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Original Research Article

Acquired and congenital urethrocutaneous fistula in pediatric population: a tertiary care centre experience

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ABSTRACT

Background: Urethro- cutaneous fistula in pediatric population is very common after hypospadias surgery ranging from 4-25% in world literature. Other acquired and congenital causes are rare. Successful repair of UCF depends on different technical factors and basic surgical principles. Considering the agony of multiple surgeries in a child and plan after failure is uttermost import and demands more challenge than the primary surgery. We are discussing our experience of UCF management in a tertiary care centre.
Methods: The study was conducted during February 2021 to January 2025 in the Department of Pediatric Surgery among 89 boys with urethrocutaneous fistula.
Results: Among 89 patients post urethroplasty (84), post traumatic (2), post circumcision (2) and 1 congenital urethrocutaneous fistula was selected. Proximal fistula (46), multiple fistula (18) and 8 among 20 distal fistula patients were repaired with Tunica Vaginalis reinforcement. TV reinforcement resulted in 87.7% success rate and only 9 patients with recurrence. Post circumcision fistula and post traumatic UCF repair is also difficult considering its location.
Conclusions: The treatment plan for a fistula must be individualized based on certain variables and basic surgical principles. The significantly improved success rate with the addition of a waterproofing layer suggests the use of this interposition layer should be done at the earliest available opportunity to prevent a reccurnce rather than to reserve it for future options during UCF repair.

Keywords: Congenital fistula, Post-circumcision fistula, Post-traumatic fistula, Tunica vaginalis flap, Urethrocutaneous fistula

INTRODUCTION

Urethrocutaneous fistula (UCF) may develop as an acquired or a congenital disorder. Congenital fistula is rare and acquired fistula is very common after hypospadias surgery (4-25%) and also seen after other traumatic events, circumcision, periurethral abscess, secondary to urethral stones in adults and very rarely neoplastic origin.[1-4] Though more than 150 procedures described during the evolvement of hypospadias surgery, UCF occurs after hypospadias surgery regardless of the location of meatus, procedure performed or experience of the surgeons.[5,6] Successful repair of UCF depends on several basic principles. Different techniques have been described for fistula repair but the simple closure bears the potential risk of recurrence.[7-10] Reinforcement with waterproofing layer (tunica vaginalis, dartos and external spermatic fascia) significantly reduces the recurrence rate of fistula.[11,12] Surgery for UCF repair is equally demanding procedure compared to primary surgery of hypospadias.[13] In this study we are going to discuss the methods of repair of UCF in different types of fistula location, size and number and also share our experience of repairing UCF and have focused the challenges of

surgical techniques following different acquired and congenital urethrocutaneous fistula patients presented in a tertiary care centre.

METHODS

This single institute based observational study was conducted in the department of Pediatric surgery, IPGMER and SSKM Hospital, Kolkata, for 4 years (1st February 2021 to 31st January 2025). All boys aged 1-12 years having congenital or acquired urethrocutaneous fistula (single or multiple) following hypospadias repair or circumcision or after trauma were included in the study. Patients with fistula (congenital) below 1year age and above 12 years age who lost follow up after fistula (acquired) were excluded from the study.

Patient sampling was done by identifying urethrocutaneous fistula clinically present either congenitally or after hypospadias or other surgery among all male patients who came to OPD for follow up. After clinical assessment of fistula site, diameter of urethral meatus and fistula, number of fistula (single or multiple), skin and tissue thickness proximal and distal to fistula (Figure 1), previous surgical scar decision of UCF repair procedure was planned. Preoperative UCF location was done by observing clinical photo and video of micturition and intra-operative by Povidone Iodine (10%) installation through meatus and observing the leaking sites (Figure 2). Preoperative application of local coconut oil for skin softening were advised, some were advised Testosterone ointment depending on skin thickening and scar prior to UCF repair.

Repair of UCF done in main OT complex of the hospital by senior surgeons of pediatric or plastic surgery department as elective procedure. Surgeries were performed under general anaesthesia and post operative caudal analgesia. 6-0 polyglactin absorbable suture (vicryl) was used for the repair. Dressing after repair was primarily done by paraffin-soaked gauge after application of ointment mupuricine and gentle compression bandage which was removed on postoperative day 5. Urethral stent (No 6 or 8 Nelaton Catheter) was kept for 10-12 days post operatively. During this period tablet Oxybutynin was given to avoid bladder irritation due to catheter. For older children Tablet Chlordiazepoxide was used for prevention of penile erection. Irritable children were kept under sedation for immediate post operative period and Paracetamol was used as analgesia. Use of Anterior urethral dilator 1/2 with adequate lubrication was advised 3 weeks after surgery. Patients have been advised to follow up in OPD with micturating video and related data was recorded and saved. All data of patients were recorded from OPD, OT register, Bed Head Tickets and results were collected, tabulated and analysed. Microsoft Word and Excel 2010 software have been used for tabulation and data analysis. Consent from parents and/or guardians have been taken before every fistula surgery routinely explaining the chance of future fistula in same or another site as hospital protocol. Institutional Ethical Approval was not required for the study as it was an observational study. Photographs are taken from patients with permission from guardians for research purpose.

