Urethral reconstruction in lichen sclerosus

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Purpose of review
Lichen sclerosus is a chronic skin disease that shows a predilection for the anogenital area and may involve anterior urethra causing stenosis. Surgical options in the management of urethral strictures caused by lichen sclerosus still represent a challenging issue.

Recent findings
Depending on the length and severity of urethral involvement, surgical management of lichen sclerosus urethral strictures can range from a simple meatotomy to a single or complex staged long repair using oral mucosa. Skin grafts or flaps are not recommended because skin could be involved by the disease. Perineal urethrostomy may represent the salvage solution in severe panurethral strictures.

Summary
One-stage or staged repairs using oral mucosa grafts are the most recommended procedures for the treatment of lichen sclerosus urethral strictures, but derivative perineal urethrostomy may play an important role in severe situations. Patients require long-term follow-up and extensive counseling that enables them to fully grasp the chronic and progressive nature of the disease and to deal with it.

Keywords
lichen sclerosus, urethra, urethral stricture, urethroplasty

INTRODUCTION
Lichen sclerosus, previously known as balanitis xerotica obliterans (BXO), is a chronic inflammatory skin disease of unknown cause, which can affect any cutaneous surface, but shows a predilection for the anogenital area [1].

In men, genital lichen sclerosus may involve urethra-causing strictures. Inappropriate treatments may lead to an extension of the stricture, which determines urinary and sexual problems that subsequently cause a dramatic reduction in the quality of life.

Depending on the extension of urethral involvement, surgical repair can range from a minimally invasive treatment as a simple meatotomy, to a more extensive genital and urethral reconstruction.

There are few studies dealing with lichen sclerosus; moreover, they report on a medium series of patients with medium-term follow-up, revealing a gap in the knowledge regarding the correct surgical management of these complex strictures.

We review the latest evidence with the aim of broadening our knowledge of this evolving field.

INCIDENCE AND PATHOGENESIS OF URETHRAL INVOLVEMENT DUE TO LICHEN SCLEROSUS
The overall true incidence of genital lichen sclerosus is unknown [2].

Similarly, the incidence of urethral lichen sclerosus in male patients with lichen sclerosus remains unknown.

Lichen sclerosus represents an emerging cause of urethral strictures. Lumen et al. [3] reported that in their experience, the rate of lichen sclerosus strictures was 4.8%; particularly, lichen sclerosus was responsible for 16% of total penile strictures and 8% of total panurethral strictures, whereas it did not cause bulbar or posterior strictures. In their series of patients with anterior strictures, Barbagli et al. [4] reported that the incidence of lichen...
sclerosus was 14% and the strictures were penile or panurethral. Patients with failed hypospadias repair showed a high incidence of lichen sclerosus [5]. Lichen sclerosus strictures usually affect adults, but children can be affected too. Kulkarni et al. [6] reported a mean age of 50 years (range 11–85).

In a retrospective review of 522 patients with lichen sclerosus, Depasquale et al. [7] discovered that the disease was limited to foreskin and glans in 57%, meatus in 4%, and involved urethra in 20% of the cases. Kulkarni et al. showed that 83.9% of their lichen sclerosus patients presented urethral involvement: specifically, the meatus in 10.6%, the penile urethra in 10.6%, and the penobulbar urethra in 62.7% [6]. However, it has to be considered that the above-mentioned authors work in referral centers where patients with genital lichen sclerosus who develop urethral involvement, surgical repair can range from a simple meatotomy to a more extensive genital and urethral reconstruction. Depending on the length and severity of urethral involvement, surgical management of lichen sclerosus urethral strictures can range from a simple meatotomy to a single or complex staged long repair using oral mucosa.

Patients with genital lichen sclerosus require long-term follow-up and extensive counseling that enables them to fully grasp the chronic and progressive nature of the disease and to deal with it.

**SURGICAL MANAGEMENT**

Depending on the extension of genital lichen sclerosus and urethral involvement, surgical repair can range from a simple circumcision with or without meatomaty, to a more extensive genital and urethral reconstruction.
Urethral reconstruction

Circumcision
Circumcision with its desiccating effect plays an important role in the management of lichen sclerosus.

Depasquale et al. [7] found that 96% of patients with lichen sclerosus limited to the glans and foreskin were successfully treated with only circumcision.

Meatotomy
Meatal strictures can be treated with a simple meatotomy, which, if performed in the early stages of the disease, prevents the extension of urethral stricture. Meatotomy showed a high success rate (>80%), increased to 100% if combined with circumcision [6,16,17].

