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ORIGINAL ARTICLE

Double dartos flap layer in tubularized incised plate urethroplasty to prevent urethrocutaneous fistula in uncircumcised patients with distal hypospadias

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Urethrocutaneous fistula may complicate hypospadias repair. We noticed that double-layered preputial dartos flaps added to tubularized incised plate urethroplasty can reduce the risk of urethrocutaneous fistula. The aim of this study was to compare the outcomes of tubularized incised plate urethroplasty with double-layered preputial dartos flaps to with single-layered local fascial flaps in preventing urethrocutaneous fistula. A retrospective cohort study was conducted between January 2017 and December 2020 at Jordan University Hospital (Amman, Jordan). Boys who were aged between 6 months and 5 years, diagnosed with distal hypospadias, and not circumcised were included. The primary outcome was the occurrence of urethrocutaneous fistula in patients who underwent tubularized incised plate urethroplasty with a double-layered fascial flap. The results showed a total of 163 boys with distal hypospadias; among them, 116 patients underwent tubularized incised plate urethroplasty with a single-layered fascial flap, and 47 underwent tubularized incised plate urethroplasty with a double-layered fascial flap. The development of urethrocutaneous fistula was higher in the group receiving tubularized incised plate urethroplasty with a single-layered fascial flap than in the group receiving tubularized incised plate urethroplasty with a double-layered fascial flap after 1 month, 6 months, and 12 months (6.9% vs 0, 10.3% vs 0, and 5.2% vs 0, respectively), and the difference after 6 months was statistically significant (P = 0.02). Asian Journal of Andrology (2023) 25, 93–97; doi: 10.4103/aja202251; published online: 16 August 2022

Keywords: distal hypospadias: double-layered preputial dartos flap; tubularized incised plate urethroplasty; urethrocutaneous fistula

INTRODUCTION

Hypospadias is a common congenital malformation referring to malpositioning of the external urethral meatus because of fusion failure of the urethral folds and foreskin on the ventral penis. The meatus may present with various degrees of malpositioning,¹ insufficient development of the penile ventral tissue, ventral penile curvature (chordee), ventral urethral opening, and excess dorsal preputial skin as anatomical manifestations.² The incidence is one in 300 live male births worldwide.² The exact cause of hypospadias has not been uncovered. However, some studies have implicated endocrine, vascular, genetic, and environmental risk factors in the hypospadias etiology.^{3,4} Hypospadias is classified according to the abnormal urethral meatus location as follows: anterior (subcoronal and glandular), middle (distal penile, proximal penile, and midshaft), and posterior (scrotal, perineal, and penoscrotal). Hypospadias surgical correction is best carried out between 6 months and 18 months of age, because at 6 months, the anesthesia and procedure are well tolerated, and at 18 months, the child is toilet trained.5

The most widely used hypospadias repair is tubularized incised plate urethroplasty (TIPU),⁶ first introduced by Snodgrass in 1999 when he described deep longitudinal incision of the plate, allowing tubularization with no additional flaps as the key modification step.7 Urethrocutaneous fistula (UCF) is the most common complication after hypospadias surgery; other recognized, and not uncommon complications include bleeding and hematoma, skin necrosis, wound infection, penile torsion, meatal stenosis, inadvertent removal of the urethral stent, and catastrophic rare wound dehiscence.8 Some of these complications might require surgical repairs if they are not healed with conservative treatment. To avoid this outcome, other techniques, such as double dartos flap (DDF) repair, have been added to TIPU to minimize the incidence of urethrocutaneous fistula. DDF repair, first introduced by Retik et al.9 in 1994, includes a subcutaneous flap to wrap the neourethra after hypospadias, and many studies over the last few years have shown excellent results of this technique in lowering complication rates, including urethrocutaneous fistula.9,10 It is believed that the double well-vascularized DDF may enhance spontaneous healing of the UCF by furnishing adequate blood supply.6

There are many techniques for hypospadias repair, but none are guaranteed to achieve adequate results. Snodgrass's TIPU, accompanied with double dartos flap technique, was implemented in most of our cases, was implemented in most of our cases. In this study, we

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showed that dartos flap coverage is a reliable technique to prevent UCF formation.

