Severe lower limb lymphoedema successfully treated with a two-stage debulking procedure: a case report

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\section*{ABSTRACT}
Lymphoedema is a chronic condition that has significant functional and psychosocial morbidity. We report a case of severe lower limb lymphoedema successfully treated with a two-stage debulking procedure, highlighting the significant improvements in function and quality of life this operation can have with the appropriate multidisciplinary support.

\section*{Introduction}
Lymphoedema is a chronic progressive disorder affecting up to 250 million people worldwide [1]. It is characterised by the impaired return of lymphatic fluid from the extremities resulting in oedema of the affected tissues. It can broadly be categorised into primary and secondary lymphoedema. Primary lymphoedema is caused by an inherent dysfunction in the lymphatic system whilst secondary lymphoedema results following an insult such as surgery, trauma or infection [2]. Filariasis, a parasitic roundworm, is the leading cause of secondary lymphoedema worldwide, however, this is primarily limited to developing countries. In the developed world, the leading cause of lymphoedema is from cancer treatments including lymph node clearance [3,4]. The chronic deposition of protein-rich lymphatic fluid in the interstitial space stimulates an inflammatory reaction leading to fibrosis and further damage to the lymphatics with a resultant cycle of deterioration [5–7]. This ultimately results in a grossly enlarged extremity with increased susceptibility to infection, impairment of mobility/function and psychosocial morbidity [8–10]. Treatment therefore aims to reduce the swelling and its associated sequelae and thus improve quality of life. Various different treatment modalities exist ranging from compression garments, exercise, skin care and manual lymphatic drainage to surgical interventions.

Surgical debulking for lymphoedema was first reported as far back as 1912 when Sir Richard Charles described excision of the affected area down to deep fascia and skin grafting the defect, with resultant immediate improvements in function [11]. A similar procedure was also described by Homan, however, here skin flaps are raised and only the underlying lymphoedematous tissue is excised [12]. With recent advances in microsurgery, lymphovenous anastomosis (LVA) and vascularised lymph node transfer (VLNT) are increasing in popularity in addition to liposuction, with traditional debulking procedures acting as a last resort due to concerns regarding subsequent morbidity [13,14].

We hereby present a case of severe lower limb lymphoedema successfully treated with a two-stage radical debulking procedure, highlighting the positive impact that this operation can have on a patient’s quality of life with the appropriate multidisciplinary team (MDT) support.

\section*{Case report}
A 56-year-old gentleman was referred to our service with a long-standing history of primary lymphoedema...
affecting his left leg and despite being compliant, con-
servative management had failed to reduce the limb
significantly (Figure 1). In 2010, he underwent a
below-knee Charles procedure at a remote unit. Whilst
this had reduced the volume of the lower leg, he was
left with multiple areas of unstable skin graft and wor-
sening swelling in his foot and thigh. The thigh swel-
ling had become so large that he found it extremely
difficult to mobilise resulting in him being house
bound. He also found it difficult to pass urine due to
the large mass of tissue medially, contributing to a
total thigh circumference of 150 cm and the leg being
219% larger than the right with an excess volume of
25,306 millilitres (mls). His comorbidities included
right-sided heart failure, sleep apnoea and atrial fibril-
ation. As part of his assessment an MRI scan was per-
duced which demonstrated diffusely oedematous
subcutaneous fat but no discrete cystic lesions.

After discussion at the lymphoedema MDT meeting
consisting of surgeons, lymphoedema therapists and
physiotherapy, the decision was made to undertake a
two-stage debulking procedure of the left thigh. He
was subsequently counselled carefully on the high
risks that general anaesthetics presented due to his
complex medical history, however, despite this, owing
to the significant impact on his quality of life, he was
keen to proceed with surgical intervention. An anaes-
thetic review was sought preoperatively.

