Urethro-cutaneous Fistula After Hypospadia Repair: A Single Institution Study

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Abstract

Urethro-cutaneous fistula (UCF) is one of the most frequently seen complications of hypospadias surgery requiring re-operation; it occurs with an incidence of between 4% and 28%. Risk factors associated with the development of UCF can be classified as preoperative, intraoperative or postoperative. The aim of this study was to determine the association of peri-operative risk factors and the development of urethrocutaneous fistula after hypospadias repair. A retrospective review of patients who had undergone hypospadias repair at Kenyatta National Hospital between 2013 and 2017 was conducted. 114 patient records were retrieved. The incidence of UCF was 47%. Risk factors that were significantly associated with UCF are hypospadias type (p=0.028), lack of a protective intermediate layer (p=0.002), and presence of postoperative complications (p=0.001). Age at surgery, suture material, type of repair and use of catheter/stents were not significant factors. Multivariate analysis showed wound infection and meatal stenosis as the most significant factors associated with UCF development.

Key words: Hypospadias, Urethro-cutaneous fistula, Risk factors, Wound infection, Meatal stenosis

Ann Afr Surg. 2019; 16(2):59–63

DOI:http://dx.doi.org/10.4314/aas.v16i2.4

Conflicts of Interest: None

Funding: None

Introduction

Urethro-cutaneous fistula (UCF) is one of the most frequently seen complications of hypospadias surgery (1).Incidence is variable but is commonly reported between 4% and 28% (2). In the Kenyan setting, fistula development remains a significant problem to the paediatric urologist and, coupled with the nature of its recurrence, these fistulae are associated with frequent redo surgeries, increased hospital costs and poor functional and anatomic outcome of the urethra and penile shaft(3). The factors that predispose to fistula development have been proposed to arise from an interplay of pre-operative, intraoperative and postoperative factors (4, 5).While some authors have identified individual factors such as the type of hypospadias and age at surgery as the most significant risk factors, contrary reports state that the surgical technique is the most significant contributor (1, 6). Variability of the surgical technique employed globally makes the association with fistula occurrence challenging.

Nevertheless, breaking down the surgical technique into measurable components allows for statistical analysis. Such components include type of repair, tissue handling, use of vascularized tissue layers, and type and size of suture material (1, 2, 7).Fistula formation is multifactorial and the risk factors for fistula development vary. The management approach to UCF requires knowledge of the factors that contribute to their formation. The purpose of this study was to determine the association of peri-operative risk factors with the development of urethro-cutaneous fistula after hypospadias repair.

Methods

This study was a retrospective review of patients who had undergone hypospadias repair at Kenyatta National Hospital (KNH), a tertiary teaching hospital in Nairobi, Kenya. Peri-operative data were collected from medical records of patients between 2013 and 2017. All records of patients who had undergone the initial hypospadias repair in KNH were included in the study. Excluding criteria were redo surgery, missing files and incomplete records. Preoperative variables included age at surgery and hypospadias type. Intraoperative variables included the type of repair, use of protective intermediate layer, suture material used, and use of stents/catheters. Data collected on postoperative complications related to fistula formation included meatal stenosis, urethral stricture and wound infection. These complications were recorded from the documented clinical record and voiding cysto-urethrogram reports. All surgeries were done by competent pediatric urologists with more than 15 years of experience in hypospadias repair. Measures to reduce infection included a perineal wash on the morning of surgery, observing aseptic technique with antibiotic prophylaxis, and site preparation with iodine solution before skin incision. Postoperatively, antibiotics were administered and the dressing

was kept for 3 days.

Data were analysed using SPSS (V.21.0 Chicago-Illinois). Univariate and multivariate analyses were used. The Chi-square test, Fisher's exact test and logistic regression were used to ascertain association among clinical variables. P-values, odds ratio (OR) and 95% confidence intervals (CI) were calculated where applicable. A p-value of less than 0.05 was considered statistically significant. Institutional approval to conduct the study was sought and granted.

