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A Review of Literature on Long-Term Outcomes of Proximal Hypospadias — Urinary, Sexual, and Psychosocial

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Abstract

Purpose of Review Proximal hypospadias management involves complex surgical interventions that can lead to the need for long-term specialized care. This review aims to explore the urinary, sexual, and psychosocial outcomes in patients with proximal hypospadias.

Recent Findings Patients undergoing surgical correction of proximal hypospadias have generally acceptable urinary flow rates but can still suffer from increased rates of urinary tract infections, voiding difficulties, and leakage as compared to controls. Sexual outcomes of surgical repair closely mimic those of controls. However, a significant proportion of patients report dissatisfaction with their penile appearance after cosmetic surgery. While patients may report hesitancy in approaching sexual or emotional relationships, overall psychosocial outcomes were close to those reports in controls.

Summary While still generally very successful, proximal hypospadias surgery has been shown to have more complications in these areas than other milder forms of hypospadias and thus care must be taken to ensure patients are being treated with a holistic mindset throughout their journey.

Keywords Proximal hypospadias · Long-term outcomes

Introduction

Hypospadias is a relatively common congenital abnormality, with an incidence of 1 in 300 male live births. However, proximal hypospadias is uncommon but has a more complex variation with the meatus presenting in the penoscrotal or perineal positions [1]. This often leads to the need for staged and more complex surgical interventions and specialized care. Surgical techniques for hypospadias have significantly evolved over time, from primarily focusing on urinary outcomes to now also focusing on cosmetic results [2].

Multi-stage procedures were initially used to straighten penile curvature and create skin grafts as needed, but this evolved into a single-stage operation with the advent of Snodgrass's tubularized incised plate (TIP) technique for distal hypospadias [3]. However, proximal hypospadias repair presents more challenges and there exists a lack of standardization for surgical corrective procedures. Two-stage approaches are still commonly used involving correcting the chordee and creating a tubularized neo-urethra at the 2nd stage [4]. Proximal hypospadias repairs are also associated with higher risk of reoperations [5]. Immediate goals of repair include straightening the penis and ensuing general functional urinary capabilities, but surgical outcomes have been shown to have significant impacts on psychosocial and sexual outcomes of patients and thus have the potential to have lasting effects [6•].

Methods

A literature search was first conducted for proximal hypospadias surgical outcomes published from 2015 to 2020. One

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hundred and seventy-two articles were retrieved and started the screening process. Seven duplicates were removed, and 165 studies moved on to the title and abstract screening. Finally, 18 full text studies were included for this review as seen in Fig. 1. Authors participated in all screening steps, and any inclusion or exclusion disputes were mediated as needed. Surgical outcomes were then classified into three categories — urinary, sexual, and psychosocial. The cohort of studies included was reviewed for relevant trends and findings in each outcome type and is reported accordingly below.

Urinary Outcomes

Functional outcomes of patients with hypospadias have predominantly been measured through uroflowmetry and postvoid residual volumes [6•]. Through a questionnaire, Örtqvist et al. found 167 hypospadias patients (mean follow-up 29 years) presenting with more voiding dysfunctions symptoms and a lower maximum urinary flow rate than controls [7]. Voiding dysfunction included prolonged voiding, difficulty voiding, leakage, hesitancy, spraying, and urine directed downward [7, 6•]. The mean maximum urinary flow rate for all patients with hypospadias was 18.8 ml/s compared to 26.2 ml/s in controls. Severe hypospadias phenotypes, including proximal hypospadias (13%), were more likely to have markedly lower maximum urinary flow rate [7, 6•]. Andersson et al. (39 patients) found 14% of proximal hypospadias patients to have uroflow $\leq$ 10 mL/s and 19% to have less than $\leq$ 5th percentile uroflow in adolescence [8]. Patients with hypospadias also reported higher rates of urinary tract infections and fistula formation [7, 6•]. Obstructive urinary flow patterns have also been observed after TIP urethroplasty for all types of hypospadias repair [6•, 8]. This is theorized to be influenced by the severity of hypospadias, abnormal elasticity of the neo-urethra, and scar tissue formation [6•] (Table 1).

In a study comparing different types of surgical procedures used in proximal hypospadias repair and its complications, Hueber et al. (50 patients, mean follow-up 10 years) contrarily showed spontaneous and progressive improvement of maximum urinary flow rate with age and during adolescence [9]. After age 13, > 80% of the cases were found to have a maximum flow rate in the normal range. They concluded that though obstructive urinary flow was frequently observed after proximal hypospadias surgery, conservative methods such as watchful waiting could prevent unnecessary future procedures as urinary flow will improve over time. Overall functional outcomes of hypospadias repair are generally acceptable but still inferior to controls [6•].

