



# Pain and quality of life in youth with inflammatory bowel disease: the role of parent and youth perspectives on family functioning

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## Abstract

**Introduction:** Daily pain experiences are a common feature of pediatric inflammatory bowel disease (IBD), which can negatively influence their health-related quality of life (HRQOL). A holistic, family systems approach is needed to further our understanding of daily pain experiences in youth with IBD and their influence on youth's HRQOL.

**Objectives:** The study's objectives were to (1) provide a detailed description of daily pain experiences in youth with IBD, (2) investigate the relative contribution of family functioning and pain in explaining youth's HRQOL, and (3) explore differences in parental and youth perspectives.

**Methods:** Sixty youth with IBD (8–17 years) and a parent completed questionnaires to assess family functioning, HRQOL, and pain experiences within the past week. A subsample of 16 youth completed an online diary (7 days) about their pain experiences.

**Results:** When including any pain experiences, higher youth-reported family satisfaction and lower pain intensity were related to better HRQOL, whereas higher parent-reported family cohesion and satisfaction indirectly related to youth HRQOL through lower pain intensity. When only accounting for abdominal pain, pain intensity related negatively with HRQOL, and only parent-reported cohesion showed an indirect relation with HRQOL through pain intensity. Diary data revealed large heterogeneity: abdominal pain, described as cramping, sharp, and/or stinging was most frequent, but other pain symptoms (eg, back pain and headache) often co-occurred.

**Conclusion:** The findings provide a rich picture of the daily pain experiences of youth with IBD and underscore the importance of a family systems approach to understand how family functioning and pain symptoms influence HRQOL.

**Keywords:** Inflammatory bowel disease, Health-related quality of life, Pain, Family functioning, Multiple perspectives, Family satisfaction, Family cohesion

## 1. Introduction

Inflammatory bowel disease (IBD), consisting of Crohn's disease (CD) and ulcerative colitis, is a chronic condition of the gastrointestinal system with uncontrolled inflammation of the intestinal mucosa as its hallmark. Inflammatory bowel disease is characterized by a relapsing and remitting course, and when active often presents with abdominal pain, diarrhea, and weight loss.<sup>3</sup> In 25% of cases, it is diagnosed in those younger than 18 years, with a pronounced impact on their physical and

psychosocial health-related quality of life (HRQOL).<sup>9,25</sup> Abdominal pain is a common feature in pediatric IBD, experienced by over 70% of youth,<sup>10</sup> irrespective of disease status. Indeed, a substantial number of patients with an inactive disease status continue to report abdominal pain symptoms (41%),<sup>11</sup> which negatively influence their HRQOL.<sup>9</sup> However, little has been reported about the exact nature and influence of pain youth with IBD experience on a daily basis, such as the frequency, co-occurrence with other pain experiences (eg, headache),

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interference, and particular affective and sensory features.<sup>9</sup> The co-occurrence with other pain symptoms, in particular, warrants further exploration given the increasingly common observation of chronic-on-acute pain or the experience of pain symptoms in youth with a chronic condition that is not explained by their organic disease.<sup>7</sup> The paediatric IBD literature mostly focuses on abdominal pain experiences, thereby lacking an understanding of potential co-occurrence with other pain symptoms. However, in youth with centrally mediated abdominal pain syndrome (CAPS), a large overlap has been found between persistent abdominal pain symptoms and other pain complaints, such as headaches. This large co-occurrence of pain symptoms could point towards central sensitization as an underlying mechanism for persistent pain experiences that could persevere into adulthood.<sup>5,36</sup> Online diary methodologies allow for precise, prospective assessment of pain experiences and could therefore be beneficial to obtain a better insight into the daily pain experiences of youth with IBD.<sup>17</sup> However, a holistic approach, which includes the influence of psychosocial factors, towards daily pain experiences of youth with IBD and their influence on HRQOL is crucial to improve our ability to identify those youth at risk for poor functioning irrespective of disease status.<sup>18,34</sup> For instance, findings by Claar et al.<sup>4</sup> revealed how pain experiences were common for pediatric patients in remission and were strongly influenced by youth's depressive symptoms.

In addition to individual psychological factors, evidence is accumulating showing the importance of family environment in understanding how youth cope with IBD. Indeed, both youth and caregiver internalizing symptoms (eg, depressive symptoms) have been found to mediate the relationship between youth HRQOL and IBD disease activity.<sup>27</sup> With respect to IBD-related pain experiences, especially during remission, a strongly influence of family stress has been observed.<sup>28</sup> Furthermore, youth with IBD, living in families with clinical-relevant difficulties with general family functioning report lower levels of general well-being and social functioning.<sup>14,15</sup> Shifting the focus from general family functioning to an exploration of the relative contribution of specific family dimensions in explaining pain experiences and HRQOL has the potential to further clarify the underlying mechanisms.<sup>26</sup> The circumplex model of family functioning, grounded in family systems theory, and integrating of 3 family dimensions (ie, cohesion, flexibility, and communication) holds promise in helping to understand the associations between family functioning and youth's outcomes.<sup>22</sup> Family systems theory postulates that families are a small set of interrelated and independent individuals who act as one unit to maintain balance,<sup>21,26</sup> with the circumplex model suggesting that extreme low and high levels of family dimensions reflect unbalanced family functioning.<sup>22</sup> Preliminary evidence in youth with chronic pain suggests that extreme unbalanced levels of family cohesion (ie, emotional bond) are related to more pain and disability.<sup>18</sup> However, the role of other potentially relevant, family dimensions such as flexibility (ie, flexibly adjusting roles and rules to changing circumstances), communication patterns, and satisfaction with family functioning is largely unexplored.