PATIENTS AND METHODS

This was a retrospective cohort study of all uncircumcised pediatric patients operated on for distal hypospadias (mid-shaft and beyond) at Jordan University Hospital in Amman, Jordan, from January 2017 to December 2020. Data were obtained from the hospital medical records. Exclusion criteria included patients circumcised before surgery, patients with a proximal type of hypospadias (proximal to mid-shaft), and patients operated on for hypospadias. Patient families were contacted to enquire about the overall satisfaction with the final penile shape. Informed consent was obtained from all legal guardians of patients to take part in the study. All patient data remained confidential. Ethical approval was obtained from the Ethics Committee and Institutional Review Board (IRB) of Jordan University Hospital (No. 10/2121/6868).

Boys in the TIPU + single-layered local fascial flap (SLLEF) group underwent TIPU repair with SLLFF, and those in the TIPU + double-layered preputial dartos flap (DLPDF) group underwent TIPU with double dartos urethroplasty. All operations were performed under general anesthesia by different surgical teams within the same department. The penis was degloved after stay sutures were inserted on the glans, a longitudinal incision was made deeply into the groove, and extreme caution was taken to incise only through the dermis on top of the distal urethra. Degloving of the shaft skin was performed while preserving intact all subdermal vascularized tissue attached to the mucosal collars, urethra, and lateral urethral corporeal groove. The neourethra was constructed over a suitably sized catheter maintained for 1 week and polydioxanone suture (PDS 6/0 or PDS 7/0, Ethicon Inc., Brussels, Belgium) used in all reconstructions. A dartos fascial flap raised from the preputial skin on the dorsal aspect was used to cover the neourethra. In the TIPU + SLLEF group, the button-hole technique was used to fit a single layer of the dartos flap, while in the TIPU + DLPDF group, the dartos flap was halved and double layered on the neourethra. UCF was defined as abnormal communication between the urethra and skin of the penis and confirmed by two or more streams during micturition.

The main measured end point was the occurrence of UCF in patients who underwent TIPU secured with DLPDF. The incidence of intraoperative and early postoperative complications such as wound infection or dehiscence, skin necrosis, bleeding, or hematoma and the incidence of long-term complications such as meatal stenosis and phallic rotation were estimated. Familial overall satisfaction with the final penile shape was recorded. Statistical analysis was carried out using the SPSS version 25.0 software program (IBM Corp., Armonk, NY, USA). Fisher's exact test was applied, with statistical significance identified as a P < 0.05.

Table 1. Type of hypospaulas, surgery, and operative th	lable	1: Type of	nypospadias,	surgery,	ana	operative	time
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RESULTS

A total of 222 boys had distal hypospadias, among which 31 were already circumcised before hypospadias surgery and were excluded. Twenty-eight boys underwent a meatal advancement and glanduloplasty (MAGPI) procedure and were excluded. The remaining 163 boys underwent TIPU: the TIPU + SLLEF group totaled 116 procedures involving TIPU with SLLFF, and the TIPU + DLPDF group totaled 47 procedures involving TIPU with DLPDF.

Coronal hypospadias was the most common subtype (66.9%). The two operative techniques were almost comparable in terms of the subtypes of distal hypospadias operated on and the median age of patients at the time of surgery. The duration (mean \pm s.d.) was greater in TIPU + DLPDF group (1.54 h \pm 0.32 h) than in TIPU + SLLFF group (0.91 h \pm 0.19 h), with a significant difference of 0.63 h (95% confidence interval [CI]: 0.53–0.73, *t* [59.477] = 12.502, *P* = 0.001), as shown in **Table 1** and **Figure 1**. There were no outliers in the data, as assessed by inspection of a boxplot. The data violated the assumption of homogeneity of variances, as assessed by Levene's test for equality of variances (*P* = 0.003), and the duration of surgery for each type of operation was normally distributed.

