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Tunneled tubularized bladder mucosal graft for long urethral reconstruction: is it a reliable option in selected complex cases in children and adolescents?

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Abstract

Aim: We analyzed the use of tubularized tunneled bladder mucosa graft (TBMG) and its application as a salvage procedure or as primary surgery for long urethroplasty in children and adolescents. Specific attention was given to a new method to harvest a long bladder mucosa graft through a minimal detrusotomy.

Methods: We analyzed the files of 10 patients who underwent TBMG urethroplasty at median age of 105 months (range 20-195 months). The indications were perineal hypospadias cripple in two, masculinizing genitoplasty for ovotesticular Ovotesticular Disorder of Sex Development (DSD) in two, perineal hypospadias in four, duplicated urethra in one, and complications of circumcision in one. Staged reconstruction was performed in all patients. The first stage was removal of all fibrous tissues and efficient treatment of curvature and skin coverage. After a minimal delay of one year, a free bladder mucosa graft was harvested through a minimal detrusotomy and tubularized. Tunneling of the graft was proceeded from the perineal urethrostomy to the glans.



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Results: The graft was successfully harvested through the minimal detrusotomy approach for variable lengths of urethroplasty; the median length was 10.5 cm (range 8-16 cm). The median follow up was 61.7 months (range 18-160 months). TBMG was the last surgery with no redo in five cases (50%). Long stricture occurred in one case of primary perineal hypospadias and needed a redo staged surgery. Two patients performed self-dilatation for distal stenosis.

Conclusion: The tunneled bladder mucosa tube graft technique represents a good alternative for a long urethroplasty in patients with a paucity of healthy skin. The minimal detrusotomy technique for graft retrieval may reduce graft harvesting morbidity.

Keywords: Hypospadias, bladder mucosa graft, hypospadias cripples, urethra reconstruction

INTRODUCTION

Redo surgery for proximal hypospadias remains a challenging procedure with inconsistent results^[1]. The majority of cases need staged reconstruction^[2,3]. Unfortunately, in some cases, the multiplicity of failed reconstruction ends with hypospadias cripples with multiple unhealthy scars, residual curvature, and inefficient urethroplasty.

It takes a surgical lifetime to learn how to deal with all of these various problems, but, even so, it is very difficult to satisfactorily categorize the treatment of such patients^[4]. In such cases, the lack of healthy skin to cover the penile shaft is challenging. The classical staged free grafts (buccal or bladder mucosa) requires, after the second stage, healthy well-vascularized skin to cover the tubularized flap.

Bladder mucosa tube graft was initially published in 1947 by Memmelaar^[5]. Its use was frequently criticized because of the high complications rate including mucosal prolapsus, proximal anastomosis stricture, and morbidity related to long detrusotomy^[6].

The tunneling approach of a tubularized free graft has been developed to minimize the rate of skin breakdown after redo surgery^[7,8]. The hypothesis was that the first stage of reconstruction, including correction of curvature, would leave an interval for revascularization of the tissues and skin coverage. Tunneling would not interrupt the vascularization of the surrounding tissues with no risk for skin dehiscence and less risk for graft necrosis.

Our aim was to analyze our cases where we used the tubularized tunneled bladder mucosa graft (TBMG) technique and its application as a salvage procedure (multiple failed reconstructions with no healthy skin or severe loss of skin) or as primary surgery for long urethroplasty (perineal hypospadias or masculinizing genitoplasty).

METHODS

We reviewed the medical files of 10 patients who underwent TBMG urethroplasty. The indications were redo perineal hypospadias in two, masculinizing genitoplasty for ovotesticular DSD in two, perineal hypospadias in four, duplicated urethra in one, and complications of circumcision in one (the detailed data are shown in [Table 1](#)).

Steps of surgery

First surgical procedure

Efficient treatment of curvature with specific procedures adapted was performed. All of our hypospadias

Table 1. Patient data and results

Case	Indication	Number of previous surgeries	Age (years)	Length of urethroplasty (cm)	Fu (months)	Complications	Treatment of complications
1	Multiple operated perineal hypospadias	7	11.3	12	97	None	
2		12	12.9	15	62	Meatal stenosis, stone	Stone laser fragmentation, self-dilatation 3 years, no recurrence
3	Ovotesticular 46XX DSD	1	12.5	14	61	Proximal fistula	Fistula closure, no further surgery
4		1	10.5	12	106	Proximal fistula	Fistula closure, no further surgery
5	Perineal hypospadias staged repair	0	1.7	8	160	Proximal stenosis	Onlay buccal mucosal graft, no further surgery
6		0	7.1	9	41	none	
7		0	2	7	30	stricture	Total redo, staged buccal mucosal graft complicated by proximal fistula needed closure without recurrence
8		0	6.6	8	6	Lost of FU (follow-up) (after 6 months free of complications)	
9	Urethral duplication	2	1.7	8	71	none	
10	Circumcision complication	2	16.3	16	18	none	Preventive self-dilatation 12 months

