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Original Article

Management of complications after hypospadias repair during childhood

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ABSTRACT

METHODS. This study aims to describe the presenting complaints, clinical findings and management strategies in men with a history of prior hypospadias surgery. Men aged 15 years or older, referred with complaints related to hypospadias, were identified through the electronic patient record system. Data were collected over a ten-year period.

RESULTS. Of 201 referrals, 136 men had a history of hypospadias surgery, with a median age at referral of 33 years (range: 15-72). The most common presenting complaints were urethral stricture (n = 63) or meatal stenosis (n = 14), which were more frequently observed in older patients. Other presenting complaints were urethrocutaneous fistula (n = 24), preputial problems (n = 8) and penile curvature (n = 7). An additional 20 men presented with various complaints such as spilt urine stream, episodes of haematuria, anejaculation or coital pain. Management strategies included reconstructive procedures (e.g., single or staged urethroplasty), minor procedures (such as Nesbit procedure or fistula closure), endoscopic interventions (urethral dilation or urethrotomy) or counselling.

CONCLUSIONS. Complaints following previous hypospadias repair are present across the entire adult age span. Therefore, caregivers of young boys must be informed following hypospadias surgery to seek medical evaluation whenever hypospadias-related complications are suspected, even decades after reconstruction.

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Hypospadias is among the most common congenital anomalies, affecting approximately one in 250 live male births [1, 2]. While the underlying causes of hypospadias remain largely unclear, the condition is characterized by a ventrally displaced urethral meatus, a deficient ventral foreskin and varying degrees of penile curvature. These features may appear in different combinations and severities [3].

Surgical correction, aimed at restoring both normal penile function and appearance, is typically performed in early childhood using a variety of surgical techniques [4]. Following surgery, most paediatric centers discharge patients from routine follow-up when stable, which may explain why most follow-up studies focus on younger boys rather than on long-term outcomes [5].

There is, however, growing evidence of significant late-presenting complications in adults who underwent hypospadias repair during childhood. Reports indicate that up to half of these individuals may experience issues such as voiding difficulties and reduced sexual function [6]. Physical changes during puberty have also been suggested as a potential contributing factor to these late-presenting complications [7].

In this retrospective cohort study, we examine the complaints, clinical findings and management strategies for men presenting with complications resulting from hypospadias repair performed during childhood.

Methods

Study design

The electronic patient record system MIDT-EPJ was reviewed to identify primary referrals of males aged 15 years or older with a diagnosis of hypospadias, as classified by ICD-10 codes (Q540, Q541, Q542). These patients were seen at the authors' congenital urology transitional clinic between 2009 and 2019. A total of 201 records with at least one relevant ICD-10 code were examined, and males aged 15 or older with a history of previous hypospadias surgery were included in the final study cohort (n = 136).

The study protocol was approved by the regional ethics committee and the Danish Patient Safety Authority (case number 3-3013-2378/1), in accordance with Danish regulations. Data collected included primary referral complaints, hypospadias grade (anterior, middle, or posterior) [8], surgical interventions and follow-up details.

Statistical analysis

According to the non-parametric distribution of data, all results are presented by their median (range) values. Comparisons between independent groups were undertaken by use of the Kruskal-Wallis analysis of variance on ranks test. $p < 0.05$ was set to signify statistical significance. All data were analysed using STATA 12.1.

Trial registration: not relevant.

Results

Study population

Over a ten-year period, 201 men aged 15 or older with hypospadias were referred to our congenital urology transitional clinic. Of these, 136 patients (67.7%) had undergone hypospadias repair prior to referral, constituting the study cohort for this paper. The remaining 65 adult men with uncorrected hypospadias have been described previously [9].

Patients were referred by other urology departments (61.8%, n = 84), followed by general practitioners (29.4%, n = 40), private urology specialists (5.1%, n = 7), plastic surgery (2.2%, n = 3) and other specialties (1.5%, n = 2). The original meatal position prior to surgery could be determined for 78 patients (57.4%) through patient record review, with middle-grade hypospadias being the most common (n = 35), followed by anterior (n = 34) and posterior grades (n = 9). There were no significant differences between patients with and without the documented original meatal position in terms of referral complaints (p = 0.57), age at referral (p = 0.50) or post-referral interventions (p = 0.53).

The median age at referral was 33 years, ranging from 15 to 72 years.

Presenting complaints

The majority of men (46.4%, n = 63) were referred due to symptoms of urethral stricture, which was confirmed through cystourethroscopy or voiding cystourethrography. Urethrocutaneous fistula was the presenting complaint for 17.6% (n = 24) of cases, while 10.3% (n = 14) were referred for meatal stenosis, 5.9% (n = 8) for preputial issues and 5.1% (n = 7) for penile curvature. The remaining 14.7% (n = 20) were referred for various other issues, including split urinary stream, episodes of haematuria, pain during intercourse and anejaculation (**Table 1**).

