LICHEN SCLEROSIS: A RARE CAUSE OF URETHRAL STRICTURE IN BLACKS, MANAGED BY BUCCAL MUCOSAL GRAFT URETHROPLASTY

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ABSTRACT

Background: Penile Lichen Sclerosus is a chronic progressive inflammatory disease of the foreskin. Balanitis Xerotica Obliterans is a traditional terminology used to refer to Lichen Sclerosus affecting the foreskin and glans penis. Involvement of the urethra by Lichen Sclerosus is quite uncommon in our region but can result in urethral stricture.
Method: We reported a case of urethral stricture secondary to Lichen Sclerosus, which was managed by Buccal Mucosal Graft Urethroplasty with a satisfactory outcome.
Conclusion: High index of suspicion helps in diagnosing this case allowing the best treatment option to be instituted. Early circumcision is one of the preventive measures.

Keywords: Buccal mucosal graft, Lichen Sclerosus, Male Urethral stricture, Outcome.

INTRODUCTION

Lichen Sclerosus (LS) can be defined as an idiopathic chronic progressive inflammatory disease of the skin. When affecting the skin of the male genitalia, it is referred to as Male Genital Lichen Sclerosus (MGLSc). LS is a rare condition affecting the male genitalia. It is synonymous with Balanitis Xerotica Obliterans (BXO), which refers to a chronic inflammatory dermatological disease of the male genitalia, often progressive and may result in phimosis and urethral stenosis affecting both urinary and sexual function,[1] as described by stuhmer in 1928. Since 1995, the American society of Dermatology[2] has recommended the replacement of the word 'Balanitis Xerotica Obliterans' by Lichen Sclerosus. The incidence of LS is uncertain in our region. In the United States it is about 1.4 per 100,000 population and predominantly among Caucasian and uncircumcised males than those circumcised.[2]

The aetiology of LS is unknown, but has been associated with risk factors such as autoimmune diseases. In a particular study, auto antibodies to extracellular matrix protein 1 (ECM-1) were detected in the serum of 67% of LS patients and only 7% of controls.[3] Other risks are infections by Mycobacterium, Spirochetes, Human papilloma Virus, Epstein-bar virus and Hepatitis C Virus and chronic irritation of the keratinized foreskin by post-micturation dribble of urine among uncircumcised males.[4] We present a case of urethral stricture secondary to LS which was managed by Buccal Mucosal Graft Urethroplasty with a satisfactory outcome.

CASE REPORT

A 26 year old male student presented with progressive difficulty in passing urine of 6 months duration. This was characterized by poor stream which improves on straining, forking with spraying of urine stream, frequency and nocturia. There were recurrent episodes of purulent urethral discharge and fever. Patient has been on self-medication with drugs purchased from patent medicine shops with no significant improvement of the Lower Urinary Tract Symptoms (LUTS) . One month prior to presentation, the symptoms culminated in acute urine retention. An attempt at urethral catheterization failed at a peripheral

hospital which necessitated Supra-pubic cystostomy after which he was referred to our centre. There was background history of blister- like lesion on the surface of the glans penis and penile shaft about 8 years prior to presentation which was associated with pruritus and minimal pain. The blister ruptured spontaneously and healed leaving multiple hypo-pigmented spots over the scar, which later coalesced into patches . There was no preceding history of trauma to the genitalia, purulent urethritis, burns, or a history suggestive of drug reaction. No sexual promiscuity or family history of similar skin lesion with urinary symptoms. He is single, not a known diabetic and not on any immunosuppressive drug.

On examination he was fit looking, not pale and afebrile with normal vital signs. Abdominal examination revealed a supra-pubic cystostomy with Foley's catheter insitu draining cloudy urine. The liver, kidneys and spleen were not enlarged, no demonstrable ascites and bowel sound was normal. Examination of the external genitalia revealed a circumcised phallus with multiple hypo-pigmented patches over an indurated scar involving the Glans penis circumferentially as shown in figure 1a. The urethral meatus was speared and patent. The remaining penile and scrotal skin and per-rectal examination were essentially normal. Review of other systems was also normal.