DSD: Disorder of sex development.

cases had severe curvature $> 45^\circ$; treatment was done by removal of all fibrous tissues and ventral corporoplasty when needed. (A ventral transverse incision was made at the point of greatest curvature through the tunica albuginea, extending from the 3 o'clock to the 9 o'clock position.) A flap of tunica vaginalis was used to cover the corpora defect. In cases of unavailable tunica vaginalis (ovotesticular DSD), multiple fairy cuts were done. All reconstructed or fibrous urethra was removed until obtaining a healthy native urethra; all of them were perineal [Figures 1 and 2].

In redo cases, penile skin was reconstructed either by local flaps or by free skin grafts. A minimum delay of one year was respected before TBMG.

Bladder mucosa graft harvesting by minimal detrusotomy

A short suprapubic transversal skin incision was done. The bladder was filled with saline and accessed by midline facial incision. A short (2 cm in length) transversal detrusotomy was done on the anterior wall of the bladder [Figure 3]. The exposed mucosa was incised vertically and retracted by 5/0 stich. Two parallel perpendicular incisions were done to isolate 2 cm width of the mucosa. Blunt and sharp dissection was done at the submucosal plane with mild retraction and progressive incision of the two parallel lines. These incisions were limited to the needed length for the urethra [Supplementary Video 1]. The mucosal defect was sutured by a running 5/0 absorbable suture, and the detrusotomy was closed after insertion of a supra pubic catheter.

Tubularization of the graft

The graft was tubularized by interrupted 6/0 absorbable polydioxanone (PDS, Ethicon) round needle. The tabularization was done around a Foley catheter of at least Ch 12 according to the age of the child. The graft



Figure 1. A 12-year-old boy (Case 2) had 12 procedures for perineal hypospadias. Sub-total loss of the penile skin.



Figure 2. First-stage surgery. (A) Persistent severe curvature after removal of all the fibrous tissues and the urethroplasty. (B and C) Large ventral cavernoplasty covered by left vaginalis flap. (D) The penis is covered by a combination of scrotal flaps (at the base) and a total skin free graft at the distal part of the shaft.

was kept hydrated with permanent irrigation of normal saline. In adolescents, the graft was tubularized around Ch 16 [Figure 4]. It was important to remove of any excess of width to have the graft fit around the chosen catheter.

Tunneling of the graft

The perineal urethrotomy was dissected free of any fibrous tissue and largely spatulated. Dissection was done close to the albuginea of the ventral surface of the corpus cavernosum. The same dissection was done distally at the base of the glans. If the glans was already constructed, a sharp midline incision was done to isolate a line of anastomosis with the deep mucosa of the glans. Efforts were made to not directly anastomose the graft with skin or fibrous glans edges. Tunneling was then done progressively with increasing diameter of metal bougie to obtain a caliber higher than the urethral catheter (e.g., a bougie 18 Fr for a 16 Ch catheter) [Figure 5].

Long forceps were introduced from proximal to distal. Then, the graft, attached to the catheter, was pulled smoothly from distal to proximal. If any resistance was felt, a new dilatation was done.

The proximal anastomosis was done by two running 6/0 PDS with the spatulated urethra. The perineal anastomosis was covered by multiple layers of corpus spongiosum and deep fascia.

The distal anastomosis was done with the inner part of the glans, with interrupted 6/0 PDS.