The first procedure consisted of placing the patient
in the right lateral position. Following infiltration of
the area with tumescent solution, an anteriorly based
fasciocutaneous flap was raised over the area of max-
imal thigh bulk through a medially placed incision
(Figure 2). Incisions were planned in order to allow
maximal debulking of the problematic area but to also
facilitate closure of the wound. Lymphoedematous tis-
sue was excised from the posterior and superolateral
thigh to the knee, down to, but not including the
deep fascia. Careful haemostasis was required due to
the presence of numerous large traversing superficial
veins. Cell salvage was also utilised with the patient
receiving 2.1 litres of blood in total. Due to the large
volume of blood loss intraoperatively the decision was
made to not address the medial area of tissue excess
during this first stage. He spent 7 days in the high
dependency unit (HDU) postoperatively where he ini-
tially required vasopressor support. He was taken back
to theatre 3 weeks following this initial procedure to
debride and close an area of wound dehiscence
around the tip of the flap. During this time the lower
leg was placed in class II compression garments with
wraps around the knee.

The second stage procedure was undertaken
11 months following the first and consisted of placing
the patient supine in a frog leg position. Tumescent
solution was once again utilised and a laterally based
flap was raised through an inferomedial incision. 3,594
grams of lymphoedematous tissue was excised primar-
ily from the medial and anterior areas of the thigh
(Figure 3). He spent 6 days in HDU and once again suf-
f ered from an area of wound dehiscence 2 weeks post-
operatively. This was debrided followed by the
application of a vacuum assisted closure dressing. No
further operations were required. Histology from both
debulking procedures demonstrated changes consist-
ent with chronic lymphoedema.

His wounds have now completely healed and he is
able to mobilise with relative ease and can pass urine
without difficulty. In addition, he feels his quality of
life is much improved and is more confident with the
appearance of his leg (Figure 4). When compared to the contralateral side the left leg is now only 22% larger with an excess volume of 2,892 mls. To maintain this reduction he now wears 2 class II compression garments (thigh and below knee) as well as a wrapping system.

**Discussion**

Lymphoedema is a debilitating condition that can have substantial physical and psychological morbidity. Treatment of these patients is extremely challenging with surgical intervention reserved for those with significant complex needs and in whom conservative measures have failed.

Various different microsurgical techniques are now available to manage lymphoedema including LVA and VLNT and these have become the surgical treatment of choice within our lymphoedema department. A smaller number of patients are offered liposuction debulking. LVA was first described in 1969 and works by diverting lymph into the venous system prior to the obstruction [15]. It is generally accepted that the earlier this procedure is performed in the disease process the more effective it is and despite numerous variations in technique being described, significant improvements have been reported [13,16–18]. VLNT is the process of transferring lymph nodes from…
elsewhere in the body to the lymphoedema affected area. Numerous different VLNTs have been described with promising results, including the supraclavicular flap, omental flap and groin flap [14,19,20]. The use of liposuction can remove excess adipose tissue and result in a significant reduction in volume [21].

In our patient, however, with such severe lymphoedema and thick fibrotic tissues, the only option to rapidly improve his symptoms was to use a more traditional debulking procedure. In recent times these procedures have fallen out of favour due to the associated high rates of morbidity and potential for mortality. In addition, large levels of intraoperative blood loss should be expected. Despite these risks, our case highlights the positive impact a traditional debulking procedure can have if combined with the appropriate expertise and specialist input available in a surgical lymphoedema MDT.

Due to the size of the swelling, our case was planned as a two-stage debulking to minimise the physiological insult, with a senior consultant anaesthetist and a four surgeon operating team. Utilising two consultants to undertake the excision, with trainees performing concurrent haemostasis improved operative time and minimised blood loss. The use of cell salvage as well as infiltration of the area with tumescent solution should also routinely be implemented in order to assist with this further. We recommend that all such patients should be admitted to HDU postoperatively during which time they can be provided with cardiovascular support. Postoperative wound care in both the inpatient and outpatient setting is also a vital component of these patients management. All nurses should have the appropriate training and experience in dealing with complex dressings and wounds due to lymphoedema, as postoperative delays in wound healing and infection should be expected. During their admission all patients should have physiotherapy input to address their complex mobility issues.

**Conclusion**

We recommend debulking surgery in cases of severe lower limb lymphoedema where other available treatment modalities are not appropriate or have been exhausted. With the correct multidisciplinary support, significant improvements in function and quality of life can be achieved.

**Disclosure of interest**

The authors report no conflict of interest and no external funding was received.

