Primary Severe Hypospadias: Comparison of Reoperation Rates and Parental Perception of Urinary Symptoms and Cosmetic Outcomes Among 4 Repairs

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Purpose: We compared complication rates, urinary symptoms and cosmetic outcomes as perceived by parents of patients undergoing 1 of 4 repairs for proximal hypospadias associated with ventral curvature.

Materials and Methods: A total of 93 patients underwent hypospadias repair between 2004 and 2010. In patients requiring no urethral plate transection the repair consisted of tubularized incised plate urethroplasty (26 patients) or onlay island flap urethroplasty (31). In patients requiring urethral plate transection the repair consisted of onlay island flap on albuginea (18 patients) or 2-stage repair (18). Complications were assessed by chart review. A customized questionnaire and the Pediatric Penile Perception Score were administered to parents to evaluate their perception of urinary symptoms and cosmetic outcomes, respectively.

Results: After a median followup of 4.5 years (range 2.2 to 8.4) complications developed in 21 patients (23%) without any difference among procedures or between patients who did and did not require urethral plate transection. Parents of 75 patients (80%) participated in the survey without differences among repairs (p = 0.35). Reported urinary symptoms were not different among repairs. For Pediatric Penile Perception Score the only difference concerned the question about penile length (p = 0.03), with the score being significantly better for the techniques requiring urethral plate transection (p = 0.05). The 2-stage repair had a significantly better score for the question about penile length and overall Pediatric Penile Perception Score than all other techniques.

Conclusions: Overall complication rates were comparable among repairs and did not increase after urethral plate transection. Urinary symptoms as reported by parents were comparable among the procedures. Perceived penile length was significantly better after urethral plate transection. The 2-stage repair yielded the best cosmetic results.

Key Words: hypospadias; penis; urethra; urologic surgical procedures, male

Abbreviations and Acronyms
OIF = onlay island flap
OIFA = onlay island flap on albuginea
PPPS = Pediatric Penile Perception Score
TIP = tubularized incised plate
UP = urethral plate

One of the most important variables in selecting the appropriate technique for primary severe hypospadias repair seems to be the need for urethral plate transection to achieve penile straightening.1 If the urethral plate can be preserved, onlay island flap urethroplasty has long been considered the standard,2,3 whereas recently the tubularized incised plate repair has been suggested to achieve a more anatomically normal neourethra and a more normal appearing penis.4 If the urethral plate is transected instead, most surgeons prefer a 2-stage repair.5–7 Nevertheless, sev-
eral other techniques exist to attempt a single stage repair under these circumstances.\(^5,9\) We have recently proposed a modified onlay island flap in which the preputial flap is sutured directly on the albuginea of the corpora cavernosa in the gap between the ends of the divided urethral plate (onlay island flap on albuginea).\(^10\)

Along such an apparently easy pathway the surgeon faces several dilemmas. The point at which UP transection should be favored over the use of dorsal shortening procedures for straightening is still somewhat subjective, and it is unclear whether preserving the UP could make the urethroplasty safer or, conversely, UP transection could make penile straightening more effective.\(^11\) In patients undergoing UP transection it is still controversial as to whether staging the repair would actually decrease the complication rate and improve outcomes or just increase the number of surgeries.\(^12\)

Few studies exist comparing different procedures to help in the decision making,\(^2,3,5-9,12-15\) and exceptionally functional outcomes are considered in addition to surgical complications.\(^16\) It has become increasingly evident that cosmetic outcomes should also be taken into account,\(^1\) and validated instruments have been developed for this purpose.\(^17,18\) We compared complication rates, urinary symptoms and cosmetic outcomes as perceived by parents of patients undergoing 1 of 4 techniques for the treatment of primary severe hypospadias to assess whether 1) UP transection increased the surgical complication rate, 2) UP transection improved penile straightening and 3) staging the repair improved the results in patients requiring UP transection.

**MATERIALS AND METHODS**

All 93 patients with primary proximal hypospadias associated with ventral curvature treated at our institution between January 2004 and June 2010 were considered for the study. All patients were treated by 2 surgeons. Preoperative evaluation was consistent during the study period.\(^19\) In instances of small penis hormonal stimulation consisting of 1 to 2 doses of 25 mg testosterone parenterally was administered monthly, ending 1 month preoperatively.

