



Long-term health-related quality of life of patients with pediatric onset intestinal failure



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ABSTRACT

Background: Despite improved survival rates of patients with pediatric intestinal failure (IF), data on health-related quality of life (HRQoL) of IF patients are still scarce. We hypothesized that I) continued parenteral nutrition, underlying intestinal motility disorder, abdominal pain and problematic bowel function would be associated with poorer HRQoL and higher parental stress levels, and II) the time intervals since the latest bowel operation, the latest episode of sepsis, and the latest inpatient care episode would be associated with better HRQoL and lower parental stress.

Methods: Patients with pediatric onset IF and their parents answered questionnaires on HRQoL, parental stress, and bowel-related symptoms. Clinical data were gathered by chart review. Controls matched for age and sex were randomly chosen by the Population Register Centre of Finland.

Results: Thirty-six (73%) IF patients participated at a median age of 9 years. Overall HRQoL was similar to healthy peers, and frequent abdominal pain was the only factor associated with poorer HRQoL. Abdominal pain and stool frequency >3 times per day were associated with higher levels of parental stress, whereas longer time intervals since the latest bowel operation or hospitalization were associated with lower parental stress levels.

Conclusion: Long-term HRQoL of pediatric IF patients is comparable to that of healthy peers. Time often eases parental stress, but frequent abdominal pain presents a challenge to the well-being of some patients and requires medical attention.

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Pediatric intestinal failure (IF) encompasses any intestinal disease requiring long-term parenteral nutrition (PN) for sustainment of sufficient nutrition, growth, and survival [1]. The two most common patient groups are short bowel syndrome (SBS) as a consequence of massive bowel resection and intestinal motility disorders [1–3]. Due to improved PN administration, enteral nutrition and surgical care, efficient infection control, and enhanced cooperation between pediatric and surgical teams, current patient survival rates for IF patients exceed 90% [1,3–8]. Thus long-term health-related quality of life (HRQoL) is emerging as a new focus of interest.

Data on HRQoL of pediatric IF patients are still scarce. Previous studies suggest HRQoL is lower than in healthy peers but the reasons for this difference remain unknown [9,10]. Possible causes discussed in earlier studies include increased frequency of stools, insecurity of adolescents, and parents' concerns about their child's current and future health. Since these issues weren't actually measured, however, discussion remains speculative. Outsiders can never precisely appreciate the value each individual gives to their unique life, as the whole concept of HRQoL refers to an individual's subjective perception of his/her

Abbreviations: ALT, alanine transaminase; BMI, body mass index; EN, enteral nutrition; HRQoL, health related quality of life; IF, intestinal failure; PN, parenteral nutrition; QRS, questionnaire for resources and stress; SBS, short bowel syndrome.

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situation. Self-assessments are consequently the preferred type of measurement, and the difference in HRQoL of patient and proxy answers is a well-known phenomenon [9,11].

We were interested in testing the hypotheses proposed by previous studies on HRQoL of IF patients. We also wanted to find other possible factors to elucidate the differences in HRQoL. Previously, HRQoL was inversely associated with disease activity in children suffering from inflammatory bowel disease [12]. After pediatric liver transplantation, we identified biliary complications and reoperations as factors associated with decreased HRQoL, and longer follow-up after liver transplantation as a factor associated with better HRQoL [11]. We therefore formulated the following hypotheses: I) continued parenteral nutrition (PN), underlying intestinal motility disorder, abdominal pain and problematic bowel function would be associated with poorer HRQoL and higher parental stress levels, and II) the time periods elapsed since weaning off PN, the latest bowel operation, the latest episode of sepsis, and the latest in-patient care episode would be associated with better HRQoL and lower parental stress.

1. Methods

1.1. Patients and controls

Between January 1984 and August 2010, 56 patients with pediatric onset IF were treated in our IF rehabilitation program, and 52 of them

are alive. IF was defined as over 50% resection of the small bowel or duration of PN over 30 days [13–15]. Eligible patients (age 1 year or older, $n = 49$) were invited to participate in this cross-sectional study.

Clinical data were gathered from patient records, including initial and current diagnoses, gestational age and weight, duration of PN and subsequent enteral nutrition, number of weekly PN infusions and percentage of energy intake from PN, absolute and percentage of predicted age-adjusted lengths of small intestine and colon [16], presence and type of current ostomies, number of blood culture positive septic episodes from birth to study date and time elapsed since the last septic episode, time since the latest surgical operation or hospitalization, and continuous use of antibiotics. Plasma levels of alanine aminotransferase (ALT) and bilirubin one month preceding the cross-sectional study were collected.

A control group of 400 healthy persons matched for age, sex, and municipalities of residence was randomly picked by the Finnish Population Register Centre. Questionnaires were mailed to controls and reminders were sent once to non-respondents.

1.2. Measures

After informed consent, children 7–18 years of age ($n = 13$) and all parents ($n = 30$) filled the PedsQL 4.0 questionnaires in 2010–2011. Investigators uninvolved in patient care delivered the questionnaires to patients and/or their parents by mail prior to a routine control at our hospital and face to face at the control visit if the questionnaire was unanswered, and handled the data.

