

## DIRECT TRANS-CERVICAL ENDOLYMPHATIC THORACIC DUCT STENT-GRAFT FOR PLASTIC BRONCHITIS

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### ABSTRACT

*Plastic bronchitis is a poorly understood and uncommon diagnosis, arising from multiple etiologies. Traditional treatment consists of steroids and vasodilators, with thoracic duct embolization emerging as a new procedural therapy. Herein, abnormal lymphatic vessels were noted on lymphangiography in an adult patient with debilitating plastic bronchitis, but anterograde lymphatic access was not feasible due to the patient's morbid obesity and non-visualization of retroperitoneal lymphatics. After trans-venous thoracic duct access could not be established, direct trans-cervical thoracic duct access was performed. A thoracic duct stent-graft was placed, excluding the abnormal bronchial lymphatics and maintaining physiologic anterograde flow through the central lymphatics. At three-month follow-up, the patient's condition had resolved.*

**Keywords:** lymphangiography, thoracic duct, stent, plastic bronchitis, endolymphatic

Plastic bronchitis is an entity characterized by expectoration of proteinaceous bronchial casts (1). Lymphatic engorgement and aberrant pulmonary lymphatic flow has been identified in these patients. Presuming a lym-

phatic component, Dori et al described thoracic duct embolization (TDE) as an alternative to surgical ligation to treat children with cardiac plastic bronchitis (2,3). In an effort to maintain anterograde flow in the central lymphatics, endolymphatic stent-grafting has emerged as an alternative to TDE although it has only been performed from trans-venous approaches (3,4). Herein, a morbidly obese patient with plastic bronchitis had non-visualization of retroperitoneal lymphatics precluding trans-abdominal access and failed retrograde trans-venous access. Direct trans-cervical access and endolymphatic stent-graft (covered stent) placement was performed to exclude plexiform lymphatics along the margins of the left mainstem bronchus, while maintaining patency of the central conducting thoracic duct.

### CASE REPORT

Institutional review board approval was granted for this publication. All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. Informed consent was obtained from the individual included in the study.

A 63-year-old male with history of morbid obesity (body mass index 57), chronic obstructive pulmonary disease, and hypertension presented with worsening dyspnea on exertion and daily coughing fits, producing caulk-like bronchial casts. Bronchoscopy confirmed the suspected diagnosis and after failing six months of medical management, interventional radiology was consulted for treatment.

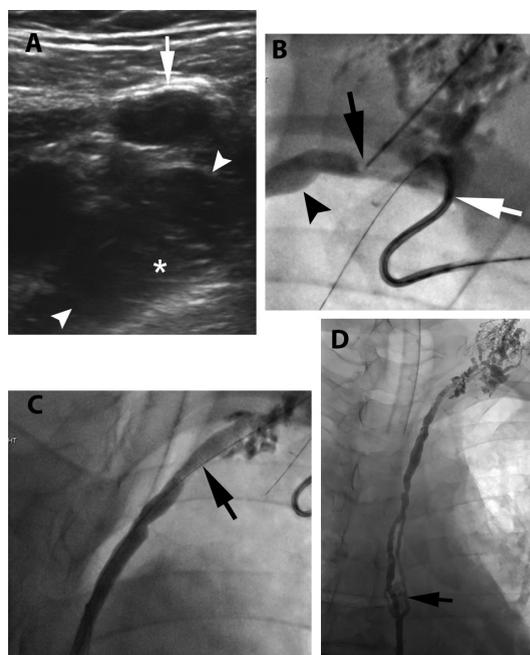
Bi-inguinal nodal lymphangiography was performed using 25 gauge spinal needles (Becton, Dickinson, and Company; Franklin Lake, New Jersey). Hand injection of ethiodized oil (Lipiodol; Guerbet, Villepinte, France) was performed at approximately 0.2-0.4 mL per minute. Fluoroscopy and spot radiographs followed the progression of ethiodized oil from lymph nodes to the retroperitoneal lymphatics. However, although ethiodized oil accumulated in the cervical portion and near the venous angle terminus of the thoracic duct, neither the cisterna chyli nor the thoracic lymphatic vessels opacified (*Fig. 1*). Anterograde abdominal access of the cisterna chyli was therefore not attempted, and retrograde trans-venous thoracic duct access was pursued. A 5 French sheath was placed in the left basilic vein and



*Fig. 1.* Lymphangiography opacifies portions of the thoracic duct and accumulates in the cervical portion of the thoracic duct to its termination at the venous angle (black arrow).

a variety of catheter and wire combinations were used to engage the thoracic duct at the left venous angle, albeit unsuccessfully due to the robust terminal valves.

