Proximal Hypospadias Repair by Using Longitudinal Preputial Double-Layered Island Flap Technique
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ABSTRACT
Background: Longitudinal Dorsal Island Flap (LDIF) can be employed to overcome the drawbacks of onlay and tubularized urethroplasty in management of hypospadias.

Objective: To improve outcome and reduce complications of cases with proximal hypospadias associated with severe chordee and deficient urethral plate by using longitudinal preputial double-layered island flap two stage technique.

Patients and Methods: We included twenty patients, who had proximal hypospadias associated with deficient urethral plate, and were admitted to Pediatric Surgery Department at Zagazig University Hospital during study period.

Results: The results of our study showed early complication after 1st stage: wound infection only in one case (5%), which was complicated by fistula formation, which was closed spontaneously. After 2nd stage the results showed 3 cases suffering from meatal stenosis managed by repeated dilatation but unfortunately one of them suffering from urethral diverticulum later on. Also, one of cases complicated with urethral fistula, which needed surgical repair. The cosmetic appearance was not good after the 1st stage, but it became better after the 2nd stage as the excessive tissue was removed and the meatus reached the position on glans penis.

Conclusion: For proximal hypospadias repairs, the longitudinal preputial island flap approach has an excellent success rate and is suitable for a wide range of cases. It is considered as good option especially in patients with underdeveloped urethral plate among proximal hypospadias patients.

Keywords: Longitudinal Preputial Double-Layered Island Flap, Proximal Hypospadias.

INTRODUCTION
Pediatric hypospadias is a frequent congenital urinary system abnormality (1). Approximately one in every 100–300 live male infants is reported to have proximal hypospadias, which is found in 20% of instances. Anteroposspadias (middle, coronal, and glanular) make up 87 percent of all hypospadias, with only 13 percent coming from the posterior (or glanular) region (2). The penoscrotal and perineal variants of proximal hypospadias account for 20% of all occurrences. A severe chordee can create a substantial curvature of the penile girdle in certain cases. To straighten the penis, the chordee must be freed (3).

Meatal site, chordee presence, prepuce availability and urethral plate quality are a few characteristics that play a role in determining the type of repair, in addition to the expertise of the surgeon (4).

For more proximal hypospadias or hypospadias with a poorly formed urethral plate, vascularized skin flaps are used for substitute urethroplasty instead of the Snodgrass (Tubularized incised plate) repair. There are a variety of ways to use vascularized preputial and penile skin flaps. Transverse island flaps from the inner prepuce have a number of drawbacks, including penile torsion/rotation and erectile dysfunction. Both inner and outer preputial layers undergo necrosis when they are separated, whereas bad appearance on the ventrum occurs when both are transferred as a single unit. The Longitudinal Dorsal Island Flap (LDIF) can be used both as an onlay and a tubularized urethroplasty to alleviate these disadvantages. It was first popularized by Scuderi and then adapted by others with a few changes (5).

While distal hypospadias is more manageable in pediatric surgery, the presence of severe chordee frequently makes the proximal form of the condition more difficult to treat and increases the risk of postoperative complications. Urethral plate deconstruction is necessary for patients who have hypospadia and significant cord damage. As a result, urethroplasty utilizing transected urethral plate tissue as well as a transverse preputial flap is becoming more popular. Urethral plate deconstruction is necessary for patients who have hypospadias and significant chordal cord transection. Because of this, urethroplasty with a transverse preputial flap in conjunction with a transected urethral plate tissue is becoming more often used (6).

During the last 30 years newer surgical techniques have evolved, with proper understanding of vascularity and healing of flaps. Advances in suture materials, catheters, dressings, hemostasis and magnification have all resulted in better cosmetic and functional results in a large percentage of cases in a single operation (7).

