

Does parental opinion differ from the health care team regarding cosmesis after hypospadias repair?

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SUMMARY

OBJECTIVE: Hypospadias is the most common malformation of the male genitalia. Surgical correction has traditionally focused on anatomic and functional outcomes, with less attention being paid to cosmetic results. Our purpose is to compare the cosmetic results of hypospadias repair among different groups of observers, namely the patient's family and the health care team, using photography and a simple rating scale.

METHODS: Prospective observational study included 9 boys undergoing Snodgrass hypospadias repair. Photographs of the penis taken before, immediately after, and six months after surgery were assessed by a panel of 15 observers (parents and health care team) and a scale including three questions with diagrams for comparison with the pictures was used. Observers also assigned an overall postoperative score for the cosmetic result.

RESULTS: Interobserver agreement was noted for the group of parents of other children with hypospadias regarding the shape of the glans ($k=0.404$; $p=0.008$) and for the group of pediatric surgeons regarding the degree of residual curvature ($k=0.467$; $p=0.005$). Two observers in the pediatrician group have indicated good performance in the assessment of residual curvature ($k=0.609$; $P=0.024$). In the overall assessment of cosmetic outcomes, the highest scores were assigned by observers in the parents group and in the pediatrician group, while the pediatric surgeons group has one of the lowest scores ($p<0.001$).

CONCLUSIONS: Photography appears to be suitable for documenting corrections of hypospadias regarding penile curvature, and postoperative cosmetic result. Surgeons seem more concerned about cosmesis than parents.

KEYWORDS: Hypospadias. Photography. Parents. Patient care team.

INTRODUCTION

Hypospadias is a common birth defect of the male genitalia, involving arrested development of the urethra, foreskin, and ventral surface of the penis¹. In patients with hypospadias, the urethral meatus is located ventrally on the penis, with or without ventral penile curvature. The incidence of hypospadias is variable and depends on regional and ethnical differences. Some papers have shown increasing hypospadias rates and others have

shown no increasing rates; but, currently, hypospadias is the most common malformation of the male genitalia¹⁻⁵.

Different surgical techniques can be used to repair hypospadias. The main goals of surgery are the construction of a urethral meatus in orthotopic (glandular) position and correction of penile curvature, if present⁶. The correction of hypospadias has traditionally focused on anatomic and functional outcomes. However, a growing concern with body image has led

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to greater attention being paid to cosmetic results. Assessment of cosmetic aspects is complicated by the absence of objective parameters. In addition, this assessment is often performed by reconstructive surgeons, who may not prioritize cosmetic results.

The use of photography could facilitate the assessment of the same patient by different people without exposing him. The objective of this study was to evaluate the cosmetic assessment of hypospadias repair by a panel of evaluators, including nurses, clinicians, surgeons, and parents, as well as to compare their opinion.

METHODS

This is a prospective observational study of consecutive children (6 months to 94 months of age/average age 14 months) undergoing hypospadias repair at a university hospital.

Photographs of the penis (frontal, lateral, and upper views) were taken before, immediately after surgery, and 6 months later. Photographs were taken using a Sony® DSC-W30 digital camera, with 3x optical zoom and resolution of 6 megapixels. All photographs followed the same parameters: there was a focal distance of 20 cm and the lateral view pictures were always performed to the left side of the patient. A set of three pages containing three photographs each (preoperative, immediate postoperative, and 6 months postoperative photographs) was placed in a folder for each individual patient. The folder was then presented to a panel of 15 independent observers (not related to the patients and not involved in the study), divided into five groups: 3 pediatric surgeons (S), 3 pediatricians (P), 3 nurses (N), 3 parents of other children with hypospadias (PH), and 3 parents of children without hypospadias (PW). The observers were instructed to use a scale developed by our research group for evaluation of position of the meatus, shape of the glans, penile curvature, and general postoperative cosmetic result. The scale proposed has three objective questions; these questions were made based on an effort to harmonize parameters used to evaluate the images among observers. The findings were then compared to the impressions recorded by the investigator when examining the child after surgery. Physical examination performed by the investigator without access to the photographs was considered the gold standard.

The surgical technique described by Snodgrass et al.⁷ was used in all cases. All surgeries were performed as routine procedures by pediatric urology surgery professionals under the supervision of a staff surgeon.

