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Reconstructive

Microsurgical Thoracic Duct Lymphovenous Bypass in the Adult Population

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Summary: Thoracic duct occlusion can lead to devastating complications, resulting in recalcitrant chylothoraces, ascites, generalized lymphedema, metabolic derangement, and death. Lymphatic extravasation has traditionally been managed conservatively and, in recent years, using minimally invasive techniques, such as thoracic duct ligation and embolization. However, these measures are often limited in application and therapeutic success, resulting in chronically difficult conditions with few modalities available for definitive management. Advances in microsurgery have allowed for surgical treatment and resolution of peripherally-based lymphatic pathology, though microsurgical intervention to address central lymphatic abnormalities is scarcely described. This report is the first series detailing experiences utilizing microsurgical thoracic duct lymphovenous bypass in a refractory adult population with thoracic duct occlusion. Four patients successfully underwent the procedure, with three achieving complete resolution of symptoms. The fourth patient enjoyed partial resolution, though ubiquitous lymphatic deformities have conferred recurrent residual lower-extremity peripheral edema requiring future intervention. Postoperatively, patent anastomoses were confirmed under magnetic resonance lymphangiography. This series demonstrates the feasibility of microsurgical thoracic duct lymphovenous bypass as a promising technique in treating patients suffering from thoracic duct occlusion. This intervention is effective for recalcitrant chylothorax, chylous ascites, and generalized lymphedema, particularly when traditional and interventional radiological techniques are unsuccessful. *(Plast Reconstr Surg Glob Open 2021;9:e3875; doi: 10.1097/GOX.0000000000003875; Published online 14 October 2021.)*

INTRODUCTION

Thoracic duct (TD) occlusion can manifest with several devastating complications, including chylothorax, cylous ascites, and generalized lymphedema.[1,2] Common etiologies include trauma, iatrogenic disruption, malignancy, and undiagnosed congenital conditions.[3] These may dramatically affect qualify of life and increase risk of mortality.[4,5]

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Traditional management has ranged from dietary adjustments and pharmacological treatment, to surgical ligation, thoracic duct embolization, and thoracic duct disruption.[6–10] Although these techniques offer efficacy, their scope of therapeutic success may be limited across certain lesion-types.[8,11–13] Increasingly advanced techniques are becoming exceedingly necessary when these interventions fail, as mortality has been reported to be anywhere from 10%–50% untreated.[2,14]

Microsurgical advancements have placed a newfound focus on employing novel techniques for lymphatic-associated pathologies.[15,16] The microsurgical specialist has seen increased involvement for refractory lymphedema and lymphatic disruption.[16–19] Additionally, the senior authors (MI, SJK) were the first to describe a novel microvascular approach involving thoracic duct lymphovenous bypass to treat recalcitrant chylothorax in the pediatric

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population.[20] Here, we present the first series of microsurgical thoracic duct-lymphovenous bypass for treatment of chylous pathology.

Perioperative Imaging

A perioperative lymphangiogram is performed through cannulation of the TD by a transabdominal or percutaneous approach, dependent upon accessibility. The perioperative lymphangiogram serves to demonstrate a real-time view of the lesion, guide dissection, and confirm anastomotic patency. (**See Video 1 [online]**, which displays preoperative lymphangiogram demonstrating retrograde lymphatic flow, extravasation, and thoracic duct occlusion.)

Operative Technique

An incision begins transversely from the anterior border of the sternocleidomastoid. The platysma is dissected, exposing the underlying carotid sheath. The carotid sheath is then entered with meticulous dissection and isolation of the internal jugular vein, with care to protect the vagus nerve and carotid artery. Subsequently, the internal jugular is traced in an inferior and posterior fashion until the TD is located (Fig. 1A). The fluoroscopic lymphangiogram confirms the presumptive identification of the TD, which is prepared for anastomosis once a suitable vein is identified (Fig. 1B).

Multiple veins serve as potential recipient candidates dependent upon the exact location of the TD and its spatial relation within the surgical field. The authors prefer the external jugular (EJ) vein due its ease in location, favorable spatial relation for the mechanical movement required for anastomosis, and because it is readily identifiable superficially to the sternocleidomastoid. However, the internal jugular, azygous, proximal cephalic, and anterior jugular veins, as well as suitable collaterals therein, are all feasible options. When selecting a vein, it is important to consider the patient's unique anatomy to select a vessel that is not only large enough for anastomosis, but will also allow vessel transposition to the TD to prevent anastomosis under tension. The vein is dissected and divided, with care to select a point that provides adequate pedicle length in order for transposition to occur without undue tension. After release, the EJ is moved in an inferolateral direction through a fenestration within the sternocleidomastoid or under the muscle, and placed into anastomotic position near the open TD. An end-to-end anastomosis is then preformed with a 2.0 or 3.0 mm venous coupler between the EJ and TD. The authors prefer an end-to-end anastomosis due to the small vessel size and likely negligible morbidity, though an end-to-side anastomosis is feasible with likely similar results. Fluoroscopic lymphangiography and isosulfan blue dye injection through the cannulated TD are used to confirm successful anastomosis. (**See Video 2 [online],** which displays lymphovenous anastomotic patency following an end-to-end anastomosis between the thoracic duct and an external jugular vein. Isosulfan blue can be seen flowing freely through the anastomosis.)

