INTRODUCTION

Lymphedema is a chronic and progressive condition, resulting from an imbalance between protein-rich interstitial fluid formation and its drainage from the tissues. This imbalance may be caused by a congenital alteration of form or function of the lymphatic vessels or nodes (primary lymphedema), or by a physical disruption of the lymphatic system due to cancer resection surgery, radiotherapy, trauma, or parasites (secondary lymphedema). For a long time this disease was thought of as incurable or refractory; and even though it is a common condition, there is still a great mystery around its causes and treatment. Many patients remain unaware that there are now different treatments available, some receive wrong information about the disease and how to treat it or even inadequate treatments, which may lead to rapid progression of the condition, depression, and frustration.1

Treatment can be divided into 2 main groups: conservative and surgical. Lymphedema has traditionally been treated conservatively by complex decongestive therapy including skin care, manual lymph drainage, and sustained compression with elastic stockings2 to suppress the progression of edema in the upper or lower limbs, but up to 40% of the patients have been reported to advance to intractable lymphedema nonresponsive to medical treatment.3,4 Regarding surgical treatment, Yamada5 in 1969 proved the effectiveness of lymphatic venous anastomosis in a series of canine models to reduce the edema of the limbs and reported the first clinical application of this procedure in an orthopedic patient with postoperative lymphedema. Since the 1970s multiple surgeries have been proposed to reduce lymphedema, which can be divided into functional procedures like lymphatic grafts, multiple lymphaticovenular anastomoses (LVAs), or vascularized lymph node transfers, and resectional procedures like liposuction of specific areas or excisional approaches of the affected tissue. All these surgical procedures have been raised as an attempt to partially or totally treat this condition, and they have significantly modified the pathophysiologic evolution of the disease and the psychosocial behavior of the patients.3

In Japan, Professor Isao Koshima started investigating the function of lymphatic vessels; the pathophysiologic evolution of lymphedema in human extremities;6 the behavior of the lymphatic system in different body areas, and the effect of lymphaticovenular anastomosis in the limbs.7 Thus, he became a leading exponent of surgical lymphedema treatment, and his efforts led to the successful treatment of secondary lymphedema in Japan.8-10

Summary: Although a large number of treatments have been developed for lymphedema, definitive management still remains a challenge for plastic surgery, because not all surgical techniques have a positive effect on every patient, and good results are difficult to reproduce. However, it is important to recognize the efforts that Professor Isao Koshima and his colleagues together with the Government of Japan are carrying out to spread information to the different countries on the newest surgical techniques available in current plastic surgery and to motivate young doctors in developing countries to create a microsurgery unit in their home country, providing them with the necessary knowledge and skills to deliver real solutions to complex problems that were previously unanswered. As a result of this program, the authors present the first case of secondary lymphedema successfully treated with lymphaticovenular anastomosis in Costa Rica, Central America. (Plast Reconstr Surg Glob Open 2018;6:e1689; doi: 10.1097/GOX.0000000000001689; Published online 27 February 2018.)

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soft tissues after LVAs in upper extremities, and their effectiveness in advanced lymphedema. LVAs have been performed by Koshima since the 1980s for the treatment of primary or secondary lymphedema with very good results. They can be performed under local anesthesia in adults, are minimally invasive, and eliminate the possibility of donor-site lymphedema after lymph node transfers.

Later on he started associating LVAs to functional procedures like vascularized lymph node transfers and resectional procedures to obtain better results. This knowledge and the high volume of patients evaluated at the University of Tokyo Hospital led to the development of a 6-month advanced surgical training program to educate young plastic surgeons from all over the world in microsurgery and supermircosurgery techniques.

JAPANESE CONTRIBUTION TO MICROSURGERY DEVELOPMENT

At the Plastic and Reconstructive Surgery Department of the University of Tokyo Hospital, a 6-month Fellowship Program for foreigners has been developed in the last few years in conjunction with the Department of Health, Labour and Welfare of Japan, to cooperate with young doctors from different countries interested in learning microsurgery and supermircosurgery techniques.

