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CONTROLLED COMPRESSION AND LIPOSUCTION TREATMENT FOR LOWER EXTREMITY LYMPHEDEMA

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ABSTRACT

In 1987 we noticed excess adipose tissue in a patient with arm lymphedema and later, objective studies confirmed this clinical finding in patients with non-pitting arm lymphedema following breast cancer. A prospective study was begun in 1993, and its long-term results (15 years) shows overall complete reduction of the excess volume in patients with non-pitting arm lymphedema and that adipose tissue dominates the excess volume. Encouraged by these results we operated on a patient with primary and secondary elephantiasis of the leg. The edema was first transferred from a pitting to a non-pitting state by controlled compression therapy. Then liposuction was performed to remove the remaining excess adipose tissue, and complete reduction was finally achieved. The patient wears compression garments continuously and during the 11 years of followup, no recurrence has occurred. This paper explains our philosophical approach: a pitting lymphedema first should be treated conservatively to remove excess fluid, then liposuction can be performed to remove remaining excess volume bothersome to the patient.

Keywords: adipose tissue, cancer, liposuction, lymphedema

Lymphedema is a condition affecting approximately 140 million people worldwide (1). Secondary lymphedema is most commonly due to the parasite Wuchereria bancrofti and in Western countries as a result of cancer treatment. Between 38 and 89% of breast cancer patients suffer from lymphedema due to axillary lymph node dissection and/or radiation. This variation can be understood by differences in definitions. measurement methods and timing, and treatment (2,3). Unilateral lymphedema occurs in up to 41% of patients after treatment of gynecologic cancer (4). A 5-66% incidence of lymphedema has been reported in patients treated for prostate cancer with incidence depending on whether staging or radical removal of lymph glands was done in addition to radiotherapy (5,6).

Primary lymphedema is caused by a defect in the lymph vessels or lymph glands and is localized to the lower extremities in most cases. Three forms can be distinguished: aplasia, hypoplasia, and hyperplasia of the lymph vessels. Aplasia, total lack of lymph vessels, is not compatible with life, and is only seen locally at the capillary level. Hypoplasia implies that the number and diameter of lymph vessels is reduced. Hyperplasia means that the vessels are wide, lymphangiectatic, and tortuous. Primary edema can present anytime from birth up to middle age, despite the pre-existence of deficient lymph vessels since birth. The congenital primary edema termed Milrov's disease is found more frequently in some families and is characterized in a subpopulation of patients by aplasia of

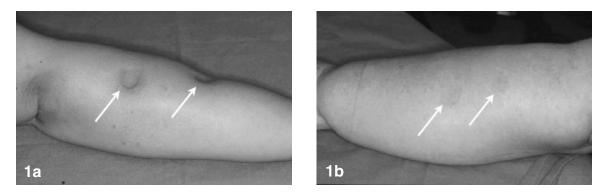


Fig. 1a. Marked lymphedema of the arm after breast cancer treatment, showing pitting several centimeters in depth (grade I edema). The arm swelling is dominated by the presence of fluid, i.e., the accumulation of lymph. b. Pronounced arm lymphedema after breast cancer treatment. There is no pitting despite hard pressure applied by the thumb for a one-minute period. A slight reddening is seen at the two spots where pressure has been exerted. The "edema" is completely dominated by adipose tissue. The term "edema" is improper at this stage since the swelling is dominated by hypertrophied adipose tissue and not by lymph, and at this stage, aspirate contains either no lymph or only a minimal amount.

small lymphatics secondary to a mutation in *VEGFR-3. Lymphedema praecox* presents before the age of 35 by definition, usually in women at puberty, and is a milder form of lymphedema constituting 80% of cases, whereas *lymphedema tardum* occurs after the age of 35.

In 1987, when the first patient with post-mastectomy arm lymphedema underwent liposuction at our hospital, we clinically noted an excess of adipose tissue in the aspirate. In 1993, we began a prospective study that showed this method, combined with controlled compression therapy (CCT), to be an effective treatment without recurrence (7-11). Most of the patients had no pitting before surgery because they had undergone conservative therapy [compression pumping, CPT (combined physiotherapy), or both] and were wearing compression garments, but they still had a considerable lymphedema in the arm.

"Pitting" is the formation of a depression after pressure is placed on the edematous tissue by the fingertip, resulting from lymph being squeezed into the surroundings (*Fig. 1a*). To standardize the pitting-test, one presses as hard as possible with the thumb for a period of 60 seconds on the region to be investigated; the amount of depression is then estimated in millimeters. Limb swelling dominated by hypertrophied adipose tissue and/or fibrosis shows little or no pitting (*Fig. 1b*) (10).

