

Mathieu vs urethral plate tubularization in circumcised Megameatus intact prepuce repair: A prospective randomized comparative study

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ABSTRACT

Objective: The objective of this study is to evaluate and compare urethral plate tubularization vs Mathieu in circumcised Megameatus intact prepuce (MIP) repair. Many techniques were described for MIP, which account for 5% of hypospadias cases and usually diagnosed at time of, or even after circumcision.

Material and methods: Forty-six circumcised MIP cases were prospectively enrolled in this prospective study, which was carried out in April 2017 and March 2020. Patients were randomly allocated into two groups. Group one operated by simple urethral plate tubularization and group two by the Mathieu technique. Hypospadias objective scoring evaluation (HOSE) scores, success rate, operative time, and the need for relaxing incision or scrotal flaps for skin closure were compared.

Results: Forty-three circumcised cases (22 in group one and 21 in group two) completed at least 6 months of follow-up. Ages ranged from 12 to 39 months (mean 18.06 ± 6.35) in group one and from 10 to 32 months (mean 19.5 ± 7.14) in group two.

There was no significant difference between cases with accepted outcome based on HOSE scores (≥ 14) of the two groups (P value = .942). Three fistulae and one meatal stenosis were the complications in group one (18.2%). In group two, two patients complicated with fistula (9.5%) (P value = .674). Significant differences were present only in the operative time (P = .001) and in the need of relaxing incision or scrotal skin flaps (P = .012) both were more in group two.

Conclusion: Mathieu and tubularized incised plate urethroplasty both are good options for circumcised MIP repair.

Keywords: Hypospadias; prospective studies; surgical flaps; urethra.

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Introduction

Megameatus intact prepuce (MIP) represents about 5% of hypospadias cases.¹ It is characterized by a complete prepuce, instead of the dorsal hood in other hypospadias variants, deficient spongiosum, and a wide thin urethral plate.^{2,3} It is usually discovered at time or even after circumcision.⁴ The need for repair in these cases may be cosmetically (the large wide or fish mouth meatus) and/or functionally (abnormal urinary stream).

Many surgical techniques were prescribed to repair this special variant of hypospadias: pyramid procedure, tubularized incised plate urethroplasty (TIPU), glans approximation procedure (GAP), and parameatal flap (Mathieu).⁴⁻⁶ Pyramid procedure is the gold standard for MIP repair with good results in circumcised and uncircumcised patients.⁷⁻¹¹ Other techniques that used in the distal hypospadias repair, e.g., TIPU, Mathieu flap, and GAP, used in MIP repair with comparable results.¹⁻³ We previously conducted a study

on MIP (uncircumcised and circumcised); in this study, patients were operated by pyramid procedure and GAP (under publication now, until now not accepted for publication). Most of series describing these techniques were retrospective in nature, including more than one technique, without randomization and including a small number for each technique. TIPU is a universally accepted technique in majority of anterior and middle hypospadias cases.⁴ Application of this technique in MIP repair was described with good results,²⁻⁴ but it carries the liability to injure the lateral edges of the plate that makes the repair more challenging.¹ Cendron¹ suggested that augmentation of this wide thin plate with Mathieu flap gives better results. But, Hill et al⁷ proposed that previous circumcision in boys with MIP could limit the use of this surgical option as scarring would be present at the site of the flip-flap. Objective evaluation of hypospadias repair can be done with different validated systems and scores: hypospadias objective scoring evaluation (HOSE) and pediatric penile perception score.^{9,10} HOSE evaluates both the surgical and cosmetic outcomes of hypospadias repair. It incorporates five main domains: meatal location, meatal shape, quality of urinary stream, state of erection, and fistula presence and complexity. It is a validated scoring system, ranged from 5 up to 16 points. It is graded as accepted and not accepted with points score ≥ 14 is accepted.⁹

We aim, in this work, to detect if Mathieu repair for circumcised MIP has a higher complication rate than urethral plate tubularization or no.

