

Repair of distal hypospadias by construction of neourethra from augmented urethral plate with two lateral strips of glans skin and coverage with dartos flap followed by skin closure with preputial flap: single center series

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Introduction Hypospadias is one of the most common anomalies in boys. Many surgical techniques for reconstruction of distal hypospadias have been described, each method having a different success rate. Our objective in this study is the assessment of the surgical technique for repair of distal hypospadias by construction of a neourethra from the augmented urethral plate with two lateral strips of glanular skin, coverage with a dartos flap, and followed by skin closure with preputial skin flap.

Material and methods From March 2016 to November 2018, repair of distal hypospadias was performed in 56 children with a mean age of 3.2 years (range 2 to 8 years old). Minimum follow-up was 12 months, maximum up to 24 months. Success was defined by cosmetic appearance of the penis, parent perception of penile appearance, and urinary function. Uroflowmetry was done in 22 children at the 12 months follow-up. Parents evaluation of procedure was done by questionnaire using the pediatric penile perception score (PPPS).

Results The results were successful with straight penile shaft, conical glans, slit-like meatus located at the tip of the glans, with no rotation, and normal micturition. Five children had urethrocutaneous fistula (8.9%) that were treated with simple closure. Parents reported 'very satisfactory' (98.2%) and 'satisfactory' (1.8%).

Conclusions The described technique of repair of distal hypospadias in children with different variants of urethral plate width and glans size showed good results, both operator's and parent's perception of the results being very satisfactory. This technique is categorised stage 2a in the IDEAL (Idea, Development, Evaluation, Assessment, and Long-term study) staging system for surgical innovations.

Key Words: hypospadias repair ◊ urethroplasty ◊ urethral plate ◊ preputial flap ◊ dartos flap ◊ surgical innovations

INTRODUCTION

Thiersch-Duplay, Mathieu, and Denis-Browne were credited to invent the early surgical techniques for repair of hypospadias. Since then, many methods for the treatment of hypospadias were described with different concepts: urethral advancement technique [1, 2, 3], Duckett's transverse preputial island flap

technique with the principle of creating a glanular channel combined with the use of a transverse preputial island flap to create a neourethra [4], meatoplasty and glanuloplasty [5, 6], onlay technique [7], tubularized incised plate (TIP) urethroplasty that was popularised by Snodgrass [9, 10, 11], augmentation of TIP with glanular skin flap and closure of the glanular wings with removal of a portion of spongiosum

tissue [11], and wide skeletonization of the incised urethral plate with standard closure of glanular wings [12]. Width of urethral plate and glans size had been considered a crucial factor for the success of surgical procedures [13, 14]. In prospectively collected data from 490 consecutive boys undergoing hypospadias repair, a small glans size, defined as width <14 mm, proximal meatal location, and re-operation were an independent risk factor for post-hypospadias repair complications [15]. Snodgrass and coworkers reported that, in patients undergoing TIP urethroplasty for distal hypospadias, the glans dehiscence occurred in 4%. Overall complication rate in patients undergoing primary distal and proximal TIP urethroplasty repair was 12%, these complications being attributed to a narrow urethral plate and small glans width [16]. Snodgrass and Bush reported that the incidence of complications increased to 32% in cases of re-operative TIP urethroplasty and small glans size <14 mm [17]. Most of the procedures for hypospadias repair reported different incidences of complications: urethrocutaneous fistula (13%), urethral stenosis (2–5%), glans dehiscence and re-operation (7%) [18, 19]. Vascularised dartos flap had been used to cover the tubularized neourethra to minimise post-operative fistula [20]. Parent's satisfaction or dissatisfaction of the cosmetic appearance of their boy's penis was a crucial factor in stratifying success of hypospadias repair [21, 22], parental perception of penile appearance and the presence of lower urinary tract symptoms being the main disappointment and regret of hypospadias operation [23, 24]. The surgical treatment of distal hypospadias aims at achieving an acceptable cosmetic outcome: a straight penis with a slit-shaped meatus at the tip of the glans which should have a conical shape. These objective criteria are the parents' expectations but there is no surgical procedure which guarantees success and fulfills all parents' wishes. The cosmetic results were judged significantly more optimistic by surgeons as compared to parents when using validated tools as the hypospadias objective penile evaluation (HOPE) score [25, 26]. Uroflowmetry is an objective non-invasive tool to evaluate functional results of hypospadias repair in boys who are toilet trained, able to void voluntarily, and having no fistula. It was proved to be a good tool for the evaluation of the neourethra after distal and mid-shaft hypospadias repair by TIP or Mathieu urethroplasty, the evaluation including maximum and average flow rate [27, 28]. Uroflowmetry of normal boys between the ages of 5 and 10 years was as follows: mean maximum flow rate (Q_{max}) 15 ± 5 cc/sec and mean average flow rate (Q_{ave}) 8 ± 3 cc/sec [29]. The innovative surgical technique had special considerations: On one

