

Psychosocial Adjustment, Health-Related Quality of Life, and Psychosexual Development of Boys with Hypospadias: A Systematic Review

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Objective A systematic review of studies on psychosocial adjustment, HRQoL (health-related quality of life), and psychosexual development of boys with hypospadias. **Methods** Research was conducted on several online bibliographic databases. Articles were selected on the basis of predefined criteria. Methodological quality was assessed by two independent reviewers who applied a standardized checklist. When possible, data analyses were performed by calculating effect sizes. **Results** Thirteen studies met the criteria for inclusion, whose methodological standard ranged from low to high quality. None of them has focused on HRQoL. Findings with regard to psychosocial and psychosexual adjustment were inconsistent, though they clearly showed that boys with hypospadias suffer from negative genital appraisal and sexual inhibitions. Overall, medical factors exerted a rather small influence. Psychosocial risk factors have hardly been examined so far. **Conclusions** The identification of psychosocial risk factors in methodologically sound studies is necessary to guarantee a comprehensive treatment for boys with hypospadias.

Key words childhood and adolescence; health-related quality of life; hypospadias; psychosocial adjustment; psychosexual development.

Hypospadias is the most common malformation of the penis. It is manifested by an abnormal position of the urethral opening that may lie anywhere between the glans and the scrotum. Severe forms are associated with a curvature of the penis (chordee) and a urethral opening that lies closer to the perineum. In these severe forms, surgical repair in two separate operations must be considered and complication rates tend to be higher. Explanations for the etiology of hypospadias have commonly considered genetic, endocrinological, and environmental factors (Duckett, 1998). Reports on the incidence of hypospadias are inconsistent, but there is evidence that it has increased over the last decades (Dolk, 2004). Dolk estimates an incidence of approximately three per 1000 male births in western countries.

The surgical goal in patients with hypospadias is to achieve a typical-looking penis, to allow urination in the standing position and, particularly in severe forms of

hypospadias, to enable normal sexual function. Progress in surgical techniques and pediatric anesthesiology over the last decades now allow successful surgical repair within the first year of life (Duckett, 1998; Manzoni, Bracka, Palminteri, & Marrocco, 2004). The section on urology of the American Academy of Pediatrics has been updating its recommendations on timing for the surgery of hypospadias. In 1975, it was assumed that the period after the third year of age would be the optimal time for surgical intervention (American Academy of Pediatrics [AAP], 1975). However, in 1996 the best time for surgery to treat hypospadias was believed to be between 6 and 12 months of age. Technical considerations were no longer considered the major limiting factors in determining the timing for the surgery of hypospadias. Considerable importance was instead given to psychological factors such as emotional, cognitive, and body image development that may be affected by the genital deformity and the reconstructive surgery. Superimposed on these

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factors, the developing sexual identity of the individual was believed to be influenced by hypospadias. It was speculated that early genital surgery could minimize disturbances in the patient's psychological development (AAP, 1996).

It is important to note that these recommendations on the timing of surgical correction are not evidence-based. Moreover, self-help groups have asked to reduce the number of surgical procedures for hypospadias because of the potential of surgical complications to cause emotional trauma (Intersex Society of North America [ISNA], 1994, 2006; M. Walker, personal communication, December 13, 2006). From a psychological point of view, it is indeed presumable that children and adolescents with hypospadias experience specific psychological stress, which might contribute to an increased risk of maladjustment and to an impaired quality of life. Apart from the stressors of hospitalization and genital surgery, the boys may grow up with a cosmetically and functionally-impaired penis. In addition, parental anxieties about the boys' future might result in an overprotection, which may limit the patients' development of autonomy and self-confidence (APA, 1996; Berg, Berg, & Svensson, 1982; Del Priore, McHugh, Picton, & Haldane, 2004; Purschke & Standke, 1993; Sandberg et al., 2001; Walker, 1998; Zavitsanakis & Gougoudi, 2004). However, according to Bracka (1999), modern management of hypospadias is much less stressful than it used to be: "There is less physical trauma, with fewer complications, and better postoperative function and cosmesis. Hospitalizations are fewer and shorter, . . ." (p. 30). Therefore, it is also to be assumed that nowadays hypospadias patients grow up with fewer psychological problems than their predecessors (Bracka, 1999).

With respect to the specific development of children with hypospadias, it is important to be not only aware of the potential negative effects of the treatment and complications but also of the potential biological factors that may influence the children's behavior. There is increasing evidence that prenatal exposure to androgens does not only induce the sexual differentiation of the human genitalia but also influences postnatal sex-typed behavior patterns by engendering sex-specific brain structures. Most of the evidence comes from studies of people with disorders of sex development (DSD), which are exposed to an atypical hormone synthesis (Cohen-Bendahan, van de Beek, & Berenbaum, 2005). Jürgensen, Hiort, Holterhus, and Thyen (2007) for instance have recently found a connection between prenatal hypoandrogenization and gender-related behavior in 33 children with XY Karyotypes and DSD. Although the

etiology of hypospadias is still hardly intelligible, it is clearly evident that the faulty embryogenesis of the penile urethra is related to abnormalities in prenatal androgen synthesis and/or androgen receptor defects (Baskin, 2004). Therefore, Sandberg et al. (1995) hypothesized that children born with hypospadias might also tend to atypical gender-role behavior. However, this hypothesis has not been confirmed yet.

Due to the ongoing controversy about the consequences of hypospadias and its treatment, we considered it important to review systematically the research on psychosocial adjustment, health-related quality of life (HRQoL) and psychosexual development of children, and adolescents with hypospadias. While psychosocial adjustment incorporates a range of outcome measures including behavioral and psychosocial factors (Achenbach, 1991), HRQoL is defined as the children's health status, weighted by the emotional response of the children themselves to their health status problems (Vogels et al., 2000). Psychosexual development, however, includes components such as the development of gender identity, gender-role behavior, body image, body inquiry, the development of sexual feelings, first sexual experiences, and sexual behavior habits (Schuhrke, 1997; von Sydow, 1993). Our aim was to answer the following questions:

- (1) Do boys with hypospadias have an impaired psychosocial adjustment, HRQoL, or psychosexual development?
- (2) Are there specific medical and psychosocial risk factors for the outcome?
- (3) How important is corrective surgery and its cosmetic outcome for the boys' adjustment?

