

SEVERE LYMPHEDEMA CAUSED BY REPEATED SELF-INJURY

M. Mihara, H. Hara, N. Murai, T. Todokoro, T. Iida, M. Narushima, I. Koshima

Department of Plastic Surgery/Aesthetic Surgery (MM,HH,TT,TI,MN,IK), University of Tokyo and Department of Vascular Surgery (NM), Saiseikai Kawaguchi General Hospital, Tokyo, Japan

ABSTRACT

Lymphedema is divided into primary and secondary forms. Primary lymphedema often develops in young people and may be caused by lymphovascular aplasia, hypoplasia, and hyperplasia. The most frequent cause of secondary lymphedema after lymphatic filariasis is regional lymph node dissection for treatment of a malignant tumor, and this complication occurs most frequently in middle aged or older patients. Here, we describe a relatively young patient (27 years old) in whom collecting lymph vessels in the upper limb were disrupted by repeated self-injury, with resultant lymphedema. There have been very few reports on lymphedema caused by self-induced trauma. This case report illustrates that secondary lymphedema should also be considered and evaluated appropriately when diagnosed in a relatively young patient without a history of cancer or infection.

Keywords: secondary lymphedema, trauma, depression, self-injury

Lymphedema is generally classified into primary and secondary forms. There are various causes of secondary lymphedema, but the most frequent causes are filarial infection and regional lymph node dissection for a malignant tumor (1,2). Development of lymphedema at a distal site has been reported after extensive burns in the limbs (3,4), radial fracture (5), and injection of a large volume

of silicone (6). However, lymphedema caused by local lymphovascular injury due to trauma other than tourniquet application has not been well described. Here, we report an unusual case of secondary lymphedema that arose at a site distal to trauma caused by repeated self-injury in the limb.

CASE REPORT

The patient was a 27-year-old woman with the chief complaints of severe lymphedema and recurrent cellulitis in the left upper limb. Her medical history included depression but she had no other physical disease or relevant family medical history. The patient had developed depression when she was 14 years old and had engaged in repeated self-injury to the left upper limb since age 23. Painless swelling of the left upper limb occurred at 25 years of age.

The patient visited our department for treatment of lymphedema at 26 years of age, at which time swelling of the upper arm distal to several scars from self-injury on the lower forearm were observed in the left upper limb (*Fig. 1*). Compared to the right upper limb, the circumference of the left upper limb was increased by 12 cm at the center of the upper arm, 9 cm in the elbow, 10 cm in the center of the forearm, and 6 cm in the wrist. No flare was noted, and there were no abnormalities in blood tests or other studies suggesting a collagen vascular disease or a malignant tumor. Secondary lymphedema



Fig. 1. Left upper limb lymphedema at presentation. Several scar wounds caused by repeated self-injury were present in the forearm and upper arm.



Fig. 2. Lymphoscintigraphy findings demonstrate lymph tracer reflux/dispersion in the skin (dermal backflow) throughout the left upper limb.

caused by repeated trauma was diagnosed based on the imaging findings described below.

Echographic findings showed thickening of subcutaneous fat tissue with a cobblestone pattern in the upper arm and forearm. Deep venous thrombosis was ruled out, and there was no edema detected in the muscle layer. Lymphoscintigraphy (*Fig. 2*) showed no apparent obstruction of the main lymph vessel in the left upper limb, and lymph flow was somewhat faster than on the healthy side. However, the radiotracer was visualized throughout the upper limb from the early phase, showing reflux/dispersion of lymph fluid into the skin and subcutaneous tissue. CT findings (*Fig. 3*) showed marked expansion of the subcutaneous fat layer and fibrosis in the left upper limb.

After diagnosis of secondary lymphedema of the upper limb caused by trauma, complex physical therapy using lymph massage and compression stockings was planned. The patient wore a stocking with assistance several times but subsequently stopped wearing it because her left upper limb became paralyzed. She received lymph massage once a month for 1 year as an outpatient. The circumference of the arm has not improved but aggravation of edema has ceased (that is, the current condition has been maintained).