Controlled Compression Therapy

Liposuction for cosmetic purposes typically leads to edema, which resolves over time. We therefore anticipated that our patients, who were operated on for lymphedema, would have even greater edema following surgery. We decided that postoperative compression would be necessary and developed CCT as an alternative method since we did not have the capacity to use CPT. Typically CPT takes 1-2 hours a day for 2-4 weeks. On the other hand, CCT is a simple conservative method where the compression garment's size is reduced by regularly taking it in, using a sewing machine, as the swelling decreases (7.8). Such adjustments can often be made by the patients themselves. When 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 method, garments are renewed three

or four times during the first year. Two sets of compression garments should always be at the patients' disposal, one being worn while the other is being washed. Thus, a garment is worn permanently and treatment is interrupted only briefly when showering and, possibly, for formal social occasions. The patient is also informed about the importance of hygienic measures and skin care.

Adipose Tissue Hypertrophy

Objective clinical findings from aspirate analysis during liposuction and volume rendered CT-images show that there is a substantial increase, 81%, in adipose tissue content in non-pitting lymphedema of the arm (12-14). There are several possible explanations for adipose tissue hypertrophy. One explanation is that there is a physiological imbalance of blood flow and lymphatic drainage, resulting in impaired clearance of lipids and their uptake by macrophages (15,16). There is increasing support, however, for the view that the fat cell is not simply a container for fat but is an endocrine organ and a cytokine-activated cell (17,18), with chronic inflammation playing a role (19-22).

Fat and Lymph

A common misconception among clinicians is that chronic lymphedema is the result of an accumulation solely of fluid, i.e., lymph (12,13,23,24). The aim then is to remove the fluid by non-invasive conservative regimens such as combined forms of physiotherapy, involving manual lymph drainage, skin care, remedial exercises, and compression bandaging, complemented by the wearing of compression garments (25). CPT works in patients with lymphedema where pitting dominates the swollen limbs in early lymphedema and in substandard conservatively treated patients where the excess volume consists of fluid and fat, thus showing pits on pressure (pitting edema) (Fig. 1a and b).

Earlier Treatment Standards

Presently, there is no cure for lymphedema. Previous surgical regimes, utilizing bridging procedures, debulking, or total excision with skin grafting, seldom achieved acceptable cosmetic and functional results. Moreover, these techniques require multiple stages, are extremely challenging, and are associated with frequent complications. Microsurgical reconstruction involving lympho-venous shunts or transplantation of lymph vessels has also been investigated (26,27). Although attractive in concept, the common failure of microsurgery and physiotherapy to provide complete reduction is due to the persistence of newly formed subcutaneous adipose tissue that cannot be removed in patients with chronic non-pitting lymphedema.

Staged subcutaneous excision, with its concomitant complications, was previously the standard technique used in our clinic for patients with leg lymphedema. After positive outcomes with our arm lymphedema patients, we wanted to reduce the morbidity of this procedure by using the less invasive liposuction technique on a patient with severe leg lymphedema who failed conservative management.

Liposuction

During recent years, liposuction technique has been refined by use of vibrating cannulas which are finer and more effective than previous equipment. Also, the novel introduction of the tourniquet and tumescent technique has lead to minimized blood loss (28). Following surgery, new tissue volume is maintained through constant (24-hour) use of compression garments. On our long-term followup of patients with arm lymphedema (11 years), no recurrence of swelling was observed (11).

The indication for liposuction is a nonpitting swelling of the extremity and a lack of volume reduction by conservative regimens.

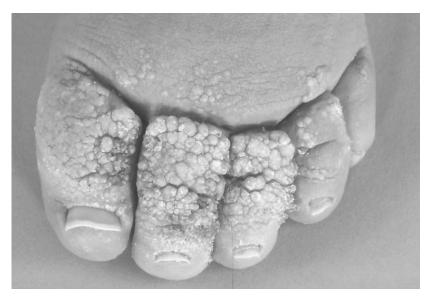


Fig. 2. Pronounced warty overgrowths on the left foot.

The first attempt to perform liposuction of a leg lymphedema in our clinic was in 1993 on a patient with unilateral primary lymphedema consisting of 4,650 ml in excess volume. She had swelling for 20 years prior to surgery and was seen once a year postoperatively until she died of myeloma 12 years later. At that time, excess volume of 1,905 ml was most likely due to the extensive fibrosis noticed at the time of surgery.