Early postoperative bleeding or hematoma formation in both techniques was low (1.2%) but still higher in TIPU + DLPDF group than those in TIPU + SLLFF group (2.1% vs 0.9%). The relative risk of developing early postoperative bleeding after TIPU + SLLFF group compared to TIPU + DLPDF group was 0.4 (95% CI: 0.026–6.344). Because of small sample size, Fisher's exact test showed no significant difference in proportions (P = 0.4). There was no skin necrosis after either technique. Early postoperative wound infection was low (3.1%) but still higher in TIPU + SLLFF group than that in TIPU + DLPDF group (3.4% vs 2.1%). The relative risk of TIPU + SLLFF group



Figure 1: The comparison of the mean operative duration time between TIPU + SLLFF group and TIPU + DLPDF group. The mean operative duration time was greater in TIPU + DLPDF than in TIPU + SLLFF with a significant difference of 0.63 h. TIPU: tubularized incised plate urethroplasty; SLLFF: single-layered local fascial flap; DLPDF: double-layered preputial dartos flap; CI: confidence interval.

Characteristic	All patients (n=163)	Operation					
		TIPU+SLLFF (n=116)	TIPU+DLPDF (n=47)	Р			
Subtype of hypospadias, n (%)				0.001*			
Glandular	37 (22.6)	25 (21.6)	12 (25.5)				
Coronal	109 (66.9)	86 (74.1)	23 (48.9)				
Distal shaft	12 (7.4)	2 (1.7)	10 (21.3)				
Mid-shaft	5 (3.1)	3 (2.6)	2 (4.3)				
Patient's age at time of operation (year), median (IQR)	1.30 (1.0)	1.16 (1.0)	1.70 (1.0)	0.278**			
Duration of surgery (h), mean±s.d.	1.09±0.37	0.91±0.19	1.54±0.32	0.001***			
Coronal Distal shaft Mid-shaft Patient's age at time of operation (year), median (IQR) Duration of surgery (h), mean±s.d.	109 (66.9) 12 (7.4) 5 (3.1) 1.30 (1.0) 1.09±0.37	86 (74.1) 2 (1.7) 3 (2.6) 1.16 (1.0) 0.91±0.19	23 (48.9) 10 (21.3) 2 (4.3) 1.70 (1.0) 1.54±0.32	0.27 0.00			

"Fisher's exact test (*r*x2); "Mann–Whitney U-test; ""independent-samples *t*-test. TIPU: tubularized incised plate urethroplasty; SLLFF: single-layered local fascial flap; DLPDF: double-layered preputial dartos flap; IQR: interquartile range; s.d.: standard deviation

compared to TIPU + DLPDF group developing early postoperative wound infection was 1.621 (95% CI: 0.186–14.123). Again, Fisher's exact test did not show a significant difference (P = 1.000) because of the small sample size.

The meatal stenosis rate was higher in TIPU + DLPDF group after 1 month and 6 months (10.6% vs 3.4%, and 6.4% vs 1.7%, respectively). The relative risk of developing meatal stenosis at 1 month and 6 months postoperatively in the TIPU + SLLFF group compared to that in the TIPU + DLPDF group was 0.324 (95% CI: 0.091–1.155) and 0.270 (95% CI: 0.047–1.565), respectively. There was no significant difference in the respective proportions of 4.2% (P = 0.1), and 1.7% (P = 0.1). The incidence of meatal stenosis of patients after 1 year was higher in TIPU + SLLFF group than that in TIPU + DLPDF group (3.4% vs 0); however, this association was not significant (P = 0.3).

The development of UCF was higher in the TIPU + SLLFF group than that in the TIPU + DLPDF group after 1 month, 6 months, and 12 months (6.9% *vs* 0, 10.3% *vs* 0, and 5.2% *vs* 0, respectively), and this

difference after 6 months was statistically significant (P = 0.02). The development of phallic rotation was also higher in patients in TIPU + SLLFF group than those in TIPU + DLPDF group at 1 month and 12 months postoperatively (1.7% vs 0, P = 1.000; and 1.7% vs 0, P = 1.000). For all cases, the most common early postoperative complication was wound infection (3.1%). The most common postoperative complication after 1 month was meatal stenosis (5.5%), followed by UCF (4.9%). The most common postoperative complication after 6 months was UCF (7.4%), followed by meatal stenosis (3.1%). The most common postoperative complication after 12 months was UCF (3.7%), followed by meatal stenosis (2.5%).