Methods

Data Sources and Search Strategy

At the beginning of October 2006, a literature search was conducted for papers that focused on psychosocial adjustment, HRQoL and the psychosexual development of children, and adolescents with hypospadias. The search was set up for the period between 1966 and October 2006. The following electronic bibliographic databases were searched: *Embase*, *PubMed*, *Medline*, *Cinahl*, *PsyINFO*, *Psyn dex*, and *Cochrane Database of Systematic Reviews*. Additionally, the databases *NDLTD*, *ProQuest Digital Dissertation*, and *Dissonline.de* were searched for relevant dissertations in the field. The searches were carried out using the keywords *hypospadias*, *psychosocial*, *adjustment*, *social*, *psychological*, *adaption*,

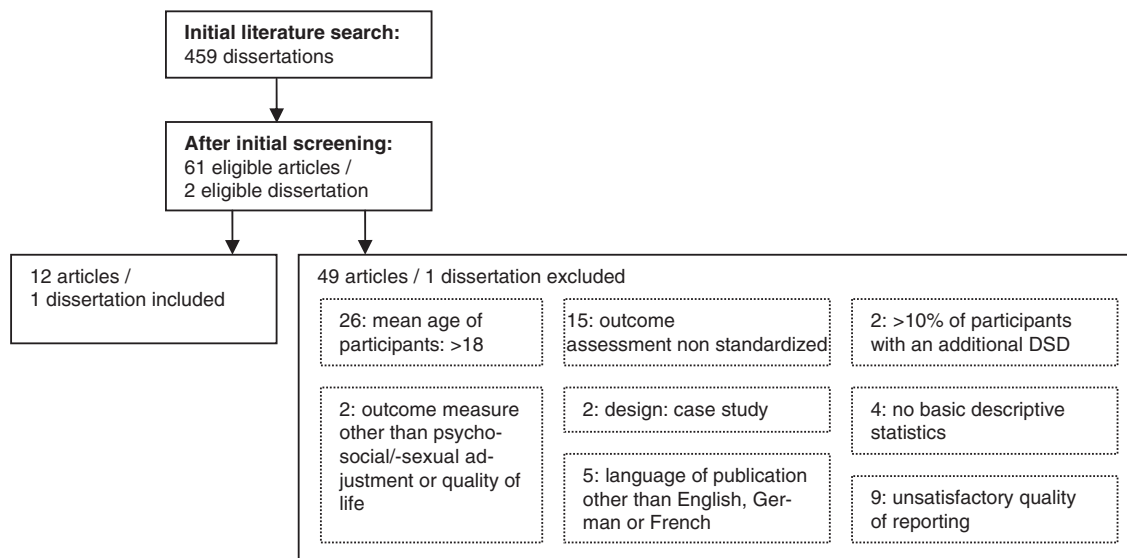


Figure 1. Flowchart of study selection. Fourteen articles have been excluded for more than one reason, e.g., due to nonstandardized outcome measure and mean age of participants >18. The 50 excluded studies are listed in the references and are marked with two asterisks. DSD, Disorder of Sex Development.

development, mental health, behavior disorder, emotional stress, anxiety, personality, quality of life, psychosexual development, sexual satisfaction, sexual behavior, sexual dysfunction, sexuality, sex role, gender identity, and follow up. The Boolean operator *and* was used to link the search term *hypospadias* with each of the residual keywords for the psychosocial and sexual outcome, whereas the Boolean operator *or* was employed to combine the latter with each other. This initial literature search yielded 364 articles and 83 dissertations. The *Cochrane Database of Systematic Reviews* provided five systematic reviews, but none of them was on the topic concerned. To augment this search result, we examined the reference lists of relevant studies and reviews. This resulted in seven additional articles. Moreover, investigators in the field were contacted for unpublished data to minimize the problem of publication bias. This contact revealed no additional eligible data.

Study Selection

First, titles and abstracts of articles were reviewed to screen for eligibility. Eligible publications were limited to trials that investigated psychosocial/-sexual adjustment or HRQoL of hypospadias patients in childhood and/or adolescence. Papers dealing only with the psychological consequences of hypospadias in adulthood or with medical conditions and surgical treatment were ruled out. Reviews, commentaries, and studies related to other urological anomalies were also eliminated. Furthermore, two articles and one dissertation were not included

because the same data were published in more than one research report.

After this initial literature review, 61 articles and two dissertations remained. Two authors (V.B.S., M.A.L.) read the selected articles independently from another, in order to determine inclusion. When necessary, corresponding authors were contacted for additional information or clarifications of inconsistencies in the texts. Papers were included if they met the following criteria: (a) >90% of study sample comprised individuals with hypospadias as solitary diagnosis (hypospadias as a concomitant phenomena in a defined DSD as for example partial androgen insensitivity syndrome, was not taken into account); (b) sex of participants: male; (c) mean age of participants: ≤ 18 years; (d) outcome: standardized self, proxy or examiner's report of psychosocial, -sexual adjustment and/or HRQoL; (e) design: case control, cross-sectional, prospective, retrospective; and (f) language of publication: English, German, French. In addition, studies were excluded if they assessed sexual function or surgical outcome only, used no basic descriptive statistics (e.g., *M*) or lacked satisfactory quality of reporting (e.g., age at follow up missing). However, discussions were ultimately required in order to conciliate existing disagreements between the two authors. Finally, the systematic study selection led to 12 articles and one dissertation (Fig. 1).

