# ORIGINAL ARTICLE

# Modified Pediatric Penile Perception Scoring After Hypospadias Repair: Comparison among Patients, Parents, and Pediatric Surgeons

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## ABSTRACT

**Introduction:** This study is aimed to compare penile perception after hypospadias repair among patients, parents, and pediatric surgeons using the modified Pediatric Penile Perception Score (mPPPS). **Methods:** This is a cross-sectional study conducted on hypospadias patients who underwent the procedure at our institution. Penile perception is evaluated with the online-based mPPPS. Comparison of the mPPPS among each group will be analyzed based on Friedman's and the Wilcoxon test (p<0.05). **Results:** The mean mPPPS in the distal hypospadias group is similar (p = 0.670). The perception of penile length, urethral meatus, glans, skin, curvature, and general appearance are no different among the assessors (p = 1.00; p = 1.00; p = 0.368; p = 0.607; p = 1.00; p = 1.00, respectively). The mean mPPPS in the proximal hypospadias group is lower in the patient and parent groups (patients vs. surgeons p = 0.005, parents vs. surgeons p = 0.007). The perception of penile length, urethral meatus, glans is not different among the assessors (p = 0.007). The perception of penile length, urethral meatus, skin, curvature, and general appearance are lower according to the patients and parents compared to the surgeons (p = 0.028, p = 0.0001, p = 0.015, p = 0.008, p = 0.003, respectively) but the perception of the glans is not different amongst them (p = 0.247). **Conclusion:** The mPPPS can be used as an alternative instrument to evaluate penile perception after hypospadias repair. Perception in the distal hypospadias group is not different among the assessors. However, patients and parents in the proximal hypospadias group are less satisfied than the pediatric surgeons.

Keywords: Hypospadias, Child, Surgery, Perception, Personal Satisfaction

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### INTRODUCTION

Hypospadias is one of the most common congenital anomalies in boys, characterized by an abnormal urethral opening on the ventral side of the penis, associated with penile curvature and a dorsal hood (1). The objective of hypospadias repair is to provide a straight penis with an external urethral opening at the tip that is functionally and cosmetically normal (2).

Despite improvements in hypospadias repair, postoperative long-term complications still pose a threat to complete recovery. Low self-esteem and negative perception of genitalia resulting from the surgical scar are faced by patients (3-6). Unsatisfied patients who have a poorer perception of their genitalia, often exhibit embarrassment, poor school performance (7,8), psychosexual function disorders, and low quality of life in adulthood (8,9).

Instruments have been developed to identify penile perception after hypospadias repair. Schwobel, et al. reported on the postoperative perception of patients and surgeons (10). Weber, et al. developed the Pediatric Penile Perception Score (PPPS), a scoring system that enables a cross-sectional evaluation and combines other measurement tools to compare the penile perception of patients, parents, and surgeons. The PPPS variables are penile length, the position and shape of the urethral meatus, shape of the glans, shape of the penile skin, penile axis, and general appearance of the penis (6).

Regular long-term follow-ups are challenging for hypospadias patients at our institution. Patients with poor socioeconomic status who live in remote districts have difficulty reaching the hospital. In particular, the many restrictions of the Covid-19 pandemic have made patients to be more unreachable for follow-up. As an alternative, we have developed a modified version of the PPPS that patients can complete during an online video call interview. Modifications are made to some variables that cannot be evaluated online. Clinical photographs are sent to multicenter pediatric surgeons for further evaluation. This study is aimed to compare penile perception after hypospadias repair among the patients, parents, and pediatric surgeons using the mPPPS.

#### MATERIALS AND METHODS

This is a cross-sectional analytical study comparing the penile perception of patients, parents, and pediatric surgeons using the mPPPS. The subjects are hypospadias patients who had undergone surgery at our institution between the year 2015 and 2020. The subjects are classified into the distal and proximal hypospadias groups. This study has obtained an ethical license number LB.02.01/X.6.5/309/2020.

Inclusion criteria in this study are hypospadias patients, aged 5 years and above until 18 years old, who had completed the operation at least 6 months before the research was carried out, no functional complications and with parental permission. Exclusion criteria in this study are patients who still have plans for further surgery, children with mental disorders who cannot communicate well, and those who cannot be contacted. All the subjects were re-examined 6 months after they had completed the surgery.

Such scoring examination has not been done before, when it was initiated in 2020, Covid's pandemic had already started. However, we still want to assess the patients we have had performed the surgery on before; therefore, the data for patients who had gone through the surgery 5 years earlier was included.

