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# Surgical management of genital lymphoedema: experience and critical considerations from a tri-center study

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## Abstract

**Aim:** Genital lymphoedema (GL) is a chronic and debilitating disease, which can severely affect the patient's quality of life with significant socio-economic impact. Nowadays, no gold standard algorithm exists for GL from diagnosis to treatment. This study proposes our therapeutic flowchart based on the three senior consultants' experience in lymphatic surgery.

**Methods:** A retrospective investigation was conducted on a prospectively maintained database (2018-2022). Inclusion criteria involved all patients who underwent surgical procedures for treating GL in three plastic surgery departments (Lausanne, Bari, and Genova). Outcomes were assessed in terms of oedema reduction, stage regression, and functional reported outcomes.



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**Results:** 16 patients with GL were included: 50% underwent debulking surgery, 18.8% microsurgery, and 31.2% debulking + microsurgery. We recorded a significant regression of the GL stage: 62.5% shifted from stage II/III to postoperative stage I. Similarly, we found an infection recurrency resolution in 50%, a scrotal oedema reduction in 62.5%, and a scrotal oedema resolution in 37.5% of the patients treated. While almost half of the patients (53.3%) with associated penile oedema described persistent postoperative penile oedema, only two patients complained of persistent lymphorrhea.

**Conclusion:** According to our clinical experience, preoperative and postoperative physical functional therapy is always recommended. For stages I and IIA, after the failure of the conservative treatment, lymph-venous shunts and lymph node transplantation surgery are proposed at the early time. When GL is already diagnosed at stages IIB and III, the debulking surgery, together with functional procedures, represents our first approach.

**Keywords:** Genital lymphoedema, debulking surgery, functional surgery, microsurgery, lymphatic venous anastomosis, multi lymphatic-venous anastomoses, lymph node transplantation

## INTRODUCTION

Genital lymphoedema (GL) localisation represents 0.6% of all lymphoedema cases on a global scale<sup>[1]</sup>. Genital lymphoedema is a chronic and evolutive disease caused by pathological fluid retention in scrotal, penis, and pubic (or vulvar) interstitial tissue as a result of imbalanced lymphatic drainage<sup>[2]</sup>. The lymph drainage reduction determines an accumulation of protein-rich interstitial fluid, which leads to further fluid retention, inducing a vicious circle of progressive fibrosis<sup>[3]</sup>.

GL can be classified by aetiology as primary or secondary. Primary (also named congenital) is often idiopathic or rarely related to genetic autosomal mutations<sup>[4]</sup>. Secondary lymphoedema is commonly related to surgical or radiotherapy cancer treatment, followed by infection causes (this aetiology is predominant in developing countries)<sup>[3]</sup>.

The diagnosis relies on the patient's history and physical examination, supported by radiological imaging, such as computed tomography, magnetic resonance imaging, and duplex ultrasonography.

Currently, the standard assessment to quantitatively evaluate the residual lymphatic function is mainly based on nuclear medicine techniques, particularly lymphoscintigraphy<sup>[5]</sup>.

From a clinical perspective, patients mainly reported pain, heaviness, skin tightness, and frequent infections in the genital area with compromised urinary and sexual functions in advanced stages, with an overall degraded quality of life<sup>[6]</sup>.

Nowadays, no specific and definitive treatment exists for GL. Early diagnosis and precocious therapy are highly recommended, particularly considering the impact on the patient quality of life and the chronic/ recurrent evolution of the disease<sup>[7]</sup>.

The gold standard care for lymphoedema is a multidisciplinary approach, which always starts with conservative treatments (complete decongestive therapy (CDT), compression bandaging, and exercises, manual lymph drainage, skin care, and treatment of the underlying conditions) followed by surgical interventions<sup>[8]</sup>.

Conservative therapy may produce satisfactory results, but these benefits are usually temporary without maintenance and continued compression<sup>[9]</sup>. In addition, compression bandaging and manual lymph drainage are shown to be less effective in the case of GL due to anatomical limitations<sup>[10]</sup>.

From a surgical perspective, the treatment options can be either reductive or functional<sup>[7]</sup>. Reductive intervention, on the one hand, involves radical excision of affected tissue (lymphangectomy), followed by reconstruction of the genital area by local flaps or skin grafting<sup>[11]</sup>.

