



Parental Psychological Factors and Quality of Life of Children With Inflammatory Bowel Disease

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Parental Psychological Factors and Quality of Life of Children with Inflammatory Bowel Disease

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Keywords: IBD; Health-Related Quality of Life; Children; Parents; Distress.

Abstract

Objectives: Parents have a central role in the management of children with inflammatory bowel disease (IBD). Alterations in parental psychological well-being may affect the patient's health-related quality of life (HRQoL). This study aimed to evaluate the correlation between maternal and paternal distress, anxiety, depression and pain catastrophizing and the HRQoL of patients with IBD.

Methods: Children with IBD aged 8-18 years and their parents were prospectively recruited. Children answered questionnaires on HRQoL while parents completed an assessment of distress, anxiety, depression and pain catastrophizing. Univariate and multivariate regression models analysis were used to evaluate correlations between parental measures and patient's HRQoL and between the factors related to children health and parental psychological suffering.

Results: One hundred patients (45 Crohn's disease, 55 ulcerative colitis), 90 mothers and 62 fathers were enrolled. Parents had high levels of distress while anxiety, depression and pain catastrophizing levels were relatively low. Parental distress had the most substantial correlation with children's HRQoL and was associated with patients' disease activity and recent flares. On multivariate regression analysis, parental factors explained less than 20% of the variance in the children's HRQoL scores. Mothers suffered from psychological alterations more frequently than fathers, but the parental inter-rater agreement was strong in regards to distress and anxiety.

Conclusions: Parental distress is high and correlates with the HRQoL of children with IBD. Interventions aimed at evaluating and managing parental distress should be considered during the management of children with IBD.

Keywords: IBD; Health-Related Quality of Life; Children; Parents; Distress.

What is known

- Parents of children with chronic illnesses report increased distress levels and experience anxiety, depression and pain catastrophizing thoughts.
- Parental psychological suffering may have a negative influence on the Health-Related Quality of Life (HRQoL) of the child.

What is new

- Parents of children with inflammatory bowel (IBD) have high levels of distress, and this has the most significant correlation with IBD children's HRQoL compared with anxiety, depression and pain catastrophizing.
- Mothers and fathers of IBD children experience similar levels of psychological suffering.

Background

Inflammatory bowel disease (IBD), including Crohn's disease (CD) and ulcerative colitis (UC), are diagnosed during childhood and adolescence in about 30% of the cases.

The chronic nature of the disorder, characterized by painful, embarrassing and disabling symptoms, requiring frequent medical evaluations and treatments, has a significant impact on the physical and psychosocial development of the young patients and on their family members, who are the first-line source of support in everyday life. (1) Indeed, the diagnosis of chronic illness in a close relative, especially that of a child, is one of the most significant stressors in a person's life. (1,2) Parents may go through negative feelings, such as guilt, anger and shame, worry about the unpredictable course of the disease and their child's future and experience a change in their way of life due to the need to support their chronically ill child.

Parents of children with IBD are reported to have high levels of distress, a negative mental status that impacts on functioning, (3-5) with an increased risk of mood disorders such as anxiety and depression. (6) Moreover, since pain is a frequent patient's issue, parents may experience pain catastrophizing thoughts, a particular cognitive distortion that makes the person always expecting the worst possible outcome. (7) Remarkably, parental anxious and depressive symptoms, exacerbated by distress and maintained by catastrophizing, negatively impact on the whole family functioning (8) and on how each family member experiences the disease. In this bidirectional relationship, parents' impaired psychosocial functioning has a negative influence on children's health-related quality of life (HRQoL) with higher adolescent depressive symptoms and more negative IBD health outcomes. (9-11)

So far, the interaction between each parental psychological domain has not been evaluated, and little is known about the correlation of parental pain catastrophizing with the HRQoL of children with IBD.

This study aimed at assessing the interactions between distress, anxiety, depression and pain catastrophizing in mothers and fathers of young patients with IBD and their correlation with children's HRQoL. As secondary outcomes, correlations between the IBD characteristic's and the parental psychological conditions and the inter-parent agreement on the psychological disturbances were evaluated.

Material and Methods

Participants

Patients aged 8-18 years with an established diagnosis of CD or UC for at least 6 months, according to the revised Porto criteria, (7) and their parents were prospectively enrolled at three pediatric gastroenterology facilities in Italy (Institute for Maternal and Child Health "Burlo Garofolo", Trieste; Ca' Foncello's Hospital, Treviso; Gaslini Children's Hospital, Genoa), from January 1st, 2018 to July 30th, 2019.

