



Single-stage salvage urethroplasty for failed distal hypospadias: comparison of Mathieu and grafted tubularised incised plate repairs

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Abstract

Aim Distal hypospadias repair is a common operation but some children require a complete salvage operation due to major complications of initial repair. The aim of this paper is to analyze the results of single-stage salvage distal hypospadias operations with salvage Mathieu (SM) and salvage grafted tubularised incised plate (SGTIP) techniques.

Patients and methods A retrospective review of 115 children from 2 centers who underwent SM or SGTIP operations for failed distal hypospadias from January 2012 to July 2021 was conducted for demographic data, success of salvage operation and complications. Children with scarred urethral plate but with healthy, mobile ventral skin underwent SM operation, while children with a supple urethral plate of width 6 mm or greater underwent SGTIP operation. Twenty-four (21%) children with pre-operative glans width < 12 mm received pre-operative intramuscular testosterone injections (2 doses, 2 mg/kg/dose). In the SM group, 22 cases with shallow, scarred glans underwent distal glans augmentation using the terminal portion of the Mathieu skin flap. Pre-operative and intra-operative parameters and post-operative results were compared between SM and SGTIP techniques. Statistical analysis was done using software; student *t* test and Fishers exact test were used, and *p* value of < 0.05 was considered significant.

Results During the study period, SM and SGTIP operations were performed in 84 and 31 children, respectively. Overall, at a median follow-up of 24 (3–72) months, the salvage operation was successful in 101/115 (87.8%) patients; complications were observed in 14/115 (12.2%). The SM and SGTIP groups were comparable with respect to the number of prior failed operations, mean age, mean length of the distal urethral defect, pre-operative testosterone use and use of dorsal plication for chordee correction. SM had significantly better success (77/84, 91.6%) than SGTIP (24/31, 77.4%, *p* = 0.044). In the SM group, urethrocutaneous fistula (UCF) was noted in 2/84 (2.3%) while this was significantly higher (*p* = 0.01) at 5/31 (16.1%) in SGTIP. No patient in SM group developed meatal/urethral stenosis while this was significantly higher (*p* = 0.0003) at 6/31 (19.3%) in SGTIP; of these, 4 children had both UCF and meatal/urethral stenosis. A higher incidence of repair breakdown/dehiscence was seen in SM group, but the difference was not statistically significant (SM 5/84, 5.9% versus SGTIP 1/31, 3.2%, *p* = 0.484). In the SM group, two fistulae were successfully repaired after 6 months, while the three cases of complete dehiscence underwent two-stage oral mucosal graft (OMG) repair. In the SGTIP group, two children underwent successful repair of UCF (one along with meatoplasty), one child with stenosis improved with urethral dilatations, while four children (three with UCF and stenosis and one with complete dehiscence) were managed with two-stage OMG repair.

Conclusions Single-stage salvage distal hypospadias repair was successful in 87.8% of patients. Both SM and SGTIP demonstrated acceptable results for salvage distal hypospadias repair. SM had better success and lower complications than SGTIP. SGTIP was associated with significantly higher post-operative UCF and meatal/neourethral stenosis.

Keywords Hypospadias · Distal · Salvage · Mathieu · Tubularised incised plate · Grafted

Introduction

Distal hypospadias repair is a common operation in pediatric urology, yet it is associated with significant complications and reoperations even in expert hands [1–3]. While urethrocutaneous fistula (UCF) is still the commonest

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complication after distal hypospadias repair, a coronal UCF after distal hypospadias repair with distal glans bridge may present a unique problem that may warrant a complete re-do repair. Reoperations needed for other complications such as neourethral stricture and glans dehiscence are also typically difficult, due to an altered urethral plate from prior surgery, a scarred and sometimes small glans, absence of prepuccial skin and scarring from previous surgery.

The management of salvage hypospadias is controversial with different techniques used; the two commonly reported techniques for salvage distal hypospadias repair are Mathieu and tubularised incised plate (TIP, with or without dorsal inlay graft) operations. We prefer grafted TIP (GTIP) to TIP, as it has been shown to have better results than TIP in re-operative cases [4]. In this paper, we compare the results of salvage Mathieu (SM) and Salvage GTIP (SGTIP) techniques for failed distal hypospadias repair. To our knowledge, there has been no previous report comparing the results of these two techniques for distal hypospadias reoperations.

