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# Post-hypospadias urethrocutaneous fistulae: no difference in repair success between proximal and distal fistulae

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**Introduction:** Urethrocutaneous fistulae are the most common complication after hypospadias repair. We sought to compare outcomes of proximal versus distal urethrocutaneous fistula repair and hypothesized that patients with proximal fistulae would have lower rates of success than those with distal fistulae. We also aimed to evaluate factors that affected these outcomes.

**Material and methods:** Current procedural terminology codes were used to identify patients undergoing urethrocutaneous fistula repair after hypospadias surgery between 2014 and 2017 at an academic, pediatric urology referral center. Characteristics for each initial hypospadias repair and each fistula repair were noted, including location of meatus, location of fistula, type of magnification, suture type, interposition layer and post-

operative stenting. The primary outcome was successful fistula repair. Univariate and multivariate analysis was performed.

**Results:** During the study period, 416 hypospadias repairs were performed. Thirty-one of these later presented with a fistula (8% fistula rate). Sixty-eight percent of fistulae were successfully closed with a single repair. There were 17 distal fistulae and 14 proximal fistulae. There was no difference in success between distal (71%) and proximal (64%) fistulae ( $p = 0.73$ ). There was no statistically significant association between the primary outcome (successful fistula repair) and fistula location ( $p = 0.71$ ), magnification ( $p = 0.38$ ), suture type ( $p = 0.49$ ), interposition coverage layer (0.43), or postoperative stenting ( $p = 0.92$ ) on univariate or multivariate analysis.

**Conclusion:** There is no difference in success when repairing distal versus proximal urethrocutaneous fistulae. Neither fistula location, type of magnification, suture type, interposition layer nor stenting affected outcomes.

**Key Words:** pediatrics, hypospadias, fistula

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## Introduction

Urethrocutaneous fistulae are the most common complication after hypospadias repair.<sup>1</sup> While there is considerable work on hypospadias repair and techniques to prevent fistulae, and other complications, the literature on management of fistulae and factors

related to successful repair is less robust. Our institution has previously demonstrated an approximately 70% success rate with each fistula repair procedure and that recurrence of a fistula does not affect the outcome of subsequent fistula repair.<sup>2</sup> This data did not specifically compare proximal and distal fistulae. Other work has shown that meatal location, however, does affect risk of fistula formation. Proximal hypospadias has nearly a two-fold risk of complication.<sup>3</sup> From this, it would follow that proximal fistula repairs have a lower success rate, but this has not been well described. We hypothesized that patients with proximal fistulae after hypospadias repair would have lower rates of successful fistula repair than those with distal fistulae.

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In this study, we sought to investigate the outcomes of fistula repair in patients who underwent distal and proximal hypospadias repairs. We also aimed to evaluate patient and surgical factors that affect outcomes in fistula repair and compare this to the current literature.<sup>4</sup>

## Materials and methods

### Design and study population

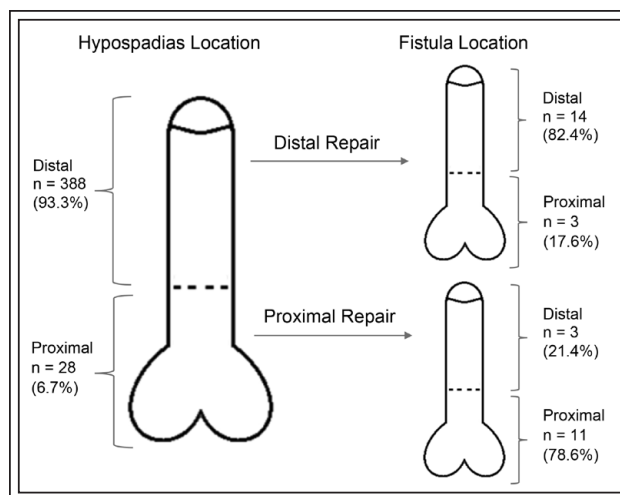
We sought to compare patients undergoing urethrocuteaneous fistula repair using a single institution, retrospective review. Current procedural terminology (CPT) codes were used to identify patients undergoing fistula repair (54340, 54344) after hypospadias surgery between 2014 and 2017 at our tertiary care pediatric urology referral center. CPT codes for all hypospadias repairs (54322, 54324, 54326, 54326, 54328, 54332, 54336) were used to identify the total number of hypospadias repairs during the same period of the initial hypospadias surgeries (2013 to 2015). This study was reviewed by the institutional review board and granted exempt status.

### Variables

Characteristics for each initial hypospadias repair and each fistula repair were noted, including location of meatus, location of fistula, type of magnification (operating microscope or loupes), suture type (polyglactin or polydioxanone), postoperative stenting, and whether an interposition layer was used. Here, we define a distal urethrocuteaneous fistula as one occurring from the glans to the midshaft and a proximal urethrocuteaneous fistula from the proximal shaft to the perineum. The outcome was successful fistula repair, defined as voiding only through the neomeatus at follow up, with either pediatric urology or the primary care physician. Time to fistula diagnosis was based on the clinic visit at which the fistula was first reported or identified.