Results

A total of 174 records of patients who had undergone hypospadias surgery were recorded. The retrieval process was electronic and could only retrieve 148 files; 26 files were missing. Only complete records were considered; 114 out of the 148 files were complete. Figure 1 shows the recruitment flow chart. Figure 2 shows the incidence grouped according to location.

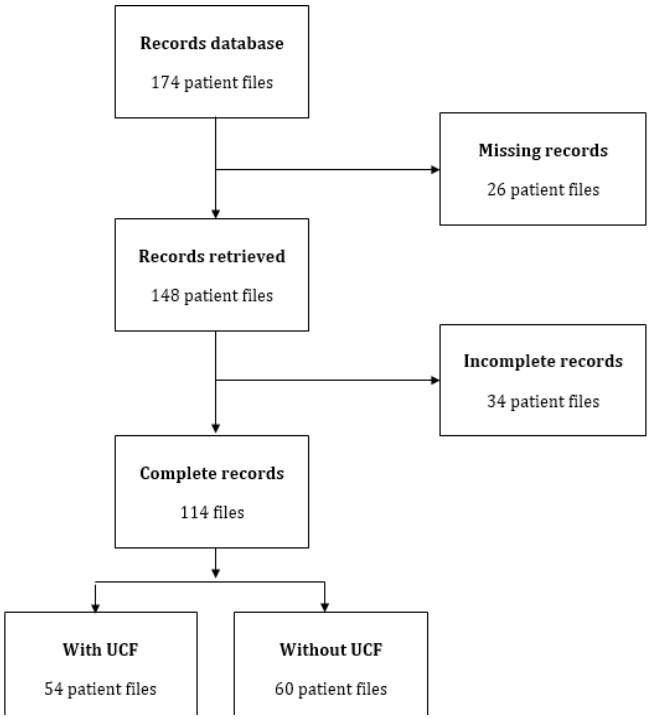

Figure 1. Records recruitment flow chart

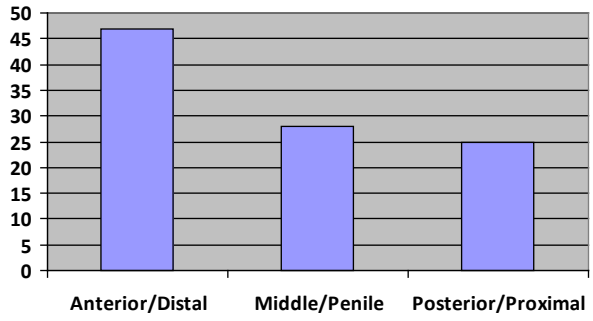

Figure 2. Incidence of UCF in proportion to significant risk factors over the 5-year period

The median age at surgery was 2 years with a range between 8 months and 16 years. The overall incidence of UCF was 47%. Table 1 summarizes the characteristics of UCF.

Table 1. Characteristics of urethro-cutaneous fistulae (UCF)

Characteristic	Frequency *n*=54	%
Time to UCF development		
<1 month	38	70.3
1–3 months	5	9.2
>3 months	11	20.3
Number of UCF		
Single	48	88.9
Multiple	6	11.1
Location of UCF		
Subcoronal	35	64.8
Penile	12	22.2
Penoscrotal	7	12.9

Table 2 shows the association between risk factors and fistulae occurrence. Type of hypospadias (p=0.028), the use of a protective intermediate layer (p=0.002) and the presence of postoperative complications (p=<0.001) were statistically significant risk factors for UCF development. Age at surgery, suture material, and type of repair and use of catheter/stents were not significant factors.

The factors that were significant in univariate analysis were entered on to a logistic regression model for multivariate analysis (Table 3). This confirmed that the presence of wound infection (OR 66.26; 95% CI 10.41–421.62) and meatal stenosis (OR 54.20; 95% CI 4.78–615.72) were the most significant risk factors for UCF development.

Trends in UCF incidence between 2013 and 2017 showed direct proportionality to the rates of wound infection and meatal stenosis, evidenced by the reduction in incidence of UCF as the rates of wound infection and meatal stenosis declined (Fig. 3).

