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# Impact of rectal dissection technique on primary-school-age outcomes for a British and Irish cohort of children with Hirschsprung disease

Benjamin S.R. Allin[a,b,*], Charles Opondo[a], Timothy Bradnock[c], Simon E. Kenny[d], Jennifer J. Kurinczuk[a], Gregor Walker[c], Marian Knight[a], For and on behalf of the NETS[2HD] collaboration[1]

[a] National Perinatal Epidemiology Unit, Nuffield Department of Population Health, University of Oxford, Old Road Campus, Headington, Oxford OX37LF, UK
[b] Chelsea and Westminster Hospital NHS Foundation Trust, Headley way, Headington, Oxford OX39DU, UK
[c] Royal Hospital for Children, Govan Road, Glasgow, Scotland G514TF, UK
[d] Alder Hey Children's Hospital, University of Liverpool, Eaton Road, Liverpool L122AP, UK

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ABSTRACT

*Background:* This prospective cohort study compared primary-school-aged outcomes between children with Hirschsprung disease (HD) following Soave, Duhamel or Swenson procedures.
*Methods:* Children with histologically proven HD were identified in British/Irish paediatric surgical centers (01/10/2010-30/09/2012). Parent/clinician outcomes were collected when children were 5–8 years old and combined with management/early outcomes data. Propensity score/covariate adjusted multiple-event-Cox and multivariable logistic regression analyses were used.
*Results:* 277 (91%) of 305 children underwent a pull-through (53% Soave, 37% Duhamel, 9% Swenson). Based upon 259 children (94%) with complete operative data, unplanned reoperation rates (95% CI) per-person year of follow-up were 0.11 (0.08–0.13), 0.34 (0.29–0.40) and 1.06 (0.86–1.31) in the Soave/Duhamel/Swenson groups respectively. Adjusted Hazard Ratios for unplanned reoperation compared with the Soave were 1.50 (95% CI 0.66-3.44, $p = 0.335$) and 7.57 (95% CI 3.39-16.93, $p < 0.001$) for the Duhamel/Swenson respectively. Of 217 post-pull-through children with 5–8 year follow-up, 62%, 55%, and 62% in Soave/Duhamel/Swenson groups reported faecal incontinence. In comparison to Soave, Duhamel was associated with lower risk of faecal incontinence (aOR 0.34,95%CI 0.13-0.89,$p = 0.028$). Of 191 children without a stoma, 42%, 59% and 30% in Soave/Duhamel/Swenson groups required assistance to maintain bowel movements; compared to Soave, the Duhamel group were more likely to require assistance (aOR 2.61,95% CI 1.03–6.60,$p = 0.043$).
*Conclusions:* Compared with Soave, Swenson was associated with increased risk of unplanned reoperation, whilst Duhamel was associated with reduced risk of faecal incontinence, but increased risk of constipation at 5–8 years of age. The risk profiles described can be used to inform consent discussions between surgeons and parents.
*Level of evidence:* Level II



## 1. Introduction

Approximately 150 children in the UK are born each year with Hirschsprung disease (HD) [1], a condition where failure of development of the intrinsic intestinal parasympathetic ganglia (aganglionosis), results in functional intestinal obstruction [2,3]. Significant variation exists in the management of children with HD, particularly in relation to the rectal dissection and anastomotic technique that is used during the child's pull-through procedure [4].

In Britain and Ireland there are three main rectal dissection techniques currently in use, the Soave (endorectal), Duhamel (posterior rectal), and Swenson (peri-rectal) techniques (Fig. 1) [4–7]. Little data exists to inform technique selection, and that which does, usually focusses on short-term outcomes reported by studies at levels 4 and 5 of the hierarchy of evidence [8,9–14]. The impact of the operative interventions used to treat HD however is lifelong, and in order to appropriately inform surgical decision making and parental counselling, level 1, 2 and 3 evidence regarding the impact of operative interventions on long-term, patient-centered outcomes is required. The overall aim of this study was therefore to

*Abbreviations:* Hirschsrung's disease, HD.

* Corresponding author at: National Perinatal Epidemiology Unit, Nuffield Department of Population Health, University of Oxford, Old Road Campus, Headington, Oxford OX37LF, UK.
*E-mail address:* Benjamin.allin@npeu.ox.ac.uk (B.S.R. Allin).

