Genital lymphoedema: pathology, reconstruction and outcomes

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Key words

Lymphoedema, genital reconstruction, scrotal lymphoedema, penile lymphoedema

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Abstract

Background: Penile and scrotal lymphoedema is a debilitating complication following lymph node dissection, radiation, or in the setting of obesity. Treatment is aimed at improving cosmesis and reducing urologic complications. **Aims:** To describe our experience with the surgical correction of peno-scrotal lymphoedema. **Methods:** A retrospective review of all patients who underwent surgical treatment for penoscrotal lymphoedema was performed. Comorbidities, technical approaches and functional outcomes were evaluated. **Results:** Twelve patients were included with an average age of 55.9 (16.6 – 74.4, standard deviation 19.4) years. Presenting symptoms included penile and scrotal oedema (seven patients), scrotal oedema (three), penile oedema (one), and suprapubic and scrotal oedema (one). Causes included obesity (four), radiation therapy (three), pelvic lymph node dissection (two) and unknown (two). Surgical procedures performed included penoplasty and scrotoplasty (eight), scrotoplasty (three), and penoplasty (one). Immediate complications developed in four patients and long-term complications in seven patients. Lymphoedema recurrence rate was seen in four (33.3%) patients out of 12, while three (25%) patients required reoperation. **Conclusion:** Our series describes a surgical technique with repeatedly favourable outcomes and highlights the necessity for multidisciplinary care to improve treatment outcomes.

Lymphoedema is characterised by the retention of lymphatic fluid in the subcutaneous compartment due to an obstruction of lymphatic flow (Schulte-Merker et al, 2011). The accumulation of this protein-rich fluid leads to dilation of lymphatic vessels, hypertrophy and hyperplasia of the connective tissue, and infiltration of fibroblasts and inflammatory cells, eventually resulting in skin thickening and chronic inflammation (Feins, 1980; Apesos and Anigian, 1991; Szuba and Rockson, 1997).

Lymphoedema can affect both males and female, and can occur in any part of the body. In males in particular, lymphoedema can affect the penis, the scrotum or the entire external genital region. The condition is generally the secondary result of surgery, radiation, obesity, infection, such as filariasis, or tumours that lead to damage of the draining lymphatics (Schulte-Merker et al, 2011; Singh et al, 2011). Regardless of its cause, penoscrotal lymphoedema can be an extremely debilitating condition that causes great discomfort and pain, as well as difficulties with voiding, potency, mobility, hygiene and an overall reduction in quality of life (Patel et al, 2015; Facio et al, 2017).

Although treatment initially involves conservative management using weight loss in cases of obesity, or lymphatic drainage and compression, once the irreversible state of tissue fibrosis has been reached, surgical debulking of the affected region must be considered. Given the relatively low prevalence of the condition, there is currently no standardised surgical approach for the treatment of penoscrotal lymphoedema. In the present study, the authors report our experience regarding the treatment of patients with this condition and describe long-term outcomes.

Methods

Data acquisition

The medical records of all patients who were surgically treated for penoscrotal lymphoedema between August 2010 and January 2017 at the authors' institution were reviewed. Patients were excluded if their lymphoedema was associated with chronic scarring as a result of suprapubic and perineal hidradenitis suppurativa or as a result of previous bariatric surgery.

Data were collected regarding patient demographics, presenting symptoms, cause of lymphoedema, peri- and postoperative management, and outcomes, including immediate and long-term complications and were then analysed using Microsoft Excel (Microsoft Office).

The study was conducted according to the Strengthening the Reporting of Observational Studies in Epidemiology (STROBE) statement (University of Bern, 2009).

Table 1. Patient information.

