Dermatology Practical & Conceptual

Saxophone Penis Secondary to Lichen Simplex Chronicus Responding to Low Dose of Methotrexate and Doxycycline

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Introduction

Our case is interesting because of a rare presentation of anogenital lichen simplex chronicus (LSC) as a saxophone penis and a good response to anti-inflammatory agents like doxycycline and methotrexate.

Case Presentation

A man in his 60s presented with complaints of severe itching over genitalia and scrotal skin thickening (lichenification) associated with overlying depigmented macules for the past 5 years. He also complained of deformity of the penis associated with deviation in urine stream during micturition for the past 3 months (Figure 1A).

He had been diagnosed with LCS, vitiligo, and non-specific pruritus by different physicians and had taken intermittent oral antihistamines and topical steroid and antifungals in the past with minimal improvement.

He had bilateral inguinal hernia for the past 6-7 years.

Skin biopsy showed psoriasiform epidermal hyperplasia with parakeratosis and focal presence of a granular layer. There was perivascular and peri-eccrine infiltrate with some interstitial spill of lymphocytes, histiocytes, plasma cells, and eosinophils. Sclerosis of collagen was noted in the dermis. Overall, features were suggestive of sclerosing mixed inflammatory tissue reaction (Figure 1 B and C).

Peripheral smear for microfilaria, Mantoux test, PCR for chlamydia (urethral and rectal), Venereal Disease Research Laboratory (VDRL) test, and stool for occult blood were negative.

He was diagnosed as a case of saxophone penis and scrotal thickening secondary to LSC and was advised to avoid scratching and treated with scrotal support, low dose methotrexate 7.5 mg/week (0.1 mg/kg/week), doxycycline 100 mg once daily along with mid potency topical steroid once daily, emollients, and antihistamines. Methotrexate and doxycycline were added because of the severity of the

disease (architectural distortion), their proven efficacy, potent anti-inflammatory action, good safety, and cost-effective profile. The patient was referred to the surgery department for the management of inguinal hernia.

There was a significant improvement in the deformity of the penis, thickening of the scrotal skin, deviated urinary stream, and itching after 3 months of therapy (Figure 1D).

Discussion and Conclusions

Complications of genital LSC are clitoral or labial hypertrophy, fibrosis, and scarring that can occur due to vigorous scratching leading to architectural distortion [1].

The inguinal hernia has been hypothesized to cause LSC by inducing itching due to the occlusive effect [2]. This might be one of the triggers for the development of LSC in our case.

There are various other causes of saxophone penis. Some of them are infective like lymphogranuloma venereum (LGV), chlamydia trachomatis infection (non-LGV), filariasis and some are non-infective like congenital hereditary lymphoedema, malignant infiltration of lymph nodes, LSC [3].

Kumaran et al gave the hypothesis of the pathogenesis of the saxophone penis by mentioning that long-standing inflammation leads to fibrosis of lymphatics, which run along the dorsal penile vein as well as surrounding connective tissue. Due to poor blood supply of the dorsal aspect of the penis, contraction of that tissue occurs leading to dorsal bending of the penis. Corpora cavernosa is attached to ischial tuberosity but bulbospongiosus and glans penis don't have such attachment leading to their diversion upwards. The ventral part (vascular part) hence becomes a dependent part leading to excessive fluid extravasation leading to edema of ventral prepuce which further pushes the glans upwards and outwards, accentuating the dorsal curvature, forming a saxophone penis [4]. This hypothesis holds true in our case as well, as chronic inflammation due to LSC might have led to the fibrosis of lymphatics and surrounding connective tissue (shown in the biopsy image) leading to a saxophone penis.

