

Original Article

Quality of Life in Swiss Paediatric Inflammatory Bowel Disease Patients: Do Patients and Their Parents Experience Disease in the Same Way?

Rebekka Mueller¹, Farah Ziade¹, Valérie Pittet², Nicolas Fournier²,
Jessica Ezri¹, Alain Schoepfer³, Susanne Schibli⁴, Johannes Spalinger^{4,5},
Christian Braegger⁶, Andreas Nydegger¹ and the Swiss IBD Cohort Study*

¹Division of Pediatric Gastroenterology, University Hospital of Lausanne, Lausanne, Switzerland ²Institute of Social and Preventive Medicine, University Hospital of Lausanne and University of Lausanne, Lausanne, Switzerland ³Division of Gastroenterology and Hepatology, University Hospital of Lausanne, Lausanne, Switzerland ⁴Division of Gastroenterology, University Children's Hospital of Bern, Bern, Switzerland ⁵Division of Gastroenterology, Children's Hospital of Lucerne, Lucerne, Switzerland ⁶Division of Gastroenterology and Nutrition, University Children's Hospital of Zurich, Zurich, Switzerland

*Members are listed in the Appendix

Corresponding author: Andreas Nydegger, MD, PD+MER, Centre Hospitalier Universitaire Vaudois, CHUV, Rue du Bugnon 46, 1011 Lausanne, Switzerland. Tel: +41 (0)79 556 60 74; fax: +41 (0)21 314 36 45; Email: andreas.nydegger@chuv.ch

Abstract

Background and Aims: Inflammatory bowel diseases (IBDs) may impair quality of life (QoL) in paediatric patients. We aimed to evaluate in a nationwide cohort whether patients experience QoL in a different way when compared with their parents.

Methods: Sociodemographic and psychosocial characteristics were prospectively acquired from paediatric patients and their parents included in the Swiss IBD Cohort Study. Disease activity was evaluated by the Paediatric Crohn's Disease Activity Index (PCDAI) and the Paediatric Ulcerative Colitis Activity Index (PUCAI). We assessed QoL using the KIDSCREEN questionnaire. The QoL domains were analysed and compared between children and parents according to type of disease, parents' age, origin, education and marital status.

Results: We included 110 children and parents (59 Crohn's disease [CD], 45 ulcerative colitis [UC], 6 IBD unclassified [IBDU]). There was no significant difference in QoL between CD and UC/IBDU, whether the disease was active or in remission. Parents perceived overall QoL, as well as 'mood', 'family' and 'friends' domains, lower than the children themselves, independently of their place of birth and education. However, better concordance was found on 'school performance' and 'physical activity' domains. Marital status and age of parents significantly influenced the evaluation of QoL. Mothers and fathers being married or cohabiting perceived significantly lower mood, family and friends domains than their children, whereas mothers living alone had a lower perception of the friends domain; fathers living alone had a lower perception of family and mood subscores.

Conclusion: Parents of Swiss paediatric IBD patients significantly underestimate overall QoL and domains of QoL of their children independently of origin and education.

Key Words: Inflammatory bowel disease; paediatric; quality of life



1. Introduction

Inflammatory bowel disease (IBD) is a chronic disease with a potentially important negative impact on quality of life (QoL). It comprises two major disorders, ulcerative colitis (UC) and Crohn's disease (CD), as well as the non-defined group of IBD unclassified (IBDU). Approximately 25% of all IBD patients are diagnosed during childhood or adolescence.^{1–3} Worldwide, the overall incidence of paediatric IBD ranges from 4 to 10 cases per 100 000 per year, and the incidence of paediatric-onset IBD, in particular of CD, seems to be increasing in both developed and developing nations, although many countries lack accurate estimates.^{1,4–10}

2 Children with IBD typically present with moderate to severe disease activity.¹¹ Initial presentation is marked by extensive involvement in up to 51% of CD and 82% of UC cases, respectively, and with rapid early progression.^{10,12} Paediatric patients often show recurrent attacks with phases of quiescence^{13,14} and most of them require numerous potentially stressful and complex long-term medications for maintaining remission and managing acute flares.¹⁵ Symptoms of IBD, such as abdominal pain and rectal bleeding, complications specific to the paediatric population, such as growth failure and pubertal delay,^{14,16} and medication adverse effects, as well as the treatment, itself are challenging for both patients and parents.¹⁷ The impact on QoL during the critical period of childhood and adolescence, including education and entry into the workforce, is often considerable.¹⁸ Assessment of the patient's estimation of their QoL is therefore essential and may have an impact on treatment strategies.¹⁹

