The THAI Journal of SURGERY 2017;38:39-45.
Official Publication of the Royal College of Surgeons of Thailand

Case Report

A Case Report of Severe Lymphedema of Lower Limb: Surgical Management

Ruch Wongtrungkapun, MD

Department of Surgery, Bamrasnaradura Infectious Disease Institute, Ministry of Public Health, Nontaburi, Thailand

Abstract

Background: **Secondary lymphedema usually resulted from damage of the lymphatic system. Common causes include surgery, trauma, radiation, or infection. One obvious example is arm edema after breast surgery or radiation treatment of some cancers. Currently, there was no definite treatment for lymphedema. Treatment goal is to restore function, reduce physical and psychological suffering, and prevent the development of complications. There are two alternative approaches for surgery: reconstructive surgery and destructive surgery.**

Objective: **To report a successful lymphedema case treated with debulking surgery of lower limb lymphedema.**

Method: **A case report**

Result and Conclusion: **Elephantiasis can divide into two types; primary and secondary. Both pathogenesis ended up with persistent edema of the tissue. Conservative measures are usually not curative for elephantiasis, while the surgery will help to restore patient's function. There are two types of surgery for elephantiasis; reconstructive of lymphatic structure and destructive debulking surgery. In this case report, debulking surgery can be an adjuvant technique for elephantiasis.**

Keywords: **Lymphedema, elephantiasis, debulking, stage surgery**

Introduction

The extensive network of lymph vessels drains out lymphatic fluid from any region of the body. Lymphedema is an abnormal collection of this high-protein fluid under the skin. This symptom usually occurs in the arm or leg. The pathogenesis of lymphedema is damage of the lymphatic vessels or destruction of lymph nodes (secondary lymphedema)[1]. Occasionally this condition occurs from impaired of lymphatic vessels or primary lymphedema. The condition has to differentiate from other type of edema such as cellulitis or water edema in volume overload[2,3]. A synonym of lymphedema is elephantiasis which is the common term used for lymphedema caused by filariasis infection.

Secondary lymphedema usually resulted from damage of the lymphatic system. Common causes include surgery, trauma, radiation, or infection. One obvious example is arm edema after breast surgery or radiation treatment of some cancers. Lymphedema has a number of stages, from mild to severe: stage 0 (non-visible, latency), stage 1 (spontaneously reversible), stage 2 (spontaneously irreversible), and stage 3 (lymphostatic elephantiasis)[2,3].

Correspondence address: *Ruch Wongtrungkapun, MD, 126 Bamrasnaradura Infectious Disease Institute, 4th Floor, 2nd Building, Soi Tiwanon 14, Tiwanon Road, Nontaburi 11000, Thailand; Telephone: +66 2590 3408; Fax: +66 2590 3411; Email: ruch_wongtrungkapun@yahoo.com*

Compromise of the lymphatic drainage can cause many complications such as skin problem, infections, discomfortability, and may be severe as elephantiasis may cause immobilization of the patient[2,4]. Currently, there was no definite treatment for lymphedema. Treatment goal is to restore function, reduce physical and psychological suffering, and prevent the development of complications. General recommendations for treatment are hygiene and skin care, physical therapy and compression. The last choice of treatment is surgery. There are two alternative approaches for surgery: reconstructive surgery and destructive surgery[5,6].

This paper reported a successful debulking surgery of lower limb lymphedema in severe left lower limb lymphedema patient by chronic leg infection. The study reviewed the cause of the lymphedema and point to consider during the destructive surgery.

Material and Method

This study represented a case report which reviewed the chart and operative records of one patient with elephantiasis left leg from chronic infection and left leg surgery who was transferred to Bamrasnaradura Infectious Diseases Institute. The 8-month hospital history was reported. Patient's informed consent was obtained. The report was approved by BIDI EC. The approval number is S015/59_Exampt (date Aug 13, 2016).

A Case Presentation

An obese 43-year-old Thai woman, who is a farmer in rural area, was referred to Bamrasnaradura Infectious Diseases Institute (BIDI) from Songkhla province with lymphedema of left leg (Figure 1).

