

Long-Term Functional, Cosmetic and Sexual Outcomes of Hypospadias Correction Performed in Childhood

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Key Words

Hypospadias · Hypospadias-repair · Functional · Flowmetry

Abstract

Objective: Hypospadias surgery, especially when performed early in life, may have a significant impact on the urinary and sexual functions in an adult. Because the literature is still limited, this paper assesses long-term functional, cosmetic and sexual results of hypospadias repair performed in childhood.

Patients and Methods: The study includes 275 patients older than 12 years treated for a hypospadias by an Onlay, Mathieu, Duplay, or Duckett's technique between January 1990 and December 2000. Flowmetry results were retrospectively obtained from patients' charts. The Paediatric Penile Perception Score (PPPS), the Hypospadias Objective Scoring Evaluation (HOSE) and the IIEF-5 score (when older than 16 years old) questionnaires were used to assess cosmetic and sexual results. The PPPS is designed to assess both penile self-perception with regard to meatus, glans, skin and general appearance. The HOSE is a five-point scoring system designed to allow an objective appraisal of the outcome of hypospadias repair, based on evaluating meatal location, meatal shape, urinary stream, straightness of erection, and the presence and complexity of any complicating urethral fistula. **Results:** Q_{\max} were within age-adjusted references, independent of the surgical technique, with median (range) Q_{\max} of 18.8 ml/s (range 3–45, $n = 136$). Patients expressed a

high satisfaction for every single item of the penile perception scale (PPPS), with mean values between 2 (satisfied) and 3 (very satisfied). Eighty-two percent were satisfied or very satisfied of the overall evaluation of penile appearance. Eighty-one percent of patients had a normal erectile function (IIEF-5 >22; $n = 35/43$). **Conclusions:** Taking into account the limitation of a small number of patients resulting from a low 21% questionnaire's response rate, the results of this study align with previous reports from the literature and confirms that hypospadias repair using standard techniques results in acceptable functional, cosmetic and sexual outcomes. This study highlights the need of developing a set of standard approved outcomes assessments tools for evaluating the long-term impact of hypospadias repair performed in infancy.

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Introduction

Hypospadias is a common congenital abnormality of the penis diagnosed in approximately 1/200 to 1/300 male newborns. Hypospadias is defined as an insufficient development of the urethral fold and the ventral foreskin, with or without the penile curvature [1–3]. The basic principles of hypospadias surgery are to reconstruct the urethra to the tip of the glans, to straighten the curvature of penis, to achieve acceptable cosmetic penile appear-

ance, and allow normal urinary and sexual function. These principles are well established and hundreds of surgical procedures have been described [2, 4–8].

Usually performed early in life, hypospadias repair may have a significant impact on the urinary and sexual function in adult life [1, 5, 6, 9, 10]. Strikingly, most studies have focussed on the short-term outcomes of hypospadias repair, and not many authors have longitudinally evaluated its long-term cosmetic and functional consequences [1, 3, 11, 12]. A recent review by Tournchi et al. [3, 4] appropriately highlights that problem and concludes that more attention should be paid to the impact of these techniques on cosmetic and psychosexual aspects in patients at the time of and after puberty, considering the impact on their adult quality of life.

In this study, we have assessed long-term functional, cosmetic and sexual results of our series of hypospadias repair using a combination of contemporary validated questionnaires and a retrospective analysis.

Patients and Methods

Our institutional database includes 583 hypospadias operated between January 1990 and December 2000. We have retrospectively selected 275 patients, 12 years old or more, whose hypospadias had been repaired by an Onlay, Mathieu, Duplay, or Duckett's technique in childhood; 78% were primary surgeries, 22% were re-do surgeries. The study has been approved by the institutional Ethical Board.

Age, history, surgical procedures, and flowmetry were retrieved from the patients' charts. Individual flow rates were plotted against age-adapted standardized curve for analysis [1, 3, 13, 14].

Patients were invited, by mail, to fill three validated questionnaires after signed informed consent: the Paediatric Penile Perception Score (PPPS), the Hypospadias Objective Scoring Evaluation (HOSE) and the International Index of Erectile Function (IIEF-5) score, when older than 16 years old.

The PPPS is designed to assess both penile self-perception with regard to meatus, glans, skin and general appearance [1, 3, 14, 15].

The HOSE is a five-point scoring system designed to allow an objective appraisal of the outcome of hypospadias repair, based on evaluating meatal location, meatal shape, urinary stream, straightness of erection, and the presence and complexity of any complicating urethral fistula [8, 16, 17]. The HOSE questionnaire is designed so that the minimum total score would be 5, corresponding to a lowest score for each variable, to a maximum total of 16, equivalent to the highest score for each variable.

The IIEF is a widely used, multidimensional self-report instrument for the evaluation of male sexual function. It has been recommended as a primary endpoint for clinical trials of erectile dysfunction (ED) and for diagnostic evaluation of ED severity. This last questionnaire was sent only to patients at least 16 years old at the date of the study.

Data were analysed using SPSS statistical package. Due to the small number of patients, only descriptive statistics was used.