On operating for hypospadias, the surgeon should keep in mind the goals of repair that include straightening of the penis, constructing a neourethra with an appropriate caliber, creating a near-normal location of the meatus, and the cosmetic final appearance of the glans and the whole penis. In the longitudinal preputial double-layered island flap technique, the prepuce is tunneled into a tube and passed ventrally to be anastomosed to the native urethra, and the tube is completed anteriorly by tubularizing the glanular urethral plate and

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anastomosing it to the distal end of the preputial tube. Those who treat this difficult problem must be well oriented with different varieties of these techniques, which reportedly number more than 200 [8].

It was the goal of this study: to improve outcome and reduce complications of cases with proximal hypospadias associated with severe chordee and deficient urethral plate by using longitudinal preputial double-layered island flap two stage technique.

SUBJECTS AND METHODS

At Pediatric Surgery Department of Zagazig University Hospitals, we conducted this clinical trial on 20 patients who had proximal hypospadias with deficient urethral plate, and were treated by the using longitudinal preputial double-layered island flap in two stages.

Ethical consent:

Research Ethics Council at Zagazig University approved the study (ZU-IRB #9634) as long as all participants’ parents provided informed consent forms. Ethics guidelines for human experimentation were adhered to by the World Medical Association's Helsinki Declaration. Inclusion criteria: Patients with proximal hypospadias (urethral meatus in peno-scrotal, scrotal or perineal) with deficient urethral plate and severe chordee (Figure 1).

Figure (1): Proximal hypospadias "different types"
Exclusion criteria: No complaint patients, previously circumcised patients, recurrent cases with absent prepuce, diabetic or immune compromised patients, and patients with complex urogenital malformations or disorder of sexual development.

All patients were subjected to:

1. A thorough review of the patient's medical history and local examination to assess type of hypospadias, degree of chordee and state of urethral plate examination.

2. All patients had full preoperative lab investigation before surgery including: Complete blood picture, random blood sugar, viral screen, coagulation studies (PT/PTT) as well as kidney and liver function tests.

3. Surgical technique:

   The procedure done in two stages: 1st stage and 2nd stage within 3 months after 1st stage.

   1st stage:

   Neourethra was designed from the preputial inner skin and lengthened by 0.5 cm from the dorsal preputial foreskin. A transverse incision of 0.5–1 cm was made in the inner skin immediately behind the glans penis. Two parallel longitudinal incisions were made in the inner skin from the ends of the transverse incision to the junction between the inner skin and the foreskin of the prepuce and extended to 0.5 cm in the foreskin on the dorsal aspect of the prepuce.

   A transverse incision was made on the dorsal aspect of the preputial skin connecting the two ends of the previously extended incisions coming from the ventral aspect of the penis. A 0.5-cm longitudinal incision (buttonhole) was made on the dorsal aspect of the penile skin 1.5–2 cm from the root of the penile shaft to allow passing of catheter through it, and later transferring the glans penis without interfering with the preputial vascularity (Figure 2).

   Figure (2): Catheter passing through a longitudinal incision in dorsal skin.

   Starting with elevation of 0.5 cm of skin from the dorsal aspect of the prepuce to elongate the preputial inner skin, the inner skin was minimally elevated from foreskin on sides to allow its tubularization without complete separation from the dorsal skin. A transverse incision was made in the short fibrous urethral plate and corpus spongiosum down to the corpora cavernosa and dissection of all fibrous tissues was carried out to straighten the hooked penis.

   A circular incision around the hypospadiac opening was made and the skin was elevated from the opening to prepare for anastomosis with the distal end of transferred inner preputial tube (neo-urethra). Tubularization of the neourethra around a 6–8 Fr catheter was started with the skin elevated from the dorsal aspect and continued to the hind skin of the inner aspect of the prepuce with 5/0 polyglycolic acid sutures. The excessive mucosa was excised, and incision was extended to reach the incision on the ventral aspect of the penis (Figure 3).

   Figure (3): Neourethra formed around the 8 Fr catheter and excised the excess mucosa after designing the tube.