An initial diagnosis of urethral stricture with Vitiligo of the glans was made to keep in view LS. Preoperative urine flow rate was 4mls per second. Urine culture grew Klebsiella species and the patient was treated with an antibiotic to which the organism was sensitive to. Retrograde urethrogram was conclusive of a long segment incomplete distal penile urethral stricture as shown in figure 1b. Hepatitis, Venereal Disease Research Laboratory (VDRL) and Retroviral screening results were negative. Abdominal Ultrasound scan, full blood count, serum electrolytes and urea were also normal. The treatment options were explained to the patient and he consented for urethroplasty.

Intra-operative findings revealed a 4cm long area of fibrosis involving the dorsal aspect of the distal penile urethra, which was excised out. Buccal Mucosal Graft (BMG) was harvested quilted, meshed and sutured on the raw area dorsally and the urethra was sewn to the graft on a stent and wound closed in layers as shown in figures 2a, 2b, 2c, 2d and 2e. Post-operative condition was satisfactory. Urethral catheter was removed after 3 weeks with a satisfactory urine flow rate of 15mls/second. The urine stream remained good 1 year postoperative as shown in figure 2e. Findings from the histology of the excised tissues include hyperkeratosis, stromaloedema of the basal cells, bands of hyalinization and angiectesia as shown in figures 3a and 3b. This is in keeping with Lichen sclerosus.

Figure 1a: Lichen Sclerosus around the glans.