RESULTS

In our study we have total 89 patients with urethrocutaneous fistula (UCF). 84 patients undergone surgery for hypospadias, 2 had fistula following trauma, 2 had fistula following circumcision and 1 was congenital. All patients in our study are between 1-12 years and according to age distribution 69 patients (77.5%) are between age group 2-6 years (Figure 9). Among total 576 cases of urethroplasty in 4 years urethrocutaneous fistula (UCF) was noted in 84 cases. Among 84 post urethroplasty fistula patients 46 had single proximal fistula, 20 single distal fistula and 18 multiple (more than 1) fistula sites. All post circumcision UCF patients had coronal location, post traumatic patient one granular and other at mid penile location and congenital UCF was distal penile fistula.

Figure 1: Preoperative clinical assessment of fistula site, number, skin around.

Figure 2: Intraoperative assessment of small fistula site by proximal tourniquet and distal instillation of Povidone iodine (10%) solution.

Figure 3: Distal urethrocutaneous fistula after urethroplasty.

Figure 4: Proximal urethrocutaneous fistula direct closure by local tissue (without TV flap).

Figure 5: Multiple urethrocutaneous fistula.

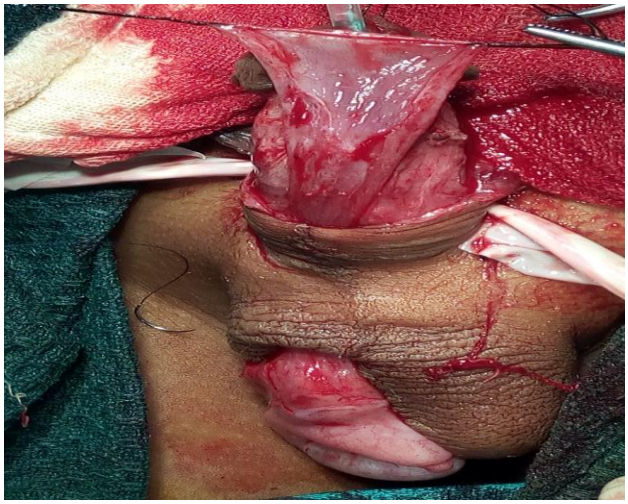

Figure 6: Reinforcement by Tunica vaginalis flap (Right side testis) after repair.

Figure 7: Post circumcision fistula showing double stream one from normal meatus (narrow stream) and another at coronal level (wide stream).

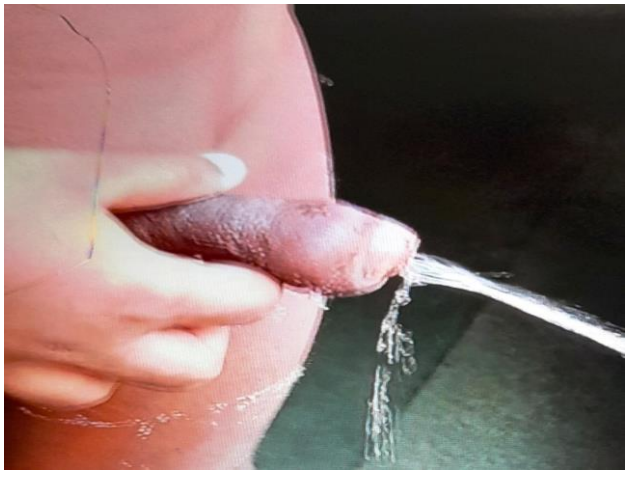

Figure 8: Post traumatic fistula at granular level.

All proximal fistula in our study following urethroplasty, patients were repaired with Tunica Vaginalis flap (TV Flap) reinforcement after primary repair and only 6 had recurrence. Among these 6 "recurrent" UCF patients 1 undergone buccal mucosal graft (BMG) staged reconstruction, 2 undergone reinforcements with opposite side TV Flap and 3 are waiting for surgery.

Among 20 patients with distal fistula after urethroplasty, 8 were repaired with TV flap reinforcement and no recurrence and rest 12 patients underwent primary repair with local tissue reinforcement (spongioplasty) and 4 had recurrence UCF. Among these 4 "recurrent" UCF patients, 1 has been repaired with TV reinforcement and other 3 are waiting.

