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Original Research Article

Single stage repair with buccal mucosal graft in hypospadias cripple patients

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ABSTRACT

Background: Hypospadias surgery has developed into a well-defined art and science. Surgeons dealing with this anomaly should have detailed understanding of various basic surgical principles and experience with delicate, precise optically assisted techniques and maintain a clinical workload that is sufficient to obtain consistently good results. Objective of this study was to evaluate the outcome of using buccal mucosal graft for single stage repair in adult hypospadias cripples.
Methods: All patients with prior failed hypospadias repair after multiple surgeries presenting in MLN Medical College, Allahabad were included in study. Dorsal onlay buccal mucosa graft with parallel turn over flap from penile skin for ventral surface method was used.
Results: Total 20 patients were included in study. Among these 12 patients presented with complete dehiscence of repair, 5 patients presented with multiple urethrocutaneous fistulas, 3 patients presented with extensive stricture formation. Same surgeon performed procedure of dorsal onlay buccal mucosa graft urethroplasty to reduce the bias. Success rate of dorsal onlay substitution free buccal mucosal graft urethroplasty in single stage is affected by scarring of surrounding penile skin. Most common complication related to donor site are transient pain and swelling and to graft site are stricture and fistulas.
Conclusions: Single stage repair with buccal mucosal graft in hypospadias cripple patients is an excellent free graft urethroplasty in cases with prior failed primary repair of hypospadias, in whom the preputial skin was lacking or insufficient and a longer urethral tube needed to be constructed.

Keywords: Dorsal onlay buccal mucosa graft urethroplasty, Hypospadias cripples, Scarring

INTRODUCTION

Hypospadias is a birth defect of male urethra that involves an abnormally placed urinary meatus. Instead of opening at the tip of glans penis, a hypospadic urethra opens anywhere along a line (the urethral groove) running from the tip along the underside (ventral aspect) of the shaft to the junction of the penis and scrotum/perineum.[1] Hypospadias is diagnosed by physical examination, first suspected by ventrally deficient prepuce and confirmed by the proximal meatus. Other abnormal ventral findings potentially include downward glans tilt, deviation of the median raphe, scrotal encroachment onto the penile shaft, midline scrotal cleft and penoscrotal hypospadias.

Anatomic classification of hypospadias recognizes the level of the meatus without taking into account curvature.[2] A more recent classification was described. This classification indicates the site of urethral meatus (before and after chordee correction), the prepuce (incomplete or complete), the glans (cleft, incomplete

cleft or flat), the width of urethral plate, the degree of penile rotation if present and the presence of scrotal transposition.

- First degree hypospadias: urethra opens on the underside of glans penis (70% cases).
- Second degree hypospadias: urethra opens on the shaft
- Third degree hypospadias: Urethra opens on the perineum.

Timing of surgery

From 18 months to approximately 3 years of age has been described as a difficult period for hospitalization, leading to a recommendation that repair be postponed to age greater than 3 years (Manzoni et al).[3]

Weber et al, compared health-related quality of life assessment after hypospadias surgery done at less than 18 months of age versus more than 18 months of age and found no differences related to age at operation.[4]

Success of repetitive operations decreases because the penis is heavily scarred, immobile, hypovascular, or significantly shortened. Failure cause in hyposadias repair can be wound infection, urine extravasation, hematoma, ischemia, necrosis of flap, graft from errors in design, technique and postoperative care during the primary repair. However, the most common complication in hypospadias repair is the formation of urethrocutaneous fistula.

Several surgical techniques have been used to reduce the rate of this complication. Horton and Devine used the term hypospadias cripple to describe the patient who had undergone multiple, unsuccessful hypospadias repair. Multiple repair attempts results in significant resultant penile deformity. Cases presented in this study have undergone the operation of hypospadias many times before and repaired area is now fully opened. However, the ventral and dorsal regions of the tissues were not of good quality. There was dense tissue scar.[5]

If local tissue cannot be used for hypospadias repair because of extensive scar formation, breakdown of the repair or a compromised vascular supply, buccal mucosa graft can provide a reliable option. Many authors have recommended buccal mucosal graft in secondary and complex hypospadias repair. Use of vascularized local penile or preputial skin has been the mainstay of urethral reconstruction for a long time. Lacking penile and preputial skin necessitated the search for new sources for tissue transfer.

