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Review Article

Assessing quality of life of patients with hypospadias: A systematic review of validated patient-reported outcome instruments

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Summary

Background
Patient-reported outcomes have the potential to provide invaluable information for evaluation of hypospadias patients, aid in decision-making, performance assessment, and improvement in quality of care. To appropriately measure patient-relevant outcomes, well-developed and validated patient-reported outcome (PRO) instruments are essential.

Objective
To identify and evaluate existing PRO instruments designed to measure quality of life and/or satisfaction of individuals with hypospadias that have been developed and validated in a hypospadias population.

Methods
A systematic search of MEDLINE, EMBASE, PsycINFO, CINAHL and Health and Psychosocial Instruments was conducted in April 2016. Two reviewers independently assessed studies and identified PRO instruments for inclusion. Data were extracted on study characteristics, instrument development and validation, and content domains.

Results
A total of 32 studies were included that used or described five PRO instruments: Hypospadias Objective Scoring Evaluation (HOSE), Pediatric Penile Perception Score (PPPS), Penile Perception Score (PPS), Genital Perception Scale (GPS) for adults, and GPS for children/adolescents. Instrument development and validation was limited. The majority of identified instruments focused on postoperative cosmetic satisfaction, with only one instrument considering urinary function, and no instruments evaluating sexual function and psychosocial sequelae.

Conclusions
While many hypospadias studies have acknowledged the necessity of a patient-reported element, few have used validated PRO instruments developed in a hypospadias population. Existing instruments to measure patient-reported outcomes in hypospadias require improvement in both the breadth of content and in their development and validation methodology.

Introduction

Patient-reported outcome (PRO) instruments are questionnaires/instruments that allow for self-reporting of the patient (or parent-proxy) experience, potentially including views of their symptoms, functional status, and health-related quality of life (QoL) [1]. While originally designed for use in research [1], PRO instruments have been adopted by healthcare professionals to support various clinical efforts, including quality improvement, performance assessment, and the tailoring of treatment plans to meet patient preferences and needs [2]. The latter is of particular importance, as healthcare professionals frequently misjudge the absolute levels of symptoms and general QoL, tending to underestimate the impact of psychological factors while emphasizing more obvious symptoms [3]. Jachuck et al. [4] observed this in their questionnaire study of hypertensive patients and their doctors, where all physicians indicated that patients had improved, while approximately half of the patients felt that there was no change or even deterioration. Physicians tended to ignore the factors that patients factored into their overall well-being, including a decline in energy, general activity, sexual inactivity, and irritability [4].

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Patient-reported outcomes are particularly useful for conditions where a large variation in care and outcomes exists, and where the impact on the patient's QoL is currently unknown. Such is the case for the congenital condition of hypospadias. While the goals of hypospadias repair are generally agreed upon — including providing the patient with satisfactory urinary function, sexual function, body image or cosmesis, and quality of life — several important variations remain. First, there is variation in surgical techniques for the same condition; for example, the use of one-stage vs. two-stage repair for proximal hypospadias [5]. While advantages have been established for each technique, the current evidence base cannot definitively identify an optimal method for individual patients [6]. Patient input, obtained through PROs, on the relative benefits may help elucidate this (e.g. does the reduced hospital stay and anesthetic risk associated with a one-stage repair offset the increased risk of complications [5,7] compared with a two-stage repair?). Additional variance also exists concerning physician recommendations and parental preference for surgical correction of distal hypospadias (i.e. glanular), where some routinely elect/recommend surgical repair and others prefer to forgo surgery in mild cases. Finally, variance can be observed in surgical success, where 15% of patients experience a complication (a value that at least doubles in patients with severe defects or prior complications) [7—9].

While it remains important to measure these results using traditional surgical outcomes, such as complications and need for reoperation, these no longer sufficiently capture all important aspects from the patient's perspective [10—12]. Rather, there is a need to capture the considerable long-term cognitive, behavioral, and self-esteem consequences that result from poor cosmetic or functional outcomes — such as negative genital perception [13,14], sexual avoidance [15,16], and poor school performance [17] — through the use of PRO measures. Comprehensive measurement of surgical outcomes requires a combination of objective and subjective measures.

Clinical outcomes research in hypospadias surgery is becoming increasingly focused on the patient's QoL and their perception of a satisfactory outcome [10]. As a result, the present systemic review of the published literature was conducted to identify and assess PRO instruments currently available for hypospadias patients or parent-proxies. The primary objective was to identify existing PRO instruments that have been developed and validated in a hypospadias population that assess patient satisfaction and/or quality of life. A secondary objective was to evaluate the development and validation of the instruments, and to assess the content of identified instruments.

Methods

Inclusion criteria

Studies were included that described PRO instruments designed to measure quality of life and/or satisfaction of individuals with hypospadias or their parent-proxy that had been developed and validated in a hypospadias population. Non-English language studies were excluded, as were conference abstracts.

Literature search

The following databases were searched: MEDLINE including In-Process & Other Non-Indexed Citations (1946-April 8 2016), EMBASE (1980-week 14 2016), PsycINFO (1806-April week 1 2016), CINAHL (April 8 2016), and Health and Psychosocial Instruments (1985-January 2016). The MEDLINE search strategy was developed by a librarian experienced in systematic review searching, and peer reviewed by a second librarian using the PRESS standard [18]. The MEDLINE search was then adapted to allow for optimal searching of other included databases. Search strategies are presented in the Appendix. Hand-searching the reference sections of relevant articles identified additional publications.

Screening

At level 1 (title and abstract), studies were screened independently in duplicate using the liberal accelerated method [19]. Two researchers then independently assessed full-text articles (level 2 screening) and compared decisions to reach consensus for final inclusion. Disagreements were resolved by discussion or third party consultation when necessary.

Development and validation criteria

Validated instruments were identified from included studies and were evaluated for their adherence to the three-stage, rigorous, gold standard methodology for the development and validation of health outcome measures developed by the Scientific Advisory Committee (SAC) of Medical Outcomes Trust International guidelines [20] and described by Cano et al. [21]. According to this methodology, stage 1 involves generating a list of items for the PRO instruments based on patient interviews, expert opinion, and review of the literature. The number of items is then reduced in stage 2 according to expert opinion, item redundancy, endorsement frequency, missing data, factor analysis, and tests of scaling assumptions. This final instrument then undergoes psychometric evaluation and validation at stage 3, including determination of acceptability, internal consistency, reliability, and validity within scale, among others [22]. All publications that detailed instrument development and validation were reviewed to determine which aspects of each stage were completed to inform the final PRO instruments. Authors of each instrument were also contacted to confirm the process of development and validation. Finally, content domains covered by the included instruments were summarized.

Results

A search of existing literature identified 1666 articles for review, with another 14 identified through grey-literature and hand searching (Fig. 1). Following removal of