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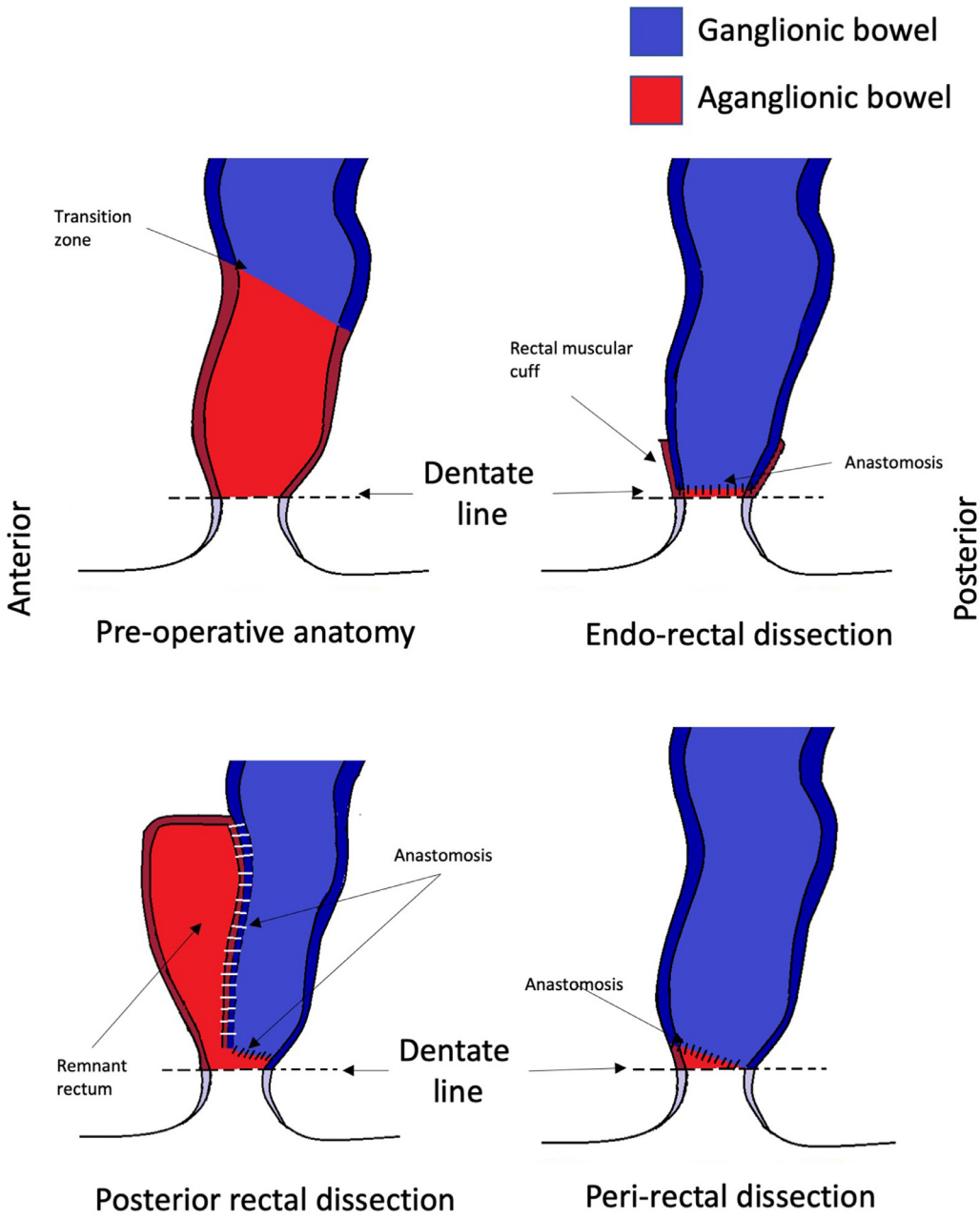


**Fig. 1.** Sagital diagrams of rectal dissection technique.

describe the management of a population-based cohort of children with HD and compare patient-centered core outcomes [15] at five to eight years of age for those who had been treated using the Soave, Duhamel, and Swenson rectal dissection techniques.

## 2. Methods

### *2.1. Ethics committee approval*

Approval for this work was obtained from the South Central (Berkshire) Research Ethics Committee (REC reference: 17/SC/0152), and the Confidentiality Advisory Group of the Health Research Authority (England and Wales) (CAG reference: 17/CAG/0052).

### *2.2. Summary*

Between October 2010 and October 2012, a prospective British and Irish cohort study was conducted using the British Association of Paediatric Surgeons Congenital Anomalies Surveillance System (BAPS-CASS) to collect data relating to the early management, and 28 day and one year outcomes of children diagnosed with HD[1]. Children from 20 of the 28 centers contributing to this cohort were followed-up to primary school age (five to eight years old), with parent and clinician reported outcomes data collected. Primary-school-age outcomes data were linked to the previously collected data. Operative management strategies for these infants were described, and outcomes for children who had been treated using Soave, Duhamel, and Swenson rectal dissection techniques were compared.

### *2.3. Participants*

All infants diagnosed with HD between 01/10/10 and 30/09/12 in one of the 28 paediatric surgical centers in the UK and Ireland, and who were less than six months of age at diagnosis, were eli-

**Box 1: Primary outcome - unplanned reoperation**

- Unplanned was defined as any procedure not considered part of routine post-intervention practice.
- Re-operation included all procedures performed as a direct result of the child's HD, ***and*** all episodes of general anaesthesia that were required as a direct result of the child's HD, regardless of whether an operative intervention was undertaken, e.g. examinations under anaesthesia.
- In the 28-day, one-year and five to eight-year data collection forms, surgeons were asked to report any operations or procedures that the infant had undergone since the last reporting period. These were reviewed against the outcome definition by members of the study steering committee to determine whether they were related to the child's Hirschsprung's disease, and against dates of previously reported procedures to ensure that duplicate procedures were not reported.

**Box 1.** Primary outcome unplanned reoperation.

gible for inclusion in the study. Infants who had not undergone a pull-through utilising one of the three rectal dissection techniques of interest prior to five years of age were excluded from the comparative analysis.

### 2.4. Intervention definition and allocation

Defining each rectal dissection technique is multi-factorial, therefore the most pragmatic method of allocating infants to intervention groups was to ask reporting surgeons to classify the utilized rectal dissection technique as 1) submucosal with formation of a muscle cuff (Soave), 2) posterior rectal (Duhamel), 3) peri-rectal (Swenson), or 4) 'other'. Infants from classifications 1–3 were allocated to the Soave/Duhamel/Swenson groups respectively. As there is no agreed cuff length differentiating the Soave from the Swenson rectal dissection technique, infants were not allocated to intervention groups based upon this measurement.