Methods

This was a retrospective review of all children who underwent salvage Mathieu (SM) or salvage grafted tubularised incised plate (SGTIP) procedures for failed distal hypospadias from January 2012 to July 2021. The children who underwent two-stage salvage operations, and children with failed proximal hypospadias are not included in the present study. Our general protocol for selecting the type of operation for salvage distal hypospadias repair without the availability of foreskin is given in Fig. 1. The three types of complications requiring salvage repairs are given

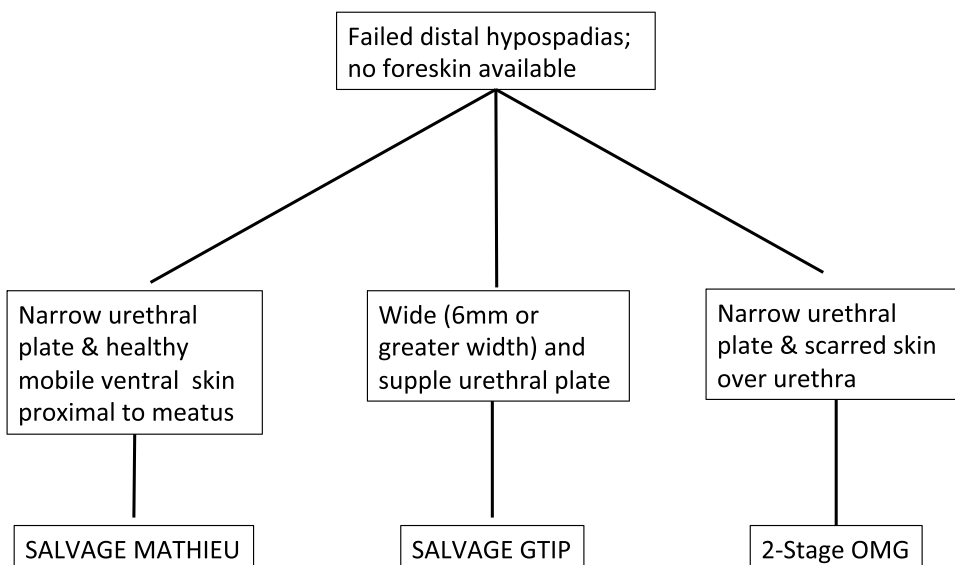
in Fig. 2. Two senior pediatric urologists from two centers performed all the procedures. Children with pre-operative glans width < 12 mm received two doses of intramuscular testosterone injections (2 mg/kg/dose) before the salvage repair, similar to our protocol for primary hypospadias repairs. Reoperation was performed at least 6 months after the previous failed operation and 4–6 weeks after testosterone injection.

Surgical technique: salvage Mathieu operation (Fig. 3)

The fundamental criterion to choose SM procedure was presence of healthy, supple and mobile ventral penile skin in the midline that could be safely elevated off the underlying urethra. Typically, these children also had a narrow and scarred urethral plate from prior surgery, which was considered unsuitable for GTIP technique. The presence of poor quality, scarred ventral midline skin and the presence of severe chordee (> 30°) were considered contraindications to the SM procedure.

The first step of the operation was to perform an artificial erection test for chordee. Mild chordee (< 15°) is typically corrected by ventral penile skin degloving, while dorsal midline plication for chordee correction was required in 8/84 (9.5%) patients with moderate (15–30°) chordee. In children with large coronal fistula and a glans bridge (Fig. 2a) or distal urethral stenosis (Fig. 2b), the ventral glans was opened in the midline to convert the case into a distal hypospadias. Mathieu flip flap was marked and developed, keeping the perimeatal base wide and keeping all the subcutaneous dartos tissue attached to the flap. Care was taken not to injure the underlying urethra. The length of the flap was kept about 0.5 cm longer than the length of the distal urethral defect.