All multiple fistulae patients were repaired with re-do urethroplasty and TV flap reinforcement and no recurrence were noted. Post circumcision and congenital UCF were repaired with primary closure. Post traumatic glanular level UCF was repaired with primary closure and mid penile level fistula with TV reinforcement. One patient with traumatic UCF at mid penile level repair underwent "recurrent" fistula and he is waiting for next surgery. Among total 89 patients 13 patients had recurrence of fistula and 7 are still waiting for further surgery during this period of study and among 73 patients with TV reinforcement repair 64 had no further fistula (Table 1).

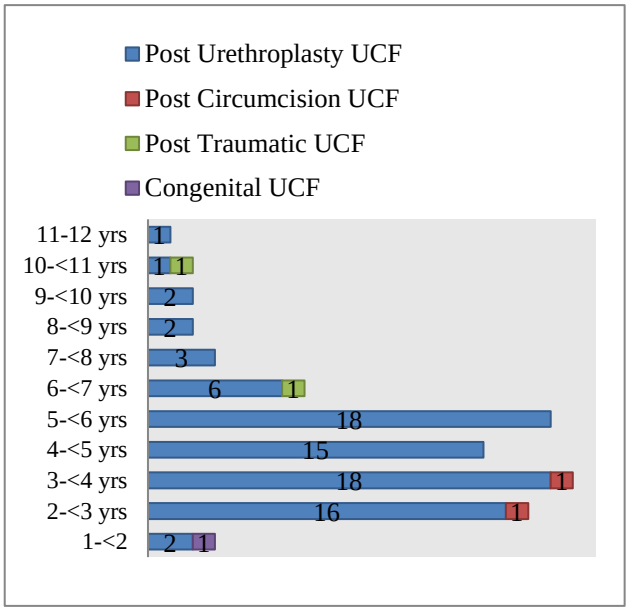

Figure 9: Age distribution table of different acquired and congenital UCF patients (boys).

Table 1: Position and types of repairs of acquired and congenital urethrocutaneous fistula.

Acquired or congenital	Types of UCF	Location of UCF	Number of patients	Repair type	Recurrence after UCF repair	Redo repair type
Acquired UCF	Post urethroplasty (hypospadias) (n=84)	Proximal penile	46	with TV	6	1 BMG 2 TV (opposite site) 3 waiting
		Distal penile	20	with TV=8 without TV=12	with TV = 0 without TV=4	TV =3 1 waiting
		Multiple	18	Open +TIP+TV	2	2 waiting
	Post circumcision (n=2)	Coronal	2	primary	0	
	Post traumatic (n=2)	Granular	1	primary	0	
		Mid penile	1	primary+TV	1	Waiting
Congenital UCF	Congenital (n=1)	Distal penile	1	Primary	0	

*UCF-urethrocutaneous fistula, TV-tunica vaginalis flap, TIP-tabularised incised plate urethroplasty, BMG-Buccal Mucosal Graft urethroplasty.

DISCUSSION

Urethrocutaneous fistula (UCF) may develop as an acquired or a congenital disorder. Congenital anterior urethrocutaneous fistula is rare and an acquired urethral fistula may result most frequently from urethral trauma, such as surgery.[1] By far the most common cause for acquired urethrocutaneous fistula is following a surgical intervention for hypospadias repair or circumcision. There are also rare reports of urethral fistula developing following an infection, neoplastic origin, periurethral abscess or secondary to urethral stone in adults.[2-4] In our study we found 89 cases of UCF during this period among which mostly after hypospadias surgery (n= 84), 2 after circumcision (both were done outside hospital as ritual circumcision), 2 after trauma (direct trauma to genitalia during playing and one following repair in emergency after trauma) and 1 found congenital UCF.

Urethrocutaneous fistula after hypospadias surgery is very common, in literature it varies from 0 to 23%.[11,12,14,15] The success of hypospadias surgery can be judged by the incidence of fistula.[16] Durham Smith in a study of hypospadias surgery noted varied fistula rates for

different procedures. Flip-flap repair varied from 2.2 to 35%, island pedicle tube flap 4 to 35%, free graft tube flap had a fistula rate of 15 to 50%.[17] In his study Durham Smith showed 3% fistula in 500 cases.[18] In our study among 576 cases in 4 years Urethro cutaneous fistula (UCF) noted in 84 cases (14.58%). Local infection, local ischaemia, inadequate procedure, poor tissue handling, distal obstruction due to distal stenosis or encrustration with severity of hypospadias has significant impact on the outcome of the primary hypospadias repair. According to literature proximal urethral fistula is common and it occurs due to distal obstruction and narrow stenotic neo meatus.[19] In our study majority of fistula (n=46) are proximal (54.76%) (Figure 3), distal fistula in 20 patients (23.8%) (Figure 4) and 18 had multiple fistula in urethra (Figure 5).

