The role of circumferential suction assisted lipectomy (liposuction) and compression in limb lymphoedema

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INTERNATIONAL LYMPHOEDEMA FRAMEWORK

Position document

BEST PRACTICE

FOR THE MANAGEMENT OF LYMPHOEDEMA - 2ND EDITION

Surgical Intervention

A position document on surgery for lymphoedema

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THE INTERNATIONAL LYMPHOEDEMA FRAMEWORK

The International Lymphoedema Framework (ILF) is a UK charity. Its aim is to improve the management of lymphoedema and related disorders worldwide through the sharing of expertise and resources, and by supporting individual countries to develop a long term strategy for lymphoedema. Such a strategy will:

- Raise the profile of lymphoedema nationally and internationally
- Place lymphoedema and its management as a priority on national health care agendas
- Lobby for appropriate financing or reimbursement of lymphoedema care
- Address issues of inequity of provision
- Implement and evaluate lymphoedema services based on best practice
- Create an international lymphoedema community that collectively strives to improve the evidence base for treatment and professional practice
- Improve the lives of lymphoedema sufferers worldwide

The standards of practice for people with lymphoedema outlined in box 1 provide a framework for the ILF and its partner organisations to work towards.

Box 1: Standards of practice for lymphoedema[i]

Standard 1
Awareness and knowledge of lymphoedema within the community

Standard 2
Identification of people at risk of or with lymphoedema

Standard 3
Empowerment of people at risk of or with lymphoedema

Standard 4
Provision of lymphoedema services that deliver high quality clinical care that is subject to continuous improvement

Standard 5
Access to appropriately trained health care professionals

Standard 6
Provision of high quality clinical care for people with cellulitis

Standard 7
Provision of optimal, individualised programmes of care

Standard 8
Provision of multi-disciplinary health and social care

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Foreword

International Lymphoedema Framework Surgical Intervention

This position document on the role of surgery in lymphoedema management forms part of the second edition of the International Lymphoedema Framework (ILF) best practice document. The ILF are very proud that this document is based on a systematic review of surgery in lymphoedema management undertaken by the American Lymphoedema Framework Project; the chapter by Dr Janice Cormier summaries their findings and recommendations. This approach follows that of the first edition of the best practice document that began with Cochrane systematic reviews of physical therapies.

This document addresses the complex and often controversial issue of surgery and attempts to present a balanced picture of how surgery fits within the total treatment paradigm using the Chronic Care model and the International Classification of Functioning, Disability and Health. It reinforces the message that lymphoedema management requires active patient self management and that the other aspects of decongestive therapy (DLT) remain critical to good outcome. New approaches including the use of circumferential suction assisted lipectomy (lipsuction) and microsurgery, provide exciting advances in practice. The importance of the multidisciplinary team and how they work with patients and carers is also explored.

The ILF hopes that this document, which will comprise part of a 12-section compendium, will help you and your fellow professionals to plan and deliver excellent care in surgical intervention for lymphoedema.

Christine Moffatt
Chair, International Lymphoedema Framework

Summary Statements

A famous lypmphologist once said that liposuction for lymphoedema is science fiction; time has shown otherwise. However, since lymphoedema is a chronic disease, an approach based on chronic disease management is mandatory. Lymphedema is not just about accumulation of lymph; it is also about hypoplasia, dysplasia, obstructed dilated lymph vessels, the accumulation of adipose tissue, and possible fibrosis

- In chronic diseases, the contribution of the patient to their own treatment is crucial; the role of the health-professional is more 'hands off' and teaching
- Self care, self management and awareness should be taught to the patient for maximum effectiveness during the maintenance phase of treatment
- As surgery for lymphoedema is often a final option, it should be integrated in a full lymphoedema service which includes early diagnosis, conservative treatment and effective follow-up using protocols
- Conservative treatment - where bandaging is the most important part - is the treatment of choice in order to remove accumulated lymph and to transfer a pitting oedema into a non-pitting state. In spite of skillful decongestive treatment, many patients do not get an acceptable reduction because the excess volume comprises not only increased adipose tissue, but also hypertrophied muscle tissue; the latter due to the heavy load that has been present for years
- Circumferential suction assisted lipectomy (CSAL) (also known as liposuction) is a well researched, effective and safe procedure for end-stage lymphoedema that has been unresponsive to conservative treatment, CSAL changes lymphoedema back into the original state; it produces a long-lasting, 100% volume reduction in limbs when proper compressions garments are used post-operatively and for life
- CSAL significantly reduces the number of episodes of erysipelas, dramatically improves quality of life and facilitates self-care
- CSAL should be embedded in a integrated lymphoedema service protocol
- While the principle of microsurgery, reconstructing the lymphatic system, is logical it does not address the reversal of hypertrophied adipose tissue. Therefore, surgery is proposed before signs of lymphoedema occur; however, this is controversial since the occurrence of lymphoedema is unpredictable
- While microsurgical approaches are developing, further work needs to be undertaken to effectively define indications for such surgery

About this document

Lymphoedema is a chronic, progressive and often debilitating condition caused by the anatomical obliteration or functional deficiency of the lymphatic system[1]. Due to its chronic nature, lymphoedema requires ongoing treatments that consider lymphatic anatomy and function[2].

It is estimated that lymphoedema impacts upon more than 120 million world-wide. Despite this prevalence, explicit assessment methodologies, effective means of treatment and comprehensive management strategies remain largely inadequate. Recent research and growing awareness of the condition has however, offered well-founded interventions for the condition[3].

In support of this growing worldwide awareness, this second edition of the Best Practice for the Management of Lymphoedema (surgery) represents an international multidisciplinary initiative led by the International Lymphoedema Framework (ILF) in association with the American Lymphedema Framework Project (ALFP) and the Canadian Lymphedema Framework (CLF). This edition only covers one aspect of decongestive lymphatic therapy (DLT), namely surgery. The ILF editorial committee believes that a large, bulky document which attempts to cover all aspects of decongestive lymphatic therapy (DLT) would not do justice to the needs of patients and practitioners world-wide. Therefore, it was decided to build a compendium of individual, in-depth documents on topics which fall under the DLT 'umbrella'. Each discreet section, building into a final 12-topic document, provides a model for best practice in the assessment, treatment and continuing management of lymphoedema. The document contains broad practice standards applicable to the international lymphoedema community for future review, consensus building, and translation. Naturally, it is expected that practitioners will have undertaken the relevant training and educational requirements before using the guidance here.

This document derives its spirit from the first edition[4]. Within the limitations outlined below, it reflects the current evidence-base. The challenge of creating and updating this document is primarily related to the paucity of randomised controlled (clinical) trials (RCTs) in the field. Where RCTs are not widely available, other sources of evidence are considered valid approaches to best practice guideline development[5]. For the purposes of this document, literature search, expert review and consensus were used.

Document terminology

As with any clinical discipline, terminology often varies between countries. While the ILF and its international framework partners are working towards a consensus on terminology in respect to lymphoedema, for the purposes of this document the following terms will be used:

- Decongestive lymphatic therapy (DLT) (also known as complete decongestive therapy (CDT) or complex decongestive physical therapy)[5]
- Inelastic bandages (also know as short-stretch bandages)

Limitations

The ILF would like to acknowledge that while the best practice statements contained within this document are as contemporaneous as possible, based on the systematic review, they are largely derived from studies published in English. For the next edition of the Best Practice Document, the ILF will be working closely with their international partners, ensuring that studies published in their respective countries will be reviewed and included where appropriate.

About this document

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CHAPTER 1
What is lymphoedema?

The lymphatic system is part of the circulatory system; it maintains the flow of fluids around the body while removing and transporting waste products from tissues[1]. Under normal conditions, venous capillaries reabsorb 90% of the fluid in the tissues, and lymphatic channels absorb the remaining 10% of lymph fluid, proteins and other molecules[2]. Lymphatic fluid passes to regional lymph nodes and empties into the venous system, most commonly by way of the thoracic duct.

Lymphoedema is an external or internal manifestation of lymphatic insufficiency and deranged lymph transport[3]. This insufficiency causes an accumulation of protein-rich interstitial fluid, leading to distention, proliferation of fatty tissue and progressive fibrosis. Skin changes such as thickening and hair loss may occur, and eventually, significant disfigurement and loss of function. Lymphoedema manifests as swelling of one or more limbs and may include the corresponding quadrant of the trunk. The head and neck, breast or genitalia may also be affected. Significant functional and psychological morbidity, such as disfigurement, pain and complications results from end-stage sequelae of lymphoedema[4,5].

Lymphoedema is generally classed as either primary (hereditary), related to congenital malformation of the lymphatic channels, or secondary, resulting from disruption to the lymphatic system.

Primary lymphoedema

Primary lymphoedema represents a heterogeneous group of disorders that includes sporadic, hereditary and syndrome-associated forms. The estimated prevalence of primary lymphoedema is 1.15 in 100,000 persons under the age of 20[6]. In children, the two main causes are Milroy disease and lymphoedema distichiasis[3].

Secondary lymphoedema

Secondary lymphoedema is a consequence of removal or damage to lymph nodes, fibrosis of the nodes (post-radiotherapy), and trauma or infection[7]. Side effects of advanced diseases such as cancer, chronic heart failure, neurological and liver disease, and end-stage renal disease can cause chronic oedema. An increase in the bariatric population has seen an increase in lymphoedema, although filarial disease, transmitted by mosquitoes, remains the most common cause of lymphoedema worldwide. The term 'chronic oedema' has been adopted by European investigators to define a population of patients with long-standing oedema (> 3 months), and perhaps a more complex underlying aetiology. Prevalence estimates for chronic oedema are between 1.3 and 1.5 per thousand[8].

Classification of the causes of secondary lymphoedema

There is a lack of consensus regarding the causes of secondary lymphoedema. In the United Kingdom (UK), the classification of causes comprises[9]:

- trauma and tissue damage (for example, burns, lymph node excision, radiotherapy, varicose vein surgery)
- malignant disease/treatment (lymph node metastases/excision, infiltrative carcinoma, lymphoma, radiotherapy, pressure from large tumours)
- venous disease (chronic venous insufficiency, venous ulceration, post-thrombotic syndrome, intravenous drug use)
- infection (cellulitis/erysipelas, lymphadenitis [inflammation of the lymph nodes], filiaris, tuberculosis)
- inflammation (rheumatoid arthritis, dermatitis, psoriasis, sarcoidosis, dermatosis with epidermal involvement)
- endocrine disease (pretibila myxoedema)
- immobility and dependency (dependency oedema, paralysis)
- factitious (self harm)

Stages of lymphoedema

Lymphoedema presents in stages *(Table 1)*; each stage may have a negative impact upon quality of life and possibly, due to recurrent tissue infection, disfigurement, pain, and impaired mobility, lead to social isolation.