Data analysis was performed using the Stata software, version 7.0⁸. Kappa coefficient was calculated for the comparison of individual observers. A generalized estimating equations (GEE) approach⁹ was used to verify whether there were

differences in means among the five observer groups. After GEE was used, the Bonferroni test¹⁰ was used for multiple comparisons of means. The Bonferroni test was proposed by Fisher in order to analyze multiple comparisons. This consists in performing a t-test for each pair of means at an error rate by comparison. In this way, the Bonferroni test protects the error rate of the family of the tests. Significance level was set at 5%. As the patients and observers were selected by convenience, and to avoid a very large folder, only nine patients were chosen.

RESULTS

Nine children treated consecutively for hypospadias were included in this study. At baseline, all patients had a hooded foreskin. Meatal position was subcoronal in five patients, distal penile in three, and midshaft penile in one. Three patients did not have penile curvature, five had mild/moderate curvature (less than 45°) and only one had severe curvature (greater than 45°). No patient received preoperative hormonal stimulation. Two patients developed postoperative subcoronal urethra cutaneous fistula and two had complete glans dehiscence. None presented residual penile curvature after surgery.

We obtained 144 evaluations using the hypospadias repair assessment scale. Table 1 describes interobserver agreement for the assessment of meatus position, penile shape, and penile curvature, based on the scale and photographic documentation. Moderate agreement ($k=0.467$; $p=0.005$) was detected among S observers regarding degree of residual curvature.

When contrasting the assessment made by each of the 15 individual observers against the gold standard (Table 2), we observed that, regarding the position of the urethral meatus, a good agreement rate was obtained for only one surgeon; nevertheless, this doesn't reach statistical significance. Regarding shape of the glans, agreement was slightly better; however, agreement was only significant for observer 1 in group S ($p<0.05$). Despite a lower agreement rate, one PW observer showed a kappa of 0.357 (fair agreement) and $p=0.015$. Evaluation of residual penile curvature revealed the highest levels of agreement in relation to the gold standard, with two observers (observers 2 and 3 in the P group) reaching 88.9%. Both showed a kappa of 0.609 (moderate to substantial) and $p=0.024$.

Regarding the overall rating of surgical results (question 4), the highest rates were attributed by the PH and P groups (9.52 and 8.67 respectively) (Figure 1). The S group mean score was 7.98. The lowest mean score was given by the PW group (7.22). The Bonferroni test revealed statistical difference between the highest and lowest mean overall rates ($p<0.001$).

DISCUSSION

Although photography has been increasingly used to evaluate hypospadias and the results of surgical correction, few studies have evaluated the usefulness of this tool in association with objective rating scales¹¹⁻¹⁷. Baskin¹¹ was the first to describe the results and complications of hypospadias surgery using photography, considering that photographic analysis could provide an unbiased assessment of cosmetic outcomes. El-Hout et al.¹² also argued in favor of digital photography as a means for evaluation of hypospadias. Nevertheless, the results of that study showed poor inter-rater agreement in relation to urethral plate ($k=0.06$), highlighting the potential subjectivity of this evaluation, regardless of the level of surgeon experience.

Like us, Holland et al.¹³ have proposed a hypospadias objective scoring evaluation (HOSE), contemplating anatomical and functional aspects. However, HOSE is based on a total score resulting from the sum of all variables, with no individual analysis of each variable. The failure to stratify each variable independently may cause confusion if the results of one or more aspects are considered poor.

The present study, based on stratified results, revealed that observers had great difficulty in locating the correct position of the meatus on the photographs, when compared to the gold standard. The difficulty in determining the position of the urethral meatus on images may be related to the presence of post-operative edema as late as 6 months after surgery. Especially in patients who developed fistulas and wound or glans dehiscence,

Table 1. Kappa coefficient and p-value of agreement within each subgroup evaluating meatus position, shape of glans and penile curvature, using the proposed hypospadias repair assessment scale and photographic documentation.

	Kappa	p		Kappa	p		Kappa	p
Q1			Q2			Q3		
observ_S	-0.044	0.584	observ_S	-0.026	0.566	observ_S	0.467	0.005*
observ_P	-0.091	0.672	observ_P	-0.344	0.973	observ_P	0.250	0.110
observ_N	-0.171	0.870	observ_N	-0.179	0.871	observ_N	0.161	0.188
observ_PH	0.030	0.431	observ_PH	0.404	0.008*	observ_PH	0.071	0.349
observ_PW	-0.115	0.781	observ_PW	-0.103	0.756	observ_PW	-0.267	0.956

Q1: question 1 (meatus position); Q2: question 2 (shape of glans); Q3: question 3 (penile curvature); Observ_S: pediatric surgeons; observ_P: pediatricians; observ_N: nurses; observ_PH: parents of other children with hypospadias; observ_PW: parents of children without hypospadias; Kappa Agreement: <0 less than chance agreement, 0.01–0.20 slight agreement, 0.21–0.40 fair agreement, 0.41–0.60 moderate agreement, 0.61–0.80 substantial agreement and 0.81–0.99 almost perfect agreement; *Indicates statistical significance.

