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REVIEW ARTICLE

Genital elephantiasis: Surgical treatment and reconstruction

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Abstract

Genital elephantiasis is a severe form of lymphedema of the groin. It is characterized by progressive enlargement and distortion of the genitals, presenting significant physical, psychological, and social challenges to the affected individuals. Although pharmacological treatment of filariasis is well-established in the medical field, the surgical management of genital elephantiasis can be varied and confusing. This review article provides an in-depth analysis of the etiology, classification, severity grading, and various effective surgical treatment and reconstructive modalities commonly employed by surgeons since the early twentieth century. We also discuss how a combination approach of ablation, soft tissue coverage, and lymphatic reconstruction is viable for treating genital elephantiasis. By examining the literature, we hope to provide insights into how surgery plays a role in the holistic management of genital elephantiasis.

Keywords: Charles procedure; Genital elephantiasis; Lymph vessel transfer; Lymphatic reconstruction; Lymphedema debulking

1. INTRODUCTION

Genital elephantiasis has been classically thought of as a chronic, severe swelling, and hypertrophy of the genital soft tissue due to lymphatic filariasis. We now know that lymphedema of any etiology, congenital, or cancer-related, if left unchecked, will result in soft tissue hypertrophy, dermal hyperkeratinization, and eventual elephantiasis.

A fraction of patients with lower extremity lymphedema will suffer from genital lymphedema. However, the true incidence of genital lymphedema and elephantiasis is poorly understood. Recently, Clinckaert et al[1] performed a systematic review to determine the prevalence of lower limb and genital lymphedema after prostate cancer treatment. From the 18 articles reviewed, the group found a prevalence of lymphedema in the lower limbs and genital regions ranging from 0% to 14% and 0% to 1% after surgery and 0% to 9% and 0% to 8% after pelvic radiation, respectively. There was a much higher prevalence in patients that underwent pelvic lymph node dissection followed by pelvic radiotherapy (18%-29% and 2%-22%). For congenital lymphedema, a study of 138 children from Children's Hospital Boston found that males were seven times more likely to have genital lymphedema than females.[2]

Genital lymphedema and elephantiasis can impart significant physical morbidity to our patients. Patients may experience multiple episodes of cellulitis, difficulty fitting clothes, swelling discomfort, and malignant transformation from repeated ulcerations. This entity can negatively impact their psychosocial well-being as well. Patients often report lowered self-esteem and poor sexual function. Features of genital elephantiasis are listed and described in Fig. 1.

In this review article, we explore the management of genital elephantiasis with an emphasis on surgical treatment and reconstruction.

2. CLASSIFICATION AND ETIOLOGY

Lymphedema can be classified into primary and secondary lymphedema. Primary lymphedema is subdivided based on the age of onset:

1. Congenital lymphedema usually presents at birth or within 2 years of life. This entity has a strong familial pattern.
2. Lymphedema praecox occurs at puberty or the beginning of the third decade of life. The majority of primary lymphedema patients belong to this group.
3. Lymphedema tarda has an onset after 35 years of age.

Primary lymphedema is most commonly associated with a hypoplastic lymphatic system. Lymphatic collectors and lymph nodes are smaller and fewer in number, and this situation is frequently seen in lymphedema praecox patients. A complete absence of lymphatic trunks is associated with congenital lymphedema, whereas hyperplastic, tortuous channels are often seen in lymphedema tarda patients.

Secondary lymphedema results from an insult to the lymphatic system resulting in sclerosis and obstruction. Although the most common cause of lymphedema worldwide is filariasis caused by infection by *Wuchereria bancrofti*, in developed countries, most secondary lymphedema cases are due to malignancy or related to the surgical extirpation and adjuvant therapy of cancer.