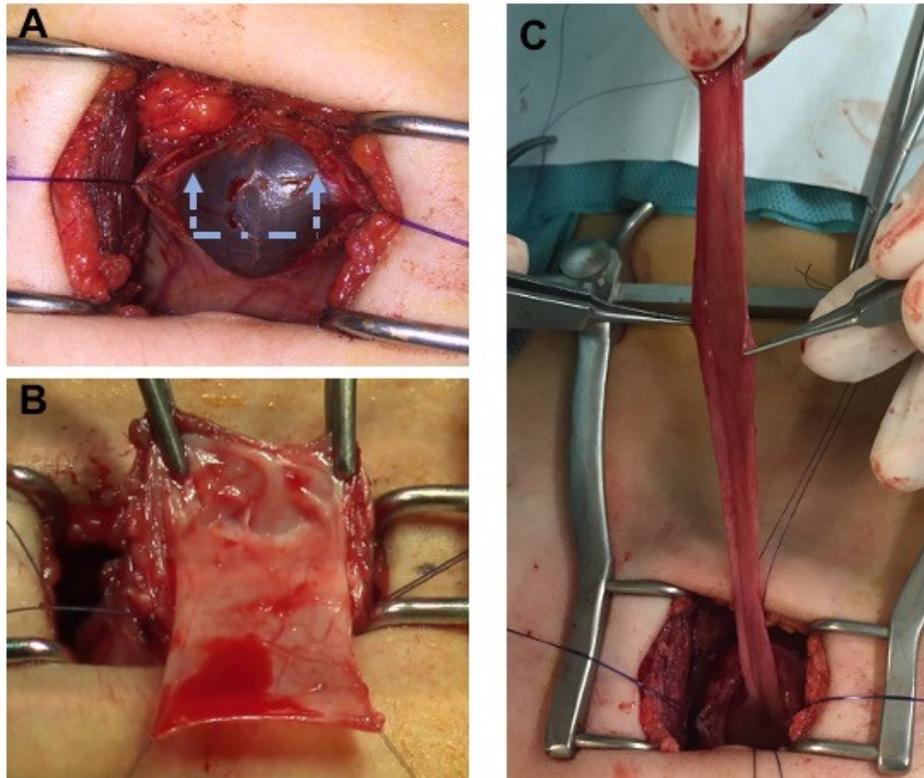


Figure 3. Harvesting of long BMG by minimal detrusotomy. (A) The detrusor is incised horizontally for 2 cm, and then two parallel incisions of the exposed mucosa are extended as far as needed. (B) The mucosa is freed from the detrusor by blunt dissection. (C) A long strip of bladder mucosa up to 16 cm can be retrieved. BMG: Bladder mucosa graft.

A compressive dressing was applied for five days and removed at the outpatient clinic. Anticholinergic treatment is routinely used. We do not use antibiotic prophylaxis other than one shot at the beginning of surgery.

After a delay of 21 days, the urethral catheter was removed, and the suprapubic catheter was closed. When voiding was easily obtained for 48 h, the suprapubic was removed. If there was any dysuria, infiltration of the perineal area, or excessive inflammation, the suprapubic was opened and antegrade urethrogram was done for eventually inserting under general anesthesia a new urethral catheter for a supplementary period until healing was obtained.

Follow up

The child was seen one month after surgery, then every three months for a year, and every year after that. At every follow-up, a flowmeter is done, and when needed cystoscopy is done under general anesthesia.

RESULTS

The graft surgery was done at median age of 105 months (range 20-195 months). The graft was successfully retrieved through the minimal detrusotomy approach for variable lengths of urethroplasty with a median length 10.5 cm (range 8-16 cm). The median follow up was 61.7 months (range 18-160 months). TBMG was the last surgery with no redo in five cases (50%) [Figure 6].



Figure 4. Second stage: tunneling of the tubularized bladder mucosa free graft. (A) With the need for a 16 cm urethroplasty, the erection test showed the absence of residual curvature. (B) Creation of the tunnel: the perineal dissection is done by sharp instrument until the exposure of the albuginea of the corpus cavernosa. A midline incision is done at the expected site of the meatus. Progressive dilatation with metallic bougie is done to achieve an easy passage of a bougie equal to Ch 18. (C) Tubularization of the free graft around a 16 Ch Foley catheter.

