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ORIGINAL RESEARCH

Urethrocutaneous fistula following hypospadias repair: recurrence rate and its determinants

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Background: Urethrocutaneous fistula (UCF) is a common complication following hypospadias repair. It may be associated with frequent recurrences after repair, ultimately increasing treatment costs and leading to poor cosmetic outcomes due to multiple surgeries.

Aim: This study aimed to evaluate the recurrence rate of post-hypospadias UCF and its determinants following repair among patients treated at the Urology Department of Kilimanjaro Christian Medical Centre (KCMC) from January 2011 to December 2023.

Methodology: We conducted a hospital-based, retrospective study that reviewed the case notes of all patients who developed UCF post-hypospadias repair. Patients were followed up for at least six months postoperatively. Various factors were analysed to assess their influence on recurrence.

Results: The study included 44 patients with UCF after hypospadias repair. The recurrence rates after repair were 30% (13/44) after the first, 30% (4/13) after the second, 50% (2/4) after the third, and none after the fourth repair. Our results demonstrated a significant association between UCF recurrence and the hypospadias grade and UCF location. Multivariate analysis using a modified Poisson regression model identified a history of grade III or IV hypospadias as an independent risk factor for UCF recurrence.

Conclusion: UCF post-hypospadias repair is associated with at least one-third of recurrence following repair. A history of grade III or IV hypospadias is an independent risk factor for this recurrence. Age at first UCF repair, catheter/stent duration, and suture material were not related to UCF recurrence after surgical repair.

Keywords: hypospadias, urethrocutaneous fistula, recurrence

Introduction

Urethrocutaneous fistula (UCF) is the abnormal connection between the urethral mucosa and the skin, such that the urine stream passes partially or entirely through that opening.[1] This abnormal opening is usually located on the underside of the penis or perineum. UCFs can result from circumcision, congenital, a complication of transurethral procedures, perineal infection, genital trauma, or malignancy.[2-5]

UCF is a common complication of hypospadias repair, with a 4–47% global incidence.[6,7] In northern Tanzania, UCF following hypospadias repair accounts for up to 40.5% of all complications of hypospadias surgery.[8] The fistula can occur immediately after removal of the urethral catheter/stent or later, and can be single, two, or multiple, with variable sizes.[7,9-11] The diagnosis is clinical, where assessment of the fistula is more accurate when performed intraoperatively. While the size is determined with a calliper, the number of UCFs is assessed by retrograde injection of methylene blue solution into the urethra through the external urethral meatus.[10,12]

UCF management can be conservative or surgical. A fistula that appears acutely and small following hypospadias repair can be managed conservatively by replacing the catheter for at least 14 days to allow it to close spontaneously.[13-15] Surgical repair options depend on the number, duration, location, fistula size, and the nature of the surrounding tissues.[16]

The UCF post-hypospadias repair should be at least six months old before the repair is done to allow the scar to mature, and also for oedema and induration to subside completely.[17-19] In the practical operational workflow, it is essential to assess the presence of meatal stenosis/distal urethral strictures during the procedure. If these conditions are identified, appropriate corrective measures should be performed to minimise the risk of fistula recurrence.[20]

Surgical repair techniques for UCFs vary from simple closure to multilayer with waterproof flap repair.[16] There are a variety of tissue flaps used to close the fistula, including penile dartos, scrotal dartos, and tunica vaginalis, which is reserved for multiple complex fistulae.[21]

UCFs tend to recur after repair, with an overall recurrence rate ranging from 0% to 28%.[12,22,23] Some fistulae may require up to 15 operations for successful closure.[24] Due to frequent recurrences, UCFs are associated with prolonged hospital stays, increased treatment costs, and imposed psychological stress due to multiple corrective surgeries.[12,18] Anatomical and technical factors, such as location, size, fistula closure technique, and distal obstruction, determine UCF recurrence after repair. Additional factors include patient age and the previous repair type.[24,25]

Despite UCFs being the most common long-term complication of hypospadias repair in Tanzania, data on its recurrence rate and contributing factors remain scarce.[8] This study aimed to provide urologists with updated insights to guide the development of targeted strategies to reduce UCF recurrence and improve surgical outcomes.

Methodology

Study site

This retrospective study involved reviewing the medical records of all patients who underwent UCF repair following hypospadias repair from January 2011 to December 2023. The study was conducted at Kilimanjaro Christian Medical Centre (KCMC), a zonal referral hospital located in Kilimanjaro, northern Tanzania. The hospital serves seven regions of the Northern Zone, with over 15 million people.[26]

As a consultant hospital in the Northern Zone, KCMC has a well-established urology institute that serves patients referred from regions within and outside its catchment area. The institute serves as a referral, training, and research centre for all urological conditions. It performs an average of one hypospadias repair each month and one UCF repair every two months. All surgeries were performed by a urologist or resident supervised by a urologist.