Results

Surgical Technique

The technique used for hypospadias repair was an Onlay in 125 patients, a Mathieu in 103, a Duckett in 30, and a Duplay in 17. The median and mean age at the time of surgery was 2.9 and 4.5 years old, respectively. Follow-up was calculated from the date of surgery to the date of the last contact. The mean (range) follow-up for the population was 102 months (1–266 months); 99 (1–201) months for Mathieu (n = 103), 93 (1–233) months for Onlay (n = 125), 152 (1–266) months for Duckett (n = 30) and 111 (1–147) months for Duplay (n = 17). The median age at the time of this study was 20.75 (n = 275 patients, 13.5–43 years).

Fistulas and Stenosis

Thirty-four (12.3%) fistula (34/275) were identified; 20/125 (16%) Onlay, 5/103 (5%) Mathieu, 4/30 (13%) Duckett, and 5/17 (29%) Duplay's. Three resolved spontaneously within a few months from diagnosis; 31 required surgical correction. Twenty-five patients (25/275, 9%) underwent a surgery for stenosis, including 15 me-atoplasty and 10 more proximal surgeries. Five stenosis were associated with a fistula.

Flowmetry

Postoperative flow charts were available for 136 (50%) of the 275 patients. Flows with fistula/breakdown were not recorded. Individual maximal flow rates (Q_{\max}) were plotted against age-adjusted standard [13, 14, 18]. Overall, individual Q_{\max} were within age-adjusted references, independent of the surgical technique, with median (range) Q_{\max} of 18.8 ml/s (range 3–45, n = 136). Q_{\max} was below the 5th percentile in 13 of 136 (9.5%) patients, not being affected by the surgical technique. Spraying, as reported in the HOSE questionnaire, was found in 17 (29%) of 58 patients. Aho et al. [19] in their series of 48 patients who had undergone the Mathieu technique in childhood, reported 63% of spraying.

Evaluation of Cosmetic Results (Tables 1, 2)

The PPPS and HOSE questionnaires were returned by 58/275 (21%) patients. Most patients expressed a high satisfaction for every single item of the penile perception scale, with mean values between 2 (satisfied) and 3 (very satisfied). Twenty-nine percent of the patients (17/58) were 'very satisfied' or their overall of penile appearance, 53% (31/58) were 'satisfied'. Only 17% of the patients (10/58) were 'poorly satisfied' of their penile appearance, the main reason being an apparent small penile size. Significant curvature (>10%) was found in 8/58 patients (13.8%, HOSE score).

Table 1. PPPS of 58/275 patients

	Mean	SD
Meatus	2.09	0.82
Glans	2.33	0.60
Shaft skin	2.02	0.68
General appearance	2.12	0.66
Overall PPPS	8.50	2.64

Table 2. HOSE results of 58/275 patients

HOSE 1: meatal localisation	Glans distal	41
	Glans proximal	13
	Coronal	3
	Penile	1
HOSE 2: meatal aspect	Vertical	43
	Circular	15
HOSE 3: flow	Simple	41
	Spraying	17
HOSE 4: curvature	No curvature	29
	<10°	21
	10–45°	7
	>45°	1
HOSE 5: fistula	None	48
	Single distal	10
	Single proximal	0
	Complex	0
HOSE: total	<10	1
	10–13	15
	14–16	42

Sexual Function

Sexual function in patients aged 16 or more was assessed using the IIEF-5 questionnaire. In total, 81% of patients had a normal erectile function (IIEF-5 >22; n = 35/43). Eight patients had a mild ED (IIEF-5 between 17 and 21; n = 8/43), including 4 patients having been operated more than 3 times.

Discussion

In contrast to the numerous studies describing hypospadias repair's surgical techniques and short-term outcomes evaluation of these surgeries, there are a very few studies reporting on long-term sexual and functional outcomes during adolescence and early adulthood [1, 3]. All these studies are retrospective reviews that have been compiled in recent systematic review [4, 5]. Table 3 summarizes the studies included in Rynja's meta-analysis [5].

In addition to being retrospective, these series have usually included a limited numbers of patients, from 19 to 196, used several patient's outcome measurement methodologies, and had low response rates in the retrospective questionnaire surveys. The present series clearly had a setback due to these limitations; this study's questionnaire response rate was 21% (58/275), thus being 'unfortunately' in line with previously reported studies [3, 11]. The follow-up is very difficult especially because the patient will experience a change of status within years, passing from child to adolescent and then to adulthood [3].

Hypospadias repair is a surgery performed early in life, with many consequences emerging in the late adolescence when cosmetic results and sexual functions are becoming more of a concern [20, 21]. In the absence of prospective 'functional' registry, studies evaluate these aspects, including this one, bearing consistent flaws. First and most importantly, hypospadias' severity and associated malformations are extremely diverse, resulting in numerous repair techniques having been used over the time, especially since most patients have been operated one or two decades ago. This is why, in this study, we have focussed on the more commonly practiced techniques, yet including all stages of hypospadias.