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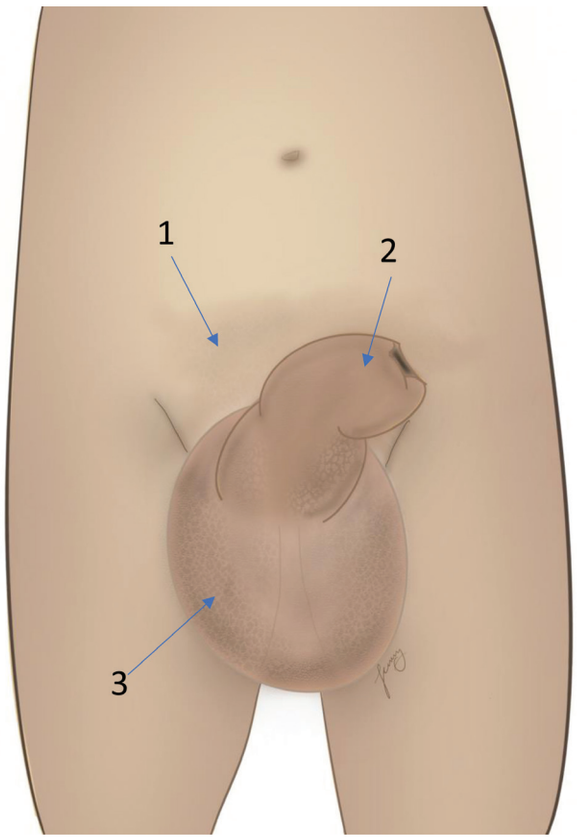

Fig. 1 Features of male genital elephantiasis are depicted in this figure. Suprapubic skin (1) thickness and undergoes verrucous hyperplasia. The prepuce can undergo edematous change and enlargement (2). This leads to hygiene problems and difficulties with urination. The penis can also become buried, which leads to the inability to perform sexual intercourse. The scrotal skin undergoes verrucous hyperplasia, and the testes enlarge due to severe hydrocele formation (3).

3. SEVERITY AND GRADING

There is no universally accepted reporting nomenclature for disease severity. Capuano and Capuano[3] described a preoperative classification based on laterality, size of the hydrocele, and degree of buried penis. Although the extent of the deformity has some correlation with the severity of the lymphedema, this relationship is nonlinear. Therefore, it is difficult to ascertain the degree of lymphosclerosis and judge the type of lymphatic surgery needed.

Indocyanine green (ICG) lymphography has changed how clinicians evaluate extremity lymphedema and can be applied to genital lymphedema. Patterns of dermal backflow such as splash, stardust, and diffuse are pathognomonic of lymphedema and can often allow for early diagnosis even before florid symptoms manifest.[4] The utility of this severity staging method is the ability to discern which patients will benefit the most from physiological lymphatic procedures like lymphovenous anastomosis (LVA) before elephantiasis occurs.

The genital lymphedema score (GLS) was developed to evaluate genital lymphedema without ICG lymphography because these specialized near-infrared scanners may not be available in all medical settings.[5] The GLS is based on subjective symptoms alone (Table 1) and has been demonstrated to correlate well with genital dermal backflow patterns and ICG lymphedema staging. The GLS can also be used as a follow-up measurement after therapeutic interventions for genital lymphedema.

4. ABLATIVE AND DEBULKING SURGERY

Excision of lymphedematous tissue is considered one of the oldest surgical treatments of lymphedema. In 1901, Sir Richard Henry Havelock Charles published in the *Indian Medical Gazette* a series of 140 consecutive patients treated successfully for scrotal lymphedema. This article was the basis for a 1912 book chapter on the same topic, whereby the eponymous "Charles procedure" was based on.[6] Although Charles mainly described his procedure for scrotal elephantiasis, the idea of the Charles procedure would evolve into what is currently understood today; a form of surgical treatment where the lymphedematous lower extremity is radically debulked and then covered with skin grafts harvested from the surgical specimen. Charles procedure is still regarded as a treatment option for chronic and advanced lymphedema where skin hypertrophy and verrucous overgrowth have resulted in elephantiasis. Over the years, many have made modifications to the original Charles procedure. These include using split-thickness skin grafts, concurrent free vascularized lymph node transfer (VLNT), and delayed skin grafting with or without negative pressure wound therapy.[7,8] Homan[9] described a modification where a longitudinal incision is made along the lateral and medial aspect of the legs lifting the dermis off the fat and preserving the subdermal plexus. The underlying fat is removed, and the skin is trimmed and closed primarily to accommodate the reduced limb volume.[9] Salgado et al[10] refined this concept by utilizing microsurgical principles to preserve skin perforators and reduce the incidence of wound healing complications. The Charles procedure stood the test of time and is still valuable for a lymphatic surgeon's armamentarium today.