Reductive surgery (also named debulking or ablative surgery in the text) aims to reduce pathologic adipose and fibrotic tissue excess and to stop lymphoedema progression. Debulking or excision techniques can involve partial or total resection of the skin and the subcutaneous tissue. Suction Assisted Lipectomy (SAL) is commonly performed for initial soft tissue accumulation, especially in the pubic area, while in the case of fibrosis, direct excision becomes necessary with more invasive procedures<sup>[12]</sup>. These can improve patient discomfort, hygiene, and quality of life<sup>[13]</sup>.

On the other hand, functional surgery (mentioned as physiologic surgery or derivative surgery along the text), such as lymphatic-venous anastomosis (LVA), multi lymphatic-venous anastomosis (MLVA) or lymph nodes transfer/transplantation (VLNT), aim to re-establish or improve the lymphatic drainage and require microsurgical expertise<sup>[13,14]</sup>.

Recent findings agree that physiologic treatments should be performed in the early stages of GL to preserve lymph drainage and avoid the progression to fibrosis. Besides that, surgical debulking is recommended in later stages, where lymphoedema has already progressed with significant fibrosis and adipose infiltration, resulting in disruption of the remaining lymphatic pathways<sup>[12,15,16]</sup>.

Despite increased interest in GL treatment in recent years, no univocal consensus exists in its approach from diagnosis to treatment choice and recurrency management. This study aims to define a therapeutic algorithm based on the experience of three senior consultants, all with extensive experience in lymphatic surgery and microsurgery.

Regarding the therapeutic strategy, here in the text, patients will be divided into two main categories as follows: (1) debulking/ablative surgery only (Ablative surgery); (2) microsurgery (Microsurgery) OR a combination of microsurgery and ablative surgery (Microsurgery + Ablative surgery).

## **MATERIAL AND METHODS**

A tri-center (Lausanne, Switzerland; Bari and Genova, Italy) retrospective investigation, based on prospectively maintained databases involving GL patients treated between January 2018 and January 2022, was performed in this study.

Patients with genital (scrotal and/or penile) lymphoedema who underwent surgical treatments were included in this study. Both primary and secondary aetiologies were considered, and eventual associated lower limb lymphoedema was not an exclusion criterion.

Moreover, all patients without a complete follow-up (at least 12 months after the last procedure) were excluded. Patient demographic data and comorbidities were gathered from medical and anaesthesiologic charts. Operative technique details, pre- and postoperative symptoms (subjective and objective), clinical

evaluation by the same consultant surgeon (in terms of pain, oedema, recurrent infections, lymphorrhoea/chylorrhoea), pre- and post-lymphoedema stage, preoperative lymphoscintigraphy records (in some cases a postoperative lymphoscintigraphy was performed too), transport index as well as postoperative complications and eventual further surgeries for each patient were evaluated and collected.

The International Society of Lymphology stage (ISL) was applied to classify the disease severity degree of the patients included in this study<sup>[17]</sup>.

The study was conducted accordingly to the guiding principles following the Declaration of Helsinki of 1975. Informed consent was obtained from all patients, including approval for scientific publication and photographic/video documentation.

## RESULTS

This retrospective study included 16 patients (all males) with a mean age of 54 years old and an average body mass index (BMI) of 29 kg/m<sup>2</sup>. The follow-up was on average 40 months (ranging from 24 to 132) [Table 1].

Regarding the aetiology of GL, primary and secondary were equally distributed: lymphatic malformation (primary, 9 patients), related to surgery (5), multifactorial/trauma (2) (secondary) [Figure 1A].

Most of the patients with GL had an associated lower limb (LL) lymphoedema (62.5%, 10 out of 16), distributed as follows: LL + penis + scrotum (56.3%), LL + scrotum (6.3%), penis + scrotum without LL counted for 6 patients (37.5%) [Figure 1B].

According to the ISL, our study included 6 patients with a preoperative stage II (37.5%), 6 preoperative stage III (37.5%), and 4 not specified.

All patients described painful oedema, while recurrent infections were associated in 87.5% of the cases; finally, active lymphorrhoea was described in 7 out of 16 patients (43.8%), always concomitant with pain, oedema, and recurrent infections [Figure 2].

