Saxophone penis: A sequel to penoscrotal hidradenitis suppurativa

Sir,

The term "saxophone penis" refers to penile twisting, along its longitudinal axis, thereby conferring to it an appearance that closely resembles a saxophone.[1]

It has been suggested that long-standing penoscrotal inflammation culminates in lymphatic fibrosis. Further, diminished blood supply to the dorsal penis heralds contraction of connective tissue that results in dorsal bending of the penis. As vascularity of the ventral penis is not compromised, blood extravasation continues, leading to edema that lifts the distal penis upward and outward, making the dorsal penile curvature more prominent that finally assumes the configuration of a saxophone.[2]

Conditions resulting in saxophone penis (reported in the past) include lymphogranuloma venereum (tertiary stage), penile tuberculosis, and primary lymphedema.[1]

We hereby report saxophone penis in a patient of penoscrotal hidradenitis suppurativa (HS).

A 47-year-old male presented with a 2-month history of gradually progressive painful lesions involving the penoscrotal region. It began as nodular lesions involving the pubic area initially, which then spread to involve the penis and scrotum. He gave history of three similar episodes in the past 2 years which improved after the intake of antibiotics. This time, however, despite the intake of antibiotics, there was no improvement. No history of exposure or trauma was obtained. Furthermore, no comorbidities were identified. His spouse was apparently healthy.

Examination revealed deep-seated nodules, abscesses, and sinuses in the penoscrotal region. The penis was grossly enlarged and had assumed the shape of a saxophone [Figure 1a]. Notable sparing of the gluteal and perianal areas was observed [Figure 1b]. No obvious lesions were detected over the axilla and groins. There was no evidence of inguinal lymphadenopathy. Routine blood examinations, including the Mantoux test, serum venereal disease research laboratory test, and HIV 1 and 2 did not demonstrate any underlying abnormality.

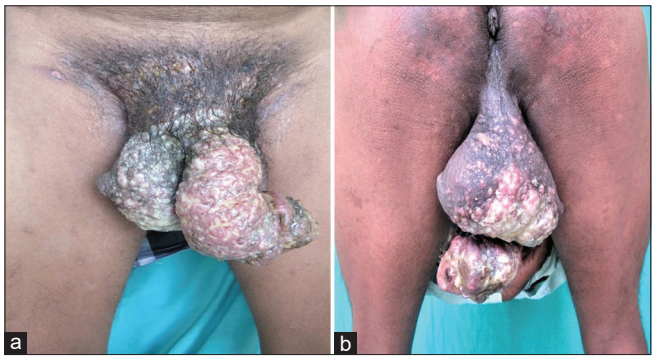

Figure 1: (a) Deep-seated nodules, abscesses, and sinuses in the penoscrotal region. The penis is grossly enlarged and has assumed the shape of a saxophone. (b) Notable sparing of the gluteal and perianal region can be observed

Serological tests for lymphogranuloma venereum like complement fixation test and microimmunofluorescence test were not done due to unavailability. Chest X-ray was normal. Although penoscrotal ultrasonography was suggested (to rule out deeper lymphatic involvement), our patient was unable to get that done. Histopathology from the scrotal lesion delineated a ruptured cyst lined by stratified squamous epithelium containing lamellated keratin and inconspicuous hair shafts. Surrounding it was a mixed inflammatory infiltrate composed predominantly of lymphocytes and histiocytes. Few giant cells, neutrophils, and plasma cells were also observed [Figure 2a and b].

Based on the above findings, a diagnosis of HS was made and the patient was started on prednisolone (20 mg once daily), clindamycin (300 mg twice daily), and rifampicin (300 mg twice daily) for 4 weeks and asked to review. The patient was subsequently lost to follow-up.

HS is a chronic, inflammatory disease characterized by painful, deep-seated, inflamed lesions in apocrine gland-bearing body parts, namely, the axilla, groins, buttocks, infra/intermammary folds, and perineal region.

HS limited to the penoscrotal area is not a common occurrence. In most cases, the disease also involves other classical sites, especially the axilla. There have been only a few reports delineating penile and scrotal HS, details of which have been elucidated in Table 1.

Based on the above publications, we infer that HS involving the penoscrotal region generally elaborates unusual and severe phenotypes.