### 2.5. Outcomes

Outcomes reported are those identified in the recently developed Hirschsprung disease core outcome set (COS)[15].

The primary outcome was unplanned reoperation (Box 1). Secondary outcomes are defined in Box 2. Two scoring systems are included in these outcomes, the pediatric Incontinence and Constipation Score (PICS) and the PedsQL quality of life score. The PICS comprises 13 questions, the answers to which are utilized to produce a constipation score (0–29, 29 = no constipation) and an incontinence score (0–32, 32 = perfect continence) for the child. Age-specific normative values for both the constipation and incontinence scores have been calculated. The PedsQL comprises 23

**Box 2: Secondary outcomes**

1. **Death** with cause classified as due to, 'a complication of treatment (excluding Hirschsprung's Associated Enterocolitis)', 'Hirschsprung's Associated Enterocolitis', an 'associated anomaly', or 'other condition'.
2. **Faecal incontinence**, defined as involuntary passage of faecal matter in an inappropriate place.
3. **Urinary incontinence**, defined as involuntary voiding of urine that was constant, associated with social problems, or requiring catheterisation.
4. **Any stoma** as a direct result of the child's HD
5. **Permanent stoma** as a direct result of the child's HD, including where the decision for a stoma had been made out of child or parental preference, or for continence management. Permanent stoma was defined as one that was created without the intention of later reversal.
6. **Hirschsprung's Associated Enterocolitis,** Clinician decision to admit and treat for Hirschsprung's Associated Enterocolitis.
7. **Objective score of bowel function**, as measured by the Paediatric Incontinence and Constipation Score (PICS).
8. **Use of any assistance (including laxatives) to maintain voluntary bowel movements**
9. **Voluntary bowel movements without need for enemas or rectal or colonic irrigation**.
10. **Quality of life**, as measured by the total scale score for the parent proxy reported PedsQL questionnaire for five to seven year olds.
11. **Psychological stress**, as measured by the psychosocial health summary score for the parent proxy reported PedsQL questionnaire for five to seven year olds.

**Box 2.** Secondary outcomes.

questions in four domains; physical functioning, emotional functioning, social functioning and school functioning. A score from 0 to 100 is produced, with higher scores representing better quality of life. Population normative values are available for reference. In addition to the outcomes identified from the COS, two additional outcomes were also described, 'presence of ***any*** stoma', and 'use of ***any*** assistance (including laxatives) to maintain voluntary bowel movements'.

Outcomes were reported by a combination of paediatric surgical consultants, nonconsultant paediatric surgeons, specialist nurses and research nurses. Some reporters had been involved in the primary management of children for whom they were reporting outcomes, and some had not.

### 2.6. Comparison of pull-through procedures

Soave is the most common technique in the UK [4] and has been taken as the reference procedure with the Duhamel and Swenson techniques compared against it. No direct comparison was made between other techniques.

A multiple-event Cox proportional hazards model was used to investigate the association between rectal dissection technique and unplanned reoperation. Propensity scores predicting a child's likelihood of allocation to the Soave/Duhamel/Swenson technique were calculated using multinomial multivariable logistic regression. Propensity score adjustment is a statistically efficient method of accounting for the impact of potential confounding factors on outcome in the analysis of observational studies [16]. The characteristics used to calculate the propensity scores were ethnicity, gestational age at birth, birthweight, sex, family history of HD, age at presentation, age at diagnosis, associated anomaly or syndrome, preoperative enterocolitis and the site of transition zone. Unadjusted hazard ratios (HR), as well as HRs adjusted for propensity score, weight at pull-through and first attempted approach to the pull-through were calculated (See supplementary material 1 for further details). Logistic regression was used to investigate the association between method of rectal dissection and binary secondary outcomes, with unadjusted Odds Ratios (ORs), and ORs adjusted for propensity score and operative confounders calculated. Complete case analysis was used throughout.

The low number of children who underwent the Swenson technique and had complete five-eight-year data meant that the Swenson technique was only compared to the Soave technique for the primary outcome, unplanned reoperation, as the survival analysis methodology allowed utilization of all data regardless of duration of follow-up. Owing to the high proportion of missing data for quality of life and PICS scores, only descriptive data are presented for these outcomes.

### 2.7. Exploration of subgroup effects

Statistical interactions between rectal dissection technique and key infant characteristics were investigated in a covariate and propensity score adjusted model describing the association between rectal dissection technique and number of unplanned reoperations. This analysis was used to investigate whether the impact of rectal dissection technique on outcome varied according to the characteristics of a child, including, location of transition zone and the presence of additional associated anomalies. This analysis was used to determine whether evidence existed to support the hypothesis that rectal dissection technique should be determined based upon the characteristics of the infant being treated, and therefore, whether techniques should be compared in defined subgroups.

