

Figure 3. (a) Preoperative lymphoscintigraphy revealed lymph stasis and dermal backflow in the right leg, megalymphatics in the thigh, and decreased inguinal lymph nodes. (b) Lymphoscintigraphy taken 3 months after the first operation revealed decreased lymph stasis and diminished dermal backflow in the leg.

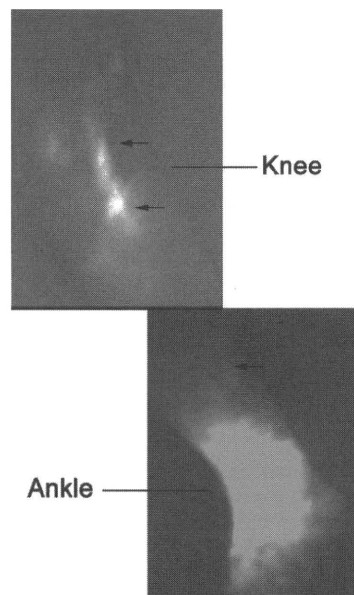


Figure 4. Indocyanine fluorescence lymphography showing dermal backflow in the leg and megalymphatics in the thigh (arrows).

cal symptoms and signs: megalymphatics, simple heman-gioma, and edema.

The underlying causes of congenital chylous reflux are still unknown. We built shunts not between the portal vein but between the great saphenous vein and the megalymphatics. In this case, chyle enters the systemic circulation of the body, and might cause some problems in the lung, for example. However, this specific patient has not developed any respiratory problems and a chest X-ray of this patient revealed no change before and after surgery.

In the case of congenital chylous reflux, valves of the lymphatics seem to be incompetent. Shunt plasty between the megalymphatics and the saphenous vein reduces the pressure in the lymphatic vessels, which improves chylous reflux, although lymph can flow in reverse because of a lack of valves in the vessels.

In cases of infants and children with localized lesions, conservative treatments such as a medium-chain triglyceride (MCT) diet or bowel rest with total parenteral nutrition (TPN) are effective in reducing chyle.<sup>4</sup> However, in cases with widespread malformations of the lymphatic system or in cases with recurrence after conventional therapies, surgical interventions should be considered. In this case, it was difficult to apply such a diet therapy before surgery because of the patient's social situation.

Campisi et al. reported 47 cases of chylous reflux including chylous ascites, chylothorax, chyluria, and chylo-rorrhea in the external genitalia or the lower extremities.<sup>5</sup> Of the 47 cases, 11 had chylo-rorrhea in the leg and external genitalia. They mentioned the usefulness of laser-microsurgical procedures as well as other surgical methods such as ligatures or excisions of incompetent dysplastic and dilated lymphatics in the treatment of several types of chylous disorders. However, details about the method used to perform the microsurgical procedures are unknown. Another report by Campisi in 2007 stated that of 12 cases with chylous ascites, 7 underwent chylo-ve-nous and lymphovenous microsurgical shunts.<sup>6</sup> Noel et al. reported 35 cases with chylous reflux in which 14 cases had chylo-rorrhea of the lower limbs. However, a ma-jority clinical finding was chylothorax in 20 of the cases.<sup>7</sup> Of the 35 cases, 21 had surgical treatment with 26 pro-cedures in which two lymphaticovenous anastomoses and two grafts were performed. They also mentioned that a segment of saphenous vein with a competent valve was used for a lymphovenous shunt but it is unclear which type of graft, where, and how the graft was applied to the patients. Instead of a segment of saphenous vein, we

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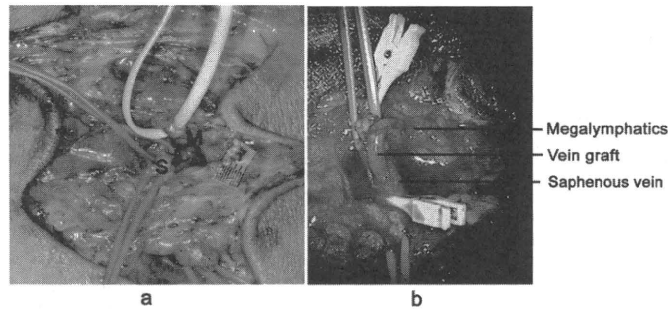


Figure 5. (a) Megalympatics (M) and the great saphenous vein (S) with external diameters of about 3 mm and 3.5 mm, respectively, exposed through an incision in the upper one third of the right thigh. (b) Subcutaneous vein graft with valves placed between the megalympatics and the great saphenous vein. [Color figure can be viewed in the online issue, which is available at [wileyonlinelibrary.com](http://wileyonlinelibrary.com).]

used a part of the subcutaneous vein with valves in the dorsum of the nonaffected foot because a vein with an adequate diameter for use as a shunt can easily be harvested and the saphenous vein should be preserved. In our experience of lymphaticovenous anastomosis for treatment of chronic lymphedema, subcutaneous ecchymosis caused by blood backflow to the lymphatic vessels sometimes occurs, which may develop postoperative inflammation or result in occlusion of the anastomosed vessels. Therefore, the valves should be included in the grafted vein to prevent the backflow of blood from the saphenous vein. Excision, ligation, and sclerotherapy have been reported to be effective methods for chylous reflux. However, drainage methods such as a lymphaticovenous shunt seem to be essential. In a case report about chylous edema in 1966, Ketterings<sup>8</sup> stated that ligation and stripping of abnormal lymphatics were effective even if done transperitoneally or retroperitoneally. Currently, we can apply microsurgical techniques to such intractable cases without great effort. In summary, a subcutaneous vein graft with valves may be considered one useful method for a shunt between incompetent and dilated lymphatics and veins instead of the saphenous vein graft in the treatment of chylous reflux in lower extremities.

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#### REFERENCES

1. Kinmonth JB, Taylor GW. Chylous reflux. *Brit Med J* 1964;1:529-532.
2. Berenji GR, Iker E, Glass EC. Lymphoscintigraphic findings in chylous reflux in a lower extremity. *Clin Nucl Med* 2007;32:725-728.
3. Ketterings C. Chylous edema of the leg: a case report. *Ann Surg* 1967;165:647-654.
4. Unger SW, Chandler JG. Chylous ascites in infants and children. *Surgery* 1983;93:455-461.
5. Campisi C, Boccardo F, Zilli A, Borrelli V. Chylous reflux pathologies: diagnosis and microsurgical treatment. *Int Angiol* 1999;18:10-13.
6. Campisi C, Bellini C, Eretta C, Zilli A, da Rin E, Davini D, Bonioli E, Boccardo F. Diagnosis and management of primary chylous ascites. *J Vasc Surg* 2006;43:1244-1248.
7. Noel AA, Gloviczki P, Bender CE, Whitley D, Stanson AW, Deschamps C. Treatment of symptomatic primary chylous disorders. *J Vasc Surg* 2001;34:785-791.
8. Kayıkçıoğlu A, Karamürsel S, Safak T, Mavili E, Erk Y. Lymphatic malformation causing intractable chylorrhagia. *Plast Reconstr Surg* 2000;105:1422-1425.

