Original Article

Individual treatment strategy for single urethrocutaneous fistula after hypospadias repair: a retrospective cohort study

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Background: Urethrocutaneous fistula (UCF) remains the most common complication after hypospadias repair, and the recurrence rate of UCF is still high if the surgical techniques is not chosen properly, which called for better approaches to this problem. UCF presents different clinical characteristics due to their different locations and sizes, therefore we retrospectively analyzed the effects of different surgical techniques on single UCF after hypospadias repair in order to reduce the recurrence rates of UCF.

Methods: A total of 602 patients diagnosed with UCF after hypospadias repair from January 2014 to December 2021 were enrolled. Baseline clinical characteristics such as age of patients, UCF location, size, surgical techniques were recorded. Patients were followed up in the outpatient clinic. The recurrence of UCF was defined as outcomes. Patients were divided according to the location of the UCF into a coronal UCF group and a non-coronal UCF group, which was then further classified according to the diameter of the UCF. The surgical technique and the recurrence rate of different types of UCF were analyzed and summarized.

Results: A total of 425 patients satisfied the inclusion criteria and 71 patients (16.7%) had recurrent UCF. Five surgical techniques were used to repair the UCF, namely tubularized incised plate (TIP) urethroplasty, Mathieu urethroplasty, double ligation, simple classical closure and trap-door procedure. The recurrence rate was 24.1%, 14.3%, 15.1%, 16.7%, and 22.2%, respectively. TIP or Mathieu urethroplasty is recommended for patients with coronal UCF with glans dehiscence or patients with coronal UCF diameter ≥4 mm without glans dehiscence. In patients with coronal UCF without glans dehiscence, double ligation is recommended for small UCF with diameter <2 mm, and simple classical closure is recommended for UCF with diameter 2–<4 mm. In patients with non-coronal UCF, double ligation is recommended for UCF with diameter <3 mm, and simple classical closure is recommended for UCF with diameter ≥3 mm.

Conclusions: Single UCF can be classified according to the location and size of the UCF. Different types of UCF should be treated with more appropriate individualized strategies, which can effectively reduce the recurrence rate of UCF.

Keywords: Urethrocutaneous fistula (UCF); hypospadias; ligation; simple closure; recurrence rate

Submitted Aug 08, 2022. Accepted for publication Sep 14, 2022.
doi: 10.21037/tau-22-559
View this article at: https://dx.doi.org/10.21037/tau-22-559

Introduction

Despite continuous improvements in surgical techniques, urethrocutaneous fistula (UCF) remains the most common complication after hypospadias repair. The incidence of UCF ranges from 7.5–50%, depending on the degree of hypospadias and the types of surgical technique (1,2). UCF can occur anywhere from the glans to the scrotum. UCF presents different clinical characteristics due to their different locations and sizes, and thus, there are various surgical techniques (3-5) to manage the different types of UCF. The choice of surgical methods is largely determined by the surgeons' own experience, and the lack of standardized and individualized treatment process also leads to a wide range of fluctuations in the success rate of UCF repair. According to literature reports, the overall success rate ranges from 66.5–94.9% (6-9). Therefore, we urgently need to summarize the therapeutic effects of different surgical techniques in the treatment of different type UCF, hoping to find out more appropriate treatment strategies to reduce the recurrence rate of UCF. Herein, we conducted a retrospective analysis of patients with UCF who were admitted to our hospital. The surgical techniques and recurrence rate of different types of UCF were analyzed with the aim of identifying the more appropriate and individual surgical treatment strategy for different type of UCF, thereby reducing the recurrence of UCF. We present the following article in accordance with the STROBE reporting checklist (available at https://tau.amegroups.com/article/view/10.21037/tau-22-559/rc).

Methods

Study design

It was a retrospective cohort study. The study was conducted in accordance with the Declaration of Helsinki (as revised in 2013). The study was approved by Institutional Review Board of The Children's Hospital, Zhejiang University School of Medicine (No. 2022-IRB-198) and individual consent for this retrospective analysis was waived. Patients with UCF after hypospadias repair who were admitted to The Children's Hospital, Zhejiang University School of Medicine, Hangzhou, Zhejiang Province from January 2014 to December 2021 were retrospectively enrolled in this study.

