

## A COMPREHENSIVE REVIEW OF SURGICAL INTERVENTIONS FOR ADVANCED LYMPHATIC FILARIASIS: CURRENT APPROACHES AND OUTCOMES

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Received : 09/12/2023  
Received in revised form : 10/01/2024  
Accepted : 25/01/2024

### Keywords:

Lymphatic surgery, lymphatic filariasis, elephantiasis, lymphedema.

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DOI: 10.47009/jamp.2024.6.1.249

Source of Support: Nil,  
Conflict of Interest: None declared

Int J Acad Med Pharm  
2024; 6 (1); 1254-1258



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### Abstract

Lymphatic Filariasis (LF) is a debilitating disease especially in the advanced stages when the quality of life of the patient is grossly affected due to the associated deformities in the limbs and the urogenital system. In these advanced stages, medical management is seldom effective in improving the deformity and hence surgical management comes into play. Various surgical and micro-surgical techniques are brought into play with varying degrees of success. These range from excisional procedures and lymphatic shunts and anastomosis to hydrocelectomy for bancroftian filariasis. However, definitive studies are lacking on the subject. Therefore, we have investigated all these various techniques across the existing literature on the topic and presented our findings. It was concluded that surgical techniques when applied to advanced grades of lymphatic filariasis, leads to improvement in disfigurement and improved quality of life in the patients. There is a need to develop surgical procedures and medical advancement for optimal outcome of this disease therapeutics until then shunt followed by reduction surgery with skin graft is the most reliable option.

## INTRODUCTION

Lymphatic filariasis (LS) is also known as elephantiasis which is a tropical parasitic disease that affects mainly the lymphatic system. It is caused by parasitic organisms, mainly *Wuchereria Bancrofti* and less commonly by *Brugia malayi* and *Brugia timori* which have affected an estimated 67.8 million patients who mainly reside in the tropics and are economically depressed (Ramaiah KD and Ottesen EA 2014).<sup>[1]</sup>

The spread occurs due to *Aedes* mosquito bite which acts as a vector for this disease. In the majority of the cases the incubation period lasts around 9-12 months followed by severe symptomatology including lymphedema of limbs and genitalia which is found to be painful and profoundly disfiguring for the patient. Quality of life is severely affected which is measured in terms of Disability Adjusted Life Years (DALYs) which takes into consideration the psychosocial and economic burden due to LF because it is a disease that contributes more towards morbidity rather than

mortality (Hotez PJ et al. 2014).<sup>[2]</sup> According to the latest Global Burden of Disease Study, LF has been attributed 2.3 million DALYs which is a modest estimate as compared to more fatal counterparts such as Tuberculosis (Addiss DG and Brady MA 2007).<sup>[3]</sup> As of recently, the Global Program to Eliminate Lymphatic Filariasis (GPELF) has been responsible for the successful implementation of multiple mass drug administration programs in more than 50 countries in the tropics which has allowed a reduction in new cases and disease burden (Ramaiah KD and Ottesen EA 2014).<sup>[4]</sup>

While the management of lymphatic filariasis is usually conservative, the surgical interventions remain a neglected area of research. Various excisional and physiologic procedures have been utilized such as Vascularised Lymph Node Transfer (VLNT), Charle's procedure, lymphaticovenular anastomosis (LVA), radical reduction with preservation of perforators (RRPP) among a few others (Ciudad et al. 2017).<sup>[5]</sup> No definitive literature exists that summarizes or explores these various

surgical avenues for LF. The scope of this review extends to various techniques utilized to treat advanced LF along with associated complications and quality of life thereafter.<sup>[6]</sup>

Conservative management includes treating the patient with the drug of choice which is diethylcarbamazine as advised by the Center of Disease Control (CDC). However due to lack of community awareness and chronicity of the disease the effectiveness of these drugs is reduced significantly. Hence surgical interventions have been considered to improve quality of life. They have mainly been advocated for advanced lymphatic filariasis to reduce the lymphedema which in turn reduces the inflammatory episode, and infections and to improve function as well as the cosmetic appearance of the limb.<sup>[7,8]</sup>

In this systematic review we intend to report on the patient reported outcomes in surgical management of patients suffering from lymphatic filariasis and bridge the knowledge gap present.<sup>[9,10]</sup>

## MATERIALS AND METHODS

### Search Strategy

This study was conducted by following the guidelines in accordance with Preferred Reporting Items for Systematic Reviews and Meta-Analysis (PRISMA). The literature search for this article was conducted through various databases including the National Library of Medicine (Pubmed), Pubmed Central (PMC) and Google Scholar. Utilization of medical subject heading (MeSH) terms was done which included all the relevant terms.