Materail and Methods

Study Design

This prospective randomized study was conducted in Department of Urology, Zagazig University Faculty of Medicine between April 2017 and March 2020. Circumcised MIP cases (Figure 1) who presented to the outpatients' clinic during the study period were prospectively enrolled in this study. We excluded uncircumcised cases, redo cases, and cases with other hypospadias variants. The nature of hypospadias as MIP in cir-

Main Points

- Megameatus intact prepuce is not uncommon hypospadias variant.
- Mathieu and tubularization techniques are among the armamentarium for Megameatus intact prepuce repair.
- There is no significant difference between the two techniques regarding the hypospadias objective system evaluation scores.
- Mathieu repair in circumcised Megameatus intact prepuce needs additional skin coverage techniques.



Figure 1. Circumcised Megameatus intact prepuce.

cumcised cases was recognized from the history by the parents stating that a complete prepuce was present before circumcision, and this large wide meatus appeared just after it. Our internal review board approved this study on February 24, 2017 (IRB number 7254). Enrollment of patients in this study was done after obtaining a signed informed consent from the patients' parents.

Sample Size

Due to the low incidence of MIP cases and the absence of previous studies reporting the outcomes of the chosen techniques in a sufficient number to calculate sample size, we calculated the sample size by formula for infinite and finite population.

For infinite sample size $SS = [Z^2 p (1 - p)]/C^2$.

For finite sample size $SS/[1 + \{(SS - 1)/Pop\}]$

where SS is the sample size, Z is the given Z value = 1.96, p is the percentage of population = 5%, C is the confidence level 95%, and Pop is the population = 120 hypospadias case.

$$SS = (1.96)^2 \times 0.05 \times (1-0.05)/(0.05)^2 = 72.9$$

$$SS = 72.9/1 + (72.9-1/120) = 45.64. \text{ The sample size is 46 patients.}$$

Randomization was done into two equal groups by random allocation using an excel sheet. We use CONSORT to prepare this RCT manuscript.

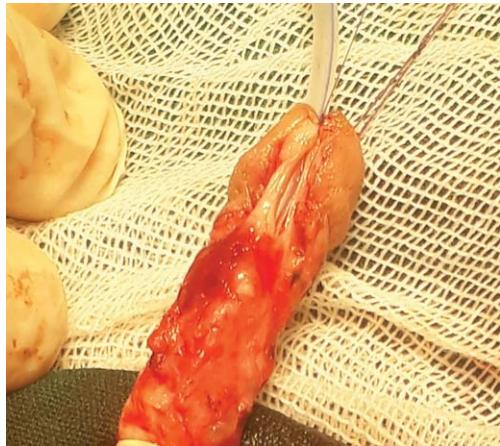


Figure 2. Urethral plate tubularization in circumcised MIP.



Figure 4. Scrotal flaps to cover penile shaft.



Figure 3. Raised Mathieu flap.

In group one (urethral plate tubularization): under general anesthesia, a stay suture in the glans penis was fixed vertically, circumferential incision 5 mm below the coronal margin dorsally and extended ventrally to meet the U-shape incision around the urethral plate was done followed by penile degloving. Penile degloving was done to facilitate harvesting a second layer dartos flap.

After the creation of the two glanular wings, urethral plate tubularization was done in two layers over 8 Fr catheter using 7/0 vicryl (Figure 2). A dartos flap as a second layer coverage was harvested

from the ventral aspect or from dorsal skin. The glans penis was closed in two layers, and finally, skin coverage was done.

In group two (Mathieu): a 1 cm width parametral flap, in a length equal to the distance from the tip of the glans to the hypospadias meatus, was designed. The flap was raised over subcutaneous dartos (Figure 3), and then penile degloving and the glans wings creation were done. The distal end of the flap was tapered into an inverted V shape, to avoid the rounded shape of the meatus, and then the two edges of the flap were sutured to the urethral plate edges by 7/0 vicryl over 8 Fr catheter. The pedicle of the flap was used to cover the suture lines. A two-layer glanular closure and, finally, skin coverage were done.