All patients and parents who fulfilled the recruitment criteria of the study were interviewed online through a video call using the mPPPS questionnaire concerning penile length, shape, and position of the urethral meatus, the shape of the glans, shape of the penile skin, curvature in the penis and general appearance of the penis. A penile axis examination cannot be conducted on a flaccid penis, and not every patient was willing or able to demonstrate an erection. Therefore, we excluded this item from the questionnaire. Patients and parents expressed their satisfaction on a 4-point scale of very dissatisfied (0), dissatisfied (1), satisfied (2), and very satisfied (3).

After the interview, photographic documentation was provided by the parents in four standardized views (oblique, lateral, anteroposterior with the penis held against the abdominal wall, and anteroposterior with the penis held straight). The completed questionnaires along with the photographs would then be sent to eight pediatric surgeons for assessment purposes.

The inter-rater agreement among patients, parents, and the pediatric surgeons was analyzed with Cohen's kappa test. A p-value < 0.05 is considered significant. The comparison of the mPPPS between patients, parents, and pediatric surgeons was analyzed using the Friedman's test and the Wilcoxon test using SPSS 18 software (SPSS Inc., Chicago, IL, USA).

### RESULTS

Of the 90 patients aged >= 5, 15 were reachable for follow-up and agreed to participate (Table I). For the group of patients with the age ranged between 5 and 15 years old, of the 15 participants, 4 had distal hypospadias and 11 had proximal hypospadias. Ten participants had a history of micropenis, and four participants had severe chordee.

#### Table I: Subjects Characteristic

Characteristics	Value	%	
Age (years)	Mean (SD): 8.3 (3.2) Range: 5 – 15		
Hypospadias type			
Distal	4	26,6%	
Proximal	11	73,3%	
Penile length			
Normal	5	33,3%	
Mircopenis	10	66,6%	
Severe penile curvature	4	26,6%	
Surgical techniques			
One stage repair	12	80%	
Two stage repairs	3	20%	

Notes: The characteristics of subjects is described in this tabel, whereas 15patients agreed to participated in this survey. We categorized the patients into two groups, distal and proximal type of hypospadias. Penile length were described whether they had history of micropenis or not.

The inter-rater agreement between the patients, parents, and the pediatric surgeon was analyzed with Cohen's kappa test (Table II). Strong agreement was observed between patients and parents in every aspect (p < 0.05), whereas poor agreement was detected between patients and parents vs. the pediatric surgeons.

In the distal hypospadias group, the mPPPS and perception of the mPPPS variables were not significantly different among the patients, parents, and pediatric surgeons (Table III). In contrast, the mPPPS in the proximal hypospadias group was lower than in the distal hypospadias group (Table IV). Patients and parents had significantly lower scores than the surgeons (patients vs. parents (p = 0.655); patients vs. surgeons (p = 0.007)). The perceptions of penile length, urethral meatus, penile skin, curvature, and general appearance were lower according to patients and parents than the surgeons (p = 0.028, p = 0.0001, p = 0.015, p = 0.008, p = 0.003, respectively).

### DISCUSSION

Genital self-perception decreases with advancing age but higher expectations begin in adolescence. The perception in childhood is relevant as a potential factor

Table II: The inter-rater	agreement of patients,	parents and	pediatric
surgeons.			

Variabel	Cohen's kappa coefficient	P value
Penile length		
Patients VS Parents	0,667	0,001
Patients VS Surgeons	0,149	0,322
Parents VS Surgeons	-0,023	0,888
Urethral meatus		
Patients VS Parents	0,884	0,000
Patients VS Surgeons	0,011	0,782
Parents VS Surgeons	0,018	0,702
Shape of glans		
Patients VS Parents	0,826	0,000
Patients VS Surgeons	-0,154	0,396
Parents VS Surgeons	-0,136	0,480
Shape of penile skin		
Patients VS Parents	0,654	0,001
Patients VS Surgeons	0,292	0,116
Parents VS Surgeons	0,228	0,123
Penile curvature		
Patients VS Parents	0,746	0,001
Patients VS Surgeons	0,009	0,935
Parents VS Surgeons	-0,132	0,252
General appearance		
Patients VS Parents	0,758	0,001
Patients VS Surgeons	0,130	0,258
Parents VS Surgeons	0,022	0,851

Notes : The inter-rater agreement between patients, parents and pediatric surgeon was analyzed with Cohen's kappa test. There was strong agreement between patients and parents in every aspect (p<0.05), conversely there are poor agreement between patients and parents vs pediatric surgeons.

