Abstract

Background: Urethro-cutaneous fistula (UCF) after hypospadias surgery occurs regardless of the location of the meatus, procedure performed or experience of the surgeon. Hypospadias repair continues to be a singularly demanding form of surgical expression with considerable artistic latitude. The commonest complication of hypospadias surgery is fistula formation, which almost always requires repeat and similarly demanding surgery.

Aims and Objectives: The aim of our study was to compare the result between simple and layered repair of UCF developing after hypospadias surgery.

Materials and Methods: This comparative type of study was carried out in the Department of Paediatric Surgery, BSMMU, Dhaka from June 2015 to August 2016 for a period of 15 (fifteen) months. Forty (40) diagnosed cases of UCF were included in the study. All the participants were randomized into two groups named Group A & Group B. Group A was assigned to the simple repair technique & Group B was assigned to the layered repair technique.

Results: The recurrent fistula formation rate was much higher in Group A (30%) than in Group B (05%), which was statistically significant in between the two groups. The majority of the fistulae recurred within a few days of the removal of the stent (from 7th to 10th POD). 6, out of 7 recurrent UCF were observed in this period. No recurrent fistula was observed beyond 4 weeks after operation.

Conclusions: This study revealed that, the layered repair technique, with the addition of vascularized dorsal dartos fascia reduces the recurrent fistula formation rate in a significant number. Though more time consuming and technically difficult, we recommend wide application of layered repair technique in the management of post hypospadias UCF.

Key words: Hypospadias, UC fistula, Dorsal dartos flap.

Introduction:

Hypospadias, a common congenital anomaly of penis, characterized by an external urethral meatus that opens on to the ventral surface of the penis, proximal to the tip of the glans. Surgical reconstruction stands the only option to correct this congenital defect. Complications are possible after any surgical procedures and these are higher in hypospadias surgery as compared to other reconstructive operations. Among the complications, UCF is the most common, challenging & difficult to manage. To overcome these postoperative complications, more than 300 different surgical techniques have been described in the literatures but the hypospadiologists are still in search of an ideal technique. The reported incidence of recurrence of UCF from 10% to 45%. This UCF not only occurs but also recurs, even after multiple reconstructive procedures. This leads to the adverse physical, financial, and psychological impacts.
on the patient and his family and, nevertheless, loss of the trust in the healthcare system. There are many factors responsible for UCF formation and its recurrence. Some of them are well explored while some are still obscured. Some causes are related to surgical techniques like rough tissue handling, the use of poorly vascularized tissues in repair, the use of very thin or fibrotic subcutaneous tissue may lead to poor healing, invites wound infection and ultimately ends with fistula formation or wound disruption. Some factors are also related to the patient himself, specifically to the periurethral plate skin, distal to the hypospadiac meatus. In the hypospadias penis, the important local healing factors, i.e., vascular-collagenous tissues, are insufficient on the ventral skin as compared with the dorsal skin. This may be responsible for the higher incidence of complications like frustrating UCF after operation.

The occurrence of UCF precludes a goal of hypospadias surgery, produces mental burden on patient and family. Unfortunately there is no single perfect technique to repair an UCF. Factors that may affect the results of their repair may be the conditions of local tissue, duration after hypospadias repair, the number, location, and size of the fistula, patient’s age, previous surgical attempts, the type of suture material used, the skills of the operating surgeon and proper inversion of the edges and coverage by second layer, etc. Some failure rate is expected in every type of repair. By providing a water-tight covering layer, the incidence of recurrence in UCF repair can be greatly reduced, especially in large urethro-cutaneous fistulas. During the last decade, different repair methods have been performed according to the location and size of the fistula.

In 1981, Goldstein and Hensle proposed a simplified technique of UCF repair with a success rate of 100%. The use of different types of well-vascularized tissue as the second interposition layer became popularized later on as a layered or multilayer repair technique. The success rate of this technique varied in different series of studies.

**Materials and Method:**
Type of study: Prospective type of comparative study.
Place of study: The study was conducted in the department of paediatric Surgery, Bangabandhu Sheikh Mujib Medical University (BSMMU), Dhaka.
Duration of the study: 15 months (June 2015 to August 2016).