### Inclusion Criterion

- Patient is suffering from advanced lymphatic filariasis with prior conservative management.
- Surgical intervention is used for the treatment of lymphatic filariasis.
- Studies conducted from the year 2000 till date.
- Studies in the English language

### Exclusion Criterion

- Studies in languages other than English
- Studies before the year 2000.
- Non availability of the full text versions of the articles.

- Studies that did not outline use of any surgical intervention for the treatment of LF.

### Eligibility and Extraction

All the relevant articles were screened in accordance with the criteria listed and assessed accordingly. The preferred language in all the articles is English. Randomized control trials, review articles, case reports and case series were included in the review. For selecting the appropriate articles, each of them was analyzed and read thoroughly.

Data extraction was done by the reviewer, which included the author's name, study type, age, gender distribution, surgical intervention, complications, follow-up time, and outcome, along with the ones mentioned in [Table 1].

## RESULTS

The papers selected were reviewed accordingly and their characteristics and the data extracted are stated in Table 1. The outcome has been favorable to the patient with improvement in the limb deformities and quality of life. The complications have not occurred and, in a few cases, seen with complications, they have been managed adequately in the subsequent follow ups.

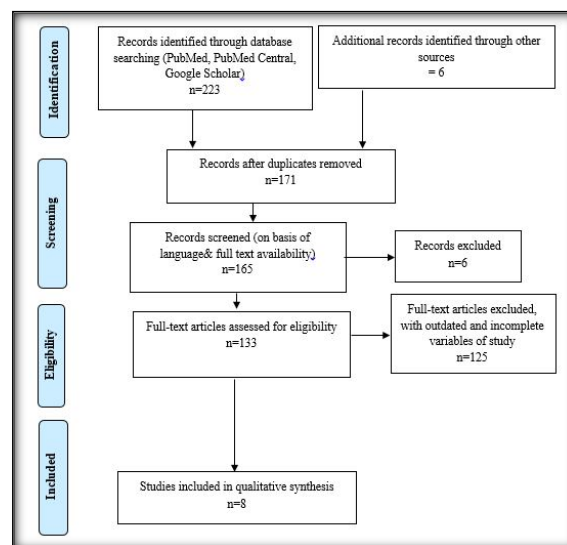


Figure 1: PRISMA

Table 1: Characteristics of Research articles included in the study

Study	Year	Location	Type of Study	Diagnosis	Surgical Intervention	Outcome	Follow up time	Complications
Victor et al.	2021	India	Retrospective Descriptive study	Grade 4 filarial lymphoedema of leg and foot	Nodovenous shunt surgery followed by debulking surgery/sculpting surgery	Reduction of 2-3 cm in the limb circumference	Till 6 to 7 years	Skin flap necrosis, seroma, cellulitis
K KN, et al.	2020	Indonesia	Case Report	Grade 4 lymphoedema of leg and foot	Excision of excess and edematous tissue followed by bandage therapy	Reduction in limb size and improved quality of life	2 years	No complications
Istranov et al.	2023	Russia	Case Report	Grade 3 bilateral lower limb lymphoedema	Lymphonodovenous shunt and removal of the affected tissues of the urogenital region,	A decrease in circumference of lowest extremities in lower leg	3 months	No complications

				and scrotal lymphedema	phalloplasty, and scrotoplasty with rotational skin flaps	area by 68 cm on the right and by 69 cm on the left. Patient's body weight reduced by 69.4 kg, and the scrotum decreased 63 cm		
Ram et al.	2014 - 2018	India	Case Report Series	Grade 2 and Grade 3 lower limb lymphedema	Vascularized lymph node transfer and serial excision	Significant reduction in lower limb size p<0.005 and reduction in episodes of acute lymphangitis	6 months	Lymph stasis, seroma, partial loss of skin graft, episodes of acute lymphangitis
Ruch et al.	2001 - 2003	Thailand	Prospective Study	Lymphedema of lower extremities	Microsurgical lymphonodovenous implantation	Reduction in circumference of affected limb	till 5 months	No complications
Terashi et al.	2003	Japan	Case Report	Filarial Chyluria	Lymphonodovenous shunt anastomosis	Clear urine and no recurrence of the chyluria and no urinary obstruction.	1 year	No complications
de Godoy et al.	2010	Brazil	Case report	Elephantiasis	Resection of excess tissue followed by bandaging	Reduction in lymphedema	Monthly	No complications
Rao et al.	2000	India	Clinical Trial	Filarial lymphoedema	Lymphonodovenous shunt anastomosis	Reduction of 25% to 50% in edema volume in 46.7% cases and more than 50% in 17.3% cases.	3 months	Hematoma (8.5%), Wound infection (13.6%), Transient lymphorrhea (13.6%)

## DISCUSSION

According to the case report, conducted by K KN et al. (2020) a 28-year-old patient complained of swelling in the right leg for 4 years which worsened in the last 3 months.<sup>[11]</sup> The examination was conducted on the leg which stated that it was edematous and hyperpigmented plaques were present. Blood examination was positive for Wuchereria Bancrofti. After medical treatment the lymphedema did not resolve due to which surgery was performed. The patient responded well to treatment and had no complications for 2 years (K KN et al. 2020).<sup>[12]</sup> This shows that the surgical intervention was a requisite where medical management could not help the young adult lead a normal and healthy life.