Patients with previous colectomy or clinical conditions that required urgent treatment were excluded from the study. For both children and parents, cognitive or developmental delay, known diagnosis of anxiety disorder or depression, or inadequate Italian language knowledge were used as additional exclusion criteria.

Data collection

Patient and parents' characteristics

Sociodemographic data were collected at the time of the enrollment. For each participating parent, information included: age, sex, education level, employment status, marital status, and the number of children. Patients information included: age, sex, disease duration, IBD phenotype according to the Paris classification,(12) presence and intensity of abdominal pain in the previous two weeks, current disease activity according to weighted Pediatric Crohn's Disease Activity Index (wPCDAI)(13) and Pediatric Ulcerative Colitis Activity Index (PUCAI) (14), fecal calprotectin, number of flares of IBD in the past 12 months (from the time of questionnaire completion), current medication, and previous IBD-related surgery.

Disease activity was defined: in remission (wPCDAI <12.5/PUCAI <10), mild (wPCDAI 12.5–40/ PUCAI 10–34), moderate (wPCDAI 40–57.5/ PUCAI 35–65), and severe (wPCDAI >57.5/ PUCAI >65). A wPCDAI>12.5 or PUCAI>10, with the need for hospitalization or treatment intensification (start of a new medical treatment or dose escalation of ongoing treatment) defined a flare of IBD.

Patient's Health-related Quality of Life

The Italian version of IMPACT III was used to assess the HRQoL in children. IMPACT III is a 35-item, validated self-administered pediatric IBD-specific questionnaire, using a 5-point Likert scale to evaluate the extent to which the patient is affected by specific issues in six domains: bowel symptoms, systemic symptoms, emotional functioning, social functioning, body image, and treatment interventions. Lower scores indicate poorer HRQoL, while higher scores indicate better HRQoL. (15) Scores on the IMPACT-III were linearly transformed to a range of 0–100. (16)

Parental Distress

Parental distress was measured using the Distress Thermometer (DT), a brief screening tool that has been shown to identify the parental distress level over the past week. (17) It consists of a "thermometer" ranging from 0 (no distress) to 10 (extreme distress) on which parents describe their overall distress. The score of 4 or higher detects clinically significant distress. (18)

Parental anxiety and depression

Parental anxious and depressive symptoms were measured with the Hospital Anxiety and Depression Scale (HADS), a questionnaire that comprises 14 items, seven related to anxiety (HADS-A) and seven to depression (HADS-D). Parents were considered to have abnormal anxiety or depression based respectively on the HADS-A or the HADS-D subscores ≥ 11 . (19)

Patients' pain and Parental pain catastrophizing

Children's pain was measured using the Numerical Rating Scale (NRS), a 0 to 10 scale to assess self-reported pain intensity over the last two weeks, with 0 indicating no pain, and 10 reflecting worst possible pain. (20)

Mother's and father's catastrophic thinking about their child's pain was evaluated by the parent version of the Pain Catastrophizing Scale (PCS-P), a validated 13-item, self-administered questionnaire that identifies different levels of catastrophizing, with greater scores indicating a more severe catastrophizing. (21,22) Scores ≥ 30 suggest a clinically relevant catastrophizing. (23)

Statistical Analyses

Continuous variables were tested for normality with the Shapiro-Wilk test, compared with the unpaired t-student test if normally distributed, otherwise with the non-parametric Mann-Whitney test. Data were summarized by median and interquartile ranges (IQR). Kruskal-Wallis test was

used to compare continuous parental scores (DT, HADS-A, HADS-D, PCS-P) respect to numbers of flares (0; 1; ≥ 2). Post-hoc tests, pairwise comparisons using the Mann-Whitney test, were conducted and corrected using the Holm method. Categorical variables were reported as absolute and relative frequencies and compared with the Chi-Squared test or Fisher exact test when appropriate.

To determine whether psychological factors and disease activity were related to HRQoL, Spearman correlation coefficient (ρ) was calculated.

To estimate the association between potential predictors and impact on HRQoL of children, univariate linear regression models were estimated for each parameter of age, sex, illness duration, disease activity related to children, and distress, anxiety, depression and catastrophizing related to parents.

The final subset of predictors was selected through the list of variables that resulted in being significantly associated with the outcome of univariable analyses ($p < 0.10$).

Subsequently, two multivariate linear regression analyses were performed: one model included only DT, HADS-A, HADS-D, PCS-P of mothers and the second the same variables of fathers.