Strength of agreement (<0.20 poor ; 0.21-0.40 fair ; 0.41-0.60 moderate ; 0.61-0.80 good ; 0.81-1.00 very good)

Table III: Comparison of mPPPS in distal hypospadias grou	arison of mPPPS in distal l	hypospadias grou	D
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Variable		mPPPS Median (rar	nge)	_
variable	Patient (I)	Parents (II)	Surgeons (III)	р
Penile length	2.5 (1–3)	2.5 (1–3)	2.5 (2-3)	1.00
Urethral meatus	2 (1-3)	2 (1–3)	2	1.00
Shape of glans	2 (2–3)	2 (2–3)	2	0.368
Shape of penile skin	2 (2–3)	2 (1–3)	2	0.607
Penile curvature	2 (1-3)	2 (1–3)	2	1.00
General appearance	2 (1-3)	2 (1–3)	2	1.00
Total mPPPS	12 (9–18)	12 (8–18)	12,5 (12–13)	0.670

**Notes:** Comparison for mPPPS between patients, parents and pediatric surgeon in distal hypospadias group were tested using Friedman test, statistically significant p<0.05. In the results, none of the variables were significant. Meaning the perception between patient, parents and surgeons are equals.

rubic rv. Comparison of fin rrs in proximal hypospaalas group	Table IV:	Comparison	of mPPPS in	n proximal	hypospadias	group
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that may affect psychosexual development. In this study, we included patients aged  $\geq$  5 years, as they are old enough to understand and judge the appearance of their genitals (6).

In 2008, Weber et al. developed the PPPS as an instrument to evaluate penile perception of patients, parents, and pediatric surgeons (6). The evaluations were usually conducted at the clinic through a questionnaire and a direct interview with the parents and patients. However, regular long-term follow-up visits after hypospadias repair remain a problem at some institutions, and particularly during the Covid-19 pandemic. An onlinebased survey may be more practical and useful. Online patient satisfaction surveys have been widely used. Internet-based and mobile apps are a solution for postoperative monitoring of surgical patients. They are ideal to use, as they are ubiquitous and accessible by the large majority of people and are starting to cross socioeconomic and geographic boundaries (31).

Although some studies have reported that this method alone cannot provide a sufficient response, most studies have reported that online follow-up is feasible and acceptable for patients and surgeons. Online follow-up increases the patient's quality of recovery and can be used to identify key areas for improvement in personcentered perioperative care. However, one critical limitation of online follow-up is that we were unable to conduct a proper physical examination (32). In our center, this is the first time we have used an online survey to gain postoperative satisfactory feedback from outpatients, and, to our knowledge, this is the first online evaluation of penile perception after hypospadias repair. There are differences in agreement between the patients, parents, and pediatric surgeons. Strong agreement between the patients and parents may reflect their same dissatisfaction. The patients might be embarrassed with their condition and parents have a higher expectation (6).

We find higher satisfaction outcomes of penile perception in the distal hypospadias group than the proximal hypospadias group. The patients and parents in the distal hypospadias group did not complain about the postoperative penile condition, perhaps

	mPPPS Median (range)			P value comparison			
	Patients (I)	Parents (II)	Surgeons (III)	р	l vs II	I vs III	II vs III
Penile length	2 (1-2)	2 (1-2)	2 (1-3)	0.028	0,564	0,063	0,025
Urethral meatus	1 (0-2)	1 (0-2)	2 (2-3)	0.0001	0.317	0.004	0.005
Shape of glans	2 (1-2)	2 (1-2)	2 (1-3)	0.247	-	-	-
Shape of penile skin	2 (1-2)	1 (1-2)	2 (1-3)	0.015	0.157	0.046	0.034
Penile curvature	2 (1-2)	2 (1-2)	2 (2-3)	0.008	1.00	0.034	0.014
General appearance	1 (1-2)	1 (1-2)	2 (1-3)	0.003	1.00	0.020	0.008
Total mPPPS	8 (5-12)	9 (5-12)	12 (10-18)	0.0001	0.655	0.005	0.007

Notes: Comparison for mPPPS between patients, parents and pediatric surgeon in proximal hypospadias group were tested using Friedman test, statistically significant p<0.05. 5 out of 6 variables were significant, therefore we continue to Wilcoxon test.

Patients and parents significantly had lower score than surgeons (patients vs parents (p=0.655); patients vs surgeons (p=0.005); parents vs surgeon (p=0.007)). The perception of penile length, urethral meatus, penile skin, curvature, and general appearance were lower according to patients and parents compared to surgeons (p=0.028, p=0.0001, p=0.015, p=0.008, p=0.003, irrespectively).

because morphological abnormalities are mild and associated with a better postoperative outcome in this type of hypospadias (13). Since the 1990s, development in the distal-type hypospadias surgical technique, such as meatal advancement and glanuloplasty, glans approximation, and tubularized incised plate urethroplasty, has rapidly improved postoperative function and outcome. These changes in procedure have led to increased expectations of surgeons and patients for the distal type of hypospadias rather than the proximal type (14-16). The development of distaltype hypospadias surgical techniques has resulted in increased success rates of 85%–95% (17-19), whereas the success in treating the proximal type of hypospadias is lower at 75%–90% (20,21).

