Urethral Reconstruction in Balanitis Xerotica Obliterans

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Introduction

Balanitis xerotica obliterans (BXO) is a progressive sclerotic process of unknown etiology that may involve the prepuce, glans, and urethra. Stuhmer [1] first described BXO in 1928 in Germany. It is also known as kraurosis penis. It is a male genital variant of lichen sclerosus et atrophicus which affects the prepuce, glans, meatus, and anterior urethra in varying proportions and combinations [2]. BXO was regarded as an exclusively adult disease till 1962 when a 7-year-old boy was diagnosed and documented to be afflicted with BXO [3]. Now it is considered to be a common cause of acquired phimosis and meatal stenosis in prepubertal boys.

The clinical presentation and severity of BXO can vary markedly. The onset and progression of disease may be insidious and indolent or aggressive and florid. Urethral involvement is seen in about 25–30% of cases [4]. It starts at the meatus, with a tendency to form superficial adhesions between the meatal lips in milder cases and then the typical dense ivory white fibrosis in more severe disease (fig. 1). In general the disease seems to spread proximally from the meatus in a confluent manner. In long-standing cases, urethral mucosal involvement and spongiosis can spread proximally as far back as the prostate. However, bladder mucosa has not been found involved so far. On physical examination penile urethra feels like a thickened cord. Occasionally mucosal skip lesions have been noted beyond the apparent posterior limit.

Key Words
Balanitis xerotica obliterans · Substitution urethroplasty · Buccal mucosal graft · Lingual mucosal graft

Abstract

Objectives: To review the results of utilizing different grafts for substitution urethroplasty for anterior urethral stricture caused by balanitis xerotica obliterans (BXO). Methods: 153 patients who underwent substitution urethroplasty for anterior urethral strictures were included in this study. The stricture length varied from 3.8 to 16.4 cm (mean 10.2 cm). In 32% of the patients (49), local genital, penile (18), perineal (16) and scrotal (15) skin grafts were used. Over the 3 years our standard treatment policy has been to utilize a free mucosal graft from a non-genital area. Buccal mucosa was the most preferred, utilized in 74 (48.3%) patients and bladder mucosa in 12 (7.8%). Recently we have used lingual mucosal grafts in 18 (11.7%) patients for substitution urethroplasty. Results: The overall success rate for non-genital mucosal graft was 92.2%. The remaining 8 patients required more than one supplementary procedure postoperatively but none required a second urethroplasty. Of 49 patients who underwent substitution urethroplasty utilizing genital skin, the success rate was only 4%. 16.3% required one and 14.3% required multiple auxiliary procedures postoperatively. 34 patients (69.4%) required subsequent urethral reconstruction. Conclusions: A free graft urethroplasty using non-genital skin is recommended for anterior urethral stricture related to BXO.

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BXO involvement of the urethra poses a surgical challenge with the goals of restoring adequate urinary flow with minimal risk of recurrence and achieving good cosmesis with nearly normal-appearing meatus. Various surgical procedures have been described in the literature for urethral reconstruction in BXO with varying degrees of success [5–7]. Excision of the involved urethra and substitution urethroplasty is the modality of choice [8]. The currently most widely accepted recommendation is the use of non-genital mucosal grafts of which buccal mucosal graft is the most popular [8, 9]. Controversies exist in the literature about advantages and disadvantages of one-stage versus two-stage repairs [10]. However, many published studies have recently confirmed equally good results with one-stage mucosal graft substitution urethroplasty [11–13].

The incidence of BXO is considerably higher in our region of practice probably on account of non-performance of neonatal circumcision as a custom. As a result we have been performing urethral reconstructions for BXO regularly using various surgical techniques. Recently the use of lingual mucosa for urethral reconstruction has been published as a pilot study [14]. We have already used lingual mucosa for urethral reconstructions in 30 patients so far of which 18 had BXO-related strictures. Herein we report our results and experience of urethral reconstruction in BXO. To the best of our knowledge this is the largest single-center study involving results of urethral reconstruction in BXO-related strictures and the first study to analyze the use of lingual mucosal grafts for urethral reconstruction in BXO.