hand, the obligatory measurements of new drug applications cannot be applied to surgical innovations, on the other hand, surgical innovations are different regarding adopting randomization-controlled trials that would pose a range of problems. The IDEAL (Idea, Development, Evaluation, Assessment, and Long-term study) system for surgical innovations recognized four stages of development which are: Stage 1: is an idea applied on patients in a single digit, highly selective, its purpose is to prove the concept, and participants are few surgeons. Stage 2a: is development, iterative modification of the procedure until reaching a stable version; the design is a single center with prospective cohort, small number of patients ranging from 10 to 100; the purpose is technical description of the procedure towards a stable optimized version, and reporting on side effects. Stage 2b: collaborative prospective data collection including many patients with broadening of indication in order to include potential beneficiaries and many surgeons; the purpose is the assessment of the procedure that is based on a large sample allowing power calculation and analysis of the learning curve. Stage 3: assessment and definitive comparison of main efficacy and safety aspects of a new technique against current best treatment. Stage 4: long term monitoring [30, 31, 32]. Objectives of the study are the description of the procedure and its results.

MATERIAL AND METHODS

The study is a prospective reporting and evaluation of patients operated for distal hypospadias, including 56 consecutive children (mean age 3.2 years, range 2 to 8 years). The study was conducted between March 2016 to November 2018. Inclusion criteria were primary distal hypospadias and intact prepuce. Children who had urethral plate width ≤ 8 mm or small glans size ≤ 14 mm were also included. Position of the native meatus was glandular in 15, coronal in 17, subcoronal in 14, and distal penile in 10 boys. Approval of the ethical committee was obtained. Minimum follow-up was for 12 months, and maximum up to 24 months. Post-operatively, the patients had a clinical assessment at 1 month, 3 months, and 6 months, and then every 6 months for 2 years. Parents were asked to fill in questionnaires before the operation and during the 12 months follow-up visits to report their satisfaction or dissatisfaction. The questionnaire pediatric penile perception score (PPPS) included the location and shape of the neomeatus, shape and skin colour the glans, urinary stream, cutaneous fistula, and cosmetic appearance of the penile shaft. Overall perception was

either very satisfactory, satisfied, dissatisfied, or very dissatisfied [19].

Uroflowmetry

The urodynamic study was done at the 12 months follow-up for boys at the age between 5 and 10 years (mean 7 years) who were trained to use the toilet and were able to hold urine until they felt that the bladder was full. The children were recruited by telephone call to their parents to whom the significance of the urodynamic study was explained. Twenty-two parents responded. Data retrieved from uroflowmetry were Q_{max} , Q_{ave} , and voided urine volume. This data was compared to a matched age group of normal boys.