The observed frequencies and percentages of postoperative complications and overall parental satisfaction for each type of surgical technique are presented in **Table 2**. For parents reached by phone, the overall satisfaction was much greater of patients in TIPU + DLPDF group than that in TIPU + SLLFF group (61.7% *vs* 46.6%). None were dissatisfied with TIPU + DLPDF, while 6.0% were dissatisfied with the TIPU + SLLFF.

Table 2.	Observed	frequencies	and	percentages of	of compl	ications	and	parental	overall	satisfaction	for	each	type	of	surgical	technique
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Post-operative complications and parental	All patients, n (%)	Operation				
satisfaction		TIPU+SLLFF, n (%)	TIPU+DLPDF, n (%)	Р		
Patients	163 (100.0)	116 (100.0)	47 (100.0)			
Bleeding or hematoma ^a				0.495*		
No	161 (98.8)	115 (99.1)	46 (97.9)			
Yes	2 (1.2)	1 (0.9)	1 (2.1)			
Skin necrosis ^a						
No	163 (100.0)	116 (100.0)	47 (100.0)			
Wound infection ^a				1.000*		
No	158 (96.9)	112 (96.6)	46 (97.9)			
Yes	5 (3.1)	4 (3.4)	1 (2.1)			
Wound dehiscence ^b				-		
No	162 (99.4)	115 (99.1)	47 (100.0)			
Yes	1 (0.6)	1 (0.9)	0 (0)			
Meatal stenosis ^b				0.122*		
No	154 (94.5)	112 (96.6)	42 (89.4)			
Yes	9 (5.5)	4 (3.4)	5 (10.6)			
UCF ^ь				0.106*		
No	155 (95.1)	108 (93.1)	47 (100.0)			
Yes	8 (4.9)	8 (6.9)	0 (0)			
Urethral stent slipped inside ^b						
No	162 (99.4)	116 (100)	46 (97.9)			
Yes	1 (0.6)	0 (0)	1 (2.1)			
Phallic rotation (>90°) ^b				1.000*		
No	161 (98.8)	114 (98.3)	47 (100.0)			
Yes	2 (1.2)	2 (1.7)	0 (0)			
Wound dehiscence (recurrence) ^c				-		
No	162 (99.4)	115 (99.1)	47 (100.0)			
Yes	1 (0.6)	1 (0.9)	0 (0)			
Meatal stenosis ^c				0.145*		
No	158 (96.9)	114 (98.3)	44 (93.6)			
Yes	5 (3.1)	2 (1.7)	3 (6.4)			
UCF°				0.020*		
No	151 (92.6)	104 (89.7)	47 (100.0)			
Yes	12 (7.4)	12 (10.3)	0 (0)			
Phallic rotation (>90°) ^c						
No	163 (100.0)	116 (100.0)	47 (100.0)			

Contd...

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Table 2: Contd...

Post-operative complications and parental	All patients, n (%)	Operation					
satisfaction		TIPU+SLLFF, n (%)	TIPU+DLPDF, n (%)	Р			
Yes	0 (0)	0 (0)	0 (0)				
Meatal stenosis (narrow projectile stream) ^d				0.325*			
No	159 (97.5)	112 (96.6)	47 (100.0)				
Yes	4 (2.5)	4 (3.4)	0 (0)				
UCF ^d				0.183*			
No	157 (96.3)	110 (94.8)	47 (100.0)				
Yes	6 (3.7)	6 (5.2)	0 (0)				
Phallic rotation (>90°) ^d				1.000*			
No	161 (98.8)	114 (98.3)	47 (100.0)				
Yes	2 (1.2)	2 (1.7)	0 (0)				
Parental satisfaction ^d				0.170**			
Satisfied	83 (50.9)	54 (46.6)	29 (61.7)				
Partially satisfied	10 (6.1)	7 (6.0)	3 (6.4)				
Unsatisfied	7 (4.3)	7 (6.0)	0 (0)				
Not reached	63 (38.7)	48 (41.4)	15 (31.9)				

