Reconstructive surgery for hypospadias: A systematic review of long-term patient satisfaction with cosmetic outcomes

Julie Adams, Aivar Bracka¹

Department of Applied Health, Faculty of Health, Edge Hill University, Ormskirk, Lancashire, 'Formerly at the Dudley Group of Hospitals, West Midlands, UK

ABSTRACT

Introduction: Research on long-term results of hypospadias has focused on surgical techniques and functional outcomes, and it is only recently that patient satisfaction with appearance and psychosocial outcomes have been considered. The aim of this study was to provide an evidence-based systematic review of adolescent and adult patient perceptions of cosmetic outcomes following childhood surgery for hypospadias.

Methods: A systematic review was performed in accordance with the PRISMA and PICO guidelines, and studies assessed using the Oxford Centre for Evidence-Based Medicine system. MEDLINE, PsycInfo, EMBASE, and CINAHL databases were searched from 1974 to 2014 for clinical studies containing patient perceptions of appearance, deformity, and social embarrassment following hypospadias surgery.

Results: A total of 495 publications were retrieved, of which 28 met the inclusion criteria. Due to study design/outcome measure, heterogeneity data were synthesized narratively. Results indicate (i) patient perceptions of penile size do not differ greatly from the norm; (ii) perceptions of appearance findings are inconsistent, partially due to improving surgical techniques; (iii) patients who are approaching, or have reached, sexual maturity hold more negative perceptions and are more critical about the cosmetic outcomes of surgery than their prepubertal counterparts; (iv) patients report high levels of perceptions of deformity and social embarrassment; and (v) there is a lack of data using validated measurement tools assessing long-term patient perceptions of cosmetic outcomes, particularly with patients who have reached genital maturity. **Conclusions:** Protocols for clinical postpuberty follow-up and methodologically sound studies, using validated assessment tools, are required for the accurate assessment of cosmetic and psychological outcomes of hypospadias surgery.

Key words: Cosmetic, genital image, genital surgery, hypospadias, long-term outcome, penis size, psychosocial, reconstructive surgery

INTRODUCTION

Hypospadias is a relatively common male genital deformity affecting about 0.3–0.4% of the population worldwide and its incidence is increasing.^[1,2] In this condition, the urethral opening is sited anywhere

For correspondence: Dr. Julie Adams,

Faculty of Health, Edge Hill University, Ormskirk,

Lancashire, L39 4QP, UK.

E-mail: guppy.j@st-annes.oxon.org

Access this article online

Quick Response Code:

Website:

www.indianjurol.com

DOI:

10.4103/0970-1591.179178

along the underside of the penis from the glans to the perineum. The aim of hypospadias repair, for which over 200 different techniques are described, [3] is to create a functional neourethra, correct any curvature, and produce a cosmetically normal penile appearance, with a slit-like meatus at the tip of the penis. [4,5] Surgical techniques have improved dramatically over the last 30 years or so, with the more modern terminalizing techniques providing successful function and an appearance which more closely matches

This is an open access article distributed under the terms of the Creative Commons Attribution-NonCommercial-ShareAlike 3.0 License, which allows others to remix, tweak, and build upon the work non-commercially, as long as the author is credited and the new creations are licensed under the identical terms.

For reprints contact: reprints@medknow.com

How to cite this article: Adams J, Bracka A. Reconstructive surgery for hypospadias: A systematic review of long-term patient satisfaction with cosmetic outcomes. Indian J Urol 2016;32:93-102.

the normal population. An average of 1623 hypospadias operations was performed in the UK during 2006–2007. [6]

Most research on the outcomes of hypospadias surgery has focused primarily on surgical techniques and functional outcomes. It is only in the last 20 years or so that the importance of the patient's perceptions of cosmetic penile appearance, and the possible psychological outcomes have begun to be considered. This study systematically reviews the data relating to constructs of patient perceptions of cosmesis following corrective surgery for hypospadias, with implications for pre- and post-surgery management and protocols for follow-up.

The aims of this systematic review were to (i) synthesize peer-reviewed studies that provided quantitative outcomes for patient perceptions of cosmesis and appearance, perceptions of deformity and social embarrassment; (ii) make comparisons between patients and controls, or those from nonclinical populations; and (iii) provide recommendations for pre- and post-surgery management and identify implications for protocols for clinical follow-up.

METHODS

A systematic review was conducted in accordance with PRISMA guidelines^[7] using MEDLINE, PsycInfo, EMBASE, and CINAHL from 1974 to November 2014. In selecting articles for inclusion in this review, the inclusion criteria outlined below were applied.

For a study to be included, it needs to:

- Include male participants who had been operated on for hypospadias in childhood
- Contain patients who had a solitary urological diagnosis of hypospadias
- Report patient perceptions of penile size, appearance, deformity/normality, satisfaction with cosmetic outcome, or measures of social embarrassment following childhood hypospadias repair
- Provide sufficient detail on the above outcome measures to allow comparison across studies
- Report primary data on patient perceptions
- Report quantitative data
- Be written in English language
- Be published in full-text and in a peer-reviewed journal.