Total (n)	12
Average age (years)	55.9 (16.6–74.4, SD 19.4)
Presenting symptoms	
Penile oedema	1 (8.3%)
Scrotal oedema	3 (25%)
Suprapubic + scrotal oedema	1 (8.3%)
Penile + scrotal oedema	7 (58.3%)
Urinary symptoms	4 (33.3%)
Changes in stream	1 (8.3%)
Urgency/incontinence	1 (8.3%)
Difficulty urinating	1 (8.3%)
Unable to aim (lack of visualisation of penis)	1 (8.3%)
Leg oedema	5 (41.6%)
Sexual symptoms	0 (0%)
Cause of lymphoedema	
Obesity	4 (33.3%)
Prostate cancer (s/p prostatectomy + PLND)	2 (16.7%)
Radiation therapy (s/p rectal cancer/lymphoma)	3 (25%)
Lymph node removal	1 (8.3%)
Not specified/Unknown	2 (16.7%)

Surgical technique

Prior to surgery, a Foley catheter is placed in sterile fashion and, if visible, the penis is sewn to the catheter with a silk suture. Surgical planning should involve the urological and reconstructive surgeons. Usually, the surgeons together define the resection margins of the lymphoedema tissue, then the urology team begins dissection and identification of the chord structures and the penis. Often, this process involves complete exposure of the penis, and release of the suspensory ligament. The goal is complete exposure through a midline approach and debulking of the penis.

After complete resection of the oedematous tissue by the urology team, the plastic surgeons begin by placing the testes in the anatomic position in the inferior groin at the site of the future neoscrotum (*Figure 1a shows patient presenting with massive penoscrotal lymphoedema and Figure 1b shows patient after resection of lymphoedematous tissue*). The testes may be fixed in place to prevent torsion. An adipofascial flap of the remaining fat and fascia is elevated on the dorsal side of the scrotum based on the perineum and incorporated into the orchipexy to provide some remaining separation between them while providing protection.

The neoscrotum is then closed in a circular fashion as the anterior scrotal flap is rotated into position using interrupted sutures (*Figures 1c & 1d*). It is critical to preserve scrotal skin and utilise it in the neoscrotum as it is the best colour match. In the suprapubic region, skin is then elevated laterally on either side, in order to advance the tissue anteriorly and is closed in layers appropriately. Drains are placed in the neoscrotal sack and in the mons area. The base of the penis is reconstructed using local tissue rearrangement, allowing a wide orifice for the penile base to pass through, and is left in the appropriate position. A split-thickness skin graft is then harvested from the lateral thigh and is placed around the shaft of the penis. Care is given to preserve a cuff of tissue around the corona to allow for inset of the split-thickness graft around the phallus.

The penis is maintained out to length during the graft application to ensure maximum functional length and skin graft take. In order to do so, a silk suture is passed through the glans and tied to the Foley, which is then pulled to extend the penis to its maximum length. A foam dressing is circumferentially wrapped around the penile shaft beginning at the base and extending to the glans. Negative pressure wound therapy (NPWT) is then applied as a bolster for 3 days, maintaing the penis at length during wound healing. Examples of the final result can be seen in *Figures 2, 3 & 4*. The scrotal skin should not be used to reconstruct the penis, as it is functionally suboptimal and leads to high rates of wound complications. Given the anatomy of the scrotal skin, reactions to temperature and sympathetic/parasympathetic variations can lead to both aesthetic and functional issues if placed along the penis. Additionally, the pubic hair and thicker skin of the scrotum can result in a step-off deformity at the penile shaft, causing decreased sensation, as well as functionality. A split-thickness skin graft that completely encircles and covers the shaft of the penis should be taken from a thigh donor site using precise measurements.

Postoperatively, patients were hospitalised for between 2–3 days for surgical site monitoring and pain control. They were initially maintained on strict bed rest and were allowed to ambulate by the third postoperative day to prevent recurrence of oedema. No formal physical therapy was conducted.

