Outcome Analysis of Redo Urethroplasty for Large and Multiple Urethrocutaneous Fistula.


1Asst. Professor, Dept of Pediatric Surgery, Rajshahi Medical College, Bangladesh.
2Professor, Dept of Biochemistry & Molecular Biology, Rajshahi University, Bangladesh
3Associate Professor, Dept of Pediatric Surgery, Rajshahi Medical College, Bangladesh.
4Professor, Department of Paediatric Surgery, Dhaka Shishu (children) Hospital, Bangladesh.
5Consultant(Jr). Dept of Gynaecology, Upazilla Health Complex, Premtoli, Godagari, Rajshahi, Bangladesh.
6Associate Professor, Dept of Pediatric Surgery, S Salimullah Medical College, Dhaka, Bangladesh

Received: March 2018
Accepted: March 2018

Copyright: © the author(s), publisher. Annals of International Medical and Dental Research (AIMDR) is an Official Publication of “Society for Health Care & Research Development”. It is an open-access article distributed under the terms of the Creative Commons Attribution Non-Commercial License, which permits unrestricted non-commercial use, distribution, and reproduction in any medium, provided the original work is properly cited.

ABSTRACT

Background: Urethrocutaneous fistula is the most common complication of hypospadias surgery. Due to the poor surrounding tissue quality, surgical treatment of these patients represents a complex problem. Objectives: Present study aimed to determine a better procedure of salvage urethroplasty for failed hypospadias, caused by persistent large (>4mm) or multiple -small (<4mm) fistulae, by a randomized comparison. Methods: This interventional study was performed in a single institute over a period of five years. Comparisons were made among three procedures of salvage urethroplasty using substitution of dorsal skin flap, Flip flap, or buccal mucosal graft in a controlled situation. Outcomes were assessed by means of objective scoring system. Results: Refistula rate, devascularization of flap and grafts and wound dehiscence rate were significantly less in Buccal mucosal graft than flip flap and dorsal transposition flap. This led to a higher success rate and better patient compliance in buccal mucosal graft. Though mean duration of hospital stay was significantly longer in buccal mucosal graft compared to other methods, it did not negatively affect patient satisfaction due to overall higher success rate. The objective scoring evaluation revealed that score gain of BM group was significantly higher than that of dorsal transposition flap and flip flap. Conclusion: Staged redo urethroplasty for large or multiple-small fistulae using substitution of buccal mucosal graft revealed as an better option for urethral reconstruction than dorsal transposition flap and flip flap procedures (group A> group B > group C).

Keywords: Buccal mucosal graft, Dorsal skin flap, Failed hypospadias, Flip-flap, Hypospadias, Urethrocutaneous fistula.

INTRODUCTION

The modern era of hypospadias surgery has seen major technical advances. This repair not only involves urethroplasty, but also has its goal in achieving good cosmetic and functional results. However, description of over 250 procedures for hypospadias repair suggests lack of uniformity of results and a complication rate that even in this era is quite high. There still is room and therefore need for further improvement in this field.[1,2] The complexity of these cases was highlighted by the largest report to date on failed hypospadias by Barbagli et al.[3] In that retrospective descriptive analysis, 1,176 patients with failed hypospadias, a median of five additional procedures (ranging from 2–23) were required to achieve a satisfactory final result.[3] Urethrocutaneous fistula after hypospadias surgery is the most common complication and remains a frustrating problem that precludes the goal of hypospadias surgery leading to failure of primary surgery. The reported incidence of urethrocutaneous fistula ranged, from 0 to 30%, varying with the severity of hypospadias, surgical technique, and experience of the operating surgeon.[4] The problem is exacerbated because urethrocutaneous fistulae not only occur but also recur, sometimes requiring many procedures in the same patient.[5] Often this difficult population of patients has been left with deformities significantly worse than the primary congenital anomaly.[6,7]

Reoperative urethroplasty after failed hypospadias surgery is usually done with principles used for primary repairs. Numerous surgical techniques have been suggested to repair the complications after failed hypospadias repair, including simple closure,
one-stage procedures (flaps-flaps, onlay flaps, tubularized preputial flaps or tubularized incised plate urethroplasties, bladder mucosa graft) or multistage procedures with penile skin or buccal mucosa. But renewed controversy exists over the best means of reconstructing the penile urethra in this difficult population of patients. The majority of publications in hypospadias literature present single-center and single-surgeon retrospective case series. High-quality randomized trials in pediatric urology are extremely challenging and therefore rarely performed. In addition, there is no generally accepted system for assessing the surgical results. This lack of an impartial method of documenting the results of hypospadias surgery has made the comparative evaluation of operative procedures inaccurate and subjective. In the present study outcome of urethrocutaneous fistula repair in failed hypospadias using buccal mucosal graft and local skin flaps were evaluated to find out a better surgical option. The outcomes were analyzed by objective scoring system and post redo surgery complication rates. Thereby a valid and balanced evaluation was made among traditional and innovative surgical procedures for the salvage repair of hypospadias.