Table 1: Lymphoedema stages[3,10]

Stage	Description
0	Sub-clinical or pre-lymphoedema. Typically includes all patients who have had lymph node dissection. Swelling is not evident, despite impaired lymph flow. This stage may last a long time.
I	Accumulation of fluid and protein in tissue is noted. Pitting may be present. Elevation may influence the limb. Mild swelling (<20% excess limb volume vs unaffected limb).
II	Includes swelling that does not reduce with elevation; pitting is present with increased adipose tissue and fibrosis. Moderate swelling (20-40% excess limb volume vs unaffected limb). Since adipose tissue accumulation can be seen within the first year after lymphoedema occurs it is important to include adipose tissue hypertrophy in the staging[11].
III	Adipose tissue and fibrotic tissue may or may not show pitting; includes skin thickening and large limb volume (elephantiasis). This morbid condition occurs when lymphostasis and chronic inflammation develop into fibrosclerosis and additional tissue swelling. Severe swelling (>40% excess limb volume vs unaffected limb). *NB: pitting can be present at all stages, also in stage III where it can dominate the swelling*[12]

Early signs of lymphoedema

Both primary lymphoedema and lymphoedema associated with non-cancer secondary causes, may initially present post-surgery as swelling, discomfort and inflammation; sensations of heaviness, tingling and aching also have been reported[13] *(Box 1)*. Both patients and practitoners need to be aware of these signs and symptoms.

Box 1: Early symptoms and signs of lymphoedema

- clothing or jewellery, e.g. sleeve, shoe or ring, becoming tighter
- feeling of heaviness, tightness, fullness or stiffness, and/or pain
- aching
- observable swelling
- tissue swelling – mild, moderate or severe; pitting or non-pitting
- skin condition – thickened, warty, bumpy, blistered, lymphorrhoeic, broken or ulcerated
- subcutaneous tissue changes – fatty/rubbery, non-pitting or hard
- shape change – normal or distorted
- frequency of cellulitis/erysipelas
- associated complications of internal organs, for example, pleural fluid, chylous ascites (accumulation of chyle in the abdominal cavity)
- movement and function – impairment of limb or general function
- psychosocial morbidity

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CHAPTER 2
The evidence base for surgery

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Introduction

The surgical treatment of lymphoedema has become increasingly publicised over the last two decades as an effective treatment. The majority of patients who have been treated surgically for their lymphoedema have had secondary, or acquired lymphoedema associated with breast cancer, which occurs with an estimated incidence of 40% among this population[1]. Other cancer survivors including patients with melanoma, gynaecological, genitourinary, and head/neck cancers have also been included in select case reports on the surgical treatment of lymphoedema.

While lymphoedema is considered to be an incurable condition, successful management of the condition can occur and is more likely following early diagnosis and intervention. The standard therapy for lymphoedema is manual lymphatic drainage (MLD) alone or as a component of decongestive lymphatic therapy (DLT). The surgical treatment of lymphoedema has been promoted more recently as an alternative or adjunctive treatment option for patients with lymphoedema[2-5] and even as a means of preventing lymphoedema[4,6]. Advances in microsurgical technique over the last decade have resulted in the introduction of a variety of techniques for the treatment of lymphoedema. However, these have not been widely studied in multi-institutional, randomised controlled trials, making interpretation difficult particularly for the majority of studies which do not include a comparison group.

Several categories of surgical procedures have been proposed for the treatment for lymphoedema.

These include:

- excisional procedures
- liposuction
- lymphatic reconstruction
- tissue transfer procedures

Recently, a systematic review of the contemporary surgical treatment of lymphoedema was performed and subsequently updated to identify the published literature related to the most frequently reported surgical procedures for the treatment of lymphoedema[7].

Methods

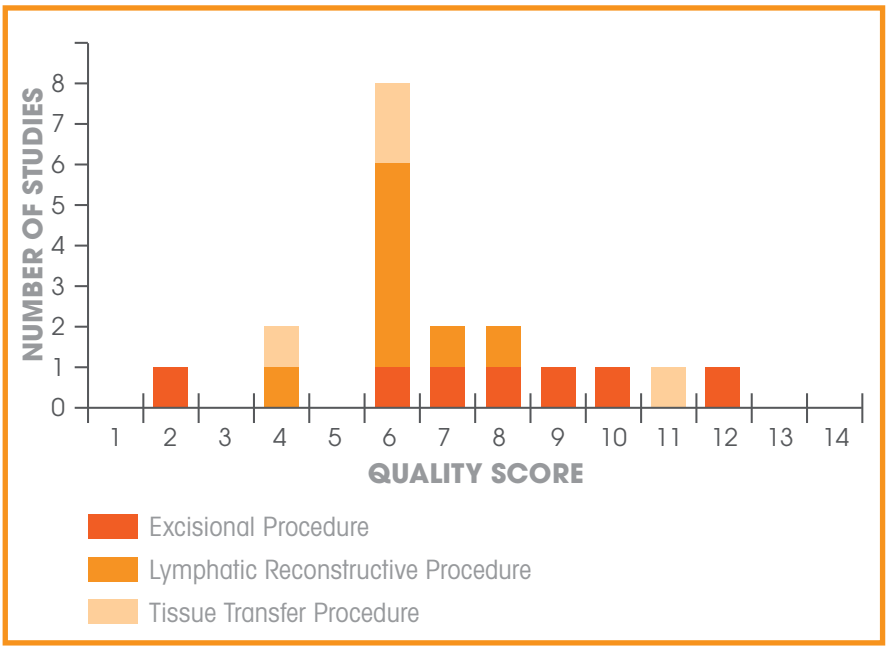
Authors completed a comprehensive search of 11 major medical indices for articles published between 2004 and 2011 using keywords related to the surgical treatment of lymphoedema[7]. Eligibility criteria included studies with at least 8 patients and published in peer-reviewed journals. Articles were categorised based on surgical treatment and data including affected anatomic region (extremity), number of patients, reported volume reduction, length of follow-up, and measurement methods. The overall weighted volume reductions were calculated based on study size.

Findings

The reported overall weighted incidence of volume reduction of lymphoedema varied from a maximum of a 118% post-operative reduction to a 13% volumetric increase. The largest reductions were noted for patients treated with excisional procedures *(Figure 1)*.

Follow-up varied widely among the studies and ranged from 6 to 120 months. Heterogeneity was noted with regards to the assessment and classification of lymphedema and the patient populations studied. The most consistent finding was the continued need for compression bandaging and physiotherapy and/or DLT following surgical treatment[8-12]. Morbidity associated with treatment was reported in only two studies[11, 13].

Figure 1: Quality scores for surgical procedures



Excisional Operations

Excisional operations and debulking have been reported as a means of achieving volume reduction for limbs with severe lymphoedema since 1912[14]. Debulking surgery removes all of the overlying skin and soft tissue above the deep fascia in the affected area with the remaining raw surfaces to be covered with grafted skin obtained from the resected area[14]. Modifications to this procedure, such as the Sistrunk procedure[15], have subsequently been introduced which reduces the bulk of tissue primarily for the treatment of elephantiasis[16]. Further modifications to this procedure include the elevation of skin flaps to maintain circulation of the skin to promote healing as well as less radical procedures for the reduction of the calf and foot[14,17]. A long list of complications have been reported with excisional procedures include haematoma, skin/flap necrosis, infection, chronic wounds/delayed wound healing, deep vein thrombosis, extensive scarring/poor cosmesis, destruction of remaining lymphatic vessels, loss of limb function and recurrent lymphoedema[17].

A total of 4 contemporary articles were identified which utilised excisional procedures for the surgical treatment of lymphedema *(Table 1)*. Only one of the studies reported outcomes of volume reduction as a percentage reduction (21%)[13]. Two studies included the radical reduction of lower-extremity lymphoedema with a follow-up time of 28 months[18,19], while one study reported radical excisional procedures for the treatment of upper extremity lymphoedema with a follow up of only 18 months[13]. The fourth study reported debulking of the male genital region using radical excision and skin grafting, with a follow up time of 48 to 72 months[41].

Liposuction

Liposuction is a surgical technique which is performed by inserting metal cannulae into small incisions to aspirate subcutaneous fat. The application of this technique has been popularised by Dr. Brorson in Sweden as an effective means of removing excess adipose tissue deposited in the lymphoedematous limb with non-pitting oedema[20,22-24]. Initially, liposuction was piloted in lymphoedema patients in whom vessels could not be identified at the time of attempted lymphovenous shunt procedures, or after such an operation failed[21]. Liposuction has been shown to be a very effective procedure in achieving limb reduction when performed following preoperative DLT to remove all excess oedema of the limb, and when used in conjunction with postoperative compression[22-24]. Complications reported from liposuction include haematoma, recurrent lymphoedema, and damage to the residual lymphatic vessels[20].

Liposuction is now the most commonly performed excisional procedure for the treatment of lymphoedema, as it is a less invasive method of removing subcutaneous fat and associated with fewer complications[17]. The physiological basis which supports liposuction as an effective treatment for lymphoedema, is that excessive adipose tissue deposits have been demonstrated to contribute to the progression of lymphoedema, in addition to increased interstitial fluid, fibrosis and muscle volume[21,25]. Four studies have been identified in the contemporary literature which reported the use of liposuction procedures for lymphoedema-associated limb volume reduction *(Table 1)*. The outcomes of these studies included a volume reduction which ranged from 18% to 118% with a weighted average for the studies of 82%. All 4 of these studies reported liposuction which was performed for lymphoedema of the upper extremity limbs[22,26-28]. The follow-up time ranged from 6 to 26 months.

Lymphatic Reconstruction

Microsurgical procedures have been proposed as a means of improving or restoring lymphatic flow in regions in which lymphatic pathways have been damaged (secondary lymphoedema) or

Table 1: Summary of published literature (2004-2010) related to excisional procedures for the treatment of lymphoedema

Author, Year	Study Design	N° Patients	Lymphoedema Site	Surgical Procedure	Follow-up time (months)	% Volume Reduction	Measurement Technique	Quality Score (Total Score=14)
Salgado *et al.* (2009)	Prospective	11	Upper Extremity	Excision with preservation perforators	17.8	21	Circumference	10
Lee *et al.* (2008)	Retrospective	22	Lower Extremity	Excision	48	not reported	Infrared optometric volumetry & circumference	9
Modolin *et al.* (2006)	Prospective	17	Penile/scrotal	Excision	72	not reported	not reported	2
Brorson *et al.* (2006)	Prospective	35	Upper Extremity	Liposuction	12	103	Water displacement	12
Qi *et al.* (2008)	Prospective	11	Upper Extremity	Liposuction, myocutaneous flap transfer	26	18	Circumference	6
Damstra *et al.* (2009)	Prospective	37	Upper Extremity	Suction-assisted lipectomy	12	118	Water displacement	8
Brorson *et al.* (2006)	Prospective	11	Upper Extremity	Liposuction	6	109	Water displacement	7
Liu *et al.* (2005)	Prospective	11	Upper Extremity	Liposuction	*	*	Circumference	*

* Abstract only available

**Quality score reflects the total number of affirmative responses to quality instrument with a potential maximum of 14.

are missing (primary lymphoedema). A number of reconstructive techniques have been attempted to create a bypass for lymphatic fluid beyond or around regions of damaged lymphatics including creating new connections or anastomoses between:

- two lymphatics (lymphatic-lymphatic anastomosis)
- veins as grafts to create channels to other lymphatics (lymphatic-venous lymphatic anastomosis)
- lymphatics to veins (lymphatico-venous anastomosis)

Lymphatic-lymphatic anastomosis is rarely performed as early postoperative failures related to scarring were often reported. With lymphatic-venous lymphatic anastomosis, a vein graft is sewn using microsurgical suturing techniques between proximal and distal lymphatic vessels to bypass the damaged areas[29,30]. The most commonly reported lymphatic reconstruction technique reported in the literature is lymphatico-or-lympho-venous anastomosis. In this procedure, fine connections (0.3–0.8 mm) are created between distal lymphatics and subdermal venules. The subdermal location of the anastomoses or connections permits the use of small incisions (<3cm) with minimal dissection. The reported complications associated with lymphatic reconstruction are few as there is minimal tissue damage and dissection, but requires surgeons that are highly trained in microsurgery.

