ORIGINAL ARTICLE



Validation of child-adapted short scales for measuring gastrointestinal-specific avoidance and anxiety

Maria Lalouni^{1,2,3} | Ola Olén^{1,4} | Johan Bjureberg^{2,5} | Marianne Bonnert^{2,5,6} | Trudie Chalder⁷ | Erik Hedman-Lagerlöf³ | Silje Endresen Reme⁸ | Eva Serlachius^{2,5} | Brjánn Ljótsson^{3,5}

Correspondence

Maria Lalouni, Department of Clinical Neuroscience, Karolinska Institutet, Nobels väg 9, 17177 Stockholm, Sweden. Email: maria.lalouni@ki.se

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Abstract

Aim: To validate child-adapted shortened versions of the Irritable Bowel Syndrome-Behavioural Responses Questionnaire (IBS-BRQ; short scale denoted BRQ-C) and the Visceral Sensitivity Index (VSI; short scale denoted VSI-C) for children with functional abdominal pain disorders (FAPDs).

Methods: A child psychologist supervised by a child gastroenterologist was responsible for shortening the scales (BRQ-C, 11 items; and VSI-C, 7 items). Then, a sample of 89 children aged 8–12 years with FAPDs was used in the validation. Construct validity was assessed with correlations. Measures included gastrointestinal symptoms, quality of life, pain intensity and anxiety. Also, internal consistency, test–retest reliability, administration time and factor structure were assessed.

Results: Internal consistency for the BRQ-C and the VSI-C was $\alpha = 0.84$ and $\alpha = 0.80$, respectively. Correlations with related scales were similar between child-adapted scales and original scales, indicating construct validity equivalence. Correlations between short scales and original scales were high. Mean administration time was reduced by 47% (BRQ-C) and 42% (VSI-C), compared with original scales. Test-retest reliability was r = 0.72 for BRQ-C and r = 0.83 for VSI-C. BRQ-C had two factors (Avoidance and Bowel control). VSI-C had a unifactorial structure.

Abbreviations: BRQ-C, Behavioural responses questionnaire-child-adapted short scale; CBT, cognitive behavioural therapy; FACES, The Faces Pain Scale Revised; FAPDs, functional abdominal pain disorders; GI-anxiety, gastrointestinal-specific anxiety; GI-avoidance, gastrointestinal-specific avoidance; IBS, irritable bowel syndrome; IBS-BRQ, Irritable Bowel Syndrome-Behavioural Responses Questionnaire; PedsQL Gastro, The Paediatric Quality of Life Inventory Gastrointestinal Symptom Scale; PedsQL QOL, Paediatric Quality of Life Inventory; SCAS-S, Spence Children Anxiety Scale Short version; VSI, Visceral Sensitivity Index; VSI-C, The Visceral Sensitivity Index-Child-adapted short scale.

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¹Department of Medicine, Solna, Karolinska Institutet, Stockholm, Sweden

²Stockholm Health Care Services, Region Stockholm, Stockholm, Sweden

³Department of Clinical Neuroscience, Karolinska Institutet, Stockholm, Sweden

⁴Department of Paediatric Gastroenterology and Nutrition, Sachs' Children's Hospital, Stockholm, Sweden

⁵Department of Clinical Neuroscience, Centre for Psychiatry Research, Karolinska Institutet, Stockholm, Sweden

⁶Department of Medical Epidemiology and Biostatistics, Karolinska Institutet, Stockholm, Sweden

⁷Department of Psychological Medicine, Institute of Psychiatry, Psychology and Neuroscience, King's College London, London, UK

⁸Department of Psychology, University of Oslo, Oslo, Norway

Conclusion: The BRQ-C and the VSI-C were found to be time-saving, reliable and valid for children with FAPDs.

