Microcannular tumescent liposuction in advanced lipedema and Dercum’s disease

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Aim. We evaluated the outcome and risks of tumescence liposuction in patients with advanced lipedema or Dercum’s disease.

Methods. Six patients treated at a single center during the years 2004 to 2008 have been included. All patients were female and obese (body mass index 34 to 41.9; mean 38.2±3.8) with an age range from 29 to 78 years (mean 55.7±20.5 years), five of them had co-morbidities.

Results. The total amount of liposuckates varied between 1 500 mL and 4 800 mL. Pain could be reduced in all four patients with Dercum’s disease. Large adipose tissue removing implies a better outcome for pain. Patient’s satisfaction was “high” or “very high” in 5 and “medium” in one. The most common adverse effect was met-hemoglobinemia (N=4).

Conclusion. Tumescence liposuction is a treatment option for lipedema and Dercum’s disease. With careful monitoring the procedure is safe even for patients in advanced stages, higher age and with co-morbidities.

Key words: Anesthesia - Lipectomy - Adiposis dolorosa.

In contrast to lymphedema lipedema is less well known and possibly much rarer. Its painful variant is also known as Dercum’s disease. Notably, the lower extremity swelling is always bilateral and symmetrical in nature and never involves the feet. Skin changes characteristic of lipedema are absent, and consistent fat pads are present anterior to the lateral malleoli in each patient. These findings are representative of lipedema, which is distinct from lymphedema and for which treatment may be different. Lipedema is a gender-related disease occurring almost exclusively in women.1, 2

The cause of lipedema remains obscure. The initial idea that the adipose tissue will cause oedema by increasing the mechanical pressure on vessels has not been substantiated by science.3 Indeed it could be shown that at least in the earlier stages of the disease lymphatic vessels transport has an even higher volume and are functional intact.4, 5 By fluorescence microlymphography multiple microlymphatic aneurysms of lymphatic capillaries were a consistent finding in the affected skin regions of patients with lipedema. Microaneurysm of a lymphatic capillary was defined as a segment exceeding at least twice the minimal individual diameter of the lymphatic vessel.6

In later stages of lipedema lower limb lymphoscintigraphy showed lymphatic insufficiency in lipedema without morphologic abnormality as seen in lymphedema.7 It has been hypothesized that an

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increased capillary permeability is responsible for the leg lipedema.\textsuperscript{8,9} This may explain the tendency to develop haematomas after mild injuries but it does not explain why the feet remain uninvolved in oedema formation. The thickened skin fold on toes known as Stemmer's sign in lymph oedema does not exist in lipedema.\textsuperscript{2}

Swelling, pain and functional impairment in lipedema have negative effects on the quality of life. Most patients report a long history of unusual diets, drug treatments and physical training programmes – all without any beneficial but sometimes harmful consequences. This can lead to psychological problems and diminished social acceptance. This frustrating experience may lead to secondary obesity.\textsuperscript{10}

The disease can be classified according to severity.\textsuperscript{2} Stage I shows a smooth skin surface, but subcutaneous tissue is thickened and gives an uneven feeling when palpating. Stage II shows nodules in the adipose tissue leading to an uneven skin surface. Stage III is characterized by large nodules and deforming fat lobes. The diagnosis of lipedema is dependent on history, inspection and palpation.\textsuperscript{11}

In the past the only treatment available for lipedema was complex decongestive physiotherapy (CDP) as in lymphedema. The principle developed by Földi consists of manual lymph drainage, mobilization and compression bandaging. This increases the transport capacity of lymphatic collectors.\textsuperscript{12} In patients with lipedema CDP can lead to a decrease of leg diameter between 4% and 12%.\textsuperscript{13} Such a treatment needs to be performed life long. It has no effect of the adipose tissue, however.