MATERIALS AND METHODS

It was an interventional study, carried out in the Department of Paediatric Surgery, Rajshahi Medical College Hospital, Rajshahi, Bangladesh, from July 2011 to June 2016, for a period of five years. Paediatric patients with previous failed hypospadias repair presented with a persistent fistula and supple dorsal skin were included in the study. A minimum of 1 year was allowed to elapse following the last failed repair. Present study was aimed to determine a better procedure of salvage urethroplasty for failed hypospadias, caused by persistent large (>4mm) or multiple -small (<4mm) fistulae, by a randomized comparison.

Patients with single pinhole or small (< 4mm) fistula and Multiple -large (>4mm) fistula; Patients with urethral diverticulum and urethral stricture; Patients with severe other surgical, medical problems; Fistula over coronal, penoscrotal, scrotal or perineal location; Patient with glans deformity and total disruption; Patients with persistent severe chordee were excluded. Simple Random sampling technique was followed to select groups for each sample by means of lottery. BM GRAFT (Buccal Mucosal Graft) was grouped as group A, DORSAL FLAP (Dorsal transposition flap) was grouped as group B, FLIP FLAP (Distally based flip-flap) was grouped as group C. Each patient in every group received a same standard schedule of management regarding preoperative assessment, Control of infection and analgesia and assessment of outcome.

Addressing ethical issues: Keeping compliance with Helsinki Declaration for Medical Research, informed consent was taken from the legal guardians of all patients under study after explanation of purpose of the study. The study design was approved by Institutional Review Committee. Preoperative evaluation: A detailed history was taken regarding position of the meatus, curvature of the penis (chordee), stream pattern, history of urinary infection, other associated anomalies, type of previous urethroplasty with number of operations. Local area was examined for site, size, number and position of fistulae, condition of local tissue. The presence of distal stenosis or obstruction was excluded by routine calibration of the urethra distal to the fistula site.

Operative techniques: Salvage urethroplasty was done in three layers under general anesthesia. Different Graft or Flaps were used in only first layer for reconstruction. Second layer was made by Tunica vaginalis wrap and third layer was made by skin in all cases.

Buccal mucosal graft: Donor sites for buccal mucosal grafts included inner lower lip and inner cheek. The harvest site was marked carefully to avoid Stensen’s duct. Haemostasis was ensured and donor site was kept open for healing by secondary intention. After the harvested mucosa was acquired, it was cleared from excess fat to have the mucosa and submucosa.

Figure 1: preparation of harvested mucosa

Figure 2: Graft implanted
Care was taken to ensure removal of scar tissue and strictured segments of the urethra to allow for a vascularized graft bed. The graft was fixed to the corporal bodies in first stage. The second stage urethroplasty was performed three months later.

Dorsal onlay skin flap: Redundancy of the dorsal penile shaft skin was ensured. A glans traction suture was placed and a circumcising incision line was marked.

Dorsal skin flap approximately 1 cm wide and longer than the length of the urethral defect was isolated and preserving vascular mesentery the flap was transposed to the ventrum of the penis at the site of the urethral defect and was tailored to correspond to urethral defect as an onlay flap and was sutured into place.

Distally based flip flap: Initially turnover flaps were marked around the fistula so as to form the lining of the urethral first layer of closure. The flap was then dissected with underlying dartos fascia. Extreme care was taken to avoid damage of the vascular pedicle of the flap as dissection approach the subcutaneous tissues adjacent to the fistula. The flap was then "flipped" distally and sutured to the edges of the urethral plate using a double layer 6/0 polygalactin running suture on each side.

Protective intermediate layer: In this study tunica vaginalis wrap from the testicular coverings was used in all three varieties of urethroplasty as a protective intermediate layer.

Postoperative care: All urethroplasties were stented for 2 weeks. The patients were kept on antibiotics for the duration of the indwelling catheter. A compressive occlusive dressing was used. The dressing was removed on day 5 to 7. Meatal dilatation was used after catheter removal for up to 6 months to avoid stenosis; patient was instructed for visits once weekly in the first month, then once monthly in the following 6 months, then every 3 months for a year.