Fig. 1 Selection of technique for salvage distal hypospadias repair



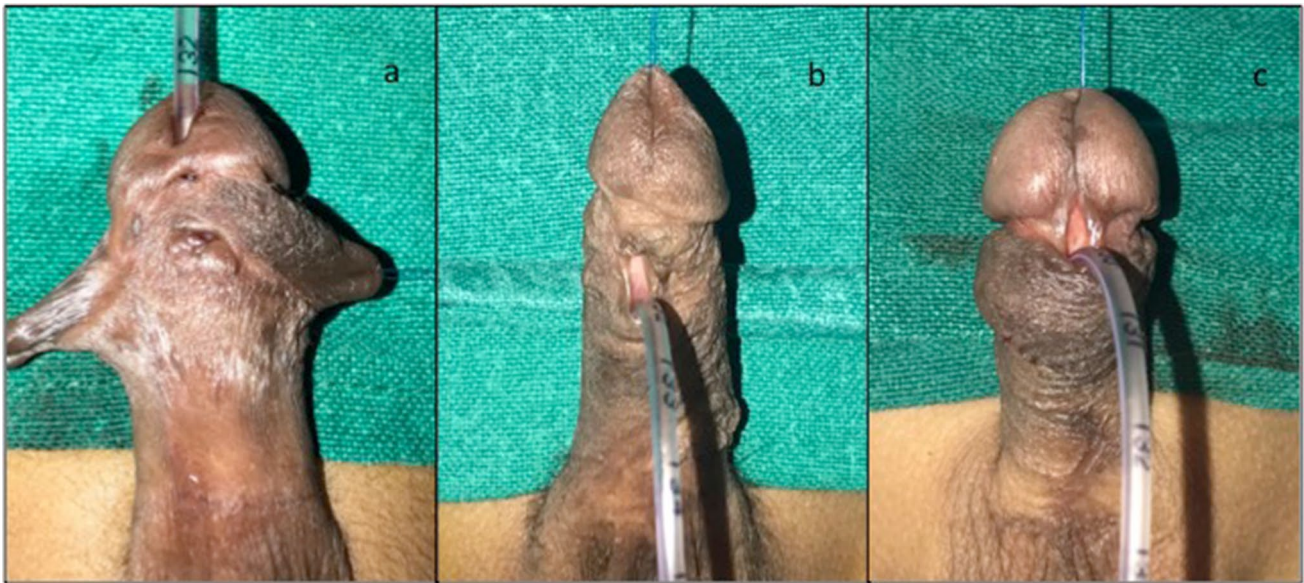


Fig. 2 Complications of distal hypospadias repair requiring complete salvage repair. Large fistula (2 fistulae in this case) with distal glans bridge (a); complete stenosis of reconstructed distal urethra with subcoronal fistula (b); complete dehiscence of repair (c)

Glans wings were developed widely, while excising the badly scarred edges on the glans to the extent possible. The mobilized Mathieu flap was sutured to either side of the urethral plate over a 7–8 F infant feeding tube using running 7/0 polydioxanone (PDS) suture. The suture line on either side was covered with the mesentery of the flap (dartos) using interrupted 7/0 PDS sutures. Glansplasty was performed in two layers, inner 6/0 PDS and epithelial 7/0 PDS stitches. Where possible, the MAVIS modification [5] was used to achieve a natural glanular meatus. In 22/84 (26%) cases with shallow, scarred glans where glans approximation over the reconstructed urethra was found to be tight, the distal glans augmentation technique was used. For this, the excess 0.5 cm of the Mathieu flap was flipped backwards and its epithelium was sutured using interrupted 7/0 PDS sutures to the distal glans edges. The edges of proximal glans at the corona were approximated in the midline with a single 6/0 PDS suture for the glans tissue and 7/0 PDS sutures for glans epithelium. The mucosal collar and penile skin closure was achieved in the most suitable way possible with interrupted 6/0 or 7/0 PDS sutures to give an acceptable cosmetic result.

Surgical technique of salvage GTIP operation (Fig. 4)

Salvage GTIP was chosen when the urethral plate was wide (6 mm or greater) and supple without excess scarring. Similar to the SM technique, presence of severe chordee ($> 30^\circ$) was a contraindication for the SGTIP procedure. Correction of mild and moderate chordee is done as described earlier for the SM operation; dorsal midline plication was performed in five (16%) children. The urethral plate was marked and

incised, and glans wings were developed. The urethral plate was then incised longitudinally in the midline and the resulting defect was covered with an inlay free graft of preputial/penile skin (8 cases) or oral mucosa (OMG, 23 cases). The graft was trimmed to the required size and sutured to the inner margins of the incised urethral plate with 7/0 polydioxanone (PDS) stitches. Quilting stitches of same suture were applied fixing the graft firmly to its bed to avoid graft extrusion or fluid collection beneath the graft. A two-layer urethroplasty was performed with continuous subepithelial 7/0 PDS suture. The suture line was covered by local dartos flap or tunica vaginalis flap. Glansplasty was done in two layers with 6/0 PDS stitches for glans tissue and 7/0 PDS for epithelial sutures. Skin closure was done in the most cosmetically acceptable manner.