KEYWORDS

 $abdominal\ pain,\ functional\ gastrointestinal\ disorders,\ irritable\ bowel\ syndrome,\ paediatric\ psychology,\ psychometrics,\ validation\ study$

1 | INTRODUCTION

Paediatric functional abdominal pain disorders (FAPDs) are highly prevalent¹ and include irritable bowel syndrome (IBS), functional dyspepsia and functional abdominal pain – not otherwise specified.² Gastrointestinal-specific anxiety (GI-anxiety) is common in FAPDs and refers to anxiety about abdominal symptoms. GI-anxiety has been found to be a predictor of IBS in adults and to occur in individuals with IBS irrespective of anxiety disorders. GI-anxiety is often accompanied by gastrointestinal-specific avoidance (GI-avoidance), which refers to avoidance of situations and stimuli that might elicit abdominal symptoms (e.g. leisure activities, being in school and certain foods). While GI-avoidance can lead to short-term reduction of abdominal symptoms and GI-anxiety, it prevents the patient from gaining experience of being able to cope in the presence of these symptoms. This then maintains a vicious circle of GI-anxiety, GI-avoidance and symptom persistence.^{4,5} Reduced GI-anxiety and GI-avoidance have been shown to mediate symptom reduction in cognitive behavioural therapy (CBT) in adult, adolescent and paediatric FAPDs.⁶⁻⁹ Accordingly, GI-anxiety and GI-avoidance may be key mechanisms involved in the emergence and maintenance of paediatric FAPDs and constitute important treatment targets.

Despite the important role of GI-anxiety and GI-avoidance in FAPDs, there are no validated measures of GI-anxiety and GI-avoidance for children with FAPDs. In adults with IBS there are validated scales for both GI-anxiety; Visceral Sensitivity Index (VSI), ¹⁰ and for GI-avoidance; Irritable Bowel Syndrome-Behavioural Responses Questionnaire (IBS-BRQ). ¹¹ The aim of this study was, therefore, to validate child-adapted versions of the VSI and the IBS-BRQ and to assess their reliability and factorial structure in children with FAPDs.

2 | METHODS

2.1 | Participants and inclusion

Data from a randomised controlled trial 12 with 89 children (8–12 years old) with FAPDs (Sample 1) were used in the validation of the child-adapted short scales (ClinicalTrials.gov NCT02873078). Data from another treatment study (Sample 2), including 68 children and adolescents (ages 8–17 years) with FAPDs (ClinicalTrials. gov NCT03252743), were included in the factor analyses to increase statistical power (total N=157). Children 8–12 years gave oral informed consent within the original studies, adolescents 13–17 years

Key notes

- Gastrointestinal-specific anxiety and avoidance are common symptoms in children with paediatric functional abdominal pain disorders (FAPDs) and important treatment targets.
- Child-adapted short versions of the Irritable Bowel Syndrome-Behavioural Responses Questionnaire and Visceral Sensitivity Index were found to be reliable, valid and time-saving compared to the original scales.
- The short scales may be used to identify children with gastrointestinal-specific avoidance and anxiety and to repeatedly assess these symptoms during treatment.

and parents gave written informed consent within the original studies. Physicians within primary, secondary or tertiary care referred participants into both samples and certified the children's functional diagnoses. The studies were approved by the Regional Ethical Review Board in Stockholm (2016/1289-31; 2017/1342-31). Self-reports of the Rome IV criteria were used to differentiate between the subdiagnoses of FAPDs. Children were excluded if they had: (1) another somatic disorder that explained their symptoms, or (2) severe psychiatric or psychosocial problems in need of immediate care elsewhere.

