Interposition of great saphenous vein on lymphatic venous anastomosis for infantile intractable chylothorax

Motoi Kato, MD, PhD,[a] Takahiro Hirayama, MD,[a] Hiroshi Kawashima, MD,[b] Azusa Watanabe, MD,[a] *and* Shoji Watanabe, MD, PhD,[a] *Saitama, Japan*

ABSTRACT

Central lymphatic diseases such as intractable chylothorax can be fatal. Lymphatic venous anastomosis at the venous angle level is expected to give a direct therapeutic effect because it opens the obstructed outlet of the main lymphatic vessels. However, the original methods resulted in some important issues, such as the potential for venous reflux. In the present case, we modified the original anastomosis method by interposing a vein graft with venous valves to increase the distance and prevent venous reflux. Collecting the lymphatic flow resulted in termination of the chylothorax with preserved postoperative patency for years, without any complications, including at the graft-harvested extremity. (J Vasc Surg Cases Innov Tech 2023;9:101332.)

Keywords: Chylothorax; Jugular vein; Lymphatic venous anastomosis; Thoracic duct; Vein graft

In treating refractory chylothorax associated with an impaired passage of the thoracic duct outlet caused by venous clots or high venous pressure, lymphatic venous anastomosis (ie, thoracic duct—jugular vein anastomosis) is expected to be effective.[1,2] However, existing reports indicate that issues, such as venous blood reflux, the distance between the anastomotic vessels, and a caliber discrepancy between the anastomotic vessels, can occur. To address these problems, we considered using the great saphenous vein (GSV), which contains a venous valve, as a graft.

CASE REPORT

A 2-month-old male infant was diagnosed with intractable chylothorax. The patient's parents provided written informed consent for the report of his case details and imaging studies. The infant was born at 37 weeks and 1 day (weight, 2408 g; length, 43 cm) by a scheduled cesarean section due to a complication of a myoma with the pelvic position. The Apgar score was full at both 1 and 5 minutes. He was diagnosed with myocardial hypertrophy at birth and was referred to our hospital for further examination at the age of 2 months. On arrival, he had depressed respiration and was diagnosed with chylothorax, which was treated with chest tube drainage in the left thoracic cavity. His medical history included hypertrophic cardiomyopathy and Noonan syndrome (PAF1 variant).

Physical findings. On arrival at the hospital, his respiratory rate was 50 breaths/min, his oxygen saturation was 90% on room air, and his body weight was 3367 g. The patient was treated with MCT (medium chain triglycerides) milk, followed by fasting, in addition to intravenous octreotide, propranolol, and steroids. However, the pleural effusion did not decrease. Two weeks after his arrival, he was referred to our department.

Lymphatic scintigraphy revealed impaired lymphatic drainage and congestion at the venous angle associated with lymphatic reflux into the thoracic cavity. Clinically, chyle pleural fluid was collected at 200 mL/d, on average, with a high risk of respiratory failure for >1 month from the initial onset of pleural fluid, even with conservative treatment. A preoperative systemic venous examination revealed no obstructions or atresia, including at the cervical venous angle and bilateral deep femoral veins. Therefore, we decided to perform lymphatic venous anastomosis at the venous angle site to improve lymphatic drainage.

Surgery. Under general anesthesia, the patient was positioned supine with the left side of his neck extended. A supraclavicular transverse incision on his left collar was made to dissect the external jugular vein, followed by dissection of the thoracic duct, with no dye or radiotracer used. No obvious clots or extravascular lesions were detected at the venous angle on the thoracic duct. However, blood backflow was present, with respiratory fluctuation, from the subclavian vein to the thoracic duct. The distal part of the external jugular vein was small for the thoracic duct, with a size discrepancy, although the proximal part was less discrepant.