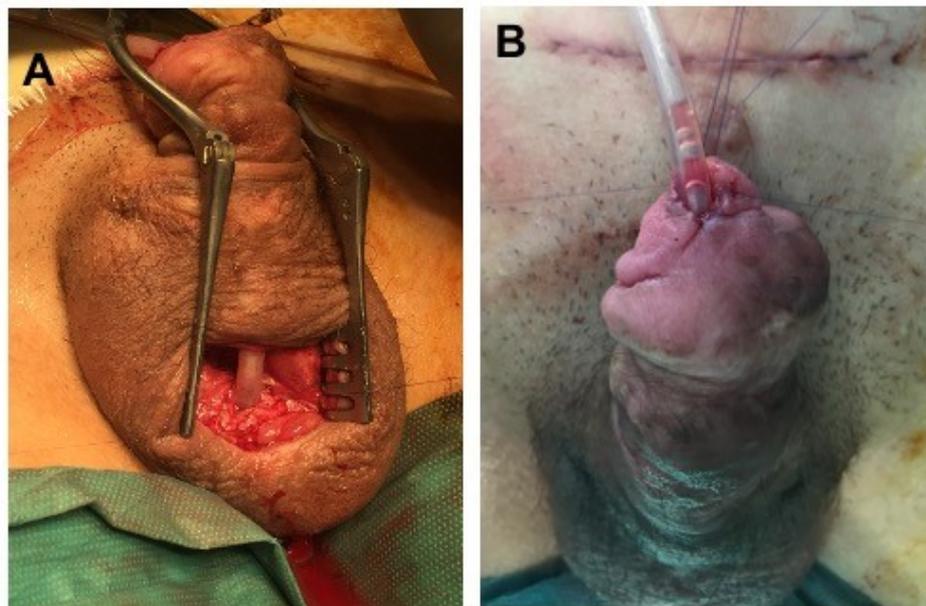


Figure 5. Tunneling of the tubularized graft from distal to proximal. Carefully positioning the suture line of the graft opposite to the corpus cavernosum, a large anastomosis is done between the graft and the spatulated perineal healthy urethra. (A and B) The distal anastomosis is done on the healthiest part of the glans.

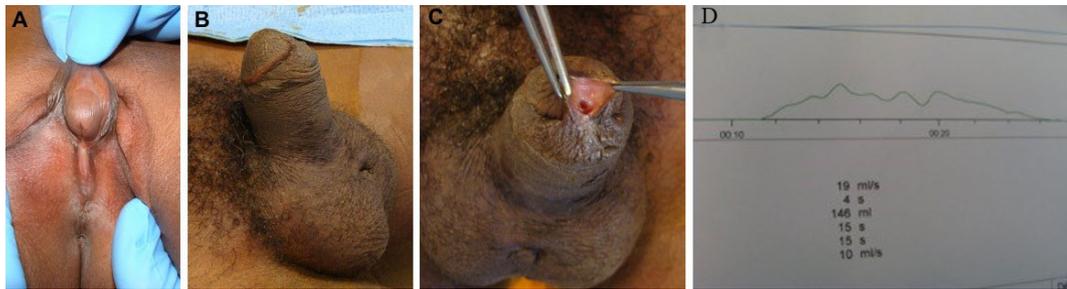


Figure 6. Results of BMG tunneling. (A) A 12-year-old ovotesticular 46XX boy, perineal urethra. (B) The aspect three years after staged urethroplasty using tunneled BMG, completed by bilateral testicular prosthesis later. (C) The glandular meatus showing the deep anastomosis between a healthy glandular mucosa and the BMG. (D) Flowmeter showing subnormal flow without postvoid residual. BMG: Bladder mucosa graft.

Redo hypospadias cripples: in one case (Case 2 in [Table 1](#)), stenosis of the distal anastomosis needed self-dilatation with progressive caliber introduced for 3 cm. The frequency was decided according to the interval of normal voiding stream. This 15-year-old adolescent preferred to keep the self-dilatation for three years. He also developed graft polyps inside the constructed urethra and stones that were voided after laser fragmentation.

Localized stenosis of the proximal anastomosis occurred in one perineal hypospadias case, and surgical treatment with onlay buccal mucosal graft successively treated the stenosis.

In one case of perineal hypospadias, an extended stricture of the graft at the penile level needed a total redo by staged approach with buccal mucosal graft. A limited prolapse of the mucosa was observed in two cases with no dysuria, which did not need further surgery.

In the case of extensive penile ischemia after circumcision (Case 10 in [Table 1](#)), the adolescent had 12 months of preventive self-dilatation of the meatus to keep good urine stream.

DISCUSSION

In our experience, the use of tunneled BMG to reconstruct the urethra remains a reliable option for a long urethroplasty in patients with a paucity of skin. Moreover, to reduce the morbidity of long graft retrieval, the minimal detrusotomy technique seemed to us an attractive solution.

There are many options to reconstruct the urethra after multiple failures of hypospadias reconstruction^[9]. Today, there is no single technique that can be used for all cases; tailored reconstruction is the optimal choice according to each case^[4]. Salvage hypospadias repair is challenging due to scarring and obliterated tissue planes, compromised blood supply, and deficient local tissue. Furthermore, patients with lengthy strictures or urethral breakdown may require augmentation or replacement of diseased urethral tissue for true surgical success. In their extensive multicenter series, Barbagli *et al.* analyzed the data from 1176 patients after failed hypospadias^[10]. They showed that a median of five additional procedures is required to achieve satisfactory final results. These cases represent two main challenges: skin covering and long segment urethroplasty.