Since 1909, a large variety of free extra genital graft tissues had been described in the literature for substitution urethroplasty e.g. ureter, tunica vaginalis, full thickness extragenital skin, bladder mucosa, oral mucosa. The first report on the application of oral mucosa as a substitute for conjunctiva dated back to 1873 when Carion S used the lip mucosa to treat conjunctival defects.[6] Humby, a plastic surgeon, first proposed and reported the use of buccal mucosa in urethral surgery for hypospadias repair in 1941.[7] Current enthusiasm for the technique was promoted by Duckett, Burger and associates, Dessanti and colleagues and Elkasaby et al.[8-11] Nowadays, buccal mucosa has become the mainstay of these tissue transfer techniques.

Current indications for the use of buccal mucosal free grafts for substitution urethroplasty in:

- Proximal hypospadias especially in circumcised patients.
- Crippled hypospadias where there is no sufficient genital skin.

METHODS

This study was a prospective study. It was carried out in the PG Department of Surgery, SRN Hospital, affiliated to MLN Medical College, Allahabad, after approval from the ethical committee and obtaining written and informed consent from the patient.

Previously operated cases of hypospadias of any age group presenting with various complications were included in the study. Patients with sub mucous fibrosis of oral cavity and other oral pathology, serious life-threatening medical illness, inability to understand and give consent, local or systemic infection and who were lost during follow-up were excluded from the study.

History of previous repairs with date of operation, duration of stay in hospital and the complication that occurred after the primary repair was recorded.

Examination of the oral cavity: buccal mucosa for submucosal fibrosis, inflammation, chronic ulcer.

Teeth deposits

- Hard (calculus)
- Soft (plaque)
- Carious teeth
- Gingivitis, periodontitis

Harvesting the oral mucosa[12]

Buccal mucosa may be harvested from inner surface of cheek, upper or lower lip. Mucosa of the cheek is preferable than that of the lip because the mucosa of the cheek is thicker and more robust than the mucosa of the lip. Also, the width of the lip limits the size of the graft.

For a single strip of buccal mucosa to be used as on Onlay patch, the adult cheek provides up to 6 cm and the lip 4 cm length with 12 to 15 mm width. It is not

advisable to continue the strip of buccal mucosa through the angle of the mouth to combine both cheek and lip segments in continuity for 10 cm. In the experience of Ransley, 1999 the only buccal mucosa donor graft site complicated by a significant contracture was at the angle of the mouth.

General anesthesia via endotracheal intubation is the preferred method of airway control to facilitate access to the oral cavity. A surgical marking pen is use to outline the extent and shape of the graft away from the parotid papilla. A graft that is at least 10% longer and 10-20% wider than actually necessary is obtained to allow for shrinkage.

Hydro-dissection of the oral mucosa from the underlying soft tissues using 1% lidocaine with 1:100,000 epinephrine and the use of a no.15 blade is recommended so that the incision is limited to the full thickness of the mucosa only. Dissection of the buccinator muscle can lead to damage of the buccal neurovascular bundle. Also, branches of the facial nerve lying deep within this muscle would not be damaged by this incision.

Donor site was sutured with 3-0 absorbable suture in interrupted fashion. Harvesting the graft is slightly painful but not disabling in the post-operative period. There is no difference in long-term post-operative morbidity whether the graft site is closed or left open however, some authors recommended to leave the buccal mucosa harvest sites unsutured. Post-operative discomfort can be lessened by a diet of soft meals and by cleansing of the wound daily with povidone iodine solution.

The buccal mucosal graft is immersed in saline to which penicillin and gentamycin have been added. The graft is then thinned to remove the excess fat. Absorbable sutures were used for closure of the donor site in case of the inner cheek whereas healing by secondary intention was allowed for inner lower lip graft sites to prevent contractures.

There is more bleeding associated with the lingual graft because the tongue is more vascular than the cheek. During graft harvesting, care should be taken to the site of the opening of Wharton's duct and the underlying lingual nerve.

Useful points to be remembered while undertaking buccal mucosa graft for reoperative hypospadias

- Grafts should be oversized by 20% to account for shrinkage.
- Meticulous “defatting” is required for good take.
- Lower lip grafts being thinner are preferred for glans and meatal area.
- Grafts should be securely quilted in position both in the center and at the edges.
- Compression “tie over” dressing for 10 days is essential for good graft take. (for staged repairs)
- Monthly review is useful to assess focal scars which can be regrafted prior to tubularisation. (for staged repairs)
- Tubularisation can be done in 4-6 months. Grafts can be left open to air without undue effects. (for staged repairs)
- Cheek donor sites can be closed primarily.
- Lower lip donor sites can be left open.
- Dairy products should be avoided for 72 hours to speed up healing.

