

Original Article



Assessment of Quality of Life and Functional Outcomes of Operated Cases of Hirschsprung Disease in a Developing Country

Arun Kumar Loganathan , Aleena Sara Mathew , and Jujju Jacob Kurian

Department of Paediatric Surgery, Christian Medical College and Hospital, Vellore, India

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Correspondence to

Jujju Jacob Kurian

Department of Paediatric Surgery, Christian Medical College and Hospital, Vellore 632 002, Tamilnadu, India.

E-mail: jujjucobkurian@gmail.com

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ORCID iDs

Arun Kumar Loganathan

<https://orcid.org/0000-0001-8671-1425>

Aleena Sara Mathew

<https://orcid.org/0000-0002-9073-3560>

Jujju Jacob Kurian

<https://orcid.org/0000-0002-4937-3524>

Conflict of Interest

The authors have no financial conflicts of interest.

ABSTRACT

Purpose: Children treated for Hirschsprung disease (HD) are adversely affected by fecal incontinence and soiling. This can be detrimental to their physical, psychosocial quality of life (QoL) and impacts the normal functioning of their family. QoL studies in HD are predominantly from developed countries. We measured general quality of life, impact on family and functional bowel status using validated questionnaires in HD children in a developing country.

Methods: Patients with HD, treated in a tertiary paediatric institution in India between 2010 and 2017, were identified. Patients and/or their proxy completed the Pediatric Quality of Life and Family Impact Module questionnaires. Functional outcomes were assessed using Rintala's score.

Results: A 86 children and their parents participated in the study. Majority had rectosigmoid disease (67.4%) and underwent Soave's endoanal pull through (74.4%). A 21% of patients had low Rintala score indicating poor functional bowel outcomes. Only 11% of children had poor QoL scores. Family functioning outcomes were also severely affected in the same subgroup of patients. There was statistically significant correlation between Rintala score and QoL scores (p -value<0.001). Disease severity, type of surgery, and duration of follow-up did not have a statistically significant impact on the QoL.

Conclusion: QoL in children with HD was comparable to the general population. Bowel dysfunction affects a notable number of children and was the most significant determinant of poor QoL.

Keywords: Hirschsprung disease; Quality of life; Fecal incontinence

INTRODUCTION

Hirschsprung disease (HD) is a congenital anomaly characterized by varying levels of aganglionosis, resulting in obstructive symptoms. Though several techniques have been described in the surgical management of HD, competent surgeons, regardless of technique, have achieved good results in the majority of patients [1]. Absence of either incontinence or constipation has been considered as markers of a successful surgery [2]. Though persistence of the symptoms mentioned above has a negative impact on the quality of life (QoL), not

much importance has been given to assessing the child's general QoL, the impact the disease has on his/her social life and the long term effect on parents. This study aims to assess the general and disease-specific QoL in children treated for HD from developing country using existing validated questionnaires.

MATERIALS AND METHODS

The study was a prospective cross-sectional follow-up of a cohort of 86 children who underwent surgical management for HD from January 2010 to December 2017 at Christian Medical College and Hospital, Vellore, Tamil Nadu, India. All children who underwent surgery for HD, including those referred to our institute for definitive surgery following primary diversion or failed attempts elsewhere, were involved in the study.

The study was approved by the Institutional Research Board and the ethics committee (IRB no: 11903 dated 06/03/2019). The clinical details, demographics, type of presentation, level of the anomaly, surgery performed, and postoperative outcomes were collected from electronic medical records. Patients were contacted either in person or over the telephone, and if consenting, the pediatric QoL and family impact module (FIM) questionnaire was administered in their local language (permission was obtained for the translated modules). Functional bowel outcome was determined using the Rintala's fecal incontinence score. Assessment of nutritional status and growth was done by measuring the weight and height and comparing it with the Indian Association of Pediatrics' (IAP) standard weight-for-age and height-for-age percentile chart.

QoL assessment was done by the Pediatric Quality of Life (PedsQL 4.0; Mapi Research Trust, Lyon, France; <https://eprovide.mapi-trust.org> and www.pedsq.org) generic core scales questionnaire. PedsQL, a 23-item measurement model for health-related QoL in children aged 2–18 years, was applied in child self-report (8 years and above) and parent proxy-report (2 years and above) versions to assess physical, emotional and social QoL along with school functioning for the previous four weeks. As the guidelines for the administration of PedsQL recommend, where applicable, children and parents completed the questionnaire independently. The PedsQL gave three separate scores: A total scale score, a physical health summary score, and a psychosocial health summary score.