The mean time between the first symptoms displayed and surgery was 11.5 years.

Regarding the type of intervention, 50% of the patients underwent excisional surgery only, 18.8% to microsurgery only (among these, one had a double physiologic treatment, consisting of MLVA first, followed by a VLNT later). Finally, a combination of ablative surgery and microsurgery was performed in 5 patients (31.2%).

All patients were treated with compressive bandages, associated with physical functional therapy (penile, scrotal and pubic lymph drainage) pre- and post-surgical procedures. Overall, anatomical lymphoedema localisation, type, and aetiology were homogeneously distributed between the three treatment groups. No significant difference in timing symptoms to surgery nor in lymphoedema stage between the three treatment groups was observed.

Our study showed a significant regression of the GL ISL stage: 10 patients (62.5%) shifted from stage II/III to a postoperative stage I, while 2 patients moved from stage III to a postoperative stage II.

**Table 1. Summarise of patient's demographic data, operative technique details, pre- and postoperative symptoms, stage, clinical evaluation, complications, and lymphoscintigraphic records**

Patient	Age	Sex	BMI	Type	Aetiology	Localisation	Duration (Y)	Stage (pre-op)	CTD (pre-op)	Symptoms pre-op	Surgical Treatment	CTD (post-op)	Outcomes	Symptoms (post-op)	Global Amelioration	Stage (post-op)	Secondary Surgical treatment	Symptoms post 2nd op	Follow-up (months)
1	50	M	29	Secondary	Multifactorial	Penis, scrotum	8	-	Y	Pain, oedema, penile shaft deformity, lymphorrhoea	MLVA	Y	Scrotal oedema resolution	Persistent penile deformity and penile oedema	Y	-	VLNT (omentum)	residual penile oedema (positive response to CT)	72
2	64	M	29.4	Secondary	Surgery	Penis, scrotum	10	-	Y	Oedema, recurrent infections	MLVA	Y	Oedema reduction	Infection (two erysipelas)	Y	-	-	-	36
3	45	M	28.6	Primary	Lymphatic malformation	Scrotum, lower limbs	11	2	Y	Oedema, scrotal chylorrhoea, lymphorrhoea, recurrent Infections	MLVA	Y	Scrotal oedema reduction	Persistent scrotal chylorrhoea, scrotal oedema	Y	2	-	-	36
4	59	M	29.5	Secondary	Multifactorial	Penis, scrotum	5	3	-	Oedema, pain, recurrent infections, penile shaft deformity	Reductive surgery	Y	Oedema reduction, infections recurrency resolution	Persistent penile deformity and penile oedema	Y	2	-	-	36
5	40	M	30.2	Primary	Idiopathic	Penis, scrotum, lower limbs	6	2	Y	Oedema, scrotal chylorrhoea, lymphorrhoea, recurrent Infections	Reductive surgery + MLVA	Y	Oedema reduction	-	Y	1	-	-	24
6	52	M	27.9	Secondary	Surgery	Penis, scrotum	5	-	Y	Oedema, lymphorrhoea, recurrent Infections	Reductive surgery + VLNT	Y	Oedema scrotal resolution	Persistent penile deformity and penile oedema	Y	-	-	-	24
7	47	M	28.8	Secondary	Surgery	Penis, scrotum	9	-	Y	Oedema, recurrent Infections	Reductive surgery + LVA	Y	Oedema scrotal resolution	-	Y	-	-	-	36
8	60	M	-	Primary	Lymphatic malformation	Penis, scrotum, lower limbs	8	3	Y	Oedema, recurrent infections	Reductive surgery	Y	Scrotal oedema reduction, infections recurrency resolution	Persistent penile oedema	Y	1	-	-	36