The next operation of this kind in our clinic was in 1994. The patient had secondary lymphedema following treatment for cervical cancer in 1958 including hysterectomy, bilateral oophorectomy, and removal of lymph nodes. Her lymphedema appeared in 1985 and before surgery, excess volume was 1,200 ml. Complete reduction was achieved 6 months postoperatively and is still maintained at last follow-up 13 years later. The reduction was measured as -725 ml, meaning that the previously lymphedematous leg was somewhat smaller than the normal leg.

Our next attempt, described in this paper, was in 1998 after having gained more experience treating arm lymphedema patients. This report also presents our philosophy on managing patients with lymphedema in general.

METHODS AND RESULTS

Patient

The male patient, born in 1968, was operated on as an infant due to congenital chylothorax in the left pleural cavity. In 1982, his left leg began to swell, and in 1984 the right one. He was treated conservatively with only elastic bandages to some effect. In 1992, an orchidectomy was performed due to a seminoma, and postoperative radiotherapy was given. Following this treatment, a rapid progression of the edema was seen in the left leg. Conservative treatment with CPT, including compression pumping, was started, but was discontinued after 6 months because it had no effect according to the patient. He was then referred to the Lymphedema Unit in our department in May 1996, at the age of 27. His left leg displayed elephantiasis, a pronounced lymphedema. The excess volume (volume of swollen leg measured by pletysmography -volume of contralateral)

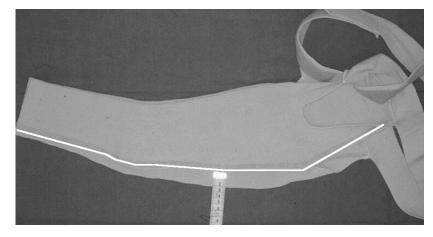


Fig. 3. Two made-to-measure compression garments laid on top of each other. The lower one has been used for 24 hours while the upper one has been washed. Note the shrinkage of several centimeters, marked by the white line.

was 14,310 ml, and there were numerous verrucae on the foot, especially dorsally on the toes (*Fig. 2*) (29).

Clinically there was pronounced pitting in the left leg with a depression of several centimeters. There was also some pitting in the lower right leg. He was socially stigmatized and could walk only with great difficulty and with pronounced heaviness and pain. At the first appointment, he said that he would like to be able to buy a pair of jeans without needing to add extra pieces of cloth to wear them.

Compression Therapy

Volume and circumference measurements were taken for two custom-made compression garments (Jobst® Elvarex BSN medical, Sweden, compression class 3, 39 mm Hg). After 2 weeks, when the compression garments were ready, CCT was started in an attempt to remove accumulated lymph and associated pitting. One garment was put on the swollen leg and worn for 24 hours, and then was removed to be washed. After taking a shower and lubricating the leg, the other garment was put on. This was repeated every day; one garment was always worn when the other was washed and dried. Washing of the garment removes salt accumulated from perspiration that could cause irritation. It also makes the garment shrink several centimeters, thus adding extra compression (*Fig. 3*).

Following 2 weeks of compression, excess volume was reduced to 11,045 ml (reduction: 3,265 ml = 23%). Garments were then taken in using an ordinary sewing-machine and measurements were taken for new ones (Elvarex, compression grade 3 Forte, 43 mm Hg). This was repeated for 3 months at which time the excess volume was reduced to 6,625 ml (reduction: 7,685 ml = 54%). Gradually tighter measurements were taken for new garments at 6, 9, and 12 months, with the sewing-machine being used in between when needed. After 12 months, excess volume was 4,190 ml (reduction: 10,120 ml = 71%) (*Figs. 4 and 5*).

At 12 months, compression was increased again (Elvarex, compression grade 4, 57 mm Hg), but at 15 and 18 months, the condition was essentially stationary, and new garments were ordered based on latest measurements. This steady state was due to the fact that all possible fluid (i.e., lymph) had been removed. The swollen leg showed no pits now upon pressure. The residual "edema" consisted of hypertrophied adipose tissue and fibrosis,