Surgical techniques

As per our local protocol, all patients diagnosed with UCF were evaluated to rule out distal urethral obstruction before surgical repair. All UCF repair surgeries were performed by both specialists and residents under supervision, and all operators had a comparable level of competence. Therefore, we considered them equivalent for analysis.

The procedure involved excision of the fistula tract at the level of its connection to the urethra, followed by tension-free closure using absorbable sutures (Vicryl, Monocryl, or Polydioxanone with sizes 4-0, 5-0, or 6-0). A local dartos flap was used to reinforce the repair site and prevent leakage, with the skin subsequently closed over this second layer. None of the patients had concomitant complications, such as distal stricture or urethral diverticula. All patients were catheterised with a urethral feeding tube, serving as a stent for a period of 7–14 days.

Following discharge, patients were routinely followed up in the outpatient clinic at one, three, and six months postoperatively, and then annually. Follow-up primarily focused on monitoring for urethral-related complications, including UCF recurrence, urethral diverticula, and urethral stricture. UCF recurrence was the primary focus of this study.

Outcome and explanatory variables

The main study outcome was defined as UCF recurrence after surgical repair. The explanatory variables were patient age at first UCF repair, using five years as a cut-off, the hypospadias grade, defined as the location of the external urethral meatus as glanular (grade I), distal (grade II), proximal (grade III), and perineal (grade IV), surgical technique used for hypospadias repair, UCF location, surgical technique used for UCF repair, suture material and size used during UCF repair, and stent/catheter duration.[27-31]

Data collection, process, and analysis

The registration numbers of all patients who underwent UCF repair during the study period were obtained from the theatre register books of the KCMC Urology Department. Thereafter, patient information was retrieved from physical files obtained from the department's medical records. The retrieved information was collected using a structured data extraction sheet. The study included all patients with UCF due to hypospadias repair during the study period. Also, patients were considered eligible if they completed a minimum postoperative follow-up of at least six months.[19,32] The study excluded all patients with other causes of UCF formation, such as circumcision, urethral stricture, and epispadias.

Data were entered into the Statistical Package for the Social Sciences (SPSS) version 25, cleaned, analysed, and summarised. Descriptive statistics were used to summarise data, whereby measures of central tendency and their respective dispersions were used to summarise continuous variables. Proportions and percentages were used to summarise categorical variables. To assess the determinants associated with UCF recurrence, binary logistic regression was used for multivariate analysis to estimate crude and adjusted relative risk (RR) with 95% confidence intervals (CI). A p-value < 0.05 was considered statistically significant.

Ethics

The College Research Ethics Review Committee of the Kilimanjaro Christian Medical University College approved the study (number PG 98/2023). No patient identifiers were used; instead, patients' identification numbers were used, and all information was kept confidential.

Results

During the study period, a total of 113 patients underwent UCF repair. However, 43 patient files (38%) could not be retrieved and were excluded from the analysis. Of the remaining 70 patients (62%) whose files were retrievable, 26 (36.2%) were excluded, as their UCFs resulted from other causes (circumcision, urethral stricture, urethral trauma, epispadias repair). The remaining 44 patients (62.8%), with a repaired, isolated UCF post-hypospadias surgery and followed up for at least six months, were included in the analysis.

Participant characteristics and factors associated with UCF recurrence

The patients' median age at first fistula surgery was four years, with the majority (29, 65.9%) being under five years. Most UCFs (27, 61.4%) were located at the mid/proximal penile, and 13 patients (30%) needed more than one repair to close the fistula tract. Most cases (34, 77.3%) underwent UCF repair more than six months after the initial hypospadias repair, with a median time of 12 months. Multilayer repair using the penile dartos and tunica vaginalis flaps (TVF) was the common UCF closure technique in 38 cases (86.4%). A urethral catheter/stent was used in all patients, with a median duration of 12 days (range 7–25) (Table I).

