

Original Research Article

Side-to-side penoscrotal anastomosis: a reliable technique for repair of recurrent urethrocutaneous fistula in male children

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ABSTRACT

Background: Surgical repair of urethrocutaneous fistulae (UCF) is relatively simple and results are often satisfactory. However, in some cases of UCF, recurrence results despite several attempts at repair. Reports are scanty on the management of such recalcitrant UCF. The aim of the study was to present our experience with recurrent UCF, and describes our technique of side-to-side penoscrotal anastomosis for repair of such fistulae.

Methods: This was a 10 years analytical comparative study of the outcome of repair of recurrent UCF in paediatric patients using the simple double layer repair technique, and our technique of anastomosing the penis to the scrotum between 2008 and 2019 in our centre. Data obtained and analysed with SPSS 21 version included the number of previous attempts at repair, number, site and size of UCF, technique of repair, and the incidence of recurrence of UCF.

Results: Nineteen patients were studied. Nine had conventional double layer repair of UCF with recurrence in 5 (55.6%). Two of the recurrent cases were added to the remaining 10 patients to make a total of 12 cases who had the staged repair by penoscrotal anastomosis (PSA) and there was no recurrence during the average follow up period of 1 year. P value was <0.05.

Conclusions: Findings in this study suggest a superior outcome when the penoscrotal anastomosis is used for repair of recurrent UCF. We think it is better to apply this technique in cases of UCF which the surgeon considers potentially difficult than to wait to have a failed attempt before deploying it.

Keywords: Urethrocutaneous fistula, Recurrent, Penoscrotal anastomosis, Repair

INTRODUCTION

Urethrocutaneous fistula (UCF) is an abnormal communication between the male urethra and the skin. It is commonly acquired but occasionally occurs as a congenital condition.¹⁻³ The commonest causes of UCF are male circumcision, complication of urethroplasty, trauma and infective urethritis.⁴

UCF is among the most common complications of urethroplasty for hypospadias. Risk factors for occurrence of UCF following urethroplasty have been highlighted in previous studies.⁵⁻⁸ Many researchers have described

measures and techniques to reduce incidence of UCF. There is a report suggesting that hypospadias repair below the age of two years is protective against UCF, but there are no other reports to support this.⁹

Double staged repair in severe hypospadias is preferred by many surgeons because of the lower rate of UCF compared to single stage.¹⁰ Whereas a few of the fistulae can close spontaneously, the majority require surgical treatment to close. Surgical closure is a relatively simple procedure and results are often satisfactory.¹¹ However, in some cases of UCF repair, despite following the standard principles of urethroplasty, there is still recurrence. In patients whose

UCF have been repaired twice or more, chances of a subsequent recurrence is high. Most reports on management of UCF describe the primary treatment of UCF, but there are limited reports on the treatment and outcome of those cases which recur after one or two previous attempts at repair.^{5,12,13}

This study focused on the subgroup of UCF in which previous attempts at repair failed and fistula seems recalcitrant to those options of treatment. We preferred a technique which involved much less dissection and yet improved outcome.

The aim of the study was to present our experience with recurrent UCF, and to compare the outcomes of our technique of side-to-side penoscrotal anastomosis with the double layer techniques of repair of such fistulae.

METHODS

This was a comparative study of outcome of repair of recurrent urethrocutaneous fistula (RUCF) using the simple double layer repair, and our technique of anastomosing the penis to the scrotum. Ethical approval was obtained from our institutional review board and informed consent from the patient and or, the parents/guardian.

The null hypothesis of our study is that there is no improvement in outcome of repair of recurrent UCF using our side-to-side penoscrotal anastomosis compared to the conventional simple double layer repair. The alternate hypothesis is that there is improvement in outcome. Outcome measure was non-recurrence of fistula within the period of follow up. Between September 2008 and August 2018, we studied the outcome of treatment of paediatric patients who had recurrent UCF. The inclusion criteria were: male patients within the age range of 18 months and 18 years who had recurrent UCF. Exclusion criteria: age outside the study age range; congenital UCF, UCF without at least two previous failed repairs, refusal to participate, and incomplete data.

Our novel technique involved two stages and were either performed or supervised by the same surgeon; the first stage involved a single layer closure of the fistula over a urethral catheter with inverting 6-0 delayed absorbable sutures after dissecting the skin away from the fistula. An incision is then made on the scrotum with similar dimensions as those of the fistula repair site on the penis. The exposed scrotal subcutaneous tissues and dartos fascia are then attached to the fistula repair site to provide a second layer coverage for the repair. A skin-to-skin anastomosis is then performed between the penis and the scrotum to achieve a side-to-side anastomosis. With healing by the 10th day, the urethral catheter was removed.

The second stage involved simple detachment of the penis from the scrotum and skin closure of the penile and scrotal wounds three to six months later. Data obtained and

analyzed included patients biodata, the etiology of the fistula, the institution where fistula first occurred, the number of previous attempts at repair, number, site and size of UCF, technique of repair, and the incidence of recurrence of UCF following repair by the conventional two-layer technique, and the penoscrotal anastomosis during a mean follow up period of one year.

