



Clinical Classification of Urethrocutaneous Fistulas Developing after Hypospadias Repair

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Abstract

Background Clinical classification of the urethrocutaneous fistulas (UCFs) was designed to help the surgeons in (1) categorizing the fistulas, (2) selecting appropriate treatments, (3) keeping record at presentation and discharge, and (4) transferring information while referring a patient with recurrent fistula to a higher center.

Methods This retrospective study comprised of 68 patients with UCFs who reported in the “Hypospadias and VVFs Clinic” between 2004 and 2016. The study was performed to determine the incidence or etiology of the UCFs. It was rather performed to classify fistulas into different categories depending on the number of fistulas: A (5 fistulas), B (16 fistulas), C-a (28 fistulas), C-b (4 fistulas), D (4 fistulas), and E (11 fistulas). Category A fistulas healed conservatively. Category B fistulas underwent transection of the fistula tracts (tractotomy), purse-string closure, or multilayered closure (fistulorraphy). Category C-a fistulas were reenforced by preputial or penile skin flaps or waterproofing flaps. Category C-b fistulas underwent re-tubularization of their neo-urethral plates and eccentric closure of peno-preputial skin. The urethral plates of category D fistulas were re-tubularized after 3 to 6 months and cover was provided by the Cecil-Culp procedure. Category E fistulas had associated hairy urethra, stricture distal urethra, stricture with diverticulum, perifistular scar-induced chordee, long narrow urethral plate, balanitis xerotica obliterans (BXO), and short reconstructed neourethra. Accordingly, appropriate corrective measures were taken. Miscellaneous category F was excluded from the study.

Keywords

- ▶ classification
- ▶ urethrocutaneous fistulas
- ▶ reconstructive ladder

Results Except for one in category D, none of the patients had any recurrence of fistula. One patient of category E had residual diverticulum.

Conclusion The designed clinical classification of UCFs is simple. Treatment was in accordance with reconstructive ladder wherein complexity of treatment paralleled with increasing complexity of fistulas.

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Introduction

Development of urethrocutaneous fistulas (UCFs) remains the main postoperative complication in repaired hypospadias ranging from 12 to 90%.¹ Different modes of treatment for UCFs had been used by different authors, but none of the studies had described such a classification that could directly point toward the characteristics of UCFs, urethra, urethral plates, peno-prepuccio-scrotal region, and the associated morbidities like the hairy urethra, urethral stricture, urethral diverticulum, urethral diverticulum with concretions, and the quality and quantity of available dispensable peno-prepuccio-scrotal tissues to provide a secured cover to the closed fistulas, thus, this could have helped the operating surgeon to select the best possible treatment option more quickly. In the present study, the treatment ranged from the simplest conservative treatment (transurethral bladder decompression) to the simplest surgical procedure by transection of the fistula tract (tractotomy), purse-string closure, or multilayered closure (fistulorrhaphy). More complex surgical repair required preputial or penile skin flaps (skin flap fistuloplasty), preputial and scrotal dartos fascial waterproofing flaps, or the tunica vaginalis (TV) flaps (waterproofing flap fistuloplasty) for reinforcement of closed fistulas. The most complex procedures were closure of fistulas or redo urethroplasty and their re-enforcement just like the Cecil-Culp procedure.²

Patients and Methods

This retrospective study comprised 68 UCFs of different categories (category A, $n = 5$; category B, $n = 16$; category C-a, $n = 28$; category C-b, $n = 4$; category D, $n = 4$; and category E, $n = 11$), developing into 68 repaired hypospadias in patients aged between 10 and 25 years, who reported for follow-ups in the "Hypospadias and VVFs Clinic" of Department of Burns and Plastic Surgery, Postgraduate Institute of Medical Sciences, Rohtak, between 2004 and 2016. Photographic records of (1) different types of UCFs, (2) different characteristics of tissues of peno-prepuccio-scrotal region (native and neourethra, native and neourethral plate, and also the preputial hood, previous preputioplasty, unused preputial hood, penile skin cover, corpora cavernosa, corpora spongiosum, quality and quantity of scrotal skin, cryptorchism, unilateral undescended testis, inguinal hernia, hydrocele, and varicocele), and thereafter (3) choosing for them the appropriate treatment protocol constituted the material and methods for this study. Based upon these three parameters, a clinical classification was designed (Table 1). The treatment plan was finalized after going through the details of (1) history, (2) previous surgical interventions and its postoperative outcome, and (3) examination of UCFs and tissues of peno-prepuccio-scrotal region. This retrospective study was performed (1) to categorize the UCFs and (2) to suggest appropriate treatment in a pattern of a reconstructive ladder. This study was not aimed to know the incidence and etiological factors of UCFs, that is, age of hypospadias, location of the original meatus, type of single or staged original repair or

the subsequent surgical repair, and the isolated characteristics of UCFs like their location or size. This clinical classification has incorporated all factors that are of clinical use to the operating surgeon while repairing UCFs. Repaired hypospadias below 10 years of age either did not report in the hypospadias clinic or would have gone to the pediatric surgery department.