2.2 | Measures

The Irritable Bowel Syndrome-Behavioural Responses Questionnaire (IBS-BRQ) is a 26 item scale for the assessment of controlling and avoidance behaviour related to IBS. The items are rated on a 7-point scale, between the endpoints: never and always. The IBS-BRQ has previously been shown to have high internal consistency ($\alpha = 0.86$) and high test-retest reliability (r = 0.81–0.85, p<0.001) in a study with adult participants. In the original validation of the scale two factors, avoidance and control, were extracted using principal component analyses. In

The Visceral Sensitivity Index (VSI) is a 15-item scale developed to assess GI-anxiety in IBS. In the VSI, the items are rated on a 6-point scale ranging from strongly disagree (1) to strongly agree (6). The scoring is then transformed to a 0-5 scale. The VSI has been found to have good psychometric properties with high internal consistency in different samples ($\alpha = 0.90-0.93$)^{10,13,14} and ability to predict IBS symptom

severity.¹⁰ The studies were in adult participants. The VSI has been found to have a unifactorial structure in several studies.^{10,13,14}

The Behavioural Responses Questionnaire-Child-adapted short scale (BRQ-C) is an 11-item scale adapted from the IBS-BRQ¹¹ within this study. The Visceral Sensitivity Index-Child-adapted short scale (VSI-C) is a 7-item scale adapted from the VSI,¹⁰ also within this study. The adaptation process is described in the section 'Development of the scales' in Appendix S1. The aims of the adaptation were to shorten the scales (to increase utility for screening, repeated assessments and use alongside other scales) and to increase the suitability for children with all FAPDs (as opposed to adults with IBS only), by changing wording when necessary and selecting the most relevant items for the target population. The first author (ML), a child psychologist with extensive experience of paediatric FAPDs and anxiety disorders was responsible for the adaptation of the scales under supervision of an experienced paediatric gastroenterologist (second author OO). The child-adapted short scales can be found in Appendix S2.

The Paediatric Quality of Life Inventory Gastrointestinal Symptom Scale (PedsQL Gastro) is a 9-item scale assessing the past month's gastrointestinal symptoms. The scale has 5 points ranging from never to almost always. The PedsQL Gastro has been found to have acceptable reliability (α = 0.77) for children with FAPDs and it has been shown to correlate with the measure of quality of life described below.¹⁵

Paediatric Quality of Life Inventory (PedsQL QOL) is a multidimensional 23-item scale assessing quality of life for children 8–12 years. The PedsQL QOL has high internal consistency ($\alpha=0.88$). The scale has also been shown to differentiate between healthy and ill children and correlates with measures of illness, indicating high discriminative and concurrent validity, respectively. ¹⁶

The Faces Pain Scale Revised (FACES) assesses pain intensity. Hand-drawn human faces with pain expressions corresponding to numbers 0 (no pain), 2, 4, 6, 8 and 10 (worst pain) are used to help the child rate their worst pain during the past week. The FACES has shown strong correlations with ratings of pain intensity (r = 0.92) on a visual analogue scale in children with chronic pain.¹⁷

Spence Children Anxiety Scale Short version (SCAS-S) is a 19-item scale that assesses anxiety in children 8–12 years. ¹⁸ The frequency of symptoms such as 'I am scared of the dark' and 'I worry about things' are assessed on a 4-point scale with answers ranging from Never to Always. SCAS-S has demonstrated a high internal consistency ($\alpha = 0.88$). ¹⁸

2.3 | Procedure

All assessments were self-administered via a secure online assessment platform.

Sample 1: The original IBS-BRQ and VSI scales and the adapted BRQ-C and VSI-C scales were assessed at screening or at pretreatment assessment in a randomised order. That is, half of the children first completed the IBS-BRQ and VSI-C at screening and then later completed the BRQ-C and VSI at pre-treatment assessment, while the other half of the children completed the scales in reverse

order. List randomiser at random.org was used to create a list for allocation to assessments at screening and pre-treatment. The other scales: PedsQL Gastro, PedsQL QOL, FACES and SCAS-S were assessed at the pre-treatment assessment. After the pre-treatment assessment, the children were randomised to internet-CBT or treatment as usual. The BRQ-C and VSI-C were then assessed with 14-day intervals. The first two assessments by the participants randomised to treatment as usual were used in test-retest reliability analyses (n = 44).