Surgery steps

Patient was laid in supine position; a stay suture was taken on glans and the penis was retracted cephalad with gentle traction. Silicone Foley’s urethral catheter was introduced through the urethral opening. The urethral plate/scarred tissue distal to urethral meatus was incised in the ventral midline until the urethral opening was reached and widened. The bleeders were cauterised.

The buccal mucosa graft was trimmed to its appropriate size, according to the length and width of the bed. The graft is thereafter sutured to the edges of the bed in continuous fashion with 5-0 Polygalactin. Two parallel incisions were made on each side of the sutured graft on shaft up to the tip of the penis, the foleys catheter was then placed over the graft and the parallel flaps are then closed over the graft with running 5-0 Polygalactin. Skin was closed with interrupted suture using Nylon 4-0/5-0.

Post OP care

- Donor site was followed up for early postoperative complication such as oral bleeding, hematoma, cheek swelling, perioral numbness.
- Urethroplasty wound was followed up for postoperative bleeding or infection.
- Urethral catheter was removed after 10-14 days postoperatively.
- All patients were followed for oral tightness, persistent oral numbness, and urethral extravasation.
- After removal of catheter patients were followed up for stream, residual chordee and cosmetic appearance as told by the patient.
- Long term follow up for any stricture formation and need for dilatation

Reference standards

We accepted the following reference standard.

For a positive test we assessed successful outcomes of use of buccal mucosa graft in secondary hypospadias repair (cases after previous unsuccessful hypospadias repairs)

For a negative test we assessed failure attempts of secondary hypospadias repair with buccal mucosa graft and their cause and the ways these could be avoided.

An arbitary period of 6 months was kept for the follow up of patients and were analysed for confirmation of a successful repair.

RESULTS

A total of 20 patients with previous failed hypospadias repair were included in the study. Position of urethral meatus was the most important factor in determining length of buccal mucosa graft and outcome of Buccal Mucosa Urethroplasty. Most patients presented with urethral meatus in midpenile region. Some of the patients had more than one fistula with stricturous passage. In such cases the proximal opening from which the patient was passing maximum urine was considered as the meatus.

Table 1: Baseline preoperative characteristics.

Characteristics	No. of patients	Percentage
Age Group (in years)		
8-11	4	20
11-14	12	60
>14	4	20
Position of urethral meatus		
Midpenile	12	60
Proximal penile	6	30
Penoscrotal	2	10
Type of penile tissue observed		
Soft and supple but deficient soft tissue	5	25
Scarred soft tissue	15	75
Number of previous surgeries done		
<3	8	40
3 and >3	12	60
Complication after previous failed surgery		
Complete dehiscence of repair	12	60
Multiple urethrocutaneous fistulas	5	25
Extensive stricture formation	3	15

Type of penile tissue observed predicted the successful outcome of graft uptake. Most patients in our study had undergone various procedures and had extensively scarred soft tissue and these patients were the one who developed complications like fistula, strictures. Those with soft and supple tissue had better graft uptake and healing. Number of previous surgeries correlated with the amount of soft tissue left for further reconstruction and the degree of scarring. Most of the patients who presented to us with previous failed hypospadias surgery had the complication of complete dehiscence of the repair with or without residual chordee.

Table 2: Intraoperative characteristics.

Characteristics	No. of patients	Percentage
Length of buccal mucosal graft		
3-4 cm	6	30
5-6 cm	12	60
>7 cm	02	10

Maximum length of buccal mucosa graft used for urethroplasty was 8 cm which was used for patients with penoscrotal hypospadias, lesser lengths needed for midpenile hypospadias.

In study of 20 patients 06 had complications following Buccal Mucosa graft urethroplasty. Of these, 1 patient had wound dehiscence and had to undergo redo procedure after 4 months. Of 4 fistula patients 2 had spontaneous healing and 2 had to undergo redo procedure. One patient with stricture was successfully managed by regular urethral dilatation.

Table 3: Outcome variables.

Characteristics	No. of patients	Percentage
Complications following Buccal mucosa graft urethroplasty		
Fistula	4	66
Stricture	1	17
Wound dehiscence	1	17
Patients questionnaire		
Do you splay when you pass urine?		
No splaying	15	75
Occasional splaying	4	20
About half the times	1	05
Usually	0	0
Always	0	0
If you splay, how bad is the splaying?		
Mild	4	75
Moderate	1	25
Severe	0	0
Are you pleased with the cosmetic appearance?		
Very pleased	12	60
Pleased	6	30
Unhappy	2	10
Very unhappy	0	0

Donor site morbidity

In all the patients who underwent Buccal mucosa graft urethroplasty the graft was taken from the lower lip. All donor sites were primarily closed with interrupted absorbable suture.