In another study conducted to evaluate the usefulness of standardized clinical classification of hydroceles in Lymphatic Filariasis endemic countries for their surgical management concluded that Standardized Clinical Classification of Hydroceles based on the Stage of enlargement of the scrotum and the Grade of burial of the penis appears to be a useful tool to guide the decision about the level of care and the surgical technique required (Capuano GP, Capuano C. 2012).<sup>[13]</sup> The debulking surgeries are performed

more in advanced cases of Lymphatic Filariasis with Grade 4 Lymphedema with nodules, ulcers and warty growths (i.e., Elephantiasis). The core principles of these surgeries were to augment the lymphatic drainage and debulk the lymphedematous & lymph producing surface thereby reducing the load. These procedures help reduce the lymphedema and improve the function of the limbs (Serville 1987).<sup>[14]</sup>

In a study VLNT (vascularized lymph node transfer) along with excisional procedure was performed in advanced LF patients and clinical efficacy was evaluated. This was performed on 17 patients with Grade 2 or Grade 3 Lymphedema in India. Swelling which resolves with elevation, pressing on may leave an impression & the swelling that does not resolve with elevation, pressing on which doesn't leave any impression correspond to Grade 2 and 3 respectively. They underwent excessive soft tissue removal and the supraclavicular lymph node was transferred to the dorsum of foot. After the procedure the patients reported reduction in heaviness of leg (p<0.005) and episodes of acute lymphangitis. The study concluded that combination of VLNT and excisional procedures are a safe and reliable approach to patients with advanced Lymphatic Filariasis (Chilgar et al. 2019).<sup>[15]</sup>

The trial of 75 patients regarding the lymphonodovenous shunt was conducted to determine the factors associated with unilateral filarial lymph edema developed among patients while considering the gender, duration, adenolymphangitis, venous reflex as well as operative impression of lymphatic nodules. Postoperative complications were found amongst the patients in minimal percentages such as 8.5% as wound hematoma, 13.6% as wound infection and decreased the volume of edema upto 46.7% and in some cases 50%. The surgical intervention was reported to be presented with best results among patients with great amount of lymphatic drainage edema and it was further elucidated that increased occurrence of adenolymphangitis episodes depicts optimal working of lymphonodovenous shunt surgical intervention (Rao et al. 1999).<sup>[16]</sup>

In another study, twenty patients with early lymphedema due to Filariasis and twenty-four patients with Elephantiasis of the lower limb were subjected to Lymph nodo-venous shunt (LNVS) and Charles operation respectively. This study concluded that while excisional surgery such as Charles operation (subcutaneous and deep fascial excision followed by full-thickness grafts) becomes necessary for late stages of lymphedema which progressed to elephantiasis, Lymphnodo-venous shunt alone is sufficient to relieve early stages of lymphedema due to Filariasis (Dandapat et al. 1986).

Grade 4 lymphedema patients with lack of treatment options surgically or any other way were selected for nodovenous shunt which would be led by the reduction surgery to produce the optimal outcome for patients to increase the viability of patients. Victor et al. conducted a study to determine the outcomes of such procedures with retrospective study design. They reported that 16 patients from 2010 to 2019 were treated surgically for this condition which mainly consisted of nodovenous shunt followed by debulking surgery and resulted in more than 5 cm reduction in the circumference of limbs after the operation which indicate significant consequences of this intervention. Shunt of the nodular venous region that is followed by reduction surgery of lymphedema is the best treatment option available to increase the quality of life as well as decrease the morbidity or secondary complications of lymphedema (Victor et al. 2021).

#### Limitations

The lack of previous research on the topic along with missing data from randomized control trials limits this study with recommendation of further trials and studies - observational and cohort. A combination of these surgical techniques should be applied on a case-to-case basis to further improve the clinical outcome, with an extended follow up period to battle the long-term complications adequately and without delay.

## CONCLUSION

Lymphatic filariasis with its parasite etiology and spread by mosquitoes has been known to affect patients significantly with limited availability of surgical and medical interventions. The condition prevails for elongated time and causes edema in limbs as well as genitals which affect the patients drastically. Nodovenous shunt and reduction surgery as well as removal of the affected tissues of the limbs and microsurgical lymphonodovenous implantation are some interventional surgical procedures opted for this disease. Nodovenous shunt followed by debulking surgery appears to be with less complications post operatively depicting it as the best route of treatment. There is need to develop more surgical procedures as well as medical approaches to decrease the suffering time of patients as well as the severity of the disease among suffering individuals.

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