Surgical reduction of scrotal massive localized lymphedema (MLL) in obesity

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KEYWORDS
Scrotal; Massive localized lymphedema; MLL; Obesity; Scrotoplasty

Summary
Background: Lymphedema of the penis and scrotum is physically and psychologically disabling. Obesity is a source of secondary lymphedema. When restricted to specific anatomical regions in obesity, this is termed massive localized lymphedema (MLL). Few surgical cases of specific scrotal MLL in obesity are reported in the literature. We present our case series to improve the management of this complicated pathology.

Methods: This is a retrospective review of obese adult patients with clinically diagnosed scrotal MLL undergoing reduction scrotoplasty by the senior author (J.R.S.) from 1992 to 2012. Medical, social, familial, surgical, and follow-up data were extracted. Prior infection of the scrotal lymphedema, surgical details, pathologic evaluation, and postoperative complications were noted. A series of the cases is presented.

Results: Four cases met the criteria for study. The average age was 35 years with an average body mass index of 53.9. Average resection at the first procedure was 3492 g. All patients were reconstructed with laterally based scrotal flaps. The pathology for each case was consistent with chronic lymphedema; no sarcomatous changes were noted. Fifty percent of the patients had recurrence of the scrotal MLL. The average total number of operations during the follow-up period for either complication or recurrence was two.

Conclusions: This is the largest case series specifically investigating surgical treatment for scrotal MLL in obesity. Lateral-based scrotal flaps (with or without mid-raphe Z-plasty) permit anatomic reconstruction. Complications are common and recurrence is frequent after surgical management. Excision with reconstruction improves urinary function and overall symptoms.

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Introduction

Lymphedema of the penis and scrotum is often physically and psychologically disabling. Painful ulcerations and infection, as well as urinary and sexual dysfunction are common sequelae. This pathology is often categorized as either congenital (primary) or acquired (secondary) lymphedema. Primary lymphedema is attributable to hypoplasia of the lymphatic system, whereas secondary lymphedema generally results from lymph obstruction. Filariasis is the most common secondary etiology worldwide, but cases of genital lymphedema secondary to radiation therapy and lymph node dissections are becoming more frequent.

Obesity is also a potential source for idiopathic secondary lymphedema. Farshid and Weiss first described massive localized lymphedema (MLL) as an enlarging lesion in the obese population due to chronic lymph obstruction. They note the most common location for this lesion is the lower extremity, followed by the abdomen/suprapubic region; the scrotum comprised only 4% of the cases. MLL has been described as a “pseudosarcoma” due to its large size, proliferation, and clinical mimicry of a sarcoma. It often presents with peau d’orange skin changes, giant swelling, and sarcoma-like inflammation; however, only anecdotal reports of MLL progressing to angiosarcoma exist.

Affected patients characteristically present for treatment due to difficulty with ambulation, impairment of activities of daily living, and excoriation. This specific lesion is described only in patients with chronic obesity. As the obesity epidemic worsens, MLL will likely increase in presentation for surgical intervention.

The nonsurgical treatment of lymphedema is typically dependent on its origin and involves elevation, physical therapy, and treatment of the underlying condition; however, this is successful only in limited disease.

Many variations in surgical interventions exist for chronic scrotal lymphedema. These methods are best separated into two categories: neo-lymphatic drainage procedures and ablation with reconstruction. Lymphovenous microanastomoses are theoretically promising yet technically complex. Application to scrotal lymphedema is not yet defined and further studies are likely required before widespread application.

The Charles procedure is historically synonymous with massive scrotal lymphedema resection and reconstruction. This procedure utilizes healthy thigh tissue for testicle coverage after excision of lymphedematous tissues. Vaught et al. advocate utilization of the posterolateral skin of the scrotum to cover the testes after excision for a natural reconstruction. Halperin et al. favor combining the anterior scrotal and posterior perineal flaps for coverage. Otsuki et al. provide a general review of an assortment of techniques since the inception of the Charles procedure. Skin grafting may also be used, but this is mostly reserved for cases of penile shaft involvement, as reduction scrotoplasty incisions typically close primarily or with local flaps.

Dandapat et al. report the largest case series of surgical treatment of infectious genital elephantiasis. They favor isolating the spermatic cord and testes through two small lateral incisions. The lymphedematous tissue is excised and the skin of the scrotal neck accommodates the testes.