   After full straightening of the penile shaft and excision of all fibrous tissues, the glans penis was passed through the dorsal skin opening (button hole) behind the catheter and the neourethra. After transferring the flap to the ventral aspect, the proximal end of the tube became distal and sutured to the remaining of urethral plate, and distal end sutured to the hypospadiac opening, here the suture line of neourethra became dorsal.

   Then the catheter was passed through the hypospadiac urethral opening and introduced into the urinary bladder. After transfer the formed neo-urethra the distal end became proximal and anastomosed to hypospadiac opening and the suture line of neo-urethra became dorsal, and the proximal end became distal and was sutured to the remnant of urethral plate at the coronary sulcus. Then a layer of subcutaneous sutures was applied to cover the anastomosis and separate it from skin sutures (Figure 4).
2nd Stage:
It was done within 3 months after the 1st stage, it was used to complete and elongate the urethra to reach the needed position on glans penis by using TIP urethroplasty technique and excise the excess skin (Figure 5). A longitudinal incision was done to separate one pedicle of transferred flap and subcutaneous tissue was used as second layer to cover the performed tube and excess redundant skin was excised (Figure 6).

During the two stages operation degloving of skin was not performed and the hemostasis was carefully controlled with electrocautery. A pressure dressing was applied at the end of procedure.

Postoperative treatment:
Prophylactic broad-spectrum antibiotic (Augmentin®) was administered for 7 days. Dressings was removed 48 hours after correction, and the penis was left exposed with the application of local antibiotic cream. Children were discharged from hospital 2–3 days after correction. Urethral catheter was kept for 7 days postoperatively. Patients were followed up for any complications and urethral calibration was performed after one month from operation every week for 1 month, and every 2 weeks for another 2–3 months.

Statistical analysis
In order to analyze the data acquired, Statistical Package for the Social Sciences (SPSS) version 20 was used to execute it on a computer. The quantitative data were presented in the form of the mean, standard deviation, and range. The qualitative data were presented as frequency and percentage.

RESULTS
Table 1 shows the mean age of studied group is 23.05±13.38 with range (9-48).

<table>
<thead>
<tr>
<th>Age in months</th>
<th></th>
</tr>
</thead>
<tbody>
<tr>
<td>Mean± SD</td>
<td>23.05±13.38</td>
</tr>
<tr>
<td>Range</td>
<td>9-48</td>
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</tbody>
</table>

Table 2 shows the distribution of types of proximal hypospadias among patients.

<table>
<thead>
<tr>
<th>Type</th>
<th>N</th>
<th>%</th>
</tr>
</thead>
<tbody>
<tr>
<td>Penoscrotal</td>
<td>13</td>
<td>65</td>
</tr>
<tr>
<td>Scrotal</td>
<td>5</td>
<td>25</td>
</tr>
<tr>
<td>Perineum</td>
<td>2</td>
<td>10</td>
</tr>
</tbody>
</table>

Figure (4): Final picture after 1st stage

Figure (5): Longitudinal incision in one pedicle and subcutaneous tissue was used as a second layer

Figure (6): Final picture after 2nd stage.
Table 3 shows the mean 1st stage operative time.  
**Table (3): 1st stage operative time**

<table>
<thead>
<tr>
<th>Operative time in minutes</th>
<th>Mean± SD</th>
<th>Range</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>69.25± 7.82</td>
<td>60-80</td>
</tr>
</tbody>
</table>

Table 4 showing the mean 2nd stage operative time.

**Table (4): 2nd stage operative time**

<table>
<thead>
<tr>
<th>Operative time in minutes</th>
<th>Mean± SD</th>
<th>Range</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>58.75±6.85</td>
<td>50-70</td>
</tr>
</tbody>
</table>

Distribution of complications:

Table 5 shows the complications after 1st stage. Wound infection occurred in one patient, regardless the appropriate management this patient was complicated by urethrocutaneous fistula, which was closed spontaneously within weeks. The cosmetic appearance after the first stage was not good as still there was an excessive tissue as well as the meatus was still not in normal position.