In paediatric IBD patients, it is crucial to also involve parents in the medical treatment process. Parents are, to an important extent, responsible for the overall development and well-being of their children and they need to make assumptions on how their child experiences its disease. Therefore, as legal representatives, parents should not only inform medical staff but also adapt psychological and physical parental support to the child according to disease activity. However, we do not know whether the parents' perception are always accurate and whether the assessment of QoL from the parents' perspective is similar to that of the diseased child.

The aim of this study was to explore the difference in perception of the QoL of paediatric IBD patients between the patients themselves and their parents, using data collected in the frame of the paediatric sub-cohort of the Swiss IBD Cohort Study (SPIBDCS). Furthermore, we aimed to study factors that may affect the perception of QoL. We hypothesized that parents and their children with IBD do not experience the disease in the same way.

2. Materials and methods

2.1. Patients

The Swiss IBD Cohort Study (SIBDCS) was initiated in 2006. The SIBDCS is a national prospective cohort study of IBD patients in Switzerland. It aims to provide up-to-date information regarding different aspects (among other QoL assessments) of IBD in Switzerland for the Swiss and international scientific community, public health authorities and medical staff.²⁰ For the paediatric sub-cohort, the Swiss Paediatric IBD Cohort Study, physician questionnaires and patient questionnaires were adapted for paediatric patients and their parents.

Up to December 2012, 196 paediatric patients ≤16 years of age, diagnosed with CD, UC or IBDU according to the international Porto criteria,²¹ were included at least 4 months after diagnosis, and registered in the SIBDCS database. As described by Pittet et al.,²⁰ patients were recruited from the following six university centres in Switzerland: Lausanne, Geneva, Bern, Basel, Zurich and St Gallen, with the

University of Lausanne as coordinating centre and database location. Ethics approval was obtained for the study protocol by the ethics committees of the cantons or regions from where the patients were included, as well as by individual consent of patient and parents. Our study sample comprised 110 paediatric patients with available and sufficiently completed questionnaires from both patients and their parents.

2.2. Data collection

We retrieved data regarding QoL and clinical reporting forms of the SPIBDCS, including demographic variables (age, gender, country of birth and schooling level) and medical items (type of IBD, Paediatric Crohn's Disease Activity Index [PCDAI] and Paediatric Ulcerative Colitis Activity Index [PUCAI]). Stratification into two age groups was initially performed (group 1 younger than 12 years; group 2, 12 years or older). Results were similar in both groups, but with a lack of statistical significance in group 1 due to low sample size ($n = 24$). Therefore, no stratification according to age groups was performed. One questionnaire was completed with the child during a structured interview conducted by a research assistant (a trained IBD nurse) without the presence of his/her parents, and another was completed by the parents separately at home. The QoL assessment was based on the KIDSCREEN-GROUP 2004 questionnaire, a well-validated five-domain self-report questionnaire of general QoL domains ('physical activity', 'children's mood', 'family life', 'friends' and 'school performance'), using a Likert scale that ranges from 0 (never) to 5 (frequent), which was developed specifically for children and adolescents between 8 and 18 years, appropriate for the life and development issues of children with chronic disease.^{22,23} Two KIDSCREEN questionnaires were thus completed – by the child and one by the parents – and the two were compared. A sociodemographic questionnaire assessing family characteristics (age, birth country, familial situation, parental education, number of children and notable life events) was completed by the parents.

