

LYMPHSPIRATION**SYSTEMIC ECHOES – CAN LYMPHATIC RECONSTRUCTION SURGERY
EXTEND ITS THERAPEUTIC REACH FROM LIMBS TO HEAD****W. F. Chen, S. Kukreja-Pandey, M. Salman**

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ABSTRACT

This case series highlights potential systemic effects of lymphaticovenular anastomosis (LVA) with conventional treatment in patients with primary lymphedema of the lower limb, with a particular focus on its implications for cerebral lymphatic function. We present two cases in which LVA performed on a lower limb led to interesting systemic improvements extending beyond the treated regions, including alleviation of preoperative cognitive symptoms. The first case involves a 37-year-old woman with diffuse body swelling, including facial edema. LVA in her left leg resulted not only in reduction of swelling but also in marked improvement in speech and mentation. The second case describes a 62-year-old woman who, following LVA in her right leg, experienced resolution of limb and facial edema, relief of oropharyngeal symptoms, and improvement in cognitive function. Although these cranial improvements were not objectively assessed due to lack of measurement before the LVAs, they were subjectively perceived by the patients post-operatively. These reports indicate the systemic nature of primary lymphedema and interestingly suggest that peripheral lymphatic surgery and treatment may positively influence cerebral lymphatic dynamics.

Keywords: lymphedema, primary lymphede-

ma, lower limb, lymphatic microsurgery, LVA, cerebral lymphatic function

INTRODUCTION

Recent advancements in lymphology have significantly enhanced treatment of both primary and secondary lymphedema. Primary lymphedema, often misconceived as localized swelling, is actually a systemic condition indicating a broader lymphatic deficiency that affects multiple body regions (1). The discovery of brain lymphatics supports the hypothesis that primary lymphedema may lead to cerebral lymphatic insufficiency (2,3). This correlation could explain cognitive symptoms, such as brain fog and memory impairments, commonly reported by primary lymphedema patients.

Understanding primary lymphedema as systemic affects the treatment approach, suggesting that lymphaticovenular anastomosis (LVA), even if performed on a limb, can have far-reaching effects. Regional surgery could enhance the lymphatic function throughout the body, including the brain, by capitalizing on the lymphatic system's interconnected nature (4).

We present two case studies demonstrating systemic improvements in lymphatic function from LVA surgery and conventional treatment suggesting that treatment for primary