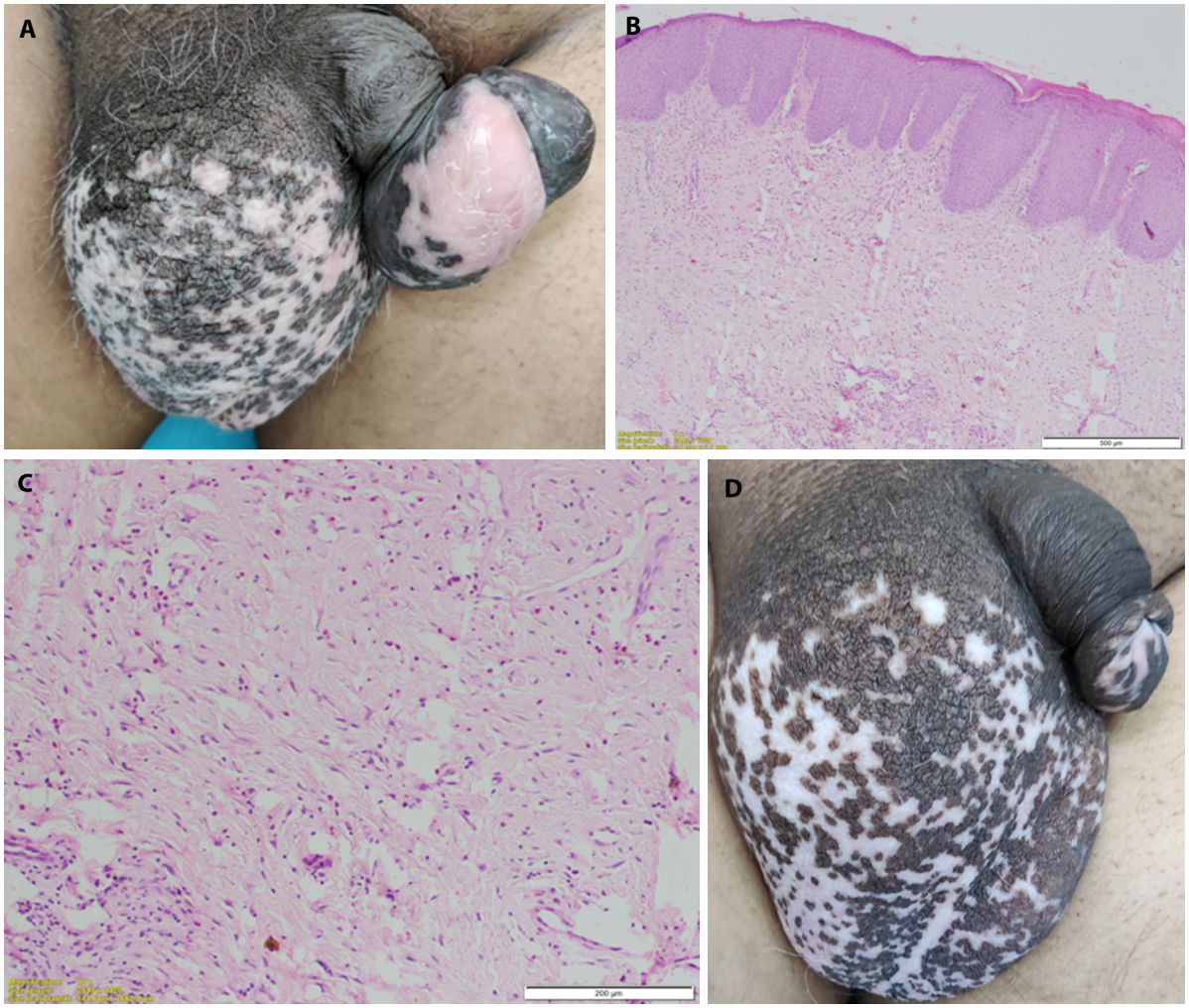

Figure 1. (A) Thickened deformed (saxophone) penis and scrotal thickening with depigmented macules. (B) Psoriasiform hyperplasia of the epidermis with parakeratosis and focal presence of a granular layer (H&E:40X). (C) Interstitial infiltrate of lymphohistiocytes, plasma cells, and eosinophils. Sclerosis of collagen was noted in the mid and deep dermis (H&E: 100x). (D) Significant improvement with correction of deformity of penis and decrease in scrotal thickening after 3 months of treatment.

Treatment includes treatment of suspected trigger like hernia, cutting off the itch-scratch-itch cycle, emollients, antihistamines, anti-inflammatory agents like doxycycline, methotrexate, topical and oral corticosteroids etc [5].

Moisturizers containing polidocanol have been found to reduce non-histaminergic itch in placebo-controlled double-blind trials. Humectants like urea, lactic acid are not recommended due to their irritant potential. Short-term high potency topical corticosteroids are considered first-line therapy for LSC. A combination of topical steroid and salicylates have also been found to be effective. Intralesional triamcinolone can be used in recalcitrant cases. Topical calcineurin inhibitors like pimecrolimus or tacrolimus are useful especially to avoid side effects related to long-term steroid use. They can cause a transient burning sensation, prior information about which should be given to the patients. Menthol (cooling effect) and pramoxine (local anesthetic) have the potential to control pruritus in LSC patients. Topical doxepin, aspirin with dichloromethane, and ketamine with lignocaine and amitriptyline are recommended in mild cases of LSC. Capsaicin has not been found to be effective.

Moderate cases can be treated with gabapentinoids, sedative antihistamines, antidepressants, cyclosporine, methotrexate, and NBUVB phototherapy. Antidepressants are useful in individuals with comorbid anxiety or depression. A randomized controlled trial found that imipramine/chlorpheniramine significantly reduced itch severity in patients with LSC. Cyclosporine and methotrexate have been shown to be effective in patients with prurigo nodularis. Hence, they are likely to be effective in LSC cases as well.

Recommended treatment options for severe cases include nemolizumab, dupilumab, Janus kinase inhibitors like tofacitinib, focused ultrasound, and transcutaneous electrical nerve stimulation [6].

We should be aware of such a complication of LSC and its management to prevent unnecessary apprehension about genital malignancy.

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