9	70	M	-	Secondary	Surgery	Penis, scrotum, lower limbs	4	3	Y	Oedema, recurrent infections	Reductive surgery	Y	Scrotal oedema reduction, infections recurrency resolution	Persistent penile oedema	Y	1	-	-	24
10	70	M	-	Primary	Lymphatic malformation	Penis, scrotum, lower limbs	4	2	Y	Pain, oedema, recurrent infections	Reductive surgery	Y	Scrotal oedema resolution	Persistent penile oedema	Y	1	-	-	24
11	70	M	-	Primary	Lymphatic malformation	Penis, scrotum, lower limbs	34	3	Y	Recurrent infections, pain, oedema	Reductive surgery	Y	Scrotal oedema resolution, lower limbs oedema reduction	Persistent penile oedema	Y	1	-	-	24
12	37	M	-	Primary	Lymphatic malformation	Penis, scrotum, lower limbs	5	2	Y	Pain, oedema	Reductive surgery	Y	Scrotal oedema resolution	Persistent penile oedema	Y	1	-	-	24
13	45	M	-	Secondary	Trauma	Penis, scrotum	15	3	Y	Pain, oedema, recurrent infections, scrotal verrucosis with lymphorrhea	Reductive surgery	Y	Scrotal oedema reduction, infection recurrency resolution	Lymphorrhea, Y persistent penile oedema	Y	1	-	-	24
14	56	M	-	Primary	Lymphatic malformation	Penis, scrotum, lower limbs	41	3	Y	Pain, recurrent infections, scrotal verrucosis with lymphorrhea	Reductive surgery + MLVA	Y	Scrotal, penile, oedema reduction, infection recurrency resolution	Lymphorrhea	Y	1	-	-	132
15	30	M	-	Primary	Lymphatic malformation	Penis, scrotum, lower limbs	15	2	Y	Pain, oedema, recurrent infections	Reductive surgery + MLVA	Y	Scrotal, penile, oedema reduction, infection recurrency resolution		Y	1	-	-	60
16	74	M	-	Secondary	Surgery	Penis, scrotum, lower limbs	4	2	Y	Pain, oedema, recurrent infections	Reductive surgery	Y	Scrotal, penile, oedema reduction, infection	-	Y	1	-	-	36

Mean	54.3	29.1	11.5	2.5	recurrency resolution	1.2	40.5
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Regarding the clinical reported outcomes, we noticed a significant improvement in objective and subjective symptoms complained by the patients. Particularly, infections recurrency resolution, and scrotal oedema reduction (or resolution) were observed in 93.8% and 46.7% of the patients, respectively.

Interestingly, penile oedema seemed more complex than scrotal oedema to resolve, (53.3% free of symptom), while surgical treatments were particularly effective on persistent lymphorrhea, which was reported postoperatively in two patients only (28.6%) [Figure 2].

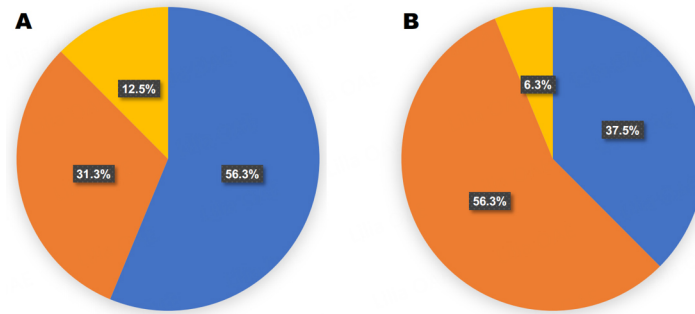
One patient (number 1) benefited from staged microsurgical procedures. The physiologic operation consisted of MLVA for active scrotal lymphorrhea and penoscrotal oedema. This resulted in a total cessation of lymphorrhea and a decrease in scrotal oedema. However, he presented after 3 years with a recurrence of scrotal-penile lymphoedema, requiring lymph node transfer by gastroepiploic vascularized lymph node transfer (GE-VLNT). Clinically, after this second intervention, the patient manifested a global amelioration with cessation of active lymphorrhea and residual penile oedema, responding to CDT [Figure 3]. A lymphoscintigraphic exam at a one-year follow-up after the second intervention showed a normalised bilateral lower limbs lymphatic drainage, and a stable scrotal dermal backflow with improved left lymphatic drainage.