She went to hospital with chronic venous ulcer on her left leg. She has underlying obesity and hypertension. She has been treated for superficial varicose veins for a half year. After she had got the venous ulcer in the gaiter area, her doctor performed a surgery. High ligation with venous stripping was done. Her left leg had infection on and off after the surgery. Her left leg started to get bigger and she was diagnosed with lymphedema grade III.

On physical examination, she is obese with severe left leg swelling (non-pitting edema). Her heart, lung, abdominal, and neurologic exams were normal. During her course in BIDI, she had undergone many investigations for lymphedema. Microfilaliasis was negative. Her other laboratory studies were as follows: Hct 31% ; Hgb 10.1mg% ; WBC 4,400 cumm (N 74%, Lym 15%); Bun 18 mg/dl; Cr 0.84 mg/dl; Na+ 136 mmol/l; K+ 3.6 mmol/l; Cl 97 mmol/l; CO2 29 mmol/l; Urine exam: no cells, WNL; EKG: HR 82/min, Normal sinus rhythm; and CXR: WNL

The diagnosis was: lymphedema from chronic infection and surgery with acute infection on top grade III; with obesity and hypertension.

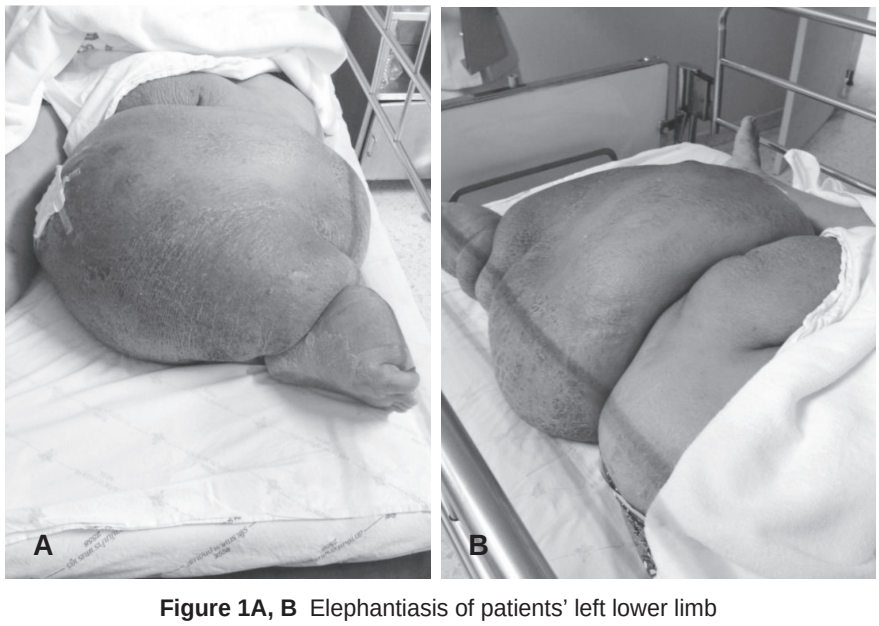

Figure 1A, B Elephantiasis of patients' left lower limb

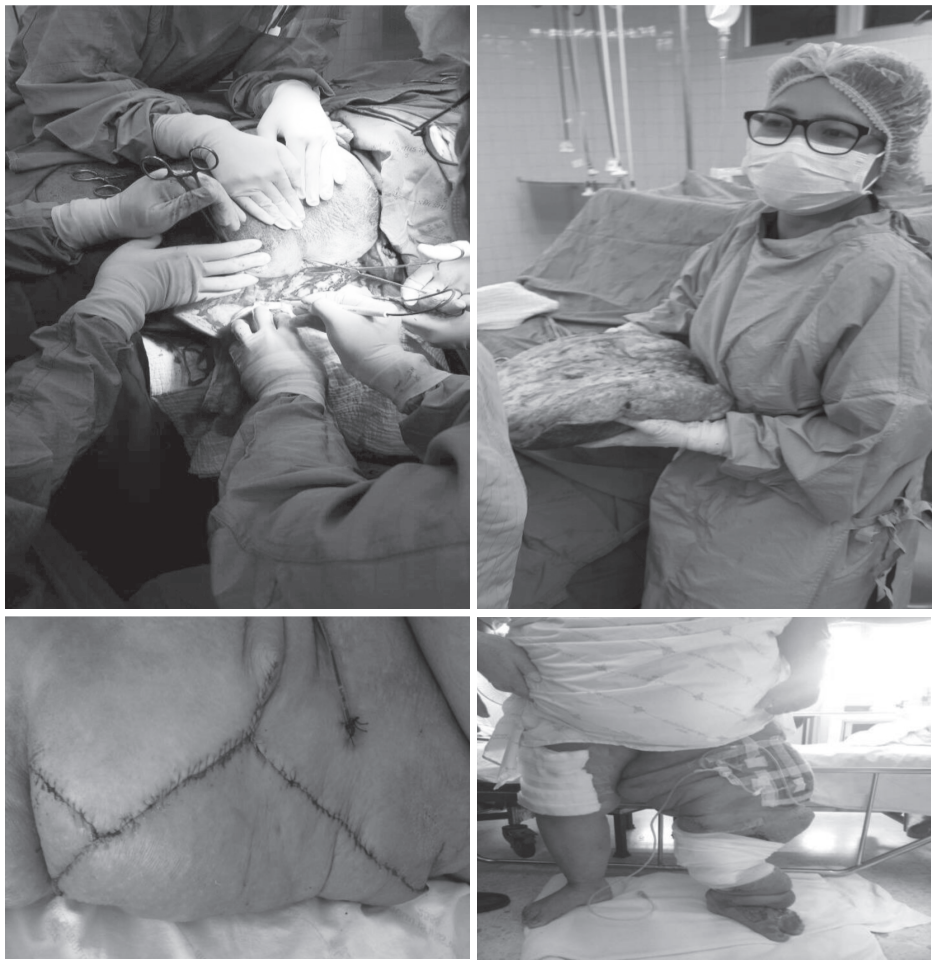

Figure 2 **A** Operating picture of patient's leg
B Debulking soft tissue from patient's leg
C Patients' leg flap wound after excision of soft tissue
D Patients' figure after stage debulking procedures

Treatment

She had been treated for infection with supportive measures such as compressive dressing, passive exercise, and warm compression. The main purpose of this period was to control infection and strengthen the physical status of the patient before surgery.

Planned stages debulking of subcuticular connective tissues were done for several times. Finally she started to walk as shown in Figure 2D.

Surgical technique for stage debulking procedure

1. Pre-operative planning