A total of 10 contemporary studies have been published which evaluate the use of lymphatic reconstruction for the treatment of lymphoedema *(Table 2)*. Six studies reported the outcomes following lymphatic reconstruction for the treatment of lower-extremity lymphoedema. Patient follow-up ranged from 9 to 87 months with a reported limb volume reduction of 11.3% to 91.7%[8,31-35]. The overall weighted limb volume reduction for all studies was as 44.8%. Two of the studies reported findings from both lower and upper extremity patients with a percentage volume reduction reported of 67% in one study[9,30]. Two of the studies reported upper extremity volume reduction of 2% to 35% volume with a follow-up ranging from 12 to 35 months; the overall weighted reduction for the 2 studies was 9.5%[36,37].

Table 2: Summary of published literature (2004-2010) related to lymphatic reconstructive procedures for the treatment of lymphoedema

Author, Year	Study Design	N° Patients	Lymphoedema Site	Specific Surgical Procedure	Follow-up time (months)	% Volume or Circumference Reduction	Measurement Technique	Quality Score (Possible Total Score=14)
Damstra *et al.* (2009)	Prospective	10	Upper extremity	Lymphatic venous anastomosis	12	2	Water displacement & Circumference	8
Demirtas *et al.* (2009)*	Prospective	42	Lower extremity	Lymphaticovenular anastomosis and/ or lymphaticovenous implantation	11.8	59.3	Circumference	6
Matsubara *et al.* (2006)	Retrospective	9	Lower extremity	Lymphatic venous anastomosis	21-87	> 5 cm (n=6), 2 cm (n=2), no effect (n=3)	Circumference	6
Koshima *et al.* (2004)	Retrospective	52	Lower extremity	Lymphatic venous anastomosis	15	42	Circumference	6
Narushima *et al.* (2010)	Prospective	14	Upper extremity (n=2), lower extremity (n=12)	Lymphatic venous anastomosis	8.9	11.3	Circumference	6
Maegawa *et al.* (2010)	Retrospective	111	Lower extremity	Lymphatic venous anastomosis	Not Reported	Percent not reported - Average 872 ml reduction	Circumference	6
Campisi *et al.* (2010)*	Retrospective	1800	Upper extremity and lower extremity	Lymphatic venous anastomosis	120	56 (83% with 67% reduction)	Water displacement	4
Chang D. (2010)	Prospective	20	Upper extremity	Lymphatic venous anastomosis	18	35	Water displacement	7

*NOTE: Selected among duplicate studies with overlapping patients

**Quality score reflects the total number of affirmative responses to quality instrument with a potential maximum of 14.

Tissue Transfer Procedures

With tissue transfer procedures, healthy lymph nodes and/ or tissue with associated lymphatic vessels are mobilised from a region of the body (i.e. omentum) or unaffected limb (most commonly the medial thigh) and transposed/rotated or transplanted into regions with damaged or missing lymphatics to create a bypass from the damaged lymphatics[20]. Reported complications associated with tissue transfer procedures include significant scarring, bleeding, infection and lymphoedema of the donor site, as well as recurrent lymphoedema of the recipient site[20]. A few studies have documented the maintenance of the volume reduction for up to 3 years[38].

A review of the literature yielded only 4 published articles with more than 8 patients related to the treatment of lymphoedema using tissue transfer procedures *(Table 3)*. A number of single patient or small case studies were not included in the review. These included primarily upper extremity lymphoedema patients with reported follow-up from 12 to 96 months. The reported volume reduction ranged from 51% to 81%[11,39,40], with a weighted volume reduction of 67%. One study of lower extremity lymphoedema, with a reported follow up of over 120 months, noted an increase in limb volume by 13%[10].

Discussion

Findings from the peer-reviewed published literature on the surgical treatment of lymphoedema indicate that these procedures are promising for select groups of patients. In general, excisional procedures were associated with the greatest volume reduction, followed by lymphatic reconstruction and then tissue transfer procedures. However, it is not possible to compare the outcomes of the various surgical techniques in order to identify one surgical technique as more effective than another because of the heterogeneity of patient characteristics and selection criteria which varied substantially among the studies. For example, it is likely that the most dramatic volume reductions were related to excisional procedures performed on limbs with massive lymphoedema, whereas patients with long-standing

Table 3: Summary of published literature (2004-2010) related to tissue transfer procedures for the treatment of lymphoedema

Author, Year	Study Design	N° Patients	Lymphoedema Site	Specifical Surgical Procedure	Follow-up time (months)	% Volume Reduction	Measurement Technique	Quality Score (Possible Total Score=14)
Lin *et al.* (2009)	Retrospective	13	Upper extremity	Vascularised lymph node transfer	56	51	Circumference	6
Hou *et al.* (2008)	Randomized control trial	15	Upper extremity	Autologous bone marrow stromal cell transplant (n=15) versus CDT (n=35)	12	81	Circumference	11
Belcaro *et al.* (2008)	Retrospective case-control	9	Lower extremity	Autologous lymphatic tissue transplant (n=9) versus control (n=8)	120	Increase 13%	Water Displacement	6
Becker *et al.* (2008)	Retrospective	24	Upper extremity	Lymph node transplant	96	Reduction to normal (n=10) Some reduction (n=10) No change (n=2)	Circumference	4

**Quality score reflects the total number of affirmative responses to quality instrument with a potential maximum of 14.

lymphoedema with associated elephantiasis and fibrosis would not likely be candidates for lymphatic reconstruction procedures.

In addition, the published studies are primarily observational studies which do not include comparison groups. The reported success of many of the surgical procedures for the treatment of lymphoedema was likely strongly influenced by the selection of patients which was not well defined in the majority of studies. Randomised clinical trials are designed to compare treatment arms among patients with similar characteristics. The primary advantage of randomised controlled clinical trials is to eliminate patient selection bias, which ensures that the findings and outcomes of the study can be attributed to the procedure or treatment itself rather than to the more favourable characteristics in a particular group of patients.

An important component of determining whether surgical treatment of lymphoedema is an option is to examine the specific procedure and individual patient risk-benefit ratio. The risk-benefit ratio considers 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 that may be life threatening. The individual goals of the patient (i.e., function vs cosmetic), the extent of the surgical procedure, and the level of expertise and experience required to perform the surgery should also be carefully considered. Additional studies are required to select appropriate patient populations who would derive the greatest benefit from surgery.

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CHAPTER 3
How surgery complements a comprehensive lymphoedema service

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Introduction

For a number of years, the approach to the diagnosis and treatment of lymphoedema was segmented. As medical doctors made the medical diagnosis, paramedical healthcare professionals often delivered conservative, treatment with the focus being centred on the reduction of swelling. Indeed, early diagnosis of lymphoedema in patients treated for cancer is generally not incorporated into post-oncological follow-up programmes. Doctors performing a surgical approach for lymphoedema often are not involved in the early diagnosis of lymphoedema or the conservative treatment of a patient.

Lymphoedema is a clinical sign of characteristic tissue swelling as an ultimate consequence of lymph transport impairment. Many inherited and acquired diseases involve the ability of the lymphatics to collect and transport lymph fluid. Lymph stasis is associated with blunted regional immune trafficking, local inflammatory changes with increase adipose tissue formation, and increased risk on infection leading to tissue damage[1].

As all these effects are persistent, lymphoedema should be considered a chronic disease. Like many chronic conditions, individuals often accumulate several of them, a phenomenon known as co-morbidity.

In 1999, the Dutch Institute for Healthcare Improvement (CBO) organised a task force on lymphoedema to evaluate the current literature and to propose evidence and expert-based recommendations suitable for inclusion in the national guidelines for the treatment of lymphoedema. The task force comprised representatives from national medical scientific organisations (for example, surgery, gynaecology, radiotherapy and dermatology, amongst others), paramedical associations and patient support groups. The guidelines were produced in 2003[2,3] and will be revised in 2012. The revision process will review the approach to lymphoedema, from diagnosis, early recognition, awareness, self management, (non) operative treatment, guidance of the patient and follow up, and reflect a new, integrated, interdisciplinary approach.

This chapter outlines the chronic care model (CCM) used in the Netherlands and reflects experiences of practitoners from the Expert Centre for Lympho-vascular Medicine in the Netherlands.

Chronic Care Model (CCM) and the International Classification of Functioning, Disability and Health (ICF)

Chronic Care Model

The principle of the CCM is active patient participation in his or her own treatment, patient empowerment and self-efficacy and a more 'hands-off' approach by health professionals. Effective electronic patient files, accessible by all workers, follow a uniform protocol to support the care process. We hope that outlining this approach can contribute to the discussion regarding provision of a comprehensive lymphoedema service, which includes surgery. Thus, lymphoedema surgery is no longer seen as a unidisciplinary therapeutic entity, rather as fully embedded in a chronic care paradigm.

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While it is well accepted that chronic illness requires a multidisciplinary approach to care, cooperation between various healthcare workers and the patients is often not obvious, or indeed in place. The chronic care model centres on pre-existing or long-term illness, as opposed to acute care, which is concerned with short term or severe illness of brief duration. The model was initially proposed by Wagner[4] in 1988 in response to the acknowledgement by health plans and provider groups, that the care of patients with chronic illness required improvement. Evidence has shown that 'usual care' is not effective for chronic condition management; sizable numbers of chronically ill patients are not receiving effective therapy, have poor disease control, and are unhappy with their care[1]. In addition, chronic medical care accounts for more than 75% of health care dollars spent in the United States, with approximately 125 million (45%) of the population faced with some type of chronic disease[5].

The chronic care model is a pragmatic approach based on the following assumptions:

1. That the patient is the centre of the care process
2. That all professionals integrate into the model and cooperate closely together
3. That the patient has an active role (care manager) rather than being a passive 'care consumer'. Aspects such as self management and self efficacy are very important
4. That healthcare workers do not merely focus on symptom control (for example, reduction of swelling), but act as 'coaches' with a 'hands-off' approach
5. That the model and integrated approach is based on guidelines, evidence based medicine and best practice documents, to which all health professionals are committed
6. That clinimetrics, standardised and validated measuring methods and questionnaires will be used to monitor the effects of treatment programmes
7. That professionals accept the correlation between health improvement, patients' health and care
8. That effective, digital electronic patient files and mutual communication between healthcare workers and patients will be ensured

Lymphoedema is a common chronic and incurable condition, particularly if it is secondary to for example, cancer treatment in which lymph node dissection and/or radiotherapy is involved. In these cases, early diagnosis and treatment is essential to prevent the progression of the disease and its complications - late stage lymphoedema may cause severe physical and psychological problems for patients owing to chronic swelling, impaired physical function, recurrent infections and disfiguring skin changes[6-8]. Secondary prevention[9], awareness of the patients and programs to reduce risk factors such as obesity[10], infection, and immobility can be beneficial[11].