2.3. Statistical analysis

We assessed the difference between overall scores and scores for each domain between paediatric patients and their parents. Differences in QoL scores were calculated as parents minus child, with a positive difference indicating a higher perception and a negative result a lower perception by parents. Parents' perception was correlated with social, psychological and demographic variables as well as with the type and severity of disease, in order to help characterize observations. For the purpose of these analyses, we grouped diagnoses of UC and IBDU in a single category, to be compared with CD. Continuous variables were described using the median, interquartile range (IQR) and range. The differences between QoL scores for paediatric patients and their parents were compared by using a Wilcoxon test for two categories and a Kruskal–Wallis test for three or more categories. The signed rank test was used to test whether the median of a single group was statistically different from a fixed value, typically 0. The effects of continuous covariates on differences in QoL were analysed using a linear regression. We considered $p < 0.05$ to be statistically significant.

3. Results

3.1. Patient characteristics

One hundred and ten paediatric patients with active IBD were included (55 males [50%]), of whom 59 (53.6%) presented with CD, 45 (40.9%) with UC and 6 (5.5%) with IBDU. Median age was 14 years (range 7–17 years). Most of the patients (71.8%) attended secondary school (Table 1).

Table 1. Demographics of children and parents.

	Child	Mother	Father
Disease			
Crohn's disease	59 (53.6%)		
Ulcerative colitis	45 (40.9%)		
IBD unclassified	6 (5.5%)		
Gender			
Male	55 (50.0%)		
Female	55 (50.0%)		
Age, years (median, IQR, range)	14 (12–15, 7–17)	44 (41–47, 30–52)	47 (44–50, 35–64)
Country of birth			
Switzerland	83 (75.5%)	72 (65.4%)	64 (58.2%)
Other	6 (5.5%)	18 (16.4%)	26 (23.6%)
Unknown	21 (19.0%)	20 (18.2%)	20 (18.2%)
Schooling level			
Primary school	4 (3.6%)		
Secondary school	79 (71.8%)		
Special school	3 (2.7%)		
Logopaedic school	1 (0.9%)		
Private school	6 (5.5%)		
Other	12 (10.9%)		
Unknown	5 (4.6%)		
Education			
None		3 (2.7%)	1 (0.9%)
Compulsory school		1 (0.9%)	7 (6.4%)
Basic formation		32 (29.1%)	30 (27.3%)
Higher formation		21 (19.1%)	11 (10.0%)
Technical school		21 (19.1%)	25 (22.7%)
University		9 (8.2%)	13 (11.8%)
Other		5 (4.5%)	4 (3.6%)
Unknown		18 (16.4%)	19 (17.3%)
Family situation			
Alone		10 (9.1%)	7 (6.4%)
Married/cohabiting		78 (70.9%)	78 (70.9%)
Unknown		22 (20.0%)	25 (22.7%)
Number of children			
0		0 (0.0%)	1 (0.9%)
1		9 (8.2%)	8 (7.3%)
2		50 (45.2%)	46 (41.8%)
3 or more		32 (29.1%)	33 (30.0%)
Unknown		19 (17.3%)	22 (20.0%)

IQR, interquartile range

3.2. Parent characteristics

When comparing mothers with fathers, there were only small differences regarding age, birth country, education level, family situation and number of children. Most of the parents were either married or cohabiting (70.9%) and the majority of parents were born in Switzerland (65.5% of mothers and 58.2% of fathers). Almost one-third of parents had basic education (29.1% of mothers and 27.3% of fathers), whereas nearly 50% of all parents had attended either higher education, technical school or university (46.4% of mothers and 44.5% of fathers). Most of the parents had 2 or more children (Table 1). The vast majority of questionnaires were completed by the mother only (78.9%), whereas 10.1% of the parents completed it together. There was, however, no statistically significant difference according to whether the mother was involved (alone or together with the father) in completing the questionnaire.

3.3. QoL differences according to patient's gender and age

Parents perceived the overall QoL of their children with IBD significantly lower, as well as the domains mood, family life and friends, compared

with the patient's own evaluation ($p < 0.001$). However, there was no significant difference in QoL scores regarding the physical well-being and school performance domains (Figure 1). This result was consistent for both genders, even though there was a trend to lower perception in girls regarding the domains of mood ($p = 0.057$) and family life ($p = 0.069$). Patient's age did not have a significant impact on evaluation of QoL.