Studies were also excluded if they only reported qualitative data, assessed surgical outcome or objective sexual function only, focused on other urological anomalies, reported a single patient case study, or were review articles. All degrees of hypospadias severity, all methods of repair, and however many operations, were included. Studies which did not have a control group (n = 12) were included. The quality of all studies was scored according to the Oxford Centre for Evidence-Based Medicine system.^[8]

All included articles were assessed for quality regarding methodological strength as per the 2009 Cochrane collaboration updated guidelines for systematic reviews.[9] Quality ratings for each study are presented in the final column of Table 1. Timmer et al.'s[10] checklist for quantitative studies, which has been demonstrated to have good construct validity, was employed and adapted for this review (three items pertaining to intervention studies were removed).[11] A total of 16 items were utilized and as per the guidelines, two points were allocated if the item was fully met, one if partially met, and none if not met at all. Up to five extra points were awarded based on the study design. A ratio score between 0 and 1 was computed, with 1 representing the highest quality score and zero the lowest. The quality of articles was assessed by two reviewers and in case of any disagreement, consensus was achieved by discussion. Articles using a duplicate patient sample were included if relevant different outcome measures were addressed. The following search terms were employed: "Hypospadias AND (perception OR psychological OR cosmesis OR (body AND image) OR psychosocial OR psychosexual OR social)." The data selection process is illustrated in Figure 1. One author performed the searches and two authors screened retrieved articles, extracted data, and summarized them to include PICOS variables (participants, interventions, comparators, outcomes, and study design). These variables and levels of evidence provide data to assess the risk of bias in individual studies. The main outcome measures were patient perceptions of penile appearance and satisfaction, perceptions of deformity, and social embarrassment.

RESULTS

Study selection and data analysis

Twenty-eight studies reported on 1699 patients were included in the systematic review and summarized in Table 1. Meta-analyses were not possible because of study heterogeneity; therefore, data analysis was undertaken by means of a thematic content analysis whereby a systematic classification process of coding for themes enabled a subjective interpretation of the data. [12] Each paper was read by two reviewers to identify specific themes related to the systematic review study objectives, following which these were organized into categories to encompass similar themes (i.e., a thematic content analysis). The themes relating to long-term patient satisfaction with cosmetic outcomes following surgery for hypospadias were independently identified by two reviewers with agreement across each of the four themes. The data synthesis process produced the following four themes: Perception of penis size, satisfaction with appearance and cosmetic outcomes, perceptions of deformity, and social embarrassment. In light of the absence of control group data in some hypospadias studies, a brief review of male penile perceptions in nonclinical populations is presented as a source of background reference.

Author (s) and date	Country	Sample size/age range	Control group	Level of evidence	Severity	Age at first operation	Validated patient	Quality
Aho <i>et al</i> . ^[37]	Finland	46/29.5 years Postpubertal	43 age-matched	III	Distal-proximal	Mean 3.6 years	rating scale	0.80
Aulagne <i>et al</i> . ^[52]	France	27/20-32 years Postpubertal	/	IV	Proximal-scrotal	Mean 3.3 years	/	0.81
Bracka ^[54]	UK	213/15-24 years Postpubertal	/	IV	Distal-proximal	Mean 5 years	/	0.79
Chertin <i>et al.</i> ^[43]	Israel	119/18+ years Postpubertal	/	IV	Glanular-proximal	Mean 2.7 years	SEAR	0.86
Frauman <i>et al</i> .[35]	USA	13/18-30 years Postpubertal	/	IV	Midshaft-proximal	Mean 14.5 months	SEAR	0.79
Hoag et al.[45]	Canada	28/16-31 years Postpubertal	/	IV	Distal-proximal	Mean 1.95 years	/	0.75
Jiao <i>et al</i> . ^[32]	China	43/21.6 years mean Postpubertal	/	IV	Distal-proximal	Mean 5.7 years	/	0.69
Jones <i>et al</i> . ^[31]	Australia	55/13-15 years Postpubertal	/	IV	Distal-proximal	Mean 1.96 years	PedsQL	0.78
Kiss <i>et al</i> . ^[39] / Merksz <i>et al</i> . ^[40]	Hungary	104/22-42 years Postpubertal	63 age-matched	III	Midshaft-proximal	4-7 years	/	0.88
Kumar and Harris ^[25]	UK	35/13-25 years Postpubertal	/	IV	Midshaft-proximal	2.3-12 years	/	0.75
Lam <i>et al</i> . ^[48]	USA	27/13-21 years Postpubertal	/	IV	Midshaft-proximal	Mean 1.7 years	/	0.87
Mondaini <i>et al</i> . ^[38]	Italy	42/18 years Postpubertal	500 age-matched	III	Distal-proximal	Not reported	MMPI	0.85
Mor <i>et al</i> . ^[50]	Israel	43/16-31 years Postpubertal	/	IV	Distal	Mean 1.7 years	/	0.79
Moriya <i>et al</i> . ^[30]	Japan	22/18-26 years Postpubertal	38 age-matched	III	Distal-proximal	Mean 3.8 years	/	0.86
Mureau <i>et al</i> . ^[27]	The Netherlands	116/9-18 years Pre- and post-pubertal	88 (mean 13.9 years)	III	Distal-proximal	Mean 4 years/ 0.3-12.3 years	/	0.91
Mureau <i>et al</i> . ^[26]	The Netherlands	116/9-18 years Pre- and post-pubertal	88 age-matched	Ш	Distal-proximal	Mean 4 years	GPS and BPS	0.91
		73/18-38 years Postpubertal	50 age-matched	Ш	Distal-proximal			
Mureau <i>et al</i> . [36]		73/18-38 years Postpubertal	50 age-matched	Ш	Distal-proximal	/	/	0.86
Mureau <i>et al</i> . ^[49]		35/9-18 years Pre- and post-pubertal	/	IV	Distal-proximal	/	GPS	0.82
Nelson <i>et al</i> . [44]	The USA	10/18+ years Postpubertal	/	IV	Severe	/	/	0.71
Ortqvist <i>et al</i> . ^[42]	Sweden	167/19-54 years Postpubertal	169 age-matched 47 (mean 24 years)	III	Distal-proximal	Median 4 years	PPS	0.82
Ruppen-Greeff et al. [41]	Swiss	45/18-41 years Postpubertal	46 age-matched	Ш	Distal-proximal	Median 4.17 years	PPS	0.88
Rynja <i>et al</i> . ^[29]	Iran	66/mean 22 years Postpubertal	151 students	III	Anterior-posterior	2.3 years	PPPS	0.77
Schonbucher et al.[46]	Switzerland	68/7-17 years Pre- and post-pubertal	68 age-matched	III	Glanular-penoscrotal	Median 3.18 years	PPPS	0.83
Vandendriessche et al.[51]	Belgium	10/11-18 years Pre- and post-pubertal	10 age-matched	III	/	<12 years	JGPS and SPPA	0.88
Wang et al.[33]	China	130/unavailable Postpubertal	50 (24-35 years)	III	Distal-proximal	<10 years, 10-18 years, 18 years	ZSRAS and ZSRDS	0.91