The ICF

Chronic lymphoedema management tends to have a medical, organic focus, particularly in relation to the prevention of infection and treatment of lymphoedema with for example, regular decongestive lymphatic therapy (DLT) or surgery. However, to help patients cope with their disease, aspects of functionality, participation in daily life, work and quality of life need to be considered. These requirements are reflected in the World Health Organization's (WHO) ICF model[12]. The *international classification of functioning, disability and health,* is based on an integrated bio-psycho-social model that allows a standardised description of functioning and disability based on individual and contextual factors. Using the ICF, influences upon a patient's functioning in three domains can be described. These domains are:

1. Body functions and structures
2. Activities of living
3. Participation in activities related to personal and environmental factors

In the management of lymphoedema, monitoring of activity of disease parameters as well as the results of treatment and follow-up is mandatory. Both healthcare professionals and the patient can undertake such monitoring, using validated tools and a protocol covering all the domains of the ICF. While the clinimetric instruments provide tools for objective measuring in the various domains of function in relation to prevention, treatment and follow-up of lymphoedema, they are not necessarily disease specific. Therefore, in the Netherlands a group of experts are working to develop an ICF core set specifically for lymphoedema.

The ICF model was recently studied by Tsauo *et al*[13] in patients treated for breast cancer related lymphoedema (BCRL). They concluded that the ICF model consisting of clinical measures for patients with BCRL, could predict their quality of life. The domains of activity and participation were the most important. Arm symptoms rather than arm volume significantly correlated with arm function. Consequently, arm function, activity and participation are very important in treating a patient, even relatively more important than just focusing on volume and lymphoedema itself.

CCM and ICF in daily practice

Within the Expert Centre for Lymphoedema, our philosophy of lymphoedema care embraces both the domains of the ICF and the concepts of the chronic care model of disease management[14]. Several clinical instruments for all the phases of treatment for the patient with or at risk of developing lymphoedema are used. For its implementation in clinical practice, the ICF-based instruments, such as the ICF core sets were developed in a standardised scientific process.

When a patient has been diagnosed with lymphoedema and the treatment started, the initial measurements are concerned with the oedema and assessment of other risk factors, such as pain, loss of joint mobility, strength, physical capacity and emotional distress. In this phase, the frequency of measurement is high.

In the maintenance phase, the desired level of activity and participation of the patient are central to therapy. To achieve this, we use measurement tools such as DASH (Disabilities of the Arm, Shoulder and Hand) and objective questionnaires regarding Health Related Quality of Life. As therapy progresses, the measurement frequency decreases and self-monitoring becomes more important.

The role of self-management

The CCM allows self-management as a core component. Self-management is the ability of an individual to cope with symptoms, treatment, physical and social consequences and lifestyle changes related to a life living with a chronic disease[15]. To achieve this goal, four domains are offered *(Box 1)*:

Box 1: Self-management domains

1. Activities focused on health improvement and build-up of physical resistance
2. Coping with healthcare providers and compliance to treatment
3. Ability to self-monitor health, draw conclusions from signs and symptoms and translate this to decisions (for example, going to see a doctor, to start self bandaging). Self-efficacy is part of self-monitoring
4. Coping with the consequences of a chronic disease and trying to get control by self-regulation of personal, behavioral and environmental factors

To stimulate a patient to become a self-regulator and to practice self-management, appropriate knowledge and skills are necessary. Consequently, treatment is much more than merely performing an operation or providing DLT. Healthcare workers should provide the patient with the skills for self-care *(Box 1)*, assess their ability to undertake them, and monitor with the patient, their continued ability and proficiency (as this may change over time as disease progresses). Ultimately, the multiprofessional team should work towards the patient being able to:

- understand their disease, its manifestations and its effect on their personal life and social context
- both manage and accept the disabilities and limitations on daily functioning caused by the disease (or its complications as in lymphoedema)
- undertake self-monitoring, self-intervention (for example, bandaging or exercise) and risk management
- cooperate with healthcare workers
- access and use appropriate resources such as care, money from insurance companies, and devices

It is important to realise that all these aspects of self-care have limitations in terms of knowledge, making judgments and decisions, and in terms of patient engagement in result-achieving actions[16].

Most lymphoedema literature relates to breast cancer-related lymphoedema. Bogan *et al*[17] described seven patients with non cancer-related lower limb related oedema who where initially treated as in-patients with DLT, and were instructed in self-exercise, self-management and symptom awareness. Participants reported the difficulty in finding a correct diagnosis and effective treatment, the importance of their inpatient experiences, and the challenges of daily self-management. The authors recommend an increase in lymphoedema awareness, promotion of inpatient treatment programs, and the requirement for effective self-management techniques.

Since 2005, Brouwer *et al*[18] have presented over fifty dedicated post-oncological treatment awareness courses (comprising two sessions) for all patients treated for breast cancer and axillary dissection. The rationale for the development of the course was based on the premise that by this stage, a patient is more concerned with returning to a functioning daily life than on cancer survival. The awareness course replaced the traditional information about the risk of lymphedema during cancer treatment. Patients were more able to incorporate the information and skills.

Organisation of care

When organising a comprehensive lymphoedema service according these new ideas, a major part of the treatment is well structured programme on awareness, prevention and self

management. The goal is to stratify patients in terms of the risk of developing lymphoedema (low risk versus high risk) and in terms of severity. A pyramid of care is proposed by the Kaiser Permanente[19]; and represents collaboration between health professionals, insurance companies, hospitals and primary care groups *(Figure 1)*. The goal is to provide integrated care for chronic diseases with a strong scope for prevention and cost reduction. Similar networks have been developed by the Mayo clinics in the United States of America, and in Canada and Europe.

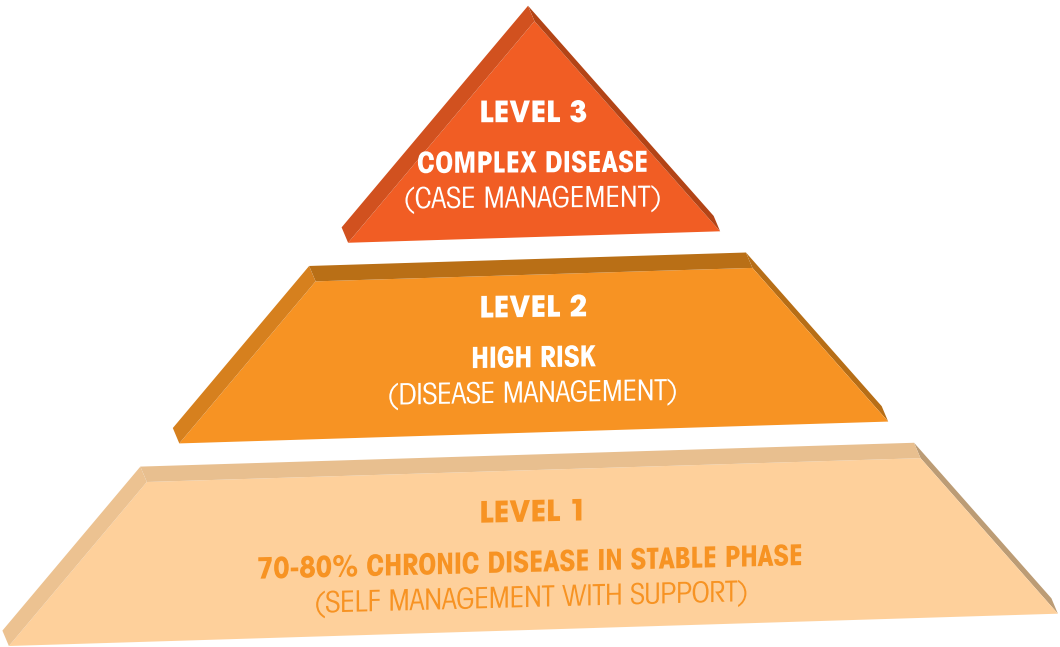
The 5 characteristics of the Pyramid of care are:

1. Attention to prevention of complications and self management
2. Use of a electronic patients file /e-health
3. Collaboration between healthcare professionals; medical doctors, paramedical, home nurses within a network
4. Medical leadership/ protocol keeping
5. Integrated funding of the whole comprehensive service

In this model, patients receive treatment and follow-up individualised to their care requirements. In order to achieve good stratification, the ICF model provides a core set of objectives and validated measurement tools on all domains of functioning such as outlined previously. When this process is done in a proper way, the therapeutic program comprises interventional tools as DLT and self management (including raising awareness, empowerment of the patient through learning to treat their own swelling with self bandaging, exercises, weight reduction, compression garments and skin care[20]. Richardson *et al*[21], in a study comparing the effectiveness of an expert patient program (EPP) in which self-care was taught in order to improve health outcomes and patients' satisfaction. The intervention comprised six 2.5 hour group sessions (8-12 individuals), covering topics such as relaxation, diet, exercise, fatigue and 'breaking the symptom cycle'. They demonstrated cost effectiveness and improvement of quality of life compared to the control group.

The Dutch Expert Centre for Lympho-vascular Medicine offers self-management courses based on EPP. All patients after axillary dissection, inguinal dissections, patients with erysipelas as first sign of lymphatic impairment shown by quantitative lymphoscintigraphy, and patients with partly reversible primary lymphoedema (Stage 1) were included[22]. To date, as a result of this programme in our centre, there is no severe breast cancer related lymphoedema and recurrence of erysipelas is below 0.2% per year.

Figure 1 Pyramid of care



At level 3, many health care professionals are involved and work with centres of excellence to provide patient-centred treatment. At level 2, treatment can be offered at the primary care level in an ambulatory setting. All hospitals practicing cancer treatment undertake early diagnostics and self management programmes. At level 1, all patients are well instructed in self-care and tend to have less contact with their healthcare professional and fewer follow-ups.

Since 1995 the centre has organised all interventions around the patients' requirements. Our focus is:

- to improve and standardise diagnostics
- early recognition of lymphoedema in the cancer department
- to start early treatment programs and stimulate self-management and awareness

More recently, clinical programmes for severe cases of lymphoedema were designed which comprised both non-surgical and surgical interventions, the latter being fully embedded in conservative treatment programmes for patients with stage 3 lymphoedema. Intensive (non) operative treatment is provided on a 12-bedded inpatient ward. Since 2009, the centre has been certified as an expert centre for end stage clinical care, secondary and tertiary referral, education and research. We try constantly to implement new concepts in chronic care and plan to implement a nationwide satellite network which will ensure all professionals will work to the same protocols, measuring devices and materials.

Interestingly, a recent study was published exploring how the chronic care model could be adjusted to the Dutch situation[23]; the recommendations from the study are reflected in our daily practice. Indeed, we are actively working with other hospitals internationally and our concept stimulated the Larissa group (Greece) to establish a multidisciplinary lymphoedema treatment programme[24]. An initiative of a multidisciplinary in Denmark[25] shows large similarities with our initial working group. Our concept of implementing ICF/CCM method into the multidisciplinary lymphoedema centre, including surgery in lymphoedema, can probably be a stimulus for others.

Mutual electronic communication and patient files

Crucial for an integrated service is a transparent and uniform communication system. Since 1998, the department has been active in utilising e-health services and developed with others, a dermatologic teleconsultation system, now embedded in the Dutch healthcare system[26,27]. This expertise was helpful in designing a dedicated system for lymphological care based on ChipSoft electronic healthcare information system (CS-EHIS) (Amsterdam, NL), which provides a complete and flexible IT solution, a high degree of configurability, overall user-friendliness and automates the hospital organisation. The CS-EHIS is comprised of functional groups (modules): we developed a separate module based on the ICF method and used by all participants; patient related data, history, physical examinations, laboratory and radiological results and pictures, are all available. The module also includes all clinimetrics and an integrated garment ordering system, all (financial) logistics, diagnoses, clinical pictures. Data, both medical and logistic are collected and easily reproduced for management and research.

