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Two simple modifications can potentially change the future of proximal hypospadias surgery. Our series and a review of the literature

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ABSTRACT

Background/Objective: To draw a hint towards two simple modifications that could potentially decrease the complication rate.
Patients and Methods: It was a single center, single operator and retrospective study. All patients with severe hypospadias operated according to koyanagi with or without modifications were presented. The surgical technique was described. Complications like fistula, stenosis, dehiscence and urethral diverticulum were studied.
Results and Limitations: Nineteen patients were included and presented in a chronological manner. The first four patients were operated according to the original koyanagi technique. The next 15 patients were operated according to koyanagi and the urethroplasty was covered by a tunica vaginalis flap. In 10 of them, glanduloplasty was done primarily and in the last 5 patients, glanduloplasty was done as a secondary procedure. The complication rates were 100% in the original koyanagi group, and 0% in the subset were glanduoplasty was deferred.
Conclusions: Systematically covering the urethroplasty with a tunica vaginalis flap and deferring the glanuloplasty might ameliorate the results of the koyanagi technique.

1. Introduction

Surgical treatment of severe hypospadias was always known to be associated with elevated rate of complications. Several surgical methods have been described as one stage or as multi-stage repairs. The koyanagi technique was first described in 1984 [1]. However, the associated postoperative complication rate was high reaching up to 50–60% [2,3]. The postoperative complications following the koyanagi technique were largely associated with vascular insufficiency [4].

Recent studies have shown that backing up the reconstructed urethra and securing a large distal urethral meatus might be associated with a better outcome [2]. However, complications were still recorded. The glanular urethra tends to narrow down with time, increasing the urethral flow pressure [2]. In our center, our experience had progressively evolved since we initiated hypospadias surgery. We progressively modified the original koyanagi technique. The first modification was systematically covering the urethroplasty by a tunica vaginalis vascularized flap. The second modification was opting for a deferred glanuloplasty. The aim of this article is to present our series of children operated for a proximal hypospadias according to koyanagi and to discuss our modifications. We also discussed the amelioration we achieved following those modifications.

2. Materials and methods

We retrospectively analyzed children operated between June

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2005 and June 2018 by the same pediatric urologist, according to koyanagi technique. Preoperatively, all children were screened by karyotype in order to rule out a primary chromosomal defect behind the disease of sexual differentiation. In addition, all patients underwent cystoscopy or cystography in the aim of searching for mullerian remnants, more specifically a prostatic utricle. All patients were followed by our endocrinology team. Patients were followed up until July 2019. Follow up was done clinically. By carefully questioning the parents and examining the children, we systematically searched for straining, retention, dribbling, fistula, dehiscence, urethral diverticula and stenosis. All had represented our main outcome. Cosmetic results were assessed subjectively. A bladder scan was done only when indicated. Different surgical technical aspects were analyzed. Complications were matched to the different surgical modifications. All patients with proximal hypospadias operated according to other techniques were excluded. Statistical analysis was done using Fisher exact test. A *P* value less than 0.05 was considered significant.

2.1. Surgical technique

At the beginning of our experience, the original technique by Koyanagi was replicated [4].

2.2. The koyanagi technique

The incision lines were done as in the original koyanagi description. Then, with an 8 Fr trans-urethral catheter in place, degloving of the penis was done. The urethral plate was released from its bed but not excised. The urethral plate was divided at the level of the corona. An erection test was always done. Chordae were corrected by degloving and urethral plate release [4]. After separating the two limbs of the "Y", the flaps were flipped over the front side. Urethroplasty was then initiated by constructing first the posterior neo-urethral wall with a running 7/0 P-dioxanone suture. The same goes for the anterior wall afterwards. Then the glans was incised with wide preparation of glans wings in order to position the urethroplasty inside. The meatus was created at the tip of the glans by 7/0 P-dioxanone separate sutures. At the beginning of our experience, the glans was systematically closed. Cutaneous closure was done using Byars flaps. Supra-pubic catheters were not used systematically. A hydrocolloid dressing was used at the end. Scrotoplasty was done at the end when indicated.

With time, two surgical modifications were added to the original koyanagi. First, we covered the anterior suture line by a vascularized tunica vaginalis flap. Second, we opted to cease practicing the glanuloplasty, leaving the patients in coronal hypospadias. Fig. 1 describes the surgery.

In cases where glanuloplasty was deferred, patients were followed with their coronal hypospadias until we judge that glans penis is large enough to accommodate a urethroplasty. Then, after patient (or parent) consent, they were operated according to the technique of Koff, described in Fig. 2.

