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ORIGINAL ARTICLE

Male Health

One-stage dorsal lingual mucosal graft urethroplasty for the treatment of failed hypospadias repair

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The aim of this study was to retrospectively investigate the outcomes of patients who underwent one-stage onlay or inlay urethroplasty using a lingual mucosal graft (LMG) after failed hypospadias repairs. Inclusion criteria included a history of failed hypospadias repair, insufficiency of the local skin that made a reoperation with skin flaps difficult, and necessity of an oral mucosal graft urethroplasty. Patients were excluded if they had undergone a failed hypospadias repair using the foreskin or a multistage repair urethroplasty. Between January 2008 and December 2012, 110 patients with failed hypospadias repairs were treated in our center. Of these patients, 56 underwent a one-stage onlay or inlay urethroplasty using LMG. The median age was 21.8 years (range: 4–45 years). Of the 56 patients, one-stage onlay LMG urethroplasty was performed in 42 patients (group 1), and a modified Snodgrass technique using one-stage inlay LMG urethroplasty was performed in 14 (group 2). The median LMG urethroplasty length was 5.6 ± 1.6 cm (range: 4–13 cm). The mean follow-up was 34.7 months (range: 10–58 months), and complications developed in 12 of 56 patients (21.4%), including urethrocutaneous fistulas in 7 (6 in group 1, 1 in group 2) and neourethral strictures in 5 (4 in group 1, 1 in group 2). The total success rate was 78.6%. Our survey suggests that one-stage onlay or inlay urethroplasty with LMG may be an effective option to treat the patients with less available skin after failed hypospadias repairs; LMG harvesting is easy and safe, irrespective of the patient's age.

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INTRODUCTION

Reoperative urethroplasty following failed hypospadias repair is one of the most challenging procedures for the reconstructive urologist, as its success requires both a functional urethra and a cosmetically acceptable glans appearance.[1–4] Most patients with failed hypospadias repairs have complicated clinical manifestations, including penile curvature, urethral contraction, urethral strictures, chordee, multiple fistulas, and diverticula. Furthermore, the insufficiency of local skin makes reoperation with skin flaps difficult. Therefore, the complication rates in cases of complex hypospadias are not optimistic.[4–6] Various surgical procedures, in one or two stages, have been described for urethral reconstruction, and each associated with varying degrees of success.[1–6]

The aim of this study was to investigate retrospectively the outcomes of patients who underwent one-stage onlay or inlay urethroplasty using LMG due to less available skin after failed hypospadias repairs.

MATERIALS AND METHODS

We performed a descriptive, observational, retrospective study of male patients with failed hypospadias repairs who underwent one-stage onlay or inlay urethroplasty using LMG for reconstructive urethral surgery. The study was approved by our Institutional Review Board.

Inclusion criteria included a history of failed hypospadias repair, the need for a substitute urethroplasty and the insufficiency of the local skin that made reoperation with skin flaps difficult.

Exclusion criteria were patients underwent failed hypospadias using foreskin flap; and patients underwent multistage repairs with buccal or skin grafts or direct repairs without grafts were not included in the study.

Between January 2008 and December 2012, a total of 110 patients with failed hypospadias repairs were treated in our institute. Of the 110 cases, 56 patients underwent one-stage onlay or inlay urethroplasty using LMG. The median age was 21.8 years (range: 4 to 45). Preoperative clinical features of the patients are summarized in **Table 1**. A total of 18 patients (32.1%) had one clinical feature; 31 patients (55.4%) had two complications; 5 patients (8.9%) had three complications; and 2 patients (3.6%) had four complications. A total of 103 complications were reported in our 56 patients.

All patients underwent a variety of past primary hypospadias corrections, ranging from 1 to 6 procedures (mean 1.9). In all, 17 patients suffered from dysuria due to urethral strictures (including meatal stenosis) and had undergone suprapubic cystostomy, while multiple fistulas had developed in ventral side of the penis as a major symptom in the other patients (**Figure 1**).

