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Intermediate-Term Followup of Proximal Hypospadias Repair Reveals High Complication Rate

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Abstract

Purpose—Results following distal hypospadias repair are favorable. Grouping proximal and distal hypospadias repair artificially increases the perceived success rate of proximal hypospadias. We identified our complication rate of proximal hypospadias repair and hypothesized a higher complication rate for 1-stage repair.

Materials and Methods—We retrospectively reviewed the records of consecutive boys who underwent proximal hypospadias from 2007 to 2014. Proximal hypospadias was defined as a urethral meatus location at or more proximal than the penoscrotal junction after penile degloving. We further stratified boys into those with planned 1-stage vs 2-stage repair. Univariate and Cox regression analyses were performed to assess associations with covariates and compare time to the first complication, respectively.

Results—A total of 167 boys met study inclusion criteria. Median followup was 31.7 months for 1-stage repair in 86 patients and staged repair in 81. The overall complication rate was 56%. Complications developed in 53 of 86 1-stage (62%) vs 40 of 81 staged (49%) repairs ($p = 0.11$). The number of unplanned procedures per patient was higher in the 1-stage than in the staged group (0.99 vs 0.69, $p = 0.06$), as was the number of patients who had at least 2 complications (29 of 86 or 33% vs 13 of 81 or 16%, $p = 0.03$). Cox regression showed no difference in time to the first complication for staged compared to 1-stage repair (HR 0.77, 95% CI 0.43–1.39).

Conclusions—Our 56% complication rate of proximal hypospadias warrants further long-term patient followup. More patients in the 1-stage group experienced at least 2 complications. However, when complications developed, they developed no differently in the 2 groups.

Keywords

urethra; hypospadias; male; reconstructive surgical procedures; postoperative complications

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Hypospadias occurs in 1 in 200 to 300 live births. Surgical correction is warranted due to concerns about cosmesis and function, specifically the ability to void while standing and the potential for sexual intercourse. Successful reconstructive surgery is defined by the ability to void with normal velocity and laminar flow, achievement of satisfactory sexual function with a straight penis and, from a cosmetic standpoint, achievement of a slit-like meatus in a well approximated glans.

Significant technical advances have allowed us to achieve a relatively high success rate in managing distal hypospadias, which is reported to be greater than 85%.¹⁻³ In the past, proximal hypospadias outcomes have been slightly worse with success rates ranging from 75% to 90%.⁴⁻⁷ Duration of followup is important to allow for the capture of complications because a significant number develop beyond the first year postoperatively.⁸⁻¹⁰

Recent publications from 3 large centers suggest a much higher complication rate for proximal hypospadias, ranging from 30% to 68%.¹¹⁻¹³ Surgical approach included 1-stage repair¹³ and planned 2-stage repair.¹¹⁻¹³ These high complication rates raise concern about our management of proximal hypospadias because boys are exposed to additional unplanned risk. A few of these boys are in danger of becoming a “hypospadias cripple,” from which recovery is difficult.

We hypothesized that the complication rate of proximal hypospadias is higher than previously reported and over application of 1-stage repair results in a higher complication rate compared to planned 2-stage repair.

MATERIALS AND METHODS

We retrospectively reviewed the records of boys who underwent primary proximal hypospadias repair with at least 6 months of followup at Children’s Hospital of Philadelphia from 2007 to 2014. Exclusion criteria included boys who did not undergo repair at our institution, those with a complicating diagnosis such as bladder exstrophy and those in whom the second stage portion of the procedure was not completed. Preoperative testosterone administration was left to the discretion of the operating surgeon and intramuscular injection was performed 5 to 6 weeks and again 2 to 3 weeks before surgery.