3. Results

A total of 19 children were operated in one center. The median age at operation was 14 months (ranged between 9 and 25 months). Nine patients had a penoscrotal hypospadias. Seven patients had a perineal hypospadiasand two patients had a proximal penile hypospadias. Three patients had a DSD. Two of them had a mixed gonadal dysgenesis, and one had a partial androgen resistance. All of the patients with a DSD had undescended testis (unilateral or bilateral) and all of them had penoscrotal transposition. Seven patients had an undescended testis, 4 of them were bilateral. Five patients had penoscrotal transposition. Patients characteristics, the degree of severity, associated anomalies and the surgical modifications used were summarized in Table 1. No hormonal stimulation was given. The median duration of urethral stent was 10 days and it ranged between 7 and 23 days. Supra-pubic catheter was inserted primarily in two patients, one of them because the patient had a bilateral congenital obstructive mega-ureter. Follow up ranged between 12 and 126 months with a median of 61 months. However, for patients who had had a tunica vaginalis flap done and with a deferred glanuloplasty, the follow up was between 6 and 22 months with a median of 17 months.

3.1. Postoperative evolution

Two of our patients were symptomatic postoperatively. Both had meatal stenosis. One patient had acute urinary retention 4 months after the surgery. The second had been straining chronically. Nine of our patients had no complications at all (47%), leaving us with 53% complication rate, as seen in Table 2. The absence of tunica vaginalis flap was associated with 100% complication rate. All patients who did not have glanduloplasty and who had a tunica

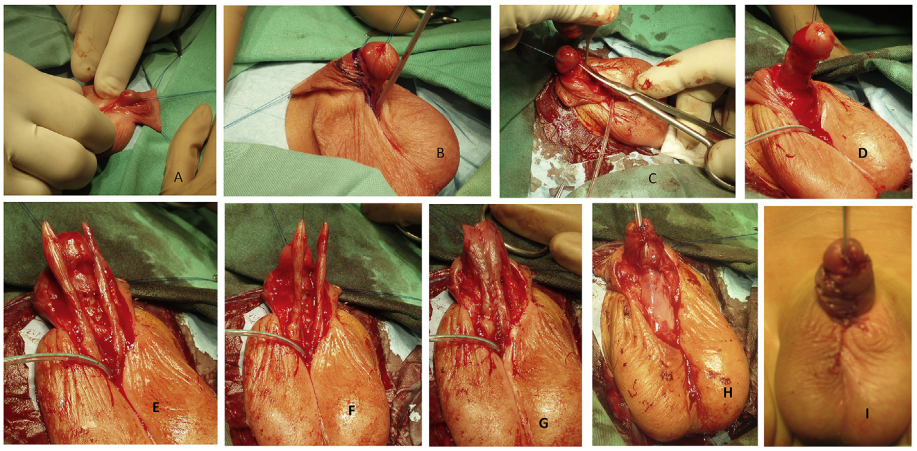

Fig. 1. Koyanagi technique.
A: Traction sutures are in place; B: incision lines are well demarcated; C: urethral plate excision in order to achieve a straight penis as in D; E-F: mobilizing preuptial flaps; G: urethral plate reconstruction (posterior plane); H: urethroplasty was done; I: final results.

Fig. 2. Advancement urethoplasty – koff technique with glanuloplasty.
A-B: aspects 4 years after the koyanagi repair, and before the urethral advancement; the asterisk represents the subcoronal meatus. C: incision line for urethral advancement (Koff) and glanuloplasty. D: urethral mobilization. E: preparation of glans wings. F: glanuloplasty. G-H: final results.

Table 1
Summarizes the profile of our population.

Characteristic	Number of patients affected
Hypospadias severity	
posterior penile hypospadias	2
Penoscrotal hypospadias	9
perineal hypospadias	7
Associated anomalies	
penoscrotal transposition	5
bilateral obstructive mega-ureter	1
mullerian remnant	2
anomaly of testicular descend	7
Congenital left diaphragmatic hernia	1
inguinal hernia	4
Associated DSD	
mixed gonadal dysgenesis	2
Partial androgen insensibility	1
Surgical modification	
Group 1: glanduloplasty with no use of tunica vaginalis	4
Group 2: glanduloplasty with tunica vaginalis flap	10
Group 3: No glanduloplasty with tunica vaginalis flap	5
Meatal location in each group	
Group 1	1 perineal, 2 penoscrotal, 1 posterior penile
Group 2	5 perineal, 4 penoscrotal, 1 posterior penile
Group 3	2 perineal, 3 penoscrotal

vaginalis flap covering their urethroplasty did not have any complications. This association was statistically significant with $P = .005$. The distribution of complications with regard to the surgical modification was summarized in Table 3.