During follow up specific questions were asked about splashing of urine on the legs and painful erections. Data was analysed with the SPSS 21 version.

RESULTS

Eighty-seven cases of UCF were seen during the study period and of these, 21 patients were diagnosed with recalcitrant urethrocutaneous fistula but 19 met the inclusion criteria and were studied.

The age range of the patients was 2.5 to 12 years with a median 5.2 years. The associated surgical conditions leading to the fistula are shown in Table 1.

Table 1: Surgical conditions leading to fistula in patients.

Etiology	Patients (N=19)	Percentages (%)
Urethroplasty for hypospadias	10	52.6
Genitoplasty for ambiguous genitalia	3	15.8
Complication of circumcision	6	31.6

Eight (42.1%) of the RUCF occurred ab initio in our centre and previous attempts at repair were also in our centre, whereas 11 (57.9%) were referred from other centres and 7 had had attempts at repair in those centres. The specific cause of the fistula was not identified in the 11 patients referred from outside our centre.

For the 8 cases which occurred in our centre, 4 were due to surgical site infection, 3 due to premature catheter blockage and 1 due to meatal stenosis. The number of previous attempts at repair is shown in Table 2.

Table 2: Number of previous attempts at repair.

Number of previous attempts at repair	Patients (N=19)	Percentages (%)
2	11	57.9
3	5	26.3
>3	3	15.8

The fistulae were all solitary and size ranged from 3-15 mm. The fistulae were located in the midline along the ventral aspect of the penis in 12 (63.2%) cases whereas 7 (36.8%) cases were located to the left or right of the midline. The site of the RUCF are shown in Figure 1.

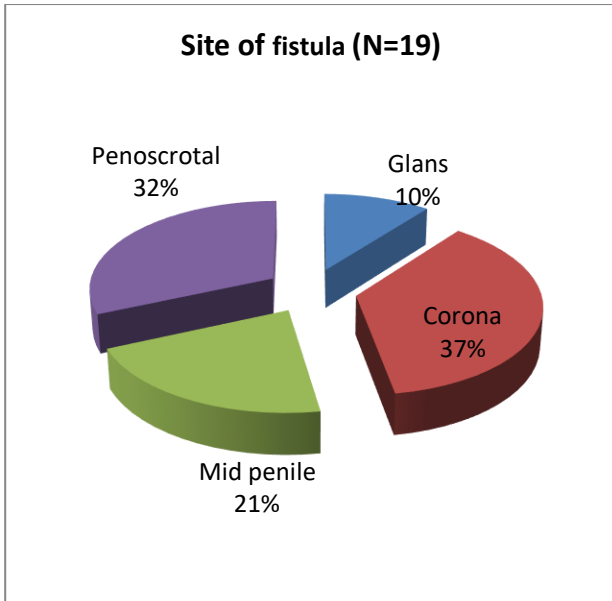


Figure 1: Distribution of patients according to site of fistula.

Of the 19 patients studied, 9 (47.4%) had conventional double layer repair of UCF, 10 (52.6%) had the staged repair by PSA (Figure 2).

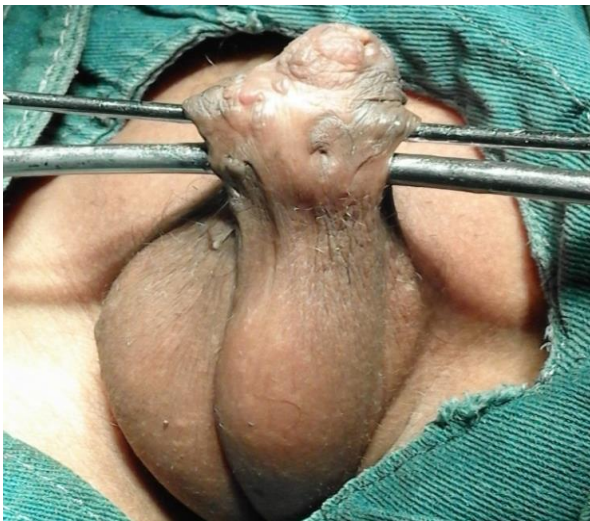


Figure 2: Penoscrotal anastomosis ready to be detached.

Of the 9 cases repaired by simple double layer technique, 8 were studied retrospectively and 1 case prospectively. There was recurrence of fistula in 5 of these 9 patients giving a recurrence rate of 55.6%. Two of the five cases of recurrence were included in the group for staged repair with penoscrotal anastomosis making a total of 12 cases repaired by this technique. These 12 cases were studied prospectively and 10 healed primarily while 2 had a transient leakage which closed in 2 and 4 weeks respectively. All fistulae were closed at the time of detachment of the scrotum 3-6 months later (2nd stage) (Figure 3 and 4).

Patients were followed up for an average period of one year. ($p < 0.05$). There were no recurrences or strictures (Figure 5).



