

A Successful Outcome after Lymphaticovenular Anastomosis Using a Contralateral Augmented Reality Mapping Technique in a Lymphedema Patient Secondary to a *Mycobacterium abscessus* Infection

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Summary: We report a case of extremity lymphedema after a *Mycobacterium abscessus* infection in a 43-year-old man with anti-interferon- γ autoantibody syndrome. Lymphaticovenular anastomosis was performed using the contralateral healthy or less severe limb as a mirror image to map the lymph vessels in the edematous limb. A satisfying outcome was provided after the surgery. No report of lymphedema caused by this condition has been previously reported in the literature. (*Plast Reconstr Surg Glob Open* 2022;10:e4554; doi: 10.1097/GOX.0000000000004554; Published online 5 October 2022.)

The most common pathogen that causes secondary lymphedema is filariasis. *Mycobacterium abscessus* (*M. abscessus*) is a rare, weakly virulent, nontuberculous mycobacterium human pathogen. It is often found as an opportunistic infection in hosts with genetic defects in the interleukin-12/interferon- γ (IFN- γ) pathway.¹ Therefore, it has a high chance of recurrence and prolonged infection.²

We present a case of extremity lymphedema secondary to a disseminated *M. abscessus* infection in a male patient with anti-IFN- γ autoantibody syndrome and the therapeutic management using contralateral augmented reality mapping technique, and impressive results after surgical treatment.

CASE REPORT

A 43-year-old man presented with a 3-year history of intermittent lymphadenitis and a prolonged fever. The lymph node biopsy and culture indicated the presence of *M. abscessus*. The patient's serological evidence for HIV infection was negative, and he did not disclose a history of immunosuppressant usage. His immunological

analysis revealed the presence of anti-IFN-antibodies. Clarithromycin and ciprofloxacin were administered intravenously and orally to treat the lymphadenitis. Throughout his 10-year follow-ups, he had no incidents of lymphadenitis.

Six months before the patient's recent admission, he developed swelling of both upper and lower extremities especially on the right side. Lymphoscintigraphy revealed a partial lymphatic obstruction. Indocyanine green lymphography (ICG-L) displayed dermal back-flow patterns without visible lymph vessels on the right side of both the upper and lower extremities. However, the left side had a linear pattern.³ Any further infections were excluded due to his clinical presentation and his normal blood and culture parameters. The patient was diagnosed with International Society of Lymphology stage II lymphedema.⁴

In this patient, we performed "mirror lymphaticovenular anastomosis" (mirror LVA) on his right arm and right leg, where ICG-L showed no linear pattern. The healthy contralateral limb was used as a guide. First, we marked the lymphatic vessels of the healthy limb, where ICG-L demonstrated a linear pattern. The picture of the marked vessels was then mirrored and projected onto the affected limb. This method enabled the lymphatic tract to be marked on the affected limb, with the vessel model being adjusted to allow for differences in the limb circumferences (Fig. 1). We also used the usual LVA for the left arm due to its linear ICG-L pattern.