In both groups, penile straightening was documented by artificial erection test after degloving. When deficient penile skin is encountered, dorsal relaxing incisions (Z-plasty or Heineke-Mikulicz technique) and/or scrotal skin flaps were raised to cover the penis (Figures 4 and 5). The dressing was removed after 3 days, and the urethral catheters were removed after 7 days postoperatively. All cases of both groups were operated by one surgeon.

Follow-up visits were scheduled at 1, 3, and 6 months postoperatively then every year. At the 6th month's postoperative visit, all cases were evaluated by an independent pediatric urologist (not knowing the type of repair technique) with 15 years of experience in hypospadias surgery using HOSE. Scores ≥ 14 were considered acceptable. Parents were asked to provide the assessor with voiding videos (to assess the flow, direction, pattern, caliber, and force) and by photos of their child penis during erection (during clothes changing or sleep).



Figure 5. Z-plasty.

The study outcome measures were as follows:

1. Primary end point was the differences between the HOSE different domains and total scores of both groups.
2. The secondary end points were the differences in operative time, number of complicated cases that needed surgical intervention, and the need for penile skin relaxing incision or scrotal flaps for skin closure between the two groups.

Statistical Analysis

Statistical Package for the Social Sciences (SPSS) version 20 (IBM SPSS Corp.; Armonk, NY, USA) was used for data analysis. Quantitative variables were described using their means and standard deviations if normally distributed, with medians (rang) values if not normally distributed. Categorical variables were described in numbers and percentages. Chi-square test, Fisher's exact test, and student T-test were used to assess the differences between the two groups. We used the Mann-Whitney U test to analyze nonparametric data. Kolmogorov-Smirnov (distribution-type) and Levene (homogeneity of variances) tests were used to verify assumptions for use in parametric tests. The level statistical significance was set at 5% ($P < .05$).

Results

Between April 2017 and March 2020, 67 boys with MIP presented to our outpatient clinics: 54 were circumcised, five declined to be enrolled in the study, three had a previous failed repair, and 13 had an intact prepuce (flow chart in Figure 6). Uncircumcised and redo cases were excluded. Forty-six of the

circumcised cases were enrolled in this study and allocated randomly in the two groups. None of our cases had penile curvature.

There were no significant differences between patients' demographics in the two groups either in the patients' ages or the site of urethral meatus (Table 1). Follow-up ranged from 6 to 23 months (mean 16.2 ± 3.1) in group one and from 6 to 21 months (mean 13.9 ± 4.3) in group two. During the follow-up, three cases were lost (one in group one and two in group two). Forty-three cases complete at least 6 months follow-up (flow chart in Figure 6).

Operative time ranged from 44 to 65 minutes (mean 55.2 ± 6.8) in group one and from 56 to 82 minutes (mean 74 ± 5.1) in group two ($P = .001$). In the second layer coverage, dartos flap was harvested from ventral aspect in 15 cases and from dorsal skin in seven cases in group one. Relaxing incisions and/or scrotal flaps were needed to facilitate skin closure in eleven cases (two in group one and nine in group two) ($P = .01$) (Table 1). For the other 32 people, simple skin closure was done in all cases. All patients were assessed for urinary stream by the voiding videos provided by their parents. Single stream was in 17/22 (86.36%) in group one and 13/21 (85.71%) in group two (P value = .273). Penile straightening was assessed with photos, and a straight penis was present in all cases in both groups (Table 2). No penile curvature or rotations were occurred after repair in both groups. There were no significant differences between the two groups on HOSE assessment either in the total scores, number of cases with the acceptable outcome, or between the individual items of the system. The scores ranged from 13 to 16 in group one and from 12 to 16 in group two. An acceptable outcome (≥ 14) was reported in 35 patients (81.39%) with no significant difference between the two groups (18/22, 81.82% vs 17/21, 80.9%), P value = .942. A slit like meatus and a single stream were more in the group one than in group two (Table 2).