We respected the requirement of a minimum of only two subjects per variable in the model to guarantee unbiased estimation of beta coefficients and R-squared. (24)

Intraclass correlation coefficients (ICCs) were determined to study the inter-rater agreement between parents. The two-way random average measures form for ICC was used. (25) The average agreement was selected when calculating the ICC. The level of agreement was evaluated by the ICC estimate's 95% confidence interval. The following classification was used: ICC values < 0.40 = poor agreement, values between 0.40 and 0.59 = fair agreement, values between 0.60 and 0.74 = good agreement, and values > 0.75 = excellent agreement. (26)

Statistical analyses were performed using R software version 3.4.3 (2017) The R Foundation for Statistical Computing, and a p-value <0.05 was considered to indicate statistical significance.

Ethical Considerations

Children and parents were asked to complete the study questionnaires after permission from the child and written informed consent from parents were obtained. A trained researcher not involved in the clinical care of the patients entered the data from the questionnaires into a database. The local Ethics Committee approved the study.

Results

Patients and parents characteristics

One hundred and five patients and their parents were screened to join the study. Four patients declined the invitation while one was excluded because she had undergone total colectomy.

One hundred patients were enrolled (response rate: 95.2%). The median age was 14.9 years (IQR 12.3-16.3); 45 children had CD, and 55 had UC. The majority of participants were male (60.0%). Most of the patients were in a quiescent state of disease, with no flares in the past 12 months: 37 (82.2%) and 34 (61.8%) children and adolescents with CD and UC were in clinical remission, respectively. There were no patients with severe disease. At the time of assessment, 21.0% of children reported abdominal pain over the last two weeks, with a median NRS of 5.0 (IQR 4.0-6.0) and a median duration of 2 months (IQR 1-3 months).

Ninety mothers and 62 fathers were recruited; the median age was 47.0 years (IQR 42.0-50.0) and 50.0 years (IQR 45.0-54.0), respectively (p<0.001). Additional patients' and parents' characteristics are reported in Table 1.

Patients' Health-related Quality of Life

Median HRQoL total score was 78.2 (IQR 67.9-85.9) with body image domain having the lowest score (median 66.7, IQR 58.3-83.3) and bowel symptoms domain registering the highest score (median 82.1, IQR 67.9-92.9). No significant difference was found between patients with CD and UC.

Parental distress, anxiety, depression and pain catastrophizing

Table 2 details parental DT, HADS and PCS-P. Median parental DT score was 5.0 (IQR 2.0-6.0) in the mothers' group and 3.0 (IQR 1.0-5.8) in the fathers' group ($p=0.05$); 57 (63.3%) mothers and 29 (46.8%) fathers reported levels of distress ≥ 4 ($p=0.04$).

Mothers had HADS-A scores ≥ 11 more commonly than fathers (18, 20.0% vs 4, 6.4%, $p = 0.02$).

Few parents had HADS-D scores ≥ 11 (8 (8.4%) mothers, 4 (6.4%) fathers, $p = 0.35$).

Median parental pain catastrophizing level by median PCS-P score was 14.0 (IQR 9.3-23.0) for mothers and 11.5 (IQR 6.0-22.8) for fathers ($p=0.40$); 9 (10.0%) mothers and 8 (12.9%) fathers showed clinically relevant pain catastrophizing ($p=0.58$).

Parental psychological conditions and patient's IBD status

All father's psychological variables showed a significant correlation with children's disease activity in UC, but not in CD: father's DT score had the strongest correlation with PUCAI score ($p=0.58$, $p < 0.001$). Mother's DT score was significantly correlated to disease activity both in UC ($p=0.36$, $p=0.009$) and CD ($p=0.33$, $p=0.03$). The remainder of the maternal variables did not show any significant correlation with disease activity.

Maternal and paternal distress was associated with calprotectin levels ($p=0.006$, $p=0.01$, respectively). The number of flares in the past year was also associated with higher DT levels both in mothers ($p < 0.001$) and fathers ($p = 0.01$): median maternal and paternal DT scores were

3.6 (IQR 1.0-5.3) and 2.0 (IQR 1.0-5.0) in the group with no flares and 5.8 (IQR 5.0-7.0) 5.0 (IQR 3.0-6.5) in the group with at least 2 flares ($p=0.01$ for mothers, $p=0.03$ for fathers).

No correlation between disease duration and all the declared variables was found.