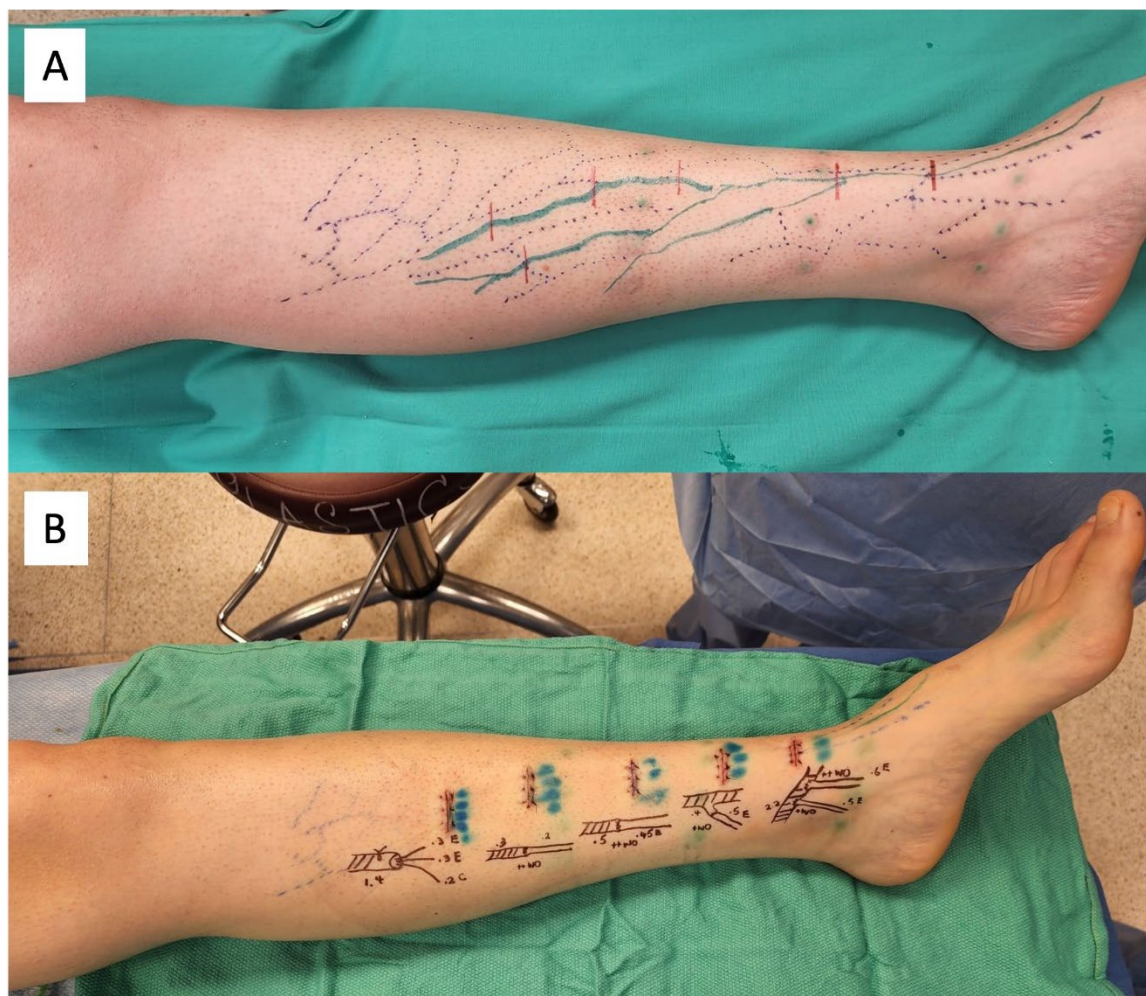


Fig. 1. Case 1: Lymphatic mapping (green lines) and vein mapping (blue dotted lines) performed using DOPSIT (distal to proximal sequential ICG injection technique) and an infrared vein finder, respectively. Incisions were strategically planned (red lines) to facilitate incorporation of both lymphatics and veins (A). A total of 8 lymphaticovenular drainage pathways were created in 5 exploratory incisions using end-to-side, end-to-end, and the "octopus" techniques (B).

lymphedema may have wider impact than previously appreciated.

CASE 1

A 37-year-old woman presented with a 2-year history of facial, gingival, truncal, and extremity swelling, showing daily fluctuations. She experienced pronounced facial and gingival swelling in the morning that improved during the day, whereas trunk and limb swelling

worsened as the day progressed. She reported severe gingival pain and a pervasive burning sensation, correlated with swelling severity. With no identifiable triggers, she was diagnosed with primary lymphedema and was managed by a certified lymphedema therapist with facial/truncal/limb weekly manual lymphatic drainage (MLD) and daytime compression garments. Even though she experienced relief with these measures, the swelling reduction was short lasting, and her symptoms

