Staged segmental urethroplasty for scrotal/perineal hypospadias; a new concept

SEVERE HYPOSPADIAS AND URETHROPLASTY

Asuka Ishiyama¹, Shogo Seo¹, Hiroshi Murakami¹, Takanori Ochi¹, Manabu Okawada¹,
Takashi Doi¹, Go Miyano¹, Hiroyuki Koga¹, Geoffrey J. Lane¹, Hidenori Haruna²,
Toshiaki Shimizu², Atsuyuki Yamataka¹.

Department of Pediatric General and Urogenital Surgery¹
Department of Pediatrics and Adolescent Medicine²
Juntendo University School of Medicine
Tokyo, Japan

Corresponding author:
Atsuyuki Yamataka
Department of Pediatric General and Urogenital Surgery
Juntendo University School of Medicine
2-1-1 Hongo, Bunkyo-ku, Tokyo 113-8421, Japan
Tel: +81-3-3813-3111
Fax: + 81-3-3817-0736
E-mail: yama@juntendo.ac.jp
ABSTRACT

PURPOSE: We report the efficacy of staged segmental urethroplasty (SSUP) versus non-staged urethroplasty (NSUP) for treating scrotal/perineal hypospadias (SPH).

METHODS: Between 1997 and 2015, 29 SPH patients underwent UP (SSUP: n=15; NSUP: n=14). Incidences of urethrocutaneous fistula (UF), stenosis of the neourethra (SNU), diverticula formation, and residual chordee (RC) were compared. Differences were statistically significant if p<.05.

RESULTS: The difference in mean age at NSUP (3.2±1.3 years) and at the final stage of SSUP (5.5±2.4 years) was significant (p<.05). Mean operative times for NSUP and SSUP (total for all stages) were not significantly different (231.5±117.5 versus 272.5±99.4 minutes), however, the incidence of postoperative complications was significantly less in SSUP (n=1; UF) compared with NSUP (n=6; 2 cases of UF, 3 cases of SNU, and 1 case of RC; (p<.05). Mean follow-up was significantly shorter in SSUP; 1.4±1.2 years versus 7.0±4.5 years in NSUP (p<.05).

CONCLUSION: SSUP would appear to be effective for treating SPH because of a significantly lower incidence of UF, SNU and RC during the first postoperative year, the period when complications have been reported to arise most frequently.

KEY WORDS:
hypospadias, staged segmental urethroplasty, scrotal/penoscrotal hypospadias, complications
INTRODUCTION

Proximal hypospadias is one of the most challenging conditions to correct. Despite multiple procedures having been reported over the years, the ideal surgical management of primary proximal hypospadias remains controversial. Because correction of scrotal/perineal hypospadias (SPH) is associated with high complication rates (14.3-60.0%) [1,2,3] we developed a staged segmental urethroplasty (SSUP) for treating SPH to improve outcome. SSUP involves performing the urethroplasty (UP) in two stages after chordectomy. In other words, our SSUP comprises chordectomy as the “first stage”, the initial UP as the “second stage”, and the final UP as the “third stage”. Here we report our technique for staged segmental urethroplasty (SSUP) developed in an attempt to decrease the incidence of complications that arise secondary to the extensive urethral reconstruction required in patients with SPH, and compare it with non-staged urethroplasty (NSUP) for efficacy of correcting SPH.

MATERIALS AND METHODS

Subjects

Data from 29 SPH patients treated by a single surgeon from January 1997 to July 2015 using SSUP (n=15) and NSUP (n=14) were collated retrospectively. Only SPH cases were reviewed in this study. All other types of hypospadias were excluded. We developed SSUP in 2010 to decrease the incidence of complications in patients who had surgery for SPH. Until then we performed conventional two stage repair (NSUP) even for severe cases. All subjects had hormone therapy with testosterone enanthate 25mg administrated intramuscularly once a month for 3 months before surgery. The length of the penis was measured at the outpatient clinic using a ruler placed on the pubic fat pad pre and post hormone therapy.