Surgical techniques

The patients were placed in a standard supine position for penile urethroplasty and in a normal lithotomy position for bulbar urethroplasty. Techniques for harvesting the lingual mucosal graft

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(LMG) have been previously described.[7] Briefly, a standard mouth opener was put into place. The apex of the tongue was passed through via a stitch for traction outside of the mouth to expose the ventrolateral surface of the tongue. In cases with urethral strictures <7.0 cm in length, grafts were harvested from the site 1.0 cm besides apex to lateral mucosal lining of the tongue. In cases with long-segment urethral strictures exceed 7.0 cm, bilateral lingual mucosa needs to be harvested, and grafts were harvested from right lateral side to left lateral side through the apex of the tongue. The required graft was measured and marked with a surgical pen. The harvest graft site was infiltrated with a mixed solution of 0.01‰ epinephrine and 0.9% normal saline. The graft edges were incised, and a full-thickness mucosal graft was harvested beginning at the anterior landmark of the graft. The donor site bed was carefully examined for bleeding, and the donor site was closed using 4–0 polydioxanone running sutures.

After being harvested, graft defatting was performed until all underlying fibrovascular tissue was completely removed, and a few small incisions were made with a blade on the graft before use. The length of the LMG used for reconstruction ranged from 4 to 13 cm, with a width of 1.0 to 2.0 cm. Two techniques were used for urethral reconstruction.

The first technique involved a total onlay LMG substitution urethroplasty in 42 patients (Group 1) with atretic strictures of the urethra, penile curvature, and multiple fistulas. With a midline ventral incision of the penis, the urethra was exposed. After resecting the atretic urethra and scar tissue, and straightening the penis, the LMG with a width of 1.5 to 2.0 cm was positioned into the urethral defect, spread-fixed to the underlying corpora cavernosa using 5-0 or 6-0 polydioxanone sutures separately to ensure close contact between the graft and cavernosa. Its margins were sutured to the mucosal edge of the residual urethra with interrupted sutures. The neo-urethra was constructed over a 10–18 Fr fenestrated silicone stent by multiple-layer coverage achieved using local dartos flaps.[7–9]

Table 1: Patient characteristics (*n*=56)

Surgical techniques	*Characteristic*	*n*
Total onlay urethroplasty	Urethral stricture	17
	Meatal stenosis	5
	Diverticula	3
	Fistula	28
	Penile curvature	32
Modified Snodgrass repair	Urethral stricture	8
	Meatal stenosis	3
	Fistula	7

Figure 1: Multiple fistulas in ventral side of the penis.

The second technique, the modified Snodgrass technique using inlay LMG urethroplasty, was performed in 14 patients with no penile skin scar or scarred urethral plate (Group 2). Parallel incisions were made along the urethral plate from the hypospadiac urethral meatus to the glans tip, and the glans wings were mobilized. A midline incision was made in the urethral plate longitudinally for the subsequent insertion of LMG (**Figure 2a** and **2b**). The LMG with a width of 1.0 to 1.5 cm was inserted between the split urethral plate and stitched to the margins of the healthy urethra using interrupted 5-0 or 6-0 polydioxanone sutures in a tension-free manner. The patch was fixed to the corpora cavernosa (**Figure 2c**). The edges of the augmented urethral plate substitute were then tubularized over an indwelling 10–18 Fr fenestrated silicone stent using 5-0 or 6-0 polydioxanone sutures (**Figure 2d** and **2e**).

Vascularized fascia was mobilized for a second layer of neourethral coverage when possible. The fascia could also be harvested from the scrotum if there were no qualifying fascia in the penile shaft (**Figure 3a–3c**). In addition, in cases underwent glansplasty, procedure began with approximation of the glans wings at the corona with subepithelial polydioxanone sutures, then the meatus was sewn to the glans at the 5- and 7-o'clock positions.

