

## Supplemental surgical treatment to end stage (stage VI-V) of chronic lymphedema

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**Aim.** When the lymphedema reaches to its end stages, the complex decongestive therapy (CDT) and/or compression therapy become less effective and increased risk of systemic/general sepsis to become potentially life threatening condition.

**Methods.** To improve its clinical management, excisional surgery was performed on 22 patients for their 33 limbs in the end stage of lymphedema as a supplemental therapy, and its efficacy was retrospectively analyzed. Diagnosis was made by radionuclide lymphoscintigraphy and basic laboratory studies (e.g. ultrasonography, magnetic resonance imaging). Twenty-two patients (mean age: 46 years; 3 male and 19 female; 5 primary and 17 secondary) submitted to the excisional surgery on 33 limbs (unilateral: 11; bilateral: 22). Surgery was indicated by further progression of the disease with recurrent sepsis despite adequate antibiotics therapy. A modified Auchincloss-Homan's operation was used to excise grotesquely disfigured soft tissue with advanced dermato-lipo-fibrosclerotic change. The normal limb contour was re-established to allow proper postoperative therapy. Postoperative CDT and compression therapy were mandatorily implemented in all cases.

**Results.** A postoperative assessment of the treatment results, at 12 months showed an overall improvement in 28 of the 33 limbs: substantial improvement on the condition of limb function and quality of life (QOL), and local and/or systemic sepsis. Eighteen patients with good compliance to maintain the postoperative CDT showed much improved clinical results and QOL through the first interim assessment (24 months).

**Conclusion.** Excisional surgery at the end stage of lymphedema seems to provide substantial improvement of clinical condition and QOL only when mandated postoperative CDT/compression therapy is well kept.

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Chronic lymphedema was once considered as relatively benign condition remaining in simple diffuse swelling of affected limb/region with minimum morbidity. But this old concept was found to be totally erroneous; this condition is rather clinically serious, steadily progressive condition not only affecting the lymphatic system itself, but also entire surrounding soft tissue, resulting in unique condition of dermato-lipo-fibrosclerosis.<sup>1</sup>

When the lymphedema reaches to its end stages (stage IV-V), it becomes disabling as well as distressing condition; lymphedema in this stage allows more frequent bacterial and fungal infection to already set in chronic inflammatory condition throughout skin and soft tissue.<sup>2, 3</sup>

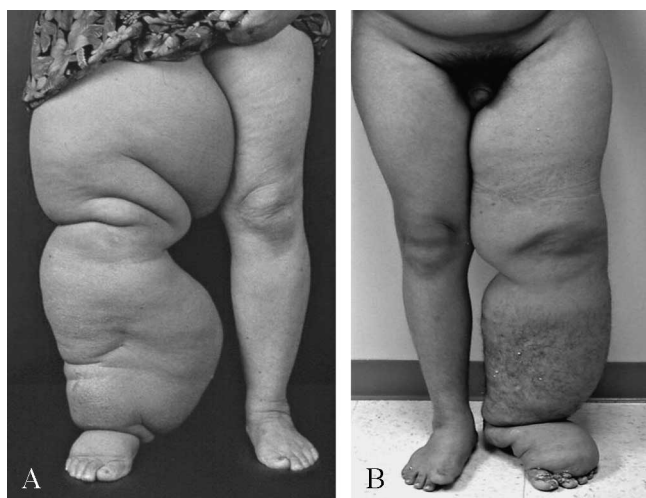


Figure 1.—Clinical appearance of lower extremities in the end stage of chronic lymphedema. Massively swollen and disfigured contours interfere appropriate application of CDT and/or compression therapy, resulting in a vicious cycle between progressive dermato-lipo-fibrosclerosis and recurrent local/systemic sepsis. CDT: complex decongestive therapy.