TABLE 1 Baseline characteristics of the study population.

Primary referral complaints	Men	Urethral stricture	Meatal stenosis	Urethrocutaneous fistula	Penile curvature	Preputial issues	Miscellaneous
Patients, n (%)	136 (100)	63 (46.4)	14 (10.3)	24 (17.6)	7 (5.1)	8 (5.9)	20 (14.7)
Age, median (range), yrs	33 (15-72)	40 (15-66)	41 (22-70)	24 (15-72)	20 (16-27)	19 (17-27)	33 (15 -68)
Uroflowmetry							
Uroflowmetry, n	-	46	12	13	6	5	14
Flow rate, median (range), ml/sec.	-	10.5 (1.0-31.0)	7.2 (1-16)	25.9 (11.0-40.0)	23.5 (21.0-30.0)	26.0 (22.0-31.0)	20.0 (1.0-42.0)
Post void, median (range), ml	-	110 (0-750)	128 (0-400)	54 (0-320)	38 (0-230)	16 (0-80)	92 (0-630)

Age dependent patterns

Men presenting with urethrocutaneous fistula (n = 24, median age 24 years, range: 15-72 years), penile curvature (n = 7, median age 20 years, range: 16-27 years) and preputial issues (n = 8, median age 19 years, range: 17-27 years) were significantly younger than those presenting with urethral stricture (n = 63, median age 40 years, range: 15-66 years) ($p < 0.05$) (Table 1). They were also significantly younger than the men presenting with meatal stenosis (n = 14, median age 41 years, range: 22-70 years) ($p < 0.05$).

Management

Urethral stricture and meatal stenosis

Urethral strictures were primarily treated with internal urethrotomy or urethral dilation (n = 36), staged urethroplasty (n = 12), single stage urethroplasty (n = 5) or ureterostomy (n = 1). One patient had combined fistula closure and internal urethrotomy. Meatal stenosis was managed by meatoplasty (n = 12) and, in one case, single-stage urethroplasty (n = 1).

Urethrocutaneous fistulae

Most urethrocutaneous fistulae were addressed with simple closure (n = 13), while five cases required urethroplasty and one patient meatotomy into the very distal fistula thereby creating a single opening.

Other issues

Penile curvature was managed using either the Nesbit penile straightening procedure (n = 2) or staged urethroplasty (n = 2). Preputial issues were typically managed by circumcision (n = 5), and one in case by preputioplasty. In the 20 men presenting with various other issues, a combination of the procedures mentioned above was used to address their complaints.

Conservative

In 18 patients presenting with issues such as urethrocutaneous fistulae, meatal stenosis, penile curvature etc., surgical intervention was initially deferred, as they were hesitant to commit to any procedures. However, during follow-up, eight patients consented to the recommended surgical treatment to address their condition (**Table 2**). The remaining patients were discharged from our care after receiving thorough counselling.

TABLE 2 Follow-up and complications after primary management in men with previously corrected hypospadias.

	Primary			Secondary		Tertiary	
Primary management	patients, n	follow-up, n	reason for re-treatment/ primary complication	management	complication	management	follow-up, mos.
Conservative	18	15	1 fistula 3 penile curvatures 2 meatal stenoses 1 buried penis 1 phimosis	3 staged urethroplasties 1 Nesbit 2 meatoplasty 1 buried penis procedure 1 preputioplasty	-	-	22.7 (3.5-88.9)
Circumcision & preputioplasty	7	6	None		-	-	12.0 (4.7-24.4)
Single urethroplasty	11	10	None		-	-	13.1 (6.3-17.2)
Staged urethroplasty	16	15	2 acquired undescended testes 1 graft failure 1 webbed penis 1 penile curvature 1 meatal stenosis	2 orchiopexies 1 re-do staged urethroplasty 1 scrotoplasty 1 Nesbit 1 meatoplasty	-	-	13.1 (6.3-17.2)
Urethrotomy/meatoplasty/ dilatation	50	42	22 recurrent strictures	8 internal urethrotomy 1 meatoplasty 6 single staged urethroplasties 8 staged urethroplasties	3 recurrent strictures	3 internal urethrotomies	16.7 (1.6-125.3)
Nesbit	2	1	None				-
Fistula closure	14	10	4 recurrent fistulas	2 single staged urethroplasties 1 declined further treatment 1 re-do fistulae closure			5.2 (2.3-24.3)

Uroflowmetry

In the subgroup of patients managed for urethral stricture at our centre with complete uroflowmetry recordings (n = 46), a statistically significant improvement in urinary maximal flow rate was observed, increasing from a median of 10.5 ml/sec. (range: 1.0-31.0 ml/sec.) before management to 18.5 ml/sec. (range: 2.0-60.0 ml/sec.) following management (p = 0.0003). Postvoid residual volumes showed a decreasing trend, from a median of 110 ml (range: 0-750 ml) to 64.5 ml (range: 0-600 ml), although this difference was not statistically significant (p = 0.1942).