**Table (5): Frequency distribution of complications after first stage**

<table>
<thead>
<tr>
<th></th>
<th>N</th>
<th>%</th>
</tr>
</thead>
<tbody>
<tr>
<td>Edema: No</td>
<td>20</td>
<td>100</td>
</tr>
<tr>
<td>Edema: Yes</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>Infection: Yes</td>
<td>1</td>
<td>5</td>
</tr>
<tr>
<td>Infection: No</td>
<td>19</td>
<td>95</td>
</tr>
<tr>
<td>Fistula: Yes</td>
<td>1</td>
<td>5</td>
</tr>
<tr>
<td>Fistula: No</td>
<td>19</td>
<td>95</td>
</tr>
<tr>
<td>Recurrent ventral curvature: Yes</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>Recurrent ventral curvature: No</td>
<td>20</td>
<td>100</td>
</tr>
<tr>
<td>Lateral deviation: Yes</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>Lateral deviation: No</td>
<td>20</td>
<td>100</td>
</tr>
<tr>
<td>Diverticulum: Yes</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>Diverticulum: No</td>
<td>20</td>
<td>100</td>
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Table 6 showing distribution of complications after second stage.

**Table (6): Frequency distribution of complications after second stage**

<table>
<thead>
<tr>
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<th>N</th>
<th>%</th>
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</thead>
<tbody>
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<td>20</td>
<td>100</td>
</tr>
<tr>
<td>Edema: Yes</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>Infection: Yes</td>
<td>1</td>
<td>5</td>
</tr>
<tr>
<td>Infection: No</td>
<td>19</td>
<td>95</td>
</tr>
<tr>
<td>Fistula: Yes</td>
<td>1</td>
<td>5</td>
</tr>
<tr>
<td>Fistula: No</td>
<td>19</td>
<td>95</td>
</tr>
<tr>
<td>Meatal stenosis: Yes</td>
<td>3</td>
<td>15</td>
</tr>
<tr>
<td>Meatal stenosis: No</td>
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<td>85</td>
</tr>
<tr>
<td>Gland dehiscence: Yes</td>
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<td>0</td>
</tr>
<tr>
<td>Gland dehiscence: No</td>
<td>20</td>
<td>100</td>
</tr>
<tr>
<td>Lateral deviation: Yes</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>Lateral deviation: No</td>
<td>20</td>
<td>100</td>
</tr>
<tr>
<td>Urethral diverticulum: Yes</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>Urethral diverticulum: No</td>
<td>20</td>
<td>100</td>
</tr>
</tbody>
</table>

DISCUSSION

To correct penoscrotal hypospadias, the urethra, penile skin, and accompanying chordee must be reconstructed in a complex manner. As a replacement for both the urethra and penile cap, the prepuce comes highly recommended (9). However, the optimum surgical strategy for severe hypospadias has yet to be determined; numerous approaches have been used to correct the accompanying abnormalities. With the exception of the Koyanagi procedure, the preputial island flap has been associated with a lower complication rate than other approaches such as the buccal mucosa transplant (10).

In this study we aimed to focus on longitudinal preputial flap in two stages for hypospadias repair, we tried to find the outcome, results and rate of complications.

In transverse preputial flap techniques, the chordee is completely dissected to correct the curvature. Hence, the transverse preputial flap is spirally rotated around the penile shaft by its pedicle to be positioned ventrally and anastomosed to the native urethra and then a urethral tube is designed. This spiral rotation may cause tethering of the opposite edge and penile rotation (8).

In 1983, Scuderi and Campus (11) introduced the dorsal vertical flap technique, which was later popularized by Perović and Vukadinović (12) and others. Because the dorsal vascularized flap is pulled down ventrally by buttonholing the mesentery, there is no torsion/rotation of the penis, a common complication following island flaps (2).

We included 20 cases of proximal hypospadias in this study, we excluded any case of penile hypospadias where sacrifice of the urethral plate was one of the steps of the procedure. Only virgin uncircumcised cases with adequate penis size
were chosen. Part of the preoperative evaluation was to exclude local infection, associated comorbidities or congenital anomalies that may make the candidate risky for anesthesia. Intersex, whenever suspected, was also carefully excluded. After inclusion, the cases were repaired by longitudinal perpetual island flap technique in two stages.

