

# The Diagnosis and Treatment of Lichen Sclerosus and Bulbous Urethral Squamous Cell Carcinoma: A Case Report

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## Abstract

Lichen sclerosus (LS) is a rare disease with malignant potential and has been identified by the European Society of Urology Guidelines as a risk factor for penile squamous cell carcinoma (SCC) cancer. LS combined with urethral squamous cell carcinoma (USCC) is extremely rare. There has been only one case report of this combination in China over the last decade. The prevalence of this combination in East Asian population is unclear. In this report, a 49-year-old patient with the presence of LS, perineal masses, and abscesses was hospitalized. He underwent anti-infectious therapy, meatotomy, perineal abscess incision, and drainage and debridement of the perineal wound. USCC was diagnosed following a series of biopsies and treated with radiation therapy. The patient remains well and alive with no recurrence of USCC 13 months after radiation treatment.

## Keywords

Lichen sclerosus, squamous cell carcinoma, chronic obstructive balanitis, urethral abscess

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Lichen sclerosus (LS) is a chronic inflammatory disease of unknown origin that occurs in the skin and mucous membranes. Male genital lichen sclerosus (MGLSc) occurs mainly in the foreskin, penis head, urethral meatus, fossa navicularis, and penile urethra, and rarely involves the bulbous urethra (Barbagli et al., 2011). LS is also a disease with malignant potential, and it has been reported in literature that a small portion of patients with MGLSc eventually develop malignant squamous cell carcinoma (SCC; Depasquale et al., 2000).

## Case Report

The 49-year-old male was admitted to the hospital for 1 week due to pain and discomfort caused by a perineal mass. His background included circumcision 10 years ago for phimosis, difficulty of urination over 2 years, and no history of urethral trauma, surgery, and stones. On examination, there were flaky white spots on the penis body, the glans, and the perineum (Figure 1); some spots were thickened with lichenoid. The urethral meatus was pin-point sized. The part of the perineum near the corpus cavernosum had a hard mass, measuring 6 × 3 cm, and no clear boundary with the urethra. No swelling was

detected in inguinal lymph nodes. A color doppler ultrasound test showed the same mass was hypoechoic before admission.

The patient's admission blood test results comprised normal white blood cell count (WBC) and prostate-specific antigen (PSA) levels, and negative human papillomavirus (HPV) and human immunodeficiency virus (HIV). The computed tomography (CT) scan showed a visibly flakey, slightly lower-density area on the corpus cavernosum in the perineal area. This area measured about 4.2 × 2.8 cm (Figure 2). The CT results suggested possible abscesses. Magnetic resonance imaging (MRI) considered a greater possibility of inflammatory lesions in the soft tissues of the perineal penis and scrotum root, which did not exclude the possible formation of small pus cavities.

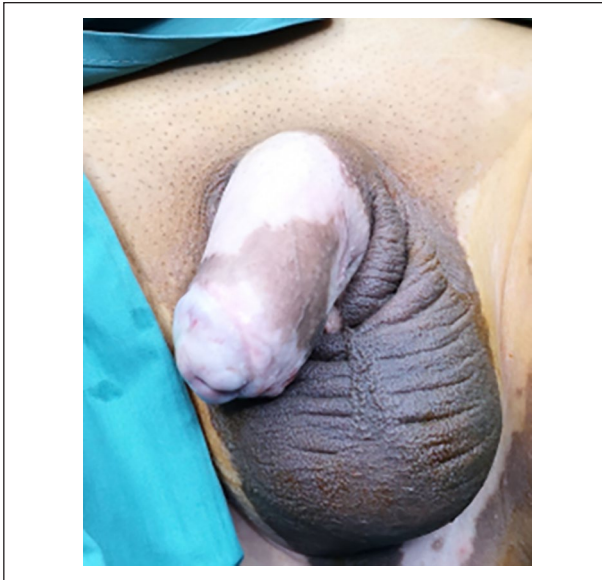
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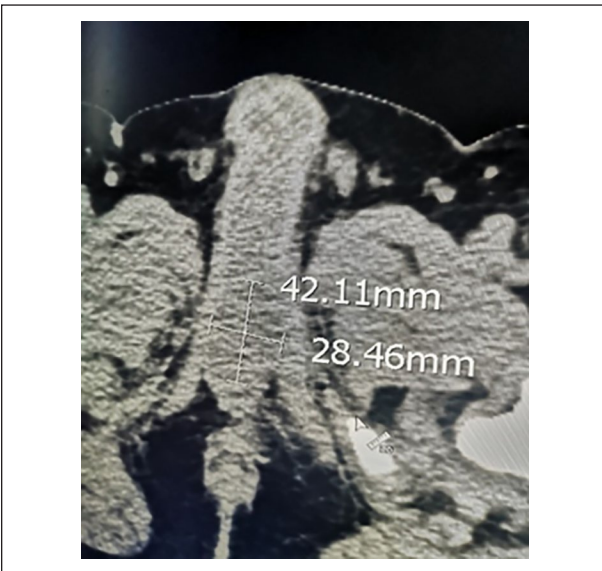
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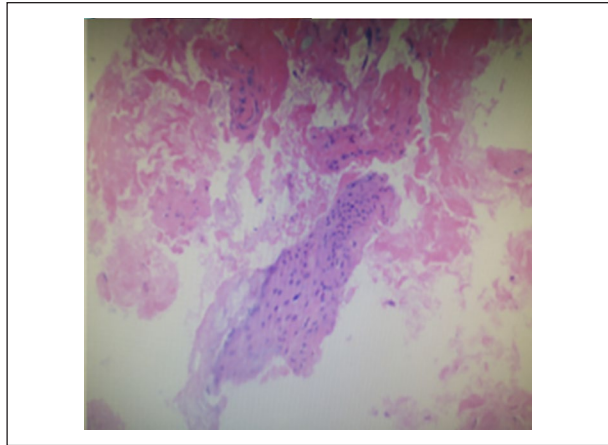