The impact on family functioning was assessed using the PedsQL 2.0 FIM questionnaire. Eight items for determining the impact of the disease on the daily activities of family members and family relationships over the previous four weeks, were administered to the caregiver. Mean scores were calculated based on a 5-point response scale for each item and transformed to a 0–100 scale, with a higher score representing better QoL and minimal impact on family function. Data was also collected from parents on whether the child was receiving any bowel management program. A Rintala's score calculated from this data was used to determine the functional bowel outcome.

Descriptive measures such as mean with SD or median with IQR were used to present continuous variables, whereas frequencies and percentages were used for categorical variables. The normality assumption of outcome variables such as QoL and Rintala was assessed by plotting a histogram. QoL, FIM, and Rintala's scores were compared across all the categorical variables using the Man Whitney U-test for dichotomous variables and

the Kruskal-Wallis test for variables with more than two categories. The analysis was done using IBM SPSS Statistics for Windows, Version 21.0 (IBM Co., Armonk, NY, USA), and p -value < 0.05 was considered statistically significant.

RESULTS

Patient demographics

Of the 192 patients operated for HD from 2010 to 2017, we could contact 86 patients (44.8%) who consented to participate in the study. Data on patient demographics, presenting complaints, length of the aganglionic segment, operative technique, need for diversion and redo surgeries were obtained from the electronic medical records and have been summarised in **Table 1**. The mean age at follow-up was 7 years (standard deviation [SD], 3.12 years) and the mean follow-up was 4.7 years (SD, 2.36 years). Males predominated at 82.6% (71/86). The majority of patients in our cohort had classical disease (67%) and underwent Soave's endoanal pull through (75%). Four patients required a redo pull-through procedure due to the failure of the primary procedure. Seven patients had postoperative enterocolitis that necessitated readmission. Three patients had a prolapse of the pulled down bowel, with two of them requiring anal mucosal trimming and the other settling over time. Three patients had cuff abscess, which was managed conservatively.

Nutritional status

The present weight and height data of all patients were plotted on the IAP growth chart. The height and weight for age were normal in 60% of the patients (>50th centile). Severe stunting was seen in 3% of children (<3rd centile) (**Fig. 1**). All the children with stunting had long segment disease.

Functional bowel outcomes

Eighty-four of the 86 parents (97.7%) reported that their child felt the urge to defecate and was able to sense the coming of a bowel movement. Fifty patients (58.1%) had regular bowel movements defined as passing stools every other day to twice a day. There were thirteen patients (15.1%) who had constipation requiring dietary management or laxatives,

Table 1. Patient characteristics of Hirschsprung disease cohort

| Characteristic | Value (n=86) |
|------------------------|------------------|
| Age surgery | 1 yr (6 mo–2 yr) |
| Age follow-up (yr) | 6 (5–9) |
| Sex (male/female) | 71/15 |
| Presenting complaints | |
| Delayed meconium | 33 (38.4) |
| Intestinal obstruction | 17 (19.8) |
| Chronic constipation | 36 (41.9) |
| Disease severity | |
| Classical | 58 (67.4) |
| Long segment | 21 (24.4) |
| Total colonic | 7 (8.1) |
| Type of surgery | |
| Soave | 64 (74.4) |
| Duhamel | 7 (8.1) |
| Laparoscopic | 11 (12.8) |
| Redo surgeries | 4 (4.7) |
| Initial diversion | 29 (33.7) |

Values are presented as median (interquartile range), number only, or number (%).

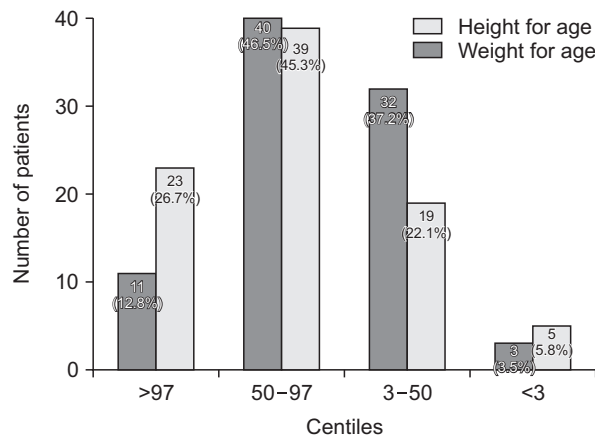


Fig. 1. Nutritional assessment. Bar diagram showing the nutritional status of the participants according to Indian association of pediatrics growth chart percentiles.