Regarding the main age of our study cases it was 23.05 months. In comparison to other studies, which repaired the proximal hypospadias cases by preputial flap, our cases mean age was similar to that of the study of Saleh et al. (8) (26.7 months), while Al Hindi and Khalaf (10) operated upon the cases in younger age, it was 16 months, while Castanon et al. (13) operated upon the cases around 37 months.

In our study, the main operative time was 69.25 minutes for 1st stage and 58.75 minutes for 2nd stage. While Zaki et al. (14) main operative time was 110 min, on other hand Al Hindi and Khalaf(10) operated in time more than 150 minutes as they were managing the cases in one stage only.

Totally, postoperative complication after 1st stage were wound infection, which occurred in one case (5%) and urethrocuteaneous fistula in one case also (5%). And after the 2nd stage the complications were meatal stenosis in 3 cases (15%), urethral diverticulum in one case (5%) and fistula in one case (5%).

Regarding the edema it did not happen in our study cases, this may due to no degloving done during the procedure as well as the tourniquet was not used during the two stages. In Saleh et al. (8) study there was a mild edema in 17.5%, on other hand the percent was 100% in Zaki et al. (14) study, it may be due to complete degloving, using of tourniquet and longer duration of operation time (110 minutes).

The incidence of urethrocuteaneous fistula in our study was 10% in total for the two stages, which is similar percent in Ali et al. (15) study (10.5%) as well as Chalapathi et al. (2) study (9.33%) and larger than in Daboos et al. (9) study (5%), who used a single stage in his study, therefore the high percent in our study apparently due to more operation sitting and stages. The fistula here was mostly between the anastomosis of neourethra with proximal hypospadias urethral tube distally.

De Badiola et al. (16) found that ventral tubularized and double faced tubularized island flap techniques both had the same rate of fistulas, according to the study (25 percent and 30 percent, respectively). In their series, the double faced tubularized flap technique had a greater incidence of diverticulum (13%) than the ventral tubularized flap approach (3 percent). Urinary leakage from the neourethra's repair or the production of an enlarged segment were both factors in the development of diverticulum, according to the researchers (17).

Only one (5 percent) case of urethral diverticulum was found in this investigation, which could be the result of four different technical improvements: (1) The tubularized flap's length and width must be precisely measured and adjusted (to avoid oversized urethra). (2) Urethral tube fixation at the suture line between the corpora and the glans of the new tube (avoiding tube laxity). (3) Proper shaping of the glans to create an oval meatus that is wide enough to swallow (avoiding stenosis of distal meatal). (4) Meatal stenosis dilatation in its early stages.

Proximal hypospadias correction must also address the problem of significant ventral curvature. Compared to urethral plate transection, simple dorsal plication was associated with a recurrence risk of roughly 37% (18).

When it came to ventral curvature (chordee), there was no recurrence in this study's follow-up period. Intraoperative chordee correction via transected urethral plate and removal of all fiber bands around the corpus spongiosum may be a possible explanation for this.

In theory, a longer urethral defect could make surgery more challenging, increasing the risk of complications. Neourethra lengths of less than 3 cm were associated with a lower risk of problems, as were lengths greater than 3 cm (19). Even though the urethral defect was lengthy, we didn't discover a greater rate of problems in this study since the repair was done in two stages.

CONCLUSION

For proximal hypospadias repairs, the longitudinal preputial island flap approach has an excellent success rate and is suitable for a wide range of cases.

Reoperations are rare with this procedure, which can be used for a wide range of hypospadias repairs in the proximal region and produces excellent results. Therefore, this operation for staged longitudinal preputial island flap procedure in cases of proximal hypospadias with deficient urethral plates is a viable choice.

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Author contribution: Authors contributed equally in the study.

REFERENCES