RESULTS

Four patients were identified, with three patients achieving complete resolution of symptoms. A fourth patient had partial resolution of her chylothorax, but developed truncal and lower-extremity lymphedema that required reoperation, including embolization and sclerotherapy in the retroperitoneum, as well as a need for future peripheral lymphovenous bypass. Specific case details are demonstrated in Table 1.

Case Presentation

A 23-year-old woman with a history notable for chronic undifferentiated abdominal pain conservatively managed with a low-fat diet presented with sudden, severe

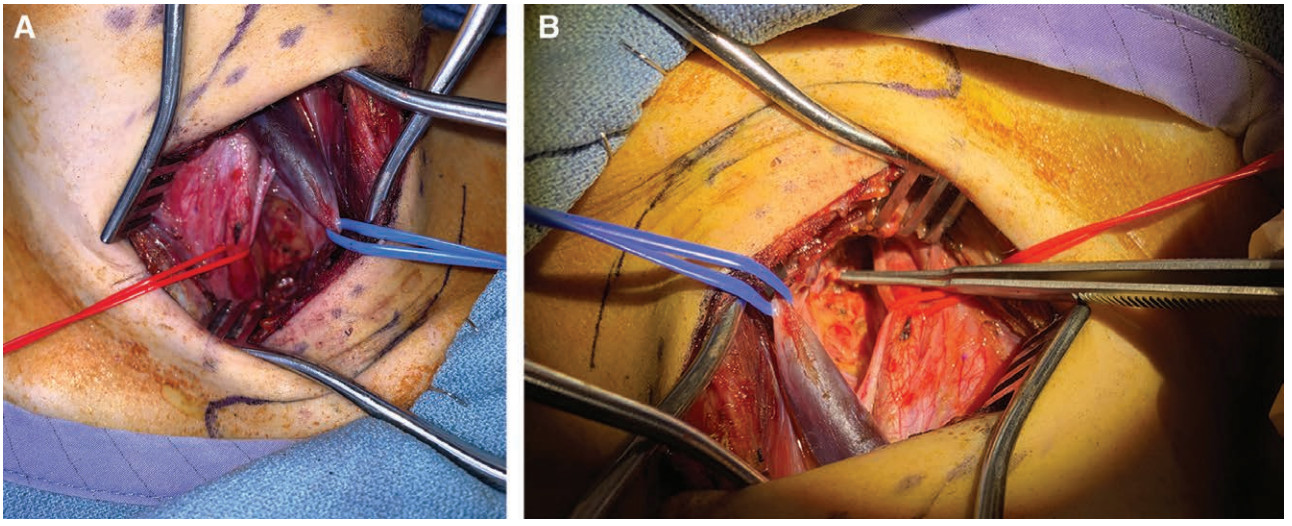

Fig. 1. A, Intraoperative dissection showing isolation of the internal jugular vein (blue) and carotid artery and vagus nerve (red). B, Intraoperative dissection demonstrating the thoracic duct after confirmation with fluoroscopic lymphangiogram (pickup forceps tip).

Table 1. Patient Characteristics, Procedural Intervention, and Postoperative Outcomes

	Patient 1	Patient 2	Patient 3	Patient 4
Age/gender	61/man	23/woman	27/woman	40/woman
Lymphatic abnormality	Chylothorax, chylous ascites, and generalized lymphedema	Chylous ascites	Chylous collections in neck, chylothorax	Chylothorax, chylous ascites, and generalized lymphedema
Etiology	Lymphatic malformation secondary to Noonan syndrome	Bifurcated thoracic duct	Cystic lymphangioma	Generalized lymphatic anomalies
Preoperative interventions	Sirolimus	None	Sirolimus, Percutaneous IR drainage of mass	Sirolimus
Intraoperative IR lymphatic access and imaging	Yes	Yes	Yes	Yes
Lymphaticovenous anastomosis	End-to-end thoracic duct-external jugular vein anastomosis	End-to-end thoracic duct-external jugular vein anastomosis	End-to-end thoracic duct-external jugular vein anastomosis	End-to-end thoracic duct to anterior jugular vein
Postoperative complications	Day 1: dyspnea requiring intubation	Day 7: dyspnea requiring admission and observation	None	Day 14: development of lower truncal and lower-extremity lymphedema
Postoperative intervention	None	None	None	Day 30: Reoperation with embolization and sclerotherapy of the lymphatic masses in the mesentery and retroperitoneum
Postoperative lymphangiogram	1 month: patent anastomosis	7 months: patent anastomosis	Awaiting imaging	Awaiting imaging
Last followup	1 year: asymptomatic	7 months: asymptomatic	7 months: asymptomatic	3 months: awaiting surgery