Patient data

Patients with UCF after hypospadias repair who were followed up for at least 6 months since the last operations were included in this study. The following exclusion criteria were applied: (I) patients with multiple UCF, because multiple UCF may have multiple repair techniques and retrospective analysis cannot determine which UCF was recurrent; (II) patients presenting with other urethral complications such as urethral diverticulums or urethral strictures; and (III) patients with coronal UCF with glans dehiscence who underwent a meatotomy procedure, since the postoperative appearance of meatotomy procedure is not satisfactory and there is no possibility of recurrence with this kind of technique.

Clinical data were collected from the medical records, including the patient's age, locations and sizes of the UCF, surgical techniques, postoperative stenting, the follow-up times, and postoperative recurrences. Patients were initially divided into a coronal UCF group and a non-coronal UCF group. These were then further divided into the following 5 subgroups according to the diameter of the UCF: <1, 1–<2, 2–<3, 3–<4, 4–<5, and ≥5 mm. The surgical techniques and recurrence rate of the different types of UCF were summarized.

Surgical techniques and methods

All patients underwent UCF repair at least 6 months after the previous surgery. If the scar around the UCF is obvious, surgery should be performed another 3–6 months later. Surgical procedures included preoperative urethroplasty (tubularized incised plate (TIP) and Mathieu urethroplasty), double ligation, simple classical closure, and trap-door procedure. The surgical techniques are similar to those reported in other literatures (3,5,10-12). Briefly, during the operation, the distal urethra was calibrated to exclude meatal stenosis, diluted povidone iodine solution was injected through a urethral catheter while compressing the urethra proximally to confirm the location and number of UCF and to confirm the presence or absence of urethral diverticulum. A ruler was used to measure the diameter of the UCF. The surgeon adopted the corresponding surgical technique according to the characteristics of the UCF and his own experience. All UCF were covered by vascularized tissue, with a local Dartos flap as the first choice and tunica vaginalis flap as the second choice when there was lake of surrounding tissue. The penile skin was closed on the other line to avoid overlapping sutures.

For most patients, urethral catheter was used for drainage and maintained for 3 to 5 days. If urethroplasty

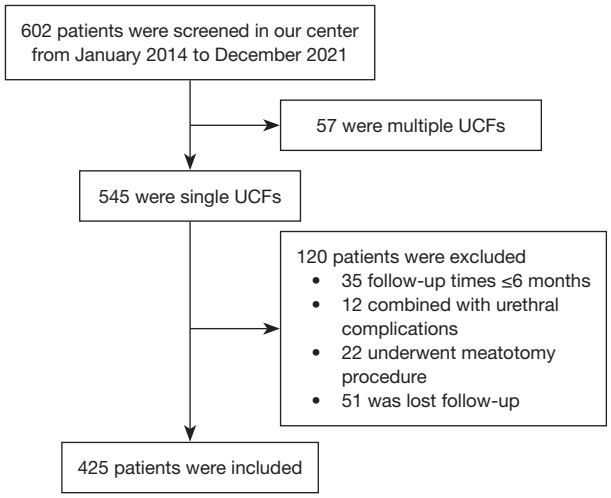

Figure 1 Flowchart of the study population. UCF, urethrocutaneous fistula.

was performed, the urethral catheter was indwelled for 7–14 days. There were some small UCF that did not require indwelling catheters after the operation. Venous antibiotics (cefuroxime or amoxicillin) were administered for 2 to 3 days, and oral antibiotics were administered continuously for 3 to 5 days.

Follow-up visits

Patients were followed up in the outpatient clinic in our hospital at 2 weeks and 6 months after surgeries, and once a year thereafter, mainly to observe the occurrence of urethral-related complications, such as the recurrence of UCF, urethral diverticulum, urethral stricture, glans dehiscence, and others. The recurrence of UCF was defined as our study outcomes.

