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*Net Letter*

# Saxophone penis with penoscrotal lymphangiomatosis secondary to lymphedema tarda

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**Quick Response Code:**

Dear Editor,

A 63-year-old male presented to our dermatology outpatient department with 8-month history of a giant verrucous cauliflower-like lesion over the penoscrotal region, which started over the

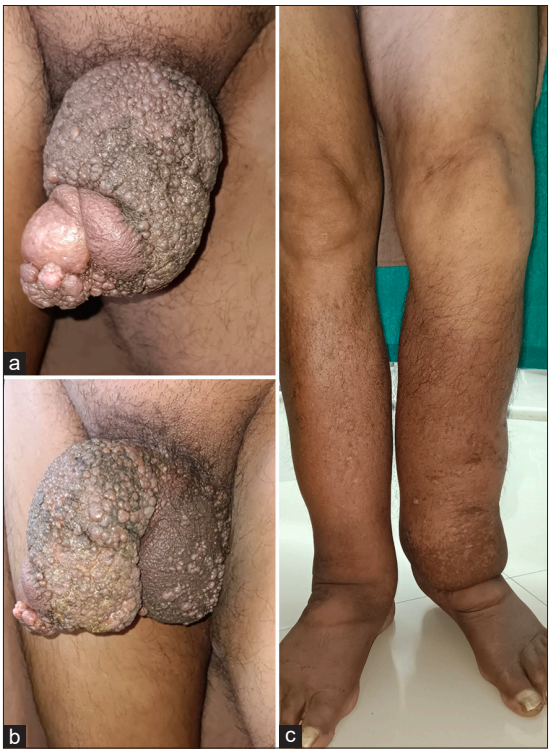


**Figure 1 (a):** A cauliflower-like swelling measuring approximately 8 × 4 × 2 cm present over the glans penis and extending to the whole shaft and part of the scrotum; (b): There are also multiple discrete verrucous papules over the scrotum and glans penis, which are of variable size and partially obstructed urethral opening; (c): An ill-defined diffuse swelling is present over both lower limbs and marked thickening of the skin.

glans penis and later progressed to involve the shaft of the penis and, lastly, the scrotum. There was associated swelling over both lower limbs, more on the left limb, for the past 12 months. There was no history of travel to a filarial endemic region, any urinary complaint, multiple sexual partners, trauma, medications, or condom catheter use.

On examination, we found a cauliflower-like swelling measuring approximately 8 × 4 × 2 cm present over the glans penis and extending to the whole shaft and part of the scrotum. There were also multiple discrete verrucous papules over the scrotum and glans penis of variable size and partially obstructing urethral opening [Figure 1a and b]. Both testes were palpable and of normal size and consistency, and no features of varicosity were present. Inguinal lymph nodes could not be palpated. An ill-defined diffuse swelling was present over both lower limbs, likely edema, which was pitting in nature, and marked thickening of skin was also noted [Figure 1c]. On clinical evaluations, differentials included a saxophone penis with penoscrotal lymphangiomatosis secondary to lymphedema, giant condylomas or Buschke-Lowenstein tumors, and verrucous squamous carcinoma.

Baseline tests, including filarial antigen testing, were normal. Ultrasonography of inguinoscrotal regions were normal. There was subcutaneous fluid along fat planes in bilateral scrotal walls and penile shaft. Right superficial inguinal nodes were enlarged, with the largest measuring approximately 5.5 mm with maintained hilum. Contrast-enhanced computerized tomography abdomen pelvis and penoscrotal region was suggestive of diffuse subcutaneous soft tissue edema in the scrotum, penile shaft, and pubic region. Associated mild subcutaneous collections in the scrotum and penile shaft are likely suggestive of cellulitis [Figure 2]. Magnetic resonance lymphangiography was suggestive of distal lymphatic obstruction [Figure 3]. Histopathological examination revealed parakeratosis with basal hyperpigmentation. Underlying demis showed loosely fibro collagenous stroma with abundant vessels [Figure 4a and b]. The patient was referred to the surgery department for a wide local excision and was lost to follow-up.