Proximal hypospadias was defined by the location of the urethral meatus after penile degloving in the operating room, specifically with a location at the penoscrotal junction or more proximal. Techniques of chordee repair included plication (Nesbit or dorsal midline), ventral “fairy” cuts, corporeal grafting (corporoplasty) with a tunica vaginalis or dermal graft and/or a combination of the techniques. The goal of surgical repair was achievement of a straight phallus with a urethral tube that would be effective at directing the urinary stream without spraying. From a cosmetic standpoint, we sought to achieve a slit-like meatus in the distal glans, although some patients were left with a coronal meatus, specifically select patients after complication repair. Followup was determined from the date of the first surgical intervention to the most recent office visit.

The use of staged vs 1-stage repair, the technique of chordee correction and the method of urethroplasty were left to the discretion of the operating surgeon. The degree of chordee was determined before and after penile degloving. Byars flaps were used for ventral penile shaft skin resurfacing in staged repair.

Complications were defined as any problem that occurred during the study inclusion period that required surgical correction. Complications included urethrocutaneous fistula, glans dehiscence, recurrent chordee, urethral diverticulum, meatal stenosis and skin concerns. Complications were determined by a combination of surgeon assessment and patient/parent reporting. We defined recurrent chordee as any degree of ventral curvature beyond 15 degrees after initial repair. Meatal stenosis was defined as meatal narrowing smaller than 6Fr and/or with evidence of stranguria, a slow urinary stream and/or elevated post-void residual volume. At the time of complication repair, cystoscopy and/or urethral calibration was performed to identify any concurrent complications.

Univariate analysis was done to determine associations between covariables and the type of repair (single vs staged). Multivariate Cox proportional hazards regression analysis was used to compare times to the development of the first complication as there were differences in followup. The proportionality of hazards assumption was satisfied by Schoenfeld residuals and inspection of log-log plots. Analyses were performed with Stata®, version 14.1, with $\alpha = 0.05$.

RESULTS

From 2007 to 2014 a total of 1,468 boys underwent hypospadias repair, of whom 167 met study inclusion criteria. One-stage repair was performed in 86 patients while planned staged repair was performed in 81 (table 1). Median age at initial surgery did not differ between the 2 groups (7.9 vs 9.2 months, $p = 0.13$). Median followup in the entire cohort was 31.7 months and it was slightly longer for 1-stage repair compared to planned staged repair (34.7 vs 27.7 months, $p = 0.09$, table 1). Median time between the stages of staged repair was 7.8 months.

Preoperative testosterone was more likely to be administered in the staged repair group (21 of 81 boys or 26% vs 5 of 86 or 6%, $p < 0.001$).

The overall complication rate was 56% since 93 of 167 boys experienced a complication after primary proximal hypospadias repair. The complication rate in the 1-stage and planned staged repair groups was 62% (53 of 86 cases) and 49% (40 of 81), respectively ($p = 0.11$). More patients in the 1-stage repair group presented with 2 or more complications compared to the staged repair group (29 of 86 or 33% vs 12 of 81 or 16%, $p = 0.03$). There was also a trend toward more unplanned procedures per patient for 1-stage vs planned staged repair (0.99 vs 0.69, $p = 0.06$, table 1).

Tables 2 and 3 show the techniques of chordee repair and urethroplasty, respectively. Plication was the most common approach used to repair chordee.

The total number of complications in the 1-stage and planned staged groups was 92 and 57, respectively (table 4). The most common complication for 1-stage and staged repairs was urethrocutaneous fistula (47% and 37%, respectively). The rates of recurrent chordee, urethral diverticulum, meatal stenosis and glans dehiscence did not differ between the 2 approaches (table 4). Skin complications, including suture sinuses, redundancy and skin bridges, occurred in 10 patients with 1-stage repair and in 6 with staged repair.

Several variables were analyzed by repair approach to determine risk factors for a shorter time to the first complication (table 5). Repair type, age at initial repair, year of initial repair and whether testosterone was administered preoperatively were not found to be associated with the interval to complication development.

DISCUSSION

The goals of penile reconstructive surgery for hypospadias are to allow the boy to void with normal velocity and laminar flow, to achieve satisfactory sexual function with a straight penis and to achieve a slit-like meatus with a well approximated glans.