Results

A total of 12 male patients were included in the study. Their average age was 55.9 (16.6–74.4, standard deviation [SD] 19.4) years and average body mass index (BMI) was 31.2 (21.8–43.7, SD 8.4). Seven (58.3%) of the authors' patients presented with penoscrotal lymphoedema, three (25%) with isolated scrotal lymphoedema, one (8.3%) with isolated penile lymphoedema, and one (8.3%) with suprapubic and scrotal lymphoedema. The diagnosis of lymphoedema was made based on clinical exam. Five (41.7%) patients had coexisting leg oedema. Four (33.3%) patients additionally presented with the following urinary symptoms: one (8.3%) changes in stream, one (8.3%) urgency and incontinence, one (8.3%) difficulty urinating, and one (8.3%) difficulty aiming due to inability to visualise the penis. None presented initially with sexual symptoms (*Table 1*).

The cause of lymphedema in these patients was obesity in four (33.3%), radiation therapy after rectal cancer or lymphoma in three (25%), prostate cancer resulting in prostatectomy and pelvic lymph node dissection (PLND) in two (16.7%), idiopathic in two (16.7%) and lymph node removal in one (8.3%) (*Table 1*). Of note, obesity was assumed to be the cause of lymphoedema after all other sources were excluded.

Eight (66.7%) patients received both a penoplasty and scrotoplasty, three (25%) received an isolated scrotoplasty and one (8.3%) received an isolated penoplasty. The following three adjunct procedures were performed in five (41.7%) patients: three (25%) panniculectomy, one (8.3%) lipectomy

and excision of a lesion, and one (8.3%) excision of a lesion. Nine (75%) patients also received a skin graft and six (50%) of those patients were treated using NPWT postoperatively. A Foley catheter was placed in all patients postoperatively for an average of 15.5 (5.0–60.0, SD 15.2) days (*Table 2*).

Immediate complications occurred in four (33.3%) of the patients and they were as follows: two (16.7%) abscess/cellulitis, one (8.3%) seroma and inability to ejaculate, and one (8.3%) wound breakdown. Long-term complications occurred in seven (58.3%) patients and they were as follows: two (16.7%) urinary strictures and retention, two (16.7%) recurrent cellulitis, one (8.3%) erectile dysfunction, one (8.3%) hypertrophic scarring and keloid, and one (8.3%) change in urinary stream. Four (33.3%) patients had partial recurrence of their lymphoedema and three (25%) received a second surgical resection. For these three patients, the average time between surgeries was 1.7 (0.4–2.3, SD 1.1) years. The average length of follow-up for our patients was 2.0 (0.1–6.4, SD 2.1) years (*Table 3*).

Table 2. Surgery details.

Surgical procedures		
	Penoplasty + scrotoplasty	8 (66.7%)
	Scrotoplasty	3 (25%)
	Penoplasty	1 (8.3%)
Skin graft		9 (75%)
Adjunct procedures		5 (41.7%)
	Panniculectomy	3 (25%)
	Lipectomy + excision of lesion	1 (8.3%)
	Excision of lesion	1 (8.3%)
Wound Vac		6 (50%)
Average time Foley in (days)		15.5 (5.0 – 60.0, SD 15.2)

Table 3. Complications and follow-up.

Immediate complications		4 (33.3%)
	Abscess/cellulitis	2 (16.7%)
	Seroma + inability to ejaculate	1 (8.3%)
	Wound breakdown	1 (8.3%)
Long-term complications		7 (58.3%)
	Urinary strictures/retention	2 (16.7%)
	Erectile dysfunction	1 (8.3%)
	Hypertrophic scarring/keloid	1 (8.3%)
	Recurrent infections/cellulitis	2 (16.7%)
	Change in urinary stream	1 (8.3%)
Recurrence		4 (33.3%)
Second resection needed		3 (25%)
Average time between first and second resection (years)		1.7 (0.4–2.3, SD 1.1)
Average Length of follow-up (years)		2.0 (0.1–6.3, SD 2.1)

Discussion

Surgical treatment of massive penoscrotal lymphoedema is generally accepted as the standard of care when the disease interferes with activities of daily living, although many different approaches have proven to be ineffective over the long term. Lymphovenous bypass, which uses vascular anastomoses to restore lymphatic drainage, can be used in mild cases with minimal stasis and no fibrosis, though recurrences are common (Sauer et al, 1998).