In both SM and SGTIP procedures, the aim was to create a neourethra of at least 10–12 F caliber. A 7 F or 8 F infant feeding tube was used as the catheter to drain the bladder into diapers. Suprapubic diversion was not used in any child. A sandwich dressing was applied using Tegaderm, sterile gauze and sticking plaster. The child was discharged after 24 h on oral antibiotics, analgesics and Oxybutynin. The dressing was removed after 1 week and the catheter was removed after 10–14 days in the outpatient clinic. The child was followed in the clinic after 1 month, 3 months, 6 months and yearly thereafter (Fig. 5).

Both the SM and SGTIP groups were compared for mean age, mean length of urethral defect, chordee, and post-operative results (success of salvage repair, fistula, meatal stenosis and glans dehiscence). Success of the operation was considered when the child voided from



Fig. 3 SM operation. Mathieu flap outlined, 0.5 cm longer than distal defect (a); flap sutured to urethral plate to construct neourethra (b); suture line on either side covered by mesentery of flap (c); completed

repair showing glans reconstruction with distal glans augmentation. Note terminal part of the Mathieu flap has been turned back and sutured to the distal glans edges (d); result after 1 month (e)

the terminal neomeatus without straining or UCF. Post-operative urethral calibration was not routinely practiced. In children with straining or difficulty in voiding, uroflow study and ultrasound examination were performed; in case of suspected meatal/neourethral stenosis, the child was subjected to urethral calibration under anesthesia. For tight meatal stenosis, meatotomy/meatoplasty was performed; severe neourethral stenosis not responding to dilatations was managed by laying open of the neourethra with staged OMG graft reconstruction later. Any complication requiring re-intervention was considered a failure. Simple UCF without associated distal stenosis were closed surgically after 6 months. UCF associated with distal stenosis were dealt with appropriately; typically, the stenosis was managed before UCF closure. Statistical analysis was done using software; student '*t*' test and Fisher's exact test were used where appropriate, and '*p*' value of <0.05 was considered significant.

Results

One hundred and fifteen children who underwent salvage distal hypospadias repair were included in this study; SM and SGTIP techniques were performed in 84 and 31 patients, respectively. Most (103, 90%) previous repairs had been performed in other centers; only 12 patients (10%) had undergone prior failed hypospadias surgery in our centers. In the majority (96/115, 83%) of cases, the primary failed operation was a TIP repair, in 8 cases, it was a meatal advancement procedure, and in 11 cases, the primary operation was unknown. Eighty-seven (76%) patients had one prior operation, while 28 (24%) patients had 2 prior operations; in most cases, the second operation was an unsuccessful UCF closure. Of the 115 children, 53 presented with a large coronal UCF with a thin distal glans bridge, 45 with complete breakdown of prior repair and 17 with severe stenosis of the distal urethra, after 1 or 2