Contd...

Table 1: Contd											
Author (s) and date	Country	Sample size/age range	Control group	Level of evidence	Severity	Age at first operation	Validated patient rating scale	Quality score			
Weber et al.[22]	Switzerland	77/6-17 years Pre- and post-pubertal	77 age-matched	III	Distal-penoscrotal	Mean 3.01 years	PPPS	0.88			
Weber et al.[23]	Switzerland	19/19-39 years Postpubertal	3 (22-28 years)	III	Distal-proximal	Mean 32.5 months/ 0-82 years	PPS	0.88			
Wouters et al.[28]		66/18+ years Postpubertal	151 age-matched	III	Distal-proximal	/	/	0.80			

BPS=Body Perception Score, GPS=Genitalia Perception Score, JGPS=Junior Genital Perception Scale, MMPI=Minnesota Multiphasic Personality Inventory, PedsQL=Pediatric Quality of Life Inventory, PPPS=Pediatric Penile Perception Score, PPS=Penile Perception Score, SEAR=Self-Esteem and Relationship Questionnaire, SPPA=Self-Perception Profile for Adolescents, ZSRAS=Zung Self-Rating Anxiety Scale, ZSRDS=Zung Self-Rating Depression Scale

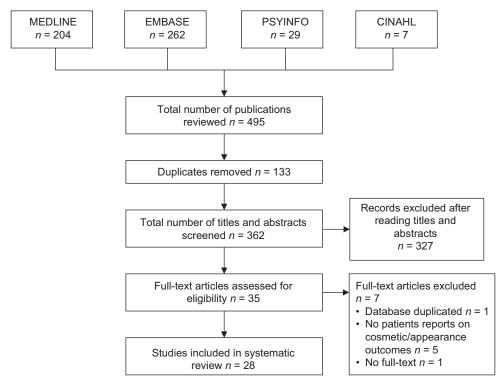


Figure 1: Flow diagram of search and selection strategy of included articles

Penile perceptions in nonclinical population

Research on penile perceptions within nonclinical groups suggests that a significant proportion perceive their penis to be below average size (7–12%),^[13,14] 30% are dissatisfied with the size of their penis,^[15] a very large percentage state that they would like a larger penis (45–60%),^[13,15-18] 61% identify their penis as an area of concern for them,^[17] and 17% report embarrassment in relation to their genitals.^[18] Further, men who perceived their penis to be small were less likely to undress in front of their partners (15%) and were more likely to hide their penis during sex (15%).^[13] While higher levels of concern regarding penis size have been reported by heterosexual men^[15,17-19] than by homosexual men,^[14,16] there is a trend toward men's perception of the ideal size exceeding their own and that of the norm in both populations.^[14,16,19] These figures provide a useful background against which to

compare the penis-related perceptions of adolescents and adults who have undergone hypospadias repairs.

Validated assessment instruments

While several validated objective scoring systems exist for use with hypospadias patients, [20,21] there are few that focus on the subjective penile perceptions of either hypospadias patients [22,23] or men from nonclinical populations. [15,18]

Cosmetic satisfaction of hypospadias patients

Table 1 presents a summary of the main studies included in the following sections of this systematic review.

Does size matter?

Early research by Bracka^[24] in the UK with 213 postpubertal (15–24 years) patients showed that over one-third felt

"inadequately endowed." In contrast, Kumar and Harris' [25] UK pilot study of 27 postpubertal, 13–25 years old found that only 7.4% reporting perceptions that their penis was too small. Small sample sizes may account for the lower results by Kumar and Harris.