### 2.8. Short segment Hirschsprung disease

As the location of the transition zone is not confirmed prior to beginning the pull-through, a surgeon's rectal dissection technique is usually selected blind to length of aganglionosis. In terms of surgical decision-making and consenting of parents, it is therefore most useful to understand the impact of choice of rectal dissection technique on outcome in a heterogeneous group of infants who mirror clinical practice, as opposed to a subset of infants with a specific length of aganglionosis. However, there is a subgroup of surgeons who vary their rectal dissection technique dependent upon the location of the transition zone identified histologically at pull-through. For these surgeons it is important to understand the relative merits of each rectal dissection technique in subgroups of infants with different lengths of aganglionosis. We therefore determined *a priori*, regardless of the results of the statistical interactions exploring subgroup effects, to conduct a subgroup analysis describing the effect of choice of rectal dissection technique on outcome in those infants with short segment Hirschsprung's disease (rectal or sigmoid transition zone), the most common form of the condition. The low numbers of infants in whom the Swenson technique was utilized prevents their inclusion in this subgroup analysis,

## 3. Results

### 3.1. Loss to follow-up and operative management

The original cohort consisted of 305 children. 279 of these children (91%) underwent a pull-through prior to five years of age, 148 (53%) using the Soave technique, 103 (37%) using the Duhamel technique, and 26 (9%) using the Swenson technique. Rectal dissection technique was unknown for two children (1%). Of the 277 children who underwent a pull-through using the Soave, Duhamel, or Swenson techniques, 259 (94%) had data available relating to the number of unplanned reoperations performed. These children's data were utilized in the primary analysis.

Thirty-six children (24%) who were treated using the Soave technique, 36 children (35%) who were treated using the Duhamel technique and 3 children (12%) who were treated using the Swenson technique had parent follow-up data returned, and 114 children (77%) who were treated using the Soave technique, 86 children (83%) who were treated using the Duhamel technique and 17 children (65%) who were treated using the Swenson techniquehad ***either*** parent ***or*** clinician follow-up data returned at five to eight years of age. Characteristics of the 217 children (78%) with parent or clinician follow-up and the 60 children (22%) who did not have ***any*** follow-up are described in supplementary material 2. The only clinically significant differences between these groups were that those who were lost to follow-up were more likely to have been treated in a low volume center (92% Vs 42%), and less likely to have an additional anomaly or syndrome (15% Vs 22%). Characteristics of the 75 children (27%) with parental follow-up and the 202 (73%) without parental follow-up are described in supplementary material 3. Operative management, loss to follow-up, and the populations in which each outcome are described are detailed in Figs. 2 and 3.

### 3.2. Intervention group characteristics

A greater proportion of children in the Duhamel group had longsegment or total colonic HD than in the other groups. There were also differences between the groups in rates of preoperative stoma formation and use of laparoscopy. Other characteristics did not appear materially different between the groups (Table 1).