3.4. QoL differences according to disease type and activity

There was no significant difference regarding the QoL differences between CD patients and UC/IBDU patients (Table 2). The severity of disease according to PCDAI and the PUCAI index was not a significant factor for difference in QoL evaluation between parents and patients either. Also, patients' hobbies did not significantly affect the difference in QoL between parents and children. ednav

3.5. QoL differences according to family situation

Regarding the family situation, the small number of single living mothers/fathers (Table 1) makes a valuable statistical

interpretation of this subgroup difficult. Still, married and cohabiting mothers had a significantly lower perception of overall QoL ($p < 0.001$) and its domains mood, family and friends (all $p < 0.001$), whereas results for mothers living alone were slightly more concordant with those for their children. In fact, the mothers' estimates were lower only for overall QoL ($p = 0.006$) and the domain friends ($p = 0.005$). Results for fathers were similar: estimates for fathers living alone were significantly lower overall QoL ($p = 0.022$) and for the domains family ($p = 0.017$) and mood ($p = 0.028$), but not friends (Figure 2). The differences between the two groups of married/cohabiting and mothers/fathers living alone were not significant (Figure 2). Regarding siblings, the lower perception of overall QoL and of the domains family and friends by parents was significant, independently of the number of siblings; the item mood was only estimated lower in the subgroup of patients with siblings (one sibling, $p < 0.001$; two or more siblings, $p = 0.002$).

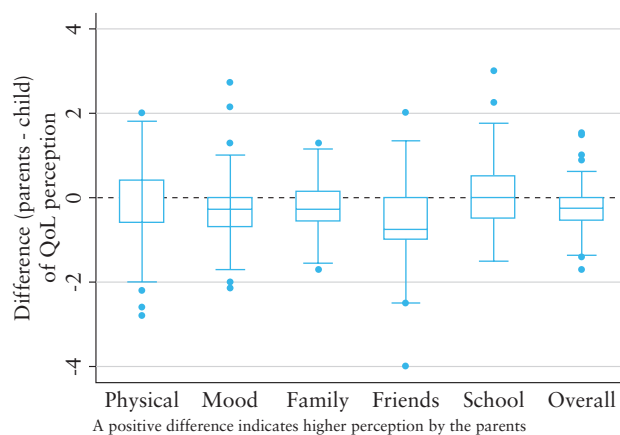


Figure 1. Difference in evaluation of patients QoL between parents and patients.

Table 2. Association of IBD type (CD, UC/IBDU) with difference in quality of life estimation between parents and patients.

Diagnosis	CD ^a	UC/IBDU ^a	<i>p</i> -value (CD vs UC/IBDU)	<i>p</i> -value (CD) = 0	<i>p</i> -value (UC) = 0
Physical	0.0	0.0	0.364	0.771	0.161
	-0.6 to 0.6	-0.8 to 0.2			
Mood	-0.3	-0.4	0.395	<0.001	<0.001
	-0.7 to 0.1	-0.9 to 0.0			
Family	-0.3	-0.	0.767	0.002	<0.001
	-0.7 to 0.1	-0.6 to 0.0			
Friends	-0.5	-0.8	0.152	<0.001	<0.001
	-1.0 to 0.0	-1.0 to -0.3			
School	0.0	0.0	0.152	0.286	0.339
	-0.3 to 0.5	-0.5 to 0.3			
Overall	-0.2	-0.3	0.152	0.001	<0.001
	-0.5 to 0.1	-0.6 to -0.0			
	-1.4 to 1.5	-1.7 to 1.5			

^aData are median of the difference (a positive number denotes overestimation, a negative number underestimation by parents), interquartile range and range (minimum–maximum).

IBD, inflammatory bowel disease; CD, Crohn's disease; UC, ulcerative colitis; IBDU, IBD unclassified.

3.6. QoL differences according to background of parents

As mentioned above, overall QoL as well as the domains mood, family and friends were perceived to be lower by all parents. When splitting the parents into groups of parents with birthplace in Switzerland and abroad, this result remained significant only in the subgroup of mothers/fathers born in Switzerland (all $p < 0.001$). There was no significant difference regarding the level of education of the parents, even though parents with a higher level of education tended to underestimate QoL more than parents with a lower level of education (Table 3).

The age of the parents had an impact on the differences observed. As they became older, mothers significantly underestimated QoL regarding friends, whereas fathers significantly overestimated QoL regarding school, compared with their children.

