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Lichen sclerosus presenting as an isolated bulbar urethral stricture

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ABSTRACT

Lichen sclerosus (LS) is a chronic inflammatory condition of the anogenital skin that can cause significant urinary and sexual dysfunction in men, particularly by means of destructive urethral disease. LS is traditionally thought to progress from the meatus with migration along the urethra proximally, however we present a case describing an isolated bulbar urethral stricture secondary to LS. To our knowledge, this has only been reported in the literature in one previous study. Clinician recognition of LS as a potential cause of isolated bulbar urethral stricture disease is important as this has ramifications on follow up and successful management.

Consent

Informed consent was gained from the patient for writing this paper.

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1. Section heading

General Urology, Inflammation/Infection.

2. Introduction

Lichen sclerosus (LS) is a chronic inflammatory condition of the anogenital skin, preferentially affecting uncircumcised men.[1–3] Genital LS usually presents with foreskin/glans involvement with evidence of cracking and fissuring, causing phimosis or meatal stenosis.[2] In 20 % of male cases, disease may progress to involve the urethra.[2] Inflammation and destructive scarring leads to urethral stricture disease.[3] The process is traditionally thought to occur in a continuous manner from distal urethra/meatus to extend proximally.[2,4,5] Isolated bulbar urethral stricture secondary to LS is rare, and to our knowledge, has only been reported in one study previously.[5] Therefore, we report a case on an isolated proximal bulbar urethral stricture secondary to LS in a patient with no prior history or anogenital manifestations.

3. Case presentation

A 77-year-old male presented with poor urinary flow, nocturia and incomplete emptying. Abdominal and digital rectal examinations were unremarkable. He was circumcised and had no external manifestations suggestive of LS. The histology of the circumcision was unknown. He had a history of transurethral resection of the prostate four years prior, and subsequently developed proximal urethral stricture disease requiring optical urethrotomy. He performed daily clean intermittent self-catherization for one month post-operatively and has been asymptomatic until this presentation. His past medical history includes atrial fibrillation and previous open appendicectomy. His International Prostate Symptoms (IPSS) score was 16 on presentation with an International Index of Erectile Function (IIEF) score of 1.

Uroflowmetry demonstrated an average flow rate of 8.2mls per second with a prolonged, intermittent flow. Retrograde cystourethrogram revealed an isolated bulbar urethral stricture (Fig. 1). Rigid cystoscopy to assess the stricture showed a 6Fr wide proximal bulbar urethral stricture approximately 2–3cm in length with surrounding pale mucosa (Fig. 2). The prostate, bladder and ureteric orifices were unremarkable.

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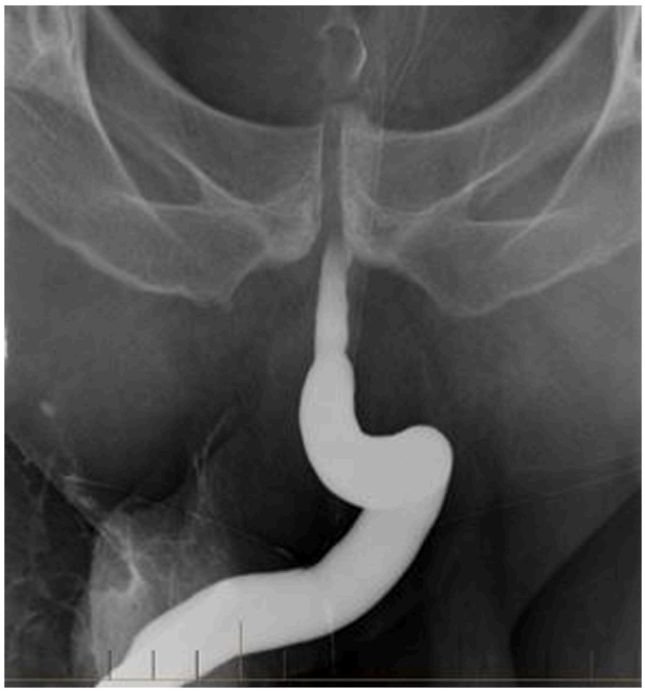

Fig. 1. Retrograde cystourethrogram showing an isolated bulbar urethral stricture.

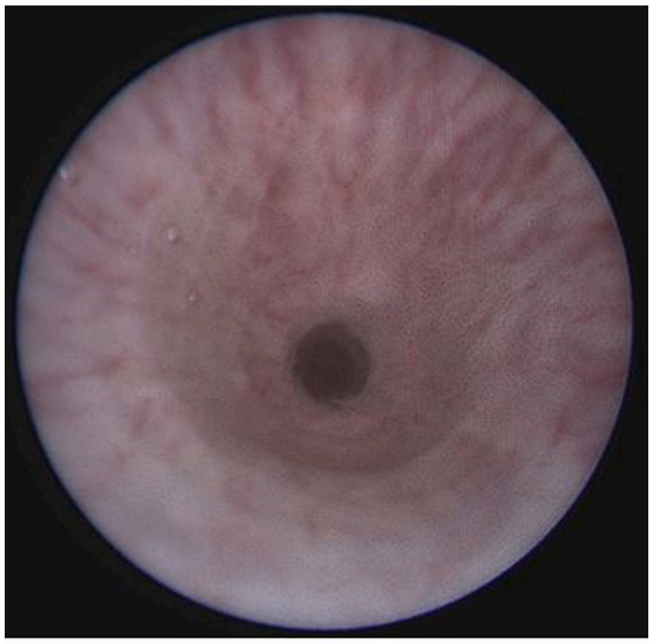

Fig. 2. Endoscopic appearance of the isolated bulbar urethral stricture.

