Long-Term Outcomes of Distal Hypospadias Repair: A Patients' Point Of View

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Purpose: To assess the long-term outcomes of patients treated for distal hypospadias. Assessment of long-term follow-up for a homogeneous population with hypospadias is difficult and there has consequently been a paucity of publications in this regard.

Materials and Methods: A retrospective review was carried out to compile cases of distal hypospadias operated at our center between 1990 and 1999 according to the MAGPI procedure. Four parameters were evaluated based on four validated questionnaires: Health-related quality of life (SF-36), Genital self-perception (PPS), Self-esteem (Rosenberg Self-Esteem Scale), and Erectile function (IIEF).

Results: A total of 77 patients who had undergone MAGPI surgery for hypospadias during the specified period were selected. Sufficient clinical data were available for 51 patients and only 15 of these patients were included, after a median follow-up of 22 years (20-26). Their outcomes were compared with those for a population of 15 matched circumcised men and 15 matched uncircumcised men. No significant difference was found between the patients and the control groups in terms of the score for quality of life ($p = .29$). There were, however, significant differences in the scores for self-perception of the penile cosmetic appearance (13.3 vs. 15.8; $p < .01$), self-esteem (30.6 vs. 35.8; $p < .01$), and erectile function (31.4 vs. 33.7; $p = .04$) between the patients and the controls. Lower self-esteem correlated with poor genital self-perception (r = .92).

Conclusion: This study confirms that adult patients operated for distal hypospadias have poor genital self-perception. This poor genital perception correlated with lower self-esteem.

Keywords: hypospadias; MAGPI; long-term outcomes; self-esteem

INTRODUCTION

Hypospadias exhibits a broad spectrum of severity, and a multitude of surgical reconstructive techniques have been devised that are aimed at correcting this congenital abnormality. Even for the specific distal type of hypospadias, the large variety of procedures reflects the absence of an established gold standard for treatment. Irrespective of the choice of the technique used, there is not a clear consensus regarding the indication for surgery in case of very distal hypospadias due to the substantial cosmetic considerations involved. There is a paucity of published data regarding the cosmetic and self-estimated outcomes of hypospadias. Most of the published series to date have been in regard to short-term cosmetic outcomes reflecting the opinions of the parents and the surgeon. Definitive urinary functional outcomes can be addressed in adolescence.[1] It is, however, more difficult to obtain data regarding sexual function and self-estimated cosmetic outcomes, as this requires long-term close follow-up.[2] There have been few reports to date of long-term outcomes, particularly in regard to distal hypospadias.[3] Similar long-term cosmetic and functional issues can be expected for operated patients and patients without hypospadias.[2]

The rationale to operate distal hypospadias during early childhood should take into account cosmetic and ethical considerations. The fundamental question is whether esthetic correction is indicated before the patient has the capacity to provide their informed consent4. There has been no large-scale study to date comparing the long-term outcomes of adult patients who have or who have not been operated for distal hypospadias. The key issue remains the self-perception of patients regarding their hypospadias. Most of the data published to date has been in regard to medium-term follow-up series with heterogeneous types of hypospadias and follow-up durations.[5,6] Of note, there has been one study that reported a similar health-related quality of life for operated patients and controls.[4] No study, however, has focused on the self-estimated outcomes for adult patients operated for distal hypospadias during childhood.

To address these issues, we focused on a particular population: patients with distal hypospadias operated with a single repair technique. The primary endpoint of the

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Table 1. Characteristics of the control group comprising unoperated and circumcised men