Table 2. Agreement between observers using the proposed hypospadias repair assessment scale and photographic documentation versus clinical assessment by surgeons performing the surgery (gold standard).

Question and observer	Agreement (%)	Kappa	p	Question and observer	Agreement (%)	Kappa	p
q1gs q1s1	66.7	0.270	0.118	q2gs q2ph2	66.7	0.342	0.147
q2gs q2s1	66.7	0.426	0.037*	q2gs q2ph3	55.6	0.265	0.134
q2gs q2p1	66.7	0.386	0.083	q2gs q2pw3	55.6	0.357	0.015*
q2gs q2p3	55.6	0.182	0.171	q3gs q3s3	77.8	0.400	0.067
q2gs q2n1	66.7	0.372	0.076	q3gs q3p2	88.9	0.609	0.024*
q2gs q2n3	66.7	0.342	0.147	q3gs q3p3	88.9	0.609	0.024*
q2gs q2ph1	55.6	0.217	0.208	q3gs q3n1	77.8	-0.125	0.646

q1: question 1 (meatus position); q2: question 2 (shape of glans); q3: question 3 (penile curvature); gs: gold standard; s1: observer_pediatic surgeon 1; s3: observer_pediatic surgeon 3; p1: observer_pediatician 1; p2: observer_pediatician 2; p3: observer_pediatician 3; n1: observer_nurse 1; n3: observer_nurse 3; ph1: observer_parent of other child with hypospadias 1; ph2: observer_parent of other child with hypospadias 2; ph3: observer_parent of other child with hypospadias 3; pw3: observer parent of child without hypospadias; *Indicates statistical significance.

the complication site was the meatal location. Another misleading factor that is not always clear on images is wound or glans dehiscence. These complications were observed in two of our patients.

Also, regarding aesthetic appraisal derived from anatomical aspects, Ververidis et al.¹⁴ have proposed an objective assessment of cosmetic results based on slide images projected onto a lecture-theater screen for approximately 1 hour, considering meatus, glans, shaft, and overall appearance. The authors compared two different methods used for surgical correction: Snodgrass technique and Mathieu/Duckett repair. The Snodgrass et al.⁷ technique (which was used in our patients) was considered the best approach. Despite using photography and evaluating cosmetic results, this study cannot be compared to ours, because the authors set different goals: the results for two different techniques were compared and a totally subjective score was used. In our study, the three first questions corresponded to anatomical analysis, facilitating the uniformity of response. Another difficulty posed by the referred study concerns the projection of images. In our study, the observers could evaluate the images individually for as long as deemed necessary.

In the study by Weber et al.¹⁵, the Pediatric Penile Perception Score (PPPS), proposed by the authors after hypospadias repair, was applied to patients, parents of patients, a control group of patients, a control group of parents, and urologists. Like the study by Ververidis et al.¹⁴, a subjective evaluation was performed by asking about the degree of satisfaction with the outcome of surgical repair. The authors concluded that the PPPS was an appropriate instrument to assess penile self-perception in children after hypospadias repair. Our study also assessed anatomical variables and, even though we did not include patient self-assessment, we evaluated the opinion of

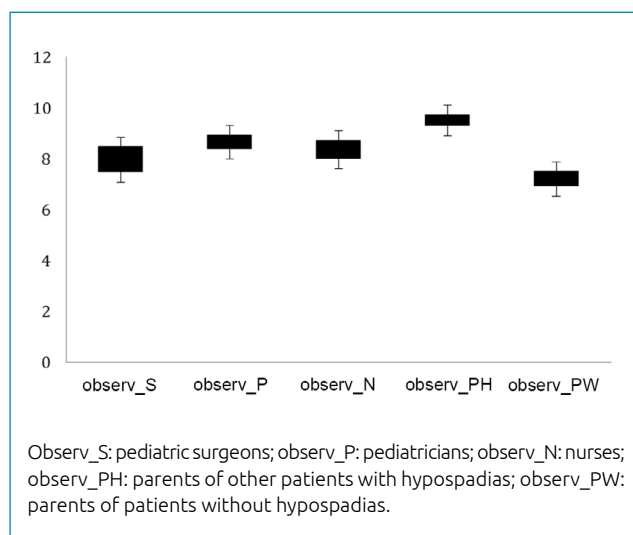


Figure 1. Mean and confidence interval for scores in question 4, according to observer group.

parents of other children with hypospadias. This group of parents ended up assigning the highest mean scores for final penile appearance. Surgery outcome was also highly evaluated by pediatricians, demonstrating that these professionals, who are the primary referring medical practitioners, were satisfied with the final results.