At this point, direct trans-cervical cannulation of the thoracic duct was undertaken. Targeted ultrasonography delineated the vasculature at the left venous angle (*Fig. 2A*) and after navigating a 22-gauge x 10 cm Chiba needle (Cook Inc.; Bloomington, Indiana) beyond the adjacent arteries and veins, fluoroscopic guidance facilitated direct puncture of the cervical thoracic duct (*Fig. 2B*). A V-18

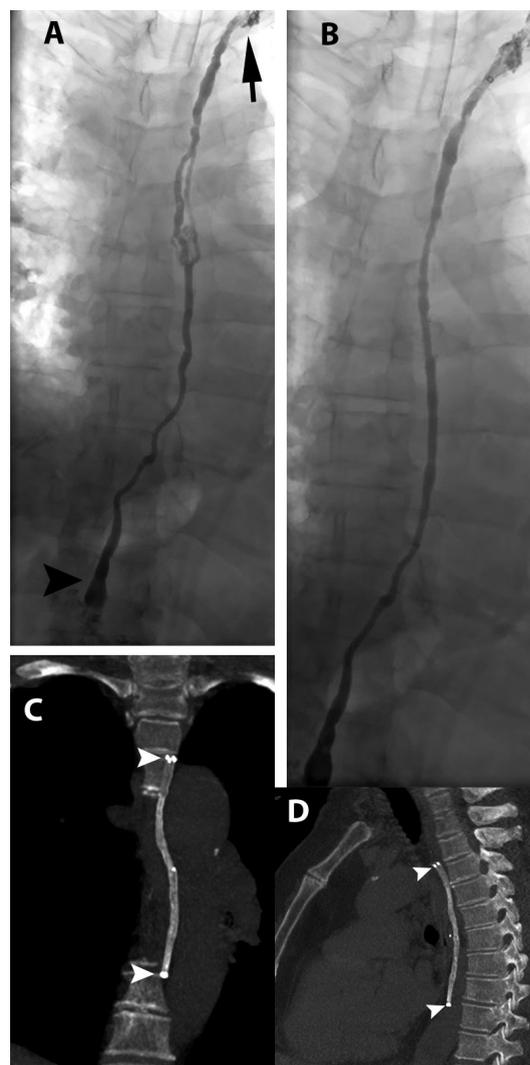


*Fig. 2A-D.* Establishing central lymphatic access. A) Ultrasonography of the left venous angle demonstrates the relationship of the left internal jugular vein (white arrow) and the terminus of the thoracic duct (white arrowheads). Speckles of increased echogenicity in the thoracic duct (asterisk) correspond to ethiodized oil in transit. B) After unsuccessful retrograde transvenous attempts with a reverse curve catheter (white arrow), a 22-gauge needle was navigated to the cervical thoracic duct (black arrowhead) and punctured it displacing contrast (black arrow). C) A V-18 microwire (black arrow) was passed in retrograde fashion through the thoracic duct. D) Central lymphangiography was performed revealing plexiform lymphatics (black arrow) in the vicinity of the left mainstem bronchus.

microwire (Boston Scientific, Marlborough, Massachusetts) was passed retrograde into the retroperitoneal lymphatics (*Fig. 2C*). A 6 French x 45 cm sheath was placed over the wire and a 4 mm x 2 cm Sterling balloon (Boston Scientific) was placed. After balloon inflation, retrograde digital subtraction lymphangiography was performed revealing plexiform lymphatic channels along the left mainstem bronchus without a definitive leak (*Fig. 2D*). In an effort to maintain antero-graduate lymphatic flow and also to exclude the abnormal vessels, endolymphatic thoracic duct stent-graft placement was pursued. A 5 mm x 15 cm Viabahn endoprosthesis (W.L. Gore & Associates, Flagstaff, Arizona), which is comprised of a self-expanding helical nitinol stent lined with polytetrafluoroethylene graft material, was chosen because it is low-profile and could conform to the size range of the patient's thoracic duct. The covered stent was centered across the plexiform lymphatics and deployed (*Fig. 3A*). Completion lymphangiography confirmed thoracic duct patency and successful exclusion of the plexiform lymphatic channels (*Fig. 3B*). No immediate complications were encountered and the patient was discharged uneventfully the next day. At initial clinic follow-up, 1-month post-procedure, the patient reported mild improvement in severity and frequency of his productive cough. At a second clinic follow-up, 3-months post-procedure, his presenting symptoms had resolved and his cough only produced clear, thin secretions. The patient declined post-procedure imaging to evaluate patency of the central lymphatics. Chest CT imaging was performed at 3 months to follow known pulmonary nodules, allowing for additional stent imaging (*Figs. 3C and 3D*).