Lymphangiectomy, however, which involves the complete resection of all affected penile and scrotal tissue superficial to Buck's fascia, can successfully be completed (Apesos and Anigian, 1991; Modolin et al, 2006; Singh et al, 2011). A major challenge associated with this method is the final reconstruction of the penile skin, for which many surgical solutions have been reported (Vaught et al, 1975; Apesos and Anigian, 1991; Favarger et al, 1991; Dumanian and Futrell, 1996; Modolin et al, 2006; Singh et al, 2011).

A combined approach consisting of a team of urologists and plastic surgeons is critical for optimally addressing both the resection and the reconstructive goals of penoscrotal lymphoedema. All aspects of the procedure should be addressed with great attention to detail, in order to provide optimal results.

The genital region has a superficial lymphatic system, which drains the penile and scrotal skin, and a deep system, which drains the testes and penile body, ultimately flowing into the inguinal and pelvic lymph nodes (Dewire and Lepor, 1992). While obstruction of the superficial chain can result in lymphoedema of the penis and scrotum, the deep system usually remains unaffected, allowing for the success of lymphangiectomy (Modolin et al, 2006). However, it must be noted that surgical resection of the oedematous tissue is not a permanent cure, as recurrence rates of up to 50% have been reported (Machol et al, 2014). Recurrent lymphoedema occurred in four (33.3%) patients, resulting in reoperation in three. The three patients that required reoperation had an average follow-up time of 4.5 years. Thus, it is possible that with longer follow-up, severe recurrence will be seen in the remaining patients as well. The overall immediate complication rate was 33.3% and all were related to wound healing.

Surprisingly, however, none of these occurred in patients whose lymphoedema was a result of obesity, and all resolved with conservative management and local wound care within 1 month of surgery. Although the long-term complication rate in this study was 58.3%, these were largely issues that were tolerable by patients and did not require operative care.

One of the limitations of this study lies in its retrospective nature, as detailed descriptions of symptoms and complications could not consistently be assessed. Another limitation lies in the small sample size, which is attributable to the low incidence of this pathology, as well as the subjectivity of the evaluation of outcomes. Unfortunately, this source of bias could not be addressed in retrospect. Despite these downfalls, this case series describes a surgical technique with repeatedly favourable outcomes for a challenging disease process. Unfortunately, due to the rare nature of the disease and the limited number of publications in the literature describing its surgical repair, the authors' results could not be adequately compared with those of