In preoperative planning, patient's physical status check up and medical consultation were done to ensure that the patient was fit for stage operation. Physiotherapist designed to improve the texture and consistency of patient's soft tissue in the lower extremity. We designed to do rehabilitation prior to surgery, because the soft tissue of lymphatic mass was firm to hard consistency with multiple skin nodule causing post-operative complications such as wound dehiscence, bleeding, and infection. The physiotherapy consisted of massage of internal iliac, groin and popliteal lymph nodes including soft tissue of lower extremities. After the massage, the warm compress was done. Along with physical support, the mental support by psychotherapist was also provided. This preparatory period was about one month.

2. Operative procedure

Surgical planning was done in stage operation for safety of circulation of lower limb and avoidance of lymphatic drainage system compromised problems.

The surgeries were done along the compartment of the leg.

First stage was planned for resection of lymphatic mass on the dorsum of the foot and coverage by spilt-thickness skin graft that harvested from lateral aspect of affected left thigh. The resected mass weight was 0.5 kgs.

Second stage and third stage were done to the lateral aspect of leg and thigh respectively in primary closure. The resected mass weight was 5.7 and 4.7 kgs, respectively.

Forth stage and fifth stage were done to the medial aspect of thigh in primary closure, in order to reduce the huge lymphatic mass of the leg which was so heavy to raise. Now the patient can start to walk. The resected mass weight was 1.5 and 2.7 kgs, respectively.

The sixth (last) stage was done to the biggest lymphatic mass in this elephantiasis of lower limb of this case. The primary closure and coverage by split-thickness skin graft from normal right thigh was performed. The resected mass weight was 22 kgs.