## DISCUSSION

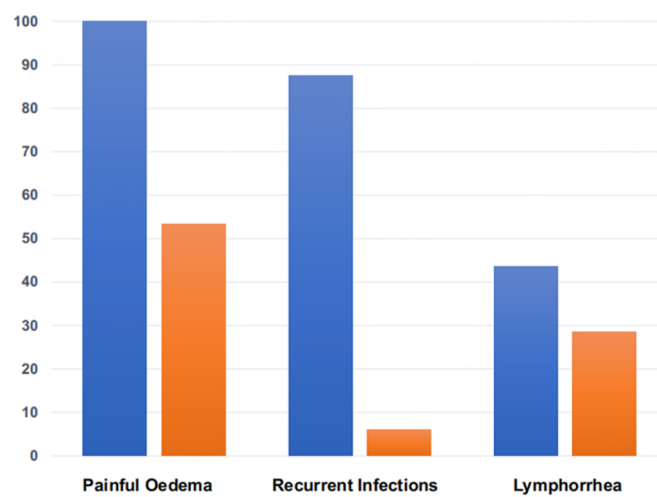
Genital lymphoedema (GL) is a chronic invalidating disease that causes a significant physical and psychological impact on patient quality of life<sup>[7]</sup>.

In patients with secondary GL, a specific external determinant (surgery, radiation, malignancy, infection, or trauma) explains the physiological lymphatic flow disruption (e.g., obstruction in the lymphatic system, lymph nodes, and/or lymphatic vessel removal or damage). By contrast, most primary GLs are caused by lymphatic malformations which arise during lymphangiogenesis<sup>[18]</sup>.

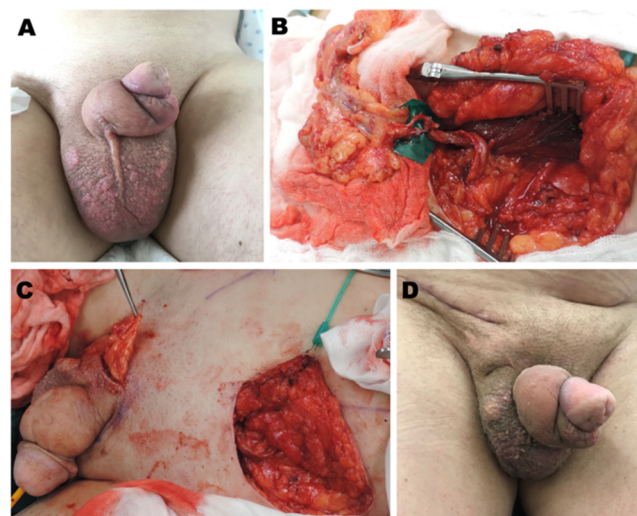
In spite of the aetiology, the classification of severity lymphoedema (ISL) is essential to choose the right treatment for GL and predicting its outcomes. Regarding the therapeutic options for GL, recent literature defines different strategies from conservative intensive physiotherapy to super microsurgery techniques, but no standardised protocols exist<sup>[7]</sup>. Finally, front-line research has recently proposed the application of stem cell therapy approaches to treat lymphoedema. Stem cells (mesenchymal stromal cells (MSC), bone marrow-derived MSC, and adipose-derived MSC) have a wide range of therapeutic effects in terms of anti-inflammation, antifibrosis, anti-oxidative stress, as well as promoting the regeneration of different tissues. These properties have been suggested as promoting factors for lymphatic vessel regeneration with interesting results in *in vitro* studies. However, at the moment, stem cell therapy has no approved clinical indication in lymphoedema treatment and multiple pre-clinical *in vitro* and *in vivo* studies are ongoing<sup>[19]</sup>.



**Figure 1.** Aetiology (A) of genital lymphoedema. lymphatic malformation (blue), surgery (orange), multifactorial/trauma (yellow); anatomical localisation (B) of genital lymphoedema. penis + scrotum + lower limbs (orange), penis + scrotum (blue), scrotum + lower limbs (yellow).



**Figure 2.** Preoperative (orange) and postoperative (blue) symptoms of genital lymphoedema.



**Figure 3.** Clinical results before (A) and after (D) the VLNT (B microsurgical anastomosis of omental vessels to inferior epigastric artery and vein; (C)inset of the omentum flap in the crural zone) to cease the scrotal lymphorrhoea and reduce the penile lymphoedema (patient number one).