A better understanding of which specific family characteristics influence pain experiences in youth with IBD and how this in turn influences their HRQOL is beneficial to further optimize family interventions in the context of pediatric IBD. After the family systems approach, it is important to solicit each member's view for an accurate perspective on the family system.<sup>1,21</sup> Consequently, the objectives were to (1) provide a detailed insight into the daily pain experiences of youth with IBD; (2) investigate the relative influence of family cohesion, flexibility, communication,

and satisfaction on HRQOL, beyond the influence of pain experiences, and (3) explore differences in parental and youth perspectives on family functioning and their subsequent influence on youth pain experiences and HRQOL.

## 2. Methods

### 2.1. Participants

Ethical approval for this study was obtained from the IWK Health Centre research ethics board. Youth were eligible to participate if they were between 8 and 17 years of age; able to speak, write, and read English; and had no hearing and/or vision impairments. Recent pain experiences (ie, in past 2 weeks) were not a requirement for inclusion. All 226 families of eligible patients registered at the Gastroenterology clinic at the IWK Health Centre were introduced to the study by a letter in name of the collaborating pediatric gastroenterologist. About 2 weeks after sending out the letter, the research team attempted to reach all families by phone to provide more details on study participation and confirm their intention to participate or not. A total of 176 families were reached by phone (78%), of which 78 families agreed to participate (44%). Of the nonparticipants, 53% declined participation and 5 (3%) were not eligible. Main reasons for declining participation were lack of interest, travel time, and other responsibilities. Of those 78 families, a total of 60 families took part in the study. Scheduling problems were the main reason for the remaining 18 families to eventually not participate in the study. It was not possible to establish whether there were any systematic differences between participating families and those who declined to participate.

All participants were given the option to participate in the diary part of the study, with a subset of 18 participants (30%) agreeing to participate in the diary part and 16 patients completing enough diary entries (ie, more than 4 days) to allow for inclusion in the analyses.<sup>3</sup> Four youth reported no pain at all throughout the week (3 of these 4 youth had an inactive disease status). Given that pain experiences were the main focus of the diary component, these 4 youth were excluded from the analyses, resulting in a final sample of  $n = 12$ .

### 2.2. Measures

The data presented in this manuscript reflects the self-reported assessment part of a larger study, which also included participation in experimental tasks (these data have not been published yet). These self-report measures were completed in the laboratory at the start of the study, before engaging in any experimental task.

#### 2.2.1. Demographic and medical data

Youth's demographic (ie, youth age and sex) and medical data (ie, diagnosis, Physician Global Assessment, months since diagnosis) were collected from the youth's medical record. The reliable and standardized Physician Global Assessment scale<sup>9</sup> was used to determine the current disease activity. Physician Global Assessment was based on the last physician visit before participation and collected from the youth medical record. Given that disease severity is variable, the PGA took place as close as possible to study participation. The average amount of days in between study participation and PGA was 7.23 days (SD = 14.73, range: 0–60), with both taking place on the same day for the majority of the participants (N = 40 or 66.67%). Parental demographic data were provided by the participating parent.

## 2.2.2. Family functioning

Family functioning was assessed with the FACES-IV,<sup>23</sup> a 62-items self-report assessment of family cohesion, flexibility, communication, and satisfaction. The 42 items assessing cohesion and flexibility and 10 items assessing communication are completed using a 5-point numeric rating scale from 1 (strongly disagree) to 5 (strongly agree). The response scale for the 10 satisfaction items ranges from 1 = very dissatisfied to 5 = extremely satisfied.<sup>23</sup>

A ratio score was calculated for both cohesion and flexibility, reflecting the relative amount of balanced vs unbalanced characteristics within family functioning. Ratio scores were calculated by dividing the score for the balanced subscale of cohesion/flexibility by the average of the 2 unbalanced subscales (ie, disengaged and enmeshed functioning for cohesion/rigid and chaotic functioning for flexibility). Ranging from 0 to 10, a ratio score of 1 reflects balanced functioning. The lower the score below 1, the more unbalanced the family functioning, whereas the higher the score above 1, the more balanced the family functioning.<sup>23</sup>

Percentile scores for the communication and satisfaction scales were used in the analyses, with higher percentile scores reflecting either positive communication patterns or more satisfaction regarding their family functioning.<sup>23</sup> The FACES-IV has been shown to be a reliable and valid assessment.<sup>2,12,23</sup> Although validity has been established for youth 12 years and older, the questionnaire has previously been used successfully in younger samples as well.<sup>6,29</sup> Further confirming the utility in younger samples, the reliability of the FACES-IV subscales did not change when only using the responses of our sample of youth 12 years and older ( $n = 45$ ). Reliability analyses in the current sample revealed acceptable to excellent reliability for both youth and parent report ( $\alpha = 0.62$ – $0.92$ ) with exception of 2 scales: youth-reported enmeshed functioning ( $\alpha = 0.43$ ) and parent-reported communication ( $\alpha = 0.41$ ). Low reliability scores for the enmeshed scale have been noted in previous studies (eg,  $\alpha = 0.65$ <sup>20</sup>) and reflect the results of the original validation study ( $\alpha = 0.77$ <sup>23</sup>). Hence, it was decided for study fidelity reasons, to include the data of all youth and all subscales, in the analyses with the FACES-IV while taking caution in interpreting the results of youth-reported cohesion and parent-reported communication.