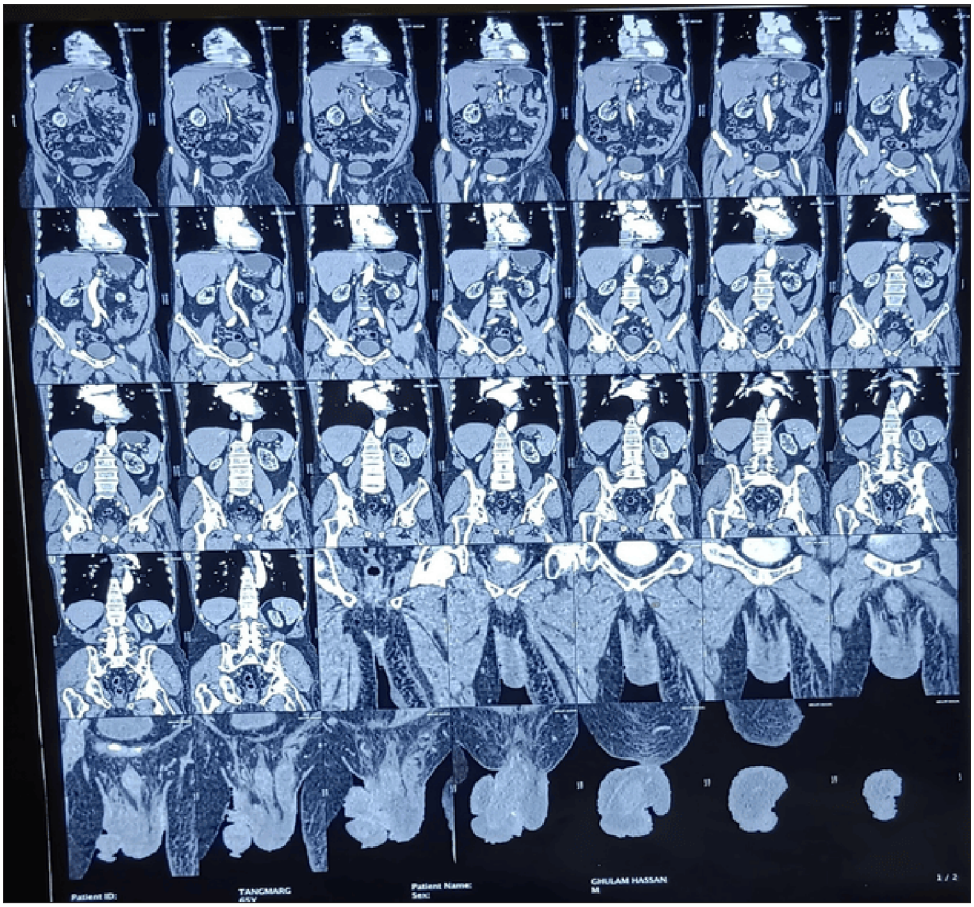


**Figure 2:** Contrast-enhanced computerized tomography abdomen pelvis a penoscrotal region suggestive of diffuse subcutaneous soft tissue edema in the scrotum, penile shaft, and pubic region. Associated mild subcutaneous collections in the scrotum and penile shaft are likely suggestive of cellulitis.