Our group prefers to use lateralized scrotal skin for coverage similar to the methods described by Charles, Vaught et al. and Kumar and Navaneethan. This skin has been compressed between the medial thigh and scrotal contents and, as a result, it is soft and pliable with less edema. There is ample tissue to allow for a primary midline closure with a suitable scrotal contour once the MLL is excised. Penile skin grafting is utilized as needed.

We apply these principles to surgically treat MLL of the scrotum and present a case series to better aid in the management of these challenging patients.

Methods

Approval from the institutional review board was obtained. A single-institution, retrospective review of patients >18 years of age undergoing scrotoplasty for chronic scrotal lymphedema was performed. Patients over a 20-year period were included, 1992–2012. These patients were identified using Current Procedural Terminology Codes (55175, 55180, 14040, 14041, and/or 14300). ICD-9-CM 608.86 “Edema of male genital organs” and clinic records from the senior surgeon, J.R.S., were also used. Patients undergoing scrotoplasty treated for trauma or acute infection management (i.e., Fournier’s gangrene) were excluded.

The charts meeting the aforementioned criteria were reviewed. Height and weight at presentation were then used to calculate the body mass index (BMI). Those not meeting the Centers for Disease Control definition of obesity (BMI ≥ 30) were excluded from review. Each patient included had the preoperative diagnosis of “secondary scrotal edema due to obesity” and clinical symptoms of MLL.

Information such as follow-up, medical, surgical, social, and family histories was extracted. Prior infection of the scrotal lymphedema was noted. Surgical details, pathologic evaluation, and postoperative complications (including number of operations required for treatment) were reviewed.

Results and patients

Four cases met the required study criteria (Tables 1 and 2). The average age was 35 years. All patients were Caucasian. All patients presented with standard MLL clinical presentations (giant swelling, inflammation, cracks or pits, infection, and lymph weeping). Fifty percent of the patients had prior surgical intervention in the groin or suprapubic region, and 75% of the subjects were previously diagnosed with buried penis syndrome. All patients had a history of lower-extremity edema and prior infection of their scrotal tissue (all cellulitis). One patient was on chronic prophylactic antibiotics. The average BMI was 53.9, and no patients were current tobacco users.

Surgical details are provided for each case (Cases 1–4). All cases were completed in conjunction with urology. A Foley catheter was placed for each patient and maintained.
until the patient was able to void without compromising surgical wounds. A midline orchiopexy was performed in each case to prevent torsion. The average estimated blood loss per primary surgery was 200 cc. Two pubic drains were placed in each patient. These were maintained until output was <20 cc per day. Surgical pathology was consistent with chronic lymphedema in each patient. No malignancy such as angiosarcoma or liposarcoma was noted. The average tissue removed at the first procedure was 3492 g. The average follow-up was 32.5 months (range 4–59 months). The patient in Case 3 died of metastatic seminoma 8 years after the initial surgery; he was discharged from scrotoplasty follow-up at 57 months.

There were surgical complications in each patient. The average number of operative procedures per patient (over the entire follow-up period) was two. Two of the patients required revision for recurrence; the MLL recurred 2 and 5 years from the initial intervention (Cases 3 and 4). All patients experienced wound-healing complications. Cases 1, 3, and 4 demonstrated superficial breakdown that was treated with dressing changes. Case 2 developed infection and posterior wound dehiscence occurred on postoperative day 7.

All patients went on to heal their wounds. Only Case 3 required prolonged catheterization due to urinary retention. No chart details specifically addressed pre- or post-operative sexual function.

**Case presentations**

**Case 1**

Case 1 involved a 49-year-old male with scrotal MLL (BMI of 54.7). The patient presented with chief complaints of buried penis and difficulty with urination, peau d’orange skin changes, and infection, all symptoms consistent with MLL (Figure 1A). Preoperative ultrasound imaging was obtained to confirm the location of the testes. Operatively, a traction suture placed in the glans (along with a Foley catheter) allowed for exposure of the penis. The scrotum was then opened in the midline, and the testes were isolated (Figure 1B). The medial lymphedematous tissue was excised (3000 g), orchiopexy was performed, and the lateral skin flaps were closed in the midline. The patient had superficial wound breakdown that healed with dressing changes. On follow-up at 10 months, all wounds were healed (Figure 1C).