PedsQL 4.0 is a generic, validated questionnaire with patient and proxy forms for children >7 and proxy forms for children <7 years [17,18]. The PedsQL consists of 23 items that are divided into four dimensions: physical, emotional, social, and school functioning. A total score of all dimensions was also calculated. In addition, emotional, social, and school functioning dimensions comprise the psychosocial functioning score. For all dimensions, higher PedsQL scores correspond with better HRQoL (maximum 100).

Parents of children <18 years also filled the Questionnaire on Resources and Stress for families with chronically ill or handicapped members (QRS) in its shortened version [19]. The QRS includes 35 true/false statements divided into seven parts: negative attitude toward index case, overcommitment/martyrdom, pessimism, lack of family integration, physical incapacitation, financial problems, and social obtrusiveness. Higher scores indicate higher levels of stress. Parents were also asked to evaluate the following four items using a visual analogue scale (scores 1–7): How worried were you at the time of your child's IF diagnosis? How worried are you currently due to your child's IF? How stressed do you feel currently due to your child's IF? How worried are you about your other children's health and everyday life?

Patients 16 years or older ($n = 8$) filled the Gastrointestinal Quality of Life Inventory (GIQLI), including 36 questions on core symptoms, physical items, psychological items, social items, and disease specific items, each with five response categories (scale 0 to 4, with highest scores indicating most desirable options). The GIQLI score was calculated as a sum of the points [20].

Patients and controls were requested information on height, weight, chronic diseases, education level and the age of classmates, and parents' level of education and employment status. We also asked about bowel function and gastrointestinal symptoms including stool frequency, stool consistency, flatus frequency, frequency of swelling, and frequency of abdominal pain.

Body mass index (BMI; weight [kg]/height [m²]) was calculated. To enable comparisons among individuals of different ages, BMI of patients and controls ages 2 to 18 was adjusted for age according to recent Finnish reference values (BMI-for-age; ISO-BMI) [21]. Subjects were then classified as underweight (BMI <19 kg/m²), normal weight (BMI 19–25 kg/m²), and overweight/obese (BMI >25 kg/m²).

1.3. Statistical analyses

Unless otherwise stated, data are presented as median (range) due to the small group size and uneven distribution of data. The Mann–Whitney U test was used for continuous variables. Frequencies were compared with Fisher's exact test because of the small group size. For ordinal variables of bowel function, gastrointestinal symptoms, and HRQoL measurements, differences between patients and controls were tested using the Wilcoxon matched pair test. Correlations were calculated using the Spearman rho two-tailed test. Statistical significance was set at 0.05.

1.4. Ethical consideration

The ethics committee of the Helsinki University Central Hospital approved the study protocol.

2. Results

The participation rate for patients was 73% (36 of 49) and 16% for controls (63 of 400). Demographic data of the patients and controls are presented in Table 1. Gender distribution and age were similar between patients and controls, but patients had lower BMI-for-age distribution than did controls.

Age, gender distribution, gestational age and weight, BMI-for-age, duration of PN, time after weaning off PN, bowel anatomy, time since the last septic episode, time since the last surgical operation, and time since the last episode of in-patient care were similar between participants and non-participants, making significant selection bias unlikely ($P > .05$ for all, data not shown).

2.1. Clinical characteristics

Causes of IF included SBS and intestinal motility disorders (Table 2). Nineteen patients (53%) were born preterm, and median gestational age was 35 weeks (range, 24–40). Nine patients (25%) had undergone total colectomy. Seven patients (19%), including six patients with extensive Hirschsprung's disease and one patient with chronic intestinal pseudo-obstruction (CIPO), had an ileostomy at the time of study. In addition to IF, 12 patients (33%) had one or several other chronic conditions: asthma or bronchopulmonary dysplasia (5), cartilage-hair hypoplasia (3), cerebral palsy (2), grade 3–4 intraventricular hemorrhage (2), and 1 each had Haddad syndrome, VATER association, and marked congenital visual impairment.

2.2. Treatment characteristics

Patients were on PN for a median of 310 days (21–7600). Eight patients (22%), of whom 6 were under 7 years, continued receiving PN after 72 PN months (21–170) in median 6 PN infusions per week (range 2–7). Only one patient was on total PN, while 3 patients acquired half of their energy intake from PN, and 4 patients only required PN to

Table 1
Demographics of 36 intestinal failure patients and 63 controls.

	Patients	Controls
Boys, n (%)	23 (64)	38 (60)
Age, y, median (range)	9 (1–27)	8 (1–32)
BMI-for-age, median (range)	20 (12–31)*	21 (14–35)
BMI categories, n (%)		
Underweight	10 (28)	3 (5)
Normal weight	15 (42)	45 (71)
Overweight/obese	4 (11)	11 (18)
Under 2 years	5 (14)	3 (5)
Missing data	2 (6)	1 (2)

* $P = .01$ between patients and controls.

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