### Materials and Methods

We retrospectively reviewed the records of all patients who underwent substitution urethral reconstructions at our institute from January 2001 to December 2006. We found records of 337 patients who had substitution urethroplasty. Out of these, 172 patients were clinically documented cases of BXO. The records of these patients were then reviewed to gather data about patient demographics; site, length and characteristics of stricture; type of procedure performed; surgical outcome; follow-up, complications and need for further procedures. Of the 172 patients, data were available for 153 patients and these were analyzed for the purpose of this study. Patients and stricture characteristics are shown in table 1. Figure 2 shows the typical appearance of anterior urethral stricture related to BXO on retrograde urethrogram.

In earlier years we used local genital skin for substitution urethroplasty but long-term results were disappointing. Over the last 3 years our standard treatment policy for BXO-related strictures has been to utilize a free mucosal graft from a non-genital areas of which buccal mucosa was the most preferred till recently when we found lingual mucosa to give equally good results with much easier harvesting and minimal donor site morbidity. Our last 18 cases have all been performed with lingual mucosal substitution.
with good short-term results. In general we have gradually shifted in our approach to dorsal onlay of free grafts as opposed to ventral onlay because of better results with dorsally placed grafts [9]. Currently our routine practice is to completely mobilize the urethra from meatus till the proximal healthy portion and then making a dorsal urethrotomy from meatus till the normal urethra proximally as described by Kulkarni et al. [15]. Invagination of glans and meatus through the perineal incision makes the meatal part of the procedure quite simple. Except for strictures limited to distal penile urethra, we routinely use a perineal approach to mobilize the urethra. For distal penile strictures a degloving circumferential incision is adequate. The majority (107/153) of procedures were performed as single-stage repairs. However, two-stage procedures were performed in 46 patients (30%) who had unsuitable local factors like severe local infection, urinary fistulae, badly scarred urethral bed and severe fibrosis with multiple areas of complete obliteration of lumen. Again, most of the two-stage procedures were performed in earlier years and our current trend is to offer single-stage repairs to all but few of the patients with local complicating attributes.

All patients underwent a thorough clinical examination which included examination of the local area along with the examination of the oral cavity to assess the condition of the buccal and lingual mucosa. Besides the routine laboratory and biochemical tests, all patients underwent a retrograde urethrogram to evaluate the location, length, and extent of the urethral stricture. Treatment options were discussed with the patient and nature of procedure with likely morbidity and complications explained to the patients. All procedures were performed under regional anesthesia and short-term nasal/oral endotracheal intubation was done during the harvesting of the graft from the oral cavity. A Penrose drain was routinely kept in all patients for 48 h postoperatively. The patients in whom oral cavity grafts are taken were permitted oral sips from the first postoperative day and clear liquids from the second postoperative day. The patients were discharged from the hospital on 4th/8th postoperative day. A pericatheter contrast study was performed on the 21st postoperative day and then the catheter was removed if no extravasation was seen. A uroflowmetry was performed at 3 months and at 1 year. The procedure was considered successful if the patient’s flow rates were >20 ml/s and no additional procedures were required postoperatively.

### Results

A total of 153 patients with BXO-related urethral strictures underwent urethral reconstruction at our center from January 2001 to December 2006. The details of various surgical procedures performed are illustrated in Table 2. The results were analyzed in terms of: overall complete success; patients requiring one minor procedure (internal urethrotomy or urethral calibration); patients requiring more than one subsequent minor procedure; complete failure with need of second reconstructive procedure, and minor and major complications associated with the procedure. The overall complete success rate was 57.5%, i.e. 88 patients were voiding with good flow without need for any auxiliary procedures. Another 18 patients (11.8%) required one minor ancillary procedure (internal urethrotomy or urethral calibration); patients requiring more than one subsequent minor procedure; complete failure with need of second reconstructive procedure, and minor and major complications associated with the procedure. The overall complete success rate was 57.5%, i.e. 88 patients were voiding with good flow without need for any auxiliary procedures. Another 18 patients (11.8%) required one minor ancillary procedure (internal urethrotomy or urethral dilation in 7 patients) which was adequate for normal voiding. 13 patients (8.5%) required multiple supplementary procedures like urethral dilatations/internal urethrotomies/self-calibrations but were spared another reconstructive procedure. 34 patients (22.2%) had complete failure of the procedure and had to undergo a second urethral reconstruction subsequently.