While ablative surgeries aim to remove tissue excess and close the residual defect, microsurgery has the purpose to re-establish lymphatic drainage, bypassing the blockage (lymphatic venous anastomosis (LVA), multiple lymphatic-venous anastomosis (MLVA), or stimulating lymphaticogenesis [vascularised lymph nodes transfer (VLNT)]<sup>[20]</sup>.

These microsurgical techniques became more popular in the last decade with promising outcomes, particularly for treating lower limb lymphoedema. However, no defined decision management has been established yet for GL<sup>[21-24]</sup>.

In our experience, surgical indication for GL generally includes an insufficient volume reduction and the ineffectiveness of conservative methods, recurrent episodes of lymphangitis/erysipelas, no responsive pain, heaviness or discomfort to CDT, and finally urinary and sexual dysfunction<sup>[18,20]</sup>.

The quantification of GL is predominately based on preoperative lymphoscintigraphy, especially for assessing the transport index (TI) according to Kleinhaus<sup>[25]</sup>, while for the GL staging, we usually adopt the ISL classification.

According to the ISL, lymphoedema stage is determined not only by the volume (partially influenced by the patient's BMI), but also by the quality/changes of the skin/soft tissue, the level of fibrosis, the functional lymph transport of the district (genital area, lower limbs, etc.).

Overall, in this study, although genital lymphoedema can be associated with an overweight patient condition, the treatment choice depends on the global lymphoedema stage.

Lymphoscintigraphy consents to the visualisation of the lymphatic flux in both deep and superficial lymphatic vessels, and the obstruction level, and also permits measurements of the transport index (TI)<sup>[25]</sup>. A score lower than 10 means a normal TI, and a score equal to or higher than 10 signifies a pathological TI. Scores are made bilaterally, even in the cases of unilateral swelling<sup>[26]</sup>. Unfortunately, the postoperative lymphoscintigraphic comparison has not been homogeneously implemented between all three centers.

Independently of the surgical approach, all our patients were followed by trained physiotherapists for regular bandages and complete multimodal physical functional therapy. Compared to preoperative conditions, patients manifested a significant reduction in scrotal painful oedema, infections, and lymphorrhea. All patients continue to apply conservative treatments such as compressive garments, but overall reduce the frequency of physiotherapy sessions.

### **Debulking surgery**

Surgical debulking is followed by skin grafting/flap coverage depending on defect size and location<sup>[27,28]</sup>. Most patients treated with this approach were the most severe cases of GL (stage IIb-III) with lymphoscintigraphic images compatible with scrotal dermal backflow and slow or absent superficial/deep lymphatic flow.

Despite the obvious immediate improvement after surgical resection of oedematous tissue, generally patients presented a higher recurrence rate compared to derivative surgery or a combination of reductive and microsurgical treatment<sup>[7,29]</sup>. This is related to the GL aetiology, which is not solved with a debulking procedure, as it is merely a palliative procedure. The persistent lymphatic obstruction, or destruction proximally, is generally the reason for the lymphoedema recurrency or its complications<sup>[30]</sup>.

In our series, after the reductive intervention, we showed a clinical resolution or at least a reduction of the pain and scrotal oedema in all the treated patients. Penile lymphoedema seemed to be more difficult to treat, showing 75% oedema persistency.

In previous literature, patients receiving debulking surgery and flap reconstructions for GL generally had a total complication rate of more than 50%<sup>[7,31,32]</sup>. Our study did not show such a complication rate with no significant postoperative complications. Then, even cosmetically, ablative surgery applied in the advanced stages of disease may be suboptimal in both donor and recipient sites with significant scarring and poor wound healing<sup>[4,24,27,33]</sup>.

### **Microsurgical or ablative surgery + microsurgery**

LVA and MLVA are microsurgical techniques in which a lymphatic channel is anastomosed to a small vein (generally one-to-one in case of LVA, or multiple lymphatics into one vein in case of MLVA) to bypass an area of reduced lymph flow and drain the lymphatic excess into the venous bloodstream<sup>[6]</sup>. Mukenge *et al.* previously reported successful treatment of advanced penile lymphoedema with anastomoses from lymphatic vessels to the pampiniform plexus veins, which are located within the spermatic cord, adjacent to the lymphatics<sup>[23]</sup>.