Study subjects: The study included children with midpenile and distal hypospadias.

Ethical consideration: Ethical clearance was taken from the Institutional Review Board (IRB) of Bangabandhu Sheikh Mujib Medical University, Dhaka.

Surgical procedure: After completion of surgical rituals urethral calibration was routinely done intra operatively with a feeding tube to exclude any distal stenosis. Thereafter, the presence, location, and number of the fistula were reassessed. Probing every pit in the skin with a probe was done to avoid missing a small fistula. In doubtful cases, methylene blue was injected under pressure from the terminal portion of the neourethra while a tourniquet was applied at the base of the penis to occlude the proximal urethra. A stay suture was given at the Glans penis by 4-0/5-0, round body prolene for penile traction. Then, a catheter or feeding tube of appropriate size was inserted into the urethra. The next operative steps were different in the two groups.

In Group A (Simple repair technique), dissection around the circumferential incision, which included the meticulous freeing of the urethra from the surrounding skin, was done. Then, the fistula was closed in an interrupted fashion with 6-0 vicryl in an inverted tension-free manner. Then, the overlying skin was closed intradermally and continuously with 6-0 vicryl.

Group B (Layered repair technique), between the urethra and skin, a well-vascularized dartos fascia was interposed. The dartos fascia was taken from the dorsum of the penile shaft. The pedicled dartos flap was separated from the skin using sharp and blunt dissection up to the penile root proximal to the 1st vascular arcades. After the preparation of the vascularized dorsal dartos flap from the dorsal penile skin, the blood supply of the flap and skin was assessed. The vascularity of the flap was assessed by color, and the number of longitudinal blood vessels was observed against the light. The penile skin vascularity was also assessed by color before and after the separation of the dartos flap. The flap was transferred ventrally as a rotational flap. The flap was then spread uniformly over the 1st layer (repaired urethra) and anchored laterally with the buck’s fascia, distally up to the repaired urethra, and proximally with the root of the penis.
Results:

In this study, the mean (+ SD) age of group A was 8.17±3.33, and that of group B was 7.00±3.21 years.

Majority of the patients had small-sized (<2 mm) and anterior UCF.

The urethral stent was functioning in all the patients in both follow-ups. The glans color was pink in all the patients in both groups. On Day 0, 2 patients developed a hematoma in Group A, and no patient developed a hematoma in Group B. On 1st POD, 2 patients developed a hematoma in Group A, and no patient developed a hematoma in Group B. On Day 0, the dressing was soaked in 2 patients in Group A and one patient in Group B. On 1st POD, the dressing was soaked in 3 patients in Group A and two in Group B. On day O, one patient was febrile in Group A and 3 patients in Group B; on 1st POD, one patient was febrile in Group A and one in Group B.

The chi-square test shows no significant difference (p-value non significant) in the follow-up parameter on day 0 and 1st POD between the two groups.

Table I: Number of recurrent UCF (n=40)

<table>
<thead>
<tr>
<th>Recurrent urethro-cutaneous fistulae</th>
<th>Group</th>
<th>P value</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Group A</td>
<td>Group B</td>
</tr>
<tr>
<td>Yes</td>
<td>n=20 (30.0%)</td>
<td>1 (5.0%)</td>
</tr>
<tr>
<td>No</td>
<td>14 (70.0%)</td>
<td>19 (95.0%)</td>
</tr>
</tbody>
</table>

A chi-square test was done to measure the level of significance
s= significant

Table II: Time of appearance of recurrent UCF (n=07)