Statistical analysis

The SPSS 20.0 software was used for statistical analyses. Age, diameter of UCF, and follow-up times are continuous variables with non-normal distribution and are represented as quartiles. Non-parametric tests (U tests) were used for comparisons between different groups. The location of UCF, postoperative stenting, recurrence rate, and surgical methods of UCF repair are categorical variables and are represented as numbers (percentages). The Chi-square test or Fisher's exact test was used for comparisons between different groups. P<0.05 (two-sided) was considered statistically significant.

Table 1 The clinical characteristics of UCF patients

Characteristics	All patients (n=425)
Age (months)	50 [31, 90.5]
Site	
Coronal	89 (20.9)
Non-Coronal	336 (79.1)
Diameter of UCF (mm)	
Coronal	2 [1, 4]
Non-Coronal	3 [2, 5]
Diameter	
<1 mm	15 (3.5)
1–<2 mm	100 (23.5)
2–<3 mm	93 (21.9)
3–<4 mm	83 (19.5)
4–<5 mm	21 (4.9)
≥5 mm	113 (26.6)
Stenting	393 (92.5)
Follow-up (months)	53 [32.5, 73]
Recurrence rate	71 (16.7)
Surgery technique	
TIP	29 (6.8)
Mathieu	7 (1.6)
Trap-door skin flap	9 (2.1)
Double ligation	146 (34.4)
Simple classical closure	234 (55.1)

Parameters are showed as median [IQR], or No. (%) as appropriate. IQR, interquartile range; TIP, tubularized incised plate; D, diameter; UCF, urethrocutaneous fistula.

Results

A total of 602 patients with UCF after hypospadias were admitted to our hospital from January 2014 to December 2021. There were 35 patients who were followed up for less than 6 months, 51 patients were lost to follow-up, 12 patients combined with urethral stricture or urethral diverticulum, 57 patients had multiple UCF, and 22 patients with coronal UCF underwent meatotomy procedure. Finally, 425 patients with single UCF were included in this study (*Figure 1*). The baseline characteristics of the patients are shown in *Table 1*. The median age of the patients was 50 months. The median follow-up time was 24.5 months

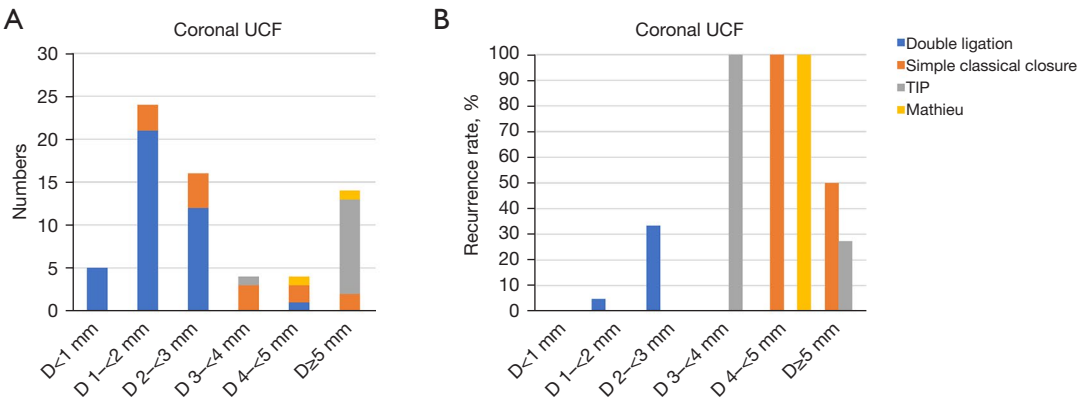

Figure 2 The associated surgical techniques and recurrence rate in different sizes of the coronal UCF without glans dehiscence. Bars are presented with numbers or proportions. D, diameter; UCF, urethrocutaneous fistula; TIP, tubularized incised plate.

(range, 6–102 months). The recurrence rate after UCF repair was 16.7%.