All the five acute ultra-micro-UCFs in category A were put on conservative treatment by uninterrupted bladder decompression using an 8-Fr infant feeding tube for a period of 10 more days after detection of leakage of urine. Five of 16 fistulas of category B were treated with transection of the fistula tracts (tractotomy) through skin incisions in coronal sulcus ($n = 2$) and the lateral side of the penile skin ($n = 3$; Fig. 1), 3 were closed using purse-string suture of the dissected ostium of fistula by 6-0 Vicryl on round-bodied needle (Fig. 2), and the remaining 8 underwent a multilayered closure of the dissected perifistular soft tissue (fistulorrhaphy; Fig. 3). Eight of 28 category C-a fistulas were closed and reinforced by preputial skin flaps designed from the tissues of previously unused skin present on either side of coronal sulcus, 5 were reinforced by penile skin flaps (skin flap fistuloplasty; Fig. 4), and 15 were provided preputial dartos flaps ($n = 4$; Fig. 5), scrotal dartos flaps ($n = 3$; Fig. 6), and TV flaps ($n = 8$; waterproofing flap fistuloplasty; Fig. 7). Five of 28 category C-a UCFs were recurrent fistulas with a history of one ($n = 3$) or more ($n = 2$) failed surgical attempts, and all responded favorably with flap closure (fistuloplasty). Another four UCFs, two cases each of mega-UCFs (Fig. 8) and near-total disruption of neourethras (Fig. 9), were kept in category C-b, wherein re-tubularization of their neourethral plates was done and the loose penile skin on either side of the re-tubularized neourethral plates was mobilized to provide a multilayered eccentric closure like the D.Smith technique³ (Fig. 10).

In four cases of category D fistula, two had multiple fistulas, one had mega-UCF, and other one was of near-total disruption of neourethra (Fig. 11). The multiple fistulas were converted into one larger fistula before repair by incising their intervening skin bridges. The urethral plates of multiple fistulas, mega-fistula, and near-total disrupted neourethra were re-tubularized after 3 to 6 months and a secured cover was provided using the Cecil-Culp procedure (Fig. 12). In near-total disruption of urethroplasty, the intact part of the glans was incised before proceeding for repair in the form of a complete redo urethroplasty, else there would be risk of occurrence of UCF at the coronal sulcus. In 11 fistulas of category E, hairy urethra was present in two, stricture of the distal urethra in two (Fig. 13), stricture of the penile urethra with a diverticulum in three (Fig. 14) of which one diverticulum had concretions, scarring, and residual chordee in one (Fig. 15), long segment of narrow urethral plate on fistulotomy in one (Fig. 16), balanitis xerotica obliterans (BXO) in one (Fig. 17), and one had short reconstructed neourethra. Accordingly, the hairy part of the neourethra was excised in one and electrodepilated in the other. A strictured urethra in both was laid open and grafted with inner preputial full-thickness skin

Table 1 Clinical classification of urethrocutaneous fistulas (UCFs) in repaired hypospadias