The DT scores were associated with the PCS-P scores, both in mothers ($\rho=0.41$, $p < 0.001$) and fathers ($\rho=0.35$, $p=0.005$) while no significant association between PCS-P scores and children's NRS scores was found (PCS mother, $p=0.71$; PCS father, $p=0.96$).

Correlations between HRQoL scores and parental psychological factors

All the declared variables showed low-moderate correlation with HRQoL ($-0.27 < \rho < -0.45$ and all statistically significant p -values). Parental distress had the strongest correlation with HRQoL ($\rho = -0.35$ considering mother's distress, $\rho = -0.45$ considering father's distress).

Univariate and multivariate regression linear model of HRQoL

The univariate linear regression models showed that mother's and father's DT, mother's and father's HADS-A, mother's HADS-D and father's PCS-P were significantly related to the HRQoL score. Mother's and father's DT had the most substantial correlation with the HRQoL score. On multivariate regression analysis, only mother's DT (standardized $\beta = -0.22$) and father's DT (standardized $\beta = -0.31$) were significantly associated with HRQoL. The model with maternal factors explained 13.7% of the variance of HRQoL score ($R = 0.37$, $p = 0.02$), the model with paternal factors explained 19.4% of the variance of HRQoL score ($R = 0.44$, $p = 0.01$) (Table 3).

Parental agreement

For 52 patients, both parents were available. The inter-rater agreement between mother and father on different psychological questionnaires are presented in Table 4.

The highest ICCs were found for the DT scale and the HADS-A scale.

Discussion

This study shows that parents, especially mothers, of children with IBD had high levels of distress and that distress was significantly associated with child's HRQoL. Pain catastrophizing was experienced by a few parents, but it was associated with higher distress levels.

The significant association with parental distress and patients' functional outcome confirms previous observations suggesting that higher levels of parental distress are related to lower levels of HRQoL in children (4,10)

Approximately half of both mothers and fathers of children with IBD reported values of clinically relevant distress, consistently with previous studies. (4,5) Levels of anxiety and depression were relatively low and, although not directly comparable with other populations of parents of children with IBD, consistent with what has been observed in parents of children with other chronic diseases. (27) No significant difference was found between the parents of children with UC and those with CD.

Parental distress was influenced by the child's clinical conditions at the time of the visit and the number of relapses in the last year but not by the duration of the disease in general. Similar findings were reported by Diederer and colleagues (5) suggesting that recent flares have a significant impact on parental distress and that it may take many months for distress level to return to a baseline.

Of note, distress levels in parents were high, even though most of the patients were in clinical remission. The excessive concern of parents could be due to a misperception about the state of health and an underestimation of the quality of life of the child (28), in particular in the emotional functioning domains,(16) to the responsibilities for management of a chronic illness (2) or the difficulties in coping with the fear of a disease evolution.(5)

About 10% of parents experienced a clinically relevant pain catastrophizing thought. Parental pain catastrophizing was associated with increased parental distress levels, as already reported (29) but did not correlate with patients' reported pain as observed in other conditions with

chronic pain, in which the parental cognitive biases seem less influential than the patients own maladaptive pain-related cognitions. (30) The correlation between pain catastrophizing levels and children's HRQoL was only moderate. To our knowledge, only one study evaluating parental pain catastrophizing in pediatric IBD has been published. It showed that parents experiencing higher levels of catastrophic cognitions are frequently distressed, and their daughters have lower rates of participation in treatment intervention programs for psychosocial difficulties. (31)

In this series, children's HRQoL levels were similar to other cohorts of pediatric IBD patients. (16,32,33) and body image scores were low as already reported in other groups of children and adolescents with chronic illnesses.(34)

In the multivariate analysis, the model including all the parental psychological factors explained less than 20% of the variance of children's HRQoL. Stronger correlations were found by examining the relationship between children's psychological factors and their functional outcomes (23,35) suggesting that children mediate the impact of parental feelings and that their HRQoL depends on their adaptative strategies more than on parental influence.

This is one of the few studies that evaluated the psychological aspects of fathers and made comparisons between parents. Fathers expressed lower levels of distress, anxiety and depression than mothers, as reported in other chronic illnesses,(27) possibly because they were not the principal caregiver of the patient. However, the inter-rater agreement analysis showed that mothers and fathers experience distress and anxiety in similar ways, as suggested by preliminary evidence. (36)

These findings should be interpreted by recognizing some limitations. The cross-sectional nature of this study exclusively allows to evaluate associations but not to drawn causal inferences. We tried to limit this bias analyzing, not only the association between the children HRQoL and the parental psychological factors, but also the relationship with the latter and the features of the child's disease.

