



The Role of Emotional and Behavioral Difficulties and Attachment Styles in Adolescent Inflammatory Bowel Disease and Functional Gastrointestinal Disorders

Alexandros Kalavris^{1*}; Kalliopi Triantafyllou¹; Stylianos Christogiorgos¹; Danae-Evangelia Mavragani¹; Niki Skopeliti³; Alexandra Papadopoulou^{2#}; Gerasimos Kolaitis^{1#}

¹Department of Child Psychiatry, School of Medicine, National and Kapodistrian University of Athens, 'Aghia Sophia' Children's Hospital, Athens, Greece.

²Division of Gastroenterology and Hepatology, First Department of Pediatrics, School of Medicine, National and Kapodistrian University of Athens, 'Aghia Sophia' Children's.

³National and Kapodistrian University of Athens, Athens, Greece.

#Co-senior authorship

*Corresponding Author(s): Alexandros Kalavris

Department of Child Psychiatry, School of Medicine, National and Kapodistrian University of Athens, 'Aghia Sophia' Children's Hospital, Thivon & Papadiamantopoulou, 11527, Athens, Greece.
 Email: kalavrisalex@med.uoa.gr

Abstract

Objective: Inflammatory Bowel Disease (IBD) and Functional Gastrointestinal Disorders (FGIDs) have been associated with emotional and behavioral difficulties in adolescence. Insecure attachment has been found to be correlated with IBD in adults. This study explores attachment disturbances and emotional and behavioral difficulties in adolescents with IBD and FGIDs.

Methods: Forty (40) adolescents with IBD (mean age: 14.4 years; 18 males), 31 with FGIDs (mean age: 13.3 years; 15 males) and 71 healthy controls (mean age: 13.1 years; 29 males) and their parents participated in the study. They filled out the Parental Bonding Instrument (PBI), the Functional Disability Inventory (FDI), the Abdominal Pain Index (API), the Pain Catastrophizing Scale (PCS) and the Strengths and Difficulties Questionnaire (SDQ).

Results: Adolescents with FGIDs reported more emotional difficulties compared to those with IBD or healthy controls ($p=0.010$ and $p=0.001$, respectively), while parents' reports from both clinical groups showed higher scores in almost all SDQ subscales. Adolescents with FGIDs experienced higher level of total SDQ difficulties and reduced functional capacity compared to those with IBD ($p=0.023$ and $p=0.004$, respectively). Adolescents with IBD and FGIDs did not report significantly different rates of insecure attachment to mother or to father when compared to control group.

Received: June 13, 2023

Accepted: Jul 25, 2023

Published Online: Jul 31, 2023

Journal: Journal of Psychiatry and Behavioral Sciences

Publisher: MedDocs Publishers LLC

Online edition: <http://meddocsonline.org/>

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Keywords: Adolescents; Attachment; Emotional/behavioral difficulties; Functional gastrointestinal disorders; Inflammatory bowel disease.

Cite this article: Kalavris A, Triantafyllou K, Christogiorgos S, Mavragani DE, Skopeliti N, et al. The Role of Emotional and Behavioral Difficulties and Attachment Styles in Adolescent Inflammatory Bowel Disease and Functional Gastrointestinal Disorders. J Psychiatry Behav Sci. 2023; 6(2): 1086.



Conclusions: Adolescents with FGIDs or IBD experience more emotional difficulties and their parents report more behavioral difficulties, compared to healthy peers. Patients with FGIDs are at higher psychosocial risk than those with IBD. Psychological assessment and early intervention where appropriate, should be incorporated in their clinical management and follow up.

Introduction

Inflammatory Bowel Disease (IBD) refers to Crohn's disease and ulcerative colitis, which are chronic gastrointestinal disorders. The pediatric prevalence of IBD has risen to 77/100,000 in 2016 [1]. Functional Gastrointestinal Disorders (FGIDs) are the most common diagnoses in gastroenterology and the mean prevalence of FGIDs for children aged over four years was 21.8% [2]. Both IBD and FGIDs in children and adolescents may have potential negative impacts on developmental processes and psychosocial well-being [3,4].