Variables (min-max)a	Total	Group 1- circumcised	Group 2- uncircumcised	*P*-value
Number	30	15	15	
Age	28.0 (21-39)	28.8 (21-38)	27.2 (21-39)	.48
Age at surgery (months)	42.4 (13-65)	42.4 (13-65)	none	
Penile score (PPPS)	15.8 (13-18)	16.5 (15-18)	15.2 (13-18)	.07
- Length	2.2 (2-3)	2.4 (2-3)	2.2 (2-3)	
- Meatus	- 2.9 (2-3)	- 3.0 (2-3)	- 2.9 (2-3)	
- Glans	- 2.6 (2-3)	- 2.8 (2-3)	- 2.6 (2-3)	
- Skin	- 2.2 (2-3)	- 2.8 (2-3)	- 2.2 (2-3)	
- Axis	- 2.6 (2-3)	- 2.8 (2-3)	- 2.6 (2-3)	
- Appearance	- 2.2 (2-3)	- 2.4 (2-3)	- 2.2 (2-3)	
Erectile function (IIEF)	33.7 (30-35)	33.8 (33-35)	33.6 (30-35)	.44
Self-esteem (Rosenberg)	35.8 (28-40)	36.5 (28-39)	34.9 (28-40)	.19
Quality of life (Euroqol5D)	91.3 (65-100)	92.1 (75-100)	90.3 (65-100)	.56

Abbreviations: PPPS: Pediatric Penile Perception Score; IIEF: International Index of Erectile Function
[a] The continuous variables were compared using a parametric test (Student's t-test) and non-a parametric test (Mann-Whitney test)

present study was to report the long-term outcomes for such patients in terms of the functional parameters, effects on sexual performance, and health-related quality of life (HRQoL) after reconstructive surgery for distal hypospadias with long-term (> 20 years) follow-up. The secondary endpoint was to compare these parameters with those of the general population without hypospadias.

METHODS

Study population

The patients had to be at least 20 years of age to be considered eligible for this study. A retrospective review was carried out to compile cases of hypospadias operated between 1990 and 1999 for distal hypospadias. This study was approved in 2019 by the ethics committee of the University Hospital of Limoges as reference number 309-2019-75.

Inclusion and exclusion criteria

The primary endpoint was the long-term outcomes of patients treated for distal hypospadias in terms of functional outcomes, effects on sexual performance, and general quality of life. A total of 138 medical files of patients who had a procedure code compatible with hypospadias repair at our institute between 1 January 1990 and 31 January 1999 were reviewed in order to select patients who were between 20 and 30 years of age in 2019 and who had undergone primary hypospadias repair. Only patients with distal hypospadias repair according to the Meatal Advancement and Glanuloplasty (MAGPI) procedure were included in the present study. We selected these patients to obtain a homogeneous popu-