Assessment of Methodological Quality

Two independent reviewers (V. B. S., M. A. L.) rated the methodological quality of each included study by using a

self-developed standardized checklist consisting of standards related to sample size, data selection, response rate, design, outcome assessment, statistical procedures, reporting of medical characteristics, and age range of sample. Each criterion was assessed on a two-point scale (0–1) with 1 indicating a better quality. Overall scores ranged from 0 (poor) to 1 (excellent). Interrater reliability was satisfactory (Cohen's $\kappa = .92$; range: 0.71–1.00). In case of disagreement, consensus was achieved by discussion.

Data Analyses

In order to be able to compare the results across the different studies, we calculated effect sizes (ES) for group differences. For continuous outcomes, we calculated standardized mean differences according to Cohen (1988), using pooled standard deviations of the two groups. For dichotomous outcomes, ES were estimated based on Hasselblad and Hedges (1995). Both dichotomous and continuous outcomes were presented with 95% confidence intervals. An ES of 0.2 is considered to be small, an ES of 0.5 to be moderate, and an ES of 0.8 to be large (Cohen, 1988). A positive ES reflects a higher score in the hypospadias group, whereas a negative score reflects a higher score in the control group. Sandberg, Meyer-Bahlburg, Aranoff, Sconzo, and Hensle (1989) presented group differences in gender-role behavior separately for five age groups. In order to summarize the data and simultaneously maintain the presentation of age effects, we combined the data into two age groups (6–8 years, 9–10 years) by calculating weighted mean differences (WMD) (Table II).

However, several studies either lacked comparison data or did not report the necessary details for the calculation of ES (Table II). Furthermore, the sample of studies included in the present review was small and the outcome measures as well as the assessed constructs differed greatly across the studies (see also Section “Study description”). Therefore, the performance of systematic meta-analyses was considered to be inadequate. Instead, we had to narratively summarize the results but gave specific emphasis on the calculated ES. In addition, we explicitly mentioned the specific constructs or measures used in the respective study in order to mirror the heterogeneity of the assessed constructs, which are not always directly comparable (e. g., personality profile and behavior problems).

Results

Study Description

The main characteristics of the included studies are summarized in Table I. Four of the 13 publications

focus on psychosocial adjustment, six on psychosexual development, while three focus on both outcome variables. However, none of the involved studies has investigated HRQoL. The articles were published between 1989 and 2005. Four come from the United States, eight from Western Europe, and one from Turkey. They were all published in English with exception of the studies by Purschke and Standke (1993) and Glaser (2004), which were both written in German. The sample sizes varied from 20 to 175 patients. Their age at follow-up ranged from 1 to 41. All of the studies but two (Mondaini et al., 2002; Purschke & Standke, 1993) incorporated only patients after corrective surgery. They were operated on at the ages of 1 month to 12 years between 1975 and 2003 (Mureau, Slijper, Slob, & Verhulst, 1997; Nelson, Bloom, Kinast, John, & Park, 2005). Except for Nelson et al., all studies included patients with both mild and severe forms of hypospadias.

With respect to the corresponding methodological quality, the studies considerably differed from each other ($M = 5.44$; range: 2–9): Studies on psychosocial adjustment showed a slightly higher methodological quality ($M = 5.86$; range: 3–9) than studies on psychosexual development ($M = 5.11$; range: 2–8). Main weaknesses of the studies included small sample sizes, lack of comparison data, the examination of only single informants, missing information about medical characteristics, and the use of nonvalidated measures. The latter mainly referred to the studies on psychosexual development apart from Mureau, Slijper, Slob, and Verhulst (1995b) and Sandberg et al. (1989, 1995). Moreover, all of the publications documented cross-sectional studies. Furthermore, the operationalizations of psychosocial and psychosexual outcome differed considerably across the studies as various constructs such as behavior problems, self-confidence, social competence, personality traits, genital appraisal, and gender-role behavior were assessed. It is also noticeable that most studies exclusively examined medical risk factors (Table II). The influence of psychosocial factors was investigated by just two research groups (Mureau et al., 1995b; Mureau et al., 1997; Sandberg et al., 2001).

Psychosocial Adjustment

Self-reports

Results of studies and effect sizes are presented in Table II. Four studies focused on the examination of self-reported psychosocial adjustment. Mureau et al. (1997) did not report any differences in self-reported psychosocial adjustment between 116 boys with hypospadias

Table I. Summary of Studies Included in the Review

Study reference/Origin	Study design/n	Age of patients (range)	Comparison data	Severity	Age at 1st op./No. of op. (range)	Outcome measures	Method quality (0–10)
Psychosocial Adjustment							
Eray et al., 2005, Turkey	cs 40	7–17	None	Distal to proximal	<1–14.0 1–4	CDI ^a STAIC ^b	5
Glaser, 2004, Germany	cs 208	1–41	None	Glanular to perineal	Not reported 1–2	Self-developed questions ^c	3
Mondaini et al., 2002, Italy	cs 42	18–18	500 healthy men	Glanular to perineal	Not reported 0->2	MMPI ^d	6
Mureau et al., 1997, Netherlands	cs 116	9–18	88 boys after hernia repair	Distal to proximal	<1–12 1–8	DPQ-J ^e SAS-C ^f YSR ^g CBCL ^h	9
Purschke and Standke, 1993, Germany	cs 47	5–13	23 boys after various operations norms	Glanular to penoscrotal	2–12 0–9	PFK 9–14 ⁱ EF ^j self-developed scales ^c	4
Sandberg et al., 1989, USA	cs 69	6–10	Norms	Glanular to perineal	Not reported 1–6	CBCL ^h CBAQ ^k	7
Sandberg et al., 2001, USA	cs 175	6–10	333 healthy boys	Glanular to perineal	Not reported 1–13	CBCL ^h	7
Psychosexual development							
Glaser, 2004, Germany	cs 20	12–41	None	Glanular to perineal	Not reported 1–2	Self-developed questions ^c	2
Kumar and Harris, 1994, Great Britain	cs 35	13–25 ^l	None	Not reported	2–12 Not reported	Self-developed questions ^c	3
Mondaini et al., 2002, Italy	cs 42	18–18	500 healthy men	Supra- to subbalanic	Not reported 0->2	Self-developed scales and questions ^c	5
Mureau et al., 1995a, Netherlands	cs 116	9–18	88 boys after hernia repair	Distal to proximal	<1–12 1–8	Self-developed scales and questions ^c	7
Mureau et al., 1995b, Netherlands	cs 116	9–18	88 boys after hernia repair	Distal to proximal	<1–12 1–8	Self-developed scales ^m	8
Mureau et al., 1996, Netherlands	cs 35	9–18	None	Distal to proximal	Not reported 1–8	Self-developed scales ^c	5
Nelson et al., 2005, USA	cs 43	4–26	None	Proximal	Not reported 1–2	Self-developed scales ^c	2
Sandberg et al., 1989, USA	cs 69	6–10	Norms	Glanular to perineal	Not reported 1–6	CBAQ ^k CGPQ ⁿ	7
Sandberg et al., 1995, USA	cs 175	6–10	333 healthy boys norms	Glanular to perineal	Not reported 1–13	CBAQ ^k CGPQ ⁿ (revised)	7