Table 2. Characteristics of the children requiring regular rectal washes

| Characteristic | Value (n=23) |
|--------------------|--------------|
| Disease severity | |
| Classical | 17 (73.9) |
| Long segment | 5 (21.7) |
| Total colonic | 3 (13.0) |
| Type of surgery | |
| Soave | 20 (87.0) |
| Duhamel | 0 (0.0) |
| Laparoscopic | 2 (8.7) |
| Redo surgeries | 3 (13.0) |
| Initial diversion | 18 (78.3) |
| Follow-up duration | 5.3 (2.1) |

Values are presented as number (%) or mean (standard deviation).

and twenty-three patients (26.7%) requiring regular saline washes or enemas for fecal incontinence. Patient characteristics of children requiring washes have been summarised in **Table 2**. While there were fifty-six parents (65.1%) who reported no bowel accidents, fifteen parents (17.4%) reported less than one accident per week, and 15 parents (17.4%) reported more than one bowel accidents per week with five of them having daily accidents. Eighteen patients (20.9%) had significantly impaired Rintala scores denoting poor functional bowel outcomes. The urinary function was not affected in any of the patients.

Quality of life outcomes

Age-appropriate PedsQL 4.0 generic core scales questionnaire for QoL assessment was administered to all patients. While parent proxy scores were made available for children up to seven years of age, children eight years and above had self-reported and parent proxy scores. The mean physical, psychosocial, and total score for each age group has been depicted in **Table 3**. It was seen that the parents had a tendency to give a higher score when compared to children but was found not statistically significant. Physical and psychosocial components showed similar scores within each age group. There was an improvement in the QoL scores as the children got older but were not statistically significant (p -value, 0.070). For further comparison, QoL scores were divided into three categories as poor (QoL < 75%), moderate (QoL 75–90%), and good (QoL > 90) (**Fig. 2**). Around 11.6% of patients were found to have poor QoL scores, while 68.6% had a QoL at par with normal healthy children.

Table 3. PedsQL 4.0 Generic Core Scale scores (physical, psychosocial and total) for child self report and parent proxy report across different age groups

| Age | Parameter | Child | Parents |
|----------------|--------------|-------------|-------------|
| 2-4 yr (n=20) | Physical | | 88.75±13.12 |
| | Psychosocial | | 88.06±15.74 |
| | Total | | 88.40±13.78 |
| 5-7 yr (n=32) | Physical | | 84.18±16.23 |
| | Psychosocial | | 83.07±18.63 |
| | Total | | 83.63±16.86 |
| 8-12 yr (n=27) | Physical | 90.63±9.69 | 92.36±8.51 |
| | Psychosocial | 89.09±11.09 | 90.88±9.35 |
| | Total | 89.86±10.05 | 91.62±8.76 |
| 13-18 yr (n=7) | Physical | 95.98±2.97 | 97.32±2.16 |
| | Psychosocial | 94.52±4.78 | 96.19±3.15 |
| | Total | 95.25±3.53 | 96.76±2.61 |

Values are presented as mean±standard deviation.
PedsQL: Pediatric Quality of Life.

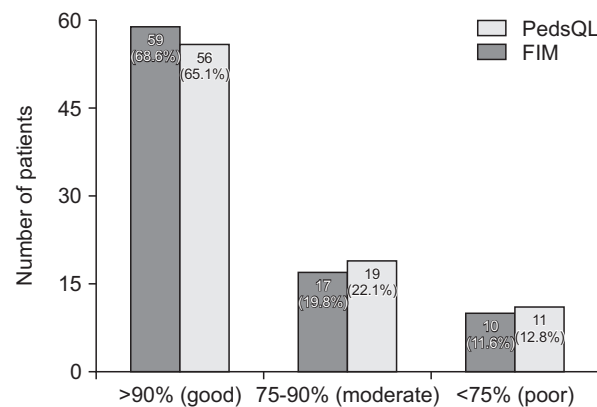


Fig. 2. Categorization of PedsQL scores and family functioning scores of the participants.
PedsQL: Pediatric Quality of Life, FIM: family impact module.

The influence of various factors such as disease severity, type of surgery, need for bowel management programs, and duration of follow-up on the QoL was assessed. It was found that the impact of the factors mentioned above on QoL scores was not statistically significant except for the need for rectal washes. Children who required regular washes were found to have a significantly impaired QoL (**Table 4**). A scatter plot comparing the Rintala score and QoL score found a statistically significant correlation between the two (p -value<0.001) (**Fig. 3**).