How surgery complements a comprehensive lymphoedema service

In general, two types of surgery for lymphoedema can be distinguished:

1. Surgery to improve lymphoedema and reduce swelling
2. Concomitant surgery in a lymphoedematous limb

Surgery for lymphoedema is thus concerned with reconstruction or debulking. Although the scientific evidence in term of efficacy and efficiency of some procedures is still not clear, none of these treatments will succeed without clear indications for intervention, a proper pre-and-post operative conservative treatment protocol and multidisciplinary cooperation, all embedded in an integrated programme. Surgery is an element of chronic care management; according to the CCM, surgery should be undertaken in an expert centre, in collaboration with all healthcare workers working on other levels of the 'pyramid' *(Figure 1)*. Generally, surgical intervention is an option only to be considered where other more conservative interventions have failed.

Education and research

The Dutch Lymphological Centre Foundation (SLCN, www.slcn.nl) is related to the Expert Centre and provides professional educational activities. This education is internal for the staff, and external for all involved in lymphoedema care in the Netherlands. These education activities are ongoing and comprise theoretical and practical issues for all types of health care personnel. We cooperated closely with all professional societies and develop dedicated courses and workshops for them. There is a close cooperation with the patients' platform, the Dutch Lymphoedema Network (NLNet), the official representative of the ILF in the Netherlands. (www.lymfoedeem.nl)

Because of the concentration of knowledge and skills in the expert centre, we perform research in many fields of interest. These include understanding mechanisms of effectiveness of compression and lymph drainage, studying new measurement devices, research in the field of operative lymphoedema treatment and studies in the field of integrating ICF and clinimetry in the lymphoedema practice. These results can hopefully contribute to best practice documents and guidelines in order to provide all patients with proper treatment of their chronic condition in a cost effective way.

Conclusion

Lymphoedema treatment has historically been provided in a solitary and unidisciplined way focusing on medical disease and swelling. We propose that a comprehensive lymphoedema service based on the chronic care model and using the ICF classification is a vehicle for determining and delivering the needs of a patient. This service includes surgery as a small, but important therapeutic option for last resort lymphoedema patients and is fully integrated in the service. Prevention, early non-operative treatment is still the majority of treatment. The Expert Centre for Lympho-vascular Medicine is integrated in the Dutch public health system.

By using the CCM for a lymphoedema service, the provision of care to the patient can be regionally/locally organised (where possible) and when necessary, in highly complex cases or when there is a need for operative treatment, should be provided in a central expert centre. Because the Expert Centre works closely with other healthcare workers and in some instances, are connected to a satellite treatment centre, according to the same protocol and connected to the same IT environment, all patients can receive good guidance and treatment.

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CHAPTER 4
The role of Circumferential Suction Assisted Lipectomy (liposuction) and compression in limb lymphoedema

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Introduction

The various types of treatment of lymphoedema are under discussion and there has been some controversy regarding liposuction for late-stage lymphoedema. In general, treatment of lymphoedema consists of two phases, the initial treatment phase and the maintenance phase. Lymphoedema treatment includes conservative and operative treatment.

A therapeutic treatment program should be individualised to the patient according to the stage of lymphoedema, patients' related co-factors such as age, co-morbidities, prognosis of (malignant) disease, psychosocial aspects, weight, hypertension and physical potential to exercise[1-3].

Presently there is no cure for lymphoedema. With early diagnosis, the majority of patients can be treated by conservative treatment, such as Decongestive Lymphatic Therapy (DLT)[4], which comprises manual lymph drainage, compression therapy, physical exercise, skin care and self-management, followed by wearing flat knitted compression garments. The effect of DLT on longstanding massive oedema with excess adipose tissue is poor, since adipose tissue does not disappear by means of compression alone. In addition, surgical intervention is reserved for patients with excess volume and heaviness causing severe strain in the shoulder and neck, functional impairment, recurrent attacks of erysipelas, and problems with clothing fit.

For the treatment of late-stage lymphoedema that does not respond to conservative treatment, circumferential suction assisted lipectomy (CSAL or `liposuction) combined with postoperative, life-long effective compression therapy has became a viable alternative. Postoperative follow-up and regular adjustment of the compression technology is mandatory.

This best practice document outlines the benefits of using CSAL in late stages of lymphoedema when excess adipose tissue dominates the swelling, and presents the evidence to support its use.

Excess subcutaneous adiposity and chronic lymphoedema

The incidence of post-mastectomy arm lymphoedema varies between 6% and 49%, depending on the combination of therapy, including mastectomy, sentinel node biopsy, standard axillary lymph node dissection and/or postoperative irradiation[5-7]. More recently, factors such as obesity, baseline general health, and limb and joint function are thought to contribute to the development of aggravation of lymphoedema[8,9].

The outcome of the surgical procedure combined with irradiation of the tissue often results in destruction of lymphatic vessels. When combined with the removal of lymph nodes and tissue scarring, the lymphatic vessels that remain are likely to be unable to remove the lymph load. The remaining lymph

collectors become dilated and overloaded and their valves become incompetent, preventing the lymphatics from performing their function. This failure spreads distally until even the most peripheral lymph vessels, draining into the affected system, also become dilated[10].

In a parallel process, the cells of the mononuclear phagocytic system of the mesenchymal tissues begin to lose their capability to remove the protein that accumulates. The accumulated interstitial proteins, as osmotically active molecules, attract fluid to the area. This accumulation of protein and fluid is usually a transitory phase, lasting between one and three weeks.

In the latent phase, which varies from about four months to ten years, there may still be no clinical signs of any discernable lymphoedema. At the end of the latent phase, pitting of the oedematous arm on pressure can be observed. This can be objectively measured by plethysmography and by decreased tissue compressibility measured using a tissue tonometer[11].

Over time, there is also an increase in the adipose tissue content of the swollen arm. One of the authors (HB) has observed this clinically since 1987, when the first lymphoedema patient in his department underwent surgery[12,13]. The enlargement of the arm leads to discomfort and complaint of heaviness, weakness, pain, tension and a sensory deficit of the limb. In addition, anxiety, psychological morbidity, maladjustment and social isolation[14] and increasing hardness of the limb[15] are seen.

There are various possible explanations for the adipose tissue hypertrophy. There is a physiological imbalance of blood flow and lymphatic drainage, resulting in the impaired clearance of lipids and their uptake by macrophages[16]. However, there is increasing support for the view that the fat cell is not simply a container of fat, but is an endocrine organ and a cytokine-activated cell[17,18] and that chronic inflammation plays a role here[17,19]. The same pathophysiology applies for primary and secondary leg lymphoedema. Recent research showed a relationship between slow lymph flow and adiposity, as well as that between structural changes in the lymphatic system and adiposity[20,21].

From a more clinical view, other indications for adipose tissue hypertrophy have been found:

- consecutive analyses of the content of aspirate showed a very high content of adipose tissue in 44 women with post-mastectomy arm lymphoedema (mean 90%, range 58– 100)[22]
- analyses with dual X-ray absorptiometry (DXA) in women with arm lymphoedema following a mastectomy showed a significant increase of adipose tissue in the non-pitting swollen arm before surgery[23]
- preoperative investigation using volume rendered computer tomography (VRCT) images in 8 patients, showed a significant preoperative increase of adipose tissue in the swollen arm, the excess volume consisting of 81% (range 68–96) fat[24]
- Damstra *et al's*[25] study of 37 patients with end-stage breast cancer related lymphoedema found that the volume of aspirate removed (when a tourniquet was used) was 953 ml (range 315-1700 ml), and the proportion of fat in the aspirate was 93% (range 59-100%)
- tonometry findings in 20 women with post-mastectomy arm lymphoedema showed postoperative changes in the upper arm, but not in the forearm, which also showed significantly higher absolute values than in the upper arm. This is probably caused by the high adipose tissue content with little or no free fluid, as in the normal arm. The thinner subcutaneous tissue in the forearm may also play a part[11]

Tonometry can distinguish if a lymphoedematous arm is harder or softer than the normal one. If a lower tissue tonicity value is recorded in the oedematous arm, it indicates that there is accumulated lymph fluid in the tissue, and thus patients are candidates for conservative treatment methods. In contrast, patients with a harder arm compared with the healthy one, have an adipose tissue excess that can successfully be removed by liposuction[11].

The increase of adipose tissue due to inflammation has also been described in other diseases. For example, an increase of adipose tissue in intestinal segments in patients with Crohn's disease, known as 'fat wrapping', has clearly shown that inflammation plays an important role[19,26,27]. In Graves' ophthalmopathy, an increase in intraorbital adipose tissue volume leading to exopthalmus has been seen. Adipocyte related immediate early genes (IEGs) are overexpressed in active ophthalmopathy and cysteine-rich, angiogenic inducer 61 (CYR61) may have a role in both orbital inflammation and adipogenesis and serve as a marker of disease activity[28].

Liposuction

Liposuction is the most common procedure in plastic surgery and is mainly performed for cosmetic purposes. To a lesser extent it has been used for reconstructive 'insulin tumours' caused by the injection of insulin into the subcutaneous fat[29], multiple familial angiolipomatosis[30], gynaecomastia[31], benign symmetricallipomatosis[32] and lipoedema[33-34].

Recently Berry *et al*[35] published an overview of the numerous techniques and recent advances. Initially, liposuction was done as a 'dry' technique, with no dilute adrenaline or anesthetics being injected into the adipose tissue beforehand[36]. A disadvantage of this technique was the large amount of blood loss.

Illouz was the first to infiltrate the subcutaneous fatty tissue when doing liposuction[37]. In the early 1980s, most surgeons used the 'wet' technique[38], which involves infiltration of 200–300ml of normal saline with or without lignocaine, adrenaline, or a combination, into the surgical area before liposuction. In 1986 the 'superwet' technique was introduced, which involves infiltration of a solution of normal saline containing adrenaline and lignocaine in an amount equal to that of the fat that is to be removed[35].

The following year, Klein described the 'tumescent' technique, which involves somewhat larger amounts of saline containing both low-dose adrenaline and lignocaine in a ratio of 2–3:1ml (infiltrate: aspirate) being injected[39]. These techniques enabled surgeons to remove large quantities of adipose tissue. By infiltrating dilute adrenaline and lignocaine into subcutaneous fat, both the excessive loss of blood and the need for general anesthesia with its associated risks are reduced[40]. Guidelines for tumescent liposuction were stated in 2006[41].

According to other authors, more than 3000ml of fat can be removed during liposuction under local anesthesia without sedation[39]. Samdal *et al* reported the amount of whole blood contained in the aspirate is roughly 2% (volume/volume) when super wet or tumescent techniques are used[42], whereas in the dry technique it is 25%[38], and in the wet technique, 15%[36].

The 'dry technique' was used initially to treat arm lymphoedema following breast cancer treatment. Later, to minimise blood loss, a tourniquet was used in combination with tumescence. Liposuction was performed up to the distal edge of the tourniquet. A sterile compression garment was put on and the tourniquet was released. The area covered by the tourniquet was infiltrated with dilute adrenaline before the liposuction was completed[25,40] *(Figure 1)*.