It was not until a few years ago that surgeons have started to pay more attention to the perception of the cosmetic results by the patients and the psychological impacts of hypospadias repair [1, 3, 14]. This was wrong since most independent studies have agreed on the conclusion that there was a significant degree of social and sexual morbidity after hypospadias surgery, probably reflecting patient dissatisfaction with the standard of repair. In particular, the importance of good cosmetic results has been underestimated, since it seems to be of greater priority to the patient than to the surgeon [1, 3, 14]. One of the conundrum of evaluation cosmetic results, however, is the lack of consensus on the best methodology to apply [8, 17]. Cosmetic and functional outcomes indeed are often subjective and quite variable between parents, doctors and patients [14, 18]. In that regard, we may assume that a self-administered questionnaire would be a better method to elicit reliable answers than a direct interview for such delicate and personal matters. When contacted directly with doctors, patients may not provide frank answers because of certain assumptions or fears [22].

In the present trial, we have used the PPPS and the HOSE score, both widely used instruments in hypospadias studies. The PPPS has developed and validated in a cross-sectional study on the psychosexual development and health-

Table 3. Long-term results (studies included in Rynja's analysis)

First author, year	Spraying	Q _{max} , ml/s	Q _{max} <95th, ml/s	Dissatisfied appearance, n (%)	Curvature, n (%)	Meatal position glandular, n (%)	Slit like, vertical meatus, n (%)	Erectile dysfunction, n (%)
Aho 1, date, 1997	25/41	–	–	6/41	2/41	–	–	–
Aho 2a, 2000	29/46	–	–	10/45	4/46	–	–	–
Aulagneb, 2010	13/27	–	–	–	5/27	26/27	15/27	0/27
Bracka, 1989	75/196	–	–	60/196	39/196	–	–	–
Hoag, 2008	10/21	–	–	8/28	–	–	–	1/26
Jones, 2009	11/53	–	4/43	10/52	6/50	46/50	41/50	–
Kumarb, 1994	14/35	26	1/27	–	5/35	25/27	–	–
Lamb, 2004	10/25	–	–	23/27	0/27	–	27/27	–
Millerb, 2005	9/19	8.4–22	–	2/19	–	–	–	4/19
Mondainia, 1997	–	–	–	11/42	1/42	–	–	–
Moriya 1a, 2006	–	–	–	9/22	5/22	–	–	0/22
Rynjaa, 2009	8/43	31.1	4/30	4/31	1/31	30/31	–	25/62
vd Werff 1, 1997	27/175	–	23/156	–	–	–	–	–
vd Werff 2, 2000	14/137	–	–	–	6/137	–	–	–
Total hypospadias	245/818	24.4	36/265	125/347	74/654	127/135	73/104	30/156
Total, %	29.95		13.58	36	11.31	94.10	71.19	19.20
Our series	17/58	19	13/136	10/58	8/58	54/58	45/58	8/43
Total, %	29		9.5	17	13	93	77.6	19

related quality of life of children and adolescents with hypospadias [15]. This was the first instrument to be available to objectively assess the surgical outcome and self-perception after hypospadias repair [15]. The HOSE is a scoring system designed to allow an objective appraisal of the outcome of hypospadias repair, based on evaluating meatal location, meatal shape, urinary stream, straightness of erection, and the presence and complexity of any complicating urethral fistula [16]. A HOSE score of more than 14 is defined as normal anatomical outcome [16]. Seventy-two percent of the patients reported a HOSE score above 14, similar to the 80% reported by the series of Jones et al. [17].

Even more difficult to interpret is the perception of the quality of the urinary flow, that reflects patient's and or parents' vague impression, keeping in mind the remarkable adaptation of the child to an impaired urethral flow [8]. In an attempt to objectify this analysis, we have reported instead a retrospective review of flowmetry results, plotted against age-adapted curve [13]. Few studies have used flowmetry for evaluation after hypospadias repair. We do not confirm the observation by Rynja et al. [5] that only patients with proximal hypospadias have lower mean Q_{max} compared to control patients.

The correct evaluation of sexual activity in adolescent and young adult is still a matter of discussion with no specific instrument having been developed for. The IIEF is a

widely used, multidimensional self-report instrument for the evaluation of male sexual function primarily developed as a primary endpoint for clinical trials on ED. This questionnaire has been developed for adults and the transposition to an adolescent population may not be exactly adequate [5]. In this study, IIEF-5 questionnaire was sent only to 223 patients aged 16 years or older at the date of the study. The response rate was 20%, and 81% of the patients reported no ED using that questionnaire. We can assume at least that these patients have a satisfactory sexual life. The signification to be given to those mentioning ED should deserve specific attention.

Conclusion

This study highlights the need of developing a set of standard approved outcomes assessments tools for evaluating the long-term impact of hypospadias repair performed in infancy. Beyond the scientific interest for the retrospective evaluation of surgical techniques, this should be inherent to any quality-controlled evaluation of departments performing such studies.

Bearing in mind these limitations, this article suggests that hypospadias repair using standard techniques results in acceptable functional, cosmetic and sexual outcomes.

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