Our patient differed from the above reports by having the disease limited to the pubis, penis, and scrotum. Besides, saxophone morphology was another peculiar finding that we observed. Further, while dealing with our patient, we realized the need for appropriate counseling regarding problems associated with obstructive lymphedema, and the requirement for regular and timely follow-up.

In our opinion, this presentation of penoscrotal HS has not been previously described. We therefore would like to highlight this unusual clinical morphology and the necessity for a cutaneous biopsy in all such cases, to enable diagnostic confirmation and prompt institution of appropriate therapy.

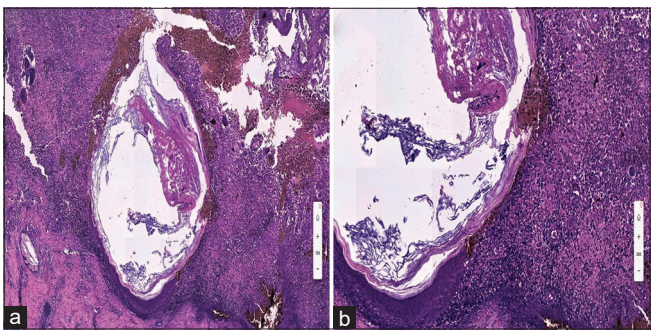

Figure 2: (a) A ruptured cyst lined by stratified squamous epithelium containing lamellated keratin, and inconspicuous hair shafts can be identified within the cyst cavity (H and E × 10). (b) A dense mixed inflammatory infiltrate composed predominantly of lymphocytes and histiocytes can be seen. Few giant cells, neutrophils, and plasma cells can also be visualized (H and E × 20)

Table 1: Previous reports of penile and scrotal hidradenitis suppurativa and their comparison with our case

Author(s), year	Patient details	Lesional morphology	Site(s) of involvement	Treatment given
Baughman and Cespedes, 2004[3]	55-year-old male Recurrent disease for nearly 20 years	Markedly indurated penis measuring 15 cm × 20 cm Ventral penis massively enlarged Inferior displacement of normal lower abdominal wall skin to the dorsal penis No fistulas identified	Apart from genitalia; inguinal, gluteal, and perianal involvement was also present	Surgical reconstruction
Gibas *et al.*, 2012[4]	45-year-old male Recurrent disease for 10 years Disease had significantly limited the patient's sexual life	Multiple abscesses Interconnected sinus tracts Diffuse involvement	Apart from penoscrotal involvement; inguinal and perianal areas were also involved	As medical treatment was unsatisfactory, staged surgery was performed with a successful outcome
Kok and Lahiri, 2012[5]	49-year-old male Recurrent disease for 13 years	Degloving penis Raw penile shaft Mass of scarred tissue was present that obstructed patient's urinary stream No fistulas were noted	Apart from genitalia, the axilla, groins, perineum, and perianal area were also affected Lesions were acutely inflamed and edematous	Conservative treatment initially to allow the settling down of acute inflammation, followed by surgical treatment
Current authors, 2021	47-year-old male Recurrent disease for 2 years	Saxophone penis Deep-seated tender nodules, abscesses, and sinuses involving the penoscrotal region	Other classical sites specific to HS were not involved	Conservative treatment instituted The patient lost to follow-up

HS = Hidradenitis suppurativa

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given his consent for his images and other clinical information to be reported in the journal. The patient understands that his name and initials will not be published and due efforts will be made to conceal his identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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References

1. Maatouk I, Moutran R. Saxophone penis. JAMA Dermatol 2013;149:802.
2. Kumaran MS, Gupta S, Ajith C, Kalra N, Sethi S, Kumar B. Saxophone penis revisited. Int J STD AIDS 2006;17:65-6.
3. Baughman SM, Cespedes RD. Unusual presentation of hidradenitis suppurativa with massive enlargement of penis. Urology 2004;64:377-8.
4. Gibas A, Matuszewski M, Michajłowski I, Krajka K. Acne inversa of the scrotum and penis – Aggressive urological treatment. Cent European J Urol 2012;65:167-9.
5. Kok K, Lahiri A. Penile degloving: An unusual presentation of hidradenitis suppurativa. Ann R Coll Surg Engl 2012;94:e101-2.

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