

# A Case of Traumatic Intractable Leg Ulcer with Lymphorrhea Diagnosed Using ICG Lymphography

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**Summary:** We report a case of traumatic lower extremity ulcers with lymphorrhea in a 37-year-old woman. She presented intractable ulcers without lymphedema for nearly 6 months after a traumatic injury. She was treated conservatively for 6 months at a referral hospital. Unfortunately, during conservative treatment, cellulitis was developed. Even though the inflammation resolved, the ulcer did not heal. We suspected persistent lymphorrhea as the cause of refractory ulcer, and for that reason, we performed indocyanine green lymphography. The lymphatic vessel damage site was identified; also, a collateral lymphatic vessel connected toward the central side was recognized. The lymph fluid drainage site was selectively ligated. The lymphorrhea and ulcers healed 1 month after ligation. Ulcers with lymphorrhea can be persistent and refractory to treatment. Indocyanine green lymphography can be a feasible diagnostic technique. It can identify the site of leakage and is thus useful for treatment. There are fewer reports on the evaluation of lymphorrhea with video presentation. Here, we report a case with video presentation. (*Plast Reconstr Surg Glob Open* 2021;9:e3489; doi: [10.1097/GOX.0000000000003489](https://doi.org/10.1097/GOX.0000000000003489); Published online 23 March 2021.)

## INTRODUCTION

Lymphorrhea can occur after inguinal or axillary lymph node dissection in different urologic, gynecologic, and dermatologic cancers; vascular reconstructive surgeries (such as a reconstructive procedure of the superficial femoral artery); development of serious lymphedema; and more.<sup>1,2</sup> Conventionally, lymphorrhea is treated with conservative methods and/or pressure dressing; however, these therapies are not always effective and may result in prolonged treatment.<sup>1</sup> In recent years, lymphaticovenous anastomosis (LVA) and negative pressure wound therapy (NPWT) have been attracting attention as new treatment methods.<sup>2-4</sup> We encountered a patient with lymphorrhea owing to lymphatic vessel damage after a traumatic injury. Throughout treatment, we could visualize the site of lymph fluid leakage using indocyanine green (ICG) lymphography. Here, we present a case with video presentation of lymphatic leakage without lymphedema, which may be helpful for clinicians involved in the care of patients with lymphorrhea.

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## CASE REPORT

A 37-year-old woman fell in the spring and sustained a bruise on the front right lower extremity. She had no remarkable medical history, including a history of obesity. The lesion became enlarged, motivating her to visit a local doctor after approximately 3 weeks. Puncture, local injection of steroids, and compression therapy were performed; however, there was no improvement. She underwent a biopsy approximately 3 months later. There was no evidence of a tumorous lesion; however, a pseudo-cystic tumor was diagnosed. Treatment was continued, but no improvement was noted. Approximately 6 months after the biopsy, the patient developed cellulitis in her right lower extremity while traveling abroad. She was administered oral antibiotics, but the ulcer did not heal, even though the inflammation resolved. After returning to Japan, she underwent incision and drainage in a local clinic, but the ulcers were refractory, and this motivated referral to our hospital. Several ulcers measuring approximately 1 cm each in diameter on the front right lower extremity, with serous exudates from them, were noted (Fig. 1). Debridement was performed at the first visit to our hospital. Conservative management was continued for approximately 2 months, but still no improvement was observed; therefore, ICG lymphography was performed. First, 0.2 mL of ICG was injected, with local anesthesia,

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**Fig. 1.** A photograph of 37-year-old female patient with right leg anterior ulcer.



