Original Research Article

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10 year institutional experience of use of buccal mucosal grafts for complex urethral reconstruction for varied indications with its outcome

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ABSTRACT

Background: The objective of the study was to evaluate long term efficacy and outcome of use of buccal mucosal graft (BMG) for urethral reconstruction in varied urologic conditions in children.

Methods: We retrospectively reviewed the medical records of 41 patients from 2009 till 2019 in our institution in which BMG was used for urethral reconstruction. Clinical findings along with surgical techniques used were noted for these patients. Postoperative outcome and complications were evaluated.

Results: Mean age was 6.8 years and mean follow up was for 4 years. Out of 41 patients, BMG for substitution urethroplasty was used in 25 cases of hypospadias, 4 cases of urethral stricture, 6 cases of 46 XY disorders of sexual disorders, 4 cases of Y-duplication of urethra, and 2 cases of redo-epispadias repair. 11 patients underwent one stage repairs with a success rate of 63% and 30 patients underwent two stage repair with a success rate of 66%. Analysis and comparison of the outcome in relation to the type of repair, meatal position and number of surgical procedures prior to BMG urethroplasty was statistically insignificant.

Conclusions: Buccal mucosa is an ideal graft substitute for urethroplasty. Two stage reconstructions has a slightly higher success rate than one stage reconstruction but the choice of the technique must be based on patients characteristics and on surgeons preference.

Keywords: Buccal mucosal graft, Two stage reconstruction, One stage reconstruction

INTRODUCTION

Pediatric surgeons and reconstructive urologist face a formidable challenge of complex urethral reconstruction in children in cases of hypospadias cripples, urethral strictures (traumatic, iatrogenic, and idiopathic etiologies), and disorders of sexual differentiation (DSD), Y-duplication of urethra and redo-epispadias repairs. Repeated attempts at surgical repair may leave the penis scarred, hypo vascular and short. Application of buccal mucosal graft (BMG) for reconstruction have emerged as reliable substitute as it has a thick non-keratinized epithelial layer and a well-vascularized and thin lamina propria, favoring early inosculation. Both single and multi-staged approaches have been described with

favorable outcomes. However, less data exists for the use of BMG in the repair of complex urethral reconstruction in pediatric patients. In our study we analyze the long term efficacy and record the complication rate along with modifications required for the use of BMG for urethral reconstruction in children.

METHODS

We retrospectively collected data from the medical records between 2009 and 2019 of all children of our institution in whom BMG was used in at least 1 urethral reconstruction procedure regardless of indication. Demographic data, urologic comorbidities, initial diagnosis, and number of procedures prior to use of BMG was noted. Perioperative

data included type of repair, number of stages necessary, graft characteristics, immediate complications, and length of time between stages were noted. We recorded duration of follow-up for each patient and also the long-term complications. For 1-stage repairs, follow-up time was calculated as time since the BMG repair. For patients undergoing 2-stage repairs, follow-up time was calculated as the time since their 2 stage.

Operative technique

Donor site

Oral mucosa was harvested from the cheek or where a long length is needed, the inner cheek strip was taken continuous with the lip. A sublingual graft was also used in 1 case. Donor graft site was outlined before harvesting, making sure that at least an 8-mm distance is maintained away from the papilla of the parotid duct and 1 to 1.5 cm behind the commisure of the mouth to prevent distortion of the vermillion border during dissection.² A retraction of about 20% of the grafted area usually occurs between stages; therefore, harvesting a graft slightly larger than required is recommended. Submucosal local anesthesia with 1:100,000 epinephrine was administered to decrease bleeding and define tissue planes. Tenotomy scissors was used to carefully dissect to the lamina propria separating the graft from the bucopharyngeal fascia. Lip donor sites were left open as this approach may reduce postoperative pain and allowed for potential re-harvest in the future. Irrespective of the site, harvested graft was appropriately de-fattened by placing the graft spread over a finger and removing the fat by sharp dissection with scissor and stored in saline for later use.

First stage buccal mucosal graft urethroplasty

During the first stage, complete release of chordee and excision of scarred skin from the ventral aspect of penis was carried out. A saline erection test was performed to make sure complete release of chordee and straightening of the penis has been achieved. Any minor residual chordee was dealt with a dorsal nesbitts procedure. Glans wings were raised with preserving of the urethral plate at the proposed neomeatus to avoid meatal stenosis. The graft was then secured to the ventral aspect of the corpora. The perimeter was sewn first and then multiple quilting stitches were placed through the graft in multiple parallel rows along the shaft so that the graft will not heap up into folds when the tension is released and also to reduce the potential risk of a hematoma. A V-shaped cut was made in the dorsal wall of the existing meatus and the graft was placed as an inset on the defect to decrease the chances of formation of circular stricture at this site. A Foleys urethral catheter was passed and a firm tie- over dressing is given to ensure that no hematoma can collect under the graft. The dressing was removed on fifth postoperative day and parents are instructed on the use of cotton bud and antiseptic ointment to make sure that the glans margins do not stick together in the early postoperative period. Steroid

and testosterone gel was applied later after 14 day postoperatively to improve graft uptake (Figure 1).