## DISCUSSION

Plastic bronchitis is an infrequent diagnosis with an uncertain etiology. Associations with tuberculosis, allergic bronchopulmonary aspergillosis, cystic fibrosis, asthma, amyloidosis, and surgical single-ventricle palliation have been documented. As such, its treatment



*Fig. 3A-D. Central lymphangiography and CT imaging pre- (A) and post- (B-D) placement of a 5 mm x 15 cm Viabahn (covered stent) endoprosthesis centered at the abnormal lymphatics. A) Pre-stent-graft placement lymphangiography was performed from the cisterna chyli (black arrowhead) to the cervical thoracic duct (black arrow). B) Post-stent graft lymphangiography confirms successful exclusion of the abnormal vessels and persistent antero-graduate flow of the central lymphatics. C) Coronal maximum intensity projection multiplanar CT reconstruction and D) Sagittal maximum intensity projection multiplanar CT images reveal the stent-graft in the expected location and configuration to the thoracic duct with radio-opaque markers (white arrowheads) marking the ends of the stent. Patency is indeterminate with this technique.*

remains inconsistent if not undefined. Current therapeutics emphasize cast expectoration by mucolytics, reduction of central venous pressure with sildenafil, or steroidal anti-inflammatories (1,3). More recently, a potential lymphatic component has been defined with conventional lymphangiography and dynamic contrast-enhanced magnetic resonance lymphangiography (DCMRL) demonstrating abnormal lymphatic flow to peribronchial lymphatics and lung parenchyma. The increase in lymphatic bronchial perfusion may result in permeation of lymph proteins and even chyle into the bronchial tree with subsequent cast formation (3).

Lymphatics are a pioneering frontier for interventional radiology. TDE for chylothorax was the first modern lymphatic intervention and has become common in adult and pediatric patients (5,6). While safe and well accepted, approximately 5-10% of patients may suffer long-term sequelae including chronic leg swelling/lymphedema, protein-losing enteropathy, chronic diarrhea, and abdominal swelling (7). Thoracic duct stent-grafting (TDSG) is an alternative approach to treat lymphatic pathology and in the case of plastic bronchitis, could potentially exclude lymphatic-bronchial branches and concurrently preserve thoracic duct patency. In prior short reports, TDSG placement has been performed from either transabdominal or retrograde transvenous access. The direct trans-cervical approach used in this case has been previously described for TDE (8). Benefits of this approach are the shallow location of the targeted lymphatic relative to the traditional access of retroperitoneal vessels, which was particularly helpful in this case given the large size of this patient. Additionally, the absence of respiratory motion and mobile bowel loops allows for easier targeting. Attention and care to avoid cervical blood vasculature is paramount. Ultimately, this access facilitated successful endolymphatic TDSG, which excluded the abnormal plexiform lymphatics, preserved anterograde lymphatic flow, and has the potential for fewer long term sequelae compared with thoracic

duct embolization.

Limitations to this report include that it was performed at a single institution with a single patient and was not compared to embolization approaches. Additionally, because TDSG is uncommonly performed there are no long-term data available on stent patency or complications. Lymphatic flow is markedly slower, less pressurized, and lower in volume than arterial or venous flow and lymphatic fluid may be more viscous, predisposing to stent occlusion. It is unclear if antiplatelet medication would potentially improve stent patency in this circulation given that the long term patency of TDSG has not been evaluated. Moreover, the mechanisms for potential stent occlusion in the lymphatic circulation may be different from those in arterial or venous stents.

In conclusion, direct trans-cervical access is a viable approach to facilitate advanced lymphatic procedures, particularly in larger patients, if retroperitoneal lymphatics do not opacify, or if they cannot be successfully accessed. TDSG may function to both treat the underlying lymphatic abnormality and potentially preserve anterograde flow in the central lymphatics. Further study and follow-up are needed.

#### *CONFLICT OF INTEREST DISCLOSURE*

All authors declare no competing financial interests exist.

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