



Lower urinary tract symptoms and sexual functions after endorectal pull-through for Hirschsprung disease: controlled long-term outcomes ^{☆,☆☆,★}



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ABSTRACT

Background/purpose: To define the prevalence of lower urinary tract symptoms (LUTS) and outcomes for sexual function after endorectal pull-through (EPT) for Hirschsprung disease (HD) compared to controls. To date, similar controlled studies are lacking.

Methods: Patients aged ≥ 4 years ($n = 123$) operated on for HD at our center between 1987 and 2011 were invited to answer questionnaires on LUTS and sexual function (aged ≥ 16 years). Patients with an intellectual disability and patients with a definitive endostomy were excluded. Patients were matched to three controls and also invited to a clinical follow-up for urological investigations including urine flow measurement, renal tract ultrasound, and urinalysis.

Results: Altogether, 59 responses concerning LUTS and 24 responses concerning sexual functions were analyzed. No significant differences were demonstrated in the overall prevalence of LUTS between patients (67%) and controls (80%), nor in the prevalence of frequent LUTS (14% vs. 16%; $P = \text{NS}$ for both). One patient (2%) had a urethral stricture after laparotomy-assisted EPT. Male patients reported sexual satisfaction and erectile function similar to controls ($P > 0.10$). Female patients were currently less in stable relationships compared to controls (25% vs. 83%, $P = 0.005$).

Conclusions: Our results support the safety of EPT in patients with HD with regard to preservation of the integrity and functioning of the genitourinary tract.

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Hirschsprung's disease (HD) is a congenital condition affecting 1:5000 live births characterized by an absence of ganglion cells in the myenteric and submucosal plexuses of the intestine. Surgical treatment involves resection of the distal aganglionic bowel segment and pull-through of the remaining ganglionic bowel down to the anus [1]. During the last decades, endorectal pull-through (EPT) techniques, including totally transanal operations have become increasingly popular approaches for the management of HD [1–5]. Historically, the original Swenson, Soave and Duhamel operations involved the entire rectal dissection via abdomen. Inadvertent injuries to the genitourinary tract or its innervation may have occasionally occurred during pelvic dissection especially outside the rectal wall in the Swenson and Duhamel operation, resulting in lower urinary tract symptoms (LUTS) and sexual

dysfunction including infertility and erectile difficulties in males [6–9]. EPT may carry less risk of these than other techniques because dissection of the distal rectum within the pelvis is carried out under direct vision on the rectal wall, and initially within a mucosal cuff in the Soave-like approach. The few uncontrolled studies available after EPT lend support to this notion [10,11]. There is very limited information concerning the outcomes for sexual function in adulthood in patients with HD [7,12,13]. This study has aimed to evaluate the long-term outcomes for LUTS and sexual function after EPT for HD in relation to age- and gender-matched peers from the general population, in order to provide further information on the safety of these procedures regarding preservation of the functional integrity of the genitourinary tract.

1. Methods

1.1. Patients

All patients aged ≥ 4 years who underwent EPT at our center after histological verification of HD between 1987 and 2011 were identified from records. Patients with an intellectual disability due to an associated syndrome and patients with a definitive endostomy were excluded from the analysis. Patients (or their caregivers) were contacted twice

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by post between 2012 and 2013 and invited to answer detailed questionnaires concerning LUTS (all patients) and sexual function (patients aged ≥ 16 years only). Voluntary participants were also invited to attend a clinical follow-up that included a clinical examination, urine flowmetry, renal tract ultrasound, and laboratory tests (infection parameters, electrolytes, creatinine, blood count, and urinalysis). An independent investigator who had not been involved in their surgical care contacted the patients. Operative details were obtained retrospectively from records. The ethics committee of the Helsinki University Hospital approved the research protocol.

1.2. Questionnaires

Of the nine items concerning LUTS (Appendix A), items three to nine were adapted from the previously validated Danish Prostatic Symptom Score [14]. Sexual function was enquired from patients aged ≥ 16 years (Appendix B), including assessment of erectile function in males according to the previously validated Erectile Hardness Score (EHS) [15].

1.3. Renal tract ultrasound

Renal tract ultrasound including residual volume was performed by a pediatric radiologist blinded to the clinical outcomes.

1.4. Urine flowmetry

Urine flowmetry was performed using a spinning disc transducer URODYN® 1000 (© Mediwatch Plc 2008). The flowmetry curves were classified as belly-shaped (normal) or tower-, plateau-, staccato- and interrupted-shaped (abnormal) [16]. A tower-shaped curve was defined as a high-amplitude curve of short duration. A plateau curve was a low amplitude and even curve. A staccato pattern was a fluctuating curve or curve with multiple peaks. An interrupted curve was defined as a curve reaching the baseline during voiding.