In all the cases the repair started with a sequential approach for penile straightening, including skin degloving, ventral dissection of the corpora cavernosa up to the bulbar urethra, UP mobilization and, eventually, UP transection. Passage from one step to the following one was done for residual curvature greater than 30 degrees, as shown by an erection test. Lower degrees of curvature were addressed using dorsal plication with or without corporeal relaxing incisions or fairy cuts to lengthen the ventral penile surface.\(^3\)

After straightening the urethroplasty was accomplished based on surgeon preference, using OIF or TIP in cases where the urethral plate continuity was preserved, or OIFA or 2-stage technique otherwise.\(^1\) In all 2-stage repairs a preputial graft was used during the first stage. All urethroplasties were performed using 6-zero monofilament absorbable sutures. Postoperatively a compressive dressing was left for 3 days and a 6Fr to 8Fr silicone catheter for 9 days.

The rate of complications requiring additional surgeries (reoperation rate) was compared among the 4 procedures. Complications were assessed by retrospective review of patient charts but all patients were also contacted during the study to ensure they did not require any additional surgeries elsewhere if more than 1 year had elapsed since the last followup visit. Complications were divided into healing complications (fistula formation and urethral breakdowns) and stricture/stenosis. Complication rates were compared among the 4 procedures and between patients who required UP transection and those who did not.

Parents of patients were also invited to participate in a survey about their perception of urinary symptoms and cosmetic outcomes. A nonvalidated questionnaire derived from recent questionnaires to assess voiding dysfunction was used to evaluate reported urinary symptoms.\(^20,21\) Parents were asked to answer yes or no to questions about the presence in their child of urinary tract infections and lower urinary tract symptoms, including hesitation, deflected stream, weak stream, straining to void, post-void dribbling and abnormal voiding frequency (more than 6 times daily or fewer than 3 times daily). The PPSS was used to assess cosmetic outcomes.\(^18\) An independent pediatric urologist administered the questionnaires via telephone.

Data were quoted as median and range (or IQR) and rate. Nonparametric tests were used throughout, including Mann-Whitney U test or Kruskal-Wallis test for nonpaired continuous values and chi-square test with trend or Fisher exact test for categorical variables. A p value of 0.05 or less was considered significant.

**RESULTS**

UP transection was not required for straightening in 57 of 93 patients (61%). Of these patients urethroplasty was performed using OIF in 31 and TIP in 26. UP transection was deemed necessary in the remaining 36 cases (39%), of which urethroplasty was performed using OIFA and a 2-stage technique in 18 cases each. A dorsal plication was performed for penile straightening in 18 of 26 TIP (70%), 23 of 31 OIF (74%), 14 of 18 OIFA (77%) and 18 of 18, 2-stage repairs (100%, p = 0.05).

Preoperatively meatal location was penoscrotal in 12 of 18, 2-stage (67%), 13 of 18 OIFA (72%), 23 of 31 OIF (74%) and 21 of 26 TIP repairs (81%), whereas meatal location was more proximal in the remainder (p = 0.8). Hormonal stimulation was used for the presence of a small penis in 10 of 18, 2-stage (56%), 10 of 18 OIFA (56%), 12 of 31 OIF (39%) and 9 of 26 TIP repairs (35%, p = 0.3).
Median postoperative followup was 6.9 years (range 3 to 8.4) for OIF, 4.2 (2.2 to 8.3) for TIP, 4.1 (2.5 to 8) for 2-stage and 2.4 (2 to 7.7) for OIFA repairs, being significantly less for the latter (p < 0.0001). Complications requiring additional surgeries developed in 21 of 93 cases (23%), including 5 of 31 OIF (16%), 4 of 18 OIFA (22%), 7 of 26 TIP (27%) and 5 of 18, 2-stage repairs (28%, p = 0.7). All complications developed within 12 months postoperatively. Complication rates were not different (p = 0.8) between cases that did (9 of 36, 25%) and did not (12 of 57, 21%) require UP transection.

Parents of 75 patients (80%) agreed to participate in the survey, including 25 of 31 (81%) undergoing OIF, 19 of 26 (73%) undergoing TIP, 14 of 18 (78%) undergoing OIFA and 17 of 18 (94%) undergoing 2-stage repairs (p = 0.35). A total of 75 patients reportedly suffered from urinary symptoms that included post-void dribbling (24, 32%), abnormal voiding frequency (19, 25%), deflected stream (12, 16%), hesitation (7, 9%), weak stream (7, 9%), urinary tract infection (6, 8%) and straining to void (2, 3%). Prevalence of urinary symptoms was not statistically different among the procedures. On the PPPS the only difference concerned the question about penile length, whereas overall PPPS had a trend toward significance (table 1). The score for the question on penile length was significantly different between techniques that did and did not require UP transection (table 2). Score for penile length was significantly better for 2-stage compared to all other repairs (table 3). It is noteworthy that the 2-stage repair also had a significantly better score for the penile axis item compared to TIP and a trend toward significance compared to OIF, despite the fact that dorsal plication was used in a significantly higher proportion of patients undergoing 2-stage repair. The 2-stage repair also had a significantly higher overall PPPS compared to all other techniques (table 3).