Due to the discrepancy in urinary outcomes between studies, recent surgical techniques performed in proximal hypospadias repair in the past 5–7 years should be further investigated with long-term follow-up.

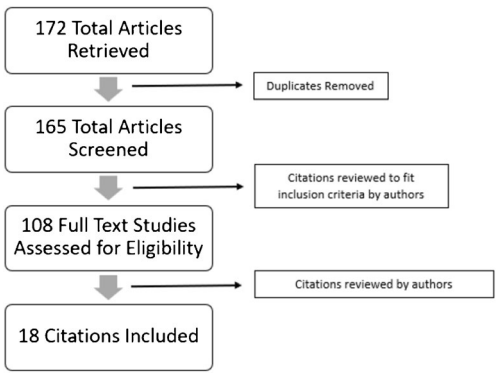

Fig. 1 Literature review PRISMA table

Sexual Outcomes

Sexual Function

Another main goal of hypospadias surgical repair is to obtain satisfactory sexual function by straightening the penis. Persistent chordee following hypospadias repair leads to recurrent complications as well as challenges with sexual outcomes (Table 2). Studies have shown patients with repaired hypospadias, including mild to severe cases, to have a sexual and marital life similar or equal to that of normal adult male population [10, 6•]. Sexual satisfaction has also been reported in more than 80% of patients [6•, 11]. However, more severe types of hypospadias, i.e., proximal, were found to have mild erectile dysfunction (75%) and premature ejaculation (66%) [12–14]. Successful achievement of intercourse was at lower rates in patients with proximal hypospadias compared to those who had milder types [12]. Patients with proximal hypospadias with curvature were also more likely to be dissatisfied with length and straightness during penile axis erection compared to patients with milder hypospadias who reported equal satisfaction with controls. Anejaculation (10% vs. 4%, control) has also been reported in patients with proximal hypospadias [14•]. From a questionnaire, glanular sensitivity was not affected even after multiple procedures [7]. Per Kanematsu et al., patients with proximal hypospadias did not achieve final success with intercourse at the rate of patients with milder hypospadias (72% vs 100%) [10]. This may be due to smaller penis size in proximal cases or an impaired pituitary axis [10, 15].

Erectile Dysfunction

The International Index of Erectile Function (IIEF-5) is a screening tool for assessing erectile dysfunction. Tack et al. reported that the severity of hypospadias had no impact on the IIEF score [16•]. The rate of discomfort in erection and problems with ejaculation were similar in patients with proximal

Table 1 Urinary outcomes for proximal hypospadias repair from current literature

Publication	Study design	Cohort size (number of patients)	Patients with proximal hypospadias	Mean age (years)	Mean follow-up time (years)	Study overview	Outcomes
Örtqvist, 2014, Sweden	Retrospective cohort study	167	13	34	29	Assess urinary and cosmetic outcomes in adolescent patients after hypospadias repair	Hypospadias patients were found more voiding dysfunction ($p = 0.003$) and lower maximum Qmax ($p = 0.001$) than controls.
Horst and Wall, 2016, Netherlands	Review Article	n/a	n/a	n/a	n/a	To explore management of hypospadias	Functional and cosmetic long-term outcomes are generally acceptable but still inferior to the situation in men without hypospadias.
Hueber, 2016, Canada	Retrospective cohort study	52	n/a	n/a	10	Compare Qmax post primary proximal hypospadias repair in TIP vs. Onlay vs. Duckett methods	84% of patients aged 3–7 years had significantly decreased Qmax, but this reduced to 21% after age 13 in all procedure types.
Anderson, 2020, Sweden	Prospective cohort study	39	39	16.5	n/a	Assess psychosocial, sexual, and urological outcomes and complications in adolescent patients after proximal hypospadias repair	14% of the proximal hypospadias patients had impaired flow rate and 19% had less than ≤ 5th percentile uroflow in adolescence.

n/a Information not provided

hypospadias and controls. Approximately 10% of patients reported having problems with mild erectile and ejaculation. However, the study found no association with erectile dysfunction, suggesting that it was likely a random occurrence [16•]. On the other hand, Kanematsu et al. argues that the IIEF-5 questionnaire does not seem to be a suitable tool for screening as it presumes that respondents have or have had regular intercourse with partners [10].