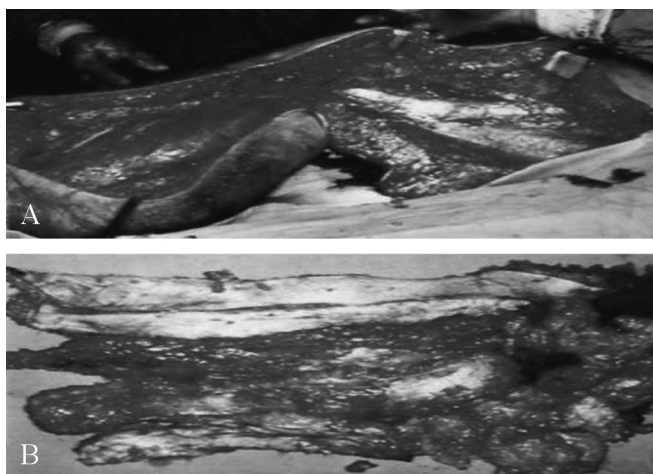


Figure 2.—Excisional surgery to the end stage of chronic lymphedema as supplemental therapy, modified Auchinloss-Homan's operation. A) Operative finding of medial approach (above-knee and below-knee) for the *en-bloc* resection of grotesquely disfigured fibroedematous tissue to restore the normal contour of the extremity for the proper CDP\* (postoperative) implication. B) Surgical specimen excised from the lower extremity; the specimen includes skin, subcutaneous tissue and underlying fascia, exposing a bare muscle to facilitate transmural absorption of lymphatic fluid. CDT: complex decongestive therapy.

Once the local/regional sepsis starts, the risk of systemic/general sepsis is also increased to become potentially life-threatening condition. This unique status is also known for the condition to predispose not only to an immunodeficiency and wasting phenomenon, but also new malignancy development: Kaposi sarcoma; lymphangiosarcoma.

The standard regimen of lymphedema management based on the complex decongestive therapy (CDT)<sup>4-6</sup> and/or compression therapy<sup>7-9</sup> becomes less effective; the compression bandage is more difficult to apply properly due to severely disfigured contour with severe deformity in general (Figure 1).

Therefore, we made revisit to this once-condemned excisional therapy<sup>3, 10, 11</sup> to re-evaluate its role with selective implementation only to the end stage of lymphedema.

## Materials and methods

Retrospective analysis was made on 22 patients for their 33 limbs in the end stage of lymphedema which underwent palliative excisional surgery to improve its management.

## Methods

Clinical evaluation with an initial detailed history taking and physical examination was followed by radionuclide lymphoscintigraphy<sup>12-14</sup> and basic laboratory studies (*e.g.* ultrasonography, magnetic resonance imaging [MRI], etc. for conclusive diagnosis<sup>15-18</sup> and proper clinical and laboratory staging,<sup>2</sup> as follows: volume measurement of swollen extremity using infrared optometric volumetry<sup>19</sup> and tape measurement of circumference of the limb at various levels along the foot, ankle, calf and thigh, venous duplex sonography, and radionuclide lymphoscintigraphy. Additional studies were further included as optional tests when indicated: air plethysmography (APG), computed tomography (CT) scan,<sup>20, 21</sup> standard MRI study.

Further studies were added to the primary lymphedema when indicated as a truncular form of lymphatic malformation,<sup>22-24</sup> mainly to rule out possibly combined other kinds of vascular malformation, especially the extratruncular forms of lymphatic malformation (*e.g.* bone involvement) and/or venous malformation.<sup>25-29</sup>

Pre- and postoperative evaluation included clinical improvement, subjectively (*e.g.* patient satisfaction index: quality of life [QOL]), as well as objectively (*e.g.* circumference measurements in centimeters, and infrared optical volume determination of limbs).<sup>2, 10</sup>

Following proper clinical and laboratory staging of the chronic lymphedema, all the patients underwent basic treatment protocol of CDT:<sup>5, 6</sup> compression bandage, therapeutic exercise regimen, compression garment, special skin care for the infection control and/or prevention, general supportive care of hygiene, nutritional counseling for diet and weight control/reduction.

Traditional compression therapy<sup>7, 8</sup> with pneumatic device was generally implemented as additional treatment when the lymphedema becomes resistant to CDT.