This study did not include siblings to control for shared familiar confounders (36) nor a control population and no direct comparison with healthy children and their parents can be drawn.

Life events not related to IBD were not taken into account but the questionnaires were focused on children's health, limiting the possibility of confounding influences on answers.

Moreover, the correlation between each parental parameter and the corresponding for the child was not evaluated, but HRQoL was chosen as the unitary patient-centred outcome.

Finally, most of the patients included were in clinical remission and were enrolled in an outpatient setting. Thus, these findings cannot be generalized to parents and patients with active disease. This limitation may also explain the low rates of clinically relevant anxiety disorders and depression.

In conclusion, the findings of this study suggest that the psychological alterations in the parents of children with IBD should be investigated in order to give support for their suffering and to provide benefit not only to the parents themselves but also to the child's HRQoL. Interventions aimed at managing parental stress should be considered as a helpful tool during the management of children with IBD.

Although studies on IBD are not yet available, cognitive behavioural and problem-solving therapies were shown to be effective in improving the mental health status of parents of children with chronic diseases.(38)

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Table1. Patients' and parents' characteristics

	IBD	CD	UC
Patients	100	45	55
Age (years) - Median (IQR)	14.9 (12.3-16.3)	14.9 (12.4-16.3)	14.6 (12.4-16.0)
Male sex - n (%)	60 (60.0)	31 (68.9)	29 (52.7)
Familiarity for IBD - n (%)			
Parents	2 (2.0)	1 (2.2)	1 (1.8)
Siblings	4 (4.0)	2 (4.4)	2 (3.6)
Duration of IBD (years) - Median (IQR)	3.1 (1.3-6.5)	3.6 (1.6-5.5)	2.8 (1.3-7.1)

Location (Paris classification)			
# - n (%)			
L1		13 (28.9)	/
L2		5 (11.1)	/
L3		26 (57.8)	/
L4a		14 (31.1)	/
L4b		5 (11.1)	/
E1		/	9 (16.4)
E2		/	5 (9.1)
E3		/	0
E4		/	41 (74.5)
Scores and inflammatory markers - Median (IQR)			
wPCDAI		7.5 (0.0-10.0)	/
PUCAI		/	0.0 (0.0-15.0)
Faecal calprotectin [§]	210.0 (30.0-825.0)	200.0 (27.0-830.0)	211.0 (33.0-819.8)
wPCDAI > 57.5, n (%)	0	0	/
PUCAI > 65 n (%)	0	/	0

Patients with flares in the past 12 months - n (%)			
No flares	56 (56.0)	28 (62.2)	28 (50.9)
1 flare	30 (30.0)	10 (22.2)	20 (36.4)
≥2 flares	14 (14.0)	7 (15.6)	7 (12.7)
Current medication for IBD° - n (%)			
Enteral Nutrition	8 (8.0)	8 (17.8)	0
Corticosteroids	8 (8.0)	4 (8.9)	4 (7.3)
5-Aminosalicylate	36 (36.0)	1 (2.2)	35 (63.6)
Thiopurine	13 (13.0)	7 (15.6)	13 (23.6)
Methotrexate	7 (7.0)	5 (11.1)	2 (3.6)
Anti-TNF	36 (36.0)	28 (62.2)	8 (14.5)
Vedolizumab	1 (1.0)	0	1 (1.8)
Thalidomide	13 (13.0)	6 (13.3)	7 (12.7)
Previous IBD-related surgery, n (%)	10 (10.0)	10.0 (22.2)	0
Parents	152	73	79

Mothers	90	41	49
Fathers	62	32	30
Age (year) median (IQR)	48.0 (43.0-51.0)	49.0 (44.0-52.0)	47.0 (43.0-50.0)
Education level, n (%)			
Degree	24 (15.8)	8 (11.0)	16 (20.3)
High school	84 (55.3)	40 (54.8)	44 (55.7)
Middle school	41 (27.0)	23 (31.5)	18 (22.8)
Primary school	3 (1.9)	2 (2.7)	1 (1.3)
Employment status, n(%)			
Self-employed	21 (13.8)	12 (16.4)	9 (11.4)
Employee	101 (66.4)	46 (63.1)	55 (69.6)
Unemployed/Housewife	25 (16.4)	12 (16.4)	13 (16.5)
Retired	5 (3.4)	3 (4.1)	2 (2.5)
Marital status, n (%)			
Unmarried	12 (7.9)	5 (6.8)	7 (8.9)
Married	119 (78.3)	54 (74.0)	65 (82.2)
Divorced	20 (13.2)	13 (17.8)	7 (8.9)
Widow/widower	1 (0.6)	1 (1.4)	0
Number of children median (IQR)	2.0 (1.0-2.0)	2.0 (1.0-2.0)	2.0 (1.0-2.0)

IBD: inflammatory bowel disease; IQR: interquartile range; CD: Crohn's disease; UC: ulcerative colitis.