Lipomatosis dolorosa (Dercum's disease) is a rare disorder resulting in painful fatty deposits around the upper legs, trunk, and upper arms. It belongs to the spectrum of lipedema but shows peculiarities such as pain and even abdominal involvement. There are no clear pathological mechanisms known, although it is suspected that there is either a metabolic or autoimmune component involved.\textsuperscript{14} Severe hyperalgesia is found on light pressure and touch. Analgesics or pain-modulating drugs usually have little or no effect. Surgical excision or liposuction of adipose tissue has shown significant reduction of pain.\textsuperscript{15-18}

Surgical procedures like liposuction have not gained wider acceptance since the functional and aesthetic outcome was uncertain and often not satisfying.\textsuperscript{12} By invention of tumescence anaesthesia a great success in surgery of lipedema was made possible.\textsuperscript{19} The wet technique provides a much better preservation of delicate structures such as blood and lymphatic vessels. Longitudinal liposuction along the course of lymphatics produced significantly less injury when compared with vertical suction (P < 0.05). In addition, tumescent liposuction is unlikely to cause major lesions of epifascial lymph vessels during suction procedures vertical to the extremity axis. Therefore, in this respect, this technique is superior to dry suction.\textsuperscript{20}

Prilocaine in contrast to lidocaine does not show vasodilatation and its resorption is more delayed compared to lidocaine. The recommended maximum dosage of prilocaine is 35 mg/kg body weight. Maximum plasma levels are seen after 5 to 6 h. Previous studies investigating large-volume liposuction procedures found maximum prilocaine levels and met-haemoglobinemia up to 12 h postoperatively, suggesting that liposuction should be performed as a hospital procedure only. This might be different for healthy patients undergoing liposuction involving less than 2 000 mL.\textsuperscript{21}

The other major step forward was the introduction of microcannulas in tumescent liposuction.\textsuperscript{22} Some authors recommend the use of vibration, water jet-assisted or ultrasound liposuction to secure lymphatic vessels.\textsuperscript{2,23}

We here present a series of patients with advanced lipedema and Dercum's disease who have been treated by tumescent liposuction and CPT.

### Materials and methods

#### Subjects

These are non-selected patients that had been treated at our department for lipedema of the upper legs. All patients had a number of pretreatments including repeated manual lymph drainage (N = 6), compression garments (N = 6), CPT (N = 5) or liposurgery (N = 1). Diagnosis was lipomatosis dolorosa Dercum (N = 4) or lipedema (N = 2).

Before treatment all patients had at least two appointments for clinical examination, discussion of the procedures and care after surgery. Photos were taken and a laboratory examination was performed (basics included blood count, met-hemoglobin, and clotting analysis). All patients got an individualized compression garment (trousers) and had been advised to wear the compression at least for six months. Patients were
Table I—Patients demographics:

<table>
<thead>
<tr>
<th>Patient</th>
<th>Age (yrs)</th>
<th>Type of lipedema</th>
<th>Grade</th>
<th>BMI</th>
<th>Comorbidities</th>
<th>Follow-up (months)</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>74</td>
<td>Lipomatosi dolorosa</td>
<td>III</td>
<td>40</td>
<td>Diabetes mellitus type 2, arterial hypertension, sick sinus syndrome (pace maker), osteoarthritis, glaucoma, varicosis, depression</td>
<td>39</td>
</tr>
<tr>
<td>2</td>
<td>78</td>
<td>Lipomatosi dolorosa</td>
<td>III</td>
<td>41.9</td>
<td>Arterial hypertension, iatrogenic hypothyroidism, varicosis</td>
<td>48</td>
</tr>
<tr>
<td>3</td>
<td>68</td>
<td>Lipomatosi dolorosa</td>
<td>III</td>
<td>40.5</td>
<td>Arterial hypertension, hyperuricemia, atopic eczema, varicosis</td>
<td>6</td>
</tr>
<tr>
<td>4</td>
<td>37</td>
<td>Lipomatosi dolorosa</td>
<td>III</td>
<td>35</td>
<td>Varicosis</td>
<td>24</td>
</tr>
<tr>
<td>5</td>
<td>29</td>
<td>Lipedema</td>
<td>II</td>
<td>34</td>
<td>Hypothyroidism</td>
<td>6</td>
</tr>
<tr>
<td>6</td>
<td>48</td>
<td>Lipedema</td>
<td>II</td>
<td>37.5</td>
<td>No comorbidities</td>
<td>48</td>
</tr>
</tbody>
</table>

informed that with increasing age skin laxity after liposuction might occur and surgery could be necessary to improve the aesthetic outcome.