Among the 44 patients who underwent UCF repair, recurrence occurred in 30% after the first attempt. Subsequent repairs showed recurrence rates of 30% for the second repair, 50% for the third repair, and no recurrence after the fourth repair. The recurrence rate was more frequent in younger children (≤ 5 years, 11/29, 37.9%),

Table I: Participant characteristics and factors associated with UCF recurrence

Variable	UCF recurrence			
	Total, *n* (%)	Yes, *n* (%)	No, *n* (%)	*p*-value†
Age (years)				0.162
≤ 5	29 (65.9)	11 (37.9)	18 (62.1)	
> 5	15 (34.1)	2 (13.3)	13 (86.7)	
Hypospadias grade, *n* = 37				0.012
I–II	21 (56.8)	3 (14.3)	18 (85.7)	
III–IV	16 (43.2)	9 (56.2)	7 (43.8)	
UCF location				0.044
Distal penile	17 (38.6)	8 (47.1)	9 (52.9)	
Mid/proximal penile	27 (61.4)	5 (18.5)	22 (81.5)	
Hypospadias repair technique				0.155
Other techniques*	31 (70.45)	7 (22.6)	24 (77.4)	
Two-stage repairs	13 (29.55)	6 (46.2)	7 (53.8)	
Time from hypospadias repair to UCF repair (months)				0.905
≤ 6	10 (22.7)	3 (30)	7 (70)	
> 6	34 (77.3)	10 (29.4)	24 (70.6)	
Duration of urethral catheter/stent (days)				0.459
≤ 7	12 (27.3)	2 (16.7)	10 (83.3)	
> 7	32 (72.7)	11 (34.4)	21 (65.6)	
First UCF repair technique				0.157
Multilayer repair	38 (86.4)	13 (34.2)	25 (65.8)	
Other techniques**	6 (13.6)	0	6 (100)	
Suture type				0.461
Monofilament	11 (25)	2 (18.2)	9 (81.8)	
Braided	33 (75)	11 (33.3)	22 (66.7)	
Suture size				0.198
< 6–0	27 (61.4)	10 (37.0)	17 (63.0)	
6–0	17 (38.6)	3 (17.7)	14 (82.3)	

† A Fisher's exact test was used to measure the level of significance, where $p < 0.005$ is significant.
* Other techniques for hypospadias repair included tabularised incised plate (*n* = 26), Duckett repair (*n* = 2), and Mathieu repair (*n* = 3).
** Other techniques for UCF repair included simple repair (*n* = 2) and granular fistula converted to grade I hypospadias and repaired (*n* = 4).
UCF – urethrocutaneous fistula

grade III–IV hypospadias (9/16, 56.2%), longer catheter duration (> 7 days, 11/32, 34.3%), multilayer fistula repair (13/38, 34.2%), and braided suture use (11/33, 33.3%). The hypospadias grade ($p = 0.012$) and fistula location ($p = 0.044$) showed significant differences in UCF recurrence between the groups. Conversely, factors such as age, hypospadias repair technique, catheter duration, and suture type or size did not show significant differences in UCF recurrence between groups ($p > 0.05$) (Table I).

Determinants of UCF recurrence after repair

Multivariate analysis using a modified Poisson regression model revealed that distal penile fistulae and grade III–IV hypospadias were significantly associated with an increased risk of UCF recurrence. Patients with distal fistulae had a 2.96-fold higher risk of recurrence (RR 2.96, 95% CI 1.06 to 8.27; $p = 0.038$), while those with grade III–IV hypospadias had a 3.93-fold higher risk (RR 3.93, 95% CI 1.25 to 12.42; $p = 0.019$), compared with their respective counterparts.

Other variables, including age at repair, prior hypospadias repair technique, the time interval between hypospadias and UCF repair, suture material, and suture size, were not significantly associated with recurrence ($p > 0.05$). After adjusting for all variables in the model, a history of grade III-IV hypospadias was identified as an independent risk factor for UCF recurrence (adjusted RR 2.38, 95% CI 1.74 to 5.94; $p = 0.028$) (Table II).

Discussion

UCF management after hypospadias repair remains a significant problem, with studies indicating variable recurrence rates, ranging from 0% to 28%, influenced by different factors.[12,22,23] This study, being the first of its kind in Tanzania, reported an overall recurrence rate of 30%, with 13/44 patients experiencing recurrence after the first repair and, subsequently, 4/13 having recurrence after the second repair. UCF recurrence was found to be determined by anatomical and technical factors, such as the hypospadias grade, UCF location, suture technique, and surgical technique of UCF repair. Our findings align with a study conducted in the United States, which documented recurrence rates of 28% after the first repair and