A series of studies by Mureau et al.[26,27] revealed that postpubertal patients (both adults and adolescents) were less satisfied with their flaccid penile size than a control group and that significantly more proximal adult hypospadias patients felt that their penis was too small^[27] (a finding echoed by others).[28,29] Of the 37% of adult patients who wished for future surgery to improve cosmesis or function, 22% of these cited small penis size as the reason. [27] While 11% of adult patients cited a smaller penis size as a reason for dissatisfaction with their penile appearance, so they did 8% of the control group. This figure sits well with earlier research on both hypospadias and nonclinical samples.[13,14,25,30] In fact, Moriya et al.[30] found that 40.9% of patients and 69% of controls who reported dissatisfaction with penile appearance quoted penis size to be the sole cause of their dissatisfaction. Perceptions about penile size are a common explanatory factor for dissatisfaction with penile appearance in both adolescents and adult patients.[30-32] Objective measurements have found that significant differences exist between adult hypospadias and control groups in terms of penile size^[29,33] and that penis length is correlated with hypospadias severity.^[24]

The above studies suggest that perceived penis size is an explanatory factor for dissatisfaction with penile appearance, and there is some evidence to suggest that objectively the penis of hypospadias patients may be smaller than the norm. However, the available research does not point to a huge variation between adult hypospadias and nonclinical adults' perceptions of their penis being too small or below average. These findings may well be explained by the fact that the vast majority of the studies does not distinguish between the different degrees of severity of hypospadias. The incidence of distal hypospadias far outnumbers that of proximal hypospadias. The objective penile length is shorter with proximal hypospadias, [24] and this is reflected in proximal hypospadias patients' dissatisfaction with size. [26-29]

Cosmetic outcomes and satisfaction with appearance

Most surgeons now regard cosmetic outcome and patient satisfaction with postoperative appearance to be just as important as functional outcome. However, many long-term studies report on the outcome of outdated surgical procedures that cannot produce a normal looking penis. While modern procedures can often achieve something close to normal function and appearance, these have not as yet formed the subject of many long-term outcome studies. Body image is also now of far more importance to men than it was a couple of decades ago. The standard of hypospadias repair that was deemed acceptable to the male patient 20 years ago would

be considered unacceptable to most young patients and surgeons today. In addition, many studies have not focused on the effect of age when considering subjective perceptions of cosmetic outcomes. Age is an important consideration because older, married patients may be less concerned with esthetics whereas younger, sexually inexperienced men are more likely to have concerns over minor cosmetic imperfections that inhibit their willingness to engage in sexual relationships.

Historically, the level of importance placed on the esthetics of a repair has been influenced by the medical specialism of the operating surgeon. Plastic surgeons by their very nature are usually inclined to focus on esthetic aspects of surgery. Adult urologists are traditionally more function orientated while pediatric surgeons have usually signed their patients off in childhood, long before they reach the age when they are at their most concerned with genital appearance. In Springer's study, nearly 60% of pediatric urologists/surgeons reported utilizing a follow-up period of <6 months, with only 10% following-up patients until and beyond puberty. Indeed, Fraumann *et al.* have recently observed that there has been a little research that has focused on cosmetic and functional outcomes after patients have gone through puberty, or indeed, in sexually active adults.

Several European and East Asian studies comparing postpubertal adolescents/adult hypospadias patients with controls have found significant differences with respect to satisfaction with penile perceptions. [26,30,33,36-42] The ranges of results include - adolescent and adult patients were significantly less satisfied with their penile appearance than controls, including flaccid penis size, glans shape, position of the meatus, and general penile appearance as measured by a standardized genital perception score questionnaire; [26] prepubertal and adult patients who were satisfied with their circumcision status had higher genital perception scores;^[26] significantly more patients have reported dissatisfaction with the appearance of their penis than controls who had been circumcised; [37,41] more adult hypospadias patients (26.2%) reported a negative genital appraisal than a control group (2%), and severity of hypospadias was related to a more negative genital appraisal;[38,43] high levels of dissatisfaction with penile appearance in both Japanese adults with hypospadias (40.9%) and control group (34.2%);[30] 28% of adult Chinese patients who underwent surgery before the age of ten reported dissatisfaction with their genital appearance compared to 8% of the control group; [33] adult hypospadias patients were less satisfied with their genital appearance than controls, but not less satisfied with their whole body image; [39] adult patients (24-42 years) in Hungary reported significantly lower genital satisfaction than age-matched controls; [40] and 10 patients aged 11-18 years reported less satisfaction with their genitals, and a lower genital perception score than controls, but this difference did not reach statistical significance. [43] Other studies in the last 10 years have also found high levels of dissatisfaction with penile appearance postsurgery, [32,44,45] with levels of dissatisfaction related to severity of hypospadias. [42] Overall, while research does indicate differences in satisfaction with appearance between patients and controls, these studies suffer from low homogeneity due to variance in type of corrective surgery, hypospadias severity, and methodological design.

Other, generally more recent, studies have reported either no significant differences between pre- and post-pubertal patients and controls or positive patient reports, in terms of penile perceptions. [23,28,31,35,46] Finding included the following: Adults reporting the cosmetic outcome of surgery as satisfactory, but less so for proximal patients; [28] no significant differences between hypospadias patients (aged 7–17 years) and controls on genital perception scores^[47] nor 6-17 years and controls on Pediatric Penile Perception Scores (PPPS);^[22] patients (aged 6-17 years) perceived the outcome of the surgery as very positive; [22] no significant differences between 7 and 17 years old hypospadias boys and control group in terms of PPPS, with both groups showing high levels of satisfaction; [46] 81% of 13-15 years old hypospadias boys were satisfied with their genital appearance; [31] 85% of 18–30 years old with severe hypospadias were satisfied with the appearance of their penis, despite 38% reporting residual curvature; [35] and 92% of a small sample of 13-21 years old patients with severe hypospadias were pleased with their penile appearance.[48]

Studies that have combined the data of pre- and post-pubertal under 18-year-old patients have tended to report relatively more positive penile perceptions.[31,46] Prepubertal patients show more positive perceptions than postpubertal ones, [23,26,46] possible because the penis has not yet acquired other social functions beyond micturition, and they have had less opportunity to compare and be aware of cosmetic differences. Following up patients until they reach sexual maturity and are sexually active should become the norm, otherwise pediatric surgeons with traditionally early discharge policies can continue to delude themselves about the quality of reconstruction that they are providing. It is only when sociosexual functions of the penis come to the fore from around puberty onward that the patients become greatly more critical of the surgical results. Some of the more positive perception findings^[35] may reflect the superior cosmetic results produced by modern surgical techniques. Nevertheless, some of the "appearance" benefits of more refined modern surgery may be partly negated by the confounding trend of ever increasing patient expectations; patients are becoming more demanding about body image and increasingly likely to seek and expect "perfection." Long-term follow-up of patients operated on children in the last 10–15 years will provide the missing data.