**DISCUSSION**

This study provides evidence that in patients with primary severe hypospadias UP transection does not increase the reoperation rate irrespective of use of a single stage repair. Prevalence of urinary symptoms is comparable among techniques. Procedures involving UP transection seem to allow for achievement of a longer penis, and the 2-stage repair yields the best cosmetic results.

The comparable reoperation rates among the 4 repairs suggest that overarching attempts at preserving UP continuity are unnecessary, at least to prevent surgical complications. Likewise, staging the repair after UP transection does not seem beneficial. In this respect we acknowledge that our patients undergoing OIFA repair had a significantly shorter followup, since this has been the latest technique incorporated in our armamentarium. There-

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**Table 1. PPPS according to surgical approach based on parent reporting**

<table>
<thead>
<tr>
<th>Procedure</th>
<th>Median OIF (IQR)</th>
<th>Median TIP (IQR)</th>
<th>Median OIFA (IQR)</th>
<th>Median 2-Stage (IQR)</th>
<th>p Value (Kruskal-Wallis test)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Penile length</td>
<td>1 (1–2)</td>
<td>2 (1–2)</td>
<td>2 (1–2)</td>
<td>2 (2–2.5)</td>
<td>0.03</td>
</tr>
<tr>
<td>Urethral meatus position + shape</td>
<td>2 (2–2)</td>
<td>2 (2–3)</td>
<td>2 (2–2.25)</td>
<td>2 (2–3)</td>
<td>0.2</td>
</tr>
<tr>
<td>Glans shape</td>
<td>2 (2–2.5)</td>
<td>2 (2–2)</td>
<td>2 (2–2.25)</td>
<td>2 (2–3)</td>
<td>0.4</td>
</tr>
<tr>
<td>Penile skin shape</td>
<td>2 (1–2)</td>
<td>2 (2–2)</td>
<td>2 (2–3)</td>
<td>2 (2–3)</td>
<td>0.3</td>
</tr>
<tr>
<td>Penile axis</td>
<td>2 (1–2)</td>
<td>2 (1–2)</td>
<td>2 (1–2.25)</td>
<td>2 (2–3)</td>
<td>0.2</td>
</tr>
<tr>
<td>General penile appearance</td>
<td>2 (2–2)</td>
<td>2 (2–2)</td>
<td>2 (2–2)</td>
<td>2 (2–3)</td>
<td>0.2</td>
</tr>
<tr>
<td>Overall PPPS</td>
<td>11 (10–12)</td>
<td>11 (11–12)</td>
<td>11 (10–12.75)</td>
<td>12 (11–16)</td>
<td>0.07</td>
</tr>
</tbody>
</table>

**Table 2. PPPS according to UP preserving or transecting procedure based on parent reporting**

<table>
<thead>
<tr>
<th>Procedure</th>
<th>UP Preserving</th>
<th>UP Transecting</th>
<th>p Value (Mann-Whitney U test)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Penile length</td>
<td>1.5 (1–2)</td>
<td>2 (1–2)</td>
<td>0.05</td>
</tr>
<tr>
<td>Urethral meatus position + shape</td>
<td>2 (2–2)</td>
<td>2 (2–3)</td>
<td>0.7</td>
</tr>
<tr>
<td>Glans shape</td>
<td>2 (2–2)</td>
<td>2 (2–3)</td>
<td>0.5</td>
</tr>
<tr>
<td>Penile skin shape</td>
<td>2 (2–2)</td>
<td>2 (2–3)</td>
<td>0.06</td>
</tr>
<tr>
<td>Penile axis</td>
<td>2 (1–2)</td>
<td>2 (1–3)</td>
<td>0.1</td>
</tr>
<tr>
<td>General penile appearance</td>
<td>2 (2–2)</td>
<td>2 (2–3)</td>
<td>0.2</td>
</tr>
<tr>
<td>Overall PPPS</td>
<td>11 (10–12)</td>
<td>12 (10–15)</td>
<td>0.1</td>
</tr>
</tbody>
</table>