^aChildren's Depression Inventory; ^bState-Trait Anxiety Inventory; ^cNo psychometric evaluation; ^dMinnesota Multiphasic Personality Inventory; ^eJunior Dutch Personality Questionnaire; ^fSocial Anxiety Scale for Children; ^gYouth Self-Report; ^hChild Behavior Checklist; ⁱPersönlichkeitsfragebogen für Kinder; ^jEncephalopathie Fragebogen;

^kChild Behavior and Attitude Questionnaire; ^lThis data is based on the original medical data base; not on the final sample of participants; ^mWith psychometric evaluation; ⁿChild Game Participation Questionnaire.

cs, cross-sectional; op., operation.

(9–18 years) and 88 age-matched healthy control subjects on several standardized measures (DPQ-J: Junior Dutch Personality Questionnaire, SAS-C: Social Anxiety Scale for Children, YSR: Youth Self-Report). However, calculation of ES showed small significant group effects that indicate hypospadias patients to be somewhat more socially competent and to have somewhat more external behavior problems compared to the controls. The study of

Mondaini et al. (2002) revealed no significant differences between 42 18-year-old youths with hypospadias and 500 healthy youths with regard to personality profile (MMPI: Minnesota Multiphasic Personality Inventory). Purschke and Standke (1993) evaluated the self-concepts of 47 children with hypospadias (5–13 years) by a German child personality questionnaire (PFK 914). Latter showed feelings of inferiority considerably more

Table II. Major Findings of Studies Included in the Review

Study reference	Outcome	Effect sizes (95%CI)	Tested associations	
Psychosocial adjustment				
Eray et al., 2005	Not examined	Incalculable (outcome not examined)	Age at final surgery: No association	
Glaser, 2004	<i>Self</i> ^a : = Psychosocial adjustment ^b <i>Self</i> ^a : + Emotional lability	Incalculable (no comparison data)	Not examined	
Mondaini et al., 2002	<i>Self</i> : = Personality Profile	0.24 ^c (-0.08-0.56)	Severity: No association No. of operations: No association Age at final surgery: No association	
Mureau et al., 1997	<i>Self</i> : = Social inadequacy <i>Self</i> : = Self-confidence <i>Self</i> : = Social anxiety <i>Self</i> : = Social competence <i>Self</i> : = Behavior problems External Internal Total	0.15 ^d (-0.12-0.43) 0.19 ^d (-0.09-0.47) 0.00 ^d (-0.28-0.28) 0.29 ^d (0.01-0.57) 0.29 ^d (0.01-0.57) 0.02 ^d (-0.26-0.30) 0.18 ^d (-0.10-0.46)	Severity: No association Surgical procedure: No association No. of operations: No association Age at final surgery: No association Age at follow-up: no association Genital perception: Pos. association with self-confidence neg. association with behavior problems Body image: Neg. association with social anxiety neg. association with behavior problems	
Purschke and Standke, 1993	<i>Proxy</i> : = Behavior problems External Internal Total	-0.07 ^d (-0.35-0.21) -0.08 ^d (-0.35 to -0.20) -0.02 ^d (-0.29-0.26)		
	<i>Self</i> : = Anxiety	-0.17 ^c (-0.67 to -0.33)	Severity: Pos. association with behavior problems	
	<i>Self</i> : = Aggressiveness	-0.38 ^c (-0.88 to -0.12)	no association with self-reported psychological adjustment	
	<i>Self</i> : = Social anxiety	0.20 ^c (-0.30-0.70)	No. of operations: Pos. association with behavior problems	
	<i>Self</i> : = Dependence on adults	0.13 ^c (-0.37-0.63)	neg. association with self-reported psychological adjustment	
Sandberg et al., 1989	<i>Self</i> : + Feelings of inferiority <i>Proxy</i> : + Behavior problems <i>Proxy</i> : + Behavior problems	0.98 ^c (0.46-1.51) Incalculable (missing information) Incalculable (missing information)	Age at 1st operation: Ideal age 5-6 years Surgical outcome: Inconsistent findings Severity: Partial positive associations with behavior problems	
	External Internal Total	0.21 ^d (-0.06-0.47) 0.29 ^d (0.03-0.55) 0.33 ^d (0.07-0.59)	Negative associations with academic achievement No association with social competency No. of hospitalizations: Inconsistent findings	
	<i>Proxy</i> : - Social competence <i>Proxy</i> : = Academic achievement	-0.29 ^d (-0.55 to -0.03) 0.10 ^d (-0.16-0.36)		
	Sandberg et al., 2001	<i>Proxy</i> : - Behavior problems External Internal Total	-0.36 ^d (-0.55 to -0.18) Incalculable (missing information) Incalculable (missing information)	Severity: No association No. of operations: Partial pos. associations with behavior problems No. of hospitalizations: Partial pos. associations with behavior problems Parents genital appraisal: Pos. association with academic achievement
		<i>Proxy</i> : - Social competence <i>Proxy</i> : = Academic achievement	0.00 ^d (-0.18 to -0.18) Incalculable (missing information)	