### Statistical analysis

Using our own historical data for fistula risk (5% after distal hypospadias repair and 50% after proximal hypospadias repair) with a power of 80% and a p value of 0.05 with a 1:1 enrollment, we calculated each arm would need 14 subjects, for a total of 28 cases. As data on risk of recurrent fistulae comes from very small series, this was used as a more robust proxy for determining sample size. Descriptive statistics are expressed as count and percent. Univariate analysis was performed using a chi-square test for categorical variables. Multivariate analysis was performed using logistic regression. Statistical significance was considered as  $p < 0.05$ . SPSS Version 25 (IBM, Armonk, NY, USA) was used for data analysis.

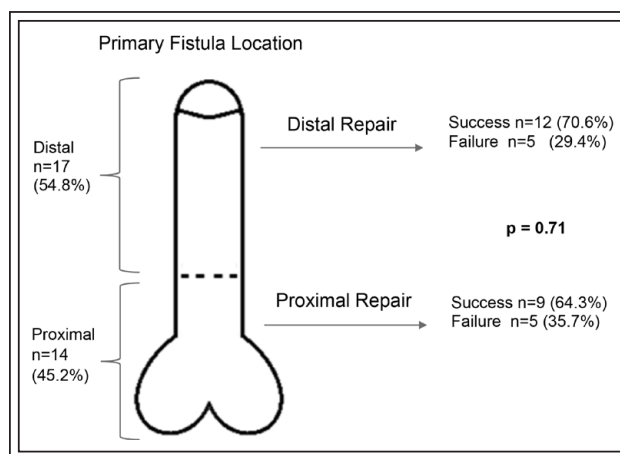


**Figure 1.** Initial hypospadias and subsequent urethrocuteaneous fistula location.

## Results

From October 2013 to August 2015 a total of 416 hypospadias repairs were performed, 31 of these later presented with a fistula, for a 7.5% overall rate of fistula. There were 17 distal fistulae (54.8%) and 14 proximal fistulae (45.2%). In the majority, fistulae were near the location of the initial hypospadiac meatus; however, three distal hypospadias presented with a more proximal fistula and three proximal hypospadias presented with distal fistulae, Figure 1. All procedures were performed by five pediatric urologists. Surgical technique was at the discretion of the surgeon, Table 1.

The median age at initial hypospadias repair was 7 months old [IQR 6.5, 11.5]. The median time to diagnosis of fistula was 7 months after initial hypospadias repair



**Figure 2.** Success by urethrocuteaneous fistula location.

TABLE 1. Descriptive data of initial hypospadias repair

	Type of repair	Magnification	Suture	Interposition layer	Stented
Total n = 31	TIP <sup>1</sup> or GAP <sup>2</sup> 28 (90.3%)	Microscope 20 (64.5%)	Polyglactin 22 (71.0%)	Yes 8 (25.8%)	Yes 16 (51.6%)
	TPIF <sup>3</sup> 3 (9.7%)	Loupes 11 (35.5%)	Polydioxanone 8 (29.0%)	No 23 (74.2%)	No 15 (48.4%)
Distal hypospadias n = 17	TIP <sup>1</sup> 15 (88.2%)	Microscope 13 (76.5%)	Polyglactin 13 (76.5%)	Yes 1 (5.9%)	Yes 4 (23.5%)
	GAP <sup>2</sup> 2 (11.8%)	Loupes 4 (23.5%)	Polydioxanone 4 (23.5%)	No 16 (94.1%)	No 13 (76.5%)
Proximal hypospadias n = 14	TIP <sup>1</sup> 11 (78.6%)	Microscope 7 (50.0%)	Polyglactin 9 (64.3%)	Yes 7 (50%)	Yes 12 (85.7%)
	TPIF <sup>2</sup> 3 (21.4%)	Loupes 7 (50.0%)	Polydioxanone 5 (35.7%)	No 7 (50%)	No 2 (14.3%)

<sup>1</sup>tubularized incised plate; <sup>2</sup>glans approximate procedure  
<sup>3</sup>transverse preputial island flap

[IQR 2, 11.5] and the median time to fistula repair from initial hypospadias repair was 9 months [IQR 6, 15.5]). The median follow up with pediatric urology from initial hypospadias repair was 20.5 months [10.25, 30.75] and the median time to follow up with pediatric urology after fistula repair was 3 months [IQR 0, 12.25]; however the median follow up with primary care from fistula repair was 32 months [IQR 23.5, 39.5]). Ten (32.2%) patients had no documented follow up.