## 2.2.3. Health-related quality of life

The IMPACT-III was used to assess youth's IBD-specific HRQOL. The IMPACT-III is a valid and reliable 35-item self-report questionnaire assessing youth's general well-being (12 items), emotional functioning (7 items), social functioning (11 items), and body image (4 items).<sup>24</sup> Each item has 5 Likert response options (eg, 0 = rarely to 4 = very often). All items scores are reverse scored and transformed to a range of 0 to 100 (0 = 100, 1 = 75, 2 = 50, 3 = 25, 4 = 0). A total score is obtained by the sum of all items divided by the number of items, with higher scores representing better quality of life. The Cronbach's alpha for the IMPACT-III in the current study was 0.89, reflecting good internal consistency.

### 2.2.3.1. General level of pain and interference

Pain intensity in the past week was assessed using the Faces Pain Scale-Revised<sup>16</sup> varying from "no pain" (0) to "very much pain" (10). In addition to completing the Faces Pain Scale-Revised, youth marked the exact location of their pain(s) using

a manikin with front and back view. Pain interference was assessed by asking youth to indicate on an 11-point numeric rating scale how much their pain had interfered with their daily activities in the past week (0 = no interference to 10 = unable to carry on activities). These pain reports covered any pain youth had experienced in the past week and were not specified to only include IBD-related pain.

## 2.2.4. Daily level of pain and interference

An adaptation of the e-Ouch diary, developed for patients with arthritis,<sup>33</sup> was used to assess daily pain characteristics and interference. This daily assessment covered any pain youth experienced and was not limited to only reporting IBD-related pain. After the timescale used in previous studies with the e-Ouch diary, youth were asked to complete the online diary twice a day (ie, upon waking and after school/before bed) for 10 consecutive days. For both entries, youth were asked to report on the current pain intensity (0 = "no pain" to 10 = "very much pain"), current pain interference (0 = "doesn't get in the way at all" to 10 = "totally gets in the way"), and the sensory-emotional characteristics of their pain by selecting the most applicable descriptors (eg, burning, cramping, pumping, like needles, sad, fearful, unbearable, and uncomfortable). In addition, during the evening diary, youth indicated the pain intensity experienced during the day, if they took any medication for the pain, and whether they had used any other self-management techniques to reduce the pain (eg, relaxation, distraction, and prayer).

## 2.3. Procedure

This study involved one visit to the Centre for Pediatric Pain Research at the IWK Health Centre. Upon arrival, participants were explained the purpose and all aspects of the study. Once consent by parent and assent by youth was given, parents and youth completed the questionnaires independently: parents completed the FACES-IV in the room adjacent to where youth completed all the above-described questionnaires assessing pain, HRQOL, and family functioning. A research assistant remained present with youth and provided assistance where needed. At the end of their participation, youth and parent were fully debriefed about the purpose of the study and were invited to participate in the online diary study. Each youth participants received a junior scientist certificate, and the family received \$30 for their participation. After everyone had completed the diary study, a raffle randomly determined the 3 youth who each received a \$25 movie gift card.

## 2.4. Statistical analysis

For the current study, the first 7 days of the 10-day diary were selected because youth were more compliant in completing the diary in the first week. This compliance observation is consistent with previous e-diary studies.<sup>8,33</sup> The 12 participants completed a total of 149 diary entries over the first 7 days. In accordance with Snijders' guidance, the 149 observations constitute an adequate sample size for obtaining reliable parameter estimates.<sup>30</sup> Descriptive statistics were used to summarize the pain characteristics reported in the diary. After the guidance by Giske et al.<sup>8</sup> on calculating reliable pain ratings and analytic techniques advised for e-Ouch data<sup>33</sup>, the daily pain ratings were averaged per week for these descriptive statistics. In addition, multilevel analyses, using maximum likelihood estimates of model parameters, were performed, using HLM version 6.01, with the aim to evaluate how

the pain experiences (ie, pain intensity now, pain intensity during the day, and pain interference) evolve throughout the week and explore the influence of the time of reporting (morning vs evening), demographic and disease characteristics, and family functioning. To this end, the variables “day” (ranging from 1 = day 1–7 = day 7) and “time of reporting” (0 = morning; 1 = evening) were entered in the first level. Age, sex, IBD type, and family functioning domains were entered in the second level. The multilevel analyses were conducted separately for each youth- and parent-reported family functioning domain.

Paired sample *t* tests were conducted to determine significant differences between youth and parent report for the various family functioning dimensions.

