Successful Relief of a Male Breast Lymphedema by Supermicrosurgical Lymphaticovenular Anastomosis

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Background: Breast lymphedema is a common issue after breast cancer surgery but remains understudied because it is difficult to be quantified. Untreated breast lymphedema may lead to severe form of delayed breast cellulitis. Supermicrosurgical lymphaticovenular anastomosis is one option for the treatment of breast cancer-related lymphedema but has not been described for the treatment of breast lymphedema. Methods: This report presented a rare case of male breast lymphedema secondary to axillary lymph node dissection for the treatment of a forearm melanoma. Deep lymphatic vessels and adjacent venules were individualized under high magnification in the periareolar area. Six lymphaticovenular anastomoses were performed using supermicrosurgical techniques. A clinical examination and a volume assessment under magnetic resonance imaging were used to assess the efficiency of surgery. Results: Postoperative outcome was uneventful. The patient was followed-up during 1 year. Swelling relief was clinically significant 3 months postoperatively. The pinch test reduction was 2 cm after 12 months. The breast volume reduction rate was 47.2%. Conclusions: Supermicrosurgical lymphaticovenular anastomosis may be efficient for the treatment of postoperative breast lymphedema, even in male patients.

Key Words: breast lymphedema, breast cancer-related lymphedema, breast cellulitis, lymphaticovenular anastomosis, supermicrosurgery

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B reast cancer-related lymphedema is a disabling morbidity after ax-illary lymph node dissection and radiotherapy. Breast edema is a common outcome in women after breast cancer surgery. However, it remains rarer than upper-limb lymphedema. The incidence ranges from 0% to 90.4% according to clinical series.¹ Breast lymphedema should not be considered as a minor issue by comparison with upper-limb lymphedema. Indeed, untreated breast edema may lead to severe form of delayed breast cellulitis.²

Nowadays, lymphaticovenular anastomosis is the most efficient option for the treatment of breast cancer-related lymphedema. This is based on supermicrosurgical techniques and commonly used for limbs lymphedema.³ However, lymphaticovenular anastomosis has not been described for the treatment of breast lymphedema.

This report presented a severe case of breast lymphedema in a male patient secondary to axillary lymph node dissection. This lymphedema was resistant to conservative methods and frequently complicated by breast cellulitis. The interest of lymphaticovenular anastomosis was assessed for the treatment of this chronic male breast lymphedema.

CASE REPORT

A 52-year-old male presented for a Campisi clinical stage IV lymphedema of his right breast secondary to lymph node dissection for the treatment of a forearm melanoma.⁴ The lymphedema duration was 4 years. No improvement occurred under physiotherapy based on manual drainage. The patient was constantly bothered by breast

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heaviness, redness, and swelling. Six episodes of breast cellulitis occurred in the last 2 years. The mean pinch test was 4.5 cm on the right breast, whereas it was 2 cm on the left breast (Fig. 1A).

Preoperative magnetic resonance imaging (MRI) was performed to assess breast edema. Breast volume was estimated by magnetic resonance volumetry with a 3-T platform MRI (Toshiba Vantage Titan 3 T; Toshiba Medical Inc, Tokyo, Japan). The estimated volume was calculated by adding up slice by slice each area delineated on the computer workstation. Breast volume was evaluated from the skin to the pectoralis major fascia between the supramammary and the inframammary folds. The estimated breast volume was 261.8 mm³ (Fig. 2A).

After approval of the institutional review board, the patient was treated by lymphaticovenular anastomosis under local anesthesia using a M525 F50 surgical microscope (Leica Microsystems Co Ltd, Heerbrugg, Switzerland) equipped with a near infrared digital video camera (FL800 NIR Sony XC-EI 50 Camera; Sony Corp, Tokyo, Japan). Periareolar lymphatic mapping was performed by indocyanine green microscopeintegrated lymphangiography. Before scrubbing, 0.5 mL of patent blue dye (2 mL; 2.5% Bleu Patenté V by Guerbet Laboratory, France) was injected subdermally in the periareolar area, using a 0.5-mL syringe and 30-gauge needle. A skin massage was performed laterally during 1 minute. A local anesthesia using 2 mL of 1% lidocaine with 1/100.000 epinephrine was performed. Lymphatic vessels and subdermal venules were dissected subdermally, through 3-cm skin incisions in the upper and lower lateral quadrants.