In particular, studies have reported emotional and behavioral difficulties in adolescents with IBD [5,6]. A systematic review with a meta-analysis [7] demonstrated the manifestation of anxiety and depressive symptomatology in pediatric patients with IBD. FGIDs also have been related to anxiety and depression. Anxiety disorders were found to be more frequent in patients with functional abdominal pain, compared to healthy controls, while lifetime risk for depressive disorder was significantly higher as well [8]. In addition, patients with Irritable Bowel Syndrome (IBS) have a three-fold increased odds of anxiety and depression, compared to healthy subjects [9].

Disease activity and adolescent internalizing symptoms found to predict pain in IBD [10]. Pain severity and pain catastrophizing may also result in poorer functioning of young patients with IBD [11]. Comparing IBD with FGIDs, adolescents with FGIDs had significantly higher rates of functional disability, abdominal pain and pain catastrophizing, compared to adolescents with IBD [12].

Moreover, the quality and the type of attachment have been known to contribute to the emergence of psychopathology [13] as well as physical illness. In particular, patients with insecure attachment types (avoidant or ambivalent) report more physical symptoms, compared to those with secure attachment type [14]. A literature review by Maunder and Hunter [15] supports the contribution of insecure attachment to the onset of physical illness. Studies in adult populations reported correlations between inadequate parenting and the development of IBD [16,17]. Also, recent findings indicate that maternal psychological factors are correlated with the onset of infant FGIDs in offspring [18]. However, attachment has been examined mainly in adults with IBD and, to the authors' knowledge, there are no studies that have examined attachment in adults or adolescents with FGIDs.

As psychogastroenterology explores further brain-gut connection, psychosocial factors have been associated with gut diseases [19]. The impact of organic as well functional gastrointestinal disorders on psychological status are also of particular interest. The aims of the present study therefore were to examine emotional and behavioral difficulties as well as attachment disturbances in adolescents with IBD and FGIDs. The relationship between functional abdominal pain, functional disability and pain catastrophizing was also examined.

Our hypotheses are summarized as follows: 1) Adolescents with IBD will report higher levels of insecure attachment and emotional and behavioral difficulties compared to healthy peers, assuming IBD activity to be correlated with the above-mentioned psychosocial correlates, and 2) Adolescents with FGIDs will present higher levels of functional abdominal pain, functional disability, emotional and behavioral difficulties compared to adolescents with IBD.

Material and Methods

Study design

This cross-sectional case-control study has been conducted in the Pediatric Gastroenterology, Hepatology and Nutrition Unit of a General Children's Hospital and in two Schools. Ethical approval for the clinical groups of the study was obtained by the Scientific Council of the General Children's Hospital prior to data collection, while ethical approval from the Ministry of Education, Research and Religions Affairs was obtained from the control group.

Participants

Forty (40) adolescents (mean age \pm SD: 14.4 \pm 1.8 years; 18 males) with IBD, ii) 31 adolescents (mean age \pm SD: 13.3 \pm 1.4 years; 15 males) with FGIDs and iii) 71 healthy high school students (mean age \pm SD: 13.1 \pm 1.6 years; 29 males) and their parents participated in the study. The clinical groups were recruited from the outpatient General Gastroenterology and IBD Clinics of the Division of Gastroenterology, Hepatology & Nutrition of the Department of Pediatrics at a General Children's Hospital, and the National Society of Crohn's disease and ulcerative colitis patients. The inclusion criteria for participation were as follows: (1) adolescents aged between 11 and 17 years; (2) diagnosis by a pediatric gastroenterologist; and (3) ability to speak language adequately. Adolescents with other chronic or acute gastrointestinal disorder were excluded from the study. All patients with IBD were classified at the time of the evaluation, as being in remission or in active state/relapse according to the Pediatric Crohn's Disease Activity Index (PCDAI) [20] or the Pediatric Ulcerative Colitis Activity Index (PUCAI) [21], which were completed by paediatric gastroenterologists. Remission is defined by a total score <10 and active state/relapse by a total score \geq 10 for both scales. For patients in relapse, disease severity was classified as mild, moderate or severe. All patients with FGIDs diagnosed based on Rome IV criteria and have not been treated with any medications.