<table>
<thead>
<tr>
<th>Time of occurrence</th>
<th>Group</th>
<th>P value</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Group A (n=20)</td>
<td>Group B (n=20)</td>
</tr>
<tr>
<td>During the stent in situ period</td>
<td>0 (0.0)</td>
<td>0 (0.0)</td>
</tr>
<tr>
<td>From 7th POD to 10th POD</td>
<td>5 (25.0)</td>
<td>1 (5.0)</td>
</tr>
<tr>
<td>11th POD to 2 weeks after operation</td>
<td>1 (5.0)</td>
<td>0 (0.0)</td>
</tr>
<tr>
<td>Two weeks to 4 weeks after operation</td>
<td>0 (0.0)</td>
<td>0 (0.0)</td>
</tr>
<tr>
<td>Four weeks to 8 weeks after operation</td>
<td>0 (0.0)</td>
<td>0 (0.0)</td>
</tr>
</tbody>
</table>

A chi-square test was done to measure the level of significance
ns= non significant
Table II shows the time of recurrent UCF appearance. Most of the fistulae appeared within a few days after the removal of the stent (within the 7th to 10th POD).

Discussion:
There is no perfect technique for UCF repair, like hypospadias surgery. Many variables could influence the surgical management and outcome, i.e., the time of occurrence after urethroplasty, the location (glandular, coronal, mid-shaft, etc.), and size (pinpoint, large), the number and the condition of local tissue. However, others reported no significant difference in outcome comparing some variables, e.g., the magnification, patient age and interval between surgery at the time of fistula repair, type of original hypospadias procedure, and number of previous fistula repairs. As no technique is effective, some failure rate is expected in every series.

Dennis and Walker proposed that the repair could be successful without transurethral stenting. Sahin et al. reported that the success rate of fistula repair was 93.3% and 94.1% in catheterized and non-catheterized groups, respectively, in 2003. As a result, they suggested that catheterless fistula repair with local anesthesia in the adult age group is an effective, safe, and inexpensive procedure. However, we have always used such a stent because we thought that such an indwelling catheter inserted for a few days could support the healing of the urethra and prevent recurrence.

Regarding the time of fistula closure, we prefer to close a fistula six months after the previous procedure to allow induration and scarring to subside. From this study, we also observed that the mean age of urethroplasty and UCF repair in our country is higher than standard practice.

The use of a microscope or loupe for magnification is now considered mandatory in hypospadias and other reconstructive surgeries. However, we did not use that in any of our procedures due to technical limitations.

The study showed that the preoperative Serum. Hb level, S. Albumin level, total WBC count, urinary pus cell count, urinary epithelial cell count, and urine bacterial growth have no negative influence on recurrent UCF formation.

The different parameters were assessed on Day 0 and 1st POD. Among these, the urethral stent was functioning in both day 0 and 1st POD in all the patients. Glan’s color was pink on day 0 and the 1st POD in all patients. In group A, one patient presented with local haemotoma in day 0 and one patient was presented with haemotoma on 1st POD. Both cases were managed conservatively, and the hematoma was resolved subsequently, and the patients did not develop UCF postoperatively. On day 0, the dressing was soaked in 2 patients in group A and one patient in group B, and on 1st POD, the dressing was soaked in 3 patients in group A and two patients in group B. The cause of the soaking was most probably persistent leakage of urine because the stent was patent in all the cases. One patient in group A and three patients in group B were febrile in day 0, and one patient in group A and one patient in group B was febrile in 1st POD. In searching for the cause of the fever, no significant pathology was found. It may be due to local tissue reaction. There was no significant difference in the above parameter between the two groups.

On the 4th, POD dressing was removed in all patients, and patients were followed up in various parameters. The wound was edematous in 3 patients in group A and two patients in group B. There was wound discharge in 1 patient of group A. The urethral stent was blocked in 1 patient in group A and one in group B. The blockage was relieved by simple irrigation of the stent with normal saline. One patient was febrile in group B. The fever was due to an associated respiratory tract infection, which subsided with conservation treatment. There was no significant difference in the above parameter between the two groups.

On the 7th POD, the urethral stent was removed in all the patients. Before removal, the stent was blocked in 1 patient in Group A, but after removal of the stent, he voided normally. No patient had a fever or wound disruption in the 7th POD. There was no significant difference in the above parameter between the two groups.

The overall recurrent UCF formation rate was in Group A, 6 (30%) and in Group B, 1 (05%). It was statistically significant between the two groups. The majority of recurrent UCF (6 out of 7) appeared within a few days of the removal of the stent (from 7th to 10th POD). No recurrent UCF was observed beyond four weeks after the operation.