IR, Interventional radiology.

abdominal pain. Exploratory-laparoscopic surgery and subsequent fluid analysis revealed excessive chylous ascites, and a follow-up lymphangiogram showed an underlying occluded bifurcated thoracic duct with formation of lymphangiectasias. Given the extent of occlusion, thoracic duct lymphovenous bypass was pursued.

The patient underwent successful operative bypass without complication using an end-to-end thoracic duct-EJ anastomosis and was deemed appropriate for discharge on postoperative day one. On postoperative day 7, the patient experienced sudden dyspnea and was admitted for work-up. Radiographic imaging revealed no underlying mechanical or physiological causes, and the patient was diagnosed with laryngeal nerve palsy secondary to the operation. At 7-month follow-up, a lymphangiogram demonstrated a patient anastomosis. The patient reports no chest pain, healthy weight-loss, and a complete resolution of the preoperative chronic abdominal pain. Specific case details for subjects are demonstrated in Table 1, with pre- and postoperative imaging added as case examples, including figures for fluoroscopic imaging (Fig. 2) and magnetic resonance lymphangiography (Fig. 3).

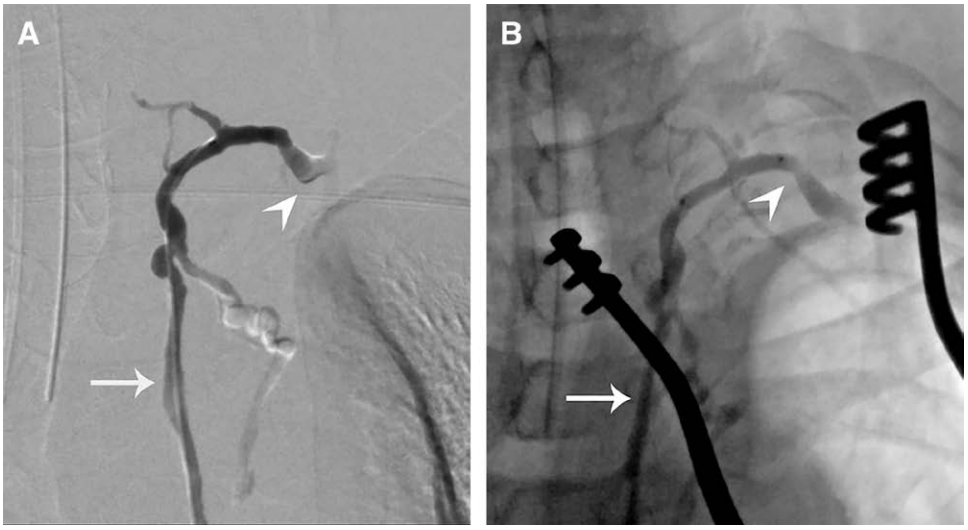

Fig. 2. Intraoperative fluoroscopy images of the injection of the contrast in the thoracic duct (arrow). A, Complete occlusion of the distal thoracic duct (arrowhead). B, Patent thoracic duct venous anastomosis (arrowhead). Reprinted from *Ann Thorac Surg* with permission from Elsevier.[22]

