

# Risk factors for fistula recurrence after urethrocutaneous fistulectomy in children with hypospadias

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## ABSTRACT

**Objective:** This study aimed to investigate the risk factors of fistula recurrence after primary urethrocutaneous fistulectomy in children with hypospadias.

**Material and methods:** The study included 63 children who underwent fistulectomy for urethrocutaneous fistula (UCFs) that occurred after urethroplasty for hypospadias, between February 2009 and December 2018. The patients were divided into 2 groups: successful group 1 and failed group 2. For data analysis, we included the demographics of the patients, the details of the previous urethroplasty (the type of hypospadias and the location of the meatus after complete chordection), the presence of meatal stenosis or urethral stricture after urethroplasty, and the size of the UCFs. The Student t-test and the chi-square test were performed to analyze the data using the Statistical Package for Social Sciences software.

**Results:** The overall success rate of primary urethrocutaneous fistulectomy was 81.0% (51/63 children). The most common location of a secondary fistula was the penoscrotal area 5 (41.6%). There were no statistically significant differences in age ( $p=0.501$ ), weight ( $p=0.063$ ), body mass index ( $p=0.924$ ), history of low birth weight ( $p=0.454$ ), and history of prematurity ( $p=0.381$ ). The type of hypospadias ( $p=0.007$ ) and urethral defect length ( $p=0.021$ ) were identified as independent risk factors for failed urethrocutaneous fistulectomy. There were no statistically significant differences in meatal stenosis ( $p=0.431$ ), postoperative stricture ( $p=0.587$ ), fistula location ( $p=0.173$ ), multiplicity ( $p=0.588$ ), and fistula size ( $p=0.530$ ).

**Conclusion:** The type of hypospadias and the length of the urethral defect are the significant risk factors for secondary fistula recurrence after primary urethrocutaneous fistulectomy.

**Keywords:** Child; fistula; hypospadias; risk factors; treatment outcomes.

## Introduction

Hypospadias is a common birth defect observed in male children. Previous studies have reported wide geographical variations in prevalence rates; the global prevalence of hypospadias for all years was around 20.9 per 10,000 births.<sup>[1]</sup> Several authors have reported an increased incidence in recent decades.<sup>[2-4]</sup>

Unfortunately, hypospadias repair is associated with a high complication rate. Complications such as urinary tract infections or wound dehiscence that develop within a month postoperatively in patients undergoing hypospadias repair tend to cause secondary complications, including urethrocutaneous fistulas (UCFs) and urethral stenosis.<sup>[5]</sup>

UCF is the most common complication in this category of patients. A previous study reported complications in 35 (11.4%) of the 307 patients enrolled, with fistula formation in 19 patients (6.2%).<sup>[6]</sup> Tension at the anastomotic suture lines, overlapping suture lines, inadequate approximation of anatomical layers, incorrect dressing, and nonfunctioning urethral catheter with extravasation predispose patients to develop postoperative fistula.<sup>[7]</sup>

At present, several techniques can be performed to repair UCF. However, repairing recurrent UCF after a previous fistula surgery is challenging and stressful for pediatric urologists and pediatric patients. A previous study reported recurrent fistula in 25% of patients who underwent a primary repair of UCF.<sup>[8]</sup>

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Some authors suggest the use of de-epithelialized skin flaps (vest-over-pant technique) to prevent fistula recurrence<sup>[9]</sup> because a simple fistula closure carries a higher risk of recurrence even in small-sized fistulas.<sup>[10]</sup> Researchers have reported that flap closure is safe for fistula repair.<sup>[11]</sup>

Several studies have investigated the risk factors of primary fistula formation after hypospadias repair; however, only a few reports describing recurrent fistula are available.<sup>[12,13]</sup> We wanted to know if the factors associated with recurrent fistula were different from those causing the primary fistula after a hypospadias repair. In this study, we investigated the factors associated with recurrent fistula after primary urethrocuteaneous fistulectomy.

## Material and methods

The study included 63 children who underwent fistulectomy for UCF, which occurred after urethroplasty for hypospadias, between February 2009 and December 2018. Repair of hypospadias and UCF were performed by a single surgeon in the pediatric urology department. The patients were divided into 2 groups: successful group 1 and failed group 2. Data analysis was performed using the demographics of the patients, the details of the previous urethroplasty, the location of the primary meatus and the type of hypospadias, the presence of meatal stenosis or urethral stricture after urethroplasty, and the size and location of the UCF.