Figure 3: Intra-operative detachment of penis from the scrotum.



Figure 4: Immediate post detachment with skin closure.



Figure 5: Common outlook on follow up.

DISCUSSION

UCF is a fairly common complication encountered in paediatric urethral surgeries in our service. Repair of these fistulae is generally easy, but same cannot be said of the repair of recurrent fistulae. The recurrence rates of these fistulae tend to increase with each failed repair, rising up to more than 50% as seen in this report. This is expected because of the increasing scar tissue and less vascularity of the tissues around the fistula. There are very few reports in literature of specific techniques to address such difficult cases of recurrent UCF.

Tokgoz et al described their technique in a single case report in which they used a transverse turn over flap of Buck's fascia of the corpus spongiosum to repair a problematic resistant UCF.¹⁴ However, they had to insert a suprapubic catheter, which was an additional morbidity, to achieve successful fistula closure. Another report described the use of a waterproof layer of tissue to cover the repair suture line before skin coverage.¹⁵ They had achieved 100% success in repair of recurrent UCF. This technique was not easy to apply to our set of patients all of whom had had recurrence more than once. Moreso, the surrounding tissues were so scarred, the waterproof layer was mostly unavailable. We anticipate similar challenges in the 'pants-over-vest' technique of repair of UCF described by Cimador et al.¹⁶

In their report they had performed triple layered procedure using double overlapping flaps to repair UCF with excellent results. However, they did not test the usefulness of their technique in cases of UCF which had recurred repeatedly. We are concerned that the extensive scars and poor vascularity in this situation may make the procedure more tedious and outcome less attractive. In another report, a scrotal septal flap was used to repair a wide UCF.¹⁷

This technique seems quite good but technically difficult for UCFs in the distal part of the urethra. The situation of increased scarring and poor peri-fistula blood supply and high potential of re-fistulation was what inspired us to consider this novel technique. The principle of this technique is similar to the Cecil Culp procedure as both take the advantage of the generous blood supply of the dartos and scrotal skin. However, the Cecil Culp procedure is a more extensive surgery in which the penis is buried in the scrotum as part of the repair of hypospadias. It is a far more extensive procedure than our technique.¹⁸ In search of improvement in the outcome, Weiss et al modified the Cecil Culp procedure leaving the penis attached to the scrotum for up to a year rather than the six weeks originally prescribed for the Cecil Culp procedure. They achieved some improvement in outcome but still had significant complications in more than 30% of their cases.¹⁹ The incorporation of virtually the entire length of the penile shaft meant more extensive dissection, and could explain the incidences of complications including scrotal abscess in their series. Routh et al, and Pescheloché et al working

independently reported their experiences with the use of tunica vaginalis flap for repair of recurrent UCF. They reported excellent results but the procedure involved extensive dissection with mobilization of flap.^{20,21} Such dissection in which the testis and adjoining structures are exposed during the procedure can put those structures at risk of injury. More so, the more distal fistulae are bound to be more technically demanding.

We relate the remarkably improved outcome of our technique of repair to the better alternative blood supply from the scrotum and more tissue available for second layer repair without tension or stenosis of the urethra, yet with limited dissection both on the penis and on the scrotum. A similar outcome of 100% successful repair was reported by Ehle et al using the Cecil Culp technique. However, of the 15 boys studied in their series, only 6 (40%) had had 2 or more previous attempts at repair unlike the present study in which all the patients had had at least two previous attempts at repair.²²

We recognize the two staged nature of this technique as a limitation, but we still consider it a better option than the recurrence which was likely to occur in those situations. The temporary distortion of appearance of the genitalia following the anastomosis was some concern for some of the patients and their parents. However, all of them accepted it for the permanent correction of their fistula. We had anticipated that the anastomosis will cause the penis to point downwards and cause patients to wet their legs while urinating. This was rather not the case as we observed most of them urinate with the stream straight and into the toilet bowl. Erection was also observed by parents to be normal and painless both during the period of the anastomosis and after detachment of the penis and follow up. The fact that this technique does not require suprapubic cystostomy, can be used even with extensive scarring around the fistula, and there is no extensive dissection or raising of flaps, makes it attractive compared to other techniques.

CONCLUSION

This study evaluated the outcome of treatment of UCF which had had at least two previous attempts at repair using the novel technique of penoscrotal anastomosis. We recognize the small volume of data as a potential drawback. A larger series is required to further corroborate or counter the findings in this study. However, findings in this study point to a superior outcome with respect to recurrence of fistula when our technique of side-to-side penoscrotal anastomosis is used compared to the conventional double layer repair in the treatment of recurrent UCF. We think that it may be better to apply this technique in cases of UCF which the surgeon considers potentially difficult than to wait to have a failed attempt before deploying it. Such potentially difficult UCF may include previous failed repair, associated extensive scarring, wide fistulae, prior circumcision before urethroplasty with inadequate skin, prior associated urethral stricture etc.

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