Data analysis: Collected data was arranged in systemic manner, presented in various tables and figures and statistical analysis was performed using SPSS software (version 16). The significance of differences and comparisons among the mean values were determined by Duncan’s multiple range tests (DMRT) formulation at 5% level, P value was reached by Chi Square test, ANOVA test and t test.

**RESULTS**

189 patients were included in present study as per inclusion and exclusion criteria and randomized in the three groups under study. Age ranged from 2 to 13 years. Out of total 189 patients BM graft consisted 61 patients, Dorsal flap comprised 69 patients and Flip flap consisted 59 patients.

Preoperative position of urethrocuteaneous fistula

Different types of urethrocuteaneous fistula included in present study were Sub coronal, Distal penile, mid
penile and Proximal penile. Other varieties of urethrocutaneous fistula were excluded from the study.

Table 1: Distribution of the studied cases according to preoperative position of urethrocutaneous fistula.

<table>
<thead>
<tr>
<th>Types of fistula</th>
<th>Methods</th>
<th>Mean ± SE</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>BM graft</td>
<td>Dorsal flap</td>
</tr>
<tr>
<td>Sub Coronal</td>
<td>14</td>
<td>16</td>
</tr>
<tr>
<td>Distal penile</td>
<td>19</td>
<td>18</td>
</tr>
<tr>
<td>Mid penile</td>
<td>15</td>
<td>20</td>
</tr>
<tr>
<td>Proximal penile</td>
<td>13</td>
<td>15</td>
</tr>
</tbody>
</table>

Analysis of variances (ANOVA) shows that significance test for the effect of preoperative position of fistulae on different methods under study at 0.01 level was non-significant. In other words, difference of the distribution of values among the three procedures under study did not significantly affect the outcome.

Duration of hospital stay (days)
The duration of hospital stays in days were calculated from the day of operation to the day of discharge from the hospital. In case of BM graft the calculated duration of hospital stays was the summation of days in two stages. The differences of mean hospital stays in days were significantly longer in BM graft (Stage-1 and 2) compared to Dorsal flap and Flip flap methods at P<0.05, according to Duncan’s multiple range tests.

Table 2: Duration of hospital stay of the patients

<table>
<thead>
<tr>
<th>Name of operation</th>
<th>BM graft (Stage 1 and 2)</th>
<th>Dorsal flap</th>
<th>Flip flap</th>
</tr>
</thead>
<tbody>
<tr>
<td>Sub Coronal</td>
<td>14.33 ± 1.15a</td>
<td>8.94 ± 0.78b</td>
<td>7.08 ± 0.75b</td>
</tr>
<tr>
<td>Distal penile</td>
<td>15.33 ± 1.20a</td>
<td>9.36 ± 1.22b</td>
<td>8.72 ± 0.91b</td>
</tr>
<tr>
<td>Mid penile</td>
<td>17.67 ± 2.07a</td>
<td>11.33 ± 1.45ab</td>
<td>7.97 ± 1.32b</td>
</tr>
<tr>
<td>Proximal penile</td>
<td>18.00 ± 2.64a</td>
<td>10.61 ± 1.86b</td>
<td>9.82 ± 1.45b</td>
</tr>
</tbody>
</table>

Values (Mean ± SE) followed by different letters (a, b, c) in a row are significantly different at P<0.05 according to Duncan’s multiple range test.

Table 3: Post operative complications for Urethral diverticulum and Anastomotic stricture

<table>
<thead>
<tr>
<th>Site</th>
<th>Methods</th>
<th>P value</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>BM graft</td>
<td>Dorsal flap</td>
</tr>
<tr>
<td>Urethral diverticulum</td>
<td>3.28</td>
<td>5.80</td>
</tr>
<tr>
<td>Anastomotic stricture</td>
<td>8.19</td>
<td>11.59</td>
</tr>
<tr>
<td>Meatal stenosis</td>
<td>4.91</td>
<td>7.24</td>
</tr>
</tbody>
</table>

NS = Not Significant. P value reached from Chi Square test.

A small number of patients developed a urethral stricture or diverticulum at the site of the anastomosis as shown in table-3. The differences of values of the post operative complications were insignificant among the three groups under study. Comparison of outcome by flap/ graft devascularization Devascularization of the flaps or graft is a major complication. The differences of devascularization of the flaps or graft rate were significantly less in Buccal Mucosal Graft group (group A) in comparison to dorsal flap method (group C), at p <0.05.

