Isolated penile lymphedema: a case report and review of the literature

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Summary

Isolated penile lymphedema is a relatively rare condition that can have a profound impact on a patient’s quality of life. Herein, we present an 18-year-old man with a two-year history of isolated penile edema that developed two weeks prior to an appendectomy for acute appendicitis. The edema began acutely, gradually worsened, and had not remitted since its onset. Physical examination revealed a grossly edematous penis with scattered ill-defined mildly erythematous patches sparing the skin folds. A cutaneous biopsy demonstrated a proliferation of dilated lymphatic vessels, revealing a diagnosis of penile lymphedema. Treatment with pimecrolimus cream has resulted in improvement of the patient’s penile erythematous patches. He is currently seeking care with a reconstructive urologist for possible lymphangiectomy.

KEY WORDS: penile; lymphedema.

Case report

An 18-year-old man with a previous history of orchitis presented with a 2-year history of persistent penile edema that had not resolved since its onset. The edema developed two weeks prior to an appendectomy for acute appendicitis. It began acutely and gradually worsened. The patient did not recall any precipitating factors. The edema was more severe in the morning, often causing mild pain with micturition. He denied a history of penile discharge or dermatoses, erectile pain, trauma, sexually transmitted infections, and weight gain. Two lymph node biopsies, previously performed by urology, did not demonstrate granuloma, necrosis, or malignancy. A recent colonoscopy was within normal limits.

Physical examination revealed edema extending from the penile base to the urethral meatus (Figure 1). Additionally, there were scattered ill-defined mildly erythematous patches with sparing of the skin folds. The mons pubis, crural and inguinal folds, scrotal sac, and the skin of the perineum were within normal limits. A punch biopsy from the penile shaft demonstrated a proliferation of dilated lymphatic vessels, which were highlighted by a D2-40 stain, and a slight increase in interstitial edema (Figure 2). A complete blood count, metabolic panel, urinalysis, and urinary culture were within normal limits. A computed tomography scan of the abdomen and pelvis did not demonstrate any abnormalities.

These results were suggestive of a diagnosis of idiopathic penile lymphedema. Treatment with pimecrolimus cream has resulted in improvement of the erythematous penile patches. He is currently seeking care with a reconstructive urologist for possible lymphangiectomy.

Comment

We are highlighting this case to review the clinical evaluation and differential diagnosis of isolated penile lymphedema. Lymphedema of the male genitalia is caused by inadequate drainage of interstitial fluid by lymphatic vessels, with subsequent enlargement of the penis. The scrotum is also commonly affected (1). Localized penile edema is characterized by severe discomfort, poor local hygiene, limited ambulation, increased risk of developing phimosis, and a progressive loss of sexual and urinary function (1-5). Hence, many patients are deeply impacted by the psychological implications of this condition (5). Given that isolated penile lymphedema is relatively rare, literature describing this entity and its management are limited. Penile lymphedema has numerous etiologies. Imaging of the lymphatic vessels may help to delineate between primary and secondary lymphedema (1). Primary lymphedema, due to hypoplastic lymphatic channels, is relatively uncommon. It may be: 1) con-
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Genital or familial (Milroy’s disease), 2) idiopathic, appearing between puberty and age 35, targeting the extremities (praeox or Meige disease), or 3) late-onset occurring after 35 years of age (tarda) (1-3). Secondary lymphedema is caused by acquired obstruction of lymphatic flow due to chronic or persistent lymphatic vessel irritation and scarring (1-3). The resultant lymphedema is attributed to destruction of the lymphatic network. Infectious causes are most commonly reported in the literature. Sexually transmitted infections such as syphilis, chlamydia, gonorrhea, herpes, lymphogranuloma venereum, and donovanosis have been reported in association with isolated penile edema (1, 3, 6). Other reported infectious causes include filariasis, coagulase positive staphylococci, hemolytic streptococci, scabies, and tuberculosis (1-4, 7, 8). Non-infectious causes include inflammatory dermatoses, trauma, obesity, thyroid disease, chronic venous insufficiency, malignant conditions, and sequelae from surgical removal of lymph nodes or radiation fibrosis (1-5). Cases of isolated penile edema with genital dermatoses have been associated

Figure 1 - Clinical image demonstrating edema of the penis extending from the penile base to the urethral meatus.

Figure 2 - Histopathology of biopsied lesion, 10x magnification, hematoxylin/eosin stain. There is a proliferation of dilated lymphatic vessels, which are highlighted by a D2-40 stain, and a slight increase in interstitial edema.
with lichen planus, Proteus syndrome, psoriasis, hidradenitis suppurativa, and metastatic Crohn’s disease (1, 9-12). Traumatic causes of localized penile lymphedema have been reported with circumcision, compulsory masturbation, and metal ring jewelry (13-16). Other causes include amyloidosis, sarcoidosis, and malignancy (11).

Management of isolated penile lymphedema is challenging. Medical treatments include the use of oral antibiotics for identified infectious pathogens, empirical antibiotics for presumed subacute genital infections, oral steroids, and topical steroid application limited to areas with cutaneous lesions (1-4). Antibiotics may also be utilized for prophylaxis in patients with recurrent infections. Multiple case series have reported partial responses to long-term antibiotic use with erythromycin, ciprofloxacin, minocycline, or combination trimethoprim and rifampin (1-4, 6-8). If patients experience an acute exacerbation of their chronic penile lymphedema, they may benefit from a short course of prednisolone (1).

Procedural intervention may be a promising alternative to medical therapy. One case report suggested that low level laser therapy (LLLT) with low intensity wavelengths between 650-1000 nanometers, used in a scanning or spot laser form, may be used to treat isolated penile edema (17-19). A 15-year-old girl with right lower extremity lymphedema praecox was reported to have decreased circumference of the affected limb after laser therapy. The Authors believed that LLLT encourages the formation of new lymphatic vessels, thereby promoting lymphatic flow (20).

Multiple surgical treatments have also been reported in the literature (21). There are two main types of surgical interventions: lymphangiolipoplasty and lymphangiectomy. In lymphangiolipoplasty, lymphatic vessels from the affected region are anastomosed with unaffected vessels of a nearby uninvolved region. In lymphangiectomy, the superficial lymphatic network is removed (22). This surgical technique may include lymphangioplasty depending on the severity and extent of involved lymphatic tissue.

In summary, isolated penile lymphedema is a relatively rare condition that can have a profound impact on the quality of life of a patient. A comprehensive clinical evaluation is important so appropriate treatment can be initiated.

Disclosure

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