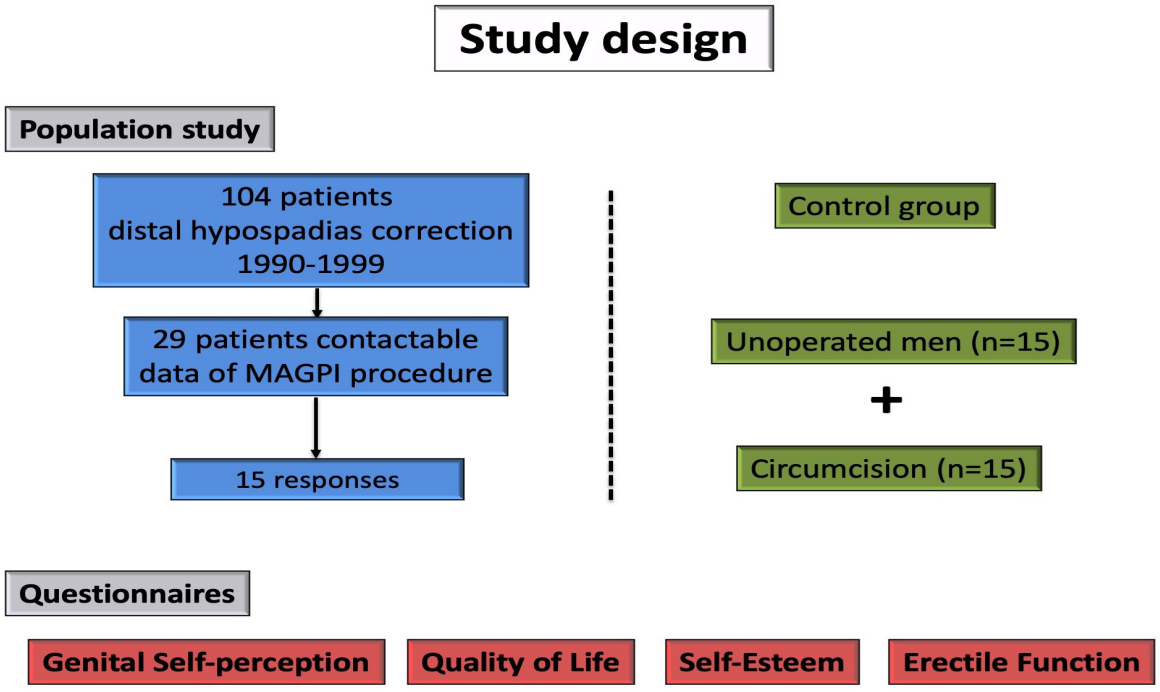

Figure 1. Study flow diagram.

Table 2. Comparison of the scores of the patients and the control group.

Variables (min-max)[a]	Patients	Control group	*P*-value
Number	15	30	
Age	24.3 (20-30)	28.0 (21-39)	.02
Age at surgery (months)	42.3 (12-78)	42.4 (13-65) (circumcision)	.86
Penile score (PPPS)	13.3 (10-17)	15.8 (13-18)	< .001
- Length	- 2.0 (1-3)	- 2.2 (2-3)	.052
- Meatus	- 2.1 (0-3)	- 2.9 (2-3)	< .001
- Glans	- 2.5 (2-3)	- 2.6 (2-3)	.0081
- Skin	- 2.2 (1-3)	- 2.3 (2-3)	
- Axis	- 2.3 (1-3)	- 2.6 (2-3)	
- Appearance	- 2.4 (2-3)	- 2.3 (2-3)	
Erectile function (IIEF)	31.4 (14-35)	33.7 (30-35)	.04
Self-esteem (Rosenberg)	30.6 (24-37)	35.8 (28-40)	.0001
Quality of life (Euroqol5D)	94.3 (80-100)	91.3 (65-100)	.29

Abbreviations: PPPS: Pediatric Penile Perception Score; IIEF: International Index of Erectile Function[a] The continuous variables were compared using a parametric test (Student's t-test) and a non-parametric test (Mann-Whitney test)

lation with high expectations regarding the cosmetic results. Distal hypospadias was defined as a preoperative meatal position distal to and including mid-penile shaft cases. These patients did not have preoperative penile curvature. Out of a total of 104 patients, 23 underwent tubularized incised plate repair, 4 underwent Mathieu repair, and 77 underwent MAGPI procedures. The only exclusion criterion was insufficient data regarding short-term follow-up of the patients.

During the follow-up, the patients were asked to complete a questionnaire regarding their genital self-perception. If they were no longer included in close follow-up, they were also invited to consult with the dedicated urology unit of our institute. The control group comprised men without hypospadias who agreed to anonymously complete the same questionnaire. Each patient was matched according to age at the time of the surgery and age in 2019 to two adult controls without hypospadias from both a circumcised and an uncircumcised patient population. The circumcised group was compared to the uncircumcised group, and the hypospadias group was compared to the total control group. Four parameters were evaluated based on four validated questionnaires.

Health-related quality of life (HRQoL)

The 36-item Medical Outcomes Study Short-Form questionnaire (SF-36) was used to assess the HRQoL.[7] This scale has been validated and has been determined to be reliable for our population (reliability coefficient of 0.75).[8] Subscales of each domain (physical function, role limitations, bodily pain, general health perception, mental health, role limitations due to emotional problems, vitality, and social function) were scored from 0 to 100, with higher scores indicating a better HRQoL.