Through two 1-cm skin incisions made on his left thigh, we collected a 3-cm-long segment of the GSV, slightly longer than the distance from the thoracic duct to the base of the external jugular vein. Immediately after the GSV segment was harvested, saline was injected to fill the lumen from the distal side (originally the inflow side), with the proximal side (originally the outflow side) clamped. Multiple varicose dilatations, typical of venous valves, were noted, and they did not flatten when the distal side was unclamped. Therefore, we confirmed that the graft contained antireflux valves. The graft was placed to function as the antivenous reflux segment. It opened a progressive pathway from the thoracic duct to external jugular vein: from

From the Department of Plastic and Reconstructive Surgery,[a] and Department of Surgery,[b] Saitama Children's Medical Center.

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Correspondence: Motoi Kato, MD, PhD, Department of Plastic and Reconstructive Surgery, Okayama University, 2-5-1, Shikata-cho, Kita-ku, Okayama City, Okayama 700-8558, Japan (e-mail: motoikato25@gmail.com).

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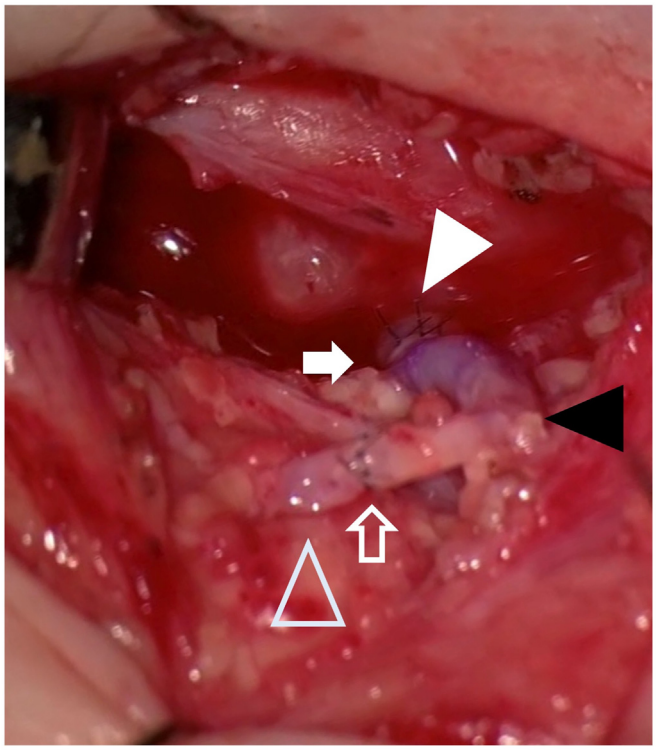

Fig 1. Thoracic duct reconstruction with great saphenous vein (GSV) interposition. The lymphatic outlet was redirected with the anastomoses. The GSV, with the venous valve included, was interposed to complete the anastomosis in an end-to-end manner between the thoracic duct and external jugular vein. *White arrowhead* indicates thoracic duct; *black arrowhead,* grafted GSV; *empty arrowhead,* external jugular vein; *white arrow,* thoracic duct to GSV anastomosis; *empty arrow,* GSV to external jugular vein.