Category and number of cases	Basis for categorization: (1) urethral calibration and methylene blue dye test, (ii) artificial penile erection, (iii) examination of native and neourethras, (iv) inspection of UCFs, urethral plate and the perifistular tissues, and (v) thorough study of peno-prepuccio-scrotal region were routinely done for categorization of UCFs	Characteristics of the given UCF/UCFs	Appropriate management following reconstructive ladder ^a
A (n = 5)	Healthy urethra and neourethra with thick perifistular healthy tissue	Acute ultra-micro-UCF with its unepithelialized tract	Conservative treatment by bladder decompression
B (n = 16)	Healthy urethra and neourethra with thick perifistular healthy tissue	Eccentrically located ultra-micro-UCFs with epithelialized tracts Centrally located ultra-micro, micro-, and macro-UCFs with epithelialized fistula/tracts (mature fistulas)	Transection of fistulous tract (tractotomy) Purse-string closure or multilayered closure (fistulorrhaphy)
Ca (n = 28) Cb (n = 4)	Healthy urethra and neourethra with thick perifistular healthy tissue and presence of preputial and/or penile skin in the coronal sulcus. Five UCFs were recurrent with a history of one or more failed surgical attempts Healthy urethra and urethral plates is fit for re-tubularization. Availability of plenty of loose and dispensable penile skin to provide secured cover of soft tissue and skin	Macro-UCF with availability of thick tissue all around or in its vicinity Mega-UCFs and near-total disruption of neourethra. Healthy reusable urethral plates	Repair and re-enforcement with skin and/or water proofing flaps (fistuloplasty) Re-tubularization of broken down neourethral plates and an eccentric closure using the D.Smith technique ³
D (n = 4)	Healthy native and neourethra and a healthy neourethral plate sufficient to reconstruct neourethra but acute deficiency of dispensable peno-preputial skin not enough to provide a secured skin and soft-tissue cover to the neourethra against re-fistulization (recurrences)	Multiple UCFs, mega-UCFs, and near-total disruption of neourethra except glans. These are complex UCFs	Multiple fistulas were converted in to one larger fistula. Six months later, tubularization of the urethral plate after dorsal median urethrotomy and a Cecil-Culp procedure for skin and soft-tissue cover (skin and waterproofing flap)
E (n = 11)	Fistulas with (i) hairy urethra, (ii) urethral stricture, (iii) urethral diverticulum, (iv) urethral metaplasia, (v) long segment of a narrow strictured urethral plate, (vi) balanitis xerotica obliterans, (vii) residual significant chordee, and (viii) short reconstructed neourethra	The characteristics of UCFs do not matter in the presence of too many other associated morbidities of the urethra. These are complex UCFs	Each one of the urethral morbidities were corrected as described in the section of patients and methods
F (n = 8)	Miscellaneous UCFs (UCFs that are congenital, iatrogenic, self-inflicted, infective, decubitus in paraplegics, and in malignancy. Preputial fistula is not UCF in the true sense	All other UCFs not related to hypospadias urethroplasty	The treatment will depend upon the etiopathogenesis and other characteristics of UCFs

^aReconstructive ladder: Treatment starts from the simplest procedure and thereafter proceeds toward the most complex procedures, that is, category A → conservative treatment; category B → tractotomy, purse-string suture, and fistulorrhaphy; category Ca and Cb → fistuloplasty (skin flaps or waterproofing flaps with eccentric multilayered closure); category D → Cecil-Culp technique; category E → redo urethroplasty cum Cecil-Culp procedure; and Category F → depending upon the etiopathogenesis and the characteristics of the fistula.

Categories A, B, Ca, and Cb are simple UCFs. Every part and tissues are normal except for the presence of UCFs, and simple surgical procedures are required.

Categories D and E are complex fistulas. These fistulas have shortage of healthy tissues and require complex surgical procedures.

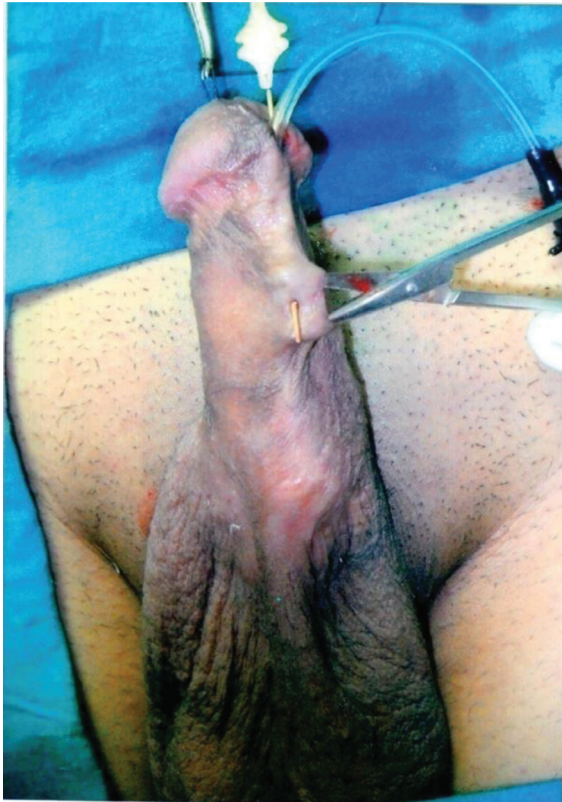


Fig. 1 Division of the fistula tract (tractotomy) through lateral penile skin incision. Unstitched wounds are left to heal spontaneously.