In this study, there were 336 patients with non-coronal UCF, including 55 (16.4%) patients who experienced recurrence. There were 89 patients with coronal UCF, including 16 (18.0%) patients who experienced recurrence. The comparison of recurrence rate between the two groups indicated no statistical difference (P=0.718). The median diameter of the UCF was 2 mm in patients with coronal UCF, and 3 mm in patients with non-coronal UCF. The diameter of the UCF between the two groups was statistically different (P=0.009).

Coronal UCF

Coronal UCF with glans dehiscence

A total of 22 patients with coronal UCF with glans dehiscence were included, and urethroplasty was performed with Mathieu and TIP procedures. The TIP procedure was performed in 17 patients, among which, 3 cases were recurrent (recurrence rate of 17.6%). The Mathieu procedure was performed in 5 patients, with no recurrence.

Coronal UCF without glans dehiscence

The sizes of the coronal UCF without glans dehiscence, as well as the associated surgical techniques and recurrence rate are shown in *Figure 2A,2B*. For the double ligation procedure, there were 5, 21, 12, and 1 patient with diameter <1, 1–<2, 2–<3, and 4–<5 mm, respectively. The recurrence rate was 0%, 4.8%, 33.3%, and 0%, respectively. For the simple classical closure, there were 3, 4, 3, 2, and 2 patients with diameter 1–<2, 2–<3, 3–<4, 4–<5, and ≥5 mm, respectively. The recurrence rate was 0%, 0%, 0%, 100%, and 50%, respectively. For TIP urethroplasty, there were 1 and 11 cases with diameter 3–<4 and ≥5 mm, respectively. The recurrence rate was 100% and 27.3%, respectively. For the Mathieu urethroplasty, there was 1 patient with diameter 4–<5 mm and 1 patient with diameter ≥5 mm. The recurrence rate was 100% and 0%, respectively.

This subgroup included 67 patients and 4 different surgical techniques were used to repair the UCF, namely TIP, Mathieu, double ligation, and simple classical closure procedure. In patients with UCF diameter <3 mm, 84.4% patients underwent operations of double ligation. The double ligation procedure had a low recurrence rate in patients with diameter <2 mm (4.8%), but a high recurrence rate in patients with diameter 2–<3 mm (33.3%). The simple classical closure procedure was mainly used in patients with UCF diameter ≥1 mm. The number of cases using simple classical closure procedure was small and thus, the recurrence rate fluctuated widely, with an overall recurrence rate of 21.4%. The Mathieu and TIP techniques were mainly used in large UCF, with recurrence rate of 50% and 25%, respectively (*Table 2*).

In summary, in patients with coronal UCF, 29 cases were treated with the TIP procedure with a recurrence rate of 24.1%, and 7 patients were treated with the Mathieu procedure, with a recurrence rate of 14.3%. The TIP and Mathieu techniques were mainly applied to coronal UCF with glans dehiscence and coronal UCF with insufficient surrounding tissue. Double ligation was mainly used in patients with coronal UCF with diameter <3 mm. The double ligation procedure had a low recurrence rate in patients with diameter <2 mm (4.8%) but a high recurrence

Table 2 The surgical techniques and the recurrence rate in patients with coronal UCF without glans dehiscence

Diameter	All patients	Double ligation			Simple classical closure			TIP			Mathieu		
		Total	Failed	Recurrence rate	Total	Failed	Recurrence rate	Total	Failed	Recurrence rate	Total	Failed	Recurrence rate
<1 mm	5	5	0	0	–	–	–	–	–	–	–	–	–
1–<2 mm	24	21	1	4.8	3	0	0	–	–	–	–	–	–
2–<3 mm	16	12	4	33.3	4	0	0	–	–	–	–	–	–
3–<4 mm	4	–	–	–	3	0	0	1	1	100.0	–	–	–
4–<5 mm	4	1	0	0	2	2	100.0	–	–	–	1	1	100.0
≥5 mm	14	–	–	–	2	1	50.0	11	3	27.3	1	0	0
Total	67	39	5	12.8	14	3	21.4	12	4	33.3	2	1	50.0

Parameters are showed as no. or percentages as appropriate. TIP, tubularized incised plate; UCF, urethrocutaneous fistula.