Patients with lichen sclerosus restricted to the foreskin and meatus showed a high surgery success rate [6,16,18].

Penile and penobulbar urethroplasty
Regarding the repair of penile or panurethral strictures, the debated issues are graft vs. flap, types of grafts, one-stage vs. staged procedures, and dorsal vs. ventral approach.

Graft vs. flap
The penile skin involvement by lichen sclerosus makes the use of penile flaps impossible. Whitson et al. [19] asserted that penile flaps are not recommended in the case of lichen sclerosus and strictures longer than 7 cm. Venn and Mundy [20] performed an analysis of 28 patients with lichen sclerosus who were treated with penile skin flap urethroplasty (12 patients) or two-stage graft urethroplasty using nongenital skin (16). All 12 urethroplasties using genital skin flaps failed. Conversely, out of 16 cases of patients who underwent nongenital skin grafts, only one was a failure.

Types of grafts
Oral mucosa has emerged as the gold standard substitute material for the repair of lichen sclerosus urethral strictures [2,6,7,21-23,24]. This mucosa is resilient to skin diseases. The most popular donor site is the cheek, allowing a repair up to 12 cm long if resorting to bilateral harvesting. Some authors have suggested adjunctive or alternative harvesting sites such as lip or tongue [25], but the minor thickness and handling of the mucosa of these sites as well as the increased risk of oral morbidity due to extensive harvesting must be taken into consideration. Numerous series have demonstrated the benefit of using oral mucosa in urethral repairs, but they remain studies with limited follow-up. Bracka [22] supports its use in lichen sclerosus strictures, but averted that it is not perfect. Some oral grafts can be subjected to contracture as a result of a suboptimal graft take, due to poor vascularity of the wound bed as in lichen sclerosus, or due to an excessive traumatization of the graft during its harvesting and trimming. Further individual biological variation may influence graft healing.

However, full reconstruction of the entire anterior urethra cannot be accomplished through the use of oral mucosa alone, as the size and the total length of the grafts are quite limited.

Meeks et al. [26] showed that in long lichen sclerosus strictures, the use of nongenital skin such as abdominal skin provides a useful alternative graft source for urethroplasty when oral mucosa or genital skin is not available or sufficient. However, their series of patients with lichen sclerosus was quite exiguous (eight patients) and the follow-up was too short, especially if we consider that lichen sclerosus strictures have a high rate of recurrence and that the recurrences develop slowly spanning the course of many years [7].

General opinion is that the use of skin (whether genital or nongenital) is contraindicated, as lichen sclerosus is a skin disease and tends to recur in skin [7,21,24]. For repairs of long lichen sclerosus strictures, Bracka [21] reported that he successfully performed bladder mucosa tube combined with a distal oral graft staged urethroplasty, but this statement is not supported by sufficient evidence in the literature [27], with the exception of experience of Depasquale et al. [7].

One-stage vs. staged procedures
One-stage graft repairs have been suggested in patients with a reasonably wide urethral plate, which could be graft-enlarged, whereas staged graft repairs are usually used for full circumference urethral reconstruction in patients with narrow and unavailable scarred urethral plate.

In one-stage graft repairs, the urethral plate is graft-enlarged using the technique of Asopa, Barbagli, or Kulkarni [6].

The Asopa technique consists of dorsal grafting via ventral urethrotomy approach: the strictured urethra is opened by a ventral midline incision; the urethral plate is longitudinally incised on the dorsal midline down to the corpora; the wings of the urethral plate are laterally mobilized to create a bed in which the graft is sutured; and finally, the graft-augmented urethra is ventrally closed [28].

The Barbagli technique consists of dorsal grafting via dorsal urethrotomy approach: the strictured
urethra is dissected from the corpora cavernosa up to the coronal sulcus and opened along the dorsal surface; the graft is fixed to the corpora; and the ventral urethral plate is rotated and sutured to the grafts [6].

Kulkarni et al. [6] reviewed their experience using dorsal graft urethroplasty in lichen sclerosus strictures, reporting a percentage of success of 100 in eight penile strictures and 91 in 88 panurethral strictures over a mean follow-up of 56 months.