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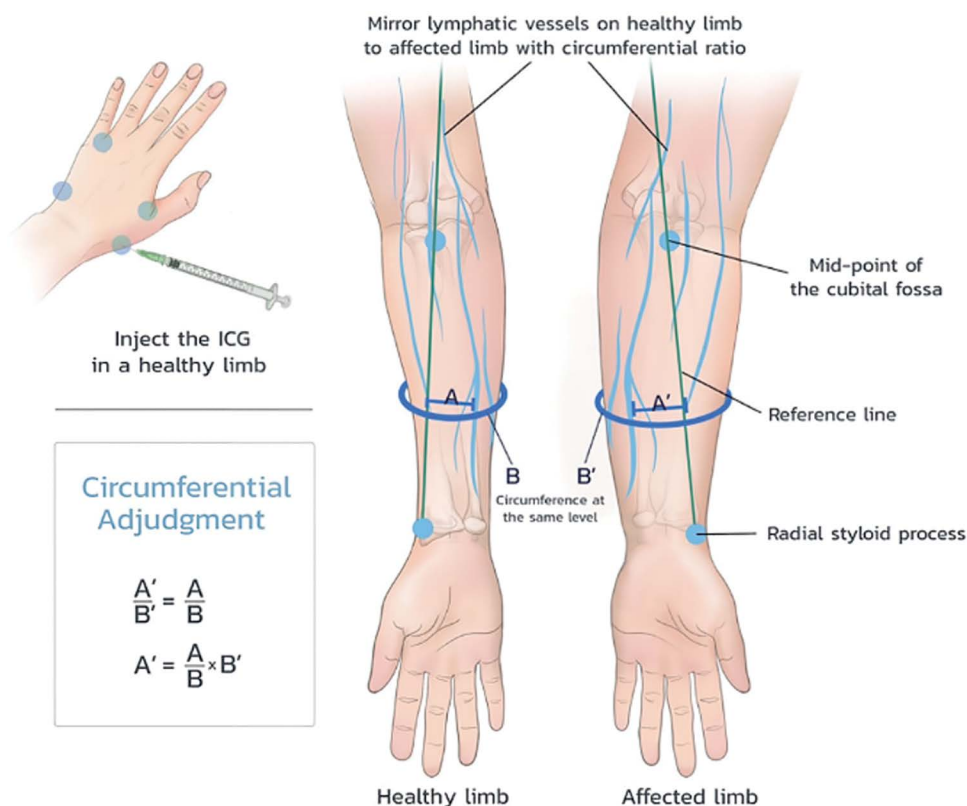


Fig. 1. Preparation for the mirror lymphaticovenular anastomosis. A straight line connecting the reference points was drawn and used as a reference line. This line was marked every 5 cm. The distance was adjusted to fit the swelling limb by using the proportion of circumference of the affected limb and the healthy limb at each level.

Table 1. The Circumferences of the Extremities on the Reference Site and the BMI of the Patient before and after the Mirror Lymphaticovenular Anastomosis Was Performed

Reference Site	Right Side			Left Side			
	Preoperative Circumference (cm)	Postoperative Circumference at 6 Months (cm)	Postoperative Circumference at 18 Months (cm)	Preoperative Circumference (cm)	Postoperative Circumference at 6 Months (cm)	Postoperative Circumference at 18 Months (cm)	
Upper extremity	10 cm above the elbow crease	38	31	32	34	29.5	30
	10 cm below the elbow crease	32	24.5	25	27	23	24
Lower extremity	10 cm above the upper border of the patella	52	44	45	37	37	37.5
	10 cm below the tibial tuberosity	41	35	36.5	32	33	33
Preoperative BMI				Postoperative BMI			
BMI (kg/m ²)	26.26			At 6 months 26.07	At 18 months 25.71		

At the 6- and 18-months follow-up examination, no complications were found. The circumferences of the affected extremities were significantly decreased (Table 1).

SURGICAL TECHNIQUE

For the preparation of mirror LVA, we designated each limb with two landmarks for the upper limb. They

were (1) the cubital fossa midpoint and (2) the midpoint between the radial styloid process and the ulnar styloid process. Two landmarks were chosen for the lower limb: the midpoint of the patella’s top border and the midpoint of the medial and lateral malleoli. To create a reference line that was marked every 5 cm, we drew a straight line linking the points. At each 5 cm distant level, we measured the distance between the reference line and the marked