The control group was recruited from two public high schools. Adolescents with developmental pervasive disorder or intellectual disability, and a comorbid chronic illness were excluded from the study. Following data collection, two pupils with reported abdominal pain were excluded.

Procedure

After taking clinician's consent to inform the family about the study, both parents and adolescents signed an informed consent and completed the questionnaires during waiting time for their appointment at the outpatient General Gastroenterology and IBD Clinics. In addition, members of the National Society of Crohn's disease and ulcerative colitis patients were invited to participate in the study completing online questionnaires.

Regarding the school sample, students from 5 classes of two public high schools were informed about the study. Information sheets and parental consent forms were distributed to students

in their classroom. Adolescents and parents/guardians who gave their consent for participation in the study, were asked to complete the questionnaires at home and return them to researchers within the next week.

Measures

The Parental Bonding Instrument (PBI) [22] is a 25 item questionnaire that assesses two dimensions of parental behavior (care and protection), as perceived by adolescents during their first 16 years. There are four types of parental bonding: optimal parenting, affectionless control, affectionate constraint and neglectful parenting. In the present study, we used the Greek-validated PBI which has demonstrated a high level of test-retest reliability [23]. Alpha coefficient is ranging in this study from 0.71 to 0.90.

The Functional Disability Inventory (child- and parent-report FDI versions) [24] assesses the physical and psychosocial functioning of children and adolescents aged 8-17 years, with a variety of pediatric health issues, over the past 2 weeks. The internal consistency reliability found to be 0.86 and 0.94 respectively in the current study.

The Abdominal Pain Index (API) [25] assesses the intensity, frequency and duration of patients' abdominal pain episodes, experienced during the previous 2 weeks. It consists of 4 items and it has been proven as a valid and reliable measure of abdominal pain in children and adolescents aged 8-18.

The Pain Catastrophizing Scale consists of two versions: child (PCS-C) [26] and parent (PCS-P) [27]. It includes three dimensions, each of them contains 13 items: Rumination, magnification and helplessness. As no Greek version was found for child- and parent-report API versions as well as PCS-C and PCS-P, the backward translation process was followed by three professional translators for the needs of the present study.

The Strengths and Difficulties Questionnaire (SDQ) [28] is a self-report screening instrument assessing mental health symptoms in children and adolescents. Cronbach's alpha was equal to or higher than 0.70 for all dimensions of both adolescents' and parents' reports in the current study.

The parents and adolescents completed all of the mentioned above questionnaires, apart from the PBI, which was completed by adolescents. The controls completed the aforementioned questionnaires except from the API, PCS-C/P and FDI.

Statistical analysis

Quantitative variables were expressed as mean (standard deviation) and as median (interquartile range). Qualitative variables were expressed as absolute and relative frequencies. For the comparison of proportions chi-square and Fisher's exact tests were used. Mann-Whitney test performed to compare quantitative variables between two clinical groups. Comparison of quantitative variables between more than two groups was performed using one-way Analysis Of Variance (ANOVA) or Kruskal-Wallis test. Bonferroni correction was conducted to control for type I error due to multiple comparisons. In order to assess the differences in SDQ, API, PCS and FDI scores among the study groups after controlling for age and gender, multiple linear regression analyses were conducted with the scores as dependent variables and age, gender, group as independent variables. In cases of non normal distribution log transformed values were used. All reported p values are two-tailed. Statistical significance was set at p<0.05 and analyses were conducted

using SPSS statistical software (version 22.0).