After multiple hypospadias repairs, the patient is often left with minimal or deficient skin, which complicates further reconstruction. There are multiple ways of augmenting skin coverage, at times employing surrounding penile shaft skin with Z-plasties and rotational flaps, but often there is no

redundant skin available for these patients. Many of these options leave extensive scarring, or, when direct transfer of scrotal skin is used, there is a large area of hair-bearing skin that will become evident after puberty, leaving the patient with a poor cosmetic result. Updates of older techniques such as the Cecil Culp technique were modified and reported by Weiss *et al.*^[11]. In their 39-patient series using a Cecil Culp-modified technique (three stages), the final complication rate remained high with 21% having long-term complications including skin breakdown.

To avoid the major complication of skin breakdown, tunneling of a tubularized graft seemed to us a good alternative in such cases with a paucity of skin and scarring covering tissues.

In our experience, none of our patients had skin breakdown. Tunneling was also, in our opinion, beneficial in preventing urethral fistula. In our experience, fistulas occurred in two patients and were treated by local closure. The easiness and efficiency of the closure was due to the position of the fistula deep in the perineum at the proximal anastomosis that allowed multiple well-vascularized available layers, in contrast with penile fistula. Positioning of the suture line of tubularization against the corpus cavernosum in a closed space created by tunneling is probably the reason for the absence of fistula in the penile shaft.

Fine *et al.* reported their experience with tunneled buccal mucosal graft in 34 children with proximal hypospadias (75% primary)^[12]. The mean length of the urethral construction was 5.3 cm (range 3-9 cm). Their overall complication rate with more than one year of follow-up was 32%, including fistula in five patients, proximal stricture in four, and meatal stenosis in two. No patients experienced skin breakdown. The rate of stricture might be underestimated with relatively short follow-up compared to other series.

Zhao *et al.* presented a series of 57 adults with failed hypospadias repair, in whom they performed tubularized buccal graft urethroplasty, which was tunneled as an initial stage and anastomosed 6-12 months later^[7].

Recently, Kim *et al.* reported their experience with tunneled buccal mucosal graft after failed hypospadias surgery in 48 cases^[8]. Only 14.7% of patients were complication free, and all others required at least one redo surgery. In their experience, the mean length of the graft was relatively short at 3.7 cm. Only 22.9% of patients were stricture free after six years, while none of their patients had skin breakdown.

In our opinion, the use of tubularized tunneled buccal mucosal graft has its limits in the length of urethral reconstruction; the extensive length needed in our cases (median 10.5 cm and maximum 16 cm) exceeded the maximal length used in the reported cases. Moreover, the morbidity of buccal mucosal graft harvesting is not negligible. Among the paucity of reports on morbidity of buccal graft harvesting, Wood *et al.* reported postoperative significant pain in 83% of patients, and the main long-term complications were persistent perioral numbness (26%)^[13].

Bladder mucosa tube graft was initially published in 1947 by Memmelaar^[5]. Modifications of the initial technique to reduce the complications were more or less successful. Ransley *et al.* combined bladder mucosa with preputial skin grafts to reduce the incidence of distal prolapses^[6].

Tunneling of the bladder mucosal graft has been rarely reported for primary hypospadias^[14] or redo for long posterior urethral stricture^[15]. Recently, Lanciotti *et al.* reported a large series of 50 children with severe hypospadias, operated by a staged bladder mucosal graft^[16]. The graft was tunneled during the second stage. The mean graft length was 5.7 cm (range 35-85 mm). At a mean follow-up of five years, the total

complication rate needing redo surgery was 46%. Stricture at the proximal anastomosis occurred in 30% of cases, and multiple surgeries were needed in 30% of patients.

In our limited experience using TBMG in severe primary hypospadias repair in four cases, one patient had major complications (long stricture) that needed a complex two-stage redo surgery. This category of hypospadias remains challenging even as a primary case, and there is no evidence in the literature of superiority of one technique of the others^[17]. Our current practice now is to proceed with one-stage repair using double-face preputial flap whenever the skin is available to reconstruct the urethra and there is a well-vascularized skin coverage^[18]. We keep the option of free graft only in the case of a paucity of skin, a condition that occurs rarely in our experience.