The technique by Kulkarni et al. [29,30] is a one-sided dorsolateral graft urethroplasty, which avoids the circumferential urethral mobilization and preserves the lateral vascular supply to the urethra. Using this new approach, Kulkarni et al. [31] reported a 92% success outcome (mean follow-up 22 months), but only 50% of the 24 patients of their series had lichen sclerosus strictures. Whether preservation of the lateral vascular supply of the urethra is of clinical significance is still under debate. Using the technique by Kulkarni et al., Dubey et al. [32] reported an 88% success rate on a series of 25 patients with lichen sclerosus panurethral strictures and Datta et al. [33] reported a 93.1% success rate on a series of 43 patients.

By the dorso or dorsolateral approach, it could be difficult to detach the scarred urethra from the corpora and it is not easy to graft the undetached glandular urethra [6].

However, it remains an important fact that in one-stage graft augmentations, the diseased urethra is not removed but only enlarged, leading to a risk for recurrence over time [7]. Therefore, many authors state that in order to obtain satisfactory long-term results, the presence of lichen sclerosus requires complete excision of the diseased urethral plate with complex staged reconstruction [7,22,34].

In staged graft repairs, the unusable urethral plate is completely removed and replaced with a graft during the first stage; the new urethral plate will be tubularized during the subsequent stage 6 months later. Depasquale et al. [7] and Bracka [35] were the first to popularize the use of the two-stage oral mucosa graft repair in lichen sclerosus penile strictures with successful results.

Levine et al. [23] reported a success rate of 80% with oral mucosa staged procedures vs. 50% of success rate with one-stage repair.

Conversely, for Kulkarni et al. [6] successful outcomes in staged repairs proved to be lower (73%) than those in one-stage repairs, but this can probably be attributed to the poor conditions of the local tissues in these cases.

Patients undergoing staged repair should be informed that more than two surgical steps could be necessary to reconstruct the urethra. Indeed, one or multiple revisions of the graft bed may be required due to an unsatisfactory healing of the transplanted tissue. This makes the term ‘multi-stage’ procedure more realistic. In their series of staged urethroplasty, Kulkarni et al. [6] showed that 20% of the patients required a revision before the final-stage procedure. Other authors reported graft contracture after the first stage [36,37]. Such results suggest the need for a significant learning curve with graft-staged procedures [22].

Dorsal vs. ventral approach

The dorsal and the dorsolateral are the more frequently used approaches in the one-stage repairs, but oppositely from the ventral approach, they cannot be converted into a staged repair during surgery.

Literature on lichen sclerosus urethral strictures is characterized by small cohort of patients, series of lichen sclerosus strictures mixed with strictures of other causes, different success criteria, and not long enough follow-up. All these factors make it hard to state conclusive sentences on the result of repairs of urethras affected by a recurrent disease as the lichen sclerosus.

However, current evidence suggests that one-stage and staged urethroplasty using oral mucosa grafts are the more frequently used procedures.

Perineal urethrostomy

Perineostomy plays an important role in the management of patients with long strictures, severely scarred urethral plate, failed prior repairs, comorbidities, and those not willing to undergo complex reconstructions [2,38]. Elliott et al. [39] advocated its utility in the modern era. Perineal urethrostomy has shown to be well accepted and with a good impact on the quality of life in patients already accustomed to seated voiding [38,40].

However, lichen sclerosus may involve perineum, causing the stenosis of the perineostomy that is difficult to manage (Fig. 1). In fact, it has been shown that the use of perineal urethrostomy is associated with multiple revisions and a lower success rate (72%) when compared to one-stage procedures [6].

Glands resurfacing

Literature has clarified the strong relationship between lichen sclerosus and penile carcinoma, leading to new surgical solutions such as the total skinning and resurfacing of gans severely involved
CONCLUSION

Genital lichen sclerosus is often associated with urethral involvement and the most appropriate surgical options still represent an open debate. In cases in which lichen sclerosus is restricted to the external meatus, simple meatotomy combined with circumcision proved to be the best option. In anterior urethral strictures, current evidence suggests one-stage or staged oral graft urethroplasty. Perineal urethrostomy plays an important role in severe situations.

Patients require long-term follow-up and extensive counseling, which enables them to fully grasp the chronic and progressive nature of the disease and to deal with it.

The authors showed that the navicularis and penile urethra had the same histologic features that lichen sclerosus presents in penile skin, but lichen sclerosus lesions were not found in the bulbar urethra. The authors showed that the navicularis and penile urethra had the same histologic features that lichen sclerosus presents in penile skin, but lichen sclerosus lesions were not found in the bulbar urethra.

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