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Complete lymph flow reconstruction: A free vascularized lymph node true perforator flap transfer with efferent lymphaticolymphatic anastomosis[☆]



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Summary Treatment of primary lower extremity lymphedema (LEL) is challenging, and lymph node transfer (LNT) can be a choice of treatment for progressive LEL. However, LNT has a risk of donor site lymphedema and possible lymph node (LN) sclerosis due to efferent lymphatic vessel (ELV) obstruction. Here, we report the first case of complete lymph flow reconstruction with true perforator LNT with efferent lymphaticolymphatic anastomosis (ELLA) for a patient with primary LEL and severe lymphosclerosis. A 49-year-old female suffered from primary progressive unilateral left LEL refractory to conservative treatments with frequent episodes of cellulitis. A true perforator LN flap was selectively harvested from the left lateral thoracic region under indocyanine green (ICG) lymphography navigation and transferred to the left groin with perforator-to-perforator anastomosis. The ELV of the transplanted LN was supermicrosurgically anastomosed to the contralateral iliac lymphatic vessel that was subcutaneously transferred to the left groin. Postoperatively, the patient experienced no episode of cellulitis with reduced degree of compression treatment, and lymphedematous volume decreased from 306 to 264 in terms of LEL index. Postoperative ICG lymphography showed evidence of reconstructed lymph flow from the left foot to the left groin and to the right inguinal LN through the transplanted LN flap and the ELLA. There were no subjective or objective

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findings of donor site lymphedema of the left arm or the right back and the lower extremity. True perforator LN flap with ELLA is a safe and effective treatment and has the potential to be a useful therapeutic option for primary unilateral LEL.

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Management of primary lymphedema is challenging because of difficulty in both diagnosis and treatment.^{1,2} Primary lower extremity lymphedema (LEL) has a wide variety of etiology, and the disease is usually diagnosed by excluding other possible causes of leg edema.^{1–4} After the diagnosis of primary LEL is confirmed, conservative treatments such as compression therapy with or without manual lymph drainage are recommended to prevent exacerbation of the affected leg. However, further surgical treatments are often required for progressive primary LEL refractory to conservative treatments.^{3–5}

With the establishment of supermicrosurgery that enables microvascular anastomosis of small vessels with diameters of 0.5 mm or smaller, supermicrosurgical lymphaticovenular anastomosis (LVA) has recently become more common for the treatment of progressive primary LEL because of its effectiveness and minimal invasiveness.^{6–13} LVA allows the diversion of congested lymph flow into the venous circulation through a 2-cm skin incision under local anesthesia, but is hardly effective for progressed lymphedema cases with severe lymphosclerosis.^{2,9,11,12} For severe lymphedema cases, vascularized lymph node transfer (LNT) is preferred to reconstruct physiological lymph flows, but is significantly more invasive than LVA; LNT requires general anesthesia and has a risk of donor site lymphedema.^{14–19} Another concern is that obstruction of the efferent lymphatic vessel (ELV) of the transferred lymph node (LN) can result in dysfunction of the transferred LN; obstruction of the ELV is reported to be a major cause of obstructive lymphedema.^{20–22}

It is therefore ideal that only a “safe” LN is selectively included in an LN flap, and that the ELV of a transferred LN is anastomosed to an intact lymphatic vessel in a recipient site for complete lymph flow reconstruction; all lymph drained into a transferred LN flows into the drainage vein and the ELV of the LN.^{22–24} However, there are no reports on such complete reconstruction of the lymph flow. In this report, we present a case of a patient with primary unilateral LEL who was successfully treated with selective LN true perforator flap transfer and supermicrosurgical anastomosis of the transferred LN’s ELV to the contralateral iliac lymphatic vessel (ILV) with efferent lymphaticolymphatic anastomosis (ELLA).

Case report

A 49-year-old female had suffered from idiopathic left lower extremity edema for the past 5 years. Edema began at the age of 44 years without any causative episode and since then had gradually worsened. The patient experienced frequent episodes of left leg cellulitis since she was

45 years old. She was not under medication and had no medical history or family history of edematous diseases. Heart failure, nephrosis, liver cirrhosis, deep venous thrombosis, varix, endocrine diseases, pelvic/inguinal malignancy, and edematous diseases other than primary LEL were excluded by thorough examinations, including physical examination, blood tests, urine tests, chest X-ray, ultrasonography, and magnetic resonance imaging, at a nearby general hospital. Although conservative treatments including wearing a compression garment set at 30–46 mmHg, skin care, and manual lymph drainage were applied to control the edematous disease at 47 years of age, her symptoms of the disease were aggravating with the progression of edema and increasing frequency of cellulitis episodes. The patient was referred to our hospital for diagnosis and treatment of the edematous condition suspected of primary LEL. Physical examination revealed that her right/left LEL indices were 224/306, respectively, indicating moderate left lower extremity edema; the LEL index was calculated by a summation of squares of circumferences (cm) at 5 points (the superior border of the patella, 10 cm above and below the superior border of patella, the lateral malleolus, and the dorsum of the foot) divided by body mass index (Figure 1).^{25,26} Preoperative indocyanine green (ICG) lymphography showed extensive dermal backflow (DB) of stardust pattern in a region extending from the left groin to the left ankle and intact lymph flow in the right lower extremity; diagnosis of primary left LEL with proximal DB (PDB) pattern was made, and the right lower extremity was confirmed to have no lymphedema.^{3,27–29} There was no lymphatic connection



Before Operation

Edema

progressing
sensation of severe tension
LEL index 306

Cellulitis

frequent (2–7 times/year)
requiring hospitalization

Conservative Treatments

garment 30–46mmHg
manual lymph drainage

Figure 1 Clinical photograph and conditions before surgery.

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