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ORIGINAL ARTICLE

# Self-reported assessment of health-related quality of life in children who underwent restorative proctocolectomy with ileal J-pouch anal anastomosis for ulcerative colitis

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**Abstract**
*Purpose* To clarify health-related quality of life (HRQOL) by self-evaluation after restorative proctocolectomy with ileal J-pouch anal anastomosis (IPAA) in children with ulcerative colitis, a questionnaire using the Pediatric Quality of Life Inventory™ 4.0 (PedsQL) was administered.
*Methods* The PedsQL was administered to 13 consecutive children (mean age 14.5 years) who underwent IPAA between 2005 and 2010 in our hospital and age-matched healthy controls. The mean duration after IPAA was 2.5 years (range 0.08–6 years) at the time of this study. Healthy children completed the same questionnaire by retrospective imaging during the past 1 month by the PedsQL evaluation policy.
*Results* Patients' total score and each functioning score after IPAA reached the same levels as those in healthy controls. Soiling, pouchitis occurrence, and bowel movements had no significant relationship to the PedsQL total score and each functioning score.
*Conclusions* Interference of physical activity, emotional status, and social life caused by refractory ulcerative colitis (UC) worsens patients' HRQOL. IPAA could resolve these problems in children with UC and result in an HRQOL comparable with that in healthy children.

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## Introduction

Inflammatory bowel disease (IBD) has recently been increasing in children worldwide [1, 2]. Pediatric ulcerative colitis (UC) is characterized by extensive intestinal involvement and rapid early progression; moreover, the colectomy rate is high in pediatric compared with adult patients [3, 4]. Pediatric IBD sometimes becomes chronic and refractory, with repeated relapses for a large portion of the patient's life. It can affect children in terms of physical, psychological, and social functioning, which are all included in the World Health Organization's Definition of Health [5, 6]. There are many issues common to all chronic illnesses that affect health-related quality of life (HRQOL) in children with IBD: school truancy, social isolation, and psychiatric problems such as anxiety, depression, and antisocial and dependent behavior [7]. Familial stress such as parental expectations, financial demands, and marital stressors are also sometimes significant [7, 8].

Several measures for HRQOL in children have been developed. However, these measures sometimes require parents' proxy assessments because of the child's age and mental development, so their reliability is low in terms of evaluation of children's HRQOL. Therefore, the development of a suitable, internationally reliable measure of HRQOL in children by self-reporting was needed. In 2001, Varni et al. [9] invented the Pediatric Quality of Life Inventory™ 4.0 (PedsQL), which is an HRQOL scale including self-reporting and proxy assessment adapted to children and adolescents 2–18 years old [9]. It has been translated to many languages. The Japanese version of the PedsQL, which was translated by Kobayashi, has a

confirmed high reliability and validity [10, 11]. Janssens [12] demonstrated that the PedsQL is a suitable measure of HRQOL in children, covers a large age range, has good psychometric properties, and covers the content of the International Classification of Functioning, Disability, and Health of the World Health Organization [13].

A previous study that evaluated HRQOL by the PedsQL for pediatric patients with IBD revealed that IBD may be detrimental to HRQOL on the physical and school functioning scales [14, 15]. The PedsQL measured HRQOL well in children after surgery such as appendectomy [16] and kidney and liver transplantation [17, 18]. Restorative proctocolectomy with ileal J-pouch anal anastomosis (IPAA) is now considered the standard surgical procedure for UC in both adults and children [19]. There are several papers about postoperative complications and functional results after surgery in children with UC; however, these researchers had no choice but to use a QOL scale for adults or original questionnaires without international validity or reliability in children because they are not a standard evaluation scale for own HRQOL by self-reporting [20–24].

The aim of this study was to investigate HRQOL after IPAA in pediatric patients with UC. A questionnaire using the Japanese translated version of the PedsQL was administered.

## Patients and controls

In this study, we enrolled 13 consecutive pediatric patients who underwent IPAA for refractory moderate or severe UC between 2005 and 2010 in our hospital. All IPAA were performed by a hand-sewn procedure.

The patient group comprised six girls and seven boys with a mean age of 14.5 years (range 9–18 years) at the time of this study. The mean duration after IPAA was 2.5 years (range 0.08–6 years) at the time of this study. Sixty-five healthy children were enrolled as controls. The control group comprised 29 girls and 36 boys with a mean age of 15.0 years (range 10–18) at the time of this study.

Table 1 demonstrates the patients' characteristics. The patients' mean age was 10.4 years (range 5–15 years) at disease onset, 12.8 years (range 8–17 years) at operation, and 14.5 years (range 9–18 years) at the time of this study. The diagnosis of UC was based on clinical features, laboratory test results indicating inflammation, and on endoscopic and histologic findings. Disease severity was defined according to the diagnostic criteria for UC severity determination established by the Research Committee of IBD of the Ministry of Health and Welfare in Japan in 1994 [25].