Swelling, bleeding and pain were the commonly encountered complications of donor site. Pain was

adequately managed by analgesics and ice fomentation. Swelling usually was transient and on an average it usually subsided by the 5th postop day. No infection or wound dehiscence was noticed after primary closure of the donor site and the donor site healed completely by the 8th postop day.

Table 4: Donor site morbidity.

Post op day	Complication noticed
Day 1	Pain, bleeding, swelling
Day 2	Pain, swelling
Day 3	Pain, swelling
Day 4	swelling
Day 5-8	Swelling subsided

Duration of hospital stay

Mean duration of hospital stay of patients was 14 days. Patient were on I/V antibiotics for 5 days and thereafter substituted by oral antibiotics.

Post op dressing was changed after 5 days and looked for any hematoma, graft necrosis, infection or wound dehiscence, and thereafter dressing was changed after every alternate day. The catheter was removed after mean duration of 12 days and stream of urine was looked for. The patient was called for follow up after every 2 weeks for evaluation of the stream and to look for delayed complications.

DISCUSSION

Secondary repair for hypospadias cripples is a difficult task as anatomical, functional and aesthetic aspects have to be taken into account while performing urethroplasty. Penile shape, appropriate meatal position and normal functional capacity have to be achieved, normal urinary outflow rates, stream and erection are to be considered. Aesthetic outcomes are to be kept in mind and the donor site impairment has to be avoided.

Goal of secondary repair of hypospadias cripples are creation of straight penis, meatus placed vertically on glans penis and to provide a functional urethra devoid of fistula, stricture or diverticula. Surgeon has to balance this procedure to achieve best cosmetic appearance with reliable function.

In order to achieve it one must restore urethral continuity upto tip of glans, straight penis without torsion. Though there are many procedures for secondary repair of hypospadias including full thickness skin grafts, bladder mucosa glue etc., the best suited option for a particular patient is critical for normal penile anatomy and function.

In the present study same surgeon performed procedure of dorsal onlay buccal mucosa graft urethroplasty to reduce the bias. 5 patients presented with multiple urethrocutaneous fistula and repair with buccal mucosa graft in these patients was successful in 4 of these patients and only 1 patient had to undergo redo procedure. Similar results were obtained by Hosseini J et al in which fistula repair using buccal mucosa graft patch has been done in 14 patients with urethrocutaneous fistula developing after hypospadias reconstruction.[13] 7 fistulas were in mid-shaft, 4 were in penoscrotal region and 3 were in coronal region. Repair was successful in 11 out of 14 patients (78.6%). In the remaining children the diameter of the fistula was smaller than that before the operation, offering a good opportunity for subsequent closure.

Beneficial characteristics of buccal mucosal graft

- Easily accessible, non-hair bearing and the supply source is constant and adequate.
- The intraoral donor site guarantees an excellent cosmetic result.
- The graft is extremely elastic and shows only a slight tendency to contraction.
- The graft has been noted to retain the elasticity of the virgin tissue.
- The meatal problems of excoriation, encrustation and protuberance that encountered with bladder mucosa, have not been a problem with buccal mucosa.
- High resistance to infection and trauma and high regenerative power.
- Buccal mucosa is characterized by thick epithelial layer and thin lamina propria that make the graft mechanically stiff and easily handled and aid in rapid vascularization of the graft.

Multiple procedures were described for correction of hypospadias with evolving techniques either single staged or two staged, but the outcomes varied on the basis of the surgical technique, the procedure used, patient selection, preoperative parameters and most importantly the position of the meatus.

Many complications occurred after primary repair of hypospadias and led to various studies for the management of hypospadias cripples.

Dessanti and associates reported only 1 case of urethral fistula out of 8 hypospadias crippled patients after 6-18 months of follow up. Another 2 patients required urethral dilatation during initial 4 weeks because of mild anastomosis stenosis. No meatal problems such as stenosis or granulomatous reaction was observed.[14]

Burger and associates in the same year used buccal mucosa in 6 patients; failed hypospadias repair, severe strictures after hypospadias repair, short urethra and epispadias in 1 patient. The results were 3 urethral fistula and 1 meatal stenosis in 3 patients. No urethral stricture or diverticulum was noted, and the final outcome was good functionally and cosmetically in all patients.