Figure 1b: Urethral stricture on RUG

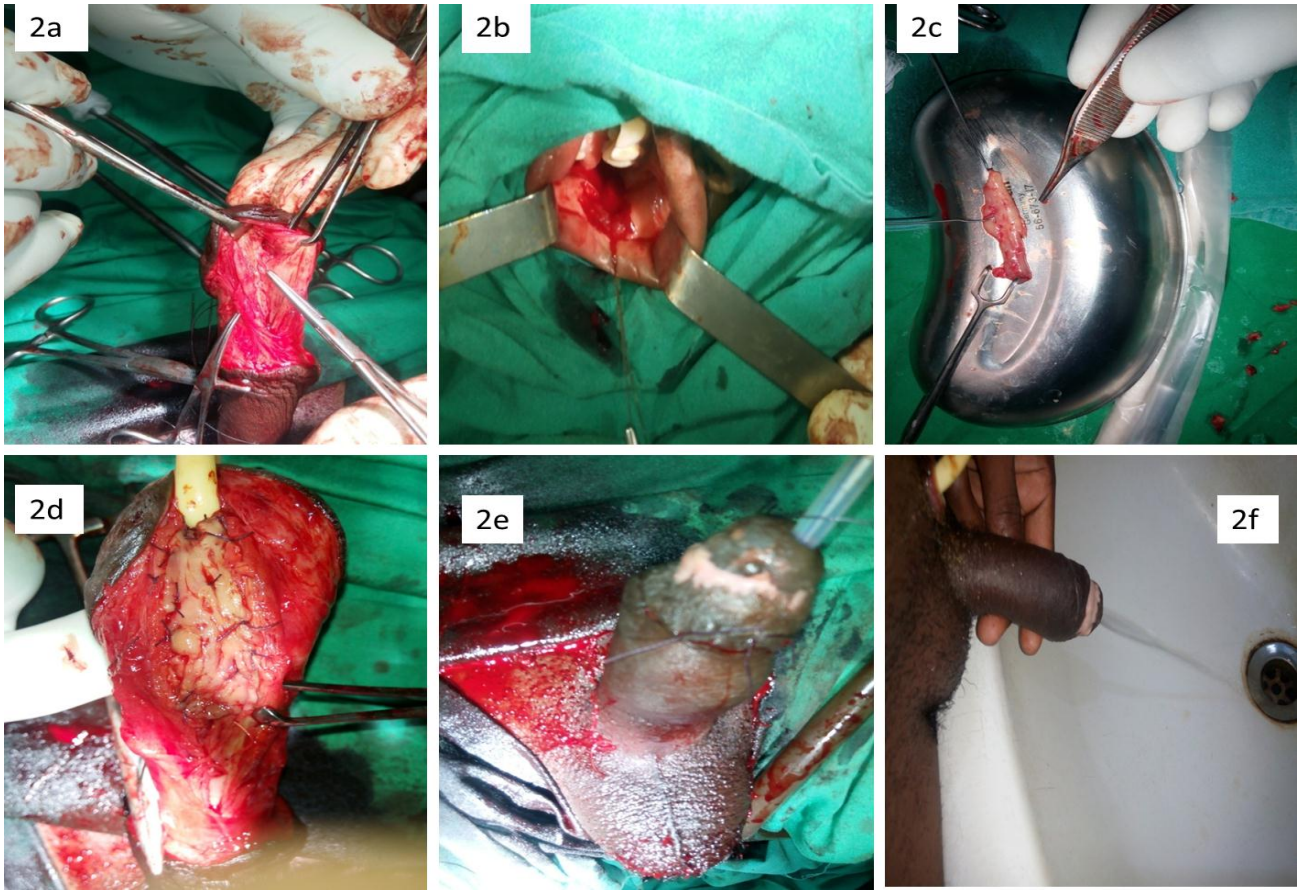

Figure 2a: LS involving the urethra. **Figure2b:** Donor site for Buccal Mucosal graft
Figure 2c: harvested BMG .**Figure 2d**: BMG sutured on the corporal body.
Figure 2e: wound closed in layers. **Figure 2f:** Good urine stream 6 months after surgery

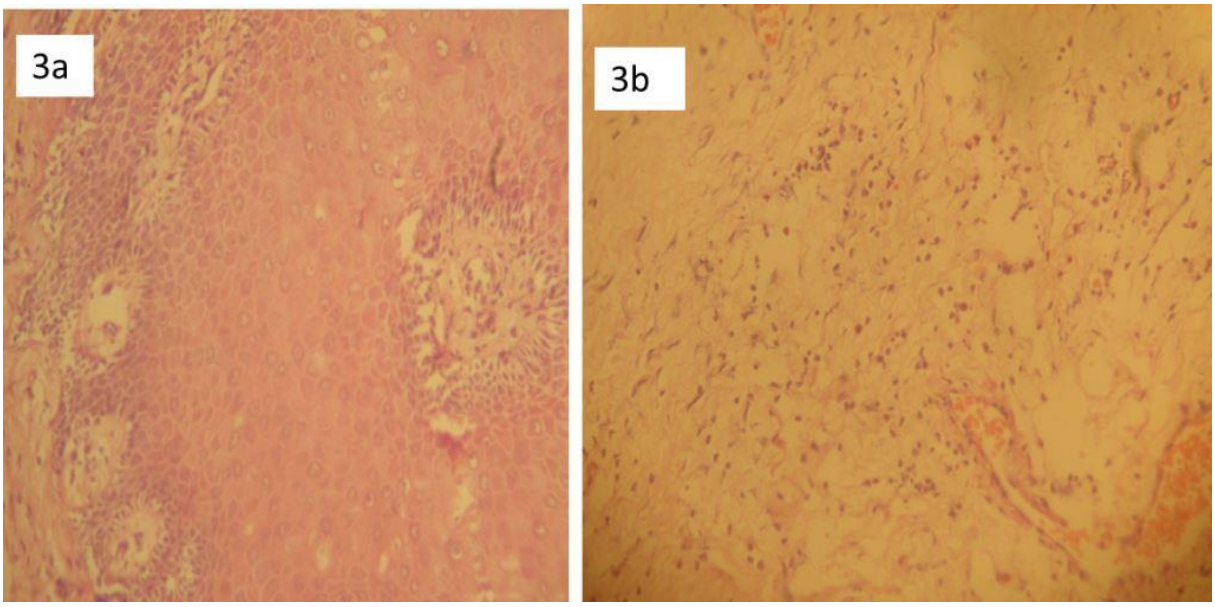

Figure3a: Histology displaying hyperkeratosis. **Figure 3b:** Stromal oedema and angiectesia.