Proximal hypospadias is challenging to all surgeons who perform penile reconstruction. Outcomes of distal hypospadias are favorable.¹⁻³ Unfortunately, extending expectations from our excellent distal hypospadias outcomes to proximal hypospadias repair has been more difficult.¹⁴ Proximal hypospadias repair has traditionally been thought to result in less successful outcomes but in satisfactory results in most patients.⁴⁻⁷ However, recent data indicate that the complication rates of proximal repair are higher than previously reported and contrast sharply with those of distal hypospadias repair.^{11-13,15,16} Of particular concern is the over extension of certain techniques, which may increase complication development. In patients with multiple complications, repair efforts focus on function, ensuring the correction of chordee and advancement of the meatus to a distal position that may not be located in the distal glans.^{14,17}

The explanation for increased complication rates center around the severity of the phenotype of proximal hypospadias. As the meatus becomes more proximal, anatomical factors such as a smaller glans, less penile shaft skin and more severe chordee increase the complexity of the repair. Smaller glans size has been associated with a higher complication rate.¹⁸ Chordee repair with plication is more likely to fail compared to corporeal grafting.¹⁹ At baseline the hypospadiac penis is anomalous. The corporeal bodies are shorter and the elasticity of the corpus spongiosum is abnormal compared to controls, adding further anatomical complexity to the repair.²⁰

Urethrocutaneous fistula was the most common complication in our patients (table 4). The rates of glans dehiscence, recurrent chordee, meatal stenosis and urethral diverticulum varied between the 2 groups but the differences did not achieve significance. We expected that the rate of recurrent chordee would be higher for 1-stage repair due to an underestimation of chordee severity or to overextension of the plication approach. Perhaps the lack of followup into puberty, when the increase in penile growth can exacerbate the degree of curvature, limited our ability to detect such a difference.^{19,21,22} Of the 9 patients in whom chordee

recurred 4 underwent skin mobilization, including 1 with skin mobilization and division of the urethral plate, and 4 with dorsal plication only. Curvature did not recur in any patients who underwent corporeal grafting. Three boys were ultimately left with a coronal meatus, which was done after reoperation for complications and necessitated by anatomical limitations. A coronal meatus leaves the patient at risk for splaying of the urinary stream and can potentially underestimate our rate of glans dehiscence.

Identification of this high complication rate warrants further investigation and risk identification to determine modifications of surgical technique to achieve satisfactory outcomes. There was a trend toward more unplanned procedures per patient in the 1-stage group. When complications occurred, patients who underwent 1-stage repair were significantly more likely to require 2 or more additional procedures to achieve satisfactory outcomes (table 1). Complications were less frequent in the staged repair approach and 84% of patients had no or 1 complication.

A limitation of our study is the lack of phenotype severity quantification, which limited comparison between groups. However, in general the patients with more severe chordee underwent the staged repair approach.²³

Our report correlates well with recent studies from 3 large centers of excellence for proximal hypospadias. Stanasel et al from Texas Children's Hospital presented their experience with 56 boys with a median followup of 34 months.¹¹ They identified a complication rate of 68% for the 2-stage repair approach.¹¹ McNamara et al from Boston Children's Hospital performed staged repair in 134 boys in a 20-year period.¹² At a median followup of 46 months the surgical complication rate was 49%. Pippi Salle et al from Toronto compared their results of 3 techniques in 140 boys during a mean followup of 30 to 48 months.¹³ Their approaches included a long tubularized incised plate, a dorsal inlay graft and staged repair for proximal hypospadias. The complication rate was highest for the long tubularized incised plate (53%) and lowest for staged repair (32%). Our outcomes parallel those in these recent reports and, given the consistency in outcomes among these institutions, raise concern about the current approach to surgical reconstruction.