Postoperative management and follow-up

The penis was wrapped with soft elastic, and a roller bandage was used to incorporate the traction suture into the dressing. Continuous wound compression was used for scrotal and perineal incisions. Patients were discharged 4 to 6 days after surgery and transferred to a health recovery hospital.

The urethral silicone stent was left indwelling for 21 days. When urethrography demonstrated that there was no evidence of strictures or fistulas, the suprapubic cystostomy tube was removed. The patients were followed-up every 2–3 months for the first 6 months, then every 3–6 months for the next year and annually thereafter. Success was

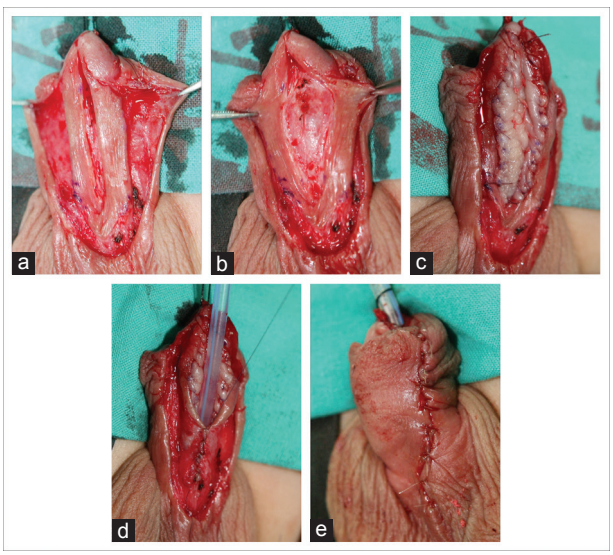

Figure 2: (**a**) Parallel incisions were made along the urethral plate from the hypospadiac urethral meatus to the glans tip. (**b**) A midline incision was made in the urethral plate longitudinally for the subsequent insertion of LMG. (**c**) The LMG was inserted between the split urethral plate and stitched to the margins of the healthy urethra. (**d**) After the glans wings were mobilized, the augmented urethral plate was tubularized over a fenestrated silicone stent to create a neourethra. (**e**) The subcutaneous tissue and skin were wrapped around the neourethra by layer.

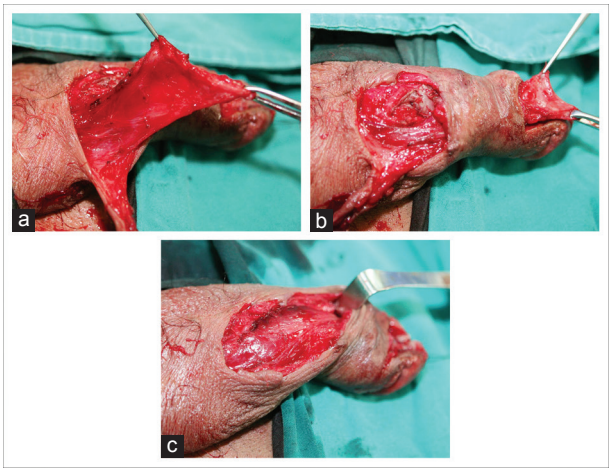

Figure 3: Transplanting of the vascularized fascia for second layer neourethral coverage. (**a**) The vascularized fascia was mobilized. (**b**) The vascularized fascia was pulled through the subcutaneous tunnel. (**c**) The neourethra was covered with vascularized fascia.

defined as having a functional urethra without fistula, stricture, or residual chordee and having a cosmetically acceptable glandular meatus after the completion of all secondary procedures.