The other category of patient in our series is the masculinizing genitoplasty for ovotesticular 46XX DSD. These patients had their surgery at a later age than the usual age for hypospadias reconstruction. The available foreskin was deficient, and the penile corpora needed cavernotomy to correct a significant curvature. TBMG allowed a long urethral segment reconstruction (12 and 14 cm) with apical meatus. Both patients had perineal fistula that occurred early after surgery and was closed by local surgery one year later without recurrence. Prolapses of the mucosa occurred in one case, and at last follow-up, nine years later, the patient was asymptomatic and no surgery was done for a minimal bladder mucosa prolapse.

In the case of hypospadias cripples, our two patients had multiples surgeries which left them with a scarring unhealthy skin coverage. After the first stage, removal of all fibrosed reconstructed urethra and treatment of significant residual curvature, the patients were left with a perineal urethrostomy at the level of their initial normal urethra. TBMG was a reliable salvage option, and none experienced skin breakdown. One of the patients, covered initially by a full-thickness free skin graft, needed self-dilatation for three years to treat distal stenosis, and then he progressively stopped the dilatation. This stenosis is probably secondary to poorly vascularized glans after multiple surgical procedures. In the other patient, who underwent seven failed surgeries, TBMG was successful without any complications, and the patient did not need any dilatation.

In some rare cases such as urethral duplication, TBMG seemed to us a good option, as the graft remains totally in the deep tissues and the distal and proximal anastomosis are well covered by vascularized local flaps. The same experience was reported by Mouttalib *et al.*, who successfully reconstructed long congenital urethral stenosis in two children^[15].

We used the same technique in a teenager with a major complication of circumcision at 15 years old. He had an ischemic sloughing of all the penile shaft skin, urethra, corpus cavernosum, and glans. He was treated somewhere else by total skin graft to cover the penis, leaving the meatus at a distance of 16 cm from the tip of the penis. TBMG as a salvage procedure was a successful option to reconstruct a 16 cm urethra without penile skin disruption. For this case, we opted to do preventive self-dilatation of the meatus for one year, and no distal stenosis was observed.

Morbidity related to bladder mucosa retrieval is one of the disadvantages of the bladder mucosa graft. The long detrusotomy is logically a source of postoperative bladder pain, hematuria, and dysfunction^[19]. In our series, we used an innovative method to retrieve the bladder mucosa graft through a minimal length of detrusotomy (2 cm). We could reach a length of 16 cm graft using this method. This approach may reduce the morbidity related to graft retrieval.

It is worth mentioning that the use of bladder mucosal graft might develop specific complications related to the nature of bladder mucosa, especially if there is an additional factor such as urine stagnation in the reconstructed urethra. Weingärtner *et al.* reported nephrogenic adenoma in six cases of reconstructed urethra using bladder mucosa graft^[20].

Our study has multiple limitations. The cohort of patients is heterogenous and of small number. The complications rates may vary according to the specific indications and duration of follow-up. In fact, if we consider one year as a minimum follow-up period to conclude success, we should exclude Case 8 from the study, which brings the valid cohort to nine patients. Consequently, the success rate would be 44% and not 50%. To give all the details of our cohort, we report clearly in [Table 1](#) all the follow-up periods and the complications of treatment. This leaves the surgeon to evaluate the eventual benefit of using our technique in their specific patient. We certainly acknowledge that these complex cases need to be reported with as long a follow-up as possible, not only one year but also long-term follow-up as complications may appear even after 10 years (after puberty and sexual activities).

Despite the heterogeneity of our cohort, our indication was mainly the same: a paucity of healthy skin coverage and the urethral defect being significantly long compared to previously published series.

In conclusion, the tunneled bladder mucosa tube graft technique represents a good alternative for a long urethroplasty in patients with a paucity of healthy skin. The minimal detrusotomy technique for graft retrieval may reduce graft harvesting morbidity.

DECLARATIONS

Authors' contributions

Made substantial contributions to conception and design of the study and performed data analysis and interpretation: El-Ghoneimi A, Peycelon M

Performed data acquisition, and material support: Lachkar AA, Lopez P, Paye-Jaouen A

Availability of data and materials

Not applicable.

Financial support and sponsorship

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Conflicts of interest

All authors declared that there are no conflicts of interest.

Ethical approval and consent to participate

The research was conducted in accordance with the principles of the Declaration of Helsinki and ethical approval has been obtained.

Consent for publication

A written informed consent for photography and publication was obtained.

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