



Unilateral lower limb lymphedema resulting from a heart surgery performed 50 years prior



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ABSTRACT

INTRODUCTION: Lymphedema is classified into two main types: secondary lymphedema accompanied by lymph node excision surgery or radiotherapy; and idiopathic lymphedema. Here we experienced a very rare case of lymphatic edema resulting from cardiac surgery that the patient underwent 50 years previously.

PRESENTATION OF CASE: A 62-year-old woman experienced progressive unilateral lower leg lymphedema for recent years. After undergoing cardiac surgery at another hospital at the age of 12 years, she gradually developed left lower leg edema. The cause of the edema was unclear and it remained untreated. Her edema symptoms gradually worsened in recent years, so she consulted the plastic surgery division of our hospital.

DISCUSSION: Perhaps the lymphatic structures of affected individuals differ prior to disease onset. If the mechanism of lymphatic edema outbreak is elucidated, patients needing conservative and surgical therapy might be more easily identified. Knowing the outbreak mechanism of lymphatic edema would definitely ease the investigation of an unconventional case like this one. Conservative treatments for lymphedema, such as self-massage and compression therapy using garments, were immediately started. With these treatments, the leg volume and edema symptoms reduced.

CONCLUSION: The research on the cause of this case may be important step for elucidating the source of secondary lymphatic edema.

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1. Introduction

Creating an experimental animal model of lymphatic edema is difficult [1]. Thus, questions remain about its outbreak mechanism, pathophysiology, and clinical course. Additionally, a consensus on the best treatment approach (surgical or nonsurgical) has yet to be reached. Lymphaticovenular anastomosis is widely performed in patients with early- to late-stage lymphedema. We previously reported the use of preventive multiple lymphaticovenular anastomosis at the ankle for the treatment of early lymphatic edema that maintained lymphatic function [2].

Lymphedema is classified into two main types: secondary lymphedema accompanied by lymph node excision surgery or radiotherapy; and idiopathic lymphedema. The detailed examination of atypical outbreak lymphedema cases not treated with lymph node excision surgery plays an important role in providing clues about its underlying cause. To our knowledge, there have been no reports of lymphedema resulting from heart surgery. Here we

experienced a very rare case of lymphatic edema resulting from cardiac surgery that the patient underwent 50 years previously. In this report, we report the patient's therapeutic course and attempted to determine the cause of the lymphatic edema outbreak.

2. Clinical case

A 62-year-old woman experienced progressive unilateral lower leg lymphedema for many years. She had undergone heart surgery (details unknown) at another hospital at the age of 12 years, and her cardiac condition improved. However, left lower leg edema developed immediately after the surgery. She had not experienced any edema symptoms at birth and before the heart surgery. Her edema symptoms gradually worsened over the past few years; therefore, she consulted the Plastic Surgery Division at our hospital. The initial diagnosis was unilateral lymphedema of the left leg, and the degree of lymphedema was found to be Campisi clinical stage II. She had a history of cellulitis. The measurements of both lower extremities at initial diagnosis were as follows (right/left [cm]): dorsalis pedis, 21/21; ankle, 21/24; 10 cm below the knee joint, 32.5/36; knee joint, 33/35; and 10 cm above the knee joint, 37/43. A star-dust pattern on indocyanine green lymphography was seen from the left dorsalis pedis to the left thigh. In contrast, a normal linear pattern was observed in the right lower leg. Lymphatic scintigraphy

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Fig. 1. Lymph duct scintigraphy. The left foot shows prepotent radiological isotope accumulation, while the right foot shows normal findings.

showed that the left leg lymphatic duct broke off near the center of thigh, and the central lymphatic course could not be confirmed. Also, the left lower leg was diffusely depicted, whereas normal lymphatic course was seen in the right foot. Lymphangiography using indocyanine green showed that the entire left leg diffuse pattern and right leg normal linear pattern. These findings confirmed the diagnosis of left lower leg lymphedema (Fig. 1). Conservative treatments for lymphedema, such as self-massage and compression therapy using garments, were immediately started. With these treatments, the leg volume and edema symptoms reduced. Two years after the start of conservative treatments, the measurements of both lower extremities were as follows (right/left [cm]): dorsalis pedis, 19.5/20.5; ankle, 18.5/20.5; 10 cm below the knee joint, 31.5/32.5; knee joint, 31.5/32.5; and 10 cm above the knee joint, 37/39.5. Currently, the degree of lymphedema is Campisi clinical stage Ib (Fig. 2).

3. Discussion

Here we report a case of unilateral lower limb lymphedema resulting from a heart surgery. Most cases of secondary lower leg lymphatic edema are caused by a lymphadenectomy of the inguinal lymph node or lymph nodes in the pelvis caused by gynecologic or urologic cancer.