The decision was made for a dorsal onlay buccal mucosal graft (BMG) urethroplasty using a Kulkarni one sided dissection.[4] A 6 × 1.5cm graft was harvested from the patient's cheek and the urethra was mobilised dorsally. Urethral biopsies of the stricture including biopsies distal and proximal to the stricture were obtained. The graft was quilted to the corpora cavernosa and closed over an 18Fr indwelling catheter. The post-operative course was unremarkable, and the indwelling catheter was removed after two weeks. There were no oral complications from the graft.

Histopathology from urethral tissue revealed submucosal scarring with a focus of hyalinization of the upper dermal collagen suggestive of lichen sclerosus (Fig. 3). Focal haemorrhage and epithelial denudation were seen with no inflammatory changes. The remaining biopsies showed dense scarring with varying degrees of acute and chronic inflammation. There was no evidence of dysplasia or carcinoma on any of the biopsies.

The patient remains asymptomatic two years post-operatively with no evidence of disease recurrence. Post-operative average urinary flow rate on uroflowmetry was 16mls per second. His IPSS score was now 7 and IIEF score unchanged.

4. Discussion

LS urethral involvement has traditionally been thought to progress distally to proximally; starting at the urethral meatus and progressing proximally along the urethra.[2,4,5] This proximal spread is believed to be from progression through urethral glands and irritative transformation of disease.[5] Therefore, isolated bulbar urethral strictures secondary to LS is rare. To our knowledge, only one prior study by Liu et al. has described isolated bulbar urethral stricture disease secondary to LS without progression from the distal urethra.[5] Liu et al. retrospectively reviewed histopathology from 70 patients who had underwent urethroplasties for isolated bulbar urethral stricture disease, revealing 7 % of patients on initial review displaying features of LS and 44 % on re-review.[5]

The pathogenesis of LS remains unknown but is likely a complex interplay of autoimmune, genetic, infective, hormonal, and Koebnerisation factors.[1,2,4] The Koebner phenomenon suggests that local trauma may be the provoking factor in the formation of LS.[2,4] In our case, the patient had undergone a previous urethrotomy and transurethral resection of the prostate, which may be the inciting factors for the isolated bulbar disease.

Treatment of urethral strictures may include dilatation, urethrotomy, urethroplasty or urethrostomy depending on the cause, extent, and location of the stricture. Urethral strictures secondary to LS remains a challenging condition to treat surgically, with suboptimal rates of recurrence, and requires complex reconstruction.[1,4] There are no standardised treatments for urethral strictures secondary to LS and surgical management should be tailored to each patient.[2] Dilatation, urethrotomy and primary anastomosis frequently lead to disease progression in LS and should be avoided.[1,5] Oral mucosal graft urethroplasty in a one- or two-staged procedure has the lowest rate of disease recurrence or need for re-intervention.[1,3] One-staged dorsal onlay BMG bulbar urethroplasty has a success rate of 91 % and was used in this case.[3] Using genital skin for the graft has a recurrence rate of 50–100 % and should be avoided as skin may already be, or become, diseased.[2–4] In complex patient populations, perineal urethrostomy can be considered with success rates of 72–100 %.[2]

Clinicians should be mindful that LS is a potential cause of bulbar urethral strictures in isolation. Management of urethral LS may warrant an oral mucosal grafted urethroplasty to minimise recurrence rates. Biopsy is recommended if LS is suspected, as LS carries a 2–8% risk of concomitant squamous cell carcinoma and requires more rigorous follow-up.[2,4] Failure of appropriate surgical intervention or follow-up may have devastating implications for patients' urinary and sexual function, or misidentification of neoplasm. Further knowledge regarding the pathogenesis and manifestations of this disease is necessary to ensure more favourable patient outcomes.

5. Conclusion

LS can be a debilitating condition, impairing sexual and urinary function. Though rare, initial presentation of LS, as in this case, can reveal an isolated bulbar urethral stricture without any external manifestations or sign of distal-to-proximal progressive disease. In such presentations, there must be a clinical suspicion of LS, as more

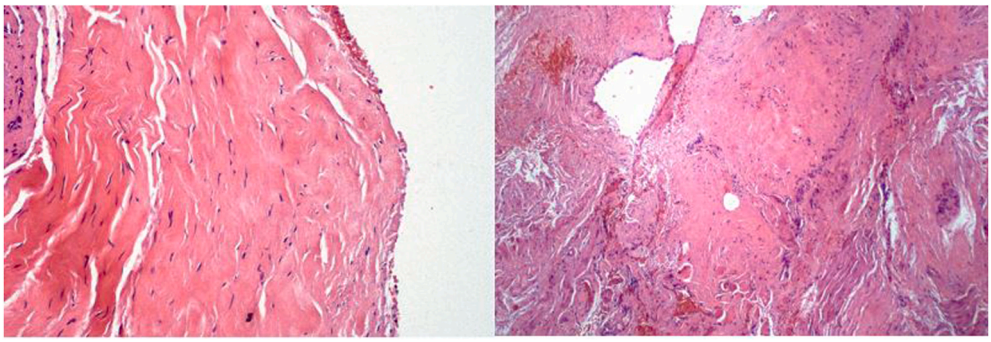

Fig. 3. Histopathology slides from the stricture biopsy showing scaring, epithelial denudation, and a focus of hyalinization suggestive of lichen sclerosus.

conservative management to treat LS urethral strictures may result in treatment failure. Consideration of BMG urethroplasty is warranted. A better understanding of LS pathogenesis and progression of disease is needed to improve patient outcomes.

Declaration of competing interest

None to declare.

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