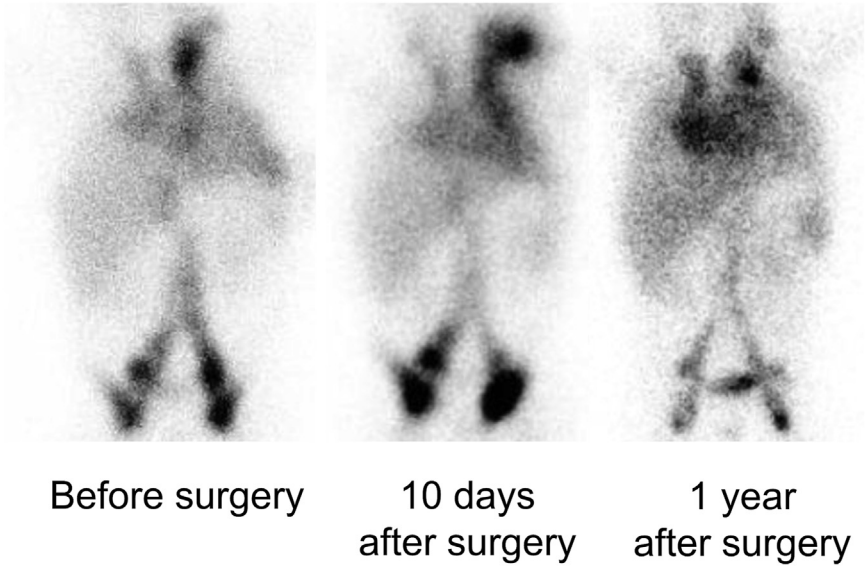

Fig 2. Lymphatic scintigraphy before and after surgery. At 45 minutes after administration of technetium-99m on bilateral dorsal pedis. Lymphatic congestion due to impaired lymphatic drainage at venous angle was present preoperatively, which improved postoperatively.

the thoracic duct (diameter, 1.3 mm) to the distal GSV segment (diameter, 1.2 mm) to the proximal GSV segment (diameter, 1.2 mm) to the external jugular vein (diameter, 1.1 mm). Anastomoses were performed using 9-0 nylon suture in an end-to-end manner under microscope magnification, with patency confirmed after each anastomosis (Fig 1).

Fig 3. Chest radiograph after 2 years postoperatively, with no signs of pleural fluid detected after discharge to home.

Postoperative course. After surgery, the patient's pleural effusion volume began to decrease and had completely resolved by postoperative day 7. Lymphatic scintigraphy on postoperative day 10 showed that the anastomosis site and thoracic duct were well opened, with mild reflux still present. Lymphatic scintigraphy was performed again at 1 year postoperatively to confirm patency. The flow had improved since the examination at 10 days postoperatively. The lymphatic reflux had largely disappeared, and the anastomotic site was completely open (Fig 2).

The patient's progress was uneventful after surgery. At 30 months postoperatively, no recurrence of pleural effusions had developed (Fig 3). The scar was relatively unremarkable, and no donor site problems had been reported (Fig 4).

Fig 4. Photographs showing unremarkable postoperative scars. At 2 years postoperatively, the scars on his neck (**Top**) and left thigh (**Bottom**) were unremarkable. No postoperative complications were detected.

DISCUSSION

Reflux of venous blood into the lymphatic vessels during lymphatic venous anastomosis can decrease postoperative patency.[3,4] Platelets in venous blood can cause inflammation of lymphatic endothelial cells[5] and related thrombus formation at the anastomotic site.[6] Therefore, preventing venous blood from flowing back into the lymphatic vessels is important to improve surgical outcomes. However, anatomically, the head and neck veins have very few functional valves.[7] Therefore, improvement of the surgical method has been required. In the present case, we performed an anastomosis using an interposition graft of the GSV. We confirmed patency of the thoracic duct reconstruction in the infant >1 year postoperatively.

The use of vein graft replacement has several advantages. First, an antireflux valve can be added to prevent possible blood reflux into the lymphatic system. Venous reflux into the lymphatic vessels increases the risk of endothelial damage to the lymphatic vessels at the anastomosis and consequent thrombus formation. Other options to prevent reflux include valvoplasty, such as suturing venous valves from the outside,[8] inserting lymphatic vessel ends into veins,[9] or using tiny venules.[10] However, they were not directly applied to thoracic duct reconstruction in our case because the previous reports aimed at peripheral anastomosis but not treating central disease.

Another benefit of using a venous graft is the longer distance, which makes anastomotic manipulation easy. Anastomosis to the main trunk of the thoracic duct must be performed in a relatively narrow surgical field covered by the clavicle and neck musculature. Interposition of the graft allows for a longer length in the anastomotic vein. This makes it easier to perform procedures such as flipping the vessel and reduces the stress on the surgeon during the anastomotic procedure. In addition, side-to-end anastomosis to other veins, such as the internal jugular vein, is possible theoretically, allowing for a backup plan in case the distal external jugular vein is not feasible for anastomosis (Fig 5).