**Figure 1.** Genital LS. LS = lichen sclerosus.



**Figure 2.** The perineal mass.

The patient was diagnosed with LS, urethral stenosis, and possibly inflammatory perineal mass. Anti-infective treatment (piperacillin-tazobactam) was given, and the mass reduced in size to  $3 \times 2$  cm. Due to the unclear boundary between the mass and surrounding tissues and concerns about the difficulty of removing the mass and complications such as urethral fistula after excision, the patient underwent meatotomy and perineal mass puncture biopsy. The postoperative pathology report showed a small amount of squamous epithelium, fat, and fibrous connective tissue (Figure 3). The patient continued

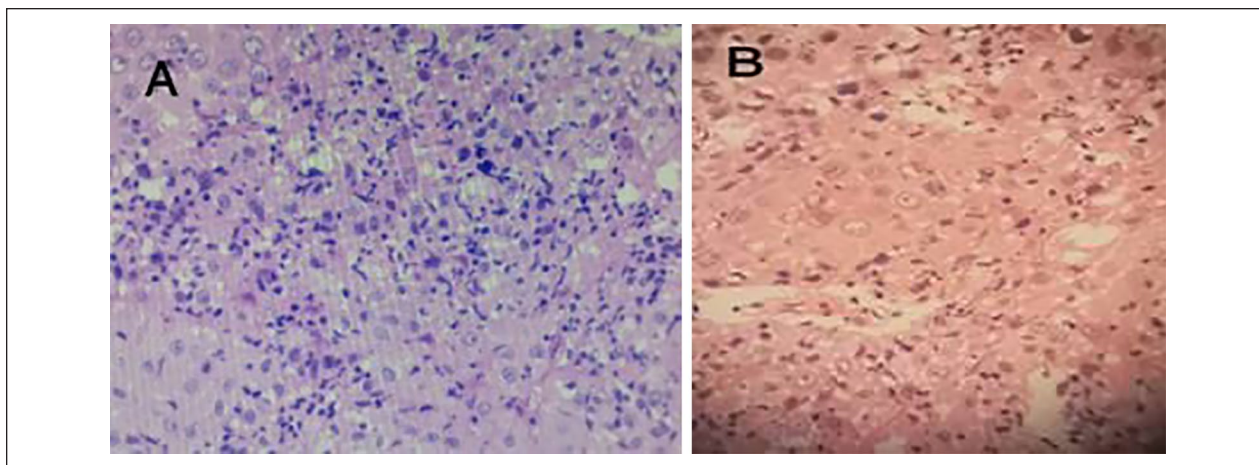


**Figure 3.** A small amount of squamous epithelium, fat, and fibrous connective tissue.

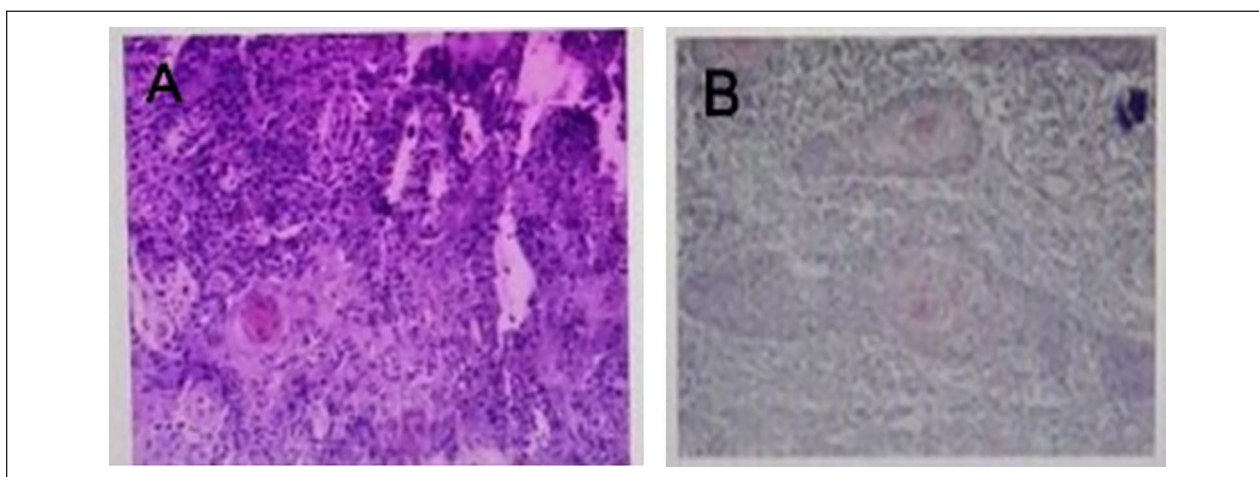
anti-infective therapy for 2 weeks, was able to urinate smoothly, and the mass was significantly reduced again; therefore, he was discharged.

Two weeks after discharge, the patient developed enlarged and painful perineal masses. He was immediately treated with piperacillin-tazobactam, which led to significant improvement. However, the patient recurred the same symptoms 1 month later. Urethroscopy and perineal mass 6-needle biopsy were performed. The urethroscope entered the bladder smoothly from the outer urethra. It was observed the anterior urethra was narrowed to varying degrees, and there were no abnormalities in the urethra and bladder. Pathological findings from perineal tissue suggested hyaline degeneration of fibrous smooth muscle tissues. Thick-walled blood vessels and proliferating fibroblasts with lymphocyte infiltration were seen. The patient developed mass ulceration and perineal discharge later. Considering perineal abscess formation with urinary fistula, perineal abscess incision and drainage were performed. A biopsy