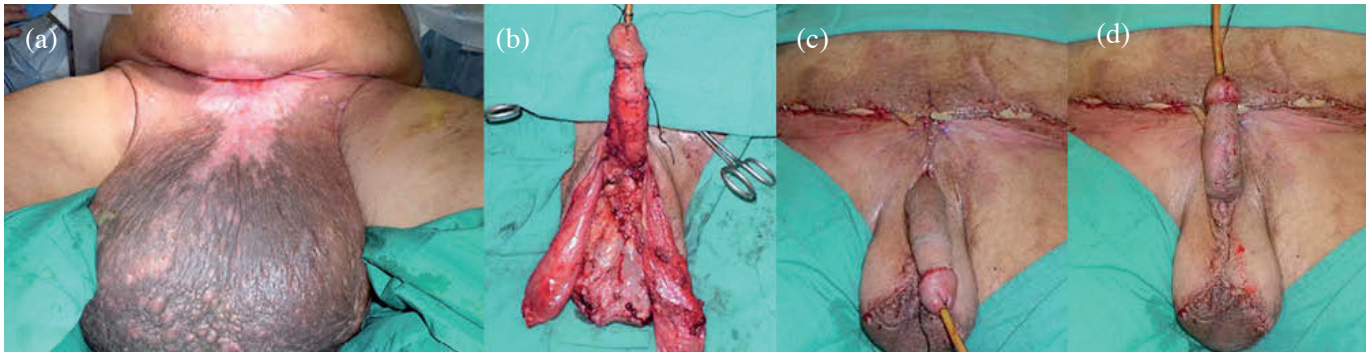

Figure 1. Patient with massive penoscrotal lymphedema (a); after resection of lymphedematous tissue (b); dorsal view postoperatively (c); ventral view postoperatively (d).

Figure 2. Patient with isolated penile lymphedema (a); postoperatively after resection of lymphoedematous tissue and reconstruction (b).

Figure 3. Penoscrotal lymphedema (a); after resection and reconstruction (b).

Figure 4. Massive penoscrotal lymphedema before reconstruction (a); after resection of lymphoedematous tissue (b).

other studies for confirmation. Although newer techniques involving lymph node transfer and lymphatic bypass may be implemented in the future, these are currently not considered a first-line therapy. However, the role of this type of supermicrosurgical intervention is being evaluated at our institution. The authors believe that the team-based approach outlined in this study can be applied by providers at other institutions for the successful treatment of penoscrotal lymphoedema.

References

Apesos J, Anigian G (1991) Reconstruction of penile and scrotal lymphedema. *Ann Plast Surg* 27(6): 570–3

Dewire D, Lepor H (1992) Anatomic considerations of the penis and its lymphatic drainage. *Urol Clin North Am* 19(2): 211–9

Dumanian GA, Futrell JW (1996) The Charles procedure: misquoted and misunderstood since 1950. *Plast Reconstr Surg* 98(7): 1258–63

Facio MF, Spessoto LC, Gatti M et al (2017) Clinical treatment of penile fibrosis after penoscrotal lymphedema. *Urol Case Rep* 11: 14–6

Favarger N, Rist M, Krupp S (1991) [Cutaneous reconstruction of external genital organs: an older method still in current use]. *Helv Chir Acta* 58(3): 301–3 [Article in French]

Feins NR (1980) A new surgical technique for lymphedema of the penis and scrotum. *J Pediatr Surg* 15(6): 787–9

Machol JA 4th, Langenstroer P, Sanger JR (2014) Surgical reduction of scrotal massive localized lymphedema (MLL) in obesity. *J Plast Reconstr Aesthet Surg* 67(12): 1719–25

Modolin M, Mitre AI, da Silva JC et al (2006) Surgical treatment of lymphedema of the penis and scrotum. *Clinics (Sao Paulo)* 61(4): 289–94

Patel KM, Lin CY, Cheng MH (2015) A prospective evaluation of lymphedema-specific quality-of-life outcomes following vascularized lymph node transfer. *Ann Surg Oncol* 22(7): 2424–30

Sauer PF, Bueschen AJ, Vasconez LO (1998) Lymphedema of the penis and scrotum. *Clin Plast Surg* 15(3): 507–11

Schulte-Merker S, Sabine A, Petrova TV (2011) Lymphatic vascular morphogenesis in development, physiology, and disease. *J Cell Biol* 193(4): 607–18

Singh V, Sinha RJ, Sankhwar SN, Kumar V (2011) Reconstructive surgery for penoscrotal filarial lymphedema: A decade of experience and follow-up. *Urology* 77(5): 1228–31

University of Bern (2009) *STROBE Statement Strengthening the Reporting of Observational Studies in Epidemiology*. Available at: https://bit.ly/2MDZELI (accessed 10.06.2019).

Szuba A, Rockson SG (1997) Lymphedema: anatomy, physiology and pathogenesis. *Vasc Med* 2(4): 321–6

Vaught SK, Litvak AS, McRoberts JW (1975) The surgical management of scrotal and penile lymphedema. *J Urol* 113(2): 204–6