RESULTS

The mean follow-up time was 38.1 months (range: 9–90 months). 44 patients voided without difficulty in the postoperative period, with urinary peak flow rates ranging from 14 to 46 ml s^{-1} (mean, 23.5 ml s^{-1}) and the overall success rate was 78.6% (44/56). Postoperative complications developed in 12 of 56 patients (21.4%), including urethrocutaneous fistulas in 7, neourethral strictures in 5 (meatal stenosis in 2, proximal anastomotic site stricture in 2, neourethral strictures in 1) (**Table 2**). Meatoplasty was performed in 2 patients with meatal stenosis, after which the patients achieved a urinary peak flow of 37.3 ml s^{-1} and 28.7 ml s^{-1} at the 15 and 12 months follow-up times, respectively. A proximal anastomotic site stenosis were developed in 2 patients, urinary peak flow rates decreased from 22 ml s^{-1} and 26 ml s^{-1} in the immediate postoperative period to 14 ml s^{-1} and 12 ml s^{-1} at that time. However, dilatation alone was performed once every month for the management of proximal anastomotic site stenosis. After 4 and 6 months respectively, the patients voided well, with a urinary peak flow of 23.1 ml s^{-1} and 25.3 ml s^{-1} at 13 and 22 months of follow-up, respectively. Six patients with urethrocutaneous fistulas were repaired with salvage procedure, succeed in 4 and failed in 2. Two patients await further reoperation.

There were no immediate postoperative complications associated with the oral donor site. All patients reported minor pain and slight difficulty in moving their tongue 3 days postoperatively (**Figure 4**).

DISCUSSION

Successful treatment of hypospadias should achieve the following objectives: correcting the penile curvature; creating a cosmetically acceptable penis; producing a normally sited meatus; ensuring that the patient voids without difficulty; and decreasing the incidence of complications.

The procedure for reoperation after failed hypospadias repair should be based on the following: the site of the abnormality, the severity of scar tissue involving the penile skin, the tension of penile skin, the amount of foreskin remaining, the site and appearance of meatus, the severity of the penile curvature, and the presence of urethral

Figure 4: Minor pain and slight difficulty in moving the tongue 3 days postoperatively.

Table 2: The surgical repair and complication

Surgical techniques	*n*	*Complication*	*n*	*Success (%)*
Total onlay urethroplasty	42	Anastomotic stenosis	2	32 (76.2)
		Meatal stenosis	2	
		Fistula	6	
Modified Snodgrass repair	14	Neourethral strictures	1	12 (85.7)
		Fistula	1	

strictures or urinary fistulas. The treatments for failed hypospadias repair include a variety of surgical techniques using different types of tissues for substitution, such as local skin flaps, free full-thickness skins or mucosal grafts. The multiple choices for the substitutes make this procedure one of the most controversial operations in the field of urology.[1–6]

The two choices of surgical procedures include the foreskin pedicle flap urethroplasty and the free grafts urethroplasty, which differ according to the different types of substitutes. It is widely believed that skin flaps, even those suffered from previous operations, have more reliable vascularity than free grafts while being placed on a poor blood bed.[1,10,11]

In cases with penile skin scar, scarred urethral plate or fistulas that lacked an available original urethra or had a shortage of the local foreskin, free grafts substitution urethroplasty was the only choice for surgical procedures. Considering the advantages of being easy to harvest and incurring minimal trauma, the lingual or buccal mucosa is part of a suitable and effective technique for these patients. Before 2008 year, we routinely performed substitution urethroplasty using BMG to treat the patients with less available skin after failed hypospadias repairs, however, size limitations associated with donor sites cannot be easily resolved, although unilateral buccal mucosa can provide a maximal length and width of 6 cm × 1.5 cm. Larger grafts can be obtained with bilateral buccal mucosa harvest, but this remarkably increases the incidence of complications, such as pain, oral numbness, functional impairment of salivary glands, difficulty in opening mouths, and lip deviation or retraction.[12,13]