Results

Demographics and clinical characteristics

There were significant differences in demographic characteristics amongst the study groups (Table 1). The clinical characteristics such as IBD disease activity, severity and duration as well as the types of IBD and FGIDs of the patient groups are shown in Table 2.

Table 1: Demographic characteristic.

		Group						P
		IBD(n=40)		FGIDs(n=31)		Control(n=71)		
		n	%	N	%	n	%	
Adolescent gender	Male	18	45.0	15	48.4	29	40.8	.764‡
	Female	22	55.0	16	51.6	42	59.2	
Adolescent age, mean (SD)		14.4 (1.8)		13.3 (1.4)		13.1 (1.6)		<.001++
Residence	Athens	20	50.0	24	80.0	71	100.0	<.001+
	Other big city	3	7.5	1	3.3	0	0.0	
	City	10	25.0	1	3.3	0	0.0	
	Village	7	17.5	4	13.3	0	0.0	

*Pearson's chi square; †Fisher's exact test; **ANOVA.

Table 2: Clinical characteristics.

		Group			
		IBD (n=40)		FGIDs (n=31)	
		n	%	n	%
IBD activity	Remission	33	82.5	-	-
	IBD Relapse	7	17.5	-	-
IBD severity	Mild	4	57.1	-	-
	Moderate	3	42.9	-	-
	Severe	0	0.0	-	-
IBD	Crohn's disease	24	60.0	-	-
	Ulcerative colitis	14	35.0	-	-
	IBD unclassified	2	5.0	-	-
FGIDs	Functional abdominal pain	-	-	22	75.9
	Functional constipation	-	-	3	10.3
	Functional dyspepsia	-	-	4	13.8
Disease duration (years)					
mean (SD)		3.7 (3.1)		3.2 (3.7)	
median (IQR)		3 (1-5)		1 (1-4)	

Strengths and Difficulties Questionnaire (SDQ) scores

According to adolescents' self-reports, patients with FGIDs presented significantly higher rates in the emotional symptoms score, compared to those with IBD (p=0.010) and healthy subjects (p<0.001). Furthermore, adolescents with FGIDs scored significantly higher in the total SDQ score (p=0.006) compared to adolescents of the control group. Moreover, the healthy control group had significantly higher scores in the prosocial behavior dimension, than patients with FGIDs (p=0.016). According to parents' reports, adolescents of the control group presented significantly lower emotional symptoms compared to patients with FGIDs (p=0.003) (Table 3). Conduct problems as well as total problems in the control group were significantly lower,

Table 3: SDQ scores as assessed by the adolescents and their parents.

	Groups						P+ IBD vs FGIDs	P+ IBD vs control	P+ FGIDs vs control
	IBD		FGIDs		Control				
	Mean (SD)	Median (IQR)	Mean (SD)	Median (IQR)	Mean (SD)	Median (IQR)			
Adolescent report									
Emotional symptoms	2.42 (2.46)	2 (0-4)	4.04 (2.44)	4 (2-6)	2.17 (1.92)	2 (1-3)	<i>.010</i>	.973	<i>.001</i>
Conduct problems	2.61 (1.05)	2 (2-3)	3.12 (1.83)	3 (2-4)	2.59 (1.53)	3 (2-3)	.348	.943	.240
Hyperactivity	2.77 (2.11)	3 (1-3)	3.88 (2.38)	4 (2-6)	2.65 (2.17)	2 (1-4)	.059	.743	.021
Peer problems	1.23 (1.23)	1 (0-2)	1.80 (1.41)	2 (1-2)	1.37 (1.47)	1 (0-2)	.093	.842	.115
Prosocial behavior	8.13 (1.52)	8 (7-9)	7.92 (1.8)	8 (7-9)	8.8 (1.47)	9 (8-10)	.775	.022	<i>.016</i>
Total SDQ score	9.03 (4.92)	7 (6-12)	12.88 (6.4)	11 (8-17.5)	8.77 (4.88)	8 (5-12)	.023	.865	<i>.006</i>
Parent report									
Emotional symptoms	3.68 (2.92)	3 (1-6)	3.96 (2.44)	4 (3-5)	2.22 (1.92)	2 (1-4)	.483	.019	<i>.003</i>
Conduct problems	2.46 (1.8)	2 (1-3)	2.72 (1.67)	3 (1-3)	1.61 (1.4)	1 (0-3)	.475	<i>.011</i>	<i>.004</i>
Hyperactivity	3.33 (2.66)	3 (1-5)	3.17 (1.9)	3 (2-4.5)	2.18 (1.81)	2 (0-4)	.977	.040	.033
Peer problems	1.67 (1.87)	1 (0-2)	2.16 (2.23)	1 (0-4)	1.03 (1.24)	1 (0-2)	.511	.110	.039
Prosocial behavior	7.79 (2.1)	8 (6-10)	7.84 (1.93)	8 (6-9)	8.91 (1.55)	9 (9-10)	.950	<i>.006</i>	<i>.006</i>
Total SDQ score	11.24 (7.24)	9 (7-14)	11.96 (6.44)	11 (7-15)	7.04 (4.43)	6 (4-10)	.464	<i>.002</i>	<i><.001</i>