Our approach to chordee repair evolved throughout the duration of this study. Early in the inclusion period, chordee correction trended toward dorsal plication or ventral fairy cuts. We have increasingly performed ventral corporoplasty with tunica vaginalis or dermal free graft, particularly when more than 15 degrees of curvature are present after degloving. Our approach now is to perform staged repair in all of these boys, separating procedures by 6 to 12 months to allow for sufficient graft healing prior to urethroplasty. Patients who underwent 1-stage repair but in whom chordee recurred required multiple additional procedures to resolve the chordee.

Use of an interposition island tube in the 1-stage group was significantly more likely to result in complication development compared to other techniques (12 of 14 patients or 86% vs 41 of 72 or 57%, $p = 0.04$). The reason for this is unclear but perhaps ischemia is more likely or the combined techniques of tubularization and anastomosis to the native urethra increase the risk.

We compared both groups to determine whether there was a difference in the interval to complication development (table 5), taking into account the differential followup. We hypothesized that if 1 approach compromised wound healing or increased tissue ischemia, this would present as a shorter interval to complication development. Type of repair had no impact on time to complication development, nor did age at repair or whether preoperative testosterone was administered. In patients in whom a complication developed requiring surgical correction, the overall median time from urethroplasty to the first operative complication was 9.7 months (IQR 2.9–21.1). For staged repair only the median time was 5.8 months (IQR 0.8–13.8) and for 1-stage repair only the time was 12.7 months (IQR 6.2–32.2). Interestingly, median followup in patients in whom a complication has yet to be identified was significantly shorter, raising concern that our complication rate could increase with the duration of followup.

Limitations of our study include its retrospective nature at a single institution and the fact that the choice of repair and administration of testosterone were left to the discretion of the operating surgeon. Some important risk factor data points were not captured, such as the number of closure layers and the use of running vs interrupted suture lines for urethroplasty, particularly early in our study inclusion period.

We did not subdivide proximal hypospadias meatal location into penoscrotal, scrotal and perineal locations. Therefore, we were unable to analyze whether precise location had an effect on outcomes. This was due to variability in our operative report documentation process throughout the study, particularly early in the inclusion period.

Patient followup was variable and left to the discretion of the surgeon, although this has since been standardized to include visits after potty training and after puberty. Lack of followup into puberty likely underestimated our failure rate. Patients were also perhaps more likely to be followed if a complication developed (table 2). However, it is not clear whether complications developed because followup was longer or followup was longer because complications developed. A total of 5 patients, including 1 with staged and 4 with 1-stage repair, have been followed beyond 3 years but have since moved out of the area and have been followed via telephone.

Lastly, we lack standardized measures of voiding function after repair. We did not measure uroflow and post-void residual volume in all patients. These studies were limited to patients in whom we suspected poor flow.

We have since standardized our documentation to facilitate objective comparison among patients. Capturing factors such as glans and urethral plate width, penile length and objective assessment of the degree of chordee will improve surgical decision making and risk stratification. This is done with scoring systems such as GMS (glans-urethral meatus-shaft) and the Penile Perception Score to determine whether we can predict the risk of complication development.^{24,25} Followup protocols have also been standardized and extended into puberty to improve the capture of complications.

CONCLUSIONS

Our results are similar to those in recently published reports of proximal hypospadias repair. Our 56% complication rate at an intermediate-term followup of 32 months warrants further long-term patient followup. Treatment of proximal hypospadias with 1-stage repair was more likely to result in more than 1 complication when a complication developed, which should be considered when deciding on a repair approach. Time to the development of complications did not differ by repair type. This higher complication rate must be integrated into the surgical management discussion with families.