The severity of lipedema was classified according to Meier-Vollrath and Schneller: grade II (N=2), grade III (N=4). All patients were scored obese according to their body mass index (BMI).

Preoperative exclusion criteria were history of deep venous thrombosis, other thrombotic events or embolism, active phlebitis, cancer and connective tissue disease.

Liposuction was performed in hospitalized patients (N=5), mainly because of comorbidities. Patient N. 6 was treated on an out-patient basis. The co-morbidities and demographic factors are shown in Table I. Patients with a genetic defect of the enzyme glucose-6-phosphatase have significant risk to develop severe methemoglobinemia due to prilocaine used for tumescence. None of our patients had this genetic defect.

Tumescence anesthesia

Patients were placed in a lying position. We preferred prilocaine for tumescence anesthesia since it shows less cardiotoxicity than lidocaine. Tumescence anesthesia was performed with 0.1% prilocarpine solution (1 mL of 1:1000 adrenaline – 50 mL of prilocarpine – 1000mL of Ringer solution) per patient using an infusion pump with a speed of 100 mL per minute.24 The maximum volume of tumescence solution was 5 L. During surgery pulse oximetry was used for monitoring.

Liposuction

All patients got a single shot of intravenous ascorbic acid (1 g) before surgery to reduce the risk of methemoglobin formation. Preoperative antibiotics was only given for the first patient only because she had a pace maker (2 g amoxicillin). Liposuction was performed in a longitudinal direction with blunt cannulas with a diameter between 2 to 5 mm depending on the anatomical situation. After removal of deeper parts of the adipose tissue with the larger cannulas, we switched to smaller ones for finer sculpturing. Suction was realized with the Mentor Contour Genesis device (Mentor Corp., Santa Barbara, CA, USA). During liposuction, a manual pressure was applied to the superficially placed cannula from outside. Incisions were adapted by polyamide sutures (3-0 Ethilon, Ethicon) but not completely closed. There was no need for drainage. Patients received compression bandages for 24 to 48 hours, then replaced by individualized compression garments (trousers). Histology was performed on a regular basis from the tissue collected by the suction machine.

Liposuction was performed in series from one to four sessions to avoid the need larger volumes of tumescence solution. After surgery blood counts and met-hemoglobin was monitored in all patients until normalization. Met-hemoglobinemia was treated with 1 g intravenous ascorbic acid once to three times a day until normalization. Adverse effects were record.
Table II.—Liposuction and outcome.

<table>
<thead>
<tr>
<th>Patient</th>
<th>N. of sessions</th>
<th>Volume of tumescent solution (L)</th>
<th>Total volume of removed adipose tissue (L)</th>
<th>Patient’s satisfaction</th>
<th>Pain reduction (Visual analogue scale)</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>1</td>
<td>4</td>
<td>1500</td>
<td>Medium</td>
<td>95±70</td>
</tr>
<tr>
<td>2</td>
<td>2</td>
<td>3 to 4</td>
<td>2550</td>
<td>High</td>
<td>90±45</td>
</tr>
<tr>
<td>3</td>
<td>4</td>
<td>3 to 4</td>
<td>4800</td>
<td>Very high</td>
<td>75±20</td>
</tr>
<tr>
<td>4</td>
<td>2</td>
<td>3 or 4</td>
<td>2600</td>
<td>Very high</td>
<td>75±21</td>
</tr>
<tr>
<td>5</td>
<td>3</td>
<td>2.6 to 4</td>
<td>3000</td>
<td>Very high</td>
<td>Not done</td>
</tr>
<tr>
<td>6</td>
<td>1</td>
<td>5</td>
<td>1800</td>
<td>High</td>
<td>Not done</td>
</tr>
</tbody>
</table>

Follow-up

Patients were asked for a regular follow-up at least during the first six months after their last liposuction. Patient’s global assessment was used to evaluate satisfaction with the outcome. Satisfaction was scored from unsatisfied, unchanged, medium, high to very high. Furthermore, the pain was assessed in patients with lipomatosi dolorosa using the visual analogue pain scale (0=no pain; 100=maximum pain).