Table II: Multivariate analysis on determinants of UCF recurrence after repair

Variable	Crude		Adjusted	
	RR (95% CI)	*p*-value	RR (95% CI)	*p*-value
Age (years)				
≤ 5	1		1	
> 5	0.35 (0.08 to 1.41)	0.140	0.39 (0.13 to 1.23)	0.11
UCF location				
Mid/proximal penile	1		1	
Distal penile	2.96 (1.06 to 8.27)	0.038	2.10 (0.74 to 5.94)	0.16
Hypospadias grade				
I–II	1		1	
III–IV	3.93 (1.25 to 12.42)	0.019	2.38 (1.74 to 5.94)	0.028
Hypospadias repair technique				
Other techniques*	1			
Two-stage repairs	2.04 (0.84 to 4.96)	0.114	1.16 (0.32 to 4.31)	0.815
Time from hypospadias repair to UCF repair (months)				
≤ 6	1		1	
> 6	0.98 (0.32 to 2.92)	0.97	2.26 (0.74 to 6.90)	0.150
Catheter duration after UCF repair (days)				
≤ 7	1		1	
> 7	2.06 (0.52 to 8.10)	0.300	1.61 (0.34 to 7.62)	0.549
First UCF repair technique				
Other techniques**	1		1	
Multilayer repair	6.88 (0.35 to 13.64)	0.2	13.04 (0.24 to 71.34)	0.209
Suture material used for UCF repair				
Monofilament	1		1	
Braided	2.5 (0.25 to 24.09)	0.428	1.39 (0.57 to 3.41)	0.472
Suture size used for UCF repair				
6–0	1		1	
< 6–0	2.12 (0.67 to 6.67)	0.207	3.45 (0.52 to 11.11)	0.326

* Other techniques for hypospadias repair included tabularised incised plate (*n* = 26), Duckett repair (*n* = 2), and Mathieu repair (*n* = 3).
** Other techniques for UCF repair included simple repair (*n* = 2) and granular fistula converted to grade I hypospadias and repaired (*n* = 4).
UCF – urethrocutaneous fistula
CI – confidence interval, RR – relative risk, UCF – urethrocutaneous fistula

33% after the second.[33] A noteworthy observation from this study is that prior UCF repair did not seem to impact the recurrence rates of subsequent repairs, which is consistent with previous published reports.[23,34]

Conversely, several prior studies reported lower recurrence rates. For instance, a study noted a 6% recurrence rate (2/32) after a TVF repair, and another study highlighted a 13% recurrence rate (2/15) following the PATIO (preserve it and turn it inside out) repair method.[10,35] The low recurrence rates in these studies may be attributed to the surgeon's familiarity with a single surgical technique, which increases standardisation and consistency. However, this discrepancy in recurrence rates emphasises the need for further research to identify the determinants influencing UCF recurrence in various populations and surgical techniques.

In this index study, UCF recurrence after repair was significantly associated with a history of higher-grade hypospadias (grades III–IV). Patients with previous grade III or IV hypospadias have a higher risk of fistula recurrence, which may be due to the complex tissue mobilisation required for neo-meatus placement at the tip. Furthermore, staged repairs, which are commonly performed for severe hypospadias, can compromise surrounding tissues needed for subsequent fistula repair.[23] Another study identified hypospadias severity as an independent risk factor for failed UCF repair, with penoscrotal hypospadias demonstrating a higher recurrence rate.[25]

It is well known that anatomical factors play an important role in UCF recurrence. In this study, distal UCFs were associated with a higher risk of recurrence after repair compared with their mid and proximal counterparts; however, this difference was not statistically significant in multivariate analysis after adjusting for other factors. The higher recurrence of distal UCFs may be attributed to anatomical and technical challenges, including the fragility of the distal penile skin, compromised blood supply from previous surgeries, and traction at the coronal sulcus – particularly during erections in the early postoperative period. Moreover, this anatomical region lacks sufficient tissue for a second-layer closure, further complicating repair efforts.[36] The use of simple repair techniques at this site has also been linked to increased recurrence.

Hence, distal UCFs, especially those located at the coronal sulcus, are particularly challenging to manage. Previous studies recommend avoiding simple repair techniques in these cases, favouring advancement or rotational flaps to improve surgical outcomes instead.[15] However, when the distal fistula is large, it is better to connect it to the distal meatus and convert it to hypospadias, which can be repaired by tabularisation of the urethral plate and multilayer coverage with healthy tissues.[24]

In our study, four granular UCFs were converted to grade I hypospadias and repaired without recurrence. Success in repairing proximal fistulae may be attributed to the meticulous surgical techniques employed and the careful selection of well-vascularised flaps. For instance, two cases of proximal UCF were successfully repaired using TVFs, which provide robust vascular support. Additionally, the low recurrence rate in this group might also be influenced by the relatively small number of patients.