Supra-pubic catheter was inserted postoperatively in two patients, one of them for a transitory retention and the other the retention was secondary to distal glanular urethral stenosis.

Table 2
Postoperative early and delayed complications.

Complications	Number of patients: 10
Isolated stenosis	2
Isolated fistula	4
Urethral divertucula with stenosis	1
Fistula with dehiscence	2
Fistula with stenosis	1

3.2. Fistula formation?

It was the most common complication in our series, representing 70% of complications. It was isolated in 4 cases and associated with stenosis, necrosis or dehiscence in the remaining 3 cases. Five fistulas were coronal; the remaining two were proximal. According to the presentation, fistula was managed either by redoing the distal urethroplasty or by direct fistula repair. The patients who had a redo urethroplasty, one was managed by duplay, two were managed by Koff advancement urethroplasty and two others were managed by Mathieu urethroplasty. We last follow up with the last patient (who had a proximal fistula associated with necrosis). Only patients who had a Mathieu urethroplasty needed a second redo operation for fistula recurrence. All fistulas happened in cases where glanuloplasty was done.

3.3. Stenosis

Stenosis was the second most common complication representing 20% of complication rate, reported in 4 patients. Two patients had an isolated stenosis, one patient had it in association with a probably secondary urethrocele, and the last patient had it in association with a fistula. All the strictures were glanular.

One of the isolated stenosis was so severe, presenting with acute urinary retention, necessitation an urgent meatotomy and

Table 3
Details of patients with complications.

Modification	Number of patients with complications	Details
A glanuloplasty with no use of tunica vaginalis flap	4 out of 4	2 patients with fistula, 1 patients with Fistula and stenosis, 1 patient with fistula and dehiscence
A glanduloplasty with a tunica vaginalis flap	6 out of 10	2 patients with stenosis, 2 patients with fistula, 1 patient with urethrocele and stenosis, 1 patient with fistula and dehiscence
No glanuloplasty with a tunica vaginalis flap	0 out of 5	NA

suprapubic catheter insertion. This same patient was then re-operated according to duplay. However, his postoperative evolution was remarkable by a distal urethra stenosis. The patient underwent another meatotomy, and a first stage buccal mucosal graft according to Bracka. The second patient with isolated stenosis presenting as chronic straining was easily managed by two dilatation sessions. The patient with stenosis associated to a fistula, was managed by advancement urethroplasty according to koff. The last patient with stenosis associated with a urethrocele was managed by urethrocele/stricture excision and primary repair. All strictures happened in cases where glanuloplasty was done.

3.4. Secondary glanuloplasty with urethral advancement

Five patients did not undergo primary glanuloplasty and hence they had a coronal hypospadias. Two of them, the eldest, underwent a second stage urethral advancement and glanuloplasty (according to koff), at the ages of 5 and 6 years, with a favorable postoperative evolution (Fig. 2). Those two patients had been followed for 19 and 22 months with no complications reported. The other 3 patients, now between 3 and 4 years old, according to our clinical impression, still have a small glans so the indication of a urethral advancement according to koff is temporarily suspended.

3.5. Reoperation rate

Nine of our patients did not need a reoperation (47%). Sixty percent of patients who needed a reoperation, were re-oprated once. The others needed more than one procedure. One patient needed 4 operations.

4. Discussion

Proximal hypospadias repair represented one of the most challenging operations even for the expert hyposadiologists [5]. Complications, what so ever the technique used, were high and troublesome [6]. Because koyanagi technique used a parameatal vascularized flaps [7], it was first described as the technique to treat most sever, even perineal, hypospadias —contrary to duckett repair- with or without peno-scrotal transposition, in one stage [8]. Moreover, this technique lacked the circular anastomosis at the base of the urethroplasty which theoretically decreased the stenosis rate [8,9]. In addition, using the koyanagi repair [7], no bulking and no apparent penile torsion were noted [10]. Moreover, following this technique, there was always enough para -meatal tissues for the urethroplasty [8]. For all those reasons, it gained popularity relatively rapidly. According to koyanagi, primitive results were highly promising, with a success rate of 87% [11]. Several authors, including Koyanagi, afterwards were able to replicate the technique but were unable to achieve the same results. Complication rates were reported high, between 47% and 62% [2,3,12]. One of the most used modifications was the Hayashi technique. However, complications were very similar to those of the original koyanagi technique [3] in nature and in incidence.