Hierarchical linear regression analyses were conducted to investigate the relative contribution of cohesion, flexibility, communication, and satisfaction in explaining youth’s pain intensity, pain interference, and HRQOL. Demographic and medical variables such as age, sex, IBD type, duration since diagnosis, and disease severity were controlled for in the first step. Level of cohesion, flexibility, communication, and satisfaction were entered in the second step. In the analyses with HRQOL, pain intensity and pain interference of the past week were also entered in the second step to allow for examination of the unique influence of pain and family functioning on HRQOL. When assumptions were met, potential indirect contributions of family functioning on HRQOL through pain experiences were explored using the PROCESS modeling tool by Hayes.<sup>13</sup> Separate analyses were conducted for parent- vs youth-reported family functioning. All regression analyses were performed for the entire sample as well as for the subsample of youth reporting abdominal pain. The variance-inflation factors of all regression analyses were acceptable (range: 1.04–3.82), revealing no problem with multicollinearity.

### 3. Results

#### 3.1. Descriptive statistics

Mean age of the youth was 13.37 years (SD = 2.54), with more males (*n* = 33, 55%) participating compared with females (*n* = 27, 45%). The majority of the youth (*n* = 41, 68.3%) were diagnosed with CD, followed by ulcerative colitis (*n* = 17; 28.3%) and IBD unclassified (UBD-U; *n* = 2; 3.3%). This distribution is reflective and representative of the population distributions of pediatric IBD<sup>10</sup> and comparable with other study samples.<sup>28,35</sup> The mean time since diagnosis was 37.42 months (SD = 33.69, range = 2–153). Most of the children had an inactive disease status at the time of study participation (*n* = 49, 81.7%) for those with an active disease status (*n* = 11, 18.3%).

Parental mean age was 45.30 years (SD = 6.13, range = 32–59 years). Most participating parents were biological mothers (*n* = 52, 86.7%), with *n* = 6 (10%) fathers and *n* = 2 (3.3%) others (ie, foster parent or custodial grandparent). Most parents were married/cohabiting (*n* = 37, 61.7%); had received education beyond the age of 18 years (*n* = 55, 91.67%); and were whites (*n* = 49, 81.7%).

The subsample of youth completing the diary component (*n* = 12), consisted of 42% (*n* = 5) females with an average age of 12.76 years. This sample did not differ from the entire participant sample (*n* = 60) on any characteristic with exception of the proportion with an active disease being slightly higher: 5 (42%) youth with an active disease status participated in the diary study.

Mean, SD, and range for all variables can be found in **Table 1**. Paired sample *t* tests revealed that youth reported significantly

lower levels of family cohesion ( $t(59) = -5.52, P < 0.001, CI: -1.78$  to  $-0.83$ ) and communication ( $t(59) = -2.91, P < 0.01, CI: -18.05$  to  $-3.35$ ), but higher levels of satisfaction ( $t(59) = 3.59, P < 0.01, CI: 5.50$ – $19.38$ ) compared with parents. There was no significant difference in levels of flexibility reported by youth and parents ( $t(59) = -1.98, P = 0.52, CI: -0.58$  to  $0.003$ ).

### 3.2. Pain experiences

#### 3.2.1. Questionnaire data

Only 6.7% of youth (*n* = 4) reported no pain in the past week. Looking only at the youth reporting pain in the past week, the average pain intensity was 4.04 (SD = 2.20, range: 1–10) with on average low pain interference (M = 1.30; SD = 2.22, range: 0–10). The most frequently reported pain was abdominal pain (*n* = 34, 60.7%), followed by lower back pain (*n* = 12, 21.4%), and headaches (*n* = 10, 17.9%) Half of the youth reporting pain (*n* = 28, 50%) reported pain in more than one location, and this was more than half (58.8%) for those youth reporting abdominal pain. The most frequent co-occurring pain symptoms were abdominal and back pain (*n* = 8; 23.5%), followed by abdominal pain and headaches (*n* = 6; 17.6%). Of those not reporting abdominal pain but who did report other pain symptoms (*n* = 19), neck pain (*n* = 5, 26.3%), headache (*n* = 4, 21.1%), and lower back pain were most commonly reported (*n* = 4, 21.1%).

#### 3.2.2. Diary data

On average, youth reported pain experiences on 4.55 days of the week, with the amount of pain days per youth ranging from 1 to 7 days. Strong correlations were found between the questionnaire-based pain reports, e-Ouch diary pain reports and HRQOL (**Table 2**). Pain in the abdominal area was most frequently reported (68%), followed by headaches (37.8%) and lower back pain (37.1%). For 30.7% of the diary entries, pain was present in 2 or more locations.