But this is a case of secondary lymphatic edema in which a lymphadenectomy was not performed. In addition, there was no

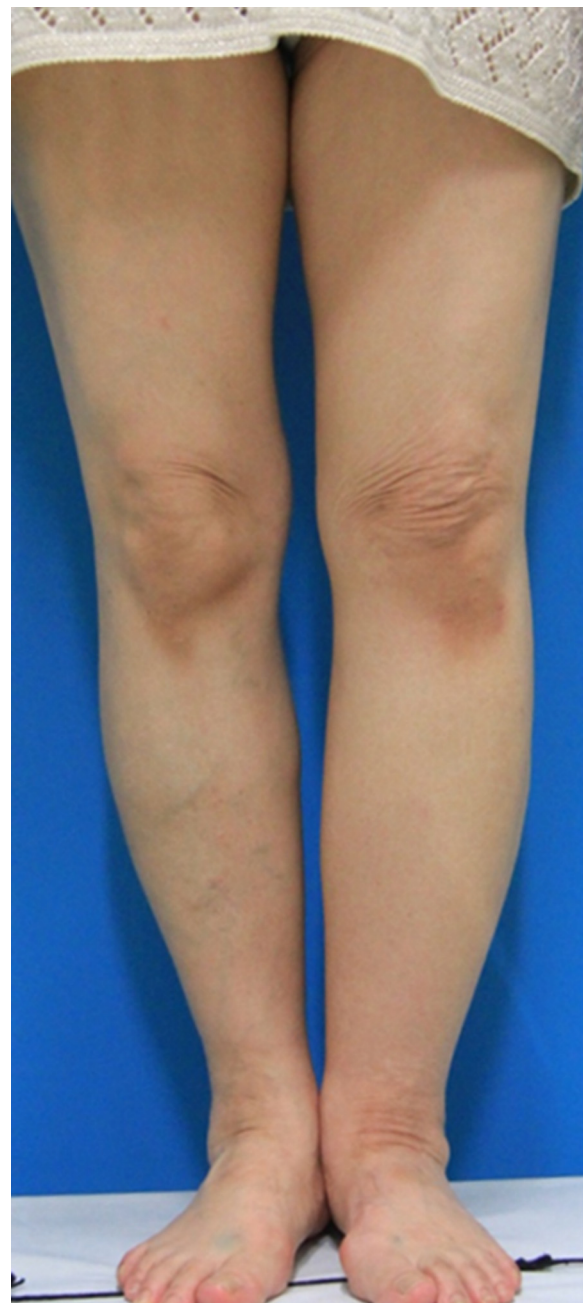


Fig. 2. Findings of the left lower leg after treatment. Unilateral edema is noted; however, typical findings, including warmth and redness, are lacking. The cutaneous state is good.

abdominal or groin surgery history. Thus, it is thought that the mechanism of the development in this case differed from that of usual cases of secondary lymphatic edema. We believe that the lymphedema in the present case resulted from the previous heart surgery because edema symptoms developed immediately after the surgery. Although the operative details are unknown, we suspected that infectious pericarditis without performing open-heart surgery, considering that the patient underwent surgery 50 years prior. Thus, it is easy to believe that this case of edema was derived from thoracic lymph duct injury or stenosis resulting from the operative procedure. In addition, individual cases vary in degree, time to edema outbreak, and progression speed. In some cases, primary lymph edema is caused by slight injury, after which edema symptoms worsen. With regard to the lymphatic system, various

anatomical examinations were accomplished in the past, but many questions remain about the outbreak and aggravation of lymphatic edema [3–5]. Perhaps the lymphatic structures of affected individuals differ prior to disease onset. If the mechanism of lymphatic edema outbreak is elucidated, patients needing conservative and surgical therapy might be more easily identified. Knowing the outbreak mechanism of lymphatic edema would definitely ease the investigation of an unconventional case like this one.

Conflicts of interest

The authors have no financial interest to declare in relation to the content of this article.

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Ethical approval

This study is not applicable.

Consent

Written informed consent was obtained from the patient for the publication of this case report and accompanying images.

Author contribution

Satoshi Onoda: performance of the treatment.
Yuki Miura: performance of the treatment.
Narushi Sugiyama: performance of the treatment.

Guarantor

We (Satoshi Onoda, Yuki Miura, Narushi Sugiyama), the author of Correction of Unilateral Lower Limb Lymphedema Resulting From a Heart Surgery Performed 50 Years Prior. We have read the current “wide for authors” of the international Journal of surgery case report.” The manuscript complies with the instruction and condition specified in the “Guide for Author.” We have examined the manuscript; We have agree to its submission with my name in authors’ list and take responsibility for the submission. We assure the material is original. The context of the manuscript has neither been published nor being submitted for publication simultaneously. If accepted, the manuscript will not be published elsewhere in the same or similar form without written consent from the copy-right holder.

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None.

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