Three urethral fistula and one meatal stenosis were the complications in group one and in group two; two cases were complicated with urethral fistula. The overall success rate was 37/43 (86.04%); 81.8% in group one and 90.5% in group two ($P = .413$). One additional procedure was needed for each complicated case to be managed in the form of fistula closure and meatoplasty.

Discussion

Juskiewenski et al¹¹ were the first to describe the MIP variant of hypospadias in 1983. Six years later, Duckett and Keating¹² gave the detailed description of the MIP variant of hypospadias and adopting the pyramid procedure for repair of these cases.

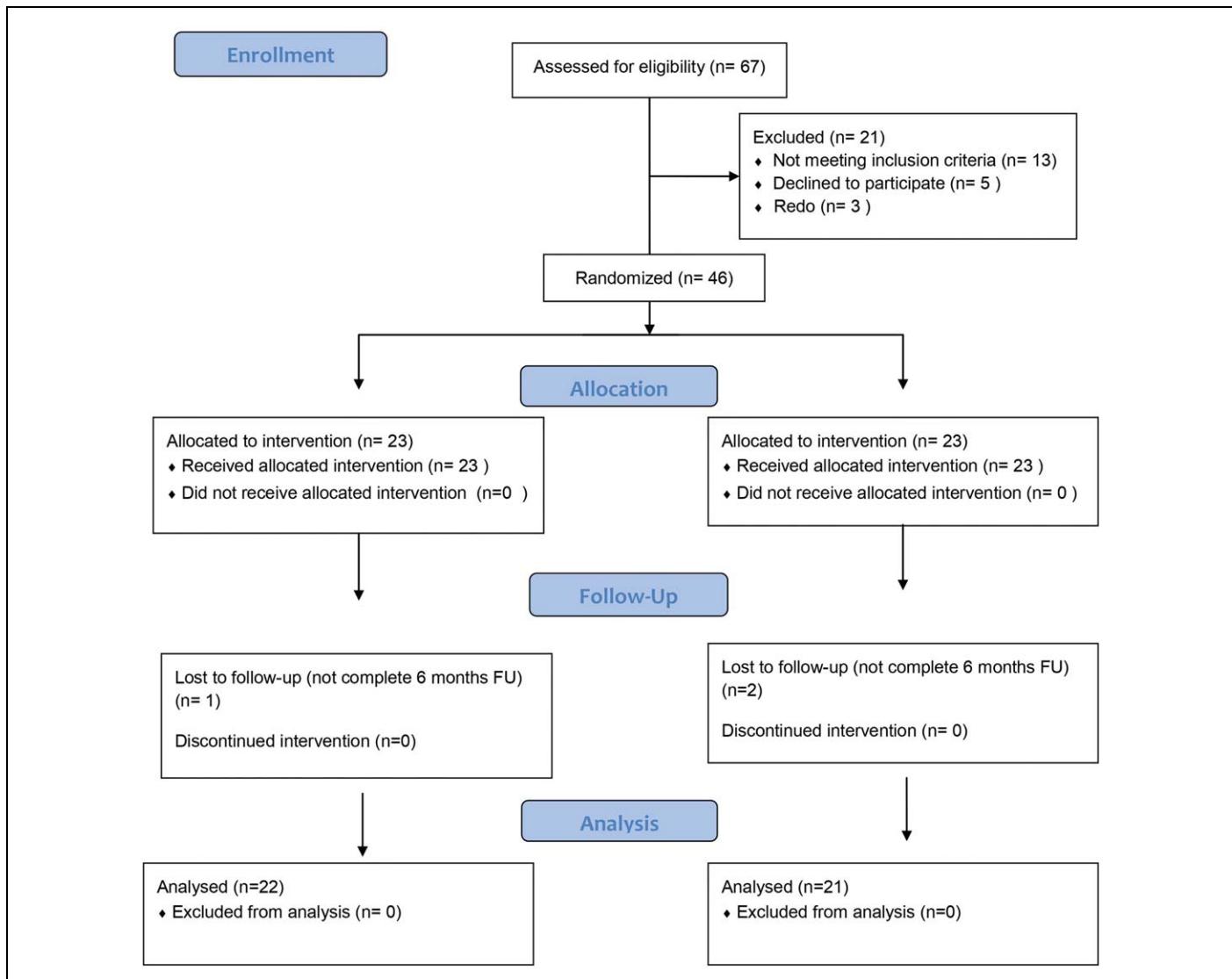


Figure 6. Flow chart.

The true incidence of these cases is not well known, as not all cases reported and some surgeons find the urethral defect is insignificant to be repaired, and the appearance of the urethral meatus varied from just a wide urethral meatus to a large fish-mouth extending to or below the coronal margin.¹

Many years ago, when this variant was discovered at the time of circumcision, the procedure was aborted, and the parents were advised to preserve the prepuce to be used in repair.^{4,13} Duckett and Keating in 1989,¹² Snodgrass and Khavari in 2006,⁴ and Pieretti et al in 2009¹³ documented that there were no differences in the success of MIP and anterior hypospadias repair between circumcised and noncircumcised cases. Based on these series, the concept of circumcision aborting in these

cases was changed although it would still be preferable to do the circumcision at the time of the MIP repair.