American Urological Association in 2006 evaluated records of 137 patients who were referred after multiple unsuccessful hypospadias repairs. Results showed urethral substitution using skin grafts and bladder mucosa a high complication rate of 32% and 37% respectively, whereas use of buccal mucosa resulted in a 15% complication rate. Hence favored the use of buccal mucosa graft for hypospadias cripples.[15]

Buccal mucosa graft urethroplasty can be done by using various techniques:

- Ventral onlay urethroplasty
- Dorsal onlay urethroplasty
- Bracka's 2 stage urethroplasty

Department of Pediatric Urology, Manchester, England retrospectively reviewed patients who underwent oral mucosal grafts for hypospadias between 1994 and 2002. 37 patients underwent oral mucosa graft consisting of an Onlay graft (30), urethral tube (5) or 2-stage Bracka's type procedure, of the patient 26 had undergone at least 1 previous operation for hypospadias. They conclude oral mucosal grafts has significant associated early complication rate of fistula and stricture, with 27% of cases requiring further surgery.[16] However majority of buccal mucosa graft have stable urethral outcomes at puberty.

Ransley and Manzoni studied 100 cases of secondary hypospadias repair with buccal mucosa graft. They reported a secondary operation rate of approximately 20% which are mostly due to minor fistulae or the need for meatal revision.[17]

In 2002 a study was concluded and reported the use of three different types of grafts (skin, bladder, buccal) in urethral reconstruction. Results showed that buccal mucosa graft had the highest success rate. Buccal mucosa graft was used in 25 patients; 10 hypospadias and 15 patients with urethral strictures.

As regard hypospadias 40% were primary and 60% were redo cases. Buccal mucosa graft was applied as a patch graft in all patients. Length of graft ranged from 2.5-6 cm. Success rate was (70%) where 3 patients showed 4 complications in the form of 2 fistulae, 1meatal stenosis and 1breakdown. Stricture was bulbar in all patients. Fistula was minute and closed spontaneously. Final success rate was 80%.

Bracka A evaluated their experience with buccal mucosa in urethral reconstruction for complex hypospadias repair in total 62 children operated since 1990 with mean follow up of 23 months.[18] The graft was then sutured in an onlay fashion to the urethral plate. Overall complication rate in this rather complex series of patients including 26 hypospadias cripples was 13%, complication involved 4 fistulas, 3 cases of graft necrosis and only 1 meatal stenosis during the follow up period. These favorable results prompted them to use buccal mucosa onlays as their current method of choice for urethral reconstruction with avoidance of tubularised repairs.

Fistula repair using buccal mucosal graft patch has been done in 14 patients with urethrocutaneous fistula developing after hypospadias reconstruction by Hosseini J et al in 2009.[19] 7 fistulas were in mid-shaft, 4 were in penoscrotal region and 3 were in coronal region. Repair was successful in 11 out of 14 patients (78.6%). In the remaining children the diameter of the fistula was smaller than that before the operation, offering a good opportunity for subsequent closure.

Comparative study between tubularized incised plate (TIP) and single stage Buccal mucosa graft urethroplasty done by Yasin M et al in patients with recurrent large urethrocutaneous fistula after hypospadias repair.[20] 59 patients with previous 1-5 failed hypospadias repair were enrolled in this study age ranging from 38 months to 29 years. Group 1 included 37 patients in which TIP procedure was done. Group 2 included 22 patients in which single stage dorsal inlay buccal mucosa graft with TIP technique. Success rate was 86.5% in group 1 with 3 fistulas and 2 urethral strictures and 90.9% in group 2 with only 1 fistula and 1 stricture. Result showed single stage dorsal inlay BMG approach combines excellent cosmetic and functional results of snodgrass technique with BMG. Given its simplicity, variability and low complication rate, it is a valuable option for complex hypospadias reoperatives. We in this study performed Dorsal onlay urethroplasty and had satisfactory results and stable neourethra and was a easier technique to perform and minimal risk of graft necrosis.

Advantages of dorsal onlay technique of urethroplasty

- Simple and quick to perform
- Does not increases risk of punch formation and post micturition dribble
- Does not increases risk of fistula or patch necrosis.
- Reduces chance of graft shrinkage and sacculation.
- Decreased risk of spongiosal bleeding
- Versatile procedure which may be combined with a pedicled flap or an augmented roof anastomotic repair
- Attractive in reoperative cases in which ventral onlay had been previously used
- Does not require extensive training in reconstruction procedures using tissue transfers.

CONCLUSION

Single stage repair with buccal mucosa graft is an excellent free graft urethroplasty in cases with prior failed primary repair of hypospadias, in whom the preputial skin was lacking or insufficient and a longer urethral tube needed to be constructed. Success rate of dorsal onlay substitution free buccal mucosal graft was affected by length of urethral tube to be reconstructed. Most common

complication related to donor site were transient pain and swelling and to graft site were stricture and fistulas which can be successfully managed by gradual urethral dilatations.

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