was performed during the operation and pathology suggested squamous papillary hyperplasia with inflammatory cell infiltration (Figure 4). Urinary drainage continued after the operation, and white pus and caseous discharge flowed out from the perineal wound; therefore, suprapubic cystostomy and dressing changes were followed.

Since the patient's sinus had not healed for an extended period of time, pathological examination was taken again after 2 months, suggesting bleeding and necrotic tissues. The patient underwent debridement of the perineal wound after dressing changes to improve the wound. The removed tissues were sent to the pathologist. Highly differentiated stage T3N0M0 urethral squamous cell carcinoma (USCC) was diagnosed according to the tumor-node-metastasis (TNM) classification system (Figure 5). The tumor was located in the bulbar



**Figure 4.** Squamous papillary hyperplasia with inflammatory cell infiltration. (A) Nuclear atypia. (B) Squamous cells.



**Figure 5.** Highly differentiated stage T3N0M0 USCC. (A) Keratin pearl. (B) Interstitial infiltration of cancer cell. USCC = urethral squamous cell carcinoma.

membrane of the urethra so radical resection of urethral carcinoma followed by chemo (gemcitabine and cisplatin) and radiation therapy (RT) was proposed. The patient did not agree to the surgical and chemotherapy approach after consultation. Only 50 Gy RT and continued dressings were given for over 5 weeks, resulting in the wound gradually healing. Additional 15 Gy RT was provided to ensure the results prior to the patient's discharge from the hospital. The patient remains in good condition and is still being followed up. There has been no recurrence of USCC for more than 13 months.

## Discussion

LS is a chronic inflammatory disease, and it has been used as a risk indicator for penile SCC by the European Society of Urology Guidelines (Algaba et al., 2002).