The number of notable life events of mothers (none, one, two or more) had no significant impact on estimation of patients' QoL.

4. Discussion

This study shows that parents in Switzerland do not experience the disease of their children with IBD in the same way as the patients themselves. They report a significantly lower overall QoL of their children with IBD, as well as of its sub-aspects of mood, family life and friends, compared with the patients themselves. Interestingly, the domains of school performance and physical activity did not differ. These findings might be due to the fact that parents can quite easily assess school performance by objective means – from school notes and physical activity – by simple observation of their child. Interpretation of a child's mood and its' interaction with family and friends, however, is more subjective and might be difficult, especially as children tend to not share all of their feelings and experiences with their parents.

Our results show that parents of Swiss paediatric IBD patients underestimate the QoL of their affected children. One might suspect that parents' concern regarding the well-being of their children is

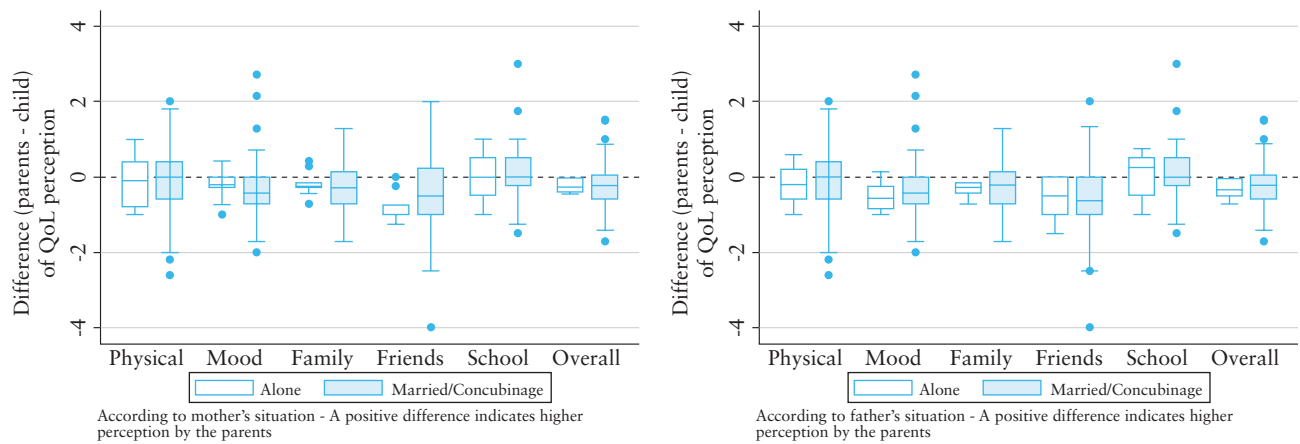


Figure 2. Impact of the parents' situation (alone, married or cohabiting) on difference in QoL estimation between parents and patients.

high. In fact, a recently published first assessment of the QoL estimation by Swiss paediatric IBD patients by Rogler et al.²⁴ also analysed the data of the same paediatric sub-cohort of the SPIBDCS; it shows moderate impairment of health-related QoL for physical well-being due to disease activity, but good mental well-being and social support and significantly higher QoL regarding autonomy and school variables in IBD patients compared with healthy children. It is possible that parents underestimate their children's QoL, leading to increased parental support and therefore to a higher QoL of paediatric IBD patients.

None of the assessed demographic, social or medical aspects of patients and their parents had a significant influence on the overall evaluation of QoL. Several factors concerning the patient (type of IBD, severity of disease, gender) and the family (parents' education, birth country, family structure) seemed to have a clear influence on the perception of the subdomains of QoL; however, these factors were non-significant. Only the parents' age seemed to affect significantly the estimation of differences in QoL. These results are comparable to the findings of Loonen et al.²⁵ in the Netherlands, who, to the best of our knowledge, were the first to investigate the degree of agreement between parents and their children with IBD regarding health-related QoL, and they assessed factors influencing parent-child agreement. In accordance with our findings, they found that parents underestimated QoL regarding social functioning, but that other components, like body complaints, cognitive functioning, autonomy and positive and negative emotions, were rated rather similarly. Another study, by Kunz et al.,²⁶ assessing adolescent IBD patients between 11 and 18 years in the USA, showed overestimation of adolescents' QoL by parents, with lowest agreement in the aspects of school and social functioning; it was hypothesized in this study that school performance may appear better outwardly (e.g. adequate grade point average), while adolescents may perceive internally their school functioning as negatively influenced by their medical condition (e.g. concentration difficulties).