In contrast, poor penile perception is observed in the proximal hypospadias group. This finding is consistent with a study by Mureau and Long, who stated that the cosmetic result of proximal hypospadias surgery is less successful than distal hypospadias surgery (25). We also found that patients and parents were less satisfied with penile length, urethral meatus shape, penile skin, curvature, general penile appearance, and total mPPPS compared to the surgeons.

Berg and Mureau reported that patients with proximal hypospadias tend to have a shorter penis than those with distal hypospadias (33,34). Other than a small preoperative penile size, it might be due to incomplete ventral lengthening and penile straightening during repair (35). Residual curvature becomes more pronounced with increasing age at puberty and with rapid penile growth. Long's study reported that penile curvature is a major concern in adulthood, as patients considered sitting to void due to spraying the urinary stream, and painful or awkward sexual function due to penile curvature can be life-threatening (36). Stojanovic showed that penile angulation not only causes potential sexual dysfunction, difficulty, and pain during intercourse, but also causes severe psychological problems (37). If a patient has residual curvature, the perception of the penile curvature will decrease with time. The patients and parents are dissatisfied with penile length and curvature in this study.

One of the goals of penile reconstruction is to achieve a slit-like meatus with a well-approximated glans (35). In this study, patients and parents in the proximal hypospadias group were not satisfied with the urethral meatus. Proximal hypospadias has been associated with glans dehiscence, meatal stricture, and redo surgery, which may contribute to the unfavorable shape of the meatus or its location below the glans tip (36).

Proximal hypospadias is associated with a smaller glans diameter. Bush and Snodgrass reported that glans width < 14 mm is associated with postoperative complications (38). The complication rate of glans dehiscence is higher in patients with proximal hypospadias (12.5%) than in those with distal hypospadias (3%). After surgery, the size of the glans decreases by an average of 2 mm in distal and proximal hypospadias groups (38). Reoperation due to glans dehiscence will affect the shape, size, and cosmetic satisfaction. In this study, all assessors share similar perceptions of the glans (mPPPS for glans, patients vs. parents vs. surgeons: 2 vs. 2 vs. 2, p = 0.247). This might be due to an acceptable glans shape and size or the expectation for it is not high in all of the assessors.

Asymmetrical, excessive skin or obvious surgical scars will affect the appearance of the penis (27,28). Unsatisfactory skin cosmetics can be caused by multiplestage surgeries or repeated operations due to surgical complications (29). Repeated operations often affect the position and shape of the meatus. This will lead to a lower perception, as seen in this study, particularly in the patient and parent groups.

The general appearance of the penis contributes to the psychosexual development of the child and can potentially interfere with the child's quality of life. Parental dissatisfaction will affect the assessment of the shape of the child's genitalia. Additionally, parents have a reference penis to compare and have higher expectations for the shape of the child's genitalia after undergoing surgery. Dissatisfaction by parents will affect the attitude of the parents towards the child, which will affect the psychosexual development of the child (6).

Compared to Weber's PPPS study in 2008, the results of this study is inversely related to the perception by patients and parents that tended to be lower than the pediatric surgeons. This result is similar to studies by Berg (1981), Mureau (1995), and Long (2016), in which patients with proximal hypospadias tend to have a lower perception of their genitals. This result indicates that patients and parents have high expectation for penile cosmetics even in cases of severe hypospadias. Otherwise, a higher mPPPS from the surgeons in the proximal hypospadias group might be due to a relatively lower cosmetic expectation, as these patients have a more severe form of anatomical anomaly, insufficient skin, complicated staged repair, and occasional redo surgery.

Despite several limitations in this study, such as small sample size, unreachable patients, and poorquality photographs, we consider that the mPPPS is advantageous for long-term follow-up after hypospadias repair. Preoperative penile biometry data, including proper clinical photographs, should be completed for comparison of the cosmetic results purposes. Parents should be notified that long-term monitoring will be conducted regularly and a definite contact number should be provided or updated if there are any changes. Further web or mobile apps-based surveys can potentially be developed.

#### CONCLUSION

The mPPPS, with some adjustments, can be used as an alternative instrument to evaluate penile perception after hypospadias repair and ensure regular and longterm postoperative follow-up. Patients and parents agreed with each other more than with the pediatric surgeons. Penile perception is not different among patients, parents, and the pediatric surgeons in the distal hypospadias group. However, patients and parents in the proximal hypospadias group are less satisfied than the pediatric surgeons. Further technical refinement will help attain a better cosmetic result after proximal hypospadias repair.

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