(continued)

Table II. Continued

Study reference	Outcome	Effect sizes (95%CI)	Tested associations
Psychosexual development			
Glaser, 2004	Self: = Masturbation frequency Self: = Intercourse-frequency	Incalculable (no comparison data)	Not examined
Kumar and Harris, 1994	Self: – Genital perception Self: – Experience of sex. intercourse	Incalculable (no comparison data)	Not examined
Mondaini et al., 2002	Self: + Sex. inhibitions	0.60 ^c (0.28–0.92)	Severity: Neg. association with genital appraisal
	Self: – Experience of sex. intercourse	–0.71 ^c (–1.03 to –0.39)	No. of operations: Most sexual inhibitions if not operated on
Mureau et al., 1995a	Self: – genital perception Self: = Age at 1st sex. Experiences ^c	1.57 ^c (1.24–1.90) –0.12 ^d (–0.40–0.16) – 0.36* (0.09–0.64)	Age at final surgery: No association Severity: No association with sexual inhibitions
	Self: = Sex. Behavior ^f	–0.09 ^d (–0.36–0.19) – 0.22 ^d (–0.06–0.49)	Surgical procedure: No association
	Self: + Sex. inhibitions Self: – Genital perception	1.53 ^c (1.17–1.89) Incalculable (missing information)	No. of operations: No association Age at final surgery: No association Age at follow-up: Neg. association with genital perception Neg. association with sexual inhibitions
Mureau et al., 1995b	Self: – Genital perception Self: = Body image	–0.52 ^d (–0.80 to –0.24) 0.19 ^d (–0.09–0.47)	Severity: No association No. of operations: No association Surgical procedure: No association Age at final surgery: No association Age at follow-up: Neg. association with body image Response from others: No association
Mureau et al., 1996	Self: – Genital perception ^g	–0.86 ^c (–1.35 to –0.37)	Penile length: No association
Nelson et al., 2005	Self: – Genital perception	Incalculable (no comparison data)	Not examined
Sandberg et al., 1989	Proxy: + Feminine behavior	6–8y.: 0.18 ^d (0.10–0.26) – 0.58 ^d (0.50–0.66) ^h	Severity: No association
		9–10y.: –0.24 ^d (–0.34 to –0.14) – 0.69 ^d (0.59–0.79) ^h	No. of hospitalizations: No association
	Proxy: = Masculine behavior	6–8y.: 0.21 ^d (0.13–0.29) – 0.37 ^d (0.29–0.45) ^h	
		9–10y.: –0.44 ^d (–0.54 to –0.34) – 0.58 ^d (0.48–0.68) ^h	
Sandberg et al., 1995	Proxy: = Feminine behavior	–0.26 ^d (–0.45–0.08) – 0.26 ^d (0.08–0.45) ^h	Severity: No association
	Proxy: + Masculine behavior	0.17 ^d (–0.02–0.35)	No. of hospitalizations: Pos. association with feminine behavior

– Indicates lower scores compared to controls/norms or indicating low scores if controls/norms are missing, + indicates higher scores compared to controls/norms or indicating high scores if controls/norms are missing, = indicates missing difference between cases and controls/norms or indicating normal scores if controls/norms are missing. Self, self-report measures; proxy, proxy-report measures.

^aIf patients were too young for the children's interview, Glaser assessed psychosocial adjustment by means of proxy-report, ^be.g. sleeping problems, anxiety, separation anxiety, ^cES was calculated according to Hasselblad and Hedges (1995) due to dichotomous outcome, ^dES was calculated according to Cohen (1988) due to continuous outcome, ^ee.g. age at first being in love, at first kiss, at first sexual intercourse, ^fe.g. sexual desire, sexual activity, sexual satisfaction, ^gcompared to surgeon's perception, ^hdependent on the specific subscale.

frequently but showed slightly less aggression than 23 boys of an age-matched control group who had received mild operations for various other medical problems. Yet, both hypospadias patients and the respective control group showed an equal level of (social) anxiety as well as dependence on adults. Likewise, Glaser (2004)¹ presented inconsistent findings: Whereas the 20 hypospadias patients showed a high level of emotional instability (unvalidated questions; ES not calculable), Glaser did not notice any further impairments of psychosocial adjustment (e.g., an increased level of separation anxiety).

The inconsistent results on self-reported psychosocial adjustment (range ESs: -0.038–0.98) might be a result of their different methodological standard. Studies with a higher standard (Mondaini et al., 2002; Mureau et al., 1997) suggested a better psychosocial adjustment. However, other differences between the studies that were related not only to their origins and the subjects' ages but also to the diversity of applied measures could also have caused the contradictory results.

Proxy-reports

Mostly, proxy-reported psychosocial adjustment was measured with the Child Behavior Checklist (CBCL) (Mureau et al., 1997; Sandberg et al., 1989, 2001). However, Sandberg et al. (1989) additionally used the Behavior Disturbance Scale of the Child Behavior and Attitude Questionnaire (CBAQ), while Purschke and Standke (1993) used the German "Encephalopathie-Fragebogen (EF)" as well as unvalidated questions (Table I). The results of proxy-reports are also inconsistent (Table II). The study of Mureau et al. (1997) showed that the parents of 116 boys with hypospadias did not report their children's behavior to be more problematic compared to the parents of 88 age-matched controls, treated for an inguinal hernia. Conversely, the findings of Purschke and Standke (1993) suggest that the 47 children with hypospadias (5–13 years) had more behavior problems than the 23 children of the same age, who had been subjected to a variety of mild operations for other medical problems (ES not calculable). In 1989, an American research group (Sandberg et al.) found more behavior problems and lower social competence in 69 children with hypospadias (6–10 years) compared to the corresponding norms of 300 healthy children between the ages of 4 and 16 years. Calculated ES were significant but small. Twelve years later, the same researchers (Sandberg

et al., 2001) published a study with a slightly better methodological standard that involved a larger sample of hypospadias patients ($N = 175$) as well as a control group of healthy school boys ($N = 333$). Although the calculated ES of mean difference suggests no group effect, multiple regression,² controlling for sociodemographic characteristics, showed that the parents of hypospadias patients (6–10 years) considered their children to be less socially competent than the parents of the comparison subjects (6–10 years). Yet, in contrast to their earlier results, boys with hypospadias were judged by their parents to have fewer externalizing behavior problems compared to the parents of the controls (ES small to moderate).