Studies in children with other chronic diseases found various results. Whilst Dey et al.²⁷ found relatively high parent-child agreement when examining three health status groups (children with mental health problems, children with physical health problems and healthy controls), other studies in children with chronic pain or cancer showed results similar to those of our study.^{28,29} Parents of children with a chronic health condition/illness perceived their children to have significantly lower QoL than did the parents of children without a chronic health condition.³⁰ Finally, Rajmil et al.³¹ showed

moderate to low parent-child agreement in healthy children as well, and the degree of agreement lessened with the children's age.

Children with IBD, as well as children with other chronic diseases like epilepsy and obesity³² and children with severe cardiovascular disease,³³ are at risk of experiencing lower health-related QoL than their healthy peers,³⁴⁻³⁶ and QoL seems to worsen with age, as shown by some authors.^{31,32} As we did not compare patients with their healthy peers, we were not able to confirm this observation. Also, a trend with increasing age could not be shown because stratification for age was not conclusive. The assessment of QoL, including aspects of physical, psychological and social functioning, therefore plays an important role in the evaluation of the burden of this chronic illness. Involvement of parents in the treatment process is essential for the optimal adaptation of medical treatment and psychological support. It is therefore important to know how parents estimate the QoL of their sick child.

The difference in the perception of the patients' QoL between the patients themselves and their parents might partially be due to under-reporting or minimizing of symptoms by young patients with IBD, as a result of an adaptive reaction in order to deny the full extent of the disease,^{35,37} or by their inability to reporting psychological symptoms.³⁸ It is also possible that patients report symptoms quite accurately to their parents, who then pathologize normal behaviour and over-report symptoms compared with parents of non-chronically ill children. Since QoL is subjective and individual, a certain difference might also be normal. In fact, in children with primarily inactive IBD, fatigue was significantly higher and health-related QoL was significantly lower than in healthy controls.³⁹ Results for children with IBD were comparable to those for children with rheumatological diseases and cancer.^{40,41} The inclusion of both patients' and parents' measures of QoL can therefore provide complementary perspectives.⁴² The discussion of differing results might also enable constructive communication between parents and children.⁴³

A holistic approach in support of paediatric IBD patients is essential. It has been known for many years that the mental well-being of paediatric IBD patients correlates well with psychological and social support.^{17,44} Studies have documented a significant correlation of adolescents' health-related QoL scores with satisfaction and degree of closeness with their social support members such as parents.⁴³ Even though increased disease severity was found to correlate with increased social dysfunction⁴⁵ and decreased QoL,^{35,39,46} psychosocial factors such as family functioning are more predictive of QoL than disease activity. Children and adolescents seem

Table 3. Association of the parents' formation (none or basic, higher) with difference in QoL estimation between parents and patients.

	Mother's education ^a		<i>p</i> -value (none vs higher)	<i>p</i> -value (none) = 0	<i>p</i> -value (higher) = 0
	None or basic	Higher			
Physical	-0.2	0.0	0.519	0.171	0.634
	-0.6 to 0.2	-0.7 to 0.5			
Mood	-0.3	-0.4	0.849	0.002	0.001
	-0.7 to 0.1	-0.7 to 0.0			
Family	-0.1	-0.4	0.137	0.025	0.001
	-0.4 to 0.1	-0.7 to 0.1			
Friends	-0.3	-0.8	0.139	0.005	<0.001
	-1.0 to 0.3	-1.0 to -0.3			
School	0.0	0.0	0.668	0.568	0.967
	-0.5 to 0.5	-0.3 to 0.5			
Overall	-0.2	-0.3	0.303	0.004	<0.001
	-0.5 to 0.0	-0.6 to 0.0			
	-1.4 to 1.5	-1.7 to 1.5			
Father's education					
	None or basic	Higher	<i>p</i> -value (none vs higher)	<i>p</i> -value (none) = 0	<i>p</i> -value (higher) = 0
Physical	0.0	-0.2	0.195	0.795	0.102
	-0.6 to 0.5	-0.8 to 0.4			
Mood	-0.3	-0.4	0.393	0.016	<0.001
	-0.7 to 0.1	-0.7 to 0.0			
Family	-0.1	-0.4	0.428	0.039	<0.001
	-0.6 to 0.1	-0.7 to 0.1			
Friends	-0.3	-0.8	0.114	0.003	<0.001
	-1.0 to 0.3	-1.1 to -0.1			
School	0.3	-0.1	0.088	0.124	0.394
	-0.3 to 0.5	-0.5 to 0.4			
Overall	-0.1	-0.3	0.052	0.081	<0.001
	-0.4 to 0.1	-0.6 to -0.1			
	-1.4 to 1.5	-1.7 to 1.5			