<table>
<thead>
<tr>
<th>Method of repair</th>
<th>Patients n (%)</th>
<th>Complete failure (need of subsequent reconstructive procedure)</th>
<th>Need for one ancillary procedure</th>
<th>Need for multiple ancillary procedure</th>
</tr>
</thead>
<tbody>
<tr>
<td>Two-stage repair</td>
<td>46 (30)</td>
<td>12</td>
<td>4</td>
<td>2</td>
</tr>
<tr>
<td>Single-stage repair</td>
<td>107 (70)</td>
<td>22</td>
<td>14</td>
<td>11</td>
</tr>
<tr>
<td>Dorsal onlay</td>
<td>105 (68.6)</td>
<td>14</td>
<td>5</td>
<td>3</td>
</tr>
<tr>
<td>Ventral onlay</td>
<td>48 (31.4)</td>
<td>20</td>
<td>13</td>
<td>10</td>
</tr>
<tr>
<td>Penile skin graft</td>
<td>18 (11.7)</td>
<td>16</td>
<td>1</td>
<td>1</td>
</tr>
<tr>
<td>Perineal skin graft</td>
<td>16 (10.4)</td>
<td>11</td>
<td>2</td>
<td>2</td>
</tr>
<tr>
<td>Scrotal skin graft</td>
<td>15 (9.8)</td>
<td>7</td>
<td>5</td>
<td>2</td>
</tr>
<tr>
<td>Bladder mucosal graft</td>
<td>12 (7.8)</td>
<td>0</td>
<td>2</td>
<td>1</td>
</tr>
<tr>
<td>Buccal mucosal graft</td>
<td>74 (48.3)</td>
<td>0</td>
<td>7</td>
<td>6</td>
</tr>
<tr>
<td>Lingual mucosal graft</td>
<td>18 (11.7)</td>
<td>0</td>
<td>1</td>
<td>1</td>
</tr>
<tr>
<td>Total</td>
<td>153 (100)</td>
<td>34</td>
<td>18</td>
<td>13</td>
</tr>
</tbody>
</table>
Of the 104 patients who underwent substitution urethroplasty utilizing non-genital tissues (mucosal grafts from oral cavity/bladder) the complete success rate was 82.6%, i.e. 86 patients were symptom-free after the procedure. Another 10 patients (9.6%) required one ancillary procedure before achieving a successful outcome. Thus the overall success rate for mucosal graft substitution urethroplasty was 92.2%. The remaining 8 patients required more than one supplementary procedure postoperatively but none required a second urethroplasty.

Of the 49 patients who underwent substitution urethroplasty with local genital tissue grafts, only 2 were symptom-free without the need of auxiliary procedures, a complete success rate of 4%. 8 patients (16.3%) had a satisfactory outcome after one ancillary procedure in this group of patients, thus giving an overall success rate of 20.4%. 5 patients required multiple auxiliary procedures postoperatively while 34 patients (69.4%) had complete failure of the procedure and required consequent urethral reconstruction.

Immediate postoperative complications were wound infection in 19 patients (12.4%), hematoma/bleeding in 6 patients (3.9%), and donor site complications like minor oral numbness (15.7%), tightness of mouth (24.8%) and mild immediate postoperative oral pain in the buccal mucosa donor site. In patients who underwent lingual mucosa graft urethroplasty, the donor site complications were noticeably minimal except for minor pain in first 48 h after surgery.

Minor urinary extravasation was seen in 13 patients during the pericatheter study at 3 weeks which responded to prolongation of an indwelling catheter by another week. 8 patients had major urinary extravasation, all of whom subsequently resulted in failure of the procedure. 24 patients had minor stenosis at the anastomotic site which responded to single internal urethrotomy subsequently. 7 patients developed meatal narrowing within 3 months and needed calibration/meatotomy. All of these patients had undergone penile skin graft urethroplasty and subsequently developed recurrent strictures.