^aThe complications measured at early postoperative; ^bthe complications measured at 1 month postoperative; ^cthe complications measured at 6 months postoperative; ^athe complications and parental satisfaction measured at 12 months postoperative. ^{*}Fisher's exact test; ^{**}Fisher's exact test (rx2). TIPU: tubularized incised plate urethroplasty; SLLFF: single-layered local fascial flap; DLPDF: double-layered preputial dartos flap; UCF: urethrocutaneous fistula

DISCUSSION

TIPU is a practiced reconstructive surgery for distal hypospadias.¹¹ However, many specialists believe that it has a high initial failure rate and unsatisfactory outcome, and we commonly need additional procedures even for adults.¹² In this series, we found coronal hypospadias to be the most common subtype of hypospadias (66.9%), followed by glandular hypospadias (22.7%). A previous study in Jordan with 2495 boys estimated the prevalence of hypospadias to be 0.92%; that of glandular hypospadias, 65.2%; and that of coronal hypospadias, 21.7%.¹³

There are conflicting data about the superiority of using double over single dartos covering flaps, as most studies are retrospective.¹⁴ However, a randomized prospective study in 2008 showed that the use of dartos flaps in hypospadias offers no significant advantage over flapless repair, although such use is better at preventing UCE.¹⁵

In a multicenter study by Bertozzi *et al.*⁶ the results of 394 patients showed that TIPU protected with a double layer of well-vascularized dartos flap has a very low overall complication rate of 5.83% that included UCF incidence (1.01%); all of these UCF healed due to very good blood supply from the two interposed layers of dartos tissue; stenosis (0.25%), mild stenosis (2.53%), dehiscence of the ventral cutis (0.50%), and penile torsion (1.26%). Nonetheless, the cosmetic results were good, and all parents were satisfied. Our study results are consistent with the previous investigation as well as with other studies¹⁶ in reporting no UCF and phallic rotations after the second technique with DLPDF. The higher rate of meatal stenosis could have been due to the added layer, which caused less space to be available and exercised more pressure on the neourethra.

UCF is the most common complication of hypospadias correction occurring at different rates. A meta-analysis by Wu *et al.*¹⁷ showed the incidence of UCF after distal hypospadias repair to be 4%. Safwat *et al.*¹⁸ compared single- and double-barrier dartos flap coverage of the neourethra. The incidence of urethrocutaneous fistula was 2 out of 28 (7.1%) cases in the single-layer group, and no UCFs occurred in the double-layer group.

A systematic review by Fahmy *et al.*¹⁹ showed a lower rate of UCF development in cases of distal penile hypospadias repaired with

DLPDF after single-dartos flap urethroplasty in primary hypospadias (0.6% *vs* 5.1%). Limitations of the study were the single-center design and the small sample size.

CONCLUSIONS

TIPU + DLPDF decreases the risk of postoperative UCF and phallic rotation. The procedure reduces the risk of long-term meatal stenosis complications, and we found it to be associated with much greater parental satisfaction. However, TIPU + DLPDF took more operative time and was associated with more bleeding and hematoma formation. Nonetheless, overall, the benefits of DLPDF outweigh its complications.

AUTHOR CONTRIBUTIONS

RT, MN, MR, AT, AJY, and JA carried out the study design and conception; RT, AJY, AT, EH, D Alshareefi, D Alenezi, and BA participated in data acquisition and classification; RT, MN, MR, AJY, and JA performed data analysis and interpretation; RT, AJY, MN, MR, and AT carried out drafting of the final manuscript; RT, MN, AJY, and JA supervised and performed critical revision. All authors read and approved the final manuscript.

COMPETING INTERESTS

All authors declare no competing interests.