Operative technique

A. Construction of neourethra

The procedure began by making a slit incision in the native meatus and by passing a 10–12 Fr urethral catheter to the bladder. A ventral U shaped incision of the penile and glanular skin was created, the base of the U incision was one centimeter proximal to the native meatus, the two limbs of the U shaped incision were longitudinal incisions in the glanular skin based on the urethral plate. Dissection began from the lateral to the medial direction to create two glanular skin strips that are based on the urethral plate. The two strips were extended distally to the tip of the penis at the proposed site of the neomeatus, then the penile skin at the sides of the urethral plate and proximal to the native meatus was dissected with its fascia to allow tension free sutures (Figure 1A). Construction of the neourethra began proximally to the native meatus and extended distally. The construction started with the closure of the penile skin strips that were based on the native meatus and the penile portion of the urethral plate. Then the two glanular skin strips were closed together to reach the location of the neomeatus at the tip of the glans. Continuous sutures were taken with 7/0 polydioxanone sutures and re-enforced with multiple interrupted sutures (Figure 1B).

B. Fashioning vascularised dartos flap and preputial flap: The prepuce was stretched with two lateral sutures, then a transverse incision was made 5 mm away of the coronal sulcus. One surface of the prepuce was de-epithelized leaving the dartos with its blood supply, afterwards the prepuce was divided vertically in the middle to form two vascularised preputial flaps (Figure 2A). The left flap was de-epithelized on both sides becoming the vascular-

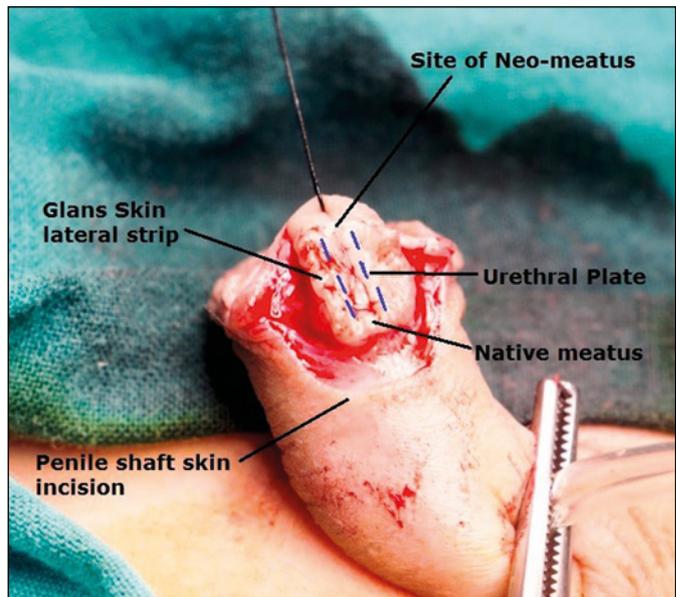


Figure 1A. Longitudinal incision of the glans skin based on the lateral edge of the urethral plate on both sides. Penile skin is included in the incision in cases of location of native meatus in distal part of the penis.

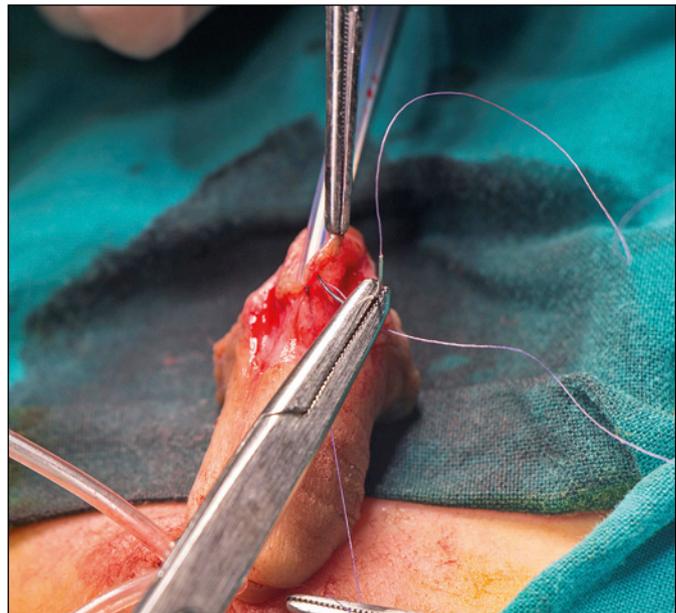


Figure 1B. Construction of the neourethra by suturing together the two lateral glans skin strips using 7/0 polydioxanone sutures. The sutures begin at the native meatus on a 10–12 Fr catheter, the neourethra is formed of the urethral plate augmented with the two glanular skin strips.

ized dartos flap that would enforce the neourethra. The right flap with its skin surface on one side and de-epithelized surface on the other side became the preputial skin flap that would cover the bare areas

in the glanular and penile shaft over the vascularized dartos flap (Figure 2B).