All the candidates were extensively reviewed preoperatively, based on their response to properly instituted treatment, as well as on the progress of their disease during the treatment over a minimum of one-year on the standard treatment regimen. The decision for additional surgical therapy was made by consensus among the multidisciplinary team for the failure of treatment to arre-

st progress of the disease, based on the results of a strict assessment.<sup>3, 10, 29</sup>

Among a total of 1 065 chronic lymphedema patients (primary: 259; secondary: 806; male [M]: 131; female [F]: 934; age range: 2 months to 82 years), 22 patients (mean age: 46 years; M=3 and F=19; primary: 5; secondary: 17) in the end stage of chronic lymphedema were selected to the palliative excisional surgery on 33 limbs (unilateral: 11; bilateral: 22).

All 33 limbs were indicated for the surgery by further progression of the disease with recurrent sepsis of more than three times per year, despite adequate antibiotic protection and to improve the efficacy of compression therapy.

A modification of Auchincloss-Homan's operation,<sup>30-33</sup> was used to excise a generous amount of grotesquely disfigured tissue with advanced dermato-lipo-fibrosclerotic change, including the whole skin layer, subcutaneous tissue and muscle fascia in order to re-establish the normal limb contour to allow proper postoperative compression therapy (Figure 2).

Inclusion criteria are the documented failure and/or technical difficulty to institute proper compression therapy due to a morbidly enlarged extremity and subsequent deterioration of limb function and QOL,<sup>2, 34, 35</sup> including increased local and/or systemic sepsis.

Postoperative CDT and compression therapy were mandatorily implemented in all cases of surgical group.

Follow-up assessments of surgical group were made at a postoperative 1, 2, 3, 6, 12, 18 and 24 months, with an additional evaluation of each episode of local and/or systemic sepsis. Evaluation for the surgical results was extended from 24 months to 48 months as the final assessment whenever possible.

A thorough periodical was made twice a year by a multidisciplinary care team; additional tests were added to the schedule for episodes of local and/or systemic sepsis with cellulitis.

In the control group, 14 limbs of 9 patients (5 bilateral and 4 unilateral) were excluded from the surgery; the exclusion criteria from the surgery were poor compliance (10/14), lack of proper support (5/14), an age of >70 (3/14), and the lack of frequent infection episodes (3/14). They underwent an identical regimen of CDT combined with compression therapy applied to surgical group.

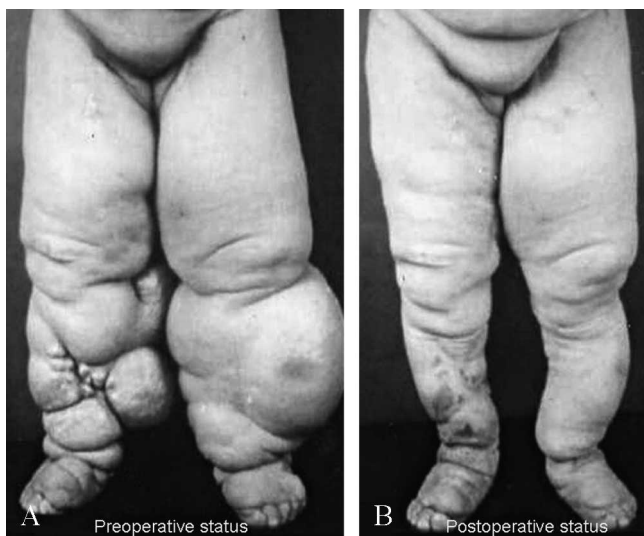


Figure 3.—Clinical outcome of excisional surgery implemented to the end stage of chronic lymphedema. A) Clinical appearance of the bilateral lower extremity lymphedema at its end stage before the resection of grotesquely disfigured fibro-edematous tissue. B) Fully restored normal contour of the extremity after the ablative surgery to improve the efficiency of CDT-based main therapy postoperatively. CDT: complex decongestive therapy.

Regular follow-up assessment for the control group was also made once a year for a minimum with additional evaluation when the sepsis should be bad enough to need a hospital care.

## Results

Among 33 limbs of 22 patients, 2 wound complications occurred: one with delayed healing with wound disruption and another with wound infection.

Initial postoperative assessment of the treatment results, 6 months and subsequently at 12 months after surgery, showed an overall improvement in 28 out of the 33 limbs, and 5 failed (Figure 3).

### *Good initial response group (N.=28)*

Twelve showed excellent response with more improvements from their immediate postoperative measurement /conditions, with an additional 10-15% reduction of limb volume from the immediate postoperative volume.