Note. p-values in *italics* indicate significant difference after Bonferroni correction +Mann-Whitney test

as parents' reported, compared to patients with IBD (p=0.011 and p=0.002 respectively) or FGIDs (p=0.004 and p<0.001 respectively). On the contrary, prosocial behavior in the control group was significantly better, according to parent's reports, compared to patients with IBD (p=0.006) or FGIDs (p=0.006). The aforementioned results were similar after adjusting for age and gender, with only exception being the hyperactivity/inattention dimension, as assessed by parents that no longer differ significantly among the three groups (p>0.05).

Abdominal Pain Index (API), Pain Catastrophizing Scale (PCS) and Functional Disability Inventory (FDI) scores

According to adolescents' and parents' reports, patients with FGIDs experienced significantly more functional abdominal pain, compared to those with IBD (p<0.001 and p=0.004 respectively), while adolescents with FGIDs reported significantly less functional ability than those with IBD (p=0.002). Furthermore, the PCS scores of patients with FGIDs were significantly higher

Table 4: API, PCS & FDI scores.

	Group				P+
	IBD		FGIDs		
	Mean (SD)	Median (IQR)	Mean (SD)	Median (IQR)	
API					
Adolescent	0.48 (0.56)	0.41 (0.00-0.78)	1.42 (1.03)	1.61 (0.25-2.05)	<.001
Parent	0.57 (0.65)	0.33 (0.00-1.03)	1.42 (1.22)	1.46 (0.00-2.4)	.004
FDI					
Adolescent	3.61 (6.32)	1.00 (0.00-4.00)	7.23 (6.60)	6.00 (4.00-9.00)	.002
Parent	5.11 (10.01)	1.00 (0.00-6.00)	5.09 (7.84)	2.00 (1.00-7.00)	.338
PCS					
Adolescent					
Rumination	5.48 (3.74)	5.00 (3.00-8.00)	8.31 (5.15)	8.00 (4.00-13.00)	.030
Magnification	2.33 (2.41)	2.00 (0.00-3.00)	3.97 (3.58)	3.00 (1.00-6.00)	.091
Helplessness	3.93 (3.62)	3.00 (1.00-5.00)	8.79 (6.87)	8.00 (2.00-13.00)	.005
Total PCS score	11.97 (8.94)	11.00 (6.00-15.00)	21.07 (14.7)	20.00 (10.00-32.00)	.011
Parent					
Rumination	12.32 (4.05)	13.50 (11.00-16.00)	12.58 (3.79)	13.00 (11.50-16.00)	.924
Magnification	5.92 (2.85)	6.00 (4.00-8.00)	5.29 (3.25)	5.50 (2.50-7.00)	.309
Helplessness	11.86 (5.94)	12.00 (7.00-15.00)	13.50 (6.28)	13.00 (9.00-18.50)	.351
Total PCS score	30.59 (10.14)	29.00 (24.00-38.00)	31.38 (11.98)	31.50 (23.50-39.00)	.756