For fistulas  $\leq 2.0$  mm in size, a simple fistulectomy technique was performed. The distal urethral obstruction was excluded by bypassing the urethral probe. To prevent missing small UCFs, indigo carmine blue solution was injected into the meatus, whereas a tourniquet was applied around the penile root to occlude the urethra. The procedure was started by making an incision around the UCF, and the tract was released from the surrounding tissues and the tunica dartos and then ligated using 7-0 polydioxanone sutures (PDS). The tunica dartos, as the second layer, was used to cover the UCF tract with a continuous suture using 6-0 PDS sutures. The skin was closed with 6-0 PDS sutures.

The rotational skin flap closing technique was used for patients with fistulas measuring  $>2.0$  mm. A circumferential incision

### Main Points:

- The severity and the condition of the urethral plate of hypospadias might be important in the recurrence of fistulas.
- Recurrence of fistula occurred in those who had long urethral defects during the initial surgery.
- Age at surgery, history of prematurity, size of glans, type of surgical repair performed, and other factors were not significantly associated with urethrocuteaneous fistula recurrence.

was made around the UCF, and the urethral wall was dissected from the penile skin. The urethral opening was closed with 7-0 PDS sutures. The dartos fascia, as the second layer, was used to cover the UCF tract with a continuous suture using 6-0 PDS. A longitudinal-proximal incision was made around the UCF to make a rotational skin flap to cover the suture line. Depending on the condition of the fistula, redo urethroplasty was performed as a fistulectomy.

### Statistical analysis

The chi-square test and the Student *t*-test were performed to analyze the data using Statistical Package for Social Sciences Statistics version 22.0 (IBM Co., Armonk, NY, USA). A *p*-value  $<0.05$  was considered statistically significant.

### Ethics committee approval

This study was approved by the ethics committee of the Pusan National University Yangsan Hospital Clinical Research Ethics Committee (05-2019-164).

## Results

The mean age of the patients in groups 1 and 2 were 82.138.5 months and 86.832.0 months, respectively. The other demographic characteristics, including body mass index (*p*=0.924), history of low birth weight (*p*=0.454), and prematurity (*p*=0.381), are shown in Table 1. Tubularized incised plate urethroplasty was performed in 28 patients (54.9%) and 8 patients (66.3%) in group 1 and group 2, respectively. Four (7.8%) patients in group 1 underwent onlay island flap urethroplasty. Mathieu hypospadias repair was performed in 8 patients (15.6%) and 1 patient (8.3%) in group 1 and group 2, respectively. Thiersch-Duplay urethroplasty was performed in 4 patients (7.8%) in group 1. A combined procedure was performed in 7 patients (13.7%) and 3 patients (25.0 %) in group 1 and group 2, respectively.

The overall success rate of primary urethrocuteaneous fistulectomy was 81.0% (51/63 children). No statistically significant differences between the 2 groups were observed in the following demographic characteristics: age (*p*=0.501), weight (*p*=0.063), body mass index (*p*=0.924), history of low birth weight (*p*=0.454), and history of prematurity (*p*=0.381). Recurrence of UCF was reported in 12 patients (19.0%) in group 2. All patients in group 2 initially had a penoscrotal hypospadias; the urethral defect length was 30.012.8 mm. By performing the Student *t*-test, the type of hypospadias (*p*=0.007) and the urethral defect length (*p*=0.021) were identified as independent risk factors for failed urethrocuteaneous fistulectomy (Table 2). By performing the chi-square test, it was determined that the location of the fistula (*p*=0.173), the type of UCF (*p*=0.588), and the size of the fistula (*p*=0.530) were not significant. Postoperative stricture was observed in 3 (5.8%) patients in group 1. In group

**Table 1. Demographic characteristics of patients**

Parameters	Success group (n=51)	Failed group (n=12)	p
Age (months), mean±SD	82.1±38.5	86.8±32.0	0.501 <sup>†</sup>
Height (cm), mean±SD	109.3±23.2	120.5±20.3	0.597 <sup>†</sup>
Weight (kg), mean±SD	20.0±12.0	25.2±13.1	0.064 <sup>†</sup>
History of low birth weight, n (%)	30 (58.8)	5(41.6)	0.454*
BMI (kg/m <sup>2</sup> ), mean±SD	16.4±6.3	16.3±3.6	0.924 <sup>†</sup>
History of prematurity, n (%)	20(39.2)	3(25.0)	0.381*

BMI: body mass index; SD: standard deviation. \*Chi-square. <sup>†</sup>Student t-test.