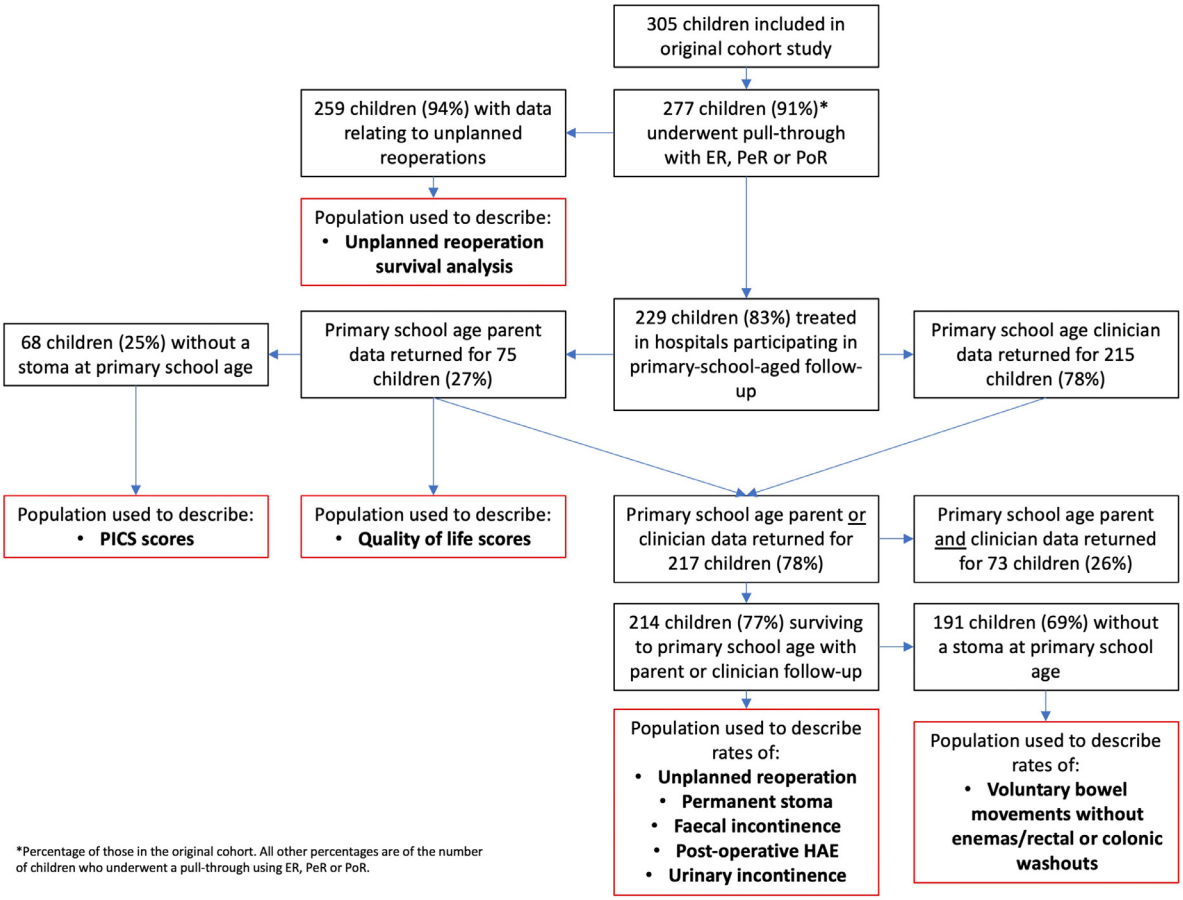


**Fig. 2.** Loss to follow up and analysis population definition.

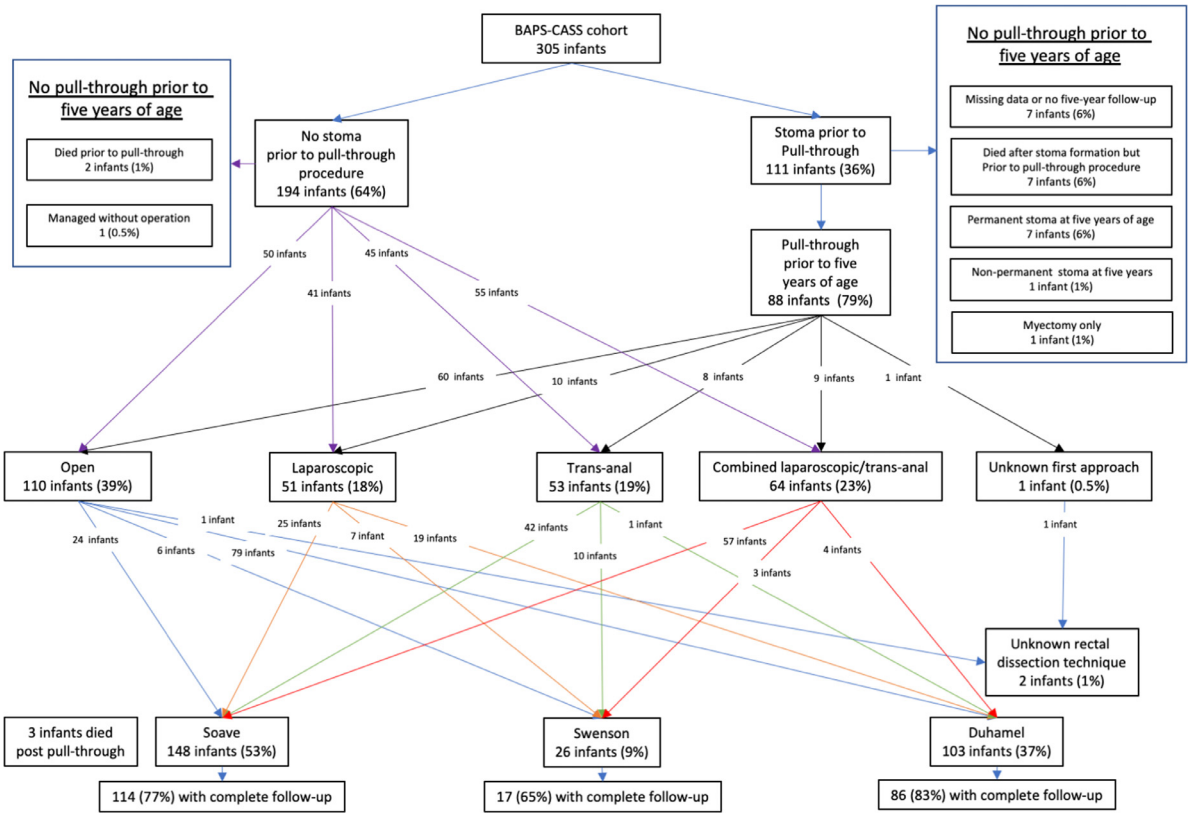


**Fig. 3.** Operative management of infants with Hirschsprung's disease.

**Table 1**
Characteristics of children with five to eight-year follow-up in each rectal dissection group.

| | ER<br>$N = 114$<br>n(%)* | PoR<br>$N = 86$<br>n(%)* | PeR<br>$N = 17$<br>n(%)* |
|---|---|---|---|
| **Low volume center (<median number of cases/year)** | | | |
| No | 66 (57.9%) | 45 (52.3%) | 14 (82.4%) |
| Yes | 48 (42.1%) | 41 (47.7%) | 3 (17.6%) |
| **Ethnicity** | | | |
| White | 101 (89.4%) | 69 (82.1%) | 16 (94.1%) |
| Nonwhite | 12 (10.6%) | 15 (17.9%) | 1 (5.9%) |
| **Gestational age at birth** | | | |
| Term | 99 (86.8%) | 76 (89.4%) | 15 (88.2%) |
| Preterm | 15 (13.2%) | 9 (10.6%) | 2 (11.8%) |
| **Birthweight** | | | |
| 2500 g or more | 96 (87.3%) | 77 (92.8%) | 15 (88.2%) |
| Less than 2500 g | 14 (12.7%) | 6 (7.2%) | 2 (11.8%) |
| **Sex** | | | |
| Male | 87 (76.3%) | 69 (80.2%) | 14 (82.4%) |
| Female | 27 (23.7%) | 17 (19.8%) | 3 (17.6%) |
| **Family history of Hirschsprung's disease** | | | |
| No | 106 (93.0%) | 79 (95.2%) | 17 (100.0%) |
| Yes | 8 (7.0%) | 4 (4.8%) | 0 (0.0%) |
| **Associated anomaly** | | | |
| Isolated HD | 87 (76.3%) | 69 (81.2%) | 13 (76.5%) |
| Syndromic | 18 (15.8%) | 13 (15.3%) | 3 (17.6%) |
| Isolated additional anomaly | 9 (7.9%) | 3 (3.5%) | 1 (5.9%) |
| **Age at presentation (days)** | | | |
| 1–7 days | 100 (87.7%) | 76 (88.4%) | 14 (82.4%) |
| 8–28 days | 2 (1.8%) | 3 (3.5%) | 3 (17.6%) |
| More than 28 days | 12 (10.5%) | 7 (8.1%) | 0 (0.0%) |
| **Age at diagnosis** | | | |
| <31 days | 98 (86.7%) | 74 (86.0%) | 15 (88.2%) |
| 31–60 days | 5 (4.4%) | 5 (5.8%) | 0 (0.0%) |
| 61–90 days | 5 (4.4%) | 4 (4.7%) | 1 (5.9%) |
| 91–120 days | 1 (0.9%) | 2 (2.3%) | 0 (0.0%) |
| >150 days | 4 (3.5%) | 1 (1.2%) | 1 (5.9%) |
| **Transition zone** | | | |
| Rectosigmoid | 92 (82.1%) | 54 (64.3%) | 12 (70.6%) |
| Long segment | 19 (17.0%) | 22 (26.2%) | 5 (29.4%) |
| Total colonic aganglionosis | 1 (0.9%) | 8 (9.5%) | 0 (0.0%) |