**Fig. 2.** Image of the same patient showing ICG fluid drainage from the ulcerated lesion.

subcutaneously into the first interdigital space of the right foot, and imaging was performed using a photodynamic eye (Hamamatsu Photonics, Japan). This method was modified from the method reported by Mihara et al.<sup>5</sup> The results showed that the lymphatic vessels on the dorsum of the foot were directed toward the lesion in the lower extremity, and fluid drainage from the ulcerated lesion was noted. Macroscopically, there was green fluid drainage from the ulcerated lesion (Fig. 2). Lymphatic vessels branched laterally from the dorsum of the foot and the lateral lymphatic vessels connected toward the central side (Fig. 3). After ICG lymphography, we suggested LVA or selective ligation. The patient decided on ligation. The drainage site was ligated under local anesthesia. Approximately 1 month after ligation, the ulcers healed and there was no recurrence at the six-month follow-up (Fig. 4). (See Video [online], which describes the clinical course of a patient having traumatic ulcers with lymphorrhea, from the first examination to treatment completion. The video includes ICG lymphography.)

## DISCUSSION

In the present case, we could visualize the lymphatic vessels and identify lymphorrhea, which led to the healing of the intractable ulcers. In this case, it was assumed that lymphatic vessel damage induced an abnormal accumulation of lymph fluid under the skin, as a lymphocele, leading

to ulceration and persistent lymphorrhea that resulted in an intractable disease. We consider that cellulitis further contributed to the delayed healing of the ulcers.

According to Abai et al., the therapies included bed rest, prophylactic antibiotics, compression dressings, multiple aspirations of the lymph cavity, instillation of sclerosing agents, radiation therapy, NPWT, intraoperative lymphatic mapping and ligation, and musculocutaneous flap closure.<sup>6</sup>

Lv et al. mentioned that “lymphatic leakage is a rare postoperative complication with controversial therapeutic methods. Many scholars devote themselves to finding the best way of treatment. However, so far, there is still no treatment guideline.”<sup>1</sup>

Previous studies have reported promising surgical procedures. Schwartz et al. reported the application of selective ligation.<sup>7</sup> Mihara et al. and Yang et al. reported performing LVA for treating lymphorrhea with lymphedema.<sup>2,4</sup> There are studies reporting the use of NPWT.<sup>1,3,6</sup>

Lymphatic vessel leakage can occur after lymphatic vessel damage irrespective of the etiology (traumatic injury or postoperative complication). The symptoms differ according to the site of injury. To improve the diagnosis and treatment, it is crucial to identify the site of the injury. The modalities used for the diagnosis of lymphatic vessel damage include computed tomography, ultrasonography, magnetic resonance imaging, lymphography, and lymphoscintigraphy.<sup>1</sup>



**Fig. 3.** Right leg lymph vessels mapping.

Unno reported ICG lymphography for lymphedema in 2007. Since then, ICG fluorescence imaging has been used for diagnosis of lymphedema, visualization of lymph vessels, and detection of sentinel lymph nodes.<sup>5,8,9</sup> ICG lymphography is safe and minimally invasive; the device is portable and easy to use, and real-time imaging can be obtained.<sup>8</sup> However, the camera used in the present study limits the detection of lymphatic vessels located at a depth of more than 2 cm from the body surface; therefore, it is difficult to identify deeply located and intra-abdominal lesions.<sup>10</sup>

In our opinion, it is important to identify the site of the leakage in the treatment of lymphorrhea. It is also important to determine whether lymphatic flow is preserved around the damaged lymphatic vessels. When lymphatic flow is preserved, healing can be achieved by closing the injured site, and the main treatment strategy is to facilitate the closure of the leakage site (by compression, ligation, NPWT, etc.). When lymphatic flow is not preserved, there is a need for improvement of the lymphatic flow, which is the main treatment strategy (by LVA, lymphatic vessel transplantation, lymphatic node transplantation, etc.).

### CONCLUSIONS

We have presented a case of intractable ulcers owing to traumatic lymphatic vessel damage with video presentation. ICG lymphography is a useful method for the diagnosis and treatment of lymphorrhea. Lymphorrhea is a



**Fig. 4.** Patient photograph showing the outcome six months after selective lymph duct ligation. The ulcer is closed. There were no recurrence, cellulitis, lymphedema, or other sequelae.

rare condition, and its treatment remains controversial. We hope that this case report will contribute to ongoing efforts regarding the treatment of lymphorrhea.

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