**Case 2**

Case 2 involved a 42-year-old male with MLL of the scrotum (BMI of 84.5). He presented with a buried penis, difficulty with urination, multiple infections, and ulceration (Figure 2A). After isolation of the penis, a midline scrotal incision was used to isolate the testes, 4535 g of scrotal tissue was removed, and the lateral skin flaps were closed in a Z-plasty lengthening fashion (Figure 2B). On postoperative day 7, he developed malodorous discharge and the posterior aspect of the wound dehisced (Figure 2C). Washout, debridement, and reclosure with the use of incisional negative pressure wound therapy was
Table 2  Patient surgical and postoperative data.

<table>
<thead>
<tr>
<th>Subject</th>
<th>Surgical details</th>
<th>EBL (cc)</th>
<th>Foley</th>
<th>Tissue excised (g)</th>
<th>Drains</th>
<th>Pathology</th>
<th>Complications</th>
<th>OR</th>
<th>Follow-up (mo)</th>
<th>Outcome</th>
<th>Sexual</th>
<th>GU Function</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>Midline excision, primary closure with orchiopexy, V-Y at penile base</td>
<td>200</td>
<td>Yes</td>
<td>3000</td>
<td>2 JP</td>
<td>“Lymphedemous tissue”</td>
<td>4 cm × 2 cm posterior open wound, superficial</td>
<td>1</td>
<td>10</td>
<td>Healed</td>
<td>NR</td>
<td>Urination improved</td>
</tr>
<tr>
<td>2</td>
<td>Midline excision, orchiopexy, lateral scrotal skin flaps with Z-plasty mid-raphe</td>
<td>300</td>
<td>Yes</td>
<td>4535</td>
<td>2 JP</td>
<td>“Skin with dermal mild interstitial edema and dilated lymphatics consistent with lymphedema”</td>
<td>32 cm dehiscence on POD#7, I&amp;D and reclosure, incisional VAC Culture: Enterococcus, resistant Proteus and Pseudomonas Superficial wound infection, resolved with IV/PO antibiotics 5 years later: recurrence, revision reduction penile/scrotoplasty</td>
<td>2</td>
<td>4</td>
<td>Healed</td>
<td>NR</td>
<td>Foley for urinary retention (resolved)</td>
</tr>
<tr>
<td>3</td>
<td>Midline excision, orchiopexy, primary closure at midline using lateral flaps, STSG to penis after degloving</td>
<td>200</td>
<td>Yes</td>
<td>735</td>
<td>2 JP</td>
<td>“Dilated lymphatic channels, consistent with lymphedema”</td>
<td></td>
<td>3</td>
<td>57</td>
<td>Healed</td>
<td>NR</td>
<td>Urination improved</td>
</tr>
<tr>
<td>4</td>
<td>Midline excision, orchiopexy, penile shaft lengthening, closure with posteriolateral scrotal skin flaps, hydrocele excision</td>
<td>300</td>
<td>Yes</td>
<td>5700</td>
<td>2 JP</td>
<td>“Scrotal lymphedema” (Gross only)</td>
<td>4 cm × 1 mm × 2 mm separation 8/2007: recurrence, revision reduction scrotoplasty with Z-plasty</td>
<td>2</td>
<td>59</td>
<td>Healed</td>
<td>NR</td>
<td>Urination improved</td>
</tr>
</tbody>
</table>

(EBL = Estimated Blood Loss; JP = Jackson–Pratt; OR = Operating Room procedures. All patients used Foley catheter. All were healed at time of listed follow-up. Sexual function was not recorded in health record).
completed (Figure 2D). Cultures grew multidrug-resistant *Proteus*, *Enterococcus* species, and *Pseudomonas* species. Follow-up at 4 months revealed healed wounds; continued catheterization for functional urinary retention was required.

### Case 3

Case 3 involved a 24-year-old male with MLL of the scrotum and the penis (BMI 38.7). The patient presented with a warty, pitting/cracking scrotum and penis. He experienced...
lymph weeping from the woody tissue and chronic bouts of cellulitis. Two prior reduction scrotoplasty procedures had been performed elsewhere. He underwent a standard mid-scrotal incision to isolate the testes. The excess scrotal tissue was excised (735 g), leaving two lateral scrotal skin flaps that were closed, creating a new midline raphe. Two years later, he re-presented with recurrent scrotal and penile MLL (Figure 3A). Again, the standard midline excision with a lateral scrotal flap closure was performed. The excision of penile lymph tissue was also performed, followed by skin grafting (Figure 3B). Unfortunately, he had another recurrence as well as the development of a large lymphocele. He was treated in a similar fashion as before, except the midline incision was carried into the left suprapubic region to achieve adequate soft tissue excision after excision of the lymphocele. At 1-month follow-up, all wounds were healed (Figure 3C).