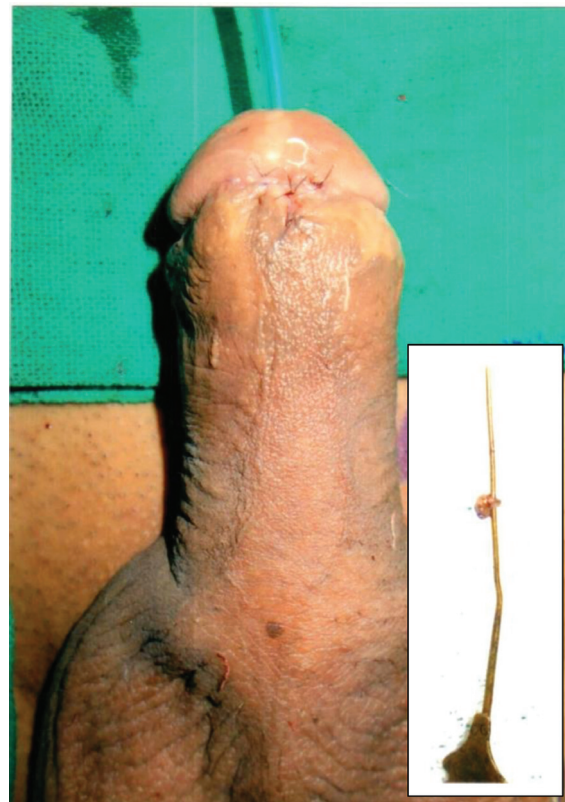


Fig. 3 Circum-excision of the fistula and its multilayered closure with 6-0 Vicryl (fistulorrhaphy). Excised ring of the urethrocutaneous fistula (UCF; inset).

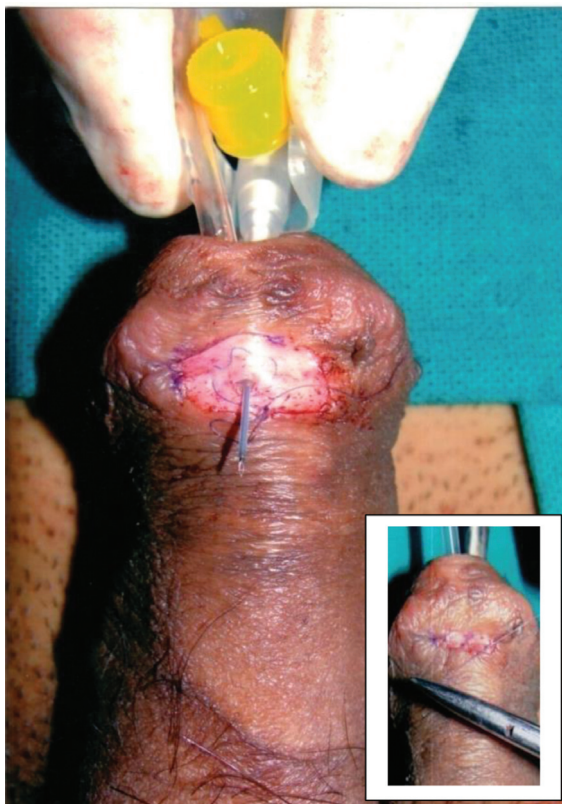


Fig. 2 Subcoronal urethrocutaneous fistula (UCF) excised. Purse-string suture inserted and tightened after removal of the cannula. Final closure is done transversally (inset).



Fig. 4 Right-sided penile skin flap designed. Flap sutured eccentrically on the left to prevent recurrences (inset).

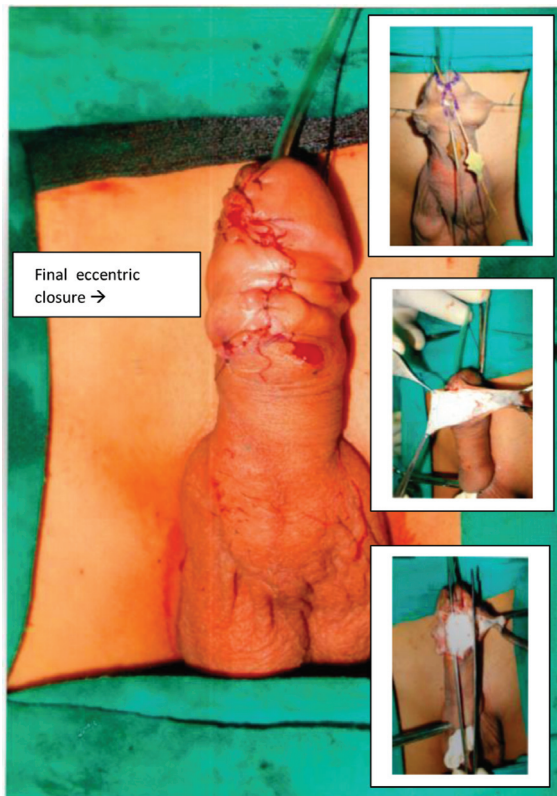


Fig. 5 Right preputial flap sutured on the left. Probed urethrocutaneous fistula (UCF; upper inset). Preputial dartos flap raised from the left skin (middle inset) and sutured on the right (lower inset).



Fig. 7 Scrotal and tunica vaginalis (TV) flaps. Proximal penile urethrocutaneous fistula (UCF) and left testis (upper inset). TV flap raised (middle inset) and sutured over closed UCF (lower inset).