Both within and across youth, various descriptors were used to express the sensory and emotional characteristics of their pain

**Table 1**  
Mean, SD, and range for all variables.

	Mean	SD	Actual range
Pain intensity in the past week	3.77	2.35	0–10
Pain interference in the past week	1.22	2.17	0–10
Current pain intensity (diary report)*	3.91	2.49	1–10
Pain intensity during the day (diary report)*	4.67	2.84	0–10
Pain interference (diary report)*	2.39	2.67	0–10
IBD-specific HRQOL	78.08	10.65	57.86–97.86
Cohesion ratio (child report)	2.90	1.59	0–7.08
Flexibility ratio (child report)	1.72	0.95	0–5.71
Communication (child report)	58.58	25.07	10–99
Satisfaction (child report)	49.95	27.23	10–99
Cohesion ratio (parent report)	4.20	1.60	0.97–8.40
Flexibility ratio (parent report)	2.01	0.73	0.61–4.00
Communication (parent report)	69.28	21.98	10–99
Satisfaction (parent report)	37.51	9.46	10–83

\* The values reported for diary variables are based on the diary entries in which pain experiences were reported.

HRQOL, health-related quality of life; IBD, inflammatory bowel disease.

**Table 2**  
Correlation analyses between questionnaire-based pain reports, e-Ouch pain reports and HRQOL.

	1	2	3	4	5	6
1. Pain intensity in the past week	—	0.51**	0.51*	0.56*	0.42	-0.49**
2. Pain interference in the past week		—	0.45	0.44	0.50*	-0.27*
3. Current pain intensity (e-Ouch report)			—	0.91**	0.80**	-0.56*
4. Pain intensity during the day (e-Ouch report)				—	0.77**	-0.54*
5. Pain interference (e-Ouch report)					—	-0.62*
6. IBD-specific HRQOL						—

HRQOL, health-related quality of life; IBD, inflammatory bowel disease.

\* $P < 0.05$ ; \*\* $P < 0.01$ .

experiences. On average, 2.36 descriptors were used to describe the pain experience (SD = 1.84, range: 0–7 descriptors). The most commonly used descriptors were cramping (48%), uncomfortable (29.3%), hurting (22.7%), sharp (18.7%), and stinging (16%).

With respect to self-management strategies for pain, youth took medicine in 66.7% of pain instances and used other techniques to reduce pain in 83.3% of instances. On average youth reported using 1.53 self-management strategies per pain experience (SD = 1.52, range: 0–6). The most frequently used techniques were distraction (38.9%), rest/sleep (38.9%), talking with friends/family (16.7%), using cold (22.2%) or heat (16.7%), and massage/rubbing (11%).

The multilevel analyses revealed that none of the included variables showed a significant influence neither on current pain intensity (all  $t$ -values < 1.60,  $ns$ ) nor on current pain interference (all  $t$ -values < 1.34,  $ns$ ). For pain intensity experienced during the day, the variable “day” showed a robust influence (coefficient = -0.07,  $t = -2.10$ ,  $P < 0.05$ ) across analyses, indicating that youth reported less-intense pain experiences further along the week. In addition, parent-reported satisfaction with family functioning (coefficient = -0.15,  $t = -2.25$ ,  $P < 0.05$ ) showed a significant influence, revealing that youth report less-intense pain experience when their parents are more satisfied with the family system. None of the other variables showed a significant influence (all  $t$ -values < 1.43,  $ns$ ).

### 3.3. Linear regression analyses

#### 3.3.1. Relative contribution of family functioning and pain experiences in explaining youth-reported health-related quality of life

Hierarchical linear regression analyses investigating the relative contribution of youth-reported pain, cohesion, flexibility, communication, and satisfaction on HRQOL revealed a significant influence for child age, pain intensity in the past week, and satisfaction with family functioning: older youth, youth with higher levels of pain in the past week, and youth reporting lower satisfaction with their family functioning describe lower levels of HRQOL (Table 3A). In the subanalyses only including youth who report abdominal pain, only a significant negative influence for pain intensity in the past week was identified, with a trend for a positive influence of time since diagnosis (Table 3B).

With respect to parent-reported family functioning, pain intensity in the past week was the only variable showing a significant contribution to explaining HRQOL: higher levels of youth-reported pain intensity were related with lower youth-reported HRQOL (Table 3A). In the subanalyses only including youth who report abdominal pain, a trend remains for a positive

influence of cohesion and a negative influence of pain intensity in the past week (Table 3B).

#### 3.3.2. Relative contribution of family functioning in explaining youth-reported pain intensity and interference

Youth-reported family functioning did not show any significant contribution to the level of reported pain intensity in the past week (Table 4), nor to reports of abdominal pain intensity (Table 5).

By contrast, the analyses with parent-reported family functioning revealed the following contributing factors in explaining pain-intensity levels in the past week: diagnosis, cohesion ratio, and satisfaction with family functioning; Table 4. These findings indicate that higher reports of pain in the past week are found for youth diagnosed with CD as well as youth who have a parent reporting more unbalanced levels of cohesion and low levels of satisfaction with family functioning. Testing for indirect effects using the PROCESS modeling tool<sup>10</sup> revealed that the contribution of parent-reported cohesion and satisfaction on youth-reported HRQOL is mediated by their contribution to explaining

**Table 3**  
Hierarchical linear regressions investigating the relative contribution of pain intensity, pain disability, cohesion, flexibility, communication, and satisfaction in explaining HRQOL.