The foreign doctors who apply for this program should have Plastic and Reconstructive Specialty and English language certifications. Once they meet the criteria and are accepted in the program, they should buy an insurance policy and apply for a Certificate of Permission of Advanced Clinical Training, which will be granted approximately 7 weeks after their arrival to the Hospital. In the meantime, they are allowed as observers in the operating rooms of the Department; they must attend and actively participate in all the conferences, lectures, article club meetings, and outpatient clinics. Although they wait for the Certificate of Permission, they are authorized to work at the Animal Laboratory at least once a week to perform multiple microvascular anastomoses on rats and perfect their microsurgical technique to help them achieve movement fluidity and surgical excellence. Once the Certificate of Permission arrives, they can participate in surgery where they are evaluated by the different surgeons and professors of the Department to determine their microsurgical abilities at the time and if considered sufficient, they are then allowed to assist the surgeons, encouraging the learning doctors to perform as many as possible supervised arterial, venous, and lymphatic anastomoses, which are very important to their final training. They are also encouraged to present surgical cases in International Conferences and Congresses held at the time they are in Japan.

The main goal of this program was to shape every foreign doctor with enough knowledge, confidence, and surgical skills to go back home and be able to develop a microsurgery unit in his/her hospital, based on the experience acquired in the 6-month visit.

In April 2014, the first female foreign doctor, Madelein Centeno-Rodríguez from Costa Rica, Central America, started the program. She finished her training in September 2014 and went back to the San Juan de Dios Hospital, where she was able to perform the first successful surgical treatment of secondary lymphedema in Costa Rica with LVAs. The case is presented below.

SURGICAL TREATMENT OF SECONDARY LYMPHEDEMA

First Case in Costa Rica

A 35-year-old female patient who was diagnosed by lymphoscintigraphy in March 2015 with stage II, 11-month-onset secondary lymphedema of her lower left extremity as a result of sequelae from hysterectomy, regional lymph node dissection, and subsequent radiotherapy because of cervical cancer treatment in 2013, was referred to the Plastic and Reconstructive Surgery Department of the San Juan de Dios Hospital in San José, Costa Rica, and evaluated on April 10, 2015.

On July 6, 2015, she became the first patient in Costa Rica to undergo surgery for the treatment of her secondary lymphedema. Because the San Juan de Dios Hospital does not have a photodynamic eye or indocyanine green to determine the exact location of lymphatic vessels, four 3-cm skin incisions were performed under general anesthesia in the region along the left great saphenous vein at the dorsum of the foot, at the lower leg, at the upper leg and lower thigh, but only 2 sites showed suitable lymphatic collectors for LVAs: at the dorsum of the foot and

Fig. 1. A, Microscopic view of the LVA performed at the dorsum of the left foot. B, Microscopic view of the LVA performed at the distal left leg.
at the lower leg. Two LVAs were performed: a termino-terminal anastomosis of a 0.8 mm lymphatic vessel to a 1.1 mm superficial vein at the distal left leg and a termino-terminal anastomosis of a 0.8 mm lymphatic vessel to a 1.2 mm superficial vein at the dorsum of the left foot. Both lymphatic vessels used for the anastomosis showed normal lymph drainage, no sclerosis or ectasia. Once both anastomoses were performed, they showed no backflow of blood and adequate lymph washout. The lymph vessel used at the dorsum of the foot showed some fibrosis of the wall. Figures 1A and B show microscopic views of both LVAs performed.

RESULTS
The patient had an uneventful recovery and was discharged the day after surgery. She was advised to wait 1 week before using her compression garments. She presented a gradual and considerable reduction of circumference measurements of the left lower limb and softening of the edematous tissue. The preoperative and 10-month postoperative anterior and posterior images are shown in Figure 2. Figure 3 shows the difference in preoperative and postoperative lower limb circumference measurements.

DISCUSSION
Secondary lymphedema is usually caused by injury to the lymphatic vessels by surgery, radiation, trauma, and other disease processes, and this type of lymphedema is 1 of the main indications for lymphaticovenous anastomosis.11 Because the lymphatic pathways are considered normal, the basic need in these cases is to create an artificial exit to the lymph forming in the affected limb before the excessive constant pressure ends up dilating and eventually degenerating the lymphatic channel smooth muscle cells, damaging the propulsion movement of the collecting lymphatics.3

LVAs are bypasses between a lymphatic vessel and a superficial vein to create an escape route for the lymph in an attempt to reduce the edema of an affected limb. They require a highly sophisticated microsurgical technique called supermicrosurgery because the vessels are usually between 0.5 and 0.8 mm in diameter.3 This type of anastomosis needs a specially trained surgeon, a surgical microscope, supermicrosurgical instruments, and suture materials.