C. Coverage of the neourethra with vascularised dartos flap: The vascularized dartos flap was rotated ventrally to cover the neourethra and was fixed in place with interrupted sutures to the tissues of the glanular wings and tunica albuginea of the penile shaft (Figures 3A and 3B).

D. Closure of glanular and penile skin defects with vascularised preputial skin flap: The pre-

putial skin flap was rotated ventrally to cover the glanular and penile skin defects by placing it over the dartos layer (Figure 4A). This way, the neourethra received twice dartos support. The preputial skin flap was sutured to the edges of the glanular wings and penile skin with 7/0 polydioxanone sutures (Figure 4B).

E. Last step of the procedure: A urethral catheter was inserted and passed to the bladder, the operative site was closed with dressing, the catheter was left for ten days, and the dressing was changed after 5 days.

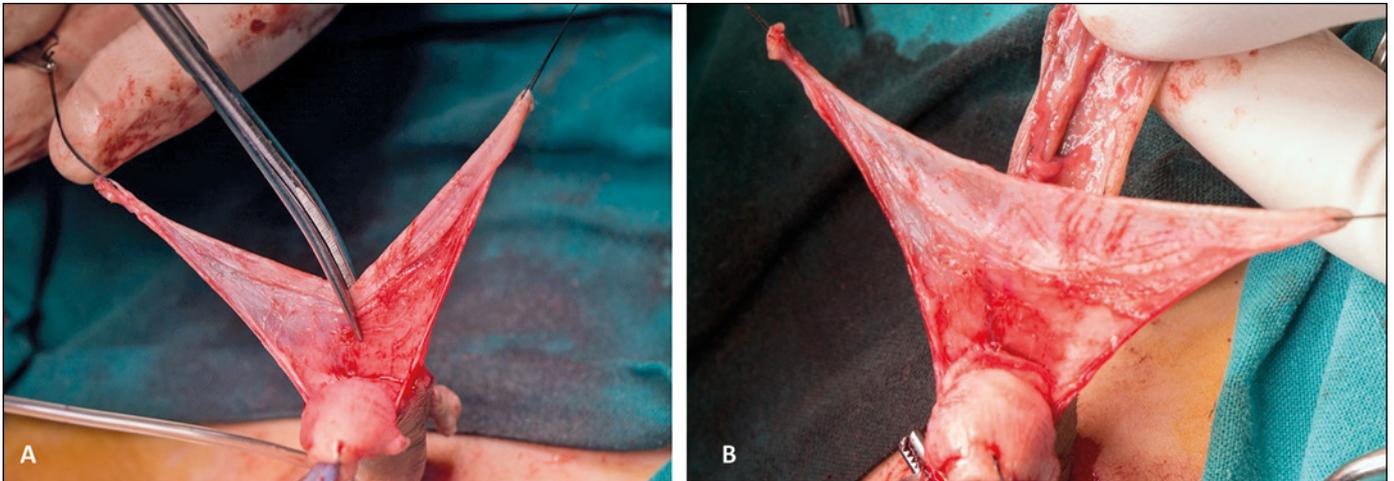


Figure 2. Fashioning vascularised dartos flap and preputial flap. **2A.** The prepuce is divided vertically in the middle to form two vascularised preputial flaps, the left flap will be de-epithelized on both sides to become vascularized dartos that will enforce the neourethra. **2B.** The right flap will be the preputial skin flap which was de-epithelized on one surface and will cover the bare areas of the glans and penile shaft.