Various techniques for correcting UCFs have been studied, demonstrating variable success rates influenced by factors such as the number, size, location, and nature of the surrounding tissues.[11,12,17,37-39] In our study, most fistulae were repaired using multilayer repair with the penile dartos as an intermediate layer – a straightforward technique. The multilayer UCF closure method is the preferred technique due to its low recurrence rate.[12,40] Surprisingly, in this study, multilayer repair had a higher recurrence rate than other techniques. However, this difference was not statistically significant. This observation may be attributed to the complexity of the fistulae associated with the use of poorly vascularised flaps/scar tissues, resulting in flap necrosis.

Despite the aforementioned limitations, we still encourage the use of multilayer repair for UCFs to improve the success rate. Various other techniques, such as PATIO, multilayer direct closure with a longitudinal relaxing incision, and the use of buccal mucosa and synthetic grafts, have shown promising results with successful UCF repair, highlighting the need for more standardised and individualised approaches in UCF repair.[22,35,39,41]

A history of hypospadias surgeries may impact the recurrence of UCFs, as extensive procedures involving significant tissue mobilisation can compromise the blood supply to the periurethral area, affecting subsequent UCF repairs. Waterman et al.[23] demonstrated that the King and Yoke technique resulted in lower fistula recurrence, likely due to reduced tissue mobilisation and preserved ventral blood supply. In our study, while the tabularised incised plate technique was predominantly utilised compared with staged repair and Mathieu and Duckett techniques, variations in these surgical approaches did not significantly influence UCF recurrence.

UCF repair is generally recommended at least six months after the primary hypospadias surgery to allow proper healing, reduce scarring, and improve skin vascularisation around the fistula site, as reported by previously published data.[12,42] In this study, the median time from the initial surgery to UCF repair was 14 months; 10 patients (22.7%) had repairs done within six months. One case repaired at two months recurred. Repairs at four and five months did not recur. No statistically significant difference in recurrence was found between those repaired before or after six months. This aligns with findings by Waterman et al.,[23] who also saw no significant difference in success rates between early (3.7–6 months) and later repairs (≥ 6 months). While our data does not show a significant statistical difference, waiting at least six months remains the preferred approach to optimise outcomes.

In the current study, the suture material type did not significantly impact UCF recurrence after repair. This result aligns with the study by Wahyudi et al.,[43] which found no significant difference in complication rates between absorbable synthetic braided and monofilament during urethral surgery. Furthermore, the suture material showed no significant impact on UCF recurrence, possibly because other factors like suturing technique, tissue handling, and tissue health play a more crucial role in fistula closure.

The use of a catheter/stent during UCF repair is debatable. Despite varying catheter/stent durations, ranging from 1 to 30 days in all UCF repairs in this study, there was no significant difference in recurrence among the different groups. Waterman et al.[23] concluded in their study that there was no apparent difference in recurrence between using a stent versus not using a stent during UCF repair. Conversely, Dekalo et al.[44] did not view the use of a catheter/stent as a preferable option for UCF repair. We used a urethral catheter/stent for all our patients undergoing UCF repair because we believe it prevents urine extravasation to the surgical site, preventing postoperative dysuria and supporting urethral healing.

Study strengths and limitations

This is the first study to report the recurrence rate of UCFs after repair in Tanzania. It also highlighted the key determinants that influence UCF recurrence after repair. Although the findings of this study are comparable to several other similar studies, these findings should be discussed considering the following limitations. Firstly, the retrospective nature of the study, which resulted in missing patient files and some variables, contributed to the small sample size and may have led to an overestimation of the proportions. Inconsistent documentation led to the omission of important variables for analysis, such as intraoperative tissue handling, fistula number and size, and suturing technique, among others, which may impact UCF repair. Secondly, the study was conducted at a single centre, limiting the generalisability of the findings to a broader population. However, the findings can be generalised to a setting with a similar context. Lastly, the follow-up period was limited to a minimum of six months; a longer follow-up period may result in higher recurrence rates.

Conclusion

This study found that UCFs following hypospadias repair is associated with a 30% recurrence rate, influenced by anatomical and technical factors, such as a history of grade III or IV hypospadias, suture material, and fistula site. Addressing these factors is crucial in developing a strategy to mitigate recurrence and improve surgical outcomes among patients undergoing UCF repair. However, further prospective studies with a large sample size should be conducted

to validate these findings and explore additional factors influencing UCF recurrence.

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Conflict of interest

The authors declare no conflict of interest.

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Ethical approval

Ethical approval was obtained from the College Research Ethics Review Committee of the Kilimanjaro Christian Medical University College (number PG 98/2023). All enrolled patients provided written, general informed consent for the future use of their data/materials for research purposes. In the case of patients younger than 18 years, consent was signed by both the patient and one of their parents.

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