Regarding the inconsistencies of proxy-reports on psychosocial adjustment, it is important to take the studies' methodological quality into account. Comparable with the findings of self-reports, studies with a methodologically higher qualified standard (Mureau et al., 1997; Sandberg et al., 2001³) suggested a somewhat better psychosocial adjustment. Calculated ES', however, tended to be small and varied less (range: -0.29 to 0.33) than ES's for self-reports. Furthermore, there are other differences between the studies (e.g., age range of subjects) that might be responsible for the contradictory findings.

Risk Factors for Psychosocial Maladjustment

Medical factors

Except for Glaser (2004), all studies on psychosocial adjustment have focused on the association with medical risk factors (Table II). The results indicate that the association between psychosocial adjustment and medical factors is weak. According to Mureau et al. (1997) and Mondaini et al. (2002), psychosocial adjustment (measured with several standardized outcome measures) and personality profile (MMPI) were not related to the severity of hypospadias, the number of operations, and the age at final surgery. Neither did the type of surgical procedure correlate with psychosocial adjustment (Mureau et al., 1997). Moreover, Mondaini et al. (2002) did not detect any difference in personality profile between treated and untreated patients. Additionally, a recent study by

²Effect sizes for multivariate analyses could not be calculated due to missing information.

³Sandberg et al. (2001) did not show a higher score in our methodological assessment than Sandberg et al. (1989), but the former involved a larger sample as well as a healthy control group and used revised measures for the assessment of gender-role behavior, which showed more favorable psychometric criteria. However, we did not consider this fact in our methodological rating due to practical reasons.

¹If patients were too young to attend the children's interview, Glaser assessed psychosocial adjustment by means of proxy-report.

Eray et al. (2005) showed that patients whose corrective surgery was completed before the age of 18 months did not differ in their level of depression and anxiety from patients who had their final operation at a later age. However, the findings of Purschke and Standke (1993) and Sandberg et al. (1989, 2001) did not coincide with those of the above mentioned authors. Namely, Purschke and Standke reported that patients with a more severe form of hypospadias and who were surgically treated more often showed a higher incidence of behavior problems. In addition, the number of operations was negatively associated with the patients' self-concept. However, the eight untreated patients displayed the lowest psychosocial adjustment. The quality of surgical outcome correlated negatively with behavior problems but positively with the patient's psychological strain. Furthermore, children operated on between the ages of 5 and 6 showed fewer behavior problems than children operated on either between the ages of 2 and 4 or between 6 and 12. The results of the two studies of Sandberg et al. (1989, 2001) are, however, inconsistent. While the first study suggested that children with a more severe form of hypospadias showed more behavior problems and better school achievement (CBCL), the second investigation did not confirm this assumption. However, in 2001 the researchers detected a positive association between the patients' behavior problems and the number of operations as well as the number of hospitalizations the children had been subjected to.

Psychosocial Factors

According to Mureau et al. (1997), boys who were more satisfied with the appearance of their genitals and their overall body image had fewer behavior problems, were more self-confident and showed less social anxiety. In addition, the subjects' age was not found to be associated with psychosocial adjustment. Sandberg et al. (2001) reported that the parents' satisfaction with the appearance of the patients' genitals were positively correlated with their academic achievement but not so with their behavior problems.

Psychosexual Development

Self-reports

Seven articles focus on at least one aspect of self-reported psychosexual development (Glaser, 2004; Kumar & Harris, 1994; Mondaini et al., 2002; Mureau et al., 1995a,b; Mureau, Slijper, Slob, Verhulst, & Nijman, 1996; Nelson et al., 2005), three of which are based on the same investigation (Mureau et al.) (Table II). The most frequently examined construct is genital perception

that has been assessed either as the satisfaction with genital appearance or as the appraisal of one's genitals to look normal. Apart from Mureau et al. (1995b), all authors used nonvalidated self-constructed measures. Authors reported between 20 and 80% of the boys with hypospadias suffered from negative genital perception, while healthy age-matched controls were significantly more satisfied with penile appearance (Kumar & Harris, 1994; Mondaini et al., 2002; Mureau et al., 1995a,b). ESs were moderate (Mondaini et al., 2002) to large (Mureau et al., 1995b.) (Table II). Interestingly, Mureau et al. (1996) did not detect any relation between the patients' and the surgeon's perception of genital appearance. Overall, the latter was more satisfied, indicated by a large ES.

With regard to sexual inhibitions, both Mondaini et al. (2002) and Mureau et al. (1995a) found that children and adolescents with hypospadias reported more sexual inhibitions than the healthy age-matched controls (moderate to large ES's). They were not only more afraid of being rejected by a potential partner due to genital appearance but also prone to hide genitals in public lavatories (Mondaini et al., 2002; Mureau et al., 1995a). However, the available studies do not clarify whether youths with hypospadias also show a delay of first sexual experiences. In the methodologically most sound study (Mureau et al., 1995a), boys with hypospadias (9–18 years) had their first sexual experiences (e.g., first kiss) at slightly higher ages than the age-matched controls treated for an inguinal hernia, except for masturbating. Yet, ES's were only small (Table II). In the study of Mondaini et al. (2002) however, the calculated ES indicates that adolescents with hypospadias have their first sexual intercourse at a considerably higher age than healthy boys. Kumar and Harris (1994) reported the same, although they did not include a control group. However, results indicate that adolescents with hypospadias do not differ from healthy youths neither with regard to sexual activity, number of coitus partners, and sexual desire, nor with regard to body image (Glaser, 2004; Mureau et al., 1995a,b).