Results

Outcome

Liposuction itself was well tolerated by all patients. We were able to remove between 500 mL and 1 800 mL adipose tissue per session. The total amount of adipose removed was 1 500 mL to 4 800 mL. There was no adverse event in the operation theatre (Table II). In patient N. 1 further procedures would have brought a better outcome but the risk for further surgical procedures was scored too high because of her cardiac problems.

Histology revealed large lobules of adipose tissue. Although the BMI did not change significantly compression trousers had to be adopted to lower leg volume during sequential procedures (Figure 1).

Hospitalization was useful to monitor laboratory changes and ensure frequent sterile wound dressing changes in particular in those patients of older age and with multiple comorbidities. In five patients who were hospitalized the stay in hospital was three to five days per session (average 3.5 days).

Patient’s satisfaction was “medium” in patient N. 1 and “high” or better in the other five patients. Pain was reduced in three of four patients with lipomatosi dolorosa and remained unchanged in patient N. 1 (Table II). The follow-up of patients was 6 to 48 months. No relapse or partial relapse was seen during this time. In three patients there was further slight improvement after six months (Figures 1-6).

The follow-up period starting from the last procedure performed in the individual patient was between 6 and 48 months. No single case of secondary lymphedema after tumescent liposuction was observed.

Adverse effects

In the post-surgery monitoring phase, five out of six patients showed a significant increase of their met-hemoglobin levels. The kinetics of that was highly variable with a peak reached with 24 to 72 hours. Among those patients who had at least two session of liposuction the amount of met-haemoglobin formation varied remarkably. There was no clear relationship to the amount of tumesence solution either.

Four patients experienced a temporary leukocytosis within 24 h with a single patient who developed a raised body temperature. Treatment was initiated in patient N. 1 only since she was a patient with high risks (pace maker etc.). None of the patients had significant decrease in haemoglobin and hematokrit levels during the procedure (Table III).

Skin laxity was observed in patient N. 2 and 3. Surgery to further improve esthetic outcome is planned in patient 3. It is obvious that in older patients with advanced lipedema skin laxity cannot completely be prevented by compression garments.

Discussion

In this study we used conventional tumescent liposuction with blunt microcannulas to treat patients with advanced (grade II-III) lipedema of the legs. Significant amounts of adipose tissue could be removed in 2-
4 sessions. Large-volume liposuction, i.e., liposuction rates in excess of five liters were not done since there is an increased risk of fat embolism in lungs and kidneys. Patients were satisfied with the results although retraction of tissue was limited in patients above 65 years of age. Although esthetics is not unimportant in medical procedures the major goal for the patients in this study was improved functionality together with decreased pain.

In a study by Schmeller and Meier-Vollrath (2006) 28 patients, who had undergone conservative therapy
over a period of years, were treated by liposuction under tumescent local anesthesia with vibrating microcannulas. Twenty-one could be re-evaluated after an average of 12.2 (1-26) months. All showed great improvement, with normalization of body proportions. Additionally, spontaneous pain, sensitivity to pressure, and bruising either disappeared completely or improved markedly. Other than minor swelling for a few days, no complications could be observed following surgery. All patients reported a tremendous increase in their quality of life. Physical therapy had to be continued to a much lower degree. Among 19 patients followed-up for eight years after tumescent liposuction there was neither secondary lymphedema nor a progression of lipedema. Other studies supported these findings. We made a similar observation in our study (Table IV).

Liposuction has been used with success in patients with lipomatosis dolorosa as well. Since the disease is rare, mostly case reports have been published. In a large study with 53 patients with lipomatosis dolorosa and liposuction coagulation and fibrinolytic factors associated with cardiovascular atherosclerosis and glucose uptake were measured. There was a slight increase in levels of coagulation factors 2 and 4 weeks postoperatively, probably in reaction to the surgical trauma. In a sample of 10 patients, glucose uptake was improved (<0.05) from a short-term perspective after surgery. In conclusion, liposuction may facilitate glucose uptake and insulin sensitivity increases from a short-term perspective. Another study in 15 obese women without lipedema could not demonstrate any significant improvement in obesity-associated metabolic abnormalities.

Weather ultrasound-assisted liposuction (UAL) may have benefits over traditional suction-assisted liposuction has been a matter of debate. Vascular damage, blood loss and tissue damage in tumescence liposuc-
TABLE III.—Adverse effects after tumescent liposuction.