The caliber of subdermal vessels was measured with a professional scaled ruler (Shinwa Sokutei Co, Ltd, Sapporo Eigyosho, Japan). The average caliber of lymphatic vessels was 0.57 mm (ranged, 0.3 to 0.7). The average caliber of subdermal venules was 1.43 mm (ranged, 0.4 to 2). Lymphaticovenular bypasses were performed with Ultrafine Microsurgical instruments (Medicon and Co, Tuttlingen, Germany), and 12-0 nylon monofilament on a 50 µm 3/8 needle (S&T; S&T AG, Neuhausen, Switzerland). Two end-to-side lymphaticovenular anastomoses were performed in the upper lateral quadrant. Four end-to-end lymphaticovenular anastomoses were performed in the lower lateral quadrant. Intraoperative lymphaticovenular anastomosis patency was checked with indocyanine green fluorescence by microscope-integrated near infrared videoangiography. A positive patency was found in each lymphaticovenular anastomosis (Figs. 3, 4).

Postoperative course was uneventful. No adverse reaction, including nausea, hypotension, hives, and anaphylaxis, occurred. The patient left the hospital after 12 hours of medical supervision. Skin healing was completed after 10 days. Pain reduction occurred 2 months postoperatively. Swelling relief was clinically significant 3 months postoperatively. The pinch test reduction was 2 cm after 12 months (Fig. 1B). Breast volume was 138.2 mm³ 12 months after surgery (Fig. 2B). The breast volume reduction rate was 47.2%.

DISCUSSION

Breast lymphedema has probably been understudied because it is difficult to be quantified by comparison with upper-limb breast cancer-related lymphedema. Various signs have been associated with breast edema such as breast heaviness, breast pain, skin swelling, skin thickening, skin redness, and peau d'orange.^{5–7} Various risk factors have also been identified. Axillary lymph node dissection, axillary staging surgery, sentinel lymph node biopsy, increasing irradiated breast volume,

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FIGURE 1. Comparison of the clinical appearance before and after surgery. A, Preoperative view: chronic lymphedema. B, Postoperative view 1 year after surgery: edema relief.

and tumor volume are major risk factors of breast lymphedema.^{1,8} Body mass index and diabetes mellitus are minor risk factors.⁹ Breast cancer in the upper lateral breast segment is recognized as the main risk factor of delayed breast cellulitis.¹⁰

A common treatment is complex physical therapy based on manual lymphatic drainage, compression therapy, skin care, and exercises. A less usual method is manual lymphatic drainage associated with deep oscillation by an intermittent electrostatic field of low frequency.¹¹ To our knowledge, this is the first case of breast lymphedema in a male patient treated by supermicrosurgery. This technique has not been reported for female breast lymphedema as well.

Lymphaticovenular anastomosis is a usual technique for chronic limb lymphedema, which derives from lymphaticovenous bypasses described by Yamada in 1969.¹² The surgical technique was refined by O'Brien¹³ and by Koshima who introduced the concept of supermicrosurgery.¹⁴ Lymph drainage is due to the difference of pressure gradient between lymphatic vessels and subdermal venules. As the differential pressure gradient between lymphatic vessels and subdermal venules is low, the rate of vascular thrombosis is decreased with supermicrosurgical lymphaticovenular anastomosis, comparing to classical lymphaticovenous bypasses.^{3,15} In this case, MRI was chosen to assess the postoperative breast volume. Magnetic resonance volumetry was used adding up slice by slice each area delineated on the computer workstation. This method derives from computed tomographic volumetry used in breast reconstruction.¹⁶ To our knowledge, this is the first report using magnetic resonance volumetry for breast volume assessment. Even if it could not identify precisely lymphatic vessels and lymphaticovenular anastomosis postoperatively, it was worthwhile to visualize edema in subcutaneous breast tissue. This method might be interesting for preoperative flap planning in microsurgical breast reconstruction in the same way as computed tomographic volumetry in near future.

CONCLUSIONS

Breast lymphedema is a disabling issue after breast cancer treatment, which impacts very negatively the quality of life. Here, we report the first case of breast lymphedema in a male patient treated by supermicrosurgical lymphaticovenular anastomosis. Postoperative outcome was very encouraging. Supermicrosurgical techniques may be also a new option for the management of female breast lymphedema.

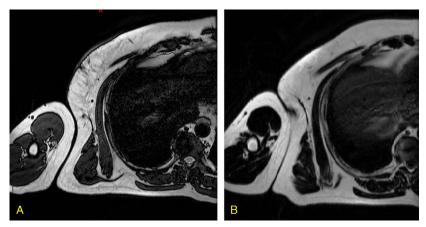


FIGURE 2. Magnetic resonance imaging comparing the right breast volume before (A) and 1 year after surgery (B) (images centered on the inferior border of the pectoralis major muscle). A, Lymphedematous subcutaneous infiltration of the breast subcutaneous tissue. The breast volume estimation was 261.8 mm³. B, Lymphedema relief 1 year after lymphaticovenular anastomosis. The breast volume estimation was 138.2 mm³.