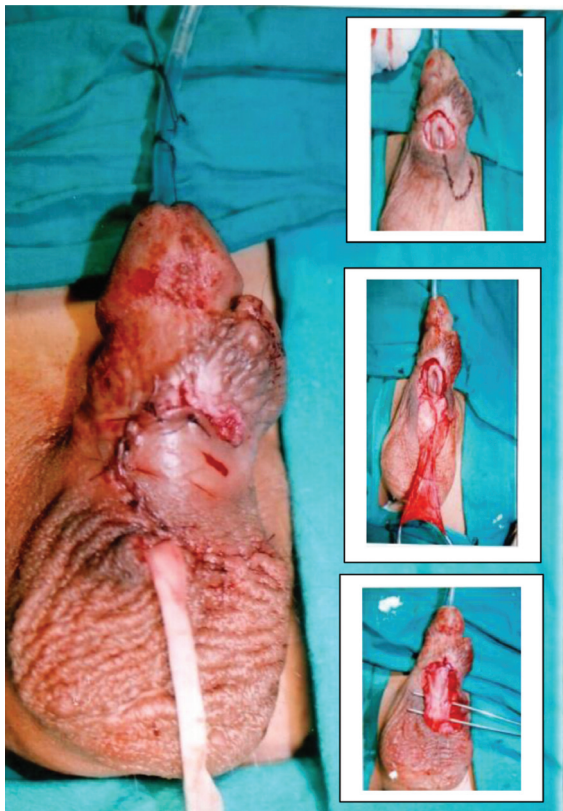


Fig. 6 Scrotal flap sutured on the right eccentrically. Urethrocutaneous fistula (UCF) and flap (upper inset). Loose scrotal dartos (middle inset) sutured over the fistula (lower inset).



Fig. 8 Mega-urethrocutaneous fistula (mega-UCF). Preputial hood and fistulorrhaphy (upper inset). Peno-preputial skin brought laterally (middle inset) and sutured on left (lower inset).

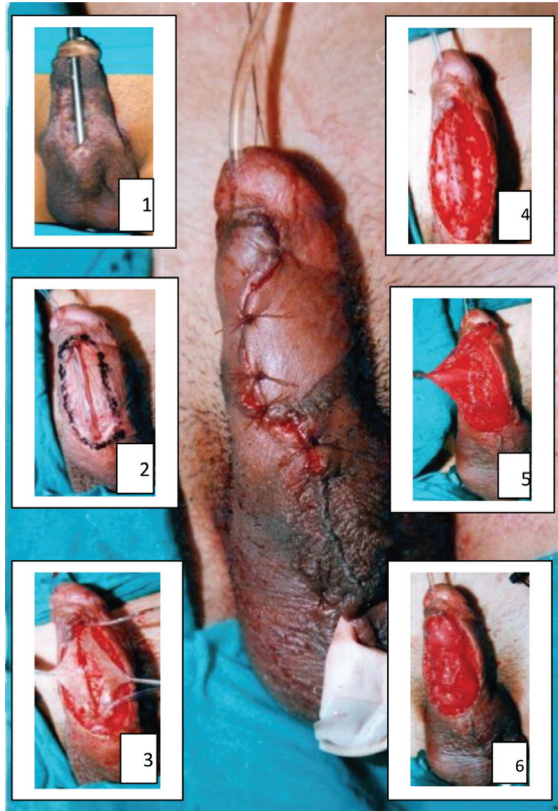


Fig. 9 Eccentric skin closure. (1) Near-total disrupted urethra. (2) Plate and urethrotomy. (3) Epithelial strips. (4) Tubularization. (5) Penile dartos (6) sutured eccentrically on the right.

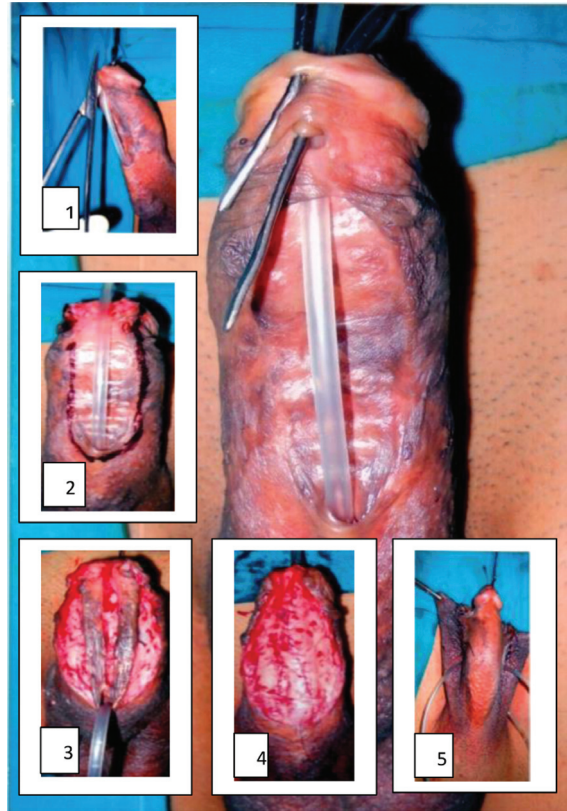


Fig. 11 Near-total disrupted urethra. (1) Intact glans cut. (2) Urethral plate. (3) Median urethrotomy. (4) Epithelial strips tubed. (5) Cecil-Culp done.