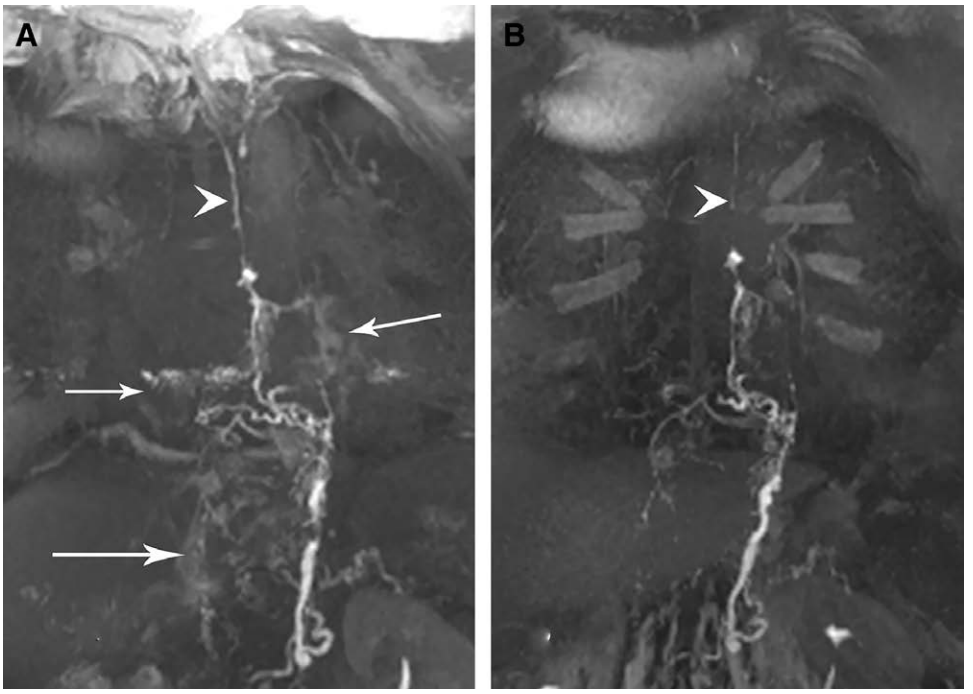

Fig. 3. A, Magnetic resonance lymphangiogram before the creation of the thoracic duct-venous demonstrated opacification of the thoracic duct (arrowhead). There is a significant lymphatic congestion/lymphatic flow reflux into the mediastinum and upper abdominal organs (arrows). B, Followup of the thoracic duct-venous anastomosis, demonstrating a significant reduction of the lymphatic congestion. The thoracic duct is notably patent (arrowhead). Reprinted from *Ann Thorac Surg* with permission from Elsevier.[22]

DISCUSSION

This is the first series reporting thoracic duct lymphovenous bypass in the adult population. Lesions of the thoracic duct can present with multisystem complications in several anatomical areas, as apparent by the range of manifestations in our patients. Anastomotic bypass allows for definitive lymphatic drainage and quality of life improvement across several variations of lymphatic abnormalities.

We hypothesize that the pathophysiological mechanism of occlusion in our patients involves an increased pressure gradient within the lymphatic system, resulting in the formation of lymphangiectasias. This, in congruence with collateral formation and overflow leakage and effusion, results in consequent fluid displacement into bodily cavities.

Utilizing bypass across the occlusion site allows for effective drainage and relief of undue pressure, and our series demonstrated successful complete or partial resolution for all subjects. Initial respiratory distress may occur postoperatively secondary to the augmentation of venous return from the now bypassed TD. We hypothesize that this risk is particularly amplified in patients with chronic disease and/or relevant comorbidities, such as patient 1 suffering from heart failure. After about 3–7 days, we believe that physiological restoration is achieved in concordance with increased venous return, and that any postoperative dyspnea or discomfort is alleviated. To help mitigate the risk of future occurrence, patients should be optimized preoperatively, including achieving cardiac clearance. Laryngeal nerve palsy is another potential etiology, which demands meticulous intubation and dissection. Literature investigating the procedure in the pediatric population has shown that severe pathology may be refractory to bypass, which was seen in one of our subjects likely due to generalized lymphatic abnormalities.[20,21]

The senior authors were the first to describe their experiences treating recalcitrant chylothorax in the pediatric population and Noonan Syndrome utilizing a microsurgical TD-bypass approach.[20–22] Our series is the first to describe successful treatment of symptomatic pulmonary and abdominal symptoms. Patients are carefully selected based on etiology of the lesion. Patients with mechanical obstruction of the TD are suitable for the procedure, while those with physiological obstructions, such as heart failure or arteriovenous fistulas, are not suitable and may have their conditions exasperated. This study involved patients that were refractory to more traditional management, including minimally invasive techniques. More specifically, thoracic duct embolization and thoracic duct disruption have reported success for a variety of etiologies.[8,10,11,13] However, these interventions may at times be limited in applicability in treating thoracic duct lesions.

The exact efficacy and safety of this technique are limited by a small sample-size and relatively short follow-up. Additionally, this procedure also requires a strong multidisciplinary approach from a radiological and microsurgical standpoint, as fluoroscopic visualization of the anatomy guides the procedure and confirms success. We utilize a hybrid-suite (magnetic resonance/operating room) to maximize surgical efficiency, with the interventional radiologist present for cannulation and imaging intraoperatively.

CONCLUSIONS

Microsurgical thoracic duct lymphovenous bypass is a promising technique in treating patients suffering from thoracic duct occlusion. This intervention warrants further investigation regarding its usage for recalcitrant chylothorax, chylous ascites, and generalized lymphedema, particularly when traditional and radiological techniques are unsuccessful.

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