**Table 2. Characteristics of urethroplasty**

Parameters	Success group (n=51), n (%)	Failed group (n=12), n (%)	p
Type of hypospadias			0.007*
Distal penile	22 (43.1)	1 (8.3)	
Mid shaft	5 (9.8)	1 (8.3)	
Proximal penile	9 (17.6)	0 (0)	
Penoscrotal	11 (21.5)	8 (66.6)	
Perineal	2 (3.9)	2 (16.6)	
Unknown	2 (3.9)	0 (0)	
Size of glans (>14 mm)	16 (31.3)	4 (33.3)	0.682 <sup>†</sup>
Location of the meatus after chordectomy			0.135*
Distal penile	11 (21.5)	0 (0)	
Mid shaft	5 (9.8)	0 (0)	
Proximal	10 (19.6)	4 (33.3)	
Penoscrotal	12 (23.5)	4 (16.6)	
Perineal	2 (3.9)	1 (8.3)	
Unknown	11 (21.5)	3 (25)	
Urethral defect lengths (mm)	20.0±15.8	30.0±12.8	0.021 <sup>†</sup>
Type of previous urethroplasty			0.098*
Mathieu	8 (15.6)	1 (8.3)	
Onlay Island urethroplasty	4 (7.8)	0 (0)	
TIP	28 (54.9)	8 (66.3)	
Thiersch-Duplay	4 (7.8)	0 (0)	
Combined	7 (13.7)	3 (25.0)	

TIP: tubularized incised plate. \*Student t-test. <sup>†</sup>Chi-square.

2, there were no patients with urethral stricture. Furthermore, meatal stenosis was found in 31 (60.7%) patients in group 1; in group 2, a mild form of meatal stenosis, with a small caliber of the urethral meatus, which resolved with dilation, was seen in

**Table 3. Characteristics of fistulectomy**

Parameters	Success group (n=51), n (%)	Failed group (n=12), n (%)	p
Location of the fistula, n (%)			0.173*
Coronal sulcus	18 (35.2)	3 (25.0)	
Distal penile	16 (31.3)	1 (8.3)	
Mid shaft	7 (13.7)	3 (25.0)	
Penoscrotal	10 (19.6)	5 (41.6)	
Type of UCF, n (%)			1.000*
Simple	44 (86.2)	11 (92.6)	
Multiple	7 (13.7)	1 (8.3)	
Size of fistula (mm), mean±SD	3.8±2.9	4.7±3.3	0.530 <sup>†</sup>
Type of closing, n (%)			0.327*
Simple	17 (33.3)	2 (16.6)	
Skin flap	26 (50.9)	7 (58.3)	
Unknown	8 (15.6)	3 (25.0)	
Postoperative stricture, n (%)	3 (5.8)	0 (0)	0.587*
Postoperative meatal stenosis, n (%)	31 (60.7)	5 (41.5)	0.431*

SD: standard deviation; UCF: urethrocutaneous fistula. \*Chi-square test. <sup>†</sup>Student t-test.

41% of patients. The results of either group were not significant (Table 3). All patients who had recurrence of fistula underwent urethrocutaneous fistulectomy with successful outcomes.

## Discussion

UCF after hypospadias repair is still challenging for pediatric urologists.<sup>[14]</sup> Recent studies have reported the risk factors of UCF after hypospadias repair; feasible approaches have been suggested to create decision-making algorithms and various management strategies for different types of UCF.<sup>[8]</sup> Unfortunately, no specific technique has been deemed suitable or effective for all patients.<sup>[15]</sup> The experience of the surgeon, the surgical technique used, the interposition of a barrier layer between the urethroplasty and the skin, and the postoperative urinary drainage are the most important factors to prevent fistula formation.<sup>[16]</sup>