Figure 1: BMG applied to form the urethral plate in the first stage.

Second stage buccal mucosal graft urethroplasty

The stage 2 was performed at 6 months after the first stage. If the graft bed is smooth and supple, 'U' shaped incision was made with the base of the curve of U right over the edge of native meatus to minimize the chance of creating the ventral pouch. The width of the graft was adjusted according to the size of the penis and size of the catheter, so as not to make patulous tubes. The second layer which was usually called the waterproofing layer' is raised from the Dartos fascia or a tunica vagnalis flap was used to cover the suture line of the first layer in such a way that the two suture lines were separated and is followed by skin closure. The catheter was removed on the tenth postoperative day, after which urinary stream is assessed. The patient was followed up at 2 weeks, 6 weeks, 3 months, 6 months and then annually for assessment of outcome of surgery and to look for any complications (Figure 2).



Figure 2: Postoperative picture after second stage BMG urethroplasty.

RESULTS

During the period from 2009 to 2019, there were 41 patients in our institution in whom BMG was used for urethral reconstruction. The mean age at the time of first stage was 6.8 years (4-16 years) with a mean follow up was 4 years (1-10 years). Table 1 illustrates the characteristics of our patients prior to their buccal urethroplasty. Out of 41 patients, buccal mucosal graft was used in 25 cases of hypospadias, 4 cases of urethral stricture, 6 cases of 46 XY disorders of sexual disorders, 4 cases of Y-duplication of urethra, 2 cases of redoepispadias repair. Four patients had associated cryptorchidism and one had inguinal hernia along with hypospadiasis. The initial meatal opening was proximal (proximal shaft, penoscrotal or perineal) in 29/41 cases (73%). Four patients of 46 XY disorders of sexual disorders and four patients of proximal hypospadias with severe chordee had a buccal urethroplasty as their first repair. Of the patients with prior repairs, 80% had at least one revision of their initial repair prior to BMG. Good urinary stream evidenced by patient's ability to urinate in standing position without spraying or dribbling with meatal opening at the tip of penis was achieved in 27 patients (66%).

Table 1: Pre-buccal graft repair characteristics.

Characteristics	Number		
Diagnosis			
Hypospadiasis	25		
Uretheral stricture	4		
DSD	6		
Uretheral duplication	4		
Extrophy epispadias	2		
Initial meatal location			
Distal penile	7		
Mid penile	4		
Proximal penile	29		
Number of procedures prior to buccal graft			
0	8		
1	15		
2	12		
3 or more	6		

A 1st stage repair was elected for 11 patients in total. Out of which 3 cases were of Y-duplication of urethra. In case 1: the long gap from root of scrotum to tip of glans was bridged with free bladder mucosal graft and the distal 1 cm of urethra was constructed with buccal mucosa graft. In case 2: patient had developed multiple strictures following ventral to dorsal urethrostomy requiring subsequent perineal urethrostomy after which he underwent a free buccal mucosal tube urethroplasty. In case 3: patient was subjected to free buccal mucosal tube urethroplasty developed perineal abscess with stricture requiring subsequent perineal uretherostomy after which he underwent a redo-free buccal mucosal tube urethroplasty.

One case of traumatic urethral stricture after undergoing failed end to end urethroplasty underwent buccal mucosal ventral onlay urethroplasty. Patient developed straining of micturation postoperatively requiring urethral dilatation. Three patient of proximal hypospadias underwent ventral onlay repair with one developing fistula which was corrected later. Two cases of redo-epispadias repair underwent inlay free mucosal graft repair to bridge the gap between transected ends of urethra. One patient of 46 XY DSD with multiple surgeries underwent urethral reconstruction using inlay BMG followed by tunica vaginalis cover with no complications. Similarly one case of distal hypospadias with multiple surgeries underwent urethroplasty using inlay BMG graft but was followed by a subcoronal fistula which was repaired. A 2nd stage repair was elected for 30 patients in total. The median interval between the stages was 6 months. The BMG uptake after the first stage was 96%. Buccal graft repair characteristics are summarized in Table 2.

Table 2: Buccal graft repair characteristics.