Table 4: Comparison of outcome by flap or graft devascularization

<table>
<thead>
<tr>
<th>Outcome measures</th>
<th>Methods</th>
<th>Flap/Graft devascularization</th>
<th>P value</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>BM graft</td>
<td>Flip flap</td>
<td></td>
</tr>
<tr>
<td></td>
<td>N (6)</td>
<td>% (6)</td>
<td>N (5)</td>
</tr>
<tr>
<td>Fistula recurrence</td>
<td>2</td>
<td>3.27 *</td>
<td>11</td>
</tr>
<tr>
<td></td>
<td>7</td>
<td>11.87</td>
<td>6</td>
</tr>
</tbody>
</table>

* Significantly different from dorsal transposition flap (group B) and distally based flip-flap (group C), p <0.05. P value reached from t test.

Assessment of outcome by number of re-fistula (urethrocutaneous fistula)
Recurrence of urethrocutaneous fistulae precludes the goal of urethroplasty. The highest re-fistula rate was reported in distally based flip-flap group (group C):32.20%, followed by Dorsal transposition flap group (group B):30.43%. On the other hand the lowest frequency was recorded in Buccal Mucosal Graft group (group A): 18.03%. The differences of re-fistula rate was significantly less in Buccal Mucosal Graft group (group A) in comparison to Distally based flip-flap group (group C) and Dorsal transposition flap group (group B), at p <0.01.

Figure 7: Outcome assessment by number of re-fistula among the different methods.

Fistula recurrence in relation to previous size and number.
This study included two major varieties of fistulae in relation preoperative to size and number. Those were; single and large (>4mm) [group 1] and multiple and small (<4mm) [group 2]. The differences of re-fistula rate was significantly less in Buccal Mucosal Graft group (group A) in
comparison to distally based flip-flap group (group C) and Dorsal transposition flap group (group B), at p < 0.05.

Table 5: Fistula recurrence in three groups; as per previous size and number.

<table>
<thead>
<tr>
<th>Methods</th>
<th>Number and Size of fistula</th>
<th>BM graft</th>
<th>Dorsal flap</th>
<th>Flip flap</th>
</tr>
</thead>
<tbody>
<tr>
<td>Multiple and small (&lt;4mm)</td>
<td>4 (14.81) * n=27</td>
<td>8 (21.05%) n=38</td>
<td>5 (19.23%) n=26</td>
<td></td>
</tr>
<tr>
<td>Single and large (&gt;4mm)</td>
<td>7 (20.59) ** n=34</td>
<td>13 (41.93%) n=31</td>
<td>14 (42.42%) n=33</td>
<td></td>
</tr>
</tbody>
</table>

*Significantly different from dorsal transposition flap (group B), p < 0.05
**Significantly different from dorsal transposition flap (group B) and distally based flip-flap (group C), p < 0.05. P value reached from Chi Square test.

Comparison of outcome by patient’s perception
Patient’s satisfaction was assessed by asking the subjects/guardians to grade their degree of agreement with predetermined statements in the questionnaire.

Table 6: Comparison of outcome by patient’s perception

<table>
<thead>
<tr>
<th>Types</th>
<th>BM graft</th>
<th>Dorsal flap</th>
<th>Flip flap</th>
</tr>
</thead>
<tbody>
<tr>
<td>N (61)</td>
<td>%</td>
<td>N (69)</td>
<td>%</td>
</tr>
<tr>
<td>Highly satisfaction</td>
<td>34</td>
<td>55.73*</td>
<td>21</td>
</tr>
<tr>
<td>Satisfaction</td>
<td>7</td>
<td>11.48*</td>
<td>6</td>
</tr>
<tr>
<td>Unsatisfaction</td>
<td>20</td>
<td>32.79**</td>
<td>42</td>
</tr>
</tbody>
</table>

* significantly different from dorsal transposition flap (group B) and distally based flip-flap (group C), p < 0.05
**Significantly different from dorsal transposition flap (group B) and distally based flip-flap (group C), p < 0.01. P value reached from Chi Square test.

Objective scoring evaluation
HOSE (Hypospadias objective scoring evaluation) system was followed in this study to allow an objective appraisal of the outcome of hypospadias repair; based on evaluating meatal location, meatal shape, urinary stream, straightness of erection, and the presence and complexity of any complicating urethral fistula. Pre operative and post operative scores and ultimate score gains were calculated and illustrated in [Figure 8].

Distributions of pre and post operative scores of the three procedures were as follows: BM graft (705) and (887); dorsal flap (790) and (902); Flip flap (682) and (771). Calculated score gain was were BM graft (182), Dorsal flap (112), Flip flap (89). Among the three techniques the highest score gain was recorded in BM graft (182) and lowest score gain was (89) for Flip flap. Duncan multiple range test (DMRT) demonstrated that the values of score gains were significantly different at p< 0.05.