It is also an important point to consider that the scar tissue left by previous surgery can compromise the success of this reconstructive surgery. Therefore, the key factors for success in this type of surgery are gentle tissue handling, the use of fine sutures, a subepithelial urethral closure and the interposition of tissue to avoid direct contact with suture lines.[20] Here the challenge of fistula repair arrives. Post-surgery scarred thick tissue requires many important considerations to prevent further fistula formation. Many efforts for prevention of fistulae are undertaken like overlapping suture lines, two-stage repair, burying repaired urethra in scrotum, dartos flap, overlapping denuded subcutaneous tissue, rotating skin flaps etc.[18,21,22]

Tunica vaginalis flap from the parietal layer of testis cover of anastomosis of urethroplasty is one more option which helps in the reduction of urethro-cutaneous fistulae.[11] TV flap is used as reinforcement after repair single or multiple fistulae. We have used TV flaps in 73 patients (Figure 6) with UCF where 64 patients had no recurrence of fistula securing success rate of 87.7%. Shanker et al, had a similar experience with the use of tunica vaginalis flap for fistula surgery.[12] They reported a success rate of 50% with fistula of >2 mm but without the use of tunica vaginalis and had only one recurrence in six cases when tunica was used in a third attempt for fistula repair. They reported success in 71% cases in first attempt for fistula repair, 70% in second attempt and 50% in third and subsequent repairs.[23] We have recurrence in 13 patients after UCF repair, among which 6 have been successfully repaired and 7 are still waiting.

Post circumcision UCF is a major and frequent complication of circumcision mostly practiced by non-qualified personnel on children aged 24-36 months. The usual presentation is micturition with a bifid stream occurring on average 3 months (ranging from 14 days to 108 months) after circumcision.[24] Coronal fistulas are the commoner location.[25] In our study 2 children presented with post circumcision UCF (Figure 7). Both were under 4 years age and location was coronal. Concerning surgical repair of UCF, some surgeons prefer a direct closure whereas others surgeons prefer skin flaps.[20] We repaired with primary closure in both the cases.

In our study, one patient presented with UCF causing double stream of urine who attended emergency 2 weeks ago with penile trauma with laceration and he had been repaired with suturing. Another patient came with penile laceration during playing and post repair UCF developed. According to literature, the incidence of urethrocutaneous fistula complicating penile trauma is not common. There may be laceration, contusion or avulsion of penile skin and urethral involvement either as contusion or laceration may result in an urethrocutaneous fistula.[26,27] One fistula was at mid penile level which was repaired with TV reinforcement and another at granular level (Figure 8) repaired primarily. In spite of tissue reinforcement, the mid penile level fistula had recurrence which is waiting for further repair.

Congenital anterior urethrocutaneous fistula (CAUF) is a very rare anomaly of the penile urethra. It is usually seen as an isolated deformity or may accompany genitourinary or other malformations.[28,29] The cause is unclear but probably reflects a focal defect in the urethral plate that prevents fusion of the urethral folds.[30,31] We received a patient with very small CAUF which was repaired primarily. Small fistulas which are <0.5 cm can be easily closed primarily after refreshing the edges and covered by skin.[32]

Distal urethral stenosis or obstruction after urethroplasty causes proximal fistula, use of anterior urethral dilator is recommended by some surgeons but considering difficulty in pediatric urethral dilatation in home and also urine itself being a good self-dilator, some are against dilatation. We have not considered urethral dilatation as inclusion criteria for the study, in future study it needs to be included. Adequate phallus length is another criteria for success after urethroplasty which is also not included in our study. Though our preoperative recommendation of toFigureal application of coconut oil or testosterone cream was mentioned, but categorisation not done prior to surgery to note the result, which also is a limitation of this study. Adequate length of follow up period is also required as we could not complete final surgery after recurrence in 7 patients.

CONCLUSION

The treatment plan of fistula repair must be individualised and to be decided according to size, location and number of fistula as well as surrounding skin or tissue scar. Minimum time interval between two surgeries for acquired urethrocutaneous fistula is also important. Tunica vaginalis as waterproofing interposition flap gives a good reinforcement for fistula repair and also significant improvement in second and third surgeries. The significantly improved success rate with the addition of a waterproofing layer suggests the

use of this interposition layer should be done at the earliest available opportunity to prevent a reccurnce rather than to reserve it for future options during UCF repair.

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