^aData in the education columns are median of the difference (a positive number denotes overestimation, a negative number underestimation by parents; inter-quartile range and range (minimum–maximum)).

to depend more on their parents' coping skills than their own.⁴³ They experience more IBD-related difficulties in school and social functioning with distressed mothers,⁴⁷ and parent distress is linked to poorer health-related QoL in adolescents with IBD.⁴⁸ These findings are also known in other chronic diseases: Hamner et al.⁴⁹ described lowered levels of physical, emotional and social functioning amongst paediatric patients with cancer associated with parental chronic stress. Similarly to our results, they also found that patients' hobbies did not significantly affect the difference in QoL estimation; this might also be related to the fact that psychosocial factors such as family life have more influence on psychological well-being than personal hobbies. There is evidence that chronic conditions such as IBD not only affect patients, but also impair parent and family functioning. In particular, mothers of children with IBD are more likely to express distress, to show

increased rates of depression and to report significantly greater family dysfunction than mothers of healthy children.⁴⁴ *Vice versa*, higher health-related QoL of patients with IBD and lower disease activity was associated with higher QoL of parents.⁵⁰ All these findings underline the importance of QoL assessments and their interpretation.

Our study has important strengths but some limitations as well. Strengths of the study are the prospective cohort design, which included 110 patients of the cohort of Swiss paediatric IBD patients, using an internationally valid health-related QoL instrument, as well as using the skills of a specially trained study nurse undertaking the interview with the child in the absence of the parents. The enrolment of paediatric patients in the Swiss IBD cohort requires the medical diagnosis of active IBD, which was performed in a hospital, as well as informed consent by the patients and parents. Since all of the

tertiary hospital centres in Switzerland participated in data collection, we assume that the cohort is representative of the overall Swiss paediatric IBD patient population. The questionnaires contained easily understandable questions. They have been validated and were available in several languages and the paediatric patient was helped by a trained IBD nurse without his/her parents being present, thus minimizing the risk of children up-scaling their responses to please their parents.

Unfortunately, 86 patients and/or their parents did not complete the questionnaires regarding QoL, which is the major limitation of our study, and no data are available for comparison if the parents/patients who did not complete the QoL questionnaire were more often depressed or less educated. However, when comparing the disease activity indexes of the entire cohort (196 patients), no significant difference could be seen between responders and non-responders ($p = 0.430$ for CD and $p = 0.618$ for UC). Even though we consider our cohort to be representative, since every single child with diagnosed IBD is invited to join the IBD cohort, we cannot exclude an undefined selection bias, as some paediatric patients might also be followed up by adult gastroenterologists not participating in the Swiss IBD cohort study. Other methodological limitations are related to the character of the questionnaires: discussions between the parents (mother and father) whilst filling in their individual questionnaire might have affected the evaluation of QoL by the parents. The fact that only 10% of questionnaires were completed by both parents together renders this risk negligible. Other methodological limitations are related to the retrospective character of the questionnaires: patients and their parents had to report facts related mainly to the past, e.g. school performance or family life of the last few months, with the risk that the remembered information provided was not accurate. Finally it cannot be excluded that children try to avoid communicating their concerns.