Figure 3. Coverage of the neourethra with vascularised dartos flap. **3A.** The Dartos flap is rotated ventrally to cover the neourethra. **3B.** The flap is fixed in place with interrupted sutures to the tissues of the glanular wings and to the tunica albuginea of the penile shaft.

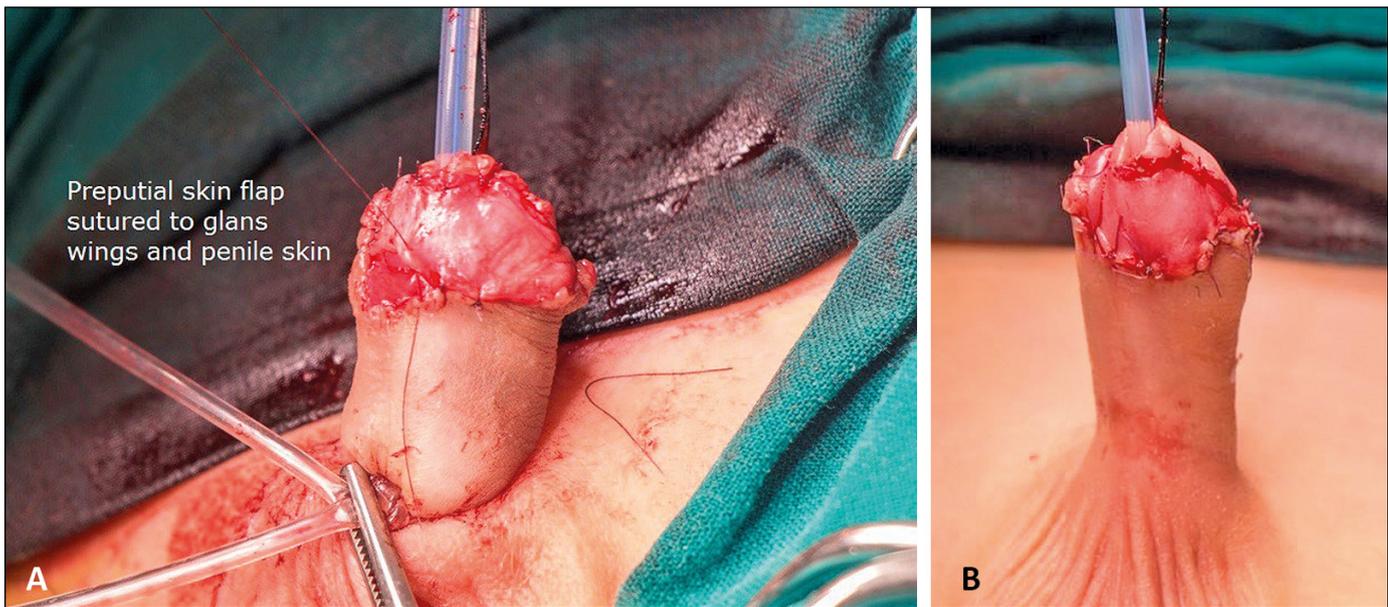


Figure 4. Closure of missing skin of glans and penile shaft with vascularised preputial skin flap. **4A.** The preputial flap is rotated ventrally to cover the glans and penile skin defects, where it is placed over the dartos layer. **4B.** The preputial flap is sutured to the edges of the glans wings and penile skin with 7/0 polydioxanone sutures.

Urethrocutaneous fistula

Urethrocutaneous fistula was reported in five children and was noticed one month to two months after the urethroplasty. The children noticed trickling of few drops of urine during micturition and the mother confirmed the findings. Clinical examination revealed the fistula during micturition. Calibration of the urethra was done with 12 Fr urethral catheter that showed free passage without meatal or urethral stenosis. Closure of the fistula was done 3 months after the hypospadias repair. The boys were admitted to hospital and examined under anaesthesia. The opening of the fistula was visualized by applying manual compression at the penoscrotal junction and injection of 3 ml sterile lubricating jelly into the meatus. The jelly protruded from the fistulous opening which was a single pinhole opening located at the sub coronal area. Closure of the fistula was done by a mini circumferential incision around the fistulous opening. There was enough tissue to close the opening and the tract in two layers with 0/6 polyglycolic sutures, one suture to tissues and a second one to the skin. No catheter was left behind, the child left the hospital the same day.