Regarding only the opinions of surgeons, Van der Toorn et al.¹⁶ proposed the Hypospadias Objective Penile Evaluation (HOPE) score, which is an objective scoring system for evaluation of cosmetic appearance in hypospadias correction. The HOPE scoring system evaluates six correctable items: the position of meatus, shape of meatus, shape of glans, shape of penile skin, and penile axis, including penile torsion and penile curvature. The study reported strong intra-rater reproducibility. Furthermore, the HOPE score may be useful to compare cosmetic outcomes obtained with different techniques. If we consider only our S group, there was high agreement regarding the degree of residual curvature and meatus position.

Haid et al.¹⁷ compared parents' and surgeons' opinions using PPPS and HOPE scores. Parents had worse results than surgeons regarding cosmesis using those scores. But there was no difference between groups when considering the general appearance of the penis. In 1996, Mureau et al.¹⁸ brought to attention that patients weren't satisfied with their penile appearance after hypospadias repair, particularly when compared to the surgeon's view. A systematic review published in 2016¹⁹ illustrated what probably is the most important measure to assure a good result: keeping patients in a long follow-up, as Springer⁶ has already mentioned. Both papers showed that results sometimes are questioned. Snodgrass et al.²⁰ indicated that parents' and surgeons' opinions of cosmetic were similar, when comparing hypospadias repair and elective circumcision. Our study suggested parents were more satisfied with cosmetic results than surgeons. It is important to remember that those parents didn't analyze their sons' results, so they didn't have any personal issues involved like previous papers cited before.

Scarpa et al.²¹ have compared objective vs. subjective assessment of cosmetic results after distal hypospadias repair. There was an excellent level of agreement among four observers (objective observer, operating surgeon, independent surgeon and parent of child). Like us, those authors also detected the highest scores among the parents of patients with hypospadias.

Another important finding from our study is that two pediatricians achieved very good agreement with the gold standard regarding residual curvature, a better performance than that of surgeons individually. This finding suggests that the professionals who are most likely to refer patients to surgeons are well trained with respect to detection of penile curvature, at least in our institution.

A possible limitation of the present work is the small number of patients. However, the number of evaluations – 144 – is more satisfactory. Each patient had nine pictures that were analyzed 15 times (panel of observers) and also by the investigator, and this total number was sufficient to ensure statistical relevance. Although we observed an unusually high number of complications after hypospadias repairs, these were not important because all observers had the same picture for analysis, and the focus of this study was on the agreement among different observers.

Another limitation of this paper is that our scale wasn't validated and it was prone to being subjective, since, because we had parents analyzing pictures, we tried to make it as simple it could be. In the other hand, it looks very easy to apply, considering it has only four questions.

CONCLUSIONS

This study shows that photography may be a good means of documenting corrections of hypospadias regarding the shape of the glans, penile curvature, and postoperative cosmetic outcomes. However, photography was not adequate to assess the shape of the glans and the position of the meatus, especially in the presence of small fistulas. Regarding this aspect, upper view photographs are not necessary, because they did not adequately show meatus position and simply meant three additional pictures per patient. Among all observers, parents of other children with hypospadias expressed the highest satisfaction with the cosmetic outcome of hypospadias repair. This finding could suggest that surgeons are more concerned with aesthetic aspects than patients' parents. At least, this trend is perceived according to data obtained at our institution, and although we

used a small sample, it was analyzed in a meticulous way. This conclusion is important to help make surgeons less concerned with aesthetic issues. Additionally, this paper suggested that pediatricians were well informed to identify penile malformations, mainly hypospadias associated with penile curvature, thus leading to appropriate referral for surgical repair. More data is necessary to assure those findings.

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Ethical approval: All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Our IRB approval is 07649.

Informed consent: informed consent was obtained from all individual participants included in the study.

AUTHORS' CONTRIBUTIONS

EC: Conceptualization, Data Curation, Formal Analysis, Project Administration, Writing – Original Draft, Writing – Review & Editing. **JPS:** Conceptualization, Writing – Original Draft, Writing – Review & Editing. **JCF:** Conceptualization, Formal Analysis, Writing – Original Draft, Writing – Review & Editing. **NR:** Data Curation.

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