Cosmetic Appearance

Penile Perception Score (PPS) is a self-assessment that rates appearance of penile length, meatus urethrae, glans, shaft skin, penile axis, and general appearance on a 4-point scale [16•]. Örtqvist et al. and Tack et al. observed differences with PPSs in patients with all forms of hypospadias compared to those without [7, 16•]. Tack et al. also noted 13.5% hypospadias patients to be dissatisfied with their penile appearance, especially in proximal hypospadias patients, compared to 6% of controls [16•]. In another study, 50% of patients with proximal or complex hypospadias reported dissatisfaction with the cosmetic outcome of their penis [6•]. On the other hand, Andersson evaluated patients with only proximal hypospadias and did not find a difference in PPS total sum [14•]. Both studies observed that patients with proximal hypospadias were often more dissatisfied with penile length compared to the distal hypospadias or controls (10 cm vs 7 cm, controls) [7, 14•]. No difference in scrotum and testicular size was observed. For proximal hypospadias patients, there was satisfaction with meatal position and shape [14•, 8]. Sexual activity even occurred at similar rates compared to the general population [14•]. In a Sexual Quality of Life-Men (SQoL-M) assessment, there was no difference found between cases and controls [16•]. However, patients with proximal hypospadias were found to have a lower score compared to the patients with distal or mid hypospadias [16•].

Psychosocial Outcomes

Psychosocial outcomes in patients with hypospadias are mainly assessed through questionnaires (Table 3). Örtqvist et al. assessed quality of life in patients with all forms of hypospadias, including proximal type, using the Psychological General Well Being (PGWB) [17]. Patients' self-perceived evaluation was found to be comparable to the general population. There was no difference in the anxiety, depression, self-control, general health, and total PGWB sum subgroups. However, patients scored lower on the well-being and vitality subgroups, which may suggest decreased quality of life in patients with all forms of hypospadias, including the proximal type.

Table 2 Sexual outcomes for proximal hypospadias repair from current literature

Publication	Study design	Cohort size (number of patients)	Patients with proximal hypospadias	Mean age (years)	Mean follow-up time (years)	Study overview	Outcomes
Kanematsu, 2016, Japan	Prospective cohort study	108	36 proximal	28	23	Assess achievement of marriage, paternity, and sexual intercourse in adult males with hypospadias	Achievement of sexual intercourse was associated with milder hypospadias (glandular $p = 0.025$; penile $p = 0.0076$) and achievement of paternity was associated with no reoperation after initial repair ($p = 0.013$).
Horst and Wall, 2016, Netherlands	Review Article	n/a	n/a	n/a	n/a	To explore management of hypospadias	80% of patients with all types of hypospadias had satisfactory sexual function.
Winship, 2017, USA	Review Article	n/a	n/a	n/a	n/a	To explore many outcomes of hypospadias repair	Patients with proximal hypospadias were found to have mild erectile dysfunction (75%) and premature ejaculation (66%).
Anderson, 2018, Sweden	Prospective cohort study	33	33	17.5	n/a	Assess psychosocial, sexual, and urological outcomes and complications in adolescent patients after proximal hypospadias repair	Patients with proximal hypospadias reported anejaculation (10%) and dissatisfaction with penile length. No difference in PPS sum.
Tack, 2020, Belgium and Austria	Cross sectional study	193	23	18.14	n/a	Analyze psychosocial and sexual outcomes and patient opinions in young adult males following hypospadias repair	Patients with proximal hypospadias were more likely to be dissatisfied with their penile appearance than their controls (13.5% vs. 6%). There was no association with erectile dysfunction.

n/a Information not provided

Table 3 Psychosocial outcomes for proximal hypospadias repair from current literature

Publication	Study design	Cohort size (number of patients)	Patients with proximal hypospadias	Mean age (years)	Mean follow-up time (years)	Study overview	Outcomes
Örtqvist, 2014, Sweden	Retrospective cohort study	167	13	34	29	Assess urinary and cosmetic outcomes in adolescent patients after hypospadias repair	No difference in self –esteem was found between patients with all types of hypospadias and controls.
Anderson, 2018, Sweden	Prospective cohort study	33	33	17.5	n/a	Assess psychosocial, sexual, and urological outcomes and complications in adolescent patients after proximal hypospadias repair	Education levels were similar to those with distal type of hypospadias and controls. However, extra tutoring was received in school. Twenty-four percent of patients reported that proximal hypospadias negatively affected their childhood.
Kanematsu, 2016, Japan	Prospective cohort study	108	36	28	23	Assess achievement of marriage, paternity, and sexual intercourse in adult males with hypospadias	Patients with all types of hypospadias were found to have similar marital life compared to the controls.
Strandqvist, 2019, Sweden	Prospective cohort study	83	10	32	n/a	Use cognitive tasks and questionnaires on childhood gender roles to assess for differences associated with hypospadias	While no differences were found between men with and without hypospadias, men with proximal hypospadias scored lower on cognitive tasks than men with distal hypospadias
Tack, 2020, Belgium and Austria	Cross sectional study	193	23	18.14	n/a	Analyze outcomes and patient opinions in young adult males following hypospadias repair	Psychosexual outcomes were most dependent on the number of surgeries and satisfaction with penile appearance. 80% of men are satisfied with their childhood operation.
Nordenvall, 2017, Sweden	Prospective cohort study	4378	n/a	n/a	n/a	Assess psychosocial outcomes in adult patients after hypospadias repair	No significant differences were found between men with and without hypospadias other than a 40% increased probability of receiving a disability pension
Örtqvist, 2017, Sweden	Case control study	167	13	34	29	Use the Psychological General Well-Being (PGWB) questionnaire and Relationship Questionnaire (RQ) to assess for a negative impact due to hypospadias	No significant differences were found between men with and without hypospadias in the mean total PGWB scores, however men with hypospadias tended to live with their parents ($p = 0.001$) and had a lower level of education ($p = 0.004$). Men with proximal hypospadias saw more negative psych outcomes as compared to mid or distal hypospadias.