Genital self-perception (PPS)

Penile cosmetic self-perception was evaluated according to the Penile Perception Score.[9] This instrument consists of the following four items for the participants to evaluate their genitals, with a 4-point scale ranging from very dissatisfied (0) to very satisfied (3): the position and shape of the meatus, the shape of the glans, the shape of the shaft skin, and the general appearance of the penis. The PPS score ranges from 0 to 12. It is a recognized score in the domain of hypospadias, although it has yet to be shown to be a validated patient-reported outcome instrument.[10] We also assessed two additional items included in the pediatric PPS because their absence is a limitation according to the literature: the length and the axis of the penis.[2, 11]

Self-esteem

The Rosenberg Self-Esteem Scale, which is a widely used self-report instrument for disorders such as sexual development, was chosen to evaluate the participants' level of self-esteem.[12] Self-esteem is tightly linked to happiness.[13] This 10-item scale measures overall self-worth by assessment of both positive and negative feelings about oneself. All of the items are rated using a 4-point Likert scale ranging from 'strongly agree' to 'strongly disagree'.

Erectile function and orgasmic function

The international index of erectile function was used to measure erectile and orgasmic functions in the past 6 months. The erectile function subscale has a maximum domain score of 30 and an organic function subscale of 10.[14] Higher scores correspond to better functions. This scoring method was validated many years ago and it is commonly used in publications relating to studies of hypospadias.[3,14]

Statistical analysis

The data were analyzed using Prism 8.2.1® Mac® software (GraphPad Software, San Diego, California, USA). The comparative statistics were performed using Fisher's exact test for the categorical variables, a parametric test (Student's t-test) for the data with a normal distribution (means and the corresponding SD are presented), and a non-parametric test (Mann-Whitney test) for the data without a normal distribution (medians and the IQR are presented). A *p*-value of less than 0.05 was considered significant.

RESULTS

A total of 77 patients operated for distal hypospadias in the specified period were selected. No postoperative complications occurred after a median follow-up of 15 months (range 1-63). No cases involving penile curvature or a short penis length were noted. Clinical data were available for 51 patients to calculate the postoperative PPS score. The patients were contacted using their parents' addresses or through social networks. The correct contact details and addresses of the patients were ultimately only available for 45 of the patients. A total of twenty-nine patients could be reached by phone, of whom twenty-two were lost to follow-up (**Figure 1**). No

additional postoperative complications were noted, nor did any further surgery take place. None of the patients indicated that they suffered from urinary functional impairment. One patient died of an unrelated cause. Five patients returned to the dedicated consultation unit for a medical examination and a uroflowmetry test.

Only 15 patients complied with the study requirements by returning the questionnaire after a median follow-up of 22 years (IQR 20,26), their median age was 24.3 years (IQR 20,27). According to surgeons, the median short-term postoperative PPS score was 17.8 (range 17-18), which was representative of the MAGPI population 17.7 (range 17-18) in this study period. Their outcomes were compared to those for a population of 15 matched circumcised men (median age 28.8 years (IQR 21,38)) and 15 matched uncircumcised men (median age 27.2 years (IQR 21,39)).

No differences were found for the scores of the four parameters that were assessed between the circumcised and the uncircumcised population (**Table 1**). Thus, we decided to compare the operated patients with all of the controls by combining both the circumcised and the uncircumcised men.

The control subjects were older than the patients (28.0 vs. 24.3 years of age; $p < .05$), although the median age at the time of the surgery was the same for both groups (**Table 2**). There was not a significant difference between the patients and the control group in terms of the quality of life ($p = .29$). There was a significant difference, however, regarding the penile cosmetic self-perception (13.3 vs. 15.8; $p < .01$). This difference was mainly due to the following items: the length (2.0 vs. 2.2; $p = .052$), the meatus (2.1 vs. 2.9; $p < .001$), and the axis (2.3 vs. 2.6; $p = .0081$) of the penis (**Table 2**). There was also a significant difference in the self-esteem score (30.6 vs. 35.8; $p < .01$) and the erectile function score (31.4 vs. 33.7; $p = .04$). Two patients had not yet had a sexual partner. Lower self-esteem correlated with poor genital self-perception ($r = .92$; $p < .01$). No association could be found between lower self-esteem and reduced erectile function.