Sample 2: The children completed the BRQ-C and VSI-C at the pre-treatment assessment and these data were combined with Sample 1 in the exploratory factor analysis. All assessments are illustrated in Figure 1.

2.4 | Statistical analyses

Statistical analyses were conducted in STATA version $13.1.^{19}$ Internal consistency for the BRQ-C, VSI-C, IBS-BRQ and VSI was examined by calculating Cronbach's α . Test-retest reliability for BRQ-C and VSI-C was calculated with Pearson's product moment correlation r. Validity of BRQ-C and VSI-C were examined in two ways. First, the short versions were correlated with their respective long version using Pearson's r. Second, the construct validity equivalence between the short and long scales was examined by

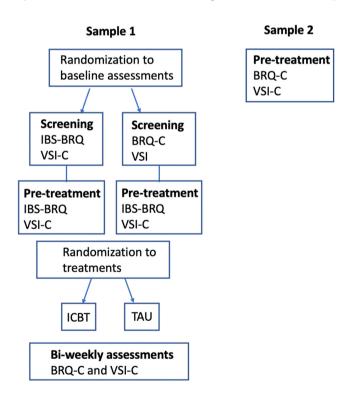


FIGURE 1 Assessments included in sample 1 and sample 2. BRQ-C, Behavioural Responses Questionnaire-Child-adapted version; ICBT, Internet-delivered Cognitive Behavioural Therapy; IBS-BRQ, Irritable Bowel Syndrome-Behavioural Responses Questionnaire; TAU, Treatment As Usual; VSI-C, Visceral Sensitivity Index-Child-adapted version; VSI, Visceral Sensitivity Index

comparing the correlations between the short scales and the measures of gastrointestinal symptoms (PedsQL Gastro), quality of life (PedsQL QOL), pain intensity (FACES) and anxiety (SCAS-S) with the correlations between the long scales and the same measures. The average time to administer the original and the child-adapted versions of the scales were extracted from the online assessment platform. Assessment times >15 min for the BRQ-IBS and BRQ-C and>10 min for the VSI and VSI-C were removed to reduce the influence of outliers on the average assessment time. Because the original scales had been adapted to a new population (i.e. children with all FAPDs vs adults with IBS), exploratory factor analyses were used to identify latent constructs. ²⁰ Eigenvalues over 1 were retained according to the Kaiser criterion. ²¹ Promax rotation, allowing factors to be correlated, was used to rotate the factors.

3 | RESULTS

3.1 | Baseline characteristics

Demographic characteristics of the participants, FAPDs diagnoses, means and standard deviations for all baseline measures included are presented in Table 1.

3.2 | Reliability

The internal consistency for the BRQ-C was $\alpha=0.84$ and for the VSI-C $\alpha=0.80$. The internal consistency of the IBS-BRQ was $\alpha=0.82$ and for VSI it was $\alpha=0.88$. The 14-day test-retest reliability (n=44) was r=0.72 for BRQ-C and r=0.83 for VSI-C.

3.3 | Validity

The correlation between the BRQ-C and the IBS-BRQ was r = 0.73, p < 0.001 and for the VSI-C and the VSI r = 0.70, p < 0.001. Correlations for the BRQ-C and the IBS-BRQ with the related measures administered were statistically significant and similar between the scales (Table 2). Likewise, the correlations for the VSI-C and the VSI with the related measures were statistically significant and similar between the scales (Table 3).