Thompson[11] first described the buried dermal flap as a treatment for advanced chronic lymphedema of the lower limb in 1959 and subsequently published his 10-year series in 1970. Long dermal adipose flaps were raised and preserved in this procedure instead of discarded. The redundant end of the flap

Table 1

GLS system based on subjective symptoms related to genital lymphedema

Subjective symptoms	No	Yes
Sensation of heaviness	0	1
Sensation of tension	0	1
Swelling	0	1
Urinary troubles due to genital edema	0	2
Cutaneous lymphatic cyst	0	2
Genital lymphorrhea	0	2
Total = GLS	Range	0-9

GLS = genital lymphedema score.

was furled into intermuscular crevices so that superficial lymphatic collectors are brought into direct contact with the main vessels of the limb and their accompanying deep lymphatic trunks, increasing the probability of lymphovenous shunts forming.[11]

Other excisional techniques are still being done by surgeons around the world, albeit with much lower frequency. Emmanuil Kondoleon observed that the deep fascia in patients with lymphedema is commonly rigid and fixed to the surrounding adipose tissue. This led to the development of the Kondoleon procedure in the early 1900s, where wide excision of the fascia was performed with concomitant partial removal of the expanded adipose tissue.[12] However, failure and early recurrence of lymphedema have been attributed to the healing and regeneration of the deep fascia. Sistrunk[13] described his technique as a modification to the Kondoleon procedure; strips of skin and soft tissue with their underlying deep fascia were removed on the lateral aspect of the lower extremity and primarily closed.

Adipose tissue expansion has been a prominent feature after lymphatic stasis and inflammation. Naturally, surgeons have applied the concept of suction lipectomy to lymphedema with varying results. Early reports of lower extremity liposuction were disappointing, resulting in minor improvements when performed without skin excisions.[14] However, liposuction techniques and equipment improvements have increased this operation's efficacy. In 2016, Brorson described excellent long-term results using power-assisted liposuction with the help of tumescent technique and tourniquet to reduce blood loss.[15] Although liposuction is also classified as an ablative surgery, cadaver, and imaging studies have demonstrated that lymphatic vessels were not disrupted when performed parallel to the limb, and lymphatic transport capacity is not further aggravated.[16,17] Postoperative compression garments are critical for reducing volume and cannot be stopped.

5. LYMPHATIC RECONSTRUCTION

The lymphatic system of the superficial soft tissue of the groin comprises an intricate system of fine lymphatic vessels that ends at the inguinal group of lymph nodes. The lymphatic networks of the left and right genitalia are closely communicating in the midline. The inguinal lymph nodes can be segmented into four quadrants, mainly the superior-medial, superior-lateral, inferior-medial, and lastly inferior-lateral groups. The largest number of lymph nodes belong to the inferior-lateral group, which drains the lower extremity. The genital region drains into the superior-medial and inferior-medial groups following the course of the external pudendal vein toward the saphenous opening. From there, the major outflow channel is through the deep inguinal nodes and subsequently into the external iliac lymph nodes.

5.1. Lymphovenous anastomosis, vascularized lymph node transfer, and lymph vessel transfer

Physiological procedures have gained prominence in the past three decades. These procedures include LVA, VLNT, and lymph vessel transfer (LVT). These procedures prominently feature microsurgical concepts and techniques to achieve the desired outcome. LVA is a minimally invasive procedure with low downtime. Due to its immediate shunting effects, it can provide symptomatic improvement and limb volume reduction in weeks.

In VLNT, physiologically normal lymph nodes and their vascular pedicles are harvested and implanted into lymphedematous portions of the limb following microsurgical vascular anastomosis. These transplanted lymph nodes directly drain the limb via a "pump" mechanism and act as the epicenter for lymphangiogenesis.[18] Unlike LVA, there is no immediate shunting effect, and surgeons can expect to see improvement only after several months.

The concept of LVT was first reported by Koshima et al.[19] The group sought to replace severely diseased lymphatic vessels with those in a lymphoadiposal flap, effectively bypassing the sclerosed obstructed segment. This lymphoadiposal flap was harvested from the first dorsal webspace of the foot. Functioning lymphatics were identified with patent blue dye injection of ICG fluorescence. Chen et al[20] expanded on this concept by using the superficial circumflex iliac artery perforator (SCIP) flap for LVT. They proposed that the SCIP flap was a better lymphoadiposal flap because of the higher density of lymphatic vessels present in the flap, superior donor site cosmesis, and well-studied anatomy.[20] Patients from both studies reported limb volume reduction and symptomatic relief.