Sixteen of these 28 limbs also showed good response with satisfactory maintenance of the immediate postoperative volume reduction with

a minimum change of 5-10% up to the 12 month assessment.

All 28 with excellent to good surgical outcome accompanied substantial improvement on the limb function in majority (25/28) and QOL (26/28). The initial responses by the sepsis to the surgery were favorable in majority although they were in a wide range: less frequent (n=18) and/or less severe (n=9), no recurrence of the local and/or systemic sepsis (n=4), no change (n=4), and further worsening (n=1) (12 months).

All 28 limbs with good results maintained excellent compression therapy postoperatively throughout 12-month assessment period.

However, among these 28 limbs with satisfactory responses at the 12-month assessment, only 18 with good compliance to continue compression therapy were able to maintain the results through the first interim assessment (24 months). These 18 limbs have shown much improved clinical results with good (12/18) to excellent (6/18) maintenance of the initial postoperative result and experienced less frequent and less severe, or no recurrence of the local and/or systemic sepsis (14/18), and a much improved QOL (16/18) (24 months).

However, the other 10 out of 28 that kept good outcome up until 12 months assessment failed to maintain proper compression therapy until the 24-month assessment and showed steady deterioration over a period of 6 months in average, when continuous postoperative compression therapy was stopped.

Among the 18 with good results on the first interim assessment (24 months), 16 were available on the final 48-month assessment (14 for full investigation and 2 for a partial one); only 6 with excellent maintenance of postoperative compression therapy were able to remain in initial good condition after the surgery, while 10 with poor compliance failed with further deterioration.

Among total of 27 with poor compliance, including 2 lost from the follow-up, 18 were available to the final assessment (48 months) and 9 were not reachable.

Eighteen available failed to maintain proper postoperative compression therapy, despite successful excision of redundant tissue to improve the physical therapy; their majority (14/18) continued to show steady deterioration of the local and systemic condition throughout the follow-up period (48 months). Another 9, dropped out from the regular

follow-up assessment through 48-month period, were also confirmed for the evidence of deterioration through frequent hospital cares for the episodes of cellulitis with/without sepsis.

#### *Poor initial response group (N.=5)*

Five among a total of 33 limbs, which failed to maintain postoperative compression therapy, were not able to maintain the initial post-excision results to 12-month follow-up. They were further deteriorated through the 24-month follow-up period: all 5 limbs regained 10% to 80% of volume reduction by excision/preoperative volume within 6 months to 1 year following the cessation of compression therapy.

The final assessment (48 months) of these 5 patients showed further deterioration in 3, continuously refusing to re-institute proper compression therapy. Other 2 with an initial moderate deterioration (over 40% regain of the initial volume reduction) at the first *interim* follow-up (24 months) had improved back to a limited regain of 20%; both were able to maintain re-instituted compression therapy and there were no disease progression until the final assessment (48 months).

#### *Control group (N.=14)*

Ten out of 14 limbs of the control (non-surgical) group, which were well maintained on the same compression therapy regimen equal to the excision group, uniformly failed to reach the same level that clinical improvement successful surgical group has made. The other 4 limbs, which failed to maintain compression therapy, deteriorated further, before reaching to the first interim follow-up assessment (24 months) and lost from the further follow-up.

A lymphoscintigraphic study, included whenever feasible for follow-up assessment in both control group and excisional surgery group, uniformly failed to provide positive findings of the improved lymphatic function that were compatible with clinical improvements through the final assessment (48 months).

## **Discussion**

Since the turn of the last century, various debulking operations were advocated for the sur-

gical management of chronic lymphedema by Charles (1912), Sistrunk (1918), Homans (1936), and Thompson (1962), etc.<sup>32, 33, 36-38</sup> These various excisional procedures, often known as reduction or debulking operations, remove scarred and disfigured lymphedematous tissue from the limb. These procedures, however, were condemned for many years because of general morbidity and significant complications in addition to its controversial effectiveness; their application had been non-selective and used for any type of lymphedema. Some of these once-abandoned operations have regained the interests lately for chronic end-stage lymphedema as alternative palliative operations, but the large numbers of long-term results have yet to be assessed.

