Role of tubularization of urethral plate in development of urethrocutaneous fistula post hypospadias repair

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INTRODUCTION

The term hypospadias is derived from the Greek words hypo meaning below and spadone meaning hole.1 Hypospadias is considered the second most common congenital malformation of the genitourinary system, which affects 1:300 newborn males.
It is defined as an abnormal urethral meatal location, found on the penis distally in 60%–65% of cases (glans penis, coronal, or subcoronal), on the middle (midpenile) in 20%–30% of patients, or proximally (posterior penile, penoscrotal, scrotal, or perineal) in 10%–15% of newborns.\textsuperscript{[1,3,5]} The severity of a case and the association of a chordee, which is the ventral curvature of the penis, is strongly correlated with the proximity of the opening, with the perineal opening being the most severe.\textsuperscript{[1]} There are consequences if the hypospadias is left uncorrected, most importantly a compromised fertility, and in severe cases, sexual intercourse may not be possible.

Correcting the defect mandates a surgical repair, which is the only definitive management. The repair is usually done at 6 months of age. The aim of the procedure is to straighten the penis after removing the chordee, transfer and fix the position of the urethral meatus on the glans, make sure that the caliber of neourethra is appropriate and allows a good stream of urine, sufficiently cover the phallus with skin, and optimize sexual function in the future.\textsuperscript{[6]}

Urethroplasty, which is the surgical repair of hypospadias, is done either by tubularized urethral plate (TUP), skin flaps, or grafts. Historically, flaps have been the most commonly used technique. However, recently, TUP with or without incision (tubularized incised plate [TIP]) has been adapted for correcting distal, proximal, and redo hypospadias. TIP offers a simpler alternative with lower complication rates and better cosmetic results.\textsuperscript{[7]}

TIP like any other procedure has some complications, most importantly urethrocutaneous fistula, meatal stenosis, recurrent urethral stricture, wound infection, and penile rotation. Urethrocutaneous fistulae in particular are the highest occurring complication with unknown etiology. Several hypotheses were raised but none confirmed.\textsuperscript{[2]} Thus, a question was raised. Will an incision of the urethral plate, just before tubularization, increase the incidence of a fistula? Therefore, the aim of our study is to compare the incidence of urethrocutaneous fistula between incised and unincised urethral plates.

**METHODOLOGY**

**Cohort description and study criteria**

The data were collected retrospectively by reviewing records of pediatric patients, who had hypospadias surgery at a tertiary care center in Jeddah, Saudi Arabia, which contains 750 beds and has an accredited training program for urology residents, between January 2000 and December 2012. Three hundred and ten files were reviewed; patients who underwent the TUP (incised/nonincised) surgical technique were included ($n = 106$) in the study. Cases that had surgical repair by different techniques such as meatal advancement glanduloplasty and Mathieu repair procedures were excluded from the study. Patients who were diagnosed with hypospadias but were not treated were also excluded as well as those who underwent meatotomy and chordee release alone. Furthermore, we excluded those who exhibited complications from surgeries done before the assigned period and patients with disorders of sexual development.

Data was collected from patients’ medical records using a predesigned data collection sheet, and the information extracted was previous history of hypospadias repair, type of hypospadias, the presence of concurrent chordee, and whether both testes were descended into the scrotum. Another section inquired about operational details such as the date of operation, name of the surgeon, dates of admission and discharge, duration of the surgery, amount of blood loss, type of suture and stent that had been used in the tubularization during surgery, date of catheterization and removal, as well as whether the urethral plate was operated on by incision or nonincision techniques. Furthermore, we also investigated hospital and postoperative complications such as urethrocutaneous fistula, meatal stenosis, penile swelling, persistent chordee, urinary tract infection, wound infection, hematoma, and others. All confounding factors were standardized including type of catheter, size of the catheter, duration of the catheter, type of the suture, and technique of surgery; however, the only factor which was not standardized was the surgeon, some of them were done by consultants, and most of them were done by senior residents supervised by the consultant.