A) Step	Predictor	$\beta$	$\Delta R^2$	Adj. $R^2$	F Change
1	Child age	-0.32*	0.15	0.07	1.81
	Child sex	-0.12			
	Child diagnosis	0.00			
	Global assessment	-0.07			
	Time since diagnosis	0.12			
2	Cohesion ratio (youth report)	0.08	0.37	0.41	6.04***
	Flexibility ratio (youth report)	0.14			
	Communication (youth report)	-0.26			
	Satisfaction (youth report)	0.48*			
	Pain intensity (past week)	-0.44**			
	Pain interference (past week)	0.10			
B) Step	Predictor	$\beta$	$\Delta R^2$	Adj. $R^2$	F Change
1	Child age	-0.22	0.15	0.07	1.81
	Child sex	-0.16			
	Child diagnosis	0.11			
	Global assessment	-0.07			
	Time since diagnosis	0.12			
2	Cohesion ratio (parent report)	0.24*	0.26	0.27	3.40**
	Flexibility ratio (parent report)	-0.15			
	Communication (parent report)	-0.17			
	Satisfaction (parent report)	0.35 <sup>+</sup>			
	Pain intensity (past week)	-0.33*			
	Pain interference (past week)	0.08			

<sup>+</sup> $P = 0.08$ , \* $P < 0.05$ , \*\* $P < 0.01$ , \*\*\* $P < 0.001$ . Standardized betas from the last step in the analyses are displayed.

HRQOL, health-related quality of life.



**Table 4**  
Hierarchical linear regressions investigating the relative contribution of cohesion, flexibility, communication, and satisfaction in explaining pain intensity.

Step	Predictor	$\beta$	$\Delta R^2$	Adj. $R^2$	F Change
1	Child age	0.09			
	Child sex	0.21			
	Child diagnosis	-0.18			
	Global assessment	0.24+			
	Time since diagnosis	-0.07	0.15	0.07	1.91
2	Cohesion ratio (youth report)	-0.20			
	Flexibility ratio (youth report)	-0.20			
	Communication (youth report)	0.36			
	Satisfaction (youth report)	-0.04	0.07	0.08	1.12
Step	Predictor	$\beta$	$\Delta R^2$	Adj. $R^2$	F Change
1	Child age	-0.11			
	Child sex	0.23+			
	Child diagnosis	-0.32*			
	Global assessment	0.19			
	Time since diagnosis	-0.07	0.15	0.07	1.91
2	Cohesion ratio (parent report)	-0.37*			
	Flexibility ratio (parent report)	0.30+			
	Communication (parent report)	0.30			
	Satisfaction (parent report)	-0.50**	0.18	0.22	3.41*

+*P* = 0.05–0.08, \**P* < 0.05, \*\**P* < 0.01. Standardized betas from the last step in the analyses are displayed.

youth-reported pain intensity in the past week (cohesion: *ab* = 0.74, SE = 0.49, CI: 0.08–1.64; satisfaction: *ab* = 0.16, SE = 0.10, CI: 0.02–0.35).

In the subanalyses for abdominal pain, similar influences for diagnosis and cohesion ratio were found, but no influence of family satisfaction was observed (Table 5). Subsequent indirect effects for cohesion revealed that the contribution of parent-reported cohesion on youth-reported HRQOL is mediated by their contribution to explaining youth-reported pain intensity in the past week: *ab* = 0.80, SE = 0.46, CI: 0.01 to 1.89.

For both, youth- and parent-reported family functioning, only the global assessment score influenced the level of youth-

**Table 5**  
Hierarchical linear regressions investigating the relative contribution of cohesion, flexibility, communication, and satisfaction in explaining abdominal pain intensity.

Step	Predictor	$\beta$	$\Delta R^2$	Adj. $R^2$	F Change
1	Child age	0.03			
	Child sex	0.26			
	Child diagnosis	-0.26			
	Global assessment	0.21			
	Time since diagnosis	-0.19	0.24	0.10	1.74
2	Cohesion ratio (youth report)	0.12			
	Flexibility ratio (youth report)	-0.51			
	Communication (youth report)	0.18			
	Satisfaction (youth report)	0.03	0.11	0.10	0.96
Step	Predictor	$\beta$	$\Delta R^2$	Adj. $R^2$	F Change
1	Child age	-0.04			
	Child sex	0.34+			
	Child diagnosis	-0.35*			
	Global assessment	0.19			
	Time since diagnosis	0.31+	0.24	0.10	1.74
2	Cohesion ratio (parent report)	-0.45*			
	Flexibility ratio (parent report)	0.33			
	Communication (parent report)	0.001			
	Satisfaction (parent report)	-0.31	0.25	0.30	2.85*

+*P* = 0.05–0.08, \**P* < 0.05, \*\**P* < 0.01. Standardized betas from the last step in the analyses are displayed.

**Table 6**  
Hierarchical linear regressions investigating the relative contribution of cohesion, flexibility, communication, and satisfaction in explaining pain interference.