Buccal graft surgical approach	Number
One stage repair	11
One stage tube	3
One stage onlay	4
One stage inlay	4
Two stage repair	30

Table 3: Analysis and comparison of the outcome and complications in relation to one stage and two stage repairs.

Variable	One stage repair, N (%)	Two stage repair	P value
Outcome			
Success	7 (63)	20 (66)	0.102
Failure	4 (36)	10 (33)	
Fistulae formation			
Yes	2 (18)	3 (10)	0.159
No	9 (81)	27 (90)	
Meatal stenosis			
Yes	1 (9)	3 (10)	0.147
No	10 (91)	27 (90)	
Restricture			
Yes	1 (9)	2 (6)	0.151
No	10 (91)	28 (93)	
Dehiscence			
Yes	0	2 (6)	0.118
No	11 (100)	28 (93)	

Of the 30 patients after second stage repair, urethral fistulae formation occurred in 3 patients, stricture occurred in 2 patients requiring revision of graft, meatal stenosis occurred in 3 patients requiring dilatation and wound dehiscence due to infection occurred in 2 patients. Overall success rate was 66% which is slightly higher than one

stage repairs (63%). No donor site abnormalities were observed in our study. Analysis and comparison of the outcome and complications in one stage and two stage repairs was statistically insignificant as summarized in Table 3.

Correlating the success rates with meatal position showed a slightly higher success rates in proximal penile hypospadias (65%). Correlating the meatal position with the complications was statistically insignificant (Table 4).

Table 4: Analysis and comparison of the outcome and complications in relation to meatal position.

Variable	Midshaft and distal, N (%)	Proximal penile, N	p value
Outcome			
Success	7 (63)	19 (65)	0.147
Failure	4 (36)	10 (34)	
Meatel stenosis			
Yes	1 (9)	3 (10)	0.131
No	10 (90)	26 (89)	
Fistulae formation			
Yes	2 (18)	3 (10)	0.155
No	9 (81)	26 (89)	
Restricture			
Yes	1 (9)	2 (6.8)	0.140
No	10 (90)	27 (93)	
Dehisence			
Yes	0	2 (6.8)	0.118
No	11 (100)	27 (93)	

Table 5: Analysis and comparison of the outcome and complications in relation to number of procedures prior to BMG urethroplasty.

Variable	<2 procedures	>2 procedures	p value
Outcome			
Success	18 (78)	9 (50)	0.25
Failure	5 (21.7)	9 (50)	
Fistulae			
formation			
Yes	2 (8.6)	3 (17)	0.070
No	21 (91)	15 (83)	
Meatal stenosis			
Yes	1 (4)	3 (17)	0.084
No	22 (96)	15 (83)	
Restricture			
Yes	1 (4)	2 (11)	0.062
No	22 (96)	16 (89)	
Dehiscence			
Yes	1 (4)	1 (5.5)	0.042
No	22 (96)	17 (94)	

Analysis of outcome in relation to the number of surgical procedures prior to BMG urethroplasty showed higher success rates (78%) in patients with less than 2 surgical procedures prior to BMG urethroplasty. When comparing the complications with the surgical procedures prior to BMG urethroplasty, the results showed statistical significance with occurrence of wound dehiscence in patients with less than 2 surgical procedures prior to BMG urethroplasty. Other complications were statistically insignificant in correlation to the number of surgical procedures prior to BMG urethroplasty as summarized in Table 5.

DISCUSSION

Reconstruction of the urethra in congenital or acquired urethral defects presents a formidable challenge. After repeated previous surgery there is lacking of penile and preputial skin and the blood supply of these skin flaps is disturbed so that free graft material is preferred. Diverse opinions have been expressed on the quality and type of ideal substitution material. Various split and full-thickness skin grafts from the scrotum, penis and extragenital sites (ureter, saphenous vein, appendix, colonic mucosa, medial upper arm, neck, lateral chest and abdomen), bladder mucosa and oral mucosa have been used.^{3,4} Buccal mucosa has a thick non-keratinized epithelial layer and a wellvascularized and thin lamina propria, favoring early inosculation. It is easily accessible, non-hair bearing, extremely elastic with high resistance to infection and trauma and high regenerative power. Buccal mucosa takes shorter time to harvest and the chances of failure and recurrence are reduced in patients with balanitis xerotica obliterans as buccal mucosa is not involved in this process.⁵ As early as 1941, Humby combined oral mucosa with a full-thickness graft for penoscrotal fistula closure and observed success with buccal mucosa.⁶ Duchett et al presented use of oral mucosa in large heterogeneous patient sample (N=18) where after 27 months 17% of the patients required corrective surgery.⁷ Dessanti et al also reported their findings from 12 cases after the first long follow-up results.⁸ The indications for the use of buccal mucosal free grafts for substitution urethroplasty are varied like in: Anterior urethral strictures, proximal hypospadias especially in circumcised patients, crippled hypospadias, DSD, Y-duplication of urethra, and epispadias when the penile skin is insufficient. This was shown in 1992 in the study by Burger and associates by using BMG in failed hypospadias repair, severe stricture after hypospadias repair, short urethra and epispadias in 1 patient. 1,3,9 The results were urethral fistulae and meatal stenosis in 3 patients each. El-Kasaby and associates also reported their experience with buccal mucosa patch graft in the management of 20 patients with anterior urethral strictures with only 2 requiring revision for recurrent stricture. 10 We have used BMG in 25 cases of hypospadias, 4 cases of uretheral stricture, 6 cases of 46 XY disorders of sexual disorders, 4 cases of Y-duplication of urethra, 2 cases of redo-epispadiase repair.