Pathology confirmed that LS was found below the tumor, and there was a clear association between LS and penile cancer. The specific existence mechanism between the two was not studied. Urethritis and urethral stricture have been reported as triggers for USCC (Gakis et al., 2013). Palminteri et al. (2007) reported that 12 cases of USCC (2.3%) were found in 522 patients with LS, which was significantly higher than the normal population (1/100,000). Primary urethral carcinoma (PUC) is a rare malignancy accounting for <1% of genitourinary cancers. Among men, the two most important prognostic factors are anatomical location and clinical stage of the tumor. Low stage tumors (T1–2) and tumors involving the fossa navicularis or the penile urethra have a better prognosis than higher stage tumors (>T2 or N+) and lesions involving the bulbomembranous urethra (Dayyani et al., 2014). Both early diagnosis and treatment of USCC

remain challenging although multimodal treatment strategies are believed to be optimal in literature.

The occurrence of MGLSc and USCC together is extremely rare. There is very little discussion on its diagnosis. Patients usually seek medical treatment with symptoms such as urethral obstruction, mass, periurethral abscess, extravasation, urethral fistula, and urethral discharge. Some patients have pain, blood sperm, or hematuria. The early diagnosis of USCC is difficult. In China, there has been only one case report of the LS and USCC combination, published in 2016 by Guo et al. The 56-year-old male had LS with the presence of perineal mass and abscess for over 4 years before the diagnosis of USCC. The 49-year-old patient in this case report was also initially diagnosed with LS of the penis and narrowing of the outer urethra based on the typical skin lesions and urethral strictures of the patient. Although the possibility of cancer was considered early, the perineal mass was significantly reduced after anti-infective therapy and no clear malignant tumor tissues were found after several biopsy examinations.

MGLSc carries a risk of USCC and may be accompanied by perineal or urethral abscesses. The patient in Guo et al.'s report had pain in the perineum with abscesses, while this patient had urethral abscesses. The 49-year-old patient underwent two perineal mass puncture biopsies and two tissue biopsies after the perineal abscess incision and drainage operation without obtaining malignant tissues. Possible reasons include: (1) insufficient tissues were taken during the conventional puncture; or (2) when the abscess was cut and drained, the wound eroded and was full of inflammatory tissues. The boundary between the tumor and surrounding tissues was unclear, which likely caused errors in tissue selection. The histopathology of large pieces of tissue taken at the time of thorough debridement indicated highly differentiated SCC. Possible reasons include: (1) more tissues can be taken than conventional puncture during thorough debridement; and (2) tissues containing tumor can be more accurately located by looking directly than puncture biopsy.

Additionally, it may be necessary for patients diagnosed with LS of the penile/genital skin to have a screening cystoscopy every 6 months to look for stricture or cancer, when difficulty for urination or a mass presents.

The two most important clinical prognostic factors for male USCC are the clinical stage and anatomical location of the tumor. Modern PUC management requires multimodal treatment including chemotherapy, RT, and surgery. Multimodal treatment is also emphasized in the latest European Association of Urology (EUA) recommendations. However, there is no randomized evidence to inform clinical practice. In the retrospective study by Guo et al. (2018), all five patients with high-stage USCC

tumors in the bulbomembranous urethra died of distant metastases between 10 and 14 months after surgery, chemotherapy, and radiotherapy, with the median being 12 months. The patient in this report had radiation treatment alone, has survived more than 13 months, and is still under follow-up. This suggests that radical treatments may not be necessary in patients with LS combined with late-stage USCC in the bulbomembranous urethra, although further research is needed.

In Guo et al.'s study, all 18 patients were aged between 52 and 69 while this patient was diagnosed at 49 years old, which indicates that the incidence can occur in younger males in the East Asian population.

There is also a possibility that the patient had LS and had an unrelated primary urethral cancer that presented as a stricture, followed by perineal fistula or abscess, given the rare incidence or association with LS.

The establishment of multi-institutional data registries of this rare disease will be the only way to accrue substantial high-quality data from which conclusions can be drawn.

## Conclusion

LS with USCC is an extremely rare disease. Early diagnosis and treatment remain challenging. Physicians should consider the possibility of malignancy in MGLSc patients with urethral or perineal mass/abscesses or spontaneous urethrocutaneous fistula, and pathological tissue biopsy should be taken to confirm the diagnosis. Surgically cut tissues are preferred when possible. There is no clear standard for the treatment of this disease. Multimodal treatment including chemotherapy, RT, and surgery is recommended, but the specific role and ideal combination of each method is not yet clear. Randomized trials are needed.

## Declaration of Conflicting Interests

The author(s) declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

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## Ethical statement

Ethics approval was not required for this study.

## Patient consent

The patient of this case report consented to the publication of his information in this article

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