In 2006, Simonato *et al*.[14] first described the use of tongue tissue as an alternative donor site for graft urethroplasty, and they achieved good functional and esthetic results. To further explore Simonato's results, we investigated the use of lingual mucosa as a novel substitute for urethral reconstruction in 10 dogs before performing this operation in patients with urethral stricture.[15] Alternatively, LMG retains some buccal mucosa advantages without apparent notable disadvantages. We used canine lateral or ventrolateral lingual mucosa, combined with buccal mucosa, for the combined urethroplasty. The results demonstrated that LMG had good biocompatibility with native urethral tissue, no signs of stricture appeared at anastomosis sites.[16] Furthermore, based on our study, LMG of up to 14.0 cm in length could be harvested in adult for urethral reconstruction,[7] while BMG is difficult to exceed 12.0 cm even with bilateral BMG harvesting. Consequently, the lingual graft urethroplasty has become our standard procedure over the past year.[7]

Our opinion was that surgeons can easily access the tongue for larger graft sizes compared with the buccal mucosa harvested. Furthermore, we harvested grafts from both the ventral and lateral mucosa of the tongue to obtain a wider graft and create stable neourethras. This reduced the risk of stricture recurrence at the distal or proximal graft anastomosis sites. Postsurgical complication rates at donor sites are relatively low, and when it occurs, resolution is available during the follow-up period.[17,18]

Our investigation showed that one-stage urethral reconstruction using LMG may be a feasible option for patients with less available skin after failed hypospadias repairs, In our group 1, 42 patients underwent a LMG onlay urethroplasty. LMG (widths of 2.0 cm under slack condition) was adopted as a substitute for the urethra to form an oblique anastomosis with the original urethra after completely excising the scar tissue and correcting the residual curvature.

The complication rates among patients with less available skin after failed hypospadias repairs were much higher than those in primary repairs, increasing the likelihood of recurrent fistulation and stricture. This observation is mainly due to the less vascularized ventral penile shaft skin, which arises from a deficient ventral dartos in hypospadias. The latter condition makes fistulation more common for a second reason, as it is more difficult to find enough dartos to interpose between the urethral and skin closures.

In our experience, a vascularized fascia needs to be transplanted to cover the neourethra or to be inserted between the neourethra and sutured skin if possible. If there is no suitable fascia in the penile shaft, the fascia can be harvested alternatively from the scrotum (**Figure 3a–3c**). Otherwise, an island flap with vascularized fascia needs to be transplanted to cover the neourethra to avoid the development of a urinary fistula. In addition, the success rate of the procedure can be increased by avoiding the excessive separation of the tissues and using tension-free sutures.

The TIP technique (Tubularized incised plate technique), a LMG inlay urethroplasty, was also a suitable and effective technique for patients with no penile skin scar or scarred urethral plate, providing a satisfactory success rate. In our experience, the TIP technique is simple to operate, has a low complication rate and reliably creates a glandular meatus of normal appearance. In our group 2, 14 patients underwent the TIP technique (LMG inlay urethroplasty), and postoperative complications developed in two patients, including urethral stenosis in one and urethrocutaneous fistula in one.

In our study, the main limitation was the small sample size, making this report more of a case series than a retrospective informative study. No statistical analysis was performed, and a larger series will require the performance of multivariable logistic analyses aimed at discovering the independent predictors of success when various materials are used.

The treatment for failed hypospadias repairs is often a difficult and challenging procedure. The most common complications are anastomotic stenosis and urethrocutaneous fistula. One-stage free graft urethroplasty using LMG may be a feasible option for patients with less available skin after failed hypospadias repairs, and LMG harvesting is easy and safe, irrespective of the patient's age.

AUTHOR CONTRIBUTIONS

YMX conceived and carried out the study. YMX, HBL, QF, YLS, JZ and HX participated in the operations. YMX, HBL performed statistical analyses and drafted the manuscript. YMX, HBL, QF, YLS, JZ and HX participated in the patient follow-up. All authors read and approved the final manuscript.

COMPETING INTERESTS

The authors declare that they have no competing interests.

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