Surgical technique

Correction of curvature

Chordee is measured intraoperatively using a goniometer after induction of artificial erection and classified as mild if curvature is between 15 and 30 degrees, moderate if between 30 and 45 degrees, and severe if over 45 degrees. In all cases, a circumcision incision is made, and the skin degloved down to the base of the penis. Any abnormal dense fibrous tissue covering the urethra is excised. Penile curvature is corrected by dorsal plication [4] if minimal to mild provided the ventral shaft is long enough to prevent shortening after plication using two or four sets of 5/0 nonabsorbable sutures (Prolene®) plication sutures placed in the tunica albuginea at
1 and 11 o’clock, respectively. Artificial erection is induced to assess residual curvature. If curvature is severe, or if the ventral shaft is short or the penis is likely to become shorter after dorsal plication, curvature is corrected by incising the tunica albuginea and using a pedicled tunica vaginalis flap [5].

_Non-staged urethroplasty (NSUP)_ [Figure 1]

NSUP is performed after correction of curvature using a Snodgrass tubularized incised plate urethroplasty or a modification of the Thiersch-Duplay technique in which a midline longitudinal relaxing incision is made before creating the entire neourethra around a 6 or 8-French urethral stent [6-8] using 7/0 PDS® single layer interrupted sutures [Figure 2]. Suture lines in the neourethra are covered entirely by pedicled soft tissue such as foreskin subcutaneous tissue, peri-meatal adipose tissue, pedicled external spermatic fascia, peri-cordal adipose tissue, scrotal adipose tissue or a combination of these [9] if the tissue of the neo-urethra is thin or if there is risk for urethrocutaneous fistula formation [9].

_Staged segmental urethroplasty (SSUP)_ [Figure 3]

For the first stage segmental urethroplasty, a U-shaped incision is made from the bottom of the urethral orifice to the penoscrotal junction and the ventral penile skin is tubularized with 7/0 PDS according to the modified Thiersch-Duplay technique we use for NSUP, except that only a segment of neourethra is created and the meatus is advanced only to the penoscrotal junction. The urethral stent is removed between 7-10 days postoperatively. The second stage segmental urethroplasty is performed after a period of 6–12 months and involves creating the rest of the urethra and advancing the meatus at the penoscrotal junction towards the glans according to the same technique used during the first stage and for NSUP [Figure. 4]. The suture line of the neourethra is covered with pedicled soft tissue if the tissue of the neo-urethra is thin or if there is risk for urethrocutaneous fistula formation. The urethral stent is removed between 7-10 days postoperatively.

_Penile cosmesis_

Satisfaction with penile cosmesis was evaluated by using a questionnaire administered three months after the last procedure during an outpatient clinic visit (see appendix). Response options were: disappointed; poorer than expected, acceptable, and happy. The questionnaire was returned anonymously to a box in the clinic and collected by the clinic clerk. Our routine follow-up schedule for SPH patients is outpatient attendance 7 days after discharge from hospital after the final procedure, then at 1, 3, 6, 12, 18, 24, 36 months, then every second year until puberty.

_Statistical analyses_
Data were evaluated by Pearson’s chi-squared test, Fisher’s exact test, Student’s t-test, and the Mann-Whitney U test. Statistical significance was determined if $p < .05$.

**Ethics**

This study was approved by the Ethics Committee of Juntendo University School of Medicine and complies with the Helsinki Declaration of 1975 (revised 1983).