**Surgical procedure**

**Tubularized plate urethroplasty**

Under general anesthesia, the penis was degloved down to the penoscrotal junction, and then, a U-shape incision was made 2 mm proximal to the meatus. After that, concurrent chordee correction was made by dorsal plication if needed. Then, an artificial erection test was performed to ensure adequate straightening of the phallus.

On the ventral glanular surface, two longitudinal parallel incisions were made between the urethral plate and glanular wings for the creation of the surgical plate. After that, a dissection was made along the corporal body to separate the glans wings. Then, tubularization of the urethral plate over an 8-F stent was done. A point that was emphasized on the distal end was not closed too tightly to prevent stenosis. Finally, creation of a vascularized dartos flap was done to cover the neourethra as a second layer. Then, the skin was closed. The stent was secured for postoperative drainage approximately 5–10 days.
Incision of urethral plate (Snodgrass modification)
A midline incision was made through the entire thickness and length of the urethral plate down to the corporal tissue after the chordee was corrected. Then, the plate is separated in the midline dorsally to provide the caliber needed for the neourethra. Finally, the plate was sutured over 6 or 8-F stent with 7-0 polydioxanone. Furthermore, to prevent stenosis, the distal end was not closed in a tight fashion leaving the neomeatus oval.

Statistical analysis
Simple descriptive statistics were produced using SPSS program version 21.0 (SPSS, Inc., Chicago, Illinois, USA). Chi-square test was used to compare the incidence between the two interventions, and P < 0.05 was considered statistically significant. The results were recorded as P values, percentages, medians, and interquartile ranges (IQRs).

RESULTS
We included 106 patients, with a median age of 2 years (IQR = 3 years), who had TUP during the study period. In Group A, we identified 87 patients (82%) who underwent TIP, and in Group B, there were 19 patients (18%) who underwent a urethral plate repair without incision. A previous history of failed hypospadias repair was reported in 26.4% of the patients (n = 28). Patients were classified according to the hypospadias position: proximal in 20.8% (n = 22), middle 13.2% (n = 14), and distal in 65.1% (n = 69). Ventral chordee was reported in 34% (n = 36). All patients required catheterization post repair with a mean duration of 7 days and a mean hospitalization period of 9 days. There was no statistically significant relationship between types of hypospadias and development of fistula (P = 0.256).

The overall incidence of urethrocutaneous fistula was 34.9% (n = 37). The incidence of fistula in Groups A and B was 36% (n = 31) and 32% (n = 6), respectively. This difference was of no statistical significance. Despite a high fistula rate, only 12 patients (11%) were required to repeat surgery.

The second most commonly reported postoperative complication after fistula was meatal stenosis in 27.4% (n = 29). Twenty-three patients (26%) had an incision in the urethral plate while six patients (32%) had urethral plate tubularization without an incision. Followed by penile swelling affecting 7.55% (n = 8) of cases, seven patients had an incision in the urethral plate and one patient without incision. Hematoma developed in 1.9% (n = 2) of patients, both belonged to Group B. A patient from each group (1.9%) (n = 2) contracted a urinary tract infection. Local infection accounted for 1.9% (n = 2) of children, with a case from each group. Furthermore, there was a statistically significant relationship between prolonged catheterization and the development of meatal stenosis with (P = 0.017).

Seventy-two patients (67.9%) who did not develop meatal stenosis in the postoperative period had a mean duration of catheterization of 7 days while 29 (27.9%) patients who suffered from meatal stenosis postoperatively had a mean period of 8 days. Unfortunately, there were five patients who lost to follow-up (4.7%). However, there was a statistically significant relationship between the type of hypospadias and concurrent chordee (P = 0.002). Of the twenty-two patients who had proximal hypospadias (20.8%), 14 (63.6%) had concurrent chordee. In addition, from the 14 patients affected by midshaft hypospadias (13.2%), six of them (42.9%) had chordee, whereas of the 69 patients (65.1%) who had distal hypospadias, 16 (23.2%) had chordee.