A key influencing factor in the measurement and interpretation of "cosmetic" outcome is the severity of

hypospadias. A patient with only mild hypospadias, who has some visible abnormality but no functional problems, may more likely seek esthetic perfection from surgery. A patient with a severe perineal hypospadias, even if the repair achieves a good esthetic outcome, may still rate it poorly on appearance because it looks significantly smaller than his peers (a factor related to the incomplete development of the penis). Nevertheless, such patients, because they have obviously deformed genitalia to start with, often have lower expectations of perfection; they are more likely to be grateful for a major improvement, even if this falls short of a truly normal outcome. One of the difficulties in making comparisons between these studies is the variation in the definition of the term "severe" hypospadias; some studies use the term to refer to mid or proximal shaft hypospadias. [35,48]

When comparing surgeon and patient ratings of penile appearance, studies have tended to produce inconsistent findings. [23,49] The method of data collection (e.g., who is interviewing the patient) may influence the results obtained. Furthermore, patients and surgeons may be influenced by different criteria. Patients will likely judge cosmetic outcome in relation to what they perceive to be peer normality. In contrast, a surgeon may judge cosmetic "excellence" not in relation to true normality but compared to prevailing surgical standards of the era or to what he can himself best achieve with his preferred technique.

Perceptions of deformity or normality

While patients may feel satisfied with the appearance of their penis, they may also separately perceive the appearance of their penis to be different from the norm. Depending on the type of technique used, a corrected penis may appear different in terms of glanular shape, penis size, and absence of foreskin or scars. To what extent do hypospadias patients feel their penis to be of a normal appearance and how important is this?

Bracka's^[24] study with 196 postpubertal patients (15–24 years) highlighted the importance of good cosmetic results: 72% of patients reported that an outcome of normal appearance was as important to them as normal functioning; 38% of patients reported that they still felt deformed; and 44% of the total sample requested further corrective surgery. Of these, 64.5% cited "abnormal appearance" as one of the reasons. This study should be viewed in context of the era in which these repairs took place. Modern techniques achieve better-looking repairs with fewer complications and can create a more natural looking meatus sited on the tip of the penis.

Kumar and Harris^[25] reported positive findings for preputial island flap terminalizing procedures performed in the UK between 1976 and 1982; 80% of their sample of 35 patients (aged 13–25 years) reported perceptions that their penis looked normal, perhaps reflecting that an apical glans meatus

was achieved in most cases. This contrasts with Mureau *et al.* Netherlands' studies,^[26,36] from a similar era, but where a large proportion of the cohort had a Van der Meulen ventralizing repair. Results showed 78% of 116 pre- and post-pubertal patients (compared to 35% of controls) and 84.7% of 73 adults (compared to 40% of controls) perceived their penile appearance to be different from other boys/men.^[26,36] Given the inherently abnormal appearance of the Van der Meulen repair, these higher dissatisfaction rates are not too surprising. Similarly, Mor *et al.*^[50] reported 46.5% of patients perceived their penile appearance as abnormal following a Browne dorsal meatoplasty (a procedure which creates an abnormal looking glans - meatus configuration).

More recent studies have also found relatively high levels of perceived abnormality among hypospadias patients: 31.4% of postpubertal 13-15 years old boys in an Australian study thought their penis appeared abnormal compared to their peers;^[31] there was a significant difference between Belgian patients and controls regarding perceptions of "normal appearance of penis" and of "penis same as peers;"[51] 74.5% of adults in a study in China perceived their penis to be different from the norm; [32] while 74% of adult patients with severe hypospadias reported that their penis looked abnormal, and 30% reported dissatisfaction because of the absence of a foreskin. [52] The problem with comparing these results is that they span three different continents, where cultural differences will come into play, and differences in the severity of hypospadias. Several papers have raised the issue of a circumcised appearance posthypospadias surgery^[53,54] and the influence of the cultural climate; most European patients will be in the minority in having a circumcised penis, and this can undoubtedly lead to perceptions of being different from peers. However, Ruppen-Greeff et al.'s[41] study still found that patients scored significantly lower than controls on genital perceptions, using the PPS, [23] even when the variable of circumcision was held constant.

Overall, research suggests that around 70% of adults perceive the appearance of their penis postcorrective surgery to be abnormal. However, perceiving one's penis as being different from the norm within a given culture is not the same as being dissatisfied with its appearance. Thus, Mureau et al.[26] found that hypospadias patients who were satisfied with their circumcision status also had higher genital perception scores. However, their study in 1995[36] reported that despite a 78% perception of "abnormality," only 25% expressed dissatisfaction with appearance. Similarly, Jones et al.,[31] who reported 31.4% of their sample perceiving their penis to be abnormal compared to their peers, also reported that 81% were satisfied with their genital appearance, and that 90% were satisfied with their overall body image. Vandendriessche et al.[51] reported that fewer patients than controls perceived their penis to be normal or the same as their peers. However, they concluded that the self-concept and body image of hypospadias adolescents were intact as there was no significant difference in genital perception scores (although trending toward significance).