Family functioning outcomes

The impact that the child's bowel problems have on the caregiver regarding their relationship with other family members, social life, and job were assessed using the FIM questionnaire. The results have been summarised in **Fig. 2**. Fifty-six parents (65.1%) reported that their child's surgery did not affect their daily activities, ability to go to work and socialize with other family members or friends. However, nineteen parents (22.1%) felt that their social life was affected but daily activities were manageable and eleven parents (12.8%) felt both their daily activities and social life were severely affected by their child's condition. This poor family functioning outcome was found to correlate with children having poor QoL and Rintala scores.

Quality of Life in Children Operated for Hirschsprung Disease

Table 4. Comparison of various factors influencing PedsQL scores and Rintala scores

| | PedsQL | | | Rintala | | |
|----------------------|---------------|-----------------------|---------|--------------|----------------------|---------|
| | Mean (SD) | Median (IQR) | p-value | Mean (SD) | Median (IQR) | p-value |
| Disease severity | | | 0.272 | | | 0.404 |
| Classical | 89.13 (13.73) | 95.58 (79.69, 98.44) | | 15.97 (4.40) | 18.00 (13.00, 20.00) | |
| Long segment | 87.93 (12.60) | 92.09 (85.32, 95.11) | | 15.71 (3.80) | 17.00 (14.00, 18.00) | |
| Total colonic | 82.79 (17.22) | 93.44 (61.39, 95.94) | | 14.29 (4.57) | 16.00 (10.00, 18.00) | |
| Type of surgery | | | 0.075 | | | 0.136 |
| Soave | 87.26 (14.12) | 93.16 (78.96, 97.61) | | 15.39 (4.36) | 17.00 (12.00, 19.50) | |
| Duhamel | 96.91 (3.60) | 99.17 (92.09, 100.00) | | 19.00 (1.53) | 20.00 (18.00, 20.00) | |
| Laparoscopic | 90.27 (15.62) | 96.78 (91.29, 99.17) | | 15.91 (4.78) | 18.00 (15.00, 20.00) | |
| Redo surgeries | 84.79 (8.41) | 85.32 (77.56, 92.03) | | 15.75 (1.89) | 16.50 (14.50, 17.00) | |
| Follow-up | | | 0.945 | | | 0.156 |
| <5 yr | 89.34 (11.41) | 93.65 (78.96, 97.61) | | 16.44 (3.97) | 18.00 (15.00, 20.00) | |
| >5 yr | 87.20 (15.92) | 93.44 (79.69, 98.34) | | 15.02 (4.47) | 16.00 (11.00, 20.00) | |
| Bowel management | | | <0.001 | | | <0.001 |
| No intervention | 95.95 (4.72) | 97.61 (93.65, 99.17) | | 18.40 (2.03) | 19.00 (17.00, 20.00) | |
| Minimal intervention | 89.16 (6.67) | 92.09 (85.32, 93.44) | | 15.46 (3.04) | 15.00 (14.00, 17.00) | |
| Enemas/washes | 71.25 (14.63) | 75.39 (61.39, 78.65) | | 10.22 (2.75) | 10.00 (8.00, 13.00) | |

PedsQL: Pediatric Quality of Life, SD: standard deviation, IQR: interquartile range.

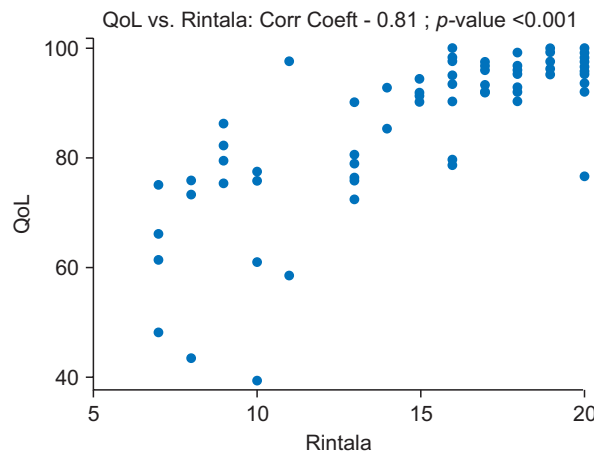


Fig. 3. Scatter plot comparing PedsQL score and Rintala score, showing significant correlation with correlation coefficient of 0.81 (p-value<0.001). PedsQL: Pediatric Quality of Life, QoL: quality of life.