Fig. 10 Right penile skin dartos flap sutured on the left. Left penile skin sutured on the right (D. Smith technique) to prevent suture line superimposition (inset).

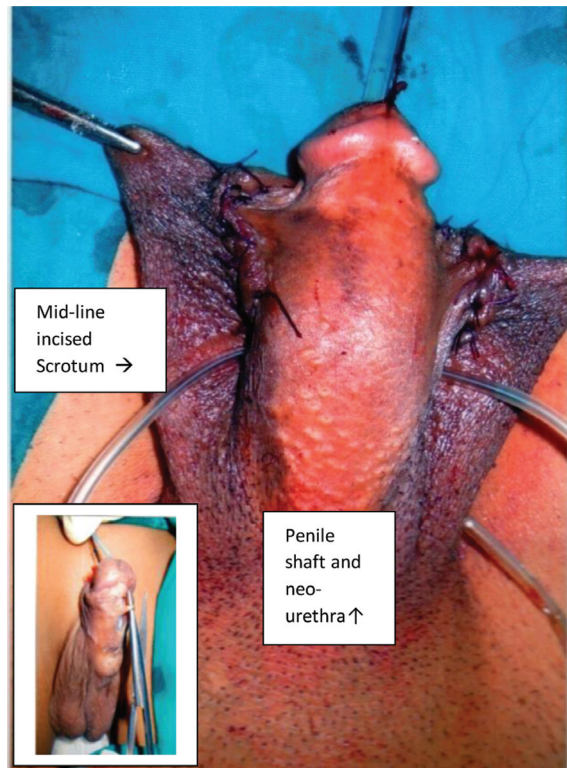


Fig. 12 Ventral penile shaft and neourethra sutured to midline incised scrotum (Cecil-Culp procedure). Multiple fistulas made one before repair (inset).



Fig. 13 Distal urethral stricture with proximal urethrocutaneous fistula (UCF). Laid open stricture and harvested inner preputial full-thickness skin graft (IPFTSG; upper inset). IPFTSG re-surfacing stricture site to construct urethral plate (lower inset).



Fig. 15 Ventral chordee on artificial erection. (1) Dorsal midline incision for dorsal tunica albuginea plication (DTAP). (2) Penile shaft straightened. (3) Final closure with drain.

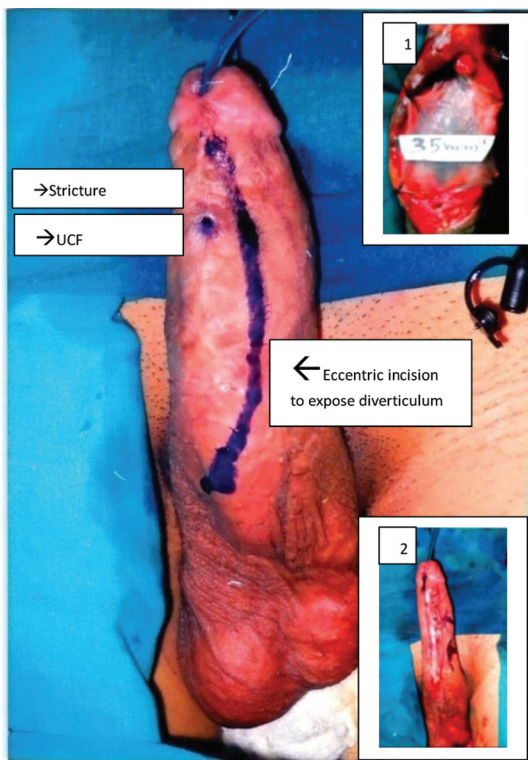


Fig. 14 Distal stricture, proximal diverticulum, urethrocutaneous fistula (UCF), and skin incision. (1) Wide diverticulum trimmed. Fistula excised, stricture incised, and grafted with trimmed lining. (2) Plate tubed and skin closed eccentrically.