3.4 | Administration time

The mean time to administer the IBS-BRQ was 5:41 (minutes:seconds) and for the BRQ-C 3:00. For the VSI the mean administration time

TABLE 1 Demographics, FAPDs diagnoses and measures

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	Sample 1 ^a	Sample 2 ^b
	(n = 89)	(n = 68)
Demographics		
Age in years, mean (range)	10.3 (8-12)	12.3 (8-17)
Gender, n (%) female	61 (69%)	51 (74%)
Duration of abdominal symptoms, years, mean (SD)	3.7 (2.1)	4.6 (3.8)
FAPDs diagnoses, n (%)		
IBS	22 (25%)	24 (35%)
FAP-NOS	7 (8%)	8 (12%)
IBS and FD	22 (25%)	15 (22%)
FD	38 (43%)	21 (31%)
Measures, mean (SD)		
BRQ-C	31.47 (12.7)	26.8 (10.8)
IBS-BRQ	30.6 (11.3)	
VSI-C	14.24 (7.9)	13.4 (8.5)
VSI	20.7 (14.4)	
PedsQL Gastro	57.8 (13.9)	
PedsQL QOL	75.3 (13.2)	
FACES	6.1 (2.3)	
SCAS-S	12.9 (7.8)	

Abbreviations: BRQ-C, Behavioural Responses Questionnaire-Child-adapted version; Faces, Faces Pain Rating Scale; FAP, Functional abdominal pain; FAPDs, Functional abdominal pain disorders; FAP-NOS, Functional abdominal pain-not otherwise specified; FD, Functional dyspepsia; IBS, Irritable bowel syndrome; IBS-BRQ, Irritable Bowel Syndrome-Behavioural Responses Questionnaire; PedsQL Gastro, Paediatric Quality of Life Inventory Gastrointestinal Symptom Scale; PedsQL QOL, Paediatric Quality of Life Inventory; SCAS-S, Spence Children Anxiety Scale Short version; SD, standard deviation; VSI, Visceral Sensitivity Index; VSI-C, Visceral Sensitivity Index-Child-adapted version.

^aSample 1. Randomised controlled trial.

^bSample 2. Treatment study with a pre-test post-test design. Data from Sample 2 was only used in the exploratory factor analyses (in combination with Sample 1).

TABLE 2 Correlations for the BRQ-C and the IBS-BRQ with measures administered at pre-treatment

	α	BRQ-C (n = 89)	IBS-BRQ (n = 89)
PedsQL Gastro	0.62	-0.39**	-0.49***
PedsQL QOL	0.88	-0.52***	-0.59***
Faces	-	0.36***	0.36***
SCAS-S	0.84	0.42***	0.47***

Abbreviations: BRQ-C, Behavioural Responses Questionnaire–Child-adapted version; Faces, Faces Pain Rating Scale; IBS-BRQ, Irritable Bowel Syndrome-Behavioural Responses Questionnaire; PedsQL Gastro, Paediatric Quality of Life Inventory Gastrointestinal Symptom Scale; PedsQL QOL, Paediatric Quality of Life Inventory; SCAS-S, Spence Children Anxiety Scale Short version; α , Cronbach's α of the measures in the sample (α was not calculated for Faces because it is a single item scale).

was 3:30 and for the VSI-C 2:01. The administration time was reduced by 47% for the BRQ-C compared with the IBS-BRQ and 42% for the VSI-C compared with the VSI.

3.5 | Exploratory factor analysis

In the exploratory factor analysis (Sample 1 and Sample 2 combined) two latent factors were found (eigenvalues 3.74 and 1.30, respectively) for BRQ-C. The items with strong loadings in factor 1 were all related to avoidance behaviours and this factor was labelled *Avoidance*. The Avoidance factor explained 70.5% of the variance in the model. The items with strong loadings in factor 2 all concerned bowel controlling behaviours and this factor was labelled *Bowel control*. This factor explained 51.5% of the variance in the model. The correlation between Avoidance and Bowel control was in the medium range (r = 0.42). VSI-C had a unifactoral structure (eigenvalue 3.05 for Factor 1). Factor 1 explained 93.7% of the variance in the model. The factor loadings for the items in the BRQ-C and the VSI-C are presented in Table 4.