5.2. Lymph-interpositional-flap transfer

LVT was further refined by marrying the concepts of lymphoadiposal flap with lymph axiality. The basis of lymph axiality was derived from direct observations of lymphatic flow restoration in multiple cases of replantation and free tissue transfer. Closely approximating the proximal and distal lymphatic stumps between the flap and the recipient site and orienting the lymphatic axes in their physiological flow direction during flap inset were the strongest predictors of restoration of lymph flow.[21] The lymph-interpositional-flap transfer (LIFT) technique utilizes these principles to achieve simultaneous soft tissue and lymphatic vessel reconstruction without supermicrosurgery.[22] The result is also a shorter and technically less demanding operation. LIFT surgery can also reduce the risk of secondary lymphedema in the recipient site limb or treat established lymphedema by draining the dermal backflow regions. When LIFT surgery was performed for soft tissue defects disrupting major lymphosomes, postoperative ICG lymphography demonstrated lymph flow restoration and fluorescence signal within the LIFT flap.

Physiological procedures, however, do not remove hypertrophic diseased skin in advanced lymphedema. Without ablative techniques, the likelihood of these skin changes and deformity reversing is low. Furthermore, the genital region is challenging to apply compression, reducing the overall effectiveness of lymphovenous anastomoses.

6. COMBINATION APPROACH: ABLATIVE AND RECONSTRUCTIVE SURGERY

In the past, treatment of genital elephantiasis commonly involved radical resection and soft tissue reconstruction without prioritizing lymph flow restoration. Even when considered, lymphatic reconstruction is usually performed as an additional procedure, for example, adding on a lymph node transfer. Yamamoto et al[23] described the radical resection and reconstruction (RRR) technique, which combines ablative surgical techniques with modern lymphatic reconstruction concepts described earlier to achieve satisfactory form and lymphatic function in one surgery. Indications for RRR include genital lymphedema with gross fibrosis and verrucous skin changes. As male genital elephantiasis is more common and challenging to manage than in females, we shall focus on RRR of male genital elephantiasis.

RRR can be divided into three main components:

1. Radical elephantiasis tissue resection.
2. Soft tissue reconstruction using chimeric SCIP flaps.
3. Lymphatic reconstruction via LIFT concept.

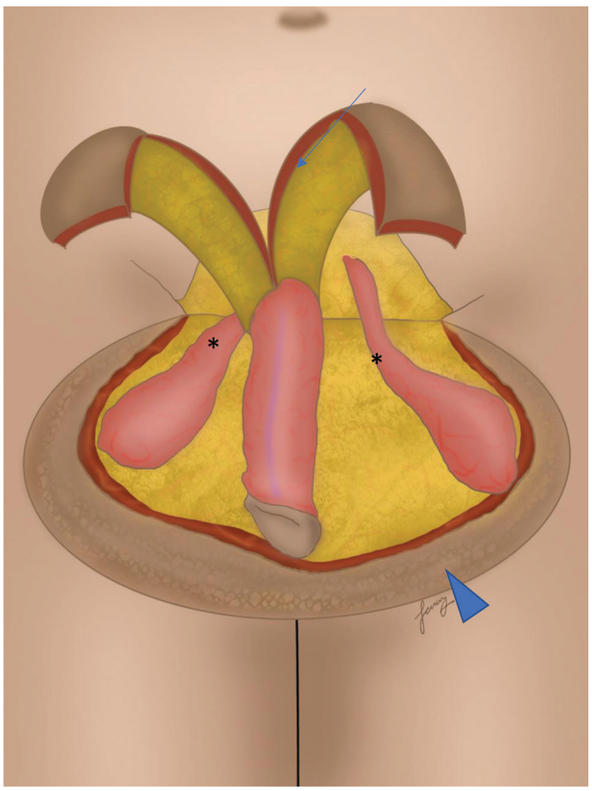

Fig. 2 Radical resection involves removing all the skin and soft tissue affected by elephantiasis. The penile skin is resected (blue arrow) above the Buck's fascia, preserving the dorsal neurovascular bundle. The spermatic cord (*) is carefully dissected and protected. All diseased scrotal skin is removed (blue arrowhead). Suprapubic skin may need to be resected if involved as well.