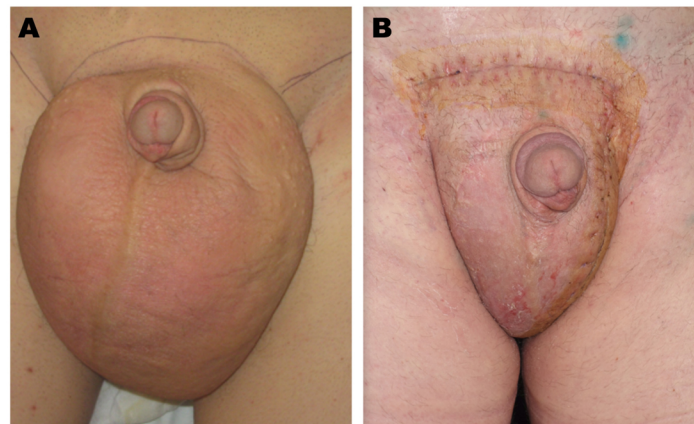
LVA and MLVA improve the long-term outcome of lymphatic microsurgery, but the efficacy, in terms of volume reduction and long-term stability, remains highly variable between surgical centres worldwide<sup>[34]</sup>. Moreover, recent reports demonstrated that the combination of reductive surgery (including less invasive liposuction) and microsurgery improves volume reduction, reduces the need for continuous compressive therapy and increases skin tone<sup>[35,36]</sup>. Similarly, excisional procedures, together with a VLNT, lead to limb circumference reduction and decrease the infection rate<sup>[12]</sup>.

Among our cases in this group, despite physiologic procedures do not remove fibrous tissue but lymph component only, we observed a reduction (or resolution) of the GL (unfortunately, only a qualitative interpretation can be extrapolated due to the lack of volume/lymphoedema quantification).

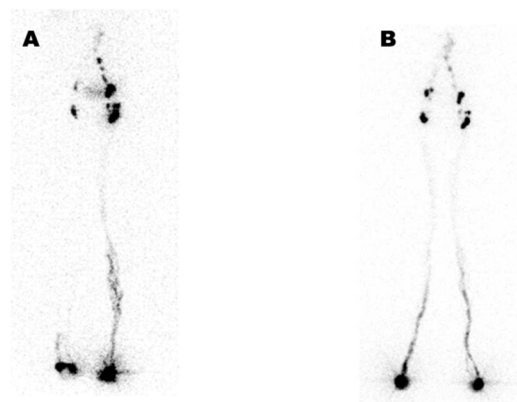
We can speculate, observing the TI, that cases treated with ablative surgery or a combination of ablative surgery and microsurgical derivation presented a more advanced/extended GL (always TI > 15) [Figure 4]. On the contrary, the GL treated only with derivative surgery showed a preoperative TI always less than 15 (with an average TI reduction of 43% when comparing pre- and postoperative lymphoscintigraphies). As an example, we reported the lymphoscintigraphic comparison pre- [Figure 5A] and post-operation at 12 months [Figure 5B] for the second patient of our series, who received only microsurgical treatment.

Besides that, no long-term complications (average follow-up longer than the previous group, 52.5 months) were found. Finally, penile lymphoedema reduction/resolution was achieved more frequently with physiologic or ablative + microsurgery than with only debulking surgery (75%).

To summarise, lymphatic-venous shunts can be indicated as the first choice in earlier stages (I or II) after conservative treatment unsuccess. Patients in these early stages have much less fibrosis of lymphatic vessels, limited skin, and subcutaneous anatomical changes. However, patients in the advanced lymphoedema stage can also obtain moderate volume reductions, meaningful symptoms, and quality of life improvement with functional surgery<sup>[6,30]</sup>.