In our study, 1-stage repair was elected for 11 patients, out of which 3 patients underwent BMG tube uretheroplasty, 4 underwent ventral onlay placement of BMG, 4 underwent ventral inlay placement of BMG. Out of which 2 patients had developed urethral fistula, 1 had urethral stricture and 1 had meatal stenosis and the outcome was not affected by the surgical technique. Similarly Barbagli et al recently compared the results of buccal mucosal urethroplasty in 50 patients with bulbar urethral strictures. BMG were placed on the ventral, dorsal and lateral bulbar urethral surface in 17, 27 and 6 cases, respectively. They reported that the placement of BMG into the ventral, dorsal or lateral surface of the bulbar urethra showed the same success rates (83 to 85%) and the outcome was not affected by the surgical technique. Moreover, stricture recurrence was uniformly distributed in all patients.¹¹

In our study, 30 patients underwent 2 stage repairs. We made 2 modifications. In the first stage glans wings were raised with preserving of the urethral plate at the proposed neomeatus. This led to slit like appearance of neomeatus and reduced the occurrence of meatal stenosis in our study. Steroid and testosterone gel was applied postoperatively. This led to improvement in graft uptake and there was only three cases of stricture postoperatively in our study. Ransley and Manzoni, in 1999 used preoperative testosterone treatment in BMG in 100 cases and recommended the use of preoperative testosterone treatment in difficult cases to improve the vascularity of the penile skin and thereby enhance the chances of success with a free graft technique. 12 On review of literature we could not find reported cases of use of local application of testosterone after the first stage of BMG urethroplasty. Complete graft take after first stage was achieved in 96% which is better when compared to the study by Haxherixha et al who reported 89% complete take. 13 Snodgrass and Elmore reported a success rate of 88% rate of complete graft take in secondary repairs. 14 Obaidullah and colleagues reported only 3% partial graft loss. 15 The reason for this difference may be the higher number of primary hypospadias cases in the later study. Fistula formation is the commonest complication especially in re-operative cases, it is higher as scarred and ischaemic fibrotic tissues have poor healing. In the original study of Bracka in 1995, fistulae were reported to be 10.5% among the re-operative cases.16 Obaidullah and Aslam applied the two-staged Bracka technique on 1206 patients (189 re-operative cases) and reported an overall fistula rate of 5.9%. 15 In our study urethral fistulae rate was 10% in two stage repairs.

Donor site complications like intraoperative hemorrhage, postoperative infection, pain, swelling, damage to the parotid duct, limitation of mouth opening eversion of the vermillion, loss of sensation or altered sensation in the cheek or lower lip and scar contracture have been mentioned in the literature. The Kamp et al and Jang et al compared donor site morbidities in both the lips and inner cheek and found significantly greater long-term complications after harvesting graft from the lower lip. 18,19 Also Greenwell et al concluded that the donor site can be

left unsutured to lessen pain.²⁰ No donor site abnormalities were observed and lip donor sites were left open in our study.

Our study reports our experience of one stage and two stage techniques in BMG uretheroplasty in children in varied cases. Two staged reconstruction is preferred when the urethral plate is unsalvageable. We performed one stage BMG reconstruction in 11 patients with a success rate of 63% and two stage reconstructions in 30 patients with a success rate of 66%. Analysis and comparison of the outcome in relation to the type of repair, meatal position and number of surgical procedures prior to BMG urethroplasty was statistically insignificant.

CONCLUSION

Buccal mucosa displays many inherent characteristics of an ideal graft substitute for urethroplasty. Two stage reconstruction has a slightly higher success rate than one stage reconstruction but the choice of the technique must be based on stricture characteristics and on surgeon's preference.

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