Social embarrassment

To what extent do hypospadias patients suffer social embarrassment through the display of an 'abnormal' appearance in public settings that involve undressing (e.g., showers after sports)?

Both Bracka's^[24] UK study and Mureau *et al.*'s^[27,36] European studies found significant cause for concern. Over 50% of Bracka's patients reported avoiding communal showers for fear of ridicule, and a similar percentage feared that they would be rejected by the opposite sex. Mureau *et al.*'s^[26,36] study reported high rates of social embarrassment about their penile appearance, receiving comments on the appearance of their penis when undressing publicly, and hiding their genitals in public lavatories. Similar social embarrassment has been reported by some^[31,33,38] but not all studies.^[25,27,51]

A lack of postoperative guidance about the condition can lead to social morbidity, [24] highlighting the importance of reassuring patients postoperatively about the normality of their penis, especially with regard to size and absence of foreskin. [50] Several authors have raised the issue of the link between cultural context and social embarrassment regarding circumcision. [47,54,55] If it is anticipated that a circumcised status is likely to cause significant social embarrassment, due to a particular cultural context, then foreskin reconstruction, which generally has a low complication rate, [56] may be considered as an option. Furthermore, one must also consider the long-term cosmetic and functional outcome of a childhood foreskin reconstruction as it must also remain satisfactory to the adolescent and adult patient. Yet, there is little adult follow-up data available.

DISCUSSION

Studies in this review vary according to methodology and surgical procedures making comparisons difficult. This is compounded by the problem of poor reporting, use of nonvalidated measuring instruments, the paucity of long-term follow-up, and the lack of reliable evidence to allow systematic comparison between groups (e.g. according to age or severity). Comparable data on the long-term outcomes of hypospadias surgery are crucial for the monitoring and improvement of clinical practice. Accurate reporting of results requires validated, standardized methods of assessment, unbiased methods of data collection, along with greater rigor in the methodological design of studies. All of these factors contribute to a degree of risk of bias across studies included in this review. While validated instruments for patient and surgeon assessment of appearance are a welcome addition,[22,23] further instruments are required to measure patients' subjective perceptions of themselves

against their perceived norms. Only when standardized methods of assessment for patient perceptions of cosmesis become the norm, it will be possible to determine its relationship to psychosocial and psychosexual outcomes in adulthood.

Few studies focus on cosmetic outcomes postpuberty, and even less in sexually active adults. Long-term follow-up beyond puberty must become standard practice as genital appraisal becomes more important during and postpuberty than childhood, and short-term outcomes may not always predict long-term ones. The development of an industry protocol for follow-up that prevents the signing-off of pediatric patients before they reach puberty or adulthood will not only improve patient care and inform clinical practice but also provide more realistic appraisal of individual surgeons' success rates.

The trends from more recent studies tend to suggest that there are not such large differences between patients and the normal population or controls. However, there is still clear evidence that a large proportion of patients perceive their penis as appearing different from that of their peers. Some of these results can be explained by the circumcision status following surgery, but not all of them. These perceptions of abnormality or "difference" may explain the continuing findings of social embarrassment at undressing in public.

All of these need to be seen within the social context of today's adolescent and young adult patients. Particularly within western cultures, the cosmetics industry is booming, with increasing numbers of young people seeking to correct perceived physical imperfections. The young hypospadias patients operated on a decade ago are now part of an adolescent generation with higher expectations than the adolescents of that era. It is a trend that seems likely to continue as successive generations of young adults continue to raise the bar for will be deemed to be a "good" cosmetic outcome. While middle-aged patients who have already proved themselves sexually may be less concerned about penis size and minor cosmetic imperfections, sexually inexperienced younger patients will understandably be more anxious about their prospects. Long-term reviews of the cosmetic outcomes for these patients are needed because although surgical techniques have improved dramatically, so have patient expectations of what is an acceptable result.

Long-term clinical management also requires the development of realistic patient and parental cosmetic expectations. Parental attitudes toward the resulting appearance of their son's penis, and attitude toward his condition, are also important as they will be a major source of reference once he reaches adolescence. Patients who are educated about their condition, who are made aware that hypospadias surgery will result in a circumcised appearance,

but will not increase penis size, and who are provided with realistic norms against which to assess themselves are more likely to be accepting of their condition. Those who are left in ignorance are more likely to develop a sense of shame and suffer social embarrassment. Pre- and post-operative counseling should be provided to reduce the potential for negative psychological impact. A positive penile appraisal is important for genital body image and psychological well-being, particularly in terms of self-esteem. The addition of longer follow-up care, with informed discussions between patients, parents, and surgeons, may assist in the development of positive penile perceptions.

CONCLUSIONS

Protocols for clinical postpuberty follow-up and methodologically sound studies, using validated assessment tools, are required for the accurate assessment of cosmetic and psychological outcomes of hypospadias surgery. While promising validated scales have been developed, findings from this review would suggest that directions for future research include the need for the development of new instruments that include the severity of hypospadias, assessment of subjective sexual function, and patients' subjective perceptions of themselves against perceived norms.

Financial support and sponsorship Nil.

Conflicts of interest

There are no conflicts of interest.