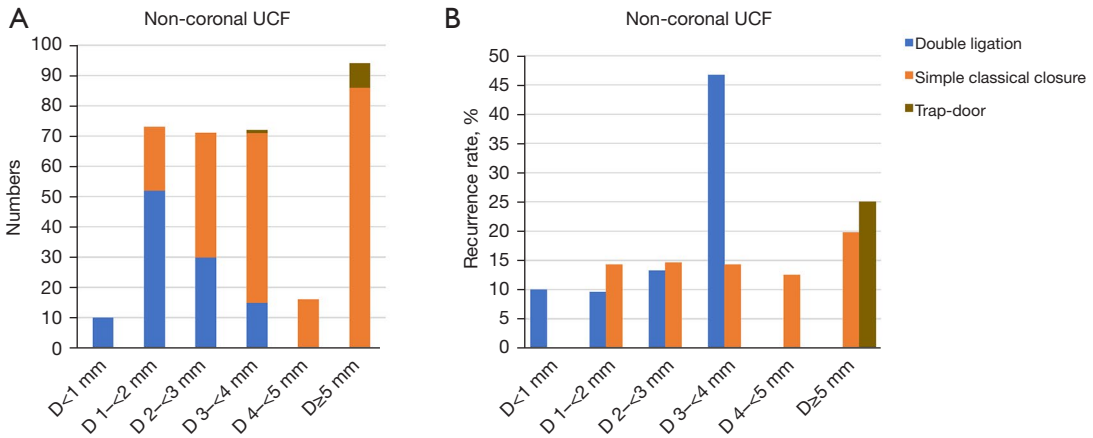

Figure 3 The associated surgical techniques and recurrence rate in different sizes of the non-coronal UCF. Bars are presented with numbers or proportions. D, diameter; UCF, urethrocutaneous fistula.

rate in patients with diameter 2–<3 mm (33.3%). The number of cases using simple classical closure procedure was small, and the overall recurrence rate was 21.4%.

Non-coronal UCF

The sizes of the non-coronal UCF, as well as the associated surgical techniques and recurrence rate are shown in *Figure 3A,3B*. This subgroup included 336 patients and 3 kinds of surgical methods for UCF repair, namely double ligation, simple classical closure, and trap-door procedure. For the double ligation procedure, there were 10, 52, 30, and 15 patients with UCF diameter <1, 1–<2, 2–<3, and 3–<4 mm, respectively. The recurrence rate was 10%, 9.6%, 13.3%, and 46.7%, respectively. For the simple classical closure procedure, there were 21, 41, 56, 16, and 86 patients with UCF diameter 1–<2, 2–<3, 3–<4, 4–<5, and ≥5 mm, respectively. The recurrence rate was 14.3%, 14.6%, 14.3%, 12.5%, and 19.8%, respectively. For the trap-door procedure, there was 1 patient with diameter 3–<4 mm and 8 patients with diameter ≥5 mm. The recurrence rate was 0% and 25%, respectively (*Table 3*).

In summary, the overall recurrence rate of non-coronal UCF was 16.4%, with double ligation giving a lower recurrence rate for UCF patients with diameter <3 mm. In all types of non-coronal UCF, the recurrence rate of simple classical closure was 14.3–19.8%. The trap-door method was mainly used in large UCF. Most patients (74.7%) with UCF diameter <2 mm were treated with double ligation, while 28.8% and 20.8% of UCF patients with sizes of 2–<3 mm and 3–<4 mm, respectively, were also treated with double ligation. The recurrence rate of UCF with diameter

Table 3 The surgical techniques and the recurrence rate in patients with non-coronal UCF

Diameter	All patients	Ligation			Simple classical closure			Trap-door		
		Total	Failed	Recurrence rate (%)	Total	Failed	Recurrence rate (%)	Total	Failed	Recurrence rate (%)
<1 mm	10	10	1	10	–	–	–	–	–	–
1–<2 mm	73	52	5	9.6	21	3	14.3	–	–	–
2–<3 mm	71	30	4	13.3	41	6	14.6	–	–	–
3–<4 mm	72	15	7	46.7	56	8	14.3	1	0	0
4–<5 mm	16	–	–	–	16	2	12.5	–	–	–
≥5 mm	94	–	–	–	86	17	19.8	8	2	25
Total	336	107	17	15.9	220	36	16.4	9	2	22.2

Parameters are showed as no. or percentages as appropriate. TIP, tubularized incised plate; UCF, urethrocutaneous fistula.