4 | DISCUSSION

The aim of the study was to validate the child-adapted short versions of the IBS-BRQ and the VSI, to be able to assess GI-avoidance and GI-anxiety in children with FAPDs. The child-adapted short scales, BRQ-C and VSI-C, showed high test–retest reliability and high internal consistency. Further, the child-adapted short scales showed equal construct validity, compared with the original scales. There was a reduction of administration time for the child-adapted short scales of 47% and 42%, which decreases the workload for children significantly. In the exploratory factor analysis we found two latent factors for BRQ-C: Avoidance and Safety behaviours. In the VSI-C we found a unifactorial structure.

The internal consistencies for the child-adapted short scales were slightly lower than for the original scales, which can be seen

TABLE 3 Correlations for the VSI-C and the VSI with measures administered at pre-treatment

	α	VSI-C (n = 89)	VSI (n = 89)
PedsQL Gastro	0.62	-0.36***	-0.47***
PedsQL QOL	0.88	-0.56***	-0.59***
Faces	-	0.33**	0.42***
SCAS-S	0.84	0.57***	0.53***

Abbreviations: BRQ-C, Behavioural Responses Questionnaire-Child-adapted version; Faces, Faces Pain Rating Scale; IBS-BRQ, Irritable Bowel Syndrome-Behavioural Responses Questionnaire; PedsQL Gastro, Paediatric Quality of Life Inventory Gastrointestinal Symptom Scale; PedsQL QOL, Paediatric Quality of Life Inventory; SCAS-S, Spence Children Anxiety Scale Short version; α , Cronbach's α of the measures in the sample (α was not calculated for Faces because it is a single item scale).

as a natural effect of decreasing the number of items.²² The correlations between the short and long scales were medium to high (r = 0.73 for the IBS-BRQ-BRQ-C and r = 0.70 for the VSI-VSI-C). Very high correlations were not expected since a main consideration in the selection of the items for the child-adapted short scales was that they should be relevant to children with different kinds of FAPDs and not only children with IBS.

In BRQ-C, the clear distribution of items into two behaviourally distinct latent factors: Avoidance (all items concern avoidance) and Bowel control (all items concern safety behaviours) supports the dimensionality found. The results also confirm the dimensionality found in the original validation of the IBS-BRQ. ¹¹ The latent factors were moderately correlated (r = 0.42), which may indicate that they represent different aspects of the same underlying construct. ²³ The unidimensional structure found in VSI-C, confirms the factorial structure found in several other studies. ^{10,13,14}

4.1 | Limitations

The sample size for the validation of the child-adapted scales was 89 children with ages 8–12 years. This is a small sample size in this context and a rather narrow age range, which is a limitation to the study. In the exploratory factor analysis, the sample size was 157, which is also rather small. Also, because the child-adapted scales were developed from adult scales, some aspects in paediatric FAPDs may be missing. However, we have found the child-adapted scales to be feasible for children to administer and in our prior studies we found they were sensitive to change, ¹² and able to capture relevant change processes. Pa larger study with a broader age range that could confirm the reliability, validity and the factorial structure of the scales in the paediatric FAPD population is recommended for future research. We also suggest future studies might assess whether the BRQ-C and the VSI-C can differentiate between children with FAPDs and healthy controls.

^{***}p < 0.001; **p < 0.01; *p < 0.05.

^{***}p < 0.001; **p < 0.01; *p < 0.05.