**Table 3. PPPS for 2-stage vs other repairs based on parent reporting (Mann-Whitney U test p values)**

<table>
<thead>
<tr>
<th>Procedure</th>
<th>2-Stage vs OIF</th>
<th>2-Stage vs TIP</th>
<th>2-Stage vs OIFA</th>
</tr>
</thead>
<tbody>
<tr>
<td>Penile length</td>
<td>0.004</td>
<td>0.03</td>
<td>0.05</td>
</tr>
<tr>
<td>Meatus</td>
<td>0.1</td>
<td>0.5</td>
<td>0.07</td>
</tr>
<tr>
<td>Glans shape</td>
<td>0.5</td>
<td>0.1</td>
<td>0.3</td>
</tr>
<tr>
<td>Penile skin</td>
<td>0.1</td>
<td>0.1</td>
<td>0.9</td>
</tr>
<tr>
<td>Penile axis</td>
<td>0.09</td>
<td>0.04</td>
<td>0.2</td>
</tr>
<tr>
<td>General appearance</td>
<td>0.1</td>
<td>0.06</td>
<td>0.08</td>
</tr>
<tr>
<td>Overall PPPS</td>
<td>0.02</td>
<td>0.03</td>
<td>0.05</td>
</tr>
</tbody>
</table>
fore, additional complications might become evident with accumulating followup. Wood et al previously reported that 23% of the fistulas in their patients were detected more than 2 years postoperatively.\textsuperscript{22} However, the shortest followup in our patients was 24 months, and in contrast to that study all of our observed complications were detected in the first 12 months postoperatively.

If UP transection does not seem to increase the reoperation rate, it seems to allow for achievement of a longer penis according to parental perception. Moreover, the better score for the 2-stage repair on the question regarding penile axis compared to TIP or OIF repair might also suggest that attempts at preserving the UP increase the risk of recurrent curvature, as the UP might cause persistent ventral tethering.\textsuperscript{5,8} Consistent with this possibility, Braga et al observed a higher rate of recurrent curvature in patients undergoing penile straightening without UP transection compared to those in whom the UP was transected (37% vs 0%, p = 0.002).\textsuperscript{11} Unfortunately the followup was too short in our patients to evaluate curvature recurrence.

We failed to observe any differences in reported urinary symptoms among patients. This finding is not surprising, as the final anatomical result of any hypospadias repair is creation of a tube of skin in place of the deficient urethra. Such neourethras never have the biological properties of the native one and are likely to share similar urodynamic properties.\textsuperscript{1} In particular the lack of spongiosum around the urethra limits the elastic properties of the reconstructed segment.

It is noteworthy that the most common symptom in our patients was post-void dribbling. Lam et al also reported that most of their patients with hypospadias needed to milk the urethra to obtain complete emptying in the long term.\textsuperscript{25} TIP repair could be an exception in this respect, as the spongiosum can be reconstructed around the urethra.\textsuperscript{24} Nevertheless, it is still controversial as to whether this approach leads to a better functioning neourethra or only to a stiffer one.\textsuperscript{16,25} Overall our results confirm that urinary symptoms are rare after any repair, at least in the short and mid term.\textsuperscript{1}

In keeping with previous series,\textsuperscript{5–7} our results support the idea that 2-stage repair allows for better overall cosmetic results in patients with severe hypospadias. A major advantage of this approach is that it allows for a more accurate reconfiguration of the penis, which includes a more accurate redistribution of the shaft skin and recreation of the penoscrotal angle. This technique might contribute to an apparently more cylindrical penis and account for the higher score for the question about penile length, although the scores for the specific questions about shape of penile skin and general appearance of the penis were, in fact, not significantly different among the repairs. It is also noteworthy that scores for questions about meatal and glanular shape were not different among repairs. This finding is quite surprising, as creation of a slit-like meatus placed well within a conical glans is generally considered a distinct advantage of TIP and 2-stage repairs. In fact, we think this observation reflects the difference between parental and surgeon views in assessing the cosmetic characteristics of the penis.