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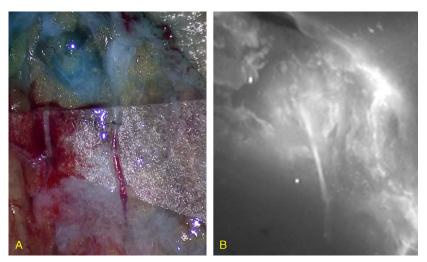


FIGURE 3. A, Intraoperative view under high magnification (27×) of an end-to-end lymphaticovenular anastomosis in the periareolar area. B, Microvascular patency of the lymphaticovenular anastomosis after indocyanine green lymphangiography.

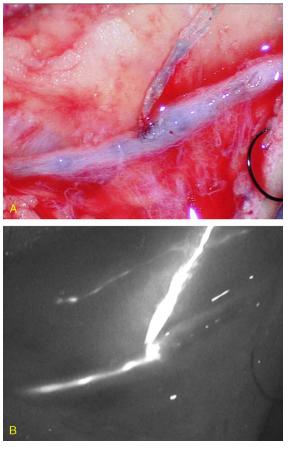


FIGURE 4. A, Intraoperative view under high magnification $(27 \times)$ of an end-to-side lymphaticovenular anastomosis in the periareolar area. B, Microvascular patency of the lymphaticovenular anastomosis after indocyanine green lymphangiography.

REFERENCES

- Verbelen H, Gebruers N, Beyers T, et al. Breast edema in breast cancer patients following breast-conserving surgery and radiotherapy: a systematic review. *Breast Cancer Res Treat*. 2014;147:463–471.
- Zippel D, Siegelmann-Danieli N, Ayalon S, et al. Delayed breast cellulitis following breast conserving operation. *Eur J Surg Oncol.* 2003;29:327–330.
- 3. Nagase T, Gonda K, Inoue K, et al. Treatment of lymphedema with lymphaticovenular anastomoses. *Int J Clin Oncol*. 2005;10:304–310.
- Campisi C, Boccardo F. Microsurgical techniques for lymphedema treatment: derivative lymphatic-venous microsurgery. *World J Surg.* 2004;28:609–613.
- Clarke D, Martinez A, Cox RS, et al. Breast edema following staging axillary node dissection in patients with breast carcinoma treated by radical radiotherapy. *Cancer*. 1982;49:2295–2299.
- Pezner RD, Patterson MP, Hill LR, et al. Breast edema in patients treated conservatively for stage I and II breast cancer. *Int J Radiat Oncol Biol Phys.* 1985;11: 1765–1768.
- Adriaenssens N, Verbelen H, Lievens P, et al. Lymphedema of the operated and irradiated breast in breast cancer patients following breast conserving surgery and radiotherapy. *Lymphology*. 2012;45:154–164.
- Boughey JC, Hoskin TL, Cheville AL, et al. Risk factors associated with breast lymphedema. *Ann Surg Oncol.* 2014;21:1202–1208.
- Barnett GC, Wilkinson JS, Moody AM, et al. The Cambridge breast intensitymodulated radiotherapy trial: patient- and treatment-related factors that influence late toxicity. *Clin Oncol (R Coll Radiol).* 2011;23:662–673.
- Rönkä RH, Pamilo MS, von Smitten KA, et al. Breast lymphedema after breast conserving treatment. Acta Oncol 2004;43:551–557.
- Jahr S, Schoppe B, Reisshauer A. Effect of treatment with low-intensity and extremely low-frequency electrostatic fields (deep oscillation) on breast tissue and pain in patients with secondary breast lymphoedema. *J Rehabil Med.* 2008;40: 645–650.
- 12. Yamada Y. Studies on lymphatic venous anastomosis in lymphedema. Nagoya J Med Sci. 1969;32:1–21.
- O'Brien BM, Sykes P, Threlfall GN, et al. Microlymphaticovenous anastomoses for obstructive lymphedema. *Plast Reconstr Surg.* 1977;60:197–211.
- 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.
- Baumeister RG, Siuda S. Treatment of lymphedemas by microsurgical lymphatic grafting: what is proved? *Plast Reconstr Surg.* 1990;85:64–74.
- Rosson GD, Shridharani SM, Magarakis M, et al. Three-dimensional computed tomographic angiography to predict weight and volume of deep inferior epigastric artery perforator flap for breast reconstruction. *Microsurgery*. 2011;31:510–516.