DISCUSSION

We encountered a patient with secondary lymphedema caused by trauma. Lymph node dissection is the most frequent cause of secondary lymphedema of the limbs, and there have been few previous reports of lymphedema caused by local lymphovascular trauma. Normally, a single incised wound rarely causes peripheral lymphedema because multiple collecting lymph vessels are distributed in parallel in the four limbs. However, our patient engaged in repeated self-injury of the four limbs, which may have gradually damaged several lymph vessels beyond repair and caused lymphedema. Therefore, this case indicates that strong

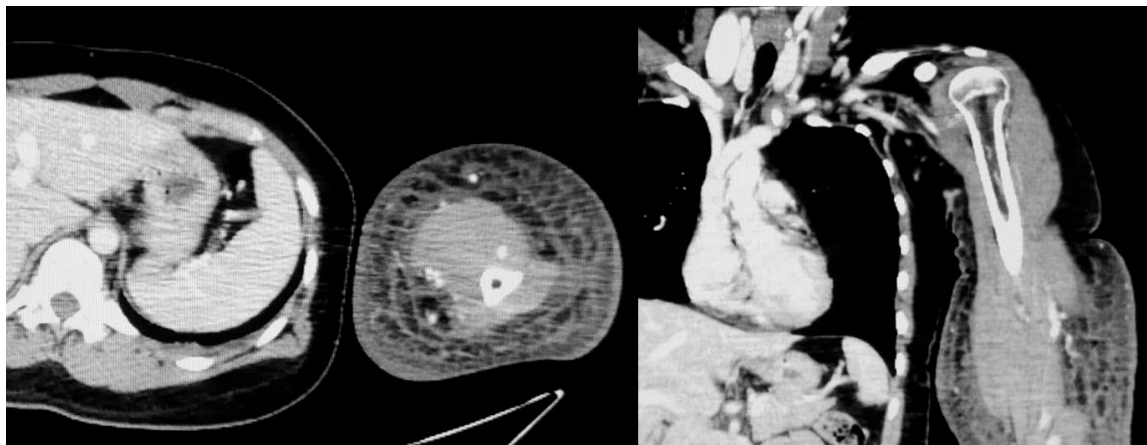


Fig. 3. CT findings in cross-sectional (left) view and coronal (right) views show expansion of the subcutaneous fat layer and fibrosis throughout the left upper limb.

contusion of regions with densely distributed collecting lymph vessels may cause lymphedema.

Lymphedema in young patients is frequently diagnosed as idiopathic lymphedema. Treatment for idiopathic lymphedema includes compression therapy using elastic stockings, complex physical therapy including lymph drainage, and surgical treatment such as lymphaticovenous anastomosis (LVA) and lymph node transplantation. However, treatment of idiopathic lymphedema is not sufficiently effective in many cases, compared to that of secondary lymphedema, since the condition is often related to lymphovascular aplasia or hypoplasia (7,8). Possible involvement of thoracic duct dysplasia in idiopathic lymphedema has also been suggested (9). However, a careful medical history by interview, as performed for our patient, may indicate that cases previously diagnosed as idiopathic lymphedema may actually be caused by trauma including self-induced. Such lymphedema caused by traumatic lymph vascular injury may need to be treated not only by control of the psychiatric condition but with LVA and remitted by preparing a drainage pathway bypassing the injured lymph vessel(s) (10-13). Therefore, careful investigation of the

medical history by interview is crucial for accurate diagnosis and subsequent design of a treatment plan in younger patients with lymphedema of the limbs.

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Makoto Mihara, MD
Assistant Professor
Department of Plastic and Reconstructive
Surgery
University of Tokyo
7-3-1, Hongo, Bunkyo-ku
Tokyo, Japan 113-8655
Tel: 81-03-3815-5411
Fax: 81-03-5800-6929
E-mail: mihara@keiseigeka.name