ER - Endorectal dissection, PoR - Posterior rectal dissection, PeR - peri–rectal dissection *Percentage of those with complete data.

The median length of muscle cuff in the Soave group was 4.5 cm (IQR 2 cm–6 cm).

### 3.3. Interaction assessment

A statistically significant interaction ($p < 0.05$ on LR testing) was identified between choice of rectal dissection technique and the presence of an associated anomaly. However, the difference was only in magnitude of treatment effect, not direction of effect and therefore this interaction was not clinically significant. No other interactions were identified. As no clinically significant interactions were identified, there was no evidence to support the hypothesis that treatment effect was different in different groups of infants with Hirschsprung's disease. No additional subgroup analyses were therefore undertaken.

### 3.4. Number of unplanned reoperations

In the Soave, Duhamel and Swenson groups, unplanned reoperation rates (95% CI) were respectively, 0.11 (0.08–0.13), 0.34 (0.29–0.40), and 1.06 (0.86–1.31) per person year of follow-up. In both unadjusted and adjusted models, choice of rectal dissection technique was statistically significantly associated with variation in rates of unplanned reoperation, $p < 0.001$ on likelihood ratio testing. Following adjustment for propensity score, weight at surgery and first approach to the pull-through procedure, in comparison to the Soave technique, the Swenson technique was associated with a statistically significantly increased risk of unplanned reoperation, adjusted HR 7.57(95% CI 3.39-16.93, $p < 0.001$). There was no difference in risk of unplanned reoperation between the Soave and Duhamel techniques, adjusted HR 1.5 (95% CI 0.66-3.44, $p = 0.335$) (Table 2). Categories of unplanned reoperations performed are described in Table 3. Examples of minor operations included botox injection and abscess drainage, examples of intermediate operations included antegrade continence enema formation, and incisional hernia repair, and examples of major/complex operations included intestinal resections and stoma formations.

### 3.5. Mortality

Two children (2%) who were treated using the Soave technique, and one (6%) who was treated using the Swenson technique died prior to five years of age. No children in the Duhamel group died prior to five years of age. Causes of death have not been reported as this could allow identification of individual children.

### 3.6. Bowel function

69 children (62%) who were treated using the Soave technique, and 47 children (55%) who were treated using the Duhamel technique reported issues with faecal continence. Following adjustment, children who were treated using the Duhamel technique were statistically significantly less likely to have faecal continence problems at five to eight years of age than those who were treated

**Table 2**
Rates of unplanned reoperation in each rectal dissection group.

| | n (children) | Number of reoperations | Person years | Event rate (95% CI) | Hazard ratio (95% CI) | *p*-value | Adj Hazard ratio (95% CI) | *p*-value |
|---|---|---|---|---|---|---|---|---|
| ER | 138 | 74 | 694.9 | 0.11 (0.08–0.13) | Ref. | Ref. | Ref. | Ref. |
| PoR | 97 | 162 | 474.4 | 0.34 (0.29–0.40) | 2.22 (1.09–4.49) | 0.026 | 1.5 (0.66–3.44) | 0.335 |
| PeR | 24 | 90 | 84.8 | 1.06 (0.86–1.31) | 8.5 (3.49 – 20.76) | <0.001 | 7.57 (3.39–16.93) | <0.001 |

**Table 3**
Detailed Bowel function.

| | ER $N = 69$ | PoR $N = 47$ | PeR $N = 10$ |
|---|---|---|---|
| Severity of faecal incontinence | n(%)* | n(%)* | n(%)* |
| Once or twice per week | 29 (46%) | 21 (47%) | 1 (10%) |
| Every day but without social problems | 18 (29%) | 15 (33%) | 3 (30%) |
| Constant, with social problems | 16 (25%) | 9 (20%) | 6 (60%) |
| | ER $N = 107$ | PoR $N = 74$ | PeR $N = 10$ |
| Assistance required to maintain voluntary bowel movements | n(%)* | n(%)* | n(%)* |
| None | 62 (58%) | 30 (41%) | 6 (67%) |
| Laxatives | 21 (20%) | 31 (42%) | 1 (11%) |
| Enemas | 11 (10%) | 6 (8%) | 1 (11%) |
| Rectal washouts (e.g. Peristeen) | 10 (9%) | 5 (7%) | 1 (11%) |
| Antegrade Continence Enema | 3 (3%) | 2 (3%) | 0 ()%) |
| | ER $N = 112$ | PoR $N = 86$ | PeR $N = 16$ |
| Number of episodes of HAE in the past 12 months | n(%)* | n(%)* | n(%)* |
| None | 108 (96%) | 80 (93%) | 16 (100%) |
| One | 4(4%) | 3 (3%) | 0 (0%) |
| Two or more | 0 (0%) | 3 (3%) | 0(0%) |

ER - Endorectal dissection, PoR - Posterior rectal dissection, PeR - peri-rectal dissection *Percentage of those with complete data.

using the Soave technique, adjusted OR 0.34 (95% CI 0.13-0.89, $p = 0.028$). Severity of faecal continence problems is described in Table 3.