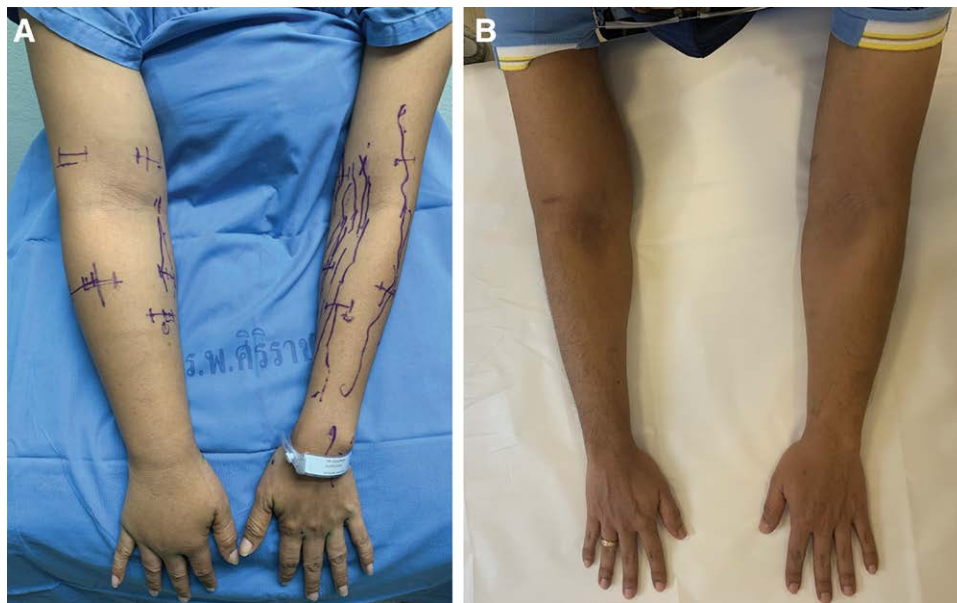


Fig. 2. Preoperative (A) and postoperative (B) pictures of the upper extremities 6 months after the operation.

lymphatic vessels on the healthy limb; this distance was adjusted to match the swollen limb.

Under local anesthesia, we injected 0.2 mL of ICG subcutaneously into each of the four designated places in each leg. Using a near-infrared camera, ICG-L was seen. We used a mobile vein visualizer app (Vein Seek Pro, Los Angeles, Calif.) to find subcutaneous veins close to the intended incision. Where the lymphatic and venous systems intersected or were close to one another, we created incisions. Consequently, each incision was made on a distinct branch of the lymphatic system. A 0.2 mL of 1 percent isosulfan blue was injected immediately proximal to the incision to help locate lymphatic vessel.

An operational microscope was then used to perform LVA through a 2.0 to 3.5 cm skin incision (OPMI Pentaro 900 and OPMI Vario S88 system, Carl Zeiss Co., Jena, Germany). Lymphatic vessels and veins were connected with 11-0 nylon. To operate this technique, end-to-end, end-to-side, side-to-side, and octopus strategies were applied. The patient had 11 anastomoses performed on his extremities, including three anastomoses on his right lower limb and four on each of his upper limbs. (See figure 1, Supplemental Digital Content 1, which demonstrates the intraoperative anastomosis. <http://links.lww.com/PRSGO/C170>) The outcomes of the operations and follow-ups were highly satisfactory (Fig. 2). (See figure 2, Supplemental Digital Content 2, which demonstrates the preoperative and postoperative picture of the lower limb. <http://links.lww.com/PRSGO/C171>).

DISCUSSION

M. abscessus is the most common pathogen proven in disseminated nontuberculous mycobacterium infections

in Thailand (34%).⁸ However, no case of an *M. abscessus* infection that has caused lymphedema has ever been reported in the literature. With our patient with anti-IFN- γ autoantibody syndrome, *M. abscessus* was isolated from his enlarged cervical lymph nodes. It was believed that his lymphedema, which had been found on his extremities, was secondary lymphedema brought on by an *M. abscessus* infection. Other systemic and local causes of the patient's swelling leg were ruled out during the differential diagnosis.⁵

Identifying lymphatic vessels is a crucial step in LVA. Unfortunately, ICG-L cannot detect lymphatic vessels deeper than 1 to 2 cm under the skin. Moreover, the deeper lymphatic vessels are obscured by pathologic dermal backflow. Nevertheless, many authors have reported that lymphatic vessels can still be found by using an anatomical approach. Some follow the anatomy of large veins, while others use the familiar anatomical location.⁶⁻⁸

In our case, we hypothesized that the human body is primarily symmetrical. We surmised that the precision of lymphatic tracking using personalized data and the mirroring technique would be superior to conventional lymphatic tracking using an anatomical approach. Our approach enabled us to identify the hidden lymphatic vessels.