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Univariate analysis of complications after single stage and planned staged proximal hypospadias repair

Table 1

	1 Stage	Planned Staged	Overall	p Value*
No. pts	86	81	167	-
Median mos age at first surgery (IQR)	7.9 (6.6-13.0)	9.2 (7.1-11.4)	8.7 (6.7-12.3)	0.13
No. preop testosterone (%):				
No	81 (94)	60 (74)	141 (84)	<0.001
Yes	5 (6)	21 (26)	26 (16)	
Median mos 1st surgery followup (IQR)	34.7 (12.6-71.4)	27.7 (13.8-43.6)	31.7 (13.0-57.9)	0.09
No. pts with complications (%):				
No	33 (38)	41 (51)	74 (44)	0.11
Yes	53 (62)	40 (49)	93 (56)	
No. complications/pt (%):				
0	33 (38)	41 (51)	74 (44)	0.03
1	24 (28)	27 (33)	51 (31)	
2+	29 (33)	13 (16)	42 (25)	
No. additional unplanned procedures/pt:				0.06
Mean ± SD	0.99 ± 0.99	0.69 ± 0.75		
Median (IQR)	1 (0-2)	1 (0-1)		

* Wilcoxon rank sum test for nonparametric continuous variables and Pearson chi-square or Fisher exact test for categorical variables.

Table 2
One-stage and planned staged chordee repair in 167 patients with or without complications

Chordee Repair	No. pts (%)	1 Stage (86 pts)		Planned Staged (81 pts)		Total No. (%)
		Complication	No Complication	Complication	No Complication	
Skin mobilization only	17 (32)	16 (48)	0	0	0	33 (20)
Skin mobilization + urethral plate division	6 (11)	2 (6)	10 (25)	9 (22)	0	27 (16)
Plication, no urethral plate division	23 (43)	15 (45)	0	0	0	38 (23)
Plication + urethral plate division	3 (6)	0	20 (50)	20 (49)	0	43 (26)
Corporoplasty + urethral plate division	0	0	6 (15)	8 (20)	0	14 (8)
Plication, + corporoplasty + urethral plate	0	0	4 (10)	4 (10)	0	8 (5)
Plication + fairy cuts	4 (8)	0	0	0	0	4 (2)
p Value (Fisher exact test)		0.22		0.97		
Median followup (mos)		56.7	12.6	37.0	15.0	31.7

For all approaches skin mobilization and chordee tissue division were performed.

Table 3

Type of urethroplasty used for hypospadias repair in patients with or without complications

Complication	No. Yes (%)	No. No (%)
1-Stage urethroplasty:	53	33
Thiersch-Duplay	2 (4)	5 (15)
Onlay island flap	15 (28)	13 (39)
Interposition island tube	12 (23)	2 (6)
Long tubularized incised plate	19 (36)	10 (30)
Interposition island tube + long tubularized incised plate	2 (4)	2 (6)
Interposition island tube + onlay island flap	1 (2)	0 (0)
Long tubularized incised plate + onlay island flap	2 (4)	1 (3)
p Value (Fisher exact test)	0.16	
Planned staged urethroplasty:	40	41
Thiersch-Duplay	39 (98)	39 (95)
Onlay island flap	1 (2)	2 (5)
p Value (Fisher exact test)	1.00	

Table 4

Univariate analysis of total complications after 1-stage and planned staged proximal hypospadias repair

	1-Stage	Planned Staged	p Value*
No. complications	92	57	–
No. pts/total No. (%):	53/86	40/81	
Urethrocutaneous fistula	40/86 (47)	30/81 (37)	0.22
Persistent chordee	6/86 (7)	3/81 (4)	0.50
Urethral diverticulum	8/86 (9)	5/81 (6)	0.57
Meatal stenosis	13/86 (15)	5/81 (6)	0.08
Glans dehiscence	4/86 (5)	6/81 (7)	0.53

Multiple complications developed in some patients.

* Pearson chi-square or Fisher exact test.

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Table 5

Multivariate Cox proportional hazards model of time to first complication after initial repair of proximal hypospadias

	HR (95% CI)	p Value
Repair:		
1 Stage	Referent	
Staged	0.77 (0.48–1.25)	0.29
Age at initial repair	1.00 (0.97–1.02)	0.86
Initial repair yr	1.01 (0.94–1.09)	0.73
Preop testosterone:		
No	Referent	
Yes	1.56 (0.83–2.92)	0.17

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