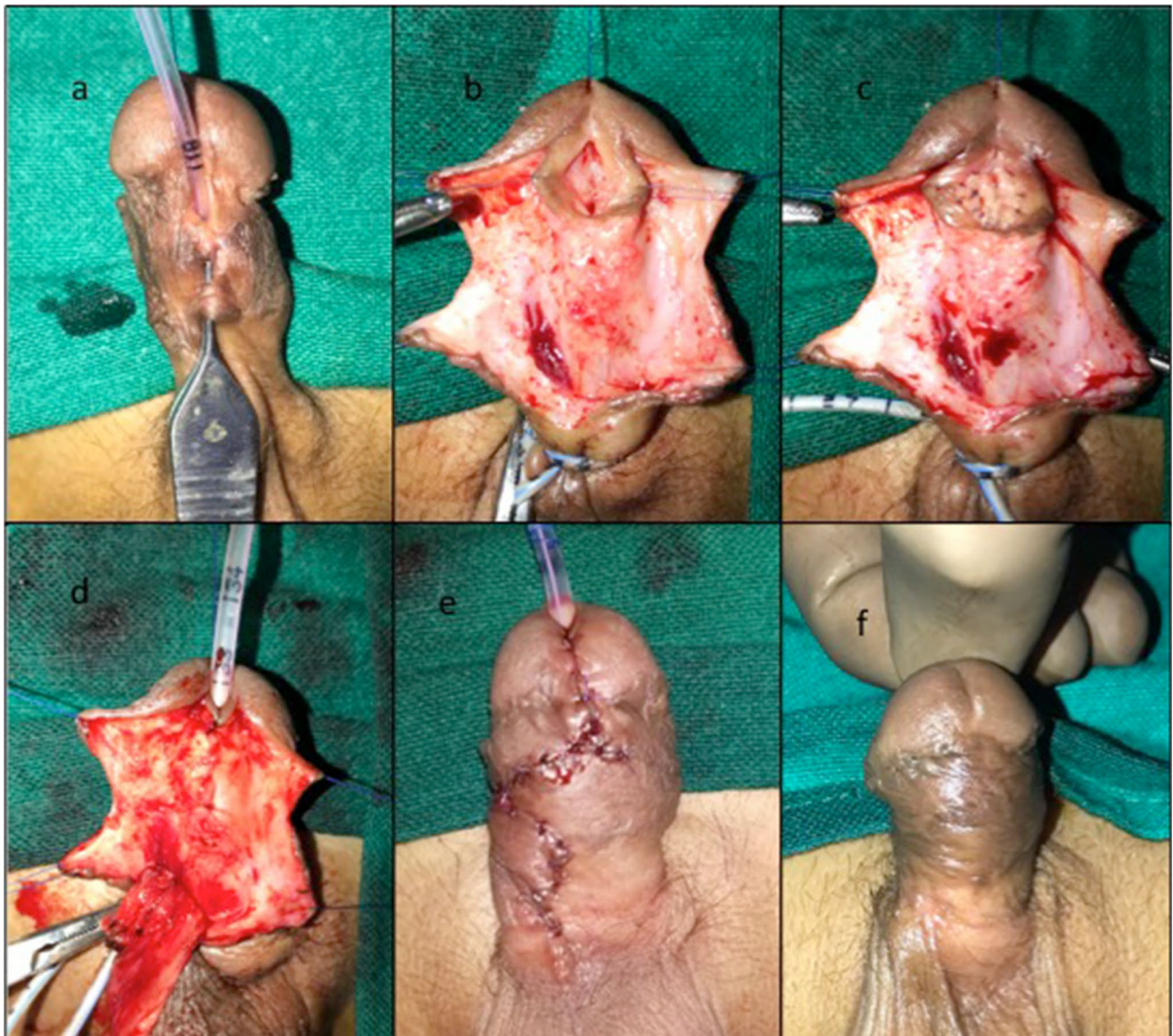


Fig. 4 SGTIP operation. Complete dehiscence of primary repair with poor quality ventral skin (a); glans wings raised and deep midline incision made in urethral plate (b); oral mucosal inlay graft quilted

into the midline defect in the urethral plate (c); urethroplasty completed, tunica vaginalis flap ready to cover the suture line (d); complete SGTIP (e); result after 6 months (f)

prior failed attempts of distal hypospadias repair (Fig. 2). Twenty-four (21%) children with pre-operative glans width < 12 mm received 2 intramuscular injections of testosterone (2 mg/kg/dose) 4–6 weeks before the salvage operation. The demographic data of the patients are given in Table 1. Both groups were comparable with respect to the number of prior failed operations, mean age, mean length of the distal urethral defect, proportion of children who received pre-operative testosterone and proportion of children who required dorsal plication for chordee correction. Overall, the salvage operation was successful in 101/115 (87.8%) of patients; complications were observed in 14/115 (12.2%).

Table 2 depicts the comparative results of the two techniques. At a median follow-up of 24 (3–72) months, SM was successful in 77/84 (91.6%) cases, while SGTIP was successful in 24/31 (77.4%) cases. Thus, SM had significantly better success than SGTIP ($p=0.044$). In the SM group UCF was noted in 2/84 (2.3%) while this was significantly higher ($p=0.01$) at 5/31 (16.1%) in SGTIP. No patient in SM group developed meatal/urethral stenosis while this was significantly higher ($p=0.0003$) at 6/31 (19.3%) in SGTIP. Of these, four children had both UCF and neomeatal/urethral stenosis. A higher incidence of repair breakdown/dehiscence was seen in SM group, but the difference was not statistically significant (SM 5/84, 5.9% versus SGTIP