Step	Predictor	$\beta$	$\Delta R^2$	Adj. $R^2$	F Change
1	Child age	0.28+			
	Child sex	0.02			
	Child diagnosis	-0.15			
	Global assessment	0.32*			
	Time since diagnosis	-0.15	0.23	0.15	3.10*
2	Cohesion ratio (youth report)	-0.14			
	Flexibility ratio (youth report)	-0.16			
	Communication (youth report)	0.05			
	Satisfaction (youth report)	0.17	0.04	0.13	0.61
Step	Predictor	$\beta$	$\Delta R^2$	Adj. $R^2$	F Change
1	Child age	0.21			
	Child sex	0.01			
	Child diagnosis	-0.23+			
	Global assessment	0.29*			
	Time since diagnosis	-0.17	0.23	0.15	3.10*
2	Cohesion ratio (parent report)	-0.06			
	Flexibility ratio (parent report)	-0.07			
	Communication (parent report)	0.22			
	Satisfaction (parent report)	-0.21	0.03	0.12	0.54

+*P* = 0.05–0.08, \**P* < 0.05, \*\**P* < 0.01. Standardized betas from the last step in the analyses are displayed.

reported pain interference (Table 6): youth with an active disease status reported more pain interference. In the subanalyses looking at interference due to abdominal pain, only a significant negative influence of diagnosis was found in the analyses with parent-reported family functioning (Table 7): youth with CD report more interference compared to youth with ulcerative colitis.

#### 4. Discussion

Despite the fact that for most sample, their IBD was currently inactive, and the findings revealed that the daily pain experiences of youth with IBD are heterogeneous with respect to frequency,

**Table 7**  
Hierarchical linear regressions investigating the relative contribution of cohesion, flexibility, communication, and satisfaction in explaining abdominal pain interference.

Step	Predictor	$\beta$	$\Delta R^2$	Adj. $R^2$	F Change
1	Child age	0.33			
	Child sex	0.22			
	Child diagnosis	-0.31			
	Global assessment	0.28			
	Time since diagnosis	-0.15	0.36	0.25	3.09*
2	Cohesion ratio (youth report)	0.16			
	Flexibility ratio (youth report)	-0.08			
	Communication (youth report)	-0.25			
	Satisfaction (youth report)	-0.04	0.04	0.18	0.43
Step	Predictor	$\beta$	$\Delta R^2$	Adj. $R^2$	F Change
1	Child age	0.34			
	Child sex	0.26			
	Child diagnosis	-0.34*			
	Global assessment	0.21			
	Time since diagnosis	0.19	0.36	0.25	3.10*
2	Cohesion ratio (parent report)	-0.11			
	Flexibility ratio (parent report)	-0.04			
	Communication (parent report)	0.16			
	Satisfaction (parent report)	-0.46	0.16	0.34	1.92

+*P* = 0.05–0.08, \**P* < 0.05, \*\**P* < 0.01. Standardized betas from the last step in the analyses are displayed.

intensity, interference, sensory and emotional characterization, as well as their self-management strategies to deal with the pain. Although youth could report on any pain experience, most pain reports were consistent with typical IBD-related symptoms of cramping, stinging, or sharp abdominal pain. However, for most youth, these abdominal pain experiences were accompanied with other pain symptoms such as headache and lower back pain. Although time of reporting (morning vs evening) showed no influence on the pain characteristics, the reporting of the pain intensity experienced throughout the day was found to decline over the course of the week. The current findings further support earlier studies revealing abdominal pain as a common symptom in youth with IBD, even when the disease is inactive,<sup>9,11</sup> and go beyond this knowledge by providing a preliminary insight in the daily characteristics of these pain experiences rather than relying on recall of the pain experiences. The large prevalence of other pain symptoms, either in the absence of or in co-occurrence with abdominal pain, further iterates the relevance of being aware of the possibility that youth with IBD can experience chronic-on-acute pain, especially in youth with IBD now in remission. It is particularly noteworthy that the most common co-occurring pain symptoms in our sample, ie, headache and lower back pain, also represent the most prevalent primary pain disorders (ie, chronic pain symptoms not explained by a medical assessment<sup>7</sup>). These findings are similar to observations in youth with centrally mediated abdominal pain syndrome<sup>36</sup> and could therefore suggest that a similar mechanism, such as central sensitization, might underlie the persistence of abdominal pain experiences and co-occurring nonabdominal pain symptoms in both populations.<sup>31</sup> However, given the small sample of participants, especially in the diary component, these findings need further replication in larger samples before drawing strong conclusions. Nevertheless, these findings underline the need for a comprehensive, multidimensional assessment of pain experiences using real-time data capture, even when IBD is inactive, to provide appropriate and individualized pain management. Given that pain intensity, interference, and location as reported using the e-Ouch diary map well onto youth's questionnaire-based pain reports, this provides preliminary confirmation of the convergent validity of the e-Ouch diary in youth with IBD. With further testing, the e-Ouch diary could become a promising, clinically useful tool to gain a comprehensive perspective on youth's daily pain experiences as well as to evaluate the use and effectiveness of pain-relieving interventions.<sup>32,33</sup>

The need to address pain symptoms, irrespective of disease status, is further stressed by our findings of the crucial role pain plays in explaining youth's HRQOL. In particular, our findings provide crucial insights into the underlying mechanism of how pain experiences and family functioning contribute to youth's functioning. Small differences were found when including all pain experiences vs only abdominal pain experiences, but overall findings can be summarized as follows: (1) lower pain intensity was related to better HRQOL; (2) higher parent-reported balanced cohesion was related to lower pain intensity, which in turn was associated with better youth's HRQOL. These findings highlight that, despite the average low-pain levels, any pain experiences in youth with IBD play a key role in explaining youth's HRQOL, irrespective of disease status. No contribution of family functioning in explaining pain interference was observed. Pain interference was only influenced by the youth's disease status or diagnosis, ie, more interference due to any pain symptoms in youth with an active disease status and more interference due to abdominal pain in youth with CD. The observed low level of pain interference might have limited our ability to detect associations with family functioning.