REFERENCES

- Snodgrass WT. Consultation with the specialist: hypospadias. *Pediatr Rev* 2004; 25: 63–7.
- 2 Satjakoesoemah AI, Situmorang GR, Wahyudi I, Rodjani A. Single-stage urethroplasty: an eight-year single-center experience and its associated factors for urethrocutaneous fistula. *J Clin Urol* 2021; 14: 190–5.
- 3 Morera AM, Valmalle AF, Asensio MJ, Chossegros L, Chauvin MA, et al. A study of risk factors for hypospadias in the Rhône-Alpes region (France). J Pediatr Urol 2006; 2: 169–77.
- 4 Haraux E, Braun K, Buisson P, Stéphan-Blanchard E, Devauchelle C, et al. Maternal exposure to domestic hair cosmetics and occupational endocrine disruptors is associated with a higher risk of hypospadias in the offspring. Int J Environ Res Public Health 2017; 14: 27.
- 5 Bhat A. General considerations in hypospadias surgery. Indian J Urol 2008; 24: 188.
- 6 Bertozzi M, Yıldız A, Kamal B, Mustafa M, Prestipino M, et al. Multicentric experience on double dartos flap protection in tubularized incised plate urethroplasty for distal and midpenile hypospadias. *Pediatr Surg Int* 2011; 27: 1331–6.
- 7 Snodgrass WT. Tubularized incised plate hypospadias repair: indications, technique,

and complications. Urology 1999; 54: 6–11.

- 8 Bhat A, Arup M. Acute postoperative complications of hypospadias repair. *Indian J Urol* 2008; 24: 241.
- 9 Retik AB, Mandell J, Bauer SB, Atala A. Meatal based hypospadias repair with the use of a dorsal subcutaneous flap to prevent urethrocutaneous fistula. J Urol 1994; 152: 1229–31.
- 10 Yigiter M, Yildiz A, Oral A, Salman AB. Comparative study to evaluate the effect of double dartos flaps in primary hypospadias repair: no fistula anymore. *Int Urol Nephrol* 2010; 42: 985–90.
- 11 Belal MT, Bhuiyun MH, Hossain MS, Chowdhury TA, Morshed MS, et al. Modified tubularised incised plate urethroplasty (TIPU) with dorsal inlay preputial skin graft: an uutcome evaluation of 21 cases. Bangladesh Med J 2018; 47: 9–12.
- 12 Myers JB, McAninch JW, Erickson BA, Breyer BN. Treatment of adults with complications from previous hypospadias surgery. J Urol 2012; 188: 459–63.
- 13 Shahin BH, Habashneh MS, Hazza IA, Switty TA, Aqqad SS, et al. Prevalence of genital disorders in jordanian school boys. Saudi J Kidney Dis Transpl 1996; 7: 383.
- 14 Maarouf AM, Shalaby EA, Khalil SA, Shahin AM. Single vs. double dartos layers for preventing fistula in a tubularized incised-plate repair of distal hypospadias. Arab J Urol 2012; 10: 408–13.
- 15 Erol A, Kayikci A, Memik O, Cam K, Akman Y. Single vs. double dartos interposition flaps in preventing urethrocutaneous fistula after tubularized incised plate urethroplasty in primary distal hypospadias: a prospective randomized study. Urol

Int 2009; 83: 354-8.

- 16 Mustafa M, Wadie BS, Abol-Enein H. Dorsal dartos flap in Snodgrass hypospadias repair: how to use it? *Urol Int* 2008; 81: 215–7.
- 17 Wu Y, Wang J, Zhao T, Wei Y, Han L, et al. Complications following primary repair of non-proximal hypospadias in children: a systematic review and meta-analysis. *Front Pediatr* 2020; 8: 750.
- 18 Safwat A, Al-Adl AM, El-Karamany T. Vascularized dartos flap in conjunction with tubularized incised plate urethroplasty: single versus double flaps for management of distal hypospadias. *Curr Urol* 2012; 6: 67–70.
- 19 Fahmy O, Khairul-Asri MG, Schwentner C, Schubert T, Stenzl A, et al. Algorithm for optimal urethral coverage in hypospadias and fistula repair: a systematic review. *Eur Urol* 2016; 70: 293–8.

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