Of those children without a stoma, in the Soave group, 45 out of 107 (42%) required assistance to maintain voluntary bowel movements, whilst in the Duhamel group, 44 out of 74 (59%) required assistance. On both adjusted and unadjusted estimates of effect, children in the Duhamel group were statistically significantly more likely to require assistance to maintain voluntary bowel movements at five to eight years of age than children in the Soave group, unadjusted OR 1.803922 (95% CI 1.01-3.22, $p = 0.046$), adjusted OR 2.61 (95% CI 1.03-6.60, $p = 0.043$). There was however no statistically significant difference between the two groups in the core outcome 'need for enemas or rectal/colonic irrigation to maintain voluntary bowel movements'. Types of assistance required are described in Table 3.

In the Swenson group, 10 children (62%) were incontinent of faeces, and 3 (33%) of the 10 without a stoma required assistance to maintain voluntary bowel movements.

All other outcomes are described in Fig. 4.

### 3.7. Parent reported outcomes

Owing to the low data return rates, no meaningful interpretation could be made for children treated using the Swenson technique, and no comparative analysis could be undertaken between children treated using the Soave technique and children treated using the Duhamel technique. Descriptive quality of life and PICS data for the Soave and Duhamel groups are shown in Table 7.

### 3.8. Short segment Hirschsprung disease(rectosigmoid transition zone)

Of the 279 children who underwent a pull-through procedure, 199 (71%) had a rectosigmoid transition zone, 73 (26%) had a transition zone proximal to the sigmoid colon, and for seven (3%), the transition zone location was not known. 114 infants (57%) with a recto-sigmoid transition zone were treated using the Soave technique, 21 infants (11%) were treated using the Swenson technique, and 63 infants (32%) were treated using the Duhamel technique. Results of the subgroup analysis were not meaningfully different from those of the primary analysis and are therefore described in supplementary material 4.

## 4. Discussion

The key message from this study is that at primary school age, there appear to be differences in core outcomes between rectal dissection techniques. Children who were treated using the Duhamel technique were more likely to be continent of faeces, but also more likely to require assistance to maintain voluntary bowel movements than those who were treated using the Soave technique, whilst those who were treated using the Swenson technique were more likely to undergo unplanned reoperations than those who were treated using the Soave technique. There is no evidence from this study to suggest that these conclusions differ for children of different ethnicities, sexes, or lengths of aganglionosis. Although differences in outcome have been identified between the rectal dissection techniques, all techniques resulted in large numbers of unplanned reoperations, and disappointing faecal continence, urinary continence and bowel evacuation outcomes. It is therefore important that regardless of rectal dissection technique utilized, the pull-through procedure is seen as only part of the management of children with Hirschsprung's disease, and appropriate post-operative support, including bowel management programmes [17] and psychological input are available.

A key strength of this study is that by describing outcomes that have been identified as important to clinicians, people with HD, and parents of children with HD, the results have direct relevance to clinical practice. In order to report these core outcomes however, it was a necessary aspiration to collect data directly from parents, not only from clinicians. Attempting to collect this data led, at least in part, to the main limitation of this study, the loss to follow-up that was experienced. The complex approvals process that collecting parent reported outcomes data entailed made it impossible to launch the study in several sites, and delayed