Case 4
Case 4 involved a 25-year-old male with scrotal MLL (BMI 37.7). He presented with a massive scrotum and buried penis. The patient complained of chronic infection, weeping, and peau d’orange skin changes (Figure 4A). A midline scrotal approach was utilized to isolate the testes per our routine. The excess midline tissue was removed (5700 g). This was closed taking the incision to the right suprapubic to ensure proper tissue excision. A vertical incision cranial to the penis was used to isolate and treat the buried penis (Figure 4B). Five years later, the condition recurred (Figure 4C). This was addressed in a similar fashion as previously (Figure 4D). He went on to heal without issue.

Discussion
MLL specifically of the scrotum is relatively rare, but incidence will likely increase with increasing obesity rates. This diagnosis is clinical and may be confirmed with pathology. Each of our patients presented with chronic infections, thickening, cracking, knobbing, and pitting of the overlying skin. Most displayed the typical peau d’orange appearance due to obstruction of dermal lymphatics. Although the combination of these findings produces a clinical suspicion of malignancy, none of our subjects demonstrated malignant changes. All patients included were diagnosed with chronic Stage 3 lymphedema. No pathologic findings of cystic changes or absent lymphatic systems were noted, thus further confirming secondary lymphedema.

Surgery is indicated when the disease interferes with activities of daily living or when certain conditions, such as recurrent infection, ulceration, or questionable malignancy, are present. We noted that the majority of our patients had buried penis syndrome secondary to the scrotal MLL and obesity. Resection of the MLL improved urination in this series.

A mean blood loss of 200 cc was reasonable, even with an average of approximately 3.5 kg of tissue excised per case. The use of lateral scrotal flaps, with optional mid-raphe Z-plasty as needed, results in an anatomic reconstruction. Skin grafting to the penile shaft after resection for penile involvement is a useful reconstructive option, as dorsal skin flaps are often too fibrotic. We strongly recommend the use of surgical drains given the lymphedematous nature of the tissue.

The average number of surgical procedures requiring an operating room was two. Complications, namely wound infection or breakdown, after scrotoplasty for MLL are common. The universal risk factors are obesity, wound location, and lymphedematous tissue. Wu et al. report similar complication rates to ours. Martinez et al. recommend continuing postoperative antibiotic treatment to decrease surgical site infection. All patients in our series eventually went on to heal their surgical sites.

One must keep in mind that surgical resection is often palliative. Half the patients returned for further surgical treatment. These subjects recurred 2–5 years after their initial scrotoplasty. The remaining patients have followed up for an average of 7 months; it is probable that they will recur as well. The use of adjunct lymphatic drainage procedures combined with reduction scrotoplasty may reduce the high recurrence rates. Furthermore, all patients did remain obese throughout the study period. Successful obesity treatment (medical or surgical) in this patient population will likely reduce recurrence as well.

The limitations of this study include number of patients, lack of sexual function appraisal, and no evaluation of long-term infection status. As reflected in prior studies, MLL of the scrotum is rare, though reports of surgical treatment for MLL are increasing. Lee et al. have the largest clinicopathologic series, yet this study does not contain specific surgical details. To our knowledge, only six cases of surgical treatment for scrotal MLL in obesity, prior to our series of four patients, have been published. The majority of these studies do not include long-term follow-up as this investigation does, nor do they discuss complications and surgical elements. Evaluation of sexual function (i.e., ability to achieve erection) would provide insight as to quality-of-life improvement. Long-term postoperative infection status would evaluate the effectiveness of reduction scrotoplasty as a successful procedure for infection control in scrotal MLL.

Conclusions
MLL of the scrotum is frequently disabling. Surgical intervention is indicated when activities of daily living are affected, for infection or ulceration, or for possible malignant changes. Lateral-based scrotal flaps (with or without a mid-raphe Z-plasty) permit anatomic reconstruction. Complications are common and recurrence is frequent after surgical management. Nevertheless, excision with reconstruction improves urinary function and relieves overall symptoms.

Ethical approval
This article is a retrospective study of patients who received treatment according to recommended standards for care at the Froedtert Memorial Lutheran Hospital, Milwaukee, WI, a fully accredited care facility.
Conflicts of interest

None.

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