RESULTS

In the 56 patients, the results of hypospadias repair were successful. The cosmetic appearance was straight penile shaft, conical glans penis, and the me-



Figure 5. Post-operative photograph at 12 months follow-up showing the site of the result of the procedure. The neomeatus is at the tip of conical glans penis, straight penile shaft, and no skin scars or discoloration.

atus was slit-shaped and located at the tip of the glans. There were neither penile rotation nor vivid scars, follow-up showed satisfactory results (Figure 5). Five children had urethrocutaneous fistula (8.9%) that were closed with one procedure. We did not report meatal or urethral stenosis, glans dehiscence, or micturition disorders. All patients had follow-up

of minimal 12 months and up to 24 months. At 12 months follow-up, the parents were asked to report their satisfaction on the results of the procedure by filling in the questionnaire of the pediatric penile perception score PPPS [19]. The items to be reported were general appearance of the penis, shape of the glans, location of the neomeatus, and evaluation of micturition. Overall perception was either very satisfactory, satisfied, dissatisfied, or very dissatisfied. Parents reported very satisfactory in 55 cases (98.2%) and satisfied in one (1.8%). The report of satisfactory and not very satisfactory was due to the appearance of the circumcision line. Five boys had a urethrocutaneous fistula that was corrected in one procedure with simple anatomical closure. Follow-up after one month and up to 12 months showed perfect results without new fistula, stenosis, or redo procedures. These fistulas occurred in the early cases of the series. With the refinement of the technique and the rising learning curve, these side events did not occur again. Other complications were not reported. Characteristics of the five patients who developed enterocutaneous fistula are shown in Table 1. Uroflowmetry was done on 22 boys, their age range was 5-10 years (mean 7 ± 3). The mean flowmetry values were maximum flow rate (Q_{max}) ± 13 ml/sec, average flow rate (Q_{ave}) ± 7 ml/sec, voided volume ± 150 ml, and no residual urine. These results of uroflowmetry are matched with normal boys who had no lower urinary tract symptoms, neurogenic dysfunction, or congenital anomalies [29].

DISCUSSION

In a prospectively reported cohort of applying innovative surgical technique for repair of distal hypospadias in 56 boys, we achieved good results. It is a safe procedure, had no morbidity or mortality, and we reported the early complication that was managed successfully. The described technique would be classified stage 2a according to IDEAL system for staging surgical innovations. In this series, we had iterative

performance of the procedure till we reached a stable version, our case series being in accordance with previous reports on surgical innovations classification which does not need a randomisation trial [30, 31, 32]. There is no standard technique for repair of hypospadias, so a comparative study was not feasible. The overall complication rate of 8.9% favourably occurred in the early cases of the series compared with other series that reported complication rates of 12% to 33% [33, 34]. Urethrocutaneous fistula occurred in 5 patients and were repaired with simple procedure. These fistulas occurred in the early cases of the series. With the refinement of the technique and the rising learning curve, these side events did not occur again. Important advantages of the described technique are the successful functional and cosmetic results which were superior to the TIP urethroplasty [8, 10, 16, 35, 36]. The favourable results of the described procedure were due to the supple tissues that were used in construction of the neourethra that was covered with two vascularised dartos flaps. The augmentation of the urethral plate with two lateral strips of glans skin based on the urethral plate, in combination with incised urethral plate had been described before, in this technique the authors excised a portion of the glans tissue to facilitate closure of the two glanular wings [11]. Compared to our procedure, we keep the glanular tissue, it is not known what the result would be of removing a part of glans tissue on the future erectile function at puberty. Pantankar described a technique with wide skeletonization of TIP urethroplasty with narrow urethral plate. They reported good results using a V incision in their technique and closure of the neourethra with the glans wings [12]. In the present technique closure of glanular wings was achieved successfully by using a preputial skin flap which avoided the complication of glans dehiscence as there is enough tissue for closure without tension. Besides, incising the undeveloped narrow urethral plate would be the site of post-operative urethrocutaneous fistula. The urethral plate width had been studied before and