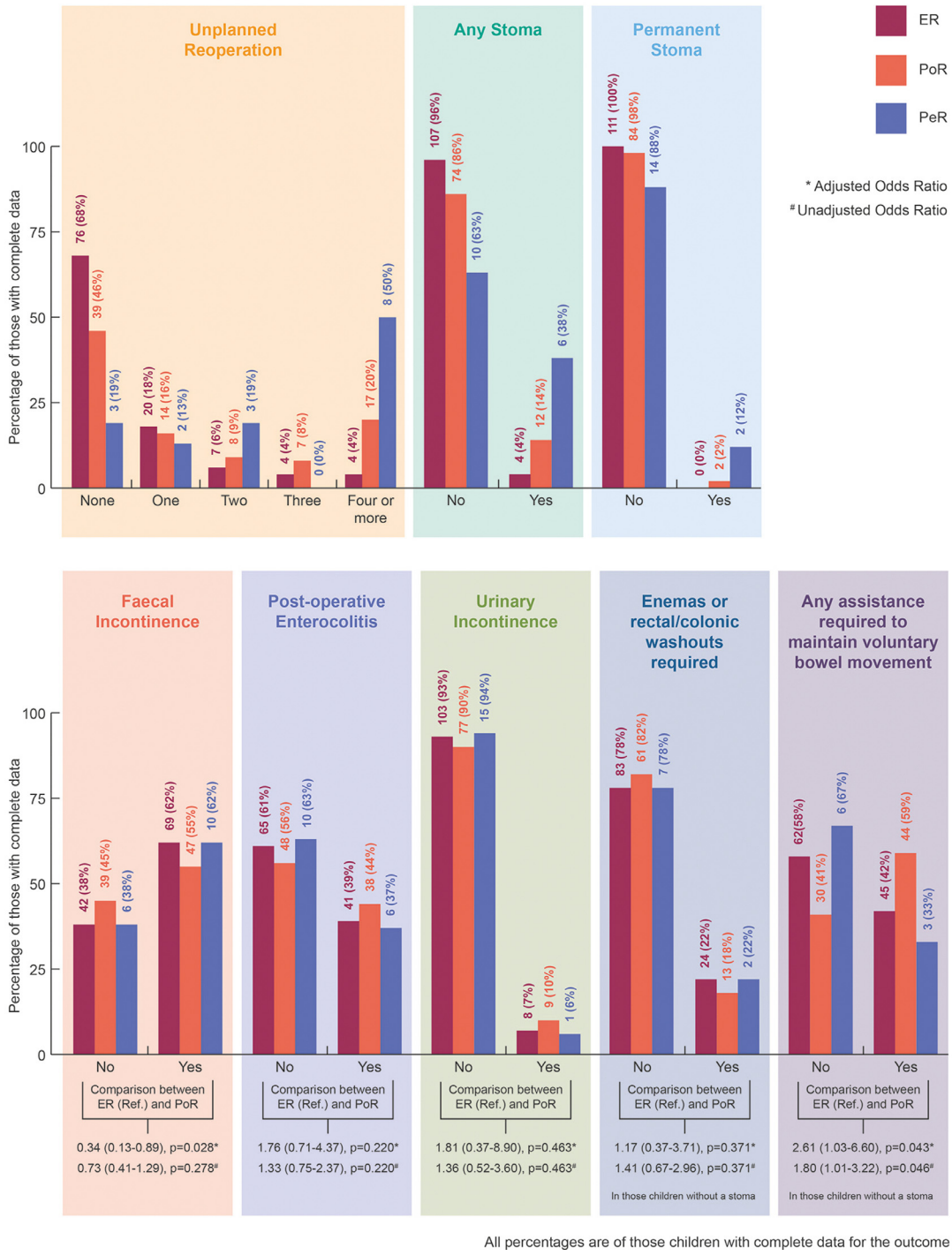


**Fig. 4.** Comparison of outcomes at five to eight years of age.

data collection in others, thereby reducing the population size, and time-period over which data could be collected. Whilst the impact of the reduced data return rate was in part mitigated for the primary outcome through the use of survival analysis methodology, and through the ability to describe many secondary outcomes based on data returned by clinicians, it prevented entirely, secondary clinician reported outcomes for the Swenson group being compared statistically to other groups, prevented comparison of any groups for parent reported outcomes, and prevented meaningful description of any parent reported outcomes for the Swenson group. We acknowledge that this limitation prevents detailed conclusions being drawn in relation to the use of the Swenson technique. It is also unclear what impact the loss to follow-up will have had on the representativeness of the cohort. As the loss to follow-up was slightly greater in the Swenson group than in the Soave and Duhamel groups, if outcomes differed between those with and without follow-up, this may have further impacted the reliability of conclusions drawn about the Swenson technique. However, as the only differences between those with and without follow-up were in the size of center in which they were treated, and whether they had a family history of HD, we do not believe there is evidence to suggest the outcomes would be different be-

tween those with and without follow-up, and therefore are confident that the conclusions of the study have not been materially affected

Prior to this study, the vast majority of data comparing rectal dissection techniques were based upon short-term outcomes reported by small, single institution studies [18–23]. As a result, widely different, and often contradictory conclusions were published [12,13]. Whilst our study is also affected by the limitations inherent to observational studies, for example heterogeneity in the intervention groups, the population-based nature of the cohort and the propensity-score/covariate adjusted analyses that were used, allowed a more robust assessment of the impact of rectal dissection technique on outcome. Importantly, these analyses are designed to account for any impact that between group variation in factors such as the location of the transition zone and presence of associated anomalies had on outcome. The specific possibility of the results being driven by between group variation in the transition zone location has been ruled out through the short segment (rectosigmoid transition zone) subgroup analysis, and the investigation of interactions between rectal dissection technique and location of the transition zone. In the absence of randomized controlled trial data, these data are therefore likely the most reliable currently available.

The results of this study can be used to allow surgeons to have more open, informed discussions of the risks and benefits of their intended rectal dissection technique when consenting parents of children with HD. At present, most surgeons have a preferred rectal dissection technique [4], which they have been taught, practiced, and utilized over a period of many years. Individual surgeons' outcomes are therefore likely, in the short-term, to be best if they continue to utilise their existing preferred technique. We also do not believe that the data presented here are conclusive enough to urge all surgeons to utilise one specific rectal dissection technique. However, we do believe that the data suggest differences in outcome between the techniques, and that these differences are significant enough to warrant discussion with parents at the time of consenting. Until the point where more robust, prospectively collected observational data or data from randomized controlled trials are available to inform evidence-based guidelines for selection of rectal dissection technique, we believe that parents of children with HD should be offered the opportunity to discuss their child's care in a multi-consultant, multi-disciplinary setting, where the pros and cons of different treatment options can be explained, and decisions made based upon a combination of clinical expertise and parental preference.

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## Supplementary materials

Supplementary material associated with this article can be found, in the online version, at doi:10.1016/j.jpedsurg.2022.05.006.

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