Regarding the size of recurrent UCF, the result was consistent with one study but was inconsistent with two study. The incidence of recurrence was
consistent with several studies. Several authors have found that anterior fistulas, especially those at coronal and granular levels, were more likely to be associated with recurrence. Tension, relative ischemia, and distal obstruction are contributing factors\textsuperscript{15,16}. However, the result was inconsistent with the report of Holland et al. in 2008. Posterior fistula was more likely to recur in their study. They mentioned the complexity of the cases referred to their institution\textsuperscript{17}. Tissue ischemia, distal obstruction of the neourethra, and postoperative infection are considered the main factors in UCF development after hypospadias surgery. However, the frequency of fistula formation has decreased due to surgeon experience, improvement in operative technique, use of appropriate suture materials and instruments, and coverage of the neourethra with well-vascularized tissue. Also, the suture lines should not overlap each other, and a dartos facial flap should be used between the suture lines to improve surgical outcomes\textsuperscript{18}. Jehangir reported some important factors contributing to fistula development after hypospadias repairs among different series, including technical differences, severity of hypospadias, opposing suture lines, suture material, length of neourethra, tissue ischemia, and distal narrowing\textsuperscript{19}. Sozubir and Snodgrass used healthy tissue from different areas to cover the neourethra to prevent fistula formation. They use a dorsal dartos flap that was transposed to the ventral aspect of the penis over a buttonhole technique. Also, some reports showed that double dartos flap coverage of the neourethra is superior to single flap coverage for preventing UCF after TIPU\textsuperscript{3,20}. Hammed et al. believed that dorsal dartos flap coverage of the neourethra is the cornerstone to prevent fistula development. Additionally, it is clear that to become more effective application of surgical technique, clinical knowledge and surgical experience is important\textsuperscript{21}. Some authors advised using tissues from an unscarred area (Tunica vaginalis or scrotal dartos layer) for recurrent fistulas\textsuperscript{22}. We selected the dorsal penile dartos fascia as a rotational flap for all the cases in group B with good results for fistula repair with a success rate of 95%. This was in agreement with previously reported data on the effectiveness of multiple layers in preventing fistula recurrence\textsuperscript{23,24,6,25}. However, the success rate of the simple repair technique, which is 70% in our study, contrasted with the first report of Goldstein and Hensle, who reported complete success with the same technique\textsuperscript{26}. Nevertheless, such a high success rate was not confirmed in later series\textsuperscript{27}. There are many possible reasons to justify such differences, e.g., patient selection, technical details not reported in the papers, and the use of urinary diversion, but it is not possible to draw firm conclusions.

The dartos flap is fibro-adipose tissue that may reach the ventral penile shaft without tension. Dartos flaps have been used for the primary waterproofing of hypospadias repair and fistula repair\textsuperscript{28}. We noticed that covering the UCF repair with a well-vascularized dorsal dartos flap after careful harvesting technique represented an excellent option for the management of UCF because it prevented the recurrence of fistulae by supporting the repair and providing the neourethra by sufficient blood supply, which might be depleted or destroyed because of previous surgery. Furthermore, it was uniformly available even in patients with previous penile or scrotal manipulation and was possible to transpose ventrally in any length to cover the urethral repair.

Though the population characteristics were homogenous between the two groups, the layered repair technique was better than the simple repair technique in reducing recurrent fistula formation. Our logical explanation for the significantly improved result in the layered repair group was the penile dorsal dartos flap coverage over urethral repair.

**Conclusion:**

This study revealed that the layered repair technique was better than the simple repair technique regarding recurrent fistula formation. Covering the repaired urethra with a well-vascularized dartos fascial flap harvested from the penile dorsum was a viable surgical option that has given satisfactory results. The significantly improved success rates in the repair with the addition of a waterproofing layer suggested that this interposition layer should be used at the earliest available opportunity to prevent a recurrence rather than to reserve it for future use.

**Conflict of interest:** None

**References:**