<table>
<thead>
<tr>
<th>Patient</th>
<th>Methb (%)</th>
<th>Leukocytes (10^9/L)</th>
<th>Other adverse effects</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Max</td>
<td>Rx</td>
<td>Rx</td>
</tr>
<tr>
<td>1</td>
<td>19.2</td>
<td>AS</td>
<td>16.4</td>
</tr>
<tr>
<td>2</td>
<td>14.3</td>
<td>AS</td>
<td>20</td>
</tr>
<tr>
<td>3</td>
<td>30.2</td>
<td>AS</td>
<td>25</td>
</tr>
<tr>
<td>4</td>
<td>1.0</td>
<td>—</td>
<td>—</td>
</tr>
<tr>
<td>5</td>
<td>29</td>
<td>AS</td>
<td>12.9</td>
</tr>
<tr>
<td>6</td>
<td>0.9</td>
<td>—</td>
<td>—</td>
</tr>
</tbody>
</table>

Normal values: Methemoglobin (Methb): 0.1 to 1.0%, leukocytes 3.8–11.6 x 10^9/L. Max: maximum Rx: treatment; AS: ascorbic acid.

TABLE IV.—Results of tumescent liposuction in lipedema and Dercum’s disease in published studies (only original paper with >5 patients, no abstracts).

<table>
<thead>
<tr>
<th>Reference</th>
<th>N</th>
<th>Age range (yrs)</th>
<th>Technique</th>
<th>Tumescence solution (L)</th>
<th>Liposaptors (L)</th>
<th>Outcome</th>
</tr>
</thead>
<tbody>
<tr>
<td>Bernstorp et al. (1996) 31</td>
<td>53</td>
<td>22-69</td>
<td>Dry technique* Microcanular</td>
<td>—</td>
<td>No data available</td>
<td>Significant weight reduction</td>
</tr>
<tr>
<td>Stutz and Krahl (2009) 23</td>
<td>30</td>
<td>21-63</td>
<td>Water jet-assisted</td>
<td>1.00-1.50</td>
<td>0.25-2.35</td>
<td>Minimal or no injury to lymphatic vessels</td>
</tr>
<tr>
<td>Schmelzer and Meiier-Vollrath (2006) 20</td>
<td>28</td>
<td>22-63</td>
<td>Power-assisted</td>
<td>2.74-13.5</td>
<td>1.06-5.50</td>
<td>Reduction in pain, sensitivity to pressure and bruising</td>
</tr>
<tr>
<td>Wollina et al. present study</td>
<td>6</td>
<td>29-78</td>
<td>Microcanular</td>
<td>3.00-5.00</td>
<td>1.50-4.80</td>
<td>Reduction in pain, high patient's satisfaction</td>
</tr>
</tbody>
</table>

*) Dry technique means without tumescence, in general, epidural or spinal anesthesia.

Function (lidocaine and epinephrine solution) was not significant different from UAL.23-30 Systematic investigations have been performed to determine whether there are differences in the intra- and postoperative course and the final cosmetic result between tumescent liposuction and UAL. The following parameter have been evaluated degree of postoperative bruising, nodularity, skin retraction, or decreased cellulite. There were no major complications attributable to either technique. These authors have not shown a significant difference in the postoperative course or the final cosmetic result when comparing tumescence liposuction with UAL.30 Although there is no doubt about that the physician will benefit from UAL what is less exhausting than conventional tumescence liposuction, the major benefits for the patient seem to be comparable.

Comparative trials with water jet-assisted liposuction have not been performed. In a single-centre trial this technique was used to treat 30 lipedema patients stage II–III, aged 21 to 63 years. Histocchemical analysis of the fat tissue aspirates demonstrated regular CD31 expression for blood vessel endothelium. Staining for lymphatic endothelium with D2–40, on the other hand was mostly absent.23 The authors argue by this finding that water jet-assisted liposuction seems to protect lymphatic vessels while removing mostly intact adipocytes with vascular components.