Table 1. Characteristics of the 5 boys who had urethrocutaneous fistula following a total of 56 hypospadias repair procedures

No.	Age (years)	Location of hypospadias	Time to diagnosis of fistula	Description of the fistula			Surgery	Result of management	Follow-up (months)
				Site	Size	Surrounding tissues of the fistula			
1	7	Subcoronal	1 month	Subcoronal	1 mm	Good	Closure	No fistula or stenosis	18
2	5	Subcoronal	2 months	Subcoronal	1 mm	Good	Closure	No fistula or stenosis	12
3	8	Subcoronal	2 months	Subcoronal	1 mm	Good	Closure	No fistula or stenosis	12
4	7	Distal penile	1 month	Subcoronal	1 mm	Good	Closure	No fistula or stenosis	18
5	6	Distal penile	2 months	Subcoronal	1 mm	Good	Closure	No fistula or stenosis	20

it was stated that width of urethral plate less than 8 millimetre in TIP procedure would carry a high risk of post-operative fistula [13, 14, 37]. In our series of 56 patients, the width of urethral plate and glans width did not affect the outcome of the procedure which has advantages over the onlay flap [7] and transverse preputial flaps [14] as it utilizes local glanular and penile tissues for construction of the neourethra. This proved to offer a higher success rate compared to other techniques that transferred preputial or penile skin to form a neourethra. The rotational flap of Mathieu was reported to have a high incidence of urethrocutaneous fistula [38]. In the technique presented here, the enforcement of the neourethra with vascularised preputial dartos fascia flap secured additional vascularity to the neourethra and supported the suture lines. This step in the technique was used by other authors [17, 39, 40]. Parents' evaluation of results of the procedure using the paediatric penile perception score showed that 98.2% reported 'very satisfactory', and 1.8% reported 'satisfactory' [19, 33]. In our study, we found that the perception of the parents on the cosmetic appearance after hypospadias repair was an important objective assessment compared to the less objective opinion of the surgeon. The surgeon's report on results of hypospadias repair is concerned with the complications of the operation. In the absence of any complication, the surgeon counts the operation as successful. In our study, we showed that the parents' concept of a successful operation was the critical point in defining success of hypospadias repair. Functional evaluation by uroflowmetry, studied on 22 boys, showed comparable results to normal age matched boys. Our results are in accordance

with previous reports that an objective assessment of hypospadias repair would be achieved with uroflowmetry [27, 28]. The mostly reported complications of TIP and Mathieu urethroplasties and their modifications lay in the glans penis leading to urethrocutaneous fistula, glans dehiscence, meatal and neourethra stenosis, and penile disfigurement. The technique presented here had minimal complications due to widening the urethral plate with two glanular skin strips that facilitated construction of the glanular part of the neourethra allowing for suturing without tension and good caliber of the neourethra. The two glanular wings were closed with interposing a vascularized preputial skin flap that led to closure without tension or compression on the neourethra. The neourethra was enforced with vascularized dartos flap. To reach stage 3 of the IDEAL staging system of surgical innovations, many operations using this technique are needed as well as expanding the indications and extending the follow-up period.

CONCLUSIONS

The procedure of repair of distal hypospadias in children by applying the technique of constructing a neourethra from augmented urethral plate with two lateral strips of glanular skin and coverage with vascularized dartos flap followed by closure of skin with vascularized preputial skin flap was successful, repetitive, and reproducible. Surgeons' and parents' perception of results were very satisfactory regarding function and cosmetic appearance.

CONFLICTS OF INTEREST

The authors declare no conflicts of interest.

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