The work of many authors supports the hypothesis that the human body is essentially symmetrical. For example, Tzou et al reported symmetry of the right and left submental lymph nodes. In addition, symmetry of the lymphatic draining pathway of the breast was demonstrated by Blumgart et al.^{9,10}

In 2014, Mihara et al reported satisfying surgical accuracy and efficiency for five severe lymphedema patients with high dermal backflow using ICG-L of the contralateral limb for guidance.⁸ A subsequent study by Servillo et

al (2017) reported contralateral mapping in 16 patients with upper extremity lymphedema.⁹

CONCLUSIONS

We have presented a case of secondary lymphedema from anti-IFN- γ autoantibody syndrome with an *M. abscessus* infection. A mirror LVA using data from the contralateral limb can be helpful in patients with severe dermal backflow in the affected limb. We hope that our case report will contribute to ongoing research regarding the natural cause of this pathogen and the urge to perform more of an innovative surgery.

REFERENCES

1. Chirasuthat P, Triyangkulsri K, Rutnin S, et al. Cutaneous nontuberculous mycobacterial infection in Thailand: a 7-year retrospective review. *Medicine (Baltimore)*. 2020;99:e19355.
2. Hong GH, Ortega-Villa AM, Hunsberger S, et al. Natural history and evolution of anti-interferon- γ autoantibody-associated immunodeficiency syndrome in Thailand and the United States. *Clin Infect Dis*. 2020;71:53–62.
3. Narushima M, Yamamoto T, Ogata F, et al. Indocyanine green lymphography findings in limb lymphedema. *J Reconstr Microsurg*. 2016;32:72–79.
4. Yamamoto T, Yamamoto N, Doi K, et al. Indocyanine green-enhanced lymphography for upper extremity lymphedema: a novel severity staging system using dermal backflow patterns. *Plast Reconstr Surg*. 2011;128:941–947.
5. O'Connor JC, André C, Wang Y, et al. Interferon-gamma and tumor necrosis factor-alpha mediate the upregulation of indoleamine 2,3-dioxygenase and the induction of depressive-like behavior in mice in response to bacillus Calmette-Guerin. *J Neurosci*. 2009;29:4200–4209.
6. Chen WF, Lensing JN, Bowen M. Indocyanine green lymphography can be falsely negative in early extremity lymphedema. *J Plast Reconstr Aesthet Surg*. 2020;73:391–407.
7. Scaglioni MF, Uyulmaz S, Arvanitakis M, et al. Intraoperatively detected but previously indocyanine green-negative lymphatic vessels may have misprized potentials and should not be neglected in lymphaticovenous bypass surgery. *Ann Plast Surg*. 2019;83:69–72.
8. Mihara M, Hara H, Kikuchi K, et al. Scarless lymphatic venous anastomosis for latent and early-stage lymphoedema using indocyanine green lymphography and non-invasive instruments for visualising subcutaneous vein. *J Plast Reconstr Aesthet Surg*. 2012;65:1551–1558.
9. Gentileschi S, Servillo M, Albanese R, et al. Lymphatic mapping of the upper limb with lymphedema before lymphatic supermicrosurgery by mirroring of the healthy limb. *Microsurgery*. 2017;37:881–889.
10. Fujita T, Miura H, Seino H, et al. Anatomical classification of breast sentinel lymph nodes using computed tomography-lymphography. *Anat Sci Int*. 2018;93:487–494.