Surgery was performed as a two- or three-staged operation. The two-staged operation comprised proctocolectomy, IPAA, and ileostomy as the first stage and ileostomy closure as the second stage. The three-staged operation comprised subtotal colectomy, sigmoid colon mucous fistula formation, and ileostomy as the first stage; proctocolectomy, IPAA, and re-ileostomy as the second stage; and ileostomy closure as the third stage.

**Table 1** Patients' characteristics

| | $N = 13$ |
|---|---|
| Mean age at onset (years, range) | 10.4 (5–15) |
| Mean age at operation (years, range) | 12.8 (8–17) |
| Mean duration from onset to operation (years, range) | 2.4 (0.08–6) |
| Disease severity (moderate/severe) | 6/7 |
| Staged operation (two-/three-) | 7/6 |
| IPAA leakage | 0 |
| Duration after stoma closure (years, range) | 2.5 (0.5–5) |
| Defecation | 5.1 (4–7) |
| Soiling | |
| Diurnal | 4 (30 %) |
| Nocturnal | 7 (54 %) |
| Pouchitis during the past 1 year prior to study | 4 (30 %) |

No patient experienced postoperative IPAA leakage or small bowel complications. Wound infection was observed in two patients. All patients were prescribed systemic medication to control bowel movements such as antidiarrhetic drugs. The mean number of defecations in 1 day was 5.1 (range 4–7). Slight diurnal and nocturnal soiling was observed in four (30 %) and seven (54 %) patients, respectively.

Pouchitis was defined as >7 points in the Pouchitis Disease Activity Index [26]. Pouchitis during 1 year prior to this study was observed in four patients (30 %) and responded well to antibiotic therapy. Other pouch-related complications such as pouch-anal fistula or pouch-vaginal fistula were not observed.

## Methods

We used the Japanese version of the PedsQL [10], which has a confirmed high reliability and validity. The PedsQL is a child self-report that measures HRQOL in healthy children and adolescents, as well as those with acute and chronic health conditions, between the ages of 2 and 18 years [9, 10]. The Generic Core Scales comprise a total of 23 items derived from four functioning scales: physical functioning in terms of the child's health and activities (eight items), emotional functioning in terms of the child's feelings (five items), social functioning in terms of how the child gets along with others (five items), and school functioning in terms of school daily life (five items). Physical functioning is evaluated about the problem to walk, to run,

to do sports, to lift, to take a bath, to do chores, to hurt, and to have low energy. Emotional functioning is evaluated about the problem to feel afraid, to feel sad, to feel angry, to have trouble sleeping, and to worry about what will happen to individual. Social functioning is evaluated about the problem to have trouble getting along with other kids, not to become friend with other kids, to be teased by other kids, not to do things that other kids of your age can do, and to keep up when you play with other kids. School functioning is evaluated as the problem with paying attention in class, forgetting things, having trouble keeping up with schoolwork, missing school because of not feeling well, and missing school to go to the doctor or hospital.

The study protocol was approved by the Mie University Hospital Review Board before approaching patients and guardians for participation. The study was explained to all patients and guardians by the same certified nurse specialist in child health nursing (A.K.). Written informed assent was obtained from all patients and healthy controls, and informed consent was obtained from guardians of all patients and healthy controls.

In the patient group, children completed the PedsQL at present. In the control group, children completed the same PedsQL questionnaire by retrospective imaging during the past 1 month by the PedsQL evaluation policy. Parents of patients and healthy controls were provided with a stamped, addressed envelope and were asked to return the questionnaire to the investigators.

Each of the 23 items was evaluated by assigning a score of 0–4, transforming the scores to a 0–100 scale at 25-point intervals, and averaging the scores to produce a total score, with higher scores reflecting better HRQOL [9, 10]. Each functioning score was produced by the same process in each functioning scale. Scores were expressed as mean ± 1 standard deviation. In statistical analysis, the Mann–Whitney *U* test was used to compare observed differences between groups. A $p$ value of $<0.05$ was statistically significant.

## Results

Table 2 shows the PedsQL total score and all functioning scores at present after IPAA and in healthy controls. The total score and each functioning score at present after IPAA did not show a statistically significant difference compared with the scores in healthy controls.

We evaluated the relationship between scores and soiling or pouchitis occurrence at present after IPAA (Table 3). There was no statistically significant difference in the PedsQL total score and each functioning score with or without soiling. Moreover, there was no statistically significant difference in the PedsQL total score and each functioning score with or without pouchitis. Bowel movements also had no relationship to the PedsQL scores (data not shown). The presence of soiling and pouchitis occurrence after surgery did not significantly affect the present total PedsQL scores in this study.