* L1: distal 1/3 ileum $\pm$ limited cecal disease; L2: colonic; L3: ileocolonic; L4a: upper disease proximal to the ligament of Treitz; L4b: upper disease distal to the ligament of Treitz and proximal to distal 1/3 ileum.

E1: proctitis; E2: left-sided disease; E3: extensive disease; E4: pancolitis.

§ Data from 12 (26.7%) CD and 15 (27.3%) UC patients were missing.

° One (2.2%) CD and 2 (3.6%) UC patients did not have any medication at the time of assessment.

Table 2. Parental distress, anxiety, depression and pain catastrophizing.

	IBD (n = 172)	CD (n = 73)	UC (n = 79)
DT			
Median (IQR)	4.0 (2.0-6.0)	5.0 (2.0-6.0)	4.0 (2.0-6.0)
HADS-A, n (%)			
No anxiety (score 0-7)	90 (59.2)	43 (59.0)	47 (59.4)
Borderline anxiety (score 8-10)	41 (27.0)	25 (34.2)	16 (20.3)
Anxiety (score ≥ 11)	21 (13.8)	5 (6.8)	16 (20.3)
HADS-D, n (%)			
No depression (score 0-7)	116 (76.3)	56 (76.7)	60 (76.0)
Borderline depression (score 8-10)	24 (15.8)	13 (17.8)	11 (13.9)
Depression (score ≥ 11)	12 (7.9)	4 (5.5)	8 (10.1)
PCS-P			
Median (IQR)	14.0 (7.0-23.0)	13.0 (8.0-22.0)	15.0 (7.0-25.0)
Clinically relevant (score ≥ 30), n (%)	17 (11.2)	6 (8.2)	11 (13.9)

IBD: inflammatory bowel disease; IQR: interquartile range; CD: Crohn's disease; UC: ulcerative colitis.

Table 3. Parental multivariate regression model

HRQoL	Beta	SE	p-value	Beta Stand
Mothers (n=90)				
constant	86.23	3.72	0.000	.
DT	-1.20	0.64	0.063	-0.22
HADS-Anxiety	-0.44	0.71	0.540	-0.10
HADS-Depression	-0.39	0.61	0.520	-0.10
PCS-P	-0.02	0.15	0.902	-0.02
Fathers (n=62)				
HRQoL	Beta	SE	p-value	Beta stand
constant	88.61	3.33	0.000	
DT	-1.47	0.65	0.027	-0.31
HADS-Anxiety	-0.43	0.64	0.505	-0.11
HADS-Depression	0.34	0.65	0.609	0.08
PCS-P	-0.24	0.18	0.191	-0.20

For mothers, the model explained 13.69% of the variance of the HRQoL score ($R=0.37$, $p=0.02$)

Only DT is associated with HRQoL ($\beta = -1.20$). For a unit increase of DT, the HRQoL score decreased by 1.20.

For fathers, the model explained 19.36% of the variance of the HRQoL score ($R=0.44$, $p=0.01$).

Only DT is associated with HRQoL ($\beta = -1.47$). For a unit increase of DT, the HRQoL score decreased by 1.47.

Table 4: Parental inter-rater agreement for questionnaires scores

	Mother and Father ratings (n=52)			
		95% Confidence Interval		
Questionnaire	ICC	Lower	Upper	p-value
TD	0.60	0.30	0.77	<0.001
HADS-A	0.60	0.29	0.77	<0.001
HADS-D	0.24	-0.31	0.57	0.16
PCS-C	0.42	-0.02	0.66	0.03

ICC = Intraclass Correlation Coefficient. Results of ICC are calculated using average agreement, two-way random-effects model. Interpretations of ICC values: values < 0.40 = poor agreement, values between 0.40 and 0.59 = fair agreement, values between 0.60 and 0.74 = good agreement, values > 0.75 = excellent agreement