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

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KEYWORDS
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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.[1,2] Primary lymphedema is attributable to hypoplasia of the lymphatic system, whereas secondary lymphedema generally results from lymph obstruction.[2] 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.[1,3]

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.[4] Brewer and Singh present a review of this pathology as well as a case report.[5] 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.[6] 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.[5,7,8] Afflicted patients characteristically present for treatment due to difficulty with ambulation, impairment of activities of daily living, and excoriation.[6] 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.[2]

Many variations in surgical interventions exist for chronic scrotal lymphedema.[9–18] These methods are best separated into two categories: neo-lymphatic drainage procedures and ablation with reconstruction.[19] 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.[20–24]

The Charles procedure is historically synonymous with massive scrotal lymphedema resection and reconstruction.[25,26] 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.[27] Halperin et al. favor combining the anterior scrotal and posterior perineal flaps for coverage.[28] Otsuki et al. provide a general review of an assortment of techniques since the inception of the Charles procedure.[19] 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.[29–31]

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.[15]

Our group prefers to use lateralized scrotal skin for coverage similar to the methods described by Charles, Vaught et al. and Kumar and Navaneethan.[26,27,32] 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.[2]

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).[33] Those not meeting the Centers for Disease Control definition of obesity (BMI $\geq$ 30) were excluded from review.[34] 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).[5] 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