Fig. 5 SM with MAVIS modification, result after 1 year

Table 1 Demographic data of both groups

Parameter	Mathieu	GTIP	<i>p</i> value
Number of patients	84	31	
Mean age y (sd)	4.7 (3.6)	5.4 (3.9)	0.73
Mean length of urethral defect mm (sd)	14 (3.6)	15 (4.1)	0.46
Prior failed operations (1/2)	61/23	26/5	0.22
Pre-operative testosterone injection	15	9	0.14
Dorsal plication for chordee correction	8 (9.5%)	5 (16%)	0.34

1/31, 3.2%, $p = 0.484$). In the SM group, two fistulae were successfully repaired after 6 months, while the three cases of complete dehiscence underwent two-stage OMG repair. Nothing further was done in the two children with distal glans dehiscence and meatal retraction. In the SGTIP group, two children underwent successful repair of UCF (one along with meatoplasty), one child with stenosis improved with

urethral dilatations, while four children (three with UCF and stenosis and one with complete dehiscence) were managed with two-stage OMG repair. The parents of all children with successful result in both groups were satisfied with the post-operative cosmetic appearance of the penis.

Discussion

The repair of failed hypospadias is a challenge, especially when the entire operation has to be repeated for a major complication, which may be the case in upto 5–10% of children after distal hypospadias repair [1–3]. Although there are individual case series of SM [6–8] and SGTIP [9, 10] techniques published for salvage distal hypospadias repair, to our knowledge, this is the first report comparing these two techniques. Our results demonstrate that both techniques are suitable for the reoperation, with SM demonstrating better success and fewer complications than SGTIP. Our overall success of 87.8%, although inferior to some reports of primary distal hypospadias repair [5, 11], is, however, comparable to other reports on salvage hypospadias repair [6–10, 15].

There are important differences between the SM and SGTIP techniques; although both these techniques are based on augmenting the urethral plate, SM uses a vascularized skin flap as ventral onlay, while SGTIP uses a dorsal inlay graft. Thus, the cases with healthy skin on the ventral aspect of the penis were selected for SM, while the cases with a supple urethral plate of reasonable width were selected for SGTIP. Assuming that the only other choice in such cases would be a two-stage OMG repair, SGTIP seems to be a reasonable single-stage salvage operation for such cases; by the use of SGTIP, over three-fourths of such cases have had a successful single-stage reconstruction. Similar observations were made by other authors [9, 10] who proposed single-stage SGTIP as a reasonable alternative to staged reconstruction for complex hypospadias reoperations.

To our knowledge, our series of SM is the largest reported till date, and it has been our preferred technique for single-stage salvage operation of failed distal hypospadias. The other authors also reported good results with SM in failed distal hypospadias [6–8]. Notably, the number

Table 2 Success and complications in both groups

Parameter	Mathieu (84)	GTIP (31)	<i>p</i> value
Success (%)	77 (91.6)	24 (77.4)	0.044
Complications (%)	7 (8.4)	7 (22.6)	0.044
Fistula (%)	2 (2.4)	5 (16.1)	0.015
Meatal/neourethral stenosis (%)	0 (0)	6 (19.3)	0.0003
Partial/complete glans breakdown (%)	5 (5.9) [2 partial, 3 complete]	1 (3.2) [complete]	0.484

of prior failed operations may be important in the success of SM operation. Bar-Yosef et al. [6] reported that SM operation was successful in 74% of 34 children, of whom 10 were prior failed TIP operations; the patients with multiple prior failed operations had only 66% success, while the success was 87% in those with a single prior failed operation. Karabulut et al. [7] observed that the success of SM decreased as the number of prior failed operations increased. They reported 57 cases of SM with 84% success; however, when patients with only 1 prior failed operation were analyzed, the success was 91.9%. The authors reported a significant negative correlation ($p=0.025$) between the number of previous operations and the success of SM urethroplasty. Hayashi et al. [8] reported 92% success with SM operation in 13 children who had undergone only 1 prior failed operation. These results are comparable to our success of 91% with SM in the present series, which consists of patients who had one or two prior failed operations only. We agree with the other authors that after multiple failed operations, the urethral plate and penile skin are typically of poor quality and may not be suitable for a successful single-stage repair. In our practice, such children with multiple prior failed operations typically receive a two-stage OMG reconstruction.

