



Review Article

Urological and sexual outcome in patients with Hirschsprung disease: A systematic review



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Summary

Purpose

There is a paucity of recent evidence regarding long-term urological and sexual outcomes following surgery for Hirschsprung disease (HD). We aimed to undertake a systematic review of all HD literature to define these outcomes.

Materials and methods

A systematic literature search was conducted on studies from 1966 to 2014. Relevant articles were assessed for urological/sexual operative complications and functional sequelae. Studies were analysed in qualitative (Rangel score) and quantitative syntheses.

Results

Initially 257 reports were assessed, with 24 studies were eligible for inclusion (1972–2014). Mean study quality was $16.5 \pm SD 4.8$ (range 6–23), indicating overall fair/poor quality. Ten studies (1021 patients) reported operative complications, with ureteric/urethral/vaginal injury occurring in seven (0.7%) patients. In three studies, the primary outcome was

urological functional assessment. From 17 studies, 52/2546 patients (2.0%) had reported urinary incontinence. In infants, absent spontaneous erections post-operatively was reported in 3/203 patients (1.5%, 5 studies); of these 3, parents did not note spontaneous erections pre-operatively either. In older patients, erectile dysfunction occurred in 6/498 (1.2%) males. Other sexual outcomes were reported in 10 studies, with 5/10 studies (416 patients) reporting no erectile dysfunction. In the other studies reports ranged from non-specified sexual dysfunction in one study to diverse sexual related problems in nine (7.8%) of their patients in another.

Conclusions

Urological/sexual outcomes are rarely reported after HD surgery (24 studies over 42 years). Study quality is usually poor and a large proportion of the studies are more than 30 years old. In the majority of series it is unclear whether urological and sexual function impairments were not present or if they were not assessed. Prospective reporting of urological/sexual outcome is required, in particular in the era of new surgical techniques/approaches to HD.

Introduction

A wide range of long-term urological and/or sexual disorders may occur as a consequence of congenital anomalies such as ano-rectal malformations (ARM), in which urological disorders have been observed in up to half of patients [1,2]. In ARM neurogenic bladder, for example, may result either as secondary to nerve injury caused by pelvic dissection, because of associated spinal malformations. Surgical trauma to the retro-rectal space is in itself well-recognised for causing urological or sexual dysfunction, such as retrograde ejaculation or impotence after abdominal rectopexy for rectal prolapsed [3]. In a recent review, associated congenital anomalies of the kidneys and the urinary tract were found to be

an underestimated problem in patients with Hirschsprung disease (HD), although a significant proportion of those reported (inguinal hernia, undescended testis) would be unlikely to influence urological function directly [4]. The authors of this review therefore advocate urological screening in all HD patients. As there is no associated spinal involvement in Hirschsprung Disease (HD), no inherent abnormalities of urological or sexual function might be expected other than secondary to surgical trauma or congenital associated anomalies of the urinary tract.

Pelvic nerves at risk during pelvic dissection are principally the superior hypogastric plexus, containing sympathetic nerves (lumbar origin) and parasympathetic pelvic splanchnic fibres [5]. These plexi are more readily

identifiable in adult patients. Injury may lead to dysfunctional detrusor and bladder neck contraction/relaxation, as well as disordered micturition. Neural trauma may also result in altered uterine, vaginal, or vulval sensation in females, or retrograde ejaculation/erectile dysfunction in males. Modification of operative techniques for rectopexy have been proposed to minimise the risk of trauma [6]. In addition, the ureters and vasa are in close proximity to the recto-sigmoid colon and may be at risk of injury.

Broadly, three operative procedures, or their modifications, remain in common use for HD. Swenson's procedure from the 1950s was the first widely used structural surgical approach for HD surgery and comprised a full-thickness trans-abdominal rectal dissection [7]. This technique had good clinical results regarding colorectal function; however, it became less popular after complications such as urinary incontinence and erectile dysfunction were reported [8]. Several other techniques were described, of which the Duhamel and Soave approaches are the ones most widely used [9,10]. In the Duhamel technique, a retro-rectal tunnel is fashioned to allow an end-to-side colo-anal anastomosis, leaving an aganglionic anterior rectal pouch. The Soave approach consists of submucosal rectal dissection, leaving an aganglionic, muscular rectal cuff. All these procedures initially were presented as trans-abdominal ones to minimise injury to other organs, and pelvic nerves and vessels. In the last two decades, there have been numerous reports of minimally invasive techniques, either using laparoscopic-assisted mobilisation, or, more recently, complete trans-anal dissection [11–14].

Besides the early reports of the trans-abdominal Swenson's approach, there is a paucity of data regarding the urological and sexual long-term outcome after HD surgery. This is particularly pertinent in an era of new surgical approaches. Therefore, we conducted a systematic review, with the aim of this study being to assess long-term difficulties in urological and sexual function as reported in current literature [2,15].

Material and methods

Guideline

The PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analyses) statement, checklist and flow-chart were used to achieve the highest standard in reporting items for a systematic review and meta-analysis [16].

Search strategy

A systematic literature search was conducted on 18 August 2014 using the PubMed, EMBASE, and Web-of-Science databases. Studies were searched in PubMed using the following search terms: *Hirschsprung AND (urological OR urologic OR urinary OR dysfunctional voiding OR gynecological OR gynaecological OR sexual OR fertility OR erectile OR ejaculation)*. For the other databases similar search terms were applied which

concerned the functional outcome of patients operated on for HD.

Eligibility criteria

Studies reporting on the urological or sexual outcome of patients with HD were selected. Urological outcome was defined as any reporting on urinary continence or other micturition difficulties. Sexual outcome was defined as any reporting on sexual function, such as erectile (dys) function, dyspareunia, or reproductive capabilities. Furthermore, post-operative urological complications were assessed (e.g. ureteric injury). Case reports presenting single patients because of their special course, treatment, or outcome were excluded. Case series that presented a special subgroup of the institution's general HD population were excluded. Different articles that presented identical variables of the same study population were excluded, and the most recent publication, the publication presenting the largest sample, or the most outcome variables was chosen. The references of each of the articles found were also reviewed to include useful studies that might have been missed with the initial literature review.

Study selection

The study selection consisted of four separate processes; 1. Study identification, 2. Study screening, 3. Study eligibility, and 4. Study inclusion. All processes were conducted by two independent reviewers (HPV, NSJ).

Quality assessment

Quality of the articles was scored using the checklist as proposed by Rangel et al. [17]. The checklist consisted of three subscales containing 30 items in total. The three subscales were: 1. Potential Clinical Relevance, 2. Quality of Study Methodology, and 3. Quality of Discussion and Stated Conclusions. A maximum of 45 points could be scored. Scores of 0–15 indicated a study of 'poor' quality, studies scoring 16–30 points were considered 'fair', and scores of 31 points or higher indicated a qualitatively 'good' study.

Data extraction

Two reviewers (HPV, NSJ) used pre-defined criteria for data extraction from included publications. The pre-defined criteria concerned study design, population, and urological/sexual function. In total, the reviewing process took 5 months (August–December 2014).

Statistical analysis

Data were stored and analysed using SPSS (version 17; SPSS, Chicago, IL, USA).

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