Counselling

Fifteen men were discharged from the transitional clinic after their initial visit, as they declined proposed treatment to manage their complaints, and were content with general information about their condition. Most of these patients (n = 9) reported issues such as split urinary stream, episodes of haematuria, pain during intercourse or anejaculation.

Follow-up

The median follow-up after each procedure performed was 4.1 months (range: 0.1-17.1 months). Overall, the median follow-up duration for all patients was 16.8 months (range: 1.6-125.3 months). Nineteen patients (16%) were lost to follow-up. Detailed follow-up outcomes following primary management are shown in Table 2.

Discussion

Long-term complications following hypospadias repair in childhood were present across all adolescent / adult age groups in our study population.

Among the men with documented information on the original meatal position, middle-grade (n = 35) and proximal-grade (n = 9) hypospadias were most prevalent. This trend aligns with findings from the largest retrospective study on failed hypospadias repair to date (n = 1,179), conducted by Bagbagli et al. [10] Similarly, multiple re-interventions were most common amongst men with severe phenotypes according to another cross-sectional study [6]. These findings suggest that proximal hypospadias carries the highest risk of post-operative

complications. However, even in milder variants of hypospadias, long-term complications are common, as evidenced by the referral of 34 men with a history of distal hypospadias managed in childhood. Our finding of a bimodal age-dependent pattern of post-surgical complications, with obstructive symptoms being more frequent in older age groups than other complications, such as urethrocutaneous fistulae presenting in younger men, has also recently been reported in other studies [9, 11].

As hypospadias is usually managed in early childhood and is primarily overseen by paediatric urologists and paediatric surgeons, most follow-up protocols tend to end abruptly with boys reaching the age of 18 years and thereby no longer subject to the responsibility of paediatric departments. This stratification between adult and paediatric healthcare can be problematic with regards to hypospadias, as complications that have not manifested in childhood may not become evident until later in life. This is notable especially for urethral stricture disease, as the onset can be insidious and progressive, leading to infravesical obstruction with all its attendant complications. Many of our patients had previously been treated by non-specialised urologists with temporising procedures or previous repeated repairs, further comprising the quality of tissues and the urethral plate.

Urethral stricture in adult men with previous hypospadias repair can also occur decades after initial surgery and may be due to genital growth during puberty, lack of spongiosum covering the neourethra or balanitis xerotica obliterans affecting the genital tissues. Management can be challenging as excision of scarred tissue, which may include part, or even the whole length of the neourethra, may be needed in order to perform substitution urethroplasty [12, 13].

In the current cohort, we applied the same surgical principles in management of long-term hypospadias complications in the adult patient as the ones employed in childhood. A similar strategy has been described in other studies with good outcomes on the short and medium term, as seen in the present study [9-11]. In general, we managed urethral strictures with endoscopic assessment and initial dilatation; however, in many cases, it was apparent that long-term success would require more extensive reconstructive surgery. Notwithstanding, many of our patients with obstructive flow opted for repeat dilatations/endoscopic management of urethral strictures, as many had reservations with regards to undergoing definitive reconstructive surgery. Repeated endoscopic interventions were rarely curative, which can be seen in the current cohort, where 60% of patients undergoing endoscopic management proceeded to further intervention (usually single or staged urethroplasty) (Table 2). Therefore, we strongly recommend definitive management of fibrotic and long urethral strictures through formal urethroplasty.

Urethrocutaneous fistulae were managed by simple closure if the location of the fistula and the surrounding tissues allowed for it, otherwise redo urethroplasty by single or staged procedure was performed. Penile curvature was managed by corporal realignment using the Nesbit principles, unless the curvature was caused by a short fibrotic neourethra in which case a staged substitution urethroplasty was employed. As for preputial issues, circumcision is usually the optimal management option in these patients.

The present study is limited by its retrospective nature. Most of our patients had no exact records of number or type of previous hypospadias-related surgery. Furthermore, the findings of this study may present outliers within the population of men with complaints following previous childhood hypospadias surgery, as not all men with problems following surgery seek medical attention.

Conclusions

Hypospadias surgery in childhood predisposes patients to a life-long risk of complications, and long-term follow-up therefore needs to be provided. The watershed gap between paediatric and adult urology departments has to

be bridged by bespoke tailored transition programs, and adolescent patients should be aware of where, whom and when to contact adult departments and specialists.

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