**References**

[1] Rockson SG, Rivera KK. Estimating the population burden of lymphedema. Ann N Y Acad Sci. 2008;1131(1):147–154.

[2] Allen RJ, Cheng MH. Lymphedema surgery: patient selection and an overview of surgical techniques. J Surg Oncol. 2016;113(8):923–931.

[3] Babu S, Nutman TB. Immunopathogenesis of lymphatic filarial disease. Semin Immunopathol. 2012;34(6):847–861.

[4] Cormier JN, Askew RL, Mungovan KS, et al. Lymphedema beyond breast cancer: a systematic
review and meta-analysis of cancer-related secondary lymphedema. Cancer. 2010;116(22):5138–5149.

[5] Avraham T, Yan A, Zampell JC, et al. Radiation therapy causes loss of dermal lymphatic vessels and interferes with lymphatic function by TGF-beta1-mediated tissue fibrosis. Am J Physiol Cell Physiol. 2010;299:589–605.

[6] Zampell JC, Yan A, Elhadad S, et al. CD4(+) cells regulate fibrosis and lymphangiogenesis in response to lymphatic fluid stasis. PLoS ONE. 2012;7(11):e49940.

[7] Cuzzone DA, Weitman ES, Albano NJ, et al. IL-6 regulates adipose deposition and homeostasis in lymphedema. Am J Physiol Heart Circ Physiol. 2014;306:1426–1434.

[8] Tobin MB, Lacey HJ, Meyer L, et al. The psychological morbidity of breast cancer-related arm swelling. Cancer. 1993;72(11):3248–3252.

[9] De Godoy JMP, Braile DM, de Fatima Godoy Jr. M, et al. Quality of life and peripheral lymphedema. Lymphology. 2002;35:72–75.

[10] McWayne J, Heiney SP. Psychologic and social sequelae of secondary lymphedema: a review. Cancer. 2005;104(3):457–466.

[11] Dumanian GA, Futrell JW. The Charles procedure: misquoted and misunderstood since 1950. Plast Reconstr Surg. 1996;98(7):1258–1263.

[12] Homan J. The treatment of elephantiasis of the legs. A preliminary report. NEJM. 1936;24:1099–1104.

[13] Chang DW, Suami H, Skoracki R. A prospective analysis of 100 consecutive lymphovenous bypass cases for treatment of extremity lymphedema. Plast Reconstr Surg. 2013;132(5):1305–1314.

[14] Lin CH, Ali R, Chen SC, et al. Vascularized groin lymph node transfer using the wrist as a recipient site for management of postmastectomy upper extremity lymphedema. Plast Reconstr Surg. 2009;123(4):1265–1275.

[15] Yamada Y. The studies on lymphatic venous anastomosis in lymphedema. Nagoya J Med Sci. 1969;32:1–21.

[16] O’Brien BM, Mellow CG, Khazanchi RK, et al. Long-term results after microlymphaticovenous anastomoses for the treatment of obstructive lymphedema. Plast Reconstr Surg. 1990;85:562–572.

[17] Campisi C, Bellini C, Campisi C, et al. Microsurgery for lymphedema: clinical research and long-term results. Microsurgery. 2010;30:256–260.

[18] Damstra RJ, Voesten HG, van Schelven WD, et al. Lymphatic venous anastomosis (LVA) for treatment of secondary arm lymphedema. A prospective study of 11 LVA procedures in 10 patients with breast cancer related lymphedema and a critical review of the literature. Breast Cancer Res Treat. 2009;113(2):199–206.

[19] Becker C, Vasile JV, Levine JL, et al. Microlymphatic surgery for the treatment of iatrogenic lymphedema. Clin Plast Surg. 2012;39(4):385–398.

[20] Cheng MH, Chen SC, Henry SL, et al. Vascularized groin lymph node flap transfer for postmastectomy upper limb lymphedema: flap anatomy, recipient sites, and outcomes. Plast Reconstr Surg. 2013;131(6):1286–1298.

[21] O’Brien BM, Khazanchi RK, Kumar PA, et al. Liposuction in the treatment of lymphoedema; a preliminary report. Br J Plast Surg. 1989;42(5):530–533.