DISCUSSION
This study was a randomized controlled trial, in a single institute over five years period. In the present study majority of patients had undergone a variety of hypospadias repairs in the past. Among the previous primary urethroplasty types “Tubularized Incised Plate” (TIP) was the commonest procedure. These findings were equivalent to our previous reports.[12,13] However, the number and types of previous procedures did not negatively influence the comparative analysis, as the differences in percentage distribution of the cases in three groups were insignificant. Similar observations were described by Yassin et al.[14] Mainly for psychological reasons, hypospadias repair was generally performed when patients are aged about 12 months.[15] Some evidence suggests that performing surgery at a younger age also decreases the risk of complications.[15,16] However, hypospadias repair may also fail many years after achieving successful functional and cosmetic results by primary repair and a urethral stricture may develop decades after the initial hypospadias surgery.[17,18] So there is no age limit for salvage urethroplasty. But, in the present study, age was limited up to thirteen years, as per admission criteria of paediatric units in the hospitals of Bangladesh, where paediatric units deals with patients up to thirteen years of age, at present.

Hospital stay and Operation times are indirect measures of economic burden of the treatment. In present study buccal mucosal graft urethroplasty was associated with a significantly longer operation time and hospital stay than other groups under study. This was reflection of the fact that buccal mucosal graft urethroplasty was done in two stages and the calculated time was the summation of two stages. On the other hand, longer operation times and hospital stay in two stages did not negatively affect patient’s compliance or outcome.

Recurrence of urethrocaneous fistulae precludes the goal of urethroplasty. The highest re-fistula rate was reported in distally based flip-flap group (group C); 32.20%, followed by dorsal transposition flap group (group B); 30.43%. On the other hand the lowest frequency was recorded in Buccal Mucosal Graft group (group A); 18.03%. The differences of re-fistula rates were significantly less in Buccal
Mucosal Graft group (group A) in comparison to distally based flip-flap group (group C) and dorsal transposition flap group (group B). High fistula rate has also been observed by others in re-operative hypospadias surgery. Shehata et al. had complication rate of 20.6%. Elicevik and colleagues documented that overall complication rate of 26%, in their study of 100 redo-surgery cases, with 18% fistula rate. Patel et al. used split onlay skin flap salvage with 54.5% postoperative fistulae.

The variability of the results was due to technical variations and difference in types of fistula and dissimilarity in tissue characteristics among the studies but the present study focused on specific varieties. Additionally, there was wide discrepancy of preoperative conditions among different studies. Devascularization of the flap or graft and wound dehiscence adversely affect success rate of the procedure and safety margin. However, Flap devascularization was more common in dorsal flap method (15.94%) Graft failure due to devascularization was significantly less (3.27%) with buccal mucosa. Contrary to the findings of present study Patel et al. reported salvage urethroplasty with split onlay skin flap with practically no flap devascularization or infection. The primary advantages with Buccal mucosa graft were more reliable and secure graft revascularisation and the ability to visually confirm the complete take and to exclude any graft contraction before considering second-stage tubularisation.

Major causes of obstructed voiding after hypospadias repair included meatal stenosis, urethral diverticulum and urethral stricture. In present study the incidences of these late complications were lowest in BM graft and differences among the three groups were insignificant. Similar findings were reported by most other researchers. Patient’s satisfaction was very subjective, wide ranging and mainly depended on the ability of the patient to cope with cosmetic and functional abnormalities. Despite multiple previous failed surgeries, both parents and patients who have experienced failed hypospadias repair often easily accepted the idea of a staged approach that allowed a safer and more reliable final reconstruction. Similar conclusions were reached by Barbagli et al. In fact without an objective scoring evaluation comparison of studies, systematic reviews and meta-analysis comparing different techniques was prone to bias, inaccuracy, and subjectiveness. In present study an objective analysis of repair techniques with a valid comparison was made by using HOSE system. Among the three techniques the highest score gain was recorded in BM graft (182). Duncan multiple range test (DMRT) demonstrated that the values of score gains were significantly different.

**CONCLUSION**

Objective assessment among procedures using substitution of flaps or buccal mucosal graft in a controlled situation revealed that staged repair using buccal mucosa was a better option for urethral reconstruction in large (>4mm) or multiple -small (<4mm) fistulae, than dorsal transposition flap and flip flap procedures.

**REFERENCES**