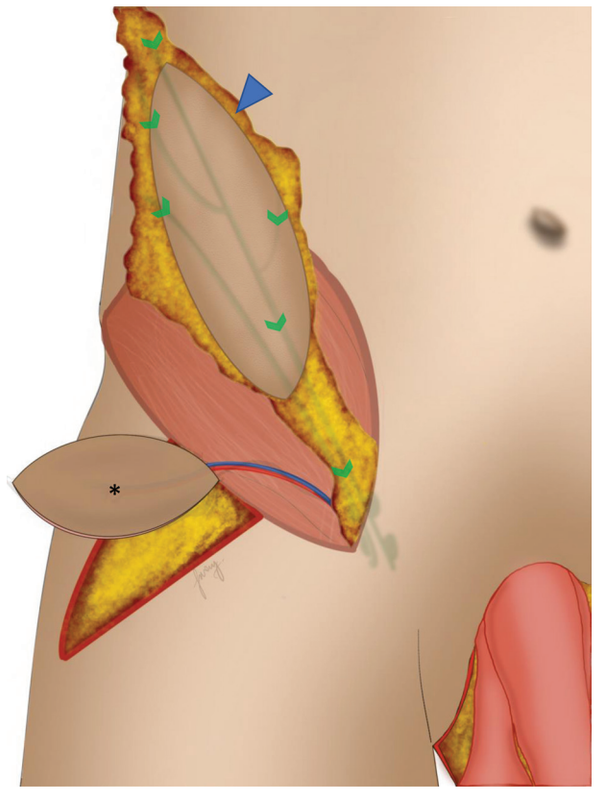

Fig. 3 In RRR, the SCIP flap (blue arrowhead) is designed to include patent lymphatic channels (green lines), which reside in the deep fat. Surrounding deep fat around the skin paddle can be recruited into the flap to extend the reach of the lymphatic reconstruction. Special attention is paid to the physiological flow of the lymph within these channels (green chevrons depict the flow direction). This concept of lymph axiality is used to bridge edematous areas on the contralateral side to drain into the right inguinal lymph nodes. A pure skin perforator flap (*) can also be harvested to resurface the penis. RRR = radical resection and reconstruction; SCIP = superficial circumflex iliac artery perforator.

6.1. Elephantiasis tissue resection

The surgeon should aim to resect all pathological skin and subcutaneous tissue, as incomplete resection can lead to lymphoceles and recurrences. Commonly resected areas include the scrotal skin, suprapubic skin, penile skin, and prepuce (Fig. 2.). Injury to critical structures, such as the dorsal neurovascular bundle, will be avoided if one stays on the suprafascial plane (above Buck's fascia). Care must be taken to isolate and protect the spermatic cords and the testes during dissection. An orchidopexy is performed to prevent torsion of the testes.

6.2. Soft tissue reconstruction

Soft tissue coverage should be planned based on the defect location and size. The SCIP flap is versatile and the workhorse flap for RRR. The SCIP flap offers several advantages:

1. Well understood anatomy.
2. Ability to raise multiple skin paddles.
3. Pure skin perforators can be harvested when thin skin coverage is required.
4. Multiple constant axial lymphatic pathways stretching from the iliac to inguinal regions are present within the flap.

A large SCIP flap resurfaces the scrotum, whereas a second SCIP pure skin perforator is raised to resurface the penis if required (Fig. 3). Sometimes part of the prepuce remains unaffected by lymphedema and can be preserved for advancement proximally. This helps to reduce the defect size. A full-thickness skin graft may be harvested for the remnant defect instead. Care must be taken not to injure the superficial inguinal lymph nodes to minimize exacerbation of lymphedema of the lower extremity. The flaps are then transposed into the defect through a subcutaneous tunnel (Fig. 4).

In our experience, the SCIP-LIFT flap is very reliable, with a low incidence of tip necrosis (<1%) due to preoperative planning and intraoperative measures. Ultrasound is used preoperatively to locate the dominant superficial circumflex iliac artery perforator. This axial vessel is sited in the center of the skin paddle to maximize tissue perfusion. Intraoperatively, the SCIP-LIFT flap is raised with the full thickness of subcutaneous fat to incorporate all levels of vascular networks. ICG angiography is sometimes performed to assess tissue perfusion to the flap tips as well. Areas with slow or no fluorescence signal are removed before inset. The two most important factors when rotating and tunneling the SCIP-LIFT flap into the scrotal defect are the amount of tension on the pedicle and the size of the subcutaneous tunnel. We repeatedly check the amount of stretch and tension on the pedicle before performing our final inset. There is no strict rule on the direction of rotation for the flap, as the surgeon should find the position with the least tension on the flap pedicle. Finally, the subcutaneous tunnel should be wide enough to accommodate the flap without excessive compression.