<3 mm treated with double ligation was 10.9%. However, for UCF with diameter 3–<4 mm, the recurrence rate of UCF treated with double ligation was as high as 46.7%. The simple classical closure was used in UCF with diameter ≥1 mm, with recurrence rate fluctuating between 12.5–19.8%, and the overall recurrence rate was 16.4%. The trap-door method was mainly used for large UCF (diameter ≥5 mm) and the overall recurrence rate was 22.2%. There was no statistical difference in the recurrence rate between the simple classical closure procedure and the trap-door procedure for large UCF (diameter ≥5 mm).

Discussion

In this study cohort, the overall recurrence rate was 16.7%, and the recurrence rate of coronal UCF was similar to that of non-coronal UCF. This result differed from other literature reporting that the recurrence rate of coronal UCF is higher than that of penile shaft UCF (11,13). In our study, the relatively low recurrence rate of coronal UCF may be related to the difference in the UCF size distribution, with the diameter of coronal UCF generally being smaller than that of non-coronal UCF. The medium coronal UCF diameter was 2 mm, and that of non-coronal UCF was 3 mm, with statistical difference between the groups. It has been reported that UCF size is a risk factor for UCF recurrence (9,14). This may be due to the fact that when the UCF is large, there is insufficient vascularization of local tissues, resulting in local tissue deficiency and high suture tension (9,15).

In the treatment of UCF, surgeons need to choose different surgical techniques according to the location, size, scarring degree, and whether the tissue around the UCF is sufficient. For coronal UCF, Snodgrass *et al.* (3) suggested that urethroplasty is appropriate for patients with glans dehiscence, while for coronal UCF without glans dehiscence, simple UCF repair would be sufficient. In our study, for patients with coronal UCF with glans dehiscence, the surgeons used urethroplasty (TIP or Mathieu) for repair, with TIP being the preferred method in our institution, possibly due to the slit-like urethral opening after TIP procedures (6).

Urethroplasty and UCF repair (double ligation and simple classical closure) is used in coronal UCF without glans dehiscence. Snodgrass *et al.* (3) reported that the simple UCF repair for coronal UCF without glans dehiscence achieved satisfactory outcomes, and the recurrence rate was only 5%. However, the diameter of the UCF included in his study was ≤3 mm, and the effect of this technique for larger UCF remains unknown. Anwar *et al.* (14) also reported the experience of simple classical closure in the treatment of coronal UCF, which achieved satisfactory outcome for UCF with diameter ≤4 mm, but reported a high recurrence rate for UCF larger than 4 mm. Therefore, urethroplasty is recommended for UCF larger than 4 mm. For patients with small UCF (<2 mm), Karakus *et al.* introduced a simple surgical repair method (double ligation) that was simple to operate and had a high success rate (5). In our study, we found that for coronal UCF with diameter <3 mm, surgeons preferred the double ligation procedure and achieved satisfactory results in patients with UCF <2 mm. However, for UCF patients with diameter 2–<3 mm, the recurrence rate of the double ligation procedure was significantly higher than of the simple classical closure method (33.3% *vs.* 0%), and this may be due to the inability of double ligation to achieve tension-free closure when the diameter is large (7). In our study,