+Mann-Whitney test.

than those of patients with IBD, in almost all dimensions including total PCS score ($p=0.011$) (Table 4). Parents' PCS scores did not differ significantly among the two groups. The aforementioned results were similar after adjusting for age and gender. API, PCS and FDI scores did not differ significantly with regards to disease activity in patients with IBD, nor did SDQ scores.

Parental Bonding Instrument (PBI) scores

PBI scores, and consequently PBI categories did not differ significantly between the three groups ($p>0.05$) (Figure 1). Furthermore, the within group analysis showed that the types of maternal bonding did not differ significantly by sex ($p=0.089$), age ($p=0.619$), disease duration ($p=0.913$) or IBD activity ($p>0.999$). The same was true for types of paternal bonding, which were comparable with regards to sex ($p=0.350$), age ($p=0.841$), disease duration ($p=0.197$) or disease activity ($p=0.185$) in patients with IBD.

Association between types of parental bonding, SDQ and FDI scores

Based on parents' assessments in the clinical groups, after Bonferroni correction for multiple comparisons, adolescents with maternal optimal parenting had significantly lower scores in the hyperactivity/inattention dimension (mean=1.80; SD=1.58), compared to those with affectionate constraint (mean=4.19; SD=1.91; $p<0.001$), or those with affectionless control (mean=6.60; SD=2.19; $p=0.001$). Furthermore, based on parents' responses, adolescents with maternal optimal parenting had significantly lower rates in the total SDQ score (mean=8.40; SD=5.19), compared to those with affectionate constraint (mean=12.86; SD=5.64; $p=0.002$) or those with affectionless control (mean=21.40; SD=10.06; $p=0.002$). Similarly, adolescents with maternal optimal parenting presented significantly less conduct problems compared to adolescents with affectionless control (mean=2.20; SD=1.19 vs mean=5.40; SD=2.51; $p=0.003$). Moreover, adolescents with paternal affectionate constraint had significantly higher scores in the emotional symptoms score compared to those with optimal parenting (mean=5.75; SD=3.14 vs mean=2.93; SD=2.10; $p=0.008$).

Focusing on every single clinical group separately, IBD patients with maternal optimal parenting showed significantly lower scores compared to those with affectionate constraint in the hyperactivity/inattention dimension (mean=1.88; SD=1.78 vs mean=4.70; SD=2.06; $p=0.004$) as well as in total SDQ score scale (mean=7.50; SD=3.03 vs mean=14.2; SD=7.35; $p=0.008$). Amongst adolescents with FGIDs, those with maternal optimal parenting, according to the parents' responses, had significantly lower scores in the hyperactivity/inattention dimension (mean=1.67; SD=1.22), compared to those with affectionate constraint (mean=3.73; SD=1.74; $p=0.014$) and those with affectionless control (mean=5.33; SD=1.15; $p=0.012$). Also, those with paternal affectionate constraint according to the parents' responses, had significantly less functional ability, compared to those with optimal parenting (mean=13.2; SD=12.32 vs mean=1.90; SD=2.60; $p=0.008$).

Discussion

This study is the first one that examined emotional and behavioral difficulties and attachment types in adolescents with IBD and FGIDs. According to parental reports, adolescents with IBD had significantly lower levels of emotional and behavioral difficulties compared to adolescents with FGIDs, while both pa-

tient groups showed significantly higher levels of emotional and behavioral difficulties compared to healthy controls. Furthermore, we showed that adolescents with IBD had comparable rates of insecure attachment compared to those with FGIDs and healthy controls. The gender and the age of adolescents, the duration of disease and the activity of IBD did not differ in regard to the type of parental bonding.