REFERENCES

- Baskin LS, Himes K, Colborn T. Hypospadias and endocrine disruption: Is there a connection? Environ Health Perspect 2001;109:1175-83.
- Wilcox D, Snodgrass W. Long-term outcome following hypospadias repair. World J Urol 2006;24:240-3.
- 3. Atala A, McAninch JW. Urethral reconstructions. Urol Clin North Am 2002:29:xiii
- 4. Mouriquand PD, Persad R, Sharma S. Hypospadias repair: Current principles and procedures. Br J Urol 1995;76 Suppl 3:9-22.
- Manzoni G, Bracka A, Palminteri E, Marrocco G. Hypospadias surgery: When, what and by whom? BJU Int 2004;94:1188-95.
- Timmons MJ. The UK primary hypospadias surgery audit 2006-2007.
 J Plast Reconstr Aesthet Surg 2010;63:1349-52.
- Moher D, Liberati A, Tetzlaff J, Altman DG; PRISMA Group. Preferred reporting items for systematic reviews and meta-analyses: The PRISMA statement. BMJ 2009;339:b2535.
- OCEBM Levels of Evidence Working Group. The Oxford Levels of Evidence; 2011. Available from: http://www.cebm.net/mod_product/ design/files/CEBM-Levels-of-Evidence-2.1.pdf. [Last accessed on 2014 May 02].
- Furlan AD, Pennick V, Bombardier C, van Tulder M; Editorial Board, Cochrane Back Review Group. 2009 updated method guidelines for systematic reviews in the Cochrane Back Review Group. Spine (Phila Pa 1976) 2009;34:1929-41.

- Timmer A, Sutherland LR, Hilsden RJ. Development and evaluation of a quality score for abstracts. BMC Med Res Methodol 2003;3:2.
- Roy M, Payette H. The body image construct among Western seniors: A systematic review of the literature. Arch Gerontol Geriatr 2012;55:505-21.
- Braun V, Clarke V. Using thematic analysis in psychology. Quant Res Psychol 2006;3:77-101.
- Lever J, Frederick DA, Peplau LA. Does size matter? Men's and women's views on penis size across the lifespan. Psychol Men Masc 2006:7:129-43.
- Grov C, Parsons JT, Bimbi DS. The association between penis size and sexual health among men who have sex with men. Arch Sex Behav 2010;39:788-97.
- 15. Veale D, Eshkevari E, Read J, Miles S, Troglia A, Phillips R, *et al.* Beliefs about penis size: Validation of a scale for men ashamed about their penis size. J Sex Med 2014;11:84-92.
- Martins Y, Tiggemann M, Churchett L. The shape of things to come: Gay men's satisfaction with specific body parts. Psychol Men Masc 2008:9:248-56.
- Tiggemann M, Martins Y, Churchett L. Beyond muscles: Unexplored parts of men's body image. J Health Psychol 2008;13:1163-72.
- Herbenick D, Schick V, Reece M, Sanders SA, Fortenberry JD. The development and validation of the Male Genital Self-Image Scale: Results from a nationally representative probability sample of men in the United States. J Sex Med 2013;10:1516-25.
- Johnston L, McLellan T, McKinlay A. (Perceived) size really does matter: Male dissatisfaction with penis size. Psychol Men Masc 2014;15:225-8.
- Holland AJ, Smith GH, Ross FI, Cass DT. HOSE: An objective scoring system for evaluating the results of hypospadias surgery. BJU Int 2001;88:255-8.
- 21. van der Toorn F, de Jong TP, de Gier RP, Callewaert PR, van der Horst EH, Steffens MG, et al. Introducing the HOPE (Hypospadias Objective Penile Evaluation)-score: A validation study of an objective scoring system for evaluating cosmetic appearance in hypospadias patients. J Pediatr Urol 2013;9(6 Pt B):1006-16.
- Weber DM, Schönbucher VB, Landolt MA, Gobet R. The pediatric penile perception score: An instrument for patient self-assessment and surgeon evaluation after hypospadias repair. J Urol 2008;180:1080-4.
- Weber DM, Landolt MA, Gobet R, Kalisch M, Greeff NK. The penile perception score: An instrument enabling evaluation by surgeons and patient self-assessment after hypospadias repair. J Urol 2013;189:189-93.
- 24. Bracka A. A long-term view of hypospadias. Br J Plast Surg 1989:42:251-5.
- Kumar MV, Harris DL. A long term review of hypospadias repaired by split preputial flap technique (Harris). Br J Plast Surg 1994;47:236-40.
- Mureau MA, Slijper FM, Slob AK, Verhulst FC. Genital perceptions of children, adolescents, and adults operated on for hypospadias: A comparative study. J Sex Res 1995; 32:289-98.
- Mureau MA, Slijper FM, van der Meulen JC, Verhulst FC, Slob AK. Psychosexual adjustment of men who underwent hypospadias repair: A norm-related study. J Urol 1995;154:1351-5.
- Wouters G, Rynja S, van Schaijk M, Kok ET, de Jong TP, de Kort LM. Long term follow up into adulthood of boys operated on for hypospadias: A study into functional and cosmetic results. J Pediatr Urol 2009;5:S56.
- Rynja SP, Wouters GA, Van Schaijk M, Kok ET, De Jong TP, De Kort LM. Long-term followup of hypospadias: Functional and cosmetic results. J Urol 2009;182 4 Suppl: 1736-43.
- Moriya K, Kakizaki H, Tanaka H, Furuno T, Higashiyama H, Sano H, et al. Long-term cosmetic and sexual outcome of hypospadias surgery: Norm related study in adolescence. J Urol 2006;176(4 Pt 2):1889-92.
- Jones BC, O'Brien M, Chase J, Southwell BR, Hutson JM. Early hypospadias surgery may lead to a better long-term psychosexual outcome. J Urol 2009;182 4 Suppl: 1744-9.

