faceted approach to the management of plastic bronchitis after cavopulmonary palliation. *Ann Thorac Surg*. 2014;98:634-40.
4. Shah SS, Drinkwater DC, Christian KG. Plastic bronchitis: is thoracic duct ligation a real surgical option? *Ann Thorac Surg*. 2006;81:2281-3.
5. Itkin M, Krishnamurthy G, Naim MY, Bird GL, Keller MS. Percutaneous thoracic duct embolization as a treatment for intrathoracic chyle leaks in infants. *Pediatrics*. 2011;128:e237-41.

EDITORIAL COMMENTARY

Etiology and new treatment options for patients with plastic bronchitis

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Ugaki and colleagues¹ 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 tissue plasminogen activator and dornase alfa, 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 lymphatic vessels. 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 died as a result of hypoxia and other comorbidities.

Until recently the etiology of PB was poorly understood, although lymphatic involvement has been considered to be part of the pathologic process.²⁻⁴ 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, applying catheter techniques such as creation of a fenestration, or they have focused on prevention of cast formation using inhaled therapies such as inhaled tissue plasminogen activator. These therapies have led to symptom improvement in some cases but usually do not lead to resolution of symptoms.⁵ Heart transplantation has been reported in some cases to result in long-term resolution of symptoms but has a high mortality risk.⁶ Recent development of new lymphatic imaging techniques has demonstrated abnormal pulmonary lymphatic perfusion in most patients who develop the disease after single ventricle palliation.⁷⁻⁹ Furthermore, embolization of the ducts responsible for this flow has been shown

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Central Message

Plastic bronchitis in patients after single-ventricle palliation has a lymphatic etiology and responds to lymphatic-based treatments.

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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 for patients with this disorder.^{8,9}

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.⁹ Selective catheterization and embolization of these abnormal lymphatic ducts has been demonstrated to resolve PB while maintaining central lymphatic flow.^{8,9} 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.¹⁰⁻¹² Similarly, in this case lymphatic disruption resulted in partial improvement of the child's symptoms and allowed him to be discharged home. We believe that complete obliteration of the abnormal pulmonary lymphatic flow by catheterization of the TD and embolization of these channels could potentially lead to long-term resolution of PB. Further studies should be conducted to determine whether lymphatic disruption could achieve the same outcomes as those that have been reported with lymphatic duct embolization.

References

1. Ugaki S, Lord DJE, Sherwood MC, Winlaw DS. Lymphangiography is a diagnostic and therapeutic intervention for patients with plastic bronchitis after the Fontan operation. *J Thorac Cardiovasc Surg.* 2016;152:e47-9.
2. Wiggins J, Sheffield E, Jeffery PK, Geddes DM, Corrin B. Bronchial casts associated with hilar lymphatic and pulmonary lymphoid abnormalities. *Thorax.* 1989;44:226-7.
3. Hug MI, Ersch J, Moenkhoff M, Burger R, Fanconi S, Bauersfeld U. Chylous bronchial casts after Fontan operation. *Circulation.* 2001;103:1031-3.
4. Parikh K, Witte MH, Samson R, Teodori M, Carpenter JB, Lowe MC, et al. Successful treatment of plastic bronchitis with low-fat diet and subsequent thoracic duct ligation in a child with Fontan physiology. *Lymphology.* 2012; 45:47-52.
5. Avitabile CM, Goldberg DJ, Dodds K, Dori Y, Ravishanker C, Rychik J. A multi-faceted approach to the management of plastic bronchitis after cavopulmonary palliation. *Ann Thorac Surg.* 2014;98:634-40.
6. Laubisch JE, Green DM, Mogayzel PJ, Reid Thompson W. Treatment of plastic bronchitis by orthotopic heart transplantation. *Pediatr Cardiol.* 2011;32:1193-5.
7. Dori Y, Zviman MM, Itkin M. Dynamic contrast-enhanced MR lymphangiography: feasibility study in swine. *Radiology.* 2014;273:410-6.
8. Dori Y, Keller MS, Rychik J, Itkin M. Successful treatment of plastic bronchitis by selective lymphatic embolization in a Fontan patient. *Pediatrics.* 2014;134: e590-5.
9. Dori Y, Keller MS, Rome JJ, Gillespie MJ, Gltaz AC, Dodds K, et al. Percutaneous lymphatic embolization of abnormal pulmonary lymphatic flow as treatment of plastic bronchitis in patients with congenital heart disease. *Circulation.* 2016;133:1160-70.
10. Itkin M, Kucharczuk JC, Kwak A, Trerotola SO, Kaiser LR. Nonoperative thoracic duct embolization for traumatic thoracic duct leak: experience in 109 patients. *J Thorac Cardiovasc Surg.* 2010;139:584-9; discussion 589-90.
11. Binkert CA, Yucel EK, Davison BD, Sugarbaker DJ, Baum RA. Percutaneous treatment of high-output chylothorax with embolization or needle disruption technique. *J Vasc Intervent Radiol.* 2005;16:1257-62.
12. Pamarthi V, Stecker MS, Schenker MP, Baum RA, Killoran TP, Suzuki Han A, et al. Thoracic duct embolization and disruption for treatment of chylous effusions: experience with 105 patients. *J Vasc Intervent Radiol.* 2014;25: 1398-404.