**RESULTS**

Our series comprised 29 SPH cases: scrotal ($n=12$) and perineal ($n=17$). Breakdown per group was NSUP: scrotal ($n=8$) and perineal ($n=6$), SSUP: scrotal ($n=4$) and perineal ($n=11$). Severity of hypospadias according to the location of the native meatus was not significantly different between the 2 groups. The difference in mean age at NSUP (3.2±1.3 years) and at the final stage of SSUP (5.5±2.4 years) was significant ($p < .05$). Mean operative times (minutes) were not significantly different; 231.5±117.5 for NSUP versus 272.5±99.4 for SSUP (total for all stages) [Table 1]. Incidence of postoperative complications was significantly less in SSUP ($n=1$; UF) compared with NSUP ($n=6$; 2 cases of UF, 3 cases of SNU, and 1 case of RC) ($p < .05$) [Table 2]. Follow-up was significantly shorter in SSUP: 1.4±1.2 (range: 0.3-3.3) years versus 7.0±4.5 (range: 0.4-12.6) years in NSUP ($p < .05$) [Table 1]. There were no statistically significant differences in preoperative parameters [Table 3]. The response rate to the penile cosmesis questionnaire was 29/29 (100%). After SSUP, outcome was acceptable (6.7%) or respondents were happy (93.3%) and after NSUP, outcome was acceptable (14.3%) or respondents were happy (85.7%) [Table 1]. Responses to the penile cosmesis questionnaire were not significantly different between the two groups.

**DISCUSSION**

The urethral meatus is located in the scrotum or perineum in approximately 10% of boys with hypospadias [10] and as a result there are few reports focusing exclusively on SPH and those that do present a spectrum of outcomes all associated with complications. Irina and Hoang reported a complication rate of 60.0% and Snodgrass and Lorenzo et al. reported a complication rate of 57.1%, while Hadidi AT et al. reported a complication rate of 14.3% [1,2,3]. Essentially, SPH poses additional problems during reconstruction because the anatomic anomalies typical of proximal hypospadias are compounded. In other words, there is almost no usable native urethra present and little supporting vasculature, and the length of neourethra that must be created to bring the neomeatus to a more normal position is long. To improve success, a two-stage approach has
become common for correcting proximal hypospadias, because of superior cosmetic and functional results with fewer complications. By staging urethral reconstruction, the length of urethra created at each operation is far more manageable at up to 3 cm, suitable for tubularization [11], and probably less likely to be associated with complications. To the best of our knowledge this is the first report of an appraisal of staged segmental urethroplasty (SSUP) for SPH repair and in the present study, our complication rate after SSUP was 6.7% and after NSUP was 42.9% which would appear to favor SSUP over NSUP for the surgical repair of SPH if complication rate is considered a valid marker of success.

Earl and Bradley et al. reported a similar procedure to our SSUP [12]. Their first stage involves dividing the urethral plate proximally at the level of the native (hypospadiac) meatus and tubularizing it using Snodgrass’ technique in a single procedure. The gap in the urethra extending from mid-shaft proximally is then created 6 months later. Our procedure however involves advancing the meatus only to the penoscrotal junction during the first stage urethroplasty then creating the rest of the urethra and advancing the meatus at the penoscrotal junction towards the glans during the second stage urethroplasty. Thus, while both are staged repairs, they involve different concepts. We believe our SSUP is easier to perform and master because we use a technique that is almost the same as conventional Snodgrass tubularized incised plate urethroplasty at each stage.

While our postoperative complication rate was less, some surgeons are concerned that multiple operations could increase morbidity because of surgical stress [3,13] and our complete SSUP involves three operations (correction of curvature followed by segmental urethral reconstruction performed in two stages). While this opinion is valid, one must also bear in mind that repair of complications such as UF, SNU or diverticulum after hypospadias surgery is problematic and often unsuccessful with a high rate of recurrence [15,16]. Erin and Anthony et al. reported a complication rate of 53% for two stage urethroplasty with 35% experiencing more than two complications on long-term follow-up [17]. Down time related to surgery and repeat surgery for complications may also impair the quality of life of both the patient and his family and cause stress because of cosmetic and functional inadequacy. As a result, our feeling is that if better results are forthcoming by waiting and performing SSUP, then our procedure of choice for SPH repair would have to be SSUP because there are less complications, and less impact on the patient and his family. Because of this, the mean age at the final stage of SSUP was significantly older than the mean age at NSUP, which was also older than the generally recommended age of 6-18 months for genital surgery in boys [14].