Discussion

The goal of hypospadias surgery is to correct the penile curvature, form a neo-urethra of adequate size, bring the new meatus to the tip of the glans, if possible, and achieve an overall acceptable cosmetic appearance (8). The occurrence of uretho-cutaneous fistula therefore prevents the achievement of this goal.

The mechanism of UCF development lies in the incorporation of urethral mucosa or neo-urethra in ventral repair with rapid migration of urethral mucosa and skin epithelium into suture tracts, usually due to infection, ischemia or both (9). The pooled incidence of UCF in our study was 47%, which is high compared with other reports in the literature (2, 4).

Table 2. Univariate analysis on risk factors for urethro-cutaneous fistula formation

Risk factor / Variable	UCF *n*=54	No UCF *n*=60
Age at surgery (p=0.229)		
< 2 years	21	16
> 2 years	33	44
Type of hypospadias(p=0.028)		
Distal		
glandular	1	10
sub-coronal	17	25
Mid		
distal penile	7	5
mid penile	5	7
proximal penile	5	3
Proximal penoscrotal	19	10
Procedure (p=0.540)		
One stage		
MAGPI	1	16
TIP	27	27
Thiersh-Duplay	9	7
Ducket Island flap	7	1
Mathieu based flap	4	0
Two stage		
Buccal mucosa flap	4	1
Preputial skin flap	2	8
Protective intermediate layer (p=0.002)		
Intermediate layer	30	40
No intermediate layer	23	4
Suture material (p=0.121)		
Monofilament	19	34
Braided	16	14
Not indicated	32	
Suture size (p=0.073)		
4.0	0	6
5.0	9	8
6.0	26	33
Not indicated	32	
Use of stent or catheter (p=0.817)		
Stent	22	28
Catheter	30	30
None	2	2
Complications (p=<0.001)		
Meatal stenosis	12	1
Neo-urethral stricture	6	0
Wound infection	29	2
None	7	57

UCF–urethro-cutaneous fistula; MAGPI–meatal advancement and glanuloplasty incorporated; TIP–tubularized incised plate

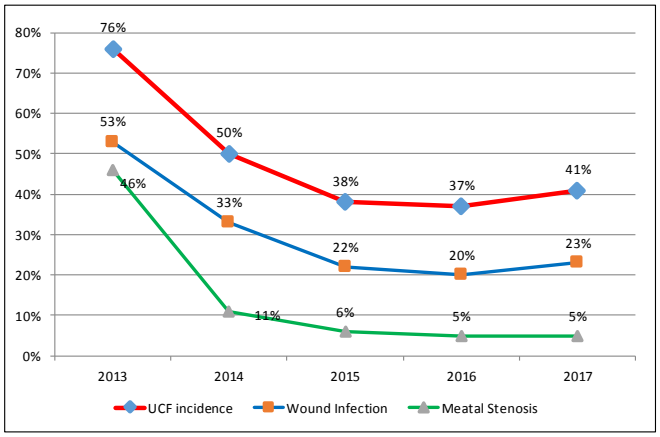

Figure 3. Trends in UCF incidence between 2013 and 2017

Most fistulae are reported to occur within the first month of surgery (2,4); similarly, we found 70.3% of UCF occurred within 30 days from time of repair. Fistulae may occur anywhere along the neo-urethra; however, common sites include the site of the original meatus and at the subcorona, which represents a region of poor vascularization and healing (2). The same principle is the basis of the higher incidence of recurrence of subcoronal fistulae. In our study 64.8% of fistulae were located at the subcorona.

Hypospadias type, lack of a protective intermediate layer and presence of postoperative factors were significantly associated with UCF formation, which compares well with the findings of Chung, who demonstrated more fistula formation with proximal hypospadias (6). Hadidi et al. also specify that UCF rates may be minimized by the use of protective vascularized intermediate layers and observation to technique, which reduce ischemia and infection (1). Other factors such as age at surgery, use of stents, suture material and type of repair have been reported in the literature as contributing to UCF (1, 4, 5, 10, 11, 12); however, these factors were not found to be statistically significant in this study. In multivariate analysis, the presence of wound infection and meatal stenosis were the only significant factors in UCF formation.