**Table 2** The after PedsQL score in patients at present after IPAA and healthy controls

| | Patients after IPAA | Healthy controls | $p$ value |
|---|---|---|---|
| Total score | 86.0 ± 14.3 | 93.0 ± 7.2 | NS |
| Physical functioning | 86.8 ± 13.9 | 95.5 ± 6.1 | NS |
| Emotional functioning | 90.4 ± 12.8 | 93.2 ± 10.0 | NS |
| Social functioning | 91.5 ± 17.8 | 96.1 ± 9.8 | NS |
| School functioning | 75.4 ± 26.6 | 86.7 ± 12.4 | NS |

*IPAA* ileal J-pouch anal anastomosis

**Table 3** PedsQL score in patients with or without soiling/pouchitis at present

| | Soiling | | $p$ value |
|---|---|---|---|
| | + | − | |
| Total score | 80.6 ± 16.5 | 92.3 ± 8.2 | NS |
| Physical functioning | 82.6 ± 13.7 | 91.7 ± 13.6 | NS |
| Emotional functioning | 86 4 ± 15.5 | 95.0 ± 7.7 | NS |
| Social functioning | 88 6 ± 22.1 | 95.0 ± 12.2 | NS |
| School functioning | 65.0 ± 28.9 | 87.5 ± 19 2 | NS |
| | Pouchitis | | $p$ value |
| | + | − | |
| Total score | 77.7 ± 21.5 | 89.7 ± 9.2 | |
| Physical functioning | 79.7 ± 15.6 | 89.9 ± 12.8 | NS |
| Emotional functioning | 82.5 ± 20.6 | 93.9 ± 6.5 | NS |
| Social functioning | 81.3 ± 28.4 | 96.1 ± 9.9 | NS |
| School functioning | 67.5 ± 32.8 | 78.9 ± 24.7 | NS |

## Discussion

This is the first study to evaluate patients' HRQOL using the PedsQL in children who underwent restorative proctocolectomy with IPAA for UC. This study demonstrated that IPAA could resolve interference of physical activity, emotional status, and school life due to refractory UC that worsens patients' HRQOL in children, resulting in a QOL comparable with that of healthy children.

The ultimate aim of surgical therapy for pediatric patients with UC is to improve their QOL. Several authors have reported a high level of satisfaction among patients

who underwent IPAA [20, 22, 27]. However, the functional results are not always perfect if postoperative surgery-related complications occur. Moreover, during the follow-up period, some patients complain of soiling, frequent bowel movements, incontinence, and urgency, and some patients develop pouchitis.

It is difficult to quantify such dysfunction and its impact on the HRQOL. However, surgeons must know how surgery impacts patients' emotional status and social life besides physical functioning [27]. To our knowledge, the PedsQL is the first suitable tool to evaluate such HRQOL using self-reporting by pediatric surgical patients with UC. The applicable QOL measurements in patients with UC under medical treatment are the Inflammatory Bowel Disease Questionnaire (IBDQ) [12, 13] for adults and the IMPACT [28] for children. However, these are not suitable for evaluation of the change in QOL before and after surgery, especially after proctocolectomy for UC, because dramatic changes occur in the patient's body condition, clinical symptoms, and bowel function after complete removal of the affected large bowel. In surgical patients, the Short Form-36 (SF-36) survey, which has been validated for evaluation of surgical results in adult populations, was used to evaluate QOL of both adults and children with UC [20, 23, 24, 27, 29, 30]. The SF-36 is a 36-item self-report measure of physical, social, and psychological functioning and well being. However, normative values have not been established for healthy individuals <18 years of age. Scarpa [27] demonstrated that patients who underwent operations experienced an improved QOL mainly because of improved emotional function, and HRQOL is influenced by drugs, stool frequency, pouchitis, and postoperative pelvic complications in adult patients with UC. An Italian study of the long-term effects 10 years after surgery in 21 children with UC showed that approximately 30 % of patients did not recover to normal in terms of emotional status and social life [31].

In this study, scores of physical, emotional, social, and school functioning after IPAA did not show a statistically significant difference compared with scores in healthy controls, as shown in previous studies [21]. Considering each item in each functioning category, these findings might demonstrate that disease-related pain, depressive feelings, and anxiety, which worsen children's QOL, were relieved by restorative proctocolectomy.

Interestingly, the presence of soiling and pouchitis occurrence after surgery had no significant relationship to the total PedsQL score and each functioning score in this study. These data may be specific to pediatric populations or associated with small numbers of enrolled individuals. Improvement in such items may require a certain time interval after surgery. Teaching children the better coping strategies will lead to an improvement in their HRQOL [32]. We must consider interventions specifically designed to improve the HRQOL in children after IPAA, including brochures about postoperative social and school life, counseling, psychological therapy, education to families, and consultation with teachers to assist with the return to school.

In the PedsQL survey, there were no supplemental questions regarding stool frequency, continence, medication taken to control bowel movements, surgical scars, or whether these factors affect school activity. These supplemental questions may clarify the relationship between these functioning factors and children's HRQOL. The PedsQL Gastrointestinal Symptoms Module for pediatric patients with functional and organic gastrointestinal diseases is undergoing testing for publication [33]. This module is expected to more accurately assess HRQOL in pediatric patients with UC.

The present study has some limitations such as non-prospective study and the small number of enrolled patients. The Japanese version of the PedsQL was just published in 2010, and this is the first pilot study at our institute. Considering the incidence of UC in the pediatric population in Japan, a multicenter, prospective study involving a large number of pediatric surgical patients with UC is needed in the future.

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