Proxy-reports

Sandberg et al. (1989, 1995) examined proxy-reported gender-role behavior measured by the gender-role behavior scales of the CBAQ and by the Child Game Participation Questionnaires (CGPQ) (Table I). Their findings were, however, contradictory (Table II). In 1989, consistent with a hormonal hypothesis of gender-role development, they observed more feminine behavior in 6–8 year old boys with hypospadias (6–10 years) than in a reference group of healthy male children. Calculated ES are small to moderate, depending on the specific

subscale. Yet, 6–8 year old children with hypospadias showed also slightly higher scores with regard to masculine behavior patterns, indicated by the small significant ES. Even more inconsistent were the findings with regard to the 9–10 year old patients, with ES ranging from negative in the moderate realm to positive in the large realm. Nevertheless, the authors reported that half of the boys even showed similar levels of feminine behavior to a reference group of 23 gender-dysphoric children (effect size not calculable due to missing information). Six years later, they published a second study that involved revised versions of the CBAQ and CGPQ. Although calculated ES indicate somewhat inconsistent findings, they suggest that the behavior patterns of 175 children with hypospadias did not considerably differ from that of the 333 boys of a healthy-control group. Moreover, multivariate analyses showed that sociodemographic control variables were stronger predictors of gender-role behavior than genital status.

Risk Factors for Psychosexual Development

Medical factors

The relationship between medical characteristics and the psychosexual development of boys with hypospadias was examined by three research groups (Mondaini et al., 2002; Mureau et al., 1995a,b; Sandberg et al., 1989, 1995). Overall, the findings imply that the association between medical characteristics and psychosexual development is rather weak (Table II). However, Mondaini et al. (2002) reported a negative effect of severity of hypospadias on genital appraisal (nonvalidated), Mureau et al., 1995a found a positive effect of severity on sexual inhibitions (nonvalidated) and Sandberg et al. (2001) detected a positive effect of number of hospitalizations on feminine behavior patterns (CBAQ, CGPQ). Furthermore, Mondaini et al. (2002) reported that untreated adolescents with hypospadias had more sexual inhibitions (nonvalidated) than treated boys. However, no association was found between psychosexual development and number of operations, age at final surgery as well as type of surgical procedure.

Psychosocial factors

Mureau et al. (1995a) reported that the boys' age was negatively related to genital perception and positively associated with sexual inhibitions. Conversely, another study by Mureau et al. (1995b) showed that age was negatively related to body image but not to genital perception. This inconsistency between the studies is assumed to be a result of the divergent assessments of genital perception. In addition, the experience of having

been teased due to genital appearance was not linked to genital perception.

Discussion

We conducted the first systematic review of studies on psychosocial adjustment, psychosexual development and HRQoL of children, and adolescents with hypospadias. Furthermore, we aimed to assess psychosocial and medical risk factors of the outcome as well as the psychological significance of corrective surgery and its cosmetic result.

Thirteen studies met the criteria for inclusion, whose methodological standard ranged from low to high quality. All studies focused on either psychosocial adjustment or psychosexual development. With regard to psychosocial adjustment, the results of the studies are inconsistent, with recent and methodologically more sound studies indicating a slightly more favorable outcome (Mondaini et al., 2002; Mureau et al., 1997; Sandberg et al., 2001). The inconsistent results may be a consequence of the variation in methodological quality. On the other hand, surgical treatment strategies have evolved over the last decades: Patients are being operated at a younger age, surgical techniques, and anesthesia have been improved and nursing practice has adapted to children's needs as for example by the rooming-in of parents (Bracka, 1999). These varying standards of treatment make direct comparisons of results difficult and may be responsible for a better psychosocial adjustment in recent studies. Yet, it is likely that the differences between the studies with regard to origin, age range of subjects, and operationalization of psychosocial adjustment contributed to contradictory findings.

The findings regarding psychosexual development, however, clearly demonstrate that boys with hypospadias suffer from negative genital appraisal and sexual inhibitions. Nevertheless, age at first sexual experiences and general sexual behavior appeared to be less affected by their penile malformation (Kumar & Harris, 1994; Mondaini et al., 2002; Mureau et al., 1995a; Mureau et al., 1995b; Nelson et al., 2005). The effects of hypospadias on gender-role behavior, however, are not clarified yet (Sandberg et al., 1989, 2001). Thus, the hypothesis that prenatal hypoandrogenization associated with hypospadias interferes with the development of gender-typical masculine behavior remains speculative and thus requires further examination.

The findings on the significance of medical factors are likewise inconsistent. Overall, they suggest that

medical characteristics bear a rather small influence on the psychosocial and psychosexual outcome. It is important to note that there is no empirical evidence that corrective surgery at the youngest possible age leads to a better psychological development. Thus, empirical results do not support the early surgical interventions, which pediatric urologist recommend for (APA, 1996; Zavitsanakis & Gougoudi, 2004).

Evidence with regard to psychosocial predictors of adjustment is scarce. The results of Mureau et al. (1997) suggest that positive genital perception and a healthy body image are crucial for the boys' psychosocial adjustment. However, it is also possible that psychosocially better adjusted children develop a more favorable genital and body perception. In addition, there is some evidence that genital appraisal of the patients' parents is connected to the patients' adjustment (Sandberg et al., 2001), which indicates that parental attitude towards hypospadias might be important for the patients' well-being. However, the findings with regard to the significance of the children's current age are contradictory (Mureau et al., 1995a,b; Mureau et al., 1997). Yet, it would be plausible if adolescents were less satisfied with penile appearance than younger boys due to a change in physical self-perception as well as the increasing importance of sexuality during puberty.