We believe the success of SSUP in our series is correlated to the period we leave between stages not only to allow recovery from surgical trauma and inflammatory changes but also to restore blood flow disrupted by
surgery and ensure that vascularity of subcutaneous tissues is as good as possible. We recommend a period of 6-12 months between the first and second stages so that each segment of neourethra has the best chance to grow.

Other factors affecting postoperative outcome generally, such as operative time were not relevant in our series. Overall operative time in SSUP cases was not significantly different from NSUP cases but was divided over time so that each stage of SSUP was shorter which we believe also contributes to success because there is less influence of fatigue during surgery on performance.

Mean follow-up is biased in this study because of a change in our routine procedure for treating SPH from NSUP to SSUP in 2010. Thus, the earliest SSUP case with the longest follow-up would be roughly 2.5 years while there are no NSUP cases with less than 2.5 years follow-up so duration of follow-up in this study is not particularly meaningful but as duration of follow-up for SSUP cases approaches the duration of NSUP cases, we will be able to compare outcomes more accurately. In particular, functional and cosmetic outcome during and after puberty are important issues that we hope to report in the future. Po and Saul reported the long-term outcome of two-stage repair using a questionnaire and 92% were pleased with the cosmetic results [18]. In the present study, there were no significant differences between the two groups with regard to cosmetic results but our questionnaire was administered three months postoperatively during an outpatient clinic visit and focused only on appearance with no opportunity to evaluate general problems and concerns, mental and physical stress related to polysurgery, or issues related to puberty. We will continue to follow-up and accumulate further data.

In conclusion, SSUP would appear to be effective for treating SPH because there were less postoperative complications (specifically, UF, SNU and RC) during the first postoperative year in our series, the period when postoperative complications are reported to arise more frequently in the literature.
APPENDIX

---Penile cosmesis questionnaire---

Q1. Did you have two urethroplasty operations?
   • Yes
   • No

Q2. Do you think your penis is straight on erection?
   • Yes
   • No

Q3. If not, please draw how your penis looks on erection

Q4. How satisfied are you with the general appearance of your penis?
   • Disappointed
   • Poorer than expected
   • Acceptable
   • Happy
REFERENCES


FIGURE LEGENDS

Figure 1
For non-staged urethroplasty, correction of curvature is performed initially, followed by urethroplasty some 6-12 months later. The neourethra is created from the native orifice to the glans during one procedure.

Figure 2
(A) For non-staged urethroplasty, after 12 months from correction of curvature, a neourethra is created from the perineum (arrow) to the glans (arrow head).
(B) Post-urethroplasty, the penis looks satisfactory.

Figure 3
For staged segmental urethroplasty, after correction of curvature, a segment of neourethra is created from the native orifice to the penoscrotal junction, then 6-12 months later, another segment of neourethra is created from the penoscrotal junction to the tip of the glans.

Figure 4
(A) After correction of curvature, a segment of neourethra is created from the native urethral orifice (arrow) to the penoscrotal junction (arrow head).
(B) Some 12 months later, the penis has recovered from surgical trauma and inflammatory changes. The position of the native urethral orifice (arrow) and the intermediary neo-orifice (arrow head) are shown.
(C) The rest of the urethra is created and the meatus is advanced from the penoscrotal junction to the tip of the glans.
(D) Some 12 months later, cosmetic and functional results are good.
Fig. 1