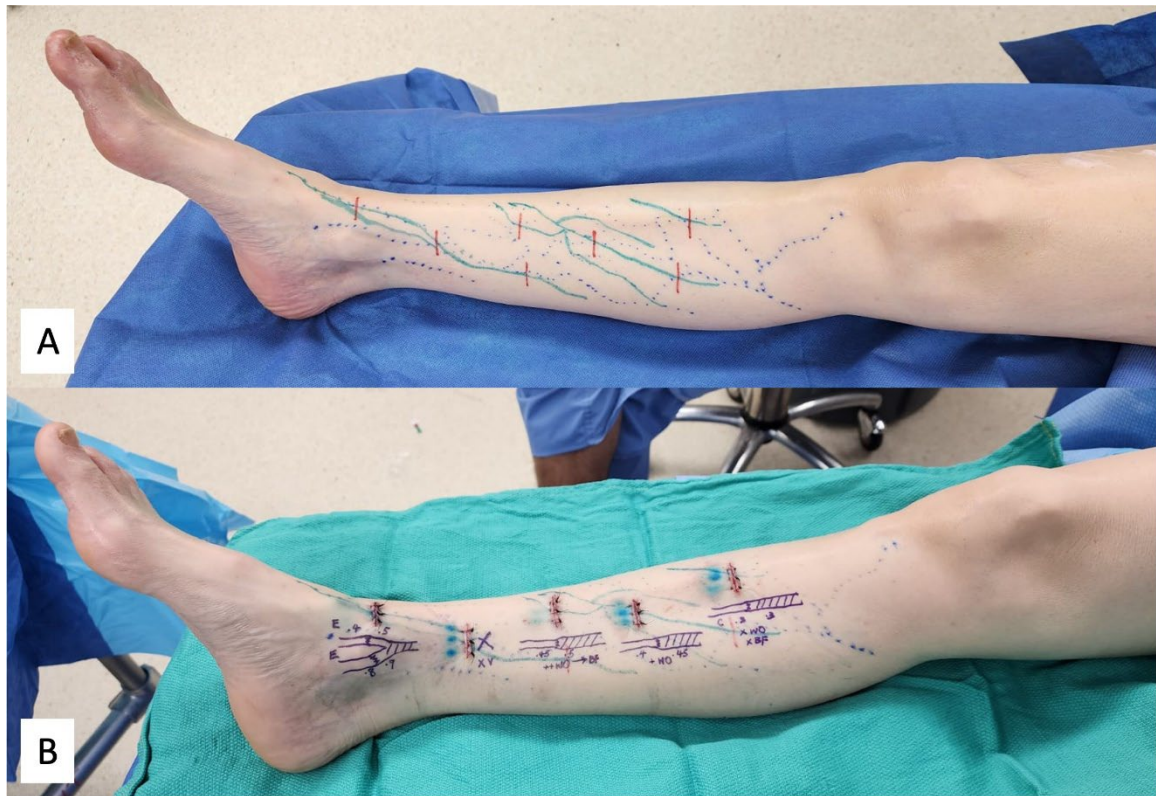


Fig. 2. Case 2: Lymphatic/vein mapping and incision placement (A) were performed as described IN Figure 1. A total of 5 lymphaticovenular drainage pathways were created in 5 exploratory incisions using end-to-end technique (B).

continued to progress.

Indocyanine green lymphography (ICGL) confirmed lymphatic functional deficits across her face and limbs, particularly in the severely compromised left leg (not shown). She underwent supermicrosurgical lymphaticovenular anastomosis (LVA) in her left leg and continued conventional treatment (Figure 1). Postoperatively, she perceived marked reductions in swelling throughout her body, although not objectively measured (data not shown), with the most notable relief from gum pain and the burning sensation. Follow-up assessments at three- and six-months post-surgery showed continued improvements in swelling and symptoms. Objectively, the operated limb volume reduced by 30% at 6 months and ICGL demonstrated accelerated lymph transit, reduced dermal backflows, and the development of new, healthy linear patterns.

At the one-year follow-up, she unexpectedly reported resolution of speech-related difficulties—specifically, problems with word-finding and flow of thoughts. She recalled that before the surgery, her speech symptoms used to worsen when using trunk compression garment, while postoperatively she did not experience those speech issues, regardless of the compression garment use. As these symptoms had not been disclosed during the preoperative evaluation, their improvement could not be objectively assessed but are interesting to consider with systemic involvement of primary lymphedema and its subsequent treatment.

CASE 2

A 62-year-old woman with a history of thyroid cancer treated with thyroidectomy and cervical cancer treated with hysterectomy