1.5. Controls

The controls were obtained from a reference pool of 594 Finnish subjects aged 4–26 years who had been randomly selected from the Population Register Centre of Finland and had answered identical questionnaires to patients [17]. From this pool, three controls matched for age and gender were randomly selected for each patient.

1.6. Operative principles

As shown in Table 1, all patients underwent either totally transanal EPT ($n = 12$) or EPT in combination with transabdominal mobilization of the colon ($n = 44$) outside the pelvis proximal to the peritoneal reflection. In all cases, dissection of the rectum within the pelvis up to the peritoneal reflection was carried out transanally. Transanal mucosectomy was commenced at approximately 5 mm proximal to the dentate line, proceeding to full-thickness dissection after 3–4 cm thereafter [18,19]. One patient with panintestinal aganglionosis underwent an end jejunostomy and colectomy [20]. The length of aganglionosis was defined by pathological examination of frozen sections. Patients were operated on by the same team of our pediatric consultant colorectal surgeons who also followed them up to adulthood.

1.7. Statistics

Unless otherwise stated, data are presented as frequencies or medians \pm interquartile range (IQR). Statistics were calculated using SPSS Version 21.0. Categorical variables were compared using Chi-square or Fisher's exact test, and continuous variables using Mann-Whitney U test. $P < 0.05$ was considered significant.

Table 1

Baseline characteristics of respondents with normal cognition ($n = 59$).

	Survey of LUTS ($n = 59$)	Survey of sexual function ($n = 24$)
Median age (years)	15 (9–21)	22 (18–24)
Sex (M:F)	43:16 (3:1)	16:8 (2:1)
Family history of HD	10 (17)	3 (13)
Age at PT surgery (weeks)	8 (3–36)	21 (11–77)
Level of aganglionosis		
Rectosigmoid	51 (86)	22 (92)
Long segment	6 (10)	2 (8)
Total colon	2 (3)	0 (0)
Preoperative decompressive enterostomy	6 (10)	4 (17)
Operation type		
EPT*	12 (20)	0 (0)
EPT* with laparotomy/laparoscopy	44 (75)	23 (96)
Ileoaanal PT with a J-pouch	3 (5)	1 (4)
Re-do PT	1 (2)	1 (4)
ACE	4 (7)	0 (0)
Recurrent enterocolitis	21 (36)	6 (25)
Botox-injections	7 (12)	1 (4)
Myectomy	4 (7)	2 (8)

Data are frequencies (percentage) or medians (IQR).

EPT = endorectal pull-through.

PT = pull-through operation.

* includes procedures with simultaneous colon biopsies through umbilical incision, and procedures with a stoma closure in conjunction.

2. Results

2.1. Patient cohort

Of a total of 126 patients, three had died and 79 (64%) returned completed questionnaires. Of these, 18 had an intellectual disability due to an associated syndrome and 2 had a definitive endostomy, leaving 59 eligible responses (median age 15 (IQR 9–21) years; 73% male) for the assessment of LUTS, and 24 responses from patients ≥ 16 years of age (median age 22 (IQR 18–24) years; 67% male) for the assessment of sexual function and fertility. The baseline characteristics of respondents are summarized in Table 1. The median age of controls ($n = 177$) for the assessment of LUTS was 14 (IQR 9–21; 73% male) years and 22 (IQR 18–24) years for the survey on sexual function ($P = 1.0$ for both vs. patients). Thirty-one out of 59 patients (53%; median age 14 (IQR 9–22)) years also participated in the clinical follow-up.

2.2. Drop-out analysis

There was no significant difference in the main patient characteristics between respondents ($n = 79$) and non-respondents ($n = 44$) in terms of age, gender, and level of aganglionosis ($P = NS$) making a significant selection bias unlikely.

2.3. Lower urinary tract symptoms

As shown in Fig. 1, the overall LUTS profiles of patients and controls were comparable for all symptoms apart from straining to void, which was reported by a higher proportion of controls than patients ($P = 0.008$). In addition, controls tended to have more symptoms of urinary urgency than patients ($P = 0.060$). Overall, 67% of patients ($n = 40/59$) and 80% of controls ($n = 141/177$) reported at least one type of LUTS ($P = 0.062$). However, most symptoms occurred only seldom in both; frequent ($> 1/\text{week}$) LUTS were reported by 14% ($n = 8/59$) of patients and 16% ($n = 28/177$) of controls (Fig. 1; $P = 0.68$) overall. The prevalence of any LUTS $> 1/\text{week}$ was 8% in patients treated with totally transanal EPT ($n = 1/12$), and 15% among patients with laparotomy- or laparoscopy-assisted EPT or IAA ($n = 7/47$, $P = 1.0$). Eighty-three per cent of patients ($n = 49/59$) and 72% of controls ($n = 128/177$) voided

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