First repair
Correction of curvature

Second repair
Urethroplasty
Fig. 3

First repair
correction of curvature

Second repair
1st urethroplasty

Third repair
2nd urethroplasty
<table>
<thead>
<tr>
<th></th>
<th>NSUP (N=14)</th>
<th>SSUP (N=15)</th>
<th>p</th>
</tr>
</thead>
<tbody>
<tr>
<td>Mean age at NSUP or final stage of SSUP (years)</td>
<td>3.2±1.3</td>
<td>5.5±2.4</td>
<td>&lt;0.05</td>
</tr>
<tr>
<td>Mean operative time (minutes)</td>
<td>231.5±117.5</td>
<td>272.5±99.4 (total)</td>
<td>0.47</td>
</tr>
<tr>
<td>Use of soft tissue interposition</td>
<td>11</td>
<td>13</td>
<td>1.0</td>
</tr>
<tr>
<td>Mean follow-up period (years)</td>
<td>7.0±4.5</td>
<td>1.4±1.2</td>
<td>&lt;0.05</td>
</tr>
<tr>
<td>Satisfaction with penile cosmesis</td>
<td>Happy: 81.8%</td>
<td>Happy: 92.3%</td>
<td></td>
</tr>
<tr>
<td></td>
<td>Acceptable: 18.2%</td>
<td>Acceptable: 7.7%</td>
<td></td>
</tr>
</tbody>
</table>

NSUP: non-staged urethroplasty, SSUP: staged segmental urethroplasty
Table 2  Details of complications

<table>
<thead>
<tr>
<th>Complications</th>
<th>NSUP (N=14)</th>
<th>SSUP (N=15)</th>
<th>p</th>
</tr>
</thead>
<tbody>
<tr>
<td>UF: 2</td>
<td>UF: 1</td>
<td>&lt;.05</td>
<td></td>
</tr>
<tr>
<td>SNU: 3</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>RC: 1</td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

UF: urethrocutaneous fistula
SNU: stenosis of the neourethra
RC: residual chordae
<table>
<thead>
<tr>
<th></th>
<th>NSUP</th>
<th>SSUP</th>
<th>( p )</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Location of the native meatus</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Scrotal</td>
<td>8</td>
<td>4</td>
<td>0.14</td>
</tr>
<tr>
<td>Perineal</td>
<td>6</td>
<td>11</td>
<td></td>
</tr>
<tr>
<td><strong>Mean length of the penis before HRT (cm)</strong></td>
<td>2.2±0.3</td>
<td>2.1±0.4</td>
<td>1.0</td>
</tr>
<tr>
<td><strong>Mean length of the penis after HRT (cm)</strong></td>
<td>3.2±0.3</td>
<td>3.3±0.2</td>
<td>0.48</td>
</tr>
<tr>
<td><strong>Type of DSD</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Specific DSD</td>
<td>2</td>
<td>4</td>
<td>0.65</td>
</tr>
<tr>
<td>Clinical DSD</td>
<td>12</td>
<td>11</td>
<td></td>
</tr>
<tr>
<td><strong>Cryptorchidism</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Yes</td>
<td>5</td>
<td>8</td>
<td>0.46</td>
</tr>
<tr>
<td><strong>Severity of chordee</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Mild</td>
<td>3</td>
<td>5</td>
<td>0.28</td>
</tr>
<tr>
<td>Moderate</td>
<td>7</td>
<td>9</td>
<td></td>
</tr>
<tr>
<td>Severe</td>
<td>4</td>
<td>1</td>
<td></td>
</tr>
</tbody>
</table>

HRT: hormone replacement therapy

DSD: disorder of sexual development
Table 2  Details of complications

<table>
<thead>
<tr>
<th>Complications</th>
<th>NSUP (N=14)</th>
<th>SSUP (N=15)</th>
<th>p</th>
</tr>
</thead>
<tbody>
<tr>
<td>UF: urethrocutaneous fistula</td>
<td>2</td>
<td>1</td>
<td>&lt;.05</td>
</tr>
<tr>
<td>SNU: stenosis of the neourethra</td>
<td>3</td>
<td></td>
<td></td>
</tr>
<tr>
<td>RC: residual chordee</td>
<td>1</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

UF: urethrocutaneous fistula
SNU: stenosis of the neourethra
RC: residual chordee