Consistent to a previous study [29], we found that adolescents with IBD reported comparable rates of emotional difficulties with the healthy controls. However, other studies [5,6] are inconsistent with our finding. As symptom activity goes along with psychological distress, a possible explanation is that in this study, the vast majority (82.5%) of patients with IBD were in remission. An important finding in our study was that, in line with previous reports [30], adolescents with FGIDs reported statistically significant more emotional symptoms, compared to those with IBD and healthy subjects, and more difficulties in total SDQ scale than healthy subjects.

However, in our study, parents' reports demonstrated that both adolescents with IBD and FGIDs had significantly higher scores in almost all dimensions of the SDQ, compared to healthy peers. A study by Shain et al. [31] provided further evidence that heightened parental emotional distress may lead to parental overestimation of the adolescent's anxiety level, reinforcing the hypothesis that differences between adolescent self-reports and parent reports are due to parental distress. It is not clear from our study whether the above findings reflect adolescents' emotional difficulties due to the presence of a chronic disease as shown by previous studies [32] or reflect parental over-reaction due to parental distress, as shown by other studies [31].

Similar to previous findings, we found that adolescents with FGIDs had significantly higher rates of functional disability, abdominal pain and pain catastrophizing, compared to adolescents with IBD. Indeed, adolescents with FGIDs report more usual pain compared to adolescents with IBD [12].

Our findings are in contrast to previous reports on adult patients with IBD, which had showed that patients with IBD exhibited a predominantly insecure attachment [17]. A potential reason for this inconsistency is that adolescents' perceptions of the parent-child relationship may be influenced by their developmental stage.

Furthermore, we showed that according to parental reports, adolescents with maternal optimal parenting corresponding to secure attachment, had significantly lower scores on hyperactivity/inattention and total SDQ score scales compared to those with affectionate constraint and affectionless control corresponding to insecure attachment. These findings are in line with a previous study [33] demonstrating that insecure attachment of youth with a history of abdominal pain predicted higher levels of mental health difficulties. Furthermore, according to parental reports, adolescents with maternal optimal parenting had significantly fewer conduct problems compared to those with affectionless control. Disturbances in early parent-child interactions have been associated with the development of behavioral difficulties and high aggression in children over 5 years of age [34]. Moreover, child disruptive behavior can lead to stress and reduced parental self-efficacy, leading to negative and inconsistent parenting practices that reinforce behavioral difficulties [35].

Concerning associations between the attachment styles and functioning of adolescents as reported by parents, we found differences among patients with FGIDs: adolescents with FGIDs who perceived their parental bond with fathers as affectionate constraint, had significantly lower functioning level compared to those with optimal parenting. In contrast to a previous study [36], we found that the above differences were not relevant to pain catastrophizing and the intensity, frequency and duration of abdominal pain.

This study has certain limitations. First, the cross-sectional design of the study and the sample size do not allow drawing conclusions on cause-effect associations. Second, the PBI may be completed by adolescents and adults but its latent variables may be perceived differently by these two age groups [37]. Additionally, since the PBI is a self-report questionnaire, it may be subject to bias. Nevertheless, the long term validity of the PBI has been proven in clinical pediatric populations [38,39].

Moreover, API was not validated in Greek population. However, it has been proven as a valid and reliable measure of abdominal pain in children and adolescents [33]. Finally, it has been shown that active, ongoing symptoms correlate with psychological distress [30]. Nevertheless, as in the present study the vast majority (82.5%) of patients with IBD were in remission, we can not predict whether adolescents with IBD who are not in remission would answer differently. Future studies should be conducted including patients with active disease.

In conclusion, the current study showed that adolescents with IBD and FGIDs experience higher level of total difficulties compared to healthy peers. Additionally, adolescents with FGIDs develop more emotional and behavioral symptoms compared to those with IBD. Our findings highlight the importance of psychological assessment of patients with IBD and FGIDs for detecting patients at higher risk for development of emotional disturbances or those with early symptoms.

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