In conclusion, the present cohort study clearly shows that Swiss IBD patients and their parents do not experience the disease of the child or adolescent in the same way. The uniform and validated questionnaires, filled in separately by paediatric patients and their parents, represent a valuable instrument in the evaluation of health-related QoL of paediatric IBD patients in Switzerland in order to further improve their care, as it opens fields of discussion for communication between parents and their affected children.

Conflict of Interest

The authors report no conflicts of interest.

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Author Contributions

All authors contributed to the conception and design of the study, acquisition of data, or analysis and interpretation of data; drafting the article or revising it critically for important intellectual content; and final approval of the version to be submitted.

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Appendix

Members of the SIBDCS study group

Claudia Anderegg, Peter Bauerfeind, Christoph Beglinger, Stefan Begré, Dominique Belli, José Bengoa, Luc Biedermann, Janek Binek, Mirjam Blattmann, Nadia Blickenstorfer, Stephan Boehm, Jan Borovicka, Christian Braegger, Patrick Bühr, Bernard Burnand, Emmanuel Burri, Sophie Buysse, Matthias Cremer, Dominique Criblez, Philippe de Saussure, Lukas Degen, Joakim Delarive, Christopher Dörig, Barbara Dora, Gian Dorta, Tobias Ehmann, Ali El Wafa, Mara Egger, Matthias Engelmann, Christian Felley, Markus Flegner, Nicolas Fournier, Montserrat Fraga, Alain Frei, Pascal Frei, Remus Frei, Michael Fried, Florian Froehlich, Raoul Furlano, Suzanne Gallot-Lavallée, Martin Geyer, Marc Girardin, Delphine Golay, Horst Haack, Tanja Grandinetti, Beat Gysi, Horst Haack, Johannes Haarer, Beat Helbling, Peter Hengstler, Denise Herzog, Cyrill Hess, Klaas Heyland, Thomas Hinterleitner, Philippe Hiroz, Claudia Hirschi, Petr Hruz, Pascal Juillerat, Rosmarie Junker, Christina Knellwolf, Christoph Knoblauch, Henrik Köhler, Rebekka Koller, Claudia Krieger, Gerd A. Kullak-Ublick, Markus Landolt, Frank Lehmann, Valérie McLin, Philippe Maerten, Michel Maillard, Christine Manser, Andrew Macpherson, Michael Manz, George Marx, Rémy Meier, Christa Meyenberger, Jonathan Meyer, Pierre Michetti, Benjamin Misselwitz, Darius Moradpour, Patrick Mosler, Christian Mottet, Christoph Müller, Pascal Müller, Beat Müllhaupt, Claudia Münger, Leilla Musso, Andreas Nagy, Cristina Nichita, Jan Niess, Natacha Noël, Andreas Nydegger, Maliza Nzabonimpa, Nicole Obialo, Carl Oneta, Cassandra Oropesa, Céline Parzanese, Laetitia-Marie Petit, Franziska Piccoli, Julia Pilz, Gaëlle Pittet, Valérie Pittet, Bruno Raffa, Ronald Rentsch, Sophie Restellini, Jean-Pierre Richterich, Silvia Rihs, Jocelyn Roduit, Daniela Rogler, Gerhard Rogler, Jean-Benoît Rossel, Markus Sagmeister, Gaby Saner, Bernhard Sauter, Mikael Sawatzki, Michael Scharl, Sylvie Scharl, Nora Schaub, Martin Schelling, Susanne Schibli, Hugo Schlauri, Daniela Schmid, Sybille Schmid, Jean-François Schnegg, Alain Schoepfer, Christiane Sokollik, Frank Seibold, Gian-Marco Semadeni, Mariam Seirafi, David Semela, Arne Senning, Marc Sidler, Johannes Spalinger, Holger Spangenberg, Philippe Stadler, Volker Stenz, Michael Steuerwald, Alex Straumann, Michael Sulz, Alexandra Suter, Michela Tempia-Caliera, Joël Thorens, Sarah Tiedemann, Marjan Timmer, Radu Tutuian, Ueli Peter, Stephan Vavricka, Francesco Viani, Roland Von Känel, Alain Vonlaufen, Dominique Vouillamoz, Rachel Vulliamy, Helene Werner, Paul Wiesel, Reiner Wiest, Tina Wylie, Jonas Zeitz, Dorothee Zimmermann.