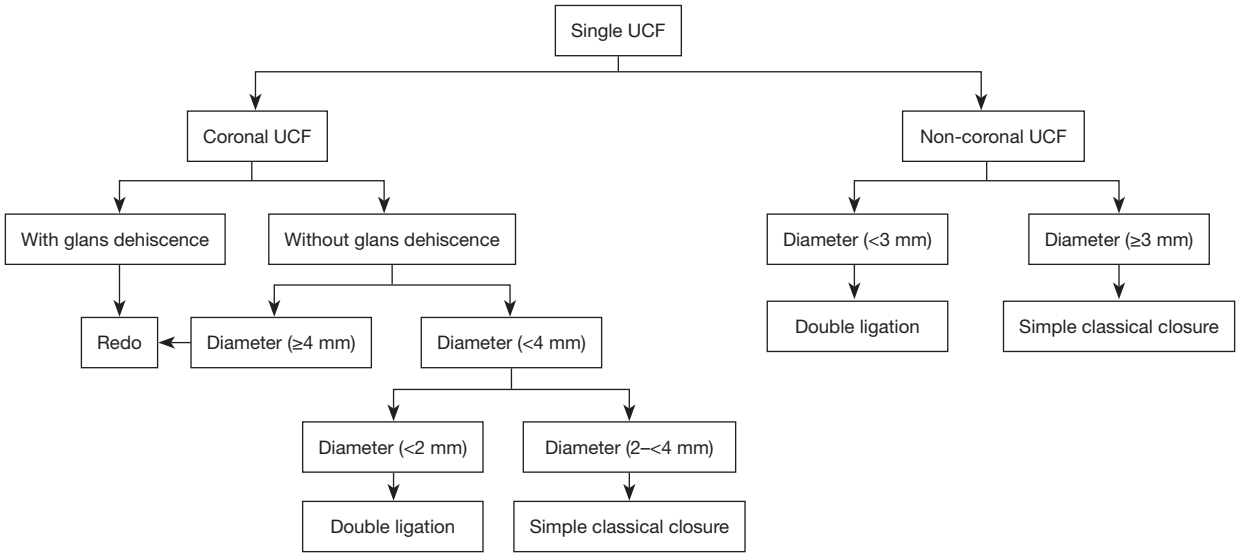

Figure 4 Flowchart of individual treatment strategies for patients with single UCF after hypospadias repair. UCF, urethrocutaneous fistula.

simple classical closure procedure was used to repair UCF ≥1 mm, and this method achieved satisfactory outcomes in UCF with diameter 1–<4 mm and the recurrence rate was 0%, although this may be related to the small number of cases. For coronal UCF with diameter ≥3 mm, some surgeons used urethroplasty to repair the UCF, with the TIP procedure accounting for a high proportion of cases. Since the number of other surgical procedures was small, it was difficult to effectively compare the recurrence rate among these surgical techniques. In summary, for patients with UFC diameter <2 mm, both double ligation and simple classical closure have achieved satisfactory outcomes. Since the double ligation procedure is technically simpler, this is recommended for such patients. However, for patients with coronal UCF with diameter 2–<4 mm, the recurrence rate of simple classical closure is significantly lower than that of double ligation procedure, and thus, simple classical closure is recommended. For patients with diameter ≥4 mm, several different surgical techniques were used. Again, since the number of cases was small, it is difficult to identify which surgical technique is superior. However, combined with data from previously published literature, urethroplasty is recommended. Future prospective studies should be performed to compare the differences in recurrence rate between different surgical techniques for large coronal UCF.

The surgical treatment strategy for non-coronal UCF patients is relatively simple. In our study, double ligation and simple classical closure were used in most non-coronal UCF patients, while the trap-door procedure was used only in a few cases. In this subgroup, we found that surgeons preferred the double ligation procedure for UCF with diameter <3 mm, and achieved a satisfactory outcome. There was no statistical difference in the recurrence rate between the double ligation procedure and the simple classical closure procedure (10.9% *vs.* 14.5%, P=0.5). However, for patients with UCF diameter 3–<4 mm, the recurrence rate of double ligation procedure was significantly higher than that of the simple classical closure procedure (46.7% *vs.*14.3%, P=0.018). In addition, the trap-door procedure was partially used in large UCF patients (diameter ≥3 mm), which had a similar recurrence rate to that of simple classical closure (22.2% *vs.* 17.1%, P=1.000). In summary, for patients with large non-coronal UCF (diameter ≥3 mm), trap-door and simple classical closure have achieved satisfactory outcomes. However, simple classical closure is technically simpler, and therefore the method of choice for these UCF. For small UCF (diameter <3 mm), the recurrence rate of double ligation is similar to that of simple classical closure, and since the procedure of double ligation is simpler, again, double ligation is recommended for such UCF.