Despite these considerations, the final role of 2-stage repair in cases requiring UP transection cannot be definitively determined based on our results. A thorough evaluation of the cost-to-benefit ratio should gauge the cosmetic benefit of this repair against the burden for the patient and family of a second procedure that can otherwise be avoided in about three-fourths of patients. Furthermore, it cannot be disregarded that additional procedures after the second stage were required in 28% of our cases and in 70% of cases reported by Shukla et al.\textsuperscript{12}

Limitations of our study include its retrospective nature, which impairs an accurate definition of the characteristics of hypospadias in study patients. Also there is a lack of consensus regarding classification to stratify cases at presentation, as the procedure necessary for penile straightening often can only be determined intraoperatively.\textsuperscript{1} In addition, given the rarity of the condition, the relatively small number of cases in each group might have underpowered the study to detect statistical significance, and the length of followup might be too short to detect some complications, such as recurrent curvature. Furthermore, we considered only complications requiring additional surgery, as this is a solid end point that can be assessed with certainty. Some complications might have been overlooked, managed conservatively or considered not to require additional surgery according to surgeon judgment or parent preference. We also relied only on parental report and did not test consistency in parent responses by test-retest, as we thought the latter might be distressing for the parents. One could question whether parents could be considered able to gauge clinical symptoms and assess cosmetic results, particularly taking into account that the questionnaire we used to assess urinary symptoms was not validated.

Nevertheless, we have always relied on parental perception in clinical practice, and particularly concerning hypospadias repair we lack criteria or diagnostic tools to define success objectively. Also potentially objective tests, such as uroflowmetry, are often abnormal in patients with hypospadias, so they are questionable as a reliable method to assess outcomes.\textsuperscript{26} Finally, we did not assess sexual function in our patients, as they were still too young. Never-
theless, to our knowledge this is the first study to compare surgical complications, urinary symptoms and cosmetic results in groups of patients with primary severe hypospadias undergoing different repairs with or without UP transection at a single institution with consistent perioperative management.

CONCLUSIONS
In our experience no repair proved to be ideal for the treatment of primary severe hypospadias. Overall complication rates were comparable among repairs. Urethral plate transection did not increase the complication rate. Urinary symptoms as reported by parents were quite rare and comparable among the procedures. Perceived penile length was significantly better in patients undergoing urethral plate transection, particularly those undergoing 2-stage repair. The latter approach also yielded the best cosmetic results overall. However, a 1-stage repair avoided additional procedures in about three-fourths of cases.

REFERENCES

EDITORIAL COMMENT
It is better to ask some of the questions than to know all the answers. One of the most important questions surrounding the repair of proximal hypospadias associated with severe ventral curvature is whether transecting the urethral plate results in improved penile length. The authors are to be commended for attempting to answer this question in a provocative study, where they mailed 2 questionnaires to parents of children who underwent 1 of 4 different hypospadias repair techniques. They concluded that urinary symptoms were quite rare and comparable among the 4 procedures. To have confi-
dence in their results, one must be assured that their questionnaires consistently measure what they are intended to measure when properly administered. In short, the questionnaires must be valid as well as reliable. In the present case face and construct validity may be lacking. The questionnaire responses were not checked against an external criterion or gold standard, such as bringing patients back after the procedure to watch them void or to check on the anatomical appearance of the repair.

The authors could also have tested for the construct validity of their questionnaire by administering it to parents of children with different severity of hypospadias. As evidence suggests that distal defects fare better in terms of outcomes than proximal ones, one would expect to see a difference in scores between distal and proximal hypospadias. Because construct validity was not checked, it becomes difficult to know whether the comparable scores seen among the 4 hypospadias procedures are truly a reflection of their outcome similarity or are simply due to the fact that the questionnaire cannot properly discriminate between the different techniques. These concerns underscore the ever present challenges in obtaining and interpreting survey data, particularly when the data are qualitative and subjective.

REPLY BY AUTHORS

The questionnaire we used to assess urinary function was not validated, and this was admittedly a limitation of our study. Although the questionnaire could not have properly discriminated between the different techniques, none of our patients required further evaluation or treatment because of urinary symptoms. Therefore, if differences were missed, they were subclinical. Moreover, the low prevalence of urinary symptoms we observed is fairly consistent with several previous series (reference 1 in article). Nevertheless, we fully agree that a validated, disease specific questionnaire to assess urinary function after hypospadias repair is much needed.

As far as validation is concerned, we question that construct validity of a disease specific questionnaire might be tested by administering it to parents of children with different severities of hypospadias. While distal hypospadias fares better than proximal variants in terms of complications and reoperation rates, urinary or ejaculatory function might be quite consistent. The glanular portion of the urethra is nevertheless the stiffest urethral segment and perhaps the one influencing urinary and ejaculatory function the most (reference 26 in article).

REFERENCES