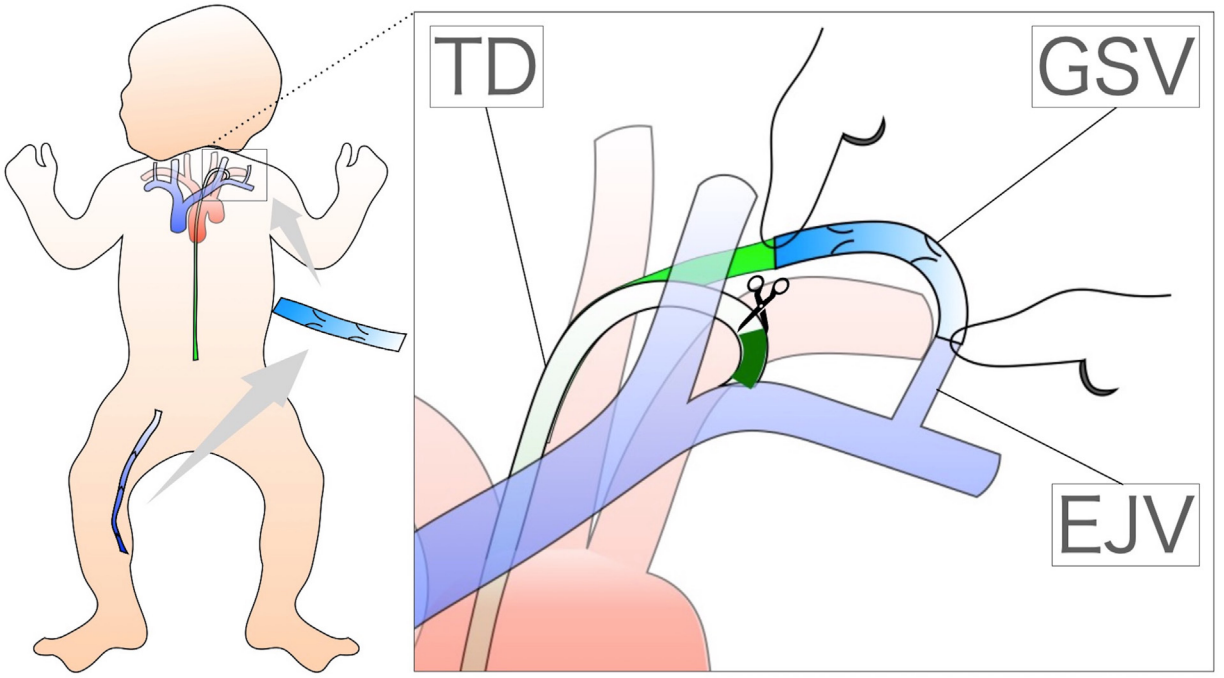

Fig 5. Conceptual diagram showing great saphenous vein (GSV) graft interposition for thoracic duct (*TD*) reconstruction. The GSV graft increases the length and included a valve to normalize lymphatic outflow. *EJV,* External jugular vein.

However, the possibility of scarring at the venous harvested site and edema of the lower extremity after harvesting cannot be denied. Also, the anastomotic site can become occluded after a long period. Therefore, careful observation should be continued. However, we considered that our patient had a relatively low risk of developing late complications caused by obstruction. One study reported that GSV grafts used in children had passed an average of 10 years without stenosis.[11] Additional studies reported the long-term results of free flap transfer in children with vascular anastomosis.[12,13] The GSV interposition graft was considered effective for lymphatic venous anastomosis in venous angle reconstruction.

CONCLUSIONS

In the case of central lymphatic conduction refractory to medical therapy, we placed a GSV graft to reconstruct a venous angle. The interposition of the GSV graft provided several advantages, including a reduced potential risk of venous reflux into the lymphatic system because of the venous valves included, reduced problems with vein shortening, and fewer issues with caliber discrepancies between the thoracic duct and draining veins.

DISCLOSURES

None.

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