Although the determination of surgical technique for UCF repair is largely influenced by the surgeon's personal experience, it is hoped that the individualized treatment strategy (*Figure 4*) for single UCF based on the above data analysis can reduce the recurrence rate. In summary, according to the characteristics of the single UCF, we recommended that single UCF be divided into

the following three groups: type A being coronal UCF with glans dehiscence, type B being coronal UCF without glans dehiscence, and type C being non-coronal UCF. Due to the lack of surrounding tissues, urethroplasty should be performed for type A UCF. Depending on the UCF diameter, type B can be subdivided into type B1 (diameter <2 mm), type B2 (2–<4 mm), and type B3 (diameter ≥4 mm). For type B1, there is no difference in the recurrence rate between double ligation and simple classical closure, and since the operation procedure of double ligation is relatively simpler, this method is recommended. For type B2, the simple classical closure is recommended, while for type B3, urethroplasty is recommended. Type C can also be subdivided according to diameter, namely type C1 (diameter <3 mm) and type C2 (diameter ≥3 mm). For type C1, double ligation is recommended, while for type C2, simple classical closure is recommended.

There were some limitations to this study. First, this study is a single-center retrospective analysis, and future prospective studies are warranted to verify these conclusions. Second, postoperative indwelling of the catheter may be a factor affecting recurrence rate. However, there is no unified standard for postoperative indwelling catheters and it remains based on the subjective judgment of the attending surgeon. Third, due to the lack of clear diagnostic criteria for urethral stricture and urethral stenosis (15), and the difficulty in finding small urethral diverticulum during outpatient follow-up, the number of postoperative urethral strictures or urethral diverticulum was not assessed in this study, and this may lead to deviations in surgical outcomes. Finally, the number of cases in some subgroup was small (for example, there was only 1 case of coronal UCF with diameter 4–<5 mm repaired with double ligation procedure) and this may have affected the statistical efficiency.

Conclusions

For single UCF patients, individualized treatment should be adopted, and appropriate surgical techniques should be determined according to the location, size, scarring degree, and condition of the penile skin around the UCF, which may greatly reduce the UCF recurrence rate. For type A and type B3 UCF, urethroplasty is recommended, double ligation is recommended for type B1 and type C1 UCF, and simple classical closure is recommended for type B2 and type C2 UCF.

Acknowledgments

Funding: This study was funded by Basic Public Welfare Research Project of Zhejiang Province (No. LGF22H050004).

Footnote

Reporting Checklist: The authors have completed the STROBE reporting checklist. Available at https://tau.amegroups.com/article/view/10.21037/tau-22-559/rc

Data Sharing Statement: Available at https://tau.amegroups.com/article/view/10.21037/tau-22-559/dss

Conflicts of Interest: All authors have completed the ICMJE uniform disclosure form (available at https://tau.amegroups.com/article/view/10.21037/tau-22-559/coif). The authors have no conflicts of interest to declare.

Ethical Statement: The authors are accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. The study was conducted in accordance with the Declaration of Helsinki (as revised in 2013). The study was approved by Institutional Review Board of The Children's Hospital, Zhejiang University School of Medicine (No. 2022-IRB-198) and individual consent for this retrospective analysis was waived.

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(English Language Editor: J. Teoh)

Cite this article as: Yang F, Ruan J, Zhao Y, Zhu L, Wang X, Chen G. Individual treatment strategy for single urethrocutaneous fistula after hypospadias repair: a retrospective cohort study. Transl Androl Urol 2022;11(9):1345-1353. doi: 10.21037/tau-22-559