Newer techniques involve ultrasonic assisted liposuction (UAL), laser-assisted liposuction (LAL), and power-assisted liposuction (PAL). UAL and LAL generate energy that is transformed into heat that can damage the skin. The authors do not use these techniques for lymphoedema. However, PAL is of great benefit as the vibrating cannula facilitates liposuction, especially in the leg.

The outcome of liposuction

The common understanding among clinicians is that the swelling of a lymphoedematous extremity is due purely to the accumulation of lymph fluid, which can be removed by use of non-invasive conservative regimens such as Decongestive lymphatic therapy (DLT).

Microsurgery and lympho-venous anastomosis (LVA) have been studied for a long time without convincing results[43]. Although

Figure 1: Liposuction of arm lymphoedema

The procedure takes about two hours. From preoperative to postoperative state (left to right). Note the tourniquet, which has been removed at the right, and the concomitant reactive hyperaemia

Figure 2a and 2b

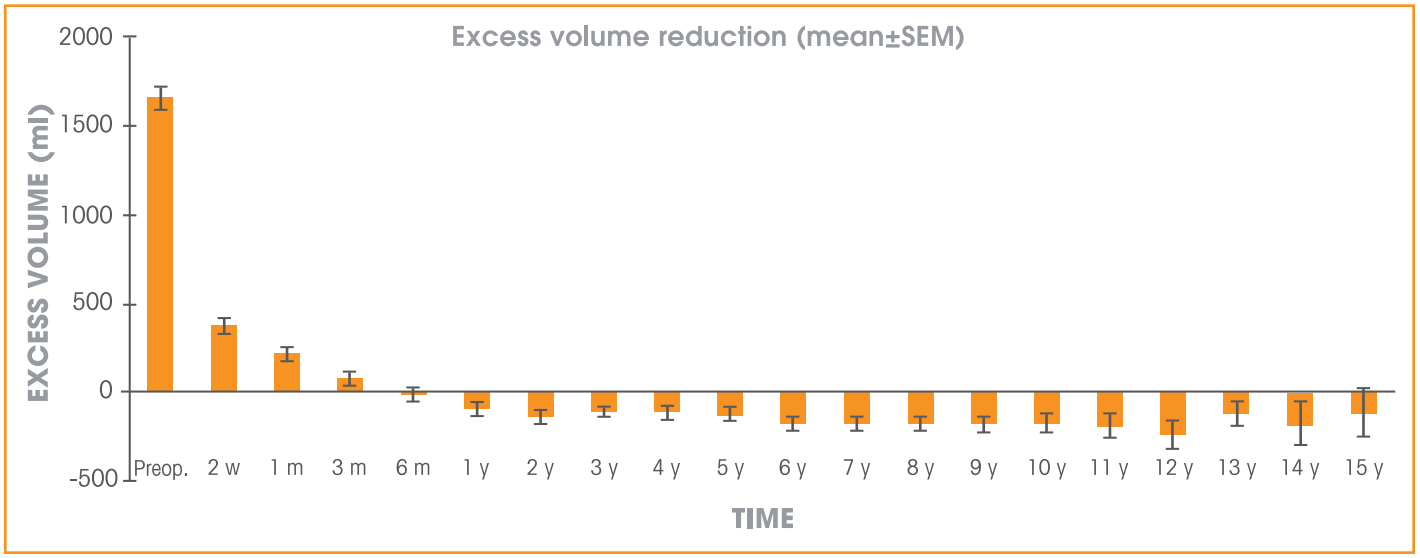
***2a:** A 74-year-old woman with a non-pitting arm lymphoedema for 15 years. Preoperative excess volume was 3090ml*
***2b:** Postoperative result*

the LVA has been performed and studied for more than three decades, this method still has not had a breakthrough and will never become a treatment of choice in daily practice. In a large overview article by Campisi *et al*[44], a positive effect was described in early stages of lymphoedema. However, for later, more irreversible stages, this therapeutic option was not suitable. Moreover, a recent study showed that the net effect of LVA was minor and that the outcome was due to the DLT performed pre-and postoperatively[45].

Brorson *et al*[13] concluded than when the excess volume is dominated by adipose tissue, supra-facial clearance by liposuction is the only method to achieve up to 100% volume reduction.

Today, chronic non-pitting arm lymphoedema of up to four litres in excess can be effectively removed by use of liposuction and compression therapy, without any further reduction in lymph transport[46]. Long-term results, up to 15 years, have not shown

Figure 3: Mean (±SEM) postoperative excess volume reduction in 116 women with arm lymphedema following breast cancer



any recurrence of the arm swelling[12,13,47] *(Figures 2a,2b,3),* In 2009 Damstra *et al*[25] reproduced these results in a large study with 37 breast cancer-related lymphoedema (BCRL) patients. A recent publication from 2012 with a 5-year follow-up in 12 patients with breast cancer related lymphoedema confirmed no recurrence with this technique[48]. Promising results can also be achieved for leg lymphoedema[47-50] *(Figures 4a, 4b).*

How to perform liposuction for lymphoedema

Surgical technique

Liposuction technique for leg lymphoedema is similar to that for the arm. By the use of liposuction, the excess hypertrophied adipose tissue is removed under bloodless conditions *(Figure 4a, b).* General anesthesia is used in most cases, but some patients with arm lymphoedema prefer nerve block in combination with a plexus and scalenus block. Neither local anesthetic nor epinephrine is injected distal to the tourniquet; hence the 'dry technique' is used. Through approximately 15–20, 3mm long incisions, the shoulder and arm are treated *(Figures 1 and 5).*

Cannulae are connected to a vacuum pump giving a negative atmospheric pressure of 0.9. The cannulae are 15cm long with an outer diameter of 3 and 4mm, and they have three openings at the tip. The finer cannula is used mainly for the hand and the distal part of the forearm, and also when irregularities are remedied. The openings differ from normal liposuction cannulae in that they take up almost half of the circumference to facilitate the liposuction, especially in lymphoedema with excess fibrosis.

Compression therapy is mandatory. For this, either bandages up to the axilla and shoulder (including the fingers) with a multilayer inelastic bandage comprising a foam layer and two short-stretch cotton bandages or made-to-measure compression garments

Figure 4a: Secondary lymphoedema:

Preoperative excess volume 7070 ml (left). Postoperative result after six months where excess volume is -445 ml, i.e. the treated leg is somewhat smaller than the normal one (right)

Figure 4B: Primary lymphoedema:

Preoperative excess volume 6630ml (left). Postoperative result after two years where excess volume is 30 ml (right)

Figure 5a, b and C

5a: *Preoperative picture showing a patient with a large lymphoedema (2865ml) and decreased mobility of the right arm*

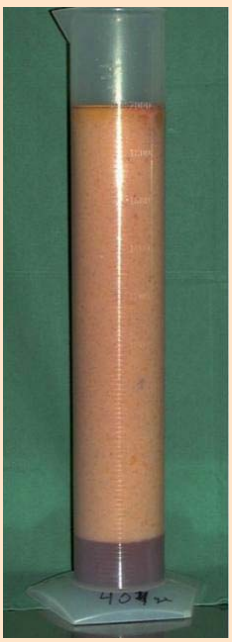
5b: *The cannula lifts the loose skin of the treated forearm*

5c: *The distal half of the forearm has been treated. Note the sharp border between treated (distal forearm) and untreated (proximal arm) area.*

(two sleeves and two gloves) can be used. In the latter case, these are ordered two weeks before surgery. The size of the garments is measured according to the size of the healthy arm and hand. In stock we always have standard interim gloves and gauntlets (a glove without fingers, but with a thumb), used as described below. Liposuction is executed circumferentially, step-by-step from hand to shoulder, and the hypertrophied fat is removed as completely as possible *(Figures 1,5,6)*.

When the arm distal to the tourniquet has been treated, a sterilised made-to-measure compression sleeve is applied (Jobst® Elvarex® BSN medical, compression class 2) on the arm to stem bleeding and postoperative oedema. A sterilised, standard interim glove (Cicatrex interim, Thuasne®, France), with the tips of the fingers cut to facilitate gripping, is put on the hand. The tourniquet is removed and the most proximal part of the upper arm is treated using the tumescent technique. Finally, the proximal part of the compression sleeve is pulled up to compress the proximal part of the upper arm. The incisions are left open to drain through the sleeve. The arm is lightly wrapped with a large absorbent compress covering the whole arm (60 x 60cm, [Cover-Dri]). The arm is kept at heart level on a large pillow. The compress is changed when needed. Another technique is the use of sterilised short-stretch bandages, which are applied during the operation and revised directly at the recovery room by a dedicated bandaging team.

Figure 6



The aspirate contains 90–100% adipose tissue in general. This picture shows the aspirate collected from the lymphedematous arm of the patient shown in Figures 1, 5 and 7 before removal of the tourniquet. The aspirate sediments into an upper adipose fraction (90%) and a lower fluid (lymph) fraction (10%)

The following day, a standard gauntlet (Jobst® Elvarex® BSN medical, compression class 2) is put over the interim glove after the thumb of the gauntlet has been cut off to ease the pressure on the thumb. If the gauntlet is put on straight after surgery, it can exert too much pressure on the hand when the patient is still not able to move the fingers after the anaesthesia.

Operating time is, on average, two hours. An isoxazolylpenicillin or a cephalosporin is given intravenously for the first 24 hours and then in tablet form until incisions are healed, about 10–14 days after surgery.

Postoperative care

The arm is held raised by the patient themselves during the hospital stay. Garments or bandages are removed 2 days postoperatively so that the patient can take a shower. When bandages are used, at the first bandage change, made-to-measure, flat knitted garment are measured and ordered. Then, the other set of garments is put on and the used set is washed and dried or the arm is re-bandaged. The patient themselves repeat this after another 2 days before being discharged. The standard glove and gauntlet is usually changed to the made-to-measure glove at the end of the stay *(Figure 7)*. When bandages are used, after 2 days a special order procedure of the garments guarantees delivery within 5 days and the patient can be discharged between 4-8 days post operatively.

The patient alternates between the two sets of garments (2 sleeves and 2 gloves) during the first two postoperative weeks, changing them daily or every other day so that a clean set is always put on after showering and lubricating the arm. After the two-week control, the garments are changed every day after being washed. Washing 'activates' the garment by increasing the compression due to shrinkage. It also removes perspired salt that can cause dry and irritated skin.

During the subsequent course, this rigorous compression regimen is maintained exactly as described below.

Compression in lymphoedema

The primary goal in lymphoedema treatment is to eliminate oedema and prevent it from recurring. Compression therapy is an effective way of reducing oedema and has been extensively studied in chronic venous insufficiency[51]. Compression therapy can be subdivided into the initial phase of treatment (phase of oedema reduction), which consists of bandaging, and the maintenance phase when hosiery is worn. Box 1 outlines the effects of compression on reducing lymphoedematous limbs and explained by the following mechanisms[52-54].