Local infection has previously been studied as a cause of UCF (5, 13). This may be as a result of the inflammatory process associated with infection, which leads to urethral mucosal migration to suture tracts (9, 14). This study revealed that wound infection is a major contributor to fistula formation. Infection sets in within the first 2 weeks after surgery, and it contributes significantly to the high number of fistulae occurring within the first month of surgery.

A decreasing trend in local infection rates is shown between 2013 and 2017, which we attribute to several factors. First, we avoid sitz baths, which we noted had increased rates of infection. This increased rate of infection has been shown to be real in gynecologic and anorectal disorders, but the evidence is still lacking in urological surgery (15). In addition, our experience of using an antibiotic ointment after the saline

Table 3. Multivariate analysis on the risk factors for urethro-cutaneous fistula formation

Risk factor / Variable	UCF (*n*=54)	No UCF (*n*= 60)	OR (95% CI)
Type of hypospadias (p=0.749)			
Distal	18 (33.3)	35 (58.3)	2.96 (0.16–54.38)
Mid	17 (31.4)	15 (25.0)	1.91 (0.13– 28.95)
Proximal	19 (35.2)	10 (16.6)	–
Type of repair (p=0.922)			
One stage	48 (88.9)	51 (85.0)	1.17 (0.05– 26.58)
Two stage	6 (11.1)	9 (15.0)	–
Intermediate layer(p=0.143)			
Layer	30(56.5)	40 (90.9)	0.2 (0.05–1.57)
None	23 (43.4)	4 (9.1)	–
Complication (p=0.001)			
Meatal stenosis	12 (22.2)	1 (1.7)	54.20 (4.78– 615.72)
Wound infection	29 (53.7)	2 (3.3)	66.26 (10.41– 421.62)
None	7 (13.0)	57 (95.0)	–

washes, usually from the third day of surgery, contributes a dual effect of reducing infection and moisturizing the site. Second, we have also set guidelines on aseptic technique aimed at reducing infection rates, which include a perineal wash with soap and water, site preparation with iodine and prophylactic antibiotics (16). Postoperatively, our patients are nursed under a cradle with sterile drapings, and any form of dressing change is under sterile conditions. These measures have been strengthened in the 5-year period, hence the decrease in infection rate which in this study was directly proportional to the incidence of fistula formation.

Distal obstruction of urine flow via the neo-urethra is a risk factor for fistula formation, usually as a result of meatal stenosis, meatal encrustation, edema and urethral stricture (3). Snodgrass attributes metal stenosis to an overzealous repair of the neo-urethra distally causing obstruction that increases tension on the repair, which leads to fistula formation (17). This is consistent with our findings in which meatal stenosis was found to be a significant contributor to fistula formation. A decreasing trend in the rate of meatal stenosis is seen in our study, which we attribute to the training workshops on hypospadias surgery that we conduct annually in collaboration with international societies of pediatric urology. The objective of the workshops is to build capacity of the residents and surgeons by developing skills and competence in pediatric urology. Specifically, the workshops have emphasized avoiding a tight closure at the distal urethra by marsupializing the ventral urethral edge to the glans and this has significantly reduced the rate of meatal stenosis. Since inception of these workshops, we have noted an overall improvement in outcome of hypospadias repair.

We acknowledge that data were collected from patient records that may have inaccurate data especially regarding surgical technique and intraoperative detail. Any errors of documentation during initial recording of these details contribute to our study limitations. Additionally, there was significant loss rate upon recruitment due to missing patient files and incomplete records. Finally, other significant factors in literature that contribute to UCF include lack of use of magnification, rough tissue handling, presence of UTI and dressing type (1, 9). These factors could not be tested in this study design due to its retrospective nature, also limiting our study.

Conclusion

We identified wound infection and meatal stenosis as the most significant risk factors for uretho-cutaneous fistula development in our set-up. Measures to reduce the rate of wound infection after hypospadias surgery and surgical technique aimed at reducing the rate of meatal stenosis will ultimately reduce the occurrence of urethro-cutaneous fistula.

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