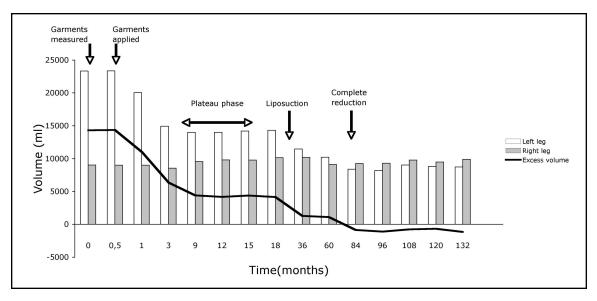


Fig. 4. The pre- and postoperative course depicting the rapid decrease in excess volume following controlled compression therapy (CCT). Note the plateau phase where pitting is no longer present. After liposuction and continuous CCT, complete reduction was achieved 4 years after surgery and has lasted 9 years after surgery, i.e., 11 years after initial treatment.

which was later confirmed during the operation. At 2 years, excess volume was 3,570 ml (reduction: 10,740 ml = 75%). This decrease was accompanied by a slight increase of the pitting edema of the right leg (*Figs. 4 and 5*).

Liposuction and Postoperative Success

The patient desired further reduction and underwent liposuction in April 1998 after 2 years of CCT. With the procedure, 3,530 ml of fat was removed from the left leg, and there were clinical signs of fibrosis. At that same time, CCT was begun on the right lower leg (knee high Elvarex, compression class 3). For the left leg, compression class 3 was used initially after surgery and later, compression class 4. The postoperative excess volume after 2 weeks was 1,535 ml (reduction: 12,775 ml = 89%).

The postoperative course is shown in *Figs. 5 and 6.* Gradually smaller sizes were obtained for the left leg by taking very firm measurements and resulting in step by step

reduction from the initial measurements. These firm measurements would be less than typical circumference measurement for volume calculations. Easy-Slide® (Arion international BV, Geleen, The Netherlands) was introduced 3 years after surgery. The effect was to increase the longevity of the garments and, as a result, the compression. Four years after liposuction, complete reduction was finally achieved without any combined physiotherapy. For the first time in many years, the patient could wear normal trousers and lead an active life. He requires a total of 12 full-length (on the left) and 4 knee-high (on the right) compression garments per year in order to maintain complete reduction. Figs. 4 and 5 show the pre- and postoperative course and Fig. 7 depicts the pre- and postoperative MRI.

DISCUSSION

Since Illouz described liposuction in 1980 (30,31), liposuction has become the most common procedure in plastic surgery (mainly

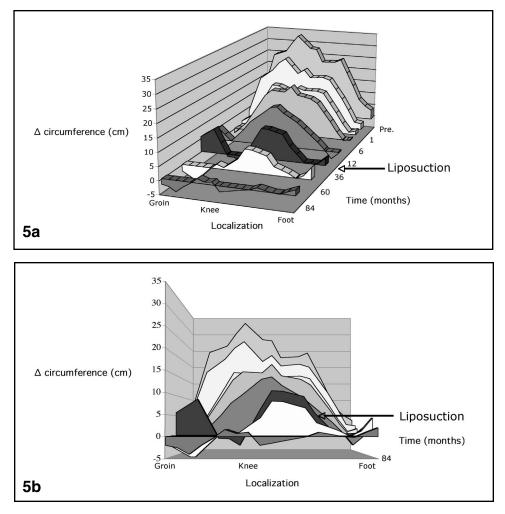


Fig. 5. 3-D graph showing the pre- and postoperative course based on the circumference difference between the lymphedematous leg and the normal leg. Note the rapid decrease in excess circumference following CCT. After liposuction and continuous CCT, complete reduction was achieved 4 years after surgery and has lasted 9 years after surgery, i.e., 11 years after initial treatment. a) oblique view, b) frontal view.

for cosmetic purposes). To a lesser extent, it has been used for reconstructive surgery (28). Complete reduction of arm lymphedema has been achieved after removing the excess adipose tissue using liposuction (7,8). There has been no sign of recurrence during 11 years of follow-up when combined with the use of custom fitted compression garments (11).

Recent studies have focused on the role of adipose hyperplasia and accumulation in the pathology of lymphedema, with chronic inflammation as a likely contributing factor (12-14,17-22). The beneficial effect of liposuction is secondary to the removal of hypertrophied adipose tissue (7-11), and it does not further diminish the already decreased lymph transport capacity (32). Increased blood flow and removal of adipose tissue could explain the reduced incidence of erysipelas seen in these patients (33). Currently, there are only five reports in the literature describing liposuction of lymphedematous legs.



Fig. 6. (a) Before treatment with CCT: excess volume 14,310 ml. (b) After 2 years of CCT and before liposuction: excess volume 3,570 ml (75% reduction). (c) Complete reduction was achieved 4 years after surgery: excess volume -865 ml, i.e., the treated leg was somewhat smaller than contralateral (106%). (d) He can now wear jeans, which was impossible before treatment.