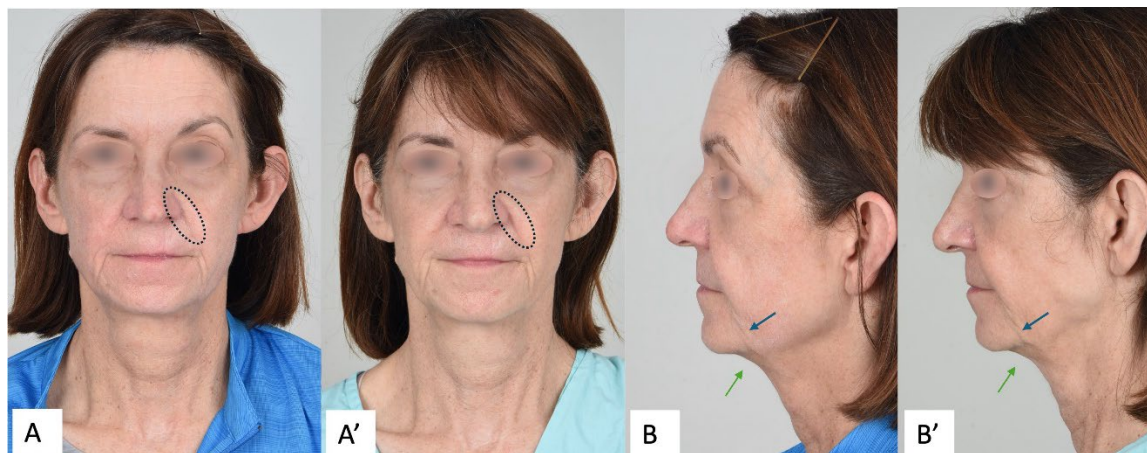


Fig. 3. Reduction in facial swelling can be appreciated when comparing preoperative frontal/profile views (A, B) with those taken at 6th postoperative month (A', B') for the second case. Secondary to swelling reduction, development of skin laxity and ptosis can be seen in periorbital regions and bilateral cheeks. Concomitantly, deepening of the nasolabial folds (dotted oval), prominence of jowling (blue arrow), and blunting of the cervicomental angle (green arrow) indicated reduction in facial volume.

and pelvic lymph node dissection, presented with swelling affecting her face, neck, right arm, and both legs. Her diffuse swelling predated the cancer diagnoses, suggesting a chronic lymphatic dysfunction and a primary etiology. Accompanying her physical symptoms were cognitive challenges, specifically impaired mental focus and memory, which she observed to worsen with facial swelling. The facial swelling was perceived as heaviness and tightness in the face with minimal change in facial appearance. She also reported persistent nasal congestion, frequent episodes of cheek biting, and difficulty in swallowing, indicative of associated mucosal edema. She was undergoing regular manual lymphatic drainage and was using a thigh high compression garment for the right lower limb, with limited relief.

Diagnostic ICGL of face and all limbs confirmed a global lymphatic insufficiency, consistent with the diagnosis of primary lymphedema (data not shown). Treatment strategy focused on her right leg which demonstrated the most serious lymphatic injury and five LVAs were meticulously created through five exploratory incisions (Figure 2). She continued right leg compression in the postoperative period and resumed MLD after

1 month. She reported immediate and significant reduction in swelling across all previously affected areas, including her face. Notably, her mucosal swelling-related functional issues also saw substantial improvement.

At six months of postoperative follow-up, she reported continued and progressive improvements in facial swelling (Figure 3). The operated limb showed 32% volume reduction. In addition to swelling reduction, she also reported enhancement of mental focus and memory. Similar to case 1, these cognitive symptoms were not preoperatively mentioned by the patient and hence could not be objectively assessed but, nonetheless, are interesting and indicate potential far-reaching effects of treatment for primary lymphedema.

DISCUSSION

Our experiences with primary lymphedema, supported by these case studies, suggest the condition's systemic nature, involving a global lymphatic dysfunction rather than being confined to isolated anatomic regions (1,5). This systemic perspective challenges the traditional view of primary lymphedema as a localized condition and highlights the inter-

connected nature of the lymphatic system.

In both cases, performing lymphaticovenular anastomosis (LVA) on a leg resulted in noticeable improvements throughout the body, including areas not directly treated by the surgery. The pronounced relief in non-treated areas, such as the gums in the first case (not objectively measured), underscores the systemic impact of localized surgical interventions. These observations affirm the lymphatic system's unified response to treatment, rather than acting as segregated, regional systems (6).

Furthermore, postoperative cognitive improvements reported by the patients in both cases introduce an intriguing aspect to our understanding of lymphedema's systemic effects, include the brain's lymphatic flow. The improvements in verbal expression, focus, and memory, experienced by our patients, even though self-reported, are not ignorable and merit reporting (7,8). It is perplexing how truncal compression that was applied in Case 1 accentuated the cognitive symptoms and it resolved after surgery. This could reflect pressure changes in the central conducting lymphatic system which impacted the brain's lymphatic drainage.

The absence of initial objective assessments of speech and cognition in our report is an obvious limitation and was due to previously unappreciated association between these symptoms and lymphatic dysfunction. While the findings of this report are suggestive rather than definitive, they underscore the potential systemic benefits of localized lymphatic interventions and pave the way for further exploration into the interconnected nature of the lymphatic system, including its cerebral components. Future studies with pre and postoperative objective data, including validated cognitive scores and brain lymphatics imaging are underway, to further explore this promising frontier in lymphatic research.

Ethical Considerations

Both patients signed consent for release of their clinical information including the photographs, for publication in a scientific journal or meeting. Both patients are covered

by our institutional IRB (22-658) for retrospective study of lymphedema patients.

CONFLICT OF INTEREST AND DISCLOSURE

All authors declare no financial conflicts of interest in any of the products, devices, or drugs mentioned in this manuscript. The study was not financially supported by any grants.

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