The total resected mass weight was 37.1 kg

The patient was in supine position under general anesthesia with orotrachea intubation. Aseptic and antiseptic techniques were prepared to the affected lower extremity. Resection area was designed for closure in W-plasty technique and done to the lateral aspect of leg and thigh and medial aspect of thigh with insertion of Radivac drain number 12. Only area on the dorsum of foot was resected and covered with a spilt-thickness skin graft that harvested from medial aspect of affected thigh. The biggest lymphatic mass of medial aspect of leg was done in primary closure and covered with a spilt-thickness skin graft that harvested from right thigh. Hemostasis is the most important point in this phase.

3. Post-operative care

The wound and drainage system were checked everyday. After two weeks of each surgery, stitch off was done. The Radivace drainage system was taken off after minimal fluid was detected.

4. Complication

Patient was safe after each surgery and the result is good. Patient starts to walk. Yet there were some complications, the wound dehiscence and disruption were detected from gravity mass and secondary wound closure was done later. The vacuum wound dressing system (Figure 3A, 3B) was used in some areas. The other concern about this patient is the site for several venous assessments and diet control to treat her obesity problem.

Follow-up and Outcomes

Patient had to stay in the hospital for six months for stage surgery. During the hospital course, there were several concerns about this patient; such as

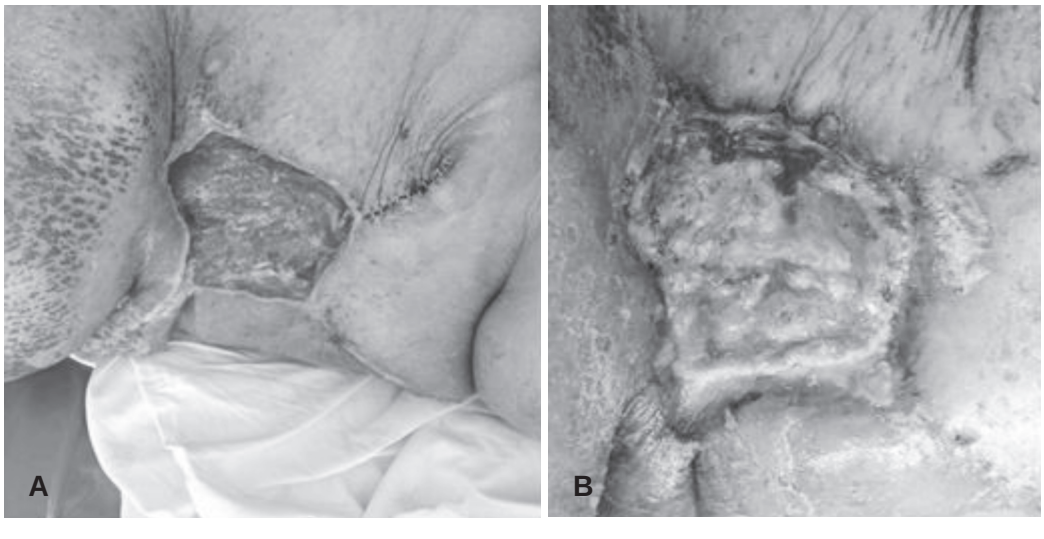

Figure 3 **A** Left leg wound dehiscence
B Left leg wound after vacuum and disruption before treatment wound dressing and coverage by split-thickness skin graft that harvested from affected thigh treatment

pressure ulcer on the leg, venous assessment, rehabilitation, mental concern and financial concern. This patient did not have any severe adverse event during her stay. The last follow-up showed good heal of the wounds and she could start to walk.

Discussion

Lymph nodes tissue transfer was reported to have successful reduction of about 47.6%[5] of edema volume and relatively significant risk for long duration of surgery, which may not fit for this patient. The debulking procedure was selected in order to save time and achieve effective reduction of the edema volume. There was a report of up to 91.1% of volume reduction of edema volume by debulking procedure[5,7]. The ultimate goal of elephantiasis limb treatment is to restore the function of the affected limb. An important component of determining whether surgical treatment is indicated is to examine the risk-benefit ratio[8]. The surgical risks or morbidity associated with an individual procedure in terms of the likelihood or frequency of a complication (such as postoperative infection) versus a rarely occurring complication may be life threatening (such as a stroke). Due to extensive and multi-stage surgery, surgeon's expertise and experience are required to perform the surgery[5,8].