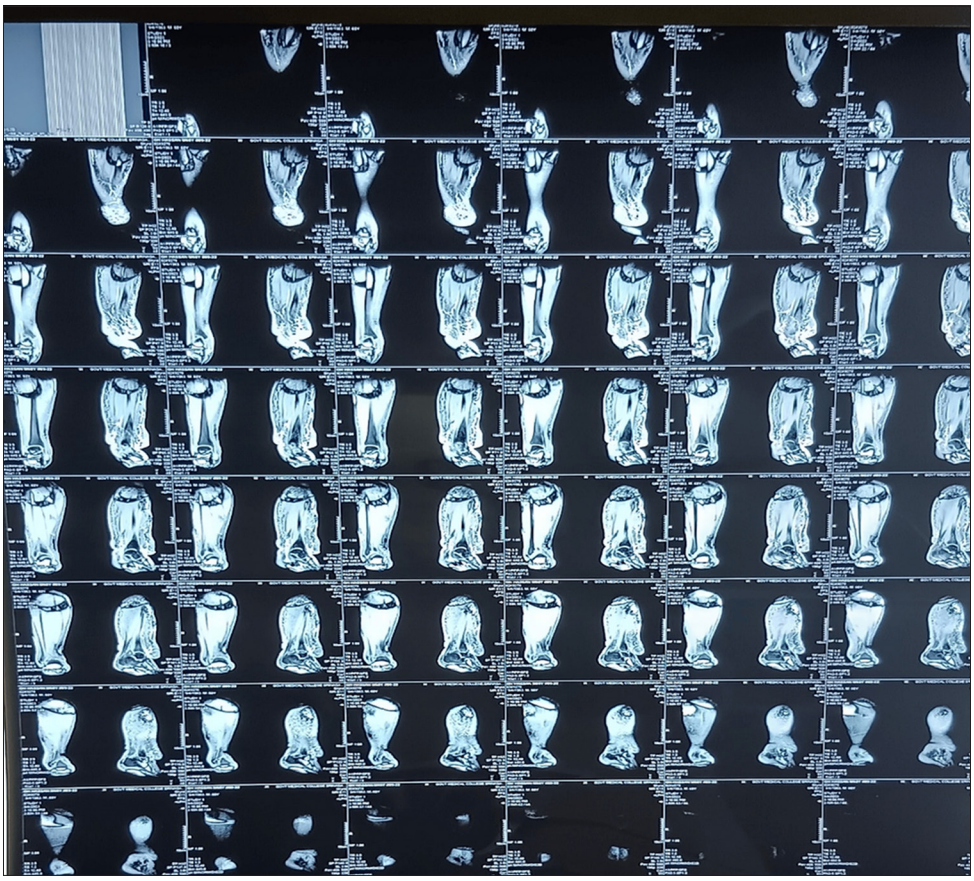

**Figure 3:** Magnetic resonance lymphangiography revealing an anteromedial and posterolateral group of lymphatic channels that are dilated, torturous with a beaded appearance and showing a reticular appearance suggestive of collateral formation. The maximum caliber of the lymphatic channel is 4.9 mm at the lower medial para-tibial location, approximately 4 cm superior to the tibiotalar joint along great saphenous vein territory, and 4.3 mm at the lower lateral para-tibial location, approximately 7.5 cm superior to the tibiotalar joint, along small saphenous vein territory. Diffuse subcutaneous edema is noted. After 1 h of contrast, administration popliteal and inguinal lymph nodes show no contrast opacification, likely suggestive of distal lymphatic obstruction.

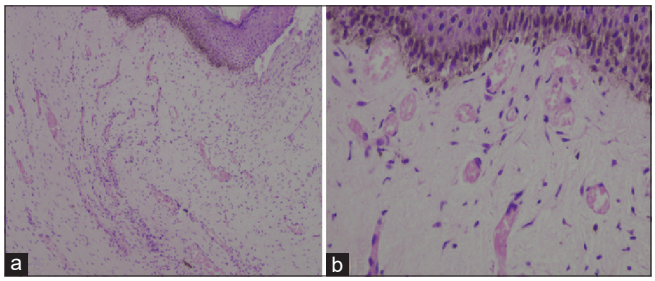


**Figure 4 (a and b):** Histopathological examination revealing parakeratosis with basal hyperpigmentation. Underlying demis showing loosely fibro collagenous stroma with abundant vessels (hematoxylin and eosin, ×10, ×40).

Lymphedema is a chronic condition involving lymphatic vessels and subsequent impairment of drainage of interstitial fluid. It can be categorized into two types: Primary lymphedema and secondary lymphedema. Primary lymphedema can be further divided into congenital (Milroy's disease), lymphedema praecox (at puberty), and lymphedema tarda (after 35 years). Secondary lymphedema is a more common entity and can be secondary to filarial or secondary sexually transmitted infections such as lymphogranuloma venereum and donovanosis, rarely following removal of local lymphatic lymph nodes, radiotherapy, and malignancy. In our report, we suspected lymphedema to be of a primary type after excluding secondary causes of lymphedema on history, clinical examination, and radiological and histopathological evaluation.[1,2]

Lymphedema tarda, also known as type III lymphedema, is a primary hereditary lymphedema characterized by onset after age 35. It is a benign condition that develops after local inflammation caused by any musculoskeletal injury, local infection, or insect bite, leading to an increased fluid load over the local site and a congenital anatomical

defect. Saxophone penis is a specific penile malformation characterized by a saxophone shape due to inflammation of the major penile lymphatic vessels that cause fibrosis of the surrounding connective tissue.[2-4]

Saxophone penis remains a disabling sequela of lymphedema tarda affecting genitalia. Proper diagnosis and taking steps to manage swelling can help to keep lymphedema from progressing and reduce complications. Close mimickers in our case included condyloma acuminata, Buschke Lowenstein tumors, and verrucous carcinoma. It is essential to diagnose these entities early as they are human papillomavirus-driven and have the potential to undergo malignancy.[2,5]

Saxophone penis can have immense physical, sexual, and psychosocial implications on the patient. It can also hinder a patient's ability to pass urine due to extension and occlusion of the external urinary meatus. Treatment is warranted for the patient's medical and cosmetic needs, thus improving quality of life. Treatment usually includes wide local excision with grafting and may require amputation if non-salvageable and treatment of the underlying main cause. Recurrences have been reported.[5]

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