DISCUSSION

Our results indicate that adult men operated on for distal hypospadias have lower self-esteem and penile perception scores compared with controls. The quality of life was comparable to that of the controls, as has also been reported for a population of patients with heterogeneous operated hypospadias.[4] This study was the first to present long-term self-perception outcomes. The authors pointed out that the limitations of their study comprised the late age at which the surgical procedure was performed (more than 4 years of age), heterogeneous types of hypospadias and surgical techniques, and the exclusive use of circumcised men as controls.

There is a paucity of studies regarding long-term follow-up of hypospadias because the transition between pediatric and adult urology is not systematic for most patients.[15-17] The most common reason for genital dissatisfaction in hypospadias patients is inadequate penile size, while the experience with masturbation appears to be similar to that of the general population.[18] It is well known that men with corrected hypospadias suffer from sexual inhibition and fear of being ridiculed, while those with proximal hypospadias are also more likely to be dissatisfied with their genital appearance.[17,19-21] No long-term-follow studies are available specifically for distal hypospadias. The present study showed that the perceived quality of life was the same for the patients, despite reduced erectile function compared to the controls. The main bias in regard to the erectile function item was that two of the patients had not yet engaged in sexual intercourse. There was no difference, however, in the overall erectile function score when these two patients were excluded from the patient group.

Recent studies have found that laypersons are not troubled by the penile appearance after hypospadias repair and that women perceive the genitals of men with distal hypospadias as being similar to the genitals of non-affected, circumcised men.[22,23] Similarly, men with non-operated hypospadias often find that some of their sexual partners or medical professionals are not aware of their hypospadias. In the present study, the patients expressed clear displeasure regarding the length, meatus, and axis of their penis, whereas the overall appearance was rated as being similar in both groups.[24,25] As described previously, these concerns did not appear to impact the overall quality of life.[4]

Particularly in case of distal hypospadias, it is likely that the main reason for the stated low level of satisfaction with their penile appearance could be considered to be part of their lower overall self-esteem. Self-esteem was distinctly lower in the patients: 8 (53%) of them had a score below 30, which means that specific psychological support is required. The underlying reason for this is not clear, however. Indeed, the role of surgical treatment may be of little or no relevance as circumcised patients do not exhibit the same psychological profile. Most of the patients contacted by phone stated that they did not remember undergoing the surgery during their childhood. However, the emotional reaction of the parents in regard to the hypospadias appears to be critically important for the patients' psychological development.[26] This information may be of considerable relevance to practitioners who treat patients with hypospadias and their families with the aim of preventing the development of feelings of shame and negative genital perception.[27,28] When these patients undergo counseling, they should be provided support to develop a positive genital self-perception, as poor genital self-perception has been shown to correlate with an impaired mental health-related quality of life.[4] Our data confirm the need for long-term follow-up to allow patients to address concerns regarding their genitals.[2,6]

The main limitations of this study are the small number of patients and the possibility of selection bias for patients who agreed to participate in the survey. Another limitation is the absence of an ideal control group of adult patients with hypospadias but who did not undergo surgical correction.

CONCLUSIONS

The findings of the current study confirm poor genital self-perception of adult patients operated from distal hypospadias. This poor genital perception correlated with lower self-esteem. Consequently, for clinical management, we suggest that adult hypospadias patients are made more aware of the fact that their penile appearance is often a non-issue for laypersons. Early and long-term follow-up of patients and their parents could prevent this negative genital and overall self-perception from taking hold. The main limitation of this study

relates to its long follow-up retrospective design with many consecutive biases. Further prospective studies with larger numbers of patients are necessary to confirm this negative perception despite a normal perception of genital appearance.

CONFLICT OF INTEREST

The authors declare that they have no conflicts of interest.

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