- 32. Jiao C, Wu R, Xu X, Yu Q. Long-term outcome of penile appearance and sexual function after hypospadias repairs: Situation and relation. Int Urol Nephrol 2011;43:47-54.
- Wang WW, Deng CH, Chen LW, Zhao LY, Mo JC, Tu XA. Psychosexual adjustment and age factors in 130 men undergone hypospadias surgery in a Chinese hospital. Andrologia 2010;42:384-8.
- 34. Springer A. Assessment of outcome in hypospadias surgery A review. Front Pediatr 2014;2:2.
- 35. Fraumann SA, Stephany HA, Clayton DB, Thomas JC, Pope JC 4th, Adams MC, *et al.* Long-term follow-up of children who underwent severe hypospadias repair using an online survey with validated questionnaires. J Pediatr Urol 2014;10:446-50.
- Mureau MA, Slijper FM, Nijman RJ, van der Meulen JC, Verhulst FC, Slob AK. Psychosexual adjustment of children and adolescents after different types of hypospadias surgery: A norm-related study. J Urol 1995;154:1902-7.
- Aho MO, Tammela OK, Somppi EM, Tammela TL. Sexual and social life of men operated in childhood for hypospadias and phimosis. A comparative study. Eur Urol 2000;37:95-100.
- Mondaini N, Ponchietti R, Bonafè M, Biscioni S, Di Loro F, Agostini P, et al. Hypospadias: Incidence and effects on psychosexual development as evaluated with the Minnesota Multiphasic Personality Inventory test in a sample of 11,649 young Italian men. Urol Int 2002;68:81-5.
- Kiss A, Sulya B, Szász AM, Romics I, Kelemen Z, Tóth J, et al. Long-term psychological and sexual outcomes of severe penile hypospadias repair. J Sex Med 2011;8:1529-39.
- Merksz M, Sulya B, Szasz M, Romics I, Kelemen Z, Nyirady P, et al. Long-term psychosexual consequences of hypospadias repair. J Pediatr Urol 2010;6:S81.
- Ruppen-Greeff NK, Weber DM, Gobet R, Landolt MA. Health-related quality of life in men with corrected hypospadias: An explorative study. J Pediatr Urol 2013;9:551-8.
- 42. Örtqvist L, Fossum M, Andersson M, Nordenström A, Frisén L, Holmdahl G, *et al.* Long-term followup of men born with hypospadias: Urological and cosmetic results. J Urol 2015;193:975-81.
- 43. Chertin B, Natsheh A, Ben-Zion I, Prat D, Kocherov S, Farkas A, *et al.* Objective and subjective sexual outcomes in adult patients after hypospadias repair performed in childhood. J Urol 2013;190 4 Suppl: 1556-60.
- Nelson CP, Bloom DA, Kinast R, Wei JT, Park JM. Patient-reported sexual function after oral mucosa graft urethroplasty for hypospadias. Urology 2005;66:1086-9.
- Hoag CC, Gotto GT, Morrison KB, Coleman GU, Macneily AE. Long-term functional outcome and satisfaction of patients with hypospadias repaired in childhood. Can Urol Assoc J 2008;2:23-31.
- Schönbucher VB, Landolt MA, Gobet R, Weber DM. Psychosexual development of children and adolescents with hypospadias. J Sex Med 2008;5:1365-73.
- Schlossberger NM, Turner RA, Irwin CE Jr. Early adolescent knowledge and attitudes about circumcision: Methods and implications for research. J Adolesc Health 1992;13:293-7.
- 48. Lam PN, Greenfield SP, Williot P. 2-stage repair in infancy for severe hypospadias with chordee: Long-term results after puberty. J Urol 2005;174(4 Pt 2):1567-72.
- Mureau MA, Slijper FM, Slob AK, Verhulst FC, Nijman RJ. Satisfaction with penile appearance after hypospadias surgery: The patient and surgeon view. J Urol 1996;155:703-6.
- Mor Y, Ramon J, Jonas P. Is only meatoplasty a legitimate surgical solution for extreme distal hypospadias? A long-term follow-up after adolescence. BJU Int 2000;85:501-3.
- 51. Vandendriessche S, Baeyens D, Van Hoecke E, Indekeu A, Hoebeke P. Body image and sexuality in adolescents after hypospadias surgery. J Pediatr Urol 2010;6:54-9.
- 52. Aulagne MB, Harper L, de Napoli-Cocci S, Bondonny JM, Dobremez E.

- Long-term outcome of severe hypospadias. J Pediatr Urol 2010;6:469-72.
- 53. Mureau MA. Psychosexual and psychosocial development of patients with hypospadias. Ned Tijdschr Geneeskd 1997;141:188-91.
- 54. Bracka A. Sexuality after hypospadias repair. BJU Int 1999;83 Suppl 3:79,33
- 55. Eray N, Dogangun B, Kayaalp L, Emir H, Soylet Y, Danismend N, *et al.* Emotional effects of hypospadias surgery on Turkish boys. J Pediatr Urol 2005;1:75-80.
- 56. Snodgrass WT, Koyle MA, Baskin LS, Caldamone AA. Foreskin preservation in penile surgery. J Urol 2006;176:711-4.