TABLE 4 Factor loadings of the explanatory factor analyses of the BRQ-C (Promax rotation) and the VSI-C (n = 157)

BRQ-C Item	Factors loadings ^a	
	Factor 1	Factor 2
	Avoidance	Bowel control
I strain when opening my bowels	-0.071	0.721
I spend more time on the toilet than I ideally would like	-0.012	0.767
I often go to the toilet to open my bowels and then do not pass anything	0.033	0.710
I avoid exercise when I have stomach pains	0.570	-0.002
I avoid certain foods because of my abdominal problems	0.405	-0.102
I avoid going out in case I have abdominal problems	0.685	0.072
I avoid making plans in case I have abdominal problems (e.g. doing things with friends after school or during the weekend)	0.729	-0.054
I avoid certain school situations (e.g. meetings, excursions) because of my abdominal problems	0.732	0.005
I avoid certain social situations (e.g. going to the cinema, eating at a friend's house) because of my abdominal problems	0.813	-0.030
After I open my bowels I wipe more than I would like	0.157	0.528
I avoid staying away from home overnight in case my abdominal problems flare up.	0.580	0.134
VSI-C Item	Factor loading	
I often worry about problems in my belly	0.794	
As soon as I feel abdominal discomfort I begin to worry and feel anxious	0.773	
I am constantly aware of the feelings I have in my belly	0.328	
As soon as I awake, I worry that I will have discomfort in my belly during the day	0.686	
When I feel discomfort in my belly, it frightens me	0.698	
In stressful situations, my belly bothers me a lot	0.572	
I constantly think about what happens inside my belly	0.653	

Abbreviations: BRQ-C, Behavioural Responses Questionnaire - Child-adapted short version; VSI-C, Visceral Sensitivity Index - Child-adapted short version.

4.2 | Clinical implications

BRQ-C and VSI-C may be used to identify children who have a high level of GI-avoidance and GI-anxiety. These children may respond particularly well to exposure-based CBT, considering that this treatment specifically targets behaviours and beliefs related to symptom-specific avoidance and anxiety. 12,24 Changes in gastrointestinal-specific avoidance and anxiety have been shown to mediate symptom improvement in CBT.⁶⁻⁹ Therefore, it may be particularly useful to administer the BRQ-C and the VSI-C repeatedly to be able to follow patients' trajectories of change during such treatment. Exacerbation of symptoms is not only present in functional abdominal disorders, but also in disorders with a clear pathology, such as inflammatory bowel disease (IBD) and celiac disease. A best practice update conclude that the psychological context is highly relevant for both FAPDs and disorders like IBD.²⁵ The BRQ-C and the VSI-C could thus also be used to assess GIavoidance and GI-anxiety in other disorders distinguished by abdominal pain.

5 | CONCLUSIONS

The current study presents the first evidence that the child-adapted short scales BRQ-C and the VSI-C are psychometrically reliable and valid measures of GI-avoidance and GI-anxiety in children with FAPDs. We conclude that the child-adapted short scales considerably lower the response burden for children compared with the original scales, particularly when repeated assessments are used.

AUTHOR CONTRIBUTIONS

ML, MB, JB, OO, ES, EHL, BL acquired the data. ML analysed and drafted the manuscript. All co-authors interpreted the data and critically revised the manuscript for important intellectual content.

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^aFactor loadings above 0.3 in bold.

CONFLICTS OF INTEREST

BL and EHL are shareholders of DahliaQomit AB, a company specialising in online psychiatric symptom assessment, and Hedman-Lagerlöf och Ljótsson psykologi AB, a company that licences cognitive behaviour therapy manuals. TC is part funded by the National Institute for Health Research (NIHR) Biomedical Research Centre at South London and Maudsley NHS Foundation Trust and King's College London. The views expressed are those of the author and not necessarily those of the NHS, the NIHR or the Department of Health and Social Care. Since this study was started, a private company has signed a licence agreement with King's College London with a view to bringing the Regul8 website product (cognitive behaviour therapy for irritable bowel syndrome) to the NHS and other international markets. TC will be a beneficiary of this licence through contracts with their respective universities. She has delivered workshops on persistent physical symptoms including bowel symptoms in the context of long-term conditions, during the conduct of the study for which she has received royalties. The remaining authors declare no conflicts of interest.

ORCID

Maria Lalouni https://orcid.org/0000-0002-6818-8156

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