Box 1: Effects of compression

1. Reduction of capillary filtration
2. Shift of fluid into non-compressed parts of the body
3. Increased lymphatic absorption in lymphatic and venous capillaries
4. Stimulation of lymph transport
5. Improvement of the venous pump leading to reduction of venous filtration to the interstitium (reduction lymphatic preload)
6. Reduction of inflammation as seen in vasculitis allergica and erythema nodosum treatment[55]

The pressure of compression is different on the legs compared to the arms. It is important to note that the hydrostatic pressure that must be overcome by external compression is much higher in the legs than in the arms. In a standing position, the venous pressure in the distal leg is equal to the weight of the blood column between the heart and the measuring point, which is about 80-100mmHg. The high intravenous pressure in the upright body position always increases the lymphatic load by promoting increased fluid extravasation. High external pressure is necessary in order to counteract this extravasation. The venous pressure in the arm is much lower than that in the leg due to the lower weight of the blood column between heart and the hand. Thus, less external compression will be needed to reduce extravasation from the venules into the tissue and to promote reabsorption of tissue fluid. The arm volume reduction from bandaging is probably due not only to a pressure-dependent shift in Starling's equilibrium but also to stimulation of lymphatic drainage.

Besides veno-dynamic issues, lympho-dynamic issues should also be considered. In healthy arms, the distance from the arm to the thoracic duct is short, and the intra-lymphatic pressure varies with the intra-thoracic pressure. Lymphatic drainage is stimulated with relatively low or even negative intra-lymphatic pressure. In BCRL, lymphatic drainage is deficient because of damage to the major lymph collectors and lymph nodes by surgery and/or radiation, leading to lymphatic congestion[56].

Figure 7

The compression garment is removed two days after surgery so that the patient can take a shower. When bandages are used, at the first bandage change, made-to-measure, flat knitted garment are ordered. Then, the other set of garments is put on and the used set is washed and dried or the arm is re-bandaged. A significant reduction of the right arm has been achieved, as compared with the preoperative condition seen in Figure 5a

Figure 8a and 8b

8a: *Marked lymphoedema of the arm after breast cancer treatment, showing pitting several centimeters in depth (grade I oedema). The arm swelling is dominated by the presence of fluid, i.e. the accumulation of lymph*

8b: *Pronounced arm lymphoedema after breast cancer treatment (grade II oedema). There is no pitting in spite of hard pressure by the thumb for one minute. A slight reddening is seen at the two spots where pressure has been exerted. The 'oedema' is completely dominated by adipose tissue. The term 'oedema' is improper at this stage since the swelling is dominated by hypertrophied adipose tissue and not by lymph. At this stage, the aspirate contains either no, or a minimal amount of lymph*

In general, two main effects of compression on the lymphatics have to be considered. The first of these is an increase in the tissue pressure, leading to stretching of the anchoring filaments attached to the initial lymphatics, which causes the opening of initial lymph capillaries. Another is enhancement of the spontaneous contractions of the lymph-collectors that normally occurs under the influence of rhythmic pressure changes[10].

Inelastic compression material exerts relatively low resting pressure and high massaging pressure peaks during movement and may promote autonomous lymphatic contractions. The pressure required to achieve optimal oedema reduction obviously depends on the underlying pathology in different body regions and is, therefore, difficult to assess.

In order to practice more evidence-based medicine, the International Compression Club (ICC) proposed guidelines to measure the pressure in lower leg compression therapy[57] to optimise the therapeutic effect of compression. This guideline was recently introduced to study compression devices and methods[58].

When maximum oedema reduction is achieved, a tailor-made compression garment is applied to maintain the optimal effect. Medical elastic compression stockings (MECS) are effective in the treatment of chronic venous insufficiency and are particularly effective under dynamic conditions. They work, among other ways, by improving venous haemodynamics and reducing oedema. This can be ascribed to their physical characteristics, the most important of which are elasticity and stiffness[59]. In lymphoedema, these characteristics are equally important. To achieve optimal effectiveness, garments should be tailor-made and always flat-knitted.

Compression in circumferential suction assisted lipectomy : the maintenance phase

A prerequisite to maintaining the effect of CSAL, and, for that matter, conservative treatment, is the continuous use of a compression garment. Compression therapy is crucial, and its application is therefore thoroughly described and discussed at the first clinical evaluation. If the patient has any doubts about continued CCT, they are not accepted for treatment. After initiating compression therapy, the custom made garment is taken in at each visit using a sewing machine, to compensate for reduced elasticity and reduced arm volume. This is most important during the first three months when the most notable changes in volume occur. At the one-and-three-month visits the arm is measured for new custom-made garments. This procedure is repeated at six, (nine) and 12 months. If complete reduction has been achieved at six months, the nine-month control may be omitted. If this is the case, remember to prescribe garments for six months, which normally means double the amount that would be needed for three months. It is important, however, to take in the garment repeatedly to compensate for wear and tear. This may require additional visits in some instances, although the patient can often make such adjustments herself. When the excess volume has decreased as much as possible and a steady state is achieved, new garments can be prescribed using the latest measurements. In this way, the garments are renewed three or four times during the first year. Two sets of sleeve and glove garments are always at the patient's disposal; one being worn while the other is washed. Thus, a garment is worn permanently, and treatment is interrupted only briefly when showering and, possibly, for formal social occasions. The patient is informed about the importance of hygiene and skin care, as all patients with lymphoedema are susceptible to infections and keeping the skin clean and soft is a prophylactic measure[60,61].

The life span of two garments worn alternately is usually four to six months. After complete reduction has been achieved, the patient is seen once a year when new garments are prescribed for the coming year, usually four garments and four gloves (or four gauntlets) or even more.

For legs, the authors' teams often use up to two to three compression garments on top of each other, depending on what is required to keep pitting away. A typical example is flat-knitted compression garments class 2 for the lower leg in combination with class 3-4. Sometimes a second leg-long class 2 might be needed. Other compression technologies include self-bandaging with short stretch bandages, using Velcro-compression devices or combinations of both.

Outcome parameters circumferential suction assisted lipectomy

As lymphoedema is a chronic disease, long-term guiding and monitoring is necessary. For that purpose, validated and objective measurements instruments are needed to perform clinimetrics on a regular basis to adjust the initial or maintenance treatment program. In general, patients with lymphoedema suffer from varying degrees of severity from swelling, limited range of motion, pain, loss of muscle strength and fatigue. Related to these problems, activities of daily living such as personal care, walking, housekeeping, sports activities and working are limited. Subsequently, the overall quality of life for people with lymphoedema is often significantly affected.

With the utilisation of the International Classification of Functioning, Disability and Health (ICF), based on the bio-psycho-social model, influences upon a patient's functioning, including body functions and structures, activities and participation in relation to personal and environmental factors, can be described[62]. Recently Tsaou *et al*[63] suggested a model for breast cancer related lymphoedema.

In the management of lymphoedema, monitoring of activity of disease parameters as well as results of treatment and follow up is mandatory. Health care professionals and the patient perform monitoring. Such checks require validated measurements in a protocolled schedule on all domains of the ICF. We use several clinical instruments for all the phases of treatment for the patient with or at risk of developing lymphoedema. After the oncological surgery phase, when the lymph system is impaired, secondary prevention consists of volumetry and Body Mass Index and is regular measured during oncological follow-up.

When a patient has been diagnosed with lymphoedema and the treatment has started, the initial measurements are aimed at the oedema itself, but also at the presence of risk factors, pain, loss of joint mobility, strength, physical capacity and emotional distress. In this phase, the frequency of measurement is high. In the maintenance phase, the desired level of activity and participation are leading for the therapy itself, but also for the instruments and the frequency of measuring. For example, the DASH (Disabilities of the Arm, Shoulder and Hand) and objective questionnaires regarding Health Related Quality of Life are utilised. The frequency of measuring decreases and the role of self-monitoring becomes more important.

Limb volume measurements

Arm volumes are recorded for each patient using the water displacement technique or the inverse volumetry method, which is validated for arm volume measurements and considered the new gold standard[64]. Both arms are always measured at each visit, and the difference in arm volumes is designated as the oedema volume. The decrease in the oedema volume is calculated as a percentage of the preoperative value.

In leg lymphoedema, water displacement is also suitable. Major downsides are that is time consuming, sometimes messy and an elevator is needed to position the leg into the water container. An adequate alternative is by opto-electric measurement which is validated for leg swelling[65,66].

Arm and leg volumes can also be calculated by measuring circumferences every 4cm along the extremity[67,68], using the volume of the truncated cone. Such volume programs can be downloaded from http://www.plasticsurg.nu

A multidisciplinary lymphoedema team

Circumferential suction assisted lipectomy in lymphoedema is a specialised treatment, which should only be performed in a dedicated, multidisciplinary expert centre. To investigate and treat patients with lymphoedema, a team comprising a (plastic/vascular) surgeon, a dermatologist, an occupational therapist, a physiotherapist, an oedematherapist, a specialised nurse, and a social welfare officer are needed. Proper diagnosis, indication for treatment and follow up is time consuming. Often one hour is reserved for each scheduled visit to the team when arm volumes are measured, other clinimetrics performed, garments adjusted or renewed, social circumstances assessed, and other matters of concern are discussed. The patient is also encouraged to contact the team whenever any unexpected problems arise, so that these can be tackled without delay. In retrospect, a working group such as this one seems to be a prerequisite both for thorough preoperative consideration and informing patients, and for successful maintenance of immediate postoperative improvements. The team also monitors the long-term outcome, and the authors' experience so far indicates that a visit once a year is necessary, in most cases, to maintain a good functional and cosmetic result after complete reduction.

A centre of excellence combining all diagnostic and therapeutic modalities, including full non operative and operatives means, has the advantage of offering all patients a treatment; particularly when a non-operative treatment program is indicated prior to an operative treatment such as CSAL. By organising an expert centre with satellite treatment centres throughout the country, a dedicated lymphoedema network can be created in which medical help can be offered regionally when possible, and complex care and in patient treatment are centralised when necessary. Self-management and awareness programs, secondary and tertiary preventive measurements and an early diagnostic program are included in this concept.

This concept of lymphological care fits perfectly in the concept of chronic care, disease management[69] and in the in the ICF model of the World Health Organization (WHO). The expert Centre for Lympho-vascular Medicine in the Netherlands is organised according to these principles.

When is CSAL indicated?

In patients with late-stage lymphoedema with irreversible changes, non-operative treatment will not provide an appropriate reduction of volume. Unfortunately, neither conservative treatment nor microsurgical procedures can remove excess adipose tissue[70-73]. Subcutaneous tissue debulking seems the only option to reduce the limb volume and lead to an improvement in the patient's quality of life. As patient treated by conservative treatment also need a life garment, this issue is the same for both groups.

A surgical approach, with the intention of removing the hypertrophied adipose tissue, seems logical when conservative treatment has not achieved satisfactory oedema reduction and the patient has subjective discomfort of a heavy arm. This condition is especially seen in chronic, large arm lymphoedema around one litre in volume, or when the volume ratio (oedematous arm/healthy arm) = 1.3.