There was a statistically significant relationship between type of urethral plate repair (incised or unincised) and the development of hematoma (P = 0.002). On the one hand, out of 87 patients who had an incision of the urethral plate, none of them developed hematoma 0% (n = 0). On the other hand, out of nineteen patients treated with unincised urethral plate repair, two developed hematoma 11% (n = 2).

DISCUSSION
Hypospadias repair is considered one of the technically challenging procedures; in fact, it needs well-trained surgeons with skilled hands and experience to correct it. The goal in correcting hypospadias is to obtain a functionally and cosmetically normal penis. In this study, the most common complication observed in our patients was urethrocutaneous fistula (34.9%); however, despite high fistula rates in the first 4 weeks postoperatively, we observed that in the follow-up period within a year, most of these fistulae closed spontaneously without any intervention. Only 12 patients (11%) needed to repeat the surgery, with a success rate of 89% (n = 94). The incidence of urethrocutaneous fistula observed in our study was significantly higher than what is reported in international journals which is around 5%. This can be explained by the fact that 26.4% of our patients had previous hypospadias repair which increased the complication rate.

To avoid this complication, we recommend adapting Snodgrass’ technique mentioned in his study by routine interposition of a barrier layer between the neourethra, overlying glans, and shaft skin closures. This coverage is best achieved using a de-epithelialized dartos pedicle flap instead of relying on local tissue. Snodgrass reported that during surgery, it is important to confine the dorsal incision to the urethral plate and that scarring could be...
due to incising distally into the glans, so suturing the plate too far distally should be avoided as it will constrict the neomeatus. Furthermore, during and after glanuloplasty, care must be taken to avoid meatal stenosis by routine fixation of stent tube in the neourethra.\cite{9,10} In addition, a statistically significant relationship between meatal stenosis and duration of catheterization was found ($P = 0.017$). It has also come to our attention that most of the complications occurred from the TIP urethroplasty were directly related to technical factors rather than patient's anatomy.\cite{11} We hypothesized that long-standing postoperative catheterization could be one of the predisposing factors that would make the urethral plate susceptible to fibrosis and scar formation, resulting in meatal stenosis. Another postoperative complication is penile swelling which affected eight patients (7.55%), with predominance in Group A, which comprised seven patients. We also concluded that incision of the urethral plate may lead to inflammatory response resulting in edema. Other complications such as urethral stricture, urethral diverticula, and wound dehiscence were not observed in our patients. Baskin \textit{et al.}, after their study of fetal hypospadias specimens, concluded that rich vascularity of the urethral plate is vital to create a healthy neourethra without postoperative stricture.\cite{12} Finally, it is crucial to focus on the techniques of hypospadias surgical repair to help limit postoperative complications.

Our study also showed that there is a strong association between proximal type of hypospadias and concurrent chordee. It was also concluded that the high fistula rate could be attributed to the involvement of training residents in performing these procedures. Therefore, a difficult surgical procedure such as hypospadias repair should not be performed by residents and should only be restricted to consultants and fellows. However, further studies are needed to compare outcomes.

**Limitations**

We experienced some limitations during our research, such as the number of patients included in this study which was less than what was expected. We attributed this to the elimination of any patient with missing information, and patients who did not exclusively undergo repair by tubularization of the urethral plate; instead, they had a previous history of hypospadias repair by another technique such as meatal advancement, glanuloplasty, and Mathieu technique, which we believed may affect our results. Finally, there were many cases that got canceled after booking for hypospadias repair due to either upper respiratory tract infection or parents’ refusal of the procedure.

**CONCLUSION**

The incidence of urethrocutaneous fistula reported in this study was significantly higher than internationally reported incidence rates. This can be explained by the fact that 26.4% of patients had a previous hypospadias repair which increased the complication rate; moreover, the procedure was done at a training institution. Incision of the urethral plate did not affect the occurrence of the urethrocutaneous fistula; therefore, it is not considered a risk factor in this cohort group.

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**Conflicts of interest**

There are no conflicts of interest.

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