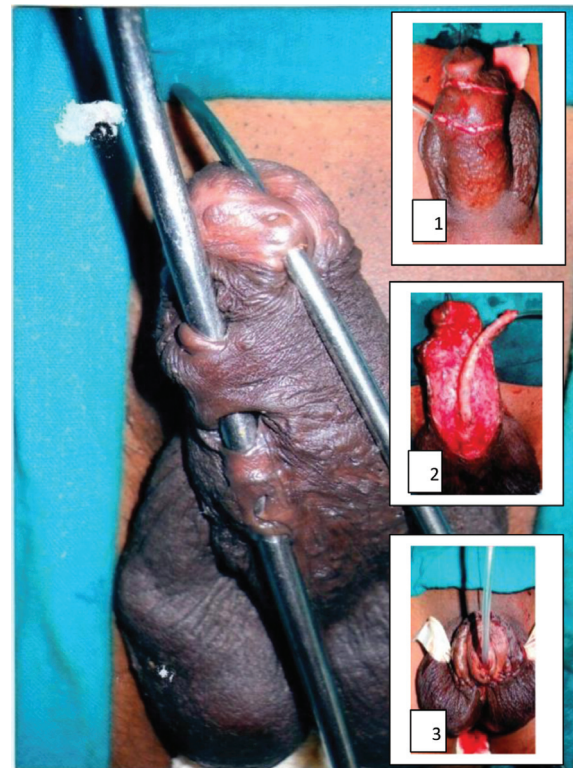


Fig. 16 Multiple urethrocutaneous fistulas (UCFs) and strictured plate. (1) Graft harvesting. (2) Scarred tissue and diseased plate excised, graft tubed, and anastomosed with native urethra. (3) Cecili-Culp procedure for cover.



Fig. 17 Balanitis xerotica obliterans (BXO) affected prepuce, urethra and fistula excised, and raw area covered by a buccal graft to make a plate. (1) Buccal graft from the right cheek. (2) Buccal graft.

graft (IPFTSG) to reconstruct the neourethral plate. The diverticulum was trimmed equal to the caliber of the urethra and the stricturotomy site was resurfaced by the trimmed lining of the diverticulum and the fistulas were excised. The concretion-affected urethra in one was excised and replaced with IPFTSG. Long, narrow, and strictured urethral plate in one case was replaced by tubed full-thickness skin graft. Excision of the BXO-affected part of the urethra and prepuce was done and replaced with the buccal mucosal graft in one case. Excision of scarred tissues and correction of a chordee by dorsal tunica albuginea plication (DTAP) was done in one case. The short urethra was transected in one case for doing subsequent interposition urethroplasty, but this patient was lost to follow-up. Tubularization of the neourethral plates was done after 3 months when tissues had matured to become soft and supple. The miscellaneous category F as detailed in **Table 1** was excluded from the present study.

Results

There was no recurrence of fistula in any of the patients in categories A, B, C-a, and C-b. One patient in category D had urinary leakage, which was closed successfully during the second stage of the Cecil-Culp procedure while separating

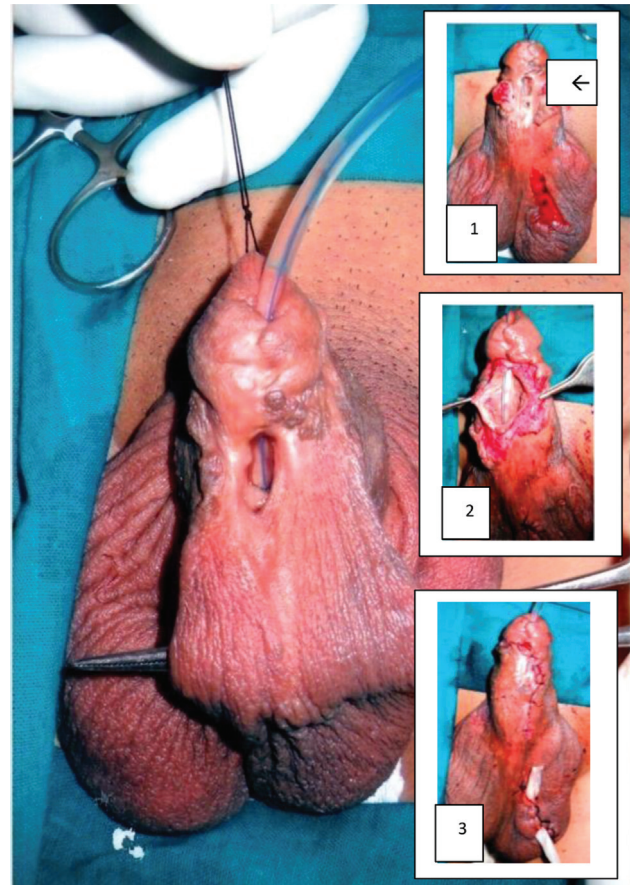


Fig. 18 Urethrocutaneous fistula (UCF) after a Cecil-Culp procedure. (1) Penis with UCF. (2) Circum-dissected fistula and median urethrotomy. (3) Multilayered closure of the fistula and scrotum.

the penile shaft from the scrotal attachment (**Fig. 18**). Urethral calibrations in follow-ups revealed normal-diameter urethra. One patient in category E had residual diverticulum and required further trimming of the diverticular wall equivalent to the dimensions of the urethra. Recurrent fistulas had successful closure after fistuloplasty. None of these patients had requested for any cosmetic correction. Five patients had reported for preputial reconstruction and the same was denied for want of adequate dispensable peno-preputial skin to reconstruct the prepuce. All had good penile erection without residual chordee. Three patients had come for getting fitness for marriage and were counseled accordingly to explain about their surgical procedures and potency to the parents of their would-be wives.