Interestingly, a prominent role of satisfaction with family functioning was only observed when looking at any pain symptoms, with youth's satisfaction being related with better HRQOL and parental satisfaction showing an indirect relation with better HRQOL through its association with less pain intensity. This role of parent-reported family function was also observed in the diary data, with higher levels of satisfaction being related to lower levels of daily pain intensity. Because of the small sample size for the subanalyses with abdominal pain, limited power could potentially explain the absence of associations with family satisfaction. On the other hand, it is possible that the family dynamics at play for youth with IBD but without current abdominal pain symptoms are more similar to youth with primary pain disorders.<sup>7</sup> Further investigations in larger samples are needed to explore such potentially difference in the role of family dynamics in explaining youth's HRQOL depending on the type of pain experiences youth with IBD experience. Although future replication of the role of family satisfaction is needed, these findings provide preliminary support for a clinical focus on obtaining satisfaction with family functioning, rather than reaching a functional level of each individual dimension of family functioning, for improving HRQOL.

Importantly, these findings are largely in line with and extend previous evidence stressing the relevance of caregiver context in understanding HRQOL.<sup>14,15,27,28</sup> Furthermore, the differences found between youth and parent perspectives on family functioning and the subsequent diverse influence on youth's outcomes highlights the importance of soliciting multiple perspectives of family functioning and underscores how the family system is more than the sum of its parts.<sup>1,21</sup> In particular, the underlying mechanisms explaining youth HRQOL seem more complex from a parental perspectives with their view on family cohesion being associated with youth's HRQOL, through their contribution in explaining pain symptoms. This could indicate that, compared to youth, it is of more importance for parents to continue to strive for a functional level of warm relationships among the family members. This systemic approach of acknowledging the different perspectives within the family has the potential to guide and individualized family-based treatment for youth with IBD, which might require different targets and strategies for parents compared to those of youth.<sup>22</sup> Given the differences identified between youth and parent reports, future research is needed to investigate the perspective of other members of the family system to gain a more complete and accurate picture of the relevant family patterns to target within clinical practice.<sup>21</sup>

The current study has several limitations. First, the sample is relatively small, and most participating parents were mothers. Father's perspectives may differ from mothers, and so, additional work should aim to explore and contrast mother vs father reports using larger samples. In addition, most participating mothers were highly educated. Although this is reflective of many study samples, this might limit generalizability of the findings to families with lower education levels, given evidence indicating how family functioning might be influenced by parental education level.<sup>1</sup> Second, although our sample characteristics are similar to previous study samples in pediatric IBD,<sup>28,35</sup> it is important to consider that, for the majority of youth in our sample, their IBD was inactive at the time of participation. Hence the findings might not generalize to the entire IBD population, especially to youth who are experiencing a flare-up. Third, the data gathered are cross-sectional and therefore does not allow for drawing any causal conclusions. Fourth, the findings solely rely on self-report of family functioning with 2 dimensions of the FACES-IV showing low levels of internal consistency in the current sample. Although these scales were not involved in any key findings, the data involving these scales need to be interpreted with caution.

Although special consideration was taken to ensure the younger participants understood all the items, the FACES-IV is only validated for youth 12 years and older, so caution needs to be taken in interpreting results involving youth-reported family functioning. In addition, although our sample's age range is consistent with previous studies in pediatric IBD,<sup>28,35</sup> the broad age range could have potentially obscured different patterns of family functioning influences depending on the youth's developmental stage. Future studies in larger samples spanning the developmental range would benefit from including more objective, observational assessments of family functioning (eg, including of family discussion tasks, see Ref. 19) to shed more light on the role of family functioning across development. Observational assessments might also provide more opportunities to overcome the practical barriers of including all family member's perspective. Finally, no data are available on why only 30% of participants decided to take part in the optional diary part of the study. One potential barrier to diary participation could have been the need for a desktop computer with internet access to complete the diaries. Furthermore, the e-Ouch diary adopted in the current study is validated for youth with juvenile idiopathic arthritis,<sup>33</sup> but not for youth with IBD. Our data provide preliminary evidence for the convergent validity of e-Ouch in youth with IBD, but further validation of this assessment tool in a larger sample of youth with IBD is warranted.

Despite these limitations, the findings illustrate the heterogeneity in prevalence and types of daily pain experiences in youth with IBD, independent of disease activity, and the key role any type of pain experience, together with family functioning, plays in understanding youth's HRQOL. Given the diverse mechanism of how youth vs parental perspectives on family functioning influence youth's outcomes, the findings underscore the need for a family systems approach to improve functioning in youth with IBD.

## Disclosures

The authors have no conflict of interest to declare.

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