DISCUSSION

Lichen sclerosus affecting the male genitalia is uncommon; it's even rarer when the lesion occur in the already circumcised male with urethral stricturing as in the index case. LS can be aggressive or insidious onset as in our index case whose skin lesions started 8 years before presentation. Penile LS usually present between the ages of 30-49years. It typically starts as an itchy patch of the white discoloration on the inner aspect of the foreskin or glans in form of patches; this may later coalesce and involve the wider skin and extending to involve the meatus and the fossa navicularis and can even ascend to involve penile urethra. The affected skin and urethral mucosa becomes inelastic, brittle and soar. This later result in scaring and narrowing of the lumen resulting in phimosis and urethral stricture[2] as seen in the patient presented.

Not all cases of LS result in urethral stricture or phimosis. Previous studies revealed isolated bulbar urethral stricture caused by LS in 44% of their cases[5] or urethral stricture as component of multiple sites penile Lichen Sclerosus in 20% of the cases.[6] Our patient had isolated LS of the glans and penile urethra spearing the meatus as shown in figures 1a and 2a.

Histology of tissue biopsy from the affected foreskin or urethral mucosa confirms the diagnosis of LS in our case. Common histological findings are hyperkeratosis of the epithelium, atrophy and loss of the rete pegs, degeneration of the basal cells, sclerosis of the sub epithelial collagen and lymphocytic infiltration of the dermis, which was oedematous initially and later become hyerlinized.[7] When lichen Sclerosus is diagnosed at early stage topical steroids such as Clobetasol propionate can be applied on the affected skin twice daily for a period of 2 to 3months. This can as well be applied into affected urethra combined with periodic urethral dilatation as a form of conservative management as described.[8] Cryotherapy and Ultraviolet phototherapy were also tried in the past. Early circumcision eliminates the progression of LS in the prepuce.

Urethroplasty and meatoplasty are the preferable treatment options for LS strictures to achieve a positive long term outcome. The current first choice of graft material for urethroplasty is BMG either as single or staged procedure and it has a success rate 91% as reported by a previous study.[9] BMG has a suitable property for being thin, and has highly vascular lamina propria that makes it a very reliable graft with excellent revascularization. Genital skins are best avoided due to high risk of recurrent strictures when the skin is used for urethroplasty.[6] Squamous Cell Carcinoma may present as long term complication of Lichen Sclerosus.[8]

CONCLUSION

Lichen sclerosus involving the penis and presenting as urethral stricture is uncommon in our region. High index of suspicion is relevant to differentiate it from other conditions such as Vitiligo and Erythroplasia of Querat. Buccal Mucosal graft urethroplasty gives a long term treatment outcome for LS strictures.

REFERENCES

1. Chong-Rui Jin, Ying-Long Sa, Yue-Min Xu, Qiang Fu. Review of the male genital lichen sclerosus and urethral involvement Int J Clin Exp Med 2016; 9(9):17069-17077
2. Pugliese JM, Morey AF and Peterson AC. Lichen sclerosus: review of the literature and current recommendations for management. J Urol 2007; 178: 2268-2276.
3. Oyama N, Chan I, Neill SM, et al. Autoantibodies to extracellular matrix protein 1 in lichen sclerosus. Lancet 2003; 362:118-23
4. Bunker CB, Shim TN. **Male Genital Lichen Sclerosus.** Indian J Dermatol. 2015 Mar-Apr; 60(2): 111–117
5. Liu JS, Walker K, Stein D, et al. Lichen Sclerosus and isolated bulbar urethral stricture disease.J Urol 2014; 192:775-9
6. Depasquale I, Park AJ, Bracka A. The treatment of Balanitis xerotica obliterans. BJU Int 2000; 86:459-65
7. Mundy AR, Andrich DE. Urethral strictures. BJU Int. 2011; 107:6–26

8. Das S, Tunuguntla HS. Balanitis Xerotica Obliterans – A review. World J Urol.2000;18:382–7.
9. Kulkarni S, Barbagli G, Kirpekar D, et al. Lichen sclerosus of the male genitalia and urethra: surgical options and results in a multicenter international experience with 215 patients. Euro Urol 2009;55:945-54