Finally, no conclusion can be drawn with regard to the importance of corrective surgery itself for the children's psychosocial and psychosexual development. Although Purschke and Standke (1993) and Mondaini et al. (2002) reported some differences in psychosocial and psychosexual adjustment between untreated and treated boys, the examined groups of untreated patients were much too small ($n=6$ and $n=8$) to generalize results.

Limitations and Directions for Future Research

There are several factors that limit the generalization of the findings. First, many studies lack methodological quality due to small sample sizes, the absence of control groups or norms and the use of nonvalidated measures, particularly with respect to psychosexual outcome. Second, the number of existing studies is too small to draw general conclusions. Ultimately, it is assumed that modern management of hypospadias is much less stressful than it used to be (Bracka, 1999): Children with hypospadias are currently operated at a younger age with more sophisticated surgical techniques which are bound to generate a better surgical outcome.

Other limitations relate to research questions, which have yet to be investigated. Surprisingly, there exists no study on HRQoL of children with hypospadias. HRQoL measurement has emerged as an important tool for analyzing health outcome in clinical trials (Eiser & Jenney, 2007). It is a multidimensional construct defined as the patient's perception of the effects of illness/disability and treatment on his physical, psychological, and social functions. It is thus not solely confined to physiological outcome and/or psychopathological symptoms (Matza, Swensen, Flood, Secnik, & Leidy, 2004). Both generic and disease-specific HRQoL-measures have been developed. While the former allows comparisons across patient groups as well as with healthy children, the latter appear to be more sensitive to the implications of a single condition (Eiser & Jenney, 2007). Furthermore, it is recommended to make both self- and proxy-ratings when measuring HRQoL: Both the child's and the parents's perspective should be assessed (Matza et al., 2004). One tool that provides both a patient's and a parent's form as well as the combination of a generic measure with disease-specific modules is the Pediatric Quality of Life Questionnaire (PedsQL) (Varni, Seid, & Rode, 1999). However, the PedsQL does not comprise a hypospadias-specific module, which would allow a more detailed assessment of HRQoL in children with hypospadias. Unfortunately, to the best of our knowledge a HRQoL measurement that is sensitive to the specific challenges of hypospadias does not exist and has yet to be developed. Ideally, it would assess the child's perception of his physical (e.g., micturition and sexual function), psychological (e.g., depressive emotions, senses of shame, and body image), and social (e.g., social withdrawal, sexual anxieties, being teased by peers, and concealment of hypospadias) functions.

Likewise, there is a lack of knowledge about the influence of psychosocial factors on the patients' development. It is presumable that family background variables (e.g., the quality of family relationships), the patients' coping behavior as well as the attitude of patients' parents towards their children's penile condition may have a greater impact on the boys' psychological development than medical characteristics. Another direction for future research would be an extended examination of the psychosexual development. The majority of existing studies examined only a certain aspect of psychosexual development. However, the construct of psychosexual development includes many components such as the development of gender identity, gender-role behavior, body inquiry, and the development of sexual feelings (Schuhrke, 1997;

von Sydow, 1993). Thus, it is important to note that the psychosexual development is not exclusively related to first sexual experiences or to sexual behavior habits. Finally, it would be interesting to shed light on the psychological development of untreated children with hypospadias, in order to be able to investigate the psychological consequences of hypospadias repair. This aspect should be raised since, on the one hand, self-help groups have recently announced the need for a reduction in the frequency of genital surgery due to the potential of emotional trauma (ISNA, 2006; M. Walker, personal communication, December 13, 2006) and, on the other hand, pediatric urologists plead for surgical correction of hypospadias in order to prevent negative repercussions on the patients' psychosexual adjustment (AAP, 1996; Zavitsanakis & Gougoudi, 2004).

A final limitation refers to the methodical procedures by which we have conducted the present systematic review. Although we have applied systematic review techniques to select relevant articles and have provided a reliable rating of methodological quality, the chosen sample of studies did not allow the conduction of systematic meta-analyses. Consequently, a sophisticated examination of trends in data, their relations to methodological quality, and specific outcome measures as well as analyses of potential differences between self- and proxy-report could not be performed. Nevertheless, by calculating effect sizes we tried to reduce subjectivity in the narrative presentation of the results.

Clinical Implications

The inconsistent results and the lack of knowledge about the influence of psychosocial factors on the psychological development of boys with hypospadias allow only limited implications for clinical practice. Based on the findings that boys with hypospadias tend to suffer from negative genital appraisal more often than healthy boys, one may assume that they might profit from psychosocial support for the development of sufficient self-confidence which would possibly encourage them to better accept their penis. Surgeons must be aware that genital appraisal does not exclusively rely on the quality of surgical outcome (Mureau et al., 1996). Thus, optimal medical care for hypospadias patients cannot merely be achieved by optimizing surgical results. In order to guarantee a comprehensive treatment concept, the incorporation of psychological factors such as the anxiety of rejection by eligible sex partners may be equally important. Moreover, it is tenable that genital appraisal and sexual inhibitions may be affected to a higher degree during adolescence

than during childhood. Hence, it is believed to be essential, as already Berg, Svensson, and Aström (1981) argued 26 years ago, that patients with hypospadias will need to be followed-up until young adulthood. Apart from regular urological medical examinations, the patients should be appropriately informed about their penile condition and to be offered psychological support.

In summary, the present review demonstrated that there is a lack of evidence regarding psychosocial adjustment, psychosexual development and HRQoL of children, and adolescents with hypospadias. Furthermore, the review pointed out that the guidelines for surgical treatment are partly based on psychological assertions that have not been empirically confirmed. Particular attention should be paid to the assumption that a surgical correction of hypospadias at the earliest age possible leads to a minimization of adverse effects on the boy's development. From a psychosocial point of view, an optimization of the treatment of boys with hypospadias ultimately requires a clarification of the patient's psychosocial and psychosexual development in methodologically sound investigations.

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References marked with one asterisk indicate studies included in the Systematic Review, references marked with two asterisks indicate studies excluded during final study selection.