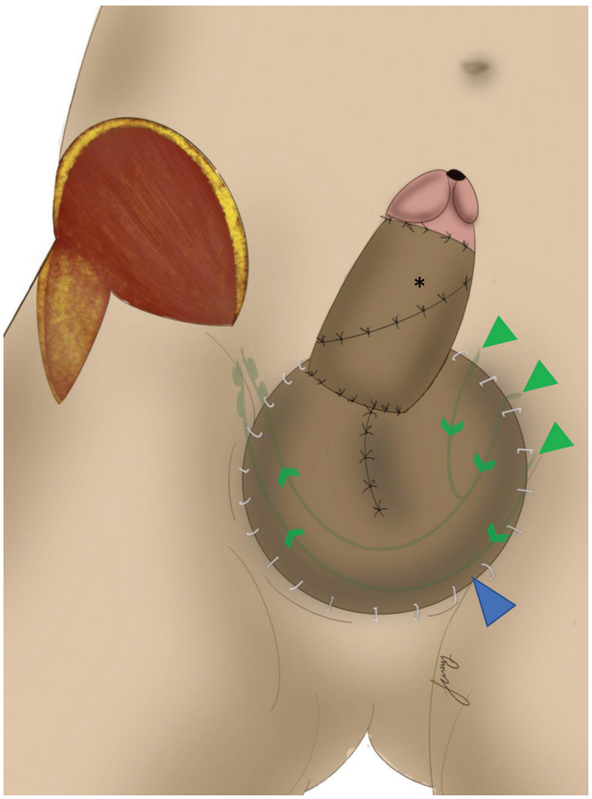

Fig. 4 The pedicled LIFT-SCIP flap (blue arrowhead) is used to reconstruct the scrotum. The flap inset is an essential aspect of LIFT to ensure the lymphatic stumps (green arrowheads) are inserted into the edematous regions. The lymphatic channels will drain the edema fluid through the flap into the inguinal lymph nodes on the right (chevrons). The pure skin perforator flap (*) is used to resurface the penis. LIFT = lymph-interpositional-flap transfer; SCIP = superficial circumflex iliac artery perforator.

6.3. Lymphatic reconstruction

Preoperatively, ICG lymphography is performed to delineate patent lymphatic channels within the SCIP flap. These channels are marked and included within the flap to be raised (Fig. 3). The main SCIP-LIFT flap is raised with deep fat intact to preserve these lymphatic channels. The flap harvest is fast and expedient with minimal perforator dissection. Inguinal lymph nodes are carefully preserved around the pedicle of the SCIP-LIFT flap. Deep fat surrounding the skin paddle is recruited into the flap to extend the lymphatic channel's reach and give an arc of movement of the lymphatic stumps to increase the probability of spontaneous reconnections. A subcutaneous tunnel is created under the suprapubic skin to allow passage of the flap into the scrotal region. The flap is lined horizontally so that the distal and lateral lymphatic stumps are in contact with the soft tissue of the lymphedematous side. It is critical that the deep fat of the flap, which contains the lymphatic stumps, is brought in contact with the deep fat of the lymphedematous areas. Lymph retained in the soft tissue of the pathological side is absorbed into the flap through new lymphatic channel connections generated via lymphangiogenesis (Fig. 4). The absorbed fluid is then drained along the lymphatic collectors in the flap and into the contralateral inguinal lymph nodes around the pedicle of the SCIP-LIFT. This can be demonstrated via ICG lymphography.

RRR helps reduce postoperative complications such as seroma, infection, poor wound healing, and recurrence of elephantiasis. As there is restoration of the genital form and function, GLS and quality of life scores have also been demonstrated to improve. It is crucial to understand that the intent of RRR is curative. Radical resection removes most of the tissue affected by elephantiasis from the genital region. However, complete ablation of all diseased tissue is often impossible. Hence, the LIFT component helps to redirect excess interstitial fluid through lymphatic reconstruction. This greatly reduces the risk of recurrences even when small amounts of lymphedema tissue are left behind.

In conclusion, genital elephantiasis, particularly the male variant, is challenging to treat and often plagued with poor wound healing and recurrences of lymphedema. Many treatment choices can be challenging for the clinician to understand. However, employing a combination approach such as RRR is often the most effective way forward as it helps improve outcomes and quality of life through a one-stage operation.

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