# Saxophone Penis: A Forgotten Manifestation of Hidradenitis Suppurativa

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## PRACTICE **POINTS**

- Hidradenitis suppurativa (HS) is a multifactorial chronic inflammatory skin disease.
- Saxophone penis is a specific penile malformation characterized by a saxophone shape due to inflammation.
- Repetitive inflammation within the context of HS may cause structural deformity of the penis, resulting in a saxophone penis.
- Early diagnosis and treatment of HS may help prevent development of this condition.

To the Editor:

Hidradenitis suppurativa (HS) is a multifactorial chronic inflammatory skin disease affecting 1% to 4% of Europeans. It is characterized by recurrent inflamed nodules, abscesses, and sinus tracts in intertriginous regions.[1] The genital area is affected in 11% of cases[2] and usually is connected to severe forms of HS in both men and women.[3] The prevalence of HS-associated genital lymphedema remains unknown.

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. Poor blood flow further causes contracture and distortion of the penile axis.[4] Saxophone penis also has been associated with primary lymphedema, lymphogranuloma venereum, filariasis,[5] and administration of paraffin injections.[6] We describe 3 men with HS who presented with saxophone penis.

A 33-year-old man with Hurley stage III HS presented with a medical history of groin lesions and progressive penoscrotal edema of 13 years' duration. He had a body mass index (BMI) of 37, no family history of HS or comorbidities, and a 15-year history of smoking 20 cigarettes per day. After repeated surgical drainage of the HS lesions as well as antibiotic treatment with clindamycin 600 mg/d and rifampicin 600 mg/d, the patient was kept on a maintenance therapy with adalimumab 40 mg/wk. Due to lack of response, treatment was discontinued at week 16. Clindamycin and rifampicin 300 mg were immediately reintroduced with no benefit on the genital lesions. The patient underwent genital reconstruction, including penile degloving, scrotoplasty, infrapubic fat pad removal, and perineoplasty (Figure 1). The patient currently is not undergoing any therapies.

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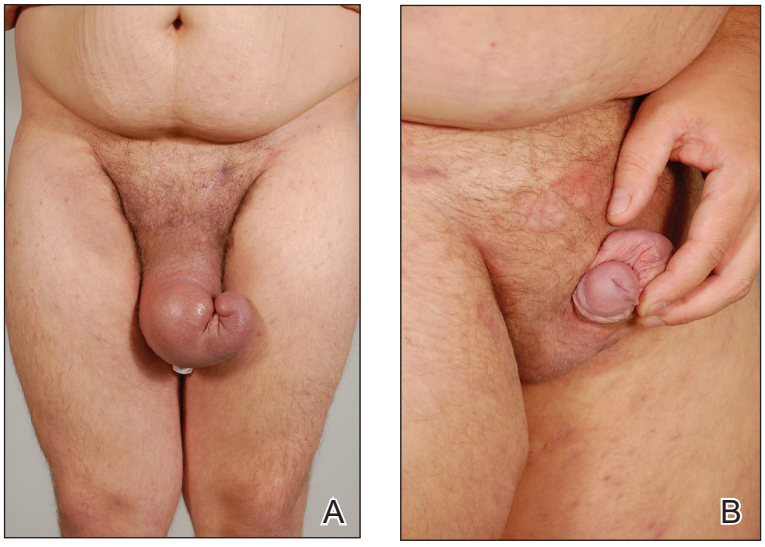


**FIGURE 1.** A, Saxophone penis in a patient with hidradenitis suppurativa treated with adalimumab. B, The patient underwent genital reconstruction, including penile degloving, scrotoplasty, infrapubic fat pad removal, and perineoplasty.

A 55-year-old man presented with Hurley stage II HS of 33 years' duration. He had a BMI of 52; a history of hypertension, hyperuricemia, severe hip and knee osteoarthritis, and orchiopexy in childhood; a smoking history of 40 cigarettes per day; and an alcohol consumption history of 200 mL per day since 18 years of age. He had radical excision of axillary lesions 8 years prior. One year later, he was treated with concomitant clindamycin and rifampicin 300 mg twice daily for 3 months with no desirable effects. Adalimumab 40 mg/wk was initiated. After 12 weeks of treatment, he experienced 80% improvement in all areas except the genital region. He continued adalimumab for 3 years with good clinical response in all HS-affected sites except the genital region.

A 66-year-old man presented with Hurley stage III HS of 37 years' duration. He had a smoking history of 10 cigarettes per day for 30 years, a BMI of 24.6, and a medical history of long-standing hypertension and hypothyroidism. A 3-month course of clindamycin and rifampicin 600 mg/d was ineffective; adalimumab 40 mg/wk was initiated. All affected areas improved, except for the saxophone penis. He continues his fifth year of therapy with adalimumab (Figure 2).

Hidradenitis suppurativa is associated with chronic pain, purulent malodor, and scarring with structural deformity. Repetitive inflammation causes fibrosis, scar formation, and soft-tissue destruction of lymphatic vessels, leading to lymphedema; primary lymphedema of the genitals in men has been reported to result in a saxophone penis.[4]

The only approved biologic treatments for moderate to severe HS are the tumor necrosis factor α inhibitor adalimumab and anti-IL-17 secukinumab.[1] All 3 of our patients with HS were treated with adalimumab with reasonable success; however, the penile condition remained refractory, which we speculate may be due to adalimumab's ability to control only active inflammatory lesions but not scars or fibrotic tissue.[7] Higher adalimumab dosages were unlikely to be beneficial for their penile condition; some improvements have been reported following fluoroquinolone therapy. To our knowledge, there is no effective medical treatment for saxophone penis. However, surgery showed good results in one of our patients. Among our 3 adalimumab-treated patients, only 1 patient had corrective surgery that resulted in improvement in the penile deformity, further confirming adalimumab's limited role in genital lymphedema.[7] Extensive resection of the lymphedematous tissue, scrotoplasty, and Charles procedure are treatment options.[8]

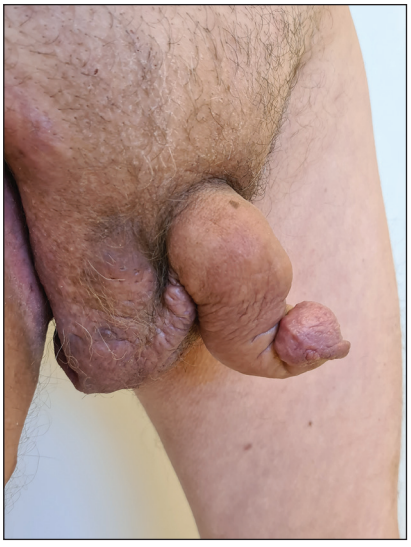

**FIGURE 2.** Saxophone penis in a patient with hidradenitis suppurativa treated with adalimumab.

Genital lymphedema has been associated with lymphangiectasia, lymphangioma circumscriptum, infections, and neoplasms such as lymphangiosarcoma and squamous cell carcinoma.[9] Our patients reported discomfort,

hygiene issues, and swelling. One patient reported micturition, and 2 patients reported sexual dysfunction.

Saxophone penis remains a disabling sequela of HS. Early diagnosis and treatment of HS may help prevent development of this condition.

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