**Figure 4.** Clinical outcomes before (A) and after (B) debulking surgery + MLVA for a stage III Genital Lymphoedema.



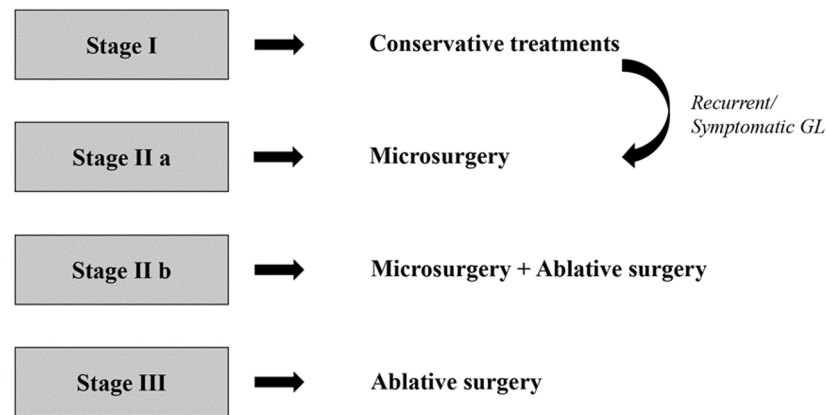
**Figure 5.** Lymphoscintigraphy pre (A) and post (B) for the second patient of our series: (A) preoperative condition; (B) Postoperative improvement of right lymphatic flow and disappearance of scrotal dermal backflow after three MLVAs at the right groin. TI = 12 preop and TI = 2 post-op.

The second option in terms of physiological treatment of GL is vascularised lymph node transfers (VLNT). This is another microsurgical technique, which consists of a lymphatic soft tissue free flap transposition from a donor site such as the groin, chest wall, neck or omentum to the affected lymphoedematous area<sup>[37]</sup>. Considering the concern of iatrogenic lymphoedema in the donor site<sup>[3,38]</sup> in this series, the omentum was the flap transfer of choice<sup>[39,40]</sup>.

When derivative procedures are not sufficient as isolated procedures<sup>[41]</sup>, we propose the combination of LVA/MLVA with VLNT. For instance, in the first patient of our series, we first performed a lymphatic LV shunt, and secondly, due to the partial blockage resolution at the level of the scrotum, but the persistence of penile oedema, an omental VLNT was performed.

### Limitations

This study has some limitations, mainly related to the type of study: being a multicenter one, diagnosis, clinical evaluation, surgery, and follow-up were performed by three different consultants; this can lead to a significant bias in data analysis, interpretation and hard comparability. Moreover, the outcomes evaluation was predominately qualitative due to the lack of systematic postoperative lymphoscintigraphy or other quantitate parameters. Finally, the retrospective nature of the study and the number of patients represent



**Figure 6.** Surgical treatment algorithm according to our experience.

the extra limitations. Further clinical studies with a prospective design with larger cohorts, standardised quantification of GL and prolonged follow-up are advocated; all these will help to clarify therapeutical protocols and improve patient outcomes.

In conclusion, for GL stage I and mild presentations, we suggest maximal conservative therapy at the first step. In stages II and above or in the presence of persisting clinical symptoms, physiological surgery should be proposed. Microsurgical options should be recommended in recurrent symptomatic GL (particularly when lymphorrhea/chylorrhea and/or recurrent perineal infections dramatically impact patient quality of life) and eventually combined with debulking.

Alternatively, in the case of chronic and extended GL stage IIb-III, excisional procedures represent our first approach, considering the fibrotic and adipose tissue and the main component to address to reduce anatomical deformity and functional impairments [Figure 6]. Finally, we always recommend postoperative CDT to all patients, in order to maintain results and prevent recurrences.

## DECLARATIONS

### Authors' contributions

Authors who performed the surgeries: di Summa PG, Campisi C, Maruccia M

Designed the study and participated in data analysis: di Summa PG

Performed data analysis, interpretation, and manuscript preparation: Guiotto M

Performed data acquisition, as well as provided administrative, technical, and material support: Elia R, Molinari L, Fresa M, Nicod Lalonde M

### Availability of data and materials

The datasets used and/or analyzed during the current study are available from the corresponding author upon reasonable request and deposited at the University of Lausanne. All figures and tables are original.

### Financial support and sponsorship

None.

### Conflicts of interest

All authors declared that there are no conflicts of interest.

### Ethical approval and consent to participate

The study was conducted accordingly to the guiding principles following the Declaration of Helsinki of 1975. Informed consent was obtained from all patients, including approval for scientific publication and photographic/video documentation.

### Consent for publication

Informed consent was obtained from all patients, including approval for scientific publication and photographic/video documentation.

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