DISCUSSION

Surgical management in HD has been directed towards ensuring that the child has regular and spontaneous bowel movements without incontinence or soiling. Several studies have looked into the QoL in children following HD surgery. However, many of these studies have had their limitations. Focussing only on the bowel dysfunction related QoL without measuring the physical and psychosocial domains was found to give a spuriously low QoL. This was because although these children had bowel dysfunction, most of them were well adjusted, especially on growing older, resulting in a good general QoL [3,4]. Further limitations include using a non-validated questionnaire, small sample size, or using mixed populations with similar issues like anorectal malformation in calculating the QoL [5]. Another issue of concern was these studies were performed in the developed world, it's application in a developing country was unclear. Developed economies have ancillary services that play a major role in taking care of these patients as they progress in life. These services are woefully short in low and middle-income economies which in theory should translate to a low QoL.

We have, in our study from a low to middle income economic stata, attempted to measure the physical, psychosocial QoL along with the severity of bowel dysfunction and the effect this illness has on the general wellbeing of the family.

Studies have mentioned a poorer QoL in children with HD when compared to the general population [6-8]. In our study, we found the majority of the children were capable of routine physical activity, academic activities, and good social relationships with their friends, resulting in good physical and psychosocial QoL. This was in concordance with other recent studies using the PedsQL 4.0 generic core scales questionnaire [5,9]. It was also seen that the parents tend to give a higher score than children, as observed by Hartman et al. [10] in their study. This confirms that parents' responses alone cannot replace the children's while assessing children's QoL and that a combination of both would be ideal.

Several factors, including age at surgery, sex, type of surgery performed, level of aganglionosis, initial stoma diversion, enterocolitis episodes, and bowel dysfunction were studied for a possible effect on the QoL. None of the factors other than bowel dysfunction was seen to impact the QoL. This was in concordance with other studies except for the level of aganglionosis affecting QoL [5,9,11]. Children with a long-segment disease or total colonic aganglionosis are expected to have frequency and soiling due to the complete or near absence of the large bowel. Studies by Ludman [12] and Moore et al. [13] have found that children with total colonic aganglionosis were less well-adjusted than their matched rectosigmoid disease pairs. Our cohort showed no difference in QoL based on the level of disease. However, we could not definitively conclude that the level of aganglionosis had no bearing on the QoL as our cohort of long-segment and total colonic aganglionosis patients were small.

The reported prevalence of fecal soiling in children post HD surgery ranges from 9.8 to 37.8% [14-16]. Our study showed similar findings, with 17% having considerable soiling. However, only two of our patients had a lack of voluntary control. This perhaps demonstrates the cause of soiling to be a result of constipation with overflow incontinence rather than true incontinence. Studies have shown worse bowel function scores to be a predictor for poor QoL, as seen in a small subset of our patients with severe bowel dysfunction [17,18]. These patients had incontinence and soiling, causing embarrassment and social ostracization due to the emanating foul odor. The resultant social withdrawal contributed negatively to their psychosocial development. However, not all incontinent patients had uniformly poor outcomes, as children on regular washes and those with impaired bowel control were found to have an acceptable QoL. We feel this was where parents play an important role in our population. Children with a poorer family function score grading had a lower QoL compared to those with a better family function score whose perceived QoL was better despite having similar symptoms. Parental perception of the disease and their response to the same play a major role in influencing the level of self-esteem that the child has, which becomes a major factor in the perceived QoL [19,20].

The phenomenon of 'response shift' has also been attributed as another factor which influences the perceived QoL in people with chronic problems [21]. These are adaptive responses by which a person adapts to what can be considered as 'normal' for him or her. An example of the same could be obtained from the study by Satoshi et al. which showed adults operated for HD leading a productive life irrespective of their bowel dysfunction status [22]. This aspect was seen in our older children who were found to have a better QoL compared to their younger counterparts. While this study provides a cross-sectional evaluation of the QoL

at a particular point in the lives of these children, longitudinal studies over a period of time will help assess whether adaptive responses help improve QoL as they get older.

Limitations of the study

There was a possibility of selection bias, as the patients recruited were mostly children still on follow-up and thus more likely to be symptomatic. Hence there could be a skew in the study towards those with worse bowel function. Our cohort predominantly included children who underwent Soaves procedure for recto-sigmoid disease, which underpowered comparison studies with other procedures and different levels of disease. Finally, our study was only a snapshot of the current status in the child's life, and long term outcomes could not be determined.

In conclusion, the majority of children with HD have a good QoL compared to the general population, even in the middle to low-income economic strata. They were also found to have a higher prevalence of bowel dysfunction when compared to the general population. In severe cases, this was seen to result in a low physical and psychosocial QoL. Longitudinal, multi-center studies with validated outcome measures would help in providing accurate information on long term prognosis, thus creating targeted care pathways leading to a better QoL in these children.

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