Fig. 7. (a,c) Magnetic resonance (MR) images before treatment: note the excess amount of adipose tissue and fluid (b,d) MR images four years after liposuction and CCT: complete reduction occurred. Note the normalization of the subcutaneous tissue. (a and b: transverse views, b and d: frontal views).

In 1989, Sando and Nahai (34) treated 10 patients with leg lymphedema, five of them with liposuction and five with combined subcutaneous resection and liposuction. An 8% reduction was reported in the group that underwent only liposuction. They stated that, due to the poor results, liposuction should mainly be used in arm lymphedema and if used for leg lymphedema, should be combined with excision. That same year O'Brien et al (35) achieved a volume reduction of 23% in five patients with a mean follow up of 10 months. They did not use tumescent solution, only tourniquet, and only half of the limb was operated on. Also in 1989, Louton and Terranova (36) published a case report where skin excision was used after liposuction. Just like O'Brien et al (35) and Sando and Nahai (34), they did not use tumescence, power assisted cannulas, or circumferential suctioning. They advocated liposuction as a means to reduce flap loss after skin excision.

In 2006, Greene et al (37) described circumferential, power-assisted tumescent liposuction of bilateral lower leg lymphedema in a patient with spina bifida, paraplegia, hydrocephalus, and a ventriculoperitoneal shunt. Based on the preoperative volume in each extremity, they achieved a 75% reduction in each leg after removing around 1,000 g of adipose tissue from below the knee. When dealing with bilateral lymphedema, the percent reduction is based on each extremity, while in unilateral swelling the percent reduction is based on the normal leg. This means that 100% reduction cannot be obtained in bilateral lymphedema because it would indicate that the leg disappeared, whereas in a unilateral swelling 100% means complete reduction (i.e., both legs are normal in volume). Liposuction of leg lymphedema was also highlighted in a 2006 paper from Cao et al (38). In their study, ultrasonic liposuction was used. Unfortunately, the authors did not publish any data about volume reduction.

In contrast to previous reports, we were able to completely remove all excess fluid and fat from the lymphedematous leg of our patient. This was achieved by the use of aggressive circumferential liposuction and without the use of power-assisted cannulas, which were not readily available in 1998. The constant use of compression garments postoperatively plays a major role in maintaining the result. We found that in order to keep the volume stable, the patient needed 2 compression garments (Elvarex CCL 4) every 2 months for the operated leg. One garment was worn while the other was washed, so that every morning a clean garment was put on the leg after showering

and lubricating the skin. Every 2 months, 2 new garments were used, resulting in a total of 12 garments needed a year. On the other leg, knee-high garments were used (Elcarex CCL 3) in a total of 4 a year, thus alternating with 2 garments for 6 months. The use of an Easy-Slide® (Arion international BV, Geleen, The Netherlands) facilitated the use of garments and increased their longevity.

An argument against aggressive circumferential liposuction is that the procedure could interfere with the blood vascular microcirculation of the skin as well the lymphatics. However, our previous studies have shown that blood flow to the lymphedematous skin is increased after liposuction (33) and so far no skin necrosis has been seen after liposuction of arm lymphedema (7-10). In addition, thorough studies using indirect lymphoscintigraphy have not found any diminution of already decreased lymph transport after liposuction in patients with arm lymphedema (32). On the other hand, performing liposuction with the cannula parallel to the extremity has been found experimentally to cause trauma to the lymphatic microcirculation in patients without lymphedema (39,40).

The combination of preoperative CCT and liposuction followed by postoperative CCT led to a successful outcome in this patient. Eleven years after the treatment was started, no recurrence of the limb swelling has been observed, most likely due to continuous use and replacement of compression garments. The constant use of compression garments is a prerequisite for maintaining size reduction. He now works full time as a mechanic, which was impossible before treatment. Because of his improvement, we now treat patients with primary and secondary leg lymphedema and are awaiting long-term results. No bouts of cellulitis have occurred during the follow-up.

This less invasive technique combining liposuction and CCT offers patients with leg lymphedema an alternative treatment to traditional surgical techniques. There should be no tension between those who favor conservative treatment and proponents of liposuction. Accumulated lymph should be removed using the well-documented conservative regimens until minimal or no pitting is seen. If the patient is still discomforted by significant excess volume at that point, it can be removed by the use of liposuction.

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