There was 67.7% of fistulae successfully closed with a single repair. There was no difference in success between distal (70.6%) and proximal (64.3%) fistulae ( $p = 0.73$ ; Figure 2). There was no statistically significant association between the primary outcome (successful fistula repair) and fistula location, magnification, suture type, interposition coverage layer, or postoperative stenting on univariate or multivariate analysis (all  $p > 0.05$ , Table 2). Proximal fistulae were more likely

TABLE 2. Association between surgical and patient characteristics for successful fistula repairs

	Unadjusted Odds ratio (95% CI)	p value	Adjusted Odds ratio (95% CI)	p value
Location of fistula				
Proximal	0.75 (0.17-3.40)	0.71	0.94 (0.17-5.04)	0.94
Distal	Reference		Reference	
Microscopic magnification				
Yes	0.50 (0.11-2.31)	0.38	0.50 (0.04-6.09)	0.58
No	Reference		Reference	
Suture type				
Polyglactin	0.57 (0.12-2.85)	0.49	0.37 (0.02-9.01)	0.54
Polydioxanone	Reference		Reference	
Interposition layer				
Yes	2.38 (0.28-19.92)	0.43	7.26 (0.37-143.71)	0.192
No	Reference		Reference	
Stented				
Yes	0.92 (0.20-4.31)	0.92	0.68 (0.12-3.99)	0.67
No	Reference		Reference	

to have had an interposition layer (71.4% versus 29.4%,  $p = 0.02$ ) than distal fistulae. Proximal fistulae with a flap ( $n = 5$ , 71.4%) recurred less often than those without an interposition layer ( $n = 2$ , 28.6%) but this was not statistically significant ( $p = 0.58$ ).

## Discussion

Urethrocutaneous fistulae are the most common complication after hypospadias repair, but the literature on outcomes of these fistulae repair are less robust. Based on our data, proximal and distal urethrocutaneous fistulae after hypospadias repair have comparable rates of successful closure. Distal fistulae had a 70.6% rate of successful repair after the first attempt and proximal fistulae had a 63.4% rate of success ( $p = 0.71$ ). This was in contrast to our hypothesis, based on data from initial hypospadias complications, that proximal fistula repair would have lower rates of success. This is, however, consistent with prior data from our institution showing overall success of fistula repair at 70% with each operation.<sup>2</sup> On univariate analysis, there was no difference in success rates with use of surgical loupes versus microscope for magnification, suture type, placement of urethral stent or use of interposition layer (microscopic magnification was also not shown to improve outcomes in our prior study).

Notably, proximal fistula repairs were more likely to utilize an interposition coverage layer than distal fistula repair (71.4% proximal, 29.4% distal  $p = 0.02$ ). Use of an interposition layer has previously been shown to decrease fistula formation after initial hypospadias repair.<sup>5,6</sup> Considering this data, it is likely surgeons are using an interposition layer when they are more concerned about fistula recurrence. This may have contributed to comparable success rates. In distal fistulae there may also be less tissue readily available for an interposition flap, especially after a prior hypospadias repair. Furthermore, the increased operative time and risk of penile torsion, chordee and devascularization of the skin must be balanced with the risk of repair failure. These same considerations made at the time of initial surgery lead many surgeons to eschew interposition flaps for initial repair of distal hypospadias.

Interestingly, three fistulae occurred proximal to the original location of the meatus. While it is possible that once the penis is degloved the meatus is in fact more proximal than initially thought, we suspect that in these cases, the ventral urethra proximal to the meatus was very thin. In such instances, the decision must be made whether to cut back to thick, healthy urethra, or leave the native tissue and use flap coverage

to buttress the area. This thin proximal urethra may serve as another factor in fistula formation.

Meatal, or distal, stenosis is another consideration when treating fistulae. All patients are catheterized with an 8 Fr at the time of repair. Only one patient had meatal stenosis at the time of fistula repair. He had an initial distal hypospadias and a distal fistula. His initial repair was a tubularized incised plate repair, and his fistula did not recur after repair. No patients have thus far had issues with meatal stenosis after repair of their fistulae.

## Limitations

Although this study was powered to detect significant differences in rates of fistula formation, it was nonetheless limited by the small sample size and retrospective nature of data. Perhaps with a larger cohort, smaller differences would be able to be detected. As with all retrospective reviews, we are limited by the data available and are subject to confounding. Surgeons may have altered technique, suture, choice to stent based on some factor we are unable to ascertain and as such affected outcomes. Another limitation of this study is the high rate of patients without documented follow up. Success was defined as no evidence of recurrent fistula on exam or no presentation to Pediatric Urology or Primary Care with a symptomatic fistula. As the only pediatric urology group in a large geographic area, over 90% of all patients with hypospadias are seen at our institution. Because of this, we feel confident that patients with complications would return or be referred back to us; however, it is possible that some patients followed up with an adult urologist or transferred care to another region. As other series have shown the median time to fistula presentation in primary hypospadias repair to be 8.5 months (range 1 month to 13.9 years), our own follow up may not be adequate.<sup>7</sup> To account for this, we reviewed not only Pediatric Urology follow up but also Primary Care follow up notes and were thus able to extend follow up reporting on the premise that these patients were still in the system and would be referred back if issues arose.

## Conclusion

Based on our data, there is no significant difference in success when repairing distal versus proximal urethrocutaneous fistulae. This is in contrast to higher success rates in initial repair of distal versus proximal hypospadias. While various surgical variables including type of magnification, suture type and stenting did not significantly affect outcomes, interposition layers were more likely to be utilized in proximal than distal fistula repairs. □

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