In 22 cases of SM where there was a difficulty in closing the glans over the Mathieu flap, we utilized the distal glans augmentation technique, with good results. A thick mesentery of the Mathieu flap, and a flat glans with significant scar tissue needing excision, are the major contributors resulting in tight glans closure. In these situations, the use of distal glans augmentation has helped avoid the complications associated with a tight glans closure; there was no case of post-operative meatal/neourethral stenosis in the SM group. Nezami et al. [12] earlier reported glans augmentation during Mathieu operation for primary hypospadias with 97% success. They utilized glans augmentation in patients with shallow urethral grooves and concluded that this technique eliminated the tension on glans wing sutures, thus reducing the risk of subsequent neourethral break down. Our results demonstrate that glans augmentation is a useful adjunct during SM operation, and it helps to avoid the potential complications arising from tight glansplasty. The only disadvantage of glans augmentation may be creation of a round meatus sitting on top of the glans, which is most evident in patients with a flat glans; in our experience, this minor cosmetic aberration is noted more by the surgeon than by the parents.

Some authors reported onlay island flap procedures for salvage hypospadias repairs [13], and compared SM and salvage onlay flap procedures. SM comprised the majority (70%) of their salvage procedures; the authors stressed appropriate case selection as the key to success [13]. In the present series, most patients had had a dorsal dartos flap (to cover the TIP repair) with circumcision during the primary

hypospadias repair; so prepuce or dorsal skin for use as island flap was rarely available for the salvage operation.

About 2 decades ago, while reporting the results of Mathieu operation for salvage hypospadias repair, the authors predicted that TIP repair would replace Mathieu technique for salvage operations [14]. However, although there have been several reports of TIP urethroplasty for secondary hypospadias, a meta-analysis [15] reported an overall complication rate of 21.8% (11.1–33.3%), despite several modifications to reduce the complications. Yang et al. [16] found that when a repeat TIP procedure was applied to urethral plates that had been altered by previous surgery, it resulted in a high incidence (58.8%) of complications, especially meatal stenosis. In the majority of patients in our series, since the prior failed operation was a TIP repair that produced some scarring and alteration of urethral plate, we considered that a repeat TIP procedure was inappropriate; hence we preferred the GTIP technique. In re-operative hypospadias, augmenting the urethral plate with a dorsal inlay graft (GTIP) has been reported to reduce complications and improve success [4]. Schwentner et al. [9] reported SGTIP in 31 patients with 84% success with 16% complication rate. The most common complication in their series was neourethral stricture at the proximal anastomosis. Ferro et al. [4] reported that stenosis after SGTIP was related to accidental early catheter expulsion post-operatively. They reported 82% success with 18% (8 of 44) complication rate after SGTIP. Ye et al. reported 85% success with SGTIP in 53 patients; their complications consisted of 5 UCF and 3 strictures. In the present series, our results were comparable with earlier reports, with a success of 77% for SGTIP in 31 children. Like others, we observed UCF and meatal/neourethral stenosis as the commonest complication of SGTIP; in four children, the UCF was associated with distal stenosis. There is no consensus regarding the ideal tissue to be used as the inlay graft; while Schwentner [9] used skin exclusively, Ye et al. [10] reported comparable success with the exclusive use of OMG. Similar to Ferro et al. [4], in the present series, we used either penile skin (where available) or OMG as graft material.

The present study has several limitations. It is a retrospective analysis, although our principles of management of failed hypospadias have not changed over the past decade. Only two senior surgeons performed all the procedures, since we believe, like others [13], that to prevent multiple failures, only experienced surgeons should treat secondary hypospadias. Hence, in the present study, we may be seeing the results in the best hands. There was also no possibility to randomize the cases between the two techniques, since the indications were different for both techniques. The median follow-up in both groups was only 24 months, and a longer follow-up till post-pubertal period is required to assess the long-term results of both these techniques. We observed

that most cases of failed distal hypospadias were suitable for the SM technique, which also yielded the best results and low complications. SGTIP may be used for cases that are not suitable for SM technique, with acceptable results. We found a higher rate of post-operative stenosis with SGTIP. The other authors [17] also observed that Mathieu technique had significantly less stenosis/strictures than TIP for primary hypospadias repair.

Conclusions

Overall, single-stage salvage distal hypospadias repair was successful in 87.8% of patients. Both SM and SGTIP demonstrated acceptable results for salvage distal hypospadias repair. SM had better success and lower complications than SGTIP. SGTIP was associated with significantly higher post-operative UCF and meatal/neourethral stenosis.

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Declarations

Conflict of interest The authors declare that they have no conflict of interest.

Ethical approval All the patients gave informed consent for the operation.

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