A previous study reported that the type of the hypospadias and the urethral tract length were not associated with UCF formation.<sup>[17]</sup> However, a different study reported that 39 (32.5%) of 120 patients developed UCF after hypospadias repair; urethral defect length and previous surgery were the main risk factors identified.<sup>[18]</sup> Bush et al.<sup>[19]</sup> reported that small-sized glans, defined as glans with width <14 mm, were an independent risk factor for primary UCF formation. Several methods have been re-

ported for UCF repair; however, no single technique can ensure successful outcomes in all types of UCF.<sup>[15]</sup> The site of the fistula and the condition of the surrounding skin are important factors in planning the surgical repair technique.<sup>[8]</sup> However, there is a high recurrence rate after a simple fistula closure, following hypospadias repair. Secondary closure was successful in 50% of patients with recurrent fistula.<sup>[20]</sup>

Ensuring multilayer coverage using well-vascularized tissue, performing tension-free closure, avoiding suture overlap, and correcting meatal stenosis are essential for a successful urethrocutaneous fistulectomy.<sup>[21-23]</sup> Successful results after hypospadias repair can be attributed to healthy tissue and rich vascular supply and may be associated with the patient's age and history of prior surgery.<sup>[24]</sup> Simple closure is easier for smaller fistulas (<2 mm), whereas local skin flap closure is performed in larger fistulas (>2 mm), with good vascularization of the surrounding skin for safety and prevention of recurrence.<sup>[11]</sup> In our study, for fistulas ≤2.0 mm, a simple fistulectomy technique was performed. Distal urethral obstruction was excluded by bypassing the urethral probe. The rotational skin flap closing technique was performed for fistulas >2.0 mm.

Traditional transposition and advancement flaps may be unreliable after primary fistula repair because the vascularity of the area around the fistula orifice may be compromised by previous surgery.<sup>[25]</sup> Several authors have reported that layered closure of coronal sulcus fistulas is challenging because the distal glandular tissue is fixed, which causes tension on the suture line, contributing to high failure rates associated with coronal sulcus fistula repair.<sup>[26]</sup> Notably, distal urethral obstruction increases the risk of recurrent UCF, although it may also affect the development of primary UCF.<sup>[27-29]</sup>

Unfortunately, only a few studies have reported secondary fistula rates. One study reported recurrent fistula in 15 (22.3%) of 67 patients.<sup>[30]</sup> The results of these studies showed a secondary fistula rate of 25% after primary fistula repair. The author observed that the method of fistula repair (rotational skin flap) and the use of suprapubic urinary diversion were associated with a lower risk of fistula recurrence.<sup>[8]</sup> In our study, although the rotational skin flap technique was performed, there was a higher recurrence of fistula in group 2 patients, who had longer urethral defects during the initial surgery.

In this study, we investigated the risk factors for secondary fistula formation in 63 children who developed primary UCF after hypospadias repair. Age at surgery, history of prematurity, size of glans, location of the meatus, meatal stenosis, location of the fistula, type of surgical repair performed, and other factors were not significantly associated with UCF recurrence. Our study found that the type of hypospadias and the urethral defect

length were independent risk factors for primary UCF formation; they might also be the main causes of secondary fistula formation. In our opinion, the severity and the condition of the urethral plate of hypospadias might be important contributors to the recurrence of fistulas.

### Study limitations

This study had some limitations. First, the number of cases were few. Second, there was a lack of important clinical information related to previous surgical procedures, actual complications, and patient satisfaction after primary repair. This limitation was mainly related to the difficulty in finding information from the patients' medical records. Moreover, fistula has many different characteristics; therefore, simplification was very difficult. Third, during our study, uroflowmetry could not be performed to exclude distal urethral obstruction. In future prospective studies, the abovementioned limitations will be taken into consideration.

In conclusion, the type of hypospadias and urethral defect length at the time of primary urethroplasty are significant risk factors for secondary fistula recurrence after primary urethrocutaneous fistulectomy.

**Ethics Committee Approval:** Ethics committee approval was received for this study from the ethics committee of Pusan National University Yangsan Hospital Clinical Research Ethics Committee (05-2019-164).

**Informed Consent:** Written informed consent was obtained from patients' parents who participated in this study.

**Peer-review:** Externally peer-reviewed.

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**Conflict of Interest:** The authors have no conflicts of interest to declare.

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