Discussion

BXO-related urethral strictures have traditionally been difficult to treat and results of conventional surgical procedures have been disappointing. Conservative management with repeated urethrotomies and dilatations in these cases usually results in worsening of the condition. The diseased segment needs to be substituted with extra-genital mucosal grafts in order to provide a long-term recurrence-free outcome. Use of genital skin grafts invariably results in restricturing, usually within the first 2–3 years and may occur up to 10 years later [8]. Venn and Mundy [5] reported an almost 100% recurrence rate for one-stage urethroplasty with genital skin flaps. Depasquale et al. [6] also observed a high restrictrate when penile skin was used for one-stage reconstruction. Therefore, they recommended a two-stage approach using non-genital skin or Buccal mucosa for BXO urethral strictures.

The results of our study validate the fact that single-stage mucosal graft substitution urethroplasty offers these patients an excellent chance for long-term satisfactory results. Use of buccal mucosa for urethral reconstruction is a well-accepted procedure and many reports have validated the use of buccal mucosa [5]. Barbagli et al. [16] first described the use of dorsally placed free grafts for repair of penile urethral strictures. Dubey et al. [10] reported excellent intermediate-term results with use of dorsal buccal mucosal grafts in BXO strictures. Brady et al. [17] in their study on 24 patients, of whom 10 had BXO, using single-stage buccal mucosal urethral reconstruction reported a success rate of 87.5%. Our results conform to these results when using non-genital mucosal grafts for urethroplasty. The perineal approach for dorsal onlay buccal mucosal graft urethroplasty was first reported by Kulkarni et al. [15]. We have found this approach especially useful for dealing with meatus which is invariably involved in cases of BXO and resultant cosmesis after dorsal onlay graft is extremely satisfying.

The use of lingual mucosal grafts for urethral reconstruction was first reported in a pilot study by Simonato et al. [14]. We have subsequently used lingual mucosal grafts in 30 patients of whom 18 had BXO-related long urethral strictures. To the best of our knowledge this is the first study to analyze the results of lingual mucosal grafts in BXO-related urethral strictures. A separate study is currently underway at our center to compare the results of buccal and lingual mucosal grafts in BXO strictures. In our initial experience, lingual mucosa is a good substitute for buccal mucosal grafts as it has similar anatomical properties of thick mucosa, high content of elastic fibers, thin lamina propria along with the ease of harvesting. Furthermore, the incidence of donor site complications is minimal and mild oral discomfort in the immediate postoperative period is all that the patients experience. However, long-term data are still lacking and further follow-up of these patients will reveal whether any long-term sequelae follow after removal of lingual mucosa.
In earlier years, genital skin was utilized for urethral reconstruction in BXO-related strictures and despite good early results, the recurrence rate of stricture was unusually high [6–8]. Our results with use of local skin flaps have similarly been disappointing and genital skin is no longer used by us for urethral reconstruction in BXO strictures. Furthermore, we have observed that even two-stage procedures utilizing local skin have a poor outcome which reinforces the fact that local skin is not a suitable substitute for BXO-related strictures as the disease recurrence is usually seen within 1–2 years. Also, sexually active young men are likely to prefer a one-stage procedure with minimum violation of the penile skin. Use of two-stage procedures is still required in certain situations like extensive local fibrosis or multiple areas of complete urethral obliteration where a primary excision of fibrotic tissues and diseased urethra is done along with application of a buccal or lingual mucosal graft which is then tubularized at a second stage after about 4 months later.

**Conclusion**

BXO involvement of the urethra presents the challenging objective of restoring adequate urinary flow whilst obviating the risk of recurrence. To treat established urethral disease, substitution urethroplasty is the modality of choice. Substitution urethroplasty with skin grafts (genital or extragenital) provides no cure. The short-term results may be excellent but if such patients are followed up long enough a recurrent BXO stricture is almost inevitable. We preferred one-stage repair with excellent results but two-stage repair was done if urethral plate or local conditions were unfavorable. We conclude that when urethroplasty is required for BXO, non-genital mucosa (buccal mucosa, lingual mucosa, bladder mucosa) should be used for substitution as the disease seems always to recur in genital skin urethroplasty. Use of lingual mucosal grafts has opened up a new avenue for these difficult-to-treat urethral strictures. However, long-term results of lingual mucosal grafts are still awaited.

**References**