In our series, there were no significant differences between the two groups regarding HOSE scores or the complications rate. Significant differences were only present in the operative time and in the need for relaxing incision and/or scrotal flaps to facilitate skin closure. The significant difference in the operative time between the two groups was due to the time consumed in harvesting of the flap, and suturing it to urethral plate in two lines. Additional factor that prolonged the operative time in group two was the need for relaxing incision and scrotal flaps in nine cases in group two vs only two in group one. HOSE assessment in our studies revealed an acceptable

Table 1. Patients' Demographic and Operative Data

	Group One (22)	Group Two (21)	P value
Age	12-39 months (median 18)	10-32 months (median 19)	.551*
Type of MIP			
Glandular	0	0	
Coronal	13	13	
Subcoronal	9	8	1†
Operative time	44-65 minutes (mean 55.2 ± 6.8)	56-82 minutes (mean 74 ± 5.1)	.001*
Second layer			
Ventral	Dartos flap	Pedicle of the Mathieu flap	
Dorsal	15 (68.18%)		
	7 (31.81%)		
Skin coverage:			
Simple skin coverage	20 (90.9%)	12 (57.14%)	.011‡
Skin mobilization	2 (9.1%)	9 (42.86%)	
Relaxing incision	2	6	
Scrotal flaps	0	3	
Follow-up period	6-23 months (median 16.5)	6-21 months (median 14)	.253*
Megameatus intact prepuce.			
*Student t-test.			
†Fisher's exact test.			
‡Chi-square test.			

outcome (≥ 14) in 35 patients (81.4%) with no significant difference between the two groups (81.81% vs 80.95%), P value = .942. Bagnara et al¹⁴ retrospectively evaluated 310 patients with distal hypospadias, and 280 of them were repaired with *in situ* tubularization of the urethral plate. HOSE system was used to evaluate the results in 234 (75.5%) patients. Acceptable outcome (scores ≥ 14) was achieved in 90.6% of these cases, and a successful repair was reported in 88.1%. In Wang et al's¹⁵ systematic review and meta-analysis of studies comparing Mathieu and TIPU repair of primary hypospadias, there were no significant differences in the complication rates, and TIPU had a significant better cosmetic outcome when objective scoring systems were used.