Four out of six patients in our series had varicose veins. In one session there was subcutaneous varicose vein bleeding. We stopped the procedure and used compression to stop the bleeding, what was successful. The role of phlebosurgery of “symptomatic” varicose veins in patients with lower extremity lymphedema or lipedema is controversial. A study was conducted in 90 patients with lower extremity lipedema who had undergone phlebosurgery for varicose veins. The outcome was disappointing since leg swelling worsened or was unchanged in greater than 90% whereas symptoms such as heaviness, fatigue, cramps were improved in less than 10%. These find-
ings support that surgery for varicose veins in the legs of patients with lipedema should be undertaken only if there is an absolute indication present (ascending phlebitis and/or bleeding). Otherwise, complete decongestive physiotherapy is still the best treatment approach for these groups of patients.36

Temporary adverse effects were met-hemoglobulinemia and leukocytosis. We preferred prilocaine for tumescence anesthesia since it shows less cardio-toxicity than lidocaine. On the other hand, prilocaine produces higher amounts of met-haemoglobin than lidocaine. This has to be monitored. A total of 242 published episodes of met-hemoglobinemia have been analyzed recently. A discrepancy between the pulse oximeter saturation (≥90%) and the arterial oxygen partial pressure (≥70 mmHg) was present in 91.8% of the episodes. The difference between oxygen saturation measured by pulse oximetry and co-oximetry varied from -6.2% to 44.7%.37 We observed the same, that pulse-oximetry was not very sensitive to select patients at risk for met-hemoglobinemia.

High concentrations of ascorbic acid are able to reduce the met-haemoglobin concentration in vitro and in vivo.37,38 Another antidote is methylene blue. Prilocaine should not be used in children younger than six-month-old, in pregnant women, or in patients taking other oxidizing drugs. In adults, the dose should be limited to 2.5 mg/kg.38 Patients with a genetic defect of the enzyme glucose-6-phosphatase have significant risk to develop severe met-hemoglobulinemia. In such patients prilocaine is contraindicated.23

Whereas the formation of met-haemoglobin is related to tumescence anesthesia the cause of leukocytosis is less obvious. Recently, it was shown that fat tissue produces and releases inflammatory cytokines, and that obesity may be regarded as a state of low-grade inflammation. In a study with 327 patients who were referred for further investigation because of leukocytosis BMI was shown to be associated with leukocytosis (P<0.0005). Obesity is recognized as a possible cause for reactive leukocytosis.39-41 During liposuction, when adipose tissue is damaged, these proinflammatory factors may result in transient leukocytosis.

In advanced stages of lipedema and older age of patients with comorbidities postsurgical monitoring has to be performed carefully. Temporary, delayed met-hemoglobulinemia and leukocytosis are the most common side effects in this group. In conclusion, tumescent liposuction has proved to be a safe and effective treatment for lipedema not only patients under 50 years of life although esthetic outcome is better in less advanced stages and younger patients.

**Riassunto**

Lipoaspirazione a microcanne tumescenti nel lipedema in studio avanzato e nella malattia di Dercum.

OBIETTIVO. Sono stati valutati i risultati e i rischi associati alla lipoaspirazione a tumescenza in pazienti affetti da lipedema in studio avanzato o da malattia di Dercum.

METODI. Sono stati inclusi nello studio sei pazienti trattati presso un singolo centro nel periodo di tempo compreso tra gli anni 2004 e 2008. Tutti i soggetti erano di sesso femminile, obesi (indice di massa corporea compreso tra 34 e 41,9; media 38,2±3,8) con un range di età compreso tra 29 e 78 anni (media 55,7±20,5 anni), cinque delle quali affette da co-morbilità.

RISULTATI. La quantità complessiva di lipoaspirato variava tra 1 500 ml e 4 800 ml. Il dolore è stato alleviato in tutte e quattro le pazienti affette dalla malattia di Dercum. La quantità di tessuto adiposo rimosso è risultata direttamente correlata al risultato in termini di alleviamento del dolore. La soddisfazione della paziente è stata "elevata" o "molto elevata" in 5 e "media" in una di loro. Il più frequente evento avverso è stato la met-hemoglobulinemia (N=4).

CONCLUSIONI. La lipoaspirazione a tumescenza rappresenta un'opzione di trattamento per il lipedema e la malattia di Dercum. Con un attento monitoraggio del paziente, la procedura è sicura anche in pazienti affetti da patologia in studio avanzato, anziani e con co-morbilità.


**References**