Discussion

Different authors have mentioned different characteristics of UCFs like the pediatric fistulas⁴; adult fistulas⁵; number and size of fistulas⁴; size, location, number, and nature of tissue surrounding the fistulas⁶; recurrent fistulas⁷; and fistulas associated with stricture and diverticulum.⁸ There is mention of different modes of treatment like conservative treatment,⁹ purse-string closure,¹⁰ multilayered closure,¹¹ simple closure, local skin flap closure or closure with

waterproofing interposition layer,⁶ and re-enforcement of closed fistula by TV flap⁷ and other soft-tissue re-enforcement interposition flaps (STRIFs)¹² or redo urethroplasty.¹³ Further, the UCFs have been divided into groups I, II, and III based upon their sizes, multiplicity, and the recurrent and persistent nature^{4,14} or the hypospadias patients have been assigned different groups as per their adopted treatment protocol.⁵ Different characteristics of UCFs have also dictated the treatment options and surgical outcome to some extent.⁶

A thorough literature search has revealed that UCFs developing after repair of male hypospadias have not been assigned any well-established classification of wide clinical use wherein due consideration had ever been given collectively to (1) all the different characteristics of UCFs; (2) associated morbidities of native and neourethral plates; (3) presence or absence of associated morbidities of native and neourethra like hairy urethra, stricture urethra, urethral diverticulum, urethral metaplasia, extensive perifistular scarring with secondary chordee, and the short reconstructed neourethra; (4) availability and dispensability of penile, preputial, and scrotal skins; and (5) the treatment plan opted for the given fistula. Little bit of literature is available about female hypospadias because of its rarity.¹⁵

The presented clinical classification of the UCFs has multiple merits. (1) It categorizes the UCFs into simple (categories A, B, C-a, and C-b) and complex (categories D and E) fistulas. Simple fistulas are those in which the native urethra, neourethra, native urethral plate, neourethral plates, and the peno-prepuccio-scrotal region are all normal and require simple treatment protocols like conservative treatment, tractotomy, purse-string closure, fistulorrhaphy, or fistuloplasty. Complex fistulas are those which alert the operating surgeon that either the diseased urethra would require correction or re-do urethroplasty or a secured cover to the repaired fistula would be required by doing Cecil-Culp procedure. (2) It incorporates almost all the characteristics of fistulas (single or multiple fistulas; simple or complex fistulas; ultra-micro, micro, macro, mega, near-total disruption of the urethra; acute or chronic fistulas; centric or eccentric fistulas; primary or recurrent UCF) that have direct bearing on choosing appropriate treatment planning. (3) It helps the treating surgeon to directly choose the appropriate treatment. (4) It helps in maintaining a detailed record of the characteristics of the fistulas at presentation, discharge, and follow-ups. (5) It will transfer detailed information of the UCF to the next surgeon just by mentioning the category and detailed diagnosis of the fistula while referring the post-hypospadias UCFs to a higher center for repair of recurrences. (6) Further, adherence to this clinical classification of UCFs will help in evaluating the results of different nonsurgical or surgical procedures attempted for the treatment of UCFs having similar characteristics or category. (7) Multicentric studies on the management of different categories of fistulas using different surgical techniques as per the expertise of the surgical team will give better evaluation of a particular

surgical technique for a specific category of fistulas and accordingly the surgeons would be able to alter their surgical planning in future to achieve better results.

Formation of complex urethrocutaneous fistulas (CUCFs) needs to be prevented by adopting the general principles of plastic surgery and repair of the fistula, because their subsequent correction could be difficult and time-consuming.

Conclusion

The classification of UCFs is of clinical significance and utility in research. Treatment planning should follow the principles of reconstructive ladder. Normal saline-induced artificial erection must be done to exclude the presence of secondary chordee. Complex fistulas respond better with a flap cover. An extragenital epithelium like the buccal mucosa must be considered where IPFTSG was not available to replace an unideal urethral plate. The Cecil-Culp procedure provides reinforcement of the fistulas by waterproofing the dartos flap and skin cover. Dispensable peno-preputial skin must be utilized to design the skin and soft-tissue flaps.

Conflict of Interest

None declared.

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