We reassessed its new role to the end stage of lymphedema, and the failure to obtain satisfactory control of the lymphedematous process or to prevent disease progression despite vigorous non-surgical treatment was a major criterion for selection.<sup>3, 10, 39</sup>

The excisional surgical therapy in our experiences is still effective as an additional method to improve clinical status along the end (late stage) stage of lymphedema with no salvageable lymphatic system left.

But, we learned that continuous physical therapy following the excision is such critical to maintain initial surgical success; a good compliance to maintain postoperative physical therapy is the precondition for the long term success.

Such devastating long-term outcome of initially successful excisional surgery among our series was due to the poor compliance to fail to meet this critical condition; 6 limbs out of a total of 33 were able to maintain initial success through postoperative CDT during the follow-up period of 48 months.

We learned a few lessons through this excisional surgery revisit.

First, there is no room in this surgery for the patient with poor compliance who cannot commit to postoperative maintenance physical therapy. Every single case who abandoned continuous physical therapy during the follow-up period failed to maintain good initial outcome. Therefore, the selection of the candidate should be more stringent with this compliance issue.

Second, the surgery should be planned as a part of multidisciplinary care including social worker

and psychiatrist/psychologist for the appropriate postoperative maintenance care.

Excisional surgery can be used as the most effective mean to control the late stage of lymphedema only when remaining a part of multidisciplinary team management with strict criteria of indication/selection and full integration with CDT following the surgery.

Third, overall dividend by the 'revisit' to once-condemned excisional surgery is, however, still worthy for the minority (N.=6) who stuck to this principle; this procedure should be condemned only to the non-compliant patients group. It is still good to the patient with good compliance to commit with a life-time maintenance of the CDT.

Therefore, the compliance remains the key to the successful surgical management of the late/end stage of lymphedema.

However, the compliance is the most difficult issue to this chronic disease; postoperative maintenance care is totally depending on the self-motivation for a home-based care. This reality affects the compliance of the patient quite negatively.

Patients at its end stage were selected for excisional surgery purely for palliative purpose to improve the local condition and facilitate proper physical therapy. All 33 limbs of 22 patients we selected were unable to carry proper CDT-based therapy preoperatively, because of the technical difficulty to apply bandages for the exercise therapy due to a massively disfigured limb contour by fibro-edematous tissue.

When the disease progresses to the end stage, recurrent sepsis and further local deterioration start a vicious cycle and, thus, it becomes technically more difficult to provide proper CDT. Therefore, this palliative surgery was implemented to our patients only when the epifascial (superficial) lymphatic system had been hopelessly destroyed with no chance of recovery or improvement of function.

The surgical aims are to improve the performance of CDT and/or compression therapy following the excision of grotesquely disfigured fibrosclerotic tissue; it effectively reduces the amount of lymphedematous tissue and subsequently reduces the incidence of sepsis. Therefore, we set a life-time commitment to the full maintenance of the CDT regimen postoperatively as a mandatory precondition to this palliative surgical therapy.

Clinical evidence indicates increased lymph

absorption through the subfascial (deep) lymphatic system following the excision of fibrosclerotic subcutaneous tissue, which includes the entire muscle fascia; the removal of the tissue barrier seems to ensure to facilitate the absorption of tissue fluid by the deeper lymphatic system within the muscles. However, this has yet to be proven lymphoscintigraphically.

## Conclusions

Excisional palliative surgery appears to be effective at initially controlling the progress of lymphedema. However, based on our limited experience over follow-up periods up to 48 months, it is not capable of maintaining the initial success without the addition of postoperative CDT and compression therapy. Thus, this surgical therapy is effective only when fully integrated with CDT and/or compressotherapy.

The commitment of a patient to life-time CDT compliance, even after surgery, is mandatory in order to achieve satisfactory long-term surgical results.

It appears that excisional surgery is likely to remain an adjunctive therapy at most, but that it will retain its critical role in improving total care management of chronic lymphedema together when combined with CDT.

Excisional surgery at the end stage of lymphedema seems to provide substantial improvement of clinical condition and QOL only when mandated postoperative CDT/compression therapy is well kept.

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