At the Department of Plastic and Reconstructive Surgery, Skåne University Hospital, Malmö, Sweden, the first liposuction of an arm lymphoedema was undertaken in 1987, but it was not until 1993 that a more detailed treatment protocol was established and a lymphoedema unit with a team was founded. The aim was arm lymphoedema after breast cancer treatment, as this is a large and common problem. There is no upper age limit to be accepted for surgery, but active tumour disease and ulcerations are contraindications. In 1994, the Expert Centre for Lympho-vascular Medicine, Nij Smellinghe Hospital, Drachten (NL) started a multidisciplinary working group for the diagnosis and non-operative treatment of lymphoedema. There is an inpatient and an outpatient clinic and patients are treated throughout

the country. In 2002, we began undertaking surgery for end-stage lymphoedema. This procedure is fully integrated into a multidisciplinary programme. In 2003, the Expert Centre started to cooperate with the Centre in Malmö and began CSAL for arms and legs. The same protocol is used with minor adjustments made in accordance with the local Dutch situation. The centre is organised according the insights of the chronic care model with emphasis on prevention of secondary lymphoedema, awareness, self-management and a focus on cooperation between all healthcare workers. On the other hand, many patients are referred for intensive diagnostics in patients with swelling or suspicion of (primary) lymphoedema. All treatment options such as conservative, operative, in and out patients clinic are applied. Long lasting follow up and guidance are an indissoluble part of the therapeutic approach. In 2012, a first roll out is starting to establish satellite treatment centers, all connected by one electronic e-health system. (see chapter 3)

Initially, lymphoedema is staged according to International Society on Lymphology (ISL)[74]:

- **Stage 1:** It starts as a swelling that shows pits on pressure: If treated immediately by conservative regimens, the swelling can disappear.
- **Stage 2:** If not, or improperly treated, the swelling increases in time and can end up in an even larger pitting oedema with concomitant adipose tissue formation
- **Stage 3:** Irreversible lymphoedema and even elephantiasis

In stages 2 and 3 in particular, there is sometimes a substantial oedema component, at times due to concomitant disease, inferior exercise or movement limitations of the limb.

The first and most important goal is to mobilise the pitting oedema and to achieve maximal result by conservative regimens such as DLT. 'Pitting' means that a depression is formed after pressure on the oedematous tissue by the fingertip, resulting in lymph being squeezed into the surroundings *(Figure 8a)*. To standardise the pitting test, one presses as hard as possible with the thumb on the region to be investigated for one minute, the amount of depression being estimated in millimeters. A swelling, which is dominated by hypertrophied adipose tissue, shows little or no pitting *(Figure 8b)*.

Another clinical feature is Stemmer's sign implies which that you can pinch the skin at the base of the toes or fingers with difficulty, or not at all. This is due to increased fibrosis and is characteristic of chronic lymphoedema. On the other hand, a negative sign does not exclude lymphedoema[75].

When a patient has been treated conservatively and shows no pitting but still there is a volume difference, CSAL can be performed. If quality of life is low, this can be especially effective. The cancer itself is a worry, but the swollen and heavy arm introduces an additional handicap for the patient from a physical, psychosocial and psychological point of view. Physical problems include pain, limited limb movement and physical mobility and problems with clothing, thus interfering with everyday activities. Also, the heavy and swollen arm is impractical and cosmetically unappealing, all of which contribute to emotional distress.

Contra indications for CSAL

CSAL should never be performed in a patient who is not maximal conservatively treated. Clinically, features show pits on pressure *(Figure 8a and b)*. In a patient with an arm lymphoedema, the authors accept around 4–5mm of pitting, and in a leg lymphoedema 6–7mm. Patients with more pitting should be treated conservatively until the pitting has been reduced. The reason for not doing liposuction in a pitting oedema is that liposuction is a method to remove fat, not fluid, even if theoretically it could remove all the accumulated fluid in a pitting lymphoedema without excess adipose tissue formation.

Other contra indications for CSAL are:

- metastatic disease or open wounds
- medical or family history of coagulation disorders or intake of drugs that affect coagulation
- physically not fit for surgery
- patient reluctant to wear compression garments continuously after surgery

Lymph transport system and liposuction

All surgery can lead to postoperative swelling due to tissue trauma and damage to the lymph and vascular systems. This swelling, depending on the type of surgery performed (ankle fractures take three to six months before the swelling disappears, free flaps tend to regenerate quickly; after a rhinoplasty, swelling can persist for more than one year, and, naturally, minor surgery, for example, after excising a mole, leads to no swelling at all), usually disappears within a few weeks when the lymphatics regenerate[76]. The same goes for liposuction performed for cosmetic reasons. In patients with lymphoedema, the lymph transport is greatly reduced. To investigate the effect of liposuction on lymph transport, an investigation using indirect lymphoscintigraphy was performed in 20 patients with post-mastectomy arm lymphoedema. Scintigraphies were performed before liposuction, with and without wearing a garment. This was repeated after 3 and 12 months. In conclusion, it was found that the already decreased lymph transport was not further reduced after liposuction[13].

Benefits to the patient

Liposuction improves patients' quality of life; particularly qualities associated with everyday activities, hence those that can be directly related to the complete 100% arm oedema reduction[15,25].

Skin blood flow after liposuction is increased and reduces the incidence of erysipelas; the annual incidence (bouts/year) of cellulitis was 0.4 before liposuction and 0.1 after, that is, a reduction of 75%[77]. Improved local skin blood flow may be an important contributing factor to the reduced episodes of arm infection. The point of bacterial entry may be a minor injury to the oedematous skin, and impaired skin blood flow may respond inadequately to counteract impending infection. Reducing the excess volume by liposuction increases skin blood flow in the arm, and decreases the reservoir of adipose tissue, which may enhance bacterial overgrowth.

Potential negative effects to the patient

Liposuction typically leads to numbness in the skin, which disappears within three to six months. Continuous, that is, lifelong wearing of compression garments is a prerequisite of maintaining the effect of any lymphoedema treatment and should not be considered as a negative effect.

Conclusion

Circumferential suction assisted lipectomy (CSAL) combined with permanent compression therapy is a proven effective treatment. The technique can be a potent therapeutic modality within an integrated, multidisciplinary lymphological care programme. Accumulated lymph should be initially removed using the well-documented conservative regimens until minimal or no pitting is seen. If there is still a significant excess volume, this can be removed by the use of liposuction. In some patients increased fibrous tissue can be present, especially in male patients and in women with a male distribution of body fat. When seen, fibrous tissue is more common in leg than in arm lymphedema, and more common in men than in women. Continuous wearing of a compression garment prevents recurrence.

Key points

- Excess arm or leg volume without pitting implies that excess adipose tissue is present
- Excess adipose tissue can be removed by the use of liposuction by CSAL. Conservative treatment and microsurgical reconstructions cannot remove adipose tissue
- As in conservative treatment, the lifelong use (24 hours a day) of custom made, flat knitted compression garments is mandatory for maintaining the effect of surgery
- Patients that are happy with an excess volume in the arm or leg are not candidates for liposuction
- CSAL is a potent therapeutical modality for special indications in persistent primary and secondary lymphoedema.The treatment is embedded within a multidisciplinary team and centralised in an expert centre

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CHAPTER 5
New developments in microsurgery

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Introduction

Microsurgery, in which surgery is performed with the aid of an operating microscope, was born in the early 1920s[1-2]. Successful microsurgical anastomosis of small vessels around 1.5mm was reported in 1960[3]. The use of microsurgical technique to treat lymphoedema by creating a bypass between lymphatic vessel and vein was first indicated in 1962[4]. The first clinical case reported about lymphatic venous anastomosis was in lower limb lymphoedema patient in 1969[5]. Since then, numerous microsurgical procedures have been attempted to treat lymphoedema over the past half century. To date, surgery has been regarded as a secondary option in treating lymphoedema patients who have failed conservative management with decongestive lymphatic therapy (DLT) and compression garments. Surgical procedures treating lymphoedema can be classified into two categories: ablative operations and physiologic operations. Microsurgery technique has been used for physiologic operations in order to drain excess tissue fluid trapped in lymphoedematous areas into other lymphatic basin or the venous circulation.

New developments in microsurgery

Lymphaticovenular bypass

Supermicrosurgery is defined as the anastomosis of smaller calibre vessels less than 0.8mm in diameter[6]. Lymphaticovenular bypass is a type of lymphovenous bypass, in which a supermicrosurgical technique is used to anastomose subdermal lymphatic vessels and adjacent venules performed through multiple small incisions using a surgical microscope[7,8]. A rationale for this approach is that, because the pressure in subdermal venules is lower than that in the deep, larger veins, there is less venous backflow, resulting in more permanent improvement of lymphoedema.

Indocyanine green (ICG) fluorescent lymphography

One of challenges of lymphaticovenular bypass is identifying functional lymphatic vessels. Recently, an indocyanine green (ICG) fluorescent lymphography was developed for visualising the lymphatic vessels[9,10]. ICG is a Federal Drug Agency (FDA) approved water-soluble compound, and it has been used for assessing cardiac output, hepatic function, and ophthalmic angiography for decades. When ICG is bound to protein in the tissue, it emits near-infrared ray. When the injected ICG is caught and streamed by the lymphatic vessels, ICG fluorescent lymphography system enables it to detect lymphatic vessels up to 2cm in depth from the skin surface.

ICG fluorescent lymphography can visualise the lymphatic vessels in the subcutaneous tissue not only before, but also during surgery, allowing surgeons to locate a functional lymphatic vessel for the lymphaticovenular bypass prior to making a skin incision. This saves substantial operating time and may contribute to improved outcomes of the operation.

Vascularised lymph node transfer

Free vascularised tissue transfer is a standard operative technique in plastic surgery in which an autologous tissue graft is harvested from a distant donor site and transplanted to the target area with its blood supply preserved by anastomosing artery and vein in the graft to vessels at the recipient site. Recent articles described transplanting composite soft tissue including lymph nodes to the lymphoedematous limb using microvascular technique[11]. Microvascular lymph node transfer is expected to result in new lymphatic vessels sprouting from the transplanted lymph node to drain the region. However, the rationale is theoretical and there have been no definitive data showing that lymphatic

vessels actually regenerate from transferred nodes and work as a lymphatic pump.

Timing of the microsurgery

Anecdotal experiences suggest that microsurgery to treat lymphoedema is more effective in the early stage before occurrence of fibrosis in the soft tissue[12]. In fact, prophylactic lymphovenous shunt operations were proposed in order to address the problem of lymphatic vessel deterioration before the manifestation of the lymphedema[13,14]. Although this concept may be feasible for retaining function in lymphatic vessels, prophylactic surgery is controversial because the occurrence of lymphoedema is unpredictable.

Conclusion

There have been several variations of microsurgical treatment for lymphoedema, including the use of healthy lymphatic vessels or a vein to interpose between peripheral and proximal lymphatic vessels in order to make a detour of the degenerated areas[15-19]. Recent refinements in microsurgical technique, instruments and improved imaging devices have allowed continued progress in microsurgical treatment for lymphedema. However, more work is needed to accurately evaluate lymphoedema, to properly define indications for surgery, and to refine the surgical procedures.

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Please note that many of the images portrayed in this document are extreme cases of lymphoedema and lymphatic filiarisis primarily seen in patients from developing countries.

The ILF Objective :

To improve the management of lymphoedema and related disorders worldwide

- To increase **awareness** by raising the profile of lymphoedema.
- To increase **knowledge** about lymphoedema by initiating and/or contributing to **Research Programmes.**
- To **disseminate** this knowledge by implementing an **international, not-for-profit, publications strategy.**
- To increase **understanding** of lymphoedema and its management by creating and/or contributing to the development **of Education Programmes.**
- To provide a cross cultural networking platform through an **Annual International Event** where all stakeholders will have the opportunity to contribute and influence the ILF agenda.
- To promote and document Best Practice with the development of an **International Minimum Dataset.**
- To facilitate and/or contribute to better **access to treatment** for patients worldwide.
- To promote and **support initiatives** whose goals are to improve the national/regional/local management of lymphoedema anywhere in the world.
- To help the Healthcare Industry understand the **real needs** of patients and practitioners, and develop and evaluate improved diagnostic tools and treatments.