Conclusion

Elephantiasis can be divided into two types; primary and secondary. Both pathogenesis ended up with persistence edema of the tissue. Conservative measures usually are not curative for elephantiasis, while the surgery will help to restore patient's function. There are two types of surgery for elephantiasis; reconstructive of lymphatic structure and destructive debulking surgery. In this case report, debulking surgery can be an adjuvant technique for elephantiasis. Although delayed postoperative wound healing problems were observed, necrectomy and vacuum assisted closure achieved a complete heal of patients' elephantiasis leg.

REFERENCES

1. Brennan MJ. Lymphedema following the surgical treatment of breast cancer: a review of pathophysiology and treatment. J Pain Symptom Manage 1992;7:110-6.
2. Földi M, Földi E, Kubik S. Textbook of lymphology: for physicians and lymphedema therapists. Munich, Germany: Elsevier; 2003.
3. The diagnosis and treatment of peripheral lymphedema: 2013 Consensus Document of the International Society of Lymphology. Lymphology 2013;46:1-11.
4. Piller N, Carati C. The diagnosis and treatment of peripheral lymphedema. Lymphology 2009 Sep;42:146-7.
5. Cormier JN, Rourke L, Crosby M, Chang D, Armer J. The surgical treatment of lymphedema: a systematic review of the contemporary literature (2004-2010). Ann Surg Oncol 2011;19:642-51.
6. Lee BB, Kim YW, Kim DI, Hwang JH, Laredo J, Neville R. Supplemental surgical treatment to end stage (stage IV-V) of chronic lymphedema. Int Angiol 2008 Oct;27(5):389-95.
7. Wollina U, Heinig B, Schonlebe J, Nowak A. Debulking surgery for elephantiasis nostras with large ectatic podoplanin-negative lymphatic vessels in patients with lipo-lymphedema. Eplasty 2014;14:e11.
8. Mehrara BJ, Zampell JC, Suami H, Chang DW. Surgical management of lymphedema: past, present, and future. Lymphat Res Biol 2011;9:159-67.

บทคัดย่อ **รายงานการรักษาสภาวะท่อน้ำเหลืองอุดตันบริเวณขาซ้าย**

นพ. รัชต์ วงศ์ตรังคพันธ์
สถาบันบำราศนราดูร

สภาวะท่อน้ำเหลืองอุดตันเกิดจากหลายสาเหตุ เช่น การติดเชื้อพยาธิโรคเท้าช้าง (ฟิลาริเอสีส) การติดเชื้อเรื้อรังของแขนหรือขา การผ่าตัดที่ทำให้การระบายของท่อน้ำเหลืองเสีย อุบัติเหตุที่ทำให้เกิด การตัดขาดของท่อน้ำเหลือง ตัวอย่างที่ชัดเจนอันหนึ่งได้แก่ การผ่าตัดต่อมน้ำเหลืองที่รักแร้ในการรักษาผู้ป่วยมะเร็งเต้านม ปัจจุบันยังไม่มีการรักษาแบบเฉพาะเจาะจงสำหรับปัญหาสภาวะต่อมน้ำเหลืองอุดตันมีการผ่าตัดแก้ไขอยู่สองแบบที่นิยมคือการทำผ่าตัดต่อท่อน้ำเหลืองใหม่และการผ่าตัดเพื่อตัดเอาเนื้อเยื่อส่วนที่บวมน้ำเหลืองออกเพื่อลดขนาดของขารายงานผู้ป่วยฉบับนี้จัดทำเพื่อรายงานการผ่าตัดผู้ป่วยที่มีสภาวะท่อน้ำเหลืองอุดตันที่บริเวณขาซ้ายจากการติดเชื้อซ้ำซ้อนเรื้อรังจากการผ่าตัดเส้นเลือดขอดที่ขาซ้ายซึ่งผู้ป่วยรายนี้ได้รับการผ่าตัดเพื่อตัดเอาเนื้อเยื่อส่วนที่บวมน้ำเหลืองออกเพื่อลดขนาดของขาหลายครั้ง จนผู้ป่วยสามารถกลับมาเดินได้อย่างปกติ