Complications reported after those types of surgeries were stenosis, fistula, urethral diverticulum, meatal regression and dehiscence [13]. Table 4 had summarized the complications reported. The complications were always frequent so that many pediatric urologists switched towards two-staged urethroplasty, like the Bracka technique [14]. Many hypotheses had linked those complications to vascularization deficiency of mostly the distal para-meatal flaps, which was probably correct [15]. With all the experts' efforts in order to create well vascularized flaps, no great advances were recorded and this technique -modified or no-was always associated with troublesome postoperative evolution [15]. However, despite the high complication rate, patients operated according to this technique did not have a high re-operation rate [3]. Kang et al. added the tunica vaginalis flap systematically, as a second layer covering the urethroplasty. They, however, used Hayashi modified koyanagi and glanuoplasty was systematic. With their modifications added to the Hayashi modified koyanagi, complications rate was at 20% comparing to 30% without the use of the tunica vaginalis flap [16]. Tunica vaginalis flap was also used by other groups, but only for treatment of urethra-cutaneous fistulas

Table 4
The complications reported in the literature.

Author	Procedure	# patients	# patients with complications	Type of complications
T. KOYANAGI et al., 1988	original koyanagi	8	5/8 (63%)	Fistula, dehiscence, infection
T. Koyanagi et al., 1994	Original Koyanagi	70	33/70 (47%)	Dehiscence, Fistula, stenosis
Snow BW and Cartwright PC 1994	Yoke hypospadias repair	4	50	fistula
EMIR et al., 2000	modified koyanagi, meatal based yoke	20	6/20 (30%)	Fistula, dehiscence
Y. HAYASHI et al., 2001	hayashi -modified koyanagi	20	6/20 (30%)	Fistula, stenosis
Elisangela de Mattos e Silva et al., 2009	original koyanagi	26	16/26 (62%)	stenosis, Fistula, urethral diverticulum dehiscence
Catti et al., 2009	Original Koyanagi hayashi -modified koyanagi	26 31	16/26 (62%) 19/31 (61%)	dehiscence, stenosis, Fistula, urethral diverticulum
Alexis A, et at. 2011	hayashi -modified koyanagi	21	16/21 (76%)	Fistula, dehiscence, chordae
Yuhong et al., 2016	modified koyanagi	21	5/21 (24%) Suivi 1 an	Fistula, necrosis

complicating mid-proximal hypospadias surgery. It was reported with a success rate reaching 100% [17]. We insisted on the importance of the well vascularized flaps, and the presence of a tunica vaginalis layer covering the urethroplasty. In addition, we hypothesized there was also the glans pressure factor. This hypothesis was established duo to our personal observation that patients with small glans had strained the most, and had the most complicated post-operative courses, even after adequate glans wings preparation. So, we ceased to do glanuloplasty, leaving our patients with coronal hypospadias. Moreover, our planned second stage for urethral advancement and glanuloplasty was a relatively simple procedure, koff procedure. Interestingly, and contrary to what might be thought, this procedure was rather easy and smooth and the surgical planes were easily dissectible. The progressive establishment of this hypothesis was the reason why, only in our last 5 patients, we opted to cease to do glanuloplasty and the tunica vaginalis flap was systemically practiced, eliminating the complications but "potentially" transforming koyanagi into a two staged procedure. We insisted on the fact that this transformation is possible and not eventual since the decision of doing a complementary glanduloplasty in the absence of functional deficiencies, in cases were the urinary jet is forward with no straining, would probably be not necessary and be only cosmetic [18].

5. Conclusion

Our modification is promising. We had a clear amelioration of our results. Taking the surgeon learning curve into consideration, our modified koyanagi repair might become finally the treatment of choice for proximal hypospadias. Larger, randomized studies, and a longer term follow up - especially after puberty -are however needed.

Ethical statement

The authors has no ethical statement to be declared.

Author statement

All authors have made substantial contributions to all of the following:

(1) The conception and design of the study, or acquisition of data, or analysis and interpretation of data.
(2) Drafting the article or revising it critically for important intellectual content.
(3) Final approval of the version to be submitted.

Declaration of competing interest

The author acknowledge that there is no conflict of interest.

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Visual abstract

Supplementary data to this article can be found online at https://doi.org/10.1016/ijpam.2020.06.005.

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