Costa Rica is a small country located in Central America. Despite being a developing country, its health system is privileged for the region. The social security system is universal and is administered by the Costa Rican Department of Social Security (in Spanish Caja Costarricense de Seguro Social). Although this health system ranked 36 according to the World Health Organization’s World Health Report 200012 (United States ranked 37), it presents certain shortcomings for the management of lymphedema such as limited budget for the purchase of medical equipment, and absence of indocyanine green in the national drug profile; there is no photodynamic eye or magnetic resonance lymphangiography for real-time diagnosis of disorders of the lymphatic system, and there are neither medical specialists in the management of lymphatic pathologies in the country nor formal education in microsurgery or supermicrosurgery. It is for these reasons that international education and Japanese collaboration play a leading role in providing young doctors in developing countries with the opportunity to take on new horizons to learn the latest advances in microsurgical techniques to return to their country of origin to improve the quality of life of their patients.

The ultimate goal of this program was for each visiting surgeon to become a change agent in his/her country

![Fig. 2. Preoperative and 10-month postoperative anterior and posterior views.](image-url)
by creating a unit of microsurgery and supermicrosurgery that will contribute to the search for advanced answers to different pathologies. This would raise the level of resolution of his/her base hospital, allowing more uniform international growth in microsurgery for the management of complex diseases such as lymphedema.

In this case, the authors present a patient with secondary lymphedema treated with the first LVAs in Costa Rica obtaining a successful outcome, based on the valuable knowledge acquired at the University of Tokyo Hospital from Professor Koshima and his colleagues.

CONCLUSIONS

Professor Isao Koshima and collaborators at the Plastic and Reconstructive Surgery Department of the University of Tokyo Hospital in conjunction with the Department of Health, Labour and Welfare of Japan have created a 6-month advanced surgical training program to educate young plastic surgeons from all over the world in the latest advancements in microsurgery and supermicrosurgery techniques to treat complex pathologies like lymphedema. As a result of this program and the Japanese contribution, it was possible for Dr. Centeno-Rodríguez to successfully perform LVAs on a female patient with secondary lymphedema for the first time in Costa Rica, Central America. It is the ambition of the program to help many other countries to achieve surgical excellence in the area of microsurgery and supermicrosurgery.

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REFERENCES

1. Champaneria MC, Neligan PC. Lymphedema: lack of solutions to a clinical problem. In: Neligan PC, Masia J, Piller NB, eds. Lymphedema: Complete Medical and Surgical Management. Boca Raton, Fla.: Taylor and Francis Group; 2016:3–23.

2. Hara H, Mihara M, Ohtsu H, et al. Indication of lymphatico-venous anastomosis for lower limb primary lymphedema. Plast Reconstr Surg. 2015;136:883–893.

3. Koshima I, Narushima M, Yamamoto Y, et al. Recent advancement on surgical treatments for lymphedema. Ann Vasc Dis. 2012;5:409–415.

4. Campisi C, Boccardo F, Zilli A, et al. Peripheral lymphedema: new advances in microsurgical treatment and long-term outcome. Microsurgery. 2003;23:522–525.
5. Yamada Y. The studies on lymphatic venous anastomosis in lymphedema. Nippon J Med Sci. 1969;32:1–21.

6. Koshima I, Kawada S, Moriguchi T, et al. Ultrastructural observations of lymphatic vessels in lymphedema in human extremities. Plast Reconstr Surg. 1996;97:397–405; discussion 406.

7. Koshima I, Inagawa K, Urushibara K, et al. Supermicrosurgical lymphaticovenular anastomosis for the treatment of lymphedema in the upper extremities. J Reconstr Microsurg. 2000;16:437–442.

8. Koshima I, Nanba Y, Tsutsui T, et al. Long-term follow-up after lymphaticovenular anastomosis for lymphedema in the leg. J Reconstr Microsurg. 2003;19:209–215.

9. Koshima I, Inagawa K, Etoh K, et al. [Supramicrosurgical lymphaticovenular anastomosis for the treatment of lymphedema in the extremities]. Nihon Geka Gakkai Zasshi. 1999;100:551–556.

10. Koshima I, Nanba Y, Tsutsui T, et al. Minimal invasive lymphaticovenular anastomosis under local anesthesia for leg lymphedema: is it effective for stage III and IV? Ann Plast Surg. 2004;53:261–266.

11. Maegawa J. Lymphatic-venous anastomosis. In: Green AK, Slavin SA, Brorson H, eds. Lymphedema: Presentation, Diagnosis, and Treatment. Cham, Switzerland: Springer International Publishing; 2015:255–268.

12. World Health Organization. The World Health Report 2000. Available at http://www.who.int/whr/2000/en/whr00_en.pdf. Accessed October 21, 2017.