Many studies have shown that patients with proximal hypospadias have more hesitancy in approaching sexual or close relationships and have a fear of being mocked when naked [1, 16•, 17]. Örtqvist et al. administered the *Relationship Questionnaire* to 167 patients (13% proximal hypospadias) which analyzed 4-group models of attachment styles in adulthood [18]. While patients with distal hypospadias did not show a significant difference compared to controls in attachment styles, patients with proximal type of hypospadias showed to have the pre-occupied attachment style where they desired to be close with others, but have fear of being emotionally hurt [17]. They were more likely to feel constrained when making new relationships, in which several patients also reported lower self-confidence. In another study with 193 hypospadias patients, 25.9% of patients were more likely to feel uncomfortable when naked due to fear of being mocked, compared to 20% of controls [16•].

In Andersson et al.'s study, 33 patients who underwent proximal hypospadias repair answered the Psychological General Well-Being Index and Body-Esteem Scale for Adolescents and Adults. Twenty-four percent of patients reported that hypospadias negatively affected their childhood [14•]. This was possibly due to the reoperations needed and the more negative appearance of the penile appearance in patients with proximal hypospadias. Örtqvist et al. found that no increased incidence of bullying was reported [17]. No difference in overall self-esteem or body image was seen between patients with different types of hypospadias and controls [7, 14•]. However, those with proximal hypospadias specifically were more likely to report negative body image [10, 1].

Many worry about fertility issues, masculinity, and male hormone levels as they enter adolescence and adulthood [16•]. Though little is known about sperm quality in men born with hypospadias, it has been shown that more severe forms of hypospadias, such as proximal, are at risk of reduced fertility [19–21]. Kumar et al. analyzed 73 patients with hypospadias, in which 14 were proximal, where a significant difference was found in semen parameters, including semen volume, concentration, and sperm motility in proximal type of hypospadias [19].

Patients overall lead rather normal psychosocial lives with comparable marital status, employment, and presence of children in the family [1, 17]. A lower degree of education was obtained in patients with hypospadias compared to controls [1, 17]. In Andersson's study with 55 patients with only proximal hypospadias, education levels were similar to patients with distal hypospadias and controls [14•]. However, patients with proximal hypospadias did receive extra tutoring in school compared to distal [14•]. In a study with 83 hypospadias patients, 10 having proximal hypospadias, Strandqvist et al. noted that patients with proximal hypospadias were more likely to score lower on cognitive tests compared to patients with distal hypospadias [1]. The patients participated in a series of battery of cognitive tests including vocabulary, spatial directions, mental rotation, episodic memory, word fluency, typing, and emotion recognition. Emotion recognition was the only task that there was no difference between patients with proximal and distal type of hypospadias. Patients with proximal hypospadias were also more often diagnosed with an intellectual disability [22]. Strandqvist et al. inferred that the lower cognitive performance and intellectual disability in patients with proximal hypospadias could possibly due to the comorbidities that may affect cognitive development [1].

In two Swedish studies, patients were also at greater risk of receiving disability pension as an adult and live with their parents for unknown reasons [23, 17]. Patients with proximal hypospadias have overall good results but should receive psychosocial support early in life. The vast majority of patients who underwent hypospadias repair were found to be satisfied having had surgery during their pediatric years [16•].

Conclusion

Surgery to correct proximal hypospadias has significant impact on patients' lives far beyond simply restoring functional urinary capabilities. While urinary outcomes like flow rate and reducing voiding dysfunctions are primary surgical objectives, it is important to consider the lasting sexual and psychosocial implications of the operation. Complications such as obstructive urinary flow, decreased sexual satisfaction, and negative body image can have negative impacts on patient outcomes. Preoperative counseling for parents and patients of an appropriate age is important to ensure positive surgical outcomes. This should be continued post-operatively with proper psychosocial supported through childhood and adolescence. While still generally very successful, proximal hypospadias surgery has been shown to have more complications in these areas than other milder forms of hypospadias and thus care must be taken to ensure patients are being treated with a holistic mindset throughout their journey. Thus, more studies focused specifically on proximal hypospadias outcomes are indeed warranted.

Declarations

Competing Interests The authors declare no competing interests.

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