By reviewing the literatures describing repair of circumcised MIP cases with Mathieu, urethral plate tubularization, or TIPU techniques, our results are comparable to the other series either in the overall success rate or the individual complication rates.^{1,2,4,5} Bar-Yosef et al² reported their experience with 24 circumcised MIP cases repair by different techniques; out of the 11 cases repaired with TIP, meatoplasty was needed in two cases and fistula closure in one case. In the retrospective study of Snodgrass et al⁴ on circumcised and non-circumcised MIP cases, 36 cases (26 circumcised and 10 noncircumcised) completed at least 6 weeks follow-up after TIPU repair. There were no significant differences between the two groups with only one circum-

cised case (3.8%) complicated with fistula.⁴ Cendron¹ retrospectively reviewed 25 cases at Boston Children's Hospital, 10 cases were repaired by urethral plate tubularization (group one) and 15 were repaired with modified Mathieu technique (group two). Better results were reported in both groups than that in our series. In the group one, injury to urethral plate during dissection occurred in six cases; one of them was complicated later on with fistula (10%), wherein in group two, no injury to urethral plate occurred with one case (6.6%) complicated with minor glans dehiscence. Also, better results were reported by Nonomura et al⁵ in 1998 when they reported no urethral complications in their nine cases with MIP: five repaired with Mathieu and four with Onlay urethroplasty preputial flap. Only one case needed excision of excess ventral skin for cosmetic reason.

Up to our knowledge, this study is the first prospective randomized comparative study on circumcised MIP variant of hypospadias. Other reported studies were either a noncomparative studies, retrospective in nature, describing more than one technique on a small number of patients and/or without randomization. Also in all the previously published series, the issue of penile skin closure in circumcised cases was not discussed. The limitations of our study are the subjective evaluation of voiding outcomes, the number of data per cell for statistical comparison is small, absence of comparison with the standard technique (Pyramid procedure), and the short follow-up period.

Table 2. Hypospadias Objective System Evaluation (HOSE) Scores of the Two Groups

	HOSE Score	Group One (22)	Group Two (21)	P
Meatal location				
Tip of glans	4	19 (86.36%)	19 (90.5%)	.674 [†]
Proximal glans	3	3 (13.64%)	2 (9.5%)	
Coronal	2	0	0	
Penile shaft	1	0	0	
Meatal shape				
Vertical slit	2	18 (81.82%)	13 (61.9%)	.146 [†]
Circular	1	4 (8.8%)	8 (38.1%)	
Urinary stream				
Single stream	2	17 (77.27%)	13 (61.9%)	.273 [†]
Spray	1	5 (22.73%)	8 (38.1%)	
Erection				
Straight	4	22	21	
Mild angulation	3	0	0	
Moderate angulation	2	0	0	
Severe angulation	1	0	0	
Fistula				
None	4	19 (86.36%)	19 (90.5%)	.674 [†]
Single proximal	3	0	0	
Single distal	2	3 (13.64%)	2 (9.5%)	
Multiple or complex	1	0	0	
Range		13-16	12-16	.50 [*]
Acceptable/total		18/22 (81.82%)	17/21 (80.9%)	.942 [†]
Median		16	15	
Mode		16	16	

HOSE, hypospadias objective scoring evaluation.

*Mann-Whitney U test.

[†]Chi-square test.

We can conclude that Mathieu urethral plasty and urethral plate tubularization in circumcised MIP are good options with no significant differences either in HOSE scores, or overall or individual complications rates. The only significant differences were in the operative time and in the need for skin mobilization technique or scrotal flaps to cover the penile shaft.

Ethics Committee Approval: Ethical committee approval was received from Zagazig University (2017/7254).

Informed Consent: Written informed consent was obtained from all participants who participated in this study.

Peer-review: Externally peer-reviewed.

Author Contributions: Concept - M.O.; Supervision - M.O.; Data Collection and/or Processing - M.A.; Analysis and/or Interpretation - H.E.G.; Writing Manuscript - A.S., I.R.

Conflict of Interest: The authors have no conflicts of interest to declare.

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